Treatment Experiences of the Care pathway for People with Cleft Lip and/or Palate Orthodontic Treatment in Context

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Treatment Experiences of the Care pathway for People with Cleft Lip and/or Palate: Orthodontic Treatment in Context

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ABSTRACT

Cleft lip and/or palate (CLP) are common deformities that are known to occur in 1 in every 700 new-born babies. The causes can be genetic and/or environmental. Its management is a complex process which involves many disciplines to ensure the delivery of the best care available.

Objectives: To explore the perspectives of people with CLP about their definitive orthodontic/orthognathic treatment within the context of the cleft care pathway and living with the condition.

Methods: This qualitative study used semi-structured in-depth interviews to explore the experiences of people with CLP who have recently finished orthodontic/orthognathic treatment. The CLP participants were recruited from two centres in the UK. This study used a purposive sampling strategy. All interviews were recorded and transcribed verbatim. Data analysis was conducted using a thematic framework developed through the analysis process.

Results: This study gives an insight into the cleft care pathway from the perspectives of those being treated within it. The first part of the study explores experiences of living with CLP on a daily basis and the impact of the care pathway within this. The results suggest that the care pathway cannot be separated from daily life. The majority of respondents in this study were born into the care pathway due to prenatal diagnosis, and grew up within the pathway. Treatment and the provision of information need to be understood within this context. There is an interrelation between experiences at home, school and work and the impact of, and satisfaction with, treatment received at different stages in the pathway.

The second part of the study focused in more detail on aspects of treatment. The results suggest that treatment choices are shaped by experiences of stigma, low self-esteem and a resulting focus on aesthetics. The definitive orthodontic/orthognathic treatment is seen within this process as the end point of treatment at which time participants would become 'normal' and leave the care pathway.

Conclusion: The study provides a new insight into the cleft care pathway from the perspective of those within it and can help to inform future developments, particularly around the provision of information and the need to be aware of the wider context of patients.

TABLE OF CONTENTS

LIST OF FIGURES

LIST OF TABLES

LIST OF ABBREVIATIONS

BCLP	Bilateral Cleft Lip and Palate
CLAPA	The Cleft Lip And Palate Association
CLO	Cleft Lip Only
CLP	Cleft Lip and/or Palate
CPO	Cleft Palate Only
MS	Multiple Sclerosis
NHS	National Health Service
OGN	Orthognathic
UCLP	Unilateral Cleft Lip and Palate
UK	United Kingdom

1. INTRODUCTION

Cleft lip and/or palate (CLP) are common deformities that are known to happen in 1 in every 700 new-born babies (Bender, 2000). The causes can be genetic and environmental. Its management is a complex process which involves many disciplines to ensure the delivery of the best care available. Treatment is long, complex and arduous and takes a considerable amount of clinical time, resource and expertise. For patients, they must deal first with living with a 'visible' long term condition, and then cope with managing the care pathway. In contrast to other long term conditions patients may move from considerable disability to improved appearance, function and social functioning as treatment progresses.

The ability to suck, chew, eat, and hear may be affected in a child with CLP, in addition to possible disturbances in speech development, dental and facial appearance. Consequently, emotional and social well-being may be affected and behavioural and learning difficulties might occur.

This study seeks to explore how people with CLP perceive their orthodontic/orthognathic treatment within the wider context of daily life and the specifics of the care pathway. Developing an understanding of the experiences of CLP patients with their treatment will enable clinicians to better understand the impact of treatment and the care pathway on their patients.

1.1 Rationale for the study

In recent years, the importance of patient centred care through the inclusion of patients' opinions and perspectives has been encouraged. Patient centred care can be defined as "respectful of and responsive to individual patient preferences, needs, and values, and ensuring that patient values guide all clinical decisions." (Institute of Medicine, 2001). Patient centred care aims to understand the patient's treatment experiences and needs, and how they perceive their treatment. It provides the framework and strategies to improve the experience of care and enhances efficiency, quality and safety. A patient centred approach to care has been shown to improve the outcomes of treatment and the

quality of life; increase patient adherence to treatment regimens and instructions; reduce anxiety and depression; address racial, ethnic and socioeconomic diversities in care; and reduce the overuse of diagnostic testing (Epstein et al., 2010). Understanding the patient's perspective is an important pre-requisite for patient centred care.

CLP are conditions that affect the individual from birth. The affected person goes through a comprehensive treatment pathway from birth until late adulthood. We have significant technical data on clinical aspects of care, but little is known about the patient's perspective on the care pathway (including the definitive orthodontic/orthognathic treatment) and how this is set within the context of living with a long term condition.

There is little detailed work that has focused on the patient experience in relation to the orthodontic management of CLP in the care pathway. Moreover, we do not know about patient's perception of the treatment they go through nor how much they understand about the different aspects of treatment. In particular we know little about how they perceive the definitive orthodontic/orthognathic stage of treatment. Knowing about patient experiences is important if we want patients to be fully involved and satisfied with their treatment. If a clinical service is to be designed to maximise patient satisfaction, then we need to understand what a care pathway means to patients who have this condition, what it means to have CLP treated and to live with it. Also there is evidence that the inclusion of patient perspectives can improve the outcomes of care (Epstein et al., 2010).

The clinical pathway has been designed from clinicians' perspectives, there is little insight into the patients' perspectives of this pathway. Several studies for example have assessed the quality of treatment from the medical and professionals' perspective (Noar, 1992, Noar, 1991, Sandy et al., 1998, Semb et al., 2005a), but fewer studies have tried to understand the concept from patients' or parents' perspectives (Hall et al., 2012, Nelson et al., 2012b). The literature is lacking an understanding of how patients have perceived and experienced their involvement in the definitive orthodontic/orthognathic treatment. Areas which lack understanding include problems with treatment, worries over treatment and the degrees of satisfaction with the definitive orthodontic treatment of their care pathway, including how the patient perceives the treatment contributes to the whole care pathway.

This study also seeks to develop an understanding of the importance of the definitive orthodontic/orthognathic treatment in a wider context including understanding its impact on the day to day life of patients. These are the gaps that this study seeks to fill.

This study has included experiences of people who have had orthodontic treatment and also orthognathic treatment. The inclusion of those who went along the orthognathic route will enable us to understand if their experience(s) are distinct from those who only have orthodontic treatment. Those who have experienced the orthognathic surgery route experience the additional orthognathic surgery and associated risks. This is a significant intervention clinically but we have little about patients' experiences of it.

The thesis will address an important gap in knowledge which could contribute to developing a more patient centred approach in CLP care. To understand the participant`s perspective and their experiences, many methods may be used. However, our aim is to develop a deeper understanding of what participants think of their experiences and thus a qualitative methodology was followed in this research. This allowed us to gather the patients' perspective using their own words and language, rather than focusing on clinical perspectives which are already well researched.

2. LITERATURE REVIEW

This research is about understanding a phenomenon from a patient's perspective. In order to understand the context of this research, it is important to have an overview of the different aspects involved. We can divide this broadly into clinical aspects and patient aspects. This literature review therefore begins with a brief outline of the development, incidence, aetiology, classification, clinical presentation and clinical management of CLP with a focused explanation of the specific role of the orthodontist in the care pathway. Understanding the role of the orthodontist is crucial to this study because one of the objectives is to understand the experiences of patients who underwent this orthodontic/orthognathic treatment stage of the care pathway. The second part of the literature will put the patient in focus, exploring the available literature on experiences of people living with this condition and how it may affect them psychologically and socially. The experiences related to the CLP care will be reviewed drawing parallels with the wider literature from the sociology of long-term conditions.

2.1 Cleft lip and/or palate

2.1.1 Incidence

CLP are common craniofacial deformities that affect both males and females. They are the most common congenital anomaly to affect the craniofacial region in human beings (Fraser, 1970).

A cleft is a partial or complete split that occurs during the development of the lip or in the palate or both. It can range in size from a small notch in the uvula to a complete bilateral lack of fusion. Isolated cleft lip and palate with no associated syndrome is known to happen in approximately 1 in 700 live births, with a prevalence that varies according to parental ethnicity, gender and the socio-economic status of the family (Bender, 2000). Cleft lip and palate is more common in boys while cleft palate only is more common in girls (Bender, 2000).

The highest incidence of cleft lip with or without cleft palate is noted within the population of Native American Indians, where the incidence is 3.6 in 1,000 infants. Japanese populations have a rate of 2.1 in 1,000 infants, followed by the Chinese with a rate of 1.7 in 1,000 infants, and then Caucasians with 1 in 1,000 infants. The lowest incidence of CLP is known to occur in African-Americans, with 0.3 in 1,000 infants (Croen et al., 1998, Tolarova and Cervenka, 1998). CLP happens more frequently in males than females with almost double the ratio at 2:1 (Tolarova, 1987).

Croen also found that the incidence of native Philippine infants born in their native country had a higher incidence of CLP compared to non-native Philippine infants who were born in the United States. This is possibly due to the higher exposure of causative environmental factors in their native country (Croen et al., 1998).

2.1.2 Embryological development of Cleft lip and/or palate:

Cleft affecting the lip with or without the alveolus

The first branchial arch gives origin to both frontonasal processes, maxillary processes and mandibular processes. The medial nasal processes and the lateral nasal processes originate within the frontonasal process. Around the fifth to sixth week in utero these processes begin to fuse in the midline. The maxillary processes fuse with the lateral and medial nasal processes and then fuse with the opposing maxillary process in the midline. In the order given, this will produce the following structures: nasal septum, the primary palate, the nasal alae and the columella around the nostrils, and the lip.

A failure of fusion between the maxillary process and the medial nasal process will lead to a cleft of the lip and alveolus. Failure of fusion in one side will lead to unilateral defect and if both sides are involved then it is referred to as a bilateral defect (Ireland and McDonald, 2003, Bender, 2000).

Cleft affecting the palate

The two lateral palatal processes develop from the maxillary processes which in turn, are derived from the first branchial arch. The hard palate forms when these two lateral palatal processes fuse. During the 6th week *in utero* these processes grow vertically and

downward around each side of the tongue and around the 8th week *in utero* when the lateral palatal processes move upward rapidly into a horizontal position above the tongue. Then they begin to fuse in the midline to form the hard palate which extends frontally from the incisive foramen until the soft palate and as far posterior as the uvula. By around week 10 *in utero*, the palatal fusion should be complete in a normally developing individual (Ireland and McDonald, 2003, Bender, 2000).

However, if one or both of the lateral palatal processes fail to move from the vertical into the horizontal position above the tongue a palatal cleft may result (Ferguson, 1981).

2.1.3 Causes:

Many factors can play a part in the aetiology of CLP. The literature suggests that the causes of clefts may be linked to genetic and environmental causes (Bender, 2000).

Genetic Factors

The search for the causes of clefts is very complex; multiple techniques are involved and have been used to discover approximately 30 genes that take part in the tissue disruption that results in various types of clefts. Some potential causative genes were identified through linkage and association studies (Schutte and Murray, 1999).

There is an association between CLP and the MSX1 gene. Also, there are major influencing genes that have to be present to cause clefts in addition to other multiple genetic factors. The identified genes that play a role in CLP formation are: Transforming growth factor alpha (TGFα), transforming growth factor beta 3 (TGFβ3), MSX1 and AP2 (Schutte and Murray, 1999).

Researchers have used mice models to provide a controlled experiment population as mice can reproduce quickly into large numbers. The MSX1, TGFβ3, and AP2 were found to be major predisposing genes for mice that developed CLP and these genes were clearly expressed during critical phases of orofacial development. The specific mechanism by which each of these genes affect the facial development varies (Schutte and Murray, 1999).

There is a strong genetic tendency to have cleft evidenced by the fact that monozygomatic twins have rates of 25-45 percent compared to 3-6 percent for dizygomatic twins (Mitchell and Risch, 1992).

Environmental Factors

It is thought that genetic contributions to the development of CLP are greater predictors than environmental ones. Four broad categories of environmental factors can lead to increased risk of developing CLP: external environment, in utero environment, drugs and nutrition. Some teratogens such as antiepileptic drugs (phenytoin, valporic acid), thalidomide and dioxin (pesticides), and retinoic acid have been linked to the facial cleft development (Bender, 2000).

Additionally, smoking and drinking alcohol have also been linked to CLP (Wyszynski and Beaty, 1996). In a month, exposure to four or more alcoholic drinks will elevate the risk of developing CLP significantly (Romitti et al., 1999).

Shaw et al., (1996) found that the exposure of the foetus to maternal smoking during the first trimester increases the risk of developing CLP. It is not yet known exactly how smoking affects the foetus to cause clefts. The authors have hypothesized that it is hypoxia caused by smoking which leads to alteration in facial development.

Malnutrition can also lead to the formation of CLP. Folic acid and vitamin B are essential for normal embryonic facial development. Deficiency of these nutrients can cause clefts (Krapels et al., 2006).

Interestingly, a study found a relation between the CLP birth incident and different seasons during the year (seasonal variation), with a significant increase in the number of cases developing cleft palates during August-September and an increased incidence of cleft lip with or without cleft palate in December-January time (Coupland and Coupland, 1988). A possible explanation could be seasonal variations in exposure to different teratogens at different times of the year.

2.1.4 Classification

There is no one classification system accepted and used worldwide due to the complexity and the variations that CLP can present clinically. There were a number of attempts to classify different types of CLP. CLP may differ in terms of the level of severity and the extent to which structures are affected. It may be as small as a notch of the uvula or as large as a complete lack of fusion. One commonly accepted classification system is based on the incisive foramen as an important embryological landmark (Kernahan and Stark, 1958). It divides cleft types into three main categories:

1- Clefts of the primary palate only: this can range from a small notch in the lip, and can extend to a complete cleft involving the lip and the alveolus up to the incisive foramen posteriorly. It can be unilateral or bilateral (rarely).
2- Clefts of the secondary palate only: this can range from a notch in the uvula and can extend to a complete cleft involving the soft and hard palate up to the incisive foramen anteriorly.
3- Clefts that involve both the primary and secondary palate: this is a complete cleft from the lip to the soft palate. Again this can be unilateral or bilateral.

In clinical situations, CLP may also be described without a classification. They can be syndromic or non-syndromic, unilateral or bilateral, complete or incomplete.

2.1.5 Clinical presentation

Skeletally

Discrepancy in the antero-posterior dimension is often noted in patients with CLP. There is controversy as to the causes of the growth potential differences and the effect on growth potential of scarring caused by early surgery during the first year of life.

One study, investigated the effects of surgery on craniofacial growth in unilateral cleft lip and palate (Mars and Houston, 1990). The study included only Sri Lankan males. Participants were divided into three subgroups: the first group did not have any repair to their cleft lip and palate. The second group had their lip repaired only during infancy but

not the palate. The last group had a lip and palate repair during infancy. These subgroups were compared to healthy non-cleft controls. The results showed that subjects who had no surgery showed better antero-posterior growth. Lip surgery without including the palate had no significant interference with antero-posterior maxillary growth. Those who had palatal surgery had a significant reduction in their antero-posterior maxillary growth (Mars and Houston, 1990).

Cleft lip and palate patients also have reduced mandibular growth potential and this may lead to smaller mandibular size when compared to healthy individuals (Bishara et al., 1976).

Transversely, different degrees and ranges of asymmetry have been noted in individuals with cleft lip and palate (Bishara et al., 1976). The face width differed consistently between CLP patients and non-cleft individuals, with cleft patient's facial width appearing to be narrower (Duffy et al., 2000).

Dentally

The development of tooth germs has a close embryological connection with the occurrence of CLP when referring to the anatomical position and timing (Stahl et al., 2006). It is suggested that the dental lamina is affected because the cleft genes cause a disturbance in various body tissues (Johnson, 1967). In many studies, it has been demonstrated that the incidence of abnormal tooth morphology is higher in people born with CLP compared to the non-cleft population (Tannure et al., 2012). Evidence proposes that people with a cleft have more dental anomalies in the permanent dentition than non-cleft individuals. Anomalies such as abnormal crown morphology, taurodontism, supernumerary teeth, tooth agenesis and microdonic upper lateral incisors are commonly noted (Küchler et al., 2010, Tannure et al., 2012).

Some studies have also reported a higher rate of occurrences of hypodontia and asymmetry in dental development linked to the increased complexity of the cleft (Eerens et al., 2001). One study investigated the relationship between dental anomalies and different CLP sub-phenotypes. The study found that agenesis of the lateral incisor on the non-cleft side may propose an incomplete form of bilateral cleft of the lip (Letra et al., 2007).

Recent studies have confirmed that oral clefts and dental anomalies share common genetic factors (Vieira et al., 2008). It is now known, for example, that teeth around the cleft area have a higher chance of developing as malformed or are missing (Ranta, 1986).

2.1.6 Management

The management of patients with CLP requires a multidisciplinary team approach reflecting the difficulty and complexity of treatment (De Ladeira and Alonso, 2012). Although it is a necessity, not every country has the luxury to be able to provide the multidisciplinary approach to treat CLP patients. In Africa, care providers face limited resources to support CLP care (Adetayo et al., 2012). In any CLP care pathway, clinical management starts from birth and extends into adulthood. A multidisciplinary team may include a paediatrician, cleft specialist nurse, cleft surgeon, orthodontist, speech and language therapist, ENT audiology specialist, paediatric dentist, restorative dentist, general dental practitioner (GDP), psychologist and clinical geneticist (Health, 1998, Nahai et al., 2005, De Ladeira and Alonso, 2012). The management will involve dental and non-dental disciplines with a number of different clinical procedures each dealing with different aspects of care. The following table (Table 1) summarises the roles of each discipline and their contribution to the CLP care pathway:

Table 1: Overview of the CLP care pathway

TIMING	Cleft Team member and procedures
At birth	**Cleft Nurse specialist:** Counselling and information for parents, Feeding advice. **Orthodontist:** Pre-surgical orthopaedics.
3-4 months	**Cleft Surgeon:** Lip repair.
6-9 months	**Cleft Surgeon:** Palate repair.
2-5 years	**Paediatric dentist:** Preventive dental advice and measures (deciduous teeth). **Speech therapist:** Speech assessment and treatment. **ENT Surgeon:** ENT assessment. **Cleft Surgeon:** Speech surgery (if needed to improve speech), fistula repair. - (Records at age 5 years).
Mixed dentition 7-10 years	**Paediatric dentist:** Continue preventive dental advice and measures. **Orthodontist:** Assessment, expansion before alveolar bone graft. **Cleft Surgeon:** Alveolar bone graft. - (Records at age 10 years).
11-15 years	**Orthodontist:** Definitive orthodontic treatment. - (Records at 15 years).
15-20 years	**Orthognathic team:** OGN surgery (if needed). **Cleft Surgeon:** If necessary (Rhinoplasty, revision surgery). **Restorative dentist:** To replace missing teeth. - (Records at 20 years).

To achieve optimal long term results CLP patients need a comprehensive multidisciplinary team approach. The responsibilities of key professions in the multidisciplinary care team are outlined briefly below (Cooke, 1989):

Cleft surgeon: To anatomically and functionally repair the cleft in the lip and/or in the palate. To improve the function of speech by performing speech surgery and possible pharyngoplasty. Also the surgeon can undertake alveolar bone graft surgery to repair the tooth bearing cleft area to help establish a continuous dental arch and to allow the cleft tooth, often the maxillary canine to erupt through the new bone. Additionally orthognathic surgery may be required if indicated to restore the skeletal and profile balance.

Paedodontist: Monitoring the dental state, with supportive dietary advice emphasising prevention and fluoride application. Also, treatment of dental diseases and repair/reshape of malformed teeth can take place.

ENT audiology/surgeon: For aural examination and hearing tests to check the child's ability to hear to facilitate communication and learning. Grommet insertion may be performed if needed.

Speech Therapist: To assess and encourage speech development and advice parents on communication problems. Cleft palate patients may have hyper-nasality, excessive nasal resonance which may be accompanied by emission of air through the nasal passageways when speaking. CLP patients can also develop speech problems unrelated to cleft such as poor language development and a stammer.

Psychologist: Psychological support for both the patient and the family through the development and engagement of strategies for coping and other psychological aids.

Prosthodontist and Restorative dentist: CLP patients might be concerned that their final dental appearance is as good as possible. Anterior dental spacing is a feature in CLP patients as teeth are usually missing in the anterior region or are unable to erupt because of lack of alveolar bone. To improve dental aesthetics and fill the spaces between teeth a Prosthodontist/Restorative dentist may prepare for prosthetic replacement of the missing teeth (Cooke, 1989).

Orthodontics: the orthodontist also plays a significant role in the management of CLP from birth until adulthood. Orthodontic treatment episodes may include: infant pre-surgical orthopaedics, early mixed dentition treatment, dento-facial orthopaedics and orthodontics, preparation for alveolar bone graft procedures and other pre surgical orthodontics including orthognathic jaw surgery (Santiago and Grayson, 2009).

Therefore, many centres have specialist craniofacial orthodontists whose practice is 'supra-specialized' in treating patients with craniofacial deformities such as CLP (McCarthy, 2009).

The goals of orthodontic treatment in CLP patients are usually similar to non-cleft orthodontic principles. That is to say: to achieve an ideal dental function, facial aesthetics and speech (Berkowitz, 2006). However, achieving this is much more difficult in CLP patients compared to non-cleft orthodontic patients. The goals according to (Berkowitz, 2006) are:

➢ Improve the relationship of the lips.
➢ Achieve harmonious balance of the dentition in the opposing jaws.
➢ Achieve favourable skeletal maxilla-mandibular jaw relationship.
➢ Achieve normal incisor overjet and overbite.
➢ Correct dental axial inclinations.
➢ Avoid the use of artificial teeth.
➢ Achieve functional dental occlusion.
➢ Achieve optimal nasal breathing.

On eruption of the permanent dentition, definitive orthodontic treatment may be provided to correct the malocclusion and align maxillary and mandibular teeth in the teenager/adolescent using fixed appliances (12+ years). Should the care pathway involve orthognathic jaw surgery to correct the occlusal relationship (in cases where the underlying skeletal discrepancy is severe), the lower arch orthodontic treatment is often delayed until growth has stopped to avoid relapse should growth continue after the completion of surgery.

Orthodontists are involved in major distinct interventions for CLP patients; the role of orthodontics has been summarised as follows (Berkowitz, 2006, Ireland and McDonald, 2003, Santiago and Grayson, 2009):

Record taking and keeping

This is important as it is essential for assessment, audit and monitoring of the treatment results. Consensus is widely agreed that records should be taken at birth, 5, 10, 15 and 20 years of age. Records should include: photographs, study models and radiographs when needed. Also, records should be taken before orthodontic intervention and casts should be prepared before constructing any devices. Records are also useful to aid patient involvement in treatment planning and deciding on different treatment plans.

Pre-surgical infant orthopaedics

This is important as sometimes it is necessary to arrange for repositioning of the segments of the cleft lip and maxilla before the surgical repair of the lip. The lip tapes and intraoral appliances help to facilitate lip closure, but the long term benefits of this technique is controversial and still not clear (Santiago and Grayson, 2009).

Correcting the anterior crossbite with removable appliance

During the early mixed dentition stage, when an anterior cross bite is identified, correction with an upper removable appliance containing springs and posterior bite plane to free the occlusion may be used to correct the incisor relationship.

Expansion of maxillary arch

During the mixed dentition stage, upper arch expansion prior to the alveolar bone graft may be involved. This can be achieved by a conventional bonded fixed brace or cemented appliance such as a tri- or quad-helix. Long term retention is necessary to maintain expansion results. Other benefits of expansion could be to improve dental function by eliminating occlusal functional shifts or to improve the nasal airway.

Reverse pull headgear

This may be considered to protract the maxilla to maintain a good jaw relationship. However, this treatment entails extensive compliance from the patient, plus it does require a long treatment time and its overall success is uncertain. The timing of the treatment is best targeted before a growth spurt.

Definitive orthodontic treatment

On eruption of the permanent dentition, the definitive orthodontic treatment is usually provided to correct the malocclusion and align the maxillary and mandibular teeth in the teenager/adolescent using fixed appliances (12+ years). Should surgery be required to correct the occlusal relationship, this should be delayed until growth has ceased in order to prevent a relapse post-surgery due to growth.

Orthognathic surgery/Distraction

Commonly the maxillary growth is deficient and a class III skeletal jaw relationship is evident. The final stages of intervention relate to the permanent dentition in preparation for orthognathic jaw surgery. In adults, when growth has stopped, a combined orthodontic plan which could include jaw surgical treatment might be needed to correct the severe malocclusion and asymmetry caused by the cleft. Orthodontic treatment may be divided into pre-surgical orthodontics and post-surgical orthodontics. By the end of the orthognathic treatment functional and aesthetic goals should be addressed. A platform for any possible final revision surgery should have been reached to enable optimum lip/nose surgical refinement.

2.2 Overview of the existing literature of the experience of cleft lip and/or palate

The review of the literature so far has focused on the CLP as a clinical entity explaining its origins, classification and its clinical management and the role the orthodontist plays in CLP treatment. It is important to understand what patients` experiences are, in relation to having the condition of CLP. This part of the review will give an overview related to three distinct areas of experiences associated with CLP. These areas are arranged into: the experiences related to psychological well-being; the experiences of having CLP on a social level; and the experiences related to the treatment pathway. The boundaries between the psychological and sociological level has sometimes been blurred in research related to CLP and is often referred to as 'psychosocial'. The author has presented the research as two different categories instead of one here to accentuate and emphasize the difference between the two approaches: psychological and sociological.

The literature was reviewed and key papers were scrutinized in relation to the CLP experience and their strength and weaknesses were assessed on an individual basis. The purpose of this is to provide a synoptic summary in addition to showing the gap that this research plans to address. This review of publications included papers which relate to the patient experience and also shows the gap in the current literature about the patient experience of the definitive orthodontic/orthognathic treatment within their CLP care pathway. Papers included can be found in the tables following its relevant section.

2.2.1 Experiences related to cleft patient psychological well being

It is important to understand the impact from a psychological view that might affect those having a CLP. There is a significant body of research which comes from a psychological perspective (Hunt et al., 2005) and there is a considerable body of work describing psychological explanations and adults' accounts of their experience of living with CLP (Table 2). This has allowed secondary analysis of findings in a number of reviews. For example Hunt et al., (2005) undertook a systematic review which included only studies that focused on children and adults with repaired (non-syndromic) CLP. They included all methods of measurement including quantitative and qualitative approaches and both self-reported and objective measures. Hunt et al., (2005) covered many of the psychological aspects of living with CLP such as: psychological functioning, self-concept, body image and facial appearance, satisfaction, speech satisfaction, behavioural problems, social functioning, anxiety and depression, attachment, development and learning. The literature in Hunt's review suggested that the psychological health of people with CLP is not greatly affected by their condition. However, some specific

problems may occur, and have been reported as dissatisfaction with facial appearance, depression and anxiety (Hunt et al., 2005). But the large variation in study designs and outcome measures in the studies and also the conflicting evidence, demonstrates that there is no definitive conclusion as to the psychological consequences of having a CLP or to being treated (and outcomes) for CLP. Hunt suggested more use of qualitative approaches for future research as this could potentially offer a greater in depth understanding.

There are a number of explanations for the conflicting reports in the literature. First, the quantitative design of the measures (i.e. questionnaires) may not enable participants to fully disclose their opinions because the issue may be sensitive. Secondly there are also methodological issues associated with the studies such as small sample size, convenience sampling, lack of social context, age range and lack of a control, which means some results lack internal and external validity. Thirdly some studies rely on proxies (i.e. Parents) rather than those with the condition. Thus the findings are seen through the lens of a non-independent observer such as a clinician or parent.

