The development and initial validation of an outcome measure for children and young people with life-limiting and life-threatening conditions

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The development and initial validation of an outcome measure for children and young people with life-limiting and life-threatening conditions.

A thesis incorporating publications submitted to King's College London for the degree of Doctor of Philosophy

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Statement of contribution

The contents of this thesis represent work conceived, planned, and undertaken by me under the supervision of Professor Richard Harding, Dr Katherine Bristowe and Dr Clare Ellis-Smith. My contribution to each of the objectives outlined in this thesis are as follows:

Objective i) – I was responsible for developing the systematic review protocol, conducting the review, disseminating the results, and writing the manuscript for paper 1 incorporated in this thesis as first author.

Objectives ii and iii) – the protocol and ethics application for the qualitative semi-structured interview study were written prior to commencement of this PhD. I attended the ethics committee and defended the protocol. I was responsible for conducting and co-ordinating recruitment, data collection, ethics amendments, data analysis/interpretation and dissemination of results. Recruitment and data collection were conducted by me, and other researchers employed on the C-POS study; data analysis and interpretation were led by myself with other C-POS team members contributing. I was responsible for dissemination of the main results, including writing the manuscripts for papers 2 and 3 included in this thesis as first author.

Objective iv) – I was responsible for developing the list of candidate items to be considered for inclusion in C-POS, using the evidence collected in objectives ii) and iii).

Objective v) – I was responsible for developing the protocol for the Delphi survey, applying for ethical approval, co-ordinating recruitment, and analysing and disseminating results. I also designed and led the item generation meeting and young person's advisory group meetings. I was responsible for writing the manuscript for this phase which has been accepted for publication (in press).

Objective vi) – I developed the cognitive interview study protocol and gained ethical approval for the study. I was also responsible for obtaining patient and public involvement (PPI) feedback on participant information sheets. I was responsible for co-ordinating recruitment and analysing and disseminating the results.

Acknowledgements

Firstly, I would like to acknowledge all the participants who contributed to the data presented in this thesis. I would like to particularly thank the children and young people who offered valuable perspectives on the development of C-POS despite being unwell. Their participation has ensured that C-POS has achieved the aim of being a child-centred measure. Also, thank you to the family members and healthcare professionals who gave up precious time to participate.

I am grateful to my supervisors, Professor Richard Harding, Dr Katherine Bristowe and Dr Clare Ellis-Smith for their support, education, and guidance throughout my PhD. I would also like to thank my thesis progression committee, Professor Fliss Murtagh, Professor Catherine Evans and Professor Matthew Maddocks for their insight, direction, and suggestions during my research. In addition, I would like to acknowledge the expertise on paediatric palliative care outcome measure development offered by Professor Julia Downing throughout my PhD.

I would like to thank the C-POS study steering group for always believing that developing C-POS was possible and for participating in steering group meetings with such enthusiasm. I would particularly like to thank the three parent members of the group - Angela Logun, Jane Green and Lydia Bate for giving up their time and offering a parent's perspective. I would also like to acknowledge the members of the young person's advisory group at Great Ormond Street Hospital for their eagerness to participate in meetings and offer their viewpoint on the development of C-POS.

My thanks go to the C-POS study research team for their help, encouragement, and friendship throughout this PhD. I would also like to acknowledge the children and young people's palliative care team at the Royal Marsden hospital for ensuring the service continued to run smoothly in my absence.

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Finally, I would like to thank my friends and family for their support throughout my PhD, particularly my husband and two children.

Table of Abbreviations

Abbreviation	Meaning
CCG	Clinical commissioning group
CCN	Children's community nurse
CoPPAR	Collaborative UK wide paediatric palliative care research network
COSMIN	COsensus-based Standards for the selection of health Measurement INstruments
C-POS	Children's palliative outcome scale
GOSH	Great Ormond Street Hospital
GP	General practitioner
MCA	Mental capacity act
MSAS	Memorial symptom assessment scale
NICE	National Institute for Health and Care Excellence
NIHR	National Institute for Health and Care Research
NHS	National Health Service
PCOM	Patient-centred outcome measure
PPI	Patient and public involvement
PROM	Patient-reported outcome measure
UK	United Kingdom
WHO	World Health Organisation
YPAG	Young person's advisory group

Abstract

Background

There is no validated outcome measure for use in children's palliative care outside of sub-Saharan Africa. Development of such a measure is required to realise the benefits of patient-centred outcome measure use that has been demonstrated in adult palliative care. Previous research into what is important to children and young people with life-limiting and life-threatening conditions has primarily focused on those with a cancer diagnosis. Much of this pre-existing research focuses on the perspectives of proxies, rather than those of the child or young person.

Aim

To develop an outcome measure, the children's palliative outcome scale (C-POS), for use by children and young people with life-limiting and life-threatening conditions and their families, and to establish face and content validity, comprehensiveness, comprehensibility, feasibility, and acceptability of use.

Methods

A sequential mixed-methods study was conducted in three phases, following the principles of patient-reported outcome measurement design described by Rothrock and the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN).

Phase 1 - gathering input

A systematic review was conducted with the aim of appraising the evidence on optimal recall period, response format and mode of administration to enable children and young people to participate in self-reporting on their health outcomes. A young person's advisory group was also consulted on the same topic.

To inform face and content validity of C-POS a semi-structured qualitative interview study was conducted to seek the perspectives of children and young people, their parents/carers and siblings, health care professionals and NHS commissioners on priority symptoms, concerns, and care priorities. Participants were also asked to identify their preferences for the design of C-POS, in terms of recall period, response scale format and administration mode.

Phase 2 - item generation

Part 1: Parents and professionals with experience in caring for a child or young person with a life-limiting or life-threatening condition participated in a three-round modified ranking-type Delphi survey with the aim of establishing which outcomes identified in phase 1 of this thesis should be included in C-POS.

Part 2: The young person's advisory group were asked to select their priority outcomes from the items ranked in rounds 2 and 3 of the Delphi survey.

Part 3: An item generation meeting was conducted with key stakeholders to develop initial C-POS versions based on the evidence collected so far.

Phase 3 - item improvement

Cross-sectional cognitive interview study to establish acceptability, comprehensiveness, and comprehension of the initial C-POS versions within the target population.

Results

Phase 1 - gathering input

Systematic review: Findings showed that children under five years old cannot validly and reliably self-report health outcomes. Face scales demonstrated better psychometric properties than visual analogue or Likert scales. Computerised and paper scales generally show equivalent construct validity and children prefer computerised measures. Children seven years old and younger often think dichotomously so may need two response options. Those over eight years old can reliably use a three-point scale.

Qualitative interview study: 106 participants were recruited: 26 children, 40 parents, 13 siblings, 15 health care professionals and 12 commissioners. Children found a short recall period and a visually appealing measure with 10 questions or fewer most acceptable. Children with life-limiting conditions were more familiar with using rating scales such as numeric and Likert than their healthy siblings and emphasised the importance of completing the measure alongside interactions with a healthcare professional. Parents assumed that electronic completion methods would be most feasible and acceptable but a small number of children preferred paper measures.

Participants described many inter-related symptoms, concerns and care priorities impacting on all aspects of life. Data revealed an overarching theme of pursuing 'normality', described as children's desire to undertake usual childhood activities. Parents need support with practical aspects of care to help realise this desire for normality.

Phase 2 - item generation

Part 1: Delphi survey (n=82). Ranking agreement between participants increased over the rounds, indicating movement towards consensus. Agreement between professional and parent ranking was poor. Professionals prioritised physical symptoms, whereas parents prioritised psychosocial and practical concerns.

Part 2: 22 children and young people attended the young person's advisory group. They prioritised items related to living a 'normal life' such as seeing friends and attending school, in addition to items prioritised by the adult participants in the Delphi survey.

Part 3: 22 participants attended the item generation meeting. Five age/developmental stage appropriate child/young person and proxy-reported versions of C-POS were drafted.

Phase 3 - item improvement

Forty-eight individuals participated (36 parents; 12 children) in cognitive testing of the C-POS versions. This revealed challenges in the acceptability of some items for parents of non-verbal children and refinements were made. C-POS content and length were acceptable, and all questions were considered important. Parents reported that completing a measure that asks about what matters may be distressing but this is anticipated and acceptable.

Conclusions

This thesis demonstrates the development of the first UK patient-centred outcome measure for use with children and young people with life-limiting and life-threatening conditions and their families. By following established methodological criteria for patient-centred outcome measure development this thesis demonstrates that C-POS has robust face and content validity and is feasible and acceptable for use within the target population.

COVID-19 Impact Statement

My PhD started in 2019 and data collection for phase 1 of this thesis was underway when the COVID-19 pandemic began. This impacted on my PhD objectives (see section 2.2 p45 for thesis aims and objectives) in the following ways:

Objectives ii and iii - when lockdown commenced data collection for the qualitative semi-structured interview study had to cease pending approval of an ethical amendment allowing interviews to be conducted online. This amendment also sought approval for data analysis and transcription to be conducted off site. This approval was received very quickly; however, it took several months to receive a university approved laptop to conduct the data analysis. Prior to the pandemic there was a requirement that analysis of all data needed to be conducted on university premises. There were challenges in participant recruitment once lockdown began as clinicians were extremely busy, and some were redeployed.

The first time the young person's advisory group was consulted regarding C-POS development was in July 2020. This was the first time the group had met virtually, having always met face to face prior to the pandemic. This may have impacted on the willingness and confidence of some members to participate, as video conferencing technology was a relatively new concept to most people early in the pandemic. Some members of the group may have been unable to attend due to lack of IT resources at home.

Objective v - the Delphi survey was conducted between November 2020 and February 2021. A further two UK lockdowns were announced during this period which impacted on the ability of parents and carers to participate. This is because vital sources of support such as respite care and education were impacted, meaning parents were experiencing a greater care burden in the home. The young person's advisory group conducted as part of this objective again had to be conducted virtually. Finally, the item generation meeting had initially been planned as a whole day face-to-face meeting. Due to social distancing rules this had to be conducted virtually and was held over half a day. This may have impacted discussion and interaction among participants and thus affected the final output which were initial drafts of the first versions of C-POS.

Objective vi - there was a significant delay in receiving NHS Trust sponsor approval for the cognitive interview study. This was because the review of non-COVID related

research studies was paused during the early part of the pandemic, creating a backlog of studies requiring review. Many research and development department staff in the sponsoring Trust had been redeployed to other areas creating further delays.

Despite these challenges all the work planned to be incorporated in this thesis has been achieved. This required me to take an extra six months to complete my PhD than initially planned.

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Publications and Presentations

Publications incorporated as part of this PhD thesis.

1. Coombes L, Bristowe K, Ellis-Smith C, Aworinde J, Fraser LK, Downing J, Bluebond-Langner M, Chambers L, Murtagh FEM, Harding R. Enhancing validity, reliability and participation in self-reported health outcome measurement for children and young people: a systematic review of recall period, response scale format, and administration modality. Qual Life Res. 2021 Jul;30(7):1803-1832.
2. Coombes L, Braybrook D, Roach A, Scott H, Harðardóttir D, Bristowe K, Ellis-Smith C, Bluebond-Langner M, Fraser LK, Downing J, Farsides B, Murtagh FEM, Harding R; C-POS. Achieving child-centred care for children and young people with life-limiting and life-threatening conditions-a qualitative interview study. Eur J Pediatr. 2022 Oct;181(10):3739-3752.
3. Coombes L, Harðardóttir D, Braybrook D, Roach A, Scott H, Bristowe K, Ellis-Smith C, Downing J, Bluebond-Langner M, Fraser LK, Murtagh FEM, Harding R. Design and Administration of Patient-Centred Outcome Measures: The Perspectives of Children and Young People with Life-Limiting or Life-Threatening Conditions and Their Family Members. Patient. 2023 May 23:1–11.

Publications in press

1. Coombes L, Harðardóttir D, Braybrook D, et al. Achieving consensus on priority items for paediatric palliative care outcome measurement: results from a modified Delphi survey, engagement with a children's research involvement group and expert item generation. Accepted for publication in Palliative Medicine.

Publications co-authored during PhD registration but not incorporated in this thesis.

1. Scott HM, Coombes L, Braybrook D, Roach A, Harðardóttir D, Bristowe K, Ellis-Smith C, Higginson I, Gao W, Bluebond-Langner M, Farsides B, Murtagh FE, Fraser LK, Harding R. COVID-19: Impact on Pediatric Palliative Care. J Pain Symptom Manage. 2022 Jul;64(1):e1-e5.
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3. Scott HM, Coombes L, Braybrook D, Harðardóttir D, Gaczkowska I, Harding R. Knowledge, attitudes and beliefs about paediatric palliative care. Ann Palliat Med. 2023 Jan;12(1):10-12.
4. Scott HM, Coombes L, Braybrook D, Roach A, Harðardóttir D, Bristowe K, Ellis-Smith C, Downing J, Murtagh FE, Farsides B, Fraser LK, Bluebond-Langner M, Harding R. Spiritual, religious, and existential concerns of children and young people with life-limiting and life-threatening conditions: A qualitative interview study. Palliat Med. 2023 Jun;37(6):856-865.

Oral conference presentations during PhD registration

1. Coombes L. The Children's Palliative Outcome Scale (C-POS) Study. Association of Paediatric Palliative Medicine Research Conference 2019.
2. Coombes L. The Children's Palliative Outcome Scale (C-POS) Study. EAPC 2020 EU studies session.
3. Coombes L. The Children's Palliative Outcome Scale (C-POS) Study. POS Workshop 2021.
4. Coombes L. Patient centred outcomes and the C-POS Study. ECHO Northern Ireland Paediatric Palliative Care Network Conference 2021.
5. Coombes L., Bristowe K., Ellis-Smith C., Downing J., Bluebond-Langner M., Fraser L., Chambers L. and Harding, R. Enhancing validity, reliability and participation in self-reported health outcome measurement for children and young people: a systematic review of recall period, response scale format, and administration modality. 5th UK National Patient Reported Outcome Measures Research Conference 2021.
6. Coombes L. and Harding R. The C-POS study. Yorkshire and Humber Children's Palliative Care Network Meeting 2022.
7. Coombes, L., Braybrook D., Scott H., Harðardóttir D., Waite F., Ellis-Smith C., Bristowe K., Downing J., Bluebond-Langner M., Fraser L., Murtagh F. and Harding R. Comprehensibility, comprehensiveness and acceptability of a novel paediatric palliative care outcome measure: cognitive interview data with children and families. European Association of Palliative Care Congress 2022.
8. Coombes L. and Harding R. The C-POS study. 12th Association of Paediatric Palliative Medicine Paediatric Palliative Care Conference 2022.
9. Coombes L. and Scott H. Involving children and young people in patient and public involvement during the C-POS study. COPPAR network webinar 2022.

10. Coombes L. Outcome measurement with children living with advanced cancer. World Cancer Congress 2022.
11. Coombes L. The C-POS study. ICPCN webinar, February 2023
12. Coombes L. Research Careers in Paediatric Palliative Care. COPPAR network webinar. March 2023
13. Coombes L. The challenges relating to measuring outcomes at the three extremes - childhood, life-limiting conditions, and older people. Invited plenary. 7th National Patient Reported Outcome Measures (PROMs) Research Conference. June 2023

Poster Conference Presentations during PhD registration

1. Coombes L., Bristowe K., Ellis-Smith C., Downing J., Bluebond-Langner M., Fraser L., Chambers L. and Harding, R. Maximising participation in patient reported outcome measures (PROMs) for children and young people: a systematic review of optimal recall period, response format and administration mode. European Association of Palliative Care Congress 2020.
2. Coombes L., Roach, A., Bristowe K., Ellis-Smith C., Bluebond-Langner, M., Downing J., Braybrook D., and Harding R. Priority outcomes for children and young people with life-limiting conditions: an in-depth qualitative interview study. European Association of Palliative Care Congress 2020.
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4. Coombes L., Roach A., Leyden D., Scott H., Harðardóttir D., Braybrook D., Ellis-Smith C., Bristowe K. and Harding R. Enabling children and young people to take patient and public involvement roles in paediatric palliative care research. European Association of Palliative Care Congress 2022.
5. Coombes L., Braybrook D., Harðardóttir D., Scott H., Gaczkowska I., Bristowe K., Ellis-Smith C., Downing R. and Harding R. Development and initial validation of a novel outcome measure for use with children and young people with life-limiting and life-threatening conditions. European Association of Palliative Care Congress 2023.

Chapter 1 Background

1.1 Paediatric Palliative Care

1.1.1 Epidemiology

It is estimated that each year 21 million children and young people with life-limiting or life-threatening conditions require input from palliative care services worldwide (1). Life-limiting conditions are those for which there is no hope of cure and from which children will die. Life-threatening conditions are those for which curative treatment may be feasible but may fail (2). Life-limiting and life-threatening conditions can be placed in to four groups (Table 1).

Table 1 Together for Short Lives categories of life-limiting and life-threatening conditions in children and young people (3).

Category	Title	Description	Example
1	Life threatening conditions for which curative treatment may be feasible but can fail	Access to palliative care services may be necessary when treatment fails. On reaching long-term remission or cure there will no longer be a need for palliative care.	Cancer, liver, kidney, or heart failure
2	Conditions where premature death is inevitable	There may be long periods of intensive disease directed treatments aimed at prolonging life. Children may be severely disabled but have long periods of relatively good health.	SMA type I, Duchenne muscular dystrophy
3	Progressive conditions without curative treatment options	Where treatment is predominantly palliative and may extend over many years.	Batten disease, mucopolysaccharidoses
4	Irreversible but non-progressive conditions causing severe disability leading to susceptibility to health complications and likelihood of premature death	Palliative care may be needed at any stage and there may be unpredictable and periodic episodes of care.	Severe cerebral palsy,

Children may move between groups or be in more than one group at any one time (3). More recently it has been proposed that there should be a fifth category, reflecting the growing awareness of the need for palliative care to start in the antenatal period when life-limiting or life-threatening conditions are diagnosed before birth:

5. Unborn children with major health problems who may not live through birth, infants who may survive for only a few hours/days, infants with birth anomalies that may threaten vital functions, and infants for whom intensive care has been appropriately applied but developed an incurable disease (4).

Within England, there are estimated to be over 86,000 children living with a life-limiting or life-threatening condition (Figure 1-1), with this number predicted to increase to between 96,000 and 121,000 over the next 10 years (5). This rise is in part due to advances in medical care such as increased survival of preterm babies, increased use of home ventilation and more aggressive management of the complications of life-limiting and life-threatening conditions in intensive care (6-8). This is resulting in increased pressure on the resources of paediatric palliative care teams, as these children and young people often have increased dependency requirements (2, 9).

Almost 400 life-limiting and life-threatening conditions have been identified as appropriate for palliative care among children and young people (10). It is estimated that deaths due to such conditions may account for 50% or more of the 5,000 deaths of children and young people in England and Wales each year (5). Mortality is highest in those under one year old, mainly due to perinatal and congenital conditions. It then decreases in middle childhood before rising again in adolescence(5). Acquired natural causes such as cancer are more prominent in these age groups (5). The prevalence of life-limiting and life-threatening conditions is highest for those with congenital abnormalities (11) (Figure 1-2). Those from Asian, Black and Bangladeshi backgrounds are more likely to have a life-limiting or life-threatening condition, with the lowest prevalence in those from Chinese backgrounds (Figure 1-3). Those from areas of higher areas of deprivation and boys are more likely to have these conditions (11).

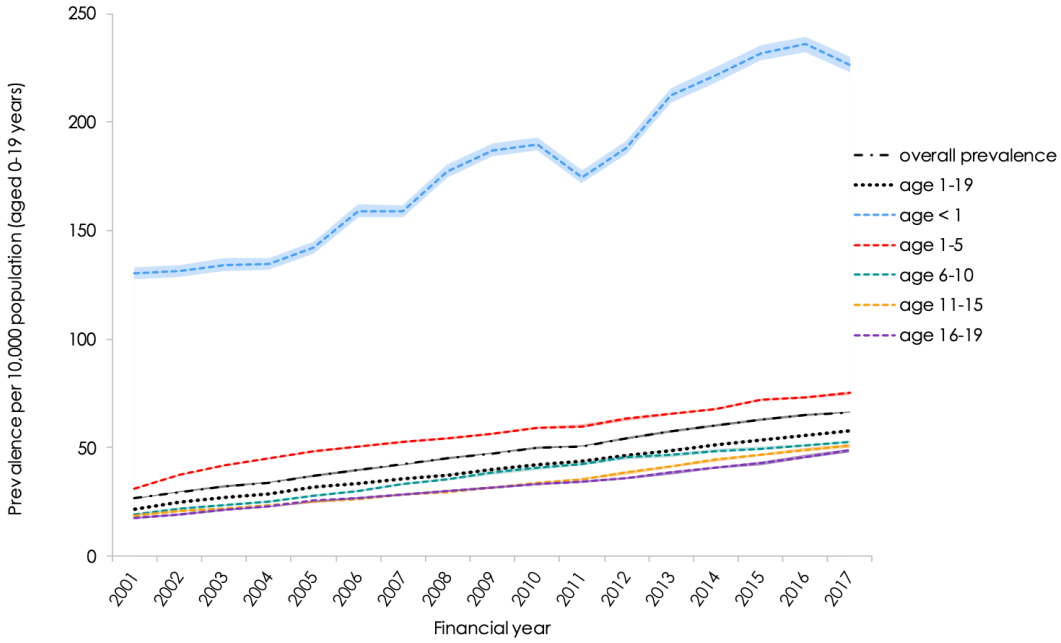


Figure 1-1 Prevalence of life-limiting/life-threatening conditions in England (with 95% confidence interval in lighter shading) in children 0-19 years overall and for age 2001/02 - 2017/18 (11).

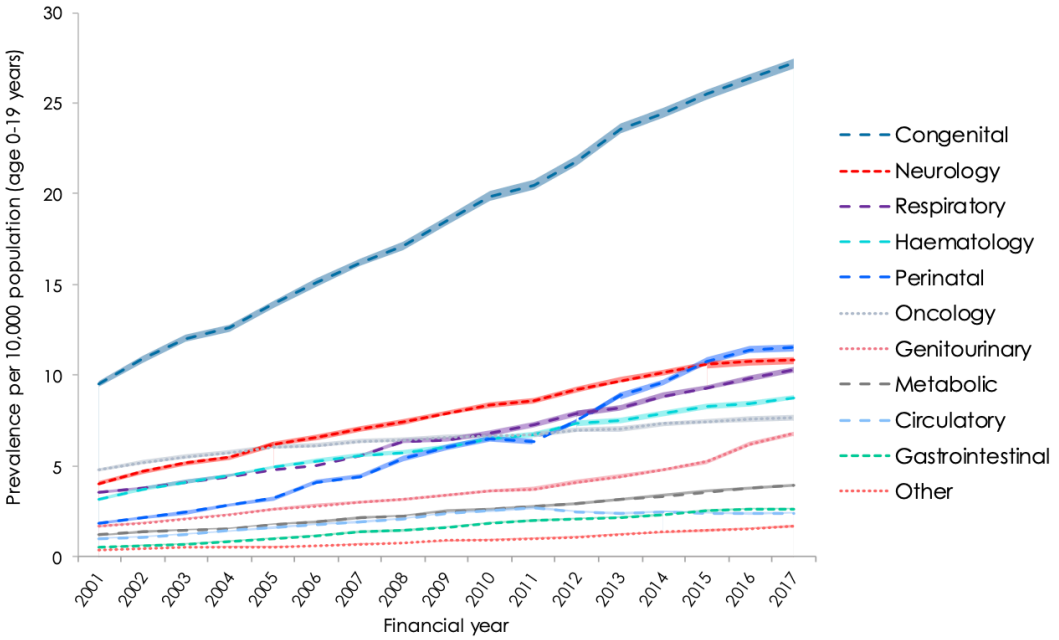


Figure 1-2. Prevalence of life-limiting/life-threatening conditions in England (with 95%) confidence intervals in lighter shading) in children age 0-19 years by diagnostic group for 2001/02-2017/18 (11).

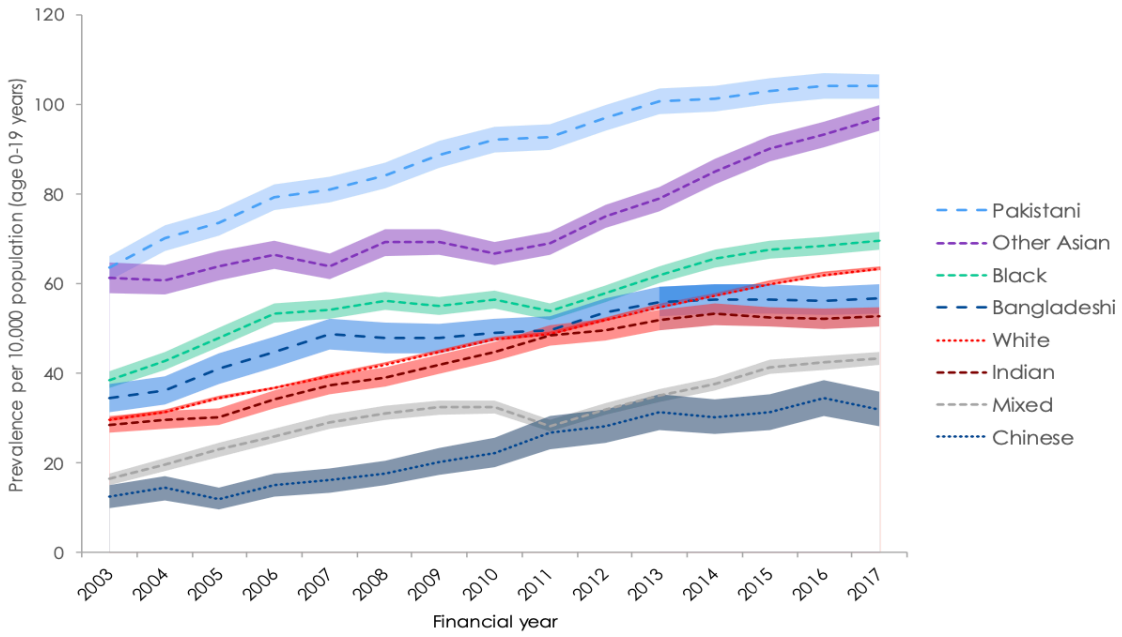


Figure 1-3. Prevalence of life-limiting/life-threatening conditions (with 95% confidence intervals in lighter shading) in children in England age 0-19 years by ethnic group for 2001/02-2017/18 (11).

The wide range of life-limiting and life-threatening conditions, along with longer survival and the use of more aggressive treatments are adding to care complexity for these children and young people. The changes that children and young people go through during childhood (physical, emotional and cognitive), the role of the family in care, and ethical and legal issues also contribute to this complexity (12). Health care professionals caring for children and young people with life-limiting and life-threatening conditions need a way to ensure that they are giving the right care to the child or young person and their family at the right time.

1.2 Symptoms and concerns in children and young people with life-limiting and life-threatening conditions

Symptoms and concerns in children and young people with life-limiting and life-threatening conditions are reported to be multidimensional, falling within physical, psychological, social, and spiritual domains, with quality of care and practical considerations also being key considerations (13). Symptoms and concerns occur across the disease trajectory and there is overlap across diagnostic groups (13). This means that the experience of symptoms and concerns for children and young people and their families is similar across the range of life-limiting and life-threatening conditions. Children and young people and their parents do not view

symptoms and concerns as independent, stand-alone constructs, rather as interrelated items that either facilitate or inhibit each other (14).

There is evidence that towards the end of life children and young people experience multiple physical symptoms including pain, lack of energy, weight loss, sleep disturbance, nausea and vomiting (15-25), with one study showing that children with cancer experienced a mean number of eleven symptoms in their last week of life (26). These symptoms are not always adequately identified or managed (27). Good control of physical symptoms is imperative and should be prioritised before any other concerns can be addressed (28).

Psychological concerns including feeling sad, feeling nervous, irritability, worry and insomnia have been reported by children and young people with life-limiting and life-threatening conditions (29). Social concerns such as isolation, stress on family relations and children and young people being concerned about the impact of their illness on their parents have also been reported (13, 30, 31). Spiritually children have been shown to worry about death and the uncertainty surrounding it (32, 33). Other reported concerns of children and young people and their families include the whole family being able to participate in normal life, and parental empowerment to be able to care for their child at home (28, 34).

Many studies of symptoms and concerns in children and young people with life-limiting and life-threatening conditions have only considered children with cancer, with children and young people with other diagnoses often not being included in research (13). There is also evidence that much of the research exploring the symptoms and concerns of children and young people with life-limiting and life-threatening conditions uses proxy reporting, with children being excluded from participating. One systematic review found that children and young people were not interviewed in 30% of such studies, instead reports from proxies such as health care professionals and parents/carers were used (13). It is important that future research into symptoms, concerns and care priorities for children and young people with life-limiting and life-threatening conditions focuses on including them as participants as they may have different views than proxies such as parent/carers and health care professionals (35). Children and young people also have experiences and relationships in their wider environment, such as school, which proxies will not be fully aware of (36, 37).

1.2.1 Paediatric Palliative Care Provision in the UK

Palliative care services for children and young people with life-limiting and life-threatening conditions in the UK are grouped into three tiers (3) – universal, core and specialist services (Figure 1-4) (38). All children in the UK have access to universal services such as GPs and education.

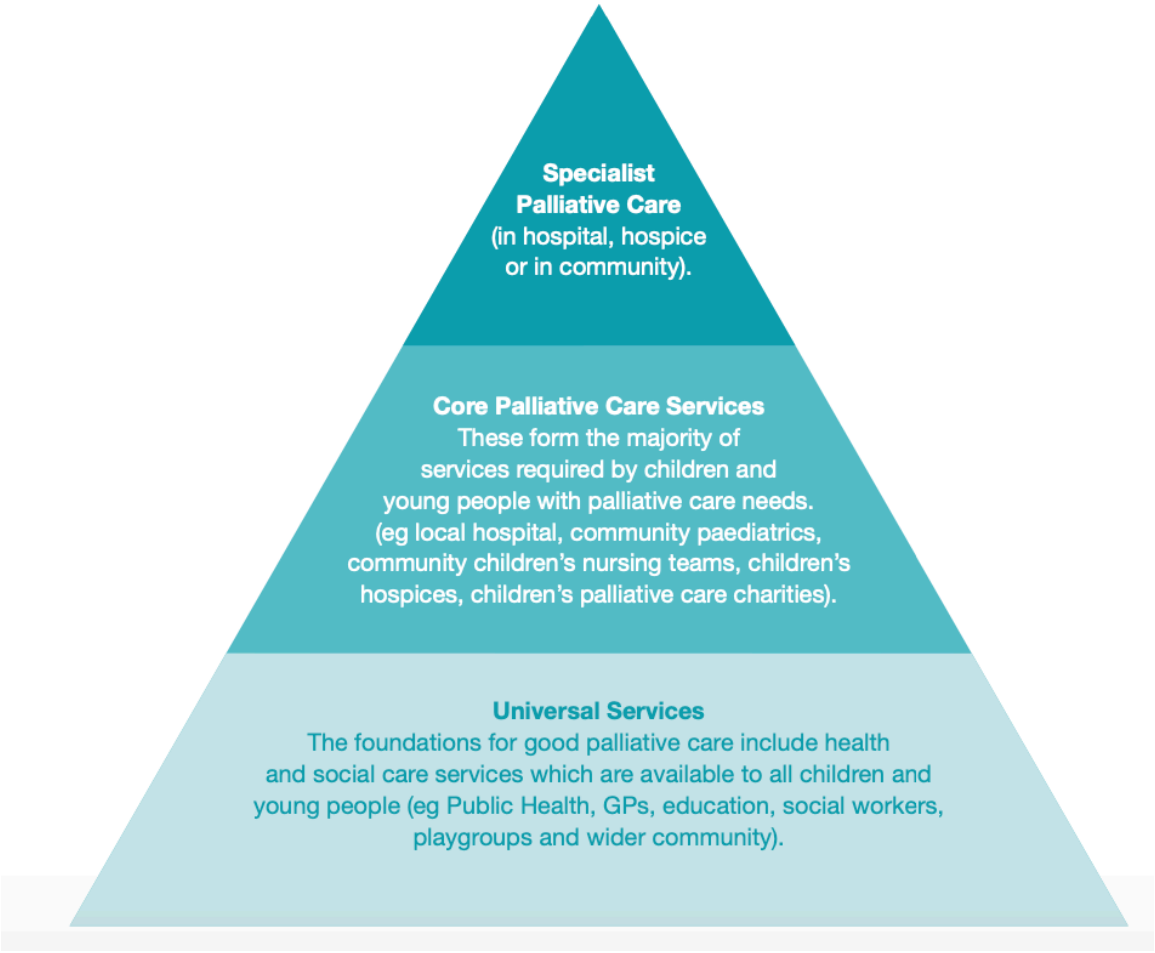


Figure 1-4. Levels of children and young people's palliative care services in the UK (3).

Most children and young people with life-limiting and life-threatening conditions will require access to core care services, which may also be available to other children with health care needs. These core services include community paediatricians, children's community nursing (CCN) teams, respite care and children's hospices. The UK currently has 54 children's hospices, some of which provide specialist paediatric palliative care services and some providing core services. There is no

data on how many hospital and community teams there are providing core children's palliative care services in the UK (39).

Recent data from England show that there is variation in access to core services, with 93% of what were at the time Clinical Commissioning Groups (CCGs; now integrated care boards) commissioning CCN teams during working hours. This number falls to 67% during nights and weekends, which has an impact on the provision end of life care in the home setting (40). This is in part because there are too few CCNs employed by the National Health Service (NHS). The Royal College of Nursing safe staffing levels suggest there needs to be 5,500 CCNs in England, but as of 2019 there were only 574 (39). The nurse vacancy rates in children's hospices are also rising as there are not enough skilled children's nurses to fill posts, and there is also a shortage of allied healthcare professionals to support children and young people with life-limiting and life-threatening conditions and their families (39). Local authorities in the UK have a legal duty to provide short breaks (respite care) to families of children with disabilities, including those with life-limiting and life-threatening conditions (41). These can be provided via centres (such as children's hospices), foster placements or direct payments. This allows them to spend time doing things that other families might do and can relieve stress on the whole family. In 2019, 21% of local authorities were failing to commission short breaks for children and young people (39). These gaps in core services have an emotional and psychological impact on children with life-limiting and life-threatening conditions and their families and may affect management of distressing symptoms and achievement of preferred place of care. There is no data available on the availability and staffing levels of core paediatric palliative care services post the COVID-19 pandemic, but it is anticipated that there are still significant challenges within the sector.

Specialist paediatric palliative care services can provide complex symptom management, respite care, end of life care in preferred place, compassionate withdrawal of life-sustaining treatment, sibling support and bereavement services (3). These services are accessed when required and children and young people and their families may require varying levels of input depending on their current level of need. The UK has provided many of the developments and innovations in children and young people's palliative care provision (42). Paediatric palliative medicine (the contribution doctors bring to the provision of children and young people's palliative care) was recognized as a sub-specialty in the UK in 2009. In 2019 there were 18

Paediatric Palliative Medicine Consultants in the UK. However, the Royal College of Paediatrics and Child Health estimate 40-60 are needed to provide high quality, equitable palliative care to all children and young people with life-limiting and life-threatening conditions who require it (43).

The National Institute for Health and Care Excellence (NICE) guidance states that a specialist paediatric palliative care team should contain the following:

- a paediatric palliative medicine consultant
- a nurse with expertise in paediatric palliative care
- a pharmacist with expertise in specialist paediatric palliative care
- experts in child and family support who have experience in end of life care (for example, in providing social, practical, emotional, psychological and spiritual support) (44).

In 2017, only 29% of CCGs (now integrated care boards) commissioned services that provided this suggested specialist paediatric palliative care multi-disciplinary team (40).

Provision of paediatric palliative care services across the UK is inconsistent and incoherent, as it is not centrally managed (45). There are finite resources within paediatric palliative care services, and no national guidelines or measures to help providers to direct these resources to those who need them the most. The impact of this is that distressing symptoms may go unmanaged, and the emotional and psychosocial impact of life-limiting and life-threatening conditions on children and young people and their family will not be addressed. Without assessing and addressing these symptoms and concerns, children and young people with life-limiting and life-threatening conditions will not be able to maximise their opportunities in life.

1.2.2 Definition of paediatric palliative care

Palliative care for children and young people with life-limiting and life-threatening conditions is described as an active and total approach to care that begins at the point of diagnosis or recognition and continues throughout the child's life and death (3). The World Health Organisation (WHO) defines children's palliative care as 'the active total care of the child's body, mind and spirit, and also involves giving support to the family' (46). Palliative care should continue regardless of whether or not a

child or young person receives treatment directed at their disease (47). It embraces physical, emotional, social, and spiritual elements, focusing on enhancing quality of life and support for the whole family. Paediatric palliative care should help children and young people and their families deal with their medical conditions, while enabling them to live life to the fullest (48, 49). This definition fits with the multi-dimensional experience of symptoms and concerns reported by children and young people with life-limiting and life-threatening conditions and their families discussed above.

The needs of children with life-limiting and life-threatening conditions are unique and usually somewhat different from those of adults requiring palliative care (50), with many such conditions being extremely rare. As children's palliative care should begin at diagnosis of a life-limiting and life-threatening condition, many tend to require input from palliative care services for extended periods of time, sometimes into adulthood (51). Children's palliative care should be active and dynamic in its approach. Children should be free from distressing symptoms, and children and young people and their families should receive support to reduce the emotional and psychosocial effects of their condition, while optimising their opportunities in life (3).

This thesis will use the above definitions of children's palliative care and consider paediatric palliative care to:

- begin at diagnosis of a life-limiting or life-threatening condition and continue regardless of whether a child is receiving potentially curative treatment.
- embrace the physical, psychological, social, and spiritual/existential aspects of care for children and young people.
- include support for both the child or young person and their family.

1.3 Centredness in Paediatric Palliative Care

Over recent decades, the concepts of person-, family- and child-centred care have been developed and used within healthcare in an effort to move the focus of care away from the more traditional biomedical model. These concepts see the patient as a person, are holistic and multidisciplinary and recognise that people may need more than one professional to support them (52, 53). This recognition that patients have needs beyond the physical or biological aspects of illness is reflected in the

definition of children's palliative care (54). It is also recognised by NICE in their recommendation that a specialist paediatric palliative care team is multi-professional (55).

1.3.1 Person-centred care

Person-centred care is an increasingly common approach used to conceptualise care and is cited as key to quality healthcare (56). Person-centred care has become one of the major goals of health policy around the world despite there being no globally accepted definition (52, 57). It emphasises participation rights, good communication, patient-provider relationships and shared decision making (52).

Children exist in the context of a family and excellent care for the child must include attention to family needs (56). The care given by a parent is the most influential for a child's long-term health and well-being (58), and this care is especially important when a child is seriously ill (59). When a child is unwell, the whole family is affected and care must be planned around the whole family (60). Parents of children with life-limiting and life-threatening conditions need more psychological support to prevent them becoming distressed, which in turn leads to benefits for their children as their home environment will be more settled and parental caring capacity will be enhanced (59). Table 2 summarises some of the most widely used concepts of person-centred care. Although most of the concepts of person-centred care outlined in Table 2 include family and friends, either explicitly, or through dimensions such as 'understanding the person as a whole', their focus is on adults with autonomy (52).

Table 2 Commonly used concepts of person-centred care

Author	Dimensions
Stewart et al (61)	1. Understanding the person as a whole 2. Agreeing to the plan for healthcare management 3. Including prevention and promotion of health 4. Focusing on the doctor/patient relationship 5. Being realistic about personal limitations 6. Exploring the experience of the illness
Picker Institute (62)	1. Respect for patients' values, preferences and expressed needs. 2. Coordination and integration of care 3. Information, communication, and education 4. Physical comfort 5. Emotional support and alleviation of fear and anxiety 6. Involvement of friends and family 7. Transition and continuity 8. Access to care
Mead and Bower (63)	1. Biopsychosocial perspective 2. Patient as a person 3. Shared power and responsibility 4. The therapeutic alliance 5. The doctor as a person
Kitson (57)	1. Patient participation and involvement 2. Relationship between the patient and healthcare professional 3. The context of where care is delivered
Sidani and Fox (64)	1. Holistic care 2. Collaborative care 3. Responsive care

1.3.2 Family-centred care

Family-centred care is a concept that is widely used within children's healthcare (65). There is no single definition of family-centred care. The advantage to this is that in can be adapted very easily to different circumstances (66). Family-centred care has been described as an approach to healthcare delivery that is dependent on an ongoing relationship between patients, their families and health care professionals and is considered a standard in paediatric care (67). Family-centred

care is based on the understanding that the family is the child's main source of strength and support and that child and family perspectives and information are important in clinical decision making (68).

For many children with life-limiting and life-threatening conditions, day to day management of care is the primary responsibility of their family, with many parents/carers having to administer medical and nursing interventions that would traditionally have been carried out in a hospital setting (69). In order to be able to do this, parents need to gain knowledge of their child's treatment and condition (69, 70), learn how to identify and respond to symptoms (69, 71) and develop relationships with health care professionals (69, 72). Parents/carers want to be involved in care decisions but do not want to be solely responsible for them (73). The child and family perspective and information are important in clinical decision making (68).

Family-centred care offers a way for parents to be involved in their child's care and is about empowerment, respecting personal autonomy and recognition of human rights (69). It has been purported to be the ideal system to involve parents and families in children's healthcare. Shelton et al (1987) developed a framework for embedding family-centred care in practice and enabling collaboration and partnership between families and healthcare professionals (74) (Figure 1-5). This framework has been criticised for focusing more on the parents than the child (75). The active members in care are the healthcare professional and parent, with the child taking a more passive role (35) and the perspective of the ill child is not very prominent (76). Family-centred care has been criticised for being poorly defined and having different meanings across professional and patient groups (73). The evidence base relating to the impact of family-centred care on parental satisfaction and care delivery is weak and outcomes are difficult to measure (60).

1. The family is a constant in the child's life.
2. Parent–professional collaboration should be facilitated across all levels of health care.
3. The racial, ethnic, cultural, and socio-economic and diversity of families should be respected.
4. Family strengths and individuality and respecting different methods of coping should be respected.
5. Complete and unbiased information should be shared with families.
6. Family-to-family support and networking should be encouraged and facilitated.
7. Healthcare practices should respond to the child and family developmental needs.
8. Policies and practices should be adopted that provide families with emotional and financial support.
9. The design of health care should be flexible (added by Johnson 1990 (77)).

Figure 1-5. Components of Family-Centred Care (74, 77)

1.3.3 Child-centred care

Since the publication of the United Nations Convention on the Rights of the Child (1989) there has been a growing focus on children and young people as social actors and agentic beings, with a right to be involved in their own healthcare decisions (36, 78). Agency in children's healthcare has been defined as children's capacity to act deliberately, speak for oneself, and actively reflect on their social

worlds, shaping their lives and the lives of others. This definition entails that multiple forms of expression can be used to speak for oneself, including speech and bodily expressions, and that the capacity of children to enact agency is not dependent on adults as facilitators (79). A child can be an agentic being even if under law they do not have the capacity to make healthcare decisions alone. Article 12 of the United Nations Convention on the Rights of the Child supports this by stating that 'every child has the right to express their views, feelings and wishes in all matters affecting them, and to have their views considered and taken seriously (78).

NICE (2021) recommends involving 'all children and young people in decisions about their healthcare, unless they do not wish (or are unable) to be involved' (80). This recognition that children need to be active agents and have a right to participate means that they need to be positioned as equal partners in their healthcare, rather than as passive recipients (36). The relatively recent concept of child-centred care means positioning children and young people and their interests at the centre of thinking, and including them as active participants in healthcare (35). Child-centred care is considered an evolution of family-centred care whereby the central importance is the child and their participation in their own health and well-being needs (35). This shifts the focus onto the family being around the child, rather than the child being part of the whole family (35, 66). Unlike a family-centred approach, taking a child-centred approach to care acknowledges the wider environment and relationships a child has outside of the family, such as friends and education, and recognises that they have their own needs and rights to privacy and dignity. It also involves acknowledging that children's views are not always the same as their parents or healthcare professionals and the best interests of the child or young person must be the paramount consideration (35). The child is focused on as a person with their own experiences and wishes which need to be respected and negotiated, and which are separate to those of the family (81). Child-centred care still recognises the central role of the family in the child's life. The Department of Health states the following principles underpin child-centred care (82):

- a holistic view of the child, seeing them as more than just their illness.
- concern for the overall experience of the child and family.
- acknowledgement of children, young people, and their parents as partners in care.

- advocacy for services to be co-ordinated around the child and family's needs.
- ensuring appropriate transition to adult services.

The provision of children and young people's palliative care fits with the above principles of child-centred care, in that it is holistic and encompasses physical, social, emotional and spiritual aspects of care for the child and family (54). It also reflects the notion that paediatric palliative care should offer support for the whole family to allow the children and young people to live their life as well as possible (48, 49). This thesis will take a child-centred approach, acknowledging that children and young people can be active agents in their own healthcare and have a right to be heard in matters that affect them. This includes being participants in research that affects them.

1.4 Outcome measurement

1.4.1 Health outcomes and their measurement

A health outcome is defined as a change in current or future health status attributable to preceding healthcare (83). This definition comes from Donabedian's widely used three-part framework for assessing quality healthcare (83, 84). The other two components of this framework are structure and process (Figure 1-6), which are considered easier to measure than outcomes (85). Structure refers to the professional and organisational resources associated with the provision of healthcare such as availability of medications, resources, equipment, service hours and staff training (83, 86). Process addresses things done to and for patients to deliver the desired outcome, for instance, hospital referrals, medication prescriptions, staff visits and information giving (83, 86). This three-part approach to healthcare is possible because good structure increases the likelihood of good process and in turn, good process increases the likelihood of good outcome (83, 87). Outcomes are considered an important part of this process as they directly affect the patient and family, allowing care to be focused on the needs of the individual and what is most important to them, thus promoting a child-centred approach to care (85). This thesis will use the Donabedian definition of an outcome – a change in current or future health status attributable to a preceding healthcare intervention (83).

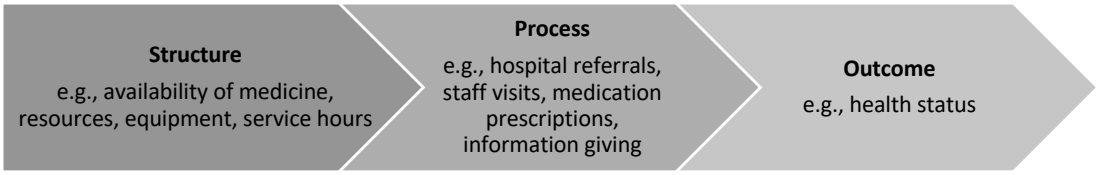


Figure 1-6. Donabedian model of structure, process, and outcome.

A patient reported outcome measure (PROM) is defined as any measure of a patient's health status, elicited directly from the patient. PROMs range from single item symptom ratings e.g., pain scales, to complex multidimensional health-related quality of life tools (88). PROMs are standardised, validated questionnaires that are completed by patients to ascertain perceptions of their health status, perceived level of impairment, disability and well-being (89, 90). PROMS are considered to be the gold standard for measuring subjective experiences, because each construct is inherent to each patient and the information comes directly from the patient (91, 92).

Many palliative care patients, including children and young people with life-limiting and life-threatening conditions will be too unwell or cognitively unable to self-report on their own health outcomes (93). Therefore, a measure which allows for proxy completion if the child is too unwell or unable to self-complete is required. Such measures are termed patient-centred outcome measures (PCOMs) (92, 93). These can be completed by a parent or proxy, and in addition to assessing patient outcomes they allow assessment of needs of unpaid carers, such as parents, in relation to concerns such as care organisation and financial constraints. This promotes a child-centred approach to care by incorporating outcome assessment for the whole family (87, 94).

The use of PCOMs in adult palliative care has been shown to improve service quality and promote patient-centred care (95) as well as leading to better symptom recognition, more discussion of quality of life and increased referrals (93). It has been recommended that outcome measures are used in palliative care to improve awareness of unmet need, understand different models of care delivery, and allow for national and international comparison (94, 96). Evidence for the use of PCOMs in paediatric palliative care is lacking, partly due to the absence of validated measures. There is some evidence from one systematic review that integrating PROMs into routine clinical care for children and young people with chronic

conditions could positively impact health-related quality of life, especially in the psychosocial and emotional domains (97). No evidence was found that PROM use changed referral rates or consultation duration in paediatric chronic conditions (97). This review included seven articles from six studies, and all included child self-report. As PCOMs for children and young people continue to be developed and validated it is imperative that research focuses on the benefits of using these in routine clinical care and where possible evidence for this is sought directly from children and young people.

It is intended that once developed and validated, the Children's Palliative Outcome Scale (C-POS) will be used as a clinical tool to assess symptoms and concerns of children and their families. This will allow the focus of current care to be on what matters most and promote a dialogue between the child family and clinician. In addition, C-POS could be used as an outcome measure in paediatric palliative care research studies, strengthening the quality of evidence generated. Another future uses of C-POS is comparison of different service models. As discussed in section 1.2.1 paediatric palliative care delivery models across the UK vary and there is currently no evidence on the best way to provide such care (45).

1.4.2 Patient-centred outcome measures in paediatric palliative care

It is important that during PCOM development the correct outcomes are chosen to be measured, as easily obtained but irrelevant outcomes are of little use (86, 92). The adult palliative care literature suggests that symptom assessment measures do not capture the broader concerns of those living with advanced illness, such as information needs, family and practical issues, and health-related quality of life measures are often weighted towards symptoms and function, which may be less relevant and applicable in palliative care (98). For a PCOM to assess paediatric palliative care outcomes it needs to incorporate not just physical symptoms but also the social, emotional and spiritual concerns of the child and family as defined by the World Health Organisation definition of paediatric palliative care (54). It also needs to be centred around the symptoms and concerns most important to the child and their family.

During the past decade, measuring outcomes in children and young people palliative care has repeatedly been identified as a research priority (99-102). Two Delphi surveys of professionals and parents of children with life-limiting and life-threatening conditions highlighted the need to measure and compare outcomes

(103, 104). The lack of outcome measures available for end-of-life and children were also highlighted as priority research areas by the Medical Research Council in 2009 (100). A 2016 systematic review to identify health-related quality of life measures that could potentially be used with children and young people with life-limiting and life-threatening conditions concluded that there were currently no ideal outcome measures available (105). Domains, recall period and response format of existing measures were not appropriate for children receiving palliative care and the methodological quality of included papers was limited (105). The most frequently used measure was the PedsQL™ generic core scale, however confirmatory factor analysis does not support its construct validity in children with life-limiting and life-threatening conditions, suggesting that development of a new measure is required (106). Other measures such as the Memorial Symptom Assessment Scale (MSAS) lack content validity as palliative care patients were excluded from validation studies and it was developed for children and young people with cancer (29, 107). Since the publication of this review, there has been further work in in the USA, Africa, and Belgium to develop a PCOM for paediatric palliative care (108-110). These are described further in section 1.4.3.

1.4.3 Existing patient-centred outcome measures used in children and young people with life-limiting and life-threatening conditions.

PediQuest

PediQuest is a computerised measure designed to capture symptoms and quality of life in children and young people with cancer (111). It incorporates the PedsQL 4.0™ (112), an adapted version of the MSAS 7-12 and 10-18 (29, 107) and a question asking how sick a child has felt (109). The MSAS was adapted to provide three age-appropriate versions – a 24 item self-report version for ages 13-18 year olds, an eight item self-report version for seven to twelve year olds with simplified response options administered alongside a parent proxy version asking the remaining 16 items. The parent proxy version is also used for two to six year olds (113). The adapted 10-18 version was shortened from 31 to 24 items in order to focus on symptoms known to be distressing in those with advanced cancer (hair loss, headache, weight loss, dizziness, taste changes, mouth sores and swelling of arms/legs removed) (109). The seven to twelve year old version was adapted by removing itch and adding dyspnoea (109). The PediQuest has been tested for feasibility, comprehensibility and acceptability in a pilot study. Validity and reliability

data for the adapted MSAS, and the PediQuest measure as a whole, have not yet been published (109). The original MSAS validation studies excluded children with advanced cancer, many of who may have different symptom experiences to those who are undergoing curative treatment (107, 114). The PedsQL™ has been reported to not be valid in a palliative care population for several reasons (115). Parents of children with life-limiting conditions reported that the content of several items was not applicable to their children - particularly items relating to physical functioning and school functioning. Items such as being able to walk 100 metres or run were not relevant for children and young people who were in a wheelchair or spent most of their time in bed. School functioning items such as being able to pay attention in class and keep up with schoolwork were also often regarded as not applicable (115). Furthermore, confirmatory factor analysis did not support the factor structure of the PedsQL in this population, suggesting that hypothesised health-related quality of life structures between children with life-limiting and life-threatening conditions and other populations may be different (115). As PediQuest is designed for those with advanced cancer it's content may not be valid for those with non-malignant life-limiting and life-threatening conditions or those near end of life.

African Children's Palliative Outcome Scale (C-POS)

The Palliative care Outcome Scale (POS) measures are a family of tools designed to measure patients' physical symptoms, psychological, emotional, spiritual, information and support needs (116). The POS measures are specifically developed for use among people severely affected by diseases such as cancer, respiratory, heart, renal or liver failure, and neurological diseases (116). Until recently all the POS measures were designed and validated for use with adults.

The African children's palliative outcome scale (APCA CPOS) began development in 2009 and was revised in 2014 (117). Expert consensus on key domains was obtained from paediatric palliative care experts and items were developed from this (Appendix A . The key domains identified for the measure were pain, symptoms, distress, quality of life, communication, and family support. The initial APCA C-POS consisted of 14 questions: nine for the child or young person and five for their parent/carer. Answers were scored using a 5-point Likert scale with numerical or descriptive labels (faces or hand scale) (118, 119). The tool was revised after pilot testing and review by paediatric palliative care experts from across sub-Saharan

Africa to include 12 questions: seven for the child or young person and five for the parent/carer (120, 121)) (Appendix B). The faces and hands scales were removed, and only verbal anchors for 0 and 5 included (121). Face and content validity, acceptability, and feasibility of use in practice has been demonstrated (122). Psychometric testing has recently been completed and is pending publication. Validation work showed that adolescents needed a separate measure to younger children (120). Development of the APCA C-POS began before recent accepted guidance on PROM development was proposed, and therefore the development process differs from this in that input on item generation was initially sought from expert children's palliative care health care professionals and not children and young people and their parents/carers, who are key stakeholders in the development of such a measure (123).

Belgian Children's Palliative Outcome Scale

The Belgian version of C-POS was adapted from the African version, starting with forward and back translation in to French, followed by a qualitative pilot study to assess face and content validity with children and parents (110) (Appendix C). The original 12-item African version was amended to include 22-items, with face and content validity in children and young people 8-18 years old in Belgium (110). The measure has the same overall domains as the African C-POS. Psychometric testing of the Belgian C-POS is pending publication (110). The need to make significant changes to the African version of C-POS may reflect the differences in population between the two regions. The Belgian version was piloted in children with cancer and neurological conditions whereas many of the children and young people included in the development of the African C-POS had conditions such as HIV and sickle cell disease. In high income countries these are seen as chronic conditions, rather than life-limiting or life-threatening and children and young people with such illnesses may have very different experiences to those with conditions such as cancer of metabolic conditions. Culturally, the two settings are also very different which may explain some of the adaptations required to the Belgian version.

1.4.4 Considerations when developing a PCOM for children and young people.

Establishing face and content validity

Content validity is defined as the degree to which the content of a measure is an adequate reflection of the construct to be measured (13). Face validity is the degree to which items of a measure look as though they are an adequate reflection of the construct to be measured (124). To develop a child-centred outcome measure for children and young people with life-limiting and life-threatening conditions that has face and content validity it is essential to find out what outcomes are important for children and young people and their families. As previously discussed, children and young people with life-limiting and life-threatening conditions are frequently excluded from taking part in research about them (13). This goes against the principles of child-centred care and children being agentic beings with the right and ability to be involved in matters that concern them. Involving children and young people with life-limiting and life-threatening conditions in research about what symptoms, concerns and care priorities are important to them during the development of a PCOM is considered an essential component of content validity (125).

Proxy reporting and inter-rater reliability

Inter-rater reliability is an important measurement property of a PCOM as they are intended to be used by proxies as well as patients. Inter-relater reliability is defined as the extent to which scores on a measure are the same with different observers e.g., child and parent. Therefore, it is important to understand similarities and differences that may be found between child and proxy reporting (124). As previously discussed, some children with life-limiting and life-threatening conditions will have illnesses that render them unable to complete a PCOM, either due to illness severity or impaired cognition. Proxy reports of symptoms are influenced not only by the child's experience, but also by the proxy's own health state and their expectations for the illness and symptom trajectory (126). Discordance is more pronounced during times of high symptom burden (126), and parents are often more negative regarding their child's health-related quality of life if their child has a chronic disease, and more positive if they are healthy (127). Patient proxy agreement is well known to be higher in more observable domains, such as mobility, than less visible domains such as emotions (128). Differences in correlation between physical

domains and psychosocial/emotional domains have also been reported. Physical and observable domains are consistently correlated more highly than social and emotional ones (129-135).

Research into parent/proxy and child correlation in report of health outcomes shows variation in correlation dependant on the child or young person's age (129-131, 133). Child and parent/proxy agreement for health-related quality of life in healthy children (129), childhood brain tumour survivors (130) and in children with epilepsy (133) found that younger children's scores correlated more closely with those of parent/proxies than those of adolescents.

In some studies children have reported having better health related quality of life than their parents have reported for them (136). However, in a systematic review of 14 studies no clear conclusion could be drawn as to how parents' perceptions of the impact of illness might differ from their child's perception (137).

A meta-analysis of nine studies examining whether a child's self-report of pain agreed with that of a parent found that a child's report of pain did not correlate strongly with the assessment of the child's pain by a parent (138). Most studies had a small sample size (less than 80) and there was a variation in age ranges between the included studies as well. This may have led to cognitive and developmental factors affecting results. Four of the nine studies included in the meta-analysis used a different pain scale for the child and parent/proxy, which may have been a cause for the weak correlation between scores.

In a population of children and young people with life-limiting and life-threatening conditions it will not always be possible to obtain child self-report. It is important when developing a PCOM for paediatric palliative care to have an understanding of the reliability of child-proxy agreement, for which the current evidence is mixed. Further evidence on inter-rater reliability between child and proxy should be obtained during psychometric testing of a PCOM to ensure the measure is working as expected.

Acceptability, feasibility, and self-report of health

Feasibility refers to the ability of a patient to be able to complete a measure (139). When developing a PCOM for children and young people, age and cognitive development must be considered (140). Children have a unique awareness of their

own experiences when it comes to reporting on their health and well-being (141) and the face validity of reports of health-related aspects is much greater when children and young people report their own perceptions (142). In order to be able to respond to PCOM questions, people must be able to go through four cognitive processing tasks: understanding and interpreting the question, retrieving the required information from memory, making a summarised judgement and reporting the judgement (143). In addition to this, people must have at least a rudimentary self-concept, understand the basic notions of health and illness, be able to pay attention, understand the questions, discriminate between the response options, recall health experiences and write a response (144). It has been proposed that people respond in one of two ways when going through these four cognitive processes: optimizing, whereby all four stages of cognitive processing described above are gone through, or satisficing, where a respondent gives superficial responses that appear reasonable (145). Two types of satisficing have been identified. Weak satisficing occurs when respondents execute all four cognitive processing stages but are less thorough in doing so (146). Less thought may be given to question meaning, memory may be searched less thoroughly, integration of retrieved information may be more careless and/or the response option may be selected more haphazardly. This results in respondents offering the first answer that they think will be acceptable to achieve the goal of responding to the survey question. Strong satisfcying occurs when respondents omit the retrieval and judgement steps altogether (146). Questions are interpreted superficially, and a response option is selected based upon the cues in the question itself. A response option is selected that seems easily defensible with very little thought (146). This may account for differences in the reliability of responses between respondents and for the effects of question wording (146). Low motivation, difficult questions or the cognitive abilities of the respondent may lead to satisficing rather than optimising. Due to age and developmental stage, children are often less cognitively able than adults, so may be more prone to satisficing, as they are less likely to understand complex questions and response formats (146, 147). Variability in children's development and ability means chronological age is not the only element for judging when children can self-report their health and complete a questionnaire. PCOMs for children and young people will need to have different versions for those with different developmental abilities to ensure feasibility, validity, and reliability. To inform PCOM development, evidence is needed on which response scales formats e.g., Likert and faces scales, children and young people can use at different

ages/developmental stages. Evidence is also needed on recall ability at different ages and developmental stages. This evidence will ensure that the measure is feasible for use in the intended population.

Acceptability of a PCOM refers to the patient's willingness to complete a measure (139). For children and young people, consideration needs to be given during measure design to preferences for administration mode (for example paper and pencil vs computerised), length and completion time to ensure acceptability. Evidence is needed on what children's preferences are for these aspects of a PCOM.

1.5 Summary

A health outcome is defined as a change in current or future health status attributable to a preceding healthcare intervention (83). This chapter has demonstrated that there is currently no valid and reliable PCOM for use with children and young people with life-limiting and life-threatening conditions in the UK. Development of such a measure has repeatedly been cited as a research priority due to the potential to improve care quality and awareness of unmet need, allow for service comparison and promote a child-centred approach to palliative care. There is a rising prevalence of life-limiting and life-threatening conditions within children and young people in the UK. This increased prevalence has not been met with an equivalent increase in healthcare resource. Development of a PCOM for this population may allow care to be focused on the symptoms and concerns that are most important to the children and young people and their family and support good care.

Development of a new PCOM for children and young people with life-limiting and life-threatening conditions is complex due to differences in age and developmental stage, the range of different life-limiting and life-threatening conditions children and young people may experience and the need for the family to be an integral part of their care. A child-centred approach to PCOM development needs to be taken, where children and young people are considered as agentic beings with a right to be involved in research and decisions regarding the healthcare they receive, while the needs of their family are also taken into consideration. This involvement of children and young people and their family in the measure development process will ensure that a PCOM is developed with robust face and content validity. Evidence is needed

on aspects of measure design such as recall period, response format and administration mode, all of which may be affected by the children and young people's age and/or ability. Ensuring these aspects are considered during the design stage will ensure that a PCOM is acceptable and feasible for use with its intended population. These aspects of measure design will be reviewed further in Chapter 4 and Chapter 5.

Chapter 2 Overview of Study Design and Methods

2.1 Introduction

This chapter outlines the aims and objectives of this thesis. It also summarises and justifies the main methodological and theoretical aspects of the study design and procedures. This includes an overview of study design, the ethical and legal considerations of conducting research with children and young people with life-limiting and life-threatening conditions and their families, and the patient and public involvement work conducted throughout this PhD.

2.2 Aims and objectives

2.2.1 Aim

To develop a child-centred outcome measure (C-POS) for use by children and young people with a life-limiting or life-threatening condition and their families, and to establish face and content validity, comprehensiveness, comprehensibility, feasibility, and acceptability of use.

2.2.2 Objectives

i. To determine optimal recall period, response format and administration mode for child-centred outcome measures in children and young people
ii. To establish child and family priorities for outcomes of care
iii. To establish healthcare professional and commissioner priorities for outcomes of care
iv. To develop a list of candidate priority outcomes to be included in C-POS
v. To gain stakeholder consensus on items to be included and construct first versions of C-POS
vi. To establish acceptability, comprehensiveness, and comprehension of C-POS versions

2.3 Overview of study Design

The design of this study is based upon the principles of PROM development described by Rothrock, and the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) (123, 148-150). Figure 2-1 shows the PROM development process described by Rothrock, incorporating the phases of this thesis. As discussed in Chapter 1, the need for a new instrument has already been identified by a previous systematic review which highlighted that there are no existing outcome measures suitable for use with children and young people with life-limiting and life-threatening conditions outside of sub-Saharan Africa (105). Having identified that development of a new measure is necessary, this thesis focuses on the Rothrock processes of gathering input, item generation and item improvement phases, using a sequential mixed methods approach, following the COSMIN standards on assessing the quality of content validity studies of PROMs (149).

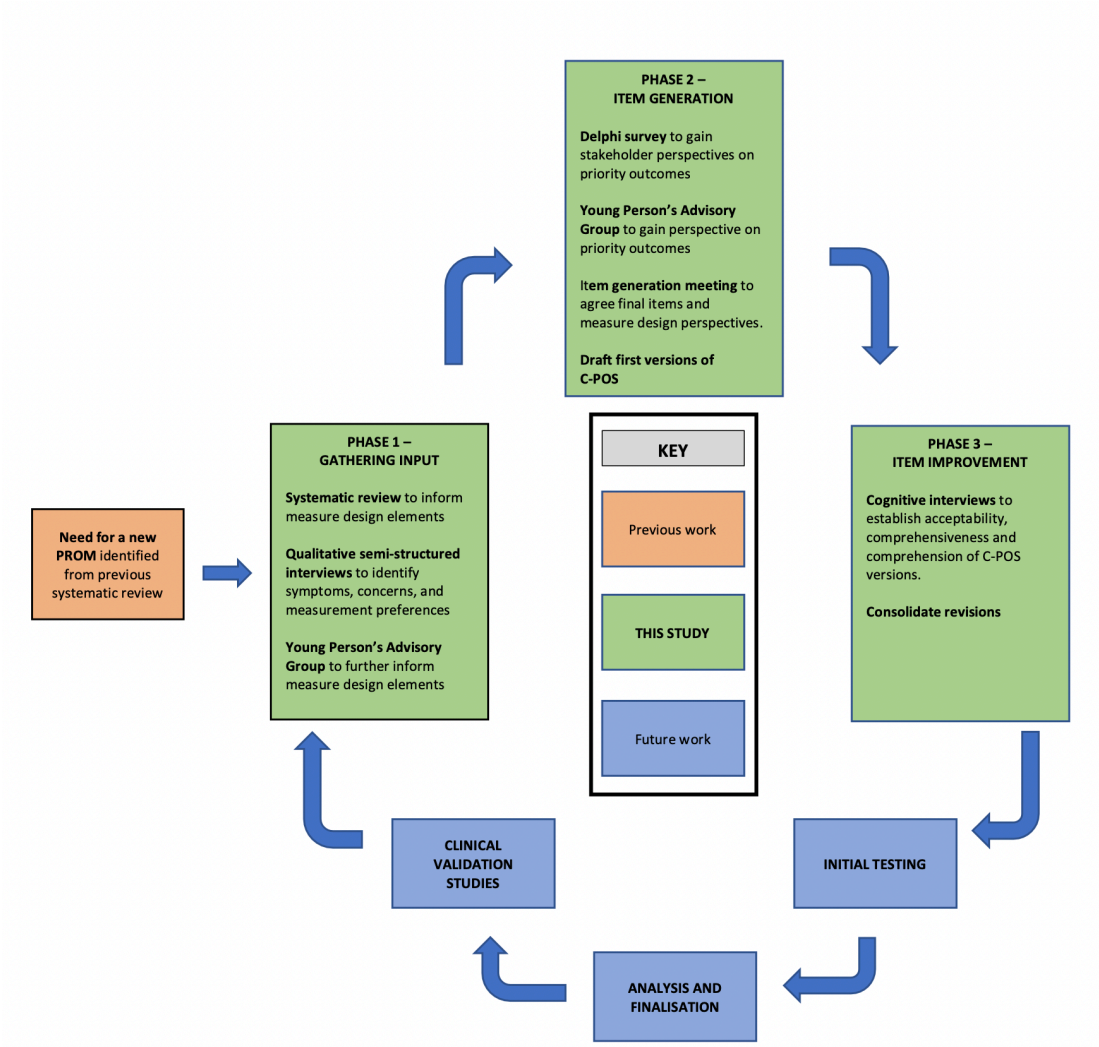


Figure 2-1 Components of PhD mapped on to Rothrock's PROM development process.

This thesis is split in to three phases (Figure 2-1):

- Phase 1 - gathering input (objectives i-iv):
    - Systematic review of optimal recall period, response format and administration mode for children and young people self-report of health outcomes.
    - Young person's advisory group (YPAG) workshop with children and young people to establish relevance, comprehensibility and acceptability of response formats, recall periods and administration modes.
    - Qualitative semi-structured interview study to establish symptoms, concerns, priority outcomes of care and measurement preferences for children and young people with life-limiting and life-threatening

conditions, their parents/carers and siblings, health and social care professionals, and service commissioners.
    - Development of a list of candidate priority outcomes to be included in C-POS
- Phase 2 – item generation (objective v):
    - Delphi survey to gain stakeholder (parent/carer and health and social care professional) perspectives on which outcomes identified in phase 1 should be included in C-POS.
    - YPAG meeting with children and young people to establish their priority outcomes from those identified in phase 1.
    - Item generation meeting with key stakeholders to agree on final items for inclusion in C-POS, along with recall period and response formats.
    - First versions of C-POS constructed based on evidence collected during phase 1 and 2.
- Phase 3 – item improvement (objective vi):
    - Cognitive interviews with children and young people with life-limiting and life-threatening conditions and their parents/carers to establish acceptability, relevance, comprehensiveness, and comprehension of initial C-POS versions.
    - Revision and retesting of C-POS versions as indicated.

The final output of this thesis will be parent/carer and child/young person versions of the C-POS with face and content validity, that are acceptable and feasible for use within the defined population, ready for psychometric testing.

2.4 C-POS and my contribution

The research presented in this thesis forms part of a larger study aiming to develop a child-centred outcome measure that can be used by children and young people affected by life-limiting and life-threatening conditions and their families, and to test its psychometric properties. Figure 2-2 shows an overview of the entire study, taken from the grant application.

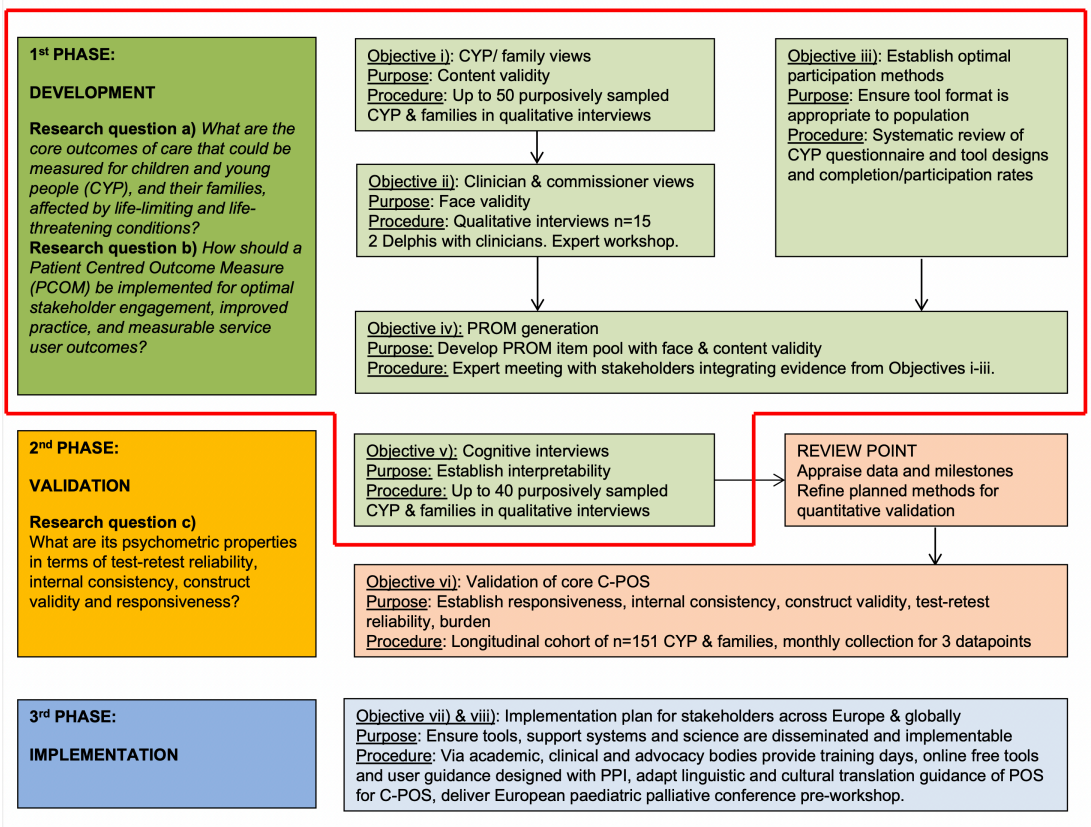


Figure 2-2 Overview of complete C-POS study

My PhD incorporates objectives i) – v) of the C-POS study outlined in Figure 2-2. My contribution is shown in the green boxes within the red outline. My contribution at each stage is outlined below by thesis objective:

- Objective i) – I was responsible for developing the systematic review protocol, conducting the review, and disseminating results.
- Objectives ii) - iv) – the protocol and ethics application for this were written prior to commencement of this PhD. I attended the ethics committee and was responsible for conducting and co-ordinating recruitment, data collection, ethics amendments, data analysis/interpretation and dissemination of results. Recruitment and data collection were conducted by me, and researchers employed on the C-POS study; data analysis and interpretation were led by myself with other C-POS team members contributing. I was responsible for dissemination of the main results.
- Objective v) – I was responsible for developing the protocol for the Delphi survey, applying for ethics approval, co-ordinating recruitment and analysing and disseminating results. I also designed and led the item generation

meeting and YPAG meetings. I was responsible for dissemination of this phase.

- Objective vi) – I developed the cognitive interview study protocol and gained ethical approval for the study. I was responsible for co-ordinating recruitment and analysing and disseminating results. I was also responsible for obtaining patient and public involvement (PPI) feedback on participant information sheets.

Objectives vi)-viii) of the original grant application do not form part of this thesis and will not be discussed further.

2.5 Mixed methods

The methods used for a research study should be informed by the underpinning research paradigm, epistemology and ontology, and the aims and objectives. Epistemology is the study of how we come to know what we think we know, what exactly we do know, how knowledge is structured and on what basis knowledge claims are made (151). Ontology is concerned with the nature of reality and what there is to know about the world (152). The aims and objectives of this thesis are best answered by using a sequential mixed methods approach, as described in the COSMIN standards for assessing the quality of content validity studies (149, 153).

Qualitative research focuses on the 'what, why and how' questions whereas quantitative research focuses on 'how many' (152). Mixed methods research is an approach whereby researchers gather both qualitative and quantitative data using the rigorous methods associated with each approach, integrate results from the two and draw interpretations based on the combined strengths of both sets of data to understand the research problem and describe different aspects of a phenomenon (154-156). This genre of research has gained traction in recent years, having first been described in the late 1980s (156, 157). It is recommended that a mixed method approach be used when developing patient-centred outcome measures (123).

A mixed methods approach has some challenges. It requires more time and resource to carry out than a study using qualitative or quantitative methodology alone. Researchers need to be trained in both qualitative and quantitative methods (153, 158). However, using a mixed methods approach also has advantages. If a study is designed well it can compensate for the weaknesses of one approach with

the strength of another, provide analytic texture to work and modify or strengthen the analytical findings when the results of each genre support, corroborate or contradict each other (155). For example, in quantitative research you don't hear the direct voice of participants, and qualitative research is not intended to enable generalisability (153, 157).

The research presented in this thesis forms an exploratory sequential mixed-methods design, with the first versions of C-POS as the output (153). The initial qualitative interview phase allowed for a deep understanding of the complexity of symptoms, concerns and lived experiences of children and young people with life-limiting and life-threatening conditions and their families. It also allowed for exploration of the relevance, comprehensibility and acceptability of different recall periods, response formats and administration mode of a PCOM. The findings from the systematic review were combined with data from the qualitative interviews and young person's advisory group to inform the recall period, response format and administration mode of C-POS. A quantitative Delphi survey allowed for a consensus of which of these symptoms, concerns and care priorities are of the most importance to stakeholders (parent/carers, and health and social care professionals) and should be considered for inclusion in C-POS. Evidence of priority of items to be included was further strengthened by work with a young person's advisory group (YPAG). The item generation meeting was conducted with expert stakeholders who were presented with the evidence from the above work and the first versions of C-POS were generated from this. Finally, cognitive testing of C-POS allowed for exploration of respondent's comprehension, retrieval, judgement, and response when using the measure to ensure that the questions are interpreted as intended and that the measure has content validity and is acceptable for use by the target population (143, 148).

2.6 Epistemological, ontological and theoretical considerations and reflexivity

All research has a philosophical foundation (158). In any research study it is important to consider ontology, epistemology and the overarching paradigm or world view, which also takes methodology in to account (152). Epistemology is ineluctably linked to research. This link becomes problematic in mixed methods research because of the philosophical differences between paradigms in the structure and confirmation of knowledge between qualitative (interpretivist paradigm) and

quantitative (positivist paradigm) studies (151). Failure to resolve this paradigmatic tension threatens the credibility of mixed methods research (151). A way to combine findings from paradigmatically distinct studies of common phenomena to maintain the integrity of methods and findings is required (151).

Within the mixed methods research community, pragmatism as a philosophical stance, has been promoted (158). Pragmatists argue that researchers should use whatever methods are needed to obtain optimum results, even if this involves switching between paradigms (159). They argue that philosophical disagreements are not fundamental and that research methods are not intrinsically linked to specific philosophical positions. Pragmatists argue that methods can be combined based on their practical utility and that paradigmatic tensions can be ignored (158). However, taking a pragmatist position may underestimate the actual influence of philosophical assumptions on research methods. This influence is particularly significant for combining qualitative and quantitative approaches (158, 159). Ontological and epistemological assumptions inevitably influence researchers' purposes and actions, and are not easily abandoned or changed (158).

An alternative philosophical approach to pragmatism for a mixed methods research study is critical realism. Critical realism combines a realist ontology (there is a real world that exists outside of our theories, perceptions and constructions) with a constructivist epistemology (our understanding of the world is a construction from our own perspectives and standpoints and there is no possibility of attaining an infallible view that is independent of any particular viewpoint) (160) Unlike pragmatism, critical realism accepts that every researcher has epistemological, ontological and axiological views which are often implicit and not easily abandoned (160). Critical realists distinguish between three levels of ontology (161-163); empirical (aspects of reality can be experienced directly or indirectly), actual (aspects of reality that occur but may not necessarily be experienced) and real (the structures and mechanisms that generate phenomena). These structures and mechanisms cannot be directly observed but can be inferred through empirical observation and theory construction (159). Critical realists believe that the choice of methods used should be dictated by the nature of the research problem (159).

A critical realist world view is consistent with a mixed method approach such as that recommended by Rothrock and COSMIN, and also the aims and objectives of the research presented here. As such it is the approach taken throughout this thesis.

The goal of critical realist research is to identify the lived experiences and beliefs of participants and develop deeper levels of understanding and explanation of the phenomena they describe. The symptoms, concerns and care priorities that influence health outcomes in children and young people with life-limiting and life-threatening conditions are complex phenomena influenced by multiple physical, social, and psychological factors (13). These need to be examined holistically and from multiple perspectives. The recruitment of participants from different stakeholder groups allows facets of reality to be examined from different perspectives (153) which is important within the context of child-centred care and outcomes.

A critical realist approach also allows for acknowledgement of the way the researcher affects the research process and outcomes (123, 164). All research is influenced by the researcher, particularly qualitative research, and there is no completely 'neutral' or 'objective' knowledge (152). Reflexivity is self-appraisal in research, and involves examining how one's own beliefs, judgements, personal experiences and behaviours influence the research process (165). It is therefore important that researchers recognise and reflect on any potential sources of bias and report on these (152). During both the qualitative and cognitive interview studies I kept reflective field notes (Appendix E This enabled me to reflect on how my prior experience as a paediatric palliative care nurse, and my assumptions and values may have influenced the interview process. They also helped identify any challenges during interviews. I also made reflexive notes throughout my PhD on supervision meetings, steering group meetings, data analysis and conversations with researchers, clinicians, patients, and carers regarding my research, to further enhance reflexivity.

2.7 COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN)

The consensus-based standards for the selection of health measurement instruments (COSMIN) criteria are methodological quality criteria (risk of bias criteria) for studies assessing the psychometric properties of PROMS. They also provide guidelines for doing methodological studies of psychometric properties of PROMS (164, 166, 167). The COSMIN checklist for assessing the methodological quality of studies on measurement properties of health status measurement instruments is used when designing a study on measurement properties (168). It

includes standards for conducting studies on validity, reliability and responsiveness (168). With regards to content validity, which is the focus of this thesis, the original COSMIN standards only considered whether things were done, not how. More recent guidance on evaluating the content validity of PROMS has been published, which contains new standards for evaluating the quality of PROM development studies (149). All COSMIN standards are based on literature reviews and subsequent consensus from an international Delphi survey of experts. The inter-rater agreement of the original COSMIN checklist is adequate and inter-rater reliability for many items is poor. It has been suggested that this is the result of variability in interpreting the checklist items (169). The inter-rater reliability of the more recent content validity standards has not been assessed. These standards are considered throughout this thesis to be the gold standard when developing and appraising content validity of PCOMS.

2.7.1 Content validity

Content validity refers to the degree to which a PROM is an adequate reflection of the construct to be measured and is often considered to be the most important measurement property (149, 170). Content validity should be established before evaluating other measurement properties (149), as lack of content validity can affect all other measurement properties of a PROM. For example, missing concepts may reduce validity and responsiveness, and irrelevant items may decrease internal consistency, structural validity and interpretability (149). Irrelevant or missing concepts may lead to lower response rates and biased responses (149). Content validity contains three aspects:

1. Relevance - all items within a PROM should be relevant for the construct of interest within the intended population and context of use.
2. Comprehensiveness – no key aspects of the construct being measured should be missing.
3. Comprehensibility – PROM items should be understood by patients (and carers) as intended (149, 171).

There are ten COSMIN criteria for good content validity, regarding item relevance, appropriateness of response options and recall period, comprehensiveness, and comprehensibility of the PROM (see Figure 2-3). Each criteria has specific standards for evaluating the quality of PROM development, updated standards for evaluating the quality of content validity studies of existing PROMs, criteria for what

constitutes good content validity, and a rating system for summarizing the evidence on a PROM's content validity and grading the quality of the evidence in systematic reviews of PROMs. The COSMIN standards for evaluating the quality of PROM content validity development have informed the methodology used throughout this thesis.

Relevance	
1	Are the included items relevant for the construct of interest?
2	Are the included items relevant for the target population of interest?
3	Are the included items relevant for the context of use of interest?
4	Are the response options appropriate?
5	Is the recall period appropriate?
Comprehensiveness	
6	Are no key concepts missing?
Comprehensibility	
7	Are the PROM instructions understood by the population of interest as intended?
8	Are the PROM items and response options understood by the population of interest as intended?
9	Are the PROM items appropriately worded?
10	Do the response options match the question?

Figure 2-3 COSMIN criteria for good content validity

2.7.2 Feasibility and acceptability

Feasibility

Feasibility is defined as the ease of application of a PROM in its intended context of use (148, 150). Aspects of feasibility include length, completion time, and type and ease of administration. Feasibility is not considered a measurement property as it is not referring to the quality of a PROM, however it is still an important consideration in measure development and is referred to in the COSMIN manual for systematic reviews of PROMS (148, 150). It is important to consider feasibility when designing a PROM in terms of both patients/carers completing it and clinicians/researchers administering it. A PROM that is easy to complete and administer in is more likely to be implemented into routine clinical practice with ease. Throughout this PhD aspects of feasibility were considered from the perspectives of children and young people, parents/carers, and clinicians by including them as research participants and involving them in aspects of study design and as members of the study steering group.

Acceptability

Acceptability is defined as the willingness to complete a PROM (139). Acceptability is not considered a measurement property in the COSMIN manual, but is referred to in the COSMIN manual on assessing the content validity of PROMS in the context of using cognitive interviews to establish relevance and comprehensibility of a measure (149). Acceptability of C-POS within the target population, and to those administering it will be considered throughout this thesis as it will have an impact on future implementation of the measure.

2.8 Study oversight

2.8.1 C-POS study core research team

The C-POS study research team consists of several researchers from the institute - the principal investigator/grant holder (PhD supervisor RH), a research associate, two research assistants, a research project co-ordination assistant, and my two other PhD supervisors (KB and CES). My PhD supervisors joined the core research team in order to ensure consistency in guidance and decision making. Each member of the team had defined roles within the wider C-POS study. The core research team met monthly throughout the work presented in this thesis.

2.8.2 Patient and Public Involvement

Patient and public involvement (PPI) in research is defined as research being carried out ‘with’ or ‘by’ members of the public, rather than ‘to’ or ‘about’ or ‘for’ them. It is an active partnership that influences and shapes the research (172). When using the term ‘public’ the National Institute for Health and care Research (NIHR) includes patients, potential patients, carers, and people who use health and social care services, as well as people from specific communities and from organisations that represent people who use services (173). Public involvement is considered a prerequisite for high-quality research, with the potential to improve its relevance, impact and quality (174).

As discussed in section 1.3.3, there is a growing focus on children’s rights, promoted by the United Nations Convention of the Rights of the Child, which outlines a child’s rights to be involved in decisions that affects their lives and have their views listened to (78, 175). This is resulting in a move towards engaging children in the research process, rather than relying on parents and professionals to

represent children (175). This involvement of children aligns with the child-centred approach of this thesis and acknowledges that children and young people may have views that differ to those of their parents and healthcare professionals and allows them to have a say in the things that matter to them (36, 37). As well as recruiting children to phases 1 and 3 of the study, there was also engagement with an NIHR funded Young Person's Advisory Group (YPAG) run by Great Ormond Street Children's Hospital at several points during the study.

A recent report on involving children in medical research has suggested that the best way to overcome many of the concerns regarding potential vulnerability is to ensure that researchers work in partnership with children and young people, and parents throughout the whole research process (176). To address this need, the C-POS steering group has three bereaved parents in its membership. Due to their direct experience of caring for a child with a life-limiting or life-threatening condition, their input into the feasibility and acceptability of study procedures and patient information sheets has been invaluable throughout the study. One parent also attended the research ethics committee meeting for the cognitive interview study. The steering group also had a representative from Together for Short Lives, a leading UK charity for children and young people affected by life-limiting and life-threatening conditions and their families.

2.8.3 Great Ormond Street Hospital Young Person's Advisory Group

In a further effort to mitigate concerns regarding participant vulnerability and ensure children had a say in how the research was designed and conducted, the YPAG from Great Ormond Street Hospital (GOSH) were consulted at three timepoints. The YPAG is part of a network of groups called Generation R which are funded by the NIHR and/or NHS. Their aim is to support design and delivery of paediatric health research throughout the UK. Work with the group involved email exchanges and meetings, which due to the COVID-19 pandemic were held on Zoom. Members of the group are between 10-21 years old and are receiving treatment at GOSH, are siblings of children and young people being treated at GOSH or are interested in a career in medicine or research. Some of the members who are patients have direct experience of participating in research studies. The group were consulted three times during the work presented in this thesis:

1. July 2020 – virtual meeting with breakout rooms. The group were given an initial overview of the team and the C-POS study. They were then asked to

think about different recall periods, administration modes and response formats that would be appropriate for C-POS. Breakout rooms were used to gain feedback and ideas on this, supervised by a YPAG facilitator and member of the C-POS team. Twenty-five children and young people aged 10-21 years attended this meeting (19 female; 6 male).

2. October 2020 – the group were emailed copies of the child participant information sheets for the cognitive interview study and asked to feedback on content and design. Ten members of the group responded to this.
3. March 2021 – virtual meeting with breakout rooms. The group were asked to consider how to name the different versions of C-POS (avoiding using chronological age which may stigmatise those with different developmental abilities) and to choose their top 10 outcomes from the list used in the C-POS Delphi survey. There were two breakout rooms, and each group was supported by a YPAG facilitator and a member of the C-POS research team. Twenty-two children and young people aged 10-21 years attended this meeting (17 female: 6 male).

Using an existing YPAG had several benefits (177). The YPAG members had already received training on health research and had previous experience of research involvement work. Parental consent to participate was already in place and the group already had an identity. Members had a rapport with each other as they had participated in previous meeting together. The group facilitators knew the members and their backgrounds well, which was helpful when the discussion involved sensitive topics.

The work conducted with the YPAG as part of C-POS was highlighted in the Great Ormond Street Hospital PPI Impact Report 2020/21 (see Appendix F

2.8.4 Study Steering Group

The C-POS study has a steering group consisting of clinicians working in children and young people's palliative care, academics with experience of outcome measure development and paediatric palliative care research, experts in the ethics of research with children and young people, bereaved parents and a representative from Together for Short Lives.

The study steering group have been involved in the development of the C-POS since the design and grant application phase. They meet with the research team at

regular intervals and have been an integral part of all decision making, interpretation and dissemination of results for the duration of the study.

2.9 Considerations related to study population.

The United Nations Convention on the Rights of the Child defines a child as anyone under 18 years old unless "under the law applicable to the child, majority is attained earlier" (78). The term ‘child’ is generally used to refer to younger children who do not have the maturity and understanding to make important decisions for themselves. The term ‘young person’ refers to older or more experienced children who are more likely to be able to make these decisions for themselves (178). The UK has ratified the United Nations Convention on the Rights of the Child and therefore this definition of a child has been used throughout this thesis to define study inclusion criteria and intended age range for use of C-POS. There are already palliative care PCOMs that have been validated in those 18 years old and over, such as the Integrated Palliative Outcome Scale (98). The term children and young people (children and young people) is used throughout this thesis to reflect the range of ages and developmental stages that C-POS is intended for.

In this thesis a parent/carer is defined as either someone with parental responsibility; or someone who cares for or looks after a child or young person with life limiting or life-threatening condition, such as other family members or adults who live within the same house (178). Only those with ‘parental responsibility’ were permitted to sign research participation consent forms for children (178). Parental responsibility is defined as someone with the rights and responsibilities that parents have in law for their child up to the age of 18 years in England, Wales and Northern Ireland, and up to 16 years in Scotland (178).

Due to the nature of many life-limiting and life-threatening conditions in children and young people, developmental age is not always congruent with chronological age. Therefore, parents/carers were encouraged to choose the most appropriate participant information sheets for their child, rather than the age specific version in phases 1 and 3 of this thesis.

2.10 Ethical considerations and approvals

2.10.1 Ethical considerations

Ethical considerations are crucial when conducting any health-related research but are even more pertinent in end-of life care studies, especially in those involving children and young people and their parents as participants. There is a perception that this is a vulnerable population, and that participation will place undue burden on participants (179, 180). Other concerns include the potential risk of coercion from the study team, the potential that children and young people are unaware of their prognosis and that parent/carers have not come to terms with the situation (179). Studies in adults show an overestimation of burden and underestimation of the benefit of participation in end-of-life research (181). There is limited evidence on the benefits and burdens of participation in paediatric palliative care research to children and young people, families, clinicians and researchers (182). Most studies of benefits and burdens of such research have been conducted with bereaved parents (183-187).

There is a paucity of studies looking at the benefits of end of life care research participation that include the voice of the child or young person (182). Benefits from the small number of studies reporting the child or young person perspective include the opportunity for their voice to be heard, leaving a legacy and facilitation of better communication with family and health and social care professionals (188-191). This research was confined to adolescents and young adults over the age of 14 years. No studies have explored the perceived benefits from the perspective of younger children and young people. The potential burdens of children and young people participating in palliative care research have only been reported from the perspectives of parents/carers who reported no adverse events to participation (192).