Table 2: The psychosocial effects of cleft lip and palate: a systematic review

The psychosocial effects of CLP: a systematic review

Author	Year	Focus	Sample size	Method	Country
(Hunt et al., 2005)	2005	the psychosocial impact of cleft lip and palate	64 articles identified as suitable for inclusion	Systematic review	Not applicable

Strength	Limitation
-Included both quantitative and qualitative methods. -Included self-reported, reported by other and both. -Associations between cleft type and the incidence of specific psychological problem. -The suggestion for future research to include semi-structured interviews with the patient to ensure topics relevant to CLP is included. -Psychosocial explained as: some of the studies examined a single psychological or social construct, while others addressed a range of psychosocial issues.	-The large variation in study design and outcome measures adopted by previous researchers meant that a collective examination using the meta-analysis technique was not possible. -Conflicting evidence exists in the literature when it comes to establishing whether children and adults with repaired CLP experience increased psychosocial problems as a result of their cleft. On the basis of currently available evidence, it is impossible to state the extent of a problem with any certainty. -More than half of the studies included in this review did not use a control group. -The results of the investigations included in the review do not help to identify the severity and duration of psychosocial problems among those with CLP. -It is important to determine whether problems experienced by children with CLP continue into adulthood or perhaps only emerge in adulthood.

Summary of findings/Conclusion
-Good quality systematic review that followed collected the data systematically using a proforma. The studies lack uniformity and consistency which proved difficult to draw a definitive conclusion. However, there is limited evidence that having cleft may face psychosocial problems.

2.2.2 Experiences related to having a cleft lip and/or palate from a sociological perspective

Few studies have evaluated what it is like to live with CLP addressing the sociological perspective (Hall et al., 2012, Hall et al., 2013, Patel and Ross, 2003, Tiemens et al., 2013, Alansari et al., 2013). This involves looking at the CLP not as just a condition in isolation but as a condition that exists in context and has a social impact. The social impact is affected by how people in the social world see and interpret the condition. The standard medical approach is to look at the CLP as something at variance from the normal which needs to be repaired. In contrast the sociological approach would focus on society as contributing to the problem. So, instead of changing the body, the focus is to identify and treat the social aspects that contributed to the problem. Several studies used such an approach to identify the social impact of having CLP. Only a qualitative methodology has been used to explore this area. These studies are detailed in tables 3 to 9 which examine these studies showing their strength, limitations and highlighting their key findings.

For example, a study conducted in South Africa that investigated the perception of adults with repaired cleft lip, cleft palate or both illustrates the sociological perspective (Table 7). Using semi-structured interviews, 20 adults aged 18-50 were interviewed in their homes and their views were explored. The investigators focused on quality of life as an assessment and in particular communication, education, employment, family and marital life, social life and emotional issues. An important finding was that participants reported that they had been greatly affected by negative social perceptions and especially the cultural myths related to the cause of the condition. Beliefs such as 'punishment from God' to 'sinning parents' were found to be common place. However, the interviews indicated satisfaction with educational achievement and after receiving treatment for speech problems, participants reported an improved quality of life. Also the CLP was not thought to be a problem for employment and marital relationships. The study also emphasised the positive role of the multidisciplinary team approach (Patel and Ross, 2003) and focused on some of the very sensitive areas of those who lived with CLP (e.g. relationships). There is the possibility that participants may have altered their responses to those which are more socially desirable, or which masked the depth of their feelings.

Another study in central Canada interviewed seven adolescent females at the preparation stage before their orthognathic surgery (Table 8). The aim of the study was to understand female CLP patients' experiences. Negative social behaviour in relation to their CLP was present including teasing, bullying and prolonged staring. Participants also battled to show the positive view of who they really were. They felt unable to engage in

community life because of the worry that people might perceive their CLP in a negative way. The study showed the importance of reconstructive surgery as it enhanced the affected individuals confidence as it improved their aesthetics (Tiemens et al., 2013).

In Sharif`s review (Table 3), none of the studies included were conducted in the UK which, due to the nature of the prescribed care pathway in the UK, might provide different findings due to a different culture and a different approach to the treatment pathway. This would have the potential to generate different results compared to other studies carried out in other settings. The review concluded with the confirmation of the paucity of the reporting of the experience of young people with CLP. It suggested that more attention was needed, both within families and in services to support affected individuals, to manage problems such as bullying and those related to perceived facial differences. The review called for a more qualitative approach for studies related to patients with CLP especially with younger people. The following tables are related to the part of the literature review that overviewed the sociological perspective.

There are a paucity of studies exploring the patient`s perspective using a qualitative approach. In a systematic review by Sharif et al., (2013), the authors explored the experiences of people with CLP in relation to being treated and living with the condition. Sharif et al., (2013) included only the qualitative literature that focused on the perspective of young adults with CLP. They were only able to identify two papers which fulfilled their inclusion criteria, which indicates the limited amount of qualitative literature in relation to the CLP patient`s perspective. Data from such studies could help clinicians to understand their patients better and provide a better plan of care which they cannot get without acknowledging the individual perspectives of those who are affected by the condition. These studies might also provide clinicians with a perspective on what it is like to have the condition in the first place.

The perspectives of children and young people living with cleft lip and palate: a review of qualitative literature					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Sharif et al., 2013)	2013	To explore the experiences of children and young people with CLP in relation to being treated for and living with the condition	2 articles identified as suitable for inclusion	Systematic review	Not applicable
Strength			*Limitation*		
-The first systematic review that used studies using a qualitative design. -Included self-reports together with parents reports. -Used the critical appraisal skills programme checklist to appraise quality.			-Very few studies identified that fit inclusion criteria (2 studies only). -Added 2 more studies which did not fit their inclusion criteria but deemed were useful to include.		
Summary of findings/Conclusion					
- There is a paucity of evidence in relation to experiences of young people with CLP. -More attention is needed within families and in service to help cleft individuals to manage everyday difficulties. -Further qualitative research is needed to address the dearth of published information on the cleft patients' perspective.					

Children's and adolescent's perspectives on CLP

Author	Year	Focus	Sample size	Method	Country
(Hall et al., 2013)	2013	explore children's and young people's perspectives of being born with a CLP	17	Interviews	UK

Strength	Limitation
- Acknowledged children's voices through the child centred qualitative approach. -Allowed a deeper insight into CLP and shows that young people can contribute their views and experiences about services. -Demonstrated the valuable contribution children and young people can make to health-related research. -Attempted to gain a deeper understanding of the impact of CLP and related treatment amongst young people.	-Children's lack of early memories relies on adults to inform them of life events. -Parents' experiences might have influenced their children`s experience through the transfer of values and attitudes to their children in relationship to cleft.

Summary of findings/Conclusion

-Demonstrated the valuable contribution young adults and children can provide for health research.

-Provided an insight into these main themes: Cleft and the biography, cleft and identity and constructing the body.

-Life stories tend to begin with the early stages of life as birth was considered a key event on the timelines.

-Parents were seen to play a key role in children accounts construction as the children are aware of that the condition was a significant event for parents.

-It is suggested that CLP is not necessarily experienced as causing a biographical disruption.

-Participants recounted CLP as part of their life stories but were not necessarily a constant feature.

-The severity and frequency of treatment are essential dimensions of patients' accounts.

-Adults had a major influence on children's opinion.

Table 5: Experience of teenagers born with CLP and interventions of the health nurse

Experience of teenagers born with CLP and interventions of the health nurse

Author	Year	Focus	Sample size	Method	Country
(Chapados, 2000)	2000	adolescents with CLP experience	10	Semi-structured interviews	Canada

Strength	Limitation
-Patient centred approach. -Qualitative interviews with young affected participants. -Offered an insight into the potential implications of CLP on social life and possible aspects of care.	-The location of the interviews was not clear. -Lack of supporting quotations for the themes described.

Summary of findings/Conclusion
- It gives important information about participants' need for information and support.   -The results enhanced the importance of the nurse in the care system and proposed the adding of a nurse to the multidisciplinary team.   -The perception that the child has of him/her self and the weight given to his/her cleft are the main influencers in the adaptation.

Table 6: Social life aspects of young adults with cleft lip and palate: grounded theory approach

Social life aspects of young adults with cleft lip and palate: grounded theory approach

Author	Year	Focus	Sample size	Method	Country
(Chetpakdeechit et al., 2009)	2009	subjective perceptions of young adults born with CL/P	12	Telephone interviews	Sweden

Strength	Limitation
-Patient centred approach. -Used grounded theory.	-Data from interviews with adults whose recollections of their childhood will be limited by their memory and maturational change. -Telephone interviews don't allow the researcher to see facial impressions and context may be lost.

Summary of findings/Conclusion
-People with cleft lip and palate or isolated cleft palate desire to be like everyone else.   -Speech difficulties and dissatisfaction with appearance affected their self-esteem negatively.   -Receiving recognition from a significant other will help them to increase their self-esteem.

Reflections on the cleft experience by South African adults: use of qualitative methodology					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Patel and Ross, 2003)	2002	Quality of life	20 adults	Interviews	South Africa

Strength	*Limitation*
-Qualitative interviews to explore quality of life related to cleft. -Mixed racial groups.	-There might be a degree of recall bias as the study is retrospective -The study focused on perceptions of respondents regarding very sensitive areas, it is possible that some participants may have furnished socially desirable responses -The small sample size with its disproportionate representation of various racial and socioeconomic groups precludes generalization of results. -Subjective interpretation of the open-ended responses could have affected the reliability of results. -The authors focused on quantifications of their findings.

Summary of findings/Conclusion

-The findings have important clinical implications for a multidisciplinary approach to treatment; counselling of patients and their families; education of the community.

-Participants were generally satisfied about their communication abilities and speech, educational achievement.

-Participants were badly affected by negative societal perceptions and prevailing cultural myths regarding aetiology of clefts.

-They reported that they were treated differently by parents with their siblings.

-The speech improvement affected their quality of life positively.

-Compassionate and competent care from the multidisciplinary team enhanced their quality of life.

Table 8: Living with difference: experiences of adolescent girls with cleft lip and palate

Living with difference: experiences of adolescent girls with cleft lip and palate					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Tiemens et al., 2013)	2013	To explore the experiences of females affected individuals before their orthognathic surgery	7	Qualitative method: semi-structured interviews	Canada
Strength			*Limitation*		
-The use of qualitative interviews. -The use of phenomenological approach which generate views and understanding from participants' perspective.			-Relatively small sample size. -Focused only on female patients before their orthognathic surgery, as their experience might change after the surgery which eventually could affect the overall long term experience of having cleft.		
Summary of findings/Conclusion					
-Highlights some of the challenges faced by cleft female individuals. -Girls with cleft lip and palate face stigma and negative social behaviour toward their cleft. -Coping strategies, social support and reconstructive surgery help them to face these negative perceptions.					

Table 9: Making sense of the cleft: young adults' accounts of growing up with a cleft and deviant speech

Making sense of the cleft :young adults' accounts of growing up with a cleft and deviant speech					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Havstam et al., 2011)	2011	To explore the experience of growing up with a cleft and speech problems	13	Semi structured interviews	Sweden
Strength			*Limitation*		
-Used patient centred qualitative approach. -Focus on the participants' active strategies for dealing with the cleft experience.			-Data from interviews with adults whose recollections of their childhood will be limited by memory and maturational change.		
Summary of findings/Conclusion					
-Two categories included; shaping the attitude to the cleft and managing being different, it describes the development of self-image in relation to the cleft.					

2.2.3 Experiences and outcome related to cleft lip and/or palate care

By looking at the available literature on CLP care and living with CLP, it is clear that there is a large amount of work related to these areas. There are papers that have looked at CLP care for the purpose of evaluation and/or to assure the quality of treatment (Semb et al., 2005a, Noar, 1992, Noar, 1991, Sandy et al., 1998, Oosterkamp et al., 2007, Becker et al., 2009). Tables 10 to 15 examined these studies showing their strength, limitations and their key findings.

For example, in the UK, a major national study assessed the outcomes of care in children born with unilateral CLP. Two groups of children were included aged 5 and 12 years. A total of 601 children were invited to participate. The study included 57 CLP centres and 105 consultant orthodontists were involved in treatment. Overall the standard of care and outcome were found to be unsatisfactory (Sandy et al., 1998) (Table 10). The authors advised a number of recommendations regarding the management of CLP. The number of centres delivering care to CLP patients was recommended to be reduced to between 8 and 15 centres. Also the authors recommended that fewer numbers of orthodontists needed to be involved and advocated a standardized care model. This work was primarily about standards of care from the clinician's perspective.

An area that has been addressed from the patient`s perspective is patient satisfaction. Satisfaction with some aspects of care and delivery of clinical services has been explored in addition to outcome (Noar, 1992, Noar, 1991, Sandy et al., 1998, Andersen et al., 2012). In these studies, parental satisfaction has also been measured. However, satisfaction was assessed very simply without considering how initial expectations matched the final results and how the actual experiences of care shaped their final opinion. These elements of satisfaction were assessed from the professions' perspective and most of the components which were assessed were framed by what clinicians thought a successful outcome would look like using data collection methods such as questionnaires or visual analogue scales.

Patient satisfaction is a complex multi-dimensional process which involves patients factors (expectations, patient characteristics and psychosocial determinants) and care variables (technical quality of care, accessibility, finance, outcome...etc) (Sitzia and Wood, 1997). Exploring deeper into patient expectations, three further categories of expectations can be identified: background, interaction and action (Sitzia and Wood, 1997). Background expectations are expectations accumulating from the learning during the treatment or the consultation process. Interaction expectations are those relating to the exchange (information, communication) that take place with the clinician. It is also

been suggested that memory may influence satisfaction (Misra et al., 2013). Action expectations are those regarding the actions the clinician will take such as giving advice, referral or prescribing a medication.

All of these factors and the contextual factors show how complex the term satisfaction can be (Sitzia and Wood, 1997). The expectations of treatment can be raised in a way which may not be realistic. Participants will view the outcome but they view it as a whole experience including transportation and treatment cost. Treatment can be disappointing and of a limited effectiveness for some with possibly unrealistic expectations. As a result, participants can think back and blame the lack of information, poor communication...etc if the outcome did not match their expectations (Sitzia and Wood, 1997). Satisfaction studies have tended to focus on clinical aspects of care using questionnaires.

For example Noar (1991) used a questionnaire based postal survey to investigate concerns and attitude of CLP patients and their parents attending five London hospitals. The age range was 16 to 25 years old. Four major areas were explored in the questionnaire; treatment aspects, speech and facial look, emotional and social aspects, and the perceived success of involving specialists. Generally speaking, participants were happy about their care and overall facial appearance and speech. Parents were also happy with the care their children received. For example in relation to the results of surgery, 85.7% of patients were satisfied while 80% of parents were satisfied. Another example is a study by Andersen et al. (2012) who compared patient satisfaction between two surgical approaches; maxillary advancement using conventional surgery versus advancement using distraction osteogenesis technique. They found that CLP patients showed a high level of satisfaction with regard to the functional and aesthetic outcome. However, the second group were less satisfied with the length and duration of treatment required by the osteogenesis distraction method.

Although similar type of studies can help to improve and expose the limitations within the CLP care pathway, this is limited as studies have tended to focus on the clinical perspective and predetermined issues of relevance to clinicians. In such cases, the voice of the patient is not heard because these methods which rely on questionnaires do not allow flexibility and the exploratory approach which a `one to one interview` may provide. Also some studies that have used questionnaires have a relatively small sample size which undermines the generalizability of such studies (Noar, 1991, Young et al., 2001). In addition the studies which investigate the impact of treatment and satisfaction start with the concept that patients with CLP must change to be accepted socially; this implies pre-judgement of the patient's perception of their condition.

Patient satisfaction may be influenced by the ability of patients to be involved in some treatment decisions. A study conducted in Sheffield in the UK investigated cleft children`s experiences and perspective of participation in the cleft care pathway (Table 17). It is the first qualitative study to look at the patient experience of participation in the CLP pathway. Eight boys and nine girls aged between 8-17 years participated in a qualitative study which used a child centred approach. The results produced a rich data source and an insight into how children can play their part in the treatment decision making process for different procedures involved as part of the care pathway. This study found that children may be either active or passive in the decision making process. The level of participation depended on the type of procedure involved. Children were active in decision making in some aspects of treatment which were related to cosmetics and treatment later on in the care pathway. Also, again later in the pathway, some refused surgical intervention as a result of fear while some also refused further treatment (Rhinoplasty) because they were happy with the result achieved (Hall et al., 2012).

Participation in decision making depended on the type of treatment; young participants felt able to decide to proceed with orthodontic treatment. However, when the decision was about a surgical procedure; children reported they needed much greater support (Hall et al., 2012). A possible limitation of this study was that of recall bias and the influence that parents can plant unconsciously into children`s memories which in turn could distort what was really retained in memory or immediately perceived. In addition, this study focuses only on Caucasians.

Hall et al., (2012) work was the first of its kind to explore children and young adults` experiences of the CLP pathway and which used a qualitative approach. The study provided an insight into some treatment aspects such as decision making and the degree to which children can make important decisions such as accepting or refusing a surgical procedure. It was however a small local study reflecting one provider of care. It is a good starting point for future studies to look at specific aspects of treatment in a more focused and detailed way.

A more recent study in Canada (Table 16) investigated how cleft patients perceived their treatment process and how the experiences and perceptions changed over time as they grew older (Alansari et al., 2013). The authors used a qualitative approach utilizing in depth semi structured interviews with 11 cleft patients. Alansari et al., (2013) found that there is a relationship between the self-perception and how patients perceive their treatment. With younger populations some have experienced negative circumstances in relation to their cleft such as peer stigmatization; this affected their self-perception and as a result they perceived their treatment as negative in many ways such as describing

it as a burden and painful (associated with surgical procedures). Whilst in late adolescence a different perspective appeared to be present. They experienced a dramatic drop in peer stigmatization which led to the improvement of self-perception. In this way participants started to see the benefits of treatment, and a more positive perspective of the care developed. It is possible that this finding is attributable to maturation.

When regarding orthodontic/orthognathic treatment, previous work has used a quantitative approach to assess patient expectations of their orthodontic treatment (Sayers and Newton, 2006) and patient experiences of the process/outcome of orthognathic treatment (Travess et al., 2004). However within the CLP care pathway, the literature lacks an understanding of the patients` perspective on their experiences of orthodontic treatment. So while there is an impressive volume of literature on the condition of CLP, most studies are quantitative and look at patient satisfaction, but looked at from the clinicians' point of view. There is also a considerable body of psychological literature on CLP but again it is predominantly quantitative and focuses on emotional affects and how people adjust to having the condition of CLP and the experiences of treatment but looked mostly from a psychological point of view. There is very little qualitative work and very little work has been carried out from a sociological perspective.

The strength of adopting the sociological approach is its broader coverage of the context; so it would not only look at the treatment but also at how the treatment impacted participants' lives and how they really see the treatment. Not from the perspective of a clinician but rather as a patient who has to undergo treatment within the context of daily life. Also, a sociological approach allows for exploration and flexibility to give a wider view rather than focussing on the treatment alone. Participants incorporate CLP treatments into their daily lives although the treatment is not the sole focus of their existence.

The benefit of using a sociological approach as proposed in this thesis, is that it will enable us to understand the definitive orthodontic/orthognathic treatment within the context of the care pathway and day to day life. This will illuminate issues which might be neglected using other approaches. This different approach may introduce us to some unexpected themes which could help to improve the care system or our understanding of participants and families who use the care system. The following tables are related to this part of the literature review and have been referred to earlier in the text.

Table 10: The clinical standards advisory group (CSAG) cleft lip and palate study

The Clinical Standards Advisory Group (CSAG) cleft lip and palate study

Author	Year	Focus	Sample size	Method	Country
(Sandy et al., 1998)	1998	Service evaluation	647	-Standardized records were collected from two age group (5year old-12 year old)	UK

Strength	Limitation
-Exposed the limitations within the cleft care pathway in UK. -Detailed review of clinical records and outcomes.	-It is from clinicians' perspective.

Summary of findings/Conclusion
-The outcome of cleft care is unsatisfactory. -A list of recommendations to cleft health care provided.

Questionnaire survey of attitudes and concerns of patients with cleft lip and palate and their parents

Author	Year	Focus	Sample size	Method	Country
(Noar, 1991)	1991	Treatment aspects, facial appearance and speech, social and emotional aspects of clefts, success of specialists involved	28 patients and 30 parents	Questionnaire	UK

Strength	Limitation
-No interview potential bias. -Included patients and parents.	-Followed a predetermined points which may not represent what patients think. -Relatively small sample size for a questionnaire study.

Summary of findings/Conclusion

-Mail questionnaires considered a useful and cost effective method.

-Patients were happy with overall facial appearance and speech.

-Patients were less satisfied with nose, lip, profile, smile and teeth.

-No display of significant emotional maladjustment or reduced body image.

-Patients and parents generally were happy about the care.

-Parents felt that their children were affected emotionally and socially and also their school results.

Questionnaire survey of attitudes and concerns of three professional groups involved in the cleft palate team

Author	Year	Focus	Sample size	Method	Country
(Noar, 1992)	1992	Treatment aspects, facial appearance and speech, social and emotional aspects of clefts, success of specialists involved	109	Questionnaire	UK

Strength	Limitation
-Investigated the opinions of three professional group (plastic surgeon, orthodontist and speech therapist).	-Followed predetermined points which may not represent what patients think.

Summary of findings/Conclusion

-Good agreement between the three groups.

-Professionals were not happy about results of their treatment and the overall facial appearance.

-Professionals felt that patients were emotionally and socially affected and teased for their cleft.

Satisfaction with treatment outcome in bilateral cleft lip and palate patients

Author	Year	Focus	Sample size	Method	Country
(Oosterkamp et al., 2007)	2007	assess satisfaction with facial appearance	43 affected and 43 control without cleft	Questionnaire	Netherlands

Strength	Limitation
-Control group. -Used open ended questions. -Used quantitative with qualitative approach.	-Bilateral cleft lip and palate only. -The quantitative part followed predetermined points. -Qualitative part doesn't allow for further exploration.

Summary of findings/Conclusion
- Patients were less satisfied with the appearance of the upper lip, the nose and their breathing. - Satisfaction with facial appearance affected the health related quality of life in a positive way. - Bilateral cleft lip and palate patients had considerable problems and concerns with speech, hearing and drinking.

A survey of cleft team patient experience in obtaining dental care

Author	Year	Focus	Sample size	Method	Country
(Becker et al., 2009)	2009	Dental care	171	Telephone interviews	USA

Strength	Limitation
-Interviewed parents or carers. -Explored the dimension of the type of insurance related.	-No patient account. -The title is misleading as interviews were performed with parents but not patients -Low response rate 45.7%.

Summary of findings/Conclusion
- 84.8% regular check-up rate. -Significantly different reported experience in obtaining care depending on insurance type. -Patients with public or no insurance funding schemes reported that it is more difficult to obtain care compared to those with private insurance.

Table 15: The Eurocleft study: intercenter study of treatment outcome in patients with complete cleft lip and palate. Part 1: introduction and treatment experience

The Eurocleft study: intercenter study of treatment outcome in patients with complete cleft lip and palate. Part 1: introduction and treatment experience

Author	Year	Focus	Sample size	Method	Country
(Semb et al., 2005a).	2005	To calculate the amount of treatment and associated travel experienced by five groups of patients treated at different centres	127	Standardised data collected	Northern Europe

Strength	Limitation
-Large study that covered a broad areas of service in a standardized way.	-Centres were from different countries which indicates different standard of treatment. -Centres names not mentioned.

Summary of findings/Conclusion
- It established a long-term follow-up of a cohort of patients with complete unilateral cleft lip and palate. -There is a large amount of variation regarding the amount of treatment provided by the five different teams and also the associated travel.

Living with cleft lip and palate: the treatment journey

Author	Year	Focus	Sample size	Method	Country
(Alansari et al., 2013)	2013	understand how individuals with cleft lip and palate perceive and experience their treatment process and how these perceptions and experiences change over the life course	11 adults with non-syndromic complete CLP	Qualitative in-depth semi-structured interviews	Canada

Strength	Limitation
-Used in depth semi structured interviews. - Acknowledged adult's voices through the patient centred qualitative approach. -Allowed a deeper insight into cleft lip and palate patients' experiences. -Suggestions were provided to improve the care. -About equal number of males and females participated. -Used an inductive grounded theory approach which generates data grounded in participants' responses.	-Generalizability is not applied here because the cleft care system is different from country to another country. -Memory bias.

Summary of findings/Conclusion
-Emphasized the importance of self-perception in several aspects of treatment: desire for treatment, how they perceive the burden versus benefits and the ultimate satisfaction. -Suggested that future studies to incorporate self-perception as an important variable and outcome in the treatment process.

Table 17: Children's experiences of participation in the cleft lip and palate care pathway

Children's experiences of participation in the cleft lip and palate care pathway

Author	Year	Focus	Sample size	Method	Country
(Hall et al., 2012)	2012	explore children's perspectives on their participation in the cleft lip and palate care pathway	17	Interviews	UK

Strength	Limitation
- Acknowledged children's voices through the child centred qualitative approach. -Enhanced the evidence of inclusion children in decisions regarding their treatment.	-Children's lack of early memories relies on adults to inform them of life events. -The sample participants had a wide age range. -No representation from children from ethnic minority groups. -Parents' experiences might have influenced their children`s experience through the transfer of values and attitudes to their children in relationship to cleft.

Summary of findings/Conclusion
- Some young people were tend for their parents to make the decisions regarding their treatment while others were more active in the decision making process. This is influenced by the nature of treatment complexity and severity. -Some children considered the multidisciplinary team clinic as daunting. -Enhanced the importance of undertaking research to understand children's perspective.

2.2.4 Experiences of the parents of a child with cleft lip and/or palate

Having a CLP not only affects individual patients, but affects parents/carers/guardians too (Nelson et al., 2012a). Several studies have explored the parents' perspective and experience of having a child with cleft (Noar, 1991, Young et al., 2001, Byrnes et al., 2003, Johansson and Ringsberg, 2004, Nusbaum et al., 2008, Stone et al., 2010, Nelson et al., 2012a, Nelson et al., 2012b). Tables 18 to 24 examined these studies showing their limitations, strength and their key findings.

The narrative review of the literature by Nelson et al., (2012a) summarised the present research in relation to the experiences of parents caring for a child with a CLP. It reports an expert opinion but this is considered to be at the bottom of the pyramid of hierarchy of evidence. The authors included both quantitative and qualitative approaches and compared the CLP literature with the wider literature of long term conditions. Nelson et al., (2012a) reviewed the emotional, social and service-related experiences of parents. Feelings of shock and anger, grief and worry as an early experience of having CLP babies was reported; also the need for parents to share their feelings and to get the appropriate emotional support from experienced professionals at the time of diagnosis. A raised level of reported emotional strain amongst parents of toddlers was reported, but there was also evidence that by the preschool level, emotional strain was similar to parents of children without clefts. Stigma related to CLP was also experienced, which generated discomfort and anxiety because of their child`s perceived difference (Nelson et al., 2012a).

Experiences of CLP services for parents were also highlighted in the review. Specifically, the need for information at birth and the way in which they had received the diagnosis from the healthcare professionals. The review found that satisfaction was investigated in a number of surveys by using simple approaches which did not necessarily reflect what parents really thought. Positive and negative aspects of care were identified such as poor access and coordination of services as a downside and the high level of confidence that parents placed in CLP team clinicians as an upside. Also the continuity of treatment from specialists was valuable to them.