The main benefits of participation from the perspective of parent/carers have been cited as altruism, reflection and reconstruction of memories/creating meaning, sense of inclusion, opportunity to tell their child's story and the therapeutic experience of sharing (191, 193-198). Lack of perceived burden is prominent in the literature but when depicted was described as including emotional intensity, fatigue, inconvenience of timing of data collection (such as not wanting to be away from a dying child and not being ready to face reality and discuss what is happening) (182, 195). When distress was experienced by parents taking part in research on their

child's end of life care, most reported also gaining some benefit (194). Parents who reported being more spiritual were more likely to experience benefit, whereas those who expressed decisional regret about their child's end of life care were more likely to report distress (194). Recent research suggests that parents of seriously unwell children can exercise their right to decline participation in research studies, with similar rates of refusal found in end of life versus non-end of life care studies (179, 196). Individuals will balance the potential harms and advantages associated with research participation according to their own personal values and beliefs (191). If parents do participate they enter the research with the expectation that some parts will be hard (197). Studies reporting on the experience of bereaved siblings participating in research found a small number reporting that participation was emotional, but no long-term effects were experienced (192, 199).

Several steps were taken during this PhD to mitigate some of the risks of paediatric palliative care research on both participants and researchers. Participants to both interview studies (phase 1 and phase 3) were initially approached by a known member of their clinical team. This allowed for someone whom they knew and trusted to provide information on the study and answer any initial questions. It was made clear that declining participation would not affect any ongoing care. There is some evidence that some families who indicate an interest to a clinical team member in taking part in paediatric palliative care research do not respond to contact attempts from the research team. This suggests that they are not always able to decline when approached by a clinician (184). By adopting a dual approach during this research, participants had opportunity to decline participation on more than one occasion, thus reducing any perceived coercion to participate.

Any participant that became distressed during an interview was offered referral to their clinical team for further support (with their consent) and a distress protocol was followed for both the qualitative interview study and cognitive interview study (Appendix G). Details of the Together for Short Lives family helpline was also provided on participant information sheets for the interview studies and on the study information page for the Delphi survey. During research interviews, there was an awareness that distress does not always signal an end to an interview, however a break was always offered if distress was apparent and participants were given the option to either cease the interview or carry on if they felt able to (192).

Studies investigating the benefits and burdens of paediatric palliative care research from the perspective of researchers show reports of emotional impact due to being immersed in emotionally laden content (such as interviews and data analysis) (200). In some studies participants have used research interviews as opportunity to gain advice/support from health and social care professional researchers (200). During all phases of this study, the research team received supervision from a qualified member of the clinical team in the university department.

2.10.2 Capacity, consent, and assent

All adult participants consented to participation. For phase 1 written informed consent was obtained up until the start of the COVID-19 pandemic and participants were given a copy of their consent form for their records. During the latter part of phase 1 and for phase 3 of this study, due to the COVID-19 pandemic, consent was taken during a virtual interview and the process was audio-recorded (after approval of an ethics amendment to allow this). The researcher electronically signed and dated consent forms, and these were emailed or posted back to participants (depending on preference).

All children and young people aged 5-15 years required consent from their parent/carer to take part in the research. They were also given the opportunity to complete an assent form, as recommended in the Declaration of Helsinki (201). The process for obtaining assent from children and young people was based on an engagement point of view, with the aim that the child or young person is involved in the decision-making process (202, 203). This process is underpinned by the principles of respect for the child and their developing autonomy, support for the development of the child or young person and support for communication between the researcher and participant (202-204). These principles demonstrate parallels with the tenets of child-centred care discussed in section 1.3.3. Children and young people were empowered to take part in decision making to the extent of their capacity by being given developmentally appropriate written information on the study. This was strengthened by the researcher giving verbal information as part of the assent process and allowing the children and young people an opportunity to ask any questions. This verbal exchange contributed to building a rapport between the researcher and participant. Allowing children and young people to take part in the decision-making process in this way has been said to promote autonomy, teach altruism and improve self-confidence (202, 203, 205). The goal was to engage the

child in the decision-making process by adjusting the information provided to a developmentally appropriate level. The process was personalised, with the child taking part in the process as much as able. If a parent consented to their child participating in the study but the child dissented, then the interview did not go ahead, thus taking a child-centred approach by recognising that children's views are not always congruent with their parents. All children and young people were given the opportunity to complete an assent form, either using pen and paper, or virtually during the COVID-19 pandemic, and were given a copy of this to keep.

In line with the Mental Capacity Act 2005 (MCA) all young people aged 16-17 years were assumed to have capacity unless this was otherwise established (206). The MCA states that an individual is unable to make a decision if they cannot:

- understand the information relevant to the decision.
- retain the information.
- use or weigh the information.
- communicate their decision (by any means).

In this case their identified parent/carer was consulted by the clinician and researcher to give advice on whether they thought participation was something the young person would like to do and complete a consent form on their behalf. All young people were also given the opportunity to complete an assent form as well.

2.10.3 Ethical approval

The Phase 1 semi-structured interview study received ethical approval from London – Bloomsbury Ethics committee (REC reference number 19/LO/0033). NHS REC approval was required as participants were being recruited via the NHS. I was not responsible for writing this ethics application but did attend the committee meeting and was responsible for subsequent amendments made to the application. After original ethical approval 11 amendments were made to the application. These included the addition of new sites to aid recruitment, minor changes to study procedures and participant information sheets, and amendments to allow virtual interviews and transcribing and analysis off site when the COVID-19 pandemic started. A full summary of amendments made to the original protocol can be seen in Appendix H

The Delphi survey (phase 2) was approved by King's College London Research Ethics Committee. This study did not require NHS ethical approval as participants

were not being recruited via NHS sites. One amendment was made to the original ethics application adding new recruitment sites. The full application, approvals and amendment can be seen in Appendix I .

The cognitive interview study (phase 3) received ethical approval from London – Bloomsbury Ethics Committee (REC reference number 21/LO/0282). Three amendments were granted for the addition of new sites and extension to the recruitment period. Full details of ethical approval can be found in Appendix L .

2.11 Summary

This chapter argues that an exploratory sequential mixed methods approach using a critical realist epistemology is the best way to achieve the aims and objectives of this thesis. Methodological issues are presented and discussed in relation to how study design was informed. Consideration has also been given to the ethical challenges of recruiting children and young people with life-limiting and life-threatening conditions and their families to research. Recruitment of participants from this population is essential to ensure robust face and content validity of C-POS. The following chapter contains more detailed methods for the different phases of this PhD.

Chapter 3 Specific methods

3.1 Introduction

This chapter describes the specific methods used to collect the data presented in this thesis, based on the methodological approach presented in Chapter 2. Considerations relating to study design, setting, population, sample, data collection and data analysis are presented for each phase of the study. The specific methods used are presented according to Rothrock's PROM development process as depicted in Figure 2-1.

3.2 Phase 1 – gathering input.

Objectives incorporated in this phase.

i. To determine optimal recall period, response format and administration mode for patient centred outcomes in children and young people
ii. To establish child and parent priorities for outcomes of care
iii. To establish healthcare professional and commissioner priorities for outcomes of care
iv. To develop a list of candidate priority outcomes to be included in C-POS

The aim of phase 1 of this study was to achieve objectives i-iv of this thesis. This was achieved by gathering input from key stakeholders to establish their priorities for care outcomes in order to inform the content validity of C-POS. Preferences regarding the recall period, response format and administration mode of C-POS, with the aim of enhancing acceptability and feasibility were also investigated. The specific methods used in this phase are discussed below.

3.2.1 Systematic review of optimal recall period, response format and administration mode for child self-reported outcomes (objective i)

The systematic review presented in this thesis aimed to appraise the current evidence on recall period, response format and administration mode required to enable children and young people to participate in valid and reliable self-report of their own health outcomes. The systematic review was conducted and reported in

accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (207).

Search strategy

PsycINFO, Medline, CINAHL and Embase, and cited by on Scopus of selected articles were searched from 1st January 1980 (when outcome measurement in children and young people began to be reported in the scientific literature (208-210)) to 15th March 2020. In addition, cited references of selected articles were searched. The full protocol containing search terms is in Appendix D

Inclusion and exclusion criteria

Inclusion criteria:

- Studies in children and young people up to and including 18 years old. If studies involved participants less than18 years old, then these were included if data for those 18 years and under was presented separately.
- Primary research of self-reported health outcomes
- Case reports ≥3 participants
- Studies looking at recall period, response scale selection, administration modality and approaches to enable children to self-report in terms of their effect on:
    - measurement properties and factor structure of instruments (validity, reliability, and responsiveness)
    - acceptability and feasibility of use
    - preference for a particular mode, recall period or response format
- Studies written in the English language

Exclusion criteria:

- Review/systematic review articles
- Discussion articles
- Editorials, reports, letters
- Small case reports ≤3 participants
- Studies solely in those over 18 years old
- Written in a language other than English

Study selection

All retrieved articles were transferred to EndNote version 9 and duplicates removed. The titles and abstracts of all retrieved articles were screened for eligibility by one reviewer. If there was not enough information within the title and abstract to determine eligibility, then the full text article was screened. Remaining full text articles were screened, with 10% screened by a second reviewer. Any discrepancies throughout the process were discussed with a third reviewer.

Data extraction

Data from eligible studies was extracted into an Excel sheet containing: study authors, publication year, geographic location, objective, study design, sample characteristics (population, size, setting), measure characteristics reported (recall period, response format, administration mode) and main findings.

Data analysis

I originally intended to use the COSMIN checklist (148) to assess study quality. As the overall aim of the review was not to appraise specific PROMs and their measurement properties however, a decision was made to use QualSyst instead (211).

QualSyst assesses the quality of studies with two scoring systems, one for qualitative research and one for quantitative research. The qualitative scale consists of ten items with scores from zero to two, yielding a maximum score of 20. The quantitative scale consists of 14 items with scores from zero to two, an option to score an item ‘not applicable’, and maximum total score of 28. Overall scores are reported as percentages. Mixed method studies receive two scores—one each for the qualitative and quantitative components (211). Inter-rater agreement was assessed for 10% of the included articles.

The findings from the systematic review were synthesised narratively to appraise the heterogeneity of included studies, any similarities or differences in findings, and to explore patterns. Recommendations were made based on the quality of the evidence of included studies and the feasibility and acceptability of using different response scale types, recall periods and modes of administration when developing patient-reported outcome measures for use by children and young people.

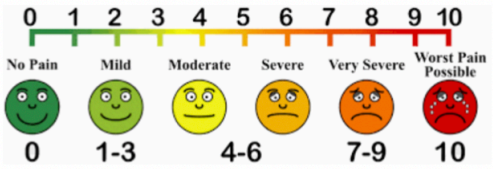
The results of this systematic review are presented in Chapter 4.

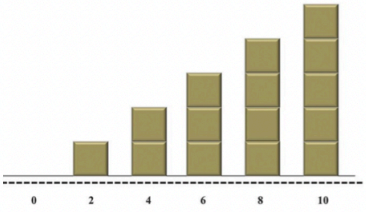
3.2.2 Young Person's Advisory Group (objective i)

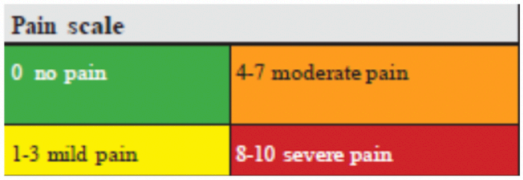
A virtual meeting was held with the young person's advisory group in July 2020, with the aim of obtaining input on appropriate administration mode, recall period and response format for C-POS. The session was presented by me, and another member of the C-POS team attended to facilitate one of the two breakout rooms. The session started with a short presentation on who we were, what the term serious illness meant, an overview of the C-POS study and why it is important. This was presented in simple, easy to understand language, with visually appealing slides. The group were then split into two breakout rooms and asked to consider the following questions, and given examples to guide discussion:

ii. How we give children the questionnaire (mode) – computer, paper, iPad.
iii. How children should answer the questions (response format) – see Figure 3-1 for examples shown to the group
iv. How far back do you think children will be able to remember (recall period) – a few days, a week, two weeks, one month, longer.

Responses were collated via the whiteboard function on Zoom and saved so they could be referred to later.

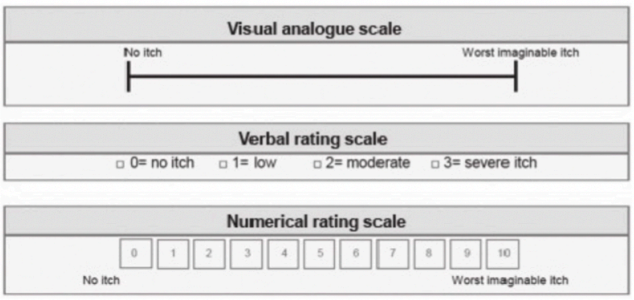






ABOUT MY HEALTH AND ACTIVITIES *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than one block	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activity or exercise	0	1	2	3	4
4. It is hard for me to lift something heavy	0	1	2	3	4
5. It is hard for me to take a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I hurt or ache	0	1	2	3	4
8. I have low energy	0	1	2	3	4

	Not at all	Sometimes	A lot
Is it hard for you to snap your fingers	☺	😐	☹



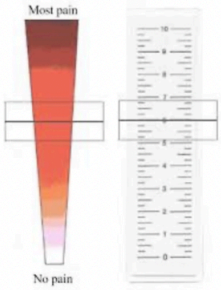


Figure 3-1 Response format examples shown to YPAG (112, 212-215)

3.2.3 Qualitative semi-structured interview study to establish symptoms, concerns, priority outcomes of care and measurement preferences (objectives ii - iv)

Study design, setting, population and sample.

This phase consisted of a qualitative, cross-sectional design using semi-structured interviews. Participants were recruited from six UK hospitals with tertiary paediatric services (King's College Hospital NHS Foundation Trust, Great Ormond Street NHS Foundation Trust, Evelina Children's Hospital, Guy's & St Thomas' NHS Foundation Trust, the Royal Marsden NHS Foundation Trust, Cambridge University Hospitals NHS Foundation Trust and Leeds Teaching Hospitals NHS Trust) and three UK children's hospices (East Anglia Children's hospice, Martin House Children's Hospice and Northern Ireland Children's Hospice).

Inclusion criteria:

- Children and young people: from 5 ≥17 years old living with a life-limiting or life-threatening condition (21).
- Parent/carers: parent or carer responsible for the primary care needs of a child of any age who is living with a life-limiting or life-threatening condition.
- Siblings: Sibling aged 5≥17 years old (by blood or relationship) of a child of any age living with a life-limiting or life-threatening condition.
- Healthcare professionals: Medicine, nursing, social work, psychology or allied health professional who has been providing paediatric palliative care for at least six months – employed at any of the recruiting sites listed above.
- Commissioners: any NHS commissioner responsible for commissioning children's palliative care services.

Exclusion criteria:

- Children and young people: unable to communicate any views or wishes via their parent/caregiver, an in-depth interview, or via "draw & talk" or play methods; speaks a language not supported by the NHS Trust translation service; currently enrolled in another study. Deemed clinically unable to give consent/assent.
- Parents/carers: Deemed clinically unable to give consent/assent.
- Siblings: Deemed clinically unable to give consent/assent.

- Participants were purposively sampled. Purposive sampling is a non-random method which aims to sample a group of people with specific characteristics (216). In this study it was used to ensure maximum variation in the key characteristics of diagnosis, age of child, geographical location, and profession (for healthcare professionals).

Recruitment, consent and data collection

Eligible parent, child and sibling participants were identified by the clinical teams at participating sites. Clinical teams were asked to use the Together for Short Lives definitions of life-limiting and life-threatening conditions described in section 1.2.2 when considering eligibility (217). For category 1 conditions (those for which curative treatment may be feasible but can fail) they were asked to only consider oncology patients with a poor prognosis who were unlikely to survive for the next 12 months. Children and young people on the organ transplant register were also considered for inclusion. NHS commissioners were recruited via recommendations from participants at the above sites, or via Together for Short Lives, the UK's leading children's palliative care advocacy charity.

NHS commissioners and health and social care professionals were contacted via telephone after their contact details had been shared with the research team, to discuss study participation. The research team then sent those that were interested a study information sheet.

A member of the clinical team approached potential child and family participants to discuss the aims of the study and what participation would involve. If the child and/or family expressed an interest in taking part, they were given an age/developmental stage appropriate written information sheet. Information sheets were developed according to the following age groups: 5-7 years, 8-10 years, 11-15 years, and 16-18 years. However, due to the heterogeneity of life-limiting and life-threatening conditions, chronological age does not always equate with developmental age, so parents/carers were asked to select the most appropriate version for their child. Parents and siblings were also given appropriate participant information sheets regarding the study.

After receiving the information sheet, all potential participants were given up to 14 days to decide whether they would like to participate. At this point they were contacted by the clinical or research team member who gave them the participant

information sheet and with their verbal consent, details (name, age, diagnosis, contact details) were passed on to the research team. The research team then contacted participants via their preferred contact method to answer any questions they had regarding taking part in the study and to arrange a time and date for the interview to take place. Prior to the COVID-19 pandemic interviews were conducted in a location of the participants choice, after this an ethics amendment was submitted to allow interviews to take place via MS Teams (Appendix H). Consent was taken as outlined in section 2.10.2.

Data was collected by conducting semi-structured interviews. Semi-structured interview schedules contain fixed questions which are be asked during the interview but allow for the participant to discuss subjects in the order most appropriate for them, and the interviewer to probe and ask questions around that. They also enable respondents to raise other relevant issues that are not covered by the schedule (216). Interviews were conducted by three members of the C-POS team. I conducted 34% of the interviews. The other two members of the team were a research assistant with an MSc in Psychology who was new to qualitative research, and a research associate with a PhD that included qualitative interview methodology. Neither are clinically trained. All interviewers received training on ethical issues in conducting research with children, communication skills for children who have additional needs and research with children who have palliative care needs. Training was delivered by experts in the respective fields. The topic guide contained an open question asking participants to describe their/their child's condition and how it affected their/their child's life. Interviews with professionals asked about the main symptoms, concerns, and care priorities of children and young people with life-limiting and life-threatening conditions. Probes ensured that all domains from the WHO definition of palliative care were discussed, along with symptoms and concerns known to be important to children and young people with life-limiting and life-threatening conditions, while allowing participants to discuss what mattered most (13, 218). Each interview began with taking consent, followed by demographic questions. Children and young people were asked about their hobbies to build rapport before the main interview began. They were also given the option of drawing on plain paper or paper cut-outs of people to enable them to talk about their views and experiences (219-221). Interviews were audio-recorded, transcribed verbatim, and pseudonymised.

Data analysis

Data analysis for phase 1 followed the five steps of Framework analysis: familiarisation, constructing a thematic framework, indexing and sorting, charting and mapping/interpretation using NVivo software (Version 12) (146, 162, 163). The framework is based on an analytical hierarchy which can be depicted as a ladder with five distinct phases of analysis (Figure 3-2) (146). The arrow above the ladder indicates that the analysis process is continuous, flexible, and iterative and the researcher moves backwards and forwards between the different stages to identify emerging themes.

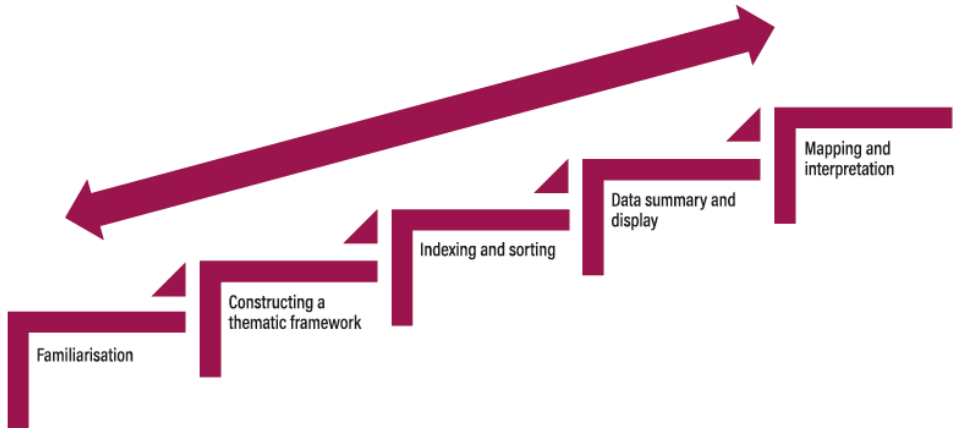


Figure 3-2 Framework analysis ladder (152)

The five steps of Framework analysis are described in more detail below.

Familiarisation

This stage involved listening to audio-recordings and reading initial transcripts and becoming immersed in the data (172). This is an important step and allowed for familiarisation with data collected by other members of the team. Three transcripts from each cohort (child, parent, sibling, health care professional and commissioner) were read by two members of the team (myself and one other), along with interview field notes and demographic data in order to obtain a broad overview of content. Notes were made in the margins of each transcript about topics/ideas that were deemed to be significant. The team had regular 'familiarisation' meetings led by me, to work through the initial transcripts together, discussing their emerging ideas and making a list of preliminary codes. This list was added to and amended at the end

of each meeting. By the end of this process the researchers felt immersed in the participants' experiences and a final list of preliminary codes were made.

Identifying/constructing an initial thematic framework

The aim of this stage was to identify all of the key issues, concepts and themes by which the data could be examined and referenced. This was achieved by drawing on *a priori* issues and questions derived from the aims and objectives of the study, as well as by issues raised by the respondents themselves, and views or experiences that recur in the data (222). The list of preliminary codes constructed during the familiarisation phase was reviewed by the research team, along with the aim of the study and the results of a previously published systematic review on symptoms and concerns in children with life-limiting/life-threatening conditions (12). Themes and subthemes were arranged in a word document (a face-to-face meeting using Post-Its or a flipchart was not possible due to the COVID-19 pandemic) in order of similarity of emerging ideas (themes and subthemes). The framework was then piloted on a further five transcripts before being uploaded into NVivo (version 12). A final thematic framework to apply to subsequent transcripts was agreed, with an 'other' category in case any subsequent data did not fit into the framework categories was found. This thematic framework was reviewed and refined as new data was collected throughout the analysis process (173).

Indexing and sorting

The thematic framework was applied to all subsequent transcripts using NVivo (version 12), thereby categorising them into the framework categories. The use of NVivo aided transparency as it leaves a clear audit trail and decisions can be tracked back to the raw data (171).

Charting

The aim of this stage was to organise the data into a more manageable format in order to facilitate mapping and interpretation. The matrix function in NVivo was used to generate matrices in which the data for each identified theme were 'charted' by case and code. Each framework category was worked through, summarising all the data indexed to that category, for each participant. Using this approach, data was also summarised by participant type e.g., child, parent, sibling.

Mapping and interpretation

This involved analysis of the key characteristics of the data and rereading of some transcripts to clarify information. Relevant and appropriate quotes were selected to highlight themes (170). This stage involved moving beyond data management towards understanding it within the context of the research question (146). Findings were brought to the C-POS project meetings and steering group for wider discussion. Themes and subthemes were developed by analysing data categories between and within cases.

The Framework approach has many similarities to thematic analysis, especially in the initial stages (167), however there is greater emphasis on making the process of data analysis transparent and linking the stages of analysis (146, 168, 169). The analytical process is systematic in nature and allows the researcher to move back and forth across the data until a coherent account emerges (146, 170). This allows for themes to be constantly refined and may lead to the development of a conceptual framework (167). It also enables comparison within and between groups. This approach is useful for studies with a large number of participants, such as this study (146). Framework analysis emphasises how both *a priori* issues and emergent data should guide the development of the thematic framework, allowing for both deductive and inductive coding. This is important in this study as there is some *a priori* knowledge of symptoms and concerns in this population, but this knowledge is largely derived from proxy-reports, and children and young people with a cancer diagnosis (13, 223).

The results of this study are presented in **Error! Reference source not found.** and Chapter 6. Using the results of this qualitative interview study, findings were used to generate a comprehensive list of outcomes that could potentially be included in C-POS.

3.3 Phase 2 – item generation (objective v)

Objectives incorporated in this phase.

v. To gain stakeholder consensus on items to be included and construct first versions of C-POS

The aim of phase 2 was to achieve objective v) of the study and gain stakeholder consensus on the items to be included in C-POS to enhance further content validity of the measure within the intended population of use, and to finalise initial versions of C-POS ready for cognitive testing. This was achieved by:

1. Conducting a Delphi survey with health and social care professionals, and parents/carers of children and young people with life-limiting and life-threatening conditions on priority outcomes derived from Phase 1. The full protocol, participant information and demographic data collection details can be found in Appendix K .
2. An online workshop with the young person's advisory group to establish children and young people priority outcomes from those identified in Phase 1.
3. An item generation meeting with the study steering group to finalise the C-POS items, recall period and response format to be used. This meeting was included in the protocol for the Delphi survey which can be found in Appendix K .

3.3.1 Study design, setting, population and sample.

Delphi survey

Design and setting

This phase of the study was a repeated online Delphi survey. A Delphi survey is an established research method used to gain reliable consensus within a group of experts when it is hard to meet face to face or there are time constraints (104, 224, 225). Delphi surveys have previously been used to gather evidence on the construct validity of measurement tools used in palliative care (225).

There are four commonly used types of Delphi methodology:

1. Classical – focuses on facts to obtain consensus.
2. Decision – focuses on preparation and decision for future directions.
3. Policy – focuses on ideas to define and differentiate views.
4. Ranking-type – focuses on identification and ranking of key factors, items or issues (224).

Either a ranking-type or classical Delphi survey methodology would have been equally appropriate for this study. In a classical Delphi survey participants are asked to score each item (in this case the outcomes identified in phase 1) from low to high priority using a Likert scale (103). In this study, there was concern that all identified outcomes could potentially be scored as high priority for inclusion, leaving too many items for C-POS. Therefore, a ranking-type Delphi methodology was chosen, using methodology similar to that proposed by Schmidt et al (226). Participants were asked to rank the outcomes in order of priority. This ensured participants made a clear decision about the relative importance of each outcome (226, 227).

A standard ranking-type Delphi survey has three phases (226, 227);

1. Brainstorming phase – experts list items that are important for the area of interest.
2. Narrowing down phase – narrowing down of the list of items developed during the brainstorming phase to a number that is manageable and reasonable for the ranking phase.
3. Ranking – the aim of this phase is to reach consensus in the ranking of selected items.

The semi-structured interviews conducted in phase 1 replaced the brainstorming phase, and the outcomes identified from these were used in the narrowing down phase of the study, along with additional outcomes identified in a systematic review of symptoms and concerns in children and young people with life-limiting and life-threatening conditions (13).

A combination of convenience and purposive sampling was used to recruit participants to the Delphi survey. Key stakeholders were purposively targeted, with convenience and snowball sampling then used within these populations. Parent/carers of children and young people with life-limiting and life-threatening conditions and health and social care professionals caring for children and young people with life-limiting and life-threatening conditions were eligible to participate. Both of these groups are considered experts in the field.

Parent/carer participants were identified through:

- Together for Short Lives (a leading UK charity for children with life-threatening & life-limiting conditions):

    - Database of 140 parent experts who want to be involved in research – the charity emailed members a link to the survey.
    - Link to survey on quarterly newsletter
- Parent group at the Royal Marsden NHS Foundation Trust – group were emailed information regarding the study along with a link to the survey web page.
- Martin House Research Centre family advisory board – via email link.
- Martin House and Northern Ireland children's hospices – email link via family Facebook page.
- Social media – a link to the survey was shared on the C-POS Twitter feed.

Health care professional participants were identified through:

- Association for Paediatric Palliative Medicine (APPM) – medical and nursing membership were emailed a link to the survey via the APPM.
- Together for Short Lives:
    - All children's hospices, hospital and community children's palliative care teams are members - the charity emailed members a link to the survey.
    - Other health care professional members - the charity emailed members a link to the survey.
- The survey link was sent to the Principal Investigators of the sites used for phase 1 of the study with a request that they disseminate to their teams and contacts.
- Association of Palliative Care Social Workers – via email link.

Social media – a link to the survey was shared on departmental and C-POS study Twitter feeds.

Upon completion of the narrowing down phase of the Delphi survey, participants were asked to recommend other experts to take part in the survey by sending them a link to the survey or sharing via social media (snowball sampling).

<u>Inclusion criteria</u>

- Any health or social care professional who had been providing care to children and young people with life-limiting and life-threatening conditions for more than 6 months.

- Parents/carers of a children and young people with a life-limiting and life-threatening condition aged 0-17 years inclusive
- Bereaved parents of children and young people with life-limiting and life-threatening condition whose child had died within 12-24 months of participation. This time period was identified as being optimal in a study with bereaved parents who felt they would still remember what had happened, how they felt and what they needed clearly. They felt that at 12 months enough time had passed so recall was not significantly painful (185).

Exclusion criteria

- Health care professionals who had been working with children and young people with life-limiting and life-threatening conditions for less than 6 months.
- Individuals who cannot complete an online survey written in English.

Sample size

The COSMIN manual for assessing content validity of PROMS recommends a sample size of >100 for a quantitative study on content validity (228). 50 to 99 participants per group is deemed adequate, whereas 100 or greater is considered very good, the highest possible rating (228). Delphi surveys in similar populations have reported varied response rates ranging from 44-80% (103, 229, 230). Therefore, the aim was to recruit >100 participants to the Delphi survey.

Young person's advisory group

A virtual meeting was held with the YPAG in March 2021 (see 2.8.3 for further details on the group). A short age-appropriate presentation on the aims of the C-POS study was given along with some simple definitions of outcome measures and life-limiting conditions, before the group was split into two. Older representatives were asked to work independently to review outcomes from those ranked during rounds two and three of the Delphi and choose their top 10. Younger representatives were asked to choose their top ten outcomes from this list as a group. Both groups were also asked to suggest names for the C-POS versions (as age bands to label measures is not appropriate in this population given common developmental delay). The groups facilitators led the session with support from a member of the research team. The intention was that working with the advisory

group would strengthen and broaden the perspectives of children in the study and ensure children's views continued to be considered in measure design, thus enhancing content validity.

Item generation meeting

The item generation meeting was a half day virtual meeting with the C-POS steering group in November 2020. See 2.8.4 for group details. The agenda was informed by previous PCOM item generation meetings and included presentation of study results so far (231). The item generation meeting was included in the protocol and ethics application for the Delphi survey (see Appendix K).

3.3.2 Data Collection

Delphi survey

The survey was designed using SmartSurvey™. This platform was chosen for its ease of use and ability to export data which are encrypted and stored within the UK. The survey was piloted with three participants (two health and social care professionals and one parent) prior to the start of recruitment. Data from these participants were not included in the results. This ensured the skip logic worked as intended and that the questions and instructions were easily understood.

The first page of the survey was similar in format to a participant information sheet and explained the aim and rationale for the study, what participation would entail, how the outcomes being prioritised were decided upon, how data would be stored and steps taken to ensure confidentiality (232). Participants were asked to complete a consent form at the beginning of each round of the survey (this was built into the survey platform) to indicate consent to participation. In the event that participants found the content of the survey distressing, they were given the contact number for the Together for Short Lives helpline on the study information page. This helpline is run by experienced staff who are used to talking to parents who are distressed.

Each round of the survey was left open for two to three weeks, depending on response rate.

The following demographic data were collected:

- Health care professionals – profession, current role, length of children and young people palliative care experience, place of work, gender and age.
- Parents/carers – age, relationship to child, child's diagnosis, child's age, ethnic background of child and parent/carer, gender of child and parent/carer and area of residence.
-

Narrowing down

Outcomes included in the narrowing down phase were those identified from the qualitative interview study conducted in phase 1, and additional items from a recent systematic review on symptoms and concerns in children and young people with life-limiting and life-threatening conditions (13). Outcomes were presented in random order and participants were asked to select the 20 that they thought were a priority for inclusion in C-POS. They were also asked to suggest outcomes that were missing. A free text box was available for choices to be justified. Items were eligible to be moved into the Delphi ranking rounds if:

- They were selected by >50% of participants (233) or,
- If more than 30 outcomes were selected by >50% of participants then only those selected by >30% of participants were included in subsequent rounds (227).
- If new outcomes were suggested during round 1, they were compared with existing items and discussed by the research team and members of the steering group to gain expert consensus on whether they should be added to round two for evaluation by participants (234, 235).

For this round of the Delphi survey, results were analysed as a whole by participant group (parents/carers and health and social care professionals). Participant email addresses were collected for invitations to subsequent rounds to be sent.

Ranking rounds (round two and beyond)

It was anticipated that two to three ranking rounds would be needed in order to obtain consensus on the final outcomes to be included in C-POS. Participants in the ranking rounds needed to have participated in previous rounds. For round two (first ranking round) participants were presented with the results from the narrowing down round outlining items that were removed, any relevant comments from participants and any new items that had been added. Participants were then asked to rank the

remaining outcomes in order of priority for inclusion in C-POS. Items were ranked in descending order, from the most to least important. Participants were asked to explain their justifications for their rankings in a free text box. Weekly reminders were sent to participants via email.

For the second ranking round onwards, participants were again sent an email with a link to the survey. They were given the following feedback from the previous round:

- the median rank of each item and where they ranked each item,
- Kendall's W coefficient of concordance (in layman's terms i.e., weak, moderate, or strong agreement),
- top half rank (the percentage of experts who ranked items in their top 50%)
- relative comments/justifications made by respondents.

Results were presented for the participants in full and stratified by group (parent/carer and professional) (234). Participants were again asked to rank outcomes based on the feedback above. From the second ranking round onwards items to be ranked were presented according to median rank rather than in random order to aide achievement of consensus (226). Participants were again asked to justify their ranking decision in a free text box. Finally, participants were asked whether they would be willing to participate in a further round if consensus has not been reached. Weekly reminders were sent to all participants.

Data was analysed in the same way as the first ranking round. If consensus has been reached (Kendall's W >0.7) then the study would stop (233). If consensus has not been reached and >50% of respondents indicated they would be willing to participate in another round a further round would be conducted. If <50% of respondents were willing to take part in a further round, the study will stop. In palliative care, perfect agreement may often not be realistic due to different values, world views and ethical dilemmas concerning medical decision making (236). There are a diverse range of life-limiting and life-threatening conditions that affect children and young people, and they are cared for in a wide range of settings which adds to this complexity. Lack of agreement could potentially have implications for the acceptability of C-POS; however it was anticipated that items that reflect the key priorities of all key stakeholders would be able to be incorporated in to the measure.

Young person's advisory group

After listening to the presentation, the group were split into two. The group containing older participants were asked to choose their top 10 outcomes from those used in the Delphi ranking rounds. Younger participants were asked to work as a group to choose their top 10 outcomes from the same list.

Item generation meeting

The meeting began with a presentation giving an overview of the study and the results from the qualitative interview study, previous developmental work (13, 237, 238), the Delphi survey, young person's advisory group and findings on measurement perspectives from our qualitative interviews. Discussion was led by the me, starting with the overarching themes found in the qualitative interview study followed by suggestions on potential wording of questions. Discussion was had regarding priority items for inclusion and measure design aspects (recall period, response format and administration mode). After the item generation versions of the C-POS were drafted ready for cognitive testing.

3.3.3 Data analysis

Delphi survey

Data were exported from SmartSurvey™ and analysed using STATA v16. Demographic data was analysed using descriptive statistics.

Narrowing down

All symptoms and concerns that were not selected by >50% of participants from each group (parents/carers and professionals) were eliminated. If more than 30 items were selected by >50% of participants, then the items selected by >30% of participants were included in subsequent rounds.

Ranking rounds

Analysis of the ranking rounds consisted of:

- the median rank of each item,
- Kendall's W (to measure the level of agreement between participants)
- Percentage of participants placing each item in the top half of their list (226, 227)

- Cohen's kappa was used to determine agreement between parent/carer and healthcare professional rankings.

Kendall's W was interpreted according to guidance by Schmidt et al (226) on conducting ranking-type Delphi surveys:

- ≥0.1 – very weak agreement
- ≥0.3 – weak agreement
- ≥0.5 – moderate agreement
- ≥0.7 – strong agreement
- ≥0.9 – unusually strong agreement

Once consensus was reached (K≥0.7) then the ranking rounds would stop. I anticipated that this would occur within two to three ranking rounds.

Free text comments were collated by symptom and concern and analysed thematically.

Results for this study are presented in Chapter 7.

3.4 Phase 3 – item improvement (objective vi).

Objectives included in this phase.
vi. To establish acceptability, comprehensiveness, and comprehension of initial C-POS versions.

This phase of the study aimed to achieve objective vi) which was to establish comprehensiveness, comprehensibility, and feasibility of C-POS within the target population. See Appendix L for the full study protocol and associated documentation. The study was conducted and reported according to the Cognitive Interview Reporting Framework (239).

The COSMIN methodology for evaluating the content validity of a PROM state that a cognitive interview study or other pilot test should be performed to evaluate comprehensibility and comprehensiveness of a PROM (149). If this was not performed the total quality of PROM development will be rated as inadequate. This phase of the study follows the COSMIN standards for evaluating the quality of a cognitive interview study (149).

Cognitive interviewing is a method which draws on cognitive theory to help understand whether or not questions in a measure can be answered, and whether

the response task is being interpreted and carried out in the way intended (145, 240). According to Tourangeau, in order to answer a question, a person must complete four distinct processes:

1. Comprehension of the question
2. Retrieval of the necessary information to answer the question.
3. Judgement about the information needed to answer the question.
4. Respond to the question (143).

It is essential that the C-POS instructions, items, response to the question and recall period are understood as intended. If they are not, the information obtained may be incorrect or respondents may not understand how to complete the C-POS (228).

3.4.1 Study design, setting, population and sample.

This phase of the study had a cross-sectional interview design. Participants were recruited from the following sites, all of whom provide care to children and young people with life-limiting and life-threatening conditions:

- Leeds Teaching Hospital NHS Trust
- Martin House Children's Hospice
- Royal Marsden NHS Foundation Trust
- East Anglia Children's Hospice
- Evelina Children's Hospital NHS Trust
- Royal Hospital for Children, Queen Elizabeth University Hospital, Glasgow
- Bradford Royal Infirmary
- East Lancashire NHS Trust
- Chestnut Tree House Children's Hospice
- Hertfordshire Community NHS Trust
- East Cheshire NHS Trust
- Forget Me Not Children's Hospice
- Leicester Children's Hospital
- Maidstone and Tunbridge Wells NHS Trust

In addition, social media was used to recruit participants via:

- The C-POS study's Twitter feed
- Twitter feeds of members of the study research and steering group.
- Together for Short Lives social media pages
- Facebook and Twitter pages of hospices and NHS services we have been collaborating with.
- Hospices and NHS sites we have working with may also share details of the study in their family newsletters or similar.

Inclusion criteria

- Children and young people: from five years old up to the age of 17 years old living with a life-limiting or life-threatening condition.
- Parents/carers: responsible for the primary care needs of a children and young people from 0-17 years living with a life-limiting or life-threatening condition.

Exclusion criteria

- Children and young people:
    - unable to communicate any views or wishes via their parent/caregiver or an interview.
    - unable to read the C-POS questions or unable to understand the questions if they are read aloud.
    - currently enrolled in another study.
    - deemed clinically unable to give consent/assent (206, 241).
    - who do not wish to participate.
- Parents:

- deemed clinically unable to give consent due to concerns regarding well-being or an underlying mental health condition (206).
- who do not wish to participate.

Sample

COSMIN recommends a sample size of at least seven for cognitive testing of a PROM (149). Therefore, the aim was to recruit a minimum of seven participants per version of C-POS to this phase of the study. If required, any amendments to the first version of the C-POS were planned to be made after four participants had completed interviews. Then it would be retested with another three participants. Any further changes would then need to be retested again as per COSMIN guidance which states the final set of items should have been cognitively tested in the intended population of use (149). Participants were purposively sampled to ensure that all versions of C-POS were tested with children and young people of varying age/developmental ability and with a range of life-limiting and life-threatening conditions and their parents/carers.

3.4.2 Data Collection

Potential participants to the study were either identified by their clinical teams or made contact with the research team via social media. Following an introductory explanation from the clinical team or research team, the children and young people and their parent/carer were provided with age/developmentally appropriate written information on the study (see Appendix L They were given a minimum of 24 hours to consider participation and if they agreed their preferred contact details were shared with the research team.

One-to-one interviews were conducted with participants either via MS Teams or face-to face, depending on participant preference and current COVID-19 guidance. Children and young people were able to have a parent/carer with them during the interview if they wished. Two of the interview team (including myself) attending a two-day training course on cognitive interviewing and disseminated our learning to the rest of the research team prior to interview commencement. Prior to commencement of the interview consent (and assent if appropriate) were taken as outlined in section 2.10.2. If interviews were conducted remotely then the children and young people and/or consenting parent/carer were asked to complete the

consent and/or assent form at the beginning of the interview. The consent form was completed via screensharing, and the conversation was audio recorded. After consent had been taken, a short demographic questionnaire was completed. Participants were then given a demonstration of the 'think aloud' technique and encouraged to take part in a practice task. This task helped to build a rapport between the interviewer and participant.

C-POS was either shared in a paper format if the interview was conducted face-to-face, or via screen sharing on MS Teams if virtual. Participants were asked to read each C-POS question out loud (younger participants could have questions read to them by the researcher or a parent/carer). They were then asked to speak out loud as they answer to elicit insights on their thought processes and decisions regarding responses (242) . Concurrent verbal probing was also used during the process. Probes were both spontaneous to explore responses and non-verbal cues such as hesitations (243), and predefined (see Appendix L for topic guide) to allow for exploration of comprehension, recall period, response options, format and missing items (143, 240). All interviews were audio-recorded and transcribed verbatim.

3.4.3 Data analysis

Audio recordings of the interviews were independently analysed by two members of the C-POS team as per COSMIN recommendations (149). Framework analysis (152) was used to identify any difficulties participants had with questions, based on Tourangeau's four-stage model of survey response – comprehension, retrieval, judgement and response (143) and Willis' coding system for classifying questionnaire problems (244), which looks at clarity of questions, knowledge required to answer questions, problems with assumptions/underlying logic, response categories, sensitivity of questions, instructions and formatting. Data was inputted into an Excel sheet using the Framework approach described by NatCen (see Appendix M for example) to allow comparison between participants and question (145). Interviews were analysed independently by two members of the core study group. I was responsible for analysing all interview data. After four interviews per C-POS version, the research team reviewed the results and discussed any difficulties participants had with items and responses. Possible changes to C-POS were discussed and consensus on whether changes were required was agreed. Demographic data was analysed using descriptive statistics. Results for this study are presented in Chapter 8.

3.5 Summary

This chapter details the specific methods used to collect the data for each phase of this thesis. The following chapters (Chapter 4-Chapter 8) present the results of these phases.

Chapter 4 Results - Systematic review of optimal recall period, response format and administration mode for child self-reported outcomes (objective i)

4.1 Introduction

When developing a PCOM it is important that all elements of the measure design are considered. Selecting appropriate recall period, response format and administration mode are components of good content validity (relevance and comprehensibility) in the COSMIN methodology for assessing content validity of PROMS (149). In addition, they contribute to the acceptability and feasibility of a PROM, reducing missing data and enhancing implementation into routine practice (245).

This chapter presents the results of a systematic review of response scale type, recall period and administration mode in children and young people to enable them to participate in valid and reliable self-report of health outcomes. This is one component of Phase 1 of this study (gathering input and defining concept) and addresses study objective i) which is to determine optimal recall period, response format and administration mode for patient-centred outcomes in children and young people. The results presented are in accepted paper format.

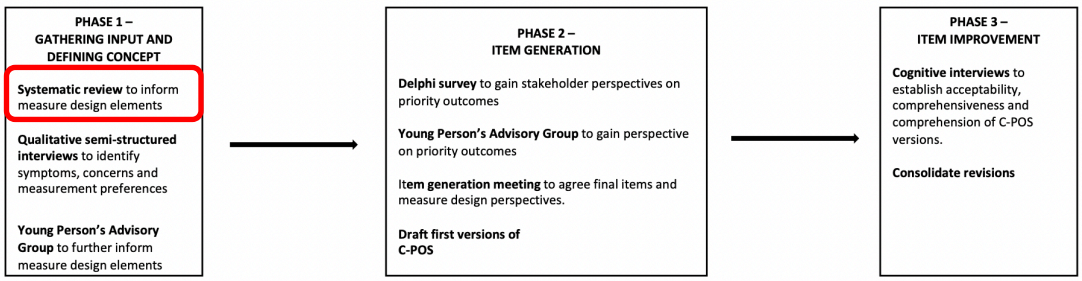


Figure 4-1 Graphic depicting where Chapter 5 fits into the overall study

4.2 Enhancing validity, reliability and participation in self-reported health outcome measurement for children and young people: a systematic review of recall period, response scale format, and administration modality [publication 1].

4.2.1 Statement of contribution [publication 1]

I was responsible for writing the systematic review protocol, conducting searches, study selection, data extraction, quality appraisal, data synthesis and preparing the manuscript for publication. JA screened 10% of the articles at the full text screening stage and 10% of articles at the quality appraisal stage. RH resolved conflicts at the full text screening and quality appraisal stages. RH, KB and CES provided supervision throughout. All authors were involved in critical review throughout the process and approved the final manuscript.

References included in publication 1: (88-91, 105, 118, 123, 147, 148, 207-211, 214, 245-335).

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Enhancing validity, reliability and participation in self-reported health outcome measurement for children and young people: a systematic review of recall period, response scale format, and administration modality

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Abstract

Introduction Self-report is the gold standard for measuring children's health-related outcomes. Design of such measures is complex and challenging. This review aims to systematically appraise the evidence on recall period, response scale format, mode of administration and approaches needed to enable children and young people < 19 years to participate in valid and reliable self-reporting of their health outcomes.

Method PsycInfo, Medline, CINAHL and Embase were searched from 1 January 1990 to 15 March 2020, and citation searching undertaken in Scopus. Articles were included if they were primary research or case reports of ≥ 3 participants reporting the following: recall period, response scale selection, administration modality. Quality was assessed using QualSyst, and results synthesised narratively. This review was conducted and reported according to PRISMA guidelines.

Results 81 of 13,215 retrieved articles met the inclusion criteria. Children < 5 years old cannot validly and reliably self-report health outcomes. Face scales demonstrate better psychometric properties than visual analogue or Likert scales. Computerised and paper scales generally show equivalent construct validity. Children prefer computerised measures. Children ≤ 7 years old think dichotomously so need two response options. Those > 8 years old can reliably use a 3-point scale.

Conclusion The results of this review have both clinical and research implications. They can be used to inform appropriate choice of PROM for use with CYP in the clinical setting. We also give eight recommendations for future development of self-reported outcome measures for children and young people.

Keywords Child · Outcome Assessment · Healthcare · Psychometrics · Cognition · Questionnaire

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Introduction

Patient-reported outcome measures (PROMs) are validated questionnaires that are completed by patients to ascertain perceptions of their health status and well-being [1, 2]. PROMs range from single-item symptom ratings e.g., pain scales, to complex multidimensional tools measuring health-related quality of life [3]. PROMs are considered to be the gold standard for measuring subjective experiences, because the information comes directly from the patient [4]. When collecting data on the health-related outcomes of children and young people (CYP) it is good practice to enable CYP to self-report whenever possible.

The design and implementation of PROMs for CYP presents methodological complexities, including consideration

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of response format, recall period and the mode of administration [5, 6]. These considerations should be addressed at the design stage to ensure PROMS are both feasible (*ability* to complete a measure) and acceptable (*willingness* to complete a measure) [7]. Acceptable modes of administration are crucial to enable CYP to engage and provide valid and reliable results [8].

Careful consideration of recall period, response scale format and administration modality during all stages of PROM design may increase response and completion rates, whilst maintaining and enhancing validity and reliability. The aim of this review is to systematically appraise the evidence on response scale type, recall period, administration modality and approaches to enable CYP < 19 years to participate in valid and reliable self-reporting of their health outcomes.

Methods

This systematic literature review was conducted and reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [9], and registered on PROSPERO (CRD42019135264).

PsycINFO, Medline, CINAHL and Embase were searched from 1st January 1980 (i.e., when outcome measurement in children began to be reported in the scientific literature [10–12]) to 15th March 2020. The search combined terms for children used in a previous systematic review [13] with those for different response scale formats, recall periods and methods of administration (* MERGEFORMAT Table 1 Search terms). Additional articles were searched using 'cited by' (Scopus), forwards and backwards referencing and consulting other experts in the field. The full Medline search strategy is reported in Supplementary Appendix 1.

Inclusion and exclusion criteria

Inclusion criteria were: (1) study population CYP ≤ 18 years old (studies reporting participants ≥ 19 years old were included if data were presented separately). Our original protocol planned to include those ≤ 17 years old but a large proportion of identified papers included 18 year olds so this was amended; (2) primary research of self-report of health outcomes among CYP; (3) studies evaluating recall period, response format, administration modality or approaches to engage CYP in self-reporting health outcomes in terms of their effect on measurement properties (validity, reliability and responsiveness) [7], acceptability (willingness to use a particular response format, administration mode or recall period), feasibility of use (ability to use a particular response format, administration mode or recall period) or preference for a particular mode, response format or recall period [7]; (4) written in the English language.

Table 1 Search terms

Children	Recall Period	Response format	Administration mode
Exp child/or exp p?ediatrics/or child* or (adolescen* or p?ediatric* or youth* or juvenile or teen* or young people or schoolchild* or school age* or kid*)	Recall period or recall interval or patient recall or recall bias	Response scale or likert scale or visual analog* scale or VAS or numerical rating scale or verbal rating scale or faces scale or dichotomous scale or yes no response or response option*	(Outcome measure adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet) or (measure adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet) or (scale adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet) or (questionnaire adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet) or (survey adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)
Combined with 'and'			

Exclusion criteria were case reports of < 3 participants (due to the risk of selection bias), discussion articles, editorials, reports, letters and reviews.

Study selection and data extraction

Citations were imported to EndNote (v9) and de-duplicated. Titles and abstracts of retrieved articles were screened for eligibility by one reviewer (LC). If there was not enough information within the title and abstract to determine eligibility, the full text article was screened. Remaining full text articles were screened by LC. 10% of the full text articles were screened by a 2nd reviewer (JA). Any discrepancies were resolved through discussion, and a third reviewer consulted as necessary (CES or RH).

Data from eligible studies were extracted into a common table: study authors, year of publication, geographic location, objective, study design, sample characteristics (population, size, setting), measure characteristics reported (recall period, response format, administration modality) and main findings.

Quality appraisal and data synthesis

QualSyst was applied rather than the COSMIN checklist in line with the overall aim of this review to examine response format, administration mode and recall period, rather than to appraise specific PROMs [14]. QualSyst assesses study quality with two scoring systems, one for qualitative and one for quantitative research. The qualitative scale consists of ten items with scores from zero to two, yielding a maximum score of 20. The quantitative scale consists of 14 items with scores from zero to two, an option to score an item 'not applicable', and maximum total score of 28. Overall scores are reported as percentages. Mixed method studies received two scores—one each for the qualitative and quantitative components [15]. Inter-rater agreement was assessed for 10% of the included articles.

Results were synthesized narratively to appraise the heterogeneity of included studies, and any similarities or differences in findings. The results were used to make recommendations on recall period, response format and administration mode when developing self-reported health outcome measures for CYP.

Results

Study selection

The search identified 13,207 articles after deduplication. A further 8 were identified via reference searching. 187 articles required full text review and 81 met the inclusion criteria. Of the articles included, 45 reported on response format [16–60], seven on recall period [61–67], 24 on administration mode [68–91], four on both recall and response format [92–95] and one on response format and administration mode [96]. The PRISMA flowchart is shown in Fig. 1 [9].

General Information on Included Studies

Tables 2, 3 and 4 summarise included studies and quality scores. Supplements 2 and 3 provide details of quality scores by item. The majority of included studies were conducted in Europe ($n = 25/81$) [17–20, 22, 26, 34, 37, 40, 41, 44, 50, 59, 65, 69, 71, 72, 75–78, 82–84, 87], the USA ($n = 31/81$) [16, 28, 29, 36, 38, 46–48, 51, 53, 54, 57, 58, 61–64, 66–68, 70, 74, 79, 80, 85, 86, 89, 90, 92, 95, 96] and Canada ($n = 18/81$) [21, 23–25, 27, 32, 39, 42, 43, 49, 52, 55, 56, 60, 81, 88, 91, 93] with two from Australia [31, 33], and one each from Japan [45], Korea [35], New Zealand [73], Kenya [94] and Jordan [30]. With respect to study design, $n = 68/81$ used quantitative methodology, $n = 11/81$ qualitative methodology and $n = 2/81$ mixed methods. Settings were predominantly home, school/nursery or hospital, and the 33,834 participants ranged from 3 to 18 years and were either healthy children ($n = 30$) or had one of a wide range of medical conditions ($n = 50$).

Quality of included studies

Study quality ranged from 38 to 96%, with 10/81 scoring less than the 55% quality inclusion threshold recommended by the QualSyst [15]. The main reasons for poor scoring were small sample size, using parametric statistical tests without stating whether data was normally distributed, treating data from Likert scales as if it was interval, using Pearson's correlation coefficient instead of intraclass correlation coefficient [97] and not stating randomisation methods. Qualitative papers rarely discussed reflexivity, the role of the researcher in the interview process or the connection to a theoretical framework. These low scoring studies were included in the review as it is often difficult to determine whether quality scoring elements were not reported rather than not taken into consideration.

Response format

50 papers investigated ability to use specific response formats [16–60, 92–96] (see Table 2 for details). The majority reported on one or more of the following pictorial scales, (faces pain scale revised (FPS-R) or Wong-Baker faces) ($n = 24$), visual analogue scales (VAS) ($n = 15$), and Likert scales (numerical or word descriptor) ($n = 14$). The methodology for these studies was mainly quantitative, assessing acceptability, feasibility, validity and reliability. Nine

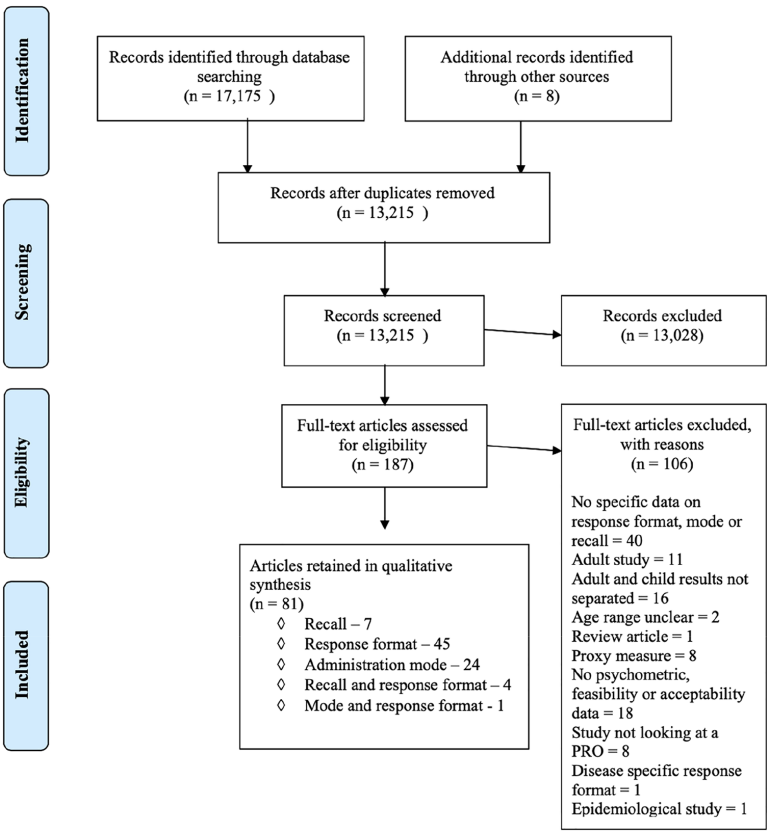

Fig. 1 PRISMA flowchart of study selection process [9]

qualitative studies used cognitive interviews to assess children's understanding of response formats.

One study demonstrated that 3-year-olds exhibited a 'yes' bias to knowledge and preference-based questions even though they knew the answer should be 'no'. By the age of 5–6 years this response bias did not exist in preference-based questions and was only weakly associated with knowledge questions regarding familiar objects [45].

Pictorial scales ($n = 24$ studies)

Most pictorial scales for children are 'faces' scales. These are generally used for self-reporting pain and show a series of faces with graded intensity from 'no pain' to 'worst pain possible' [24]. Children are asked to point to the face that best shows how they are feeling. Most studies in this review have used either the Wong-Baker Faces scale ($n = 5$) or the FPS-R (n-19). The Wong-Baker scale has six cartoon-like, hand drawn faces ranging from smiling to crying with a score of 0–5 [98]. The FPS-R was adapted from the original FPS which had seven faces [99]. The FPS-R excludes smiles and tears and has six hand-drawn faces rather than seven so that it can be scored from 0 to 5 allowing scoring to be in line with other pain measures [32]. There is also a simplified version of the FPS (S-FPS), designed for children 3–5 years old, which first asks the child if they are in pain and if they respond 'yes' then they are shown a three-point faces scale [27].

From the age of seven, the use of six-point faces scales shows construct (convergent and discriminant) validity [16, 41, 49, 56, 96]. Convergent validity was found with numerical/verbal rating scales, VAS and the Poker Chip Tool in children 6–8 years old ($r > 0.7$ or $p < 0.001$) [22]). The Poker Chip (known as Pieces of Hurt) tool involves children being asked to pick the number of Poker Chips that represent their level of pain. One chip represents a small amount of pain and four the most amount.

Cognitive interview studies showed that children of 7 and over are generally able to understand and complete faces measures [96]. In younger children, the evidence on ability

Table 2 Summary of studies on response format

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Baxter (2011) [16]; USA; Quantitative; Prospective; Feasibility, construct validity, responsiveness	To create and validate a pictorial scale with regular incremental levels between scores depicting increasing nausea intensity (BARF scale)	*N* = 127; Hospital; 7–18 years; Emergency department and surgery	The Spearman correlation coefficient of the first paired BARF and VAS for nausea scores was 0.93. VAS for nausea and BARF scores ($P = .20$) were significantly higher in patients requiring antiemetic agents and decreased significantly after treatment, while posttreatment pain scores ($P = .47$) for patients receiving only antiemetic agents did not. All patients understood the pictorial faces scales	68
Benson (2016) [17]; UK; Quantitative; Prospective; Construct validity	To test items, identified through previous qualitative interviews, that might form the basis of a new Malocclusion Impact Questionnaire for young people	*N* = 184; Hospital; 10–16 years ; Dental outpatients	Using Rasch analysis it was shown that all but one item had disordered thresholds, indicating response categories were not functioning as expected. The original 5-point response scale format was reduced to 3 points	60
Berntson (2001) [18]; Sweden; Quantitative; Cross-sectional; Acceptability, construct validity reliability	To evaluate the concordance between pain assessments made on a VAS[a] and a 4-point verbal descriptor scale and establish scale preference	*N* = 12; Hospital; 10–18 years; juvenile arthritis	Slight pain on verbal scale corresponded to a wide interval of 7–65 on VAS suggesting VAS was difficult to interpret. Preference was for VAS (69%) but this did not show the most reliable results	68
Borgers (2003) [19]; Netherlands; Quantitative; Prospective; Feasibility, reliability	To investigate the effects of partially labelled response options and vague quantifiers in response stability compared to completely labelled response options and the use of clearly quantified words in children of different ages	*N* = 91; Home; 8–16 years; Healthy	No effect on stability over time was found when offering vague quantifiers in the response options ($p > 0.05$). Young children do not benefit from the extra information of completely labelled response options. Offering different types of response option can lead to substantially different structural models	75
Borgers (2004) [20]; Netherlands; Quantitative; Prospective Feasibility, reliability	To examine the effects of negatively formulated questions, number of response options and offering a neutral midpoint as response option question characteristics on the reliability of responses	*N* = 222; Home; 8–16 years; Healthy;	Negatively formulated questions had no effect on reliability, although children respond consistently differently to negatively formulated questions as opposed to positively formulated ones. Offering about 4 response options is optimal (reliability increased up to 6, more than 7 caused a decrease)	80

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Campbell (2011) [21]; Canada; Quantitative; Cross-sectional; Feasibility	To investigate the utility of a VAS[a] to measure peer conflict resolution knowledge in children with language impairment (LI) and typically developing peers (TLD). Are children with varying language status able to express nuances in social knowledge by marking responses along the full VAS	*N* = 26; School; 9–12 years; Healthy	Those with TLD used the whole VAS; most (83%) with LI relied solely on scale anchors	59
Castarlenas (2013) [22]; Spain; Quantitative; Cross-sectional; Acceptability, construct validity	To assess whether the NRS-11[b] is a valid tool with 6–8 year old children when presented verbally	*N* = 126; School; 6–8 years; Healthy	The NRS-11 showed high convergent construct validity with VAS[a], FPS-R[c] and CAS[d] (r = 0.73–0.86), adequate discriminant validity (z = 2.05–5.55) and adequate criterion validity (r = 0.45–0.70). Preference order = CAS > NRS > FPS-R > VAS	75
Chambers (1998) [23]; Canada; Quantitative; Cross-sectional; Feasibility	To examine the potentially biasing impact of neutral or smiling face as a no pain anchor on children's reports of pain in response to a series of vignettes	*N* = 100; Childcare centres; 5–12 years; Healthy	Children who use a smile anchored scale had significantly higher pain scores for no pain and pain negative emotions ($p < 0.001$) and significantly lower faces pain scores for pain/positive vignettes than children who use a neutral anchored face scale ($p < 0.001$). Faces scales that use smiling anchors may confound affective states with pain ratings	63
Chambers (1999) [24]; Canada; Quantitative; Cross-sectional; Acceptability, feasibility	To examine the potential for bias in children's' self-report of pain when using scales with smiling rather than neutral anchors and to establish preference of type of faces scale	*N* = 75; Hospital; 5–12 years ; Endocrine/ diabetes	Scores across scales were highly correlated (r = 0.81–0.93). There was no age or gender interaction effect. Pain was rated significantly higher when scales with a smiling, rather than neutral, anchor were used ($p = 0.001$). 52.1% of children preferred scales they perceived to be happy and cartoon-like	75

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Chambers (2005) [25]; Canada; Quantitative; Cross-sectional; Acceptability, feasibility	To determine whether scales beginning with a smiling rather than neutral "no pain" face would produce higher ratings in the assessment of postoperative pain intensity in children and to compare ratings using different faces. Preference also asked	*N* = 78; Hospital; 5–13 years ; Post-surgical	Children's ratings of postoperative pain intensity are influenced by the presence of smiling "no pain" face at the beginning of faces scales, with such scales producing significantly higher ratings than scales with neutral "no pain" faces ($p < 0.01$). Ratings on the independent CAS[d] measure were more comparable to those provided on faces scales with neutral "no pain" faces. 55.6% preferred Wong Baker faces scale despite it giving the highest pain scores	83
Decruynaere (2009) [26]; Belgium; Quantitative; Cross-sectional; Construct validity, feasibility	To examine with the rating scale model how a sample of healthy children from 4–7 distinguish different faces when rating imaginary painful situations	*N* = 121/76; Schools and sports centres; 4–7 years ; General	Children performed better on a 3-point faces scale than 6-point scale. Ability improves with age on a 3-point faces scale. 4–5-year-olds could only distinguish 2 response categories	70
Emmott (2017) [27]; Canada; Quantitative; Cross-sectional; Construct validity, feasibility	To evaluate validity and feasibility of 2 simplified pain scales—S-FPS and S-COS in pre-school age children	*N* = 180; Hospital; 3–6 years ; Venepuncture	The ability to discriminate pain vs no pain was improved with S-FPS[d] and S-COS[f] ($p = 0.858$) compared with FPS-R[c] ($p = 0.036$ with S-FPS and $p = 0.022$ with C-COS) within 4–6-year olds but not 3-year olds. Quantitative pain rating remains challenging for 3-year-olds	88
Fanciullo (2007) [28]; USA; Quantitative; Cross-sectional; Acceptability, construct validity, feasibility	To determine initial psychometric properties and feasibility of a new Computer Face Scale for measuring children's pain	*N* = 54; Hospital; 3–17 years ; Hospitalised in pain/healthy	76% of children from3 years preferred moveable online faces to select their degree of pain over paper and pen static faces. Paired t tests showed significantly more hospitalised children reported pain than non-hospitalised ($p < 0.001$). Correlation with Wong-Baker faces scale $r = -0.72$	75

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Fritz (1994) [29]; USA; Quantitative; Prospective; Feasibility	To determine whether the use of pictorial anchors improved a VAS[a] designed to assess asthma episodes	*N* = 77; Summer camp; 8–15 years; Asthma	The mean VAS scores increased by 64% using the pictorial VAS while the mean PEFRs[g] in the 2 years were almost identical, suggesting that changes on the VAS were not due to differences in pulmonary functioning. For boys, the increase in individual mean VAS score in year 2 using the pictorial VAS was 44%; for girls, the increase in individual mean VAS scores was 112%. Use of a pictorial anchor led to greater usage of the full range of the scale	77
Gharaibeh (2002) [30]; Jordan; Quantitative; Cross-sectional; Acceptability, construct validity, reliability	To test the reliability and cultural validity of the following three pain assessment scales: Faces Scale, the Word Description Scale, and the Poker Chip Scale	*N* = 95; Hospital; 3–14 years; Venepuncture	55.8% of children preferred the Poker Chip Tool to the Faces Scale and the Word Descriptive Scale. There was significant convergent validity ($p < 0.01$) and test–retest reliability ($p < 0.01$) between the three scales	60
Goodenough (1997) [31]; Australia; Quantitative; Cross sectional; Construct validity, feasibility	To compare the utility of the Faces Pain Scale with three other self-report measures (VAS[a], Poker Chip, VRS[h]) of pain severity. These four scales were compared and contrasted in terms of the facility of application and comprehension by young children and their relative response frequency distributions	*N* = 50; Hospital; 4–7 years; Immunisation	Scores on all 4 scales correlated well ($r > 0.7$). The scales seemed to be measuring the same construct of pain. The faces scale was well understood. 12% had difficulty with the Visual analogue toy scale. The faces pain scale was skewed to low possibly because there are too many response options for the age group causing them to choose the extreme options	60
Gulur (2009) [58]; USA; Quantitative; Prospective; Acceptability, construct validity, reliability	1) to determine whether children understood the link between the facial expressions of smiling and frowning and the subjective feelings of happiness and pain/hurt. 2) to determine whether children understood that relative degrees of smiling or frowning were linked to relative degrees of happiness and pain/ hurt. 3) to determine the concurrent validity of the Computer Face Scale with the Wong-Baker Faces Scale. 4) to determine the test–retest reliability of the Computer Face Scale	*N* = 79/50; Hospital; 3–17 years; Study 1 Surgical; Study 2 general inpatients	The computerised scale showed concurrent validity with Wong-Baker faces ($r = -0.68$). 15-min test–retest reliability $r = 0.77$. 77% preferred the computerised faces scale. Participants were able to use both scales	45

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Gupta (2016) [96]; USA; Qualitative; Cross-sectional; Acceptability, feasibility	To evaluate comprehension and usability of a modified electronic version of the electronic version of the FPS-R[c] for children aged 4–17 years with sickle cell disease	*N* = 22; Unclear; 4–17 years; Sickle cell	Children age 4–6 years were generally unable to demonstrate understanding of the FPS-R and its response scale. Children > 7 years understood the scale and could complete it electronically. Those aged 7–8 years often needed parental assistance	55
Hicks (2001) [32]; Canada; Quantitative; Cross-sectional; Construct validity	1) to revise the FPS[i] from 7 faces to 6 to make scores comparable to other measures (0–5 or 0–10). 2) to evaluate the validity of the revised version	*N* = 76/45; Ear piercing/Hospital; 4–12 years; Healthy/hospital	The validity of the revised scale is supported by a strong positive correlation ($r = 0.93$) with a VAS[a] measure in healthy children aged 5–12 years. In hospitalised children the revised scale correlated with the VAS ($r = 0.92$) and CAS[d] ($r = 0.84$)	60
Hunter (2000) [33]; Australia; Quantitative; Cross-sectional, Feasibility	To further investigate the psychometric properties of the faces pain scale	*N* = 135; School; 3.5–6.5 years; Healthy	All children were capable of making meaningful discriminations. Children had difficulties with the middle of the scale suggesting that it formed an acceptable series but could not be considered an interval scale. The scale is best reserved for school age children	50
Irwin (2009) [95]; USA; Qualitative; Cross-sectional; Feasibility	To conduct cognitive interviews with children and adolescents to gain feedback on items measuring physical functioning, emotional health, social health, fatigue, pain and asthma specific symptoms for PROMIS item bank	*N* = 77; Hospital/community; 8–17 years; Healthy/asthma	Response options were understood by the majority of participants (up to 5 options). Children could clearly identify variable levels of functioning. Younger children misunderstood the word difficulty, so it was changed to trouble	65
Joffer (2016) [34]; Sweden; Qualitative; Cross-sectional; Feasibility	To explore how adolescents interpret and reason when answering a question on self-rated health	*N* = 58; School; 12–18 years; Healthy	Participants' understandings of the response alternative "Neither good, nor bad" varied. Some regarded it as normal and "in the middle", some as a negative alternative, and others as a passive state. The five response options all demonstrated differences in self-rated health	60

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Jung (2018) [35]; Korea; Quantitative; Prospective; Construct validity	To develop and validate the "Pain Block" concrete ordinal scale for 4- to 7-year-old children. Psychometric properties were compared with the FPS-R[c]	*N* = 163; Hospital; 4–7 years; Emergency dept	Agreement between the 2 pain scales was acceptable, with 95% of the values within the predetermined limit ($r = 0.82$). The pain scores for both pain scales were significantly decreased when analgesics or pain-relieving procedures were administered ($p < 0.001$). The Pain Block pain scale could be used to assess pain in 4- to 7-year-old children capable of understanding and counting up to the number 5, even if they do not understand the FPS-R pain scale	68
Keck (1996) [36]; USA; Quantitative; Prospective; Acceptability, construct validity, reliability	To investigate the Faces and modified Word Descriptor Scale for concurrent validity, discriminant validity and test retest reliability	*N* = 118; Hospital; 3–18 years; Haematology and oncology; venepuncture	Both the word descriptor and faces scales demonstrated discriminant validity ($p < 0.001$ for scores before and after painful procedure) and concurrent validity ($r > 0.71$) and test–retest reliability (faces $r = 0.9$ and word scale $r = 0.92$). All children understood the scales. The majority of children in all age groups preferred the faces scale (65.1%)	50
Klassen (2015) [60]; Canada; Mixed methods; Cross-sectional; Acceptability, construct validity, feasibility, reliability,	1) to conduct individual cognitive interviews with adolescents age 12–18 with different health conditions to obtain their feedback on the instructions, response options and items of a transition questionnaire (Transition-Q) with a 5-point Likert response option and to identify any missing content and to revise the scale as necessary. 2) conduct a large-scale field test to examine reliability and validity	*N* = 32/37; Hospital; 12–18 years; Chronic conditions	Item response option thresholds weren't ordered for 13 of 18 items. Items were rescored in to 3 response options. 14 participants did not like the agree/disagree response format. It was changed to frequency (never, sometimes, often and always). This was preferred by 8/9 in the second round. Cronbach's $\alpha = 0.85$. Test–retest reliability = 0.9	90 (quant) 55 (qual)
Lawford (2001) [37]; UK; Quantitative; Cross-sectional; Feasibility, reliability	To provide an empirical basis for selecting the response format of a QOL measure for 3–8-year olds (4 point Likert scale vs 4 coloured circles)	*N* = 28; Nursery school; 4–5 years; Healthy	The Likert scale took significantly longer to complete ($p < 0.005$). The coloured circle format had higher internal consistency than the Likert scale ($\alpha = 0.7$ vs 0.48)	65

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Leske (2015) [38]; USA; Quantitative; Cross-sectional; Construct validity	To use Rasch analysis to refine the Intermittent Exotropia Questionnaire, removing items that do not contribute meaningful information and ensure response options are properly interpreted	*N* = 575; Eye clinics; 8–17 years; Intermittent exotropia	Performance of the child and adult versions were enhanced by reducing the number of response options from 5 to 3	80
Locker (2007) [39]; Canada; Quantitative; Cross-sectional; Construct validity	To assess the performance of negatively and positively worded items in questionnaires to measure child perceptions of child oral health-related quality of life	*N* = 91; Dental clinics; 10–14 years; Dental/oro-facial	Positively worded items elicited significantly more 'don't know' responses and missing values. The performance of positively worded items was unsatisfactory	85
Luffy (2003) [57]; USA; Quantitative; Cross-sectional; Acceptability, construct validity, reliability	To compare the validity, reliability and preference of pain intensity measurement tools—the African American Oucher scale, Wong-Baker Faces scale and VAS[a]	*N* = 100; Outpatient clinics; 3–18 years; Sickle cell	Faces and African American Oucher are valid (no significant difference in scores between Oucher and Wong-Baker faces) and reliable (test–retest $p < 0.005$) tools for measuring pain in children. The VAS was not. 56% preferred the faces scale	50
Maïano (2009) [40]; France; Quantitative; Cross-sectional; Construct validity, reliability	To test the factor validity and reliability of 2 versions (graphical scale vs Likert scale) of the Very Short Form of the Physical Self-Inventory (PSI-VSF), with a sample of adolescents with mild to moderate intellectual disability	*N* = 342; School; 12–18 years; Learning difficulties	Both versions showed good structural validity, with the graphical version being superior. The graphical faces scale version had higher internal consistency ($\alpha = 0.7$–0.74 vs 0.65–0.67) than the Likert scale	80
McGrath (1996) [55]; Canada; Quantitative; Cross-sectional; Construct validity, feasibility	To determine the validity of the CAS[d] as a pain measure for children by evaluating the psychometric properties of the scale and comparing them to the properties of the VAS[a]	*N* = 104; 5–16 years; Routine check-up/pain clinics; Healthy/recurrent headache	There was no significant difference in pain scores between the VAS and CAS for the same event. Higher mean scores were reported for severe tissue damage injuries such as broken bones than for minor bruises. 87% found the CAS very easy to use whereas 22% found the VAS easy to use	70
Miro (2004) [41]; Spain; Quantitative; Prospective; Acceptability, construct validity, reliability	1) determine the initial psychometric properties of the Catalan version of the FPS-R[c] 2) compare patients' opinion of the FPS-R with the CAS[d]	*N* = 371; Hospital/school; 7–15 years; Hospitalised/healthy	Correlations between the FPS-R and CAS ranged from $r = 0.83$–0.9. Relationship between pain and affective state $r = 0.32$. Test–retest ranged from $r = 0.26$–0.7. The proportion of children that preferred the FPS-R was significantly higher than the proportion that preferred the CAS (66–68%)	46

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Morley (2014) [42]; Canada; Qualitative—cognitive interviews; Cross-sectional; Feasibility, acceptability	To cognitively test the Pediatric Advanced Care Quality of Life Scale (PAC-QoL) to establish whether the items and response options were understood	*N* = 34; Hospital; 8–18 years; Oncology;	Response scale was accurately interpreted in 88–93% of cases. When participants had trouble distinguishing between responses it involved options in the middle of the 4-point scale (sometimes and often)	65
O'Sullivan (2014) [43]; Canada; Qualitative; Cross-sectional; Feasibility	To evaluate and refine a new instrument for cancer symptom screening (SSPedi), including evaluating understanding of the response scale	*N* = 30; Hospital; 8–18 years; Oncology	Response options (5-point Likert) were understood by 90% of children	60
Ogden (2008) [44]; UK; Mixed methods; Cross-sectional; Acceptability, feasibility	To identify changes needed to adapt the IMPACT instrument for use in British children with inflammatory bowel disease and to see whether children preferred the Likert scale or the VAS[a]	*N* = 20; Outpatients; 8–16 years; Gastroenterology	Participants distinguished between the responses in the Likert scale and related their answers to the response options proficiently. Some children only guessed that 'moderate' meant 'in the middle' because of its position in the scale (5 point). 75% preferred the Likert scale to the VAS as it was easier and quicker to complete ($p < 0.01$)	55 quant 45 qual
Okanda (2010) [45]; Japan; Quantitative; Cross-sectional; Feasibility	To investigate whether 3–6-year-old children exhibit a 'yes' bias to various yes–no questions and whether their knowledge status affects the production of a yes bias	*N* = 135; Kindergarten/ nursery; 3–6 years; Healthy	3-year-olds had a strong tendency to exhibit a yes bias to both preference-object and knowledge object yes–no questions (even though they know the answer, $p < 0.01$). 4-year-olds could appropriately answer preference questions but showed a yes bias to knowledge questions ($p < 0.1$). 5- and 6-year-olds did not show a response bias to yes questions but showed a weak tendency to say yes to knowledge questions regarding familiar objects	55
Ortqvist (2012) [46]; USA; Qualitative; Cross-sectional; Feasibility	To examine how well the Knee Injury Osteoarthritis Outcome Score for Children (KOOS-Child) is understood	*N* = 34; Outpatient clinics; 10–16 years; Knee injury	Most children understood how to use a 5-point Likert response scale. The response option 'moderate' was persistently perceived as confusing. Most could interpret the meaning of the word by its location in the scale but could not define the word and suggested replacing it with the word 'some'	70

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Pagé (2012) [56]; Canada; Quantitative; Prospective; Acceptability, construct validity, feasibility	To evaluate the convergent and discriminant validity of the NRS[b] for pain intensity and unpleasantness in children after surgery	*N* = 83/69; Hospital; 8–18 years; Orthopaedic/general surgery	The NRS correlated highly with the VRS[b] and FPS-R[c] ($p < 0.001$). Scores were significantly higher at 48–72 h post-surgery than at 2 weeks ($p < 0.001$). Children found the faces scale easiest to use (51%). The VRS was least liked (13%) and hardest to use	82
Rebok (2001) [92]; USA; Qualitative—cognitive interviews; Cross-sectional; Acceptability, feasibility	(1) to determine whether children can answer health survey items. (2) to test the feasibility of a pictorial questionnaire format using cartoon drawings of a child. (3) to examine several types and numbers of response formats to see which are preferred and most easily understood. (4) to test children's understanding of specific concepts of health and wording of different response formats	*N* = 114; School/kindergarten; 5–11 years; Healthy	74% preferred circle responses to VAS[a], with 68% preferring graduated circles. 74% preferred 4 rather than 3 circles. 100% preferred a horizontal presentation. Younger children gave a significantly higher number of extreme responses. Younger children effectively reduced a 5-point response format to 3 points by using only the middle and extremes. 67% preferred the 5-point response format (rather than 4 point)	70
Shields (2003) [47]; USA; Quantitative; Cross-sectional; Feasibility	To identify demographic and cognitive variables that would maximise the accuracy of predicting children's abilities to use a VAS[a]	*N* = 40; Kindergarten; 5–7 years; Healthy	Only 42% of participants could use a VAS. Cognitive ability (IQ ≥ 100) combined with chronological age (≥ 5.6 years) was the best predictor of accurate use	80
Shields (2005) [48]; USA; Quantitative; Cross-sectional; Feasibility	To determine whether age, combined with estimated IQ, is an accurate predictor of a child's successful use of a VAS[a] in a non-clinical situation vs an acute, clinically emergent situation	*N* = 104; Hospital; 5–11 years; Healthy/lacerations	Estimated IQ and the ability to do a seriation task were the best predictors of 5–6-year-olds ability to accurately use the VAS ($p < 0.001$). Estimated IQ was not as important as chronological age and ability to perform a seriation task in those 7 years and over	83

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Stanford (2006) [49]; Canada; Quantitative; Cross-sectional; Feasibility	To examine variations in 3- to 6-year-old children's ability to accurately use a common self-report scale to rate pain in hypothetical vignettes (faces pain scale revised)	*N* = 112; Community; 3–6 years; Healthy	5- and 6-year-old children were significantly more accurate (40% errors) in their use of the FPS-R[c] in response to the vignettes than 4-year-old children, who in turn were significantly more accurate than 3-year-old children (60% errors). Over half of 6-year-olds demonstrated difficulty using the FPS-R in response to the vignettes. Child age was the only significant predictor of children's ability to use the scale in response to the vignettes ($p < 0.001$). The ability to use the scale improved with age	65
Staphorst (2017) [50]; Netherlands; Mixed methods; Cross-sectional; Acceptability, construct validity feasibility	To develop a generic, short and child-friendly instrument: the DISCO-RC questionnaire (DISCOmfort in Research with Children)	*N* = 46; Outpatients; 6–18 years; Unclear	Children preferred a 5-point Likert scale as a response option. The 5-point Likert scale coloured numeric VAS[a] and simple VAS were strongly correlated ($r = 0.76 - 0.99$)	60 (quant) 65(qual)
Tesler (1991) [51]; USA; Quantitative; Cross sectional; Acceptability, construct validity, reliability, responsiveness	A program of studies designed to select and test a pain intensity scale for inclusion in a multidimensional pain assessment tool for children, focusing on determining each scale's reliability, validity, ease of use and preference.5 scales were tested: a word graphic scale. VAS[a], graded graphic rating scale, 0–10 magnitude estimation scale and CAS[d]	*N* = 1223; Hospital, outpatient, school; 8–18 years; Acute/healthy/chronic illness	Convergent validity for the 5 scales was supported ($r = 0.66–0.84$). The word graphic rating scale (Likert) was preferred by 47% of sick children. When used in a multidimensional pain assessment tool it showed test–retest reliability ($r = 0.68–0.97$) also showed sensitivity to change ($p = 0.002$)	65
Tomlinson (2019) [93]; Canada; Qualitative; Cross-sectional; Feasibility	To develop a new self-report symptom screening tool for children receiving cancer treatments who are 4–7 years of age (mini-SSPedi), based on SSPedi	*N* = 100; Hospital; 4–7 years; Oncology	Dichotomous response scale (yes/no) was understood by all participants. 80% understood the Wong-Baker faces, 70% understood FPS-R[c] and 65% understood the pieces of hurt scales	60
van Laerhoven[59] (2004); Netherlands; Quantitative; Cross-sectional; Acceptability, feasibility	To examine which response options children prefer and which they find easiest to use (VAS[a] vs Likert). To examine the relative reliability of the different response options	*N* = 122; Outpatients; 6–12 years; Not specified	Children preferred the Likert scale. They considered the Likert scale easiest to fill out. Results of the different response options correlated strongly with each other ($r = 0.67 – 0.90$)	59

Table 2 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
von Baeyer (2013) [52]; Canada; Quantitative; Cross sectional; Feasibility	To evaluate a binary question followed by simple response options for pain assessment in young children (FPS-R)	*N* = 184; Preschool/day care; 3–5 years; Healthy	3- and 4-year-olds performed significantly better using the simplified task than the FPS-R[a] ($p < 0.001$). The simplified pain task made no difference to the 5-year olds who had almost identical mean scores using both methods. Response bias is common in children under 5	68
Vreeman (2014) [94]; Kenya; Qualitative—cognitive interviews; Cross-sectional; Acceptability, feasibility	To improve the understandability of paediatric antiretroviral adherence measurement items through cognitive interviewing with paediatric caregivers and HIV-infected adolescents	*N* = 10; HIV clinic; 13–18 years; HIV	Participants inconsistently quantified the differences between 4-point Likert response options. Visual analogue scales and the addition of a response option to give 5-points yielded more divergence and were considered hard to understand. It was suggested that VAS[a] would require pictorial cues to orientate the participant to scale meaning	70
Watson (2006) [53]; USA; Quantitative; Cross-sectional; Feasibility	To evaluate the psychometric properties of the fruit and vegetable self-efficacy (FVSEQ) questionnaire	*N* = 1477; School; 9–10 years; General	Item response modelling showed that the 5-point response scale was not fully utilised	86
West (1994) [54]; USA; Quantitative; Cross-sectional; Feasibility, construct and convergent validity	To identify a clinically feasible and accurate method of measuring pain intensity in paediatric oncology patients in the ITU (FPS and Poker chip)	*N* = 30; Intensive care; 5–13 years; Oncology	Pain rating scales on the two tools were correlated (faces pain scale and Poker Chip, r = 0.67). 91.6% preferred the faces pain scale to the poker chip tool	50

Table 3 Summary of studies on recall period

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Chogle (2012) [61]; USA; Quantitative; Prospective; Acceptability, feasibility	To assess ability to accurately recall abdominal pain in children—comparison of daily reports vs one-month recall	*N* = 63; Outpatients; 8–17 years; Functional gastro-intestinal disorders	Most children reported a lower frequency of abdominal pain by recall than daily diaries (r = 0.4; CI 0.17–0.59%). Children 8–11 years had a higher correlation (r = 0.59) than those 12–18 (r = 0.26). Similar correlations were found to just the past 7 days (r = 0.47)	68
Heyer (2014) [62]; USA; Quantitative; Prospective; Feasibility, reliability	To compare the 90 day and 30-day recall of paediatric migraine disability assessment (PedMIDAS) elements and headache frequency against daily entries from an internet headache diary	*N* = 52; Outpatients; 10–18 years; Migraine	Reliability improved at 30-day recall compared to 90 days. 90-day diary: PedMIDAD r = 0.65; headaches r = 0.8330-day diary: PedMIDAD r = 086; headaches r = 0.88. Age and confidence in ability to answer were poor predictors of recall accuracy	86
Irwin (2009) [95]; USA; Qualitative; Cross-sectional; Feasibility	To conduct cognitive interviews with children and adolescents to gain feedback on items measuring physical functioning, emotional health, social health, fatigue, pain and asthma specific symptoms for PROMIS item bank	*N* = 100; Hospital; 4–7 years; Oncology	All children reported that the 7-day recall period meant the past 7 days and responded to items accordingly	60
Jacobson (2015) [67]; USA; Qualitative; Cognitive interviews; Cross-sectional; Feasibility	To develop and evaluate item candidates for new PROMIS Pediatric Pain Quality and Pain Behavior item banks, and Pain Intensity items	*N* = 34; Hospital; 8–18 years; Chronic pain	Participants from 8–18 years old understood that the recall period referred to the past week. There was a need to reiterate the 7-day time period to some younger children	70
Okupa (2013) [63]; USA; Quantitative; Prospective; Feasibility, reliability	To compare daily diaries vs retrospective questionnaires to assess asthma control	*N* = 88; Asthma Research and Education Network Centres; 6–17 years; Asthma	Asthma control days correlated better with daily diary information from the last 2 weeks of a 4-week recall (r = 0.46) than from the first 2 weeks	68
Ravens-Sieberer (2014) [66]; USA; Qualitative; Cognitive interviews; Acceptability and feasibility	To (1) conceptualize children's subjective well-being and (2) produce item pools with excellent content validity for calibration and use in computerized adaptive testing	*N* = 37; Not stated; 8–17 years; Healthy and chronic conditions	Cognitive interviews supported children's capacity to use a 7-day recall period for positive affect and a 4 week recall period for life satisfaction	65

Table 3 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Rebok (2001) [92]; USA; Qualitative—cognitive interviews; Cross-sectional; Acceptability, feasibility	1) to determine whether children can answer health survey items. 2) to test the feasibility of a pictorial questionnaire format using cartoon drawings of a child. 3) to examine several types and numbers of response formats to see which are preferred and most easily understood. 4) to test children's understanding of specific concepts of health and wording of different response formats	*N* = 114; School/kindergarten; 5–11 years; Healthy	80% of participants could accurately use a 4 week recall period. Younger children did not understand the concept of a week and may not have used the 4-week time interval appropriately	70
Self (2015) [64]; USA; Quantitative; Prospective; Feasibility, reliability	To evaluate correspondence between retrospective questionnaire and prospective diary data for children and adolescents with IBS	*N* = 50; Outpatients; 8–18 years; Irritable bowel	For pain days ICC = 0.83 and days without bowel movement ICC = 0.74. Maximum pain score ICC = 0.8 and days with diarrhoea = -0.03. Although under conditions likely to facilitate agreement and with individual variation observed. Age was not significantly related to difference scores	70
Tomlinson (2019) [93]; Canada; Qualitative—cognitive interviews; Cross-sectional; Feasibility	To develop a new self-report symptom screening tool for children receiving cancer treatments who are 4–7 years of age (mini-SSPedi), based on SSPedi	*N* = 100; Hospital; 4–7 years; Oncology	Only 40% understood the time frame yesterday, so today was chosen for the measure	60
van den Brink(2001) [65]; Netherlands; Quantitative; Prospective; Feasibility, reliability	To investigate whether children and adolescents can recall prior headache complaints accurately and to study whether age, gender, headache severity, preferred coping strategies, depression, somatization, and trait anxiety are related to recall errors, causing recall bias	*N* = 100; School; 9–16 years; Headache	Compared to daily diary, retrospective questions led to overestimation of headache intensity and duration (r = 0.16). Lower age and increased headache severity were statistically related to recall errors	50
Vreeman (2014) [94]; Kenya; Qualitative; Cross-sectional; Acceptability, feasibility	To improve the understandability of paediatric antiretroviral adherence measurement items through cognitive interviewing with paediatric caregivers and HIV-infected adolescents	*N* = 10; HIV clinics; 13–18 years; HIV	Adolescents preferred either a 24-h recall period for ease of remembering or a 1 month recall as clinic appointments were monthly	70

Table 4 Summary of studies on administration mode

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Bender (2007) [68]; USA; Quantitative; Prospective; Reliability	To test the effect of reporting mode on accuracy of inhaled cortico-steroid adherence reporting in children with asthma and their parents under conditions similar to those of an asthma clinical trial	*N* = 104; Outpatients; 8–18 years; Asthma	All methods led to over-reporting compared to electronic device on asthma pump. More than half of children over- reported adherence by > 25% Discrepancy was greatest in computer interview condition	77
Castarlenas (2015) [69] Spain; Quantitative; Cross-sectional; Acceptability, construct validity	To examine the agreement between verbally and electronically administered NRS-11[b] (eNRS) for pain	*N* = 191; School; 12–18 years; Healthy	Bland Altman LOA fell outside the a priori limit for 95%. LOA at 80% fell inside the maximum limit established a priori. K-coefficients ranged from 0.786–0.912 indicating almost perfect agreement. 83% preferred the eNRS	77
Eaton (2010) [89]; USA; Quantitative; Cross-sectional; Construct validity, feasibility	To examine whether paper and pencil surveys and web surveys yield equivalent risk behaviour prevalence estimates when using the Youth Risk Behaviour Survey	*N* = 5227; School; Unclear; Healthy	Prevalence estimates from paper and pencil and web-based surveys were generally equivalent. Questionnaire mode was only significantly ($p < 0.05$) associated with 7 of 74 risk behaviours	82
Fouladi (2006) [70]; USA; Quantitative; Cross-sectional; Construct validity, feasibility	To examine systematic differences in the responses of 4th, 5th, and 6th graders to measures of stress, coping, and humour among three modes of assessment: paper-and-pencil questionnaires, computer-assisted self-interviewing (CASI), or a combination of paper-and-pencil and CASI. Scales used – feel bad scale, school agers coping strategies inventory, the multi-dimensional sense of humour scale	*N* = 1245; School; 9–12 years; General	CASI means and medians were higher ($p < 0.002$) and correlations between CASI measures tended to be lower than those obtained with paper and pencil and mixed modes. CASI variances were lower	65
Geerdink (2009) [71]; Netherlands; Quantitative; Cross-sectional; Acceptability, construct validity, feasibility	To develop a reliable and user-friendly digital child health assessment questionnaire (CHAQ) to complete systematically at the outpatient paediatric rheumatology clinic	*N* = 51; Outpatients; Unclear; Juvenile arthritis	Correlation between the digital and paper versions was high ($r = 0.974$). No statistically significantly differences in median outcome were found in visual analogue scale (VAS) pain (25.6 vs 25.9 mm) and VAS well-being (20.1 vs 19.5 mm). Although the mean time (5.06 min) to complete the digital CHAQ was significantly longer than the mean time (3.75 min) to complete the paper form, the majority of patients (75%) preferred the digital version. User-friendliness received maximum positive score	59