Nelson's review demonstrated that there is variable quality in the published literature. Most studies use a cross sectional design, small sample sizes, and focus mainly on mothers. Fathers` perspectives are rarely reported upon and few studies employ a qualitative approach.

Nelson et al., (2012a) called for more research to examine the parents` perspective and needs at different stages of their children's lives and as they move through CLP treatment together with more research to address the perspective of both parents.

While the review by Nelson et al., (2012a) is not a systematic review, it reflects the diversity of evidence from different countries, different settings, different care programmes and different approaches to exploring the phenomena. Nelson`s review relies heavily on quantitative research, but as Sharif et al. (2013) noted there are very few qualitative research studies conducted on this theme. The danger of over reliance on this quantitative evidence is that the design is usually determined by what researchers and clinicians think is important. There is little opportunity for the voice of the parent (and his/her concerns) to be heard. This 'black box' approach means that only 'known knowns' are studied and new insights from parents 'unknown unknowns' have no way of emerging using a quantitative approach. Nelson et al., (2012a) and Sharif et al. (2013) recognised the limitations of current approaches and call for more research which allows the patients` and parents` voice to be heard. They strongly advocate the use of qualitative research to bridge this important gap. The work of Hall et al., (2012) , though small and confined to one care centre, illustrates the potential for this approach to produce new insights.

However, from all of these studies, there is powerful evidence that having a CLP might affect the day to day life of a patient and his/her parents/carers/guardians.

Parents' experiences of caring for a child with a CLP: a review of the literature

Author	Year	Focus	Sample size	Method	Country
(Nelson et al., 2012a)	2011	the existing quantitative and qualitative research evidence about the experiences of parents caring for a child with a cleft	57 articles identified as suitable for inclusion	Review of the literature	Not applicable

Strength	Limitation
-Included both qualitative and quantitative studies. -Appraised the included studies. -Noted the similarities with outcome with other long term conditions. -Identified the lack of research to examine parents' experiences and needs at different stages of their children's lives.	-Quantitative research in the cleft field has comprised mainly cross-sectional surveys with relatively small sample sizes focused mainly on mothers. -Little qualitative research. -Variable quality of research to date.

Summary of findings/Conclusion
-Lack of literature to assess parents' experiences and needs at different stages of their children`s lives and also as they move through the care pathway. -The need of research to know how both mothers and fathers experience having a child with cleft.

Parents' experiences of having a child with cleft lip and palate

Author	Year	Focus	Sample size	Method	Country
(Johansson and Ringsberg, 2004)	2004	Investigate parents' experiences of having a child born with a cleft lip or CLP	20 mothers and 12 fathers	interviews	Sweden

Strength	Limitation
-Used qualitative interviews approach. -Good sample size number for qualitative study. -Third researcher tested the credibility of the results. -Illustrates how the parents experience the first meeting with their child. -Documents parents' experiences of living with a child with cleft lip and palate.	-Variation in the time of experiencing living with a child with CLP; some have the experience of 5 years and some for only six weeks. This in turn will lead to different experiences. -Retrospective which rely on memory of participants

Summary of findings/Conclusion
-The study provided an insight and description of the social and medical related experiences of parents who have a child with a cleft. - Parents did not look upon their child as handicapped but as having a congenital defect.

Table 20: Parents' emotional and social experiences of caring for a child through cleft treatment

Parents' emotional and social experiences of caring for a child through cleft treatment					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Nelson et al., 2012b)	2011	Experiences of parents caring for a child through long-term treatment for CLP	35	interviews	UK

Strength	*Limitation*
-Good sample size number for qualitative study. -The qualitative interviews approach.	-Cross sectional so examining changes in parents' experiences across the treatment trajectory was not possible. -Interviews are mixed between joint and separate interviews with parents.

Summary of findings/Conclusion
-Parents experienced conflicting emotions about their child having cleft. -Parents experienced uncertainty related to aspects of treatment. -Parents experienced stigma related to their child condition. -They suggest a routine assessment of parents 'emotional and social well-being as part of the cleft care pathway.

Table 21: What information do parents of newborns with cleft lip, palate, or both want to know?

What information do parents of newborns with cleft lip, palate, or both want to know?					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Young et al., 2001)	2001	Information	40	Questionnaire	USA

Strength	*Limitation*
-The information provided in this study provides an important initial step toward identifying what information these parents want and need to hear.	-Small sample size. - A predetermined points which may not represent what patients think.

Summary of findings/Conclusion
-Parents of newly born cleft babies need basic information at that time promptly. -Information about feeding and how to recognise an illness were a prime importance for parents. -The study suggests that parents are not receiving adequate information.

<u>Table 22: Improving quality of life of children with oral clefts: perspectives of parents</u>

Improving quality of life of children with oral clefts: perspectives of parents					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Stone et al., 2010)	2010	Quality of life	20	2 Focus group	USA
Strength			*Limitation*		
-Using focus group can provide a rich exploratory approach. -Parents from these focus groups provided important information about several issue affecting their quality of life and their affected children and they suggested some enhancer of the quality of life.			-Opinions could be influenced from other participants in focus group. -The study included only parents of a child with an isolated cleft. Parent's experience of receiving diagnosis of facial cleft as part of a genetic syndrome may be different. -Fewer male (fathers) participants than female (mothers) participants. -Relatively small sample size and few sessions of focus groups.		
Summary of findings/Conclusion					
-Important issues were identified such as the early need for support for reliable information and for daily life. -Framework was provided for interventions that focus to enhance of quality of life at selected milestones.					

<u>Table 23: Parental evaluation of informing interviews for cleft lip and/or palate</u>

Parental evaluation of informing interviews for cleft lip and/or palate					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Byrnes et al., 2003)	2003	information	98 parents	Questionnaire	USA
Strength			*Limitation*		
-Explored the information theme in relation to cleft. -Good sample size. -Questionnaire through emails which allowed recruiting good number of participants.			-Sample mostly one ethnicity. -Participants 99% female (mother). -Informing interviews are influenced by time and experience due to the retrospective approach of the study.		
Summary of findings/Conclusion					
-Parents were not happy with many aspects related to informing professionals during interviews. Also the study suggests ways to improve the information gathered from interviews.					

Table 24: A qualitative description of receiving a diagnosis of clefting in the prenatal or postnatal period

A qualitative description of receiving a diagnosis of clefting in the prenatal or postnatal period					
Author	*Year*	*Focus*	*Sample size*	*Method*	*Country*
(Nusbaum et al., 2008)	2008	the experience of receiving a diagnosis of clefting in the prenatal or postnatal period	20 parents	interviews	USA
Strength			*Limitation*		
-Used qualitative interviews. -Comparative study.			-The study included only parents of a child with an isolated cleft. Parent's experience of receiving diagnosis of facial cleft as part of a genetic syndrome may be different. -Fewer male (fathers) participants than female (mothers) participants.		
Summary of findings/Conclusion					
- The diagnosis should be made with sensitivity to the parents, and additional information should be available, beside the referrals to a multidisciplinary cleft centre. -Parents who received the diagnosis before birth were happy to know at that time.					

2.3 Understanding cleft lip and/or palate as a long term condition

The review of the literature suggests that there is a considerable body exploring CLP from a psychological perspective focusing particularly on the emotional ramifications of living with, or having a child born with a CLP. As mentioned earlier, there is also a body of literature looking at satisfaction from the point of view of the CLP team professionals. Very few studies look at the experiences of CLP from a sociological perspective and few studies make use of qualitative research methods to explore the area. One possible way of exploring the experiences of young people with CLP from this perspective is to draw on the wider literature and on the experiences of patients living with other long-term conditions. This approach gives a way of exploring CLP in context.

Long term conditions extend over a prolonged period of time and affect multiple areas of day to day life. The Department of Health defines long term conditions as "a condition that cannot be cured but can be managed through medication and/or therapy" (Health)(DH, May 2012). Long-term conditions can affect any system in the human body, can be fatal or not and can be stable or progressive. Most long-term conditions are not, at the current time, curable and so medical interventions are focused on symptomatic treatment and helping people to live with these conditions on a day to day basis (Scambler, 2008). Whilst CLP is not a disease or illness, it is a condition which lasts over a life time, cannot be cured and may impact on functioning, self-esteem and quality of life in the long term (Hunt et al., 2005).

CLP is a congenital facial deformity that people live with from birth until adulthood, it does not progress with time but nor does it disappear. The effects of the condition generally improve with time (i.e. appearance and speech) as the affected person goes through complex treatment. However, CLP patients may be functionally affected for a long time until speech is corrected. Affected young people and their families live for an extended period of time with the consequences of dealing with this condition. Nelson et al., (2012a) noted the similarity between the CLP literature and the long term condition literature in relation to emotional, social and service related experiences. For example, the recognition of the early emotional and social support at the time of the child`s diagnosis. It could be reasonably expected that they would share some of the experiences of those who live with other long term conditions, as explored within the sociology of long term conditions.

2.3.1 The sociology of long term conditions

Within the sociological study of chronic or long-term conditions there is a vast body of work exploring the experiences of people living with a range of conditions (Scambler, 2012). The evidence suggests that long-term conditions affect multiple areas of day to day life and affect both the individual and their family and friends.

Empirical work also suggests that people living with long-term conditions can experience a range of social impacts stemming directly or indirectly from their condition. These include unemployment, social isolation, lower career prospects and estrangement in their social life. In some cases, coping or dealing with the symptoms of a long-term condition may take up considerable time each day, and may impact on the family and friends of the affected person as well as on the individual themselves.

The sociology of long-term conditions has developed almost as a challenge to traditional biomedical approaches to acute illness and the treatment of 'ill' people. This is based on the idea that people living with these types of condition will remain 'ill' for an extended period of time if not indefinitely (Scambler, 2012) and that the main focus of their attention, and that of the clinicians providing care for them, becomes living with, or managing, the condition on a daily basis rather than seeking a cure *per se*. Understanding the lived experiences of people living with long-term conditions gives clinicians and others involved in the care pathway an insight into the impact of the condition, and its treatment, on participants.

The sociological study of long-term conditions spans 60 years and within that time a range of theoretical approaches have been used to inform empirical research. Conflict theorists have focused on the power relationships inherent in the doctor/patient relationships whilst functionalists explored the societal implications of the sick-role and its corresponding rights and responsibilities. The studies that are most relevant to this work, however, are interpretivist in approach and have focused on the ways that people understand health and illness and its impact on their daily lives (Scambler, 2012).

2.3.2 Major themes of research in long term condition

Interpretivist work has explored the lived experiences of people with a range of long-term conditions, focusing on the ways in which they make sense of the symptoms and the label attached to it. For example, Robinson (1988) and the implications of the condition on daily life. On exploration of the (already significant) body of empirical research exploring long-term conditions from a sociological perspective, Conrad (1987) developed a list of key themes which impact on the lives of people living with a range of long-term conditions. These themes are: uncertainty; stigma; family relations; biographical disruption and reconstitution of self; information, awareness and sharing and managing medical regimes (Scambler, 2008). It is proposed these themes can be used to help shape a contextualised understanding of CLP from the perspective of young people living with the condition. The main implications of each theme are outlined below along with their potential relevance to the study of CLP.

2.3.2.1 Uncertainty

One of the key themes to emerge from studies across a range of long term conditions is uncertainty. In many cases the uncertainty proceeds diagnosis and continues throughout the course of the condition. Initial symptoms, (for example, when looking at multiple sclerosis), of long-term conditions, are non-specific and it may take months, or even years, to reach a diagnosis (Scambler, 2008, Robinson, 1988). With multiple sclerosis the delay between the first appearance of symptoms and the final diagnosis can be as long as 15 years (Robinson, 1988, Mullins et al., 2001).

Uncertainty goes beyond diagnosis, however, and symptom onset, degree and impact of individual symptoms, along with the overall disease trajectory, may vary even between people with the same diagnosis. For some, symptoms fluctuate on a daily, or even hourly, basis. For example, in his early study on the experiences of people living with rheumatoid arthritis, Bury (1988) found that participants were unable to plan events in advance as they were not sure how much pain they would be in at any given time. Further, he found that symptoms were often compounded by external factors such as the weather.

Cleft is routinely diagnosed prenatally now in the UK with the use of high resolution ultrasound as a routine procedure in pregnancy screening. 3D ultrasound has been proven to be a very effective diagnostic technique to diagnose cleft prenatally and almost eliminate uncertainty about diagnosis (Chen et al., 2001).

Whilst there is little uncertainty around diagnosis now for most parents (at least in developed countries) in relation to their children's CLP, uncertainty may still be a factor throughout the subsequent phases. Uncertainty may remain in relation to what a diagnosis of CLP means, how it will affect the child (functionally, aesthetically and socially) and what will be involved in both the treatment for, and living with CLP.

Children born with CLP are born with an obvious facial difference and grow up with the knowledge of that difference. Uncertainty here may be related to treatment options, outcomes, and the functioning ability after the treatment, facial aesthetics post intervention and the impact of all of these things on family life. In a qualitative study that used interviews with parents of children with CLP to explore emotional and social experiences across the treatment regimen; one of the main themes that emerged was uncertainty associated with the extent of impairment. This included its causes, treatment type, duration, extent and treatment outcome (Nelson et al., 2012b). Uncertainty also can be related to different aspects of participants understanding of their care.

2.3.2.2 Stigma

Another theme that has emerged from long term condition studies is stigma. A definition of stigma produced by Goffman is "the process by which the reaction of others spoils normal identity". He identified three different forms of stigma: a form related to mental illness fact, another form related to physical form of deformity or unpleasant difference, and the third form which is associated with a particular ethnicity or race, religion and belief (Goffman, 1986). It is also useful to bear in mind that attributes or characteristics of social stigma can be different in different parts of the world.

Social stigma is generated when people are labelled as 'different' and experience negative discrimination. A response which is not positive from the outside world may lead to this. When the affected person thinks, feels or behaves in a way that is related to their physical appearance or is a result of their physical appearance then thus is termed 'body image attitudes' (Sousa et al., 2009). Social activities such as establishing relationships at different life stages are significantly affected by physical attractiveness. It is possibly the relationship between the beauty of external shape and look of the body and social acceptance may affect the person with CLP (Sousa et al., 2009).

One study showed that having a colostomy alongside a stigmatized condition (cancer) was 'overstressing' (MacDonald and Anderson, 1984). Another study was conducted looking at adolescents with acquired and congenital facial differences and their parents,

to explain their stigma experiences. The study found that stigma experiences were reported frequently by adolescents with facial differences and their reported experiences were similar to those reported by parents although reported independently. The authors suggested there was a need to reduce discrimination and prejudice in relation to facial difference (Strauss et al., 2007). Nelson et al. (2012b) also suggest that there is evidence of social exclusion of children with CLP and the family, indicating the presence of stigma. Given this evidence, it would be interesting to know if there is any stigma associated with the treatment care pathway for CLP patients and this proposed study has the potential to understand this aspect.

2.3.2.3 Family relations

Another theme of the research on long term illnesses is how family relations are affected. Living with a long term condition can be resource consuming. It is a burden on the affected person and their family or carer. Care and support is needed by CLP patients and that can be a strain on their family. The family may be affected financially, travelling to the hospital or the treatment centre can be exhausting especially if the family live far away and their journey time is long. In a study in North Carolina state, the authors found that almost half of respondents to a mail/phone survey travelled more than one hour to access CLP care (Cassell et al., 2013). In many cases, getting time off from work for patients or parents can be difficult. This is especially so if they have no one to replace them at work and these appointments may also impact parents' commitments to other children or to other family members. Sometimes the affected individual might need extra treatment or special equipment and this could lead to an increased financial burden on the family.

The family can be affected by the transfer of stigma from the affected family member. In a qualitative study that examined the courtesy stigma for the family members of affected persons with Alzheimer's disease; the results showed that carers or members of the family had experienced courtesy stigma (MacRae, 1999). Baker et al. (2009) suggests that there are psychosocial compensatory mechanisms which develop within the family environment of CLP individuals.

Conflicting emotions through participation in CLP treatment pathways have been noted (Nelson et al., 2012b). Parents tend to overcome and resolve the emotional tension they experienced by adhering closely to treatments recommended by CLP team specialists. By doing this they reported they felt secure and were relieved. This emphasises the importance of family support within the CLP care pathway.

2.3.2.4 Reconstitution of self and psychosocial effects of cleft lip and/or palate

The biographical work and the reconstitution of self is another theme. All long term illnesses can pose a threat to identity (Bury, 1988). The receipt of a diagnosis calls into question the past, future and present which might cause a disruption into personal biography and self-concept. If we think about our life story as a narrative story which is partially written but not yet complete. The past which is passed and recorded as vague or explicit memories, the future which incorporates our life objectives, dreams and hopes. The present which is still constructing and developing and could alter our perception about our past or future. Biographical disruption means rethinking the identity to incorporate the diagnosed illness, so the biography is reorganized in order to account for the onset of illness. Thus the past is reinterpreted in line with what we know or in line with the diagnosis given.

When diagnosis or a label is received, the way people think about themselves changes. So for example: in epilepsy, it was found that when people were given the diagnosis of epilepsy they became 'an epileptic' so their identity changed and their diagnosis became the first part of their identity. It caused them to rethink how they saw themselves and how they behaved and where they were going and what they thought they would do in the future (Iphofen, 1990).

There are other studies of people living with multiple sclerosis (MS) which have shown a similar concept (Boeije et al., 2002). Once people got their diagnosis, they looked back over how things had been in their past and they started to reinterpret those according to their diagnosis. Individuals avoided socializing because of the MS and anticipated avoiding activities in the future because of the limitation associated. So a chronic condition disrupts and can change the way that patients understand themselves.

This concept of reconstitution of the self may be relevant to the CLP groups because participants' expectations and understanding of their identity could be changed by receiving treatment. What are they going to look like and what is their future going to be like, and how participants' identities are affected by facial appearance and how society treats them.

In the literature, it has been acknowledged that living with a visibly different face as a result of disease or injury is not easy. Deformities in the face have been significantly correlated with a negative social perception. In a study that compared the 'normal' face to the 'abnormal' face judged by adults, traits evaluated were honesty, trustworthiness and employability. The 'abnormal' face scored less in all of these characteristics (Rankin and Borah, 2003).

It is known that the acceptance of society can depend on the individual`s physical look, this relationship between social acceptability and outside beauty may help to explain the difficulties experienced by affected people with CLP (Sousa et al., 2009).

Turner et al., (1997) carried out a study to show the value of identifying psychological outcome. They explored the psychological status of CLP patients and their parents. They used a standardized interview to assess satisfaction with treatment. Their results indicated a high satisfaction level with treatment but also 73% of patients reported that self-confidence is highly affected as a result of their CLP. Around 60% of participants were teased because their cleft and cleft related features. A good number also expressed that they were not included in decisions regarding their treatment.

For individuals with a CLP, some particular problems may take place, it is been reported that some difficulties have been associated with satisfaction with facial appearance, depression and anxiety (Hunt et al., 2005). In one study, Kappsimon (1986) compared the self-concept of school children aged 5 to 9 years old. They found that children with cleft lip and palate had a self-concept significantly lower in relation to their controls. Moreover, it is reported in children and adults that the level of confidence of cleft patients has been affected because of the cleft (Noar, 1991, Turner et al., 1997). CLP individuals are reported to be twice as likely to be exposed to anxiety and depression compared with non CLP controls (Ramstad et al., 1995a). Social functioning issues have been reported from the professionals team care perspective, indicating that many cleft patients are socially affected (Noar, 1992). Also research suggests that children with clefts may have fewer friends and relations than children not affected with cleft (Noar, 1991, Ramstad et al., 1995b). It is also reported that cleft patients are less likely to marry and should they do so, they tend to marry later in life (Ramstad et al., 1995b).

This conflict in evidence suggests that individuals with CLP may or may not face psychosocial problems which will lead to issues when identifying self. One of this study`s area of interests is to understand what effects orthodontic treatment has on the affected individual`s day to day life and whether the results of orthodontic treatment impact positively or negatively.

2.3.2.5 Information, awareness and sharing

Another important theme is the work on information. For all patients with long term conditions, information is an invaluable resource. They need to know about the illness they are experiencing; What it is?; How will it progress? What are the options for treatment? How can they reduce its symptoms? Information can help an individual to understand the illness and allow them to develop a plan for managing the illness during everyday life (Scambler, 2008). Information is considered as an invaluable resource. The more information patients have, the less uncertainty occurs. It helps the patient to understand the illness and allows for developing effective and efficient strategies to manage the illness in everyday life. It provides the affected people with a sense of control over their illness as it holds the basis of action (Scambler, 2008).

People with long term conditions may have different types of relationship with their physicians. People are becoming/developing into relative experts in their individual condition. People with long term condition over time gather and accumulate a significant volume of information by themselves about the condition and become very informed. They often know a lot more about their condition than the average patient presenting for the first time with an acute condition. They may know what they want to find out and often they know about different treatment options that are available and they want to find out from their doctors if these are appropriate for them. People living with a long term condition might come prepared with the information that they want to get from their clinicians. Information and awareness is about more than just giving of information to patients. Do they get enough information or if they have received the information in the right way?

Research shows that people with long term illness are less satisfied with their relationship with doctors (Fitzpatrick, 1990, MacDonald and Anderson, 1984) and are dissatisfied about the amount of information provided by their doctors (MacDonald and Anderson, 1984).

In relation to the cleft condition, most studies have a particular interest in the patient`s information experiences and needs. It is a traumatic experience for parents to discover that their baby has a cleft. So information should be available to parents at this time. A lack of ready information at diagnosis was reported by many parents (Young et al., 2001). In a retrospective study to explore what information was critical to parents of a newly born cleft baby; the authors concluded that parents need to know about basic information, particularly information related to feeding and recognizing illness (Young et al., 2001). Also, parents prefer to get the information verbally at this time from a specialist

and one study suggested that carers should adapt a better way of informing interviews with parents (Byrnes et al., 2003).

Another study has looked at parents` experience of the timing on receiving a diagnosis of cleft, during the prenatal and postnatal period (Nusbaum et al., 2008). Using a qualitative methodology, researchers interviewed 20 parents of CLP patients with a variety of different types of clefts. In the prenatal group, participants were happy that they have learned about the CLP before giving birth to the child. On the other hand, some participants from the postnatal group preferred to have known about the diagnosis prenatally. The authors concluded that the diagnosis of CLP should be performed with respect and with sensitivity to parents. Information about CLP should be available at the time and referral to a CLP centre should be arranged. Also, they concluded that it is better to prepare parents and let them know about the possibility of identifying abnormality before undergoing the ultrasound. Generally speaking, the way parents received the diagnosis was more significant to them than the timing. While knowing about the CLP diagnosis affected the pregnancy period negatively, parents were satisfied to know about the CLP before labour and delivery.

In long term conditions, people sharing experiences and ideas with other people that have the same condition is helpful. They want to share their experiences with the people that really understand and no one understands the condition better than those who already have it. The Cleft Lip and Palate Association (CLAPA) is a UK based voluntary organization that supports people living in UK with CLP and their family. They provide information and social support. For example, they arrange for teenagers with CLP to attend confidence building camps which aim to help them to improve self-confidence (CLAPA). Such organizations provide, support, create and enhance the environment of sharing the experience of having the illness. Some CLP centres in the UK also run what they call a 'school change' day where CLP patients are invited and meet each other.

In relation to their orthodontic treatment, the literature is lacking information on the opinions of patients about the extent and quality of information they received before and during the orthodontic phase of their treatment or information related to different aspects of treatment. While previous work has focused on information to parents on early diagnosis, this study will explore participants' information needs within the care pathway and in particular around the definitive orthodontic/orthognathic treatment and care.

2.3.2.6 Managing the medical regimen

The last theme is managing treatment and the impact of this on everyday life. The subject of treatment and its impact is an important one. While professionals of a care team might have their own objective way of assessing the success of the treatment, it is important to understand how the patient experienced the treatments they have received. Patients must learn how to manage their treatment so as to control symptoms during everyday life. With a lot of long term conditions the things that patients have to do in order to treat or manage their condition actually take over some specific aspects of their lives. For example, people with diabetes alter their diet content and also eating patterns. They may only eat at certain times to control their blood glucose level. Treatment forces them to build everything else around it. This can be time consuming for such patients.

Treatment can be part of the problem as well as part of the solution. Treatment may place a burden on individuals and the family as well (Bury, 1991). People with long term illness learn to manage their symptoms and their complex medical regimen within their everyday life, and the time they spend to do so cannot be spent on other, perhaps more enjoyable things (Locker, 2008).

Abed Al Jawad et al., (2011) noted the enforced change of diet as a result of having a brace for normal orthodontic treatment. Drawing a parallel in CLP, we may ask how people changed their eating habits as a result of wearing a brace, how oral hygiene, types of food, being embarrassed to eat in public or taking times off from school to attend their appointments and having a brace impacted their lives.

By exploring the experiences related to the long term condition literature, it allows us to focus on the impact of CLP on daily life and will potentially give a greater understanding of the relationship between the patients, the condition, the care pathway, clinical outcomes and patient satisfaction. And by viewing CLP as a long term condition and trying to compare it with the long term condition literature it will give us insight about and more understanding of CLP patient, a perspective not presented previously in the literature in a comprehensive way.

2.4 Conclusion

What we see from this literature, is that a considerable amount is known about the treatment of CLP and there is a very clear well established care pathway. We know from a psychological perspective about what it is like to live with CLP and some of the impacts CLP can have on individual and their family. There remains very little qualitative work in this area and this study seeks to address this. What is also missing is an in depth understanding of the definitive orthodontic/orthognathic treatment from a patient and parent perspective. We want to look at orthodontics/orthognathic within the CLP care pathway; because orthodontic treatment does not come as separate independent stage which has no link with the pathway, but comes as part of the package provided by the CLP care system.

This study seeks to look at orthodontic treatment for CLP patients within the wider contexts of the CLP care pathway and their daily lives.

3. RESEARCH AIM AND OBJECTIVES

Sharif et al. (2013) identified the paucity of qualitative evidence in their review in relation to the experience of people living with CLP. This also includes the experiences and perception of the long term treatment of CLP care. Understanding the perspective of people living with CLP may help us to understand issues related to treatment such as adherence and satisfaction etc, in addition to an alignment with the concept of patient centred care.

CLP care in the UK was restructured after the damning report of The Clinical Standards Advisory Group (Sandy et al., 1998). Understanding of the participant's perspective and patient experience of the newly centralized CLP services is lacking in the literature. Qualitative studies which involved interviewing parents or carers are present in the literature (Nelson et al., 2012a, Nelson et al., 2012b) though far fewer studies involved children participants (Hall et al., 2012, Hall et al., 2013).

The focus on the experience of the definitive orthodontic and/or orthognathic treatment is justified for several reasons. First, it is placed at the end or near the end of the CLP care pathway therefore participants at that stage will have experienced a great deal of the CLP care pathway and will be able to comment on the whole pathway. Another reason is that orthodontic/orthognathic treatment involves the major contact hours between participants and clinicians as treatment can last up to 3 or 4 years with regular visits. Furthermore, there is a gap in the literature about the CLP participant's experiences of their orthodontic/orthognathic treatment within the care pathway and within their day to day life.

Little or nothing is known about affected young people`s experiences of the orthodontic/orthognathic treatment as part of CLP care and as part of this wider context. A better understanding of the experiences of people living with CLP about their treatment process may enable clinicians to plan better care and to enable the provision of more patient-centred care which is likely to improve patient satisfaction. Although measuring clinical outcome objectively can give an idea of how successful the treatment is clinically, assessing patients` perceptions of care within the wider context will enable members of the multidisciplinary team involved in the care of this group to understand better the non-clinical impact of the condition, and of the care pathway, on their patients.