Table 4 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Jensen (2010) [72]; Denmark; Quantitative; Prospective; Acceptability	To examine the assessments and priorities by children and adolescents of health care in a paediatric outpatient clinic, to examine the influence of the time factor on assessments and priorities by children and adolescents of health care, and to determine their preferred method of evaluation	*N* = 346; Outpatients; 11–17 years; Range of diagnoses	50.1% of children and adolescents preferred to complete an electronic questionnaire to a paper one. They did not want to receive questionnaires by email	45
Jones (2010) [73]; New Zealand; Quantitative; Prospective; Acceptability, construct validity, reliability	To investigate the reliability and validity of a computerised anxiety assessment (smiley faces program revised (SFP-R)) and to explore children's preferences for the method of anxiety assessment	*N* = 206; School; 5–13 years; Healthy	The online SFP-R demonstrated good reliability ($\alpha = 0.75$) and strong convergent validity with the modified children's dental anxiety scale ($r = 0.75$). Test–retest reliability $r = 0.67$. Children preferred the computerised assessment to pen and paper methods	54
Knight (2007) [74]; USA; Quantitative; Cross-sectional; Acceptability	To determine adolescents' preferences for method of substance abuse screening	*N* = 2133; Outpatients; 12–18 years; General medicine	Paper was the preferred method (mean rank (MR) = 2.92, 95%CI 2.87–2.96) vs. computer (MR = 2.38, 2.33–2.43), nurse (MR = 2.43, 2.39–2.47), and doctor (MR = 2.30, 2.25–2.35). Participants stated they were more likely to be honest with paper followed by computer, rather than responding to questions administered by a doctor or nurse. Those reporting on the computer were significantly more likely to report drug and alcohol use	67
Lloyd (2011) [75]; UK; Quantitative; Cross-sectional; Construct validity, feasibility, Reliability,	To examine the psychometric properties of an Internet version of a children and young persons' quality of life measure (Kid's Life and Times) originally designed as a paper questionnaire	*N* = 3440; School; 10–11 years; Healthy	Exploratory principal component analysis supported 5 components, in line with the paper version. Items loaded on to the expected components. Internal consistency was similar to that reported for the paper version (α all > 0.76). Domain scores were similar to those reported in the literature for the paper version. Non-response was lower with the online version (1% vs 1.72–3.83%)	72

Table 4 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Magnus (2016) [90]; USA; Quantitative; Cross-sectional; Construct validity	To test the equivalence of scores obtained with the PROMIS paediatric depressive symptoms, fatigue and mobility measures across computer and telephone administration	*N* = 377; Home; 8–17 years; Healthy	There were high correlations between the two modes of administration (0.71–0.94), although fatigue scores were affected by mode of administration, but the differences in scores were sufficiently small that they would not affect overall interpretation of results	77
Mangunkusumo (2005) [76]; Netherlands; Quantitative; Cross-sectional; Acceptability, construct validity	To assess whether scores of an internet administered adolescent health questionnaire (KIVPA) are equivalent to those obtained via paper and pencil. To compare adolescents' evaluation of administration modes	*N* = 565; School; 13–17 years; Healthy	Internet questionnaire generally resulted in equal scores to pen and paper mode. Adolescents in the internet one-item mode group more frequently reported satisfaction with appearance compared with the Internet multiple items mode ($p \leq .01$). The internet group had more adolescents reporting that they had a sufficient number of friends compared to the paper mode ($p \leq .01$)	77
Mangunkusumo (2006) [77]; Netherlands; Quantitative; Cross-sectional; Construct validity, feasibility	To compare the feasibility, presence of score differences and subjective evaluations by children between Internet and identical paper questionnaires (International study of asthma and allergies in childhood questionnaire)	*N* = 249; School; 10–12 years; Healthy	There were similar mean scores between administration modes. ICC 0.64–0.9. One third of items showed moderate agreement between modes (kappa 0.43–0.6). The remaining items had very good agreement (kappa 0.61–0.95). There were fewer missing data with the internet version	82
Mauz (2018) [78]; Germany; Cross-sectional; Acceptability, construct validity, feasibility	To determine whether prevalence rates or mean values of self-reported health indicators for children and adolescents age 11–17 years differ between self-administered paper-based questionnaires and self-administered web-based questionnaires (German Health Interview and Examination Survey for Children and Adolescents)	*N* = 1194; Home; 11–17 years; Healthy	Most questions showed mode equivalence except for alcohol consumption. Higher levels of consumption were reported online ($p < 0.001$). Male adolescents preferred the online mode. Those choosing the web-based response format were more likely to have higher household income and higher educational attainment (actual data not reported)	71
McCabe (2005) [79]; USA; Quantitative; Cross-sectional; Construct validity, feasibility	To examine the feasibility and mode effects of using a web form vs paper form survey to collect alcohol and tobacco data from 3rd and 4th grade students	*N* = 323; School; Not specified (3/4 grade); Healthy	There were minimal differences between survey modes. (future alcohol use and lifetime alcohol use showed significant difference, $p < 0.05$))	55

Table 4 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Moskowitz (2004) [80]; USA; Quantitative; Cross-sectional; Construct validity, feasibility	To assess the effect of telephone audio computer-assisted self-interviewing (A-CASI) and computer-assisted telephone interviewing (T-ACASI), on self-reports of smoking behaviour and smoking susceptibility among adolescents 12–17 years of age (adapted from Youth Attitudes and Practices Survey)	*N* = 2444; Home; 12–17 years; Healthy	Adjusted estimates of current smoking were higher in the self-administered T-ACASI (8.3% vs 4.5%). The commitment not to smoke among those who had never smoked was also higher in the T-ACASI (45% vs 34.9%). Parental presence was negatively associated with smoking. T-ACASI survey had more missing data than CATI	77
Nitikman (2017) [81]; Canada; Quantitative; Prospective; Construct validity, feasibility, reliability	To validate and test the reliability of using the Internet as a method of administering health-related quality of life questionnaires in a paediatric spine population (Scoliosis Research Society 30 (SRS-30) and Pediatric Outcomes Data Collection Instrument (PODCI))	*N* = 96; Outpatients; 11–18 years; Scoliosis	There was no significant difference in scores between methods of administration at the 2 time points (p = 0.206). Patients expressed a preference for the internet option (84%)	63
Raat (2007) [82]; Netherlands; Quantitative; Cross-sectional; Construct validity, feasibility, reliability	To evaluate the indicators of feasibility, reliability and validity of the Child Health Questionnaire-Child Form (CHQ-CF). To compare the results in those of those who complete the standard paper version compared to an internet version	*N* = 933; School; 13–17 years; Healthy	The internet version resulted in fewer missing answers. All scales clearly discriminated between adolescents with no, a few or many self-reported chronic conditions. The paper administration resulted in statistically significant, higher scores on 4 of 10 CHQ-CF scales compared with the internet administration ($P < 0.05$), but Cohen's effect sizes d were ≤ 0.21. Mode of administration interacted significantly with age ($P < 0.05$) on four CHQ-CF scales, but Cohen's effect sizes for these differences were also ≤ 0.21	96
Raat (2007) [83]; Netherlands; Quantitative; Cross-sectional; Construct validity, feasibility	To compare the results from written and internet questionnaires about respiratory symptoms to find out if both forms yielded the same responses (International Study of Asthma and Allergies in Childhood (ISAAC) questionnaire)	*N* = 933; School; 13–17 years; Healthy	The Internet version showed fewer missing answers not statistically significant). The respiratory items did not show statistically significant score differences between the Internet and written modes of administration. Both approaches yielded equal results	96

Table 4 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Moskowitz (2004) [80]; USA; Quantitative; Cross-sectional; Construct validity, feasibility	To assess the effect of telephone audio computer-assisted self-interviewing (A-CASI) and computer-assisted telephone interviewing (T-ACASI), on self-reports of smoking behaviour and smoking susceptibility among adolescents 12–17 years of age (adapted from Youth Attitudes and Practices Survey)	*N* = 2444; Home; 12–17 years; Healthy	Adjusted estimates of current smoking were higher in the self-administered T-ACASI (8.3% vs 4.5%). The commitment not to smoke among those who had never smoked was also higher in the T-ACASI (45% vs 34.9%). Parental presence was negatively associated with smoking. T-ACASI survey had more missing data than CATI	77
Nitikman (2017) [81]; Canada; Quantitative; Prospective; Construct validity, feasibility, reliability	To validate and test the reliability of using the Internet as a method of administering health-related quality of life questionnaires in a paediatric spine population (Scoliosis Research Society 30 (SRS-30) and Pediatric Outcomes Data Collection Instrument (PODCI))	*N* = 96; Outpatients; 11–18 years; Scoliosis	There was no significant difference in scores between methods of administration at the 2 time points (p = 0.206). Patients expressed a preference for the internet option (84%)	63
Raat (2007) [82]; Netherlands; Quantitative; Cross-sectional; Construct validity, feasibility, reliability	To evaluate the indicators of feasibility, reliability and validity of the Child Health Questionnaire-Child Form (CHQ-CF). To compare the results in those of those who complete the standard paper version compared to an internet version	*N* = 933; School; 13–17 years; Healthy	The internet version resulted in fewer missing answers. All scales clearly discriminated between adolescents with no, a few or many self-reported chronic conditions. The paper administration resulted in statistically significant, higher scores on 4 of 10 CHQ-CF scales compared with the internet administration ($P < 0.05$), but Cohen's effect sizes d were ≤ 0.21. Mode of administration interacted significantly with age ($P < 0.05$) on four CHQ-CF scales, but Cohen's effect sizes for these differences were also ≤ 0.21	96
Raat (2007) [83]; Netherlands; Quantitative; Cross-sectional; Construct validity, feasibility	To compare the results from written and internet questionnaires about respiratory symptoms to find out if both forms yielded the same responses (International Study of Asthma and Allergies in Childhood (ISAAC) questionnaire)	*N* = 933; School; 13–17 years; Healthy	The Internet version showed fewer missing answers not statistically significant). The respiratory items did not show statistically significant score differences between the Internet and written modes of administration. Both approaches yielded equal results	96

Table 4 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Robles (2015) [84]; Spain; Quantitative; Cross sectional; Construct validity, feasibility,	To develop web-based Spanish and Catalan versions of the EQ-5D-Y, and to compare scores and psychometric properties with the paper version	*N* = 715; School; 8–18 years; Healthy	Both formats of EQ-5D-Y showed low percentages of missing values ($n = 2$, and 4 to 9 for web and paper versions respectively), and a high ceiling effect by dimension (range from 79 to 96%). Percent agreement for EQ-5D-Y dimensions on the web and paper versions was acceptable (range 89% to 97%), and k ranged from 0.55 (0.48–0.61, usual activities dimension) to 0.75 (0.68–0.82, mobility dimension). Mean score difference on the VAS was 0.07, and the ICC for VAS scores on the two formats was 0.84 (0.82–0.86). Both formats showed acceptable ability to discriminate according to self-perceived health, reporting chronic conditions, and mental health status	83
Sun (2015) [91]; Canada; Quantitative; Longitudinal; Acceptability, construct validity, feasibility	To evaluate agreement between electronic (called Panda) and paper versions of the faces pain scale revised (FPS-R) and colour analogue scale (CAS)	*N* = 62; Hospital; 4–18 years; Surgical	Panda scores correlated strongly with original scores at T0 and T30 ($r > 0.93$ for FPS-R; $r > 0.87$ for CAS). Most participants expressed a preference for the iPod Panda version (76–81%)	67
Trapl (2013) [85]; USA; Quantitative; Cross sectional; Acceptability, feasibility	To examine the impact of 3 data collection modes (paper, PDA, audiPDA (APDA)) on the number of questions answered, data quality, and student preference	*N* = 275; School; Not specified (7th grade); Healthy	APDA respondents completed significantly more questions compared to paper and PDA ($p < 0.001$). PDA and APDA had significantly fewer missing data than did paper ($p < 0.001$). No differences were found for student evaluation	63
Varni (2009) [86]; USA; Quantitative; Cross-sectional; Construct validity	To implement the multigroup confirmatory factor analysis (CFA) method for invariance testing across mode of administration for children's self-reported health-related quality of life (in person, mail and telephone) using PedsQLTM 4.0 Generic Core Scales	*N* = 3741; Home or clinic; 5–18 years; Chronic illness	Strong factorial invariance across the mode of administration groups was demonstrated based on stability of the Comparative Fit Index (CFI) between the models, and several additional indices of practical fit including the Root Mean Squared Error of Approximation (RMSEA), the Non-Normed Fit Index (NNFI), and the Parsimony Normed Fit Index (PNFI). Children across the three modes of administration groups interpreted items on the PedsQLTM 4.0 Generic Core Scales in a similar manner	75

Table 4 (continued)

Author (date); Country; Study Design; Measurement properties evaluated	Objective	Sample size (*N*); Setting; Age; Population	Main findings	QualSyst Score (%)
Wood (2011) [87]; France; Quantitative; Cross-sectional; Acceptability, construct validity, feasibility	To compare concordance and preference for electronic and paper versions of the faces pain scale revised, and to determine whether the electronic version can be used by children 4 years and over	*N* = 234; Hospital; 4–12 years; Inpatients	Overall weighted kappa = 0.846 and Spearman's correlation between scores on the 2 versions was 0.91. The mean difference between scores was neither clinically nor statistically significant. 83.2% chose the same face on both versions. The PDA was preferred by 87.4% of participants	88
Young (2009) [88]; Canada; Quantitative; Prospective; Construct validity, feasibility	To test the impact of web administration on well-established measures of children's physical function and quality of life using the ASK and PedsQL measures	*N* = 91 time 1 *N* = 69 time 2; Hospital; 8–14 years; Chronic illness	Both measures were highly reliable in web and paper format. Inter-method ICC = 0.98 for ASK and 0.64 for PedsQL compared to ICC of 0.99 and 0.94 respectively for paper formats. The web ASK seems to be valid compared to paper format. Consistency in administration mode may be more important when using the PedsQL	88

to use faces scales is mixed. Two studies report that six-point faces scales are valid (convergent validity $r > 0.71$ with word descriptor scale; discriminant validity $p < 0.001$ before and after a painful procedure) and reliable (test–retest reliability $r = 0.9$, $p < 0.005$) in children as young as three. These studies had relatively low quality scores and data on 3–7-year olds was analysed together [36, 57]. Other studies have shown that not all children under 7 years are able to understand six-point faces scales, and some have difficulty in using the middle of the scale [33, 49, 93, 96]. There is no evidence that ability to use faces scales differs between healthy children and those with underlying conditions.

Although faces scales tended to demonstrate convergent validity with other response formats such as VAS and the Poker Chip tool in children between 4 and 7 years, scores tend to be skewed low, suggesting children are scoring at the extremes and are unable to use the middle response option [31]. Studies of the S-FPS suggest that from 4 years, a three-point faces scale can be used reliably, although 4-year-olds tend to use the scale anchors thus rendering it dichotomous [26, 27].

Scales with smiling anchors lead to reporting of higher pain scores in 5–13-year-olds, compared to those with neutral face anchors, although scores between the two scales correlate [23–25]. Children aged 5–12 years expressed a preference for cartoon like faces in one study [24].

Likert scales (*n* = 14 studies)

These studies were carried out with children 8 years and over, except one which had a lower age limit of 6 years [59]. Most showed that children from 8 years old can understand and use a 4 or 5-point Likert scale [20, 34, 42, 43, 46, 95], with scores correlating strongly with a VAS [59]. Cognitive interview studies (5–18 years) demonstrated that if children struggled with Likert scales, it was usually with the middle points of a scale [34, 42, 92] with the term 'moderate' being perceived as confusing [44, 46]. One study found that children 13–18 years old could not use a 4-point Likert scale as they were unable to quantify the differences between response options. Addition of a fifth point created more divergence and was harder to understand [94]. Four studies in children 8–18 years used item response theory to examine scale performance [17, 38, 53, 60]. Three found that using a five-point scale led to disordered thresholds and performance was enhanced by using a three-point scale [17, 38, 60]. One study in 9–10-year-olds showed that a five-point scale was not fully utilised [53]. Negatively formulated questions were shown to have no effect on reliability in one study [20]. As with faces scales, there is no evidence that ability to use a Likert scale differs between healthy and unwell children.

Visual analogue scales (*n* = 15 studies)

A visual analogue scale is usually a 100 mm long horizontal line with verbal descriptors at each end expressing extremes of feeling. Respondents mark a point on the line that best corresponds to the severity of their symptom or feeling [100].

At all ages the VAS seems to be less valid and reliable to use than faces or Likert scales, with slight pain on a verbal rating scale corresponding to a wide interval of 7–65 on a VAS scale [18, 57]. In children aged 5–7 years, cognitive ability, chronological age and the ability to conduct a seriation task (arranging circles in order of size) seems to be the best predictor of ability to use a VAS [47, 48]. Cognitive ability was less important after the age of seven [48]. This finding is supported by a study in children 9–12 years with learning impairment who only used the scale anchors, whereas children without learning impairment of the same age were able to use the whole VAS [21]. One study suggests that for those over 8 years old, the addition of pictorial anchors allowed children to make greater use of the full scale [29].

Other scales (*n* = 6)

The Pain Block Scale is a pictorial ordered block scale with a score between 0 and 10. This demonstrates agreement with the FPS-R and has discriminant validity in children from the age of 4–7 years who can count to five [35].

Two studies in children 3–14 years showed that the Poker Chip tool has convergent validity with faces scales ($r = 0.67$; $p < 0.001$) [30, 54] and one in children 4–7 years old showed convergent validity with VAS and VRS ($r = 0.7$) [31]. One study showed that 65% of 4–7-year olds understood the scale [93].

The coloured analogue scale (CAS) resembles a ruler, with one side showing a wedge-shaped figure filled with colour that progresses from white to red as the figure widens. The other side shows corresponding numerical ratings from 1 to 10 cm. One study demonstrated discriminant and construct validity with the VAS, and children from 5 to 16 years found the CAS easier to use than the VAS [55].

Preference of scale (*n* = 13)

13 studies asked children 3–18 years their preference of scale [18, 22, 30, 36, 41, 44, 50, 51, 54–56, 59]. In all studies using a faces scale this was preferred to VAS and Likert scales [22, 30, 36, 41, 54, 56, 57]. In all but one study, Likert scales were preferred to VAS [36, 50, 51, 59]. Four studies examined preference for the CAS, and in three it was preferred to FPS-R, VAS and Likert scales [22, 51, 55]. The FPS-R was preferred to the CAS in one study [41].

Recall period (*n* = 11)

11 studies reported on recall period [61–67, 92–95] (see Table 3 for details). Of these, 5/9 compared daily diary reports to retrospective questionnaires. Four of these were conducted in children 8 years and over and one in children from 6 years old. They showed that shorter recall periods lead to better correlation with daily diaries, with 7–14 days being optimal [61–65]. The other six studies were cognitive interview studies. These suggest that children under 8 years old cannot understand the concept of a week [92] and some could not understand the term 'yesterday' [93]. Those over 8 years could use both 7 day and 4-week recall periods [66, 67, 92, 95]. One study asked children 13–18 years old their recall preference and they suggested that 24 h was preferable but that one month would be easy to remember as they had monthly clinic appointments [94].

Administration mode (*n* = 24)

24 studies reported on administration mode with children aged 4–18 years [68, 70–91, 96] (see Table 4). The majority compared paper and pencil PROMs with an identical computerised version. Most studies showed moderate to strong correlation between paper and computerised versions [71, 75, 76, 81, 83, 84, 87–89, 91]. All studies that asked preference for mode showed preference for computer-based measures [71–73, 78, 81, 87, 91]. Sensitive subjects such as stress, coping, alcohol and tobacco use were more likely to be reported using web-based measures in children 8–18 years [70, 74, 78, 79]. One study showed that those under 8 years needed help completing a computerised measure [96]. There was fewer missing data with computerised measures. It was not always clear whether this was due to the inability to move on until a question was completed [75, 82, 85]. Strong factorial invariance was found across telephone, face to face and mail [86], and computer and telephone methods were also shown to be strongly correlated [90].

Discussion

This review provides evidence that CYP over 5 years old can meaningfully report on aspects of their own health, providing consideration is given to age, response format and recall period. CYP as young as 4 years old expressed a preference for completing measures regarding their health via a computerised method.

To self-report health-outcomes, children must have at least a rudimentary self-concept and ability to express this, understand the basic notions of health and illness, be able to pay attention, discriminate between the response options, recall health experiences and write a response [92]. Until

4–5 years old, children's language and thought processes are limited, so their ability to go through these process is also limited [101]. Children as young as 3 years of age were included in some of the studies in this review but results were presented alongside those of children ranging from 6 to 17 years old. The results of this review suggest that most children over five are able to reliably self-report on their health to some degree, with children younger than this exhibiting a 'yes' bias in response to questions [45].

Response format

Up until 6–7 years old, children view themselves in predominantly physical terms and their response to questionnaires is mainly dichotomous [102]. This is demonstrated in studies of 3–7-year-olds using a 3-point faces scale where only the anchors were used [26, 27]. Evidence on the ability of CYP over 7 years old to use 5- or 6-point response formats is mixed. This may be a reflection of variability in children's development, with chronological age having less of an influence than cognitive ability [5]. Difficulty with the middle of scales was found in cognitive interview studies in those 5–18 years using Likert scales [42, 44, 92, 94]. In contrast, evidence from other cognitive interview and validity and reliability studies showed that those over 8 years old can understand 5-point Likert scales [20, 34, 42, 43, 46, 95] and that children over the age of 7 years can validly and reliably use scales with six faces [16, 33, 49, 93, 96]. However, item response theory studies show that the use of 5-point Likert scales led to disordered thresholds and 3-point scales functioned better in those 8–18 years old [17, 38, 60]. As data for all ages was usually presented together, it is not possible to ascertain whether older children can reliably use a 5-point response format. The VAS was less reliable and valid than Likert or faces across the age span [18, 57] and functions better with pictorial anchors [29]. There was an overwhelming preference at all ages for faces scales, with the VAS being the least preferred, suggesting that children are motivated by visually appealing response formats. It is recommended that when developing PROMS for CYP consideration is given to making them visually appealing to improve acceptability. It is also recommended that a dichotomous response format is used for those aged 5–7 years and a 3-point response format should be considered for those seven and over. Validity of response formats should not be evaluated solely in terms of convergent and discriminant validity of the measure, as this will often be high. Cognitive interview studies should also be undertaken, to give greater insight into how response format is understood. This review found no evidence that children who had underlying health conditions, were able to more reliably use any of the response formats described than their healthy peers.

Recall period

Evidence on recall period is limited, with only 11 studies reporting on this. These suggest that recall period should be kept to 24–48 h for those under 8 [92, 93]. Those over 8 years old are able to respond reliably to events that occur over the past 7–14 days [66, 67, 92, 95]. It is recommended that when developing PROMs for CYP the recall period is kept to no more than 48 h for those under 8 years. From 8 years old CYP seem to be able to recall the past 14 days, but due to data being presented for wide age ranges is unclear from what age CYP may be able to recall further than this.

Administration mode

Online and paper-and-pencil response formats demonstrated moderate to strong correlation [71, 75, 76, 81, 83, 84, 87–89, 91], similar to findings in adults [103] and there was an overwhelming preference for a computerised format [71–73, 78, 81, 87, 91]. Sensitive questions are more likely to be answered honestly in a computerised measure, probably as this method of data collection is perceived as more anonymous [70, 74, 78, 79]. There was fewer missing data on computerised versions of measures, possibly because children were not allowed to move to the next question if a response was left unanswered [75, 82, 85]. Those under 8 years old may need help from an adult to complete computerised outcome measures [96]. It is recommended that PROMS developed for CYP of all ages include a computerised version to enhance acceptability.

Strengths and limitations

This systematic review provides evidence of children's ability to self-report on their health outcomes in terms of recall period, response format and administration mode of measures but has some limitations. The inclusion criteria only incorporated articles published in the English language and searches were carried out in health-related databases; further evidence may be found in educational research. There were relatively few studies on recall period ($n=11$) and the effects of cognitive ability rather than chronological age ($n=2$) which highlight areas for future research. This review identified 13,215 articles for screening, another eight were included as a result of hand-searching and communication with experts. The assessment of recall period, response format and administration mode was a small part of these studies and as such, was not included in the paper keywords. The quality of included studies was poor in some instances which could have affected the reported results. These were included as it is often not possible to assess which aspects were addressed but not reported in the published paper. This is particularly relevant

for older studies that were published before current reporting guidance was developed. Sample size was sometimes small, but it is well known that recruiting to paediatric research, particularly when this includes children with an underlying health condition, can be challenging [104]. A large number of studies were researching pain focused measures, rather than having a multi-dimensional focus.

Most included studies did not stratify their results by age, presenting data for wide age ranges. This makes it impossible to distinguish variation in ability by age group. As cognitive ability usually improves with age, it is recommended that when developing PROMs, psychometric testing is stratified by age and/or cognitive ability. PROM developers should also consider having different versions for different age groups or developmental ability to account for this. Future research could also take further steps to appraise the reliability of CYP self-report by using multi-indicator approaches, such as lack of response variability, excessive response variation and extreme, inconsistent or improbable response patterns, to assess invalid responses at the individual level [105].

Implications for developing PROMS for CYP.

From this systematic review we make eight recommendations for developing PROMS for CYP. These are:

1. Proxy measures should be used for those under 5 years old.
2. Measures should be visually appealing, to improve acceptability.
3. PROM studies should be analysed and reported in developmentally appropriate age bands.
4. Developers should consider different versions of a measure for different age groups.
5. Development should include both cognitive interview studies, and psychometric testing to enhance understanding of how children formulate answers.
6. 5–7 years olds should be given a dichotomous response format; those 7 years and over should be given a three-point response format.
7. Recall period should be kept short, no more than 48 h for those 5–7 years.
8. PROMS should have a computerised version.

We propose that these recommendations are used alongside the COSMIN and Rothrock [14, 106] guidance on PROM development and validation.

Conclusion

Development of PROMS for CYP is complex and challenging due to diversity in developmental stage and cognitive ability. Children < 5 years old are unable to reliably report on their own health outcomes. Children < 8 years old cannot accurately recall beyond the past 48 h and can only reliably use a dichotomous response format. Children find visually appealing measures, in a computerised format more acceptable to use. Future work should focus on the impact of cognitive ability on self-report in CYP, reporting results of validation studies in smaller age ranges and establishing whether CYP with underlying health conditions are more able to report on their own health outcomes than their healthy peers. The results of this review have both clinical and research implications. They can be used to inform appropriate choice of PROM in the clinical setting. Our eight recommendations for developing PROMS for CYP can be used to further research in PROM development for CYP.

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Data availability The data that supports the findings of this review are available in the supplementary material.

Declarations

Conflict of interest There are no conflicts of interest to declare.

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Supplementary Appendix 1. Full search strategy

Children

1 exp child/
2 exp p?ediatrics/
3 (child* or adolescen* or p?ediatric* or youth* or juvenile or teen* or young people or schoolchild* or school age* or kid*).ti,ab.
4 1 or 2 or 3

Response Scale format

5 (response scale or likert scale or visual analog* scale or VAS or numerical rating scale or verbal rating scale or faces scale or dichotomous scale or yes no response or response option*).ti,ab.

Recall period

6 (recall period or recall interval or patient recall or recall bias).ti,ab.

Method of administration

7 (outcome measure adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.
8 (measure adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.
9 (scale adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.
10 (questionnaire adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.
11 (survey adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.
12 7 or 8 or 9 or 10 or 11

Combine the above

13 14 or 15 or 21
14 4 and 13 and 22

Exclusion criteria(21)

24 (addresses or biography or comment or directory or editorial or interview or festschrift or lectures or legal cases or legislation or letter or news or newspaper article or patient education handout or popular works or congresses or consensus development conference or practice guideline).pt
25 23 not 24
26 (limit to 1980-current; humans; English language; all child 0-18 years).

Supplementary data 2. QualSyst Scores for Quantitative Studies

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Baxter (2011)[16]	1	1	1	1	NA	NA	NA	2	2	1	2	0	2	2	68
Bender (2007) [68]	2	2	1	1	1	2	NA	2	1	2	2	0	2	2	77
Benson (2016)[17]	0	2	1	1	NA	NA	NA	1	1	1	2	NA	2	1	60
Berntson (2001)[18]	2	2	2	0	NA	NA	NA	2	0	2	2	0	1	2	68
Borgers (2003)[19]	2	1	2	1	NA	NA	NA	1	2	1	2	NA	2	2	75
Borgers (2004)[20]	2	1	2	1	NA	NA	NA	1	2	1	2	NA	2	2	80
Campbell (2011)[21]	1	2	0	0	NA	NA	NA	2	0	2	2	0	2	2	59
Castarlenas (2013)[22]	2	2	1	1	NA	NA	NA	1	2	2	0	NA	2	2	75

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Castarlenas (2015)[69]	2	2	1	1	NA	NA	NA	2	2	2	2	1	2	0	77
Chambers (1998)[23]	2	1	1	0	1	NA	NA	2	1	1	2	0	2	2	63
Chambers (1999)[24]	2	1	1	1	1	NA	NA	1	1	2	2	2	2	2	75
Chambers (2005)[25]	2	2	1	1	1	NA	NA	2	2	2	2	1	2	2	83
Chogle (2012)[61]	2	2	2	1	NA	NA	NA	2	1	0	2	0	2	1	68
Decruynaere (2009)[26]	2	2	1	1	NA	NA	NA	1	1	2	NA	0	2	2	70
Eaton (2010)[89]	2	2	1	0	NA	NA	NA	2	2	2	2	2	2	1	82
Emmott (2017)[27]	2	2	2	2	NA	0	NA	2	2	1	2	2	2	2	88

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Fanciullo (2007) [28]	2	2	1	1	NA	NA	NA	2	1	1	2	NA	1	2	75
Fouladi (2006)[70]	2	0	1	1	0	NA	NA	1	2	1	2	NA	2	1	65
Fritz (1994)[29]	2	2	1	1	NA	NA	NA	2	1	1	2	1	2	2	77
Geerdink (2009)[71]	1	1	1	1	1	NA	NA	2	1	1	2	NA	1	1	59
Gharaibeh (2002)[30]	2	2	2	1	NA	NA	NA	1	1	1	0	NA	1	1	60
Goodenough (1997)[31]	1	1	1	1	0	NA	NA	1	1	1	0	NA	2	1	45
Gulur (2009)[58]	2	1	1	1	NA	NA	NA	2	1	1	2	NA	1	0	60
Heyer (2014)[62]	1	2	2	2	NA	NA	NA	2	1	2	2	1	2	2	86
Hicks (2001)[32]	1	1	0	0	NA	NA	NA	1	1	2	2	NA	2	2	60

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Hunter (2000)[33]	1	1	1	0	NA	NA	NA	2	0	1	1	0	2	2	50
Jensen (2010)[72]	1	1	1	1	0	NA	NA	1	1	1	1	NA	1	1	45
Jones (2010)[73]	1	1	1	1	0	NA	NA	1	1	1	2	2	1	1	54
Jung (2018)[35]	2	2	1	1	NA	NA	2	1	1	1	2	NA	1	1	68
Keck (1996)[36]	2	1	1	0	NA	NA	NA	1	1	1	0	NA	1	1	50
Klassen (2015)[60]	2	2	1	2	NA	NA	NA	1	2	2	2	NA	2	2	90
Knight (2007)[74]	1	1	1	2	0	NA	NA	1	1	2	2	2	2	1	67
Lawford (2001)[37]	1	2	1	0	NA	NA	NA	1	1	1	2	NA	2	2	65
Leske (2015)[38]	1	2	1	2	NA	NA	NA	1	2	2	2	NA	1	2	80

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Lloyd (2011)[75]	2	1	2	1	NA	NA	NA	2	2	1	2	1	1	1	72
Locker (2007)[39]	2	2	1	1	NA	NA	NA	2	1	2	2	NA	2	2	85
Luffy (2003)[57]	2	1	0	0	NA	NA	NA	2	1	1	0	NA	2	1	50
Magnus (2016)[90]	2	1	1	2	1	NA	NA	2	2	1	2	NA	2	1	77
Maïano (2009)[40]	2	0	2	0	NA	NA	NA	2	2	2	2	NA	2	2	80

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Mangunkusumo (2005)[76]	2	2	1	1	1	NA	NA	1	2	1	2	NA	2	2	77
Mangunkusumo (2006)[77]	2	2	1	1	1	NA	NA	1	2	2	2	NA	2	2	82
Mauz (2018)[78]	2	2	1	1	1	NA	NA	1	1	2	0	2	2	2	71
McCabe (2005)[79]	1	1	1	1	1	NA	NA	1	1	1	0	2	1	1	55
McGrath (1996)[55]	2	1	1	0	NA	NA	NA	2	1	1	2	NA	2	2	70
Miro (2004)[41]	2	1	1	0	0	NA	NA	1	1	1	2	0	1	1	46

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Moskowitz (2004)[80]	2	1	1	2	1	NA	NA	2	1	0	2	2	2	1	77
Nitikman (2017)[81]	2	1	1	1	2	NA	NA	1	0	1	2	1	2	1	63
Ogden (2008)[44]	1	1	1	1	NA	NA	NA	2	0	1	2	NA	1	1	55
Okanda (2010)[45]	1	1	1	0	NA	NA	NA	1	1	2	2	NA	2	0	**55**
Okupa (2013)[63]	1	2	1	2	1	1	1	1	1	1	2	1	2	2	68
Pagé (2012)[56]	2	2	0	2	2	NA	NA	2	1	1	2	NA	2	2	82
Raatt (2007)[82]	2	2	1	2	2	NA	NA	2	2	2	2	2	2	2	96
Raat (2007)[83]	2	2	1	2	2	NA	NA	2	2	2	2	2	2	2	96

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Robles (2015)[84]	2	2	1	2	1	NA	NA	2	2	2	2	0	2	2	83
Self (2015)[64]	2	2	1	0	NA	NA	NA	0	1	2	2	NA	2	2	70
Shields (2003)[47]	2	2	1	1	NA	NA	NA	1	1	2	2	NA	2	2	80
Shields (2005)[48]	2	2	1	2	1	NA	NA	2	0	2	2	2	2	2	83
Stanford (2006)[49]	1	1	0	1	NA	NA	NA	2	1	1	2	NA	2	2	65
Staphorst (2017) [50]															
Sun (2015)[91]	2	2	1	1	1	NA	NA	1	1	1	1	1	2	2	67
Tesler (1991)[51]	1	1	1	1	NA	NA	NA	1	2	1	2	NA	1	2	65

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Trapl (2013)[85]	1	1	1	1	1	NA	NA	1	1	1	2	2	2	1	63
van der Brink(2001)[65]	2	1	0	0	NA	NA	NA	0	2	1	1	NA	1	2	50
van Laerhoven (2004)[59]	2	1	1	1	0	NA	NA	2	1	2	0	NA	2	1	59
Varni (2009)[86]	2	1	1	1	0	NA	NA	2	2	2	NA	0	2	2	75
von Baeyer (2013)[52]	2	1	1	1	1	NA	NA	2	1	2	2	NA	1	1	68
Watson (2006)[53]	2	2	1	1	NA	NA	NA	1	2	2	2	2	2	2	86
West (1994)[54]	2	1	1	1	NA	NA	NA	1	1	1	0	NA	1	1	50
Wood (2011)[87]	2	2	1	2	2	NA	NA	1	1	2	2	2	2	2	88

Author	Objective	Design	Subject selection	Subject characteristics	Random allocation	Investigator blinding	Subject blinding	Outcome measures	Sample size	Analysis	Estimate of variance	Confounding	Reporting of results	Conclusion	Score (%)
Young (2009)[88]	2	2	1	2	2	NA	NA	2	1	2	2	2	2	1	88

Supplement 3 – QualSyst scores for qualitative studies

Author	Objective	Design	Context	Connection to literature	Sampling	Data collection	Analysis	Credibility	Conclusions	Reflexivity	Score (%)
Gupta (2016) [96]	1	2	1	1	1	2	1	0	2	0	55
Irwin (2009) [95]	2	2	2	0	2	2	1	0	2	0	65
Jacobson (2015) [67]	2	2	2	1	1	1	2	0	2	1	70
Joffer (2016) [34]	1	1	1	1	1	2	2	2	1	0	60
Klassen (2015) [60]	2	1	2	0	1	2	2	0	1	0	55
Morley (2014) [42]	2	2	2	0	1	2	1	2	1	0	65
Ogden (2008) [44]	1	1	2	1	1	1	1	0	1	0	45
OSullivan (2014) [43]	2	1	2	0	1	2	2	0	2	0	60
Ortqvist (2012) [46]	2	2	2	0	1	2	2	2	1	0	70
Ravens-Sieberer (2014) [66]	2	2	1	1	0	1	1	2	2	1	65
Rebok (2001) [92]	2	2	2	1	1	2	2	0	2	0	70
Staphorst (2017) [50]	1	2	2	1	0	1	2	2	2	0	65
Tomlinson (2019) [93]	1	2	2	1	1	1	2	1	1	0	60

Vreeman (2014) [94]	2	1	2	1	1	2	1	2	2	0	70

4.3 Summary

This chapter presents the results of a systematic review aiming to evaluate the evidence on response format, recall period and administration mode required to enable children and young people to validly and reliably self-report on their own health. The results have informed the development of C-POS, as outlined in the rest of this thesis. The evidence strongly suggests that children and young people under the age of 5-years-old are unable to self-report on their own health, and therefore a proxy-version of the measure is required for this population. It also highlights that children and young people prefer measures with visually appealing response formats.

This systematic review also highlights several knowledge gaps in the existing literature. Only eight of the 81 included articles recruited children and young people with life-limiting and life-threatening conditions (301, 305, 324, 328, 336-339), and of these only two recruited participants with non-malignant conditions (328, 340). Although the data suggests that ability to self-report on health does not differ between healthy and unwell children, this needs further exploration in relation to those with life-limiting and life-threatening conditions, due to the small number of articles including such participants. In addition, many children and young people with life-limiting and life-threatening conditions have cognitive impairments because of their condition. Only two papers (254, 341) included in this review included children and young people with cognitive impairment, highlighting another gap in the literature.

The next chapter in this thesis, along with results from cognitive testing of C-POS (Chapter 8) will aim to address some of the gaps in knowledge with regards to response format, recall period and administration mode highlighted above in children and young people with life-limiting and life-threatening conditions and thus inform development of C-POS.

Chapter 5 Children and young people's perspectives on measure design (objective i)

5.1 Introduction

This chapter aims to further address phase 1, objective i) of this thesis, by identifying children and young people's preferences regarding design and administration of C-POS (Figure 5-1). Data on these elements collected as part of the semi- qualitative interview study conducted in phase 1 are presented in accepted paper format. A summary of the workshop conducted with the young person's advisory group aiming to establish relevance, comprehensibility and acceptability of different response formats, administration modes and recall periods used in PROMs designed for children and young people is also presented.

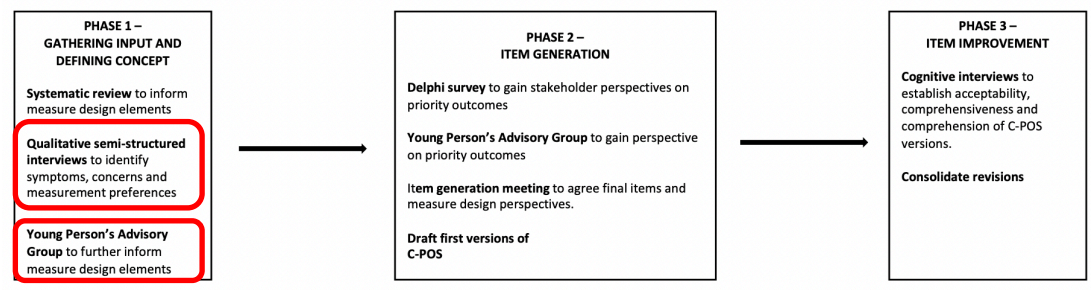

Figure 5-1 Graphic depicting where Chapter 5 fits into the overall study.

5.2 Design and administration of patient-centred outcome measures: the perspectives and preferences of children and young people with life-limiting/life-threatening conditions and their family members [paper 2].

5.2.1 Statement of contribution [publication 2]

RH wrote the initial protocol for this study. I was responsible for amendments to the initial protocol, leading on and conducting data collection, leading on and conducting data analysis, interpretation of the data and preparing the manuscript for publication. Other authors contributions: data collection: DB and AR; data analysis: DB, AR, DH and HS. RH, KB and CES provided supervision throughout. All authors were involved in critical review throughout the process and approved the final manuscript.

References included in publication 2: (1, 3, 10, 13, 88-90, 93, 105, 112, 122, 123, 148-150, 152, 168, 182, 212, 214, 218, 237, 279-281, 283, 304, 313, 314, 322, 324, 334, 342-361).

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ORIGINAL RESEARCH ARTICLE

Design and Administration of Patient-Centred Outcome Measures: The Perspectives of Children and Young People with Life-Limiting or Life-Threatening Conditions and Their Family Members

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Abstract

Background Self-reported health data from children with life-limiting conditions is rarely collected. To improve acceptability and feasibility of child and family-centred outcome measures for children, they need to be designed in a way that reflects preferences, priorities and abilities.

Objectives The aim was to identify preferences for patient-reported outcome measure design (recall period, response format, length, administration mode) to improve the feasibility, acceptability, comprehensibility and relevance of a child and family-centred outcome measure, among children with life-limiting conditions and their family members.

Method A semi-structured qualitative interview study seeking the perspectives of children with life-limiting conditions, their siblings and parents on measure design was conducted. Participants were purposively sampled and recruited from nine UK sites. Verbatim transcripts were analysed using framework analysis.

Results A total of 79 participants were recruited: 39 children aged 5–17 years (26 living with a life-limiting condition; 13 healthy siblings) and 40 parents (of children aged 0–17 years). Children found a short recall period and a visually appealing measure with ten questions or fewer most acceptable. Children with life-limiting conditions were more familiar with using rating scales such as numeric and Likert than their healthy siblings. Children emphasised the importance of completing the measure alongside interactions with a healthcare professional to enable them to talk about their responses. While parents assumed that electronic completion methods would be most feasible and acceptable, a small number of children preferred paper.

Conclusions This study demonstrates that children with life-limiting conditions can engage in communicating preferences regarding the design of a patient-centred outcome measure. Where possible, children should be given the opportunity to participate in the measure development process to enhance acceptability and uptake in clinical practice. Results of this study should be considered in future research on outcome measure development in children.

1 Introduction

A patient-reported outcome measure (PROM) is defined as any measure of a patient's health status, elicited directly from the patient without interpretation of the patient's response by a clinician or anyone else [1]. PROMs are standardised, validated questionnaires that are completed by patients to ascertain perceptions of their health status, perceived level of impairment, disability and well-being [2, 3]. Many palliative care patients, including children with life-limiting conditions, are too unwell or cognitively unable to self-report on their own health outcomes [4]. A measure that allows for proxy completion is required. Together PROMs and proxy-reported measures are termed patient-centered outcome measures (PCOMs) [5]. Within adult palliative care, PCOMs have been shown to improve service quality, increase referrals and lead to better symptom recognition and quality of life [4]. World-wide there are approximately 21 million children and young people (hereafter 'children') with life-limiting and life-threatening conditions (hereafter 'life-limiting') who could benefit from palliative care [5].

Extended author information available on the last page of the article

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Key Points for Decision Makers

Children with life-limiting conditions find brief measures with a short recall and visually appealing response format most relevant, acceptable and feasible for use.

Patient-reported outcome measures for children with life-limiting conditions should be available in paper and electronic formats.

To enhance acceptability of patient-centred outcome measure use in children with life-limiting conditions, they should be administered in conjunction with a face-to-face interaction with a health or social care professional.

There are currently no suitable PCOMs to measure palliative care symptoms and concerns in this population outside of sub-Saharan Africa [6, 7]. Development of a validated measure for children is needed to realise the benefits of PCOM use that have been demonstrated with adults [8].

Guidance on methodological standards and quality criteria for PROM development have been published by the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) and Rothrock [9–11]. The COSMIN standards on evaluating the content validity of PROMs consider three aspects: relevance, comprehensiveness and comprehensibility [9]. During PROM development, it is also important to ascertain acceptability and feasibility within the population it is intended for [9, 11, 12]. Attention to the preferences and ability of the target population with regard to recall period, response options/format, mode of administration, length and ease of completion and administration increases the likelihood of use and implementation in routine practice [3].

Children with life-limiting conditions are often excluded from research participation due to the presumption that it will result in undue burden [13–15]. This presumption is not supported by empirical data [16] and has resulted in very little primary evidence on symptoms, concerns and care priorities in this population [14]. Past healthcare experience may impact upon opinions of intrinsic features of outcome measures; thus, it is important to involve children with life-limiting conditions in developing a PCOM. However, much of the existing data reflects the proxy opinions of parents, carers and healthcare professionals [17].

A recent systematic review showed that evidence regarding recall period, response scale format and administration mode is largely confined to either healthy children or those with chronic or oncological conditions with a good prognosis [18]. This (albeit limited) evidence suggests that children prefer visually appealing measures, require a short recall period of a few days to a week and should be offered the option of electronic measures [18]. Children with health conditions have different conceptions of health and illness compared to their healthy peers, due to greater exposure to medical care [19], and the nature and therapeutic interventions of different diseases [20]. They may also need different considerations in order to practically and conceptually engage in measure completion. Therefore, it is important to design measures that are suitable for their use and can capture their experience.

This study is part of a programme of work to develop the Children's Palliative care Outcome Scale (C-POS), a child-centred outcome measure for use in paediatric palliative care within the UK. Previous sequential outputs include two systematic reviews (establishing the need for a new PCOM [8], identifying response formats and administration modes used in PCOMs for children [10, 21]) and primary qualitative data identifying symptoms, concerns and care priorities (the sample included children and young people, health and social care professionals, siblings, parents and commissioners) [22]. The aim of this analysis of the primary data is to identify preferences for PCOM design (recall period, response format, length, administration mode) to improve feasibility, acceptability, comprehensibility and relevance of a child and family-centred outcome measure, among children with life-limiting conditions and their family members. The results will be used to inform C-POS measure development, prior to cognitive testing and psychometric validation.

2 Methods

2.1 Study Design

A cross-sectional, semi-structured, qualitative interview study was conducted. This is reported in accordance with the Consolidated Criteria for Reporting Qualitative Studies (COREQ, see supplementary file 1) [23]. This study was conducted from a critical realist perspective, which allows the researcher to move beyond preferences shared by multiple participants, towards understanding the reasons for these preferences [24].

2.2 Setting

Participants were recruited from six hospitals and three children's hospices in England and Northern Ireland.

2.3 Sampling and Recruitment

Inclusion criteria were as follows: children and young people (5–17 years old) with any life-limiting condition [25]; parents/carers responsible for the primary care needs of a child < 18 years old with a life-limiting condition; siblings (5–17 years old) of children and young people with a life-limiting condition. Siblings were included in order to gain the perspectives of healthy children and those who live with children with life-limiting conditions. Participants did not have to be recruited as family units.

2.4 Exclusion Criteria

The exclusion criteria for children and young people were as follows: unable to communicate via an in-depth interview, use of 'draw and talk' or play methods or via their parents; spoke a language not supported by NHS translation services; currently enrolled in any other study; unable to give consent/assent.

The exclusion criteria for parents/carers and siblings were as follows: unable to give consent/assent; spoke a language not supported by NHS translation services.

Purposive sampling was used to ensure maximum variation in the key characteristics of age and diagnosis.

2.5 Data Collection

Semi-structured interviews were conducted using a topic guide (supplementary file 2) that began with rapport-building questions, followed by questions about what mattered most (symptoms, concerns and care priorities) to individuals and their family, in order to inform content validity of the C-POS. Play and drawing were offered to children to aid interviews [26]. Following this, participants were asked how we could best measure the things that mattered in terms of response format, recall period and measure administration. Participants were given examples of response formats to help frame their answers and explore their interpretation and preference of these. These included a 0- to 10-point numerical rating scale, the Wong-Baker faces scale (a series of six faces ranging from a happy face at 0 to a crying face at 10) [27], Likert scales anchored by numbers and faces [28, 29] and the pain block scale (concrete ordinal picture-based scale, shaped as toy blocks) [30]. Participants were also asked to suggest other response formats. With respect to recall period, participants were asked how far back they/their child could remember. Examples of paper and pencil, computerised or app-based administration modes were given. The aspects explored with participants are shown in more detail in Table 1.

The topic guide was reviewed by the study steering group (healthcare professionals, parents and researchers). Interviews were conducted by LC (experienced children's palliative care nurse, new to qualitative research), AR (experienced in working with children but new to qualitative research) and DB (experienced qualitative researcher). Interviewers did not have any previous relationship with participants. All interviewers received training and supervision on conducting interviews with children, including communication, and legal and ethical issues. Interviews were audio-recorded, transcribed verbatim and pseudonymised.

2.6 Data Analysis

Transcripts were analysed by LC, DH, AR, DB and HS (all female) using deductive and inductive coding (from the World Health Organisation domains of palliative care [31] and COSMIN taxonomy [9]) [32, 33]. Analysis followed the five steps of framework analysis: familiarisation, constructing a thematic framework, indexing and sorting, charting and mapping/interpretation [32–34] using NVivo software (Version 12). All researchers received training on the use of Nvivo and framework analysis. Regular meetings were held to discuss emerging themes and resolve any differences (20% of transcripts were independently coded by two researchers [32]). KB, CES and RH were consulted if needed to resolve discrepancies. Analysis was reviewed by the study steering group throughout the study.

2.7 Ethical Approval

Ethical approval was granted by the Bloomsbury research ethics committee (HRA:19/LO/0033). Participants 16 years old and over provided written informed consent. Those with parental responsibility provided written informed consent for participants < 16 years. Those < 16 years provided written or verbal assent.

3 Results

3.1 Participant Characteristics

Seventy-six interviews were conducted (April 2019–September 2020) with 79 participants: 39 children aged 5–17 years (26 living with a life-limiting condition; 13 healthy siblings) and 40 parents (of children aged 0–17 years). Two sets of parents and one set of siblings were interviewed together. International Classification of Diseases 10th Revision (ICD-10) chapter headings are reported for pseudonymity, as some children had rare conditions. Interviews were carried out face-to-face in a location of the participant's choosing, with the exception of 13 interviews that were conducted remotely via video call due to the coronavirus disease 2019 (COVID-19) pandemic [35]. No participants required the use of an interpreter. Table 2 shows participant demographic data.

Table 1 Aspects of measure design explored in interviews using COSMIN recommended measurement properties [9]

Relevance	Comprehensibility	Acceptability	Feasibility
Recall period relevant to C-POS aim of measuring symptoms and concerns	Understanding of recall periods	Measure appearance	Measure length
Response format relevant to C-POS aim of measuring symptoms and concerns	Understanding of response formats	Recall period acceptable to C-POS aim	Completion time
		Response format acceptable to C-POS aim	Type and ease of administration
		Type and ease of administration	Ability to use recall periods
		Willingness to complete a measure	Ability to use response formats

COSMIN Consensus-based Standards for the selection of health Measurement Instruments, *C-POS* Children's Palliative care Outcome Scale

3.2 Main Findings

Participants spoke about aspects of PCOM recall period, response format and measure administration that encompassed the COSMIN content validity standards of relevance and comprehensibility. They also discussed aspects of feasibility and acceptability of a PCOM designed to measure health outcomes in children with life-limiting conditions, such as length and number of questions. Table 3 shows the findings of this study mapped onto these themes.

3.3 Response Format

Children with life-limiting conditions as young as 8 years old were familiar with the numerical rating scale, and seemed to comprehend how to use this, especially in relation to pain assessment. They were also able to use the scales to report on other symptoms, such as worry and sleep.

> *"They usually ask me like 'on a scale of 1 to 10'" (Child, 10 years old, respiratory condition)*

Most children found visually appealing response formats more relevant and acceptable, predominantly the 6-point Likert faces scale. Children as young as 5 years old seemed to understand how to use scales anchored with faces. However, a small minority of teenage participants stated that numerical rating scales were more acceptable for them and felt that faces were more appropriate for younger children.

> *'[Investigator] I: 'So, these are different faces, so again the smiley face would be no hurt and then that really sad face, do you know what that would be? [Participant] P:'Umm that really hurts, and that one, that would hurt a little bit, that would hurt a little bit as well, and that would hurt a little bit more and that would hurt a whole lot….' (Child, 5 years old, gastrointestinal condition)*

> *'I think those are like more my age, and then like the faces could be like for younger kids.' (Child, 15 years old, gastrointestinal condition)*

One child felt that use of the faces scale could lead to ambiguous interpretations about how they felt. There was a concern that one could be experiencing a high level of pain or distress, but that this would not necessarily be reflected in the selected facial expression. This led to the concern that people would think your symptoms were not as bad as they were:

> *'Say I felt like…like 0 and I was like this but actually in the inside I'm 10? Mm because erm…sometimes like people could be hurting out of 10 but then people could say, it's not hurting out of 10 […]…because the guy isn't crying and like you're not exactly like the face but then you don't have to like (makes noise) squeeze out a tear or…' (Child, 10 years old, gastrointestinal condition)*

None of the participants had seen or used the colour block scale before. Only one child found it acceptable for use as it was similar to a computer game they played:

> 'I: If you had to choose one?
> *P: That one.*
> *I: The blocks? Yeah, and why do you like the blocks? Do you know why you like that one more than the others?*
> *P: Because there's something called number blocks*
> I*: Oh, do you use them at school?*
> *P: No there's a programme and it…and it…and erm… it has numbers all the way up and it keeps going up and up and I saw one what said, one hundred'*

In contrast to children with life-limiting conditions, healthy siblings were less familiar with rating scales and struggled to comprehend them, particularly those under 11

Table 2 Participant demographics

Participant-group demographics	
Children with life-limiting conditions ($n = 26$)	
Age (years), mean (range)	12 (5–17)
Gender (F:M), *n*	17:9
Diagnosis, *n*	
Congenital	3
Neurological	5
Gastrointestinal	10
Metabolic	1
Cancer	6
Respiratory	1
Interview duration (min), mean (range; SD)	37 (12–81; 19.1)
Parents/carers ($n = 40$)	
Age (years), mean (range)	40 (21–65)
Gender (F:M), *n*	30:10
Relationship to child, *n*	
Mother	30
Father	10
Diagnosis of child, *n*	
Congenital	7
Neurological	10
Gastrointestinal	4
Metabolic	9
Cancer	6
Perinatal	1
Genitourinary	1
Infectious disease	2
Age of child with life-limiting condition (years), mean (range)	12 (0–17)
Interview duration (min), mean (range; SD)	63 (33–161; 28.3)
Siblings ($n = 13$)	
Age (years), mean (range)	9 (5–15)
Gender (F:M), *n*	7:6
Diagnosis of child, *n*	
Congenital	3
Neurological	7
Gastrointestinal	2
Metabolic	1
Age of child with life-limiting condition (years), mean (range)	10 (3–16)
Interview duration (min), mean (range; SD)	26 (8–37; 10.2)

F female, *M* male

years old. Those older than 11 were more able to use common rating scales.

> *'The sad faces are for not important and the angry faces for so not important' (Sibling, 7 years old, of a child with a neurological condition)*
> *'May be ask like erm…what's worrying you on a scale of 1–10 how is…how is this making you feel. Erm, how much are you worrying about this problem?' (Sibling, 12 years old, of a child with a neurological condition)*

Parents and carers were generally in agreement with the children that visually appealing response formats would be more acceptable, relevant and comprehensible than numerical rating scales for the children. Some also expressed that older children and teenagers may find a numerical rating scale more acceptable, with emojis being suggested to anchor scales, as all children were familiar with them. One parent expressed concern about the acceptability of using traffic light colours for scales, as these are often used for behaviour management in schools:

Table 3 Main findings mapped on to COSMIN properties of content validity, feasibility and acceptability [9]

PROM design aspect	Relevance	Comprehensibility	Acceptability	Feasibility
Recall	A few days to a week so focus is on current symptoms	A few days to a week was understood by all participants	Long recall periods less acceptable, as participants did not want to dwell on past periods of ill health	Most participants were able to recall up to the past week
Response format	Visually appealing response formats most relevant	All participants understood the faces scales, pain block scale and colour scale; those ≥ 8 years could use a numerical rating scale	Visually appealing response formats were most acceptable; use of emojis to anchor Likert scales; colours less acceptable, as traffic lights often used for behaviour management in schools; colour block scale least acceptable	≥ 5 years could use faces scales; ≥ 8 years could use a numerical rating scale
Measure administration	Adults assumed computerised measures would be more relevant to CYP; some CYP preferred paper based	All participants understood how to use iPad/computerised and paper and pencil-based measures	Most participants found computerised measures acceptable; a small number expressed a preference for paper and pencil	Option for both computerised and paper and pencil measure should be given
Measure length	Not applicable	Not applicable	10 questions optimal	10 questions maximum
Completion time	Not applicable	Not applicable	5–15 min optimal	No more than 10 min, due to concerns regarding illness fatigue and attention

COSMIN Consensus-based Standards for the selection of health Measurement Instruments, *CYP* children and young people, *PROM* patient-reported outcome measure

'They use a lot of that for like autistic children and things for behaviour and at school. At [patient] and [sibling's] school, you know, their behaviour chart is green, amber and red.' (Parent of child with cancer)

3.4 Recall Period

Most children with life-limiting conditions and their healthy siblings proposed an acceptable recall period of a few days up to a week. Overall, there was no difference in recall period preference with respect to age, although some older children and teenagers stated that they could only remember the past few days, while others suggested a week or more. Some participants felt they would forget the past week:

'P: Like today one of the Drs asked how long have I been having my headaches.
I: Ok, and do you find it easy to remember about the last week?
P: No, I forget very easily.' (Child, 12 years old, gastrointestinal condition)

Some children with life-limiting conditions did not want to reflect back further than a few weeks, wanting to put the past behind them, which suggests a long recall period is less acceptable. The reasons for this included not wanting to be reminded of past periods of ill health, and a desire to have current symptoms and concerns addressed so that the focus could be on undertaking normal childhood activities such as attending school and seeing friends.

'[E]rm I suppose…the more recent is the better because, I don't know, I think sometimes people want to put like past things sort of behind them you know and move on. And because things change so much and I think most, the more recent are better because it's easier to remember and to focus on what you're going through at that stage rather than things that have happened' (Child, 14 years old, cancer).

One participant suggested that recalling salient aspects of their illness was easier than remembering usual activities of daily life, suggesting that acceptable and feasible recall periods may differ depending on the question.

'It depends on like the thing, if it's like, what I had for dinner last night or like the last night before that, I can't really remember (laughter), but then like if it's like what happens when I was ill, I can pretty much remember that exactly' (Child, 13 years old, gastrointestinal condition).

3.5 Length/Completion Time

There was broad consensus across participants that a short measure, with enough questions to elicit how a child was feeling, but not placing undue burden in terms of completion time, was most feasible. When asked to specify what length would be acceptable for a measure, ten questions was the most frequent response, with optimal completion time ranging from 5 to 15 min. The majority of participants felt that 10 min was an optimal completion time.

'I: How many questions do you think?
P: Probably like 10?' (Child, 14 years old, congenital condition)
'Yeah, I suppose for the kids then something like 10 minutes' (Mother of child with cancer)

3.6 Administration Mode

Parents overwhelmingly thought that children would find electronic modes of administration more acceptable and feasible because children are familiar with technology.

'P: What an app that you can log into or something?
I: Yeah, like what do you think would be better for the kids?
P: Well kids are more technological nowadays anyway, so that would probably suit them better.' (Father of child with a gastrointestinal condition)

Whilst this was the preference of some children, several expressed a strong preference for a paper-based measure or had no preference. Most siblings also found computerised administration modes more acceptable, with a few expressing a preference for pen and paper. The preference for pen and paper seemed to be in part because they thought they would be more likely to have someone with them during measure completion to help if they did not understand what to do:

'I: *And then if we had a questionnaire like that, do you think it would be better to give it to you on a pen and paper* or an *iPad or a laptop…?*
P: A pen and paper' (Child, 9 years old, gastrointestinal condition)
'Umm…I wouldn't really mind if I'm honest. Like writing would be a lot better because sometimes things online, you can't…you don't really understand and this is like, if someone's in front of you they'll tell you what to do then…' (Sibling, 11 years old, of child with a congenital condition)

3.7 The Need to have Someone to Talk to in Measurement

Throughout the interviews, children expressed the importance of having someone from the healthcare team to talk to during or after measure completion. There were several reasons cited for this. Some children wanted to be able to clarify potential concerns regarding comprehension and interpretation of questions in a measure. Other reasons included ensuring children were honest in answering questions and to give the child's healthcare experience a 'human and compassionate' feel. Like children with life-limiting conditions, siblings preferred to have someone to talk to about how they were feeling either in addition to choosing response options on a measure or instead of measure completion.

'I'm thinking like the patient should fill something out but then on top of that, you know have the discussion with the…with the person, the healthcare assistant or healthcare professional…erm because umm…the… when the patients filling out the form, may…the…the patient may not…either may not be erm…like fully honest or…or they may not understand the erm…the question, because that actually happens.' (Child, 17 years old, gastrointestinal condition)
'[Y]ou know like, you can't just substitute the healthcare professional for a robot, you want to kind of have that human feel, so that's…that's important because otherwise the patient may not want to say anything.' (Child, 17 years old, gastrointestinal condition)
'You could just put it in front of them and ask their opinion and just ask them to circle it and…and ask them if they wanted to expand a bit more about their… how they're feeling' (Sibling, 11 years old, of a child with a congenital condition)

4 Discussion

This study provides evidence on the acceptability, feasibility, relevance and comprehensiveness of PCOM design properties for children with life-limiting conditions. We found that children with life-limiting conditions find brief measures more acceptable and feasible to use, and shorter recall period to be acceptable and relevant. Most stated that electronic measures are more acceptable for use, although differences in preferences indicate that measures should also be available in paper and pen format. Additionally, whilst children with life-limiting conditions can comprehend numerical rating scales and use them to report on their health, most find visually appealing response formats using emotive faces more acceptable. Finally, we found that children want to

complete a measure alongside a conversation with healthcare professionals about their care needs and priorities.

The findings of this study largely support a recent systematic review on recall period, response scale format and administration mode in healthy children, and those with acute or chronic conditions with a good prognosis [18]. However, contrary to previous investigations, we found that a small number of participants had a strong preference for paper-based measures, indicating that it is important to offer various modes of administration [36–42]. Notably, this finding also conflicts with parent/carer beliefs that all children would find electronic modes of administration more acceptable, demonstrating the importance of asking children with life-limiting conditions about their own preferences and not relying exclusively on proxy reports.

Our study also highlights that children with life-limiting conditions have a desire to talk about how they are feeling directly with healthcare professionals, in addition to PCOM completion, to ensure their healthcare experience is 'human and compassionate'. This will enhance acceptability of the measure. Similarly, in adults, PCOMs facilitate patient-centred communication by providing overview and insight and by prompting discussions about topics that are important to patients [43, 44]. Our findings with children need to be considered when PCOMs are implemented into practice within this population, so that the intended goals are achieved and care is focused on the child and family and what is important to them.

Children with life-limiting conditions demonstrated more familiarity with and understanding of common rating scales compared to their healthy counterparts. This was evident in our study where healthy siblings, who were less familiar with commonly used rating scales, often struggled to comprehend them. This is likely to be because children with life-limiting conditions have more exposure to describing their health than other children when being asked about pain and other symptoms. The difference between children with life-limiting conditions and other ill children and/or healthy peers also underscores the need to understand the needs of this population and not rely on outcome measures created for use in other illness populations or healthy children.

Participants expressed variable preferences in recall period, with most stating a short recall period of between a day and a week was most relevant and acceptable. Previous studies have found that recall should be kept to 24–48 h for those under 8 years [18, 45, 46], with those over 8 years being able to reliably recall events from the past 7–14 days [18, 45, 47–49]. The difference in our findings may reflect the variability in cognitive ability among children with life-limiting conditions [14]. Some participants suggested that when it comes to recalling salient events regarding health, such as episodes of being more unwell, they can remember further back than they can for details of day-to-day life. Previous studies have reported that children found it easier to remember what had happened between specific events, such as clinic appointments [50]. This may indicate that PCOM questions regarding physical symptoms such as pain may be easier for children to respond to than questions regarding whether they were able to undertake their usual day-to-day activities. However, despite episodes of past ill health being more salient to children with life-limiting conditions, many did not want to reflect that far back. They either did not want to be reminded of a negative experience, did not think it was relevant or wanted to move on and focus on the present. This calls attention to what constitutes respondent burden and the need to address it when developing PCOMs for this population to ensure acceptability in the target population.

4.1 Strengths and Limitations

The strengths of this study include the involvement of children from the age of 5 years old, with a range of life-limiting conditions. We also included the perspectives of healthy siblings and parents. Many studies reporting on this population rely on proxy reports from parents/carers and healthcare professionals, or focus on children with a cancer diagnosis [14]. Our sample size was relatively large in comparison to other studies that include children with life-limiting conditions, and the geographical spread of participant recruitment covered several areas of the UK, across two countries. We were also able to compare the perspectives of children with life-limiting conditions with those of their healthy siblings.

Our study has some limitations. The siblings of children with life-limiting conditions were likely to have had more exposure to conversations regarding healthcare than other children of the same age, so caution should be taken in extrapolating this finding to other healthy children. Although we included children as young as 5 years in our study, relatively few children with life-limiting conditions under 8 years old were recruited, meaning the data presented cover to a greater extent the perspectives of those who were towards the older age limit. One site recruited only children with gastrointestinal diagnoses, reflected in the higher number of participants from this group. There are almost 400 different life-limiting conditions known to affect children, so not all could be included [51]. Our parent sample was predominantly female, which reflects other paediatric palliative care research studies, where fathers are under-represented [52]. No participants were recruited who required an interpreter, and data on ethnicity were not collected. Therefore, our findings may not reflect culturally diverse perspectives. Finally, some interviews with children were short due to difficulty keeping them engaged, or due to illness related fatigue, which further highlights the need for a short, brief PCOM for this population. All participants discussed some aspects of recall period, response format and administration mode in their

interviews. However, sometimes this was a small part of the overall interview as these questions were asked at the end. Therefore, the perspectives presented may be from those who were more willing or able to respond to a PCOM.

4.2 Implications for Research and Practice

This study endorses some of the findings of a recent systematic review on recall period, response format and administration mode in predominantly healthy children. Recall periods of PCOMs should be kept short due to the difficulty children have in remembering too far back, and to reduce respondent burden of having to dwell too much on periods of past ill health. This study also provides some additional insights into PCOM development in children with life-limiting conditions [18]. When developing, validating and implementing PCOMs for children with life-limiting conditions, consideration should be given to ensuring that, where possible, they are given the opportunity to discuss their responses with a healthcare professional during or soon after completion. Children need to know that their responses are seen and considered. This supports a child-centred approach whereby children are regarded as active and equal partners in their care [53]. In contrast to findings in healthy children, a choice of electronic or paper and pencil format should be given to those with life-limiting conditions where possible, to ensure acceptability and feasibility of use of PCOMs.

4.3 Next Steps

Further research is required to generate initial versions of the C-POS and demonstrate comprehensiveness, comprehensibility and acceptability using cognitive interviews. This will be followed by psychometric testing.

5 Conclusions

Children with life-limiting conditions are able to describe their health outcomes [22] and can communicate their preferences regarding PCOM design. Incorporating these preferences should improve acceptability of the measure and enhance its uptake in clinical practice [3]. Children with life-limiting conditions expressed a strong desire for the opportunity to be able to discuss their symptoms and care concerns with healthcare professionals alongside PCOM completion and valued the opportunity to report on this. Children's views and preferences should be included early on and throughout measure development to improve design and enhance valid and reliable self-report.

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Declarations

Conflict of Interest The authors have no competing interests to declare.

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Ethical Approval Ethical approval was granted by the Bloomsbury research ethics committee (HRA:19/LO/0033).

Consent to Participate Participants over 16 years old provided written informed consent. Those with parental responsibility provided written informed consent for participants < 16 years. Those < 16 years provided written assent.

Consent for Publication All authors have reviewed this version and consent to publication.

Data Availability Due to the nature of the research, supporting data are not available.

Code availability Not applicable.

Author Contributions All authors: conception and design of the work. LC, DB and AR: data collection. LC, DB, AR, DH and HS: data analysis. All authors: interpretation of data. LC and DH: draft of paper. All authors: critical review and revision of article.

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5.3 Young Person's Advisory Group

The young person's advisory group (YPAG) was held in July 2020. The aim was to establish relevance, comprehensibility, and acceptability of response formats, recall periods and administration modes in children and young people. The methods used for this are described in section 3.2.2.

Administration mode

The majority of participants expressed a preference for a computerised or app-based measure. The reasons for choosing this approach included inclusivity for those who were unable to write, feeling more comfortable answering questions virtually than face-to-face and the ease of using an app when feeling less well and unable to leave the house. One participant expressed a preference for a paper-based measure that was completed face-to-face with a healthcare professional as it was a more personal approach.

Response format

Participants suggested that using a 0-10 numerical rating scale could be challenging as the choice of number could be subjective and having ten numbers was too many to choose from. The colour analogue scale and visual analogue scale were considered too complex to use by the group.

Some participants liked the Likert-type scales but suggested that these were easier to understand if anchored with faces. It was suggested that a Likert scale should always have a clear middle option, and that three options may not be enough to choose from. Several participants suggested anchoring Likert-type scales with emojis as all children were familiar with these.

Other suggestions included children drawing how they felt, colouring in the answers or turning the measure in to a game. These suggestions were not considered to be practical for C-POS as PROMs need a clear scoring system so that changes due to health care interventions can be measured.

Recall period

Most participants agreed that a few days was an optimal recall period for children and young people to use when talking about their health. They felt that a recall of longer than a week would lead to problems if symptoms fluctuated over time.

5.4 Summary

The findings presented in this chapter, along with those presented in Chapter 4 provide evidence on feasible and acceptable recall period, response format and administration mode for C-POS. Children and young people overwhelmingly prefer visually appealing measures with a short recall period. These findings were used to inform the first versions of C-POS ready for cognitive testing.

The Young Person's Advisory Group has demonstrated that children and young people are capable of expressing their preferences on measure design and should be given the chance to participate in child-centred research, particularly as their views are not always congruent with those of adults.

The evidence presented here and in Chapter 4 demonstrates a lack of evidence on what age/developmental stage children and young people may be able to use longer recall periods and more complex response formats. It was aimed to address this gap evidence during cognitive testing of C-POS (see Chapter 8).

Chapter 6 Results – Establishing priority outcomes (objectives ii - iv).

6.1 Introduction

This chapter presents the results from phase 1 of this study, addressing objectives ii- iv):

ii. To establish child and parent priorities for outcomes of care.
iii. To establish healthcare professional and commissioner priorities for outcomes of care.
iv. To develop a list of candidate priority outcomes to be included in C-POS

This chapter also aims to address the lack of evidence on symptoms and concerns elicited directly from children and young people and those with non-malignant conditions. Results are presented in published paper format. Figure 6-1 shows where this fits in to the overall study.

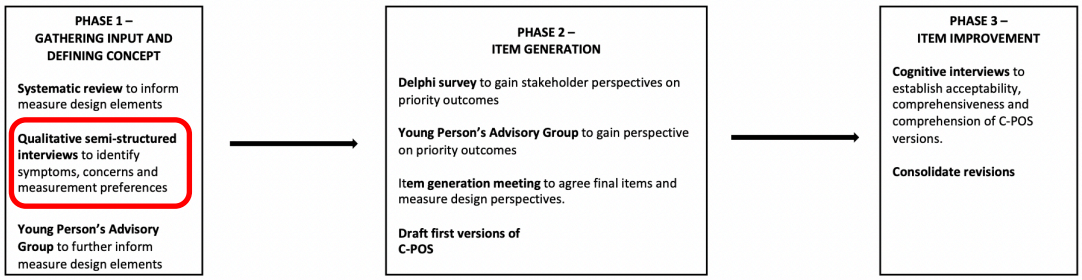

Figure 6-1 Graphic depicting where Chapter 7 fits into the overall study.

6.2 Achieving child-centred care for children and young people with life-limiting and life-threatening conditions—a qualitative interview study [publication 3]

6.2.1 Statement of contribution [publication 3]

RH wrote the initial protocol for this study. I was responsible for amendments to the initial protocol, leading on and conducting data collection, leading on and conducting data analysis, interpretation of the data and preparing the manuscript for publication. Other authors contributions: data collection: DB and AR; data analysis: DB, AR, DH and HS. RH, KB and CES provided supervision throughout. All authors were involved in critical review throughout the process and approved the final manuscript.

References included in publication 3: (1, 3, 10, 13, 14, 36, 37, 39, 78, 81, 96, 99-102, 105, 108, 152, 343, 346, 353, 355, 357, 358, 362-382).

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RESEARCH

Achieving child-centred care for children and young people with life-limiting and life-threatening conditions—a qualitative interview study

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Abstract

This study aims to identify the symptoms, concerns, and care priorities of children with life-limiting conditions and their families. A semi-structured qualitative interview study was conducted, seeking perspectives from multiple stakeholders on symptoms, other concerns, and care priorities of children and young people with life limiting and life-threatening conditions and their families. Participants were recruited from six hospitals and three children's hospices in the UK. Verbatim transcripts were analysed using framework analysis. A total of 106 participants were recruited: 26 children (5–17 years), 40 parents (of children 0–17 years), 13 siblings (5–17 years), 15 health and social care professionals, 12 commissioners. Participants described many inter-related symptoms, concerns, and care priorities impacting on all aspects of life. Burdensome symptoms included pain and seizures. Participants spoke of the emotional and social impacts of living with life-limiting conditions, such as being able to see friends, and accessing education and psychological support. Spiritual/existential concerns included the meaning of illness and planning for an uncertain future. Data revealed an overarching theme of pursuing 'normality', described as children's desire to undertake usual childhood activities. Parents need support with practical aspects of care to help realise this desire for normality.

Conclusion: Children with life-limiting conditions and their families experience a wide range of inter-related symptoms, concerns, and care priorities. A holistic, child-centred approach to care is needed, allowing focus on pursuit of normal childhood activities. Improvements in accessibility, co-ordination, and availability of health services are required to achieve this.

What is Known:

- *Existing evidence regarding symptoms, concerns, and care priorities for children with life-limiting conditions is largely limited to proxy-reported data and those with a cancer diagnosis.*
- *Child-centred care provision must be directed by children's perspectives on their priorities for care.*

What is New:

- *Social and educational activities are more important to children with life-limiting conditions than their medical concerns.*
- *A holistic approach to care is required that extends beyond addressing medical needs, in order to support children with life-limiting conditions to focus on pursuit of normal childhood activities.*

Communicated by Gregorio Milani

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Keywords Paediatrics · Palliative care · Normality · End of life care · Children · Symptom assessment

Abbreviations

Children	Children and young people
Healthcare professional	Health and social care professional
Life-limiting condition	Life-limiting and life-threatening condition
Parents	Parents and carers

Introduction

Worldwide there are approximately 21 million children and young people aged 0–19 years (hereafter 'children') with life-limiting and life-threatening conditions (hereafter 'life-limiting') [1]. Life-limiting conditions are those for which there is no hope of cure and from which children will die [2]. Life-threatening conditions are those for which curative treatment may be feasible but may fail [2].

Due to medical advances, increasing numbers of children are living with life-limiting conditions [3, 4]. However, provision of children's palliative care varies geographically, and increased prevalence has not been met with an equivalent increase in healthcare resource [3, 5].

Palliative care for adults is effective and cost-effective, reducing unplanned admissions and futile treatments [6–8], while improving quality of life, care quality, and survival [9–11]. There are almost 400 conditions that affect children for which palliative care could be beneficial [3, 12]. However, evidence for effectiveness of children's palliative care is limited in part due to a lack of a valid and reliable outcome measure [13, 14]. Development of such a measure has repeatedly been identified as a research priority [15–17]. A measure is in development in sub-Saharan Africa and Belgium, but primary data to inform measurement has not been generated outside Africa [18–20].

Outcome measure development for children with life-limiting conditions is complex due to differences in age and developmental stage, the range of conditions [12], and the role of family in care provision. To establish face and content validity, it is imperative to understand which symptoms and concerns matter the most. However, most studies focus on children with cancer [21], or rely on proxy reports of parent/carers (hereafter 'parents') or health and social care (healthcare) professionals [21]. This exclusion of children from participating in primary research directly contradicts the growing focus on children having agency, with a right to be involved in their own healthcare decisions [22, 23] as active partners in their healthcare, not passive recipients [22, 24]. This study aimed to identify the symptoms, concerns, and care priorities of children with life-limiting conditions and their families.

Methods

Study design

Semi-structured, qualitative interview study reported in accordance with the consolidated criteria for reporting qualitative studies (COREQ) [25].

Setting

Children, parents, and healthcare professionals were recruited from six hospitals and three children's hospices within three UK countries.

Commissioners were recruited via recommendations from healthcare professionals and the UK's national children's palliative care advocacy charity.

Sampling and recruitment

Inclusion criteria

Children (5–17 years) with any life-limiting condition; parents/carers with a child < 18 years old with a life-limiting condition; siblings (5–17 years) of children with a life-limiting condition; healthcare professionals with > 6 months experience of caring for children with life-limiting conditions; commissioners of UK paediatric palliative care services.

Exclusion criteria

Children: unable to communicate via an in-depth interview, using 'draw and talk' or play methods or via their parents; speaks a language not supported by NHS translation services; currently enrolled in another study; unable to give consent/assent.

Parents/carers and siblings: unable to give consent/assent, speaks a language not supported by NHS translation services.

Purposive sampling was used to ensure maximum variation in key demographics such as age and condition. Given the heterogeneity of the sample, the concept of pragmatic saturation was used to determine the required sample size in order for the dataset to have the required diversity and depth to meet the aims and objectives of the study [26].

Data collection

Semi-structured interviews were conducted using a topic guide informed by a systematic review of symptoms and concerns in children with life-limiting conditions [21] and the World Health Organisation (WHO) definition of paediatric palliative care [27]. The topic guide was reviewed by the study steering group (healthcare professionals, parents, and

researchers). Interviews were conducted by LC (experienced children's palliative care nurse, new to qualitative research), AR (new to qualitative research), and DB (experienced qualitative researcher). All interviewers received training and supervision on conducting interviews with children, including communication, legal, and ethical issues.

Interviews commenced with demographic questions and children were asked about their hobbies and interests to build rapport. Play and drawing were used to aid interviews where required. The topic guide contained an open question asking participants to describe their/their child's condition and how it affects their/their child's life. Interviews with professionals asked about the main symptoms, concerns, and care priorities of children with life-limiting conditions. Probes ensured that all domains from the WHO definition of palliative care were discussed, while allowing participants to discuss what mattered most. Interviews were audio-recorded, transcribed verbatim, and pseudonymised.

Data analysis

Transcripts were analysed by LC, DB, AR, DH, and HS using deductive (from the WHO domains of palliative care [27]) and inductive coding [28, 29]. Analysis followed the five steps of framework analysis: familiarisation, constructing a thematic framework, indexing and sorting, charting and mapping/interpretation [28–30] using NVivo software (Version 12). Using framework analysis allowed the authors to compare and contrast the findings from each theme overall and by participant group. Regular meetings were held to discuss emerging themes and resolve any differences (20% of transcripts were independently coded by two researchers). RH, KB, and CES were consulted if discrepancies could not be resolved. Analysis was reviewed by the study steering group throughout the study.

Ethical approval

Ethical approval was granted by the Bloomsbury research ethics committee (HRA:19/LO/0033). Participants over 16 years old provided written informed consent. Those with parental responsibility provided written informed consent for participants < 16 years. Those < 16 years provided written assent.

Results

Participant characteristics

A total of 103 interviews were conducted (April 2019–September 2020) with 106 participants: 26 children, 40 parents, 13 siblings, 15 health and social care professionals and 12 commissioners (see Table 1). Two sets of parents and one set of siblings were interviewed together. ICD-10-chapter headings are reporting for pseudonymity as some children reported rare conditions. Most interviews were carried out face-to-face in a location of the participant's choosing. Due to the COVID-19 pandemic, 13 interviews were conducted remotely (telephone or video call) [31].

Priority healthcare outcomes

The priority healthcare outcomes of children with life-limiting conditions and their families fitted into five themes—physical, spiritual and existential, emotional and psychological, social and practical, and pursuing normality. Table 2 shows these themes and the subthemes that comprise them. Illustrative quotes are presented in Tables 3, 4, and 5 and supplementary Table 1 (S1). Themes and subthemes were often closely inter-related.

Physical symptoms and concerns

All participants spoke of the importance of managing pain and other physical symptoms (such as seizures and infection), and the impact of multiple medical interventions. Symptom management and children being 'comfortable' was important to parents and professionals (T3Q1). Pain and other symptoms were often linked to other themes. For example, if physical symptoms were well managed, then children were more likely to be happy, have reduced anxiety, and be able to participate in normal childhood activities. Professionals discussed symptom management in relation to managing expectations of care and setting realistic goals (T3Q2). Seizures were particularly distressing and often described as difficult to manage by parents (T3Q3), sometimes being triggered by noise and over excitement (T3Q4), meaning siblings had to play quietly.

Participants from all groups spoke of the difficulties children had with eating and drinking. Some children described feeling under pressure to maintain weight (T3Q5), and others required artificial feeding. Healthy siblings spoke of feeling guilty about consuming treats in front of a sibling who was unable to eat (T3Q6).

Tiredness and fatigue were a concern for both children and parents. Parents spoke of lack of sleep and exhaustion which impacted on ability to care for their child (T3Q7). Children spoke of overwhelming fatigue causing lack of stamina and the need to take daytime naps (T3Q8).

Siblings and children with life-limiting conditions were very aware of changes in physical appearance which impacted on school attendance, seeing friends, and social activities (T3Q9).

Table 1 Demographic details of participants

	n or mean (range)
Children (*n* = 26)	
Age (yrs)	12 (5–17)
Gender	
Female:male	17:9
Diagnosis	
Cancer	6
Congenital	3
Gastrointestinal	10
Metabolic	1
Neurological	5
Respiratory	1
Interview duration (mins)	37 (12–81)
Parent/carers (*n* = 40)	
Age (yrs)	40 (21–65)
Gender	
Female:male	30:10
Relationship to child	
Mother	30
Father	10
Diagnosis of child	
Cancer	6
Congenital	7
Gastrointestinal	4
Genitourinary	1
Infectious disease	2
Metabolic	9
Neurological	10
Perinatal	1
Age of child with life-limiting condition (years)	12 (0–17)
Interview duration (mins)	63 (33–161)
Siblings (*n* = 13)	
Age (yrs)	9 (5–15)
Gender	
Female:male	7:6
Diagnosis of child	
Congenital	3
Gastrointestinal	2
Metabolic	1
Neurological	7
Age of child with life-limiting condition (yrs)	10 (3–16)
Interview duration (mins)	26 (8–37)
Health and social care professionals (*n* = 15)	
Gender	
Female:male	14:1
Profession	
*Doctor**	3
*Nurse***	7
Social worker	1
Chaplain	1
Psychologist	1
Play specialist	1
Physiotherapist	1
Interview duration (mins)	55 (38–82)
Commissioners (*n* = 12)	
Gender	
Female:male	11:1
Geographical location	
Southeast England	4
Greater London	1
East England	2
Northwest England	1
Yorkshire and Humber	4
Interview duration (mins)	53 (33–86)

*1 paediatric palliative medicine consultant, 1 haematology consultant, 1 general paediatrician

**4 palliative care nurse specialists, 1 children's community nurse, 1 hospice nurse, 1 ward sister

Spiritual and existential

Professionals spoke of lack of confidence in discussing spiritual and existential issues (T3Q10). For some patients and families, faith offered a source of comfort (T3Q11, S1Q1), whereas for others, it was a potential cause of conflict (T3Q12). Some moved more towards faith, for example, by having their child christened 'just in case' (T3Q12). Faith was also important in decisions about future care, with one participant describing how hospital policy on death registration and care of the body conflicted with her own culture (T3Q14).

Participants from all groups spoke about the uncertainty surrounding length of life (T3Q15), with children wanting to plan for their future regardless of their prognosis (T3Q16). Children were often determined to overcome and survive (T3Q17, S1Q3). Parents spoke of adjusting their hopes and dreams for a child who would be unlikely to reach typical life-course milestones (T3Q18) and questioned the meaning of illness ('why me/why my child?') (T3Q19). They expressed a desire for their child to live life as fully as possible, to their full potential, experience relationships with others, and have things to hope for and look forward to (T3Q20).

Emotional and psychological

All participants described many psychological and emotional impacts of living with a life-limiting condition. Where children had been diagnosed during childhood, rather than at birth, they spoke of an awareness of being different and having different life experiences (T4Q21). For some siblings, their experience led to desires to pursue caring careers (T4Q22), while children with life-limiting conditions sought out others with similar experiences (T4Q23).

Table 2 Inter-related domains and themes—symptoms, concerns, and care priorities ($n = 106$)

Themes	Pursuing normality	Physical	Spiritual/existential	Emotional/psychological	Social	Practical
Subthemes	Not knowing any different	Pain	Life unlived	Awareness of difference	Loneliness and isolation	Minimising hospital stays—preventing unplanned admissions, timely discharge
	Regaining normality	Other symptoms, e.g., seizures, infection, breathing difficulties, nausea, and vomiting	Religious beliefs and needs	Need to meet others the same	Access to social support	Service provision and availability, e.g., 24/7 care at home, access to respite, care continuity and co-ordination, and facilities
	Adjusting to a new normal	Management of symptoms	Hopes for and uncertainty about the future	Control and independence,	Communication and decision making (including building trust and respect, managing discord, managing goals, and expectations)	Burden and logistics of care
		Medical interventions, e.g., minor procedures, surgery, feeding tube insertion, and blood tests	Living a full life	Protecting family members	Balancing needs of family	Information needs
		Eating and drinking	Determination to overcome condition	Emotions, e.g., worry and anger sadness	Employment, housing, and financial concerns	Changing needs
		Sleep, fatigue, and tiredness	Meaning of life	Memory making and wishes	Access to technology and social media	Advance care planning
		Changes in physical appearance		Loss of self-confidence	Enjoying usual childhood activities, e.g., hobbies, play, school, and friendships	Transitions (care settings, change of school, child and adult services)
				Impact on family life	Restrictions on day-to-day life	Access to equipment
				Psychological and emotional support		
				Memory making and wishes		
				Privacy and dignity		

Table 3 Participant quotes—physical symptoms and concerns, and spiritual and existential concerns

Quote number	Quote	Participant details
Physical symptoms and concerns		
Q1	'…and that's what we live for, we just carry on for her smiles. Because she doesn't have a great value of life, this is (child's) life mainly but she is happy…erm and she's not in discomfort, so I can't really ask for anything more than that.'	Mother of an 8-year-old with a neurological condition
Q2	'So, it's about being realistic but reassuring them that we have different medications we can use for different situations and that we will continuously try and control symptoms. Obviously not promising that we can get everything under control, but we will try our hardest'	Nurse
Q3	'Now it's about trying to control seizures the best we can, we know we can't totally control them'	Mother of a 14-year-old with a metabolic condition
Q4	'P: Well, she has seizures and they're triggered easily, pretty easily…umm I: Do you know what sort of things trigger them? P: Umm…her being excited, like going to do like a sport that will trigger it, like swimming that could…'	Sibling of a child with a neurological condition
Q5	They say if you don't eat then you need a nose tube. I don't like them	11-year-old with cancer
Q6	'…erm or when we have anything from our treat box, it…I kind of feel sorry for him because he can't…he's watching us eat it and he can't eat any of 'em'	Sibling of a child with a gastrointestinal condition
Q7	'its very difficult when people say 'well can't you just put him in his wheelchair and take him for a walk round the block?' and I'm like 'I haven't slept for fourteen hours'. I don't wanna get him in his chair and take him for a walk around the block because I…I'm exhausted and it's not because I'm lazy, its because I'm physically exhausted'	Mother of 14-year-old with metabolic condition
Q8	'I get worn out a lot quicker, so I can't like run around for long or stand for long…. or like go on long walks'	14-year-old, congenital condition
Q9	'…sometimes you see like, when you…when like you're at the park or something, like you see people staring and you just think…oh honestly, I couldn't really care any less. Because if she didn't have the pipe, she'd just be a normal person and she is a normal person now. It's just that she has… medical reasons'	Sibling of a child with a congenital condition
Spiritual and existential concerns		
Q10	'I think its variable. It's um, I think sometimes it's not necessarily a question that we are very good at asking. I think it's one that we miss out on.'	Nurse
Q11	'…as I've gone through all of these…all of this and I've been in hospital…erm I always remember that, you know there as someone who suffered even worse for me and that, you know gives me peace because I know that you know I can suffer…you know I can go through all these things but nothing is gonna like keep me down and that yeah I'm always gonna continue to get back up on my feet and even if…even if something happens that, you know I'm in hospital for a very long time and things don't get better, I know that you know, that there's a greater hope and like the greater hope is in Jesus and that I trust in that. You know even whatever happens, whether you know I die or whether I live, it's for "him" and you know I'm just gonna continue to live a life according to his grace'	17-year-old, gastrointestinal condition

Table 3 (continued)

Quote number	Quote	Participant details
Q12	'I'm just thinking about parents that…that talk about usually losing their faith actually when it comes to end of life. I mean some find their faith and some lose it'	Psychologist
Q13	'I'm not godly, I don't believe that there's a higher being out there I don't believe anything like that but I'm not a hundred percent certain and I just felt it was the right thing to do because I got told that my son was gonna die. I need to get him christened just in case'	Mother of a 14-year-old with metabolic condition
Q14	'So, in [country], if you're [tribe] if someone dies, someone stays with the body until they are buried. And that is built into the system. But here if [child] was to die in hospital either after hours or a weekend or bank holiday, the body would be moved to the morgue alone and I wouldn't be able to be with him until a death certificate was issued, which can only be done by a person who works in the morgue who isn't want to be there on a bank holiday, after hours or on a bank holiday weekend. Um so we have it in our care plan that [child] is not to die in hospital.'	Mother of a 2-year-old with a metabolic condition
Q15	'They haven't told me, after the year, they don't know if I'm going to live or everyone knows what's the other, they've said they can only tell what is going to happen now.'	13-year-old, cancer
Q16	'The teenager that died recently, I mean she was still going to do her GCSE's this summer. And she died much quicker than we thought. But no, she was definitely going to still do them.'	Nurse
Q17	'…just …remember that even if I have this disease, I want to live my life normally and it will get better. I mean the treatments already started so now I will get more en…I will have more energy and I'm looking forward to just enjoying what…what is coming'	15-year-old with metabolic condition
Q18	'So, I dreamt of you know doing having the lifestyle with [child] like I'd had. Being a beach bum, you know sort of rock pooling. And you know sort of that, and you know you had all these dreams and aspirations and things. But they didn't pan out'	Mother of 10-year-old with neurological condition
Q19	'You know 'why me?' and we had a lot of anger first off, again the issue I just said 'Oh you know 'eat your veg, fruit and veg, you know you'll be big and strong' you know', 'drink lots of water because it's good for you' erm…and initially we had the "well you lied to me, why…you know why, why me. Why, what have I done wrong?"'	Father of 13-year-old child with a gastrointestinal condition
Q20	'It's I guess it's not about you know, her, her being, you know, her physical, you know if she's if she has physical issues. It's more I guess about her learning and development you know. Making sure that she can, not necessarily develop at the same pace as everyone else but she's still developing. So that you know, hopefully she can you know, she can experience love, relationships, work and you know, she has you know what we consider to be the standard things.'	Father of a 1-year-old child with an infectious disease

Table 4 Participant quotes—emotional and psychological concerns, and social concerns

Emotional and psychological concerns		
Q21	'I can't do as much as other people. I can't go out as often. You know… I can't um… go and hang out with friends or go to the town because… I get worn out quickly. And if something was to happen to me no one would know what to do.'	14-year-old with a congenital condition
Q22	'I mean she works in (child's) old school on a Saturday now, she's got a Saturday job down in err (area in London) and they said to her 'you know, what you know…is there things that you wouldn't want to do for the…' she said 'I'll do anything' she said 'you know I'll…I'll change their pads' she said "I…I will do anything that makes them happy, to get a smile out of them or to just know that I am helping them"'	Mother of a 3-year-old child with a neurological condition
Q23	'We haven't met that many with the same sort of symptoms… and I think it's good for (child) to see that… its good for us to meet other families I think'	Mother of a 15-year-old child with a neurological condition
Q24	'It's a long, sometimes painful, sometimes heart-breaking but it's an ocean of emotions that you go through. You're in this boat and it's your diagnosis with you and imagine you're in this boat, you're in this ocean of emotions and that boat is your diagnosis, the boat sometimes breaks apart but you've just, you just have help from the sunlight'	13-year-old with cancer
Q25	'It's hard work, its hard you know for the whole family. It has an effect on everybody, because everyone's trying to help and everyone's worried and you know trying to also make sure she's okay and so it is…it does, it is…it affects everybody in the family definitely.'	Mother of a 4-year-old with a congenital condition
Q26	'I: What would you say are your main care and support needs for (child)? P: For (child)… is that he's happy and safe and that he has an enriched life as much as possible'	Mother of a 12-year-old with a congenital condition
Q27	'We had a young girl who, she couldn't go to the bathroom on her own…umm at the end and she wanted the carers to take her rather than her mum and it was because she was a 14-year-old girl and she just wanted that…and her mum was very, she was a little bit upset by it initially…erm because her mum just wanted to do everything for her'	Commissioner
Q28	'I can't really have that much privacy because we don't know whether or not I'm going to have a seizure or not'	17-year-old with cancer
Q29	'….it is a bit strange just sort of often having so many people in your house. Erm, it does feel a bit of a loss of sort of privacy but, again, that's just something that we've got used to really.'	Mother of an 8-year-old with a congenital condition
Q30	'I don't always talk to my Mum, I don't like talking to her because I don't like making people upset or anything like that of how I am feeling.'	15-year-old with cancer
Q31	'And my husband did see, my husband saw [psychologist] here for a little while. But again, he found it really tricky, because he's not, he only comes in on a weekend cause he started seeing her when he was off, when she was initially ill. But he went back to work so he couldn't get up to see her.'	Mother of a 12-year-old with cancer
Q32	'One of the young people who we lost quite recently, the carers just supported mum to do things like make a memory box and just sit and read stories with the young person and it was just giving the young person and the family those memories really.'	Commissioner
Social concerns		
Q33	'It's definitely affected my social life because I spent most of the year in hospital receiving my chemotherapy and radiotherapy so I wasn't able to go to school'	17-year-old with cancer

Table 4 (continued)

Emotional and psychological concerns		
Q34	'Okay…erm so the education and the provision of education in its broader sense for children with special needs and how the cur…you know it doesn't feel like the current system is set up for children to achieve their potential. ….So, we spent an enormous amount of time ensuring that he gets the right provision in terms of education and associated therapy services, you know so physio, OT, speech and language all that sort of stuff…erm but that's a constant battle and dealing with the local authority is absolutely exhausting because they can't…don't function.'	Father of an 8-year-old with a congenital condition
Q35	'Erm so personally I found socially, I really, really felt isolated… erm for quite a long time…erm tried to find places to take him, groups to go to…'	Mother of a 3-year-old with a neurological condition
Q36	'I just miss like [pause] the environment of school and like, talking with people, because it gets lonely as well'	15-year-old with a gastrointestinal condition
Q37	'I: So, how do you manage those expectations? P: I think it's being honest. I think it's telling them what can be expected… umm that there are times when you might be a bit behind getting all these things and the reason why you will be, is about being safe but that you will get there.'	Nurse
Q38	'Um and it's difficult to trust people because, erm particularly er considering that we've had quite an adversarial relationship with our local authority at times, um then you're not always completely sort of clear erm how independent people are and who's on your side.'	Mother of an 8-year-old with a congenital condition
Q39	'It's very much a full time job for me. And I've, I had to give up my job… and I've never worked as hard as I am now.'	Mother of a 4-year-old with a metabolic condition
Q40	'Yeah, and some…and you wouldn't believe how many people I see funding stuff themselves. 'How much is this, how much is that? Do you know, if its broken, how do we get it repaired… umm it needs a service, do you…can I have the number for the service of you know the suction machines'. And I think, goodness why are you paying for this stuff yourselves?'	Mother of a 4-year-old with a congenital condition

All participants spoke of the life-altering impact of living with a life-limiting condition (T4Q24). They described anger, worry, sadness (T4Q25, S1Q4), and an overwhelming desire for children to be happy (T4Q26, S1Q5). Older children spoke of loss of privacy, control, and independence (T4Q27–28, S1Q6–7). Parents also faced a loss of privacy due to having professionals in their home, and the wish to maintain some control over their child's care and condition (T4Q29, S1Q8–9).

There was a sense of children and parents wanting to protect each other from how they were feeling, specifically around discussion of prognosis (T4Q30). Parents found accessing psychological support for themselves and siblings challenging, as this is often hospital-based and does not fit around work and school hours (T4Q31). Individuals also spoke of the importance of memory making (T4Q432).

Social concerns

Children were focused on being able to undertake usual childhood activities such as seeing friends, pursuing hobbies, and playing. School was important to parents and children for maintaining friendships, retaining a sense of normality and planning for a future by preparing for exams (T4Q33, S1Q10). Parents spoke of difficulty in accessing suitable education for their child due to complex medical needs (T4Q34). Many parents and children experienced loneliness and isolation due to absence from school and not being able to find suitable activities for their child to take part in (T4Q35–36, S1Q11). Unclear communication about symptom management goals and service availability often led to unrealistic expectations, causing discord between professionals and families. This impacted on decision-making, trust and respect, and continuity and coordination of care (T4Q37–38, S1Q12).

Table 5 Participant quotes—practical concerns and normality

Practical concerns		
Q41	'I was absolutely terrified that she'd go to hospital and either, one die in hospital which we don't want or two they do things to her that we didn't want to happen. So, I never took her to hospital, just kept her out and then when they…once they did the DNR and…and all of our wishes…erm that's when I…I felt more comfortable to be able to take her in.'	Mother of an 8-year-old with a neurological condition
Q42	'I had a parent who said to me, '(participant) you said we have a choice, we don't have a choice. The choice…the choice isn't there' and that's because a hospice refused to take a patient with a central-line and the parents did not want the sub cut line'	Nurse
Q43	'Sometimes some very…you know people just don't die overnight, children just don't die overnight or often don't die within a couple of days. They have a…you know a trajectory that's days to weeks, to months sometimes and actually, for the parents to be able to deliver, we expect parents to do a lot these days and we have more and more gaps and you know we sometimes need to plan around the fact that we don't have anybody who could go out to change a pump.'	Doctor
Q44	'I think the family stuff, they do get more concerns as they get older. When they are older and bigger its more stress and pressure… physically on the parents and carers.'	Nurse
Q45	'Umm…yeah its…its fairly frequent, yeah (wife) tends to book the…book the respite hours…err yeah and we…I mean (child's) we'll have the respite care and we'll have a long weekend, well not a long weekend…err maybe from Friday through to Sunday and that enables us to go and take (sibling) out and sort of do normal…yeah normal sort of family things… it's not often we do stuff as a four, you know a foursome, because he is so difficult to manage or take him out…'	Father of a 12-year-old with a congenital condition
Q46	'I: Out of everything what do you think matters most to you? P: Getting home.'	12-year-old with cancer
Q47	'I: And how do you feel when you're in hospital? P: Well, I'm happy because I get better, but then I'm sad because I miss school, miss my friends, miss my family, yeah'	12-year-old with a respiratory condition
Q48	'I: So, do you have any questions about your illness and how you are cared for? P: Uh, I know pretty much what happens and things like that and what will happen. So not really…'	15-year-old with cancer
Q49	'I: Is there anything else you want to tell me about when [brother] was in hospital? And what you thought, what they told you? P: Mmmm, no thank you. They didn't really tell me anything. I: What, no one told you what's going on? P: They didn't tell me what was going on, but they did tell my parents. I: Yeah. Do you wish they did tell you what was going on? P: Yup.'	Sibling of a child with a metabolic condition
Normality		
Q50	'P: No, it was um just about not caring about my condition. Just ignoring it. I: So, just ignoring your condition and do you think that's just because you want to forget about it? P: No, I don't really, I don't really care about it. I don't really let it get in my way so….'	11-year-old with neurological condition
Q51	'I was just brought up like this. I don't really remember anything different.'	14-year-old with a congenital condition
Q52	'So, I started going back to school a little bit and my mum…I just want… because I just love school. I just wanted to go back and get back to normal and everything and then my mum was like, 'okay just like do half days' and everything and I was like, 'no please let me do a whole day'. I was like (laughter) begging her to do it'	13-year-old with a gastrointestinal condition
Q53	'I want to be a normal person. Sure, normal is a harsh word that some people may not like using, oh my gosh I can't believe this person is using this word, but what other words could I use'	13-year-old with cancer
Q54	'I often, yeah, I do feel worried about things. I think mostly, I'm more worried about my normal… like going back to normal. I really want to just be normal, I'm just scared that the more time I spend in hospital, the less I'm normal, the less I'm gonna be like all the other kids my age, yeah'	17-year-old with cancer

Table 5 (continued)

Practical concerns		
Q55	'Erm and but back then about 18 months ago I asked her [doctor], I said, you know 'is he really poorly?'. You know I couldn't grasp it because giving him these recovery meds, it was just run of the mill, it's what we did you know. And I am thinking is he really poorly? And she [doctor] said- 'The only reason that [child] is still here is because of the amount of medication he's on'. But erm you know and her making me realise that this is not the norm you know. There aren't kids in the community having this level, kids that need this level of medication are generally in hospital.'	Mother of 10-year-old with neurological condition
Q56	'…sometimes you see like, when you…when like you're at the park or something, like you see people staring and you just think…oh honestly, I couldn't really care any less. Because if she didn't have the pipe, she'd just be a normal person and she is a normal person now. It's just that she has… medical reasons'	Sibling of a child with a congenital condition

Parents and professionals spoke of the financial impact of having a child with a life-limiting condition in terms of having to give up work, the expense of hospital stays, and self-funding equipment due to lack of availability (T4Q39–40, S1Q13).

Practical concerns

Parents and professionals were concerned with many practical aspects of care. These included care quality, advance care planning, service availability and facilities (T5Q41–42, S1Q14), the huge familial burden of care, and the logistics of managing this (T5Q43, S1Q15). The physical burden of care increased as children grew older (T5Q44). Access to respite care was essential to many parents of children without a cancer diagnosis, allowing them to have uninterrupted sleep and spend time with other children (T5Q45). Children did not share these concerns and were more interested in being at home (not hospital), being able to see their friends and carry on with their usual activities (T5Q46–47, S1Q16–17).

Parents and children felt well informed about the condition, treatment, and available services, which was considered important (T5Q48, S1Q18). Siblings often felt less well informed and not included in care (T5Q49).

Normality

The theme of normality was cross-cutting across all other themes. Children wanted to live life as normally as possible, focusing on being a child first, with their condition secondary to this (T5Q50). They described the importance of seeing friends, attending school, and making plans for the future. To achieve this, physical symptoms need to be well managed. Children with varying diagnoses described normality in different ways, with all wanting to pursue normal childhood activities. When a condition had been present since birth or soon after, children spoke of feeling normal and not knowing any different (T5Q51). Those that had been diagnosed later in childhood spoke of having to adjust to a new normal such as having carers in the home (T5Q52). Those with an uncertain prognosis, such as cancer, wanted life to return to pre-diagnosis normality and desired to be like their healthy peers (T5Q53–54). Parents who had been caring for a child with a life-limiting condition for many years had often adjusted to their child's care needs and had to remind themselves of their unique situation (T5Q55, S1Q19). Siblings spoke of seeing their unwell sibling as normal but with different needs (T5Q56, S1Q20).

Discussion

This study provides novel evidence of inter-related symptoms, concerns, and care priorities for children with a wide range of life-limiting conditions and their families, from the perspectives of multiple stakeholders (including children). This is an area of knowledge not previously well described [21]. Symptoms and concerns were broadly the same across the spectrum of life-limiting conditions, which is a finding previously reported [21, 32]. Most were evident across participant groups, except practical aspects of care, which were not a priority for children.

The concept of child-centred care encourages healthcare professionals to place the child and their interests at the centre of thinking and, where able, include them as active participants [22]. The focus of care is on the child in the context of the family, while acknowledging the child's wider environment and relationships [22, 33]. Previous studies have found that children with cancer and their families try to adjust to a 'new normal', and those with severe neurological impairment were able to regain some normality with input from a paediatric palliative care team [34–36]. Our

study adds to the concept of pursuing normality within the context of children living with life-limiting conditions, demonstrating that a child-centred approach to care needs to take an individual and holistic view of the child, ensuring that physical, emotional, social, practical, and spiritual concerns are addressed. This enables children to pursue normal childhood activities such as attending school and seeing friends. Children in our study wanted to be seen as children first, with their condition coming second to this, reinforcing that children do not want to be defined by their condition [37].

We found children wanted the opportunity to make plans for a longer-term future, even if these would not be realised, adding to the concept of pursuing normality. In contrast, a previous study found that children with neuro-disability only want to plan for the present or near future [38]. This difference may be due to the older age of the sample of participants with neurodisability meaning they had a better understanding of their condition. The heterogeneity of conditions in our study may also have contributed to our finding, as curative treatment for some life-limiting conditions is feasible, but may fail [39].

Taking a child-centred approach to care for children with life-limiting conditions needs to incorporate support for the family, while ensuring that the child remains the focus of care [40, 41]. This is important for families of children with life-limiting conditions, as this study demonstrates that they often have to provide complex, burdensome care. Many life-limited children are unable to communicate their needs due to their condition, and parents will need to advocate for their best interests. Parents require access to adequate holistic services, particularly respite care and practical support to enable them to provide care. Parents and siblings need time and space to undertake their own normal activities such as self-care, spending time as a family, and seeing friends. In our study, this was not always achieved, with insufficient or inaccessible practical, psychological, educational, and respite support often highlighted, along with lack of co-ordination and communication between services. To attempt to address this pursuit of normality and accomplish child-centred care, services need to be co-ordinated around child and family needs [40, 42], and this should be considered in the design of future health services for those with life-limiting conditions.

In our study, we found that children as young as five wanted to be informed about their condition, supporting a child-centred approach to care where the child is, where able, encouraged, and supported to be an active participant. Other studies have found that the desire to be informed about a condition is associated with adolescence, rather than younger children [21]. Siblings wanted to be informed, which is a finding previously reported in children whose parents have a life-limiting illness [43].

Strengths and limitations

As far as the authors are aware, this is one of the largest studies conducted exploring symptoms and concerns of children with a range of life-limiting conditions from multiple stakeholder perspectives. We have demonstrated that verbal children from the age of five years old are willing and able to participate in research and share their perspectives on their condition. This study's strengths include our large sample, wide range of stakeholder participants, and the range of life-limiting conditions. Fathers, who are often underrepresented in palliative care research, represented 25% of our parent sample [44].

Our study has several limitations. Recruitment took place in a small number of UK sites and data on ethnicity was not collected. One site recruited only children with gastrointestinal diagnoses, and this is reflected in the higher number of participants from this group. There are almost 400 different life-limiting conditions known to affect children, so not all could be included [12]. Many children with life-limiting conditions are non-verbal and cannot meaningfully share their perspectives and parent/proxy-reporting has to be used. The findings presented here reflect those of children who were able to participate. As a child-centred approach to care should include support for the family, care must enable them to use their knowledge and experience of their child in order to advocate for them. The child's needs and interests should always be at the centre of care and decisions [42].

Clinical and research implications

This study provides a comprehensive insight into what symptoms, concerns, and care priorities are important to children with life-limiting conditions and their families, to enable healthcare professionals to support them to be viewed as children, rather than their condition, within a child-centred model of care. We have demonstrated that children can be meaningfully involved in such studies [45]. Findings will be used to develop the construct for a valid child-centred outcome measure for use in this population.

Conclusions

Children want to focus on pursuing normal childhood activities, but need a holistic approach in addressing their care needs to achieve this. Improvements in accessibility, availability, and co-ordination of relevant health services are required.

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Declarations

Ethical approval Ethical approval was granted by the Bloomsbury research ethics committee (HRA:19/LO/0033).

Consent to participate Participants over 16 years old provided written informed consent. Those with parental responsibility provided written informed consent for participants < 16 years. Those < 16 years provided written assent.

Consent for publication All authors have reviewed this version and consent to publication.

Conflict of interest The authors declare no competing interests.

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Supplementary table 1

Quote Number	Quote	Participant details
	Spiritual and existential concerns	
Q1	"We do occasionally go up to the chapel and she's been up and stuff. And I'll often go up and spend some time on my own."	Mother of a 12-year-old with cancer
Q2	"P: Well, I do even though I'm gonna be a Police I: You're gonna be the Police, are you? Why do you want to be in the Police? P: Umm…because I want to…erm like…umm have a big job arresting people"	5-year-old with gastrointestinal condition
Q3	"With cancer you want to live, you want to show people that you can overcome this"	13-year-old with cancer
	Emotional and psychological concerns	
Q4	"It makes me feel upset because erm…when I talk about him, it's kind of like I feel… I feel like I'm the unusual one at school"	Sibling of a child with a gastrointestinal condition
Q5	"I: What would you say are your main care and support needs for (child)? P: For (child)… is that he's happy and safe and that he has an enriched life as much as possible"	Mother of a 12-year-old with a congenital condition
Q6	"And a couple of them have actually asked the parents to call an ambulance right at the end and died in hospital. I think it was because they were scared, and they didn't want their parents to have to struggle on."	Nurse
Q7	I: Do the nurses ever come and wake you up in the night? P: Yeah! I: Yeah, do you like that? P: No!	6-year-old with a gastrointestinal condition
Q8	"I think for us it's important that we live very sort of separate lives to a degree… one of us will do something with (sibling), someone does something with (child) and vice versa, so (partner) and I are never a couple, and I think yeah, that needs to be changed"	Mother of a 12-year-old with a congenital condition
Q9	"I think this year has been a big thing with her not being right and not knowing and not being able to do anything to help has been horrible, because it's been totally taken out of our hands and out of our control. So, yeah that's probably one of our biggest concerns, the not knowing and not being able to be in control of it both"	Mother of a 13-year-old with a metabolic condition
	Social concerns	
Q10	"I was crying at some of the exams because like, I'm very like, a very studious and like [pause] I like to achieve like high grades and that so, just [pause] disappointing"	15-year old with gastrointestinal condition
Q11	"P: Sometimes in the night…some…sometimes I have the separate bedroom from (sibling) and (mum) is…is three bedrooms away from mine, so I feel very alone I: Ah, so you like to be in the bedroom with (sibling)? P: Yeah"	Sibling of 8 year old with a congenital condition (talking about staying in a hospice)
Q12	"There is also the families that are quite, understandably they are going through a really stressful situation, but they can be sometimes quite obstructive and difficult as well. So I think that that sometimes impacts communication because if you're, for example, very obstructive, we have an obstructive grandmother at the moment who is there all the time and actually the nurses are really nervous about talking to her because they're not too sure what way she'll go either time, so as a result of that, that impedes the communication and also impacts patient care to some extent as well. So, I think that's a real challenge."	Nurse
Q13	"Yes and there's all the erm…hospital appointments as well, so you're driving there all the time. The one in London, and I mean if you're paying for that hotel every time as well and it's kind of…yeah a trip to London's probably and an overnight with everything surrounding and if you count driving there, because I drive to the hotel. It's probably about five hundred pounds a time. So, you have to think actually, you know you…."	Mother of a 15-year-old with a neurological condition
	Practical concerns	
Q14	"…yes when she…she had erm…a five-night telemetry…erm last time at the inpatient there and but they had just…there was a…erm kind of a bench on the side of her room so we could sleep on there. It was hot, it was hot, the air conditioning wasn't working."	Mother of a 15-year-old with a neurological condition
Q15	"We are basically sort of managing the care ourselves from home but there's a lot of emails and phone calls going around with all the doctors."	Mother of a 4-year-old with a metabolic condition
Q16	"I: And how do you feel when you're in hospital?	12-year-old with a respiratory condition

6.3 Outcomes identified from the qualitative interview study.

The findings of the semi-structured interview study described above were used to generate a comprehensive list of outcomes that were important to children with life-limiting conditions and their families (objective iv). This step was necessary as the findings presented in this chapter describe many symptoms, concerns and care priorities that were important to participants. Not all of these could be aligned with health outcomes, with participants often discussing aspects of care experience and quality. This outcome list was generated during a meeting with the scientific members of the C-POS study steering group (academics and clinical academics with expertise in paediatric palliative care and PCOM development). The list of outcomes identified that were considered for inclusion in C-POS are shown in Table 3. These outcomes have some similarities with the items on the adult integrated palliative outcome scale, particularly in relation to symptoms, anxiety and worry, and information needs (383). However, there are also differences in the outcomes that are important to children and young people and their families. These include accessing education, maintaining peer relations and participating in memory making opportunities.

Table 3 Outcomes identified in qualitative interview study.

Outcomes identified	
Pain	Financial burden of care
Having sufficient support from HSCPs	Agitation
Reducing the impact of illness on family life/burden of care	Bowel problems
Child being able to do things they enjoy	Changes to appetite and/or eating
Ability to live life to the fullest	Changes in physical appearance
Breathing and respiratory difficulties	Having spiritual needs met
Tiredness or fatigue	Changes in behaviour
Emotional impact of illness	Infections and/or impaired immunity
Being able to maintain relationships with peers	Impact of illness on cognition
Being supported/enabled to express emotions and feelings	Having cultural needs addressed
Having a plan for future care (advance care planning)	Having religious and faith needs met
Being able to take part in memory making opportunities	Cough
Having as much information as needed	Changes in consciousness
Sleeping difficulties	Changes to self-outlook
Nausea and/or vomiting	Skin concerns
Having psychological needs met	Weight changes
Having social support needs addressed	Opportunity to explore the meaning of life
Being able to access and undertake education	Being able to leave a legacy
Seizures	Low blood counts
Dystonia/muscle spasm	Setting and achieving life goals
Changes to physical function	Fertility concerns

6.4 Summary

This chapter presents findings on the symptoms, concerns, and care priorities of key stakeholders. It provides comprehensive evidence on outcomes that are relevant to the C-POS target population thus ensuring the measure will have robust content validity. An inventory of all potential C-POS items was generated from the evidence. The final items in C-POS need to capture the holistic nature of paediatric palliative care and enable children and young people to participate in normal childhood activities where possible.

As highlighted in Chapter 4 and **Error! Reference source not found.**, children and young people prefer a brief measure that does not take too long to complete in order to promote acceptability. The next step in this study is to gain stakeholder consensus on which outcomes identified in this chapter are the priority for inclusion in C-POS in order to achieve this. This is presented in Chapter 7.

Chapter 7 Results – C-POS item generation (objective v)

7.1 Introduction

This chapter presents the results of Phase 2 (objective v) of this study, which set out to establish stakeholder consensus on items to be included in C-POS, agree final items, response format and recall period and finalise the first versions of C-POS ready for cognitive testing. The paper presenting the results of this chapter has been accepted for publication in Palliative Medicine and is in press. The manuscript can be found in Appendix N Figure 7-1 shows where this work fits in to the overall study.

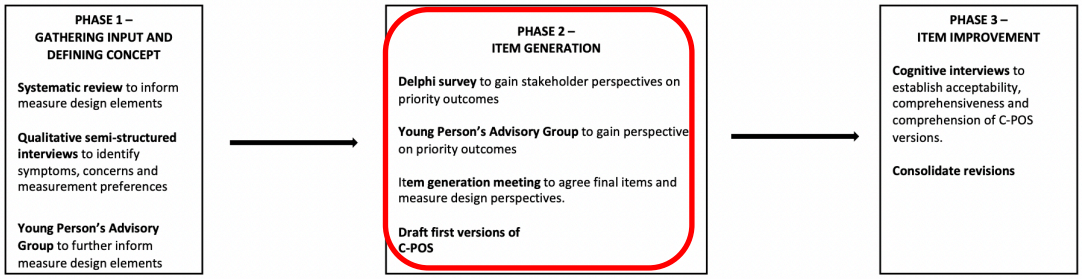


Figure 7-1 Graphic depicting where Chapter 8 fits into the overall study.

7.2 Delphi survey results

7.2.1 Round 1 - narrowing down

Eighty-two individuals participated (59 healthcare professionals, 23 parents/carers (one of which was bereaved)). See Table 4 for participant demographic details.

Table 4 Participant demographics Delphi round 1 - narrowing down

Health and social care professionals (n=59)		**Parent/carers (n=23)**	
Gender (male:female)	8:50 (1 preferred not to answer)	**Gender (male:female)**	0:23
Profession	16 Doctor 32 Nurse 1 Physiotherapist 2 Social work 4 Health care assistant 4 Counsellor/therapist	**Child's diagnosis**	4 Metabolic 5 Congenital 8 Neurological 3 Circulatory 1 Cancer 2 Genitourinary
Place of work	17 Hospital 30 Hospice 5 Community 7 Multiple settings	**Child's age years (mean; range)**	8.9 (1-17)
UK region	5 England-Northeast 7 England – Southeast 6 England – Southwest 3 England – West Midlands 4 England – Yorkshire and Humber 25 England – East 2 Wales 1 England – Greater London 3 England – East Midlands 2 England – Northwest 1 Scotland	**UK region**	8 England – Southeast 2 England – Yorkshire and Humber 9 England – East 3 England – Northwest 1 Scotland
Experience years (mean; range)	11.8; 1-30	**Ethnic background**	23 white British (parent/carer) 4 mixed race: 19 white British (child)

Twenty-one outcomes were selected by over 50% of participants. In addition, two further outcomes were selected by over 50% of the healthcare professional group, and three by the parent/carer group. See Table 5 for results of the narrowing down round. Twenty-three suggestions were made for additional outcomes. Most of these were deemed to be incorporated in existing items, except for an outcome regarding siblings. This was added to the ranking rounds, along with the 26 outcomes outlined above.

Table 5 Results Delphi round 1 - narrowing down

Outcome	**Frequency n (%)**		
	Overall (n=82)	***Parent/carer (n=23)***	***HSCPs (n=59)***
Pain*	73 (89.0)	18 (78.3)	55 (93.2)
Having sufficient support from health and social care professionals*	70 (85.4)	19 (82.6)	51 (86.4)
Reducing the impact of illness on family life/burden of care*	68 (82.9)	22 (95.7)	46 (78.0)
Child being able to do things they enjoy*	68 (82.9)	22 (95.7)	46 (78.0)
Ability to live life to the fullest*	67 (81.7)	22 (95.7)	45 (76.3)
Breathing and respiratory difficulties*	63 (76.8)	14 (60.9)	49 (83.1)
Tiredness or fatigue*	62 (75.6)	19 (82.6)	43 (72.9)
Emotional impact of illness*	59 (72.0)	20 (87.0)	39 (66.1)
Being able to maintain relationships with peers*	59 (72.0)	19 (82.6)	40 (67.8)
Being supported/enabled to express emotions and feelings*	57 (69.5)	17 (73.9)	40 (67.8)
Having a plan for future care*	55 (67.1)	19 (82.6)	36 (61.0)
Being able to take part in memory making opportunities*	54 (65.9)	19 (82.6)	35 (59.3)
Having as much information as needed*	54 (65.9)	17 (73.9)	37 (62.7)
Sleeping difficulties*	53 (64.6)	12 (52.2)	41 (69.5)
Nausea and/or vomiting*	52 (63.4)	10 (43.5)	42 (71.2)
Having psychological needs met*	49 (59.8)	16 (69.6)	33 (55.9)
Having social support needs addressed*	48 (58.5)	18 (78.3)	30 (50.9)
Being able to access and undertake education*	48 (58.5)	11 (47.8)	37 (62.7)
Seizures*	45 (54.9)	10 (43.5)	35 (59.3)
Dystonia/muscle spasm*	43 (52.4)	8 (34.8)	35 (59.3)
Changes to physical function*	42 (51.2)	8 (34.8)	34 (57.6)
Setting and achieving life goals*	40 (48.8)	13 (56.5)	27 (45.8)
Financial burden of care*	38 (46.3)	19 (82.6)	19 (32.2)
Agitation*	37 (45.1)	4 (17.4)	33 (55.9)
Bowel problems*	37 (45.1)	6 (26.1)	31 (52.5)
Changes to appetite and/or eating	33 (40.2)	7 (30.4)	26 (44.1)
Changes in physical appearance	27 (32.9)	3 (13.0)	24 (40.7)
Having spiritual needs met	26 (31.7)	2 (8.7)	24 (40.7)
Changes in behaviour	25 (30.5)	9 (39.1)	16 (27.1)
Infections and/or impaired immunity*	25 (30.5)	12 (52.2)	13 (27.1)
Impact of illness on cognition	24 (29.3)	9 (39.1)	15 (25.4)
Having cultural needs addressed	21 (25.6)	0	21 (35.6)
Having religious and faith needs met	16 (19.5)	0	16 (27.1)
Cough	16 (19.5)	3 (13.0)	13 (22.0)
Changes in consciousness	15 (18.3)	3 (13.0)	12 (20.3)
Changes to self-outlook	14 (17.1)	5 (21.7)	9 (15.3)
Skin concerns	13 (15.9)	4 (17.4)	9 (15.3)
Weight changes	10 (12.2)	5 (21.7)	5 (8.5)
Opportunity to explore the meaning of life	9 (11.0)	4 (17.4)	5 (8.5)
Being able to leave a legacy	6 (7.3)	4 (17.4)	2 (3.4)
Low blood counts	5 (6.1)	4 (17.4)	1 (1.7)
Fertility concerns	4 (4.9)	1 (0.2)	3 (5.1)

* = items moved to ranking rounds (n=27)

7.2.2 Round 2 - ranking round i)

Sixty individuals participated (47 healthcare professionals, 13 parents) in ranking the 27 outcomes identified in the narrowing down round. See Table 6 for demographic details of participants in this round.

Table 6 Participant demographics Delphi round 2 - ranking round i)

Health and social care professionals (n=47)		**Parent/carers (n=13)**	
Gender (male:female)	6:41	**Gender (male:female)**	0:13
Profession	11 Doctor 28 Nurse 1 Physiotherapist 2 Health care assistant 5 Counsellor/therapist	**Child's diagnosis**	4 Metabolic 5 Congenital 3 Neurological 1 Genitourinary
Place of work	15 Hospital 25 Hospice 4 Community 3 Multiple settings	**Child's age years (mean; range)**	9.3 (1-16)
UK region	5 England-Northeast 4 England – Southeast 4 England – Southwest 3 England – West Midlands 3 England – Yorkshire and Humber 22 England – East 2 Wales 2 England – East Midlands 2 England – Northwest	**UK region**	4 England – Southeast 2 England – Yorkshire and Humber 6 England – East 1 Scotland
Experience years (mean; range)	13.2; 1-36	**Ethnic background**	13 white British (parent/carer) 3 mixed ethnic group: 10 white British (child)

Overall, there was weak agreement on ranking (Kendall's W = 0.17). There was also weak agreement between parents' rankings alone (W=0.16) and those of health and social care professionals (W = 0.21). Cohen's kappa between parents and healthcare professionals was 0.08. See Table 7 for results of ranking round i).

Table 7 Delphi results round 2 – ranking round i)

Outcome (n=27)	**Overall median rank (% ranking in top 50%) (n=60)**	**Parent median rank (% ranking in top 50%) (n=13)**	**HSCP median ranking (% ranking in top 50%) (n=47)**
Pain	5.5 (88.3)	7 (84.6)	1 (89.4)
Ability to live life to the fullest	6.5 (66.7)	5 (76.9)	5 (63.8)
Breathing and respiratory difficulties	7 (80.0)	12 (69.2)	2 (83.0)
Child/young person being able to do things they enjoy	8 (73.3)	6 (69.2)	3 (74.5)
Having sufficient support from health and social care professionals	9 (68.3)	9 (76.9)	6 (66.0)
Having a plan for future care	9.5 (68.3)	14 (61.5)	4 (70.2)
Dystonia/muscle spasms	11.5(60.0)	18 (38.5)	9 (66.0)
Being supported/enabled to express emotions and feelings	12 (58.3)	11 (53.8)	10 (59.8)
Sleeping difficulties	12.5 (58.3)	12 (76.9)	12 (53.2)
Setting and achieving life goals	12.5(50.0)	13 (53.8)	19 (48.9)
Having psychological needs met	12.5 (53.3)	9 (61.5)	16 (51.1)
Nausea and vomiting	13 (58.3)	19 (23.1)	7 (68.1)
Tiredness or fatigue	13.5 (56.7)	14 (61.5)	11 (55.3)
Reducing the impact of illness on family life/care burden	13.5 (53.3)	14 (53.8)	15 (53.2)
Emotional impact of illness	14 (55.0)	11 (53.8)	14 (55.5)
Seizures	14 (56.7)	14 (46.1)	8 (59.6)
Agitation	15.5 (51.2)	20 (15.4)	13 (61.7)
Siblings being supported and having their needs met	16(38.3)	14 (61.5)	21 (31.9)
Changes to physical function	16.5 (41.2)	14 (53.8)	20 (38.3)
Bowel problems	17 (43.3)	19 (23.1)	18 (48.9)
Having as much information as needed	17 (48.3)	17 (46.2)	17 (48.9)
Being able to maintain relationships with peers	18 (36.7)	15 (46.2)	23 (34.0)
Being able to take part in memory making opportunities	19.5 (33.3)	20 (30.8)	22 (34.0)
Financial burden of care	20 (25.0)	15 (46.2)	25 (19.1)
Infections and/or impaired immunity	20 (26.7)	19 (38.5)	24 (23.4)
Having social support needs addressed	20.5 (23.3)	17 (38.5)	26 (19.1)
Being able to access and undertake education	22.5 (26.7)	22 (38.5)	27 (59.6)
Kendall's W	**0.1671**	**0.1595**	**0.2053**

7.2.3 Round 3 - ranking round ii)

Thirty individuals participated in round three (26 healthcare professionals and four parents) and the 27 items ranked in the previous round were ranked again. See Table 8 for demographic details of participants.

Table 8 Participant demographics Delphi round 2 - ranking round ii)

Health and social care professionals (n=26)		**Parent/carers (n=4)**	
Gender (male:female)		**Gender (male:female)**	0:4
Profession	9 Doctor 14 Nurse 1 Physiotherapist 1 Health care assistant 1 Counsellor/therapist	**Child's diagnosis**	1 Metabolic 1 Congenital 1 Neurological 1 genitourinary
Place of work	9 Hospital 11 Hospice 4 Community 2 Multiple settings	**Child's age years (mean; range)**	12.0 (2-16)
UK region	3 England-Northeast 2 England – Southeast 1 England – Southwest 2 England – West Midlands 1 England – Yorkshire and Humber 12 England – East 2 Wales 2 England – East Midlands 1 Greater London	**UK region**	1 England – Southeast 1 England – Yorkshire and Humber 2 England – East
Experience years (mean; range)	13.3; 1.5-36	**Ethnic background**	4 white British (parent/carer) 1 mixed ethnic group: 3 white British (child)

Agreement between participants was moderate (Kendall's W = 0.61). There was also moderate agreement between the healthcare professional group alone (W=0.68) and parent group alone (W = 0.64). Cohen's kappa between parent and health and social care professionals was 0.13 (poor agreement). See Table 9 for results of this round.

As Kendall's W had increased from weak to moderate agreement it was decided to stop the study at this point. This was due to concerns regarding potential gain and the feasibility of another round given the attrition of parent/carer respondents between the two ranking rounds.

7.3 Younger Person's Advisory Group

Twenty-two children (17 female; six male) aged 10-21 years attended the meeting. The responses given by two groups are shown in Table 9. Both groups suggested naming the C-POS versions after planets to avoid any stigma associated with using chronological age on a measure designed for use in a population with a high variation in ability.

Table 9 Delphi results round 3 - ranking round ii) and responses from young person's group

Outcome	**Overall median rank (% ranking in top 50%) (n=30)**	**Parent median rank (% ranking in top 50%) (n=4)**	**HSCP median rank (% ranking in top 50%) (n=26)**	**Times item selected in top 5 by older children and young people in YPAG (11 representatives)**	**Item selected by younger children and young people in YPAG in overall top 13 (11 representatives)**
Pain	1 (90.0)	9.5 (50.0)	1 (96.2)	7	Yes
Ability to live life to the fullest	2 (96.7)	1.5 (100)	2.5 (96.2)	3	Yes
Breathing and respiratory difficulties	3 (96.7)	6.5 (100)	3 (96.2)	2	Yes
Child/young person being able to do things they enjoy	4 (96.7)	4 (100)	4 (96.2)	5	Yes
Having sufficient support from HSCPs	5 (93.3)	5.5 (75)	5 (92.3)	3	No
Having a plan for future care	6 (90.0)	9.5 (25)	6 (92.3)	1	No
Dystonia/muscle spasms	8 (76.7)	20 (25)	7 (84.6)	0	No
Being supported/enabled to express emotions & feelings	9 (80.0)	8 (100)	9.5 (76.9)	2	No
Sleeping difficulties	10.5 (86.7)	10.5 (75)	10.5 (88.5)	3	No
Having psychological needs met	10.5 (76.7)	9.5 (100)	11 (73.1)	5	No
Nausea and vomiting	12 (76.7)	17 (50)	11.5 (80.8)	1	Yes
Setting and achieving life goals	12 (73.3)	8.5 (100)	12 (69.2)	1	No
Tiredness or fatigue	13 (80.0)	5.5 (100)	13 (76.9)	3	No
Reducing the impact of illness on family life/care burden	14.5 (50.0)	13.5 (75)	15.5 (46.2)	2	Yes
Agitation	16 (36.7)	20 (0)	16 (42.3)	0	No
Seizures	16 (36.7)	16.5 (0)	16 (42.3)	0	Yes
Emotional impact of illness	16 (23.3)	10.5 (75)	17 (15.4)	3	Yes

Siblings being supported and having their needs met	18 (16.7)	22 (0)	18 (19.2)	0	No
Changes to physical function	19 (16.7)	10.5 (75)	19 (7.7)	2	Yes
Having as much information as needed	20 (13.3)	20.5 (0)	20 (15.4)	0	No
Bowel problems	20.5 (23.3)	20.5 (0)	20.5 (26.9)	0	No
Being able to maintain relationships with peers	22 (13.3)	22 (0)	22 (15.4)	5	Yes
Being able to take part in memory making opportunities	23 (13.3)	16 (25)	23 (11.5)	0	Yes
Infections and/or impaired immunity	24 (6.7)	20.5 (25)	24 (3.8)	1	No
Financial burden of care	25 (20.0)	24.5 (25)	25 (19.2)	0	No
Having social support needs addressed	26 (10.0)	21.5 (25)	26 (7.7)	0	No
Being able to access and undertake education	27 (6.7)	23.5 (0)	27 (7.7)	1	Yes
Kendall's W	**W=0.61**	**W=0.68**	**W = 0.64**	**-**	**-**

7.4 Item generation meeting

Twenty-two members attended the item generation meeting – nine paediatric palliative care clinicians, six research team members, five clinical academics with expertise in PCOM development and two bereaved parents. After the initial presentations, each domain from our qualitative interview study was discussed and potential C-POS items were mapped onto these (122, 238). Previous work had suggested children's care priorities differed from parents, particularly regarding practical aspects of care. It was agreed that C-POS would have self-report items regarding children's symptoms and concerns, and separate questions for parents to answer regarding family concerns (238). It was further agreed that there would be proxy versions of the measure for parents to answer on behalf of their child if they were unable to respond themselves. Proxy versions would contain the same items as the self-report versions.

Five versions of the measure were drafted, each with eight questions about the child and five about the family: (1) parent/carer of child less than two years old, (2) parent/carer of child 2 years old and over, (3) child five to seven years old, (4) child 8-12 years old and (5) young person 13-18 years old. The intention was that the child/young person and their family would choose the version most suitable to their ability to complete. The number of items was informed by previous work which suggested that children should have 10 items or fewer to respond to (384). These versions were named after planets (Mercury, Saturn and Neptune), as suggested by the young person's advisory group. Items were the same across versions but were worded differently in consideration of age/ability. For example, using the term 'hurt' rather than 'pain'. Recall period and response format were based on previous evidence, with shorter recall and a three-point Likert scale for younger/less able children, and a longer recall and five-point Likert scale for older/more able children (237, 384). The Likert scales on the child versions were anchored with emojis. Table 10 shows domains and agreed items for C-POS.

Due to the number and heterogeneity of life-limiting conditions (10), ensuring suitability of all items for the entire population proved challenging. Several physical symptoms (e.g., dystonia and breathing difficulties) were prioritised in the Delphi survey, but not all children with life-limiting conditions experience these. Only pain was common across the population. Hence a decision was taken to have a generic question regarding symptoms other than pain. The item regarding siblings was not

relevant to all families, so a question regarding the impact of the child's condition on the family was worded to incorporate relevant family members.

Table 10 Mapping of agreed C-POS items onto domains from previous qualitative interview study and systematic review(122, 385)

Child self-report items	
Domain	**Question item**
Physical	Pain Other symptoms
Social and practical	Being able to ask questions Being able to undertake usual activities
Emotional and psychological	Worry Sharing feelings Being able to do things you enjoy
Spiritual/existential	Being able to do things you enjoy Living life to the fullest
Parent/carer items	
Physical	Getting enough sleep
Social/practical	Access to information about child's condition Support needed to care for child Support to plan for future care
Emotional/psychological	Impact of child's condition on family
Spiritual/existential	Support to plan future care

7.5 Initial versions of C-POS

The outcome of the paper presented above was the generation of five versions of C-POS ready for cognitive testing. The wording of the items in these versions is shown in Table 15 . More evidence was required to inform recall period and response format for children eight years old and over, which is explored more in Chapter 8. A decision was made at this stage to name the child self-report C-POS versions after planets (Mercury, Saturn and Neptune).

Table 11 Initial C-POS items

Items regarding the child or young person				
Mercury version **Child 5-7 years (or equivalent ability)**	**Saturn Version** **Child 8-12 years (or equivalent ability)**	**Neptune version** **Young person 13-17 years (or equivalent ability)**	**Proxy version A** **Parent/carer of child < 2years**	**Proxy version B** **Parent/carer of child > 2 years**
How much have you hurt yesterday and today?	How much have you hurt ...?	How much have you been affected by pain..?	How much has your child been affected by pain in the past week?	How much has your child been affected by pain over the past week?
How much have you had other problems with your body yesterday and today?	How much have you had other problems with your body ....?	How much have you been affected by other problems with your body..?	How much has your child been affected by other symptoms in the past week?	How much has your child been affected by other symptoms in the past week?
Have you felt worried yesterday or today?	How much have you worried ...?	How much have you felt worried ...?	Has your child cried more than usual over the past week?	How much has your child worried over the past week?
How much have you been able to do the things that are fun yesterday and today?	How much have you been able to do the things that you enjoy/are important to you...?	How much have you been able to do the things that you enjoy/are important to you...?	How much has your child been able to do the things that they enjoy in the past week?	How much has your child been able to do the things that they enjoy/are important to them in the past week?
Have you been able to enjoy your life as much as possible yesterday and today?	How much have you been able to live your life to the fullest...?	How much have you been able to live your life to the fullest...?	How much has your child been able to reach their full potential in the past week?	How much has your child been able to live life to the fullest in the past week?
Have you been able to talk to people about how you feel yesterday and today?	How often have you had the opportunity to share your feelings..?	How often have you had the opportunity to share your feelings..?	Has your child been able to express their feelings in the past week?	How often has your child had the opportunity to share their feelings in the past week?
Have you been able to ask questions about your illness yesterday or today?	How often have you been able to ask the questions you wanted to...?	How often have you been able to ask the questions you wanted to...?	How often has your child been able to communicate their needs in the past week?	How often has your child been able to ask the questions they wanted to in the past week?
Have you been able to do the things you normally would yesterday and today?	Have you been able to do the things you usually would ...?	Have you been able to do the things you usually would ...?	How often has your child been able to do the things they usually would in the past week?	How often has your child been able to do the things they usually would in the past week?
Items regarding the family				
Have you had as much information as you wanted about your child's illness over the past week?				
Have you had the support you needed to care for your child over the past week?				
How often has you child's illness impacted family members during the past week?				
In the past week, to what extent have you felt supported in planning for your child's future care?				
Have you been able to get enough rest and sleep over the past week?				

7.6 Summary

This chapter presents the results from phase 2 (objective v) of this study, the item generation stage of C-POS development. Items included in the initial C-POS versions demonstrate robust content validity, relevance, and comprehensiveness within the target population. The next step in this study is to cognitively test the five versions of C-POS for relevance, comprehensiveness, comprehensibility, and acceptability within the target population.

Chapter 8 Results – Cognitive testing of initial C-POS versions (objective vi).

8.1 Introduction

This chapter presents the results of Phase 3 (objective vi) of this study. The aim of objective vi was to cognitively test the five versions of C-POS described in Table 15 with children and young people and their parents/carers to ensure relevance, comprehensiveness, comprehensibility, feasibility, and acceptability. Figure 8-1 shows where this fits into the overall study.

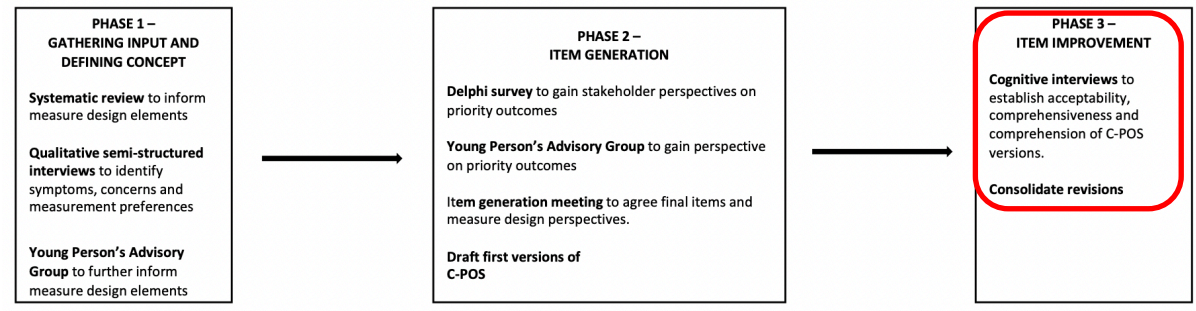


Figure 8-1 Graphic depicting where Chapter 9 fits into the overall study.

C-POS version selection for child participants was guided by developmental age, allowing the child and their parent to choose the most appropriate version. The systematic review presented in Chapter 4 demonstrated challenges in ascertaining at what chronological or developmental age children can reliably use longer recall periods and more complex response formats (237). In all children eight years old and over (or equivalent ability), both a three- and five-point response scale format was tested, along with recall periods of the past week, and yesterday and today (

Figure 8-2). Children under eight years old (or equivalent ability) were given a three-point response format and a recall of yesterday and today. The use of emojis to anchor response formats was based on evidence presented in chapters Chapter 4 and Chapter 5, with a smiling emoji representing the positive outcome for the item (237, 384). All child versions of C-POS had eight questions (Table 11).

Parent/proxy versions of C-POS had a recall period of the past week and a 5-point Likert response format (Never, Almost never, Sometimes, Often, All of the time). Parent/proxy versions contained eight questions about the child (identical concepts to child self-report items), and five about the family (Table 11).

Never	Sometimes	Most of the time
☐	☐	☐
😀	😐	☹️

Never	Almost never	Sometimes	Often	Most of the time
☐	☐	☐	☐	☐
😄	🙂	😐	😕	☹️

Figure 8-2 Response formats tested with children and young people

8.2 Cognitive testing of the Children's Palliative Outcome Scale (C-POS) with children and young people and their parent/carers.

8.2.1 Sample characteristics

Forty-eight individuals (36 parents; 12 children) participated in cognitive interviews between June 2021 and April 2022 (see Table 12 for demographics). Diagnoses are classified according to ICD-10 chapter headings to preserve participant anonymity, as many life-threatening conditions are rare (11).

Table 12. Sample characteristics

Parent/carer of child testing < 2years C-POS version (n=10;)

Gender	7 female: 3 male
Age (years) (mean; range)	34.9 (30-41)
Age of child (months) (mean; range)	26.1 (2.5-108)*
Diagnosis of child	1 Cancer 5 Congenital 1 Metabolic 3 Neurological
Ethnic background	2 Asian/Asian British 8 White British
Length of interview (minutes) (mean; range)	74.2 (37-144)
Parent/carer of child testing > 2years C-POS version (n= 26)	
Gender	22 female: 4 male
Age (years) (mean; range)	45.4 (32-60)
Age of child (years) (mean; range)	10.7 (2-17)
Diagnosis of child	4 Cancer 6 Congenital 3 Metabolic 13 Neurological
Ethnic background	1 Black/Black British 1 Other 24 White British
Length of interview (minutes) (mean; range)	60.6 (25-121)
Children testing 5- 7 years C-POS version(n=3)	
Gender	1 female: 2 male
Age (years) (mean; range)	11 (7-16)*
Diagnosis	1 Congenital 2 Neurological
Ethnic background	3 White British
Length of interview (minutes) (mean; range)	32.7 (26-42)
Child testing 8-12 years C-POS version (n=6)	
Gender	6 female
Age (years) (mean; range)	11.5 (10-13)*
Diagnosis	2 Cancer 1 Congenital 3 Neurological
Ethnic background	1 Other 5 White British
Length of interview (minutes) (mean; range)	38.0 (13.5**-83)
Young person testing 13-17 years C-POS version (n=3)	
Gender	1 female; 2 male
Age (years) (mean; range)	15.0 (14-16)
Diagnosis	1 Cancer 1 Congenital 1 Neurological
Ethnic background	3 White British
Length of interview (minutes) (mean; range)	53.5 (39.0-69.5)

*Version was tested according to ability not chronological age; ** one participant only completed three questions and was too unwell to continue.

8.2.2 Main findings

C-POS was tested over two to seven rounds dependent on version. Interview results and subsequent changes made to C-POS are displayed in Table 13 and Table 14. All

participants were able to participate in the cognitive interview process after the practice task. Some children under eight years old needed direction and explanation from a parent during the first few questions. Understanding of the ‘think aloud’ task then improved. Participants less than 8 years old (or equivalent ability) frequently needed a verbal explanation of measure items prior to responding.

Findings related to all versions.

In round one participants responded to items in terms of frequency alone, so quantifiers (how much, how often) were removed from the stem of questions so that they began with ‘Have you been affected by’…'. In subsequent rounds, participants responded in terms of how much they had been affected by a symptom or concern.

The original response format in the child versions was amended so that 'most of the time' became 'always'. Children felt this fitted with 'never' at the other end of the scale and wanted definitive always and never response options.

8.2.3 Child and young person versions

The child versions of C-POS were tested over two rounds.

Comprehension. In round one, two items posed comprehension problems, and these were amended and tested in round two. Children less than 12 years old (or equivalent ability) did not understand the term 'live life to the fullest'. This was amended to 'enjoy life as much as possible'. Those less than eight years old (or equivalent ability) found difficulty understanding the term 'sharing feelings'. This was changed to 'been able to talk to people'. These changes were understood well.

> *'I: And so, when we ask about sort of live your life to the fullest, what are you thinking about in those things?*
>
> *P: I don't know.' (13-year-old with a congenital condition)*

Retrieval. All participants 13 years old and over (or equivalent ability) could recall the past week and were able to respond to items using this recall period. They also preferred this option to 'yesterday and today'.

> *'I think again it [yesterday and today] isn’t a long enough timescale so I do prefer the past week' (15-year-old with a neuromuscular condition)*

Retrieval ability varied in eight to twelve year olds (or equivalent ability), with some only being able to recall yesterday and today, and some responding to things that had happened since the start of the week or weekend, rather than the past seven days. Those under eight years old sometimes struggled with the concept of yesterday, reporting only on the current day. No changes were made to the recall period as intention is in practice to use the version most appropriate for the child's developmental ability.

Judgement. Participants under eight years old (or equivalent ability) initially needed some support from their parent to formulate a response. The ability to respond independently improved as they moved through the measure. The questions regarding 'usual activities' and 'things you enjoy' posed difficulties for some participants. There was uncertainty about whether the benchmark for responding should be: activities undertaken pre-diagnosis, current activities, or activities they were able to undertake before the COVID-19 pandemic.

> *'Like, have you been able to do the things that you usually would, before having your disease? Or like, as in, like you, that you usually do, with your disease, but you've just had another thing happened.' (12-year-old with cancer)*

Response. All participants 13 years old and over (or equivalent ability) could use a five-point response format and expressed preference for this option over the three-point response format.

> *'Um, because there's more options. Easier to find one that… makes sense.' (14-year-old with cancer)*

There was variability in those eight to twelve years old (or equivalent ability), with some being able to use a five-point response format and some managing better with a three-point format.

Participants were able to describe in which circumstances they would choose specific response options, suggesting understanding of how to use these:

> *'Because, in my week, I get, I have Monday to Friday of radiotherapy, so that's every single day, and then I get Saturday and Sunday off. So then I think, I get like, a good few days of doing things that I really enjoy. But then, if I think about yesterday or today, that could be a Sunday or a Saturday, and I would say, Yeah, that's often. Then I would say, if it was a Tuesday or Wednesday, I would say sometimes.' (12-year-old with cancer)*

Acceptability. All children found the measure content and number of questions acceptable and all questions were reported to be important. The emojis used to anchor the Likert response scales were well-liked and made it easier to select a response option.

> *'All of them [the questions] were important to me.' (7-year-old with a congenital condition)*

> *'Thank you. It's been good. It's been good to have someone ask questions that are, like, asking about how I actually feel about everything. Not just, are you in pain? Do you need some paracetamol?' (12-year-old with cancer)*

8.2.4 Parent/carer versions

The CPOS version for parents of children two years old and over was tested over seven rounds; the version for parents of children less than two years old over four rounds.

Comprehension. Most questions were understood by participants, with two posing comprehension problems. When responding to the questions regarding 'reaching full potential' (under two years old) and 'being able to 'live life to the fullest' (two years old and over), participants compared their child to healthy children. It was intended that this question was answered within the context of their child's life-limiting condition. Amending this to 'live life to *their* fullest' across both proxy versions allowed greater comprehension. The item 'having support planning for care' was intended to ask whether participants felt supported in advance care planning decisions. However, this was interpreted to mean planning for day-to-day care needs. Amending this to 'planning for future care' allowed the item to be understood as intended.

Retrieval. Participants had no problems with retrieval. Some suggested a longer recall for the 'information needs' item as these needs are often higher at diagnosis. This was not changed as recall period is the same for all C-POS items.

Judgement. There were several issues with judgement, particularly for parents of children who are non-verbal. Terms such as 'sharing feelings' and 'asking questions' were changed to 'express feelings' and 'having appropriate information' to be more inclusive of the range of children with life-limiting conditions. Judgement difficulties were also found with the 'crying more than usual' item. Participants were unsure whether to include crying due to frustration, temper or falling over in their response. They also had difficulty deciding what 'more than usual' meant, as babies and young children cry less with age. This item was amended to 'displayed signs of worry or anxiety' which improved judgement.

> *'Well… cried, it's probably almost never, but he is getting more upset and frustrated with things. But that's, not crying, that's different. So probably almost don't. Um no, I think, if we're looking at just proper crying...' (Father of a 19-month-old with a congenital condition)*

The item 'tiredness and fatigue' also posed difficulty. These were felt to be two different concepts requiring different responses. The item was changed to 'been able to get enough sleep'.

> *'So, I think tiredness and fatigue, sometimes can mean a bit different. Like I think tiredness can, like, you could be like lack of sleep. And fatigue could just be like exhaustion....'(Mother of a 23-month-old with a congenital condition)*

Response. The 5-point Likert scale was easy to use. Some participants suggested that a 'not appropriate to my child' option was added for those who felt that their child had no understanding of, or could not articulate, concepts such as worry and information needs.

Acceptability. Measure length and number of questions was acceptable. Participants found some questions upsetting to answer (particularly 'planning future care'), however none felt any questions should be removed from the measure. They were all important to ask.

> *'You can't not ask those questions because they're important questions.' (Father of a 10-week-old with a congenital condition)*

> *'Some of them [questions] could be upsetting but that goes back to what I said, this is just an upsetting situation.' (Mother of a 2-year-old with a congenital condition)*

Several participants suggested adding an additional item regarding psychological or emotional support for the family. They felt this was not incorporated in the question regarding support needed to care for their child. This was added as question 14.

Finally, participants wanted the research team to know that when completing a PCOM about their child, they expected it to be emotive at times. Despite this, participants all questions were important and needed to be asked. They also felt that the process of cognitive interviewing made the questions more emotive than they would be in a clinical scenario.

> *'I think that anyone who's got a child in palliative care, who's agreed to be in a study about palliative care, should know that they might get a little bit upset while answering questions cos it's sad, you know?' (Mother of a 2-year-old with a congenital condition)*

'But it's because of the way that we're having to discuss our thought processing about why we're -- that is -- that -- that makes it more emotive in this scenario than it might have otherwise done' (Mother of a 2-year-old with a neurological condition)

8.3 Final C-POS versions

All versions of C-POS were tested in their final format as per COSMIN recommendations (149). Details of the final versions are shown Table 15. Recall period and response format for each version are informed by the results of cognitive testing which showed that younger children require a short recall and simple response format and children over 12 years old (or equivalent ability) can recall the past week and use a five-point response format. Children eight to 12 years old (or equivalent ability) showed variability in which recall and response format they could use, recalling the past week seemed to be easier than using a five-point response scale.

Table 13 Main findings from cognitive interviews with children and young people

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
1	Hurt (5-12 years) Pain (13-17 years)	Good comprehension in those over 8-years-old*. Younger children* understood after a verbal explanation. Question answered in terms of frequency, rather than severity or impact pain had on day-to-day life.	The majority of those >8 years* could recall the past week. Some interpreted this to mean since Monday or the start of the weekend. Some <8 years* struggled with yesterday and could only report on the current day.	Those <8 years * needed some help from a parent to integrate their thoughts into a response. Those >8 years* had no difficulties.	Some concerns that those <8 years* chose the response they thought the interviewer wanted to hear. Emojis made choosing easier. 8-12 years* showed variability in ability to use a 5-point response format. Those >13 years* all preferred and could use 5-point response format.	'Most of the time' replaced with 'All of the time.	Quantifiers removed from beginning of question (How much, how often etc) to allow severity and impact to be reported in addition to frequency.
2	Other problems with your body	Well-understood by those >8-years old*. Young children sometimes needed a verbal explanation. One participant included emotional problems.	The majority of those >8 years* could recall the past week. One <8-year-old* could only	No problems integrating thoughts into a response.	Those <8 years* could use the 3-point response format. 8-12 years* showed variability in	As above	Quantifiers as above

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
		Question answered in terms of frequency, rather than severity or impact symptoms had on day-to-day life.	remember the current day.		ability to use a 5-point response format. Those >13 years* all preferred and could use 5-point response format.		
3	Worry	Good comprehension in all participants except one 5–7-year-old*.	All those >8 years* could recall the past week. One participant <8 years* could not understand the recall period yesterday and today and discussed salient events in the recent past.	All but one participant (5-7 years*) could integrate their thoughts into an appropriate response.	As above	As above	Quantifiers as above
4	Sharing feelings	Good comprehension. One 5–7-year-old* required a verbal explanation from parent to understand the question.	All participants could retrieve the information required. All those >8 years* except one participant could recall the past week.	No problems integrating thoughts into an appropriate response.	As above	As above	Quantifiers as above 'Sharing feelings' changed to 'been able to talk to people' in 5-7 year old* version

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
5	Being able to do the things you usually would	Good comprehension.	One child <8years* could only recall the current day. One >8 years* thought back to the start of the weekend (interview was a Wednesday).	Several children >8-years-old* wanted clarity regarding whether usual things were those done currently, pre-diagnosis or pre COVID-19 pandemic.	As above	As above	Quantifiers as above
6	Being able to do things that are fun (5-7 years) Being able to do things you enjoy (8-17 years)	Good comprehension – all could explain the difference between 'usual activities' and 'fun things/things you enjoy'.	Those <8 years* could recall yesterday and today. Those >8 years* could recall the past week, although one referred back to start of the week (interview was mid-week).	No major problems integrating thoughts into a response. One participant asked same question as above regarding pre/post diagnosis and COVID-19 pandemic.	As above	As above	Quantifiers as above

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
7	Enjoying life as much as possible (5-7 years) Living life to the fullest (8-17 years)	Younger children understood 'enjoying life as much as possible'. For children aged 8-12* only half understood 'living life to the fullest'. The rest preferred the 5–7-year* question. Children > 13 years* could comprehend and explain what living life to the fullest meant to them.	One child <8 years* answered generally without relating response to required recall period. Those > 8years * could recall the past week. One 8-12-year-old* expressed a preference for a recall of yesterday and today.	No problems integrating thoughts in to a response.	As above	As above	Quantifiers as above 'Living life to the fullest' changed to 'Enjoy life as much as possible' in 8–12-year-old* version
8	Being able to ask important questions	No problems with comprehension in those >8 years*. Younger children needed a verbal explanation of what the question meant.	One <8-year-old* could only recall the current day. Those >8-years* could recall the past week.	No problems integrating thoughts into a response.	As above	As above	Quantifiers as above

* Or equivalent ability

Table 14 Main findings from cognitive interviews with parent/proxies

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
Parent/proxy items							
1	Pain	No problems	No problems	No problems. Those with children <2 years or non-verbal children could all formulate a response based on child's behavioural cues.	No problems. Answered in terms of frequency, rather than severity and distress.	Extra response option added 'Not appropriate to my child'	Quantifiers removed from beginning of question (How much, how many etc) to allow severity and impact to be reported in addition to frequency.
2	Other symptoms	No problems	No major problems. A few participants spoke of answering based on events of approximately the past week, as if your child is unwell days all merge together.	No problems arriving at a response.	No problems	As above	Quantifiers as above

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
3	Crying more than usual (<2 years) Affected by worry (>2 years)	No problems	No problems	<2 years - Participants had difficulty judging which episodes of crying to include when formulating a response i.e., crying due to frustration, tantrums, pain, falling over, hunger. Also, participants struggled to judge what 'more than usual' meant as crying often becomes less frequent as babies get older. >2years – parents of children who were non-verbal had difficulty formulating a response. This question worked much better when wording was changed to 'expressed anxiety or worry'.	All response options understood. Participants struggled to choose an option due to issues with judgement.	As above	Quantifiers as above. <2-years changed to 'displayed signs of worry or anxiety over the past week e.g., by being more irritable, sad, clingy or withdraw''. >2 years changed to 'expressed anxiety and worry'

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
4	Express feelings (<2 years) Opportunity to share feelings (>2 years)	No problems.	No problems	Some difficulty in judging which response to pick for children who were non-verbal or who had developmental delay as they could not verbally share feelings. The question worked better when it was changed to 'opportunity to express feelings'.	All response options understood. This question worked better for parents of children who could not express feelings in a meaningful way when the 'not appropriate to my child option' was added.	As above	Quantifiers as above. >2years changed to 'opportunity to express feelings'
5	Being able to do the things child usually would	One participant struggled to articulate the difference between this item and the following item about things the child enjoys doing. All other participants could describe and understand the difference.	No problems	No problems formulating a response	No problems	As above	Quantifiers as above.
6	Being able to do things child enjoys	No problems	No problems	Most participants no problems formulating a response. One participant questioned whether the item was appropriate for a 5-month-old baby and	No problems	As above	Quantifiers as above.

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
				struggled to formulate a response.			
7	Reach full potential (<2 years) Live life to the fullest (>2 years)	The majority of participants whose child was <2years understood 'full potential' to mean in comparison to a healthy child. Only two interpreted it as being in the context of their child's condition. >2 years - Participants interpreted 'live life to the fullest' as comparing their child to healthy children.	No problems	<2-year version - Some difficulty due to problems comprehending the question. Some participants were unsure what their child's 'full potential' was and whether this was related to physical development or ability. >2 years - Amending to 'live life to *their* fullest' worked better and allowed participants to talk about the child in the context of their condition.	<2 years - Response options were understood but most participants struggled to choose one due to comprehension and judgement issues. This improved when 'not appropriate to my child' as added as a response option. >2 years no problems.	As above	Quantifiers as above. Both versions changed to 'Has your child been able to live life to *their* fullest'

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
8	Communicate needs (<2 years) Ask questions (>2 years)	No problems	No problems	<2 years No problems >2 years - participants struggled to formulate an answer if their child was non-verbal. This improved when the wording was changed to 'had the appropriate information for them about their condition'.	No problems	As above	Quantifiers as above. >2years changed to 'had the appropriate information for them about their condition'
Items about the family							
9	Information about child's illness	No problem. Several participants suggested the term 'illness' should be changed to 'condition'.	No problems recalling the past week. Some participants felt that the recall period should be longer for this question, as the need for information reduces after initial diagnosis	Most participants had no issues with judgement. Two struggled to decide what type of information they should include when formulating their response e.g., medication management, condition-specific.	Most participants understood the responses. One suggested a dichotomous yes/no response option would be better.	No changes	Quantifiers as above. 'Illness' changed to 'condition'.
10	Support needed to provide care	No problems	No problems	No problems. Most participants discussed medical, psycho-emotional and practical support.	No problems	No changes	Quantifiers as above.

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
11	Planning for care	Participants interpreted this item to be about planning child's immediate care, such as respite stays and home care. When the term 'future' was added this changed to understanding it to be about anticipatory planning for the future.	No problems	No problems with judging a response. When the term 'future' was added to the question, participants spoke about advance care planning and transition.	No problems	No changes	Quantifiers as above. Changed to 'planning for future care' as question was intended to ask about advance care planning.
12	Impact of child's illness on family	No problem. Several participants suggested the term 'illness' should be changed to 'condition'.	No problems	No problems	No problems	No changes	Quantifiers as above. Term 'illness' replaced with 'condition'
13	Parent/carer tiredness/fatigue	No problems	No problems	Most participants felt fatigue and tiredness were two different concepts and should be asked about separately. Judgement was improved when question was amended to 'able to get enough sleep'.	Difficult to choose a response due to judgement issues regarding using the terms fatigue and tiredness in the same question. Response was improved when question was	No changes	Quantifiers as above. Changed to 'able to get enough sleep'.

Item number	C-POS item	Comprehension	Retrieval	Judgement	Response	Changes made to response format	Changes made to question
					changed to asking about sleep.		
14	Access to psychological and emotional support (question added to final round of cognitive testing)	No problems	No problems	Participants discussed both formal psychological and emotional support, as well as informal support from family and friends.	No problems	No changes	No changes

Table 15 Final C-POS versions

Version	Respondent	Number of questions	Recall period	Response format	Total number of participants version cognitively tested with	Number of rounds of cognitive testing	Number of participants final version tested in
A	Parent/carer of younger and non-communicative children	14 (8 about the child; 6 about the family)	Past week	5-point Likert scale + not appropriate to my child	10	4	3
B	Parent/carer of older children and those who can communicate	14 (8 about the child; 6 about the family)	Past week	5-point Likert scale + not appropriate to my child	26	7	5
Mercury	Child self-report - appropriate for children with a development age of 5-7 years	8	Yesterday and today	3-point Likert anchored with emojis	3	2	2
Saturn	Child self-report - appropriate for children with a developmental age of 8-12 years	8	Past week	3-point Likert anchored with emojis	6	2	1
Neptune	Child self-report - appropriate for children with a development age of 13-17 years	8	Past week	5-point Likert anchored with emojis	3	2	2

8.4 C-POS versions after cognitive testing

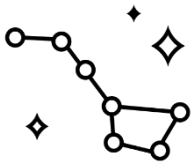
This section shows the final output of this thesis, which is five cognitively tested versions of C-POS which demonstrate face and content validity as well acceptability and feasibility within the target population. Table 16 shows the number of participants recuited across the whole of C-POS development process presented in this thesis. It demonstrates that the experiences of children and young people with a range of life-limiting and life-threatening conditions have been captured.

Table 16 Participants recruited by diagnostic category.

ICD-10 chapter heading	**Number of participants**
Cancer	21 (9 children; 12 parents)
Circulatory	3 (3 parents)
Congenital	32 (6 children; 23 parents; 3 siblings)
Gastro-intestinal	16 (10 children; 4 parents; 2 siblings)
Genito-urinary	3 (3 parents)
Infectious disease	2 (2 parents)
Metabolic	19 (1 child; 17 parents; 1 sibling)
Neurological	52 (11 children; 34 parents; 7 siblings)
Perinatal	1 (1 parent)
Respiratory	1 (1 child)

8.4.1 Mercury version

C-POS – Mercury version (5-7 years or equivalent ability)



PART A: QUESTIONS FOR CHILD ABOUT SYMPTOMS AND CONCERNS

1) Has your body hurt yesterday or today?

Never	Sometimes	All of the time
☐	☐	☐
		

2) Have you had other problems with your body yesterday or today?

Never	Sometimes	All of the time
☐	☐	☐
		

3) Have you felt worried yesterday or today?

Never	Sometimes	All of the time
☐	☐	☐
		

4) Have you been able to talk to people about how you feel yesterday or today?

Never	Sometimes	All of the time
☐	☐	☐
		

5) Have you been able to do the things that are fun yesterday or today?

Never	Sometimes	All of the time
☐	☐	☐
		

6) Have you been able to do the things that you usually would yesterday or today?

Never	Sometimes	All of the time
☐	☐	☐
		

7) Have you been able to enjoy your life as much as possible yesterday and today?

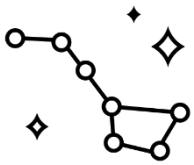
Never	Sometimes	All of the time
☐	☐	☐
		

8) Have you been able to ask questions about your illness yesterday or today?

Never	Sometimes	All of the time
☐	☐	☐
		

8.4.2 Saturn version

C-POS – Saturn version (8-12 years or equivalent ability)



PART A: QUESTIONS FOR CHILD ABOUT SYMPTOMS AND CONCERNS

1 Has your body hurt over the past week?

Never	Sometimes	All of the time
☐	☐	☐
😃	😐	😟

2) Have you had other problems with your body over the past week?

Never	Sometimes	All of the time
☐	☐	☐
😃	😐	😟

3) Have you felt worried over the past week?

Never	Sometimes	All of the time
☐	☐	☐
😃	😐	😟

4) Have you had the opportunity to share your feelings over the past week?

Never	Sometimes	All of the time
☐	☐	☐
😟	😐	😃

5) Have you been able to do the things that you usually would over the past week?

Never	Sometimes	All of the time
☐	☐	☐
😟	😐	😃

6) Have you been able to do the things that you enjoy over the past week?

Never	Sometimes	All of the time
☐	☐	☐
🙁	😐	😃

7) Have you been able to enjoy your life as much as possible over the past week?

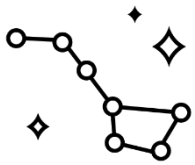
Never	Sometimes	All of the time
☐	☐	☐
🙁	😐	😃

8) Have you been able to ask the questions that are important to you over the past week?

Never	Sometimes	All of the time
☐	☐	☐
🙁	😐	😃

8.4.3 Neptune version

C-POS – Neptune version (13-17 years or equivalent ability)



PART A: QUESTIONS FOR CHILD OR YOUNG PERSON ABOUT SYMPTOMS AND CONCERNS

1) Have you been affected by pain over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

2) Have you been affected by other problems with your body over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

3) Have you felt worried over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

4) Have you had the opportunity to share your feelings over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

5) Have you been able to do the things that you enjoy over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

6) Have you been able to do the things that you usually would over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐
				

7) Have you been able to live your life to the fullest over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐
				

8) Have you been able to ask the questions that are important to you over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐
				

8.4.4 Parent carer version A (child < 2 years or equivalent ability)

PART A: QUESTIONS ABOUT YOUR CHILD

1) Has your child been affected by pain over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

2) Has your child been affected by other symptoms over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

3) Has your child displayed signs of worry or anxiety over the past week e.g., by being more irritable, sad, clingy or withdrawn?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

4) Has your child been able to express their feelings over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

5) Has your child been able to do the things they enjoy over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

6) Has your child been able to do the things they usually would over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

7) Has your child been able to live life to *their* fullest over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

8) Has your child been able to communicate their needs over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

PART B: QUESTIONS ABOUT YOU AND YOUR FAMILY

9) Have you had access to the information you needed about your child's condition over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

10) Have you had the support you needed to care for your child over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

11) Have you felt supported in planning for your child's future care over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

12) Have you had access to the emotional support you needed over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

13) Has your child's condition impacted you or your family over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

14) Have you been able to get enough sleep over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

8.4.5 C-POS parent/carer proxy-report for children 2-17 years old or equivalent ability

PART A: QUESTIONS ABOUT YOUR CHILD

1) Has your child been affected by pain over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

2) Has your child been affected by other symptoms over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

3) Has your child expressed anxiety or worry over the past week?

Not appropriate to my child*	Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐	☐

4) Has your child had the opportunity to express their feelings about their condition over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

5) Has your child been able to do the things they enjoy over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

6) Has your child been able to do the things they usually would over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

7) Has your child been able to live life to *their* fullest over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

8) Has your child had the appropriate information for them about their condition over the past week?

Never	Almost never	Sometimes	Often	All of the time	Not appropriate to my child*
☐	☐	☐	☐	☐	☐

PART B: QUESTIONS ABOUT YOU AND YOUR FAMILY

9) Have you had access to the information you needed about your child's condition over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

10) Have you had the support you needed to care for your child over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

11) Have you felt supported in planning for your child's future care over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

12) Have you had access to the emotional support you needed over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

13) Has your child's condition impacted you or your family over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

14) Have you been able to get enough sleep over the past week?

Never	Almost never	Sometimes	Often	All of the time
☐	☐	☐	☐	☐

Chapter 9 Discussion and integration of main findings

9.1 Summary of main findings and scientific contribution

This thesis presents the robust development and initial testing of a child-centred outcome measure (C-POS) for use with children and young people with life-limiting and life-threatening conditions and their families from birth up until their 18th birthday. Face and content validity, and acceptability and feasibility of use of C-POS have been demonstrated in this research. Measure content was informed by semi-structured qualitative interviews with key stakeholders (including children and young people), a systematic review to inform recall period, response format and administration mode, and work with a young person's advisory group. Item generation was informed by a Delphi survey with key stakeholders, further work with the young person's advisory group and an item generation meeting. Finally, item improvement was achieved by testing the C-POS versions with children and young people and their parent/carers in a cognitive interview study. Throughout each phase of this thesis, the COSMIN methodology for assessing the quality of studies on PROM content validity were followed (149). This has ensured robust evidence of face and content validity, comprehensiveness, and comprehensibility. Table 17 highlights how the aspects of the COSMIN criteria for good content validity were achieved within this thesis. Items 1-5 and 7 in Table 17 also contribute to the demonstration of acceptability of C-POS. Items 8-10 demonstrate feasibility within the target population.

Table 17 Aspects of thesis mapped on to COSMIN criteria for good content validity.

Relevance		
	COSMIN content validity criteria	*How aspect was achieved in this thesis*
1.	Are the included items relevant for the construct of interest?	Phase 1 - qualitative interview study (objective ii and iii). Phase 2 - Delphi survey and item generation meeting (objective v) Phase 3 - cognitive interview study (objective vi)
2.	Are the included items relevant for the target population of interest?	Phase 1 - qualitative interview study (objective ii and iii). Phase 2 - Delphi survey and item generation meeting (objective v) Phase 3 - cognitive interview study (objective vi)
3.	Are the included items relevant for the context of use of interest?	Phase 1 - qualitative interview study (objective ii and iii). Phase 2 - Delphi survey and item generation meeting (objective v) Phase 3 - cognitive interview study (objective vi)
4.	Are the response options appropriate?	Phase 1 - qualitative interview study, YPAG group meeting and systematic review (objective i). Phase 2 - item generation meeting (objective v) Phase 3 - cognitive interview study (objective vi)
5.	Is the recall period appropriate?	Phase 1 - qualitative interview study, YPAG group meeting and systematic review (objective i). Phase 2 - item generation meeting (objective v) Phase 3 - cognitive interview study (objective vi)
Comprehensiveness		
	COSMIN content validity criteria	
6.	Are no key concepts missing?	Phase 2 - Delphi survey (objective v) Phase 3 - cognitive interview study (objective vi)
Comprehensibility		
	COSMIN content validity criteria	Phase 3 - cognitive interview study (objective vi)
7.	Are the PROM instructions understood by the population of interest as intended?	Phase 3 - cognitive interview study (objective vi)
8.	Are the PROM items and response options understood by the population of interest as intended?	Phase 3 - cognitive interview study (objective vi)
9.	Are the PROM items appropriately worded?	Phase 3 - cognitive interview study (objective vi)
10	Do the response options match the questions?	Phase 3 - cognitive interview study (objective vi)

9.2 Objective i) - To determine optimal recall period, response format and administration mode for patient-centred outcome measures in children and young people.

This objective was achieved in three ways. Firstly, I conducted a systematic review that aimed to appraise the evidence on response scale type, recall period, administration mode and approaches to enable children and young people under 18 years old to participate in valid and reliable self-reporting of their health outcomes. Secondly, participants in the semi-structured qualitative interview study were asked questions on their preferences regarding recall period, response format and administration mode of C-POS. Finally, the young person's advisory group were consulted for their perspectives on optimal recall period, response format and administration mode for the measure. Using the research evidence and engagement from these three sources has enhanced the acceptability and feasibility of C-POS for children to self-report.

In order to be able to self-report on their own health outcomes, children must have an idea of self-concept and be able to express this. They must also be able to understand the basic notions of health and illness, have sufficient ability to focus on completing a measure and be able to discriminate between available response options, recall their health experiences and formulate a response (314). Evidence suggests that until the age of four to five years old, children have limited language and thought processes, so their ability to do this is limited (252).

Most of the studies included in the systematic review reported on response format, predominantly Likert type scales anchored with words, numbers, or faces. Evidence shows that most children can self-report to some degree from the age of five years old. This finding was corroborated in the semi-structured qualitative interview study and informed the lower age limit of five years old for self-completion of C-POS. Evidence in the systematic review suggested that from the age of eight years old children can use Likert scales anchored with faces (253, 282, 301, 305, 338, 386). There was mixed evidence for those under eight years old being able to do this, with some studies suggesting that children as young as three could use a six-point faces scale (263, 266). However, data in many of the eligible studies was aggregated in wide age ranges making it difficult to discern whether children across the age span C-POS is intended for could do this. As evidence for those under seven years old was limited, with some studies suggesting children score at the extremes, even with a three-point scale, a decision was made to cognitively test only a three-point format

with this age group (263, 266). Data showed mixed evidence of the ability of children eight years old and over to use a four to five-point Likert scale, with the majority of papers suggesting this was feasible. This may be due to variability in children's development at this age, with cognitive ability having more influence than chronological age (302). The findings on response format from the systematic review were largely supported by the qualitative interview study findings presented in this thesis (384). It was found that those eight years old and over were able to use 5-point Likert style faces scales. However, we found that healthy siblings in our qualitative interview study were less familiar with such scales, with sibling participants under 11 years old sometimes struggling to use a five-point response scale format. This suggests that children with life-limiting conditions have more exposure to using Likert-type scales than their healthy peers and therefore they are more able to utilise them. This may have implications if C-POS is used to measure outcomes in children and young people with a newly diagnosed life-limiting condition, as they will be less familiar with this type of response format. This will be an important consideration in the subsequent implementation and any associated guidance.

Due to the mixed evidence discussed above, both a three- and -point Likert-type scale were tested in children eight years old and over to contribute to the limited evidence base. It is important that response options are understood by respondents of PCOMs as problems could raise questions regarding the validity, reliability and responsiveness of scores (387). Visual analogue scales were found to be less valid and reliable across the age range than Likert or faces scales and the young person's group found these challenging to interpret. Other scales such as coloured analogue scales and block scales have limited evidence for use and were considered too complex to use by the young person's advisory group. Children also expressed a preference for faces and Likert scales, with participants in both our interview study and young person's advisory group suggesting that Likert scales should be anchored with emojis. Therefore, visual analogue, coloured analogue and block scales were not considered as response options for C-POS and a decision was made to use emojis to anchor the response scales.

There was limited evidence on recall period in the systematic review, with data suggesting that children over eight years old require a short recall period of no more than seven to 14 days (280, 313, 314, 338). Younger children could usually manage a recall of the past 24 hours, however some papers suggested they could only recall

events that had occurred during the current day (314, 324). Most participants in the young person's advisory group and semi-structured qualitative interview study agreed that a few days to a week was an optimal recall period, which supports the evidence from the systematic review. As C-POS is designed to capture current symptoms and concerns it was decided to test yesterday and today in those five to seven years old to try to add to the limited evidence available and enhance the usability of C-POS. A recall period of yesterday and today, and the past week were tested in those eight years old and over to add to the evidence on ability in this age, as well as acceptability and feasibility of this within a population of children with life-limiting conditions. This demonstrated that young people aged 13 years old and over (or equivalent ability) were able to recall the past week. Recall ability varied in those aged eight to twelve years old (or equivalent ability), with some able to recall the past week and some only being able to recall yesterday and today. This finding informed the decision to allow children and young people and their parents/carers to select the most appropriate C-POS version, rather than relying on chronological age to guide choice.

There was an overwhelming preference for a computerised measure over a paper and pencil one in the systematic review findings and also within the young person's advisory group. Within the qualitative interview study preference was mixed, with some child participants expressing a strong desire for a paper-based measure. This conflicted with parent/carer beliefs that all children would find electronic modes of administration more acceptable. This desire for a paper-based measure may be linked to the finding from the qualitative interview study that children and young people with life-limiting and life-threatening conditions have a desire to talk about their health outcomes directly with a healthcare professional alongside completing a PCOM. Similar findings in adult studies demonstrate that PCOMs facilitate patient-centred communication by providing overview and insight and by prompting discussions about topics that are important to patients (348, 359). Evidence suggests that correlation is strong between paper and pencil and computerised measures (265, 290, 295, 304, 312, 315, 322, 334, 335, 347). Therefore, it was decided for pragmatic reasons to develop C-POS initially as a paper-based measure with a view to developing a computerised version at a later date. Future development of a computerised version of C-POS may enhance acceptability of C-POS for some respondents.

Evidence from the systematic review suggested that different versions of C-POS would be required for different age/developmental stages, due to differences in ability to use recall periods and response formats across the age/developmental stage span. There was a clear delineation between those seven years old and under and those eight years and older in terms of ability to use more complex recall periods and response formats in both the qualitative interview study and systematic review. A decision was made at this stage of the study that those under eight years old would need a simpler C-POS version to ensure feasibility and acceptability of a self-completed PCOM.

9.3 Objectives ii and iii). To establish child, parent, healthcare professional and commissioner priorities for outcomes of care and develop a list of candidate priority outcomes

Main findings

The semi-structured qualitative interview study conducted as part of this thesis aimed to establish priority care outcomes from key stakeholders (children and young people, parents/carers, siblings, health and social care professionals and NHS commissioners). Four themes were identified across stakeholder groups - physical symptoms and concerns, spiritual/existential concerns, emotional and psychological concerns, and social and practical concerns. A fifth cross-cutting theme of pursuing normality was also identified. All themes were inter-related, with an increase in concerns in one often leading to an increase in concerns across other themes. These findings support previous work showing that symptoms and concerns in children and young people with life-limiting conditions are known to be multidimensional and burdensome, falling within the physical, psychological, social and spiritual domains, with quality of care and practical concerns also being key considerations (13). These findings are consistent with the definitions of paediatric palliative care given by Together for Short Lives and the World Health Organisation (3, 218). Similarly to previous studies, the qualitative semi-structured interview study found that there is considerable overlap of symptoms and concerns across diagnostic groups supporting the use of one patient-centred outcome measure across the range of life-limiting and life-threatening conditions experienced by children and young people (13). Due to the sample size and wide range of conditions represented it was beyond the scope of this study to conclude whether

the symptoms and concerns identified are more prevalent in certain diagnostic groups or age brackets.

Physical symptoms and concerns

Participants discussed a multitude of physical symptoms, including pain, which were considered for inclusion in C-POS, many of which have been reported previously (15-24). Unidentified and poorly managed symptoms affected all aspects of daily life for the child or young person, including psychosocial, spiritual/existential and social/practical domains. This finding reflects the notion of ‘total pain’ coined by Dame Cicely Saunders which characterises the multidimensional nature of the palliative care patient's pain experience to include the physical, psychological, social, and spiritual domains (388). By incorporating items regarding symptoms into C-POS it is anticipated that this will open a dialogue between children, families and health care professionals regarding symptom experience and management thus improving health outcomes.

Many participants expressed a desire for general symptom management and for children and young people to be 'comfortable', rather than citing specific symptoms. Participants from professional backgrounds (health and social care professionals and NHS commissioners) aligned good symptom management with setting realistic expectations of the outcome of symptom management interventions with families. Unrealistic expectations of symptom management and disease directed treatment were reported to often cause conflict between professionals and families. Examples of this include the expectation that a child or young person would always be free of symptoms. Previous studies have reported that unprepared parents, unrealistic parental expectations, and parental fear of hastening death contribute to conflict between parents and professionals in paediatric end of life care (389-391). This conflict can result in a major barrier to providing palliative care (392). Including items on symptom experience in C-POS will allow for early identification and open a dialogue between professionals and children and their parents in order to set realistic expectations for symptom management. This may in some part ameliorate any potential conflict and improve the overall healthcare outcomes and experience for both the child and their family.

In addition to general symptom management and the commonly reported symptoms cited above, participants in this study discussed the impact that medical interventions had on their daily life. Burdensome medical interventions reported

included taking medications, chemotherapy side effects, surgery, blood tests, enteral feeding tubes and central venous access devices. Previous research with children with cancer has found that having central lines accessed and taking medication makes children feel 'bad' (393). If children and families are struggling with the burden of medical interventions, the inclusion of questions about physical symptoms and concerns in C-POS could highlight this. This could prompt clinicians to have conversations about the benefits, risks, and potential futility of further such interventions, and allow viable alternatives to be explored. Opening this dialogue will keep the child and their experiences at the forefront of decisions regarding treatment thus promoting child-centred care.

Spiritual and existential concerns

Spiritual and existential concerns are a core component of adult palliative care (394). The European Association of Palliative Care define spirituality as 'the dynamic dimension of human life that relates to the way persons (individual and community) experience, express and/or seek meaning, purpose and transcendence, and the way they connect to the moment, to self, to others, to nature, to the significant and/or the sacred (395). Spirituality is described as being multidimensional and includes existential concerns such as meaning of life, joy suffering and hope; value based considerations such as what matters most to a person; and religious considerations such as faith, beliefs and religious practices. Despite the spiritual domain being included in the definition of palliative care for children, existing research has focused on religious considerations (rather than spiritual or existential) aspects among children and young people with cancer, from the perspectives of proxies (13, 396). This thesis adds to this knowledge by incorporating the perspectives of children and young people, and those with a non-cancer diagnosis. Participants discussed spiritual and existential concerns (beyond religion) in the context of being able to live a full life and take part in enjoyable activities. Other aspects included questioning the meaning of life and illness, determination to survive and a life unlived. Religious beliefs and needs were also discussed, with participants describing both moving towards and away from faith. Professionals articulated a lack of confidence in discussing spiritual and existential aspects of care with children and their families, which highlights an important gap in professional education and knowledge.

All participants in this study identified the importance of children and young people being able to live a full life by participating in activities they enjoy, such as seeing

friends and family, attending school or nursery, and pursuing hobbies. This finding has links with children and young people with life-limiting and life-threatening conditions wanting to pursue normality and be seen as children first and foremost, with their condition being seen as secondary to this. Prognostic uncertainty was found to impact on the ability of children and young people to make plans to undertake activities they enjoy and live a full life. The data on spiritual and existential concerns identified during the qualitative interview study has been published as a separate paper (see Appendix P Scott, Coombes, Braybrook et al. 2023).

To facilitate a child-centred approach to palliative care, C-POS items need to identify concerns that will enable children and young people to live a full and meaningful life. The wider environment and relationships outside of the home and family need to be acknowledged, as children and young people want to attend school and spend time with friends (81). By asking whether children are able to undertake activities they enjoy, and whether they are able to live a full life within the context of their condition, professionals will be able to explore any concerns children and young people may have in these areas. This may allow issues to be resolved and more open discussions regarding prognostic uncertainty to be had.

Emotional and psychological concerns

Participants in this study spoke about a wide range of emotions experienced by children and young people with life-limiting and life-threatening conditions and their families. This supports the findings of previous research where psychological concerns including feeling sad, feeling nervous, irritability, worry, concentration and insomnia were reported by children and young people with life-limiting and life-threatening conditions and their families (14, 29, 397-399). Children and young people described the psychological and emotional impact of their illness in terms of their ability to engage in usual activities such as play, having fun and attending school (354).

In this study, children and young people frequently experienced a loss of independence. Loss of privacy was a concern for both children and young people and their families. Parents and carers spoke about the impact of their child's condition on all aspects of family life.

Children and parents often described 'mutual protection' whereby they wanted to shield each other from their concerns and worries, particularly regarding prognosis.

Previous research has shown that the majority of parents do not talk about death with their terminally ill child (400, 401). However, when such discussions do take place, parents did not regret having them (400). This highlights the importance of ensuring a child-centred approach to care, whereby both the needs of the child and their family are assessed and addressed.

Access to psychological support was sometimes challenging for family members who were in paid employment or at school during normal working hours. Taking a child-centred approach to care means that services should be co-ordinated around both the child and family (82). Inclusion of items regarding support for family members within C-POS could allow important gaps in services to be highlighted. This could potentially improve future care and improve the health outcomes of children and their families.

The findings discussed above highlight the need to include items in C-POS that measure outcomes from the perspective of both the child or young person with a life-limiting or life-threatening condition and their immediate family members. This will allow opportunity for professionals to address concerns regarding both the child and their family and thus support a child-centred approach to care.

Social and practical concerns

Participants spoke of many social and practical concerns that living with a life-limiting or life-threatening condition brings, such as the impact on the ability to see friends, attend school and undertake hobbies. Factors such as treatment, isolation, stress on family relations, care burden and children and young people being concerned about the impact of their illness on their parents were also identified and have previously been reported (13, 30, 31).

The need for information regarding a child's condition and its treatment was also important to participants, with siblings reporting that they were less well-informed than parents and unwell children. Siblings of children with life-limiting conditions are often forgotten in the midst of a family trying to cope and the consequences of this can be significant (402, 403). Evidence shows that when siblings needs are unmet every aspect of their life from education to family life, can be affected (404). This highlights the importance of assessing outcomes from the perspective of the whole family, in order to achieve the principles of child-centred care.