3.1 Aim

The aim of this research is to undertake a qualitative study to explore late adolescent`s and adults' with CLP perspectives about their CLP care within the context of living with CLP and with a focus on their definitive orthodontic/orthognathic treatment. The areas to be explored in this study are the impact of the treatment on a day to day basis, expectations and experiences of the care pathway and the definitive orthodontic/orthognathic treatment.

3.2 Research questions

The research questions are:

- What is the experience of living with CLP and what impact does treatment have on the daily lives of patients living with CLP?

- How do participants perceive/experience the care pathway with a focus on the definitive orthodontic/orthognathic treatment?

3.3 The research objectives

The research objectives are:

- To conduct in-depth semi-structured qualitative interviews to CLP patients using a purposive sampling strategy.

- To answer the research questions by conducting a comprehensive data analysis after transcription.

- To report a detailed description and analysis of the data.

- To use the findings of the study to make recommendations about how to improve the care pathway in light of the experiences of those within it.

4. METHODOLOGY AND METHODS

This study was designed to address the research questions and objectives that were presented in the previous chapter. In this chapter the rationale for using this methodology will be discussed as well as an explanation of the steps and procedures involved. The chapter will incorporate reviewing the protocol, describing the procedures for obtaining ethical approval, the recruitment process, data collection and data analysis. The chapter begins by presenting a justification for the research approach used.

4.1 Justifications for the research approach and study design

We know that there is a large body of quantitative research in this area and the scarcity of qualitative research has been identified (Sharif et al., 2013, Hunt et al., 2005). Qualitative approaches are common in social science and psychological research and in recent decades, qualitative methods are becoming more popular in health related research (Pope and Mays, 2006).

Qualitative interviews allow for an understanding of the patient perspective in depth and with flexibility to probe into new areas. This flexibility cannot happen with quantitative methods such as using questionnaires. In orthodontics in particular, the use of qualitative research methods is increasing in popularity and many studies use a qualitative methods for data collection (Travess et al., 2004, Ryan et al., 2009b, Lee et al., 2011, Masood et al., 2011, Abed Al Jawad et al., 2011).

Qualitative research

To be able to explore the experiences and the impact of the orthodontic treatment for CLP patients, a qualitative research inquiry is needed. There are many different definitions of qualitative research that are accepted in the literature. A straightforward definition was provided which draws attention to the difference between qualitative and quantitative. It is stated '' *By the term 'qualitative research' we mean any type of research that produces findings not arrived at by statistical procedures or other means of quantification*' (Strauss and Corbin, 1998).

Ritchie and Lewis (2011) highlighted some key elements that shape qualitative research`s distinct character. According to them, qualitative research provides an in depth understanding and interpretation of the research participants social world by

learning about participants experiences and perspectives. The sample size is small and purposively selected (as will be explained later under the sampling section), methods of data collection provide flexibility which allow for emergence of data and involve close contact with participants, the results data should be very complete and provide thick, detailed and rich information. The data analysis is open to new ideas and concepts.

Many developments in qualitative research have occurred as it has evolved. As a result, there are several approaches and traditions which have emerged in different schools (Ritchie and Lewis, 2011).

For example, narrative research, phenomenological research, grounded theory, ethnography and case studies. In the present study, the phenomenological approach was adopted.

Phenomenology is where the researcher describes the data or the experiences of the lived phenomena as described by participants. It is focused on the perspective of the first person and this coincides closely with the study aims and objectives (Ritchie and Lewis, 2011, Creswell, 2014). In the present study, the phenomenon is the experience of treatment, specifically of the orthodontic treatment for CLP patients.

The functions of qualitative research can be 'contextual' which can be called 'descriptive', 'explanatory',' evaluative' or 'generative'. Contextual is concerned with identifying and describing what is existing by displaying the phenomena from the participants' perspective. Explanatory is concerned with answering the why questions in relation to the phenomena under study by understanding the reasons and associations that exist. Evaluative is concerned about how good the work or the system is functioning by collecting information about the process and the outcome. Generative is when the research provide new ideas or recommendations or aiding to develop or modify a theory. The present research is not purely focussing on one function of qualitative research, however, the results will have a combination of all of these functions at different levels.

Using a solely qualitative approach is needed when the subject is complex, ill-defined or new. These features apply to CLP patient experiences of their definitive orthodontic treatment contextualised within the CLP care and their day to day life. This multi-layered subject provides the necessity to adopt a qualitative approach.

In qualitative research, there are a range of methods or tool-kits that are used to collect the data. Generally speaking, the data can be divided into naturally occurring data and generated data. On the one hand, naturally occurring data is needed when we need to study the phenomena in its' own natural settings such as the study of behaviours or

interactions. Some examples of naturally occurring data collection methods include observation, document analysis or conversation analysis. Conversely, generated data is needed when we need to understand the experience, attitude, or event from the participant's perspective. The two most commonly used data collection methods are individual interviews (one to one interview) and focus groups (group discussion).

In focus groups, data are generated and participants can share and discuss their experiences and views through interaction. The idea of a focus group is to synergise the group by working together to generate the data (Stewart and Shamdasi, 1990).

The most commonly used data collection method in qualitative research is in-depth interviews. In the health care setting, it is the most commonly used method (Pope and Mays, 2006). Interviews can be defined as a 'conversation with purpose' (Webb and Webb, 1932). A topic guide is necessary before conducting the interviews. Interviews have the substantial strength of combining structure with flexibility and allowing probing to gain depth. Interviews can be face to face or through telephone or other network communication methods such as Skype or FaceTime. The interviewer should encourage the participant to speak by starting with initial questions that requires surface level initial responses from the participant. Then the interviewer can probe for further depth and clarifications (Ritchie and Lewis, 2011). Focus groups have strength when encouraging a shy member to speak (Trulsson et al., 2002) but it has been suggested by Ritchie and Lewis (2011) that one to one interviews are more suitable for sensitive topics.

This study was conducted using semi-structured interviews with participants to explore participants' perspective of CLP and care they had received. Adopting this data collection method assisted in obtaining a deeper understanding of the participants' perspectives as there is the flexibility for participants to shape the interviews and express the experiences in their own words, This is not possible using solely quantitative methods (Ritchie and Lewis, 2011). One of the functions of qualitative research is explanatory. It is concerned with human experience and can facilitate examining subjects in depth and answering questions that explain what lies behind the patients` perspectives and why (Ritchie and Lewis, 2011, Draper, 2004).

Using face to face interviews in this qualitative study will allow the researcher to investigate the complex experiences of participants. By combining structure with flexibility it will allow the researcher to use a range of probes and other techniques to achieve depth and detail in terms of the investigation of data, exploration and explanation. This might not be possible when using focus groups. As this study is centred on in depth data acquisition and exploration/explanation, one to one interviews is the

most appropriate approach (Ritchie and Lewis, 2011). Interviews also provide a more relaxed atmosphere for participants (Travess et al., 2004). The disadvantage of using interviews is that the researcher is relying on the participants' memory and there will be different levels of recall among participants. Interviews are time consuming; arranging, conducting, transcribing and analysing them can be an exhausting process.

Using a qualitative interviews method has its limitations. Limitations are discussed in the discussion chapter.

4.2 Methods

This qualitative study has used semi-structured in-depth interviews to explore the experiences of people with CLP who have undergone their definitive orthodontic/orthognathic treatment. This section will explain how the topic guide was generated, the researcher interview practice, participant identification, consent procedures and interviewing.

4.2.1 Reviewing the protocol

This study was carried out by a postgraduate research PhD student (RA). All of the student's three supervisors reviewed the research protocol and have provided support in developing it. In addition, the protocol was formally peer reviewed at the Unit of Social and Behavioural Sciences at Kings College London-The Dental Institute (Peer review research meeting on the 22nd May 2012).

4.2.2 Ethical approval

As this research involved human participants, ethical approval for the study was sought and granted from National Research and Ethics Service Committee in London- City and East, REC reference number 12/LO/1022 (please see Appendix).

Research and Development approval was granted from both research sites R&D Department.

Ethical issues related to this study can be summarised as follows:

Confidentiality and Anonymity

Participants' responses would remain anonymous and confidential. Participants were assured of this. No individual would be identified in any reports written.

Right of Withdrawal

All participants were invited to participate with a full description of the study. This included establishing that choosing not to take part or expressing negative views in interviews would not in any way affect future treatment. The researcher explained to participants that they could consider withdrawal from the research process at any time without any

impact on their care. They were reassured of this from the start of the study and in all information provided (both written and verbal).

Sensitive topic

The topic of this study had the potential of raising some sensitive, embarrassing or upsetting topics. Participants could have felt embarrassed or uncomfortable about discussing their negative experiences of dental care.

As a result, it was emphasized that participation was voluntary, anonymous and confidential to all participants. Participants were reassured that all their data would be anonymised and it would not be possible to identify individuals from any published reports.

Benefits and risks to participants

Participants would not experience a direct benefit. However, the findings from this study may lead to greater future understanding of the patient's and parent's perspective on their experiences of orthodontic treatment. It could lead to improvements in care of patients undergoing CLP repair and greater patient satisfaction. There were no serious risks associated with this study. The researcher was aware that if the topic caused any distress to a participant during the interview, they would be reassured and any necessary follow up help identified. However, all interviews were successfully run with no apparent distress associated.

Data storage

All transcribed interviews were kept in a secure locked storage facility in King`s College London. No names were used on transcribed interviews. Only codes were used to identify transcripts which were traceable to participants. Data were completely anonymized and stored on password protected encrypted (NHS and trust standards of encryption) computers. Only the research team had access to the material.

4.2.3 Topic guide

In any research using interviews, a topic guide is essential to act as an interview agenda. In this study, the interviews followed an interview schedule (topic guide) which ensured the key areas were covered but had the flexibility to allow participants to explore and explain their experiences in their own words. The topic guide was a written document which was created to be used during the interview process. It was generated after

61

considering the research questions and incorporating the opinions of all supervisors and the opinion of the lead orthodontist in the CLP centre. It was flexible to adjust to the new themes that were raised during the interviews. As this study did not do a pilot study as such, but the practice interviews and first few interviews were acting as a pilot study. After the interview training and the first few interviews of the study, the topic guide was reviewed in terms of sequence and phrasing of questions to encourage more open ended questions. Also, if a new theme/concept was raised, this theme was added on the side of the original topic guide and used as a prompt in future interviews. The original topic guide was used throughout the study (see appendix). The topic guide started with a general rapport question such as (can you tell me about yourself or what do you do?). Then moving to broader questions such as (what do you know about the CLP or how is it living with CLP?). The purpose of beginning with general questions was to encourage the participant to start speaking. Then a more focused question related to the CLP treatment was asked and finally questions about the orthodontic treatment experience followed in order to concentrate on the specific aspects of CLP care.

4.2.4 Interview techniques training and practice

The main researcher (RA) underwent significant and targeted training. He attended several courses in well recognised schools in UK for training on how to conduct qualitative interviews. These courses were intensive, comprehensive and designed to train professional researchers on how to conduct appropriate and effective qualitative research interviews. The researcher also carried out several practice interviews with nurses and colleagues. These practice sessions took place under the supervision of an experienced qualitative researcher and the researcher received 30-45 minutes evaluation feedback following the practice interview. The advice included how to explore and use probing, how to use open ended questions and to be less leading of the participant. The researcher gained the skills required and assessed by the supervisors, as sufficiently skilled to conduct the interview without supervision. The topic guide was useful to act as an aide memoir of the areas to be questioned. Minor amendments were added to the topic guide following the practice interviews.

4.2.5 Sample strategy and sample size selection

As this was a qualitative study, the sampling did not aim to provide a sample that was statistically representative of the general population. We needed to study and explore the dimensions of phenomena. As a result, the preferred sampling strategy was planned non-probability sampling as the aim was not to know the distribution or the prevalence of the condition or phenomena, but to understand the phenomena from the affected participants.

Several sampling strategies may be used in qualitative research. For example: purposive sampling, theoretical sampling, convenience sampling and snow ball sampling (Ritchie and Lewis, 2011).

This study used a purposive sampling strategy in which the sample criteria were prescribed. This was deliberately non-random sampling, which aimed to sample a group of people with particular characteristics, where respondents were selected because they have knowledge that is valuable to the research questions. Table 2 presents the inclusion/exclusion criteria. Patients who recently finished their orthodontic/orthognathic treatment (Maximum 18 months post treatment) or were about to finish were invited to participate. The definitive orthodontic/orthognathic treatment is placed at the final stages of CLP care therefore participants could reflect and could provide personal accounts about their overall care.

Ritchie and Lewis (2011) stated that sample size in qualitative research is usually small because of three reasons. Firstly, the high number of participants would be unnecessary if the data achieved saturation early during data collection or the data analysis process. Secondly, it was not the aim of qualitative research to represent the population statistically. Finally, a large sample size would be difficult to manage and to analyse the data as qualitative research is highly time consuming reflecting the rich and detailed data it provides.

Patients with a declared learning disability or significant medical conditions/associated craniofacial syndromes were excluded from the study to eliminate the chance of confusion. The approval for an interpreter was granted in cases of the participant being a non-English language speaker. A total number of 110 were invited to participate in this study, of which 28 patients accepted to participate into the study. At the point of completing the interviews in these 28 participants, data saturation was deemed to be reached in relation to orthodontic treatment experience.

In qualitative research, when the data reach the point where there are no more new themes or concepts arising, a point of saturation is said to have been achieved and the data collection process ended (Coyne, 1997). However, a sample of about 30 participants is still sufficient to identify common themes and suggest transferability of findings (Gott et al., 2011) whilst retaining the depth of information needed to fully explore experiences and understanding of the treatment process (Crouch and McKenzie, 2006).

Patients were recruited using two CLP services centres. The CLP services were under the South Thames CLP services unit. The data collection period extended for 18 months (Completed in September 2014).

Table 25: Inclusion and Exclusion criteria

Inclusion Criteria	Exclusion Criteria
-Participants having any form of CLP -Participants who have recently finished their definitive orthodontic/orthognathic treatment (Maximum 18 months post treatment) or were about to finish -Minimum age 15 years but any age after that as long as other criteria were met -Equal number of male and female participants -English speakers and non-English speakers	-Patients with other significant medical condition or a declared learning disability or any associated craniofacial syndromes

4.2.6 Identifying and approaching suitable participants

To be able to identify potential suitable participants, the researcher focused on identifying those patients who were debonded or who were coming for their retainer review for the past 2 years. This was possible by checking the laboratory records to identify the debonded patients; clinicians always book for same day retainer construction. Furthermore, the CLP unit audit coordinator helped the researcher initially to identify future debonding patients as the clinicians fill an audit form after each visit and to write what treatment will be provided on the next visit.

Suitable participants were identified by the researcher. Participants who fulfilled the above inclusion criteria were approached and requested if they would be willing to take part in the research and if they agreed they were asked to sign a consent form.

For those who had recently completed treatment, they were approached either by letter to their home address or by approaching them in clinical waiting area directly. If approached by letter, the letter contained a letter of invitation. In the case of the patients who were below 16 years old, then two different participant information sheets for both the affected young person and the Parent/Carer/Guardian, a reply form with a stamped addressed envelope were also included (please see appendix). Potential participants were asked if they preferred to contact the researcher by telephone to have the study explained further and to arrange an appointment for an interview should they be interested in taking part.

For young people due to attend the clinic to have their brace removed at their next scheduled appointment or who were coming for a retainer review appointment the approaches were by letter to their home address before their appointment. The letter contained a letter of invitation, two different participant information sheets for both the affected young person and their Parent/Carer/Guardian. Each participant information sheet was designed specifically to the targeted group. In case of the gap between sending the letter and the appointment being too short, so the participant was at risk of not receiving the letter, they were approached in the clinical waiting area with patient information sheets and with a full description of the study.

In spite of these useful tools to identify participants, the recruiting process proved to be slow. Reaching sufficient people in the study time frame using this way was not possible. As a consequence, the researcher decided to be more proactive and adopted a more efficient search strategy for potential suitable participants (still within the approved Ethics consent). We extended our method to recruit to be visible within the clinics. Although this method was time consuming, it is a well-established method of recruiting. The researcher

would be physically present and a member of the care team approached patients in the waiting area at the CLP centre and asked them if they were interested to participate in a study. The researcher was present at the CLP clinic every day to identify suitable participants. Also, the main researcher asked his colleagues and staff to identify potential participants and notify him about when these potential participants would be attending their clinic.

A number of reasons for the slow recruitment for participation in this study can be suggested. First, the recent restructuring of the centre made it difficult to identify patients who had moved to the centre from other services. There is an established, audited, data base available which covers patients under the age of 10 but less information is available for older patients. This led to the need to include an additional cleft service to ensure that sufficient respondents were recruited. Another possible reason for the slow recruitment is that cleft patients are a heavily researched group which may have made potential respondents reluctant to take part. Also, qualitative interviews need a time commitment from both researcher and participants as it involves arranging a meeting that may last up to 2 hours. This study did not use any incentives to encourage participants to take part, nor was travel reimbursed and this may also be a contributing factor. Many participants were initially happy to participate but subsequently decided not to participate once the requirements of the interview were explained.

If the participant indicated they were happy to participate then they were introduced to the researcher. The researcher then checked if they had finished or nearly finished their definitive orthodontic treatment or their orthognathic treatment. If they were suitable he then explained the study in detail. In addition the appointment system was reviewed manually to look at each patient in the patient list of all CLP clinics types (orthodontics, plastic surgery, paediatric, speech therapy) for the following week. Potential indicators that the researcher was looking for were the patient's age and the history of previous appointments as frequently the receptionists would write some description of the coming appointment such as (debond) under the comment column in the system. This was helpful but the patient flow was still low. Please see (Figure 1).
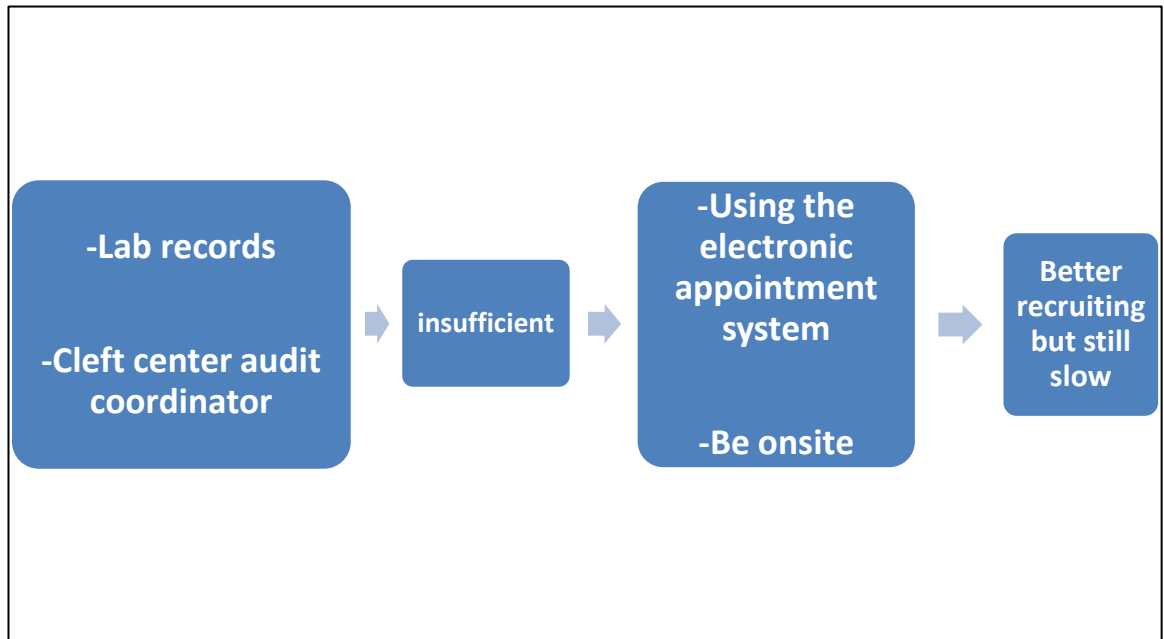
Figure 1: Patients identifying process

4.2.7 Consent procedures

Patient information leaflets were prepared for children of fourteen years of age and over, young adults and parents. Young people and their Parents/Carers/Guardians had the opportunity to receive an explanation of the study. Children had the study explained to them using more child friendly formats.

Participants had the opportunity to read a patient information sheet that explained and outlined the details of the study and had the opportunity to have the study explained further verbally. They were given sufficient time to decide if they wished to take part. After being given a suitable time to decide, the participants were asked if they wish to participate and to sign a consent form indicating their consent.

Before consenting, the author made sure that the participants understood the study clearly by oral explanation using language used by a layman; also the patient information sheet was written clearly using simple words and sentences. This was validated by non-participant individuals. Then participants were asked separately to sign a consent form if they decided to participate. In consultation with the direct care team on all occasions, if it was felt that the participant did not understand what was involved in participation, the recruitment process was terminated. All clinical postgraduate students attend a clinical induction day that covers the consent taking process.

All consent forms were completed and consent was taken by the main investigator. The participants who needed more time to think about joining the study were given one week to consider their involvement. They were contacted one week later and invited to participate. Communications were either via email and/or telephone. A number of potential participants ignored this communication and failed to reply. On a few occasions, the researcher travelled outside London to meet the participants in an agreed appointment (n=6).

An appointment was made for the interview and participants were asked to sign the consent form on commencement of the interview. No child was approached without first seeking permission from the parent/responsible adult. If the patient was under 16, then a parent's signature was obtained. Both the patient and one of their parents were required to sign the consent in order to participate. In cases of the child refusing to participate, the wish of the child was accepted even where the parent was happy for the child to participate. As the participants included a wide age range which included children under the age of 16, an assent form were signed by all children participants besides their parents/cares/guardians consent.

4.2.8 Interviewing

All interviews were conducted face to face by the main investigator (RA) and recorded, with the permission of the participant, using two digital voice recorders. An appointment was made to conduct the interview after or before their clinical appointment visit or on another date and time at the participant's convenience. Participants were given a telephone number which they could call to change or cancel an appointment. They were reminded of the interview date and time 1 week and also 24 hours to 3 days prior to the interview.

For participants' convenience, they were given the option to choose the location of the interview (hospital or at the participant's home). The interviews were held in a non-clinical setting to provide a more relaxed and friendly environment than the clinics. Rooms were booked in advance according to the agreed time between the researcher and the participant. Although participants were given the option to choose the location, most participants chose to arrange the interview after their next appointment. Few arranged a specific date just for the interview. All interviews took place in a non-clinical room within the cleft centre. Whilst we did not ask why participants did not request interviews at home, it may be that being interviewed whilst already attending the clinic was most convenient to the participants.

Participants were told that if they found the questions of a sensitive nature or not pleasant then they did not have to answer. Also they were reminded that they could excuse themselves from the interview at any stage and withdraw from the study at any point.

The minimum interview length was 19 minutes and the maximum interview length was 108 minutes. The average time for all interviews length was (51 min, 25 seconds).

The interview started by giving reassurances to participants about the confidentiality of their responses. Then asking a general open ended question like (can you tell me about yourself?). Participants were encouraged to introduce themselves in their own terms and specific sociodemographic details were not collected. The reason for this was to enable participants to describe and introduce themselves. As the purpose of this study was exploratory, we did not feel that additional sociodemographic details were necessary. The interviews then moved on to cover the topics in the topic guide. Starting with questions related to participants understanding of the CLP, moving to the asking participants about the experience of living with CLP. Then asking questions related to the CLP care starting with for example (what do you think about the CLP care?) and then adopting a flexible approach to explore further based on the participant`s answer. Then questions related to the experience of having the definitive orthodontic/orthognathic treatment were asked. The researcher (RA) used a probing technique with questions to explore further including "Can you tell me more about...?" or, for a clarification, "What did you mean when you said...?). Also, following probing, a prompting technique was also used to raise issues mentioned in previous interviews.

4.3 Data analysis

Thematic framework analysis was used to analyse the data. This method of data analysis was developed and followed by the National Centre for Social Research (NatCen) and used to provide detailed in depth information of the participants' experiences. This approach is increasingly being used in relation to dentistry. Also it is been popularised by some in orthodontics (Pabari et al., 2011, Lee et al., 2011, Ryan et al., 2009a, Trulsson et al., 2002).

All interviews were recorded and transcribed verbatim. All interviews were coded to ensure confidentiality and anonymity. The transcripts were coded independently by two members of the research team (RA) and (SS).   The other members of the research team (BD) and (FM) coded independently a random 5 interviews. Interviews transcription was carried with interview analysis concurrently. The main investigator transcribed eight interviews then a professional transcription service was used (Essential Secretary Ltd) for the rest of the interviews (n=20). Interviews transcribed by external service were checked by the main investigator before commencing their analysis. Data analysis was conducted using a thematic framework derived from the data and developed through the analysis process. Thematic content analysis was undertaken. This involved data management starting with familiarisation, when the researcher familiarized himself with the data by reading and re-reading it several times, then recurrent themes were identified and then were sorted under main themes to create an overall framework. Indexing of the data was then undertaken which was to apply the thematic framework to all data and assign the data to the developed themes through labelling the original data to the appropriate theme. The next stage was charting. This was where the data were arranged according to the appropriate part of the thematic framework. This was done by creating a matrix for each theme with the columns representing the subthemes and the rows representing each participant. This provided a summary of the data into the matrix cells. The researcher tried different tools to help arranging the data such as: Microsoft Excel spread sheet and (NVIVO) software. Then the data analysis moved into describing through identifying elements, classifying data, establishing typologies and then developing explanation by trying to make sense of the data (Ritchie and Lewis, 2011). (Figure 2).
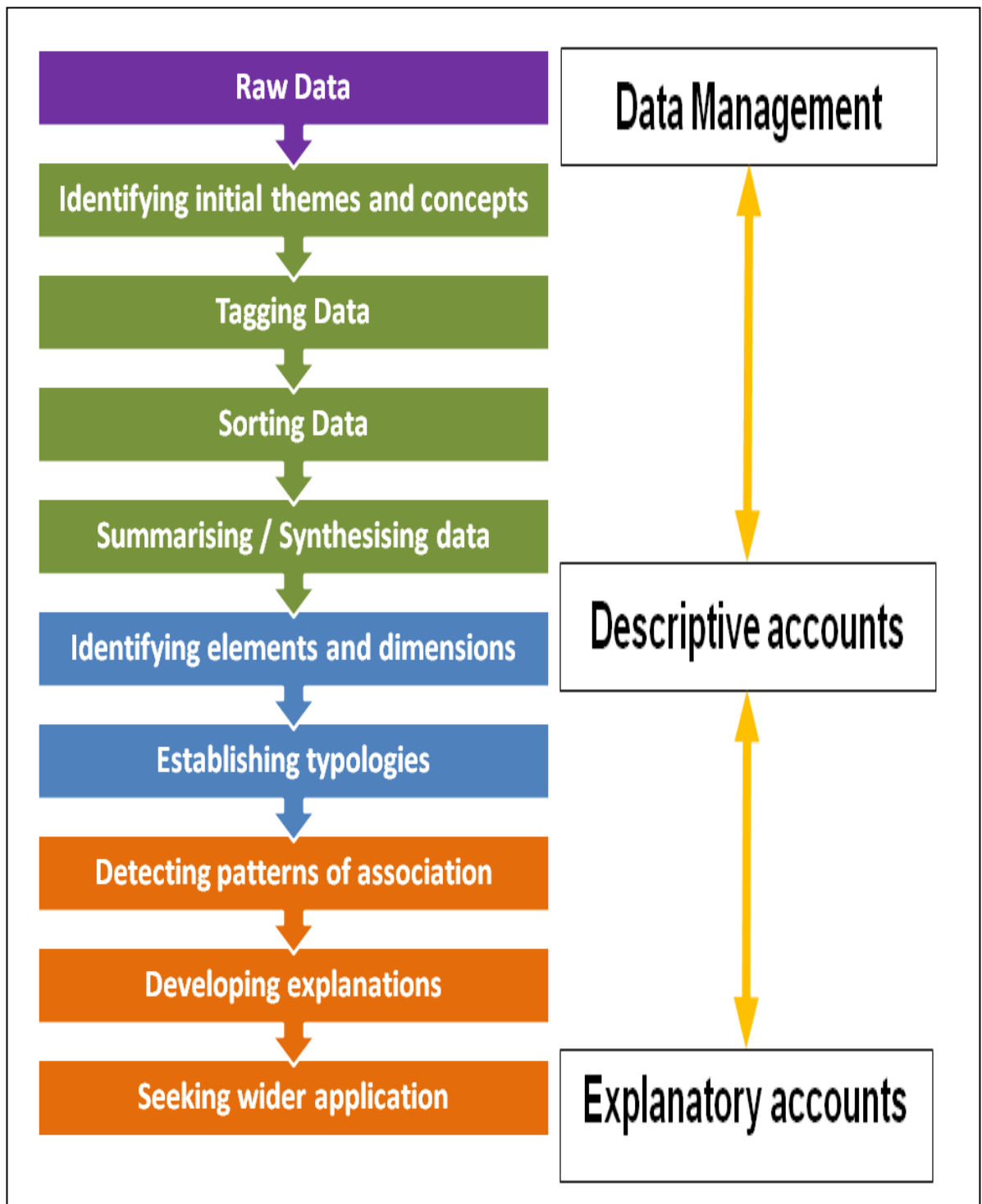
Figure 2: Steps involved in data analysis

4.4 Quality of research

The validity of this research was enhanced and assured by the use of rigorous and explicit data analysis methods described in detail by Ritchie and Lewis (2011). Respondent validation was also used during the interview where a new theme was raised or where the researcher needed to clarify a point. To ensure reliability of the data analysis, the transcripts were double coded independently by two members of the research team. The inclusion of a different age range from males and females plus recruiting different ethnicities and different types of CLP allowed for diversity of perspectives to understand "fair dealing".