Adult participants in this study discussed many practical aspects of care. This included familial care burden and the logistics of managing this. These findings support those in previous studies (405). This study found that children did not share these concerns, once again highlighting that children have their own needs, which are often different to those of their family. However, within the context of a child-centred approach to care, parents and carers need to be supported in manging the practical aspects of their child's care. This will ensure that children with life-limiting and life-threatening conditions can pursue and enjoy usual childhood activities and maintain a sense of normality in their lives. Therefore, it is important that when outcomes are assessed parents and carers are given the opportunity to respond to items that affect them, as their well-being and ability to cope is important in its own right, but also impacts on the outcomes of the child or young person affected by a life-limiting condition.

The opportunity to plan for the future and have advance care planning conversations was important for many adult participants in our study. No child or young person discussed this despite the United Nations Convention on the Rights of the Child recognising the right for children to be involved in medical decision making (78). Within the UK legislation allows decisions to be made about children up until the age of 16 years old according to best interests. However, it also recognises that young people should be involved in their own care decisions (206, 406). Research suggests that the optimal time to initiate an advance care plan with a young person is in their mid-teenage years (407). Asking parents and carers about planning for their child's future as part of a child-centred outcome measure could facilitate conversations with professionals on if and how to have these conversations with their child, thus promoting emotional well-being and outcomes (408).

Pursuing normality

The cross-cutting theme of pursuing normality found in the qualitative semi-structured interview study was an important consideration throughout the development of C-POS. Children with life-limiting and life-threatening conditions want the focus to be on them as children, with their condition being seen as secondary to this. The theme of pursing normality was influenced by the other four themes. If physical symptoms and concerns, emotional/psychological, social, practical, and spiritual/existential concerns were not adequately addressed and managed, this impacted on their ability to undertake usual childhood activities. It

was therefore important that symptoms and concerns that impacted on this pursuit of normality were included in C-POS.

Previous research has identified similar themes, whereby families experience a disrupted normality at diagnosis and then attempt to reconstruct this and start to adjust to a new normality by re-organising family life based on the needs of the sick child. This enables a sense of control over the disease by understanding the illness and treatment (409, 410). Families, to varying degrees, strive toward the incorporation of normalisation, depending upon their definition of the situation, in the way they manage their children's complex care needs, and perceive future consequences of a child's condition (411). Most families who have children with serious illnesses eventually view their children and their lives as normal (411). Children are also reported wanting to gain control by striving for a normal life, fitting in at school and taking part in activities with other children (410).

The evidence discussed above demonstrates that a holistic, child-centred approach to address the care needs of children and young people with life-limiting and life-threatening conditions is required. C-POS needs to contain outcomes that identify priority symptoms and concerns and allow for these to be addressed so that children and young people can achieve this pursuit of normality.

9.4 Objective iv). To develop a list of candidate priority outcomes to be included in C-POS

One output of the qualitative interview study was a list of 42 outcomes that could potentially have been included in C-POS. These are discussed in Chapter 6 and were taken to a Delphi survey for prioritisation by key stakeholders.

Both Rothrock and COSMIN state that the construct that a PROM is designed to measure needs to be well defined and clearly described (123, 149). In this thesis the construct measured by C-POS was not defined *a priori*. It was the intention that it would be defined based on the results of the semi-structured qualitative interview study which aimed to establish priorities for outcomes of care with key stakeholders.

One candidate construct for C-POS was health-related quality of life. The term 'health-related quality of life' is used with the intention of narrowing the focus of 'quality of life' to the effects of illness, health and treatment on quality of life (412). Health-related quality of life measures are often weighted towards symptoms and function and these concepts may be less relevant in advanced illness (96).

Commonly used models of health-related quality of life contain biomedical and social science domains and purport that environmental and individual factors are associated with outcomes (412, 413). Individual factors may include demographic and developmental aspects such as gender, family history and ethnicity (412). Environmental factors can include social aspects such as the influence of family and friends, as well as physical aspects such as housing and locally provided amenities (412). These environmental and physical factors are not amenable to intervention by a multi-disciplinary paediatric palliative care team and thus are not measured by C-POS. Therefore, C-POS cannot be considered to measure the construct of health-related quality of life.

By capturing what is important to children with life-limiting conditions and their families across the five themes discussed above, C-POS was developed as a measure of symptoms and concerns which could benefit from intervention by members of a multi-professional paediatric palliative care team, as defined by NHS England and NICE (55, 414) . Although C-POS is not measuring health-related quality of life, it could benefit this construct by leading to improved recognition and management of symptoms and other important concerns. C-POS will allow holistic assessment of child and family needs by identifying common and important symptoms and concerns that affect children and young people affected by life-limiting and life-threatening conditions and their families. Future construct validity testing will establish whether C-POS improves other constructs such as pain and health-related quality of life by assessing concurrent and discriminant validity with other commonly used measures (125).

9.5 Objective v). To gain stakeholder consensus on items to be included in C-POS and construct first versions

This objective was achieved by conducting a modified Delphi survey with parent/carers and health and social care professionals working in paediatric palliative care to establish which outcomes identified in objectives ii and iii were a priority for inclusion in C-POS. A young person's advisory group were also consulted on priority outcomes. The results of previous work presented in this thesis were presented at an item generation meeting with key stakeholders where initial CPOS versions were developed.

The item generation phase of PCOM development is rarely well described (231). There is little guidance given by COSMIN or Rothrock on what steps should be

taken between conducting a qualitative interview study and/or systematic review to inform content validity of a PCOM and cognitively testing it in the target population (123, 149). The COSMIN manual on methodology for assessing PROM content validity has only one standard for assessing the quality of quantitative studies used to identify relevant PROM content. This advises that the sample size should be over 100. In contrast there are seven boxes to assess the methodological quality of qualitative studies (149). This thesis addresses the lack of a clearly described process of item generation in PCOM development by detailing the scientific and rigorous steps taken during C-POS development in detail.

The population of children and young people living with life-limiting conditions in the UK is heterogenous in terms of diagnosis, age and ethnicity. A strength of the data presented in this thesis is that it represents a range of ages and life-limiting/life-threatening conditions. However, most participants were White British, so the final outcomes may not be fully representative of this diverse population. Due to the heterogeneity of the population of children and young people with life-limiting and life-threatening conditions, it was anticipated that the qualitative interview study would generate a large number of potential outcomes that could have been included in C-POS. As the aim of C-POS development was to develop a measure for use across the range of life-limiting conditions children and young people experience, the final items needed to reflect those most important to the target population. i.e., C-POS is a core outcome scale.

In addition to the evidence on symptoms and concerns generated, data from the qualitative interview study demonstrated that children with life-limiting conditions found a short measure with 10 questions or fewer, taking no more than 10 minutes to complete most acceptable (384). Therefore, it was important to ensure that from the outcomes identified in the qualitative interview study, those that were most important to the majority of the target population were included in C-POS. COSMIN guidance on PROM development states that experts (including patients) should be included in measure development to ensure face and content validity (166). Evidence shows that healthcare professionals need more education on the use and implementation of PCOMs in clinical practice, and suggests that engaging professionals in the measure development processes can help to achieve this (90). We included parents/carers of children and young people with life-limiting and life-threatening conditions as experts in the Delphi survey, as well as health and social care professionals to enhance validity and ensure clinical relevance. Delphi surveys

are not frequently used in PCOM development but some studies have reported the benefit of their use in item generation (235). This thesis contributes to the methodological science of PCOM development by using Delphi methodology during the item generation stage. Conducting the Delphi survey allowed potential items to be reduced to an acceptable number, while ensuring that those most important to the target population were included.

Most healthcare research that utilises Delphi surveys uses classical style Delphi methodology (103). This thesis contains one of the very few instances of a ranking-type Delphi being used in healthcare research. This methodology best met the aims of this phase of C-POS development, whereby key outcomes needed to be identified. The use of a classical style Delphi survey could have resulted in participants selecting all potential items as important for inclusion, which would have made C-POS unacceptable to target users (149, 224). The use of a ranking-type Delphi survey in this thesis contributes to the science for its use within a healthcare setting.

There is no uniform definition for consensus in Delphi surveys. This study used a stopping criterion of W>0.7 which is often cited as a stopping criterion in ranking-type Delphi surveys (226). However, most studies do not actually achieve this, reporting a moderate final consensus rate of W=0.5-0.7 (227, 415). Our Kendall's W coefficient of concordance increased from weak to moderate between rounds two and three, suggesting a move towards consensus. This move towards consensus could potentially have been due to the increase in proportion of health care professionals compared to parents in the final ranking round, however Kendall's W increased for each stakeholder group (parents and professionals) during the ranking rounds, suggesting this may not be the case.

It was important that throughout the development of C-POS the perspectives of children and young people were sought to maintain the focus on developing a child-centred PCOM that reflected the perspectives of all key stakeholders, and to ensure robust face and content validity. It was not possible to include children in the Delphi survey for ethical and logistical reasons. To maintain the child-centred focus of C-POS children's perspectives on priority outcomes were sought via the young people's advisory group. The group were given fewer items to prioritise than Delphi participants, being asked to identify their top ten outcomes from the items included in the ranking rounds. This made the task less overwhelming and easier to achieve

in the time given, but it is also possible that items that were excluded during the narrowing down round could have been important to the group.

Involvement of key stakeholders in C-POS item generation has demonstrated many similarities and some important differences in the priority healthcare outcomes identified by children, parents, and healthcare professionals in paediatric palliative care. Pain management was a priority for all stakeholder groups. Pain can influence the daily life of children in a multitude of ways and has been linked to poor school performance, difficulties socialising, sleep issues and reduced overall quality of life (416). Therefore, it was apparent at this stage in C-POS development that an item about pain would be essential. Other common priorities included children being able to live life to the fullest and do things that they enjoy. These findings reflect the overarching concept of pursuing normality found in the qualitative interview study.

Healthcare professionals tended to give a higher priority to managing symptoms that were amenable to treatment with medication. Pain was an exception to this in that it was a priority for all stakeholder groups. This is a finding that has been previously reported (417, 418). There may be several reasons for this. The majority of participants in the Delphi survey were doctors and nurses. Their professional focus is often on managing physical symptoms that can be ameliorated with medication. Management of physical symptoms is often perceived to be more within the boundaries of their job role than management of emotional, social and psychological concerns, hence why they may have been prioritised for inclusion in C-POS.

Parents were more likely to prioritise psychosocial concerns such as the emotional impact of a life-limiting condition and their child’s physical function. These outcomes were probably identified as a priority because they impact family care burden and participation in activities outside the home. Research shows that there is a significant association between carer strain and depression, and unmet needs relating to emotional and practical support in parents of children and young people with life-limiting and life-threatening conditions (419). This may be why our sample highlighted these particular concerns as an outcome priority.

The young people's advisory group also identified being able to access education and maintain peer relations as priority outcomes. These items were not highlighted as a priority by parents or professionals. This finding corroborates the findings from the C-POS qualitative interview study and previous research that identified the importance of addressing not only physical needs but also supporting pursuit of activities which are part of normalcy for children (362, 364, 405, 420). The finding

that children and young people have different priority health outcomes further highlights the need to ensure that children have a voice in research that affects them. It also further promotes the need to take a child-centred approach to care by acknowledging that children's views are not always the same as those of their parents and health care professionals (82).

During the item generation meeting all the evidence generated (as described above) was presented to the study steering group and discussion was had about outcome items to include within each domain identified in the qualitative interview study (physical, social/practical, emotional/psychological and spiritual/existential). It was important that outcomes that were important to all stakeholder groups were incorporated into C-POS in order to optimise measure acceptability and future implementation. Engagement of UK paediatric palliative care professionals was a priority throughout this thesis, with each recruiting site being invited to have one participant in the study steering group. Evidence suggests that engaging paediatric healthcare professionals in the development of PCOMs facilitates future implementation (421).

Evidence presented from the systematic review and qualitative data on recall period and response format highlighted the requirement for different C-POS versions for those of different ages/developmental stages, with proxy versions for those who were unable to self-report. The different priorities expressed by parents regarding practical aspects of care also led to agreement that there needed to be items regarding the child, and separate items that reflected family outcomes. This decision further strengthens the child-centredness of C-POS as it acknowledges children's views and opinions while also taking the needs of the wider family in to account (66). Particular issues that were discussed during the item generation meeting were the inclusion of items regarding symptoms beyond pain, and how to ensure that items reflected the needs of siblings. It was agreed to have a global symptom item, rather than asking about multiple individual symptoms, in order to reduce the risk of missing data if symptoms were irrelevant to individual respondents. Likewise, despite sibling outcomes being a priority for all stakeholders throughout the research presented, it was agreed to have a generic question about impact on the family to avoid missing data from families whose child with a life-limiting condition had no siblings.

The item generation phase of C-POS development has ensured that by involving key stakeholders the measure has excellent face and content validity for the

construct being measured, the target population and context of use (149). C-POS items capture all domains covered in the World Health Organisation's definition of paediatric palliative care. The robust and transparent description of the item generation process of C-POS contributes to the science of outcome measure development. It also demonstrates that such research can be done with children and young people with life-limiting and life-threatening conditions and their families.

9.6 Objective vi). To establish acceptability, comprehensiveness, and comprehension of C-POS versions

This objective was achieved by conducting cognitive interviews with children with life-limiting and life-threatening conditions and their parents/carers using 'think aloud' and verbal probing techniques. Cognitive testing of C-POS revealed some problems with the initial versions, particularly when parents of non-verbal children were providing proxy reports. For example, initial C-POS versions asked about concepts such as whether a child had been 'able to share their feelings' or 'ask questions'. Changing the wording to use terms such as 'express feelings' and 'had appropriate information for them' allowed C-POS to be more inclusive of the range of children with life-limiting conditions, many of whom have communication difficulties (10). An option for 'not appropriate to my child' was added for those who could not respond in order to avoid missing data in future psychometric testing. Making these changes enabled C-POS to maintain its child-centred focus by ensuring that all children with life-limiting or life-threatening conditions receive a holistic assessment of their care outcomes in a way that is meaningful to their individual development and ability.

In the original C-POS versions developed in the item generation meeting many of the items began with 'How much...', or 'how often...' During initial cognitive testing it was found that participants were responding to these items based on the frequency of an outcome occurring - e.g., the amount of time they were in pain. C-POS is intended to measure symptoms and concerns in terms of the impact on a child and family's life, so it was important that items measured more than just frequency. By removing these quantifiers further cognitive testing revealed that participants were responding based on the impact of items on their life and how this affected their ability to pursue normality, which is the intention.

The parent/carer version of C-POS (version B) intended for parents of verbal children and those over two years old was cognitively tested over seven rounds. This could be perceived to be a large number of cognitive testing rounds for a

PCOM, however previous studies have not reported the number of rounds conducted, or whether the items were cognitively tested in their final version (231, 243). It was imperative that the wording of C-POS was right for the intended population of use and this rigorous cognitive testing is a strength of the measure development process presented in this thesis.

Adult participants in the cognitive interview study also highlighted the need for the addition of a question regarding psychological and emotional support for the family in C-POS. They felt this was not incorporated into the existing item regarding 'having the support needed to care your child'. The addition of this question supports the concept of a child-centred approach to care by acknowledging the overall experience of not just the child, but their family as well (35, 82).

COSMIN recommends that PCOM items are cognitively tested with at least seven participants and that all items are tested in their final format (149). It does not specify how many times the final format should be tested with the target population. All C-POS versions were tested in their final form, with the parent proxy versions being tested in 3-5 participants and the child self-report versions being tested in one to two participants. By this point we had sufficient information power to suggest no further revisions were required. Within the child versions, two changes were made between version 1 and 2. One change was to question eight in the eight to twelve year old and 13-17 year old C-POS where ‘Have you been able to ask the questions you wanted to over the past week/yesterday or today?’ was changed to ‘Have you been able to ask the questions that are important to you over the past week/yesterday or today?’. The wording was the same for both versions so was actually tested in its final format with three children in total. This limited cognitive testing of the final C-POS child versions could potentially affect the measure's content validity, impacting on future psychometric testing of C-POS (149, 422). Irrelevant items could decrease internal consistency, structural validity and interpretability. Missing concepts could decrease validity and responsiveness (149).

The second change made to the child versions was to the response format, where 'most of the time' at the end of the scale was changed to ‘always' as participants felt that this fitted better with the 'never' response at the other end of the scale. The initial response format for C-POS was based on evidence from the systematic review and qualitative interview study conducted in phase 1. The response formats of other commonly used measures in paediatric healthcare were also considered, including the Pediatric Quality of Life Inventory (PedsQL^{TM}) (423). The PedsQL^{TM}

has been widely used in research studies and there is good evidence of reliability and validity in both healthy and unwell children (424). The PedsQL™ uses 'Almost always' rather than 'Always' at the end of its response scale and no problems have ever been identified with this in psychometric or cognitive testing, which does not support the findings presented here.

The length of C-POS and the number of items included is based on the findings of the systematic review and qualitative interview study conducted in phase 1 of this thesis (384). During cognitive testing, all participants found the number of questions and the time taken to complete them acceptable. It is important that the length of a PCOM aligns with the preferences of the target population, as otherwise they may lose focus or motivation to complete it (139). Cognitive testing has demonstrated that children and young people with life-limiting and life-threatening conditions and their families did not find C-POS too long or burdensome to complete.

Cognitive testing demonstrated some issues in children aged five to seven years old (or those with similar ability). They usually needed an adult to explain questions to them before they could choose an appropriate response and undertake the 'think aloud' technique required for the study. This supports findings from previous research and from the qualitative interview study conducted in phase 1 of this thesis that show that when younger children are completing PCOMs they need to be administered with initial adult support so that more difficult concepts can be explained (425). This finding may have implications for C-POS if it is developed as a computerised measure in the future. It is important that younger children are not expected to self-complete a computerised measure without the support of a parent or healthcare professional.

Children aged five to seven years old (or similar ability) also demonstrated some instances of social desirability bias during cognitive testing of C-POS. For example, there were instances of participants saying that they had not experienced any pain, when in fact they had. This finding corroborates previous research which has shown that children this age can be suggestible when participating in survey research (252). This has implications for measuring outcomes in this age group, and for the implementation of this measure, as children need to be told that there are no right or wrong answers. With encouragement children were able to express their own thoughts and experiences as the cognitive interview progressed and fully participate in the research process.

Children aged five to seven years old (or similar ability sometimes responded to items in terms of the ‘here and now’, rather than ‘yesterday or today’. This supports evidence that young children may not always understand the difference between the past, present and future and sometimes engage in 'scripting' whereby they respond about what usually happens and regularly occurring events (426). In the systematic review conducted in phase 1 of this thesis evidence suggested that children seven years old and younger think dichotomously and are unable to use three-point Likert type response formats. Cognitive testing of C-POS demonstrated that all children were able to use a three-point Likert response format, a finding which contributes to the evidence base in this area. We were keen to test whether younger children five to seven years old could use a three-point Likert scale, as a dichotomous format limits the responsiveness of an outcome measure (314). Future inter-rater reliability testing of C-POS will help to establish whether any additional recall and response format issues are occurring, particularly in younger children where the evidence presented above is not clear cut.

A small number of parent participants found some C-POS items upsetting to answer during cognitive testing but acknowledged that they were in a challenging and emotive situation, so this was to be expected. They did not feel that such items should be removed from C-POS as they felt they were still important to discuss and address with the health care team. Parents also reflected that the cognitive interviewing process meant they had to think about items in more detail than they would do in a clinical scenario which could have exacerbated any upset. Parents were keen to share that they expected to experience some mild distress when taking part in a palliative care research study, and they found this acceptable. This finding has implications for ethical review of future studies and supports previous findings that caregivers can find taking part in paediatric palliative care research hard at times, but this is acceptable (182, 196).

9.7 Strengths, challenges, and limitations

9.7.1 Measure development process

C-POS has been developed following accepted PROM development guidance outlined by both Rothrock and COSMIN (123, 149). This mixed methods approach has ensured that the measure has robust face and content validity within the target population by demonstrating relevance, comprehensiveness, and comprehensibility. This thesis has demonstrated that all items within C-POS are relevant to measuring

symptoms and concerns in children with life-limiting and life-threatening conditions and their families. This was achieved by establishing relevant outcomes in phase 1 of this thesis, gaining consensus on which were a priority for inclusion in C-POS in phase 2 and then further testing the final items for relevance with cognitive interviews in phase 3. Comprehensiveness of C-POS was established by undertaking the Delphi survey and young person's group in phase 2 to ensure no important relevant items were missing. This was assessed again in the cognitive interview study by asking participants whether any items were missing. Comprehensibility was assessed in the phase 3 cognitive interview study to make sure the target population understood the items, recall period and response format as intended.

Feasibility and acceptability of C-POS within the target population have also been demonstrated. The cognitive interview study demonstrated that children with life-limiting and life-threatening conditions and their parents are able and willing to complete C-POS.

Very few PROM development studies explicitly describe the item generation process (231). A strength and methodological contribution of this thesis is that this process is clearly outlined. Delphi survey methodology was used to determine consensus on which outcomes were a priority for inclusion, a method which is very rarely used in PROM development. Use of this methodology, along with engagement from the young person's advisory group allowed for a robust and transparent item generation process for C-POS.

Another strength of this thesis is the use of mixed methods to develop C-POS. This has enhanced the face and content validity of the measure by harnessing the strengths of both qualitative and quantitate research. However, to a novice researcher, using mixed methods to develop C-POS posed some challenges. Learning was required in the different data collection and analysis approaches used throughout this thesis.

Both the Rothrock and COSMIN PROM development processes used in the development of C-POS are designed for self-reported outcome measures (123, 149). C-POS is a patient-centred outcome measure (PCOM), which allows for proxy-reporting when a child or young person is unable to self-report. Some items in the Rothrock and COSMIN content validity processes state that you should speak directly with patients and do not incorporate such child or patient-centred measure

development. In this study these recommendations were adapted to allow children and young people as well as other key stakeholders to inform measure development. All other aspects of both the Rothrock and COSMIN measure development processes fitted well in the development of a measure for children and young people with life-limiting and life-threatening conditions, as they allowed researchers to select the most appropriate methods for the population and research aims.

9.7.2 Study samples

A strength and contribution of this thesis is that it presents the perspectives of children and young people with a range of life-limiting and life-threatening conditions. Previous paediatric palliative care research has focused on children with cancer, often from the perspective of proxies, rather than children and young people themselves (13). A total of 38 children with life-limiting conditions participated in the research presented in this thesis, 22 further children and young people participated in the young person's advisory group, and 13 siblings took part in the semi-structured interview study.

To protect participant anonymity as many life-limiting conditions are extremely rare, all demographic data is presented using ICD-10-chapter headings, an approach used in previous studies (5). Across all phases of C-POS development the perspectives of a wide-range of diagnostic categories and life-limiting conditions has been captured, further strengthening face and content validity. **Error! Reference source not found.** shows overall recruitment by diagnostic category.

One limitation to C-POS development presented in this thesis is that only one participant was recruited to the perinatal diagnostic category, and only four parents of children under one year old were recruited across the whole research process (see Table 16). Prevalence of life-limiting conditions in the UK is highest in those under one year old, where perinatal conditions are more common (5). This has implications for the face and content validity of C-POS in children under one year old as very few participants were recruited from this group. Further work will be required with this population to establish this.

Our sample did not contain many children under seven years old (three in the qualitative interview study; one in the cognitive interview study), although two participants in the cognitive interview study chose to use the C-POS version intended for five to seven year olds as this best suited their preference and ability.

This may have impacted findings both in terms of establishing priority outcomes in this age group and the comprehensibility of subjective C-POS items such as worry. It could also have impacted the finding in the cognitive interview study that children in this age group could use a three-point Likert response format. Exploration of future psychometric data is required to affirm these findings.

In many young people with life-limiting and life-threatening conditions chronological and developmental age are not always congruent and it could therefore be argued that C-POS could be used with young adults with a developmental age of under 18 years old. However, C-POS has been designed and tested as a measure to be used from birth up until a young person's 18th birthday, and this is the population that it's use will be recommended in. Further research would be required to ensure that C-POS is valid and reliable in a young adult population.

Participants that contributed to C-POS development were recruited from across a wide geographical area, incorporating all four UK countries. There is geographical variation in UK paediatric palliative care service provision, and widespread recruitment allowed for differences in perspectives based on provision to be accounted for which is a strength of the work presented in this thesis (39).

The majority of participants recruited to the Delphi survey and cognitive interview study were white British. This is not reflective of the population of children and young people with life-limiting and life-threatening conditions in the UK, which are more prevalent in those from Asian, Black and Bangladeshi backgrounds (5). Unfortunately, collection of ethnicity data was not part of the ethical approval for the semi-structured qualitative interview study which may in part explain participant demographics. Due to the lack of diversity across all studies presented in this thesis, C-POS items may not have such robust face and content validity for those from minority ethnic backgrounds. Further work is required after C-POS has been psychometrically tested in the English language to translate it into other languages and retest its psychometric properties.

The parent participants that contributed to the development of C-POS were predominantly female. This is consistent with much of paediatric palliative care research, i.e. fathers are often under-represented (355). This may have implications for the face and content validity of C-POS in fathers and other male carers.

9.7.3 Recruitment, sample size and data saturation

Recruitment

Recruitment to the qualitative interview study was initially slow, taking three months from the study opening to recruiting the first non-professional participant. This experience resonates with that of other paediatric palliative care research studies where there was been significant non-invitation of eligible participants (427). Evidence suggests that clinicians are often reluctant to refer families for palliative care studies due to perceived burden of participation, fear of upset caused by the term 'life-limiting' and anxiety that their clinical performance would be in some way evaluated (192, 428). Clinicians who have good relationships with a family are more likely to invite them to participate, and if a child's condition was unstable or a child was felt to be close to death then this was also a barrier (429). The C-POS study opened to recruitment at some sites at the same time as two other national paediatric palliative care studies which had very similar inclusion criteria. This and our exclusion criteria stating that participants should not be enrolled in another study may also have impacted recruitment. During recruitment to C-POS no data was collected on how many participants were eligible to participate at each site, how many were approached and how many declined. Therefore, the extent of any clinician gatekeeping was unclear.

In this research, in addition to probable clinician gatekeeping, we also found that for many of our recruitment sites, this was their first experience of inviting children/young people and families to participate in a palliative care research study, and they were hesitant about how, when and if to do this. Interventions such as regularly attending team meetings and providing promotional recruitment posters (Appendix O) and information sheets with suggested phrases to use when approaching families boosted recruitment (Appendix P and helped improve the confidence of recruiting teams. We are unable to recruit as many children and young people as initially intended, due to these initial recruitment problems and the impact of the COVID-19 pandemic, so sought an ethics amendment to be able to recruit more parents and carers in order to get a wider perspective on priority outcomes from those who lived with the daily impact of a life-limiting condition. An extension to the data collection time periods also had to be sought for the same reasons (Appendix H).

During the course of this PhD there were many opportunities to speak about the C-POS study and its development at national meetings, such as those run by the Association of Paediatric Palliative Medicine, Together for Short Lives and CoPPAR (UK paediatric palliative care research network). This raised awareness of C-POS among paediatric palliative care professionals within the UK. It also generated interest from teams who wanted to be involved in further C-POS development research. This raised awareness and eagerness from sites who wanted to be involved in future research meaning that recruitment to the Delphi survey and cognitive interview study were much more straightforward than to the original qualitative interview study. The work outlined in this thesis has done much for the appetite for research within the UK paediatric palliative care sector, particularly with regards to C-POS. Many sites who had never conducted or recruited to paediatric palliative care research have become involved. As such, the research presented in this thesis has contributed to improving 'research readiness' within the speciality. Another benefit of raising awareness of C-POS development within the UK paediatric palliative care workforce is the impact it will have on future implementation of the measure. Clinician education about a measure, and understanding why it is needed and how it will benefit their practice have been shown to benefit implementation (90).

Sample size and data saturation

In this thesis it was necessary to have a large sample size in the semi-structured qualitative interview study for several reasons. These include the multiple stakeholder groups eligible for participation and the heterogeneous nature of the population of children with life-limiting and life-threatening conditions (10). It needed to be ensured that there was enough representation from each group to capture comprehensive data on priority outcomes from all stakeholders. As there were multiple team members collecting the data and indexing transcripts (led by myself) this was manageable. The decision on sample sizes for this study were made pragmatically, based on expectations of how many participants it would be feasible to recruit within a population of unwell children.

The COSMIN content validity standards for evaluating the quality of studies on PROM development state that evidence should be provided that data collection continued until saturation was reached (149). Data saturation, also termed information redundancy, evolved from the notion of theoretical saturation in grounded theory (430). It is defined as 'the point at which the properties of

categories and the relationships between them are comprehensively explained' (431). In the sense intended, saturation means more than the usually used concept of 'no new ideas emerging from the data' but also the notion of a conceptually dense theoretical account of the field of interest where all the categories are fully accounted for, the variations within them explained and all relationships between categories are established, tested and validated for a range of settings (432). In more recent years the use of ‘saturation’ outside of the context of grounded theory has been questioned. The challenge with saturation as a construct is that there is always potential for new theoretical insights to be made as long as data continues to be collected and analysed (433, 434). Also, funders, sponsors, and ethics committees want to know the details of sample size in advance of providing funding and approvals and nearly all research is a pragmatic activity shaped by time and resource (430). Within this thesis, the aim of the qualitative interview study was to establish 'priority' outcomes from key stakeholders and define the concept of symptoms and concerns in a population of children and young people with life-limiting and life-threatening conditions and their families. Theory development was not required to inform CPOS development. Exploring meaning across the datasets, as opposed to individual experiences enabled generalisations to be made about the target populations 'reality' and allowed for generation of key outcomes that could have been included in CPOS. The coding frame went through multiple iterations as data was analysed. The final version of the coding frame was agreed after 69 transcripts had been coded. No further themes were identified during coding of the final 37 transcripts, which included transcripts from all five stakeholder groups. Thus, given the aim of identifying 'priority outcomes' it was unlikely that further interviews would have yielded symptoms and concerns that were a priority for children and young people with life-limiting conditions and their families.

9.7.4 Steering group and young person's advisory group

Another strength of this thesis is the wide range of stakeholders represented in C-POS development. The study steering group had a diverse group of paediatric palliative health care professionals, three bereaved parents of children with life-limiting conditions, experts in patient-centred outcome development and experts in qualitative research methodology and psychometrics. The young person’s group, working in an advisory capacity, rather than a patient and public involvement capacity, provided valuable insights and suggestions from the perspective of children and young people. This thesis demonstrates that it is feasible and

acceptable to work with children and young people as advisors in paediatric palliative care studies.

The healthcare professionals on the steering group represented all sites that recruited to the semi-structured qualitative interview study and the cognitive interview study. Holding regular steering group meetings and acknowledging them in publications and conference presentations strengthened their sense of ownership in the development of C-POS and likely boosted their enthusiasm to drive recruitment within their site. Their insights into how C-POS would be used in day-to-day care and the benefits it would have for patient outcomes also ensured that the acceptability of C-POS to end-users was considered throughout the study.

The parent representatives were all bereaved of children with life-limiting conditions. Their children had died between three and five years prior to the commencement of this study. They offered valuable insights into the relevance of results presented in meetings, research procedures and how these would impact children and families, and analysis and interpretation of results. One of the parent representatives also attended the ethics committee meeting for the cognitive interview study. As all of our parent representatives were bereaved, there is a possibility that some of their insights were affected by recall bias. The development of C-POS may have been strengthened by having parents who were still caring for a child with a life-limiting or life-threatening condition in the steering group, or who were more recently bereaved. However, ethically this was not considered appropriate due to the additional burden or distress it could cause them.

Another way that this study could have been strengthened is by including children and young people in further patient and public involvement work, beyond the involvement of the young person's advisory group. This may have added further context to the results presented and enhanced the design of C-POS. In adult palliative care there are challenges to conducting meaningful patient and public involvement, and these are amplified by ethical concerns in paediatric palliative care research (177, 435). However, there are examples of successful patient and public involvement work with children and young people with life-limiting conditions (177). Participants report that they have a desire to be involved in patient and public involvement work in paediatric palliative care research, and recognise the importance of this (436).

9.7.5 Non-verbal children and the concept of child-centred care

Many children with life-limiting and life-threatening conditions are non-verbal due to the nature of their illness. The C-POS study inclusion criteria did allow for those who could communicate via their parents, or with the use of augmentative or alternative communication aides, to participate. Due to the predominantly qualitative methodology used in C-POS development, it was not possible to include the perspectives of children with severe communication impairment. Therefore, it is possible that the items included in C-POS do not reflect the symptoms, concerns and care priorities of these children and young people. Provision of child-centred care for non-verbal children is just as important as for verbal children. Although they may have difficulty expressing their needs and preferences, children who are non-verbal still have a right to be involved in their care and to receive care tailored to their needs (35). They also have a right to be involved in research if at all possible. Professionals should respect a child's autonomy and involve them in decision making to the extent possible (66). Health care professionals and parents are usually attentive to non-verbal cues such as facial expressions, gestures, and body language in non-verbal children. In addition, parents of non-verbal children with life-limiting and life-threatening conditions are experts in understanding their child's needs and preferences. In providing child-centred care professionals should observe a child's behaviour and listen to parental concerns and insights into their child's needs and preferences (35). An environment should be created that supports a child's emotional and social well-being and allows them to feel secure and at ease (81). By including healthcare professionals and parents in the development of C-POS, some insight into the perspectives of non-verbal children was gained.

9.8 Implications for research

This thesis presents the development of the novel UK children's palliative outcome scale and demonstrates its face and content validity, as well as acceptability and feasibility of use within the target population. The implications and contributions to research methodology are discussed below, followed by identified evidence gaps requiring future research.

Research with children with life-limiting conditions and their families

This thesis demonstrates that it is feasible and acceptable to recruit children and young people with a range of life-limiting and life-threatening conditions beyond cancer and their families to palliative care research studies. Engaging children as

young as five years old to take part in research interviews was successful in the development of C-POS. Several strategies were used to achieve this. Allowing children to have a carer present during interviews allowed them to feel more comfortable talking to the researcher. Beginning each interview with a rapport building process such as playing a game or asking participants about their hobbies and interests enhanced further engagement. There was concern that when interviews moved online during the COVID-19 pandemic that children would struggle to engage, as there would be no opportunity to play with toys during interviews and it would be harder to employ techniques such as colouring and drawing. However, this was not the case. All children that participated in virtual interviews were able to sit and engage in the process and give valuable and insightful responses. This has implications for future research with children as this thesis demonstrates that it is feasible and acceptable to conduct qualitative research virtually, which could save valuable time and resources.

It was anticipated that there would be some difficulty in gaining ethical approval for the qualitative interview study and cognitive interview study. However, in both instances the research ethics committee was very supportive of this research. No children that participated in the research presented in this thesis became upset or distressed during interviews, with only one asking to stop as she felt unwell. This does not support the perception that children and young people with life-limiting and life-threatening conditions should not participate in research as it will cause undue burden (179). In contrast, many participants shared that they enjoyed taking part in the interviews, with some teenagers expressing a wish to make a difference to those going through similar experiences in the future. This suggests it is not only appropriate to include children and young people in palliative care research, but it may also afford them the same benefits recognised in adult research.

Some parent participants became upset during research interviews. All were offered an opportunity to take a break or stop the interview, but all wanted to carry on. Parents shared that they expected interviews to provoke some distress, due to the nature of the research study. However, they all found the level of distress experienced acceptable. This has implications for ethical review of future paediatric palliative care research studies.

Implications for recruitment to paediatric palliative care research

At the commencement of C-POS development, most research sites had no experience of recruiting children and young people with life-limiting and life-threatening conditions and their families to research studies outside of clinical trials of investigational medicinal products. During C-POS development the 'research readiness' of recruiting sites was enhanced using several strategies. Working with busy clinical teams to develop strategies that instilled confidence in them to discuss research participation with children and their family's enhanced recruitment. These strategies included providing posters and information sheets with suggested phrases that could be used and ensuring regular communication between the research team and recruiting team. Attendance at clinical team meetings (both in person and virtually), and regular email updates outlining recruitment by site also helped. Finally, instilling a sense of ownership and participation in the overall study by inviting a representative from each participating site to join the study steering group and be acknowledged in publications has enhanced recruitment. All of these strategies are straightforward to implement and will help future researchers to recruit to future paediatric palliative care studies.

Development of C-POS has had an impact on the way paediatric palliative care research is viewed by the UK paediatric palliative care workforce. During this thesis the paediatric palliative care clinical and research workforce within the UK have been regularly updated on the C-POS development process and research findings at national conferences and meetings. This has raised awareness of the C-POS study and the relative success of recruitment and evidence generation. This has sparked huge interest from further sites who want to be involved in future C-POS studies and has resulted in over 70 sites gaining approval to recruit to the next phase of C-POS development (psychometric testing). This demonstrates how much of an impact the work presented in this thesis has had in enhancing the research readiness of the UK paediatric palliative care sector.

Areas for future research

During the semi-structured interview study conducted in phase 1 of this thesis (437, 438) participants spoke about many aspects related to quality and experience of care. This data is beyond the scope of C-POS development and this thesis, but while listening to these experiences in interviews and analysing transcripts it became clear that there is a need for the development of a care quality measure

and patient-reported experience measure within paediatric palliative care. The experiences of care at the end of life can have a large impact on the subsequent bereavement experiences of both parents and siblings and a recent scoping review demonstrated that existing measures have gaps in important domains such as cultural aspects of care, grief and economic costs (438).

Another area for future research that has been highlighted in this thesis is the development of strategies to increase the cultural and ethnical diversity of participants to paediatric palliative care research.

Finally, further research is needed to explore the benefits and burdens of participation in research from the perspective of children and young people with life-limiting and life-threatening conditions. This could have been achieved during the research presented in this thesis if ethical approval had been sought to ask questions about participant experience at the end of either the semi-structured or cognitive interview studies. Although this would have made the interview slightly longer, thus increasing participant burden, it would have given valuable insights into the experience of participants and contributed to the current lack of evidence on this. Future researchers could consider incorporating such questions into their studies.

9.9 Implications for clinical care

This study demonstrates that children and young people with life-limiting and life-threatening conditions and their families have multidimensional symptoms and concerns. Healthcare professionals need to be able to assess and manage these symptoms and concerns in order to support children and young people to be able to pursue normal childhood activities. The focus should be on children being supported and encouraged to be children, rather than focusing on their health condition. This is best achieved by working within a child-centred model of care, where it is acknowledged that children exist both within and outside of their family. In addition, families of children and young people with life-limiting and life-threatening conditions need support to provide often complex and burdensome care for their child.

This research also demonstrates that when implementing PCOMs into clinical practice with children and young people with life-limiting and life-threatening conditions, consideration should be given to ensuring that they are given the opportunity to discuss their responses with a healthcare professional during or soon after measure completion. Children need to know that their responses are seen and

considered and to ensure that their healthcare experience has a human and compassionate feel. This further supports a child-centred approach whereby children are regarded as active and equal partners in their care (35). In addition, taking this approach should enhance the acceptability and uptake of PCOMs in clinical practice.

This research also demonstrates that consideration needs to be given to a more flexible approach to service provision for children with life-limiting conditions and their families. Taking a child-centred approach to care means services should be co-ordinated around the needs of children and their families (35). Participants in this study often struggled to access psychological support that fitted around caring and work responsibilities. Spiritual and existential support was often hard to access due to lack of clinician awareness and education. Participants also found it challenging to find suitable education for children with complex medical needs and to access to vital equipment needed for their child's care. Equipment was often self-funded by families due to the lack of available resources within health and social care.

9.10 Reflections, personal development, and learning.

Prior to commencing my PhD, I had been working in clinical practice for 20 years, 11 of those in paediatric palliative care. I had received some research training while undertaking my MSc in 2012 and conducted and published a systematic review for my dissertation. I had very little first-hand experience of conducting and analysing primary research data. Prior to data collection for the semi-structured interview study in phase 1 of this thesis I received training on interviewing children and young people with life-limiting and life-threatening conditions, and ethical considerations in paediatric palliative care research. I was also able to access training on qualitative data analysis and the use of NVivo, which was invaluable in analysing the large qualitative semi-structured interview dataset.

The COVID-19 pandemic began while I was planning the Delphi survey and most training courses had been postponed or cancelled. Therefore, to analyse the quantitative data from the Delphi survey I had to learn how to use STATA via online videos and a textbook. This was challenging but extremely rewarding once achieved.

Prior to developing the cognitive interview study protocol, I was able to attend a two day online cognitive interviewing course which strengthened my knowledge of the cognitive interviewing process and analysing the data.

I came to my PhD with my own set of beliefs and assumptions regarding what outcomes were important to children and young people with life-limiting and life-threatening conditions and their families. My clinical background had allowed me to spend time with children and families in their own homes to assess symptoms, prescribe medication and provide tertiary level palliative care. I had also spent some time researching and reading the current evidence within the field of paediatric palliative care and outcomes during my MSc, and prior to commencing my PhD. However, during the research process I was afforded the opportunity of being able to spend more time talking to children and families about what was most important to them than I have the ability to do in clinical practice. This opened my eyes to their experiences, and I realised that very little of what a child and family experience on a day-to-day basis is shared during a short clinical visit, which tends to focus on symptom management and a broad discussion of psychosocial support. I believe my experience of conducting research with this population has enhanced my clinical skills and I am now able to offer a much more holistic approach to patient care. I also learned that it was often better not to share my clinical background with interview participants as they often made an assumption that I already knew and understood their experiences, and I found that I obtained much richer data when this was not disclosed.

I conducted my PhD part-time, whilst also working clinically. Although balancing my priorities was challenging at times, I feel that this clinical academic approach also benefited the research I conducted. Within the UK the field of paediatric palliative care is relatively small. I was able to use my clinical contacts to drive recruitment to all phases of my research, both by reaching out to key contacts at recruiting sites and approaching contacts at potential new sites. My clinical background also gave me an insight into the challenges busy clinical teams experience when recruiting to research. This helped develop some of the strategies to improve recruitment described in this thesis.

9.11 Next steps

This PhD has outlined the development and initial validation of five versions of C-POS - three age /developmental stage appropriate versions for child and young

person self-report, and two parent/proxy versions, following COSMIN and Rothrock guidance (123, 149). This thesis has demonstrated that these five versions of CPOS have robust face and content validity and are acceptable and feasible for use with the target population. The next stage in PCOM development requires that measures are psychometrically tested within the target population to establish construct, concurrent, structural, and convergent/discriminant validity, and reliability. This study is already in progress. Following this, CPOS will require implementation into routine practice. Research is in progress to develop a strategy for implementing C-POS into routine practice in paediatric palliative care and to appraise its mechanisms, processes and potential benefits.

9.12 Conclusions

This study describes the development and initial validation of the first patient-centred outcome measure for use with children and young people with life-limiting and life-threatening conditions and their families in the UK. C-POS has been developed as a child-centred measure of symptoms and concerns that can be used from the diagnosis of a life-limiting condition and throughout the illness trajectory. Items reflect children and young people's desire to focus on the pursuit of normal childhood activities. C-POS has undergone a robust development process using accepted methodological guidance on PROM development. This has ensured items within the measure reflect the construct set out to be measured, and that they have face and content validity within the target population. C-POS has been demonstrated to be relevant, comprehensive, comprehensible, acceptable, and feasible for use with children and young people with life-limiting and life-threatening conditions and their families.

This study also demonstrates that children and young people with life-limiting and life-threatening conditions can engage in research, in this case communicating preferences regarding the development and design of a patient-centred outcome measure. Important differences were found in priority outcomes identified by different stakeholder groups, highlighting the importance of involving all key stakeholders, including children and young people in patient-centred outcome measure development.

Further research is required to establish the psychometric properties of C-POS within the target population, and to develop an implementation strategy. It is also recommended that further research is conducted into ways to engage minority

ethnic participants and fathers into future paediatric palliative care research, in order to ensure a range of perspectives are represented.

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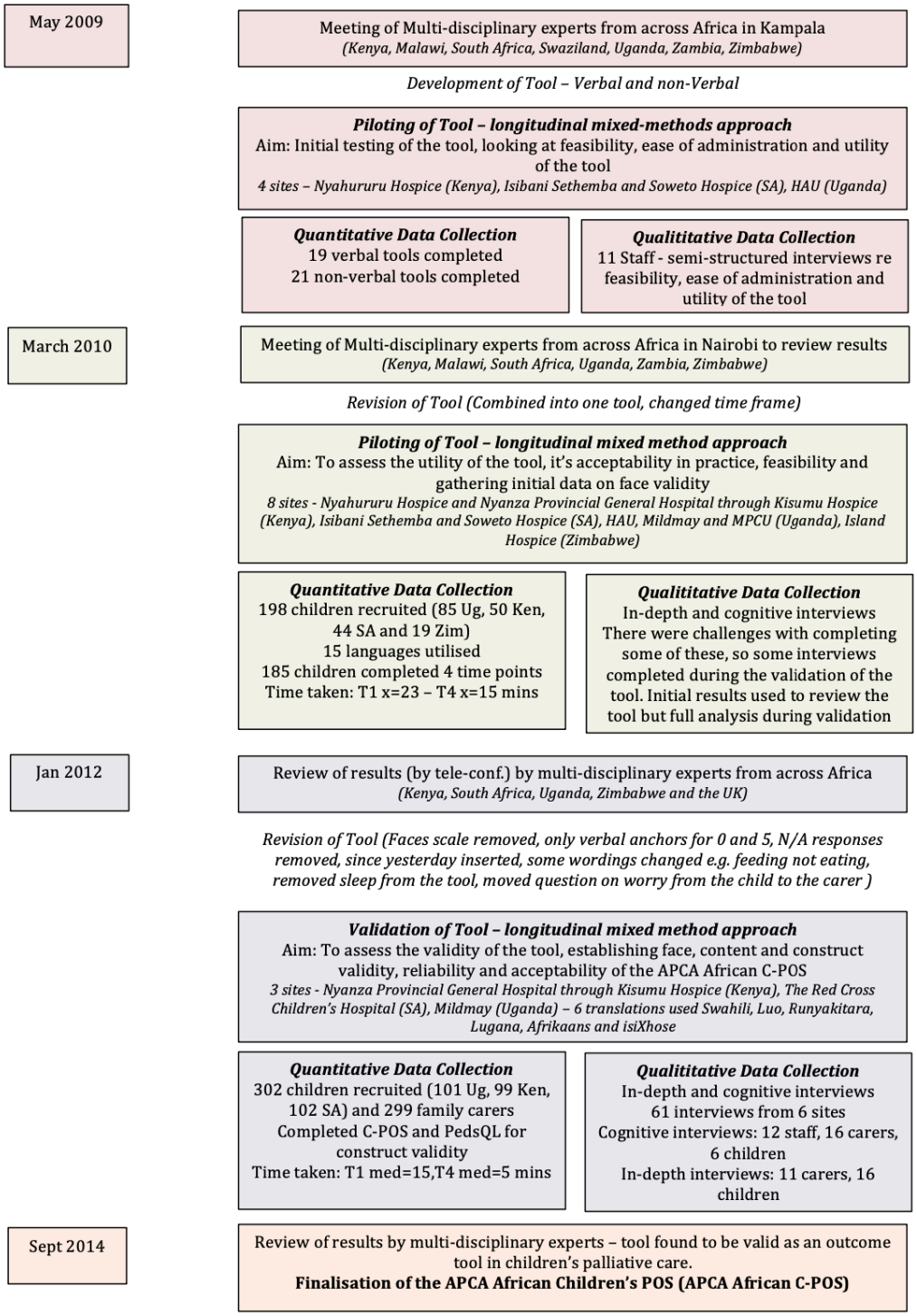
Appendix A Development of the African C-POS

Process of the Development and Validation of the APCA African C-POS



Appendix B African C-POS

Study (patient) Reference Number:		***POS***...../100					**Date:**	**Visit 1**				
								Visit 2				
								Visit 3				
								Visit 4				
	QUESTIONS TO BE ASKED TO THE CHILD		**Visit 1**	**Visit 2**	**Visit 3**	**Visit 4**	**QUESTIONS TO BE ASKED TO THE CARER OR NURSE IF THE CHILD IS UNABLE TO RESPOND**		**Visit 1**	**Visit 2**	**Visit 3**	**Visit 4**
	Question	**POSSIBLE RESPONSES**					**Question**	**POSSIBLE RESPONSES**				
SECTION A: ABOUT THE CHILD												
Q1.	Can you tell me how much pain you have had since yesterday?	0 (No pain) – 5 (The worst pain you can imagine)					Can you tell me how much pain your child has had since yesterday?	0 (No pain) – 5 (The worst pain you can imagine)				
Q2.	How much have other problems with your body been troubling you since yesterday? *(Prompt only if needed:* e.g. being sick, going to the toilet a lot)?	0 (No other problems with my body have been troubling me) – 5 (Other problems with my body have been troubling me very much)					How much have other problems with their body been troubling your child since yesterday (*Prompt only if needed:* e.g. vomiting, diarrhoea, skin problems etc.)	0 (No other problems with their body have been troubling my child) – 5 (Other problems with their body have been troubling my child very much)				

Q3.	Can you tell me how much you have been feeding since yesterday?	0 (Not feeding at all) – 5 (Feeding enough)					Since yesterday, how much has your child been feeding?	0 (Not feeding at all) – 5 (Feeding enough				
Q4.	Can you tell me how much you have cried since yesterday?	0 (Not cried at all) – 5 (Cried all the time)					Since yesterday, how much has your child cried?	0 (Not cried at all) – 5 (Cried all the time)				
Q5.	Can you tell me how often you have felt happy since yesterday?	0 Happy all the time) 5 (Not happy at all)					Since yesterday, how much has your child felt happy?	0 (Happy all the time)- 5 (Not happy at all) –				
Q6.	How much have you felt like playing since yesterday?	0 (Felt like playing all the time) 5 (Have not felt like playing at all)					Since yesterday, how much has your child felt like playing?	0 (Felt like playing all the time) 5 (Have not felt like playing at all)				
Q7.	How much have your questions about your sickness been answered since yesterday?	0 (As much as I wanted) 5 (Have not been answered at all)					How much have your questions about your child's sickness been answered since yesterday?	0 (As much as wanted) 5 (Have not been answered at all)				

SECTION B. QUESTIONS ABOUT FAMILY / CARER (Note: The time period is since yesterday)						
Q8.	How much have you been feeling worried about your child's illness?	0 (Not at all worried) – 5 (Worried all of the time)				
Q9.	Have you been able to share how you are feeling about your child's illness with others when you have wanted to?	0 (Not at all) – 5 (Talked freely)				
Q10.	How much information have you and your family been given about your child's illness?	0 (None) – 5 (As much as wanted)				
Q11.	Have you had enough help and advice for your family to plan for the future with regards to your child's illness?	0 (None) – 5 (As much as wanted)				
Q12.	How confident does the family feel caring for the child?	0 (Not at all) – 5 (Very confident)				

Appendix C Belgian C-POS (English translation)

Section A: Regarding the CHILD. Please indicate 0, 1, 2, 3, 4, or 5 or "I don't know" for every question asked, thinking about the last few days.

	Question	*Answers*	*Question*	*Answers*
Q1	Can you tell me if you have had any pain?	0 1 2 3 4 5 No pain at all A lot of pain ○ I don't know	Has your child had any pain?	0 1 2 3 4 5 no pain at all Yes, a lot • I don't know
Q2	Have you experienced any problems in (with?) your body that have been bothering you? *(Suggest if necessary: vomiting, diarrhea, nausea, insomnia)*	0 1 2 3 4 5 0 No problems in my body 5 Problems in my body are bothering me • I don't know	Are they any physical problems that have been bothering your child? *(Suggest if necessary: vomiting, diarrhea, nausea, insomnia)*	0 1 2 3 4 5 0 No physical problems bothered him/her 5 Yes, a lot of physical problems bothered my child • I don't know
Q3	Is there anything about food that has been bothering you?	0 1 2 3 4 5 Nothing Yes, a lot • I don't know	Is there anything about food that has been bothering your child?	0 1 2 3 4 5 Nothing Yes, a lot • I don't know
Q4	Can you tell me if you have been sad?	0 1 2 3 4 5 Not at all Yes, very sad • I don't know	According to you, has your child been sad?	0 1 2 3 4 5 Not sad at all Very sad • I don't know
Q5	Can you tell me if you have been happy?	0 1 2 3 4 5 Not happy at all Very happy • I don't know	According to you, has your child been happy?	0 1 2 3 4 5 Not happy at all Very happy • I don't know
Q6	Have you had fun? Have you been playing?	0 1 2 3 4 5 Never Always • I don't know	Has your child had fun? Has he/she been playing?	0 1 2 3 4 5 Never Always • I don't know
Q7	Do you still have any questions (about your illness)?	0 1 2 3 4 5 No, none Yes, a lot • I don't know	Do you feel that your child still has some unanswered questions (about his or her illness)?	0 1 2 3 4 5 No, none Yes, a lot • I don't know
Q8	Do you sleep well?	0 1 2 3 4 5 Not at all Very good • I don't know	Does your child sleep well?	0 1 2 3 4 5 Not at all Very good • I don't know
Q9	When something bothers you, can you talk to someone about it?	0 1 2 3 4 5 No, never Yes, always • I don't know	When something is bothering your child, do you feel that he or she can express it to someone? (talk to someone about it?)	0 1 2 3 4 5 No, never Yes, always • I don't know
Q10	Do you have as much contact with friends as you would like?	0 1 2 3 4 5 No, never Yes, always • I don't know	Do you feel that your child has as many contacts with his/her friends/peers as he/she would like?	0 1 2 3 4 5 No never Yes, always • I don't know
Q11	If you had a magic wand, is there something you would you like to change in your family?	0 1 2 3 4 5 Not at all Yes, absolutely • I don't know	Do you feel that your child would like to change something in your family?	0 1 2 3 4 5 Not at all Yes, absolutely • I don't know
Q12	Do you feel loved? *this question is to be introduced cautiously, in the case the child is anxious	0 1 2 3 4 5 No, not at all Yes, absolutely • I don't know	Do you feel that your child feels loved?	0 1 2 3 4 5 Not at all Yes, absolutely • I don't know

Comments/observations: Section B. Questions to address only to the Parent "In the last few days, …"	
Q13 To what extent do your concerns (worries?) have an impact on your daily life, sleep, work...?	0 1 2 3 4 5 Not at all A lot • I don't know
Q14 To what extent can you share with others how you feel about your child's illness?	0 1 2 3 4 5 Not at all A lot • I don't know
Q15 Do you and your family receive as much information as needed (about your child's illness)?	0 1 2 3 4 5 Not enough at all Receive as much as needed • I don't know
Q16 To what extent do you receive enough help and advice to plan for your child's future?	0 1 2 3 4 5 Not enough at all Receive as much as needed • I don't know
Q17 To what extent do you feel confident about caring for your child?	0 1 2 3 4 5 Not confident at all Completely confident • I don't know
Q18 To what extent do you receive the support and guidance you need for yourself?	0 1 2 3 4 5 Not at all All the support needed • I don't know
Q19 To what extent do administrative procedures (equipment, treatment, care) represent a burden for you?	0 1 2 3 4 5 Not at all A lot • I don't know
Q20 Do you ever worry about the financial aspects linked to your child's illness?	0 1 2 3 4 5 Not at all A lot • I don't know
Q21 To what extent does the medical care you provide to your child represent an overload of work for you?	0 1 2 3 4 5 Not at all A lot • I don't know
Q22 How do you currently evaluate your own quality of life?	0 1 2 3 4 5 Very bad Very good • I don't know

Appendix D Systematic review protocol

Background and Rationale

When collecting data on health-related outcomes it is widely accepted that it is good practice to obtain children's self-report whenever possible. This is because health-related quality of life is generally understood as a latent, not directly observable construct, and it contains the perceptions and evaluations of someone's life from the subjective view of the individual, as well as the individual's subjective well-being and affective mood(439). Studies looking at correlation between child self-report of health-related quality of life and those of a parent/carer show a higher correlation for observable constructs such as physical symptoms, and a lower correlation for non-observable constructs, such as emotional concerns (362, 440). It has also been observed that children conceptualise quality of life differently for their parents/carers and clinicians. Patient reported outcomes developed for adults are unlikely to capture the realities of childhood or be sensitive to developmental change (245) and children's conceptualisation of health-related quality of life is likely to be different to that of adults.

Developing measures intended for child self-report comes with methodological complexities that need to be considered during the design stage. These include the types of response format children are able to use, appropriate recall period and the mode of administration of the measure (214, 302). Researchers need to be aware that chronological age may not always be the main consideration when judging whether children can self-report, due to variability in development and cognition. If a measure is being designed for use in a child or young person with a life-limiting or life-threatening (life-limiting and life-threatening) condition, development and cognition may be impaired due to the underlying illness, which adds further challenge (302).

A recent systematic review of health-related quality of life outcome measures that could be used in children with life-limiting and life-threatening illnesses identified 27 potential instruments and examined their feasibility of use and their psychometric properties. Their psychometric properties were analysed using the COSMIN methodological checklist(105). No measures scored at least 'fair' methodological quality or higher on all characteristics. Moreover, the domains, recall period and response format of included measures were not always considered appropriate for children with life-limiting and life-threatening conditions. Measures that were

included in the review had recall periods ranging from the 'current moment' to 4 weeks.

The mode of administration was predominantly pen and paper, with one measure validated for use over the telephone (328).

Research conducted in the 1980s and 1990s has demonstrated that children as young as 5 years old can self-report pain using an age-appropriate visual analogue scale (441, 442). However, pain is a single domain and health-related quality of life (HRQOL) is a subjective, multidimensional and dynamic construct that comprises physical, psychological and social functioning. Assessing health-related quality of life as an outcome involves considering the impact of a condition and its management on emotional, physical, social and spiritual functioning as well as lifestyle. Thus, HRQOL can be seen as a holistic and subjective construct. It is generally accepted that children can report on more concrete domains of health related quality of life between 4 and 6 years of age but more subjective concepts may be more difficult to comprehend and articulate(443).

Children as young as four years old have been shown to be able to validly and reliability self-report on their own HRQOL when using an age-appropriate measure (444), although elsewhere it has been suggested that children younger than seven years old do not have sufficient cognitive skills for this (445). Children aged 4–7 years are highly suggestible and will often give interviewers the responses they think they want to hear (252).

It has been shown that from the age of 8 years children can use 5-7 point Likert response scales (314) with younger children either being unable to use these or requiring fewer response options (314, 443, 446). There are also age differences in children's ability to report on their health within specific recall periods (314, 446). Given that HRQOL can change quite rapidly in a child or young person with a chronic or life-limiting and life-threatening condition, using measures with a shorter recall period may elicit more accurate responses (314).

When measuring outcomes in children and young people the mode of administration needs to be considered, so that children can be engaged in the process in a way that is acceptable to them. One large study using multi-group confirmatory factor analysis for invariance testing showed that items on the PedsQL$^{TM\ were}$ interpreted in a similar manner by children aged 5-18 years across three modes of administration; telephone, face to face and in the mail (328). With advances in computer and

mobile technology it is now possible to enhance patient reported outcome measures with graphical, video and audio content in order to improve both understanding and engagement (245). Using pictures that depict specific symptoms and activities has been shown to improve younger children's engagement in outcome reporting as well as their response reliability (447-449). It is imperative that attention is paid to completion methods and tool format at the measure design stage to reduce incompletion and non-response rates.

The aim of this review is to systematically appraise the evidence on response scale type, recall period, mode of administration and approaches to engage children in self-report for measuring patient-reported outcomes in children and young people up to the age of 18 years to inform measure development.

Aim and Objectives

Aim

To review the evidence on response scale types, recall period, appropriate mode(s) of administration and approaches to engaging children in self report when developing and implementing patient reported outcome measures for use with children and young people up to the age of 18 years.

Objectives

Identify the literature on response scale types, recall period, mode(s) of administration and approaches to enable participation in self-report for patient reported outcomes in children and young people up to the age of 18 years.

Extract data on response scale types, recall period, mode(s) of administration and approaches to enable participation in self-report for patient reported outcomes in children and young people up to the age of 18 years.

Appraise the quality of the evidence on response scale types, recall period, mode of administration and approaches to enable children to participate in self-report for outcome measures for use with children and young people up to the age of 18 years in terms of feasibility, acceptability and effect on measurement properties.

To synthesize the findings of the review and make recommendations on selecting response scale type, recall period, mode of administration and methods to enable self-report when developing patient-reported outcome measures for use by children

and young people, both those who are cognitively able and those who have learning and/or communication difficulties.

Identify gaps in the literature to make recommendations for future research.

Methods

Search Strategy

This review will be conducted and reported in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (207).

Databases to be searched

PsycINFO, Medline, CINAHL and Embase, cited by on Scopus of selected articles. In addition, cited references of selected articles will be searched and any key journals that are identified will be hand-searched.

Search Terms

Children

1 exp child/

2 exp p?ediatrics/

3 (child* or adolescen* or p?ediatric* or youth* or juvenile or teen* or young people or schoolchild* or school age* or kid*).ti,ab.

4 1 or 2 or 3

Response Scale format

5 (response scale or likert scale or visual analog* scale or VAS or numerical rating scale or verbal rating scale or faces scale or dichotomous scale or yes no response or response option*).ti,ab.

Recall period

6 (recall period or recall interval or patient recall or recall bias).ti,ab.

Method of administration

7 (outcome measure adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.

8 (measure adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.

9 (scale adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.

10 (questionnaire adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.

11 (survey adj2 (paper or (paper and pen) or tablet or tablet computer or app or application or telephone or face to face or internet)).ti,ab.

12 7 or 8 or 9 or 10 or 11

Combine the above

13 14 or 15 or 21

14 4 and 13 and 22

Exclusion criteria(450)

24 (addresses or biography or comment or directory or editorial or interview or festschrift or lectures or legal cases or legislation or letter or news or newspaper article or patient education handout or popular works or congresses or consensus development conference or practice guideline).pt

25 23 not 24

26 (limit to 1980-current; humans; English language; all child 0-18 years).

Inclusion Criteria

- Studies in children and young people up to 18 years old. If studies involve participants >18 years old then they will be included if data for those under 18 years is presented separately.
- Primary research
- Case reports ≥3 participants

- Studies looking at recall period, response scale selection, administration modality and approaches to enable children to self-report in terms of their effect on:
    - measurement properties and factor structure of instruments,
    - acceptability of use
    - feasibility of use.
- Studies written in the English language

Exclusion Criteria

- Review/systematic review articles
- Discussion articles
- Editorials, reports, letters
- Small case reports ≤3 participants
- Studies solely in those 18 years and over
- Written in a language other than English

Study selection

All retrieved articles will be transferred to EndNote version 8 and duplicates removed. The titles and abstracts of all retrieved articles will be screened for eligibility by LC. If there is not enough information within the title and abstract to determine eligibility, then the full text article will be screened by LC. Remaining full text articles will be screened by LC and 10% will be screened by a 2nd reviewer (CES). Any discrepancies throughout the process will be discussed with a third reviewer. Studies excluded at the full text stage will record the reason for exclusion.

Data extraction

Data from included articles will be extracted into an Excel spreadsheet and will include the following: title, authors, date, country, aim, study design, sample (including population, inclusion/exclusion criteria, size and setting), which measure characteristics the paper reports (recall period, response format, administration modality, approaches to engage children in self-report) and quality score.

Study quality assessment

For studies on measurement properties the COSMIN risk of bias checklist will be used (148). For studies using qualitative or quantitative methodology the ‘Standard

quality assessment criteria for evaluating primary research papers from a variety of fields (QualSyst)' will be used (211).

Analysis

Evidence will be tabulated in Excel and ordered by primary aim (i.e. whether the primary aim was to collect evidence on recall period, response format, administration modality or methods to enable participation in self-report). Results will also be synthesized narratively to discuss heterogeneity of included studies, similarities and differences in findings of selected studies and to explore patterns. Differentiation will be made across groups – such as those healthy children and those with learning disabilities. Recommendations will be made based on the quality of the evidence of included studies and the feasibility and acceptability of using different response scale types, recall periods and modes of administration when developing patient-reported outcome measures for use by children and young people.

Output

The results of this review will be submitted for publication in a peer reviewed journal and along with results of primary qualitative interviews, will also inform the development of the Children's Palliative Outcome Scale

Appendix E Reflective field note template

General

Setting (including things affecting depth/length of the interview):

Content (summary of key points in case something happens to the recording & to help think about saturation):

Reflections

How did it go?

My own emotions and reflections:

Key themes and reflections on saturation:

Any surprises?

Anything in line with what I expected?

Most memorable part of the interview?

Best interview question?

What I'd ask / ask differently next time?

How did my thoughts / attitudes change?

Other thoughts

Appendix F GOSH YPAG PPI Impact Case Report

5. The C-POS Study

What matters to children and young people facing serious illness

About the researchers and their research

The researchers are a unique multidisciplinary collaboration ; including Great Ormond Street Hospital and the main researcher that we are working with is a palliative care nurse and PhD student from the Royal Marsden Hospital.

The C-POS study is an exciting children's research study aiming to develop the first person-centred outcome measure. An outcome measure is a questionnaire that can be used in routine practice by children, young people and families affected by life-limiting and life-threatening conditions (LLLTC) which addresses the symptoms and concerns that matter most to them.

A recent systematic review has highlighted that no measures suitable for use in this population currently exist. In addition, development of such a measure has been highlighted as an international research priority. The researchers seek to engage children within the research process, rather than relying on proxy data.

So far the researchers have conducted interviews with:

- ill children
- their parents and brothers/sisters
- grown ups whose job is to look after them in hospital or at home

How does the research benefit patients?

The C-POS study addresses a current gap in both methods and evidence: repeated reviews and policies have called for scientific advancement to develop, validate and implement Person-Centred Outcome Measures (PCOMs) for Children and Young People and their families facing LLLTC.

The team will develop a person-centred outcome measure that can be used by children and youngpeople and their families affected by LLLTC, and to test its psychometric properties. They will also be developing implementation guidance once the measure is finalised.

Where in the Research Project Lifecycle did PPI take place?

PPI took place at the very early stages of questionnaire design and again at a second meeting to shape it further. PPI will be intrinsic throughout this project at every step going forwards.

Who were involved?

- GOSH Young Persons' Advisory Group (YPAG) for research and representatives from Southampton YPAG (at meeting 2)

Meeting 1:
July 2020 (25 young people attended)

Researchers consulted with the group on 3 things:

1. How they give children the questionnaire
2. How children should answer the questions
3. How far back you think children will be able to remember

I felt like it was both fun and important to consider how this questionnaire would be received by young and older children. YPAG had to think about how children would answer the questions and how far back the children could remember.

GOSH YPAG member Sandra

Meeting 2:
March 2021 (22 Young people attended)

Researchers consulted with the group on 2 things:

1. How should we label the versions (e.g., Version A , Version B, etc.) – children with serious illness have different levels of understanding so we don't want to use age to label the versions
2. What would be your top 10 questions from the list on the next slide?

What Impact has PPI had on the study?

The research team have already incorporated changes and chosen different ways of answering the questions based on your help from last time.

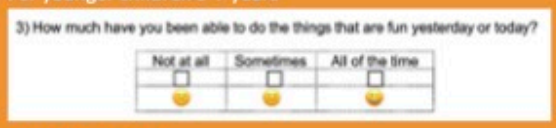
Example of YPAG feedback:

Do you prefer descriptors + numbers or the smiley faces?

- I think smiley faces is good but there is not enough options (3 is limited and not enough)
- Faces with more info would be better
- Faces is less confusing, the other one is more confusing, and I would struggle to choose one
- I think 5 faces are needed
- for no pain, / or for worst possible pain

Changes made:

For younger children 5-7 years



For older children

Feedback from Meeting 2

Label version ideas: could name after animals (panda, wolf, monkey, dogs – different types, polar bear, penguin, cats, tiger, lynx, dolphin, peacock, raven, hedgehog and bunny) or colours (warm colours for compete, gold, red, blue, yellow, pink, green, purple, sunset colours, orange blue in different shades).

Could also name after mystical creatures or Disney characters (merida) or cartoon characters (peppa pig) or star wars or marvel or movie characters (lion king, black panther, spiderman, the minions), different areas (rainforests, oceans, deserts, Antarctica) or hobbies (football, arts), movie series (harry potter, star wars) or seasons or emojis.

Have to be careful that different animals aren't associated with certain personalities because it might upset some children.

Green is a difficult colour as can be associated with sickness. Olive green could work. Blue is quite a calming colour.

What are the outcomes so far and what's happening in the future?

GOSH YPAG have featured in the C-POS Newsletter

GOSH YPAG representative has written blogpost for GenerationR website

GOSH YPAG representative will be prepared to attend an Ethics Committee Meeting with the team:

> We will have another phase of the study going to ethics later this year/early next year and would love for one of the group to join us for that
>
> *C-POS research team member*

Appendix G Distress protocols

Distress Protocol – C-POS qualitative interview study

Pre Interview **Explanation**	Explain to the participant (child/sibling/parent or caregiver or healthcare professional) that due to the sensitive nature of the research it is possible that they might find some questions / discussions upsetting and difficult to answer. Reiterate to the participant that: • They do not have to answer any questions that they do not want to answer. • They can say that they want to have a break at any time. • If they want to stop the interview, they can do so without giving any reason. When speaking to the participant's parent / carer, acknowledge that they too may find involvement upsetting. Explain that there is information available to signpost them to appropriate support.
During Interview **Recognising Distress**	A participant indicates that they are distressed or exhibits behaviours which would indicate distress – crying, becoming quiet, fidgeting, shouting, sudden change in their presenting behaviour, shaking, moving away from the researcher, seeking their parent / carer.
During Interview **Step 1 – Acknowledge Distress**	Stop the interview questions or discussion and acknowledge the distress e.g. 'I can see that talking about this is making you upset…'
During Interview **Step 2 – Offer to Stop the Interview**	Remind the participant that: they may stop for a break, stop the interview if they are finding it too upsetting, or withdraw completely from the study.
During Interview **Step 3 - Review**	• If the participant wishes to continue, carry on with the interview. • If the participant wishes to have a break, offer to give them some time or to return on another day (ensure there are drinks, crayons, paper and other small, age appropriate, play intervention activities available for the participant to engage in to help de-escalate their distress). • If the participant does not wish to continue, terminate the interview.
Post Interview **Step 4 – Offer Support**	At the end of the interview, or on terminating the interview, acknowledge again that the participant found the interview distressing.

	• Inform the participant that their parent / carer will be informed that they have been distressed during the interview. • Talk to the participant's parent / carer about the child / young person's distress. • Communicate back to the clinical team about the child / young person's distress. • Talk to the participant and their parent / carer, offer any required support and ensure that they are all comfortable before leaving. • Provide the participant/parent/carer with information about support services via Together for short lives links
Post Interview **Step 5 – Feedback –** **Discuss with senior research team**	After each interview, where the distress protocol has been used, discuss the interview with the Principal Investigator for the research, outlining: the cause of the distress, actions taken, any ongoing concerns, and agree any further action required.
Research Governance **Step 6 – Record in Research Log / Diary**	Record all instances where the distress protocol has been used in a research log or diary. These anonymised records will be used to inform the development of future studies and topic guides.

Distress Protocol – C-POS Cognitive Interview Study

Pre Interview **Explanation**	Explain to the participant (child/sibling or parent/caregiver) that due to the sensitive nature of the research it is possible that they might find some questions / discussions upsetting and difficult to answer. Reiterate to the participant that: • They do not have to answer any questions that they do not want to answer. • They can say that they want to have a break at any time. • If they want to stop the interview, they can do so without giving any reason. When speaking to the participant's parent / carer, acknowledge that they too may find involvement upsetting. Explain that there is information available to signpost them to appropriate support e.g., Together for Short Lives or via their clinical team.
During Interview **Recognising Distress**	A participant indicates that they are distressed or exhibits behaviours which would indicate distress – crying, becoming quiet, fidgeting, shouting, sudden change in their presenting behaviour, shaking, moving away from the researcher, seeking their parent / carer.
During Interview **Step 1 – Acknowledge Distress**	Stop the interview questions or discussion and acknowledge the distress e.g. 'I can see that talking about this is making you upset…'
During Interview **Step 2 – Offer to Stop the Interview**	Remind the participant that: they may stop for a break, stop the interview if they are finding it too upsetting, or withdraw completely from the study.
During Interview **Step 3 - Review**	• If the participant wishes to continue, carry on with the interview. • If the participant wishes to have a break, offer to give them some time or to return on another day (ensure there are drinks, crayons, paper and other small, age appropriate, play intervention activities available for the participant to engage in to help de-escalate their distress). • If the participant does not wish to continue, terminate the interview.

Post Interview **Step 4 – Offer Support**	At the end of the interview, or on terminating the interview, acknowledge again that the participant found the interview distressing and offer support as outlined above. • Inform the participant that their parent / carer will be informed that they have been distressed during the interview. • Talk to the participant's parent / carer about the child / young person's distress. • Communicate back to the clinical team about the child / young person's distress. • Talk to the participant and their parent / carer, offer any required support and ensure that they are all comfortable before leaving. • Provide the participant/parent/carer with information about support services via Together for short lives links
Post Interview **Step 5 – Feedback –** **Discuss with senior research team**	After each interview, where the distress protocol has been used, discuss the interview with the Principal Investigator for the research, outlining: the cause of the distress, actions taken, any ongoing concerns, and agree any further action required.
Research Governance **Step 6 – Record in Research Log / Diary**	Record all instances where the distress protocol has been used in a research log or diary. These anonymised records will be used to inform the development of future studies and topic guides.

Appendix H Summary of ethics amendments Phase 1

	Date sent	Categorisation	Description	Documents	Comments
1	06 March 2019	C	Minor correction to Professional information sheets (taken our mention of "your child" and PALS information)	Professional information sheet version 5.2	
2	11 April 2019	C	Alteration to the protocol so that clinicians can share information sheets with potential participants	Protocol version 5.2	
3	03 July 2019	A	Alteration to the protocol so that non palliative-care paediatric clinicians can approach participants who have a life-limiting or life-threatening illness but are not necessarily under a palliative care team	Protocol version 5.3	GOSH have not approved this amendment so will continue to work from protocol 5.2
4	07 August 2019	New sites	Alteration to the protocol to open new sites – East Anglia Children's Hospices, Martin House Children's Hospice, Cambridge University Hospitals NHS Foundation Trust and Leeds Teaching Hospitals Trust	Protocol version 5.4	
5	10 September 2019	A	No cost extension to recruitment. Change End date from 30th November 2019 to 30th March 2020	Statement of activities version 5.1	
6	24th September 2019	C	Correction to parent, child 16-18 and sibling 16-18 information sheet to explain that data will be given to KCL researchers (in line with IRAS and protocol) Changes to protocol to explicitly outline researchers receiving data are from KCL Minor correction to	Protocol version 5.5 Information sheet- Child 16-18 – 100919 – v5.2 Information sheet – Sibling 16-18 – 100919 – v5.2	

				Information sheet – Parent or caregiver being interviewed – 100919 – v5.2 Information sheet – Parent signing for child – 100919 – v5.2 Information sheet – Parent signing for sibling – 100919 – v5.2	
19 September 2019: Not official amendment- Confirmed via Ashley Totenhofer at HRA			*Corrections to typos in sibling facing documents, e.g. "your care" changed to "your sibling's care"*	*Same version numbers as originals as typos*	*Have kept email evidence*
Substantial - 1	6th December 2019	**Substantial**	Increase the recruitment upper limit for parents	Protocol version 5.6 Signed amendment form	
7	11th December 2019	New sites	Addition of Northern Ireland Children's Hospice	Protocol v5.7 041119	
8	9th December 2019	C	Rewording of parent signing for sibling information sheet deleting mention of palliative care	Information sheet – Parent signing for sibling 091219 – v5.3	
9	19th February 2020	A	No cost extension to recruitment. Change End date from 30th March 2020 to 30th September 2020	Rationale for extending recruitment	

10	16th March 2020	C	Changes to protocol following COVID 19 - Transcription off site - Phone call and video interviewing	Protocol v5.8 160320	*Due to COVID-19 this did not need to be approved by HRA instead KCL and KCH confirmed this was a category C amendment to be implemented immediately*
New process of submitting amendments via IRAS (from June 2nd 2020)					
11	29th May 2020	C	Changes to patient information sheets to reflect COVID-19 protocol changes - Transcription off site detailed - Mention of phone-call and video interviewing	Information sheet – Child aged 5-7 – 290520 – v5.2 Information sheet – Child aged 8-10 – 290520 – v5.2 Information sheet – Child aged 11-15 – 290520 – v5.2 Information sheet – Child aged 16-18 – 290520 – v5.3 Information sheet – Parent being interviewed – 290520 – v5.3 Information sheet – Parent signing for child – 290520 – v5.3 Information sheet – Parent signing for sibling – 290520 – v5.3	*Approved via the new IRAS system and email confirmation sent to sites*

				Information sheet – Professionals – 290520 – v5.3 Information sheet – Sibling aged 5-7 – 290520 – v5.2 Information sheet – Sibling aged 8-10 – 290520 – v5.2 Information sheet – Sibling aged 11-15 – 290520 – v5.2 Information sheet – Sibling aged 16-18 – 290520 – v5.3	

Appendix I Ethics application, approval and amendments for Delphi survey and item generation meeting

Minimal Risk Registration Form

Section A: Confirm that your study requires KCL ethical clearance

Before completing a Minimal Risk Registration Form, you may find it useful to read through the accompanying guidance.

IMPORTANT NOTICE RELATING TO COVID-19 – Whist researchers are permitted to register new projects involving face-to-face interactions, such data collection is currently not permitted to commence unless it falls under one of the exemptions and fulfils the criteria outlined by the College Research Ethics Committee at the link below:

https://internal.kcl.ac.uk/innovation/research/ethics/applications/COVID-19-Update-for-Researchers

1 Is your study considered research as defined in the guidance icon information?
Please note studies deemed to be either a service evaluation or audit do not require ethical clearance.

- (•) Yes
- ( ) No

2 Does your study require external ethical review by either the Health Research Authority (which includes the NHS REC and Social Care REC) or the Ministry of Defence REC?
See guidance icon for further information on the HRA and MOD REC ethical review remit.

- ( ) Yes
- (•) No

3 Please indicate which of the following data collection methods your study involves:
Please note: You should only select the methods that you are certain will be employed for the project and you are able to outline in section 20(d).

- [ ] Interviews
- [ ] Focus Groups/Workshop
- [x] Questionnaires/Surveys/App based research tool
- [ ] Non-interventional classroom observations
- [ ] All other non-interventional observations
- [ ] Physical procedures (e.g taking body temperature, wearing a virtual reality headset, taking pulse)

Please note: Analysis of pre-existing data is not eligible for the Minimal Risk Registration Process. Before continuing, if your study involves the analysis of pre-existing data, please visit our 'Analysis of pre-existing data' page for advice on whether or not you will need to complete a 'Full Application Form' for ethical approval

Project ID: 18826

Section B: Confirm that your study does not require High Risk review

6 Does your study present any of the following risks to participants?

- ☐ a) Vulnerability: Does the study involve participants who are vulnerable, lack capacity to give informed consent, or are in a dependent position (e.g. vulnerable children, people with learning difficulties, people with mental health problems, people with diminishing capacity to consent, young offenders, people in care facilities, offenders in prison)?
- ☐ b) Consent and deception: Will participants be asked to take part in the study without their informed consent or knowledge at the time or will deception of any sort be involved?
- ☐ c) Participant disclosures: Is there a risk that the highly sensitive nature of the research topic might lead to disclosures from the participant concerning their own involvement in illegal activities or other activities that represent a threat to themselves or others (e.g. sexual activity, drug use, or professional misconduct)?
- ☐ d) Stress and anxiety: Could the study induce psychological stress or anxiety, or produce humiliation or cause harm or negative consequences beyond the risks encountered in a participant's usual, everyday life?
- ☐ e) Urgent mental health risks: Participation in this research may identify urgent mental health risks, including, but not limited to, suicidal ideation and/or self-harm intent.

You should only select the statement below if you have not selected any of the above. Your application will be invalid if you select the below statement in addition to any of the above.

- ☑ I have answered no to all questions in the risk checklist above and I do not believe that my research is high risk

Section C: Confirm that your study does not require Low Risk review

7 Will any participants be under the age of 16?

- ○ Yes
- ◉ No

8 Will you be asking participants to disclose any information of a personally sensitive nature that you cannot assume they would be otherwise willing to discuss in public? *See information icon for further guidance.*

- ○ Yes, I will be asking participants to disclose personally sensitive information
- ◉ No, I will not be asking participants to disclose any personally sensitive information

9 Do you have a current or prior relationship with any potential participants? (This includes professional and/ or personal relationships)

- ○ Yes, I do have a current or prior relationships with potential participants.
- ◉ No, I do not have any current or prior relationships with potential participants.

10 Gatekeeper Permission: Will you require an individual or organisation to grant you permission to approach/ access your intended participants? This includes gatekeepers contacting participants on your behalf.

- ◉ Yes, I will be using a gatekeeper to access potential participants
- ○ No, I will not be using a gatekeeper to access potential participants

10a Will any gatekeeper used be in a position of authority over potential participants?

- ○ Yes, the gatekeeper is in a position of influence or authority over participants
- ◉ No, the gatekeeper is not in a position of influence or authority over participants

11 Can you confirm that the participants will not be subjected to any undue incentives to participate? (Receipt of a project report or a reasonable reimbursement such as travel/lunch costs or a voucher would not be considered undue incentives.)

- ○ Yes, participants will be subject to undue incentives
- ◉ No, participants will not be subjected to any undue incentives

12 You have determined that your project is eligible for minimal risk registration. Before completing your minimal risk registration form you must confirm that you have read the Minimal Risk Guiding Principles at the link below and that you will adhere to these principles in the conduct of your research.

Minimal Ethical Risk Guiding Principles

☑ I confirm that I have read the Minimal Risk Guiding Principles and will adhere to the principles within.

Section D: General Information

13 Applicant Details

Title	First Name	Surname
Mrs	Lucy	Coombes

Department: Palliative Care, Policy and Rehabilitation

Email: lucy.coombes@kcl.ac.uk

14 Faculty/Institute/School

Please refer to the information icon if you are unsure of your Faculty/Institute/School.

Nursing and Midwifery

15 Applicant Status

MPhil / PhD / Special Doctorate

16 Supervisor Details

Title	First Name	Surname
Professor	Richard	Harding

Department: Palliative Care, Policy and Rehabilitation

Email: lucy.coombes@kcl.ac.uk

17 Are there any other investigators/collaborators involved in the study?

○ Yes
◉ No

Section F: Study Overview

18 Project Title

A working title for your project that accurately reflects its aims

Children's Palliative Outcome Scale (C-POS) - Delphi survey and item generation meeting

19 Anticipated start date for the collection of data: 07/09/2020

20 Study Overview: Briefly describe your research project, as you would to a potential participant, highlighting the aims of your research, who your participants are (type of people rather than individual names), how you will recruit them and what will be asked of them (try to use no more than 50 words for each section).

a) What are the aims of your study?

You should explain what the principal research question is and the specific objectives of the study

The aim of this study is to gain consensus on items to be included in C-POS, which is a child and family centred outcome measure for use in paediatric palliative care. The objectives are:
a) To conduct a Delphi survey to obtain stakeholder consensus on items to be included in the C-POS measure, thus further establishing face and content validity. The content of this Delphi survey will be informed by previously conducted qualitative interviews with children and young people with life-limiting conditions and their parents/carers and siblings and a systematic review of symptoms and concerns in this population. Health care professionals and NHS commissioners were also interviewed about their priorities for outcomes of care in this group.
b) To conduct an item generation meeting to finalise initial parent/carer and age-appropriate child and young person versions of the C-POS.

b) Who are your potential research participants?

This should outline any specific criteria participants must meet in order to be eligible take part (e.g. age, ethnicity, gender, members of a specific group etc.)

Delphi Survey

• Health care professionals:
Doctors - who have been providing care to CYP with LLC/LTC for > 6 months.
Nurses - who have been providing care to CYP with LLC/LTC for > 6 months.
Social workers - who have been providing care to CYP with LLC/LTC for > 6 months.
Psychologists - who have been providing care to CYP with LLC/LTC for > 6 months.
Other allied health professionals e.g., physiotherapists, play specialists, occupational specialists, spiritual care providers - who have been providing palliative care to CYP with LLC/LTC for > 6 months.
• Parents/carers of a CYP aged 0-18 years (up to their 18th birthday) who have a LLC/LTC.
• Bereaved parents of CYP with LLC/LTC whose child has died within 12-24 months of participation.

Exclusion criteria
o Health care professionals who have been working with CYP with LLC/LTC for less than 6 months.
o Individuals who cannot complete an online survey written in English.

The item generation meeting will include members of the study steering group, including existing PPI representatives.

c) How will participants be recruited?

This should address how participants will be identified and approached in the first instance. For example, if approaching potential participants in person you should explain the circumstances in which this will take place or if approaching potential participants by email you should explain how you will obtain email addresses.

Recruitment and sampling technique
The Delphi survey will involve two key stakeholder populations – parents/carers of CYP with LLC/LTC and healthcare professionals who care for children with LLC/LTC. Both are considered experts in the symptoms and concerns that CYP with LLC/LTC experience. This will further enhance the content validity of C-POS.

Parent/carer participants will be identified through:
• Together for Short Lives (a leading UK charity for children with life-threatening & life-limiting conditions):
-Database of 140 parent experts who want to be involved in research – the charity will email members a link to the survey.
-Link to survey on quarterly newsletter
• Parent groups at the Royal Marsden – groups will be emailed information regarding the study along with a link to the survey web page.
• Martin House Research Centre family advisory board – via email link.
• Martin House and Northern Ireland children's hospices – email link via family Facebook page.
• Social media – a link to the survey will be shared on the study's Twitter feed.

Health care professional participants will be identified through:
• Association for Paediatric Palliative Medicine (APPM) – medical and nursing membership will be emailed a link to the survey via the APPM. The research team will not have access to individual contact details.
• Together for Short Lives:
-All children's hospices, hospital and community children's palliative care teams are members - the charity will email members a link to the survey.
-Other health care professional members - the charity will email members a link to the survey.
• The link will be sent to the Principal Investigators of the sites used for the previous qualitative interviews with a request that they disseminate to their teams and contacts (Royal Marsden Hospital, Evelina Children's Hospital, King's College Hospital, Great Ormond Street Children's Hospital, Cambridge University Hospital, East Anglia Children's Hospice, Northern Ireland Children's Hospice, Leeds Teaching Hospital Trust, Martin House Children's Hospice).
• Association of Palliative Care Social Workers – via email link.
• Children's Cancer Network – via email link.
• Children's Cancer and Leukaemia Group – via email link.
• Hospice UK membership – via email link.
• Social media – a link to the survey will be shared on the study's Twitter feed.

d) What will participation involve?

Briefly explain how each data collection method, indicated in Section 3, will be used to collect data. This should include what participants will be asked to do and an example of the types of questions they may be asked.
Please note: Failure to address each method indicated in Section 3 will result in your form being invalidated.

The Delphi study is a repeated online survey (3-4 rounds proposed). Items to be included in the survey will be symptoms, concerns and priorities for care identified from the previous qualitative interview study and from a recent systematic review on symptoms and concerns in children and young people (CYP) with life-limiting/life-threatening conditions (LLC/LTC). A matrix has been created by the research team that demonstrates the evidence source for each item included.

Study Procedure
A 3-4 round survey will be designed using SmartSurvey (an online survey platform). The survey and instructions will be piloted in a small group (3-5) of participants from the target population before use to ensure that the questionnaire and instructions are understood. The SmartSurvey platform has been chosen for its ease of use and ability to export data which is encrypted and stored within the UK. The research team will not have direct contact with potential participants.

The first page of the online survey will explain what the survey is for and how the survey items were decided upon. Reminders for each round will be sent after 1 week to maximise participation. Participants will be asked to share the link with others who could be eligible to participate in order to maximise the expertise of the panel. Demographic data will be collected as follows:
• Health care professionals – profession, current role, length of CYP palliative care experience, generic place of work e.g., hospital, hospice, community, gender and age.
• Parents/carers – age, relationship to the child, child's diagnosis, child's age, the ethnic background of child and parent/carer, the gender of the child and parent/carer and area of the country they live in.

Narrowing down (round 1)

Items for inclusion in round 1 of the Delphi will be chosen from:
1. Symptoms and concerns in CYP with LLC/LTC identified in the research team's previous qualitative interview study with CYP with LLC/LTC, their parents/carers and siblings, health care professionals and NHS commissioners.
2. Results of a systematic review of symptoms and concerns in CYP with LLC/LTC.

Symptoms and concerns will be presented in random order. Participants will be asked to select the 20 symptoms and concerns that they believe to be the most important to be included in the C-POS. They will also be asked to suggest items that are missing and be asked to justify and explain their choices in a free text box. Items will be selected for inclusion in subsequent rounds as follows:
• Items that are selected by >50% of participants will be included in the subsequent ranking rounds.
• If more than 30 items are selected by >50% of participants then the items selected by >30% of participants will be included in subsequent rounds(44).
• If new items are suggested during round 1, they will be compared with the existing items and discussed by the research team and members of the steering group (including PPI representatives) to gain expert consensus on whether they should be added to round two for evaluation by participants.

For this phase of the study, results will be analysed by participant group (parents/carers and professionals and by both groups combined). At this stage of the study, an academic and PPI steering group meeting will be held to review the data and make decisions on items to be carried forwards to the next round of the Delphi survey. This will include discussion of the suggested new items and free text justification of choices. This meeting will ensure that if there are differences between priorities identified by parents/carers and health care professionals these are reconciled by an expert panel.
Participant email addresses will be collected (with consent) during this round so that participants can be sent invitations to participate in further rounds. These will be kept in a password-protected file on a secure server.

Ranking (rounds 2 and 3)
Round 2 will occur 1-2 weeks after the close of round 1 and will be open for 2 weeks. Round 3 will occur a 1-2 weeks after round 2 closes and again will be open for 2 weeks. Participants in rounds 2 and 3 will need to have participated in round 1. Participants will be presented with the results from the previous round outlining items that were removed for round 2, any relevant comments from participants and any new items that have been added. Participants will then be asked to rank the symptoms and concerns remaining from round 1 in order of priority for inclusion in the C-POS measure. Items will be ranked in descending order, from the most to least important. Participants will be asked to explain their justifications for their rankings in a free text box. Reminders will be sent at 1 week.

For round 3, participants will again be sent an email with a link to the survey. They will be given the following feedback from the previous round:
• the median rank of each item from round 2 and where they ranked each item,
• Kendall's W coefficient of concordance (in layman's terms i.e. weak, moderate or strong agreement),
• top half rank (the percentage of experts who ranked items in their top 50%)
• relative comments/justifications made by respondents.

Results will be presented for the participants in full (parent/carer and professional data) and stratified by group. Participants will again be asked to rank symptoms and concerns based on the feedback above. This time items to be ranked will be presented according to median rank rather than randomly in order to aide the achievement of consensus. Participants will again be asked to justify their ranking decision in a free text box. Participants will be asked a final question on whether they would be willing to participate in a further round if consensus has not been reached. Reminders will be sent again at 1 week.