Reflexivity

The researcher`s awareness of his role is important in qualitative research. As the author is an orthodontist clinician, conducting the research with extreme care can minimise the impact that might influence the data collected (reflexivity). Extra measures were taken to avoid the bias and influence during the interview process by the awareness that the participants should say whatever they feel; they should not be corrected nor be influenced by the researcher even if the researcher totally disagrees with their views or if the participant say something clinically 'wrong'. The researcher was neutral and would not interfere or influence the participants' responses directly or indirectly. Prior assumptions, thoughts, opinions and experiences of the researcher (particularly clinical and orthodontic related) should have been completely ignored and the researcher was prepared to be mentally open to any arising themes or issues. The researcher was careful not to use language that was too clinical and was aware not to make clinical comments during the interview. The same approach was followed during the data analysis stage and throughout the research process.

4.5 Summary of research methods

Construct a topic guide to perform the interviews

Modify topic

Practice the interview

Suitable participants will be identified by a member of the clinical team

Completed treatment

Ongoing treatment/review

-Letter of invitation

-2 Participant information sheet (patient, parent)

-Reply form

-Letter of invitation

-2 Participant information sheet (patient, parent)

To contact the researcher if they be interested in taking part

Will be approached in person by a member of the direct care team who will then introduce the researcher

Recruitment (consent, arrange appointment)

Conduct the interviews with participants

Data Saturation

Qualitative analysis of the interviews

Report results

4.6 Summary

In this chapter, the rationale and justification of the methodology were explained. The ethical issues related to the study were presented; and the sample, data collection and analysis methods were described. The next chapter presents the results of the study.

5. RESULTS – CHAPTER ONE

5.1 Introduction to findings

The data presented here provide a detailed description of the experiences of people living with CLP incorporating the day to day impact of living with the condition as well as a detailed exploration of experiences of the care pathway.

The results of this study are considered in two chapters. The first chapter looks at the day to day experiences of people living with CLP providing the context with which to understand the experiences of care. This chapter is organised to build a picture of the impact of CLP from the perspective of the respondents, starting with their understanding of the condition and its causes before moving on to look at the impact of living with CLP. This will include perceptions and recall of the care received through the pathway. In addition the impact of the condition on daily life will be evaluated.

It is against this background that the second chapter then looks in more detail at the CLP care pathway from the perspective of the respondents, incorporating the novel, rich and detailed description of the definitive orthodontic treatment as part of this pathway. In the second chapter the data is presented chronologically according to the stages or steps of the care pathway. The treatment related themes are grouped into three major time points (before treatment, around or during treatment, after treatment).

In the case of the CLP care pathway, and the experience of the definitive orthodontic care contained in the pathway, it can only be understood within the context of the understanding and experiences of people living with the condition. This in turn needs to be understood as a temporal experience. The concept of time emerges repeatedly through the data and can be used to help explain the experiences presented. CLP is diagnosed prenatally and the participants in this study have grown up with the condition. The understanding and experiences presented need to be understood in the context of the ageing process.

This first chapter starts with an overview of participants within the study. This is followed by an outline of the main themes to emerge from the data. Each theme is then analysed in turn and explored in more detail, using quotes from the relevant data sources to illustrate the findings of the study.

Participants Profile

The participants were selected using the purposive sampling criteria that were presented in the methods chapter. To reflect on the diversity of the possible experiences, the sample included patients of different genders, ages, ethnicity and different types of CLP. In total, twenty eight (fourteen males; fourteen females) participated in the interviews (Table 1).

All but four of the participants were Caucasian. The rest included Afro-Caribbean (n=2), Chinese (n=1) and Indian (n=1) ethnic backgrounds. They ranged in age from 15.8 to 45 years. The sample included a range of different CLP types and all participants experienced orthodontic treatment as part of an overall treatment strategy. Twelve participants had received orthognathic treatment. (All abbreviations used are listed in the glossary).

Table 26: Participants demographic information and information related to results

Interview Number	Gender	Age	Interview length	Ethnicity	Type	OGN
1	M	23.1	77 min	Afro Caribbean	BCLP	Yes
2	F	16.8	48 min	Caucasian	CPO	No
3	F	17.5	58 min	Caucasian	UCLP	Yes
4	F	15.8	36 min	Caucasian	CLO	No
5	M	21.5	57 min	Caucasian	BCL	No
6	M	19.11	55 min	Caucasian	UCLP	Yes
7	F	27	62 min	Caucasian	UCLP	Yes
8	F	17.5	64 min	Caucasian	UCLP	No
9	F	18	46 min	Caucasian	BCLP	Yes
10	F	23.9	64 min	Caucasian	UCLP	Yes
11	M	22.2	54 min	Caucasian	UCLP	No
12	F	45	60 min	Caucasian	UCLP	Yes
13	F	20.1	49 min	Caucasian	UCLP	No
14	M	18	30 min	Afro Caribbean	UCLP-Van der Woude left	Awaiting
15	F	17	19 min	Caucasian	CPO-Isolated alveolar cleft	No
16	M	22.6	24 min	Caucasian	UCLP	No
17	M	23	108 min	Caucasian	BCLP	Yes
18	M	16.2	34 min	Caucasian	CPO	No
19	M	18.5	26 min	Caucasian	UCLP	No
20	F	20.4	40 min	Indian	UCLP	No
21	F	18.9	64 min	Caucasian	BCLP	Awaiting
22	M	17	32 min	Caucasian	CPO	No
23	M	20	52 min	Chinese	UCLP	Yes
24	M	18	33 min	Caucasian	CPO	No
25	F	17.11	24 min	Caucasian	CPO	No
26	M	21	73 min	Caucasian	UCLP	Yes
27	F	21	65 min	Caucasian	UCLP	Yes
28	M	20	81 min	Caucasian	UCLP	Yes

5.2 The experiences of people living with cleft lip and/or palate

5.2.1 Overview of the key themes

A number of areas were covered in the interviews. These included the amount of understanding which participants had about the condition itself and its causes, the impact of the condition socially, aesthetically, functionally and personally across time, and the role of memory in treatment recall. The following areas were explored in detail:

- Understanding of CLP
- Feelings about living with CLP
- Impact of living with CLP
- Perceptions of the care pathway
- Memories of treatment
- The impact of treatment on day to day life

Throughout the remainder of this chapter the results will be presented within these themes. Quotes will be used to illustrate the points made and, where useful, findings will be presented visually as well as textually.

5.2.2 Understanding of cleft lip and/or palate

The first part of the results explored what participants understood about their CLP. Of the participants interviewed within this study, two major groups were identified. The first group were able to give some explanation about the condition. This included information about what CLP is and how common it is, along with timescales as to when it happens, as well as causes and effects of the condition. The other group are those who felt unable to explain or lacked knowledge related to the condition. These responses will be presented in detail. The main findings are presented in (Figure 3).
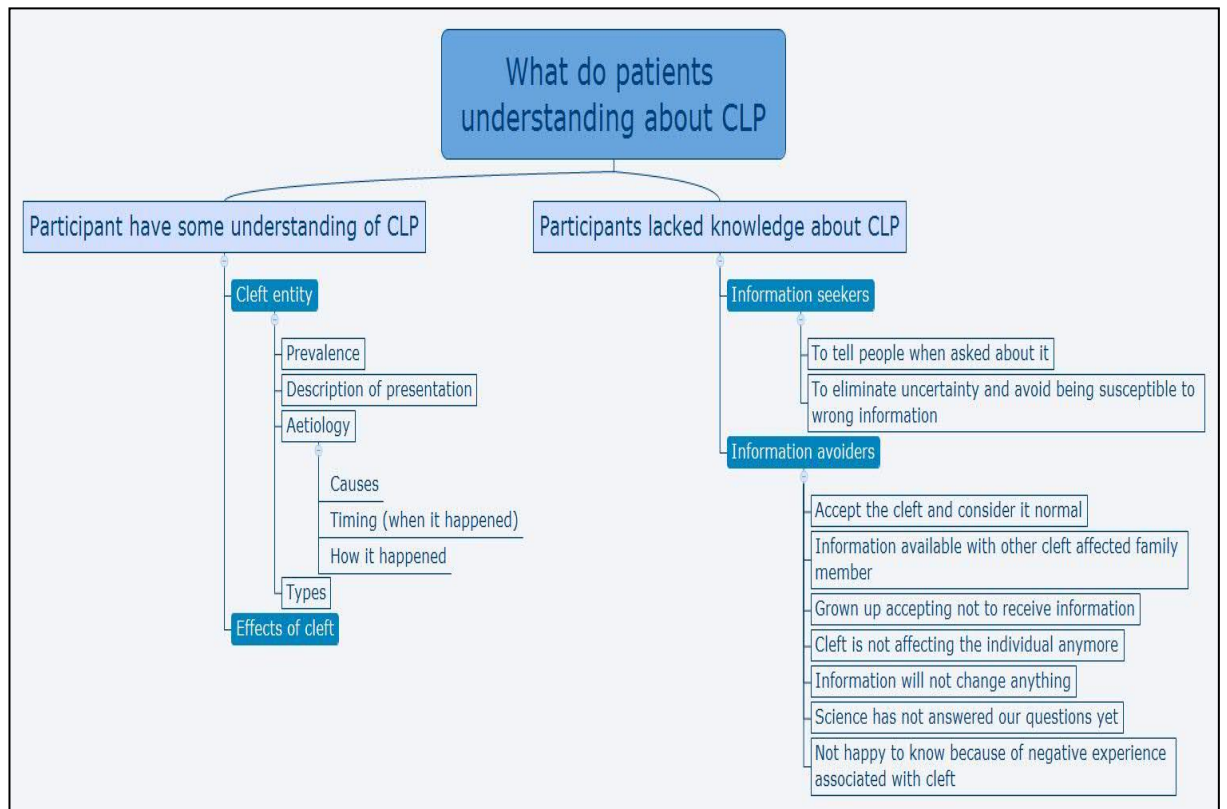
Figure 3: What do patients understand about CLP

When asked to explain what they knew about CLP, it appeared from the data that participants held a variable degree of knowledge about the condition at the time of interview. A range of descriptions about understanding the CLP were reported. Descriptions ranged from describing the condition itself, its aetiology, its effects on the individual to descriptions that incorporated the care pathway and treatment. Descriptions included both personal (presentation of the CLP) and more generalised information about the condition (prevalence and process).

General descriptions incorporated both the process that had occurred, or failed to occur, and the timescale within which it had happened:

> *"It ... a cleft occurs when in the womb the bones don't fuse together as they ought to. That just leaves a gap in your palate and your lip."* **INT#3.F**

> *"it's a condition where basically when you're a baby, your body is not, it's supposed to be fused right in the middle so I didn't get that fusion where it stops from the lip, that's what I'm assuming, am I right or am I wrong?"* **INT#23.M**

Also, the actual physical description of the condition was combined with information about the incidence of cleft:

> *"I know that it's fairly common, something is it 1 in 700, something like that"* **INT#10.F**

Personal accounts contained broadly similar information. Descriptions included CLP presentation in addition to a timescale although there seemed to be some confusion over whether the condition developed within the womb or at the point of birth.

> *"Well as much as I know about it is that just after I was born I had a hole in the roof of my mouth which actually I don't know the specifics of it but that had to be closed up and they took skin from the back of my throat I think to seal it up or something like that, I'm not aware really of the specifics of it."* **INT#24.M**

> *"I know that I was born with two large holes in my palate and with a skin bit of my upper lip missing…"* **INT#6.M**

When asked about the causes of CLP, opinions were divided and there was some confusion about whether and to what extent, genetics played a role in the development of the condition:

> *"Well, it's not known whether it's a genetic or environmental condition. It could be a mixture of both. And sometimes you get people in families having clefts but it's not always the case."* **INT#3.F**

> *"I know it's not genetics"* **INT#23.M**

As a result, alternative explanations about the cause of the condition may have developed within one generation and be passed down to other generations:

> *"It can be caused by the environment, like my Mum always thinks, she was scared by a dog when I was early development in her tummy, she was knocked over by a dog or something, she's always said it must be that, but obviously that's just a story, but I'm not*

*really how it all comes about but yeah, I'm not, yeah I should probably read up a bit more on I guess, its quite an interesting subject"***INT#11.M**

There was also an acknowledgment that there are different types of CLP. CLP was described both from a medical perspective and other descriptions were provided in lay terms:

*"I know that there are different stages of cleft lip and palate, or just the palate, deformity. So, in my case I got both, the cleft lip and the palate, as you can probably tell just by looking at me, someone in your field can probably pick you out quite quickly."***INT#17.M**

*"I only had the one side as opposed to the full lip. That's pretty much what I know. And the palette obviously is another, you can either have the full palette or not so."***INT#10.F**

Some descriptions focused on the effects of the condition when asked about what the participants knew about CLP. Rather than describing the process by which the condition developed or potential causes, they described the aesthetic and functional issues:

*"Yes, the only thing I know about it you end up with bent teeth (laughing)."***INT#16.M**

This may suggest that understanding of the condition may not easily be separated from individual/personal experiences of it.

The other major group were participants who self-reported that they did not know much about CLP and provided a range of responses varied from 'knows nothing about it' to 'not much details'. This group could be further sub-divided into two smaller groups. The first group were those who do not know much about CLP but would like to know more about it (information seekers) whilst the second group were those that did not want, or feel the need, to have more information (information avoiders).

Information Seekers

Information seekers provided two reasons to explain their response. Both of the reasons given for wanting further information involved social situations. One reason was to be able to share information with others whilst the other was to combat misinformation and potential stigmatisation or bullying. The need to provide information to children rather than just parents to enable them both to understand their own condition and to share that understanding with others was identified in the following passage:

"... what's going on, like you know what you have but don't know or understand like obviously when they're ... I can sit back and yeah, your mum will tell you this and kind of

81

*break it down, you'd be like ... they wouldn't bother but, if you could go back it would be a bit more better you can ... when I was younger I didn't really knew about cleft lip and palate and it would have been nice to know about it when you was younger because but obviously you have a lot of young people that also don't know about it and obviously I didn't really know a lot about it so I couldn't really tell them about it ..."**INT#14.M**

The other reason, which can build on the first, was where a participant felt vulnerable to the wrong information. As in this example, a participant talked about feeling 'very susceptible' because when he was young and he lacked information about CLP he tended to believe whatever other people told him. This left him feeling vulnerable:

*"As it is such a minor thing it does affect what you do but it does affect your mentality and what it is in the way that other people treat you. That is the main issue. The main issue is growing up with something and not being able to fully understand it yourself, those people don't really know why people have cleft palates. Which means when other people tell you something about it you then believe that because you don't know yourself and you are too young to understand it anyway so you become very susceptible, I wouldn't say nasty but darker sides of what you feel you are because of your cleft palate. So there's a very dark, I think it all depends on your parents as well and the good people around you."**INT#28.M**

In both of these examples the participants would have benefitted from further information. From a personal perspective, building their confidence about themselves and how and why they found themselves living with CLP in turn appeared to enable them to challenge incorrect information. Socially it could provide the tools to educate others and combat bullying and misinformation.

Information Avoiders

In contrast, there are several reasons which are grounded in the data that may explain why one group was either happy with a minimal amount of information and/or showed no interest in plugging any gaps in knowledge that they were aware that they had. Reasons varied from a simple acceptance of CLP as 'what is' to avoidance of any discussion about CLP because of the trauma of bullying.

One reason offered during interview was the perception that CLP was just something that was there and had to be accepted and dealt with as part of normal life. There was no desire for seeking information about the CLP because the CLP was not perceived as an issue but rather accepted or considered as 'normal'.

"It never really, it never really, never really have that kind of desire to learn a lot about it, I read on like charity websites that go out and do repairs like they did say that it's quite common but nothing really beyond that, nothing like why did it happen, I mean I understand that when I was born it didn't repair properly but that's really all I really know..........I think, I didn't think of it as anything very special that I felt like I had to go and then know about it like why did it happen to me, I think like me and my family we accepted it and it was like normal for us." **INT#20.F**

In the second part of this quote this participant goes further to suggest not only that she didn't need to know more but that CLP was 'normal', not seen as different or requiring specific knowledge. This is interesting in light of the care pathway that the participant will have been part of from birth and the extensive treatment that she will have undergone.

Another participant was aware that she had gaps in her knowledge about the condition, and had looked into it to a certain extent as she grew older, she felt no real need to know more than she did:

"I mean obviously I've looked into it in later years but it just shows how ignorant when you asked me what type I had I've just not really thought too much about it, I've got it but yes, you know I just know that part of my lip and my palate didn't form properly and I've just had surgery to repair it throughout the years." **INT#12.F**

"I think, I didn't think of it as anything very special that I felt like I had to go and then know about it like why did it happen to me, I think like me and my family we accepted it and it was like normal for us. You know I know it's hard [unclear-0:03:34.9] but I didn't think it was something that I had to go and find everything it, you know if I didn't find out about it I'm not going to be satisfied or something I kind of accepted it like I have, I was born with it and I'm going to go through the process that I need to go through and I'm not going to worry about it too much and go on about it I'm going to you know live my life and not think about my cleft." **INT#20.F**

This suggested an acceptance of CLP as simply something that 'is', a part of life that has always been there.

Another reason provided was that CLP ran in the family and so was normalised. In a case of another family member having CLP, there was less interest to seek information about the CLP condition because as this example shows the information was already available:

"Because my mum had it, it was just something, because my mum was the first one, there's no other records in the family of anyone else having a cleft lip and palate and because my mum had it, I just followed in her footsteps in a way, because she was telling me her experiences through having it, so I never really thought of looking it up." **INT#13.F**

83

A feature of not obtaining or enquiring about supportive information may relate to the acceptance that data was never provided to children as patients. It was only provided to the responsible adults/carers of the child patients:

> *"I don't know a lot about it actually because as a child, sort of forty five years ago we were just put in hospital and not really told what we were having done so I just grew up to accept it and we weren't sort of given counselling or explained a lot really and I've never felt that I really needed to know much about it. ."***INT#12.F**

Another possible reason is because the cleft was either lip or palate seen as mild, causing no functional or speech issues. The individual lost interest or motivation to know more about the cleft because the cleft is not affecting them anymore, possibly being corrected during the earlier surgical procedures as with a cleft palate only. If the case was mild and the earliest surgery was successful, then the individual would probably not face future speech difficulties. This was similar, in a case of 'cleft lip only' where the scar is only mildly visible.

> *"Well it was explained to me once but I sort of understand it, it's ... but I don't really, I really feel I need to understand it completely because it doesn't really affect me any more."***INT#15.F**

The feeling that knowing about CLP would not actually change anything may be another reason that could encourage avoiding information. Even if the individual knows everything about CLP they may still face the problems associated with CLP which in turn will still affect them in one way or another.

> *"I think, I didn't think of it as anything very special that I felt like I had to go and then know about it like why did it happen to me, I think like me and my family we accepted it and it was like normal for us. You know I know it's hard [unclear-0:03:34.9] but I didn't think it was something that I had to go and find everything it, you know if I didn't find out about it I'm not going to be satisfied or something I kind of accepted it like I have, I was born with it and I'm going to go through the process that I need to go through and I'm not going to worry about it too much and go on about it I'm going to you know live my life and not think about my cleft."***INT#20.F**

Furthermore, the feeling in which science has not completely answered the question 'why people are born with CLP?' may lead to avoidance of obtaining information. The following example shows that a participant avoided information because they felt that there was no new information worth having; scientists do not yet know why people are born with CLP:

> *"You are born with it, other than that nothing really. I mean I know there's a 2 minute gap in your mum's womb where this has to sew up, or come together or whatever and it doesn't so that causes all the implications. But they don't know why that happens. So I don't really know much about it myself."* **INT#28.M**

Building on this, there is a need to know what causes the CLP.

> *"Until they know why people are born with cleft palates that is the only question I've ever had. And they don't know why, they know how it happened, they don't know why. So there's nothing really."* **INT28.M**

The final reason to avoid information was related to negative experiences. From the presented example, one participant explained that she was not happy talking about CLP and avoided the subject because of negative experiences at school. It was made clear that this feeling was related to bullying experiences at school:

> *"Because I don't really like talking about it, I don't like telling other people why. Sometimes I get funny looks and strange looks and I think to myself well that's just how I am. Sometimes I feel like I'm getting treated differently as a person, I feel like I get judged by my looks. I went through primary school and secondary school being bullied from it and it made me feel like I couldn't meet any friends because of the way I was. I got called lots of names and nobody took me for who I was really and then as I got older I started to understand that this condition isn't my fault or anybody else's fault, so I just had to live with it and get on with it."* **INT#27.F**

The data showed that whilst some patients may have some degree of understanding about CLP, in contrast, others did not. From the latter group, patients may have needed to know more about CLP or may not have any interest to acquire further information/material. These results tell us that some patients needed more information and thought sometimes they did not have enough information. The data also showed that there may be a linkage between the experiences of bullying and the avoidance of information. Bullying and stigmatisation came up consistently when participants were asked about their feelings and experiences of living with CLP. This is explored further in the next sections. In this section, an insight into the patients understanding of CLP was obtained. The next section provides an overview of how CLP patients feel about it.

5.2.3 Feelings about cleft lip and/or palate

Following questions about the nature of CLP, participants were asked to describe how they felt about having the condition. Views were mixed. (See figure 4).

Negative views
- Difficult for parents
- Feel different (not normal)
- Ordeal
- Hates it because its negative impact on life

Positive views
- No impact as treated early
- It is normal
- Better than other more severe cleft
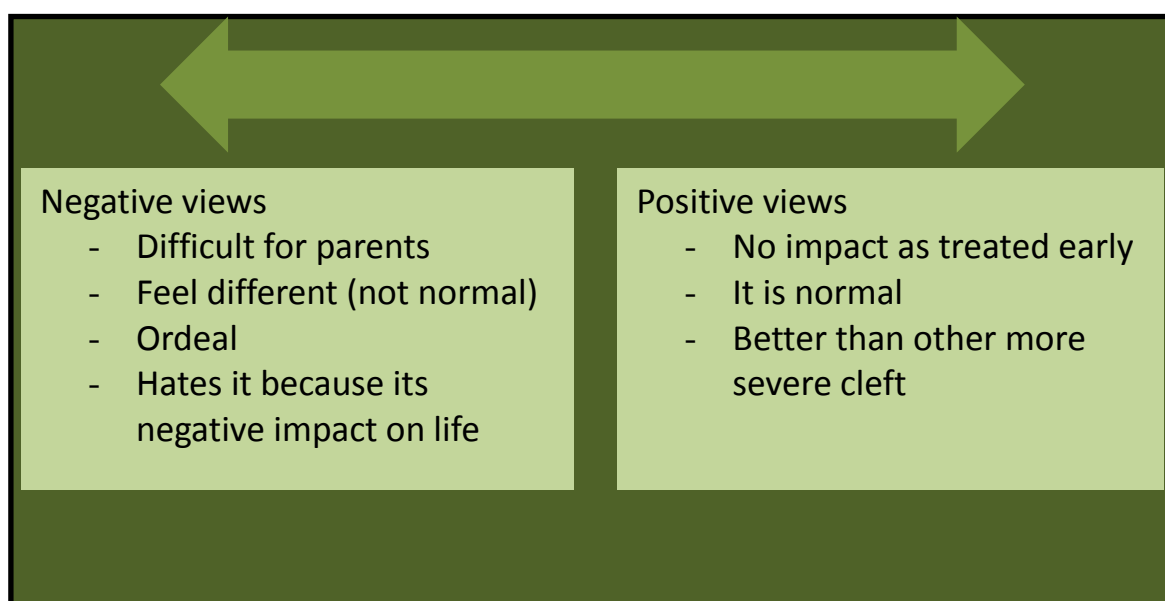
Figure 4: The range of participants views about their CLP

Several reasons were provided in relation to negative feelings about CLP.

Knowing the fact that having a child with a CLP is difficult for parents may make the individual perceive the CLP negatively.

> *"My mum just told me through, it's difficult, it can be difficult for parents, if it's their first child with a cleft lip, because everything's different, they have to get certain bottles, and they find things a lot harder"**INT#13.F***

Another reason reported was feeling different (not normal) to others because of the aesthetic appearance which could also make a CLP individual feel negative about the condition.

> *"There's some days where you feel down about having this because you just think that this is the only thing that stops you from being normal and I'm guessing that a lot of other people would be going through the same thing that I would be going through and then they'll ask themselves why did this happen to them"**INT#23.M***

Participants may hold a stronger negative feeling toward CLP, when seen in the wider context of the treatment pathway, perceiving it is an ordeal.

*"If you are not born with that you don't go with that, you don't have to deal with that. It becomes a long ordeal if you are born with something regardless. It's an extra little thing you have to do throughout your life. I am 20 no so for 20 years I've had this so it's a course that's going to be a long ordeal. It is a nuisance, it's a pain"**INT#28.M***

The effects of having a CLP may cause the negative outlook towards it. In the example below, a participant reported that she simply hates it because of the effects of the CLP that still exist even though there has been some significant treatment and improvement of the condition.

*"I hate it. I don't like looking in the mirror, I don't like hearing myself. I can't, I don't like people taking pictures of me because the way my nose is, the way my lips are and I don't like my speech and I don't, I just don't, I hate my looks, I'm a really insecure person and never have liked my looks. And it's just, it affects the way I feel as well about myself because I feel like I'm not confident enough to go out and make new friends or people who are going to take me for who I am."**INT#27.F***

In this example the negative association related to aesthetic, functional, social and psychological.