Data will be analysed in the same way as round 2. If consensus has been reached (Kendall's W >0.7) then the study will stop. If consensus has not been reached (W<0.7), a McNemar test will be performed to see whether there is a difference in median ranking between rounds 2 and 3. If there is no significant difference in median rankings between the two rounds the study will stop(52). If consensus has not been reached and there is a significant difference in median ranking between rounds a further round will be considered if >50% of respondents indicate they would be willing to participate in another round. If <50% of respondents were willing to take part in a fourth round, the study will stop. In palliative care, perfect agreement may often not be realistic due to different values, world views and ethical dilemmas concerning medical decision making. There are a diverse range of LLC/LTC that affect CYP and they are cared for in a wide range of settings which adds to this complexity. Results from this Delphi exercise will be taken forwards to an item generation to gain further expert agreement on items to be included in the C-POS in order to further evidence face and content validity.

This study will be reported according to guidance on Conducting and REporting of DElphi Studies in palliative care CREDES and recommendations by Paré et al on ensuring rigour in ranking-type Delphi surveys. All data will be anonymised.

Sample of items to be ranked in the Delphi:
Physical concerns:
1. Skin issues
2. Bowel problems
3. Appetite
4. Being physically unable to eat or drink as much as normal
5. Changes in weight
6. Infections and/or impaired immunity
7. Impact of medical interventions and treatment such as central lines, prosthesis
8. Muscle weakness
9. Impaired growth
10. Reduced physical function
11. Changes in consciousness

12. Dystonia
13. Seizures
14. Pain
15. Breathing difficulties
16. Cough
17. Excess respiratory secretions
18. Sleeping difficulties
19. Tiredness or fatigue
20. Appearance
21. Low blood counts
22. Impaired growth
23. Agitation
24. Nausea
25. Vomiting
26. Ability to do usual self-care
27. Side effects of medications
28. Fertility concerns
29. Hair loss

Social and practical concerns
1. Having access to technology to stay connected
2. Opportunity to discuss advance care planning and/or resuscitation
3. Discussion of preferred place of care
4. Discussion of preferred place of death
5. Discussion of wishes for life/death
6. Opportunity to set individual goals or outcomes
7. Avoiding unplanned hospital admissions
8. Identifying wishes for life
9. Care needs changing over time
10. Balancing needs of child with rest of family
11. Burden of medication regime
12. Communication difficulties – child
13. Communication with health and social care professionals
14. Eating
15. Friendship – adaptation to dynamics and challenges due to illness
16. Impact on play and hobbies
17. Impact on school life
18. Financial concerns –benefits, cost of hospital visits and stays, equipment etc
19. Impact on work life
20. Information provision – about illness, services, medication, developmentally appropriate, understanding illness, not having enough information
21. Meeting cultural needs
22. Access to 24/7 care at home
23. Access to 24/7 telephone advice
24. Care co-ordination
25. Quality of care
26. Access to equipment
27. Access to holistic care
28. Timely hospital discharge
29. Transitions – school, services
30. Fluctuating needs
31. Logistics of care e.g., organising appointments/carer rotas
32. Decision making
33. Discord between family members
34. Discord between service providers and family
35. Having to do things differently – eating, play/hobbies, routines, family life
36. Housing
37. Wanting to get back to normality
38. Adjusting to a new normal
39. Relationships
40. Equal opportunities with other children
41. Achieve life goals
42. Initiate and maintain sexual relationships
43. Breaking bad news
44. Shared decision making
45. Missing home

Spiritual and existential
1. Determination to live life to the fullest
2. Determination to survive
3. Meaning of life

4. Religion and faith
5. Spiritual support – access and availability
6. Uncertainty of the future – fear of life unlived, length of life
7. Worry about death
8. Will I be remembered
9. Am I dying?
10. Life devoid of meaning
11. Connected with God or something larger than self
12. Appreciate life as a gift
13. Keeping the spirit alive
14. Must survive the hard bits of illness

Emotional and psychological
1. Behaviour regression
2. Being different
3. Looking different
4. Challenges to independence
5. Problems with cognitive ability and learning needs
6. Loss of control (child)
7. Parents wanting to have control over their child's health
8. Impaired ability to express and/or understand emotions and feelings
9. Opportunity to meet people in a similar situation
10. Privacy
11. Grumpy/moody
12. Anger
13. Feeling like a burden
14. Interruption to family life
15. Worry about family
16. Wanting to protect family
17. Fear, worry and anxiety
18. Irritation or annoyance
19. Low mood, sadness
20. Memory making
21. Support for family members
22. Trust
23. Access to/availability of psychological support
24. Treatment related anxiety and worry
25. Happiness
26. Feeling stupid
27. Declining school performance
28. Reduced concentration
29. Lack of self-worth
30. Aggression
31. Non-adherence to treatment

Item generation meeting
In order to generate the final items for the C-POS, an item generation meeting will be held after the Delphi survey to further gain expert stakeholder input to enhance face and content validity of the C-POS. The C-POS steering group (including PPI representatives) will be given a brief presentation on the use of patient-reported outcome measures in healthcare and the evidence for the need of the C-POS. They will then be presented with the findings from:
• The two systematic reviews carried out by the research team on a) symptoms and concerns in children and young people with LLC/LTC(16) and b) enhancing validity, reliability and participation in self-reported health outcome measurement for children and young people: a systematic review of optimal recall period, response scale format and administration modality.
• Feedback from the Young Person's Advisory Group at Great Ormond Street Children's Hospital on response format, recall period and administration mode of outcome measures for CYP.
• The results of the main findings from the qualitative interview study on symptoms and concerns in CYP with LLC/LTC including the analysis framework.
• The results of the above Delphi survey including item mean rank (overall and stratified by participant group), Kendall's W, the percentage of participants placing each item in the top half of ranked list and any free-text comments provided by participants. Item mean rank will also be presented by domain. Domains were created using the World Health Organisation's definition of paediatric palliative care (see appendix 1 for items) (55) and the results of the previously conducted qualitative interviews. The domains are physical, social and practical, emotional and psychological, spiritual and existential and normality.

After the presentations, the symptoms and concerns ordered by domain and mean rank from the Delphi will be presented by the PhD candidate (LC) and discussed with the stakeholder group to gain final agreement for which items to include in the C-POS. Discussion will include whether consensus was reached during the Delphi and any disparities between groups (parent/carer and health care professionals) in order to reconcile any differences. Any disagreement will be worked through as a group with the PI (RH) acting as chair.
Once items for inclusion have been agreed there will be a discussion regarding response format, recall period and administration mode(s) of the C-POS, led by the study PhD candidate and based on discussions with the Great Ormond Street Young Person's Advisory Group and the results of the aforementioned systematic review of evidence of these in children. A presentation will be given

on suggestions for these. There will be an opportunity for discussion of these suggestions, again chaired by RH. These will be agreed for both parents/carers and child age. PPI representatives will be briefed before the meeting on the content and will be offered support if needed afterwards, either by discussion with the research team or by referral to the Together for Short Lives advice line.
After the meeting, LC (PhD candidate) will take the list of agreed symptoms and concerns for inclusion and draft the first parent/carer and age-appropriate versions (version1) of C-POS. Response format, mode and recall will be based on the systematic review results and outcomes of the stakeholder group discussion. These will be reviewed and finalised by the research team to ensure they represent the discussion held in the stakeholder group. A summary of the stakeholder group discussions will be sent electronically to the group participants along with version 1 (parent/carer and child versions) of the C-POS for final comment. Minor changes will be made, if required, prior to the next phase of the overall C-POS study (cognitive interviews, subject to a subsequent ethics application via IRAS).

21 Confirm which of the following consent processes will be used:

- [x] Written Consent: A written description of the research will be provided to all potential participants and written consent will be recorded in either paper or electronic form in advance of participation.
- [ ] Verbal Consent: I am able to demonstrate that written information and consent is not practical, or not appropriate, so I confirm that I will follow College guidance on providing information and gaining consent from participants verbally.
- [ ] Anonymous submission of survey/questionnaire/app based research tool data: A written description of the research will be provided to all potential participants and it will be made clear that the submission of a completed survey/questionnaire/app data implies consent.
- [ ] Non-invasive observations that do not involve any interaction with participants and no identifying information will be recorded.

(•) Provisions for written consent: I confirm that I will follow the KCL Information Sheet and Consent Form templates and I will provide appropriate researcher contact details for purposes of questions, complaints or withdrawal requests.
KCL guidelines and for participant recruitment templates

22 Does the project involve the collection and/or use of personally identifiable information at any point during the research project?
Personally identifiable information is any data collected during the course of the project which could potentially lead to the identification of an individual. Please see the guidance icon for examples of the type of data which could be considered personally identifiable. ***Please note: this includes audio recordings such as interview recordings.***

Please indicate which of the following applies:

( ) a) The project involves the collection and/or use of personally identifiable information
(•) b) Personally identifiable information will only be obtained in order to contact potential participants (e.g. using phone numbers/ email addresses) but no identifiable information will be collected beyond this point and it will not be possible to link any research data to participants.
( ) c) No personally identifiable data will be collected and/or used for this project.

22b Please indicate which of the following applies:

(•) The personal data used for recruitment purposes will not be linked to the anonymous data collected from participants and will not be held for any longer than is necessary for the purposes of recruitment.
( ) The identifiable information used for recruitment purposes will be linked to the data collected from the corresponding participant/s (this includes linking participant details to raw data for withdrawal purposes).

(•) I confirm that I understand that it is the responsibility of the researcher to ensure that all research data is appropriately handled and stored during and after the study in compliance with College guidelines:
KCL Research Data Management Guidelines

Section G: Declaration and Signatures

23 Researcher/Applicant Declaration:
IMPORTANT NOTE FOR STUDENTS: Please ensure that you have signed the form in this section before requesting your Supervisor's signature in section 24 below.

By signing this form I confirm the following:

- The information supplied above is to the best of my knowledge accurate.
- I have read the Minimal Ethical Risk Guiding Principles and clearly understand my obligations and the rights of participants, particularly as regards obtaining informed consent.
- The participant selection and recruitment procedures, including the recruitment documents to be provided and the manner of obtaining informed consent, are appropriate and the ethical issues arising from the project have been considered (and agreed with my Supervisor if applicable).
- I understand that I must not commence research with human participants until I have received confirmation of minimal risk registration.

Please note that in order to authorise your application you must sign off using your KCL email address i.e. joe.bloggs@kcl.ac.uk and your KCL email password.

24 Supervisor Declaration:

Once you have electronically signed the form in section 23, you should then request your Supervisor's signature using the below 'Request' button. Once your Supervisor has also signed the form the form will be automatically submitted for registration.

Please note: Following submission for registration you must wait until you have received a letter from REMAS confirming that your project has been registered before commencing data collection.

Request Supervisor's Signature:

Appendix I Ethics application, approval and amendments for Delphi survey and item generation meeting

Research Ethics
Office

Franklin Wilkins Building
5.9 Waterloo Bridge Wing
Waterloo Road
London SE1 9NH
Telephone 020 7848 4020/4070/4077
rec@kcl.ac.uk

KING'S College LONDON

20/08/2020

Lucy Coombes

Dear Lucy

Children's Palliative Outcome Scale (C-POS) Phase 2 - Delphi survey and item generation meeting

Thank you for submitting your Minimal Risk Self-Registration Form. This letter acknowledges confirmation of your registration; your registration confirmation reference number is MRSP-19/20-18826

IMPORTANT CORONAVIRUS UPDATE: In light of the COVID-19 pandemic, the College Research Ethics Committee has temporarily suspended all primary data collection involving face to face participant interactions, unless the data collection fall under one of the exemptions and fulfils the criteria outlined by CREC at the link below:

https://internal.kcl.ac.uk/innovation/research/ethics/applications/COVID-19-Update-for-Researchers

Ethical clearance for this project is granted. However, the clearance outlined in the attached letter is contingent on your adherence to the latest College measures when conducting your research. Please do not commence data collection until you have carefully reviewed the update and made any necessary project changes.

Ethical clearance is granted for a period of **three years** from today's date and you may now commence data collection. However, it is important that you have read through the information provided below before commencing data collection:

As the Minimal Risk Registration Process is based on self-registration, your form has not been reviewed by the College Research Ethics Committee. It is thereforeyour responsibility to ensure that your project adheres to the Minimal Risk Guiding Principles **and the agreed protocol does not fall outside of the criteria for Minimal Risk Registration. Your project may be subject to audit by the College Research Ethics Committee and any instances in which the registration process is deemed to have been used inappropriately will be handled as a breach of good practice and investigated accordingly.**

Record Keeping:

Please be sure to keep a record of your registration number and include it in any materials associated with this research. It is the responsibility of the researcher to ensure that any other permissions or approvals (i.e. R&D, gatekeepers, etc.) relevant to their research are in place, prior to conducting the research.

In addition, you are expected to keep records of your process of informed consent and the dates and relevant details of research covered by this application. For example, depending on the type of research that you are doing, you might keep:

- A record of all data collected and all mechanisms of disseminated results.
- Documentation of your informed consent process. This may include written information sheets or in cases where it is not appropriate to provide written information, the verbal script,or introductory material provided at the start of an online survey.
  Please note: For projects involving the use of an Information Sheet and Consent Form for recruitment purposes, please ensure that you use the KCL GDPR compliant Information Sheet & Consent Form Templates
- Where appropriate, records of consent, e.g. copies of signed consent forms or emails where participants agree to be interviewed.

Audit:

You may be selected for an audit, to see how researchers are implementing this process. If audited, you and your Supervisor will be asked to attend a short meeting where you will be expected to explain how your research meets the eligibility criteria of the minimal risk process and how the project abides by the general principles of ethical research. In particular, you will be expected to provide a general summary of your review of the possible risks involved in your research, as well as to provide basic research records (as above in Record Keeping) and to describe the process by which participants agreed to participate in your research.

Remember that if you at any point have any questions about the ethical conduct of your research, or believe you may have gained the incorrect level of ethical clearance, please contact your supervisor or the Research Ethics Office.

We wish you every success with your project moving forward.
With best wishes,

The Research Ethics Office

On behalf of the College Research Ethics Committee

Modification Request Form For Minimal Risk Registered Studies

Modification Details

Researchers should complete this form in order to register any significant changes to their research aims and objectives, data collection methods, participant groups, recruitment methods or consent process that were not outlined in their original minimal risk registration form.

If the proposed changes to the study mean that you should change you answers to any of the low risk or high risk criteria question in your original Minimal Risk Registration Form, a modification request cannot be submitted and a new 'Full Application Form' for ethical approval must be submitted.

Registration extensions are granted for a period of one year and should be sought before the original period of registration has ended. If the original registration has already expired, researchers will need to provide assurance that no data collection has been conducted beyond the original registration period and explain why an extension request could not be submitted before the original registration period expired.

1. Provide details of the type of modification proposed. Select all that apply.

- ☐ Study title
- ☐ Changes to the research aims & objectives
- ☑ Changes to recruitment methods
- ☐ Changes to research methods
- ☐ Changes to participant group(s)
- ☐ Extension to period of registration
- ☐ Changes to the consent process/ how study information is disseminated
- ☐ Additional researchers
- ☐ Other

2 Provide details of the modification:

We would like to add the following sites to our recruitment:
1. Northern Ireland Children's Hospice - to post a link to the survey on their parents Facebook page
2. Chestnut Tree House Children's Hospice - to share the link to the survey with parents/carers under their service via email and social media
3. Demelza Children's Hospice - to share the link to the survey with parents/carers under their service via email and social media
4. Glasgow Royal Hospital for Children and Children's Hospices Across Scotland - to share the link to the survey with parents/carers under their service via email and social media
5. Welsh Network for Children's Palliative Care - members will be asked to share the survey link with parents/carers they work with via word of mouth, social media or email.

Project ID: 18826

3. Provide a brief justification for the modification

Our original survey had a fairly poor response from parents and the demographic was 100% white British. All parents were from England, predominantly East England. We would like to get a UK wide perspective with more varied demographics. Therefore, we would like to reopen the first round of the survey for parents/carers and try to target those in different regions with a more varied ethnic background.

Signature

3. I confirm that I have revisited the Minimal Risk Registration criteria and the changes I have outlined above do not mean that my study is no longer eligible for Minimal Risk Registration. The information in this form is accurate to the best of my knowledge and I take full responsibility for it. I consider that it would be reasonable for the proposed modification to be implemented.

Signed: This form was signed by Lucy Coombes (lucy.coombes@kcl.ac.uk) on 07/10/2020 16:16

Appendix I Ethics application, approval and amendments for Delphi survey and item generation meeting

Research Ethics
Office

Franklin Wilkins Building
5.9 Waterloo Bridge Wing
Waterloo Road
London SE1 9NH
Telephone 020 7848 4020/4070/4077
rec@kcl.ac.uk

KING'S College LONDON

14/10/2020

Dear Lucy ,

Reference Number: MRM-19/20-18826
Study Title: Children's Palliative Outcome Scale (C-POS) Phase 2 - Delphi survey and item generation meeting

Confirmation of Registration for Minimal risk Modification Request

Thank you for submitting a modification request for the above study. This is a letter to confirm that your request has now been registered and you may continue with your study. If you have any questions regarding your application please contact the Research Ethics Office at rec@kcl.ac.uk.

Kind regards

Miss Elizabeth Chuck

Research Ethics Office

Appendix J Ethics application, approval and amendments for cognitive interview study

IRAS Form Reference: IRAS Version 5.19

Welcome to the Integrated Research Application System

IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please complete the questions in order. If you change the response to a question, please select 'Save' and review all the questions as your change may have affected subsequent questions.

Please enter a short title for this project (maximum 70 characters)
C-POS cognitive interview study

1. Is your project research?

(●) Yes ( ) No

2. Select one category from the list below:

( ) Clinical trial of an investigational medicinal product
( ) Clinical investigation or other study of a medical device
( ) Combined trial of an investigational medicinal product and an investigational medical device
( ) Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
( ) Basic science study involving procedures with human participants
(●) Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
( ) Study involving qualitative methods only
( ) Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
( ) Study limited to working with data (specific project only)
( ) Research tissue bank
( ) Research database

If your work does not fit any of these categories, select the option below:

( ) Other study

2a. Please answer the following question(s):

a) Does the study involve the use of any ionising radiation? ( ) Yes (●) No

b) Will you be taking new human tissue samples (or other human biological samples)? ( ) Yes (●) No

c) Will you be using existing human tissue samples (or other human biological samples)? ( ) Yes (●) No

3. In which countries of the UK will the research sites be located?*(Tick all that apply)*

☑ England

☐ Scotland
☐ Wales
☐ Northern Ireland

3a. In which country of the UK will the lead NHS R&D office be located:

◉ England
○ Scotland
○ Wales
○ Northern Ireland
○ This study does not involve the NHS

4. Which applications do you require?

☑ IRAS Form
☐ Confidentiality Advisory Group (CAG)
☐ Her Majesty's Prison and Probation Service (HMPPS)

Most research projects require review by a REC within the UK Health Departments' Research Ethics Service. Is your study exempt from REC review?

○ Yes ◉ No

5. Will any research sites in this study be NHS organisations?

◉ Yes ○ No

5a. Are all the research costs and infrastructure costs (funding for the support and facilities needed to carry out the research e.g. NHS support costs) for this study provided by a NIHR Biomedical Research Centre (BRC), NIHR Applied Research Collaboration (ARC), NIHR Patient Safety Translational Research Centre (PSTRC), or an NIHR Medtech and In Vitro Diagnostic Co-operative (MIC) in all study sites?

Please see information button for further details.

○ Yes ◉ No

Please see information button for further details.

5b. Do you wish to make an application for the study to be considered for NIHR Clinical Research Network (CRN) Support and inclusion in the NIHR Clinical Research Network Portfolio?

Please see information button for further details.

◉ Yes ○ No

The NIHR Clinical Research Network (CRN) provides researchers with the practical support they need to make clinical studies happen in the NHS in England e.g. by providing access to the people and facilities needed to carry out research "on the ground".

If you select yes to this question, information from your IRAS submission will automatically be shared with the NIHR CRN. ***Submission of a Portfolio Application Form (PAF) is no longer required.***

6. Do you plan to include any participants who are children?

(●) Yes ( ) No

7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?

( ) Yes (●) No

Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the Confidentiality Advisory Group to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.

8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?

( ) Yes (●) No

9. Is the study or any part of it being undertaken as an educational project?

(●) Yes ( ) No

Please describe briefly the involvement of the student(s):
This study includes a PhD project. The PhD student has designed the protocol and will be leading on data collection and analysis.

9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?

(●) Yes ( ) No

10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?

( ) Yes (●) No

11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?

( ) Yes (●) No

Appendix J Ethics application, approval and amendments for cognitive interview study

Integrated Research Application System
Application Form for Research administering questionnaires/interviews for quantitative analysis or mixed methodology study

IRAS Form (project information)

Please refer to the E-Submission and Checklist tabs for instructions on submitting this application.

The Chief Investigator should complete this form. Guidance on the questions is available wherever you see this symbol displayed. We recommend reading the guidance first. The complete guidance and a glossary are available by selecting Help.

Please define any terms or acronyms that might not be familar to lay reviewers of the application.

Short title and version number: (maximum 70 characters - this will be inserted as header on all forms)
C-POS cognitive interview study

Please complete these details after you have booked the REC application for review.

REC Name:

REC Reference Number: **Submission date:**

PART A: Core study information

1. ADMINISTRATIVE DETAILS

A1. Full title of the research:

Children's Palliative Outcome Scale (C-POS) – cognitive interview study to determine comprehensibility, comprehensiveness and feasibility.

A2-1. Educational projects

Name and contact details of student(s):

Student 1

	Title	Forename/Initials	Surname
	Mrs	Lucy Helen	Coombes
Address	Department of Palliative Care, Policy and Rehabilitation		
	Cicely Saunders Institute		
	Bessemer Road		
Post Code	SE5 9RS		
E-mail	lucy.coombes@kcl.ac.uk		
Telephone	07482484414		
Fax			

Give details of the educational course or degree for which this research is being undertaken:
Name and level of course/ degree:

PhD - The development and initial validation of an outcome measure for use in children with life-limiting and life-threatening conditions – the Children's Palliative Outcome Scale (C-POS)

Name of educational establishment:
King's College London

Name and contact details of academic supervisor(s):

Academic supervisor 1

	Title	Forename/Initials	Surname
	Professor	Richard	Harding
Address	Department of Palliative Care, Policy and Rehabilitation		
	Cicely Saunders Institute		
	Bessemer Road		
Post Code	SE5 9RS		
E-mail	richard.harding@kcl.ac.uk		
Telephone	02078485518		
Fax			

Please state which academic supervisor(s) has responsibility for which student(s):
Please click "Save now" before completing this table. This will ensure that all of the student and academic supervisor details are shown correctly.

Student(s)	Academic supervisor(s)
Student 1 Mrs Lucy Helen Coombes	☐ Professor Richard Harding

A copy of a current CV for the student and the academic supervisor (maximum 2 pages of A4) must be submitted with the application.

A2-2. Who will act as Chief Investigator for this study?

- ○ Student
- ◉ Academic supervisor
- ○ Other

A3-1. Chief Investigator:

	Title	Forename/Initials	Surname
	Professor	Richard	Harding
Post	Herbert Dunhill Chair in Palliative Care and Rehabilitation		
Qualifications	BSc (Joint Hons) MSc DipSW PhD		
ORCID ID	0000 0001 9653 8689		
Employer	King's College London		
Work Address	Cicely Saunders Institute		
	Bessemer Road		
	London		
Post Code	SE5 9PJ		
Work E-mail	richard.harding@kcl.ac.uk		

* Personal E-mail	
Work Telephone	02078485518
* Personal Telephone/Mobile	07814918959
Fax	

** This information is optional. It will not be placed in the public domain or disclosed to any other third party without prior consent.*
A copy of a current CV (maximum 2 pages of A4) for the Chief Investigator must be submitted with the application.

A4. Who is the contact on behalf of the sponsor for all correspondence relating to applications for this project?
This contact will receive copies of all correspondence from REC and HRA/R&D reviewers that is sent to the CI.

	Title	Forename/Initials	Surname
	Professor	Reza	Ravazi
Address	Director of Research Management and Director of Administration (Health Schools)		
	Room 5.31, James Clerk Maxwell Building		
	57 Waterloo Road, London		
Post Code	SE1 8WA		
E-mail	reza.razavi@kcl.ac.uk		
Telephone	02078483224		
Fax			

A5-1. Research reference numbers. *Please give any relevant references for your study:*

Applicant's/organisation's own reference number, e.g. R & D (if available):	N/A
Sponsor's/protocol number:	N/A
Protocol Version:	v1
Protocol Date:	02/11/2020
Funder's reference number (enter the reference number or state not applicable):	772635
Project website:	https://www.kcl.ac.uk/cicelysaunders/research/outcome/pos/childre n%27s-p

Additional reference number(s):

Ref.Number Description	Reference Number

Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you have registered your study please give details in the "Additional reference number(s)" section.

A5-2. Is this application linked to a previous study or another current application?

(•) Yes ( ) No

Please give brief details and reference numbers.
C-POS phase 1. IRAS number 250470. Qualitative interviews have been carried out with children and young people with life-limiting conditions, siblings, parents/carers, health care professionals and NHS commissioners to inform the content of the C-POS. This study seeks to further establish face and content validity as well as ascertaining comprehensibility, comprehensiveness and feasibility of the initial versions of the C-POS.

* Personal E-mail	
Work Telephone	02078485518
* Personal Telephone/Mobile	07814918959
Fax	

** This information is optional. It will not be placed in the public domain or disclosed to any other third party without prior consent.*
A copy of a current CV (maximum 2 pages of A4) for the Chief Investigator must be submitted with the application.

A4. Who is the contact on behalf of the sponsor for all correspondence relating to applications for this project?
This contact will receive copies of all correspondence from REC and HRA/R&D reviewers that is sent to the CI.

	Title	Forename/Initials	Surname
	Professor	Reza	Ravazi
Address	Director of Research Management and Director of Administration (Health Schools)		
	Room 5.31, James Clerk Maxwell Building		
	57 Waterloo Road, London		
Post Code	SE1 8WA		
E-mail	reza.razavi@kcl.ac.uk		
Telephone	02078483224		
Fax			

A5-1. Research reference numbers. *Please give any relevant references for your study:*

Applicant's/organisation's own reference number, e.g. R & D (if available):	N/A
Sponsor's/protocol number:	N/A
Protocol Version:	v1
Protocol Date:	02/11/2020
Funder's reference number (enter the reference number or state not applicable):	772635
Project website:	https://www.kcl.ac.uk/cicelysaunders/research/outcome/pos/childre n%27s-p

Additional reference number(s):

Ref.Number Description	Reference Number

Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you have registered your study please give details in the "Additional reference number(s)" section.

A5-2. Is this application linked to a previous study or another current application?

(•) Yes ( ) No

Please give brief details and reference numbers.
C-POS phase 1. IRAS number 250470. Qualitative interviews have been carried out with children and young people with life-limiting conditions, siblings, parents/carers, health care professionals and NHS commissioners to inform the content of the C-POS. This study seeks to further establish face and content validity as well as ascertaining comprehensibility, comprehensiveness and feasibility of the initial versions of the C-POS.

Appendix J Ethics application, approval and amendments for cognitive interview study

2. OVERVIEW OF THE RESEARCH

To provide all the information required by review bodies and research information systems, we ask a number of specific questions. This section invites you to give an overview using language comprehensible to lay reviewers and members of the public. Please read the guidance notes for advice on this section.

A6-1. Summary of the study. *Please provide a brief summary of the research (maximum 300 words) using language easily understood by lay reviewers and members of the public. Where the research is reviewed by a REC within the UK Health Departments' Research Ethics Service, this summary will be published on the Health Research Authority (HRA) website following the ethical review. Please refer to the question specific guidance for this question.*

Children and young people (CYP) with life-limiting conditions (LLC) are living longer with more complex health care needs. It is important that health care professionals are asking these children and their parents/carers about the aspects of their life and the care they receive that are most important to them so that any areas of concern can be addressed. This can be done by using a patient-reported outcome measure (PROM). These are questionnaires measuring a patients view of their health status. Where a patient is unable to answer these questions due to their age, understanding or medical condition then someone else, such as a parent, may be asked these questions on their behalf.

This study is the second phase of a larger project aiming to develop a PROM (called C-POS) for use with CYP who have a LLC and their parents/carers. Interviews with children with LLC, health care professionals, siblings and parents have already been conducted to find out which symptoms and concerns are most important to them. The symptoms and concerns identified have then been evaluated by parents/carers of CYP with LLC and health care professionals in an online survey, and they prioritised which one should be included in our PROM. We now have version 1 of C-POS ready to test with CYP and their parents/carers. This part of the study (phase 2) aims to find out whether the questions in C-POS are understood and cognitively processed in the way the research team intend. This will be done by asking parents and children to complete the C-POS while telling us what they are thinking and why they choose the response they do as they answer the questions (called cognitive interviewing). An interviewer will ask questions while the C-POS is being completed. These interviews will be audio recorded.

A6-2. Summary of main issues. *Please summarise the main ethical, legal, or management issues arising from your study and say how you have addressed them.*

Not all studies raise significant issues. Some studies may have straightforward ethical or other issues that can be identified and managed routinely. Others may present significant issues requiring further consideration by a REC, HRA, or other review body (as appropriate to the issue). Studies that present a minimal risk to participants may raise complex organisational or legal issues. You should try to consider all the types of issues that the different reviewers may need to consider.

PURPOSE AND DESIGN
This research is needed because it has previously been established that there is a need for an outcome measure for use with children with life-limiting and life-threatening conditions and that nothing suitable already exists. There have also been calls for such a measure in publications on research priorities within paediatric palliative care. Such a measure will be an invaluable clinical decision-making tool and allow paediatric palliative care teams to evaluate new interventions and services. This will enhance the care and experience of this patient population by allowing children, their parent/carers and their healthcare team to focus on the symptoms and concerns that are most important to them. This study builds on previous work by the team:
1. Three systematic reviews - a) showing there are no suitable measures already in existence, b)symptoms and concerns of children with life-limiting conditions, c)appropriate recall periods, response formats and administration modality when asking children to self-report.
2. 106 qualitative interviews with children and young people with life-limiting conditions, their parents/carers, siblings, paediatric palliative care healthcare professionals and NHS commissioners to establish symptoms and concerns in this population..

The study steering group includes parents whose child has a life-limiting condition, representatives from the UK's leading charity for children with life-limiting conditions (Together for Short Lives) and Professor Bobbie Farsides, Professor of Clinical and Biomedical Ethics, Brighton and Sussex Medical School. We have an external ethics advisor, Dr Sara Fovargue, from Lancaster University. The research team who will be conducting the cognitive interviews have received training in qualitative interviewing in Paediatric Palliative Care from Professor Myra Bluebond-Langner, as well as sessions in communicating with children with additional needs and the ethics of interviewing children in palliative care. The researchers are all experienced qualitative interviewers and have experience of working with this population, having been involved in the previous qualitative interview study.

RECRUITMENT
The clinical teams at our participating sites will be responsible for initially approaching children and families to participate in the cognitive interviews. They will explain the study and give written information. We will also be using social media such as the Twitter and Facebook feeds of Together for Short Lives and those of our participating sites to recruit. This recruitment method worked very well for our previous online survey. It will be made clear that children and parents do not have to participate if they do not want to, and not participating will not in any way affect any care they receive. Each participant will be given a minimum of 24 hours to consider whether they would like to take part. If they agree their contact details will be given to the research team via secure NHS e-mail. The research team will then make contact and answer any questions participants may have and if participants agree an interview will be arranged. This will be either in a location of the participants choice if COVID-19 restrictions allow, otherwise interviews will be conducted online using Teams, Zoom or similar. Participants will be informed that Teams is our preferred option for interviews as it is NHS approved. If they do not have access to Teams then other platforms such as Zoom will be used and participants will be informed that it is not an NHS approved tool and confidentiality has not been formally assessed. The research team have experience of conducting interviews with this population via online platforms, having done this for some of our previous interviews in phase 1 of this study. The only data that will be shared between the clinical team and the research team will be the age of the child, their diagnosis and telephone number/email address and this will only be shared after the family have given verbal consent for this to happen.

INCLUSION/EXCLUSION
Inclusion criteria
• Children and young people: from age 5 up to the age of 17 who are living with a LLC/LTC.
• Parents/carers: responsible for the primary care needs of a child of any age who is living with a LLC/LTC.

Exclusion criteria
• Children and young people:
-unable to communicate any views or wishes via their parent/caregiver or an interview
-unable to read the C-POS questions or unable to understand the questions if they are read aloud
-speaks a language not supported by the NHS Trust's translation service;
-currently enrolled in another study.
-deemed clinically unable to give consent/assent.
-who do not wish to participate.

• Parents:
-deemed clinically unable to give consent
-who do not wish to participate.
- speak a language not supported by the NHS translation service

CONSENT
All participants will be given age/cognitively appropriate written material explaining the research. The researcher or person responsible for recruitment from participating sites will ensure that potential participants understand the nature of participation and will ensure that expression of interest in the study does not assume consent. The researcher/recruiter will clarify again how the data will be collected (i.e. one-to-one interview) and the steps to maintain confidentiality (i.e. that no identifying information will be used in publications or presentations to external audiences). Consent forms with names and study numbers will be kept in a locked cupboard within the Cicely Saunders Institute or in a secure file on a secure server while COVID-19 restrictions do not allow attending the office. Demographic data sheets will be kept in another locked cupboard or secure online file. Audio-recordings will be done on encrypted, password-protected devices.
Parents will be required to provide written consent for their child's participation up to and including the age of 15 years old, and the child will provide either verbal consent or written assent if they are willing/able. From age 16-17 the young person may give written consent if their treating clinician believes that the individual has sufficient understanding and can give full consent independent of their parents/caregivers. Parents/carers will also be required to provide written consent for their own participation. While COVID-19 restrictions remain and interviews are conducted online, participants will be asked to complete their consent/assent forms at the beginning of the call. They will then be given the choice of emailing the researcher a photograph or scanned copy via NHS email. If this is not possible then they will be asked to post this to the Cicely Saunders Institute.
Children and young people will, following approval by their parent/caregiver, have the study explained to them in appropriate language, following prior guidance from the parent/caregiver on the timing and manner for optimal comprehension and most clear explanation of the meaning of participation. Those 16 years and over will be asked to give their own written consent.

A potential participant with limited ability to indicate written consent on the form may give verbal consent in the presence of their parent/caregiver, who will sign alongside the researcher signature as witnesses to that consent.
For all participants, a copy of the information and consent forms will be left with/sent to them after data collection.
Information sheets will inform participants of the Data Protection Officer's contact details, how to complain, how long

data will be retained, and the right to withdraw consent and/or data.
Given that this is a single qualitative interview, we will assume capacity/competence is not lost between consent and interview as these will usually occur concurrently on the same day.

Consent forms with names and study numbers will be kept in a locked cupboard within the Cicely Saunders Institute or in a secure file on a secure server while COVID-19 restrictions do not allow attending the office. Demographic data sheets will be kept in another locked cupboard or secure online file. The secure online folders are KCL Sharepoint folders which will be password protected. A copy of the consent form will be sent to participants after the interview if it is face to face we won't always have access to a photocopier (i.e., in a participants home) and if it is a remote interview we will need to check they understand what participation entails before we countersign. If scanned and emailed it can be the same day, but not if posted.

RISKS, BURDENS AND BENEFITS
Children and young people and their families are considered a vulnerable population, even more so when they have a life-limiting condition. Participation in research is viewed by some as an undue burden. Children with life-limiting conditions are rarely included in research studies and it is important that they are given a chance to express their views. Not allowing CYP to participate can also be seen as discriminatory and unjust and in contravention to Article 12 of the UN Convention of the Rights on the Child which states that any child capable of forming their own views should have the right to express these. There can be concerns regarding coercion from clinical teams to participate but findings suggest that parents are able to say no if supported to do so. Avoiding using terms such as palliative care and end of life care can alleviate concerns from parents that their child is unaware of their prognosis. Ensuring that researchers work in partnership with child and their parents throughout the research process, including obtaining meaningful assent from the child, using age-appropriate written information and ensuring child and their family have free and informed choice regarding participation have been recommended. There is also evidence that allowing children and parents to participate in research, despite advancing illness, is often viewed as a positive and rewarding experience.

The research team are all experienced in interviewing children and parents with life-limiting conditions. Younger children will be offered the opportunity to play or draw while they are participating if they so wish. Participants will be made aware that the interview can be stopped or paused at any time if they wish. No information regarding prognosis will be shared as a result of participation. If a participant becomes unduly distressed during an interview their clinical team will be informed (with consent) so that further support can be provided.

CONFIDENTIALITY
No child or parent identifiable data will be shared in any dissemination of results. Any quotes used from the interviews will be anonymised for dissemination.

3. PURPOSE AND DESIGN OF THE RESEARCH

A7. Select the appropriate methodology description for this research. *Please tick all that apply*:

- ☐ Case series/ case note review
- ☐ Case control
- ☐ Cohort observation
- ☐ Controlled trial without randomisation
- ☑ Cross-sectional study
- ☐ Database analysis
- ☐ Epidemiology
- ☐ Feasibility/ pilot study
- ☐ Laboratory study
- ☐ Metanalysis
- ☑ Qualitative research
- ☐ Questionnaire, interview or observation study
- ☐ Randomised controlled trial
- ☐ Other (please specify)

A10. What is the principal research question/objective? *Please put this in language comprehensible to a lay person.*

To conduct cognitive interviews with CYP and families to establish feasibility, comprehensibility and comprehensiveness of the C-POS measure.

A11. What are the secondary research questions/objectives if applicable? *Please put this in language comprehensible to a lay person.*

NA

A12. What is the scientific justification for the research? *Please put this in language comprehensible to a lay person.*

The overall aim of C-POS is to develop a patient-reported outcome measure (PROM) for children and young people (CYP) affected by life-limiting and life-threatening conditions (LLLTC). There have been repeated calls for scientific advancement to develop, validate and implement PROMs for CYP and their families facing LLLTC. Currently, no valid tool exists, largely due to the complexities of self-report among children who often have profound communication difficulties, sensitivities around the subject matter, and lack of evidence on their symptoms and concerns. Therefore, this population have been neglected in terms of research activity and evidence.

Within the UK, there are estimated to be over 86,000 children living with a life-limiting or life-threatening condition(LLLTC), many of whom would benefit from palliative care services due to complex symptoms, social and emotional needs and the unpredictability of their condition. The number of children with LLLTC conditions in the UK is rising due to advances in medical care leading to slower deterioration. For these children, dependency requirements are increasing due to the increased use of medical technology such as home ventilation. This is putting increased pressure on the resources of paediatric palliative care teams. Within England and Wales more than 5000 CYP die each year from all causes. It is estimated that deaths due to LLLTC may account for 50% or more of these deaths. Mortality is highest in those under one year of age (peaking in the neonatal period) and decreases in middle childhood before rising again in adolescence. There is a significantly higher prevalence of LLLTC conditions in CYP from both ethnic minority backgrounds and higher areas of deprivation in the UK..

This study seeks to build on previous work by the team to develop a PROM for use in this population by testing the initial versions of the C-POS with parents/carers and CYP to ensure the questions and response format are understood and used as intended.

A13. Please summarise your design and methodology. *It should be clear exactly what will happen to the research participant, how many times and in what order. Please complete this section in language comprehensible to the lay person. Do not simply reproduce or refer to the protocol. Further guidance is available in the guidance notes.*

Parents/carers and children/young people eligible for participation will be approached by their clinical team and given written information regarding the study. They will be given a minimum of 24 hours to consider the study and if they agree after this time the clinical team will share their contact details with the research team via secure NHS mail. Invitation to participate in the study will also be posted on the social media pages of the Cicely Saunders Institute, Together for Short Lives and other paediatric palliative care providers who agree to share the details. These providers may also share details via their family newsletters. The research team will contact potential participants and answer any questions they may have regarding the study. If they agree to participate the researcher will arrange an interview time and location with the participant, which due to current COVID-19 restrictions may be via Zoom or similar.

Before the interview, informed consent will be taken from parents/carers and CYP 16 years and over. Younger children will be given the opportunity to provide written and/or verbal assent. Consent/assent will either be taken by the researcher at the time of the interview, or by the recruiting team (if appropriately trained) prior to the interview. All participants will be asked to complete the C-POS measure while talking aloud about what they understand by each question and why they have chosen their answers. The researcher will ask questions throughout the process in order to establish what participants understand by each question and how they chose each answer and why. These interviews will be audio-recorded.

It is anticipated that there will be 5 initial versions of C-POS (approximately 5-7 years; 8-12 years; 13-18 years, a parent/carer version for CYP≥5 years and a parent/carer version for CYP <2 years). Initially 4 cognitive interviews per version will be conducted. Changes will then be made to C-POS depending on any difficulties participants encountered with the measure. These amended versions will then be tested with another three participants. If further amendments are required these will be tested with another three participants.

A14-1. In which aspects of the research process have you actively involved, or will you involve, patients, service users, and/or their carers, or members of the public?

- [x] Design of the research
- [x] Management of the research
- [ ] Undertaking the research
- [x] Analysis of results
- [x] Dissemination of findings
- [ ] None of the above

Give details of involvement, or if none please justify the absence of involvement.
We have parents/carers of children with life-limiting conditions on our steering group. They attend our steering group meetings and provide feedback on our research design, recruitment, analysis and dissemination.

We are also working with the Young People's Advisory Group at Great Ormond Street Children's hospital who have loooked at our study information sheets and the C-POS measure in order to provide feedback.

4. RISKS AND ETHICAL ISSUES

RESEARCH PARTICIPANTS

A15. What is the sample group or cohort to be studied in this research?

Select all that apply:

- [ ] Blood
- [x] Cancer
- [ ] Cardiovascular
- [x] Congenital Disorders
- [x] Dementias and Neurodegenerative Diseases
- [ ] Diabetes
- [ ] Ear
- [ ] Eye
- [x] Generic Health Relevance
- [ ] Infection
- [x] Inflammatory and Immune System
- [ ] Injuries and Accidents
- [ ] Mental Health
- [x] Metabolic and Endocrine
- [x] Musculoskeletal
- [x] Neurological
- [x] Oral and Gastrointestinal
- [x] Paediatrics
- [x] Renal and Urogenital
- [ ] Reproductive Health and Childbirth
- [x] Respiratory

☐ Skin
☐ Stroke

Gender:	Male and female participants
Lower age limit: 5	Years
Upper age limit: 99	Years

A17-1. Please list the principal inclusion criteria (list the most important, max 5000 characters).

- Children and young people: from age 5 up to the age of 17 who are living with a LLLTC.
- Parents/carers: responsible for the primary care needs of a child of any age who is living with a LLLTC.

A17-2. Please list the principal exclusion criteria (list the most important, max 5000 characters).

• Children and young people:
o unable to communicate any views or wishes via their parent/caregiver or an interview
o unable to read the C-POS questions or unable to understand the questions if they are read aloud
o speaks a language not supported by the NHS Trust's translation service;
o currently enrolled in another study.
o deemed clinically unable to give consent/assent.
o Who do not wish to participate.

• Parents:
o Deemed clinically unable to give consent
o Who do not wish to participate.
o Who do not speak a language supported by the NHS translation service

RESEARCH PROCEDURES, RISKS AND BENEFITS

A18. Give details of all non-clinical intervention(s) or procedure(s) that will be received by participants as part of the research protocol. *These include seeking consent, interviews, non-clinical observations and use of questionnaires.*

Please complete the columns for each intervention/procedure as follows:

1. Total number of interventions/procedures to be received by each participant as part of the research protocol.
2. If this intervention/procedure would be routinely given to participants as part of their care outside the research, how many of the total would be routine?
3. Average time taken per intervention/procedure (minutes, hours or days)
4. Details of who will conduct the intervention/procedure, and where it will take place.

Intervention or procedure	1	2	3	4
Cognitive interview	1	0	Up to 90 minutes	Trained study-specific researcher; location of participants choice or via Zoom or similar.
Consent for cognitive interview	1	0	15 minutes	Trained study-specific researcher; location of participants choice or via Zoom or similar.
Demographic questionnaire	1	0	10	Trained study-specific researcher; location of participants choice or via Zoom or similar.

A21. How long do you expect each participant to be in the study in total?

Approximately a 60-90 minute interview plus 15 minutes prior to this to take consent and build rapport.

A22. What are the potential risks and burdens for research participants and how will you minimise them?

For all studies, describe any potential adverse effects, pain, discomfort, distress, intrusion, inconvenience or changes to lifestyle. Only describe risks or burdens that could occur as a result of participation in the research. Say what steps would be taken to minimise risks and burdens as far as possible.

There is a risk that participants will become upset during the study. They will be made aware before the interview begins that they can stop or take a break at any time. Participant information sheets will outline exactly what is involved in participation, so there should be no surprises.

Participants will be given at least 24 hours to decide whether they would like to take part in the study, and will also have the opportunity to ask the researcher any questions before the interview. It is not the role of the research team to disclose any information regarding diagnosis or prognosis and any such questions that do arise will be referred back to the clinical team.

Interviews will be conducted at a time and location of the participants choice.

A23. Will interviews/ questionnaires or group discussions include topics that might be sensitive, embarrassing or upsetting, or is it possible that criminal or other disclosures requiring action could occur during the study?

(●) Yes ( ) No

If Yes, please give details of procedures in place to deal with these issues:

There is a distress protocol for the study. If participants become upset during an interview the researcher will offer to stop or take a break. If any participant becomes unduly distressed this will be fed back to their clinical team, with their consent, so that they can provide further support. Participants will also be offered the telephone number of Together for Short Lives Family Helpline.

A24. What is the potential for benefit to research participants?

There will be no direct benefit to research participants. However there is evidence that children and young people with life-limiting conditions and their parents/carers who have participated in other studies have perceived that they are able to potentially help others in a similar situation and felt rewarded by this.

A26. What are the potential risks for the researchers themselves? *(if any)*

There is a risk that the interviewers may become distressed due to the nature of the study. The researchers are experienced in working with this population and have received training in order to do this. The department operates a 'buddy' system to ensure any home visits are conducted safely and that the researchers whereabouts are known at all times. Researchers also have access to supervision with a trained member of the clinical team.
KCL lone worker policy will be adhered to. If interviews are able to be carried out face to face during the COVID pandemic screening questions will be asked prior to the visit (anyone in the household having a high temperature, continuous cough or loss/change of taste/smell, anyone in the house testing positive in the past few 10 days or being asked to self-isolate). Appropriate PPE will be worn if needed during a home visit (mask, apron and gloves as per NHS guidance).

RECRUITMENT AND INFORMED CONSENT

In this section we ask you to describe the recruitment procedures for the study. Please give separate details for different study groups where appropriate.

A27-1. How will potential participants, records or samples be identified? Who will carry this out and what resources will be used?*For example, identification may involve a disease register, computerised search of GP records, or review of medical records. Indicate whether this will be done by the direct healthcare team or by researchers acting under arrangements with the responsible care organisation(s).*

Participants for the cognitive interviews will be identified by the clinical teams at participating sites during their team

meetings. Participants may also express interest in taking part in the study by contacting the research team directly in response to social media posts and/or information on their care providers family newsletter.

A27-2. Will the identification of potential participants involve reviewing or screening the identifiable personal information of patients, service users or any other person?

○ Yes ◉ No

Please give details below:

A28. Will any participants be recruited by publicity through posters, leaflets, adverts or websites?

◉ Yes ○ No

If Yes, please give details of how and where publicity will be conducted, and enclose copy of all advertising material (with version numbers and dates).

The study will be advertised via the study Twitter feed, Together for Short Lives social media feeds and family newsletters, as well as newsletters and social media feeds of UK children's hopsices and paediatric palliative care teams that are willing to share the information.

A29. How and by whom will potential participants first be approached?

Eligible participants will be identified by their clinical teams, participating site research team, or self-identify to the team via an organisation's newsletter or social media feed (see A28). If they are approached by a member of the clinical team the child and their parent/carer will have the study explained to them and be given age/cognitively appropriate written information about the study.

A30-1. Will you obtain informed consent from or on behalf of research participants?

◉ Yes ○ No

If you will be obtaining consent from adult participants, please give details of who will take consent and how it will be done, with details of any steps to provide information (a written information sheet, videos, or interactive material). Arrangements for adults unable to consent for themselves should be described separately in Part B Section 6, and for children in Part B Section 7.

If you plan to seek informed consent from vulnerable groups, say how you will ensure that consent is voluntary and fully informed.

Before the interview commences, the researcher will ensure that potential participants understand the nature of participation and willl clarify how the data will be collected (i.e. one-to-one interview) and the steps to maintain confidentiality (i.e. that no identifying information will be used in publications or presentations to external audiences). Parents will be required to provide written consent for their child's participation up to and including the age of 15 years, and the child will provide a statement of informed voluntary participation/permission. From the age of 16 years the young person may give written consent if their treating clinicianand/or parent/carer believes that the individual has sufficient understanding and can give full consent. Parents/carers will also be required to provide written consent for their own participation.

Children and young people will, following approval by their parent/caregiver, have the study explained to them in appropriate language, following prior guidance from the parent/caregiver on the timing and manner for optimal comprehension and most clear explanation of the meaning of participation. Those 16 years and over will be asked to give their own written consent.

A potential participant with limited ability to indicate written consent on the form may give verbal consent in the presence of their parent/caregiver, who will sign alongside the researcher signature as witnesses to that consent.

For all participants, a copy of the information and consent forms will be left with them after data collection. Information sheets will inform participants of the Data Protection Officer's contact details, how to complain, how long data will be retained, and the right to withdraw consent and/or data.

Given that this is a single qualitative interview, we will assume capacity/competence is not lost between consent and interview as these will usually occur concurrently on the same day.

If remote interviews are conducted due to the COVID-19 pandemic, the participant will complete the consent form with the researcher during the video call. This will then be emailed to the researcher as a photo/scanned document. If this is not possible the participant will be asked to post this to the Cicely Saunders Institute.

If you are not obtaining consent, please explain why not.

Please enclose a copy of the information sheet(s) and consent form(s).

A30-2. Will you record informed consent (or advice from consultees) in writing?

(●) Yes ( ) No

A31. How long will you allow potential participants to decide whether or not to take part?

If they are given details by their clinical team, participants will be given a minimum of 24 hours to decide whether they would like to participate.

If the CYP and family contact the research team directly in response to a social media post, a member of the research team will make contact and explain the study to them and what participation will involve. Following the introductory explanation, the CYP and their parent/carer will be provided age specific written information on the study, either by post, email or via the clinical team. They will be given a minimum of 24 hours to consider participation.

If a CYP and/or their parent/carer are keen to participate, and it would be more convenient for them to do this less than 24 hours after being approached (for instance they are due to be discharged from hospital) then they will be allowed to waive the 24 hour period.

A33-1. What arrangements have been made for persons who might not adequately understand verbal explanations or written information given in English, or who have special communication needs?*(e.g. translation, use of interpreters)*

If participants would like to have written information in a different language then we will commission this via an NHS-approved interpretation service. Interviews may also be carried out via an NHS approved interpreter.

A35. What steps would you take if a participant, who has given informed consent, loses capacity to consent during the study? *Tick one option only.*

( ) The participant and all identifiable data or tissue collected would be withdrawn from the study. Data or tissue which is not identifiable to the research team may be retained.

( ) The participant would be withdrawn from the study. Identifiable data or tissue already collected with consent would be retained and used in the study. No further data or tissue would be collected or any other research procedures carried out on or in relation to the participant.

( ) The participant would continue to be included in the study.

( ) Not applicable – informed consent will not be sought from any participants in this research.

(●) Not applicable – it is not practicable for the research team to monitor capacity and continued capacity will be assumed.

Further details:

This is a single, qualitative interview and consent will be taken immediately prior to the interview, Therefore, we will assume that capacity is not lost between consent and interview.

CONFIDENTIALITY

In this section, personal data means any data relating to a participant who could potentially be identified. It includes pseudonymised data capable of being linked to a participant through a unique code number.

Storage and use of personal data during the study

A36. Will you be undertaking any of the following activities at any stage (including in the identification of potential

participants)?*(Tick as appropriate)*

- [x] Access to medical records by those outside the direct healthcare team
- [ ] Access to social care records by those outside the direct social care team
- [x] Electronic transfer by magnetic or optical media, email or computer networks
- [ ] Sharing of personal data with other organisations
- [ ] Export of personal data outside the EEA
- [x] Use of personal addresses, postcodes, faxes, emails or telephone numbers
- [x] Publication of direct quotations from respondents
- [ ] Publication of data that might allow identification of individuals
- [x] Use of audio/visual recording devices
- [ ] Storage of personal data on any of the following:
  - [x] Manual files (includes paper or film)
  - [ ] NHS computers
  - [ ] Social Care Service computers
  - [ ] Home or other personal computers
  - [x] University computers
  - [ ] Private company computers
  - [ ] Laptop computers

Further details:
USE OF TELEPHONE NUMBERS: Those individuals who express an interest in participation will have their telephone number sent by secure NHS email to the study researcher.
DIRECT QUOTATIONS: Direct qualitative quotes will be used with identifying information removed (i.e. pseudonymised)
AUDIO RECORDING DEVICES: We will use digital recording devices with built in encryption and secure file deletion. All studies are subject to audit by representatives of the sponsor outside of the direct care team, this includes access to medical records where necessary.

A37. Please describe the physical security arrangements for storage of personal data during the study?

Signed consent forms will be stored in a locked cabinet separately from data files, at KCL (as consent forms will be signed in most cases by family members at home rather than at the NHS site). Consent forms will be stored in a cabinet in a card-protected academic department in a locked office. Study ID forms (that allocate a study ID to participant name) will be stored separately in locked cabinet separate from consent forms.

During the COVID-19 pandemic, consent forms may be sent electronically to the research team by participants. These will be stored on a secure server in a password protected folder until such time that they can be printed out and stored on site as outlined above.

Transcriptions will be pseudonymised (i.e. names places and attributable events removed) and stored on password protected study PCs. Transcripts (in Word files) will use an anonymous study ID number.

Transcription will be completed using two approaches: 1) by the research study team, on computers at the Cicely Saunders Institute or remotely on secure computers on the KCL managed environment; or 2) by the KCL preferred supplier for transcription – Clear Voice. Clear Voice utilise a secure site for researchers to upload password protected audio files. Passwords for audio files would be sent via the secure email service, Egress.com. At this stage all data will be pseudonymised, in that names, places, dates of birth (and any other potentially identifying information) will be removed from transcripts. Audio recordings will be transferred in encrypted files with no accompanying information.

A38. How will you ensure the confidentiality of personal data?*Please provide a general statement of the policy and procedures for ensuring confidentiality, e.g. anonymisation or pseudonymisation of data.*

Transcripts will only contain study ID and no names. Qualitative data will be pseudonymised. Transcription will be completed using two approaches: 1) by the research study team, on computers at the Cicely Saunders Institute or remotely on secure computers on the KCL managed environment; or 2) by the KCL preferred supplier for transcription – Clear Voice. Clear Voice utilise a secure site for researchers to upload password protected audio files. Passwords for audio files would be sent via the secure email service, Egress.com. At this stage all data will be pseudonymised, in that names, places, dates of birth (and any other potentially identifying information) will be removed from transcripts. Audio recordings will be transferred in encrypted files with no accompanying information.

DATA MANAGEMENT PLAN.
The EXCEL spreadsheet of participants will be managed by the study cognitive interview lead LUCY COOMBES. She will be the sole person responsible for updating the ECCEL file. She will back this up to the KCL secure server with a new date and version after each addition of a participant. The data will be stored separately in a different password-protected file and folder. The password will be held by the study PI PROFESSOR HARDING.

The pseudonym breaksheet will also be stored in a password protected file separately from data and participant record sheet. The password will be held by COOMBES and HARDING.

A40. Who will have access to participants' personal data during the study? *Where access is by individuals outside the direct care team, please justify and say whether consent will be sought.*

The research team will have access to participant names and telephone numbers in order to contact them and arrange interviews.

Storage and use of data after the end of the study

A41. Where will the data generated by the study be analysed and by whom?

The data will be analysed by the study team: Professor Harding (PI), Research Associate (Dr Debbie Braybrook), Research Assistants (Hannah Scott and Daney Haroardottir) and PhD candidate (Lucy Coombes). This will either me analysed on site at the Cicely Saunders Institute, King's College London, or remotely on KCL encrypted laptops by the study team (during the COVID pandemic).

A42. Who will have control of and act as the custodian for the data generated by the study?

	Title	Forename/Initials	Surname
	Professor	Richard	Harding
Post Qualifications			
Work Address	Department of Palliative Care, Policy and Rehabilitation Cicely Saunders Institute Bessemer Road		
Post Code	SE5 9RS		
Work Email	richard.harding@kcl.ac.uk		
Work Telephone	02078485518		
Fax			

A43. How long will personal data be stored or accessed after the study has ended?

- ○ Less than 3 months
- ○ 3 – 6 months
- ◉ 6 – 12 months
- ○ 12 months – 3 years
- ○ Over 3 years

A44. For how long will you store research data generated by the study?

Years: 8
Months: 0

A45. Please give details of the long term arrangements for storage of research data after the study has ended. *Say where data will be stored, who will have access and the arrangements to ensure security.*

Data will be stored in the secure, locked data archive at the Cicely Saunders Institute, King's College London. The archive box specific to this study will detail the destruction date.
Access to research data will be on reasonable request to the PI.

INCENTIVES AND PAYMENTS

A46. Will research participants receive any payments, reimbursement of expenses or any other benefits or incentives for taking part in this research?

(•) Yes ( ) No

If Yes, please give details. For monetary payments, indicate how much and on what basis this has been determined.
If participants wish to be interviewed somewhere other than their home then reasonable travel expenses will be reimbursed in cash at the interview. No other payment will be provided for participation.

A47. Will individual researchers receive any personal payment over and above normal salary, or any other benefits or incentives, for taking part in this research?

( ) Yes (•) No

A48. Does the Chief Investigator or any other investigator/collaborator have any direct personal involvement (e.g. financial, share holding, personal relationship etc.) in the organisations sponsoring or funding the research that may give rise to a possible conflict of interest?

( ) Yes (•) No

NOTIFICATION OF OTHER PROFESSIONALS

A49-1. Will you inform the participants' General Practitioners (and/or any other health or care professional responsible for their care) that they are taking part in the study?

( ) Yes (•) No

If Yes, please enclose a copy of the information sheet/letter for the GP/health professional with a version number and date.

PUBLICATION AND DISSEMINATION

A50. Will the research be registered on a public database?

(•) Yes ( ) No

Please give details, or justify if not registering the research.

NIHR portfolio and research registry.com

Registration of research studies is encouraged wherever possible.
You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you are aware of a suitable register or other method of publication, please give details. If not, you may indicate that no suitable register exists. Please ensure that you have entered registry reference number(s) in question A5-1.

A51. How do you intend to report and disseminate the results of the study? *Tick as appropriate:*

- [x] Peer reviewed scientific journals
- [ ] Internal report
- [x] Conference presentation
- [x] Publication on website
- [ ] Other publication
- [ ] Submission to regulatory authorities
- [ ] Access to raw data and right to publish freely by all investigators in study or by Independent Steering Committee on behalf of all investigators
- [ ] No plans to report or disseminate the results
- [ ] Other (please specify)

A52. If you will be using identifiable personal data, how will you ensure that anonymity will be maintained when publishing the results?

We will take all reasonable steps to pseudonymise qualitative data, removing clearly identifiable information such as names and places. As some participants may have extremely rare conditions these will be reported under ICD10 headings.

A53. How and when will you inform participants of the study results?

If there will be no arrangements in place to inform participants please justify this.
It is possible that participating children will have died before reporting and the research team will not be aware of this. We are also collecting minimal patient identifiable data so will not have access to addresses/email details for dissemination. However, our partner Together for Short Lives (leading UK charity for families with children with life-limiting conditions) will be sent updates and key results to publish in their news letter to parents/carers of children with life-limiting conditions.

5. Scientific and Statistical Review

A54. How has the scientific quality of the research been assessed? *Tick as appropriate:*

- [x] Independent external review
- [ ] Review within a company
- [ ] Review within a multi−centre research group
- [x] Review within the Chief Investigator's institution or host organisation
- [ ] Review within the research team
- [x] Review by educational supervisor
- [ ] Other

Justify and describe the review process and outcome. If the review has been undertaken but not seen by the researcher, give details of the body which has undertaken the review:

This study has received competitive funding via "Consolidator Award" from the European Research Council (ERC). This involved a full research grant application, external peer review, shortlisting, interview with a panel in Brussels, and ethical review commissioned by the ERC. All PhD projects are subject to thorough review at King's College London.

For all studies except non-doctoral student research, please enclose a copy of any available scientific critique reports, together with any related correspondence.

For non-doctoral student research, please enclose a copy of the assessment from your educational supervisor/ institution.

A56. How have the statistical aspects of the research been reviewed? *Tick as appropriate:*

- ☐ Review by independent statistician commissioned by funder or sponsor
- ☐ Other review by independent statistician
- ☐ Review by company statistician
- ☐ Review by a statistician within the Chief Investigator's institution
- ☐ Review by a statistician within the research team or multi−centre group
- ☐ Review by educational supervisor
- ☐ Other review by individual with relevant statistical expertise
- ☑ No review necessary as only frequencies and associations will be assessed – details of statistical input not required

In all cases please give details below of the individual responsible for reviewing the statistical aspects. If advice has been provided in confidence, give details of the department and institution concerned.

Title Forename/Initials Surname

Department
Institution
Work Address

Post Code
Telephone
Fax
Mobile
E-mail

Please enclose a copy of any available comments or reports from a statistician.

A57. What is the primary outcome measure for the study?

Cognitive interviews - NA as qualitative.

A58. What are the secondary outcome measures? *(if any)*

NA

A59. What is the sample size for the research? *How many participants/samples/data records do you plan to study in total? If there is more than one group, please give further details below.*

Total UK sample size: 50
Total international sample size (including UK): 50

Total in European Economic Area: 50

Further details:

It is anticipated that there will be 4 initial versions of the C-POS (approximately 5-7 years; 8-12 years; 13-18 years and a parent/carer version) but this will be guided by previous research. There will be at least 7 participants per version in the cognitive interview study. The actual number of participants will depend on how many changes need to be made to C-POS after each round of interviews.

A60. How was the sample size decided upon? *If a formal sample size calculation was used, indicate how this was done, giving sufficient information to justify and reproduce the calculation.*

The sample sizes were decided upon by following accepted guidance on developing patient reported outcome measures published by the COSMIN (COnsensus-based Standards for the selection of health Measurement Instruments) group.

A61. Will participants be allocated to groups at random?

○ Yes ◉ No

A62. Please describe the methods of analysis (statistical or other appropriate methods, e.g. for qualitative research) by which the data will be evaluated to meet the study objectives.

Audio recordings of the interviews will be listened to by two members of the research team. The completed C-POS measure will be read alongside this and interview note templates reviewed. Data will be tabulated in Excel by participant and item. This will be reviewed by the research team after four interviews have been conducted for each C-POS version (child and parent proxy versions). Consensus will be reached on whether changes to version 1 of the C-POS need to be made. If changes are made, the new version will then be piloted with another three participants from each cohort and analysed in the same manner. Demographic data will be presented using descriptive statistics. If the research team feel there is sufficiently rich data in the interviews, they will be transcribed and analysed using framework analysis.

6. MANAGEMENT OF THE RESEARCH

A63. Other key investigators/collaborators. *Please include all grant co−applicants, protocol co−authors and other key members of the Chief Investigator's team, including non-doctoral student researchers.*

	Title	Forename/Initials	Surname
	Professor	Myra	Bluebond-Langner
Post	True Colours Chair in Palliative Care for Children and Young People		
Qualifications	BA MA PhD		
Employer	University College London		
Work Address	Institute for Child Health 30 Guildford Street		
Post Code	WC1N 1EH		
Telephone	02079052366		
Fax			
Mobile			
Work Email	bluebond@ucl.ac.uk		

	Title	Forename/Initials	Surname
	Professor	Irene J	Higginson
Post	Professor of Palliative Care and Policy		
Qualifications	BMedSci MSc PhD FMedSci		
Employer	King's College London		
Work Address	Cicely Saunders Institute		
	Bessemer Road		
	London		
Post Code	SE5 9PJ		
Telephone	02078485516		
Fax			
Mobile			
Work Email	irene.higginson@kcl.ac.uk		

	Title	Forename/Initials	Surname
	Professor	Lorna	Fraser
Post	Director of the Martin House Research Centre		
Qualifications	PGCAP, PhD, MMedSci, MSc, MRCPCH, MBChB		
Employer	University of York		
Work Address	Department of Health Sciences, Area 2 Seebohm Rowntree Building		
	Heslington		
	York		
Post Code			
Telephone	01904321889		
Fax			
Mobile			
Work Email	lorna.fraser@york.ac.uk		

	Title	Forename/Initials	Surname
	Professor	Bobbie	Farsides
Post	Professor of Clinical and Biomedical Ethics		
Qualifications	BA PhD		
Employer	Sussex University		
Work Address	Medical Teaching Building 3.04		
	Brighton and Sussex Medical School		
Post Code	BN1 9PX		
Telephone	01273877630		
Fax			
Mobile			
Work Email	b.farsides@sussex.ac.uk		

	Title	Forename/Initials	Surname
	Professor	FM	Murtagh
Post	Professor of Palliative Medicine		
Qualifications	BMedSci MSc PhD		
Employer	Hull York Medical School		
Work Address	Allum Medical Building		
	University of Hull		
	Hull		

Post Code	HU6 7RX
Telephone	01482463164
Fax	
Mobile	
Work Email	fliss.murtagh@hyms.ac.uk

A64. Details of research sponsor(s)

A64-1. Sponsor

Lead Sponsor

Status: ○ NHS or HSC care organisation ◉ Academic ○ Pharmaceutical industry ○ Medical device industry ○ Local Authority ○ Other social care provider (including voluntary sector or private organisation) ○ Other

Commercial status: Non-Commercial

If Other, please specify:

Contact person

Name of organisation	King's College London
Given name	Reza
Family name	Razavi
Address	Director of Research Management and Administration (Health Schools), 5.31 JCMB, 57 Waterloo Road
Town/city	London
Post code	SE1 8WA
Country	United Kingdom
Telephone	02078483224
Fax	
E-mail	reza.razavi@kcl.ac.uk

Co-Sponsor

Status: ◉ NHS or HSC care organisation ○ Academic ○ Pharmaceutical industry ○ Medical device industry ○ Local Authority ○ Other social care provider (including voluntary sector or private organisation)

Commercial status:

◯ Other

If Other, please specify:

Contact person

Name of organisation	GSTT
Given name	Rachel
Family name	Fay
Address	Guy's & St Thomas' Foundation NHS Trust R&D Department 16th Floor, Tower Wing, Great Maze pond
Town/city	London
Post code	SE1 9RT
Country	United Kingdom
Telephone	02071885733
Fax	02071883472
E-mail	R&D@gstt.nhs.uk

A64-2. Please explain how the responsibilities of sponsorship will be assigned between the co-sponsors listed in A64-1

The lead sponsor, King's College London, will take primary responsibility for ensuring that the design of the study meets appropriate standards and that arrangements are in place to ensure appropriate conduct and reporting. King's College London also provides cover under its No Fault Compensation Insurance, which provides for payment of damages or compensation in respect of any claim made by a research subject for bodily injury arising out of participation in a clinical trial or healthy volunteer study (with certain restrictions). The co-sponsor, Guy's & St Thomas' Foundation NHS Trust, take ultimate responsibility for arranging the initiation and management of this research, and will take responsibility for ensuring that appropriate standards, conduct and reporting are adhered to regarding its facilities and staff involved with the project.

A65. Has external funding for the research been secured?

Please tick at least one check box.

☑ Funding secured from one or more funders

☐ External funding application to one or more funders in progress

☐ No application for external funding will be made

What type of research project is this?

◉ Standalone project

◯ Project that is part of a programme grant

◯ Project that is part of a Centre grant

◯ Project that is part of a fellowship/ personal award/ research training award

◯ Other

Other – please state:

Please give details of funding applications.

Organisation	European Research Council
Address	European Commission ERC Executive Agency COV2 Brussels
Post Code	BE-1049
Telephone	
Fax	
Mobile	
Email	

Funding Application Status: (•) Secured ( ) In progress

Amount: 1.8m Euros

Duration
Years: 4
Months: 6

If applicable, please specify the programme/ funding stream:

What is the funding stream/ programme for this research project?
Consolidator Award

A66. Has responsibility for any specific research activities or procedures been delegated to a subcontractor (other than a co-sponsor listed in A64-1) ? *Please give details of subcontractors if applicable.*

( ) Yes (•) No

A67. Has this or a similar application been previously rejected by a Research Ethics Committee in the UK or another country?

( ) Yes (•) No

Please provide a copy of the unfavourable opinion letter(s). You should explain in your answer to question A6-2 how the reasons for the unfavourable opinion have been addressed in this application.

A68-1. Give details of the lead NHS R&D contact for this research:

	Title	Forename/Initials	Surname
	Ms	Rachel	Fay

Organisation	Guy's & St Thomas' Foundation NHS Trust
Address	R and D department, 16th Floor, Tower Wing, Great Maze Pond, London
Post Code	SE1 9RT
Work Email	R&D@gstt.nhs.uk
Telephone	02071887188
Fax	02071883472
Mobile	

Details can be obtained from the NHS R&D Forum website: http://www.rdforum.nhs.uk

A68-2. Select Local Clinical Research Network for NHS Organisation identified in A68-1:

South London

For more information, please refer to the question specific guidance.

A69-1. How long do you expect the study to last in the UK?

Planned start date: 01/03/2021
Planned end date: 28/02/2022
Total duration:
Years: 0 Months: 11 Days: 28

A71-1. Is this study?

- ○ Single centre
- ◉ Multicentre

A71-2. Where will the research take place? *(Tick as appropriate)*

- ☑ England
- ☐ Scotland
- ☐ Wales
- ☑ Northern Ireland
- ☐ Other countries in European Economic Area

Total UK sites in study 5

Does this trial involve countries outside the EU?

○ Yes ◉ No

A72. Which organisations in the UK will host the research?*Please indicate the type of organisation by ticking the box and give approximate numbers if known:*

☑ NHS organisations in England	3
☐ NHS organisations in Wales	
☐ NHS organisations in Scotland	
☐ HSC organisations in Northern Ireland	
☐ GP practices in England	
☐ GP practices in Wales	
☐ GP practices in Scotland	
☐ GP practices in Northern Ireland	
☐ Joint health and social care agencies (eg community mental health teams)	
☐ Local authorities	
☐ Phase 1 trial units	

☐ Prison establishments	
☐ Probation areas	
☑ Independent (private or voluntary sector) organisations	3
☐ Educational establishments	
☐ Independent research units	
☐ Other (give details)	
Total UK sites in study:	6

A73-1. Will potential participants be identified through any organisations other than the research sites listed above?

◉ Yes ○ No

A73-2. If yes, will any of these organisations be NHS organisations?

○ Yes ◉ No

If yes, details should be given in Part C.

A74. What arrangements are in place for monitoring and auditing the conduct of the research?

The Chief Investigator will be responsible for the ongoing management of the study. The Sponsor will monitor and conduct audits on a selection of studies in its clinical research portfolio. Monitoring and auditing will be conducted in accordance with the UK Policy Framework for Health and Social Care 2017 and in accordance with the Sponsor's monitoring and audit procedures

A76. Insurance/ indemnity to meet potential legal liabilities

***Note:* in this question to NHS indemnity schemes include equivalent schemes provided by Health and Social Care (HSC) in Northern Ireland**

A76-1. What arrangements will be made for insurance and/or indemnity to meet the potential legal liability of the sponsor(s) for harm to participants arising from the management of the research? *Please tick box(es) as applicable.*

Note: Where a NHS organisation has agreed to act as sponsor or co-sponsor, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For all other sponsors, please describe the arrangements and provide evidence.

☑ NHS indemnity scheme will apply (NHS sponsors only)

☐ Other insurance or indemnity arrangements will apply (give details below)

Please enclose a copy of relevant documents.

A76-2. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of the sponsor(s) or employer(s) for harm to participants arising from the design of the research? *Please tick box(es) as applicable.*

Note: Where researchers with substantive NHS employment contracts have designed the research, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For other protocol authors (e.g. company employees, university members), please describe the arrangements and provide evidence.

☑ NHS indemnity scheme will apply (protocol authors with NHS contracts only)

☑ Other insurance or indemnity arrangements will apply (give details below)

King's College London indemnity applies.

Please enclose a copy of relevant documents.

A76-3. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of investigators/collaborators arising from harm to participants in the conduct of the research?

Note: Where the participants are NHS patients, indemnity is provided through the NHS schemes or through professional indemnity. Indicate if this applies to the whole study (there is no need to provide documentary evidence). Where non-NHS sites are to be included in the research, including private practices, please describe the arrangements which will be made at these sites and provide evidence.

☑ NHS indemnity scheme or professional indemnity will apply (participants recruited at NHS sites only)

☐ Research includes non-NHS sites (give details of insurance/ indemnity arrangements for these sites below)

Research includes non-NHS sites as PIC sites only.

Please enclose a copy of relevant documents.

A78. Could the research lead to the development of a new product/process or the generation of intellectual property?

○ Yes ◉ No ○ Not sure

PART B: Section 7 - Children

1. Please specify the potential age range of children under 16 who will be included and give reasons for carrying out the research in this age group.

5-16 years. The C-POS is being developed for children from 5 - 18 years and therefore needs to be cognitively tested with them to ensure that they understand the questions and response format as intended.

2. Indicate whether any children under 16 will be recruited as controls and give further details.

NA

3-2. Please describe the arrangements for seeking informed consent from a person with parental responsibility and/or from children able to give consent for themselves.

Children and young people will, following approval by their parent/caregiver, have the study explained to them in appropriate language, following prior guidance from the parent/caregiver on the timing and manner for optimal comprehension and most clear explanation of the meaning of participation. Those 16 years and over will be asked to give their own written consent.
A potential participant with limited ability to indicate written consent on the form may give verbal consent in the presence of their parent/caregiver, who will sign alongside the researcher signature as witnesses to that consent.

Parents will be required to provide written consent for their child's participation up to and including the age of 15, and the child will provide a statement of informed voluntary participation/permission. From age 16-17 the young person may give written consent if their treating clinician believes that the individual has sufficient understanding and can give full consent independent of their parents/caregivers.

4. If you intend to provide children under 16 with information about the research and seek their consent or agreement, please outline how this process will vary according to their age and level of understanding.

Please see 3-2 above. We have developed age appropriate information sheets for children explaining the study and what will be expected of them.

Copies of written information sheet(s) for parents and children, consent/assent form(s) and any other explanatory material should be enclosed with the application.

Please see 3-2 above. We have developed age appropriate information sheets for children explaining the study and what will be expected of them.

Copies of written information sheet(s) for parents and children, consent/assent form(s) and any other explanatory material should be enclosed with the application.

PART C: Overview of research sites

Please enter details of the host organisations (Local Authority, NHS or other) in the UK that will be responsible for the research sites. *For further information please refer to guidance.*

Investigator identifier	Research site		Investigator Name	
IN1	(•) NHS/HSC Site ( ) Non-NHS/HSC Site		Forename Middle name Family name Email	Susan Picton susan.picton@nhs.net
	Organisation name	LEEDS TEACHING HOSPITALS NHS TRUST	Qualification (MD...)	Consultant Paediatric Oncologist
	Address	ST. JAMES'S UNIVERSITY HOSPITAL BECKETT STREET LEEDS	Country	United Kingdom
	Post Code	LS9 7TF		
	Country	ENGLAND		
IN2	(•) NHS/HSC Site ( ) Non-NHS/HSC Site		Forename Middle name Family name Email	Anna-Karenia Anderson Anna-karenia.anderson@nhs.net
	Organisation name	THE ROYAL MARSDEN NHS FOUNDATION TRUST	Qualification (MD...)	Consultant in Paediatric Palliative Medicine
	Address	FULHAM ROAD LONDON GREATER LONDON	Country	United Kingdom
	Post Code	SW3 6JJ		
	Country	ENGLAND		
IN3	(•) NHS/HSC Site ( ) Non-NHS/HSC Site		Forename Middle name Family name Email	Joanna Laddie Joanna.laddie@gstt.nhs.uk
	Organisation name	GUY'S AND ST THOMAS' NHS FOUNDATION TRUST	Qualification (MD...)	Consultant in Paediatric Palliative Medicine
	Address	ST THOMAS' HOSPITAL WESTMINSTER BRIDGE ROAD LONDON	Country	United Kingdom
	Post Code	SE1 7EH		

Country ENGLAND

IN4

NHS/HSC Site
Non-NHS/HSC Site

Institution name: Martin House Children's Hospice
Department name:
Street address: Grove Road
Town/city: Boston Spa, Wetherby
Post Code: LS23 6TX
Country: United Kingdom

Forename: Michelle
Middle name:
Family name: Hills
Email: Michelle.hills@nhs.net
Qualification (MD...): Consultant in Paediatric Palliative Medicine
Country: United Kingdom

IN5

NHS/HSC Site
Non-NHS/HSC Site

Institution name: East Anglia Children's Hospice
Department name: The Nook
Street address: Pigot Lane, Farmington Earl
Town/city: Norfolk
Post Code: NR14 7PX
Country: United Kingdom

Forename: Linda
Middle name:
Family name: Maynard
Email: linda.maynard@each.org.uk
Qualification (MD...): PhD
Country: United Kingdom

IN6

NHS/HSC Site
Non-NHS/HSC Site

Institution name: Northern Ireland Children's Hospice
Department name: Horizon House
Street address: 18 O'Neill Road
Town/city: Newtownabbey
Post Code: BT36 6WB
Country: United Kingdom

Forename: Deborah
Middle name:
Family name: Burns
Email: Deborah.burns@nihospice.org
Qualification (MD...): Physiotherapist
Country:

PART D: Declarations

D1. Declaration by Chief Investigator

1. The information in this form is accurate to the best of my knowledge and belief and I take full responsibility for it.
2. I undertake to fulfil the responsibilities of the chief investigator for this study as set out in the UK Policy Framework for Health and Social Care Research.
3. I undertake to abide by the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research.
4. If the research is approved I undertake to adhere to the study protocol, the terms of the full application as approved and any conditions set out by review bodies in giving approval.
5. I undertake to notify review bodies of substantial amendments to the protocol or the terms of the approved application, and to seek a favourable opinion from the main REC before implementing the amendment.
6. I undertake to submit annual progress reports setting out the progress of the research, as required by review bodies.
7. I am aware of my responsibility to be up to date and comply with the requirements of the law and relevant guidelines relating to security and confidentiality of patient or other personal data, including the need to register when necessary with the appropriate Data Protection Officer. I understand that I am not permitted to disclose identifiable data to third parties unless the disclosure has the consent of the data subject or, in the case of patient data in England and Wales, the disclosure is covered by the terms of an approval under Section 251 of the NHS Act 2006.
8. I understand that research records/data may be subject to inspection by review bodies for audit purposes if required.
9. I understand that any personal data in this application will be held by review bodies and their operational managers and that this will be managed according to the principles established in the Data Protection Act 2018.
10. I understand that the information contained in this application, any supporting documentation and all correspondence with review bodies or their operational managers relating to the application:
    - Will be held by the REC (where applicable) until at least 3 years after the end of the study; and by NHS R&D offices (where the research requires NHS management permission) in accordance with the NHS Code of Practice on Records Management.
    - May be disclosed to the operational managers of review bodies, or the appointing authority for the REC (where applicable), in order to check that the application has been processed correctly or to investigate any complaint.
    - May be seen by auditors appointed to undertake accreditation of RECs (where applicable).
    - Will be subject to the provisions of the Freedom of Information Acts and may be disclosed in response to requests made under the Acts except where statutory exemptions apply.
    - May be sent by email to REC members.
11. I understand that information relating to this research, including the contact details on this application, may be held on national research information systems, and that this will be managed according to the principles established in the Data Protection Act 2018.
12. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the Health Research Authority (HRA) together with the contact point for enquiries named below. Publication will take place no earlier than 3 months after the issue of the ethics committee's final opinion or the withdrawal of the application.

Contact point for publication*(Not applicable for R&D Forms)*

HRA would like to include a contact point with the published summary of the study for those wishing to seek further

information. We would be grateful if you would indicate one of the contact points below.

- ○ Chief Investigator
- ○ Sponsor
- ○ Study co-ordinator
- ◉ Student
- ○ Other – please give details
- ○ None

Access to application for training purposes *(Not applicable for R&D Forms)*

Optional – please tick as appropriate:

☑ I would be content for members of other RECs to have access to the information in the application in confidence for training purposes. All personal identifiers and references to sponsors, funders and research units would be removed.

This section was signed electronically by Dr Richard Harding on 15/03/2021 19:28.

Job Title/Post:	Professor
Organisation:	King's College London
Email:	richard.harding@kcl.ac.uk

D2. Declaration by the sponsor's representative

If there is more than one sponsor, this declaration should be signed on behalf of the co−sponsors by a representative of the lead sponsor named at A64-1.

I confirm that:

1. This research proposal has been discussed with the Chief Investigator and agreement in principle to sponsor the research is in place.
2. An appropriate process of scientific critique has demonstrated that this research proposal is worthwhile and of high scientific quality.
3. Any necessary indemnity or insurance arrangements, as described in question A76, will be in place before this research starts. Insurance or indemnity policies will be renewed for the duration of the study where necessary.
4. Arrangements will be in place before the study starts for the research team to access resources and support to deliver the research as proposed.
5. Arrangements to allocate responsibilities for the management, monitoring and reporting of the research will be in place before the research starts.
6. The responsibilities of sponsors set out in the UK Policy Framework for Health and Social Care Research will be fulfilled in relation to this research.

   Please note: *The declarations below do not form part of the application for approval above. They will not be considered by the Research Ethics Committee.*
7. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named in this application. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.
8. Specifically, for submissions to the Research Ethics Committees (RECs) I declare that any and all clinical trials approved by the HRA since 30th September 2013 (as defined on IRAS categories as clinical trials of medicines, devices, combination of medicines and devices or other clinical trials) have been registered on a publically accessible register in compliance with the HRA registration requirements for the UK, or that any deferral granted by the HRA still applies.

This section was signed electronically by Prof Reza Razavi on 16/03/2021 10:53.

Job Title/Post:	Vice Principal (Research)
Organisation:	King's College London
Email:	reza.razavi@kcl.ac.uk

D3. Declaration for student projects by academic supervisor(s)

1. I have read and approved both the research proposal and this application. I am satisfied that the scientific content of the research is satisfactory for an educational qualification at this level.

2. I undertake to fulfil the responsibilities of the supervisor for this study as set out in the UK Policy Framework for Health and Social Care Research.

3. I take responsibility for ensuring that this study is conducted in accordance with the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research, in conjunction with clinical supervisors as appropriate.

4. I take responsibility for ensuring that the applicant is up to date and complies with the requirements of the law and relevant guidelines relating to security and confidentiality of patient and other personal data, in conjunction with clinical supervisors as appropriate.

Academic supervisor 1

This section was signed electronically by Dr Richard Harding on 16/03/2021 09:32.

Job Title/Post:	Professor
Organisation:	King's College London
Email:	richard.harding@kcl.ac.uk

Appendix J Ethics application, approval and amendments for cognitive interview study

NHS
Health Research Authority
London - Bloomsbury Research Ethics Committee
HRA RES Centre Manchester
3rd Floor Barlow House
4 Minshull Street
Manchester
M1 3DZ

Telephone: 02071048285

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

12 May 2021

Professor Richard Harding
Department of Palliative Care, Policy and Rehabilitation
Cicely Saunders Institute
Bessemer Road
SE5 9RS

Dear Professor Harding

Study title:	**Children's Palliative Outcome Scale (C-POS) – cognitive interview study to determine comprehensibility, comprehensiveness and feasibility.**
REC reference:	**21/LO/0282**
Protocol number:	**N/A**
IRAS project ID:	**282412**

Thank you for your letter of 19 April 2021, responding to the Research Ethics Committee's (REC) request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair, together with the Lead Reviewer, Ms Monica King, and Second Reviewer, Miss Selina Tsai.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Good practice principles and responsibilities

The UK Policy Framework for Health and Social Care Research sets out principles of good practice in the management and conduct of health and social care research. It also outlines the responsibilities of individuals and organisations, including those related to the four elements of research transparency:

1. registering research studies
2. reporting results
3. informing participants
4. sharing study data and tissue

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or NHS management permission (in Scotland) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All research should be registered in a publicly accessible database and we expect all researchers, research sponsors and others to meet this fundamental best practice standard.

It is a condition of the REC favourable opinion that **all clinical trials are registered** on a publicly accessible database within six weeks of recruiting the first research participant. For this purpose, 'clinical trials' are defined as the first four project categories in IRAS project filter question 2. Failure to register a clinical trial is a breach of these approval conditions, unless a deferral has been agreed by or on behalf of the Research Ethics Committee (see here for more information on requesting a deferral: https://www.hra.nhs.uk/planning-and-improving-research/research-planning/research-registration-research-project-identifiers/

If you have not already included registration details in your IRAS application form, you should notify the REC of the registration details as soon as possible.

Further guidance on registration is available at:
https://www.hra.nhs.uk/planning-and-improving-research/research-planning/transparency-responsibilities/

Publication of Your Research Summary

We will publish your research summary for the above study on the research summaries section of our website, together with your contact details, no earlier than three months from the date of this favourable opinion letter.

Should you wish to provide a substitute contact point, make a request to defer, or require further information, please visit:
https://www.hra.nhs.uk/planning-and-improving-research/application-summaries/research-summaries/

N.B. If your study is related to COVID-19 we will aim to publish your research summary within 3 days rather than three months.

During this public health emergency, it is vital that everyone can promptly identify all relevant research related to COVID-19 that is taking place globally. If you haven't already done so, please register your study on a public registry as soon as possible and provide the REC with the registration detail, which will be posted alongside other information relating to your project. We are also asking sponsors not to request deferral of publication of research summary for any projects relating to COVID-19. In addition, to facilitate finding and extracting studies related to COVID-19 from public databases, please enter the WHO official acronym for the coronavirus disease (COVID-19) in the full title of your study. Approved COVID-19 studies can be found at:
https://www.hra.nhs.uk/covid-19-research/approved-covid-19-research/

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

After ethical review: Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study, including early termination of the study
- Final report
- Reporting results

The latest guidance on these topics can be found at
https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/.

Ethical review of research sites

NHS/HSC sites

The favourable opinion applies to all NHS/HSC sites taking part in the study, subject to confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or management permission (in Scotland) being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS/HSC sites

I am pleased to confirm that the favourable opinion applies to any non-NHS/HSC sites listed in the application, subject to site management permission being obtained prior to the start of the study at the site.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Copies of materials calling attention of potential participants to the research [Example social media recruitment post]	2	19 April 2021
Covering letter on headed paper [Response letter]	1	19 April 2021
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only)		
Interview schedules or topic guides for participants [Topic guide]	1	10 March 2021
IRAS Application Form [IRAS_Form_17032021]		17 March 2021
Letter from funder		
Non-validated questionnaire [C-POS 5-7 years]	1	22 February 2021
Non-validated questionnaire [C-POS 8-12 years]	1	22 February 2021
Non-validated questionnaire [C-POS parent/carer]	1	22 February 2021
Non-validated questionnaire [C-POS parent/carer less that 2yrs]	1	22 February 2021
Non-validated questionnaire [C-POS 13-18 years]	1	22 February 2021
Other [Distress protocol]	1	15 October 2020
Other [Insurance]		
Other [Insurance]		
Participant consent form [Assent 11-15yrs]	1	15 October 2020
Participant consent form [Assent 5-7yrs]	1	15 October 2020
Participant consent form [Assent 8-10yrs]	1	15 October 2020
Participant consent form [Consent 16-18yrs]	2	19 April 2021
Participant consent form [Consent parent/carer signing for child]	2	19 April 2021
Participant consent form [Parent/carer consent]	2	19 April 2021
Participant information sheet (PIS) [Parent/carer PIS]	2	19 April 2021
Participant information sheet (PIS) [Parent/carer signing for child PIS]	2	19 April 2021
Participant information sheet (PIS) [Child 16-18yrs PIS]	2	19 April 2021
Participant information sheet (PIS) [Child 11-15yrs PIS]	2	19 April 2021
Participant information sheet (PIS) [Child 8-10yrs PIS]	2	19 April 2021
Participant information sheet (PIS) [Child 5-7yrs PIS]	2	19 April 2021
Protocol [Cognitive Interview Protocol - Clean]	3	05 May 2021

Protocol [Cognitive Interview Protocol - Tracked]	3	05 May 2021
Referee's report or other scientific critique report [Feedback from young people on PIS']		05 March 2021
Summary CV for Chief Investigator (CI) [CV for CI]		01 October 2020
Summary CV for student		
Summary CV for supervisor (student research)		

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

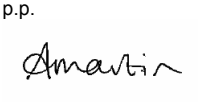
HRA Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities– see details at:
https://www.hra.nhs.uk/planning-and-improving-research/learning/

IRAS project ID: 282412 Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely
p.p.



Dr Paul Gorczynski
Chair

Email:bloomsbury.rec@hra.nhs.uk

Enclosures: "After ethical review – guidance for researchers" *[SL-AR2]*

Copy to: Professor Reza Ravazi

Appendix J Ethics application, approval and amendments for cognitive interview study

Professor Richard Harding
Department of Palliative Care, Policy and Rehabilitation
Cicely Saunders Institute
Bessemer Road
SE5 9RSN

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

12 May 2021

Dear Professor Harding

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Children's Palliative Outcome Scale (C-POS) – cognitive interview study to determine comprehensibility, comprehensiveness and feasibility.**
IRAS project ID:	**282412**
Protocol number:	**N/A**
REC reference:	**21/LO/0282**
Sponsor	**King's College London**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **282412**. Please quote this on all correspondence.

Yours sincerely,
Damilola Odunlami

Approvals Specialist

Email: approvals@hra.nhs.uk

Copy to: Professor Reza Ravazi

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	*Version*	*Date*
Copies of materials calling attention of potential participants to the research [Example social media recruitment post]	2	19 April 2021
Covering letter on headed paper [Response letter]	1	19 April 2021
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only)		
Interview schedules or topic guides for participants [Topic guide]	1	10 March 2021
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Protocol [Cognitive Interview Protocol - Tracked]	3	05 May 2021
Referee's report or other scientific critique report [Feedback from young people on PIS']		05 March 2021
Schedule of Events or SoECAT [SoE]	1	11 March 2021
Summary CV for Chief Investigator (CI) [CV for CI]		01 October 2020
Summary CV for student		
Summary CV for supervisor (student research)		

Appendix J Ethics application, approval and amendments for cognitive interview study

IRAS project ID	282412

Information to support study set up

The below provides all parties with information to support the arranging and confirming of capacity and capability with participating NHS organisations in England and Wales. This is intended to be an accurate reflection of the study at the time of issue of this letter.

Types of participating NHS organisation	Expectations related to confirmation of capacity and capability	Agreement to be used	Funding arrangements	Oversight expectations	HR Good Practice Resource Pack expectations
All sites will perform the same research activities therefore there is only onesite type.	Research activities should not commence at participating NHS organisations in England or Wales prior to their formal confirmation of capacity and capability to deliver the study.	An Organisation Information Document has been submitted and the sponsor is not requesting and does not expect any other site agreement to be used.	No study funding will be provided to sites as per the Organisation Information Document.	A Principal Investigator should be appointed at study sites.	No Honorary Research Contracts, Letters of Access or pre-engagement checks are expected for local staff employed by the participating NHS organisations. Where arrangements are not already in place, research staff not employed by the NHS host organisation undertaking any of the research activities listed in the research application would be expected to obtain a Letter of Access based on standard DBS checks and occupational health clearance.