In contrast, positive views were also reported. This is clear when the CLP was treated early in the participants' life and they didn't experience the effect of CLP in the longer term. For example the following passage in a mild, cleft palate only, case.

 *""Not much. I don't really think about it much, I just live with it. It doesn't affect me so much right now but I've been told it could affect me in later years and that it has affected me when I was a baby. Right now, at this stage, it's not a problem. I don't really think about it that much."**INT#2.F***

Another reason for the positive outlook was when CLP was perceived as a normal thing or was ignored and/or avoided.

*"I suppose it is what it is. It is what it is (laughing). I managed. I just lift my head up and just carried on. That's all I've done. I've not let nothing get in my way. Nothing will bring me down. I've just lift my head up and carried on."**INT#16.M***

Comparing a milder condition to other more severe CLP forms may lead to viewing CLP in a more positive way.

*"Definitely, yeah I think, also having that, other people with the cleft, I think I look much better than them"**INT#7.F***

The results showed that CLP patients may perceive the CLP itself or the CLP care through different lenses ranging from a positive lens all the way to a negative view. The data shows that these outlooks are influenced largely by how having CLP or its treatment affected daily life. These effects include CLP individuals considering themselves as 'not feeling normal' or effects on those socially associated with the CLP patient such as the family. In this section, the data showed some possible explanations as to why participants may view CLP in a positive or negative way. What was obvious from these explanations was that all are related to the experience that participants have of living with CLP. The outlook is strongly related to the living experience. This leads us to the next part which explains in detail the impacts of living with CLP lip and/or palate from the participant's perspectives.

5.2.4 Impact of living with cleft lip and/or palate

In this section the data demonstrates how individuals with CLP are affected by CLP. This will be discussed on the personal level as well as the social level. These impacts can be grouped under two groupings: physical and non-physical. These groups can then be further sub-divided under four major clusters: aesthetic, functional, psychological and social impact (Figure 5). It is also important to note that these impacts may interrelate.
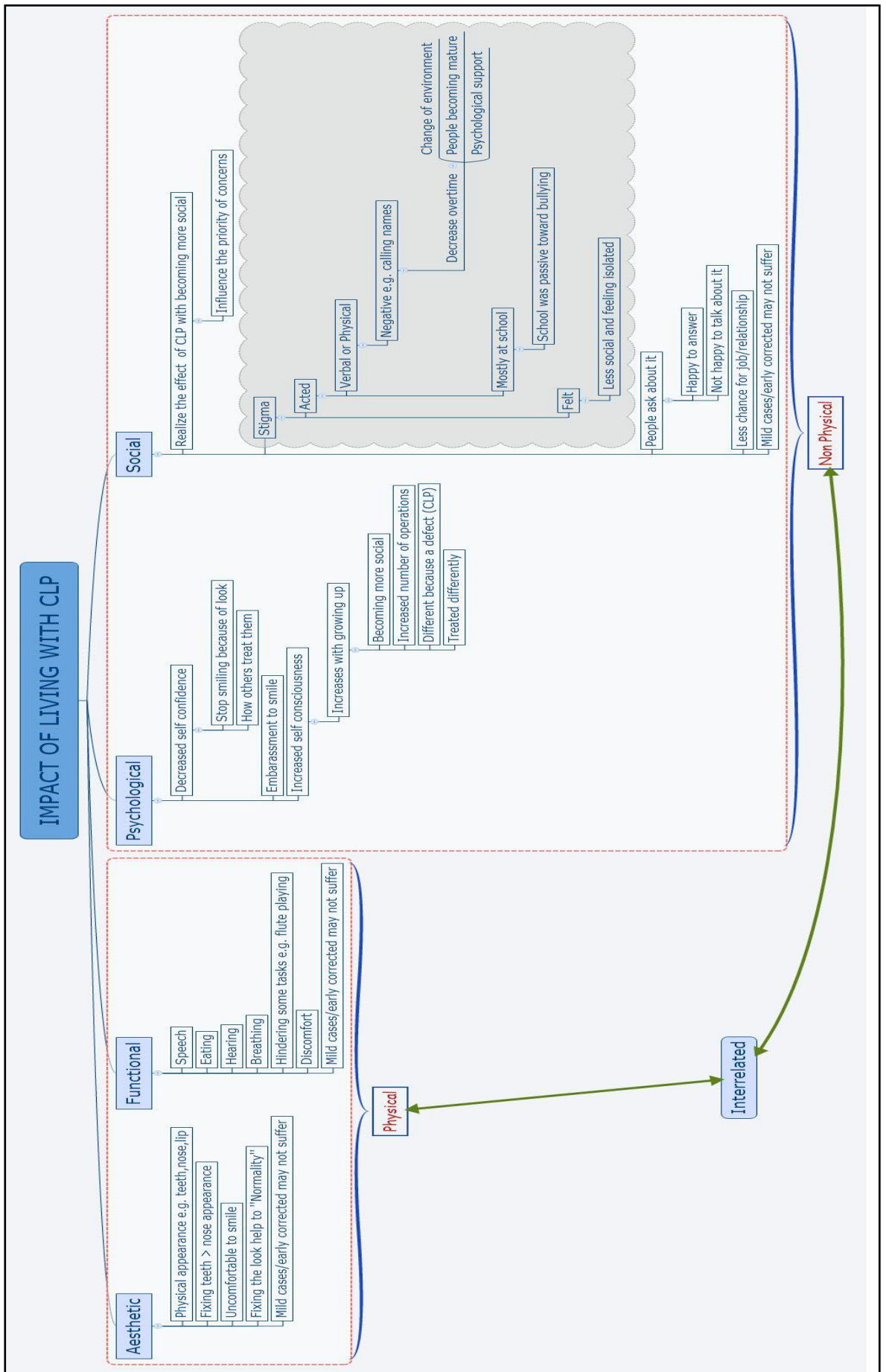
Figure 5: Impact of living with CLP

Aesthetic impact

Aesthetic impact encompasses anything relating to the look of the physical body of the individual. The impact can affect the appearance of different parts of the face including the teeth, nose and/or lip scar. Surprisingly it was reported that correcting the aesthetics of the teeth is more important than that of the nose. This is perhaps because fully 'correcting' the teeth is known to be achievable and more realistic than the complete 'correction' of the nose. Alternatively there is a dislike of the dental appearance due to the influence of modern society in which people are now more conscious about their dental appearance.

> *"Yes it's just cosmetically really I don't know whether it's because people have, no it's not because people are far more conscious nowadays, I don't like the look of my teeth and I know I'll never have the perfect smile, you know but yes I am conscious of my teeth especially if I see a picture of myself I go oh you know and it's never at my nose it's just at my teeth that I don't like. Like I say I can accept you know that my nose won't be straight or anything but my teeth I just haven't been happy with and I know there are people with worse teeth than mine as well."* **INT#12.F**

The aesthetics of the teeth can make the person feel different and uncomfortable with the simple act of smiling. It was also reported that having a CLP affected their social behaviour with less tendency to smile.

> "so *"I felt like I never felt comfortable smiling or anything, because people the first thing they notice was my teeth"* **INT#13.F**

> *"I always felt I'm different to everyone else, because they had perfect teeth and they look at mine and they go what's wrong with yours, why do your look so odd, and then I had to describe the whole.."* **INT#13.F**

Correcting the appearance may lead to the feeling of being 'normalized'. It was stated that correcting the look 'can help to become normal'.

> *"I will put the looks are the main thing I would say, me a person because it's like, it's like, I don't like to say but it's like, when I look normal yeah, if it's like, it would ... not by look it just makes me feel like me as a person I could be"* **INT#14.M**

However in a cleft lip only case where the scar was barely visible, the individual could be happy and claimed that the cleft did not affect their appearance.

> *"not particularly I mean…few of my friends didn't really notice…like scar or odd I ever have cleft lip…I mean people have asked me and that's been fine..and it's not…it hasn't really like inhibited my social life at alL"* **INT#4.F**

Again this suggests the importance of taking the severity of the CLP into account.

Functional impact

Functional impact is related to problems that affect the expected function of part of the body. Based on the data; these include problems with speech, eating, hearing and to a lesser degree breathing problems as well as some general discomfort irrespective of the functional issues.

Speech and hearing problems might hinder the person from being fully socially integrated, as well as being less accepted by others. This may lead to stopping the individuals from having friends with avoidance of social interaction and contact. As a result the individual may feel different from everyone else.

> *"Socially, because I had problems with my hearing as well, I think that was a, I couldn't hear what people were saying sometimes, my speech probably stopped me from making friends as how I could do, if you know what I mean, so that was something that obviously, affected me socially, being different from everyone else and then not talking to me, maybe, not talking to me but not making friends as easy as it would be, if I didn't have it, so people look at you differently."**INT#18.M**

Eating problems were identified as present:

> *" Okay. Having the cleft itself was never any problem. It was just my jaw, it was just that it looked different, my eating was different I couldn't when I ... Like I said I couldn't bit into things properly. Which again I adapted over the years of having this and I was just very much more self conscious."**INT#10.F**

In addition there were problems associated with the balancing function of air pressure identified with abnormal/unusual functions of their inner ears.

> *" Yeah, well you have your breathing problems and your ears pop more than the average person and you wear a brace for a number of years and you have a couple of surgeries but its, yeah I think it might have been worse for my mum having to look after a child with a cleft and going through surgeries than myself really."**INT#7.F**

Speech issues could hinder the sociability of the affected individual. It was reported that one participant found it hard to talk to other people because they were afraid of not being understood.

> *"I think I talk really loudly but I don't think I do, just for the pure reason, that I don't think I socialised properly just for the pure reason that I was scared what people would think, so in that way I backed out, but I don't do that now, just when I was younger, because things were a lot worse than they are now."* **INT#21.F**

In less severe cases of cleft palate only, where the early closure of the palate was successful, the cleft may not have significant functional impact.

> *"I don't think there's anything ... I don't think it's anything to do with it"* **INT#15.F**

In cleft lip only patients it may seem less likely that there will be functional problems. However it may have some effects that hinder the individual from performing certain tasks in everyday life.

> *"when I play the flute....and its quiet hard...to play the flute with cleft lip...it's like...the mouth shape...and so that being difficult...to...its quiet difficult to learn how to play a flute...but I overcome that and so...as sometimes it's difficult to get by some of the notes out but...it's not too bad..."* **INT#4.F**

Psychological impact

A psychological impact was identified from the data. Appearance may have a strong influence on decreased levels of self-confidence possibly associated with CLP. This may cause participants to be embarrassed when they smile.

> *"If you're not embarrassed then you've got problems (laughing). Who wants bent teeth and show it off like you're happy with it."* **INT#16.M**

Lack of confidence and feeling insecure also appeared to be related to the physical defects associated with a CLP. For example, a patient may stop smiling because of the look of the teeth caused by the CLP condition.

> *"When I used to talk people used to stare at my teeth and, or when I, I never used to smile and in any pictures I used to keep my mouth closed, because I didn't want to open them, because they maybe look a bit different to everyone else."* **INT#13.F**

> *"I will say the yeah, I will put the looks are the main thing I would say, me a person because it's like, it's like, I don't like to say but it's like, when I look normal yeah, if it's like, it would ... not by look it just makes me feel like me as a person I could be, I'd be, well again well I'm not like, I sit in with the crowd but I'll be a bit like, how can I put it ... it will just ... it will just give me that, that confidence like whereas where I feel insecure,*

*sometimes I feel a bit insecure about myself sometimes. Not about how I look, it's about the whole thing and you attend ..."***INT#14.M***

Furthermore, how other people treat the individual because of their look can also affect their confidence in a negative manner.

*"I don't feel good enough and I don't know, it's just like some friends have taken me for who I am and some haven't. The ones that haven't make me feel like, it knocks my confidence."***INT#27.F***

The reasons for an increased self-consciousness were partly explained in the data obtained. One possible reason was because of the ageing affect on the participants as they develop to become an adult with more social interaction and dining out in public.

*"I mean sometimes the braces were a bit painful and sore so that wasn't nice and I was very self-conscious because obviously as an adult I ate out a lot more than I did as a child so I was really paranoid about eating in public."***INT#12.F***

A further reason may be because individuals may go through multiple surgical operations related to correcting their appearance due to the condition.

*"it maybe I'm quite self conscious with the surgeries and things but that reduces."***INT#11.M***

Further explanations may be related to the patient having an initial and significant defect leading on to them looking and functioning differently throughout life.

*"I guess I am more self-conscious about my appearance because of it. Like I said before, other people don't notice it but I still notice it."***INT#26.M***

*"Well, a lot of other people don't have them for a start and so there's a difference there. And then, I don't know, it's hard to explain, it's like ... I don't know, because like where I've got my lip's obviously different there and so ... yeah, it just looks different from other people, I guess, and to me that makes it stand out and so I'm more aware of it than other people would be if they don't have it, if that makes sense."***INT#26.M***

The final psychological effect could potentially be the feeling of increased self-consciousness due to the patient being treated differently or feeling that they were being treated differently.

".. sometimes I felt slightly different. I felt that people judged me slightly differently just because I had a, what's the word when people see you've got a scar and it's like, ooh and then they ask questions. I was more conscious of, less conscious of the scar but

*more conscious of my jaw itself because of having a very much an under bite that's what I really found tricky."**INT#10.F***

This ties in with feelings of difference and the desire for normality identified previously.

Social impact

The data suggest that having CLP may have some negative impacts on an individual's social life. These social impacts were mainly related to enacted, or felt, stigma but also (as shown in the previous sections) it is sometimes related to a malfunction or affected aesthetics. This is complicated by ageing which can also influence how the individual may be socially affected.

The degree of social involvement may have an influence on when or if an individual realizes the social impact of CLP. The more socially involved the participant was, the more they started to realize the effects of CLP. It is not until the individuals face a particular social point where the effects of the CLP are more obvious and emphasised within a social activity of social incident.

> *"It was more, when I was at college, it didn't affect me as much, because I weren't talking as much as what I used to, but once I got to, when I started my job, it, I was more interacting with customers and my colleagues, trying to talk to them, and some of the customers that come in, fine, but when you get certain customers that try to tell you everything and you're trying to explain something to them in detail, that's when I found it more harder because I was talking more and then once I'd finished talking, my mouth just started drying up, so I had to go and get water, so it was just more."**INT#13.F***

> *"not aware of it but it wasn't really anything I thought about particularly until I was about 11 really, and that was sort of ... I went to secondary school and I started travelling lots more and meeting lots of new people. And I don't know, that year I can remember just noticing that I did look quite different and my sound is quite different."**INT#3.F***

Social involvement may also influence how the concerns change in relation to treatment priorities. For example, the shift from being more concerned about the look when younger, to being more concerned about speech improvement in order to become more socially involved and accepted within society. This also showed how the maturational effects of ageing presented.

> *"well for me I was more concerned about the physical aspect and how I looked, but more recently I've been more concerned, I don't know, more hopeful that they'll be changing my speech, just because I do keep meeting lots of people and it does sometimes get*

*difficult when they don't quite understand me or I have to say something again. And that has been quite a recurring thing throughout my life and it has been quite frustrating"**INT#3.F***

It is evident in the data that participants were socially affected by the CLP in one way or another. The range of stigma associated with CLP was broad and ranged between the two extremes from staring to physical abuse as a consequence of the CLP.

*"Well, maybe they're staring at you because they're bored and they're not paying attention," you know, things like that"**INT#17.M***

*"When I used to talk people used to stare at my teeth"**INT#13.F***

The experience of enacted stigma or bullying was evident; it was experienced not only in the form of verbal but also physical bullying. The data showed that the school environment was a place where different forms of enacted stigma took place from both students and school staff.

For example, one participant stated that her teacher was not sure how to treat her and approached the mother asking if there were any precautions or advice when interacting with her daughter. The same participant was excluded from sport at school because of her CLP but she stated clearly that this exclusion was her decision although it was also supported when she was given permission to be excluded because of the CLP.

*"when I was very little, about six and I come from a very small town and obviously the knowledge about cleft or anything that's not normal, in inverted commas, probed a lot of scepticism and questions and, so my primary school teacher asked my mum if she should treat me as a normal child and not really knowing that, its not a mental thing at all, its just some speech problems but nothing else really"**INT#7.F***

Another example was a child called unpleasant names who subsequently was physically insulted/assaulted because of the CLP.

*"In terms of comments, there'd be things like, I think 'spay-face' was one, I think 'flat-face' was another, 'lippy' was another because my lip would stick out, things like that. There were a couple of violent incidents. A kid once … there was one particular kid who hated me, he absolutely couldn't stand me."**INT#17.M***

*" at that point, other children became more conscious about being something different about my mouth and my speech, such as, I started to get bullied quite a lot at that age. When I was about fourteen, it got really, really horrific"**INT#17.M***

Schools could play an important role in managing social interaction focused around facial stigma. Surprisingly, as evident in the data, schools had responded negatively to enacted stigma such as bullying e.g. at one school teachers failed to stop bullying and ignored it.

> *"I used to go to my head teacher and all my teachers and they just told me to ignore it and from the minute I'd be in school to when school finished every day, I used to get bullied, constant bullying all the time."***INT#27.F***

> *"He once hit me in the face in a classroom and I had a bruised black eye. And the thing that I felt, that made me very angry was the staff at the school wouldn't really do anything about it. And this teacher saw me get hit in the face and she sent both of us out of the classroom, and I was shaking, I was crying, and I was only fourteen, and I was in a real state. And they then had to take this kid and move him into another classroom because he was then trying to get me in this classroom while we were alone. And I was yelling and shouting, "Somebody get this guy away from me now." **INT#17.M***

In contrast, some schools played a more positive role to limit and stop bullying. For example, one school helped a participant and stopped the harassment.

> *'' Because I sat and told them about it, because they didn't know what it was and I went into a lesson and I had the final straw with them and I told my tutor about it and two of the three boys happened to be in my tutor group, so my tutor pulled them in one morning and I just came in the morning to sit and read my book or whatever it was I was doing and one of them said that they were sorry and they just wanted to know what it was, so I sat and explained it to them and once they knew about it they wouldn't stop being sorry and they couldn't do enough for me. So yeah that was it really that was the end of it and after that they grew up."***INT#8.F***

Negative enacted stigma and 'bullying' had a significant impact on the individual's self-confidence and, as mentioned earlier, it was also reported that this social stigma increased the individual's self-consciousness.

> *"obviously you didn't feel very good, it's embarrassing especially when it's in front of your friends and you know it makes you feel, well it knocks your confidence doesn't it because you go along thinking that you're in your normal world and then someone's making a comment on you about just how you look"***INT#12.F***

The feeling of isolation was also initiated from the impact of having CLP.

> *"and not really sure about who I was, why I was being treated this way and feeling very different and very isolated"***INT#17.M***

In relation to facing stigma, people responded differently, in terms of approach, to enacted stigma or 'bullying' summarized in (Figure 6).
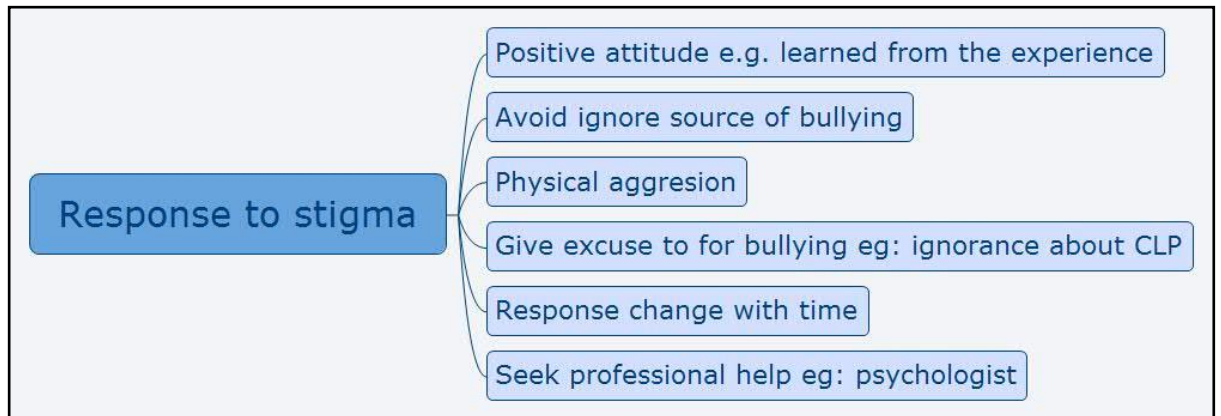
Figure 6: Different responses to stigma

Perceiving previous experiences of stigma through a positive mind-set was the way one participant managed the experience. It was stated the affected individual learned from the experience and actually bullying made them stronger and affected their personality in a positive way.

> *" Yeah, I've never really found it too taxing maybe, I was never really bullied for it, it maybe I'm quite self conscious with the surgeries and things but that reduces. I don't know, I think it's probably given me a better sense of humour maybe. I've learnt to be able to defend myself quickly with wit and stuff instead of just being quiet and suffering from it because yeah it's never really, I've never really been bullied. People don't really notice it anymore. Maybe as kids when you were less serious people would just come up to you and say, what happened to your lip and stuff and I'd just tell them but its never really been a problem because mine isn't really that bad, I've seen worse, but yeah I forget its there really."* **INT#11.M**

Avoidance of the source of the stigma was another approach. For example, trying to avoid the people that were the cause of bullying or just ignoring was another tactic used.

> *" As I was growing up I had people pick on me, but I think as you're growing up, people going through changes and people tend to be more immature when they're younger and people don't necessarily understand, but as I've got older, I think people have got mature and they became more sensitive, all my friends understand and are a lot more sensitive, you do have the occasional person who will say that rude comment, and you just have to ignore it and then at the time you will be like oh god why did he say that, and then you will be like oh god is it really that bad, but then after a while you just have to get over it*

but when I was younger, the comments were a lot worse but now things are better, and."INT#21.F

Another direct approach to cope with bullying was through physical aggression and fighting.

" For me, I am thick boned, I am tough skinned, I would knock them out, simple as. [Laughs] But, because I used to play rugby as well, I can take care of myself. But I think for a lot of people they don't have the tough background that I have so when someone does that it's quite scarring mentally to your soul as well, to your aspect of how you yourself, if someone tells you you are ugly because you have a cleft palate and you hear that enough for 20 years you are going to believe it, simple as. So, and I think it all depends on your own understanding whatever psychology is going on with their life and knowing that does not have to impact yours, simple as."INT#28.M

Interestingly, a lack of information and ignorance about CLP was given as a reason for bullying by one victim. This fuelled the participant's desire to promote information about CLP as they believed that people needed to know more about the condition. One participant worked for CLAPA to help publicise information about CLP. This also linked to the example in the first section where information seeker behaviour was related to gathering and disseminating information of CLP for the public.

" I didn't like the fact that people decided to make a joke out of it, but they didn't know what it was. So I found plenty opportunities since to promote CLAPA and Smile Train and they are sixth form charity. So I do quite a lot of work for them so that people in my school do know what it is, they might not notice that I have one but at least if they see it they then think well it might be there, so therefore if people want to question me about it they don't have a total faint idea of what it is, then you roughly know about it. But I like … because it did get to the point where I didn't want to be around those people and I avoided them on purpose because they were so disrespectful about it, something that I couldn't help and they just thought … I don't go out my way to bother anybody, and they were just being immature and they thought it was funny to taunt me about it."INT#8.F

The ageing effect influenced how participants responded to bullying. While they were fighting as a response to bullying at a younger age, they tended to ignore the issues as they became more mature. The improvement in speech, due to treatment, appeared to make social life easier as the patient matured.

"I look at things differently so when people ask me about my lip now, I'd be like ... I will try and tell them, I'll explain it to them, whereas when I was younger I'd be ignorant and have a fight with them"INT#14.M

> *"you know as I got older when the treatment has been working it's been a lot better I think when I was younger my speech was a lot worse so therefore it was a bit harder to communicate with other people"INT#1.M*

It is also important to emphasize (as will be expanded in the second chapter) the role of the psychologist within the CLP care pathway. Psychological support can be beneficial in overcoming the negative effect of bullying. For example, a participant was in an extremely bad psychological state at one point during his life when he was a victim of excessive bullying. He clearly stated that the psychologist at that time had been a major support for him.

> *"So I had psychiatric help, which was very important at the time because I was quite shaken and, on the wire, shall we say, a familiar phrase, and not really sure about who I was, why I was being treated this way and feeling very different and very isolated."INT#17.M*

People asking the participants about their CLP were also evident in the participant narrative. While some participants may have been happy to answer questions about the CLP, this was not the case for all participants. As stated earlier, one participant stated clearly that she was not happy to speak about her condition. Yet, conversely, another participant reported that they needed to know more about it to be able to tell people about the condition.

Other social impacts included the chance of getting a job and relationships e.g. a participant believed that his chance of getting some type of work was lower than others because of his CLP.

> *"Obviously from bully to side comments, all through everything. It doesn't matter what you do, if you are in an interview like now, whether it's for a job or something I am pretty sure that the other person is contemplating hiring you and your cleft palate comes into that equation. It is a whole thing, it is you and you have a cleft palate, it's always regarding the cleft palate or considering you have a cleft palate. So it's as much of you as it is everything else and it tends to be the defining point of you and other people, as a leverage to other peoples' acceptance. And some accept you have a cleft palate and some can't so it defines where you go in life, but not all the time. But it does have a hindrance to where you can achieve."INT#28.M*

Relationships may also have been affected as the CLP condition appeared to have a variable influence on the individual's aesthetics.

> *"Relationships, I've never really had a boyfriend but I don't know whether that's because of just me or my, the way I look I don't know. But I don't think it affects that."INT#10.F*

In contrast to social stigma, milder cases may not face any social effects because of the CLP especially where early treatments have resolved the problem. With respect to social impact that is related to the look, it tended to correlate with the severity of the influence on the aesthetics.

> *"I think specially like the image of my lip and stuff is not as obvious as maybe others I mean people can still notice obviously the scar and stuff but…it's never…unless I tend to mention it people…done ever really even ask like what is the scarring caused by or anything…because I think it's not a big issue as it is to other people who have had it…so for me it doesn't affect anything really…I wouldn't let it I guess because there is no need for it to…so its fine…"INT#5.M*

Based on the data, there are several influencers on the degree to which participants were affected by the CLP:

- Firstly, the personal experience faced because of the CLP such as the experience of bullying and how the experience added a negative dimension to the overall CLP experience.
- Secondly, the degree of severity of the CLP and/or realisation of the treatment results and outcomes such as mild cleft palate only and cleft lip only cases where no significant impact was reported by participants.
- Thirdly, the personal attitude toward CLP, influenced by how the participant was supported during their social development and how much support was provided especially with respect to psychological support.

Fortunately, bullying tended to decrease over time for the following reasons: The change of environment from school to work (a more mature environment) coincided with the change of the participants from less mature children to mature adults. In addition the positive impact of psychological support could have contributed to how participants face bullying and make it less likely to happen.

> *" I think because of quite a few different reasons that all sort of merge together and create this one specific, I don't know, situation. But for the most part, I started noticing improvements when I was round about eighteen. But for the most part it was because, one, I think a lot of people grew out of the situations, you know, the mentalities that they were in. But secondly, I think I learnt how to deal with it and learnt not to take people seriously. And then not to get offended by people. And it's something that took a while to get used to.*
>
> *And when I was about nineteen I went to – I was, no, I was 20, my mistake – I was 20, I went to university and I had already worked for a year doing some other stuff, so I'd met a lot of different people and I'd become far more confident about things. And when I went*

there I realised that nobody really cared about things like, I don't know, Geordie-face, or anything like that, everybody had this different attitude. It was a more mature crowd. People weren't so judgemental, people were very accepting of other people and I realised, also I think I realised nobody really notices it anymore. These aren't fifteen year-old kids that will go up to you and will notice it straightaway. They are adults, like yourself, who just want to make friends and meet new people. So going into that environment completely shifted my perspective and made me realise that there are a lot of nice people on this planet and you haven't got to worry about things like the way you look."INT#17.M

In this section the data presented the impacts of living with CLP. These included the aesthetic, functional, psychological and social impacts. It was also evident in the data that CLP treatment could be considered as part of the impact of living with CLP as it may have caused a disturbance within day to day life. CLP involves a long and complex treatment which could be viewed sometimes as a burden and therefore can be a negative impact of living with the CLP condition. In the next section, how participants perceive their care will be explored.

5.2.5 How do cleft participants perceive the cleft care pathway?

The views on how the participants perceived treatment for CLP could be divided broadly into two groups, broadly as positive or negative (Figure 7).
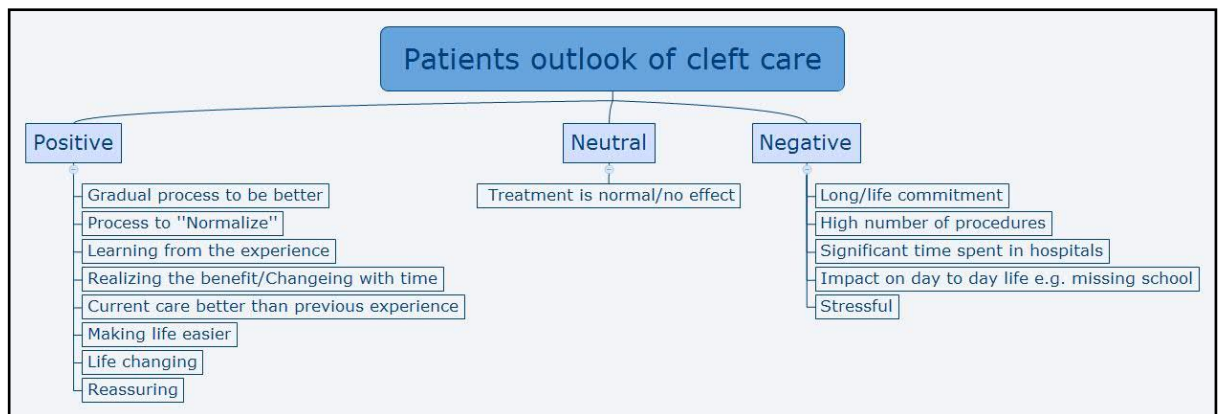
Figure 7: Patients outlook of care

Many reasons could explain why participants perceived treatment as positive. Looking at the care as a gradual process of improvement over time can encourage positive outlook. As described by one participant:

" just got better over the years..I know it is a gradual process it doesn't happen in a year in a space of a year or anything like that...I know that is through that years I don't know ...I just know that series operations is more gradual and through that my speech became better and so i can communicate with people and..I think I'm a lot less nasally as well"INT#1.M

In addition, another reason is that treatment is helping them to become 'normal'.

" That's because afterwards a lot of things did change. There were no more questions about my jaw; I felt like I could finally be a normal person. I already was normal but, you know, properly, properly normal, and things like that."INT#17.M

This picks up on early discussion about feelings of difference and wanting to be 'normal'.

Living with the experience was considered as another possible way in which the participant 'coped', and took something positive from the experience.

" so that's all part of character building as I said... you learn from what you did before...but I'm really happy I had the operation .."INT#1.M

The lens of perceiving treatment can change with the ageing effect. One participant suggested that young people tend to think short-term while older individuals think of the longer term benefits and therefore their view toward the treatment tends to be positive. When participants can see the difference after treatment, this could assist in the perception of treatment as being positive.

" very..no negatives to feedback on…all positives…whether that's helped with the fact that maybe yeah because I'm a young adult as such…where is obviously..when I would be in the waiting room and see lots of younger younger people like little children in for treatment and stuff…obviously maybe they wouldn't quiet healthy have the live ahead of it…because I suppose when you are younger younger you only think short term…whereas me I always thought of the long term benefits so..always just though as a positive…never negative always good…"INT#5.M

Comparing the current care with an previous care or poorer care system may help to perceive current treatment as positive because of the comparison they are making.

" It's been very, very positive, definitely. I mean I haven't the comparison, what I experienced before, it has been pretty much plain sailing."INT#7.F

Treatment was also perceived as making life easier.

" Oh it has made my life considerably easier definitely."INT#8.F

Or as a life changer:

> " and then I went all the way through school with that and as soon as I left school I had reached 18 and I'd stopped growing so then I had my final operation to correct my jaw. And that has changed my life I think. I really am a completely different person. I like the way I look. I'm happy when I look in the mirror and yeah, I guess it has changed me."*INT#10.F*

Furthermore, the CLP care pathway was described as reassuring for CLP participant because of the ongoing provision of care.

> " It was reassuring, it reassured me that they would always be there, even now they keep saying if this goes wrong or if you ever feel the need to get a different colour tooth caps you can always come back and that's lovely to know that there's always someone going to be there and that I more or less wont have to pay or wait because they'd understand the context …"*INT#11.M*

This shows the importance of continuity, as will be explained in more detailed in the next chapter.

One participant pointed out that treatment was neither positive nor negative, just a continuous part of life.

> " I suppose it's just all the appointments and having to keep going but it didn't really impact my life having a brace that much because so many people have braces now, it's just normal,"*INT#9.F*

In this example, treatment and being part of the care pathway had become normalised.

Negative views were reported because of the following reasons. One reason was that CLP patients received treatment all of their lives in a chronic way with a significant number of operations.

> " If you are not born with that you don't go with that, you don't have to deal with that. It becomes a long ordeal if you are born with something regardless. It's an extra little thing you have to do throughout your life. I am 20 no so for 20 years I've had this so it's a course that's going to be a long ordeal."*INT#28.M*

The large number of operations led on to participants spending significantly more time in hospitals than non cleft peers.

> " its not a mental thing at all, its just some speech problems but nothing else really, but from my experience, I guess you spend more time in hospital than the average child,"*INT#7.F*

In addition, the impact of treatment on day-to-day life such as missing school had a negative impact.

> " the only thing it has affected it my exams, a couple of my exams last year were on the same day as the orthodontist so I had to say I have got an exam and I need to come another day and there might not be an appointment for another week which will mean that someone else will have to take the day off work to take me which will mean I will miss another lesson in school. But it was worth it in the end so it hasn't really … its quite nice coming here."**INT#8.F**

It was also reported that treatment could be stressful and long in duration with significant disruption in time planning and commitment.

> " It's quite stressful, especially with school and college, because I had to miss a lot,"**INT#9.F**

In this part, it is apparent that a range of views toward the CLP care were present. The data presented the different reasons that explained why participants perceived care differently. This was mainly influenced by how the care affected their lives and how the care was helping participants to change their life, highlighting the importance of understanding the care pathway in context. In the next section the data will explore what participants remember from their treatment.

5.2.6 What cleft patients remember about their treatment

What we can see based on the data is that memories related to treatment can be grouped into three major parts (figure 8):
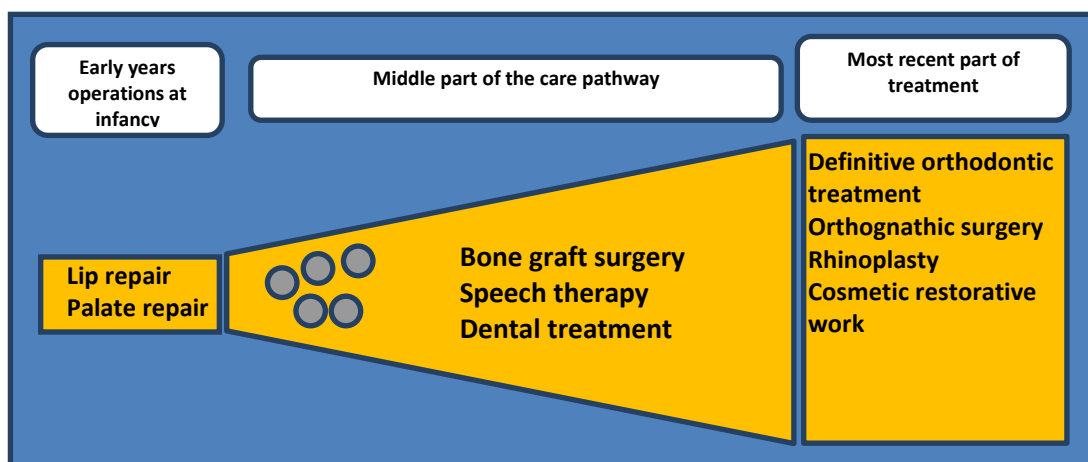
Figure 8: Conceptual representation of what CLP patients may remember from their treatment

Operations at infancy

Participants did not remember aspects of care related to procedures around infancy but they incorporated it in their accounts based on what they were told by parents.

> *"My parents told me when I was a baby that it was difficult for them because I had to have some kind of massive brace thing. I don't know what it is called medically, but I know that it was difficult for them and for me as a baby"* **INT#7.F**

> *"Well at the beginning I suppose I had the operation. I think I had a few, I was a baby, I can't remember. I suppose"* **INT#20.M**

> *"Yeah, it was, I think I had, I must have, I think I had more than one operation, I think the first one was when I was quite young, it was unsuccessful, I think I was in hospital for quite a lot of time when I was quite young, I remember my dad saying, but I remember having a, I remember in photographs that I had things on my arms, casts, so that I couldn't put my hands in my mouth, but the operation wasn't successful and my speech didn't resolve"* **INT#5.M**

With respect to the feeling of not remembering the early years surgeries, it was stated that this was a positive feeling because the patient doesn't really want to remember these times and the pain and trauma associated.

"but actual pain of it or sort of lying in bed and being in pain that's all goes away really which is a good thing because I don't remember it if I remember the pain when I was that young but I know it was painful because it was an operation but I don't really remember the pain." **INT#1.M**

These are of the most invasive parts of treatment is only remembered through proxy recollections of family members. This may affect the overall view of treatment.

Middle part of the care pathway

Memory and recall increased with age with participants having wider (but often vague) memories of procedures focussing on and around bone grafting surgery. Participants also tended to lose the sequence of treatment or the exact timing of specific procedures. Also some participants mentioned that during childhood they had not vividly remembered the actual major procedure (e.g. bone graft surgery) however they did remember brief memories of side events related to the surgery such as collection from school on the day of the surgery, the play room or what they used to eat or drink at the hospital.

"I think I remember like a little glances of being in the hospital..so I remember a toast I remember like a Nesquick..I remember being in the [games]room I remember bits of being lying in hospital but I really I don't remember things a part from that..I remember having what you call it...it's called having a bone graft...I had a bone graft in year 7 or 8 I remember that because I was on the list to be older to be 12 ...so remember not being able to walk that well and then being fine in a couple of month" **INT#17.M**

"the one when I was about 3 I do remember the day like when I went for it…and obviously I stayed in hospital I think it was overnight…but I mean too young to really understand a great deal and I can't really say I remember many feelings or anything about that I just remember the moment when my mum and my dad came and collected me from nursery school and we went there…so other than that I don't remember any feeling or anything regarding that it's just I remember the moment of having it done…but so other than that…I don't really remember much experience in terms of the care path from when I was younger its only then when I started the orthodontic work" **INT#3.M**

This is interesting as again it suggests the importance of the context around the care pathway.

Most recent treatment

Finally participants gave more detailed accounts of their recent orthodontic or orthognathic treatment; they could remember first-hand the procedures clearly and in some instances could recall the date of the surgery. However this was built on the early proxy memories of parents.

> *"It felt really, I don't know how to explain it, it felt really tight and my teeth felt very heavy I think if I can recall, and yes it rubbed a lot on my mouth, it was very uncomfortable, it hurt to eat because you know it felt very tight on my teeth and also like caring for my teeth I had to be very careful and I have to be very thorough when I brush my teeth, I have to make sure I have the mini toothbrush thing to clean everything and make sure I take care of my teeth and not have food or drink that will, that they recommend you having."* **INT#12.F**

> *"…and the last one which was last year on the [Date] yeah last year that the one when I had the jaw surgery so I remember that very well"* **INT#17.M**

What people remember and how they recall different events is clearly important both when looking at experiences, and also if trying to measure satisfaction over time.

5.2.7 The impact of treatment on the day to day life of cleft patients

The long term commitment to CLP care can impact on the day to day life of the CLP individuals. The continuous and frequent appointments cause disruption to school attendance and thus affect individual work commitments; recovery after operations may also negatively affect school and work. This in turn may cause a negative impact on family members.

This section will present issues related to the impact of treatment on school and work, the possible burden on parents and family and finally the apparent important timelines for the patients that were evident in the data and that appear patients tend to plan their treatment around. (Figure 9).
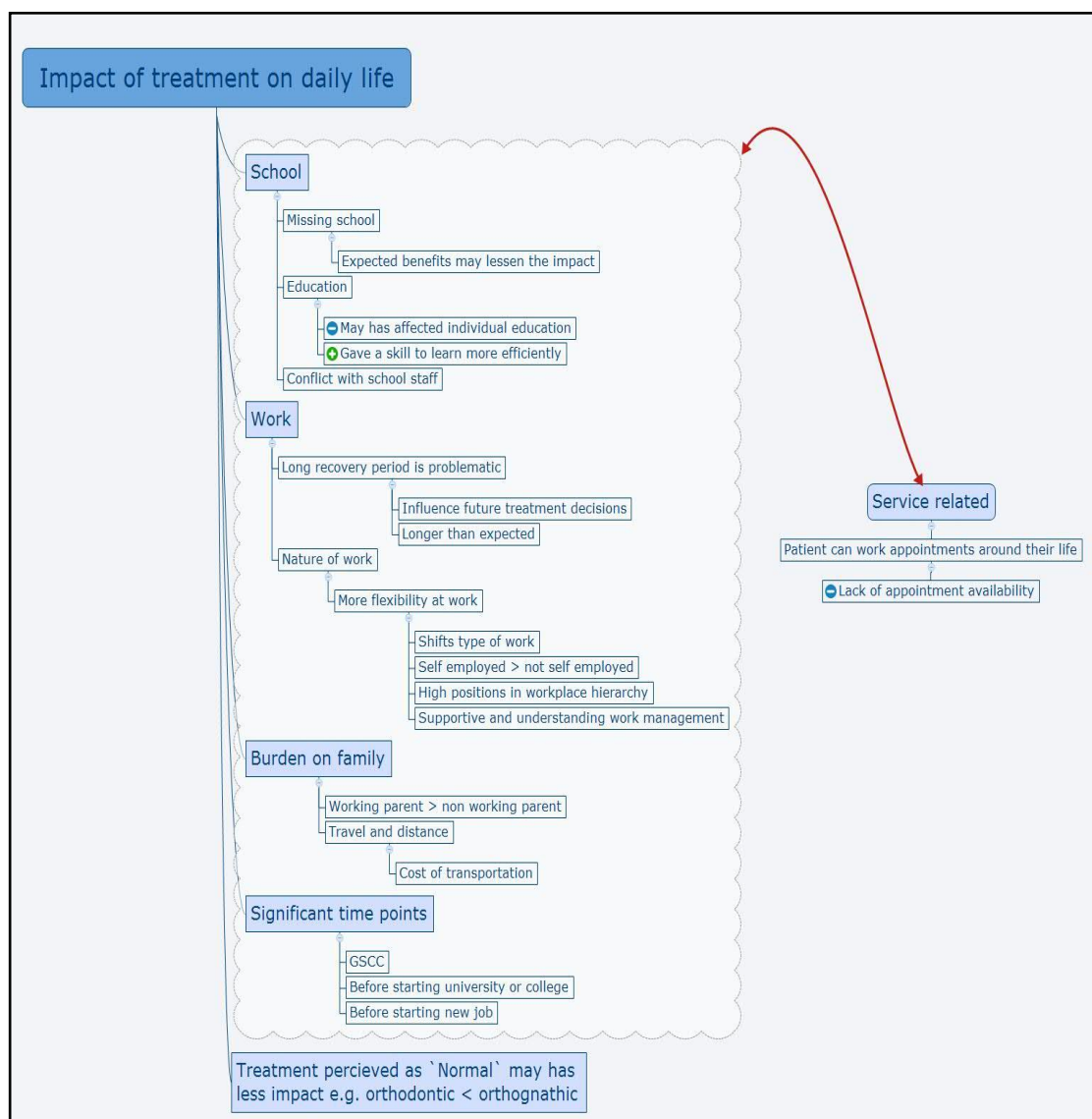
Figure 9: The impact of CLP care on day to day life

In relation to school, attendance at treatment appointments conflicts with school time which then leads to missing school days or missing some lessons.

> *''well…I would rather to have it like in the half terms…or in the holidays so I don't have to miss schools…or like after schools but I didn't…I was never able to get them after school…so i think generally I get them like first thing…so I would be able like the first appointment and then goes to the school from …''***INT#1.M**

> *'' Yeah, I suppose sometimes I have to miss school because the appointments are in the middle of the day there's ... I have ... I can't make it back to school and stuff like that but not particularly. But I suppose it did a little bit, yeah.''***INT#15.F**

Some aspects of treatment, with a long time commitment, can have a significant impact on school attendance. For example, it was reported that 'distraction osteogenesis' caused a significant disruption for education because it required a longer period for recovery due to the necessary commitment to turn the screws regularly and the pain associated.

> *'' I think the two big-ish procedures I remember having have been quite recently, and the first was the distraction, where I had to break my jaw a little bit each day for about a month, I think. And the actual operation was fine, that was easy to cope with, and I actually went back to school a week after I had the operation, but it was actually that week that I started turning the screws and started actually the process of breaking my jaw, and it became a lot harder after that and I did have to go home to recover, and it was quite painful. Also just because you couldn't really have a chance to break a bit and then let it all heal and break it again, you kept breaking it. So that one was really, really hard.''***INT#9.F**

Missing school lessons affected participants' education, both in terms of workload and friendships.

> *'' to be honest with you ... schooldays ... treatments, I never really got to settle in to school. I never got my place, do you know what I mean, I never really fitted in. I fitted in but I was not always there every day ... coming treatment obviously then operations ... take more time out of school ... but I wouldn't say I'm not bonding other people, I'm losing my work so obviously I'm not learning properly, I'm not ... I'm always behind, I'm not ahead so yeah that did affect me and in ... yeah it did, it did really affect me, yeah.''***INT#16.M**

In contrast, others believed that their education had not been affected.

> *'' well I have to miss like…quite a few lessons…to come here and it take pretty take about like half a day…to come here and go back and so…like when I was going to my gcse it was…annoying because I needed to catch home work and it was hard like maybe to*

*understand something sometimes…and…that it wasn't wrong but…I've…it's not like affected me too much"I**NT#4.F**

*'' A small, small, small amount, I wouldn't want to say it affected it negatively because there were opportunities for me to catch up like I've missed a lesson that's it, there were opportunities for me to catch up and I should have taken more you know sort of stuff into my own hands to catch up rather than blame it on cleft."**INT#24.M***

Surprisingly, it was reported by a participant that missing time in education helped to develop skills around time management and efficiency in order to keep up to date. As stated by a participant:

*'' RES: Yeah, that was a positive thing because now I'm able to pick up things very quickly … and I'm not a systematic person and I never was probably because of that, because I was off sick a number of times at school for long periods and also at work and that probably made me catch up much quicker than the average person because you have to, well you have to catch up if you're ambitious and if you want to get somewhere in life, that's what it takes, so yeah."**INT#7.F***

Committing to treatment could also be a significant burden for parents. If schools do not show understanding with parents of affected individuals, this may add greater stress for parents. As the example shows, a participant reported that his mother used to argue with his teacher because of his continuous absence from school because of the ongoing treatment.

*'' well I remember he had an argument with my mother because my mother was more concerned about my health care but the teacher was more concerned about my education …and I was very on the fence..you know I just thought it was a silly argument because you know I am going to have the healthcare one way or another and it's not affecting my education so much"**INT#6.M***

Travel and long distances to attend appointments at CLP centres can also be an issue. Long travel times to get to the CLP centre may lessen the chance of their individual to go back to school after his/her appointments and may contribute to more missed education.

*''It was more coming up to [name of hospital] for my second lot of treatment that has caused more disruption because I've had to have time out of work and you know get the train from [Name of city] to [location] which is over an hour's travel."**INT#12.F***

The expected benefit of the treatment could help the individual to be patient about all the strategic hassles and frustrations related to her treatment including missing school classes.

'' I did decide in the end to have all the operations and everything, that was me because obviously its my body, but again back then I saw pictures of myself and then I saw my friends or my family and if there was anyway that I could look a little bit more like those people then I was going to do it and if it meant that I had to miss a bit of school here and there or give up three or four days to have an operation and I was going to be knocked out anyway then it didn't matter too much to me. So I wanted to look more like them so that is why I did it really.'' **INT#8.F**

In relation to work, a long recovery period could affect jobs negatively. This, or any other day to day life task, may influence the decision about whether and when to go through any future operations. A more stable workplace environment would be considered together with placing the treatment whenever it would be convenient for the participant's day to day life. Also, orthognathic surgery recovery periods appear to be longer than expected and have a more negative impact on an individual's day to day life. This was emphasized when one participant talked about taking time off work for recovery and then realising that the time is not enough. This placed the individual under huge pressure when they needed to go back to work as soon as possible without being fully recovered especially in contemporary workforce pressures.

'' I mean if I have the operation around then you know I'll be in a stable environment...like you know I'll be used to everything..I'll do it and then..you know if done..I'll take a couple of weeks off of work..and then I know exactly how it works at work...you know say right now still learning I'm still developing so right now it will be a bad time of having an operation like I've learned from my last time...the last time I recently started the job I had the operation I was still recovering when I went back...they didn't really know me that well so it was all a bit of nightmare really..it was a bit of train crash that whole experience so I want to learn from that and so yeah and sort of adjust myself a little bit so ill have a good time for myself you know because at the end of the day work is work I don't work to live you know..no... I don't live to work..I work to live ..so I want to do the operation at a good time for myself but not having a bad effect for my job...so I take time off ..I come back and start working again.. as opposed to ..you know..'' **INT#1.M**

'' so it was literally like normal and I didn't really [register] you know I thought I will just take 2 weeks off and then be back to normal but it wasn't the case you know I really stopped move you know ..It was like going at a hundred mile at hour hitting a brick wall you know after that I couldn't do anything for ages...I went back to work too early...I was a bit..You know I can barely talk... you know and you work in advertising so you need to talk...so it was a bit of nightmare...you know the whole sort of impact of the operation..So...'' **INT#1.M**

The nature of the type of work may influence the impact of treatment within day to day life considerations. Flexibility of working times could be helpful. For example, working in shifts and the ability to swap shifts was seen as advantageous.

> '' basically my work was a lot of shift work so it was…I kind of have a lot of flexibility with my shifts anyway so…obviously you find of your appointment a lot in advance so…I never had to have anything conflicted …and then obviously the day that I would have to have the appointment..I would still just treat it as a general day off anyway''**INT#5.M**

Also, the data suggested that self-employed individuals may possess more flexibility to manage their work within the framework of treatment. Self-employed individuals would have more control over their time and this could help control and plan ahead for major aspects of their treatment.

> '' like I say even taking time off work because I'm self-employed it wasn't too bad because I wasn't actually using up annual leave to have an afternoon off, I'd just rearrange my hours and Monday's normally my day off''**INT#12.F**

Although the financial penalties of taking time off could be greater.

Having a supportive working environment and supportive and understanding senior managers, may also help to lessen the impact of treatment to day to day life.

> '' you think you can do things but you can't really and the time then comes much quicker and at that time I had a very understanding boss as well, so when I came after my sick leave, after that month off, for the first week I came only, I think it was four and then six hours a day, so I was able to get back into it on a slower pace and I could avoid commuting times where the trains were packed, I can always sit down and have a relaxing journey to and from work and that was very helpful as well that I was able to get a couple of hours of work in the morning and then go into the office which I found very helpful as well.''**INT#7.F**

As previously stated, long recovery periods can be problematic with jobs especially if the information provided was not reflective of the actual experience. For example, one participant has faced a negative experience in relation to his surgery in that he stated that he had lost his job because of the long recovery period that was associated with his treatment.

> '' Well I think because I had an operation which meant I couldn't eat, which meant I was very lacklustre of energy, which meant applying myself into a job is a task because you don't have the energy yet to do that, which meant your standards slipped which meant they fired you. What can you do? Get another job like any other normal person. And I now work in a sushi bar making sushi. And I am going to university in September. So it all

depends on your own mind-set. If someone wants to fire you that's their issue. You know fully, full well of your capability, of what you can do.'' **INT#28.M**

Treatment also had an impact on employment for parents of CLP children. It can be helpful, if one of the parents is not working, to take the child patient to treatment.

'' well it's not been too bad because she has not been working…and so sometime she needs to get back [back of her] from time…or…like recently she has start working and…so like sometimes if my mom unable to take me here…I would come with my dad…and then he go after like he takes sometimes off work maybe and he gets back to his work at the end'' **INT#4.F**

In addition, being in a higher position in the workplace hierarchy would give more flexibility to come to clinical appointments.

'' The regular, I missed, it didn't really matter but it was only primary school but if I missed Friday afternoons off school, I suppose if people cared enough about their school life then it might affect them in that way. I suppose my dad had, my, luckily my dad was the Manager of where he worked, so he could get the time off, so he'd take the afternoon off and I'd get the afternoon off school but that would have affected, anyone else finds it hard to get the time off work or is self-employed, it might have been difficult for them to have to come'' **INT#11.F**

Some aspects of treatment can place a burden on the affected individual`s family especially in relation to travel and distance. The patient may appreciate if an option was given in regards to which centre they preferred to have part or all of the orthodontic treatment in as chosen, more logistic centre to reduce the distance issue.

'' I think maybe the option to be seen somewhere closer to home so with my orthodontist thing maybe they could have referred me to a hospital more local to me rather than having to come over here, but I know now that they're having me see my local orthodontist now in the future but I think maybe it might have been more convenient to maybe pick up my retainers at my local hospital.'' **INT#20.F**

The cost of travel to the CLP centre can also be a burden to a family with a cleft individual with it being reported that it is expensive to come to the centre.

'' But I guess the cost of it, coming up here. That's another thing you have to think about. The train ticket up here and back, yeah the cost of it really.'' **INT#10.F**

Lack of appointment availability or lack of flexibility to reschedule, or move the appointment to another day of the week, to avoid conflict with other life activity may add more difficulties to the impact of treatment into day to day life.

*'' This has been generally good except having to wait for appointments some times.''***INT#15.F***

*'' It wasn't too much of a problem, sometimes it was a little bit frustrating because [name of orthodontist] would say go and book an appointment for four weeks' time you know we'll tighten them in four weeks and then [name of staff] would say we haven't got anything for six or eight weeks, so you know sometimes your treatment was slowed down a bit just because of availability of appointments which obviously when you want your braces off you just think I wish I could have that appointment sooner.''***INT#12.F***

*'' I think it's just booking appointments just so like you could, like they could be in on like a Thursday so you can phone in on that day just to make it a little bit more easier instead of waiting the whole week so just given us different options, different days to phone in instead of only phoning in on a Tuesday.''***INT#22.M***

It was important to notice that CLP care was strongly integrated into CLP patients' lives. For example, one participant reported that some treatment happened on their birthday. Whenever there was a conflict between treatment and day-to-day life commitments, the patient would work around it by managing priorities.