Other information to aid study set-up and delivery

This details any other information that may be helpful to sponsors and participating NHS organisations in England and Wales in study set-up.
The applicant has indicated they intend to apply for inclusion on the NIHR CRN Portfolio.

Appendix J Ethics application, approval and amendments for cognitive interview study

C-POS Cognitive Interview Study Ethics Amendments

	Date sent	Categorisation	Description	Documents	Comments
1	14/07/2021	Non-substantial	Adding Glasgow as a site	Protocol v3.1	
2	8/11/2021	Non-substantial	Adding Hertfordshire, Bradford RI, East Lancashire, Chestnut Tree House, East Cheshire, Forget me Not, Leicester RI.	Protocol v3.2	
3	26/11/2021	Non-substantial	Adding Maidstone and Tunbridge Wells NHS Trust as a site. Extension until 30/4/2021.	Protocol v3.3	

Appendix K Delphi survey protocol, participant information sheet and demographic data collection sheet

FULL/LONG TITLE OF THE STUDY Children's Palliative Outcome Scale

SHORT STUDY TITLE / ACRONYM C-POS

PROTOCOL VERSION NUMBER AND DATE V0.10 17/8/2020

RESEARCH REFERENCE NUMBERS

IRAS Number:	The unique identifier generated by Integrated Research Application System (IRAS) for the project. This will be the primary reference number used by Research Ethics Committee, Health Research Authority and sites to identify the project and should be quoted in all project related correspondence as well as on all participant literature.
SPONSORS Number:	Generated by the Sponsor. Enter if applicable
FUNDERS Number:	772635

Appendix K Delphi survey protocol, participant information sheet and demographic data collection sheet

SIGNATURE PAGE

The undersigned confirm that the following protocol has been agreed and accepted and that the Chief Investigator agrees to conduct the study in compliance with the approved protocol and will adhere to the principles outlined in the Declaration of Helsinki, the Sponsor's SOPs, and other regulatory requirement.

I agree to ensure that the confidential information contained in this document will not be used for any other purpose other than the evaluation or conduct of the investigation without the prior written consent of the Sponsor

I also confirm that I will make the findings of the study publicly available through publication or other dissemination tools without any unnecessary delay and that an honest accurate and transparent account of the study will be given; and that any discrepancies from the study as planned in this protocol will be explained.

For and on behalf of the Study Sponsor:

Signature: Date:
...../...../.....

.....

Name (please print):

.....

Position:
.....

Chief Investigator:

Signature: Date:
..../...../.....

Name: (please print):

.....

LIST of CONTENTS

Appendix K Delphi survey protocol, participant information sheet and demographic data collection sheet

KEY STUDY CONTACTS

Chief Investigator	Professor Richard Harding Email richard.harding@kcl.ac.uk Tel: 0207 858 5518 Fax 0207 848 5518
Study Co-ordinator	Anna Roach anna.e.roach@kcl.ac.uk 0207 848 5536
Sponsor	King's College London Director of Research Management and Administration 5.31 JCMB, 57 Waterloo Road London 02078483224 Reza.razavi@kcl.ac.uk
Joint-sponsor(s)/co-sponsor(s)	King's College Hospital NHS Foundation Trust Anne-Marie Murtagh R&I Manager 161 Denmark Hill London SE5 9RF 02032993841 annemariemurtagh@kch.nhs.net
Funder(s)	European Research Council COV2 Brussels BE-1049
Key Protocol Contributors	Lucy Coombes (lucy.coombes@kcl.ac.uk) Professor Richard Harding (Richard.harding@kcl.ac.uk) Professor Wei Gao (wei.gao@kcl.ac.uk)

Appendix K Delphi survey protocol, participant information sheet and demographic data collection sheet

STUDY SUMMARY

Study Title	Children's Palliative Outcome Scale – phase 2
Internal ref. no. (or short title)	C-POS 2
Study Design	Delphi survey
Study Participants	a) Parents of children with life-limiting/life-threatening conditions b) Health care professionals
Planned Size of Sample (if applicable)	Up to 200
Follow up duration (if applicable)	N/A
Planned Study Period	3 months
Research Question/Aim(s)	Aim: To gain consensus on items to be included in C-POS (child and family-centred outcome measure for use in paediatric palliative care). Objectives: a) To conduct a Delphi survey to obtain stakeholder consensus on items to be included in the C-POS measure, thus further establishing face and content validity. b) To conduct an item generation meeting to finalise initial parent/carer and age appropriate child and young person versions of the C-POS.

FUNDING AND SUPPORT IN KIND

FUNDER(S) (Names and contact details of ALL organisations providing funding and/or support in kind for this study)	**FINANCIAL AND NON FINANCIALSUPPORT GIVEN**
European Research Council	**1.8 million Euros**

ROLE OF STUDY SPONSOR AND FUNDER

The Funder (European Research Council) and sponsor (King's College London and King's College Hospital NHS Foundation Trust) had no role in study design, conduct, data analysis and interpretation, manuscript writing, and dissemination of results.

ROLES AND RESPONSIBILITIES OF STUDY MANAGEMENT COMMITEES/GROUPS & INDIVIDUALS

The **Study Steering Group** consists of PI Professor Harding (Cicely Saunders Institute, King's College London) co-apps Professor Lorna Fraser (Martin House Research Centre, University of York), Professor Murtagh (Wolfson Palliative Care Research Centre, Hull York Medical School), Professor Wei Gao (Cicely Saunders Institute), Professor Higginson (King's College London), Professor Farsides (Brighton and Sussex Medical School), Professor Bluebond-Langner (Louis Dundas Centre, University College London). These members are funded by the study and responsible for delivery of the protocol. Independent members of the Steering Group are Dr Anna-Karenia Anderson (Consultant in Paediatric Palliative Medicine, Royal Marsden), Lydia Bate(PPI), Dr Debbie Braybrook (King's College London), Dr Katherine Bristowe (King's College London), Dr Rachel Burman (Palliative Care Consultant, King's College Hospital), Lizzie Chambers (Together for Short Lives), Lucy Coombes (PhD candidate, King's College London), Professor Sir Alan Craft, Dr Finella Craig (Consultant in Paediatric Palliative Medicine, Great Ormond Street Hospital), Professor Julia Downing (International Children's Palliative Care Network), Dr Clare Ellis-Smith (King's College London), Dr Sara Fovargue (Lancaster University), Dr Ann Goldman (Consultant in Paediatric Palliative Medicine), Jane Green (PPI member), Dr Michelle Hills (Consultant in Paediatric Palliative Medicine, Martin House Children's Hospice), Dr Gillian Hughes (Consultant in Paediatric Palliative Medicine, Evelina Children's Hospital), Dr Joanna Laddie (Consultant in Paediatric Palliative Medicine, Evelina Children's Hospital), Dr Steven Marshall (King's College London), Angela Logan (PPI), Dr Linda Maynard (Nurse Consultant, East Anglia Children's Hospice), Renee McCulloch (Consultant in Paediatric Palliative Medicine, Great Ormond Street Hospital), Dr Eve Namisango (African Palliative Care Association), Dr Sue Picton (Consultant Paediatric Oncologist, Leeds Teaching Hospital) and Anna Roach (King's College London). These individuals will be responsible for reviewing progress, delivery and quality of the research.

PROTOCOL CONTRIBUTORS

Appendix K Delphi survey protocol, participant information sheet and demographic data collection sheet

The protocol has been led by PI Professor Harding, PhD Candidate Lucy Coombes and Professor Wei Gao, and contributed to by steering group members (Dr Bristowe, Dr Ellis-Smith, Professor Murtagh, Professor Fraser, Professor Farsides and Dr Namisango).

KEY WORDS: Paediatrics, outcome assessment, child, cognitive interviews, Delphi survey, outcome measure

STUDY PROTOCOL

Children's Palliative Outcome Scale (C-POS)

1. BACKGROUND

Life-limiting conditions (LLC) in children and young people are defined as conditions for which there is no reasonable hope of cure and from which children or young people will die, such as Duchenne muscular dystrophy. Life-threatening conditions (LTC) are those for which curative treatment may be feasible but can fail, such as cancer(1, 2).

Epidemiology

Worldwide, it is estimated that each year 21 million children and young people (children and young people) with LLC/LTC conditions require input from palliative care services(3). Within the UK, there are estimated to be over 86,000 children and families living with a LLC/LTC condition, many of whom would benefit from palliative care services due to complex symptoms, social and psychological needs and the unpredictability of their condition(4). Almost 400 LLC/LTC conditions have been identified as appropriate for palliative care among children and young people(5). The number of children with LLC/LTC conditions in the UK is rising due to advances in medical care leading to slower deterioration. For these children, complexity of care is rising due to the increased use of medical technology such as home ventilation(2, 6). This is putting increased pressure on the resources of paediatric palliative care teams. Within England and Wales more than 5000 children and young people die each year from all causes(7). It is estimated that deaths due to LLC/LTC may account for 50% or more of these deaths(7). Mortality is highest in those under one year of age (peaking in the neonatal period) and decreases in middle childhood before rising again in adolescence(7). There is a significantly higher prevalence of LLC/LTC conditions in children and young people from both ethnic minority backgrounds and higher areas of deprivation in the UK(1).

Patient-reported outcome measurement in children and young people

A patient reported outcome measure (PROM) is defined as any measure of a patient's health status, elicited directly from the patient(8). PROM's range from single item symptom ratings e.g. pain scales, to complex multidimensional health-related quality of life tools(9). Patient reported outcome instruments may be either generic or disease specific(10). Generic measures are useful for comparing general outcomes across different populations e.g., cancer vs. cardiac disease or for assessing interventions across a wide range of conditions(11). Some of these measures are used to

assess health-related quality of life in healthy children so are more likely to have been validated based on large samples but may lack sensitivity for sick children and young people. They may also be too long for very unwell children to complete(12). Generic measures allow you to compare outcomes across groups of children and young people with different illnesses, which is essential for a discipline as wide and varied as children and young people palliative care. Disease specific instruments, on the other hand, are used to compare quality of life within a given condition e.g., cancer. Disease specific measures are assumed to be more sensitive to the implications of different illnesses and may be more appropriate for evaluating interventions or different treatments within children and young people with the same illness(12). Some patient reported outcome measures have been developed combining both approaches giving rise to generic measures with disease specific modules(13). In children and young people palliative care, an outcome measure is required that can capture symptoms and concerns that are important to children and young people with a wide range of conditions but is specific to the unique experience of this population(14).

Previous work and current study

This study is informed by and builds upon previous work conducted by the research team, following the principles of outcome measure development described by Rothrock(15). A systematic review has been carried out which identified that there is currently no suitable outcome measure for use with children and young people with palliative care needs(14). 102 qualitative, semi-structured interviews have been conducted with children and young people with LLC/LTC, their parents/carers, siblings, health care professionals and NHS commissioners to establish their priorities for outcomes of care This is one of the largest and most comprehensive qualitative interview studies to have been conducted in this population. Approval was received from the Bloomsbury ethics committee in 2019 (REC reference 19/LO/0033). In addition, systematic reviews regarding a) symptoms and concerns in children and young people with LLC/LTC (16) and b) optimal recall period, response format and administration mode for use with children and young people (pending publication) have also been conducted. Together, this previous work will inform the content of the Delphi survey and this and the previous work conducted will be used to develop initial parent/carer and age appropriate child versions of the C-POS.

2. RATIONALE

In adult settings routine use of PROMs has been shown to improve awareness of symptoms and concerns, identify unrecognised symptoms, increase the monitoring of symptoms, improve patient satisfaction and experience as well as having a positive effect on patient-clinician communication(17-22). Measuring outcomes in children and young people

palliative care has repeatedly been identified as a research priority(23-25). However, a recent systematic review concluded that there is currently no suitable outcome measure available for use in children and young people palliative care, with domains of existing generic health-related quality-of-life measures not being relevant to all children with LLC/LTC and some domains within disease- specific measures are only relevant for that specific population. (14).

A high quality PROM that includes physical, emotional, psychosocial and spiritual elements that contains sensitive indicators specific to children with LLC/LTC illness is required(26). The development of a validated outcome measure for children and young people will help address the current gap in high quality research in a population for whom there is currently very little evidence for good practice(27). It will also allow palliative care teams responsible for providing services to an increasing population of children and young people with complex needs a way to evaluate new interventions, compare treatments or services and aid clinical decision making(9).

Recent studies have highlighted that much of the research carried out with children with LLC/LTC conditions does not include them as participants(16, 28-32). One third of studies did not include children and young people as participants in one recent systematic review looking at symptoms and concerns in children and young people with LLC/LLC. Child self-report should be considered the gold standard for measuring symptoms and concerns in children and young people palliative care due to the subjective nature of many of the questions(33).

The overall development of the C-POS aims to address these gaps by including children and young people with a range of LLC/LTC conditions as participants both in the previous qualitative interviews and future cognitive testing. This will establish the comprehensiveness, comprehensibility and feasibility of the C-POS measure. From this, an evidence-based outcome measure for use with children and young people and their parents/carers with LLC/LTC conditions that is comprehensive and comprehensible with face and content validity will be developed. Ensuring that a PROM is relevant for the intended population and has clinical utility has been shown to be a facilitator in its implementation(34, 35). The inclusion of clinicians and parents in qualitative interviews and the Delphi survey and item generation meeting will provide further evidence of content validity of the C-POS. This will further ensure that the content is relevant and meaningful, which should aid implementation of the C-POS when it has been further validated.

The overall aim of C-POS is the development and validation of an outcome measure for use with children and young people with LLC/LTC and their families. The qualitative interviews and systematic review on response format, administration mode and recall period are complete. This protocol details

the Delphi survey to be used to obtain consensus on items to be included in the measure. There will be subsequent applications for a cognitive interview study and validation of the measure.

3. THEORETICAL FRAMEWORK

This study is conducted in line with the principles of outcome measure development proposed by Rothrock and the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN)(15, 36, 37). Figure 1 shows the patient reported outcome development process described by Rothrock with this study mapped on to it(15). The protocol we present in this application for ethical review incorporates the item generation part of the process (shown in green in the figure).

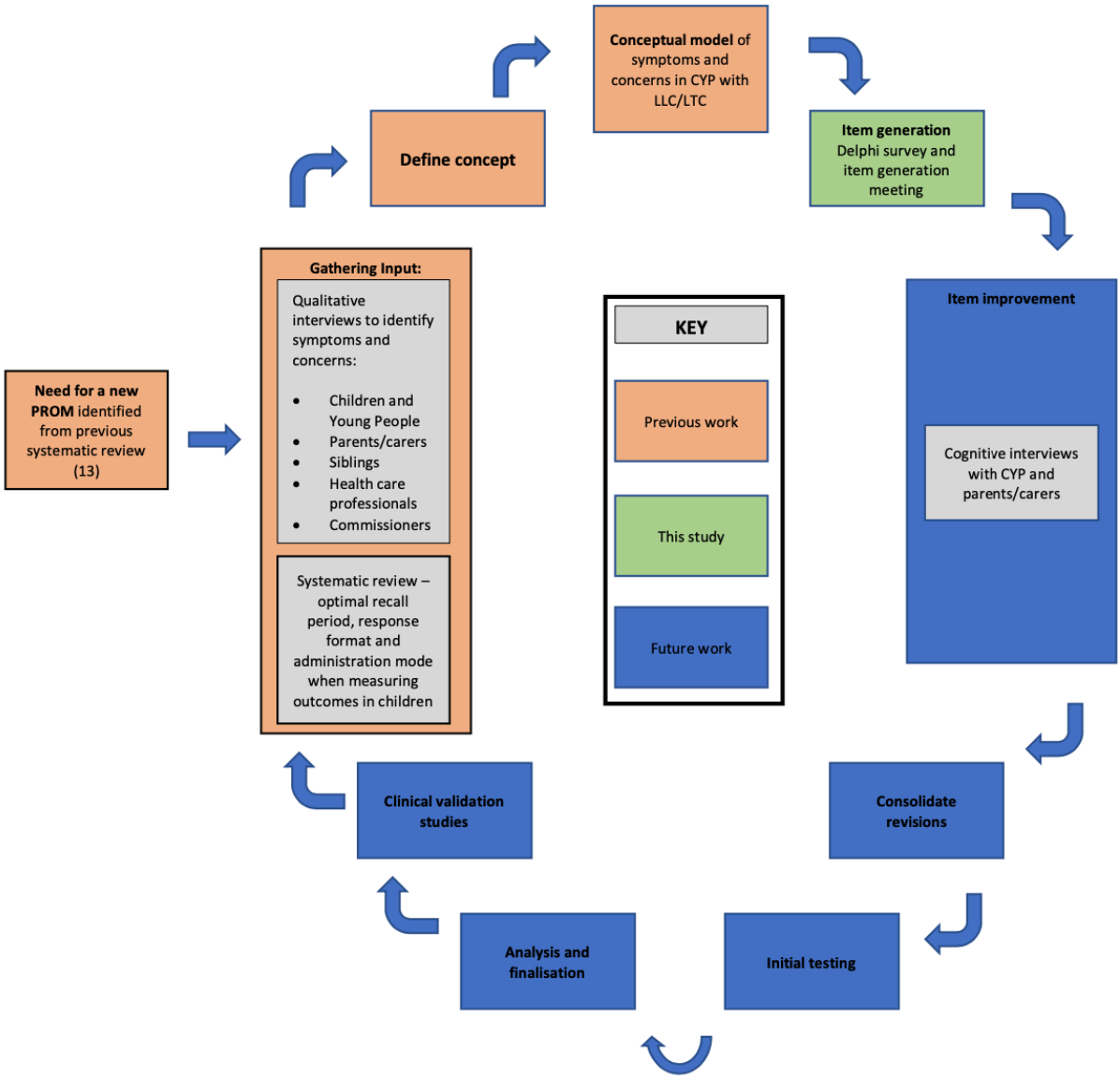

Figure 1 Patient reported outcome development process and the C-POS study

4. RESEARCH QUESTION/AIM(S)

To gain expert consensus on items to be included in the C-POS, which will be a patient-reported outcome measure (PROM) for children and young people (children and young people) with life-limiting and life-threatening conditions (LLC/LTC) and their parents/carers.

4.1 Objectives

a) To obtain stakeholder consensus on items to be included in the C-POS measure, thus further establishing face and content validity.
b) To finalise initial parent/carer and age appropriate children and young people versions of the C-POS.

4.2 Output

The key output from this study will be initial age appropriate and parent/proxy versions of the C-POS (version 1) ready for cognitive testing.

5 STUDY DESIGN and METHODS of DATA COLLECTION AND DATA ANALYIS

The objective of a Delphi survey is to achieve reliable consensus within a group of experts(38). Using a Delphi survey enables researchers to collect judgements from experts when it is hard to meet them face-to-face due to time constraints or distance(39, 40). Participants take part in several rounds, receiving feedback in between. Within the field of measurement, Delphi studies are well-recognised and commonly used to gather evidence of validity(40). The method has been used successfully in palliative care tool development to obtain international evidence of content validity(41).

There are four different types of Delphi methodology:

1. Classical – focuses on facts to obtain consensus.
2. Decision – focuses on preparation and decision for future directions.
3. Policy – focuses on ideas to define and differentiate views.
4. Ranking-type – focuses on identification and ranking of key factors, items or issues(38).

In this study, a ranking-type Delphi survey will be carried out to establish further evidence of content validity of the C-POS, using methodology similar to that proposed by Schmidt(38, 42, 43). Ranking-type Delphi surveys are used to reach a group consensus about the relative importance of issues or concerns(43, 44). In this protocol, the issues group consensus is required upon are the symptoms and concerns identified in previous qualitative interviews (see Figure 1)(38). For all measurement

instruments, it is important that content validity is assessed by clinical experts in the relevant field(45). For patient-reported outcomes, patients and representatives of the target population, are considered experts. They are the most able to assess the relevance of items and whether important aspects are missing(45).

The standard ranking-type Delphi survey has three phases(43, 44);

1. Brain storming phase – experts list items that are important for the area of interest.
2. Narrowing down phase – narrowing down of the list of items developed during the brainstorming phase to a number that is manageable and reasonable for the ranking phase.
3. Ranking – the aim of this phase is to reach consensus in the ranking of selected items.

102 semi-structured interviews to identify symptoms and concerns in children and young people with LLC/LTC have already been conducted, which will replace the brainstorming stage in this study. This Delphi survey will have 3-4 rounds, one to narrow down the number of symptoms and concerns, followed by 2-3 rounds aiming to achieve consensus on the rank importance of these. The Delphi survey will be followed by a face-to-face item generation meeting to finalise the content and format of the C-POS(46). It has been suggested that a three round Delphi is optimal to ensure that results are meaningful without creating participant fatigue and thus high levels of attrition between rounds(47).

Delphi Survey Design

This study is a repeated online survey (3-4 rounds proposed). Items to be included in the survey will be symptoms, concerns and priorities for care identified from the previous qualitative interview study and from a recent systematic review on symptoms and concerns in children and young people with LLC/LTC(16). A matrix has been created by the research team that demonstrates the evidence source for each item included.

Recruitment and sampling technique

This Delphi survey will involve two key stakeholder populations – parents/carers of children and young people with LLC/LTC and healthcare professionals who care for children with LLC/LTC. Both are considered experts in the symptoms and concerns that children and young people with LLC/LTC experience(36). This will further enhance the content validity of C-POS.

Parent/carer participants will be identified through:

- Together for Short Lives (a leading UK charity for children with life-threatening & life-limiting conditions):
    - Database of 140 parent experts who want to be involved in research – the charity will email members a link to the survey.
    - Link to survey on quarterly newsletter
- Parent groups at the Royal Marsden – groups will be emailed information regarding the study along with a link to the survey web page.
- Martin House Research Centre family advisory board – via email link.
- Martin House and Northern Ireland children's hospices – email link via family Facebook page.
- Social media – a link to the survey will be shared on the study's Twitter feed.

Health care professional participants will be identified through:

- Association for Paediatric Palliative Medicine (APPM) – medical and nursing membership will be emailed a link to the survey via the APPM. The research team will not have access to individual contact details.
- Together for Short Lives:
    - All children's hospices, hospital and community children's palliative care teams are members - the charity will email members a link to the survey.
    - Other health care professional members - the charity will email members a link to the survey.
- The link will be sent to the Principal Investigators of the sites used for the previous qualitative interviews with a request that they disseminate to their teams and contacts (Royal Marsden Hospital, Evelina Children's Hospital, King's College Hospital, Great Ormond Street Children's Hospital, Cambridge University Hospital, East Anglia Children's Hospice, Northern Ireland Children's Hospice, Leeds Teaching Hospital Trust, Martin House Children's Hospice).
- Association of Palliative Care Social Workers – via email link.
- Children's Cancer Network – via email link.
- Children's Cancer and Leukaemia Group – via email link.
- Hospice UK membership – via email link.
- Social media – a link to the survey will be shared on the departmental. And study Twitter feeds.

Upon completion of round 1 of the survey, participants will be asked to recommend other experts to participate in the study by sending them a link to the survey or sharing the link on their social media (snowball sampling).

Eligibility criteria

Inclusion criteria

- Health care professionals:
    - Doctors - who have been providing care to children and young people with LLC/LTC for > 6 months.
    - Nurses - who have been providing care to children and young people with LLC/LTC for > 6 months.
    - Social workers - who have been providing care to children and young people with LLC/LTC for > 6 months.
    - Psychologists - who have been providing care to children and young people with LLC/LTC for > 6 months.
    - Other allied health professionals e.g., physiotherapists, play specialists, occupational specialists, spiritual care providers - who have been providing palliative care to children and young people with LLC/LTC for > 6 months.
- Parents/carers of a children and young people aged 0-18 years (up to their 18th birthday) who have a LLC/LTC.
- Bereaved parents of children and young people with LLC/LTC whose child has died within 12-24 months of participation. This time period was identified as being optimal in a study with bereaved parents who felt they would still remember what had happened, how they felt and what they needed clearly. They felt that at 12 months enough time had passed so recall was not significantly painful(48).

Exclusion criteria

- Health care professionals who have been working with children and young people with LLC/LTC for less than 6 months.
- Individuals who cannot complete an online survey written in English.

Sample size

COSMIN recommend a sample size of >100 for a quantitative study on content validity of a patient reported outcome measure(36). 50 to 99 participants per group is deemed adequate, whereas 100 or greater is considered very good, the highest possible rating (36). Delphi surveys in similar populations have reported varied response rates ranging from 44->80%(41, 49, 50). This study anticipates a sample size of 100-200 based on these reports of previous response rates and the numbers of participants that will be reached by the process described above. Reminders emails will be sent to potential participants at 1 week. Social media invitations will be posted weekly.

Study Procedure

A 3-4 round survey will be designed using SmartSurvey (an online survey platform). The survey and instructions will be piloted in a small group (3-5) of participants from the target population before use to ensure that the questionnaire and instructions are understood(51). The SmartSurvey platform has been chosen for its ease of use and ability to export data which is encrypted and stored within the UK. The research team will not have direct contact with potential participants.

The first page of the online survey will explain what the survey is for and how the survey items were decided upon. Reminders for each round will be sent after 1 week to maximise participation. Participants will be asked to recommend others that could be invited to participate in order to maximise the expertise of the panel. Demographic data will be collected as follows:

- Health care professionals – profession, current role, length of children and young people palliative care experience, generic place of work e.g., hospital, hospice, community, gender and age.
- Parents/carers – age, relationship to child, child's diagnosis, child's age, ethnic background of child and parent/carer, gender of child and parent/carer and area of the country they live in.

Narrowing down (round 1)

Items for inclusion in round 1 of the Delphi will be chosen from:

1. Symptoms and concerns in children and young people with LLC/LTC identified in the research team's previous qualitative interview study with children and young people with LLC/LTC, their parents/carers and siblings, health care professionals and NHS commissioners.
2. Results of a systematic review on symptoms and concerns in children and young people with LLC/LTC(16).

See appendix 4 for an example of the items to be included. Symptoms and concerns will be presented in random order(43, 44). Participants will be asked to select the 20 symptoms and concerns that they believe to be the most important to be included in the C-POS. They will also be asked to suggest items that are missing and be asked to justify and explain their choices in a free text box. Items will be selected for inclusion in subsequent rounds as follows:

- Items that are selected by >50% of participants will be included in the subsequent ranking rounds(52).
- If more than 30 items are selected by >50% of participants then the items selected by >30% of participants will be included in subsequent rounds(44).
- If new items are suggested during round 1, they will be compared with the existing items and discussed by the research team and members of the steering group (including PPI representatives) to gain expert consensus on whether they should be added to round two for evaluation by participants (34, 46).

For this phase of the study, results will be analysed by participant group (parents/carers and professionals and by both groups combined). At this stage of the study an academic and PPI steering group meeting will be held to review the data and make decisions on items to be carried forwards to the next round of the Delphi survey. This will include discussion of the suggested new items and free text justification of choices. This meeting will ensure that if there are differences between priorities identified by parents/carers and health care professionals these are reconciled by an expert panel.

Participant email addresses will be collected (with consent) during this round so that participants can be sent invitations to participate in further rounds. These will be kept in a password protected file on a secure server.

Ranking (rounds 2 and 3)

Round 2 will occur 1-2 weeks after the close of round 1 and will be open for 2 weeks. Round 3 will occur a 1-2 weeks after round 2 closes and again will be open for 2 weeks. Participants in rounds 2 and 3 will need to have participated in round 1. Participants will be presented with the results from the previous round outlining items that were removed for round 2, any relevant comments from participants and any new items that have been added. Participants will then be asked to rank the symptoms and concerns remaining from round 1 in order of priority for inclusion in the C-POS measure. Items will be ranked in descending order, from the most to least important. Participants will be asked to explain their justifications for their rankings in a free text box. Reminders will be sent at 1 weeks.

For round 3, participants will again be sent an email with a link to the survey. They will be given the following feedback from the previous round:

- the median rank of each item from round 2 and where they ranked each item,
- Kendall's W coefficient of concordance (in layman's terms i.e. weak, moderate or strong agreement),
- top half rank (the percentage of experts who ranked items in their top 50%)

- relative comments/justifications made by respondents.

Results will be presented for the participants in full (parent/carer and professional data) and stratified by group(34). Participants will again be asked to rank symptoms and concerns based on the feedback above. This time items to be ranked will be presented according to median rank rather than randomly in order to aide achievement of consensus(53). Participants will again be asked to justify their ranking decision in a free text box. Participants will be asked a final question on whether they would be willing to participate in a further round if consensus has not been reached. Reminders will be sent again at 1 week.

Data will be analysed in the same way as round 2. If consensus has been reached (Kendall's W >0.7) then the study will stop. If consensus has not been reached (W<0.7), a McNemar test will be performed to see whether there is a difference in median ranking between rounds 2 and 3. If there is no significant difference in median rankings between the two rounds the study will stop(52). If consensus has not been reached and there is a significant difference in median ranking between rounds a further round will be considered if >50% of respondents indicate they would be willing to participate in another round. If <50% of respondents were willing to take part in a fourth round, the study will stop. In palliative care, perfect agreement may often not be realistic due to different values, world views and ethical dilemmas concerning medical decision making(51). There are a diverse range of LLC/LTC that affect children and young people and they are cared for in a wide range of settings which adds to this complexity. Results from this Delphi exercise will be taken forwards to an item generation to gain further expert agreement on items to be included in the C-POS in order to further evidence face and content validity (see item generation meeting section of this protocol).

This study will be reported according to guidance on Conducting and REporting of DElphi Studies in palliative care CREDES(51) and recommendations by Paré et al on ensuring rigor in ranking-type Delphi surveys(44). All data will be anonymised.

Consent

Written information about the purpose of the Delphi survey will be included at the beginning of each round (appendix 3 and 4). Information will be given on the rationale for the study, what participation will entail, how data will be used and stored and steps that will be taken to ensure confidentiality(54). This information has been reviewed by our PPI stakeholders. No participant identifiable information will be used in any publication or in the dissemination of results. Participants will be asked to complete a consent form at the beginning of each round of the survey (included at the beginning of the survey) to indicate that they consent to participation (appendix 5). In the unlikely event that participants find the content of the survey distressing, they will be given the contact number for the

Together for Short Lives helpline on the study information page. This helpline is run by experienced staff who are used to talking to parents who are distressed.

Analysis

Analysis will be undertaken using SPSSv25 or equivalent. Demographic data of participants will be analysed using descriptive statistics.

Round 1 (narrowing down)

All symptoms and concerns that are not selected by >50% of participants from each group (parents/carers and professionals) will be eliminated. If more than 30 items are selected by >50% of participants, then the items selected by >30% of participants will be included in subsequent rounds.

Round 2 (ranking)

After round two and three analysis will consist of:

- the median rank of each item,
- Kendall's W (to measure the level of agreement between participants)
- Percentage of participants placing each item in the top half of their list(43, 44).

Kendall's W will be interpreted according to guidance by Schmidt et al(43) on conducting ranking type Delphi surveys:

- ≥0.1 – very weak agreement
- ≥0.3 – weak agreement
- ≥0.5 – moderate agreement
- ≥0.7 – strong agreement
- ≥0.9 – unusually strong agreement

If Kendall's W is not ≥ 0.7 after round 3 a McNemar test will be conducted to see whether there is a difference in median ranking between rounds.

Free text comments will be collated by symptom and concern and analysed thematically. These will be presented and discussed at the item generation meeting (see below).

Data will be analysed by the C-POS PhD candidate (LC) on a laptop and/or desktop computer either within the Cicely Saunders Institute or off site. Data within the Smart Survey site is encrypted and stored within the UK. Once it has been extracted participant email addresses will be stored in a locked file on the Cicely Saunders Institute SharePoint. All other data will be anonymised and stored on SharePoint in an SPSS file.

Item generation meeting

In order to generate the final items for the C-POS, an item generation meeting will be held after the Delphi survey to further gain expert stakeholder input to enhance face and content validity of the C-POS. The C-POS steering group (including PPI representatives) will be given a brief presentation on the use of patient-reported outcome measures in healthcare and the evidence for the need of the C-POS. They will then be presented with the findings from:

- The two systematic reviews carried out by the research team on a) symptoms and concerns in children and young people with LLC/LTC(16) and b) enhancing validity, reliability and participation in self-reported health outcome measurement for children and young people: a systematic review of optimal recall period, response scale format and administration modality.
- Feedback from the Young Person's Advisory Group at Great Ormond Street Children's Hospital on response format, recall period and administration mode of outcome measures for children and young people.
- The results of the main findings from the qualitative interview study on symptoms and concerns in children and young people with LLC/LTC including the analysis framework.
- The results of the above Delphi survey including item mean rank (overall and stratified by participant group), Kendall's W, the percentage of participants placing each item in the top half of ranked list and any free text comments provided by participants. Item mean rank will also be presented by domain. Domains were created using the World Health Organisation's definition of paediatric palliative care (see appendix 1 for items) (55) and the results of the previously conducted qualitative interviews. The domains are physical, social and practical, emotional and psychological, spiritual and existential and normality.

After the presentations the symptoms and concerns ordered by domain and mean rank from the Delphi will be presented by the PhD candidate (LC) and discussed with the stakeholder group to gain final agreement for which items to include in the C-POS. Discussion will include whether consensus was reached during the Delphi and any disparities between groups (parent/carer and health care professionals) in order to reconcile any differences. Any disagreement will be worked through as a group with the PI (RH) acting as chair.

Once items for inclusion have been agreed there will be a discussion regarding response format, recall period and administration mode(s) of the C-POS, led by the study PhD candidate and based on discussions with the Great Ormond Street Young Person's Advisory Group and the results of the aforementioned systematic review of evidence of these in children. A presentation will be given on suggestions for these. There will be opportunity for discussion of these suggestions, again chaired by

RH. These will be agreed for both parents/carers and child age. PPI representatives will be briefed before the meeting on the content and will be offered support if needed afterwards, either by discussion with the research team or by referral to the Together for Short Lives advice line.

After the meeting LC (PhD candidate) will take the list of agreed symptoms and concerns for inclusion and draft the first parent/carer and age appropriate versions (version1) of C-POS. Response format, mode and recall will be based on the systematic review results and outcomes of the stakeholder group discussion. These will be reviewed and finalised by the research team to ensure they represent the discussion held in the stakeholder group. A summary of the stakeholder group discussions will be sent electronically to the group participants along with version 1 (parent/carer and child versions) of the C-POS for final comment. Minor changes will be made, if required, prior to the next phase of the overall C-POS study (cognitive interviews, subject to a subsequent ethics application via IRAS).

6 ETHICAL AND REGULATORY CONSIDERATIONS

6.1 Assessment and management of risk

Co-applicant on the study, Professor Bobbie Farsides (Professor of Clinical and Biomedical Ethics), has led the ethical dimensions of this study. Professor Farsides has 20 years' experience of speaking to health care professionals about the most ethically challenging aspects of their work. She will be attending the item generation meeting. Professor Farsides has provided a 2 hour training session to summarise the recommendations and their translation to research practice of the report she led "Children and clinical research: ethical issues" http://nuffieldbioethics.org/wp-content/uploads/Children-and-clinical-research-full-report.pdf. Researchers have also completed training in communication skills, paediatric palliative care and the role and skills of play therapy in research studies.

We have appointed an external ethics reviewer, Dr Sara Fovargue of Lancaster University who has dual qualifications in ethics and medical law and was a member of the Nuffield Council committee on ethics and research with children http://www.lancaster.ac.uk/law/people/sara-fovargue. She authors and approves our annual ethical check to the European Research Council.

Parents will be directed to our partner Together for Short Lives if they require additional support and guidance with respect to their child's condition and to access support for their child.

Information and consent forms will clarify the content of the Delphi survey to ensure that full information is provided and there are no surprises regarding content.

There will be no direct benefit to participants, although parents recruited form this population who have participated in other studies have acknowledged their perceived positive experience and personal reward. The participant information sheet explains the content of the survey and includes the contact details for the Together for Short Lives helpline.

Regulatory Review & Compliance

Before any site can enrol patients into the study, the Principal Investigator (Professor Harding) will ensure that appropriate approvals from participating organisations are in place.

For any amendment to the study, the Chief Investigator or designee, in agreement with the sponsor will submit information to the appropriate body in order for them to issue approval for the amendment. The Chief Investigator or designee will work with KCL R&D department so they can put the necessary arrangements in place to implement the amendment to confirm their support for the study as amended.

Amendments

The process for making amendments will be led by the study PI (Harding) and taken to full Steering Group (or by email approval of the majority if not meeting is due) and with PPI consultation. It will be submitted to KCL for review and no change initiated until full approval.

Any approved amendments will be incorporated into study materials with revised version number and the change noted and stored in the Site File.

6.2 Peer review

Peer review was conducted by the Funder (the European Research Council) and the PI presented the study at a grant panel in Brussels to a multiprofessional decision making panel of approximately 30 scientists from across Europe who individually and independently scored the propossal.

6.3 Patient & Public Involvement

To date our PPI involvement has been through the NGO “Together for Short Lives” and through PPI parent representatives on our group. They have commented on the aims, methods and dissemination plan of all phases of the C-POS study so far.

These groups are also represented on our Steering Group. They will be part of all decision making, interpretation and dissemination activities.

6.4 Protocol compliance

Accidental protocol deviations can happen at any time. They must be adequately documented on the relevant forms and reported to the Chief Investigator and Sponsor immediately.

Deviations from the protocol which are found to frequently recur are not acceptable, will require immediate action and could potentially be classified as a serious breach.

6.5 Data protection and patient confidentiality

Each study participant will have their email address/name/study ID code sheet stored separately to their survey responses'. Data will be stored for a period of 8 years within a secure computer file. Future secondary analysis of the data will require a further application for ethical approval in on the presumption that it is seen as compatible processing. Secondary analysis will be subject to the participant's prior enactment of their right to withdraw consent and data.

Participants' demographic data will be stored in Excel files in password protected folders on the server. No research study staff will have access to NHS patient files.

Participants will be able to have their data removed from the study dataset or archive at any point, and for it to be removed from results up until these are presented/published and in the public domain. This will also include the removal of consent forms, participant name from the study code ID list, and removal from all recruitment records. Removal can be requested by email or letter.

The maintenance of Site Files, data management plans and adherence to these, and reporting will be overseen by the Faculty Research Development Manager. Data Protection Officer at KCL will conduct a review of the full protocol for the ERC reporting and will provide guidance and issue a letter of review. Any breaches will be reported immediately (same day as the breach becomes known) to the Data Protection Officer.

6.6 Indemnity

The lead sponsor, King's College London, will take primary responsibility for ensuring that the design of the study meets appropriate standards and that arrangements are in place to ensure appropriate conduct and reporting. King's College London also provides cover under it's No Fault Compensation Insurance, which provides for payment of damages or compensation in respect of any claim made by

a research subject for bodily injury arising out of participation in a clinical trial or healthy volunteer study (with certain restrictions).

6.7 Access to the final study dataset

The study research team: Professor Richard Harding as PI, Professor Wei Gao as study statistician, Dr Debbie Braybrook (Research Associate), Anna Roach (research assistant), Lucy Coombes (PhD student), Dr Clare Ellis-Smith and Dr Katherine Bristowe will have access to the full dataset.

The PI will allow site investigators to access the full dataset if a formal request describing their plans is approved by the steering group.

7 DISSEMINIATION POLICY

7.1 Dissemination policy

KCL owns the data arising from the study.

On completion of the study, the data will be analysed and tabulated, and a Final Study Report prepared.

The full study report will be provided to the Ethics Committee and the findings will available in peer review publications. The PI will lead publications with agreement for lead authorship by co-applicants. All data will be submitted for peer review within 6 months of data collection.

We will acknowledge our funder and clinical and community partners in the manuscript.

We will keep the stakeholder community informed via a newsletter that will be distributed via our project website and through Together for Short Lives. We will give the URL to all participants.

We will post the outputs of the research to any participant/family who requests it.

Following publication, the data will be available for use upon reasonable request to the PI (subject to consent of participants).

7.2 Authorship eligibility guidelines and any intended use of professional writers

We will adhere to The International Committee of Medical Journal Editors defined authorship criteria for manuscripts submitted for publication. This can be found at ***http://www.icmje.org/recommendations/browse/roles-and-responsibilities/defining-the-role-of-authors-and-contributors.html***

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Appendix 1- Study Information Sheet – Parents/Carers

C-POS- what matters to children, young people and their families facing serious illness?

What is the aim of this research study?
We would like to invite you to be part of an online research survey to help us prioritise which symptoms and concerns matter to children and their families facing serious illness. The goal of this study is to help us develop a "checklist". This would contain a brief list of the things that often matter most to children and their families. The idea is that when you see health professionals, we can try and make sure that you are asked about the things that matter to you, and that we check that these things are being addressed.

What does taking part involve?
This study involves completing an online survey on three separate occasions over the next 2-3 months. Questions in the survey are based on symptoms and concerns previously identified by children and young people with life-limiting/life-threatening conditions, their carers, siblings, health care professionals and NHS commissioners during face to face interviews. You will be asked whether each symptom or concern identified is a priority to be included in the final measure. This type of survey is called a Delphi survey, and it will try to reach agreement on items to be included in the checklist. We will also ask you for some basic information regarding you and your child.

How will we collect information?
Information will be collected by an online survey. The survey site used is encrypted (which means no-one outside this study can read responses) and all data is stored on UK servers. We will ask for your email address during the survey so that we can send you a link to complete rounds two and three.

What will happen to that information?
The information collected will be transferred to secure files by the research team. All data will be anonymous, and we will not be asking for your name. When we publish or present the findings it will be done in such a way as to minimise the potential of you being identifiable.

Do I have to take part?
You do not have to take part in this research study- it is entirely voluntary. You can take your time in deciding whether to take part, and you may want to talk it over with your care team or others.

Can I change my mind after saying yes?
If you do decide to take part you can change your mind at any time before or during the survey. If you complete the first survey (round 1), you will be sent a link to round 2 in a few weeks. There will be a third round a few weeks after round 2. If you do not want to, you do not have to complete the subsequent rounds.

Are there any direct benefits to taking part?
Taking part will not provide any direct benefits to you or your child, although we hope that there will be future improvements to care for others.

What if there is a problem?
If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions [Professor Richard Harding, 02078485518 or richard.harding@kcl.ac.uk]. In the event that something does go wrong and you are harmed during the research you may have grounds for legal action for compensation against King's College Hospital NHS Foundation Trust and/or King's College London, but you may have to pay your legal costs. If you

find participation in the survey distressing in any way and you feel that you need to speak to someone for support then please contact the Together for Short Lives helpline on: 0808 8088 100.

Our compliance with legal requirements regarding your data

King's College London will keep your contact details confidential. King's College London will use this information as needed, to contact you about the research study and to oversee the quality of the study. Certain individuals from King's College London and regulatory organisations may look at your research records to check the accuracy of the research study. King's College London will only receive information without any identifying information. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details.

King's College London is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. Our lawful basis for processing your personal data under the General Data Protection Regulation (GDPR) is 'task in the public interest' (as a university, doing research is part of our public task). Similarly, we will be processing your health data (which is a special category of personal data under the GDPR) because it is 'necessary for scientific or historical research purposes'. King's College London will keep identifiable information about you for 8 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

You can find out more about how we use your information by reading King's College London's core privacy notice at https://www.kcl.ac.uk/terms/privacy.aspx, or by contacting Albert Chan (the Data Protection Officer for King's College London) on email at info-compliance@kcl.ac.uk or telephone at 0207 848 7816.

Thank you for reading this information and for considering our study.

Appendix 2 - Study Information Sheet – Professionals

C-POS- what matters to children and young people with serious illness?

What is the aim of this research study?
We would like to invite you to be part of an online research survey to help us prioritise which symptoms and concerns matter to children and their families facing serious illness. The goal of this study is to help us develop a "checklist". This would contain a brief list of the things that often matter most to children and their families. The idea is that when they see health professionals, we can try and make sure that they are asked about the things that matter to them, and that we check that these things are being addressed.

What does taking part involve?
This study involves completing an online survey on three separate occasions over the next 2-3 months. Questions in the survey are based on symptoms and concerns previously identified by children and young people with life-limiting/life-threatening conditions, their carers, siblings, health care professionals and NHS commissioners during face to face interviews. You will be asked whether each symptom or concern identified is a priority to be included in the final measure. This type of survey is called a Delphi survey, and it will try to reach agreement on items to be included in the checklist. We will also ask you for some basic information regarding your professional role.

How will we collect information?
Information will be collected by an online survey. The survey site used is encrypted (which means no-one outside this study can read responses) and all data is stored on UK servers. We will ask for your email address during the survey so that we can send you a link to complete rounds two and three.

What will happen to that information?
The information collected will be transferred to secure files by the research team. All data will be anonymous, and we will not be asking for your name. When we publish or present the findings it will be done in such a way as to minimise the potential of you being identifiable.

Do I have to take part?
You do not have to take part in this research study- it is entirely voluntary. You can take your time in deciding whether to take part, and you may want to talk it over with others before deciding.

Can I change my mind after saying yes?
If you do decide to take part you can change your mind at any time before or during the survey. If you complete the first survey (round 1), you will be sent a link to round 2 in a few weeks. There will be a third round a few weeks after round 2. If you do not want to, you do not have to complete the subsequent rounds.

Are there any direct benefits to taking part?
Taking part will not provide any direct benefits to you, although we hope that there will be future improvements to care for others.

What if there is a problem?
If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions [Professor Richard Harding, 02078485518 or richard.harding@kcl.ac.uk]. In the event that something does go wrong and you are harmed during the research you may have grounds for legal action for compensation against King's College Hospital

NHS Foundation Trust and/or King's College London, but you may have to pay your legal costs.

Our compliance with legal requirements regarding your data

King's College London will keep your contact details confidential. King's College London will use this information as needed, to contact you about the research study and to oversee the quality of the study. Certain individuals from King's College London and regulatory organisations may look at your research records to check the accuracy of the research study. King's College London will only receive information without any identifying information. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details.

King's College London is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. Our lawful basis for processing your personal data under the General Data Protection Regulation (GDPR) is 'task in the public interest' (as a university, doing research is part of our public task). Similarly, we will be processing your health data (which is a special category of personal data under the GDPR) because it is 'necessary for scientific or historical research purposes'. King's College London will keep identifiable information about you for 8 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

You can find out more about how we use your information by reading King's College London's core privacy notice at https://www.kcl.ac.uk/terms/privacy.aspx, or by contacting Albert Chan (the Data Protection Officer for King's College London) on email at info-compliance@kcl.ac.uk or telephone at 0207 848 7816.

Thank you for reading this information and for considering our study.

Appendix 3 - Consent Form

1. I confirm that I have read the information regarding this study. *

- ☐ Yes
- ☐ No

2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason. *

- ☐ Yes
- ☐ No

3. I understand that information held about me by King's College London (including my email address), will be used to invite me to participate in future rounds of this study. *

- ☐ Yes
- ☐ No

4. I give permission for anonymous data collected about me to be used in the study report, publications and presentations. *

- ☐ Yes
- ☐ No

5. I consent to participation in this study. *

- ☐ Yes
- ☐ No

Appendix 4 – Sample of symptoms and concerns to be included in Delphi

1. Skin issues
2. Bowel problems
3. Appetite
4. Being physically unable to eat or drink as much as normal
5. Changes in weight
6. Infections and/or impaired immunity
7. Impact of medical interventions and treatment such as central lines, prosthesis
8. Muscle weakness
9. Impaired growth
10. Reduced physical function
11. Changes in consciousness
12. Dystonia
13. Seizures
14. Pain
15. Breathing difficulties
16. Cough
17. Excess respiratory secretions
18. Sleeping difficulties
19. Tiredness or fatigue
20. Appearance
21. Low blood counts
22. Impaired growth
23. Agitation
24. Nausea
25. Vomiting
26. Ability to do usual self-care
27. Side effects of medications
28. Fertility concerns
29. Hair loss

<u>Social and practical concerns</u>

1. Having access to technology to stay connected
2. Opportunity to discuss advance care planning and/or resuscitation
3. Discussion of preferred place of care
4. Discussion of preferred place of death
5. Discussion of wishes for life/death
6. Opportunity to set individual goals or outcomes
7. Avoiding unplanned hospital admissions
8. Identifying wishes for life
9. Care needs changing over time
10. Balancing needs of child with rest of family
11. Burden of medication regime
12. Communication difficulties – child
13. Communication with health and social care professionals
14. Eating
15. Friendship – adaptation to dynamics and challenges due to illness
16. Impact on play and hobbies
17. Impact on school life
18. Financial concerns –benefits, cost of hospital visits and stays, equipment etc

19. Impact on work life
20. Information provision – about illness, services, medication, developmentally appropriate, understanding illness, not having enough information
21. Meeting cultural needs
22. Access to 24/7 care at home
23. Access to 24/7 telephone advice
24. Care co-ordination
25. Quality of care
26. Access to equipment
27. Access to holistic care
28. Timely hospital discharge
29. Transitions – school, services
30. Fluctuating needs
31. Logistics of care e.g., organising appointments/carer rotas
32. Decision making
33. Discord between family members
34. Discord between service providers and family
35. Having to do things differently – eating, play/hobbies, routines, family life
36. Housing
37. Wanting to get back to normality
38. Adjusting to a new normal
39. Relationships
40. Equal opportunities with other children
41. Achieve life goals
42. Initiate and maintain sexual relationships
43. Breaking bad news
44. Shared decision making
45. Missing home

Spiritual and existential

1. Determination to live life to the fullest
2. Determination to survive
3. Meaning of life
4. Religion and faith
5. Spiritual support – access and availability
6. Uncertainty of the future – fear of life unlived, length of life
7. Worry about death
8. Will I be remembered
9. Am I dying?
10. Life devoid of meaning
11. Connected with God or something larger than self
12. Appreciate life as a gift
13. Keeping the spirit alive
14. Must survive the hard bits of illness

Emotional and psychological

1. Behaviour regression
2. Being different

3. Looking different
4. Challenges to independence
5. Problems with cognitive ability and learning needs
6. Loss of control (child)
7. Parents wanting to have control over their child's health
8. Impaired ability to express and/or understand emotions and feelings
9. Opportunity to meet people in a similar situation
10. Privacy
11. Grumpy/moody
12. Anger
13. Feeling like a burden
14. Interruption to family life
15. Worry about family
16. Wanting to protect family
17. Fear, worry and anxiety
18. Irritation or annoyance
19. Low mood, sadness
20. Memory making
21. Support for family members
22. Trust
23. Access to/availability of psychological support
24. Treatment related anxiety and worry
25. Happiness
26. Feeling stupid
27. Declining school performance
28. Reduced concentration
29. Lack of self-worth
30. Aggression
31. Non-adherence to treatment

Appendix 5– Amendment History

Amendment No.	Protocol version no.	Date issued	Author(s) of changes	Details of changes made

List details of all protocol amendments here whenever a new version of the protocol is produced.

Protocol amendments must be submitted to the Sponsor for approval.

Appendix L Cognitive interview protocol, patient information sheets, consent forms and demographic collection sheet

FULL/LONG TITLE OF THE STUDY Children's Palliative Outcome Scale (C-POS) – cognitive interview study to determine comprehensibility, comprehensiveness and feasibility.

SHORT STUDY TITLE / ACRONYM C-POS cognitive interview study

PROTOCOL VERSION NUMBER AND DATE V3.3 17/11/2021

RESEARCH REFERENCE NUMBERS

IRAS Number: 282412

FUNDERS Number: 772635

LIST of CONTENTS

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STUDY SUMMARY

Study Title	Children's Palliative Outcome Scale – Cognitive Interview Study
Internal ref. no. (or short title)	C-POS
Study Design	Qualitative –cognitive interviews
Study Participants	c) Children and young people affected by a life-limiting or life-threatening condition d) Parents/carers of these children
Planned Size of Sample (if applicable)	Cognitive interviews – up to 50
Follow up duration (if applicable)	N/A
Planned Study Period	1 months
Research Question/Aim(s)	Aim: To cognitively test version 1 of C-POS with children and young people with life-limiting and life-threatening conditions and their parents/carers. Objectives: c) To conduct cognitive interviews with children and young people and families to establish feasibility, comprehensibility and comprehensiveness of the C-POS measure.

FUNDING AND SUPPORT IN KIND

FUNDER(S) (Names and contact details of ALL organisations providing funding and/or support in kind for this study)	**FINANCIAL AND NON FINANCIALSUPPORT GIVEN**
European Research Council	**1.8 million Euros**

ROLES AND RESPONSIBILITIES OF STUDY MANAGEMENT COMMITEES/GROUPS & INDIVIDUALS

The **Study Steering Group** consists of PI Professor Harding (Cicely Saunders Institute, King's College London) co-apps Professor Lorna Fraser (Martin House Research Centre, University of York), Professor Murtagh (Wolfson Palliative Care Research Centre, Hull York Medical School), Professor Wei Gao (Cicely Saunders Institute), Professor Higginson (King's College London), Professor Farsides (Brighton and Sussex Medical School), Professor Bluebond-Langner (Louis Dundas Centre, University College London). These members are funded by the study and responsible for delivery of the protocol. Independent members of the Steering Group are Dr Anna-Karenia Anderson (Consultant in Paediatric Palliative Medicine, Royal Marsden), Lydia Bate(PPI), Dr Debbie Braybrook (King's College London), Dr Katherine Bristowe (King's College London), Dr Rachel Burman (Palliative Care Consultant, King's College Hospital), Lizzie Chambers (Together for Short Lives), Lucy Coombes (PhD candidate, King's College London), Professor Sir Alan Craft, Dr Finella Craig (Consultant in Paediatric Palliative Medicine, Great Ormond Street Hospital), Professor Julia Downing (International Children's Palliative Care Network), Dr Clare Ellis-Smith (King's College London), Dr Sara Fovargue (Lancaster University), Dr Ann Goldman (Consultant in Paediatric Palliative Medicine), Jane Green (PPI member), Dr Michelle Hills (Consultant in Paediatric Palliative Medicine, Martin House Children's Hospice), Dr Gillian Hughes (Consultant in Paediatric Palliative Medicine, Evelina Children's Hospital), Dr Joanna Laddie (Consultant in Paediatric Palliative Medicine, Evelina Children's Hospital), Dr Steven Marshall (King's College London), Angela Logan (PPI), Dr Linda Maynard (Nurse Consultant, East Anglia Children's Hospice), Renee McCulloch (Consultant in Paediatric Palliative Medicine, Great Ormond Street Hospital), Dr Eve Namisango (African Palliative Care Association), Dr Sue Picton (Consultant Paediatric Oncologist, Leeds Teaching Hospital) and Anna Roach (King's College London). These individuals will be responsible for reviewing progress, delivery and quality of the research.

PROTOCOL CONTRIBUTORS

The protocol has been led by PI Professor Harding, PhD Candidate Lucy Coombes and contributed to by steering group members (Dr Bristowe, Dr Ellis-Smith, Dr Braybrook, Professor Murtagh, Professor Fraser).

KEY WORDS: Paediatrics, outcome assessment, child, cognitive interviews, outcome measure

STUDY PROTOCOL

Children's Palliative Outcome Scale (C-POS)

1 BACKGROUND

Life-limiting conditions (LLC) in children and young people are defined as conditions for which there is no reasonable hope of cure and from which children or young people will die, such as Duchenne muscular dystrophy. Life-threatening conditions (LTC) are those for which curative treatment may be feasible but can fail, such as cancer(1, 2).

Epidemiology

Worldwide, it is estimated that each year 21 million children and young people (children and young people) with LLC/LTC conditions require input from palliative care services(3). Within the UK, there are estimated to be over 86,000 children and families living with a LLC/LTC condition, many of whom would benefit from palliative care services due to complex symptoms, social and psychological needs and the unpredictability of their condition(2). Almost 400 LLC/LTC conditions have been identified as appropriate for palliative care among children and young people(4). The number of children with LLC/LTC conditions in the UK is rising due to advances in medical care leading to slower deterioration. For these children, complexity of care is rising due to the increased use of medical technology such as home ventilation(1, 5). This is putting increased pressure on the resources of paediatric palliative care teams. Within England and Wales more than 5000 children and young people die each year from all causes(6). It is estimated that deaths due to LLC/LTC may account for 50% or more of these deaths(6). Mortality is highest in those under one year of age (peaking in the neonatal period) and decreases in middle childhood before rising again in adolescence(6). There is a significantly higher prevalence of LLC/LTC conditions in children and young people from both ethnic minority backgrounds and higher areas of deprivation in the UK(7).

Patient-reported outcome measurement in children and young people

A patient reported outcome measure (PROM) is defined as any measure of a patient's health status, elicited directly from the patient(8). PROM's range from single item symptom ratings e.g. pain scales, to complex multidimensional health-related quality of life tools(9). Patient reported outcome instruments may be either generic or disease specific(10).

Generic measures are useful for comparing general outcomes across different populations e.g., cancer vs. cardiac disease or for assessing interventions across a wide range of conditions(11). Some of these measures are used to assess health-related quality of life in healthy children so are more likely to have been validated based on large samples but may lack sensitivity for sick children and young people. They may also be too long for very unwell children to complete(12). Generic measures allow comparison of outcomes across groups of children and young people with different illnesses, which is essential for a discipline as wide and varied as children and young people palliative care. Disease specific instruments, on the other hand, are used to compare quality of life within a given condition e.g., cancer. Disease specific measures are assumed to be more sensitive to the implications of different illnesses and may be more appropriate for evaluating interventions or different treatments within children and young people with the same illness(12). Some patient reported outcome measures have been developed combining both approaches giving rise to generic measures with disease specific modules(13). In children and young people palliative care, an outcome measure is required that can capture symptoms and concerns that are important to children and young people with a wide range of conditions but is specific to the unique experience of this population(14).

Previous work and current study

This study is informed by and builds upon previous work conducted by the research team, following the principles of outcome measure development described by Rothrock(8). A systematic review has been carried out which identified that there is currently no suitable outcome measure for use with children and young people with palliative care needs(9). 106 qualitative, semi-structured interviews have been conducted with children and young people with LLC/LTC, their parents/carers, siblings, health care professionals and NHS commissioners to establish their priorities for outcomes of care This is one of the largest and most comprehensive qualitative interview studies to have been conducted in this population. Approval was received from the Bloomsbury ethics committee in 2019 (REC reference 19/LO/0033). In addition, systematic reviews regarding a) symptoms and concerns in children and young people with LLC/LTC (10) and b) optimal recall period, response format and administration mode for use with children and young people (pending publication) have also been conducted. A Delphi survey and item generation meeting have also been conducted, to establish parent/carer and health and social care professional priorities for which items identified in the semi-structured interviews should be included in version 1 of C-POS. This study seeks to cognitively test version 1 of C-POS with the target population to ensure it is comprehensible and feasible for use in practice.

2 RATIONALE

In adult settings routine use of PROMs has been shown to improve awareness of symptoms and concerns, identify unrecognised symptoms, increase the monitoring of symptoms, improve patient satisfaction and experience as well as having a positive effect on patient-clinician communication(11-16). Measuring outcomes in children and young people palliative care has repeatedly been identified as a research priority(17-19). However, a recent systematic review concluded that there is currently no suitable outcome measure available for use in children and young people palliative care, with domains of existing generic health-related quality-of-life measures not being relevant to all children with LLC/LTC and some domains within disease- specific measures are only relevant for that specific population(9).

A high quality PROM that includes physical, emotional, psychosocial and spiritual elements that contains sensitive indicators specific to children with LLC/LTC illness is required(20). The development of a validated outcome measure for children and young people will help address the current gap in high quality research in a population for whom there is currently very little evidence for good practice(21). It will also allow palliative care teams responsible for providing services to an increasing population of children and young people with complex needs a way to evaluate new interventions, compare treatments or services and aid clinical decision making(22).

Recent studies have highlighted that much of the research carried out with children with LLC/LTC conditions does not include them as participants(10, 23-27). One third of studies did not include children and young people as participants in one recent systematic review looking at symptoms and concerns in children and young people with LLC/LLC. Child self-report should be considered the gold standard for measuring symptoms and concerns in children and young people palliative care due to the subjective nature of many of the questions(28).

The overall development of the C-POS aims to address these gaps by including children and young people with a range of LLC/LTC conditions as participants both in the previous qualitative interviews and cognitive testing. This will establish the comprehensiveness, comprehensibility and feasibility of the C-POS measure. From this, an evidence-based outcome measure for use with children and young people and their parents/carers with

LLC/LTC conditions that is comprehensive and comprehensible with face and content validity will be developed. Ensuring that a PROM is relevant for the intended population and has clinical utility has been shown to be a facilitator in its implementation(29, 30). The inclusion of clinicians and parents in qualitative interviews and the Delphi survey and item generation meeting has provided further evidence of content validity of the C-POS. This will further ensure that the content is relevant and meaningful, which should aid implementation of the C-POS when it has been further validated.

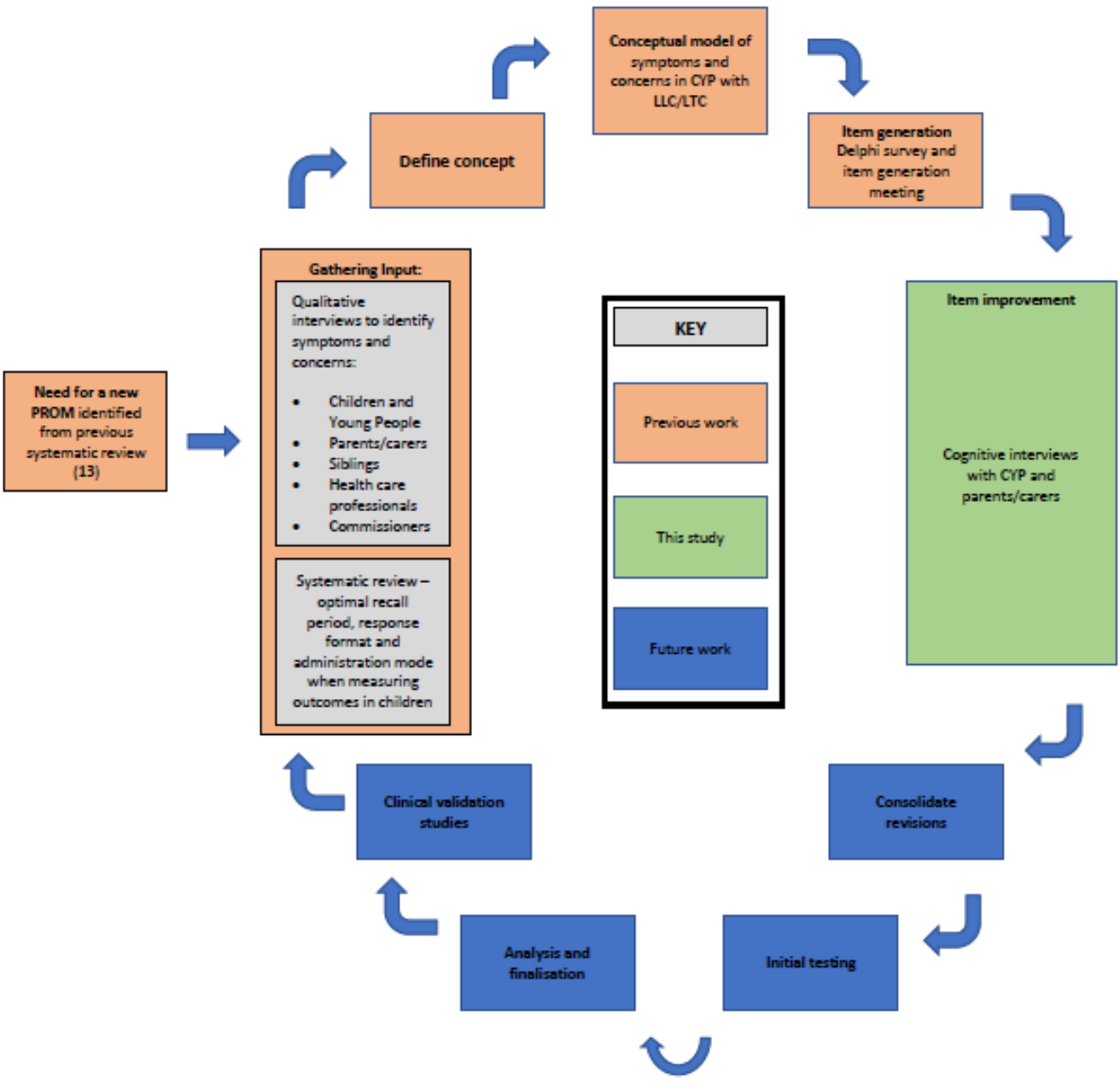
The overall aim of C-POS is the development and validation of an outcome measure for use with children and young people with LLC/LTC and their families. The qualitative interviews and systematic review on response format, administration mode and recall period are complete.

This protocol outlines the cognitive interview phase of the C-POS study. Cognitive interviews should be carried out to evaluate a PROM for comprehensibility, comprehensiveness and feasibility(31). This involves ensuring that the PROM instructions, items, response options and recall period are understood by the target population(32).

3 THEORETICAL FRAMEWORK

This study is conducted in line with the principles of outcome measure development proposed by Rothrock and the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN)(8, 31, 33). Figure 1 shows the patient reported outcome development process described by Rothrock with this study mapped on to it(8). This protocol incorporates the item improvement part of the process (shown in green).

Figure 1. Patient reported outcome development process and the C-POS study(8)



4 RESEARCH QUESTION/AIM(S)

To test the C-POS for comprehensibility, feasibility and comprehensiveness with children and young people with life-limiting and life-threatening conditions and their parents/carers.

4.1 Objectives

To conduct cognitive interviews with children and young people and families to establish feasibility, comprehensibility and comprehensiveness of the C-POS measure.

4.2 Output

The key output from this study will be initial age appropriate and parent/proxy versions of the C-POS (version 1) ready for further psychometric testing.

5 STUDY DESIGN and METHODS of DATA COLLECTION AND DATA ANALYIS

This study is a cross-sectional interview design.

Recruitment and sampling techniques

In order to ensure the C-POS is comprehensive, comprehensible and feasible for use with children and young peoples from 0-18 years (up to their 18th birthday) with any LLC/LTC and their parents/carers purposive sampling will be used.

Participants will be identified by health care professionals at the following participating sites during their weekly team meeting:

- Leeds Teaching Hospital NHS Trust
- Martin House Children's Hospice
- Royal Marsden NHS Foundation Trust
- East Anglia Children's Hospice
- Evelina Children's Hospital NHS Trust
- Royal Hospital for Children, Queen Elizabeth University Hospital, Glasgow
- Bradford Royal Infirmary
- East Lancashire NHS Trust

- Chestnut Tree House Children's Hospice
- Hertfordshire Community NHS Trust
- East Cheshire NHS Trust
- Forget Me Not Children's Hospice
- Leicester Children's Hospital
- Maidstone and Tunbridge Wells NHS Trust

When the recruiting clinical team next sees the children and young people and their family they will speak with them about the study and what participation will involve.

Social media will also be used to recruit potential participants via the following:

- The C-POS study's Twitter feed
- Twitter feeds of members of the study steering group.
- Together for Short Lives social media pages
- Facebook and Twitter pages of hospices and NHS services we have been collaborating with.
- Hospices and NHS sites we have working with may also share details of the study in their family newsletters or similar.

If the children and young people and family contact the research team directly in response to a social media post, a member of the research team will make contact and explain the study to them and what participation will involve. Following the introductory explanation, the children and young people and their parent/carer will be provided age specific written information on the study, either by post, email or via the clinical team. They will be given a minimum of 24 hours to consider participation. If after this time they agree, the healthcare team will pass on their preferred contact details to the C-POS research team.

No child will be recruited below age 5, although adult carers of children under 5 may be recruited. We have developed information sheets about the study for children and young people according to the following age groups: 5-7 years, 8-10 years, 11-15 years, 16-17 years. However, given the heterogeneity of this population, we may need to use versions for children based on developmental age rather than chronological age. We will ask parent/caregivers to advise which information sheet is most appropriate.

No participant will be paid to participate in this study, although reasonable (standard class) travel costs will be provided if the participant wishes to travel to the study researcher, and the restrictions of the COVID-19 pandemic allow. Researchers can provide cash reimbursement of travel costs for participants at the interview.

Eligibility criteria

Inclusion criteria

- Children and young people: from age 5 up to the age of 17 who are living with a LLC/LTC.
- Parents/carers: responsible for the primary care needs of a children and young people of any age who is living with a LLC/LTC.

Exclusion criteria

- Children and young people:
    - unable to communicate any views or wishes via their parent/caregiver or an interview
    - unable to read the C-POS questions or unable to understand the questions if they are read aloud
    - speaks a language not supported by the NHS Trust's translation service.
    - currently enrolled in another study.
    - deemed clinically unable to give consent/assent (34, 35).
    - who do not wish to participate.
- Parents:
    - deemed clinically unable to give consent due to concerns regarding well-being or an underlying mental health condition (35).
    - who do not wish to participate.

Sample size

It is anticipated that there will be at least seven participants per version of the C-POS, as per COSMIN recommendations(31). If required, amendments to the first version of the C-POS will be made after four participants have completed interviews. Any difficulties with the measures and potential amendments will be discussed by the scientific members of the study steering group and agreed as a team. The next three participants per group will then be given the amended version. A decision will need to be made at this point as to whether further changes and cognitive testing is required.

Cognitive interviews will be conducted using version 1 of the C-POS to further test for comprehensibility, comprehensiveness and feasibility of the final measure (objective c). It is essential that the C-POS instructions, items, response options and recall period are understood as intended. If they are not, the information obtained may be incorrect or respondents may not understand how to complete the C-POS(31).

Method

This will be a qualitative, cross-sectional study, similar to methods described by Murtagh(36). For this study think aloud and probing methods will be used, which involves the participant sharing their thoughts while answering each question, and the researcher asking probing questions to clarify understanding and reasons for selecting a score(37). Questions will draw upon the cognitive processing model described by Tourangeau – understanding the question, recall of relevant facts, making a judgement and selecting a response(38). There are different versions of C-POS:

- children and young people 5-7 years (or cognitive equivalent)
- children and young people 8-12 years (or cognitive equivalent)
- children and young people 13-18 years (or cognitive equivalent)
- Parent/carer of a child ≥ 2 years old
- Parent/carer of a child < 2 years old

For the children and young people versions 8-12 years and 13 -18 years different recall periods and response formats will be tested. If the children and young people finds a harder version difficult, we will move to an easier version during the interview. Versions will be amended as the study progresses, in line with any difficulties encountered with the cognitive interview process.

One-to-one interviews will be conducted with children and young people with LLC/LTC conditions and their parents/carers. children and young people will be able to have their parent/carer with them during the interview if they wish. These will be carried out either face-to-face in a location of the participants choice, or virtually via video call software (i.e. Zoom, MS Teams or Skype). The interviews will be conducted by members of the team that have been trained in conducting qualitative interviews with children with LLC/LTC and their parents/carers and have previous experience of doing this. Participants will be purposively sampled to ensure that there are between four and seven cognitive interviews conducted for each version of the C-POS. Sampling will also ensure that children with both malignant and

non-malignant diagnoses cognitively test each version. The number of participants recruited to cognitive testing will be dependent on the number of versions of C-POS developed. It is anticipated that there will be a parent/carer version and three or four age-appropriate versions.

Participants will be asked to read each C-POS question out loud (younger participants may need the questions reading to them by the researcher or parent). They will then be asked to speak out loud as they answer to elicit insights on their thought processes and decisions regarding responses(39) . This will provide an understanding of the cognitive processes used to formulate answers and checks how questions have been interpreted(40). The researcher will use concurrent verbal probing during the process. These probes will be both spontaneous to explore responses and non-verbal cues such as hesitations(41), and include predefined areas for exploration. The interview guide will contain probes regarding comprehension, different recall periods, the use of different response options, format and missing items(36, 38). In some instances, if interviews are conducted remotely, the research team may have to enlist a child's parent/carer to help facilitate the interview. Notes will be made during the interview using a notes template based on the structure of the question and probe sheet. All interviews will be digitally audio-recorded and transcribed verbatim, either within the Cicely Saunders Institute, King's College London, or remotely on secure departmental laptops. All audio files and transcripts will be password protected and stored on a secure server.

Consent

Consent and assent will either be taken by the researcher prior to conducting the interview, or by a team member of the recruiting site if they have been appropriately trained to do so e.g., up to date Good Clinical Practice training. The person taking consent will ensure that potential participants understand the nature of participation and will ensure that expression of interest in the study does not assume consent. The person taking consent will clarify again how the data will be collected (i.e. one-to-one interview) and the steps to maintain confidentiality (i.e. that no identifying information will be used in publications or presentations to external audiences).

Parents will be required to provide written consent for their child's participation up to and including the age of 15, and the child will provide a statement of informed voluntary participation/permission. From age 16-17 the young person may give written consent if their treating clinician believes that the individual has sufficient understanding and can give full consent independent of their parents/caregivers. Parents/carers will also be required to

provide written consent for their own participation. If interviews are conducted remotely then parents/carers will be asked to complete the consent form at the beginning of the interview so that the researcher can go through it with them and ensure they understand it, and then either send the research team a photograph or scanned copy via email or post it.

Children and young people will, following approval by their parent/caregiver, have the study explained to them in appropriate language, following prior guidance from the parent/caregiver on the timing and manner for optimal comprehension. Those 16 years and over will be asked to give their own written consent.

A potential participant with limited ability to indicate written consent on the form may give verbal consent in the presence of their parent/caregiver, who will sign alongside the researcher signature as witnesses to that consent. Children under 16 years old will be given the opportunity to complete an assent form.

If interviews are conducted remotely then the children and young people and/or consenting parent/carer will be asked to complete the consent and/or assent form at the beginning of the interview so that the researcher can go through it with them and ensure they understand it, and then either send the research team a photograph or scanned copy via email or post it. If posted, the researcher will ensure that the participant is sent a copy to keep for their own records. Alternatively, a trained member of the local recruiting team may take consent (see above) and forward the consent forms to the research team prior to the interview taking place. Participants will have a copy of the appropriate patient information sheet(s) and consent form available at the point of consent for them to keep a copy of.

Information sheets will inform participants of the Data Protection Officer's contact details, how to complain, how long data will be retained, and the right to withdraw consent and/or data.

Given that this is a single qualitative interview, we will assume capacity/competence(34, 35) is not lost between consent and interview as these will usually occur concurrently on the same day.

Analysis

Audio recordings of the interviews will be listened to by two members of the research team. The completed C-POS measure will be read alongside this and interview note templates reviewed. Data from the cognitive interviews will be tabulated in Excel by participant and item using the coding system for classifying questionnaire problems described by Willis(42)

which looks at clarity of questions, knowledge required to answer questions, problems with assumptions/underlying logic, response categories, sensitivity of questions, instructions and formatting. This will be reviewed by the research team after four interviews have been conducted for each C-POS version (child and parent proxy versions). Consensus will be reached on whether changes to version 1 of the C-POS need to be made. If difficulties are found with any of the questions, recall periods or response formats the C-POS versions will be amended in line with these difficulties. If changes are made, the new version will then be piloted with another three participants from each cohort and analysed in the same manner. If difficulties are found, further changes will be made. Demographic data will be presented using descriptive statistics.

By the end of this stage of the study version 2 of the C-POS will be ready for further psychometric testing.

6 ETHICAL AND REGULATORY CONSIDERATIONS

6.1 Assessment and management of risk

children and young people and their families are considered a vulnerable population(43), with participation in research being viewed by some as an undue burden(44). As discussed earlier, children and young people with LLC/LTC are rarely included in research studies and it is important that they are given a chance to express their views. Not allowing children and young people to participate can also be seen as discriminatory and unjust(45) and in contravention to article 12 of the UN Convention of the Rights on the Child which states that any child capable of forming their own views should have the right to express these(46). There can be concerns regarding coercion from clinical teams to participate(47) but findings suggest that parents are able to say no if supported to do so(48). Avoiding using terms such as 'palliative care' and 'end of life care' can alleviate concerns from parents that their child is unaware of their prognosis(47). Researchers will work in partnership with children and young people and their parents throughout the research process, including recommendations such as obtaining meaningful assent from the children and young people, using age appropriate written information and ensuring children and young people and their family have free and informed choice regarding participation(49).

Co-applicant on the study, Professor Bobby Farsides (Professor of Clinical and Biomedical Ethics), has led the ethical dimensions of this study. All study staff are experienced in conducting qualitative interviews with this population. Professor Farsides has provided a 2 hour training session to summarise the recommendations and their translation to research practice of the report she led "Children and clinical research: ethical issues" http://nuffieldbioethics.org/wp-content/uploads/Children-and-clinical-research-full-report.pdf. Researchers have also completed training in communication skills, paediatric palliative care and the role and skills of play therapy in research studies.

We have appointed an external ethics reviewer, Dr Sara Fovargue of Lancaster University who has dual qualifications in ethics and medical law and was a member of the Nuffield Council committee on ethics and research with children http://www.lancaster.ac.uk/law/people/sara-fovargue. She authors and approves our annual ethical check to the European Research Council.

The research team will work to ensure that the research environment is conducive to promoting and protecting children's interests while also seeing them as participants rather than subjects. We recognise the potential burden of distress associated with discussion of symptoms and concerns (including physical and psychological concerns) and the potentially poor prognosis.

Please see our distress protocol for specific details, however in summary (drawing on the approved ethical approval for our co-applicant's study of children with high risk brain tumours and the Nuffield Guidance on research with children) the training and supervision received by the researchers will ensure that potential vulnerability is recognised in all interactions, dealt with in line with the protocol, and emergent issues dealt with immediately through the senior team(49).

Parents will be directed to our partner Together for Short Lives if they require additional support and guidance with respect to their child's condition and to access support for their child. The clinical team will be approached to offer additional support if the family requests it, and any issues of safety will be raised with the clinical team and the family will be informed.

Information and consent forms will clarify the content of the interview topics to ensure that full information is provided and there are no surprises regarding content. A minimum of 24 hours will be given following provision of the participant information sheet to decide upon participation.

No information regarding diagnosis or prognosis will be shared as a result of participation in the research interview.

We will offer participants the right to choose where interviews are conducted, to choose to express themselves through play or third person narratives, and to ensure that researchers are well trained in observing when to offer a pause or to terminate interviews.

We recognise that while protecting the child in terms of vulnerability, there is a body of evidence that demonstrates children's willingness to participate in research in the face of serious illness, seeing it as a positive, rewarding experience, and seeing benefits for future practice as cited above. However, we also recognise that some individuals may become distressed, hence our detailed plan to manage this.

There is risk of psychological burden as discussed above but we will: a) Give clarity in the information and consent process of the interview content; b) use of language will ensure that no information is disclosed that will cause additional grief (e.g. potential prognosis, future disease development); the distress protocol will ensure that researchers will recognise signs of distress and to acknowledge this, pause and to give the opportunity to restart after an appropriate time or to terminate the interview.

Any apparent distress will be responded to as above and all participants will have the opportunity to have their concerns raised with their clinical team, to be signposted to community support via Together for Short Lives links, and all interviews will be concluded with at least 10 minutes (according to need) of debriefing with recording apparatus switched off. As above, please note that the current literature suggests that this population value the opportunity to express their feelings.

This approach is applied to parents and the children and young people with serious illness. As stated in the training outline, we recognise that these groups have their own potential sources of distress.

To reduce the burden of participation by the child or their family members we will ensure that interviews are arranged at the preferred time and place for the interviewee.

There will be no direct benefit to participants, although as noted above children and young people recruited form this population who have participated in other studies have acknowledged their perceived positive experience and personal reward.

There is a risk of distress to interviewers. They will be trained in line with the programme detailed above, only experienced researchers will be recruited, we use a researcher "buddy" scheme" to ensure that all home visits are monitored, and we run a researchers' group for peer support. Study clinical colleagues will provide any additional one-to-one support self-identified by researchers. King's College lone and remote worker policy will be adhered to.

6.2 Research Ethics Committee (REC) and other Regulatory review & reports

Before the start of the study, a favourable opinion will be sought from a REC and the HRA for the study protocol, informed consent forms and other relevant documents.

Substantial amendments that require review by NHS REC will not be implemented until that review is in place and other mechanisms are in place to implement at site.

All correspondence with the REC will be retained.

It is the Chief Investigator's responsibility to produce the annual reports as required.

The Chief Investigator will notify the REC of the end of the study.

An annual progress report (APR) will be submitted to the REC within 30 days of the anniversary date on which the favourable opinion was given, and annually until the study is declared ended.

If the study is ended prematurely, the Chief Investigator will notify the REC, including the reasons for the premature termination.

Within one year after the end of the study, the Chief Investigator will submit a final report with the results, including any publications/abstracts, to the REC.

Regulatory Review & Compliance

Before any site can enrol patients into the study, the Chief Investigator (Professor Harding) will ensure that appropriate approvals from participating organisations are in place.

For any amendment to the study, the Chief Investigator or designee, in agreement with the sponsor will submit information to the appropriate body in order for them to issue approval for the amendment. The Chief Investigator or designee will work with sites (R&D departments at NHS sites as well as the study delivery team) so they can put the necessary

arrangements in place to implement the amendment to confirm their support for the study as amended.

Amendments

The process for making amendments will be led by the study CI (Harding) and taken to full Steering Group (or by email approval of the majority if not meeting is due) and with PPI consultation. It will be submitted to the approving REC for review and no change initiated until full REC approval for the change has been given and the R&D department at all partners sites have been informed and approved.

Any approved amendments will be incorporated into study materials with revised version number and the change noted and stored in the Site File.

6.3 Peer review

Peer review was conducted by the Funder (the European Research Council) and the CI presented the study at a grant panel in Brussels to a multiprofessional decision making panel of approximately 30 scientists from across Europe who individually and independently scored the propossal. The study is informing part of a King's College London PhD and is subject to King's College London approvals and review processes.

6.4 Patient & Public Involvement

To date our PPI involvement has been through the NGO "Together for Short Lives" and through PPI parent representatives on our group. They have commented on the aims, methods and dissemination plan of all phases of the C-POS study so far.

These groups are also represented on our Steering Group. They will be part of all decision making, interpretation and dissemination activities.

We are also working with the Young People's Advisory Group at Great Ormond Street Children's hospital who have looked at our study information sheets and the C-POS measure in order to provide feedback (see appendix 3).

6.5 Protocol compliance

Accidental protocol deviations can happen at any time. They must be adequately documented on the relevant forms and reported to the Chief Investigator and Sponsor immediately.

Deviations from the protocol which are found to frequently recur are not acceptable, will require immediate action and could potentially be classified as a serious breach.

6.6 Data protection and patient confidentiality

Each study participant will have their name/study ID code sheet, consent forms, and demographic sheet with study ID number, and transcript, stored separately in separate locked filing cabinets or in a secure folder on Sharepoint if access to the office is not possible due to COVID-19 restrictions. These will then be stored in a locked filing cabinet at the earliest opportunity and the electronic copies will be destroyed. Eventually, the transcript will be the only information stored on the study PC. All materials will be stored at the Cicely Saunders Institute, King's College London in the research area that is swipe-access only for research staff, with data storage in the dedicated researcher area. Data will be stored within the building for a period of 8 years within the secure archive. Future secondary analysis of the data will require a further application for ethical approval in on the presumption that it is seen as compatible processing. Secondary analysis will be subject to the participant's prior enactment of their right to withdraw consent and data. Transcription will be completed using two approaches: 1) by the research study team, on computers at the Cicely Saunders Institute or remotely on secure computers on the KCL managed environment; or 2) by the KCL preferred supplier for transcription – Clear Voice. Clear Voice utilise a secure site for researchers to upload password protected audio files. Passwords for audio files would be sent via the secure email service, Egress.com. At this stage all data will be pseudonymised, in that names, places, dates of birth (and any other potentially identifying information) will be removed from transcripts. Audio recordings will be transferred in encrypted files with no accompanying information.

After the recordings are transcribed, the pseudonymised qualitative data will be stored in Word documents in password protected files on the King's College London server which are only accessible by the research team. Participants' demographic data will be stored in Excel files in password protected folders on the server. No research study staff will have access to NHS patient files.

Participants will be able to have their data removed from the study dataset at any point, and for it to be removed from results up until these are presented/published and in the public domain. Minimal personal data will be retained in a study withdrawal log, with reasons for withdrawal and a file note to confirm that data has been removed and why.

The maintenance of Site Files, data management plans and adherence to these, and reporting will be overseen by the Faculty Research Development Manager. Data Protection Officer at KCL will conduct a review of the full protocol for the ERC reporting and will provide guidance and issue a letter of review. Any breaches will be reported immediately (same day as the breach becomes known) to the Data Protection Officer.

6.7 Indemnity

The lead sponsor, King's College London, will take primary responsibility for ensuring that the design of the study meets appropriate standards and that arrangements are in place to ensure appropriate conduct and reporting. King's College London also provides cover under it's No Fault Compensation Insurance, which provides for payment of damages or compensation in respect of any claim made by a research subject for bodily injury arising out of participation in a clinical trial or healthy volunteer study (with certain restrictions). NHS sites are covered for clinical negligence and conduct at site through the NHS Risk Pooling Scheme.

6.8 Access to the final study dataset

The study research team at King's College London and Guy's and St Thomas' will have access to the full dataset.

The CI will allow site investigators to access an anonymised dataset if a formal request describing their plans is approved by the steering group. All data transfers will be discussed with the KCL contracts team to assess whether data transfer agreements need to be in place before release.

7 DISSEMINIATION POLICY

The co-sponsors KCL and GSTT will own all arising data from this study.

On completion of the study, the data will be analysed and tabulated and a Final Study Report prepared.

The full study report will be provided to the Ethics Committee and the findings will available in peer review publications. The PI will lead publications with agreement for lead authorship by co-applicants. All data will be submitted for peer review within 6 months of data collection.

We will acknowledge our funder and clinical and community partners in the manuscript.

We will keep the stakeholder community informed via a newsletter that will be distributed via our project website and through Together for Short Lives. We will give the URL to all participants.

We will post the outputs of the research to any participant/family who requests it.

Following publication the data will be available for use upon reasonable request to the PI (subject to consent of participants).

7.2 Authorship eligibility guidelines and any intended use of professional writers

We will adhere to The International Committee of Medical Journal Editors defined authorship criteria for manuscripts submitted for publication. This can be found at ***http://www.icmje.org/recommendations/browse/roles-and-responsibilities/defining-the-role-of-authors-and-contributors.html***

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11 APPENDICIES

11.1 Appendix 1. Required documentation

Assent form - child 5-7 years

If you would like to take part in this project, please let your grown up know what you think about the questions

We would like to talk to you about whether you understand our questionnaire. Are you OK about this? (please tick)

☐ **Yes** ☐ **No**

You can stop taking part in the project at any time and you do not have to say why. Are you OK with this? (please tick)

☐ **Yes** ☐ **No**

Would you like to take part in the project? (please tick)

☐ **Yes** ☐ **No**

Your name	Date	Signature/verbal consent
Name of parent	Date	Signature
Researcher	Date	Signature

Assent form – child 8-12 years

If you would like to take part in this project, please let your grown up know what you think about the questions

We would like to talk to you about whether you understand our questionnaire. Are you OK about this? (please tick)

☐ **Yes** ☐ **No**

You can stop taking part in the project at any time and you do not have to say why. Are you OK with this? (please tick)

☐ **Yes** ☐ **No**

Would you like to take part in the project? (please tick)

☐ **Yes** ☐ **No**

Your name	Date	Signature/verbal consent
Name of parent	Date	Signature
Researcher	Date	Signature

Assent form child 11-15 years

If you would like to take part in this project, please let your grown up know what you think about the questions

 **We would like to talk to you about whether you understand our questionniare. Are you OK about this? (please tick)**

☐ **Yes** ☐ **No**

 **You can stop taking part in the project at any time and you do not have to say why. Are you OK with this? (please tick)**

☐ **Yes** ☐ **No**

 **Would you like to take part in the project? (please tick)**

☐ **Yes** ☐ **No**

Your name	Date	Signature/verbalconsent
Name of parent	Date	Signature
Researcher	Date	Signature

Consent form 16-18 years

Study number:

Name of Chief Investigator: Professor Richard Harding

Project title: C-POS-cognitive interview study

Please initial box:

1. I confirm that I have read the information sheet for young people 16-18 years, dated 19/04/2021 version 2 for the above study and have had the opportunity to ask questions

2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my care being affected

3. If I become distressed at any point I can stop at any time and support will be available to me.

4. I give permission for the interview to be audio recorded

5. I give permission for you to report what was discussed without using my name

6. I understand that relevant sections of my medical notes and data collected during the study, may be looked at by individuals from the Sponsor organisations King's College London and Guy's and St Thomas' NHS trust, from regulatory authorities or from the NHS Trust/private Trust, where it is relevant to us taking part in this research, for example checking the quality of the research. I give permission for these individuals to have access to my records.

☐ 7. I understand that the information collected about me and will be used to support other research in the future and may be shared anonymously with other researchers.

☐ 8. I agree to take part in the above study

_____	_____	_____
Name of child	Date	Signature
_____	_____	_____
Researcher	Date	Signature

*IF CHILD NOT ABLE TO GIVE INFORMED CONSENT:

_____	_____	_____
Name of parent	Date	Signature

Keep one copy for the patient, one for their medical notes and one for the site file

Consent form parent being interviewed

Guy's and St Thomas' NHS
NHS Foundation Trust

Study number:

Name of Chief Investigator: Professor Richard Harding

Project title: C-POS- cognitive interview study

Please initial box:

☐ 1. I confirm that I have read the information sheet for parents/carers being interviewed, dated 19/04/21 version 2 for the above study and have had the opportunity to ask questions

☐ 2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my or my child's care being affected

☐ 3. If I become distressed at any point I can stop at any time and support will be available to me.

☐ 4. I give permission for the interview to be audio recorded

☐ 5. I give permission for anonymised direct quotes to be used in the report, publications and presentations

☐ 6. I understand that relevant sections of my child's medical notes and data collected during the study, may be looked at by individuals from the Sponsor organisations King's College London and Guy's and St Thomas' NHS trust, from regulatory authorities or from the NHS Trust/private Trust, where it is relevant to us taking part in this research, for example checking the quality of the research. I give permission for these individuals to have access to my child's records.

☐ 7. I understand that the information collected about me and my child will be used to support other research in the future and may be shared anonymously with other researchers.

☐ 8. I agree to take part in the above study

_____	_____	_____
Name of parent	Date	Signature
_____	_____	_____
Researcher	Date	Signature

Keep one copy for the participant and one for the site file

Consent for parent signing for child

Study number:

Name of Chief Investigator: Professor Richard Harding

Project title: C-POS- cognitive interview study

Please initial box:

☐	1. I confirm that I have read the information sheet parent/carer signing for child dated 19/04/2021 version 2 for the above study and have had the opportunity to ask questions
☐	2. I understand that my child's participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my or my child's care being affected
☐	3. If I become distressed at any point I can stop at any time and support will be available to me.
☐	4. I give permission for the interview to be audio recorded
☐	5. I give permission for anonymised direct quotes to be used in the report, publications and presentations
☐	6. I understand that relevant sections of my child's medical notes and data collected during the study, may be looked at by individuals from the Sponsor organisations King's College London and Guy's and St Thomas' NHS trust, from regulatory authorities or from the NHS Trust/private Trust, where it is relevant to us taking part in this research, for example checking the quality of the research. I give permission for these individuals to have access to my child's records.

☐ 7. I understand that the information collected about me and my child will be used to support other research in the future and may be shared anonymously with other researchers.

☐ 8. I agree for my child to take part in the above study.

_____	_____	_____
Name of parent	Date	Signature
_____	_____	_____
Researcher	Date	Signature

Keep one copy for the participant and one for the site file

Information sheet – parent being interviewed

Information about the research for parents

C-POS- cognitive interview study

What is the aim of this research study?

We would like to invite you to be part of a research study to help us find out whether a questionnaire of symptoms and concerns we have developed for children with a serious illness and their parents/carers is easy to understand and fill in. We also want to make sure that there are no important items missing from the questionnaire.

What will happen if I take part?

One of our study research team will meet with you at a time and place that is convenient to you. If you were recruited from an NHS site and travel is permitted, we can interview you at the site if you wish. If you were recruited from a non-NHS site, then we will not be able to conduct interviews on their premises, but can choose another location convenient to you, such as your home. If you choose to travel, then we can pay your travel expenses. We can also use video meeting software such as Zoom, if travel is not possible.

Our researchers will ask you to fill in the questionnaire and explain what you understand by the questions and why you have chosen your answers as you do this. The questionnaire will ask you about physical symptoms your child may have experienced as well as questions about how they are feeling and how you have been managing.

How will information be collected?

We will complete a simple form with you that describes you and your child, then we will audio record the conversation between you and the researcher, as you complete the questionnaire. We will also ask you to complete a consent form before the interview starts,

to ensure that you understand what the study is about. You will be provided with a countersigned copy of this after the interview. The interview should take approximately 1 hour and is a one-off.

The information collected by the researcher (the audio recording of the interview and notes) will be kept locked in a secured location.

To analyse the interviews in detail, we need to transcribe them (type up the full text of the interview word by word). If you agree, we will use staff from outside the research team, hired specifically for transcription, who will complete this at the Cicely Saunders Institute or off-site on a secure computer. If you do not agree to this, you may still take part and the research team will transcribe the interview themselves.

What will happen to that information?

Either the team at the Cicely Saunders Institute, or their preferred supplier will type up the interview, removing any information that is identifiable such as dates, names, places and then when it is typed up, we will delete the audio recording within 12 months of the end of the study. If we send the files to our preferred supplier this will be sent securely as an encrypted file. When we publish or present the findings, it will be done in such a way as to minimise the potential of you being identifiable. Anything you tell us will be confidential, unless we are concerned for you or your child's safety in which case we will share that information with your clinical team, and we will tell you that we are doing so.

How to access results of this study?

If you would like to access the results of this study when it is complete please contact a member of the research team (contact details are at the end of this information sheet).

Do I have to take part?

You do not have to take part in this research study - it is entirely voluntary. If you choose not to take part it will not affect your current or future care. You can take your time in deciding whether to take part, and you may want to talk it over with your care team or others.

If during the interview the researcher becomes aware that you appear distressed, or exhibit behaviours that would indicate distress, either as a result of our questions, or for another reason that may not seem clear at the time, we will take steps to remedy that and may need to stop the interview, and we will communicate with your child's clinical team.

Can I change my mind after saying yes?

If you do decide to take part, you can change your mind at any time before or during the interview. If you change your mind after the interview, we will only keep the minimal personal information needed to confirm why and when you withdrew, for audit purposes. If we have already presented and published data this cannot be removed. If you would like your data withdrawn from the study, then please contact the research team. Our contact details can be found at the end of this information sheet.

Are there any direct benefits to taking part?

Taking part will not provide any direct benefits to you or your child, although we hope that there will be future improvements to care for others.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions (Professor Richard Harding, 02078485518 or richard.harding@kcl.ac.uk). If you remain unhappy and wish to complain formally, you can do this through the Guy's and St Thomas' Patients Advice and Liaison Service (PALS) on 020 7188 8801, pals@gstt.nhs.uk. The PALS team are based in the main entrance on the ground floor at St Thomas' Hospital and on the ground floor at Guy's Hospital in the Tower Wing.

In the event that something does go wrong and you are harmed during the research you may have grounds for legal action for compensation against Guy's and St Thomas' NHS Foundation Trust and/or King's College London but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you (if appropriate).

How will we use information about you?

We will need to use information from you for this research project. This information will include your name, contact details and demographic data. People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead. We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

Your data will be kept for 8 years within King's College London. Audio-recordings will be kept for 12 months after the end of the study. No identifiable data will be shared outside of the research team.

This study also forms part of a PhD project at King's College London.

What are your choices about how your information is used?

You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.

We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

You can find out more about how we use your information

- at www.hra.nhs.uk/information-about-patients/
- by asking one of the research team
- By contacting the data protection officer at King's College London or Guy's and St Thomas' NHS Foundation Trust:
    - For GSTT: Nick Murphy-O'Kane, Contact: DPO@gstt.nhs.uk.
    - For KCL: Albert Chan, Contact: info-compliance@kcl.ac.uk

Links to the Trust and college privacy notices can be found here:

GSTT: www.guysandstthomas.nhs.uk/research/patients/use-of-data.aspx

KCL: www.kcl.ac.uk/research/support/research-ethics/kings-college-london-statement-on-use-of-personal-data-in-research

Contact details of the research team

Professor Richard Harding (Chief Investigator) – Richard.harding@kcl.ac.uk

Lucy Coombes (PhD candidate) – lucy.coombes@kcl.ac.uk or 07867 785582

Dr Debbie Braybrook (Research Associate) – Debbie.braybrook@kcl.ac.uk

Thank you for reading this information and for considering our study.

Information sheet for parent whose child is being interviewed

Information about the research (parent/carer of a child under 16 who has assented)

C-POS- cognitive interview study?

What is the aim of this research study?

We would like to invite your child to be part of a research study to help us find out whether children and young people who are unwell understand a questionnaire we have developed. This questionnaire asks how your child has been feeling. The goal of this study is to help us develop a questionnaire for children and their families to ensure that when they see health professionals, they are asked about the things that matter and that we check that these things are being addressed.

What will happen if my child takes part?

One of our study research team will meet with you and your child at a time and place that is convenient to you. If your child was recruited from an NHS site and travel is permitted, we can interview them at the site if you wish. If your child was recruited from a non-NHS site, then we will not be able to conduct interviews on their premises, but can choose another location convenient to you, such as your home. If you choose to travel then we can pay your travel expenses. We are also able to use video meeting software. Our researchers will talk with your child and ask them to fill in the questionnaire. While they are doing this the researcher will ask them what they think each question means, and why they choose the answers they do. They will ask your child in a conversational style but your child can also use drawing and playing with toys to help them express themselves. As their parent/carer you can also be present to help them express themselves. The questionnaire will ask your child about physical symptoms they may have experienced as well as questions about how they are feeling.

How will information be collected?

We will complete a simple form with you that describes you and your child, then we will audio record the conversation between you, the researcher and your child. We will also ask you to complete a consent form before the interview starts, to ensure that you understand what the study is about. You will be provided with a countersigned copy of this after the interview. The interview should take approximately 1 hour, and is a one-off.

The information collected by the researcher (the audio recording of the interview and notes) will be kept locked in a secured location.

To analyse the interviews in detail, we need to transcribe them (type up the full text of the interview word by word). If you agree, we will use staff from outside the research team, hired specifically for transcription, who will complete this at the Cicely Saunders Institute or off-site on a secure computer. If you do not agree to this, you may still take part and the research team will transcribe the interview themselves.

What will happen to that information?

Either the team at the Cicely Saunders Institute, or their preferred supplier will type up the interview, removing any information that is identifiable such as dates, names, places and then when it is typed up, we will delete the audio recording within 12 months of the end of the study. If we send the files to our preferred supplier this will be sent securely as an encrypted file. When we publish or present the findings, it will be done in such a way as to minimise the potential of you being identifiable. Anything you or your child tells us will be confidential, unless we are concerned for you or your child's safety in which case we will share that information with your clinical team, and we will tell you that we are doing so.

How to access results of this study?

If you would like to access the results of this study when it is complete please contact a member of the research team (contact details are at the end of this information sheet).

Do I have to take part?

You do not have to take part in this research study - it is entirely voluntary. If you choose not to take part it will not affect your current or future care. You can take your time in deciding whether to take part, and you may want to talk it over with your care team or others.

If during the interview the researcher becomes aware that your child appears to indicate that they are distressed, or exhibit behaviours that would indicate distress, either as a result of our questions, or for another reason that may not seem clear at the time, we will take steps

to remedy that and may need to stop the interview, and we will communicate with your child's clinical team.

Can I change my mind after saying yes?

If you do decide to take part, you can change your mind at any time before or during the interview. If you change your mind after the interview, we will only keep the minimal personal information needed to confirm why and when you withdrew, for audit purposes. If we have already presented and published data this cannot be removed.

If your child becomes unwell during the interview or any other incident occurs and we have to stop recording, we would be led by you as to whether the interview can continue either when the incident has resolved, or at a later date. If we have collected usable data and it is felt that the interview should not continue we would seek permission from you before using this data in our study.

If you would like your child's data withdrawn from the study, then please contact the research team. Our contact details can be found at the end of this information sheet.

Are there any direct benefits to taking part?

Taking part will not provide any direct benefits to you or your child, although we hope that there will be future improvements to care for others.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions (Professor Richard Harding, 02078485518 or richard.harding@kcl.ac.uk). If you remain unhappy and wish to complain formally, you can do this through the Guy's and St Thomas' Patients Advice and Liaison Service (PALS) on 020 7188 8801, pals@gstt.nhs.uk. The PALS team are based in the main entrance on the ground floor at St Thomas' Hospital and on the ground floor at Guy's Hospital in the Tower Wing.

In the event that something does go wrong and you are harmed during the research you may have grounds for legal action for compensation against Guy's and St Thomas' NHS Foundation Trust and/or King's College London but you may have to pay your legal costs.

The normal National Health Service complaints mechanisms will still be available to you (if appropriate).

How will we use information about you?

We will need to use information from you for this research project. This information will include your name, contact details and demographic data. People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead. We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

Your data will be kept for 8 years within King's College London. Audio-recordings will be kept for 12 months after the end of the study. No identifiable data will be shared outside of the research team.

This study also forms part of a PhD project at King's College London.

What are your choices about how your information is used?

You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.

We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

You can find out more about how we use your information

- at www.hra.nhs.uk/information-about-patients/
- by asking one of the research team
- By contacting the data protection officer at King's College London or Guy's and St Thomas' NHS Foundation Trust:
    - For GSTT: Nick Murphy-O'Kane, Contact: DPO@gstt.nhs.uk.
    - For KCL: Albert Chan, Contact: info-compliance@kcl.ac.uk

Links to the Trust and college privacy notices can be found here:

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Contact details of the research team

Professor Richard Harding (Chief Investigator) – Richard.harding@kcl.ac.uk

Lucy Coombes (PhD candidate) – lucy.coombes@kcl.ac.uk or 07867 785582

Dr Debbie Braybrook (Research Associate) – Debbie.braybrook@kcl.ac.uk

Thank you for reading this information and for considering our study.

Information sheet child 5-7 years

Guy's and St Thomas' NHS
NHS Foundation Trust

KING'S College LONDON

The C-POS Study

Information Sheet for Children aged 5-7 years

We want to ask you to be part of some work we are doing.

We have written a questionnaire that asks children who have not been very well how they are feeling. We want to find out if you find the questions easy to answer.

We will ask the grown up you live with if it is ok for you to do this.

If your grown up agrees we will come and see you at a time you like, or we will chat to you on the computer. A grown up can be with you.

We will ask you to fill in the questionnaire with a grown up. While you do this, we will ask you what you think the questions mean and why you have chosen your answers.

You can also draw pictures and play if this helps you tell us what you think. We can talk for up to an hour.

If you get tired while we are talking you can ask to stop or take a break.

It is up to you if you want to take part. You can change your mind and no-one will be cross. If you have any questions, then you can ask your grown-ups at home to call us.

Information sheet child 8-10 years

A questionnaire for children and young people who have been unwell

A leaflet that can be read aloud by a parent or carer

Who are we?

Hello! We are researchers who want to find out whether you understand a questionnaire that we have made for children who are unwell. This leaflet will tell you all about our project. Please ask as many questions as you'd like.

What is this project all about?

We want to try and find out whether children understand our questionnaire about how they have been feeling when they are unwell. We also want to find out whether children like using our questionnaire, and that it includes everything that is important to them.

What do we want to do?

If you agree to talk to us, one of our research team will come to see you or video call you. They will ask you to fill in the questionnaire and ask you about it as you fill it in. Your grown-up (parent or carer) can be there too, if you want.

What would happen if I say yes to being part of this?

We will come and see you in a place that you think would be best to talk. Or can talk on a video call. We will ask you to fill in the questionnaire with us and tell us what you think each question means. We will also ask you to tell us why you choose the answers you do.

How would you do that?

We would ask what you think and record what you say. We can use paper and pens to draw to help you think. Or we can also use toys to help you think. You can ask your grown-ups to help you say what you want.

Do I have to do this?

It is up to you to choose whether you want to do this or not. You can tell your grown up what you want to do. They will also have to say if it is OK. If you get tired, we can stop. If you say no, then no-one will mind. It is up to you and nothing will be different if you say no.

Can I change my mind?

Yes, you can change your mind at any time and we can stop if you want to. We will also get rid of anything you have told us.

If when we meet you we become worried that you are very upset we may need to stop the interview, but we will always let your clinical team (doctor, nurses and team) know.

What will happen to the things I tell you?

We will not tell anyone that you took part in this. When we tell people what we have found, we will not tell them who it was that spoke to us.

Can I take some time to decide?

Yes, you can take your time, ask any grownups or friends what they think.

Information sheet child 11-15 years

Do children and young people who are unwell understand a questionnaire about how they are feeling?

This leaflet tells you about our research project. Please feel free to ask us any questions you have.

Who are we?

We are a team of researchers from King's College London.

Who can I contact if I have questions?

If you have any questions you can ask your parent/carers or the study researchers.

What is the project about?

This project is trying to find out whether children and young people who are unwell understand our questionnaire about how they have been feeling.

We want to find out whether you understand the questions, whether you like the way the questionnaire looks and whether we are asking the right questions.

By finding this out we can try and ensure that the questionnaire is able to be used regularly so that care teams can make sure that any problems are addressed.

What does taking part involve?

If you decide to take part, one of our team will arrange a time and place to meet you that is most convenient for you, and where you would feel most comfortable to talk.. We can also

do this using video call. We would ask you to fill in the questionnaire and tell us what you think each question means. We will also ask why you choose the answers you do. Your parent/carer can be with you if you would like. We will record the interview so that we can remember what you have said.

Who has checked this study?

This study has been checked to make sure that we work in a fair way. We have been told we can do this research by X ethics committee.

How do I choose whether to take part?

Please feel free to take your time deciding. You may want to talk to your care team, family or friends to help you decide. Choosing to say no will not affect anything for you.

Can I change my mind?

Yes. If you decide you want to take part then you can change your mind at any time. You can also choose for us to remove any information about you from the study. If you change your mind and decide that you do not want to take part, nothing will change in the care that you get.

If when we meet you we become worried that you are very upset we may need to stop the interview, but we will always let your clinical team (doctor, nurses and team) know.

What happens to the information I give you?

We will keep your name separate from any other information you give us. When we report what we find in this study (in reports and in presentations) we will be sure to remove any information that could be linked to you (like names, places, dates). We will not share anything that you say directly, but if we felt that you were in danger then we would let you know that we were planning to share that with a member of your care team.

Information sheet child 16-18 years

Do children and young people who are unwell understand a questionnaire about how they are feeling?

This leaflet tells you about our research project. Please feel free to ask us any questions you have?

Who are we?

We are a team of researchers from King's College London. Professor Richard Harding is leading this study.

Who can I contact if I have questions?

If you have any questions you can ask your parent/carers, the study researchers, our care team, or you can call Richard Harding on 0207 848 5518 or email him on richard.harding@kcl.ac.uk

What is the project about?

This project aims to find out whether children and young people who are unwell understand a questionnaire we have developed. The questionnaire will ask you about physical symptoms as well as questions about how you are feeling.

We want to find out whether you understand the questions and like the way the questionnaire looks and whether we are asking the right questions.

By finding this out we can try and ensure that the questionnaire is able to be used regularly so that care teams can make sure that any problems are addressed.

What does taking part involve?

If you decide to take part, one of our team will arrange a time and place to meet you, for a one-off interview, in a place that is most comfortable for you to talk and most convenient for you. If you were recruited from an NHS site and travel is permitted, we can interview you at the site if you wish. If you were recruited from a non-NHS site, then we will not be able to conduct interviews on their premises, but can choose another location convenient to you, such as your home. We can also do this using video call. We will ask you to complete a consent form before the interview starts, to ensure that you understand what the study is about. You will be provided with a countersigned copy of this after the interview. We would have a conversation with you while you fill in the questionnaire, asking you what each question means and why you chose the answers you did. Your parent/carer can be with you if you would like. We will audio record the interview. The interview should take about 1 hour.

Who has checked this study?

This study has been checked to make sure that we work in a fair way. We have been told we can do this research by Bloomsbury ethics committee.

How do I choose whether to take part?

Please feel free to take your time deciding. You may want to talk to your care team, family or friends to help you decide. If you chose not to, then nothing will change in your care – choosing to say no will not affect anything for you.

Can I change my mind?

If you do decide to take part, you can change your mind at any time before or during the interview. If you change your mind after the interview, we will only keep the minimal personal information needed to confirm why and when you withdrew, for audit purposes. If we have already presented and published data this cannot be removed.

If during our interview the researcher becomes aware that you appear to be distressed and upset, either as a result of our questions, or for another reason that may not seem clear at the time, we will take steps to remedy that and may need to stop the interview, but we will always communicate with your clinical team. If you would like your data withdrawn from the

study, then please contact the research team. Our contact details can be found at the end of this information sheet.

What happens to the information I give you?

Either the team at the Cicely Saunders Institute, or their preferred supplier will type up the interview, removing any information that is identifiable such as dates, names, places and then when it is typed up, we will delete the audio recording within 12 months of the end of the study. If we send the files to our preferred supplier this will be sent securely as an encrypted file. We will keep your name separate from any other information you give us. When we report what we found in this study (in reports, in presentations) we will be sure to remove any information that could be linked to you (like names, places, dates). We will not share anything that you say directly, but if we felt that you were in danger then we would let you know that we were planning to share that with a member of your care team.

How to access results of this study?

If you would like to access the results of this study when it is complete please contact a member of the research team (contact details are at the end of this information sheet).

How will we use information about you?

We will need to use information from you for this research project. This information will include your name, contact details and demographic data. People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead. We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study. Your data will be kept for 8 years within King's College London. Audio-recordings will be kept for 12 months after the end of the study. No identifiable data will be shared outside of the research team.

This study also forms part of a PhD project at King's College London.

What are your choices about how your information is used?

You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.

We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

You can find out more about how we use your information

- at www.hra.nhs.uk/information-about-patients/
- by asking one of the research team
- By contacting the data protection officer at King's College London or Guy's and St Thomas' NHS Foundation Trust:
    - For GSTT: Nick Murphy-O'Kane, Contact: DPO@gstt.nhs.uk.
    - For KCL: Albert Chan, Contact: info-compliance@kcl.ac.uk

Links to the Trust and college privacy notices can be found here:

GSTT: www.guysandstthomas.nhs.uk/research/patients/use-of-data.aspx

KCL: www.kcl.ac.uk/research/support/research-ethics/kings-college-london-statement-on-use-of-personal-data-in-research

You can find out more about how we use your information by reading King's College London's core privacy notice at https://www.kcl.ac.uk/terms/privacy.aspx, or by contacting Albert Chan (the Data Protection Officer for King's College London) on email at info-compliance@kcl.ac.uk or telephone at 0207 848 7816.

Contact details of the research team

Professor Richard Harding (Chief Investigator) – Richard.harding@kcl.ac.uk

Lucy Coombes (PhD candidate) – lucy.coombes@kcl.ac.uk or 07867 785582

Dr Debbie Braybrook (Research Associate) – Debbie.braybrook@kcl.ac.uk

11.2 Appendix 2. Feedback from GOSH YPAG group on information sheets

Member 1

I have managed to have a little bit of a look at the information sheets. I definitely think they work well for each age group. Its hard to remember being that small but from what I can see the sheets for younger children seem very clear and the language is basic enough for smaller children to understand. I also really like that the one for older children (e.g 16-18) has quite a lot of detail in it as I know that if I was part of this study I would like to know a lot about it so I could really understand why it was being done and why these questions were being asked. An improvement could be a little box next to each piece of information on the younger age groups so they could maybe draw a smiley face to show that they understand what each point is saying. I think it would make the experience more fun for them so they are more likely to engage and also helps to make sure they understand why they are taking part in the study and so they understand what it going to happen to them. Hope this all makes sense!

Member 2

C-POS 16-18 y/o sheet feedback

•Overall, sheets make sense and can understand.

•Might be worth placing somewhere whether they can take part if do a video call but without the cameras on? Also I think some reassurance that they will audio record the interview as I know I'm quite self-conscious when it comes to hearing back my own voice (not that they would, but reassuring who will hear this – e.g researchers only, will be stored safely etc)

•Some reassurance in terms of "danger" and letting the care team know, perhaps like " but if we felt like you were in danger, then we would let you know that we were planning to share that with a member of your care team so you can get the best support for your health"

•I skimmed over the compliance with legal requirements regarding the data, not too sure whether this can be presented in a more succinct manner?

Member 3

11-15 years

1. I believe that the language used would fit perfectly fine for children of this age, but may need assistance from their parents.

2. The questions are fine. I just believe they need to structure them in a better format.

8-10 years

1. I believe that the language used is ok, but the general feeling I feel would be quite confusing for an eight year old

Member 4

The C-pos Study Information Sheet - 5 to 7:

The sheet is very clear and simple,

the pictures are interesting and fun while explaining what the text is about, It was easy to understand and simple too follow. I

think its very suitable for 5-7 year olds.

The C-pos Study Information Sheet- 8 to 10:

The font is a bit small and the writing is very bunched together and could be difficult to read. The questions are answered very clearly and an 8-10 year old would be able to understand. The pictures also make it more interesting and fun.

The C-pos Study Information Sheet- 11 to 15:

There is too much technical detail for the younger part of the age group.
Perhaps a bigger font would make it less daunting to read.

Member 5

Comments on the information sheet for 16-18:

•"Do children and young people who are been unwell understand a questionnaire about how they are feeling?" - a little too wordy, I needed to reread it a couple times before I understood what it meant. Also, the typo makes me unsure whether it's talking about people who have been unwell, or are currently unwell

•On P6, "we work in a fair way" is a little too ambiguous - a more specific phrase, or a sentence of description could help

•There are two 'P6's

•The answer to the question "what happens to the information I give you" isn't actually answering the question. It might need to be split into two questions, e.g. "what is my information used for" (and then talk about the reports/presentations) and "how is my

information kept private/safe" (as this later question seems to be what the written answer is actually answering).

Member 6

Comments on the information sheet for 16-18:

I looked at the age 16-18 questionnaire and thought it was really good. I thought it was simple but also age appropriate and answered any questions I had.

Member 7

This was sent as the original document for 8-10 year olds. It has the reviewers name on the track changes so the original cannot be shared.

11.3 Appendix 3. Social media post

The main message will be as below, but exact wording may change dependent on response and demographic of previous respondents.

The C-POS team @CSI_KCL are looking for children and young people with life-limiting conditions and/or their parent/carers to take part in an interview to help test our new questionnaire. Please see details below if you are interested. #pedpc #patientandpublicinvolvement.

The C-POS team at King's college London are looking for children and young people with life-limiting/life-threatening conditions and/or their parent/carers to help us test our new questionnaire. Participation will take approximately 1 hour and will involve you answering the questions in the measure while telling us what you think they mean and how you chose your answers. The conversation will be audio recorded and can be conducted via video calling. The questions will ask about physical symptoms such as pain that you/your child might have been experiencing, as well as how they have been feeling. If you are interested in participation or would like more information, then please contact:
Lucy Coombes – lucy.coombes@kcl.ac.uk
Dr Debbie Braybrook – Debbie.braybrook@kcl.ac.uk

11.4 Appendix 4. Interview guide

Interview Guide for Cognitive Interview

Objective: To explore the cognitive processes used by respondents when reading, interpreting and responding to items on the C-POS.

Consent and demographics:

- Hi my name is …. Thank you for agreeing to talk to me today.
- Have you had a chance to read the information sheet about the study?
- As the sheet says, we have developed a questionnaire asking about outcomes for parents/carers of children and young people with a serious illness. The aim of this interview is to see if the questions are understood in the way we intended, and whether the way we have presented the answers is clear. It doesn't matter if they are not, because we have time to improve the questions. What you tell us will help us to improve the questionnaire.
- We are also keen to find out whether you think there are any important questions missing or any that aren't relevant or useful. There are no right or wrong answers to any of the questions.
- We will be recording the interview and anything that you tell us is confidential.
- If you want me to stop the interview or stop recording at any time, then just let me know. It's fine if you need to take a break or don't want to continue.
- Go through assent and/or consent forms and sign. (Ask consenting person to complete and return either photo/scanned copy or post paper copy). **Please record conversation as separate audio-file.**
- Ask participant demographic questions (*share screen and fill out separate sheet*).

Introduction to interview:

- So, I'm going to show you some questions (either on the screen/on paper) and I would like you to read & answer them one at a time
- We will stop and talk about each question before moving onto the next.
- As you answer I'd like you to try to 'think out loud' as you read and answer the question.
- Is it ok if we practice with a question first? I'll demonstrate and then you can have a go.
- [Demonstrate dummy question.] *Either for:*
  - *Children:* 'Can you tell me what you have in the room where you sleep?'
  - *Adults/Young people*: 'How many windows do you have in your home?'
    - How about you try with the same question?
- Remember to try to 'think out loud' as you read and answer the questions.
- [Ask prompts as needed for dummy question.]
- Thank them for responding and give feedback.
- During the interview I will also ask you some more specific things about each question.
- Apologies if the questions get repetitive
- In this study we are less interested in your answers to the questions, but how you arrive at the answers – what you think the question means, and the things you were thinking about when you chose your answer. There are no right or wrong answers.

- You can tell me any thoughts or views you might have about the questions as we go along.
- Ask permission to start recording

---------- START RECORDING ----------

General:

- What were you thinking about when you answered that question?
- (*If there was any hesitation, follow-up*) – You seemed to hesitate – can I ask what were you thinking about then?

Comprehension: **What does the respondent believe the question to be asking?**

- What does the question mean to you, in your own words?
- What does the word/term XXXXXX mean to you? (*Refer to corresponding sheet for flagged words, OR raise if certain words seem problematic*)
- How easy or difficult was it to understand this question?
- (*If problem*) How would you change this question?

Retrieval: **Could they recall the information required by the question? Was the time frame suitable?**

- How easy or difficult was it to remember your/your child's experience when answering this question?
- Was it easy or difficult to think about ["*the past week" / "yesterday or today"*] when answering this question?
- Would there be a different time period that would be easier to understand?

Judgement: **Is the respondent able to make an evaluation based on the information recalled?**

- How did you arrive at your answer to that question?
- Was that easy or hard to arrive at your answer? Why do you say that?
- How did you choose your answer?
- How sure are you of the answer to this question?

***(NOTE**: Consider using easier version for 8-12yrs and 13-18yrs if they struggle with a 5-point response format.)*

Response: **Is the respondent able to map their internally generated answer to a response option?**

- How did you choose your answer to this question?
- Was it hard or easy to select an answer from the options given?

- Did all options make sense for this question?
- What do you think the difference is between (chosen option – e.g. "*sometimes*") and (option next to it – e.g. "*often*")?
- If there are different response formats given – ask which format they preferred and why? Which was easiest to use?

Other:

- Is there anything else you would like to say about this question? / The questionnaire as a whole?
- Did you find any of the questions upsetting? / embarrassing? / inappropriate?
- Are there any questions that you would leave out of this questionnaire?
- Are there any questions that you would add to this questionnaire?

---------- THANKS + STOP RECORDING ----------

Appendix M Example of cognitive interview data extraction sheet

Q4 - Have you been able to talk to people about how you feel yesterday or today?										
Answer choices - Never, Sometimes, Most of the time										
Participant ID	**Response**	**Observations**			**Comprehension**	**Retrieval**	**Judgement**	**Response**	**Other**	**Notes**
Insert ID number	*Select answer from drop down*	*Did the participant hesitate or ask for the question to be repeated?*	*Did the participant change their answer at any point?*	*Did the participant appear comfortable answering the question?*	*Meaning of question, meaning of specific words, ease or difficulty understanding question, suggested changes to question*	*Ease of difficulty remembering experience, ease or difficulty with recall period, suggestions for different recall period*	*Thoughts when answering question, how was the answer arrived at, was this easy or hard, how was response chosen, how sure is participant of their answer.*	*How was response chosen, was selecting from the options easy or hard, did all response options make sense.*	*Any other comments, was the question upsetting, embarrasing or inappropriate.*	

Appendix N Achieving consensus on priority items for paediatric palliative care outcome measurement – in press

AUTHORS

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TITLE

Achieving consensus on priority items for paediatric palliative care outcome measurement: results from a modified Delphi survey, engagement with a children's research involvement group and expert item generation

AFFILIATIONS

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Abstract

Background

There is no validated outcome measure for use in children's palliative care outside sub-Saharan Africa. Stakeholders must be involved in the development of such measures to ensure face and content validity.

Aim

To gain expert stakeholder consensus on items for inclusion in a paediatric palliative care outcome measure to establish face and content validity.

Design

This study was conducted in three phases following Rothrock and COSMIN guidance on patient-reported outcome measure development. Phase 1: Three-round modified Delphi survey to establish consensus on priority items. Phase 2: A young person's advisory group was consulted on priority outcomes. Phase 3: Item generation meeting with key stakeholders to develop initial measure versions.

Setting and participants

Delphi survey: Parents and professionals with experience of caring for a child with a life-limiting condition. Young persons's advisory group: young people age 10-20 years. Item generation meeting: bereaved parents, academics and clinicans.

Results

Phase 1: Delphi survey (n=82). Ranking agreement increased from Kendall's W=0.17 to W=0.61, indicating movement towards consensus. Agreement between professional and parent ranking was poor (Cohen's kappa 0.13). Professionals were more likely to prioritise physical symptoms, whereas parents prioritised psychosocial and practical concerns. Phase 2: Children (n=22) prioritised items related to living a 'normal life' in addition to items prioritised by adult participants. Phase 3: Five age/developmental stage appropriate child and proxy-reported versions of C-POS, containing 13 items, were drafted using the results from Phases 1 and 2 (n=22).

Conclusions

This study highlights the importance and feasibility of involving key stakeholders in PROM item generation, as important differences were found in the priority outcomes identified by children, parents and professionals.

Key words - Outcome assessment; Delphi survey; Public participation; Palliative Care; Children

What is already known about this topic?

- Children and young people with life-limiting and life-threatening conditions experience many inter-related symptoms, concerns and care priorities that require a holistic approach to care.
- There is currently no validated patient-centred outcome measure (PCOM) for use in paediatric palliative care outside of sub-Saharan Africa.
- Development of such a measure has repeatedly been highlighted as a clinical and research priority.

What this paper adds

- This study describes the item generation phase of the development of a novel PCOM with demonstrated face and content validity for use in paediatric palliative care (C-POS).
- Involvement of key stakeholders in item generation has demonstrated important differences in the priority healthcare outcomes identified by children, parents and healthcare professionals in paediatric palliative care.
- Five versions of C-POS have been developed that reflect variation in age/developmental stages of the target population and allow for proxy reporting if required.

Implications for practice, theory or policy

- A PCOM that considers psychosocial domains will support professionals to assess needs more holistically.
- Further research is required to test C-POS cognitively and psychometrically prior to implementation.

Background

It is estimated that each year 21 million children and young people worldwide (hereafter 'children') with life-limiting or life-threatening ('life-limiting') conditions require input from palliative care services[1]. Life-limiting conditions are those for which there is no hope of cure, and from which children will die. Life-threatening conditions are those for which curative treatment may be feasible, but may fail[2]. With advances in medical care, increasing numbers of children are living longer with life-limiting conditions[3, 4]. Provision of children's palliative care varies geographically, and increased prevalence of life-limiting conditions has not been met with an equivalent increase in healthcare resource allocation[3, 5]. Children with life-limiting conditions experience a multitude of inter-related symptoms, concerns and care priorities that impact on all aspects of daily life[6]. This requires a holistic, child-centred approach to care.

A patient-reported outcome measure (PROM) is defined as a measure of a patient's health status, elicited directly from the patient. Many palliative care patients, including children with life-limiting conditions, are too unwell or cognitively unable to self-report on their own health outcomes[7]. A measure which allows for proxy completion is required. Together PROMs and proxy-reported measures are termed patient-centred outcome measures (PCOMs)[7, 8]. The use of PCOMs in adult palliative care has been shown to improve service quality and promote patient-centred care[9], as well as lead to better symptom recognition, more discussion of quality of life and increased palliative care referrals[7]. PCOMs have been advocated for improving awareness of unmet need, understanding different models of care delivery and allowing national and international comparison[10, 11].

Evidence of the use of PCOMs in paediatric palliative care is lacking due to absence of a validated measure[12]. Development of a PCOM for use in this population has been repeatedly highlighted as a priority[13-17]. A psychometrically validated measure exists in sub-saharan Africa (recently adapted in Belgium) where the sample informing content validity predominantly had a HIV diagnosis[18, 19] This measure was developed before current PCOM development guidance had been established [20, 21] . The Belgian version has undergone initial face and content validation but further psychometric data is not available[22].

This study is part of a programme of work to develop the Children's Palliative Outcome Scale (C-POS), a child-centred outcome measure for use in paediatric palliative care. This measure is being developed within the UK healthcare context, with parallel processes to develop C-POS in other regions. Previous sequential outputs are two systematic reviews (establishing the need for a new PCOM[12], identifying response formats and administration modes used in PCOMs for children[23, 24]) and primary qualitative data identifying symptoms, concerns and care priorities (the sample included children and young people, health and social care professionals, siblings, parents, and commissioners) [25, 26]. This previous work has demonstrated that several versions of C-POS will be required to reflect the age/developmental stages of children with life-limiting conditions. The aims of the study presented here were to: gain expert stakeholder consensus on items to be included in C-POS; further enhance face and content validity and finalise initial versions of C-POS for cognitive testing.

Methods

C-POS is being developed following the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) and Rothrock guidance on PROM development[21, 27, 28]. This paper reports on a Delphi survey, engagement with a young person's advisory group, and an item generation meeting. A flow chart of the study is shown in Figure 1.

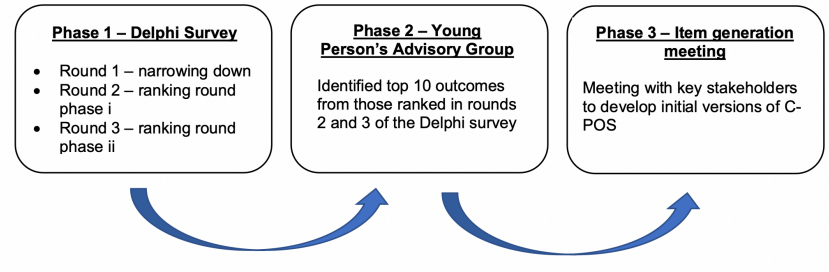

Figure 1 Study flow diagram

Phase 1 – Delphi Survey

Study design

A modified Delphi ranking survey was conducted and reported in accordance with CREDES, between November 2020 and February 2021[29]. A typical ranking Delphi survey has three phases: a) 'brainstorming' – experts list items important for the area of interest, b) 'narrowing down' – items identified in step one are narrowed down and c) 'ranking' – experts rank the remaining items over multiple rounds, with the aim of reaching consensus[30, 31]. Our previous work identifying symptoms, concerns and care priorities for children with life-limiting conditions and their families served as the 'brain-storming' phase[25, 32]. This paper reports on the 'narrowing down' and 'ranking' phases conducted using SmartSurvey™.

Study procedure

COSMIN guidance on PROM development states that experts (including patients) should be included in measure development to ensure face and content validity [20]. We included parents/carers ('parents') of children with life-limiting conditions as experts, and health and social care professionals ('professionals') to enhance validity and ensure clinical relevance.

Eligibility criteria Professionals with >6 months experience of caring for children with life-limiting conditions; parents of children 0>18 years with a life-limiting condition; bereaved parents whose child (0>18 years) had died of a life-limiting condition 12-24 months prior to consenting to participate.

Recruitment Professionals were recruited via the Association of Paediatric Palliative Medicine (UK doctors, nurses and allied health professionals), social media (UK paediatric palliative care charities, and researcher and institute Twitter pages) and the sites that participated in the previous qualitative study for this project [32]. Parents were recruited via a UK a children's palliative care charity, parents' groups and social media.

Data collection

Round 1-'Narrowing down'. The 42 outcomes identified from our previous work were presented in random order to each participant[6]. Participants were asked to select the

20 items most important for inclusion in C-POS, and to suggest any items they thought were missing. A free text box allowed participants to explain their choices.

Rounds 2-3-'Ranking'. Participants from the previous rounds were presented with the results in plain English terms. Participants were asked to rank the outcomes retained from round 1 in order of priority for inclusion in C-POS from most to least important. Items were presented in random order for the first ranking round and according to mean rank in subsequent rounds[31]. A free text box allowed participants to explain their rankings. Weekly reminder emails were sent to those who had not responded. Each round was open for 2-3 weeks.

Data analysis

Round 1-'Narrowing down'. Items selected by >50% of participants were moved to the ranking rounds[31]. Data were analysed as a whole group, and separately for professionals and parents. New suggested items were compared with existing items and discussed by the research team and study steering group to gain expert consensus on whether they should be included in round two[33, 34]. The study steering group comprises parents whose child had died of a life-limiting condition, academics with expertise in PROM development, and professionals who care for children with life-limiting conditions. The steering group is responsible for reviewing the progress, quality and delivery of the C-POS study.

Rounds 2-3-'Ranking'. Kendall's W coefficient of concordance and top half rank (percentage of participants who ranked items in their top 50%). Kendall's W was interpreted as follows: weak<0.5, moderate 0.5-0.7, strong>0.7[30]. Cohen's kappa was used to determine agreement between parent and professional rankings.

Stopping criteria. Data were analysed as per the previous round. If consensus was reached (Kendall's W>0.7) then no further rounds would be undertaken.

Data analysis was conducted using Stata (v16, StataCorp LLC, College Station, TX).

Ethics and consent

Ethical approval was obtained from King's College London (MRSP-19/20-18826). Participants received written study information and completed a consent form at the beginning of each round.

194 Phase 2 – Young People's Advisory Group

195 The research team worked with a young person's advisory group at a UK tertiary
196 children's hospital. The group comprised children and young people aged 10-21
197 years with a life-limiting condition, siblings of children with life-limiting conditions or
198 those interested in a career in healthcare or research. During a virtual advisory group
199 meeting in March 2021 the group were given a short, age-appropriate presentation
200 on the C-POS study aims and some simple definitions of outcome measures and
201 life-limiting conditions. The group was then divided in two by age. Older
202 representatives were asked to work independently to review outcomes from those
203 ranked during rounds two and three of the Delphi and choose their top 10 (Table 3).
204 Younger representatives were asked to choose their top ten outcomes from this list
205 as a group. Both groups were also asked to suggest names for the C-POS versions
206 (as age bands to label measures is not appropriate in this population given common
207 developmental delay). The groups facilitators led the session with support from a
208 member of the research team. The intention was that working with the advisory
209 group would strengthen and broaden the perspectives of children in the study and
210 ensure children's views continued to be considered in measure design.

211 Representatives were providing patient and public involvement and thus ethical
212 approval was not requried[35]. Involvement is reported in line with GRIPP2 (short-
213 form) guidance[36].

214 Phase 3 – Item generation meeting

215 This consisted of a half-day virtual meeting with the C-POS steering group. The
216 agenda was informed by previous PROM item generation meetings[37]. The meeting
217 began with a presentation from the research team including: an overview of the
218 study and the results from previous development work[23, 25, 32], the Delphi survey, and
219 findings on aspects of measure design (recall period, response format,
220 administration mode) from our qualitative interviews. Discussion was led by the
221 research team, starting with the construct to be measured and the corresponding
222 overarching themes found in our interview study (physical symptoms,
223 spiritual/existential, social/practical and emotional/psychological), followed by
224 suggestions on potential wording of questions. Also discussed were priority items for

inclusion and aspects of measure design. After the item generation meeting, versions of C-POS were drafted for future cognitive and psychometric testing.

Results

Phase 1 Delphi survey

Round 1 – narrowing down

Eighty-two individuals participated (59 healthcare professionals, 23 parents/carers (one bereaved)). See Table 1.

Table 1 Participant demographics: Delphi round 1 – 'Narrowing down'

Health and social care professionals (n=59)		**Parent/carers (n=23)**	
Gender (male:female)	8:50 (1 preferred not to answer)	**Gender (male:female)**	0:23
Profession	4 Counsellor/therapist 16 Doctor 4 Health care assistant 32 Nurse 1 Physiotherapist 2 Social work	**Child's diagnosis**	1 Cancer 3 Circulatory 5 Congenital 2 Genitourinary 4 Metabolic 8 Neurological
Place of work	5 Community 30 Hospice 17 Hospital 7 Multiple settings	**Child's age in years (mean; range)**	8.9 (1-17)
Experience in years (mean; range)	11.8; (1-30)	**Ethnic background**	4 mixed ethnic group 23 white British (parent/carer) 19 white British (child)

Twenty-one outcomes were selected by >50% of participants. Two additional outcomes were selected by >50% of the professional group, and three by the parent/carer group (Table 2). Twenty-three suggestions were made for additional outcomes. Most suggestions were thought to be incorporated in existing outcomes, except for one regarding siblings (suggested by 22% of parent participants).

242 Table 2 Results Delphi round 1 – 'Narrowing down'

Outcome			
	Overall (n=82)	***Parent/carer (n=23)***	***HSCPs (n=59)***
Pain*	73 (89.0)	18 (78.3)	55 (93.2)
Having sufficient support from health and social care professionals*	70 (85.4)	19 (82.6)	51 (86.4)
Reducing the impact of illness on family life/burden of care*	68 (82.9)	22 (95.7)	46 (78.0)
Child being able to do things they enjoy*	68 (82.9)	22 (95.7)	46 (78.0)
Ability to live life to the fullest*	67 (81.7)	22 (95.7)	45 (76.3)
Breathing and respiratory difficulties*	63 (76.8)	14 (60.9)	49 (83.1)
Tiredness or fatigue*	62 (75.6)	19 (82.6)	43 (72.9)
Emotional impact of illness*	59 (72.0)	20 (87.0)	39 (66.1)
Being able to maintain relationships with peers*	59 (72.0)	19 (82.6)	40 (67.8)
Being supported/enabled to express emotions and feelings*	57 (69.5)	17 (73.9)	40 (67.8)
Having a plan for future care*	55 (67.1)	19 (82.6)	36 (61.0)
Being able to take part in memory making opportunities*	54 (65.9)	19 (82.6)	35 (59.3)
Having as much information as needed*	54 (65.9)	17 (73.9)	37 (62.7)
Sleeping difficulties*	53 (64.6)	12 (52.2)	41 (69.5)
Nausea and/or vomiting*	52 (63.4)	10 (43.5)	42 (71.2)
Having psychological needs met*	49 (59.8)	16 (69.6)	33 (55.9)
Having social support needs addressed*	48 (58.5)	18 (78.3)	30 (50.9)
Being able to access and undertake education*	48 (58.5)	11 (47.8)	37 (62.7)
Seizures*	45 (54.9)	10 (43.5)	35 (59.3)
Dystonia/muscle spasm*	43 (52.4)	8 (34.8)	35 (59.3)
Changes to physical function*	42 (51.2)	8 (34.8)	34 (57.6)
Setting and achieving life goals*	40 (48.8)	13 (56.5)	27 (45.8)
Financial burden of care*	38 (46.3)	19 (82.6)	19 (32.2)
Agitation*	37 (45.1)	4 (17.4)	33 (55.9)
Bowel problems*	37 (45.1)	6 (26.1)	31 (52.5)
Changes to appetite and/or eating	33 (40.2)	7 (30.4)	26 (44.1)
Changes in physical appearance	27 (32.9)	3 (13.0)	24 (40.7)
Having spiritual needs met	26 (31.7)	2 (8.7)	24 (40.7)
Changes in behaviour	25 (30.5)	9 (39.1)	16 (27.1)
Infections and/or impaired immunity*	25 (30.5)	12 (52.2)	13 (27.1)
Impact of illness on cognition	24 (29.3)	9 (39.1)	15 (25.4)
Having cultural needs addressed	21 (25.6)	0	21 (35.6)
Having religious and faith needs met	16 (19.5)	0	16 (27.1)
Cough	16 (19.5)	3 (13.0)	13 (22.0)
Changes in consciousness	15 (18.3)	3 (13.0)	12 (20.3)
Changes to self-outlook	14 (17.1)	5 (21.7)	9 (15.3)
Skin concerns	13 (15.9)	4 (17.4)	9 (15.3)
Weight changes	10 (12.2)	5 (21.7)	5 (8.5)
Opportunity to explore the meaning of life	9 (11.0)	4 (17.4)	5 (8.5)
Being able to leave a legacy	6 (7.3)	4 (17.4)	2 (3.4)
Low blood counts	5 (6.1)	4 (17.4)	1 (1.7)
Fertility concerns	4 (4.9)	1 (0.2)	3 (5.1)

* = items moved to ranking rounds (n=27)

Round 2–'Ranking' round phase i

Sixty individuals (47 professionals; 13 parents) participated in ranking the 27 retained items. See supplementary table 2 for demographics. There was weak overall agreement on ranking (W=0.12). There was also weak agreement between parents' rankings alone (W=0.16) and professionals alone (W=0.21). Cohen's kappa between parents and professionals was 0.08 (Table 3).

Table 3 Delphi results round 2 – ranking phase i

Outcome (n=27)	**Overall median rank (% ranking in top 50%) (n=60)**	**Parent median rank (% ranking in top 50%) (n=13)**	**HSCP median ranking (% ranking in top 50%) (n=47)**
Pain	5.5 (88.3)	7 (84.6)	1 (89.4)
Ability to live life to the fullest	6.5 (66.7)	5 (76.9)	5 (63.8)
Breathing and respiratory difficulties	7 (80.0)	12 (69.2)	2 (83.0)
Child/young person being able to do things they enjoy	8 (73.3)	6 (69.2)	3 (74.5)
Having sufficient support from health and social care professionals	9 (68.3)	9 (76.9)	6 (66.0)
Having a plan for future care	9.5 (68.3)	14 (61.5)	4 (70.2)
Dystonia/muscle spasms	11.5(60.0)	18 (38.5)	9 (66.0)
Being supported/enabled to express emotions and feelings	12 (58.3)	11 (53.8)	10 (59.8)
Sleeping difficulties	12.5 (58.3)	12 (76.9)	12 (53.2)
Setting and achieving life goals	12.5(50.0)	13 (53.8)	19 (48.9)
Having psychological needs met	12.5 (53.3)	9 (61.5)	16 (51.1)
Nausea and vomiting	13 (58.3)	19 (23.1)	7 (68.1)
Tiredness or fatigue	13.5 (56.7)	14 (61.5)	11 (55.3)
Reducing the impact of illness on family life/care burden	13.5 (53.3)	14 (53.8)	15 (53.2)
Emotional impact of illness	14 (55.0)	11 (53.8)	14 (55.5)
Seizures	14 (56.7)	14 (46.1)	8 (59.6)
Agitation	15.5 (51.2)	20 (15.4)	13 (61.7)
Siblings being supported and having their needs met	16(38.3)	14 (61.5)	21 (31.9)
Changes to physical function	16.5 (41.2)	14 (53.8)	20 (38.3)

Bowel problems	17 (43.3)	19 (23.1)	18 (48.9)
Having as much information as needed	17 (48.3)	17 (46.2)	17 (48.9)
Being able to maintain relationships with peers	18 (36.7)	15 (46.2)	23 (34.0)
Being able to take part in memory making opportunities	19.5 (33.3)	20 (30.8)	22 (34.0)
Financial burden of care	20 (25.0)	15 (46.2)	25 (19.1)
Infections and/or impaired immunity	20 (26.7)	19 (38.5)	24 (23.4)
Having social support needs addressed	20.5 (23.3)	17 (38.5)	26 (19.1)
Being able to access and undertake education	22.5 (26.7)	22 (38.5)	27 (59.6)
Kendall's W	**0.1671**	**0.1595**	**0.2053**

253

254 Round 3–'Ranking' round phase ii

255 Thirty individuals participated in round 3 (26 professionals; 4 parents) and the 27
256 items ranked in the previous round were ranked again. See supplementary table 3
257 for demographics. Overall, agreement between participants was moderate (W=0.61).
258 There was also moderate agreement between the professional group alone
259 (W=0.68) and parent group alone (W=0.64). Cohen's kappa between parent and
260 professionals =0.13 (poor agreement) (Table 4).

261 As Kendall's W had increased from weak to moderate agreement the decision was
262 taken to stop the study at this point due to concerns regarding potential gain and
263 feasibility of conducting another round.

264 Phase 2- Younger Person's Advisory Group Meeting

265 Twenty-two children (17 female; 6 male) aged 10-21 years attended the meeting.
266 The responses given by two groups are shown in Table 4. Both groups suggested
267 naming the C-POS versions after planets to avoid any stigma using chronological
268 age. Measure selection will be dependent on developmental stage.

269 Table 4 Delphi results round 3 – 'Ranking' round phase ii

Outcome	Overall median rank (% ranking in top 50%) (n=30)	Parent median rank (% ranking in top 50%) (n=4)	HSCP median rank (% ranking in top 50%) (n=26)	Times identified in top 5 by older patient and public involvement (11 representatives)	Item identified by younger patient and public involvement in overall top 13 (11 representatives)
Pain	1 (90.0)	9.5 (50.0)	1 (96.2)	7	Yes
Ability to live life to the fullest	2 (96.7)	1.5 (100)	2.5 (96.2)	3	Yes
Breathing and respiratory difficulties	3 (96.7)	6.5 (100)	3 (96.2)	2	Yes
Child/young person being able to do things they enjoy	4 (96.7)	4 (100)	4 (96.2)	5	Yes
Having sufficient support from HSCPs	5 (93.3)	5.5 (75)	5 (92.3)	3	No
Having a plan for future care	6 (90.0)	9.5 (25)	6 (92.3)	1	No
Dystonia/muscle spasms	8 (76.7)	20 (25)	7 (84.6)	0	No
Being supported/enabled to express emotions & feelings	9 (80.0)	8 (100)	9.5 (76.9)	2	No
Sleeping difficulties	10.5 (86.7)	10.5 (75)	10.5 (88.5)	3	No
Having psychological needs met	10.5 (76.7)	9.5 (100)	11 (73.1)	5	No
Nausea and vomiting	12 (76.7)	17 (50)	11.5 (80.8)	1	Yes
Setting and achieving life goals	12 (73.3)	8.5 (100)	12 (69.2)	1	No
Tiredness or fatigue	13 (80.0)	5.5 (100)	13 (76.9)	3	No
Reducing the impact of illness on family life/care burden	14.5 (50.0)	13.5 (75)	15.5 (46.2)	2	Yes
Agitation	16 (36.7)	20 (0)	16 (42.3)	0	No
Seizures	16 (36.7)	16.5 (0)	16 (42.3)	0	Yes
Emotional impact of illness	16 (23.3)	10.5 (75)	17 (15.4)	3	Yes
Siblings being supported and having their needs met	18 (16.7)	22 (0)	18 (19.2)	0	No

13

Changes to physical function	19 (16.7)	10.5 (75)	19 (7.7)	2	Yes
Having as much information as needed	20 (13.3)	20.5 (0)	20 (15.4)	0	No
Bowel problems	20.5 (23.3)	20.5 (0)	20.5 (26.9)	0	No
Being able to maintain relationships with peers	22 (13.3)	22 (0)	22 (15.4)	5	Yes
Being able to take part in memory making opportunities	23 (13.3)	16 (25)	23 (11.5)	0	Yes
Infections and/or impaired immunity	24 (6.7)	20.5 (25)	24 (3.8)	1	No
Financial burden of care	25 (20.0)	24.5 (25)	25 (19.2)	0	No
Having social support needs addressed	26 (10.0)	21.5 (25)	26 (7.7)	0	No
Being able to access and undertake education	27 (6.7)	23.5 (0)	27 (7.7)	1	Yes
Kendall's W	**W=0.61**	**W=0.68**	**W = 0.64**	**-**	**-**

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14

Phase 3 – Item generation meeting

Twenty-two members attended the item generation meeting – nine paediatric palliative care clinicians, six research team members, five clinical academics with expertise in PCOM development and two bereaved parents. After the initial presentations, each domain from our qualitative interview study was discussed and potential C-POS items were mapped onto these[26, 38]. Previous work had suggested children's care priorities differed from parents, particularly regarding practical aspects of care. It was agreed that C-POS would have self-report items regarding children's symptoms and concerns, and separate questions for parents to answer regarding family concerns[32]. It was further agreed that there would be proxy versions of the measure for parents to answer on behalf of their child if they were unable to respond themselves. Proxy versions would contain the same items as the self-report versions.

Five versions of the measure were drafted, each with eight questions about the child and five about the family: (1) parent/carer of child<2 years, (2) parent/carer of child≥2 years, (3) child 5-7, (4) 8-12 and (5) 13-18 years (or cognitive equivalent). The number of items was informed by previous work which suggested that children should have 10 items or fewer to respond to[24]. These versions were named after planets, as suggested by the young person's advisory group. Items were the same across versions but were worded differently in consideration of age/developmental stage. For example, using the term 'hurt' rather than 'pain'. Recall period and response format were based on previous evidence, with shorter recall and a 3-point Likert scale for younger/less cognitively able children, and a longer recall and 5-point Likert scale for older/more cognitively able children[23, 24]. The Likert scales on the child versions were anchored with emojis. Table 5 shows domains and agreed items for C-POS.

Due to the number and heterogeneity of life-limiting conditions[39], ensuring suitability of all items for the entire population proved challenging. Several physical symptoms (e.g., dystonia and breathing difficulties) were prioritised in the Delphi survey, but not all children with life-limiting conditions experience these. Only pain was common across the population. Hence a decision was taken to have a generic question regarding symptoms other than pain. The item regarding siblings was not relevant to

all families, so a question regarding the impact of the child's condition on the family was worded to incorporate relevant family members.

Table 5 Mapping of C-POS items onto domains from previous qualitative interview study and systematic review[6, 38]

Child symptom and concern items (self-reported or proxy-reported)	
Domain	**Question item**
Physical	Pain Other symptoms
Social and practical	Being able to ask questions Being able to undertake usual activities
Emotional and psychological	Worry Sharing feelings Being able to do things you enjoy
Spiritual/existential	Being able to do things you enjoy Living life to the fullest
Parent/carer items	
Physical	Getting enough sleep
Social/practical	Access to information about child's condition Support needed to care for child Support to plan for future care
Emotional/psychological	Impact of child's condition on family
Spiritual/existential	Support to plan future care

Discussion

This paper reports on the development of the first parent-proxy and age/developmental stage appropriate child versions of an outcome measure for children with life-limiting conditions and their families outside of sub-Saharan Africa[21, 28]. The Delphi survey, young person's advisory group, and item generation meeting have together established face and content validity of the proposed C-POS. This research ensures that the proposed items to undergo further psychometric testing reflect the construct we intend to measure, i.e., priority multidimensional palliative

care outcomes for children with a range of life-limiting conditions, their families and the professionals caring for them. Importantly, C-POS items capture all domains covered in the World Health Organisation's definition of paediatric palliative care[40].

Parent and professional Delphi rankings contained many similarities, but there were some differences, resulting in low inter-relater reliability between the two groups. Professionals were more likely to prioritise physical symptoms such as pain, respiratory difficulties and dystonia. Parents were more likely to prioritise psychosocial concerns such as memory making and the emotional impact of a life-limiting condition. Parents were also more likely to prioritise their child's physical function, possibly because these impact family care burden as well as participation in activities outside the home, some of which are important to siblings. While many elements of palliative care are important to both professionals and parents[41], some studies indicate that professionals put greater emphasis on physical well-being[42]. The final C-POS versions address these differences by incorporating items that were highlighted as a priority by either and both stakeholder groups.

Consultation with members of the young person's advisory group identified similarities between the Delphi results and the selection of priority items by adult participants, particularly in relation to managing physical symptoms such as pain, being able to live life to the fullest and undertake activities that provide enjoyment. However, the group also identified the importance of being able to access education and maintain peer relations. These items were not ranked in the top 50% by parents or professionals. This finding corroborates previous research that identified the importance of addressing not only physical needs but also supporting pursuit of activities which are part of normalcy for children[32, 43-45]. Input from the group informed the C-POS item regarding ability to undertake usual activities. It also highlights the importance of input from all stakeholder groups in the development of PCOMs. The involvement of children and young people affirms that it is both possible and vital for children to have the opportunity to participate in the development of PCOMs intended for their use, and not rely on proxy reporting alone[46, 47].

What this study adds

Our robust, sequential approach to the development of C-POS has ensured that items are an accurate reflection of the outcomes that are important to children with life-limiting conditions and their families[27]. Involving professionals in the measure development process has helped raise awareness of the development of C-POS and the use of PCOMs in clinical practice. Evidence shows that healthcare professionals need more education on the use and implementation of PCOMs in clinical practice, and suggests that engaging professionals in measure development processes should help to achieve this[48].

Strengths and limitations

The C-POS development process follows outcome measure development guidance from COSMIN and Rothrock[21, 27]. This has ensured that by involving key stakeholders C-POS has excellent face and content validity for the construct being measured, the target population and context of use[28]. Delphi participants were recruited from across three of the four UK nations, and from multiple regions in England. There is geographical variation in UK paediatric palliative care service provision, and widespread recruitment allowed for differences in priority based on provision to be accounted for[5]. We recruited a relatively large number of participants, with many Delphi surveys recruiting less than 50 participants[49].

The lack of ethnic diversity of parents recruited to the Delphi survey is not reflective of the population of children who require palliative care in the UK. Those from Asian, Black and Bangladeshi backgrounds are more likely to have life-limiting conditions[4]. Our parent participants all identified as white British, with four saying their child was of mixed ethnic group. Future research should focus on ways to increase ethnic diversity in paediatric palliative care research, and we will seek to recruit participants from minoritised groups in future C-POS validation work. All of our parent participants were female and this is consistent with much of paediatric palliative care research, i.e. fathers are often under-represented[50].

By round 3 of the Delphi survey only 36.5% of original participants responded. This attrition rate is similar to other Delphi surveys in paediatric palliative care where parents and professionals were included as participants[16]. In our study, attrition was particularly high in parents, with parents forming 15% of the sample in round 3. This

can be attributed to two national COVID-19 pandemic lockdowns during recruitment. These lockdowns led to loss of vital social support and disruption to essential healthcare services, placing additional care burden on families of children with life-limiting conditions[51]. As a result of attrition and concerns about the feasibility of a further round and potential gain, it was decided to stop the Delphi survey before reaching the predetermined criteria (W>0.7)[30]. There is no uniform definition for consensus in Delphi surveys. Although achieving W>0.7 is often used as a stopping criterion, most ranking-type Delphi's report a moderate final consensus rate (W=0.5-0.7)[31, 49]. Our Kendall's W coefficient of concordance increased from weak to moderate between rounds 2 and 3, suggesting a move towards consensus. The increase in proportion of health care professionals in the final ranking round could potentially have contributed to this increase in consensus.

Next steps

Further research is required to demonstrate the comprehensiveness, comprehensibility and acceptability of C-POS using cognitive interviews, followed by psychometric testing.

Conclusions

C-POS has undergone a robust development process using accepted methodological guidance on PROM development. This has ensured items within the measure reflect the construct set out to be measured, and that they have face and content validity within the target population. Important differences were found in priority outcomes identified by different stakeholder groups, highlighting the importance of involving all key stakeholders in PCOM development.

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Author contributions

LC, DB, KB, CES, LF, FM and RH contributed to study concept and design, analysis and interpretation of the data, drafting of the manuscript and critical revision of the manuscript for important intellectual content. DH, HS, JD and MBL contributed to analysis and interpretation of the data, drafting of the manuscript and critical revision of the manuscript for important intellectual content. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Declaration of conflicting interests

There are no conflicts of interest to declare.

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Appendix O Qualitative interview study recruitment poster

KING'S College LONDON

Children's Palliative Outcome Scale (C-POS)

Cicely Saunders International
Better care at the end of life

We are currently recruiting for Phase 1 (tool development stage) of the C-POS study.
The aims of this phase are:

- To identify priorities of children and young people and their families regarding care and questionnaire completion methods.
- To identify clinician and commissioner priorities for patient reported outcome measure (PROM) items.

This will enable us to develop a draft tool for validation and implementation.

We are recruiting the following:

Children and Young People

- 5-17years
- Referred to the palliative care (PC) team and had at least 1 consultation
- Living with a life-limiting/life threatening (LL/LT) condition
- Able to assent/consent to the study
- Able to communicate views via parent/carer, in depth interview or draw and talk or play methods.

Parents

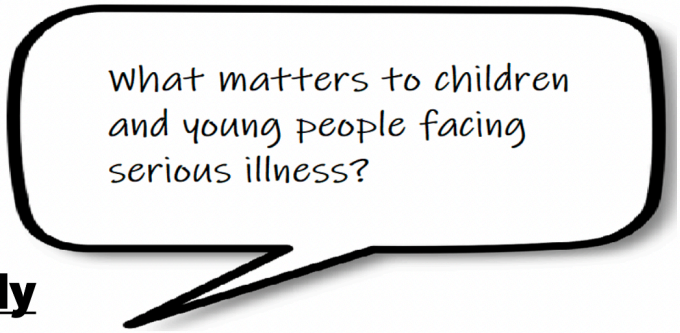
- Of children 0-17years
- Whose child has been referred to the PC team and had at least 1 consultation and has a LL/LT condition
- Are able to provide informed consent

Siblings

- Of children 0-17 years who have been referred to the PC team and had at least 1 consultation and have a LL/LT condition
- Are able to provide assent/ consent

If you are seeing children/families who meet the above criteria please discuss this study with them at their next consultation. If they verbally agree to receiving further information from the research team then please contact Lucy Coombes at lucy.coombes@kcl.ac.uk or 0207 848 5559 or Anna Roach at anna.e.roach@kcl.ac.uk or 02078485536

Appendix P Recruitment support flier



The C-POS Study

Thank you for helping recruit to our study exploring what is important to children and young people with serious illness.

We are looking for children and young people aged 5-17 who are living with a life-limiting or life-threatening condition to take part in a one-off interview.

When research involves children, it is not unusual to be worried about the possibility of causing upset. Parents may be concerned about their child being involved in research and the impact it could have. However, there is currently a large gap in research and the voices of children are not being acknowledged.

Children have a right to express their feelings and wishes, especially in matters that affect them. There is also evidence that children and young people appreciate being included in research and having their opinions heard.

We are asking you, as clinicians, to identify any children aged 5-17 living with a life-limiting or life-threatening condition, to give our study information sheet to their parent/carer, and ask permission for the adult's contact details to be shared with the C-POS Team. We will then follow up with families to answer any questions and to ask if they are interested in participating.

Here is a possible way to introduce the study to potential participants:

"Could I tell you about a research project that colleagues from King's are currently undertaking? They are interested in talking to children and young people who are living with serious illness to find out what is important to them. The King's team has asked me to talk to parents/carers and their children about this research. Could I give you this information sheet and give your contact details to the team who will answer any questions and see if it's something you'd be interested in?"

Here are answers to some Frequently Asked Questions:

- We are happy to meet with parents beforehand and discuss any concerns.
- Children will not be given any information beyond what they already know. The purpose of the study is simply to ask children and young people who have been living with illness what matters most to them.
- Even if parents are keen for their child to take part, the child will have the opportunity to refuse and this will be respected.
- We have a detailed plan if the child does become distressed in any way or if the child tells us something that is worrying.
- Interviews tend to last around an hour and are always arranged at a time and place convenient to patients and families.
- The interviews will be anonymised, so the patient and their family will not be recognisable.
- Findings from the interviews will be used to develop an outcome measure for use with children with serious illness (C-POS).
- Your role as clinicians is to identify potential participants, give them an information sheet and ask their permission to pass their contact details to the C-POS Study – the C-POS Team will do the rest!

For more information, or to pass on details of potential participants, please contact one of the study team.

Thank you for your help with recruiting to the C-POS study!

Appendix Q Spiritual, religious, and existential concerns paper

PALLIATIVE MEDICINE

Original Article

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Spiritual, religious, and existential concerns of children and young people with life-limiting and life-threatening conditions: A qualitative interview study

Hannah May Scott[1], Lucy Coombes[1,2], Debbie Braybrook[1], Anna Roach[1,3], Daney Harðardóttir[1], Katherine Bristowe[1], Clare Ellis-Smith[1], Julia Downing[1,4], Fliss EM Murtagh[5], Bobbie Farsides[6], Lorna K Fraser[7], Myra Bluebond-Langner[3,8] and Richard Harding[1]; On Behalf of C-POS

Abstract

Background: Despite being a core domain of palliative care, primary data on spiritual and existential concerns has rarely been collected among children with life-limiting and life-threatening conditions and their families. Existing evidence has tended to focus on the religious aspects among children with cancer.

Aim: To identify the spiritual needs of children with life-limiting and life-threatening conditions.

Design: Cross-sectional semi-structured, qualitative interview study with children, families and health and social care professionals. Verbatim transcripts were analysed using Framework analysis

Setting/participants: Purposively sampled children with life-limiting and life-threatening conditions, their parents and siblings, health and social care professionals recruited from six hospitals and three children's hospices in the UK, and commissioners of paediatric palliative care services recruited through networks and a national charity.

Results: One hundred six participants were interviewed: 26 children (5–17 years), 53 family members (parents/carers of children 0–17 years and siblings (5–17 years)), 27 professionals (health and social care professionals and commissioners of paediatric palliative care). Themes included: living life to the fullest, meaning of life and leaving a legacy, uncertainty about the future, determination to survive, accepting or fighting the future and role of religion. Children as young as 5 years old identified needs or concerns in the spiritual domain of care.

Conclusions: Addressing spiritual concerns is essential to providing child- and family-centred palliative care. Eliciting spiritual concerns may enable health and social care professionals to identify the things that can support and enhance a meaningful life and legacy for children and their families.

Keywords

Child, palliative care, spiritual concerns, existential concerns, religious concerns, terminal illness

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What is already known about the topic?

- Although spiritual concerns are recognised as a core component of palliative care for children, there is a paucity of primary data.
- Self-report data from children is rare, and existing evidence is largely proxy data from parents or health and social care professionals and mainly focused on the religious aspect of spiritual care for cancer patients.

What this paper adds?

- Specific spiritual concerns among children with a range of life-limiting and life-threatening conditions and their families (parents and siblings) included: living life to the fullest, meaning of life and leaving a legacy, uncertainty about the future, determination to survive, accepting or fighting the future and role of religion.
- This work broadens understanding of the spiritual domain for these children beyond religious needs to existential and value-based spiritual concerns.
- Recognition of the way in which children conceptualise spirituality and being able to identify their spiritual concerns is essential for child- and family-centred holistic palliative and end-of-life care.

Implications for practice, theory or policy:

- Professionals can optimise children and family's wellbeing through identification of the things that provide meaning for them, and working together to set goals and actions towards achieving them.
- Such concerns must be assessed beyond religious considerations.
- Simple tools and training to support professional may be useful in implementing this.

Background

Children and young people living with life-limiting and life-threatening conditions have many complex symptoms and concerns that span health and social care domains.[1] Paediatric palliative care is a holistic approach that aims to address and manage the symptoms and concerns of children and young people with life-limiting and life-threatening conditions and their families.[2] The goal is to identify and improve symptoms and concerns across four core domains: physical, psychological, social and spiritual.[3]

Whilst there has been considerable debate as to what constitutes spirituality in adult palliative care, one of the more accepted definitions is that: 'Spirituality is the aspect of humanity that refers to the way individuals seek and express meaning and purpose and the way they experience their connectedness to the moment, to self, to others, to nature, and to the significant or sacred'.[4] Moreover consensus work by the European Association of Palliative Care (EAPC) Reference Group on Spiritual Care states that spiritual care requires consideration of people's values (e.g. what is most important for each person), faith and beliefs, and existential concerns (e.g. concerns surrounding meaning of life, hope and death).[5] However spirituality in children and thus the extent to which this definition meets the needs of children with life-limiting and life-threatening conditions is less well understood.[6]

Spirituality for dying children is often linked to the losses they face relating to their sense of normality or that which is normal for them.[6] Thus spiritual care should be about supporting children and families with meaning-making and redefining hope, whether that is in a religious sense or not.[6] Spiritual care for children and young people has also been defined as that which addresses and resolves spiritual or existential distress such as fear or questions like 'why me?', as well as supporting them to find meaning and exploring legacy.[7] This paper accepts the definition of spirituality as put forward and agreed upon for adult palliative care[4,8,9] but extends it to also recognise the importance of hope and normality in spirituality for children.[6,7]

In a recent systematic review on spiritual care in palliative care, only two of 53 included studies were of paediatric populations and neither study included primary data from children.[10] Similarly, a systematic review of symptoms and concerns of children with life-limiting and life-threatening conditions found that of the 37/81 studies that included spiritual concerns, nearly all focused on cancer and recruited professionals or parents as proxies. Moreover, much of the existing literature has focused more on religious aspects of spirituality.[11,12] As part of a larger study to develop and test outcome measurement for children and young people facing life-limiting illness[13] this article aims to identify and describe the components of the spiritual domain of palliative care for children with life-limiting and life-threatening conditions as expressed by the children themselves, their families, professionals and commissioners.

Methods

Research question

What are the core spiritual, existential and religious concerns of children, young people and their families facing life-threatening or terminal illness?

Design

A semi-structured, qualitative interview study was conducted from a critical realist perspective[14] and reported in accordance with the consolidated criteria for reporting qualitative studies (COREQ).[15] This data was collected as part a study aiming to identify priority outcomes for care for children with life-limiting and life-threatening conditions and their families[13] as part of the larger Children's Palliative care Outcome Scale (C-POS) study aimed at developing and validating a child- and family-centred outcome measure for implementation into routine paediatric palliative care.

Population

Inclusion criteria: Children (5–17 years) with any life-limiting condition; parents/carers with a child $<$18 years old with a life-limiting condition; siblings (5–17 years) of children with a life-limiting condition; healthcare professionals with $>$6 months experience of caring for children with life-limiting conditions; commissioners of UK paediatric palliative care services.

Exclusion criteria: children unable to communicate via an in-depth interview or by using 'draw and talk' or play methods or via their parents, and/or speak a language not supported by NHS translation services, and/or currently enrolled in another study, and/or unable to give consent/assent; parents/carers and siblings unable to give consent/assent, and/or speak a language not supported by NHS translation services.

Setting

Six hospitals and three children's hospices in the UK across England and Northern Ireland.

Sampling

Children and their families were purposively sampled to ensure representation of a wide range of ages and conditions as evidence suggests that the symptoms and concerns that matter to children and young people vary by age and diagnosis.[1]

Recruitment

Children and their families were identified by their care teams at the nine recruiting sites during their weekly multidisciplinary team meetings, during ward rounds or during outpatient appointments. Eligible families were then approached by a member of their care team who would introduce the study to them and request them to consider participation. If they expressed an interest to learn more about the study, child age-specific information sheets and the parent/caregiver information sheets were provided. Families had 2 weeks to decide whether to proceed and clarify if they would like to contact the researcher direct (email, telephone), or whether they would like the researcher from King's College London to make contact.

Healthcare professionals were identified by the site service manager who approached staff with information on the study and took verbal consent to share their contact details with research team members. Commissioners of UK paediatric palliative care services were recruited via the study's partner professionals and a national children's palliative care non-governmental organisation.

Data collection

Semi-structured interviews were conducted by LC (experienced children's palliative care nurse, new to qualitative research), AR (experienced in working with children, new to qualitative research) and DB (experienced qualitative researcher) between April 2019 and September 2020. All interviewers received training and supervision on conducting interviews with children, including the legal, ethical and communication issues.

Interviews commenced with demographic questions and children were asked about their hobbies and interests to build rapport. 'Draw and talk' and play methods were used, with toys, paper and pencils provided to children to aid interviews where required to facilitate the interviews. A topic guide with open questions informed by a systematic review of symptoms and concerns in children with life-limiting conditions[1] and the World Health Organisation (WHO) definition of paediatric palliative care[16] was used to ensure all domains of the WHO definition of palliative care was discussed.

Interviews were audio-recorded, transcribed verbatim and pseudonymised.

Data analysis

Full transcripts were deductively and inductively coded[17,18] by LC, DB, AR, HS (experienced qualitative researcher, experienced in working with children), and DH (experienced in working with children, new to qualitative research) using NVivo 12 Software.

Analysis followed the five-step Framework method[17,18]; familiarisation, coding, developing an analytical framework, applying the framework, charting and interpretation. Interviews were analysed individually, and the within stakeholder groups, across age ranges, and then across stakeholder groups. The use of Framework analysis

enabled comparison and contrast of findings across the sample both within and across stakeholder groups. As part of robust and valid outcome measurement development, recommendations[19] and guidelines[20] emphasise the importance of key stakeholder views to ensure face and content validity. It is only by looking across different stakeholder groups that similarities and differences between groups can be identified. Importantly, some groups may overlook something that they do not consider important or may find difficult to talk about. Thus, whilst different participants both within and across groups may view or understand the construct of spirituality in different ways, it is important to understand it from all of their perspectives to support the delivery of person-centred care for children that addresses spiritual concerns.

Regular meetings were held to discuss emerging themes and resolve any differences (20% of transcripts were independently coded by two researchers). RH, KB and CES were consulted and resolved if disagreement in coding and interpretation arose. Analysis was reviewed by the study steering group throughout the study.

Ethical issues

Ethical approval was granted by the Bloomsbury research ethics committee (HRA:19/LO/0033). Participants over 16 years old provided written informed consent. Those with parental responsibility provided written informed consent for participants<16 years. Those <16 years provided written assent.

Results

Sample characteristics

One hundred and four interviews were conducted with *N* = 106 participants (two parents and two siblings were interviewed together): *n* = 26 children, *n* = 53 family members, *n* = 15 health and social care professionals and *n* = 12 commissioners (see Table 1). To protect anonymity of children with rare conditions, International Classification of Diseases-10 (ICD-10) chapter headings are reported in lieu of precise diagnoses.[13]

Main findings

The framework analysis identified seven themes that relate to the three dimensions of the spiritual domain of care as defined by the EAPC Reference Group on Spiritual Care[5]: (1) Relating to the term 'spiritual', (2) living life to the fullest, (3) meaning of life and leaving a legacy, (4) uncertainty of the future, (5) determination to survive, (6) accepting or fighting the future, and (7) role of religion. Figure 1 shows how these themes relate to the three dimensions of the spiritual domain of care as defined by the EAPC reference group.[5]

Table 1. Participant demographic characteristic.

	N or mean (range)
Children (*n* = 26)	
Age (years)	12 (5–17)
Gender	
Female:Male	17:9
Diagnosis	
Gastrointestinal	10
Cancer	6
Neurological	5
Congenital	3
Metabolic	1
Respiratory	1
Family members (*n* = 53)	
Age (years)	32 (5–65)
Gender	3
Female:Male	7:16
Relationship to child	
Mother	29
Father	10
Sister	7
Brother	6
Sibling Caregiver	1
Diagnosis of child	
Neurological	17
Congenital	10
Metabolic	10
Cancer	6
Gastrointestinal	6
Infectious disease	2
Genitourinary	1
Perinatal	1
Age of child with life-limiting condition (years)	10 (0–17)
Professionals (*n* = 27)	
Gender	
Female:Male	25:2
Profession	
Commissioner of paediatric palliative care	12
Nurse	7
Doctor	3
Chaplain	1
Physiotherapist	1
Play specialist	1
Psychologist	1
Social worker	1

Dimension 1: Personal values

Theme 1: Relating to the term 'spiritual'

Some health and social care professionals discussed a lack of confidence in asking about spiritual concerns, and noted that in terms of asking children and families about spiritual needs, this often focused only on religious needs. This was also reflected in parents understanding of

spiritual needs with parents often responding to questions about spiritual needs in terms of religion

> I think sometimes when we say spiritual needs I think er we look more at the religious side but that does not always reflect spiritual needs *[Oncology nurse]*

> Erm yeah [parent] and I are not remotely sort of religious. So erm, no we don't really have any. I mean, spiritual sort of needs *[Mother of child aged 8 with congenital condition]*

When asked directly about spiritual concerns, parents stated that they and their children did not have needs in this domain

> Interviewer: Do you think that (child) has any spiritual concerns or any like concept of spirituality?

> No, I don't think he has any. . .any concept of that whatsoever, not that I've. . .no I don't think, no. . .no *[Father of child aged 12 with congenital condition]*

Theme 2: Living life to the fullest

Being able to take part in activities they enjoyed was very important to children. Being able to do the things that made their life meaningful, and retained some degree of normality, contributed to 'living life to the fullest'.

> Well I'm happy because I get better, but then I'm sad because I miss school, miss my friends, miss my family *[Child aged 12 with Respiratory condition]*

> Interviewer: what do you mean by living your life

> Doing what I want in my life *[Child aged 9 with gastrointestinal condition]*

Health and social care professionals and commissioners recognised this theme as important, and wanted to help enable children to live their lives to the fullest through goal setting and supporting them to continue to do the things that were important to them.

> you know that they're en. . .enabled to live the. . .the fullest life that they're able to do. Do you know what I mean that they. . .they can achieve some of their goals and development and. . .and as appropriate to them *[Commissioner]*

> for their quality of life it is about going to school and being around friends and trying to be normal *[Psychologist]*

For parents this was often talked about in terms of ensuring that their children had quality of life, particularly in terms of comfort and happiness.

> We can't change his length of life, but I can do everything I can to make sure his quality of life is good and that he lives his best life possible *[Mother of child aged 2.5 with metabolic condition]*

Dimension 2: Existential concerns

Theme 3: Meaning of life and leaving a legacy

Parents and children often had questions such as 'why me?'. Professionals and parents also highlighted that children expressed concern that they had done something wrong to deserve their illness

> You know 'why me?' and we had a lot of anger first off, again the issue I just said 'Oh you know "eat your veg, fruit and veg, you know you'll be big and strong" you know', 'drink lots of water because it's good for you' erm. . .and initially we had the "well you lied to me, why. . .you know why, why me. Why, what have I done wrong?"' *[Father of 13-year-old child with a gastrointestinal condition]*

> I think one of the things that children will often worry about. . .about things that they feel they've done wrong [. . .] those sort of things really do play very heavily on erm. . .small children and particularly if they'd had some kind of religious background maybe from, you know perhaps going to a faith school or. . .or having RE lessons at school. *[Hospital Chaplain]*

Children also wanted their life to mean something and to leave a legacy after they had died. This included setting up charity initiatives, inspiring other children and leaving messages for their family members

> I don't think there's much point in going through life and just supporting yourself, because when you go through. . .at the end of life, you're gonna think, what have I done for other people? Because those are the people, you're gonna miss and you're think, what have I actually done for them. If you haven't done anything for them, you may regret it [. . .] I can write a story, I can include that and hopefully help other people to make that kind of change *[Child aged 17 with gastrointestinal condition]*

> I did quite a lot of work about her bucket list and wanting to and her legacy and wanting to, she wanted to leave messages for her brother and other people in her family *[Social worker and family therapist]*

Theme 4: Uncertainty about the future

Uncertainty about the future was a concern for both children and their families.

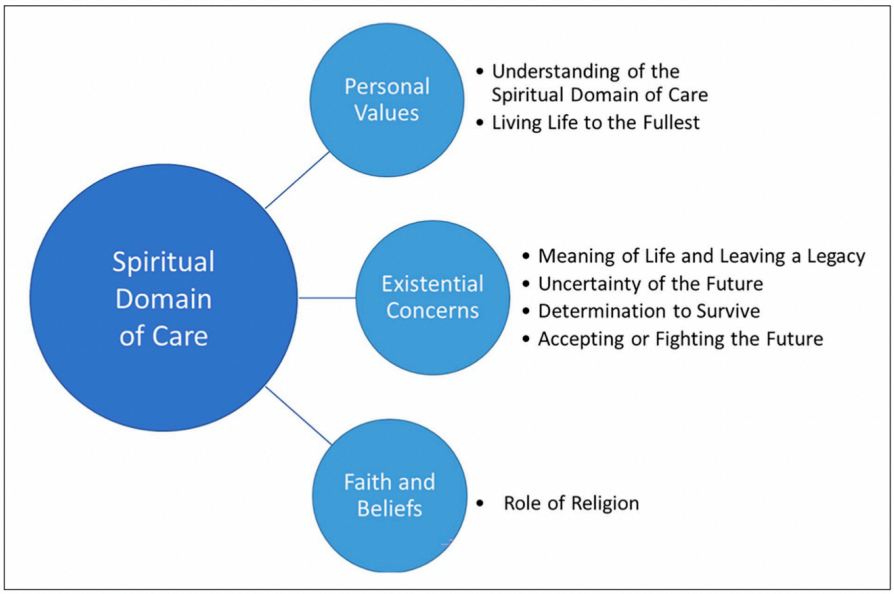

Figure 1. Spiritual domain of care.

There was longer-term uncertainty around their prognosis, how long they had to live, and worry about dying, particularly for older children. Parents and one sibling also expressed similar concerns

> I felt worried when I was, like really sick in [month]. Umm, yeah, I was just worried like [long pause] and then [pause] like, was I gonna get through it and like stuff like that. Because, that time was really [pause] wasn't the best *[Child aged 15 with gastrointestinal condition]*

> And I think particularly the older kids, although there's sort of an understanding that their life, well they're 15 or they're 16, they understand what's going on, they are often the most frightened because they have complete understanding. *[Transplant Nurse]*

> They don't know how long he has *[Mother of child aged 1 with congenital condition]*

> Interviewer: Okay and what would you say worries you and what's your biggest worry about. . .?

> Umm . . .that erm. . .she might die *[Sibling of Child aged 14 with Neurological condition]*

However, the unpredictable nature of life-limiting and life-threatening conditions often created uncertainty in the more immediate short-term. This made it difficult for children and their families to make plans due to unpredictable changes in symptoms

> You can't really arrange things because you might arrange something for this week or the next week and then you get to that week and you have got an infection and you are in hospital or something like that. So it changes. . . so you can't really plan much. *[Child aged 15 with cancer]*

> we literally take every day as it comes. We don't plan anything. We just do day by day which is all you have to do with a sick child *[Mother of child aged 4 with congenital condition]*

Theme 5: Determination to survive

Children voiced a determination to live their lives and survive despite their health conditions, wanting to be brave and overcome their prognosis

> It's ok because I am brave *[Child aged 5 with Gastrointestinal condition]*

> with cancer you want to live, you want to show people that you can overcome this *[Child aged 13 with cancer]*

> the fact that I'm still here means that my journey hasn't finished *[Child aged 17 with metabolic condition]*

Parents also spoke of their children's determination in terms of being brave or fighting, despite their condition and prognosis

> she's classed as an end of life stage now, but she's plodding on, she's still got her fight *[Mother of child aged 8 with neurological condition]*

> I think part of it is that she feels like she doesn't want people to know she's not coping or she, you know, she doesn't want people to know that she's been upset or you know. There's still that stigma behind it, about being brave *[Mother of child aged 12 with cancer]*

> He has got such a strong heart. He keeps fighting back. He is a fighter. And you know, you have to give him a lot of credit for that. He's a strong little soul *[Mother of 10 year old with neurological condition]*

Theme 6: Accepting or fighting the future

Older children were often more aware of the life-limiting nature of their condition and prognosis. Whilst some were accepting others expressed internal conflict over accepting the possibility or actuality of death whilst simultaneously wanting to fight to keep going and survive

> you just keep going until you reach that final choice or decision of what's going to happen to you, either you can overcome this or a different consequence that everyone hopes will never happen *[Child aged 13 with Cancer]*

> If they realise that they are, that they are going to die, you know, they feel distraught cos they are not going to have a life that others have. They are more aware. The young ones are not so aware of what they are going to miss out on, what they are not going to achieve, whereas the older ones, they know. They can see, they know what they're going to lose *[Oncology nurse]*

> if you talk about (child) dying he's just like 'I don't wanna talk about it because it's not gonna happen yet' do you know what I mean *[Mother of 14 year old with metabolic condition]*

Parents also discussed difficulty in accepting the life-limiting aspect of their child's diagnosis, and how challenging it was to come to terms with knowing their child would die

> the biggie is obviously the. . .the life-limiting diagnosis and the fact that we're gonna lose him *[Mother of 7 year old with metabolic condition]*

Irrespective of their condition and prognosis, children still wanted to think about and make plans both for the short-term and the longer-term future

> I wanna be grown up *[Child aged 15 with neurological condition]*

> I would wanna travel like around the world *[Child aged 15 with gastrointestinal condition]*

> The teenager that died recently, I mean she was still going to do her GCSE's this summer. And she died much quicker than we thought. But no, she was definitely going to still do them. *[Nurse]*

Dimension 3: Faith and religion

Theme 7: Role of religion

A number of parents and health and social care professionals discussed children's religious beliefs and needs. Whilst some professionals and parents spoke of children wanting to continue to be able to attend church, many parents spoke in terms of passive identification of their own or their children's beliefs rather than in terms of unmet religious needs

> I had one boy that was very concerned about. . .he wanted to spend time with umm. . .his priest. . .erm and I found that. . .that was. . .that was unusual for me because he was quite young, so that really sort of jarred for me. I've had old. . .I've had adolescents who have really wanted to make sure they could still attend. . .erm church and things like that *[Oncology Clinical Nurse Specialist]*

> I think she'd put herself down as Buddhist but, obviously not practicing *[Father of child aged 16 with cancer]*

Only one child participant spoke about their religious faith and beliefs. They considered their religion a source of strength and comfort and it was important for them to be able to continue to attend church

> I'm a Christian, so I believe in Jesus and that he's my lord and saviour [. . .] as I've gone through all of these. . .all of this and I've been in hospital. . .erm I always remember that, you know there as someone who suffered even worse for me [. . .]there's a greater hope and like the greater hope is in Jesus and that I trust in that. You know even whatever happens, whether you know I die or whether I live, it's for 'Him' and you know I'm just gonna continue to live a life according to his grace *[Child aged 17 with gastrointestinal condition]*

Similarly for some parents, religion could be a source of comfort and their faith became stronger, even influencing their willingness to discuss their child's care. However other parents and health and social care professionals spoke of parents having a crisis of faith or losing their faith altogether due to their child's ill health. In cases when one

parent held onto their faith and the other lost it, religion could become a source of conflict

> the families don't want to discuss anything because their faith is so strong. They are going to be saved *[Children's Community Nurse]*

> I do believe in God and you know sort of and erm the guys here do know I have a faith even though I have thought you know long and hard at times. And had sort of inward arguments with you know with whoever it is that's up there. But I have to hold on to something *[Mother of child aged 10 with neurological condition]*

> I mean there's been recent erm. . .situation where there's been a Greek. . .Greek Orthodox family and they were. . . erm going through a period of great spiritual distress and also confusion. . .erm because within the family, one of the people was Greek Ortho. . .Orthodox, the other one wasn't. . .erm in just the period of the child's illness, one parent had become more religious, the other one had become less religious *[Hospital Chaplain]*

For some families, religion could also be a source of anger (particularly when they did not have religious belief). This was often linked to existential concerns and questions relating to the meaning or reason for developing a life-limiting or life-threatening condition

> (child) finds it really frustrating when people say 'oh I'll pray for you'. . . because actually it's [. . .] 'what God would do this'. . . so when people start saying 'We'll pray for you' and things like that, we think. . .. And often we both just ignore it, we don't comment on it but I know (child) does find it a bit frustrating *[Father of child aged 16 with cancer]*

Discussion

Main findings

This novel primary data provided by children, their family members and professionals has given detailed understanding of a previously under-explored domain. When asked directly about spiritual concerns parent/carers tended to state that they and/or their children did not have spiritual concerns, or responded to the question in terms of religion. However, when probed further, particularly in relation to worries, hopes or other things that were important to them, children and their families discussed many concerns that demonstrate the experience of the dimensions of spirituality in line with the definition of spirituality for children discussed at the beginning of this article.[6,8]

Professionals tended to lack confidence in asking about spiritual concerns, particularly existential concerns and tended to focus more on the religious aspects of spirituality. However, the data revealed that even if asked using broad terms of 'spirituality', families may interpret this to mean religion. The data demonstrate that children and families do have spiritual concerns to be addressed (although notably, only one sibling discussed spiritual concerns in the interviews). These concerns may need to be asked about in a less direct way; for example, asking them about the things that are important to them, their hopes, and worries, rather than if they have any spiritual or religious needs or concerns.

What this study adds

Whilst some of the themes echoed those identified in a recent systematic review,[1] such as meaning of life and determination to survive, other novel, important themes were identified. It was particularly important for children to live life to the fullest through being enabled and supported to continue to engage in activities that were important to them and that gave their lives meaning. These included taking part in their hobbies, going to school and seeing friends and family. These highlight the importance of continuing normality and maintaining meaning as core components of spiritual wellbeing.[13]

Understanding the way in which children and families conceptualise and express spiritual concerns is essential to assessment and planning. Additional training for professionals may also be beneficial to develop their confidence and support them in asking about spiritual concerns. As is a strength of this work in including the voices of children in an area where their perspectives are often lacking, it is recommend that the perspectives of children be included in training and development for professionals to help overcome the pervasive view that children do not have spiritual needs. Fear of causing distress, or not having a solution, should never be a reason for clinicians not asking about spiritual concerns. Instead of directly asking if children and their families have spiritual or existential concerns, professionals should consider shaping discussions about spiritual and existential needs in the context of what matters to the child, and what they value in terms of their usual routines, in order to talk about their life, future and spiritual or existential concerns and build confidence in talking about and addressing these concerns. Moreover, professionals need to be aware of the potential for faith to become a source of conflict between family members in order to be able to best support families.

Strengths and weaknesses

This research explored a previously under-researched area despite the well documented ethical challenges in conducting primary research with this population relating to difficulties in gaining ethics approval and clinician gatekeeping.[21,22] Previous studies in this area have tended to

rely on proxy reports from parents/carers and professionals, or focused solely on children with a cancer diagnosis.[1] A major strength of this study is the involvement of children themselves from the age of 5 years old, and with a range of life-limiting conditions. The sample size was also relatively large in comparison to other studies that include children with life-limiting conditions, and the geographical spread of participant recruitment across the UK. However there were also limitations. There were only a small number of UK recruitment sites and one site only recruited children with gastrointestinal diagnoses, reflected in the larger number of participants from this group. Moreover a lack of ability to extensively probe in interviews with children within a larger study to identify the breadth of their symptoms and concerns means that some concerns may have been missed or underexplored. Whilst interviews were adapted for children's age and developmental stage, additional child-centred techniques (such as using photographs or puppets) may have been able to better elicit children's concerns and needs.[23–25]

Conclusion

If care is to be child and family-centred, then spiritual concerns should be identified and addressed equally to other domains of care. Children have spiritual concerns when facing life-limiting and life-threatening conditions, and, when explored appropriately, have the capacity to articulate these. For professionals in practice, it is important work with children and their families to identify the things that are most important to them and that give their lives meaning, and to work together to set achievable goals that can support a meaningful life and legacy.

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Author contributions

All authors: conception and design of the work. LC, DB and AR: data collection. LC, DB, AR, DH and HS: data analysis. All authors: interpretation of data. HS prepared draft of paper. All authors: critical review and revision of article.

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