*'' I had lots of operations to put grommets in my ears. I once had an operation to remove one of my teeth. Can't really ... I don't remember why but I did. And I think that was actually on my birthday''***INT#3.F***

*'' It's hard to sort of balance that because you only have certain days when the orthodontist or a surgeon is in, and you try to make the time as convenient as possible but sometimes you have to just miss a few lessons which can be disruptive, and you have to catch up later, but I was normally pretty good at doing that so it just became something I got used to really.''***INT#3.F***

Frequent orthodontic visits may be perceived as a normal thing with a lesser impact than orthognathic surgery or other major sections of treatment that required longer recovery periods. This may indicate that what is considered 'normal' may hold less impact on day to day life.

*'' because the braces is part of my life a normality but at the time when I had the operation there is a lot of change going on..''***INT#1.M***

Based on the data, it appeared that there were a significant number of time points when the patients tended to time their treatment. The aging process is also evident in determining the important time points as the patient grow and these develop to become ready for treatment.

The time when children undertake their General Certificates of Secondary Education (GCSE) examinations is a significant point in education and can be considered as an important time point for CLP individuals.

> '' It's purely because I'm a bit forgetful but these two years have been massively – at school it's been really hard. We've had our GCSEs and all that and so when I go to a doctor, I just feel like I need to get the appointment over and done with, just get the information or get the treatment and go home, go back to work or whatever. Now that I've finished that massive piece of work the questions are all starting to get to me now. Now that the treatment is over, in terms of braces, I feel like I should have asked these questions before.''**INT#2.F**

> '' I mean while I have had to have lots of time off which has been difficult, particularly with my friends and trying to navigate that, and also school work, particularly as I came up to my GCSEs, that's been quite a challenge to work out really.''**INT#3.F**

After that point, the period focussing around the interval before they are starting university or college after school acts as another key time. Any major treatment that may need a long recovery time may be considered and planned for before they start the course/next stage of career development.

> '' and so I specifically took a year off from…before going to university because I thought..since all that the orthodontic treatment and the surgery was during that year''**INT#6.M**

The same applies to the first professional appointment after graduation especially when there is significant workplace competition for posts.

> '' Okay, I said if you can keep me, I have a surgery because I got a new job offer and I was about to postpone the surgery because of the new job but my old boss said that he was very keen to keep me and I said here's the deal, I can stay but I need to have as much time off as I can during my surgery, being fully paid for that, yeah, sorry''**INT#7.F**

Knowing this, it would be helpful for the clinicians to consider these points when discussing the major aspects of treatment such as surgical interventions.

In this part, the data presented suggests that treatment has an impact on participants' lives. This includes the impact on school, jobs and their family. It also showed that significant time points from participants' perspectives may affect treatment as they plan their treatment around them. This shows that participants do not simply see treatment as only clinics but as 'clinics' within their lives.

5.3 Summary

In this chapter, the findings showed the participants understanding and feelings about the condition of CLP. It highlighted the impact of CLP on day to day life including aesthetic, functional, psychological and social impacts. In addition, it showed how participants perceived the CLP care and what patients remember from their treatment. Finally, it concluded with examining how CLP care impacted on the day to day life of the affected individuals (Figure 10). Exploring these areas provided a good understanding of participants' backgrounds and how they influenced different aspects of the CLP care. This is evident in the data presented in the second chapter which focuses entirely on treatment and the experience of the definitive orthodontic/orthognathic treatment within the CLP care pathway.
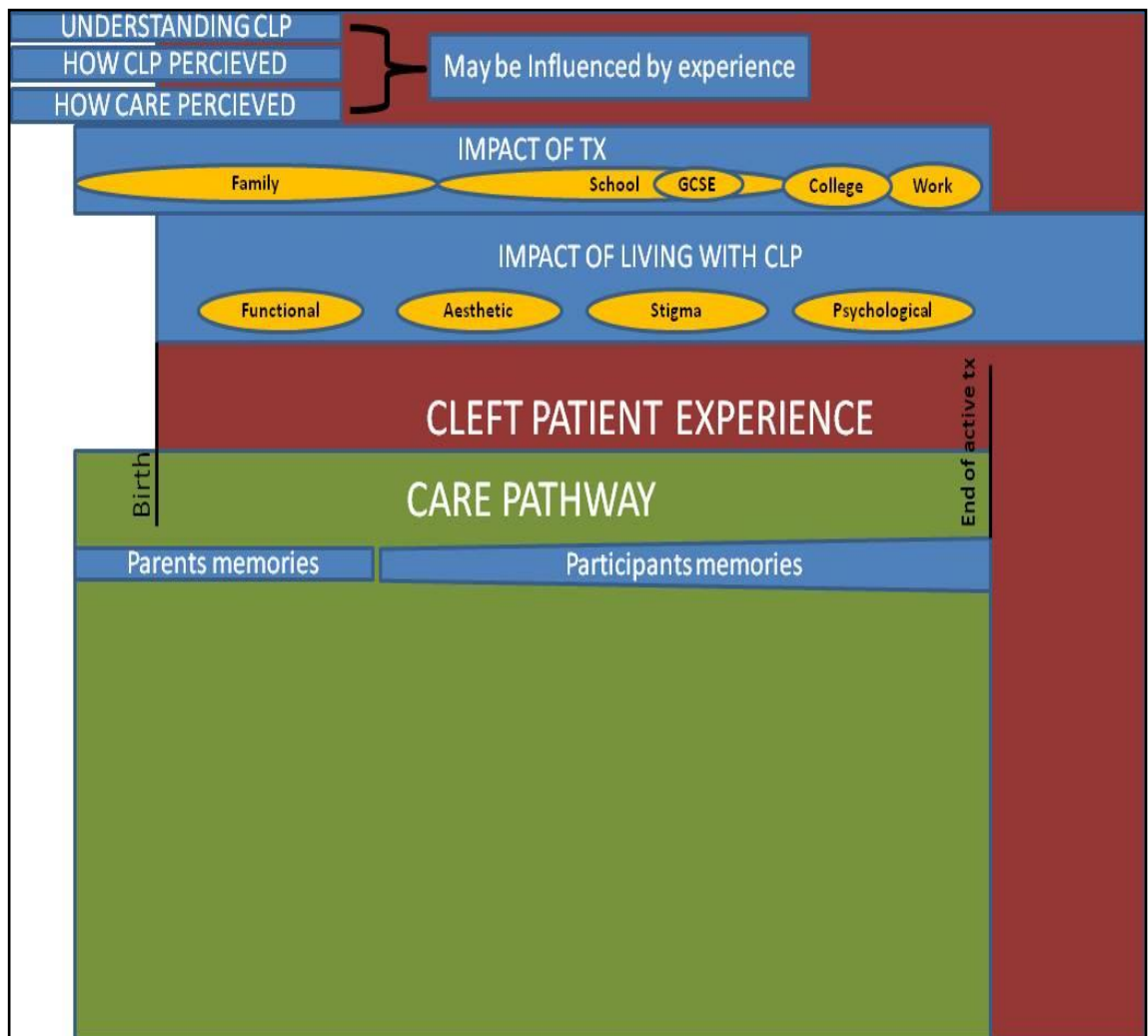
Figure 10: Summary of chapter 1

6. RESULTS – CHAPTER TWO

6.1 The care pathway and cleft treatment

The results presented in the previous chapter focused on the experiences and understanding of people living with CLP and the impact of the condition on daily life. This chapter builds on this to focus specifically on CLP care and experiences of the CLP care pathway including the definitive orthodontic/orthognathic treatment. It is worth bearing in mind here that alongside general questions about treatment, participants were asked specifically about their orthodontic treatment and so results here may be skewed towards this particular aspect of treatment. The results in this chapter are presented chronologically moving through the different stages of treatment and are centred around three time points; before, during and after treatment. The final part of this chapter draws together the daily life and care pathway experiences to highlight overarching themes and build a more nuanced and holistic picture of CLP care in context from the perspective of those who have been through the process.

It is important to note that the information in this chapter focuses on experiences of treatment rather than on movement through the care pathway *per se*. The vast majority of children and young adults living with CLP have been on the care pathway from before birth with the widespread prenatal screening programmes that are currently in place. Thus the care pathway can be seen, in the broadest terms, as a part of daily life from birth to young adulthood when active treatment usually stops. This is reflected in the results presented in the previous chapter. This chapter, then, focuses specifically on episodes of treatment, looking at the experiences of participants before, during and after treatment and culminating in the definitive orthodontic treatment that, in most cases, heralds the end of the journey through the care pathway.

6.1.1 Overview of the key themes

The treatment related themes were conceptually grouped into three major themes (before treatment, around or during treatment, after treatment). Under each of these three major themes, there will be a list of subthemes relevant to this arrangement:

Before treatment

- Reasons for treatment

- Expectations

- Concerns

Around or during treatment

- Hospital environment

- Staff

- Information

- Decision/Choice

- Effects of treatment (orthodontic/orthognathic treatment)

- Coping

After treatment

- Benefits of treatment

- Satisfaction

6.1.2 Before treatment

This first section explores the reasons that active treatment is undertaken and expectations and/or concerns about the treatment prior to its commencement. The data is split into three sections:

- Reasons for undertaking treatment
- Expectations
- Concerns about the treatment

Each sub-theme is presented in turn and illustrated with quotes from the interviews.

6.1.2.1 Reasons for treatment

In this part, the reasons given can be grouped into three main areas: aesthetic concerns; function; treatment availability and cost (Figure 11). Whilst improving function would seem the most obvious reason for seeking treatment from a clinical perspective, it is interesting that reasons relating to aesthetics were cited consistently by participants.
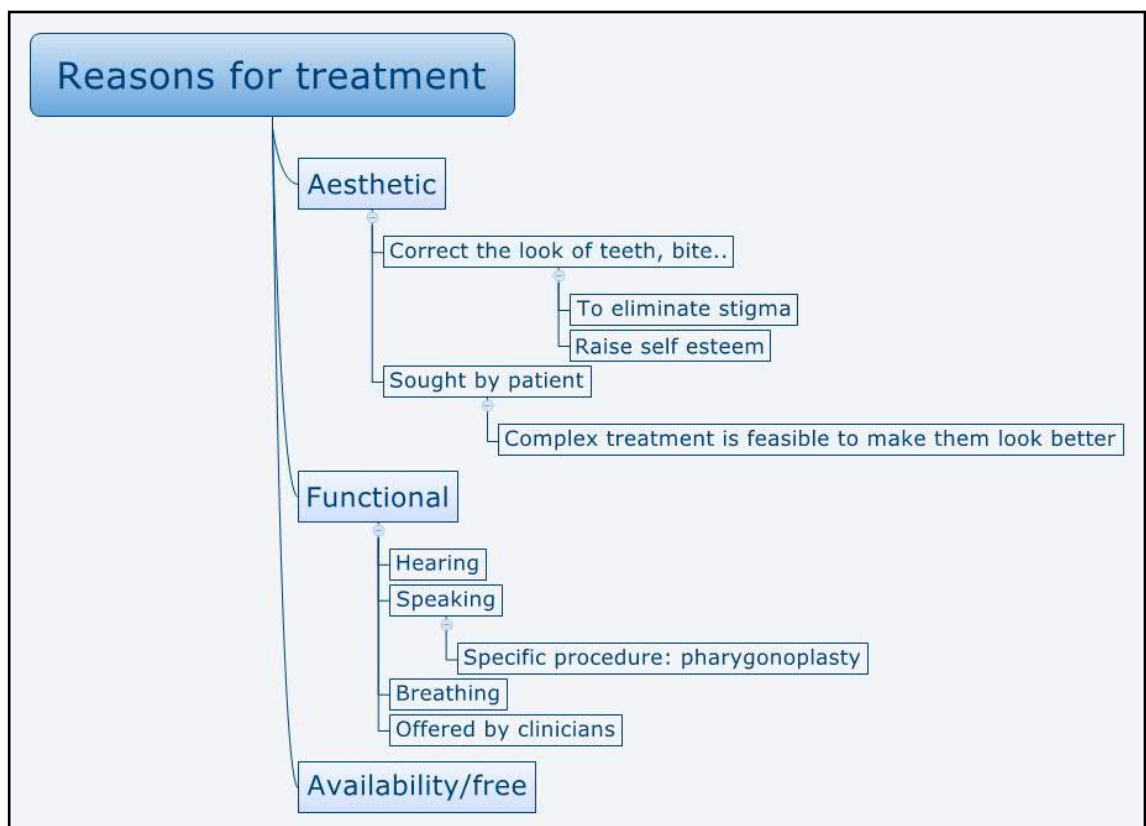
Figure 11: Reasons for treatment

Aesthetic reasons for treatment are present throughout the data and there is a focus on the look of teeth and the importance of orthodontic treatment. Orthodontic work and the look of the teeth were raised as issues far more frequently than other aesthetic issues around the structure of the face and nose. This may simply be an influence of the study introduction and information sheet or may be because orthognathic treatment is needed less often, reflecting the profile of the participants in this study. It could also suggest the importance of the teeth in people's overall view of the aesthetics of their face. This is evident in the number of participants who focus on the need to correct the look of the teeth through orthodontic treatment, and also, to correct the bite and profile of the face through orthognathic treatment.

> *" I prefer my teeth being…a lot of it was vainly to do with aesthetics just because I remember my mouth was possibly the part of my mouth which I was least happy with as it were so..if I was able to have braces and sort out the under bite then I just felt I wanted to and now I prefer how I look both with the jaw and with my teeth I prefer it…in practical terms, in terms of eating and speaking…I don't think it made incredible amount of difference…I mean it may have done..it may have made it easier for me to eat and speak but because I spent my entire life before that eating and speaking in a certain way I was entirely used to it and so didn't seem any to be any difficulty I was perfectly comfortable eating and speaking as I did…and so now I've been change I've just kept doing again…and so it's not been so easy or like it's been noticeable it's been the same in practical terms so…but aesthetically its very different and happy with it it's what I wanted so.."***INT#6.M***

> *" Because it needed vast improvements, my teeth were horrible they were crooked, they were a funny colour, I have had some of them whitened they were all coming up in the wrong places. ………. Yeah so that is why I needed care because there was no way I would live with those teeth now."***INT#8.F***

> *" All my teeth were quite bad, I had quite a few gaps and like baby teeth and adult teeth and stuff like that so it was quite ... I was bad like always that [unclear - 00:10:17…attendant] just so that because I wanted my teeth to be nice. "***INT#13.F***

> *" It's ... my teeth all over the place. I'd prefer to have nice straight nice teeth. Otherwise my teeth isn't right, my smile isn't right (laughing)."***INT#16.M***

> *" Well I knew that my teeth were, there was something really wrong with them, because I had teeth coming down, they were like fangs, they were horrible, and I knew I had to get braces, because people said stuff about it, they said that I should get braces and I was so glad when it happened to be honest, because I knew that one day they would be straight and it would help me in the future."***INT#18.M***

One participant explained that correcting the look of the teeth was more important than the look of her nose.

> *"Yes it's just cosmetically really I don't know whether it's because people have, no it's not because people are far more conscious nowadays, I don't like the look of my teeth and I know I'll never have the perfect smile, you know but yes I am conscious of my teeth especially if I see a picture of myself I go oh you know and it's never at my nose it's just at my teeth that I don't like. Like I say I can accept you know that my nose won't be straight or anything but my teeth I just haven't been happy with and I know there are people with worse teeth than mine as well."* **INT#12.F**

This illustrates the importance of teeth for appearance, and expectations of how teeth should look. The importance of appearance in relation to treatment also ties in with the results presented in the previous chapter which highlight the impact of appearance on self-esteem and also on the likelihood of experiencing felt or enacted stigma. One participant focused on the stigma associated with looking different.

> *" Reason? My reason is what everyone other's reason is, as I said everyone else, if people didn't bother making it into an issue or into a thing or whatever you yourself would not have to take it upon yourself to come in here and sort it all out. If people weren't to judge a book by a cover you wouldn't have your jobs, surgeons here wouldn't have their jobs to fix cleft palates. It's because other people make you feel lesser than them, like just not as good because you have something, thus you wish to sort it out so you don't have to go through all that. That's why you're here."* **INT#28.M**

And another highlighted the impact on self-esteem:

> *"Because I wanted to look better, I'm not unhappy with the way I look right now, it's not like I have really low self esteem, I have my up and down days but if I can do the orthognathic surgery and it's going to make me look better, in the long run, then why not take it."* **INT#21.F**

This demonstrates the importance of understanding treatment choices, expectations and experiences within the wider context of people's lives. It is easier to see the importance of aesthetics when understood in the context of participants' accounts of bullying and stigma, particularly when the timeframe is taken into account. Participants were most likely to report bullying and/or stigma whilst at school, and this tied in, for many participants, with the start of orthodontic treatment.

Participants also showed a willingness to undertake more complex treatment, such as surgery, in the quest for aesthetics.

" Well I guess always when you have an option in life of doing something else, definitely you should go for looks always better, I think that more than anything else, I knew that the whole treatment would be more lengthy and probably painful and time consuming but in effect it was the better solution long term. So yeah I wasn't happy about my braces but I think what helped was that, pardon me, I got the lower brace a few months before the upper brace, so I wasn't just given train tracks on both of my jaws."INT#7.F

" Yeah, well I didn't really mind. I did mind how I looked but it didn't, I didn't, not enough to go for surgery and then they said, then they showed me what it could look like and he said I could just do that and that and he had a way of describing it to make it sound quite appealing and in the end I'm very happy I got it, but it took some convincing and even then I wasn't sure as I was going under with the anaesthetic, I was still thinking do I really want to do this but when I woke up I was very happy with the outcome, it was a surprise. So I'm, yeah I'm, yeah glad I did it."INT#11.M

" At the end of the day what do you want? Do you want to feel a bit of pain or are you going to walk around with bent teeth? Because I know surely I'd prefer to have a little bit of pain in my teeth knowing the end result's going to end up straight."INT#16.M

The data suggest that participants expected the treatment received to improve the look of their teeth. The quotes presented above suggest that participants were eager to engage in this part of the treatment process, if not actively seeking it out, and so it could be seen as patient led.

In contrast, treatment aimed at correcting functional problems could be seen as clinician led. This does not mean that participants were not interested in improving functionality, but it was not persistent when participants highlighted function as a key issue in their narratives around treatment. Of those that did, one participant spoke specifically about a functional issue that 'no one can see it because it's internal':

" I think that's more to do with the fact that my jaw is very back and I have a very small mouth, like it didn't develop. I can tell that younger children have a bigger mouth than me, but no one can see it because it's internal. As you get older your mouth grows, your teeth grow, your jaw grows; I think. But mine grew a very small amount and I feel sometimes I can't breathe that well. It's not a problem too much, it's just something I've lived with and have become used to it."INT#2.F

Another participant highlighted function alongside aesthetics:

" I think many people in the medical professional strive for perfection, or at least aim for the most conventional appearance and aesthetic as they can. And that's also what I wanted. I think it was both function and appearance, just to get the line up correct. And also I think the plan was that it would help me to eat a little bit better. I don't recall having

*any issues eating before, but I guess I could have been compensating [inaudible-29:02] very good at that. But yeah, and I think it was also [inaudible-29:09] surgery it would make it easier to not have to work on the teeth, just to move it forward and then the teeth would fit together and then you could ... I think I must have had braces on then. Then I could use the elastic to keep the teeth in the right place so they wouldn't move and keep the jaw from moving back too much. So, yeah. Function and aesthetic."**INT#3.F**

The data here suggests that participants were most interested in appearance and aesthetics. It is worth noting, however, that the majority of the functional work was carried out when the participants were very young. This links with results presented in the previous chapter on memory and how early treatment episodes are remembered. It would be interesting to see if parents had the same or different priorities in relation to treatment and the reasons for undergoing it, as in most cases, it would have been parents who made the key decisions about treatment related to function.

The final reason that was highlighted within the data about why participants underwent treatment was related to cost and availability. Participants highlighted the desire to accept whatever treatment was offered:

*"well…I wanted to have them…yeah…I really wanted to have them…I mean it's not like my scar bother me now…but like…it's because…maybe later when I'm older…it might bother maybe and…it will be I have to pay for it…in like 10 years time…where is now I get it for the NHS…"**INT#4.F**

*" and then followed the standard cleft lip and palate path with all the orthodontic treatment, pulled a tooth down from my jaw with a chain brace. I've had the full works really and plus a rhinoplasty just to sort out my nose, just everything that they've offered I've taken really because it's free and it if helps, so."**INT#11.M**

This was both because the treatment was seen as beneficial but also through an awareness that the treatment was being offered free at source on the NHS. When looking at the reasons participants gave for engaging in treatment, the data suggest that participants seek treatment predominantly for aesthetic reasons. It is important to note, however, that the participants in this study were adults, but that many of the treatment decisions were taken when they were babies or very young children and were taken by parents or carers at the earliest stages of the care pathway. The reasons for treatment presented here may only represent one part of the story. An understanding of some of the reasoning behind treatment choices may help to contextualise expectations surrounding the outcomes of the treatment. These expectations are explore in more detail in the next section. Also adults are more aesthetically aware influenced by the media and friends.

6.1.2.2 Expectations

Participants were asked to reflect not just on the reasons behind their treatment choices within the care pathway but also on their expectations of that treatment once it was undertaken. When asked about expectations responses fell into three broad categories:

- Outcomes focus

- Context/process focus

- No expectation/Laissez-faire approach

The first category related directly to the results of the treatment and expected physical and non-physical outcomes. The second set of expectations related to the processes of treatment (timings, length of treatment, information needs etc.) rather than about the outcomes *per se*. What is interesting here is that the answers that fell into this category were about the nature of expectation itself, rather than the expectations of treatment outcomes. This may reflect that participants' expectations can be unclear or even that they had not thought about it. The third category related to participants who stated that they had no expectations, either through a lack of information or as a deliberate strategy to avoid disappointment. Each category is taken in turn below and illustrated with quotes from the interviews. (Figure 12).
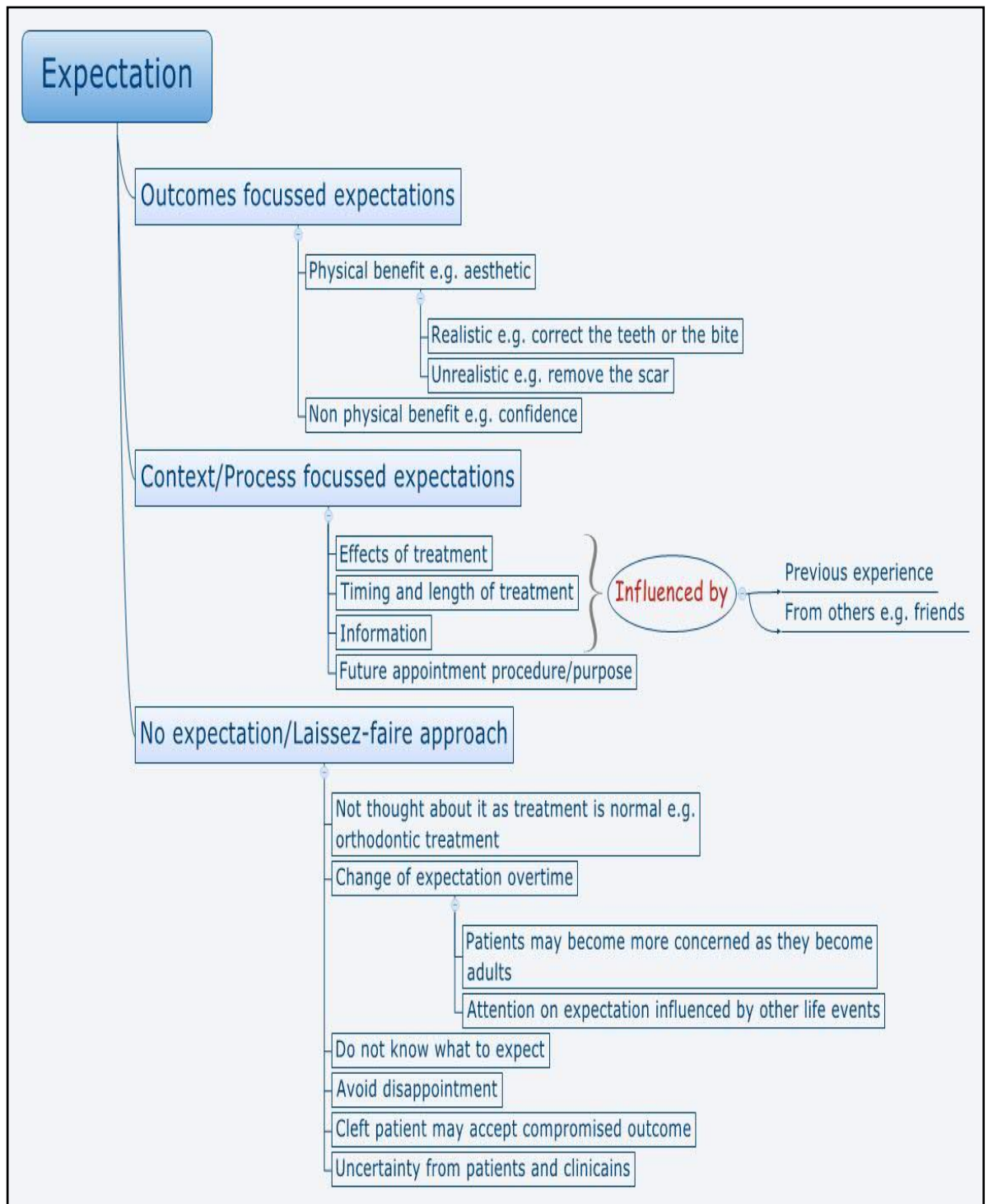
Figure 12: Expectations of treatment

Outcomes Focused Expectations

The data suggest that participants' expectations of outcome were mainly related to expectations that revolved around aesthetics. This ties in with the findings related to the reasons for seeking treatment presented above. Expectations focused on correcting the

look of the teeth or the jaw/face or on the minimisation of scars. The specific expectation was dependent on the type of treatment being undertaken.

Expectations of orthodontic treatment revolved around the look of the teeth, for example:

> *''To just have straight teeth, and I got that, so it was good but I think the amount of time that, I think they were straight before, no, hold on, I think they were straight a few years before I actually got the braces took off, so that was quite frustrating that I was happy when they took them off and they were all straight, so they lived up to my expectations.''**INT#9.F***

> *'' Of the orthodontic side? I thought I would have perfect teeth for ever more. But I don't and nobody has perfect teeth. I mean I am happy with the outcome, I am but there are things like not quite in line any more. It used to be I've got a bar behind at the moment round here [demonstrating]. Yes so they have moved slightly but ... and I've been back to see someone a little while ago and they just said you can have it altered but it doesn't, it won't necessarily stay. So I guess I've just accepted that. So my expectation of it has not really, it's not bad at all. I'm happy with it.'' **INT#10.F***

Whilst expectations related to the look of the jaw or face where related to orthognathic surgery:

> *'' It's going to make my bite better, which will be good, because at the moment I have an under bite, is that right, an under bite, I think he said, yeah, it's an under bite, yeah, it's a crooked bite, and I just want to get that fixed, and I've seen pictures of before and after, and it just looks like so much better, and I'm hoping that's what mine will come out like, but the surgeons here are really good, cleft house [ph] is really good.''**INT#21.F***

One participant stated that she will be undergoing future surgery to remove the scar of the cleft.

> *'' .. I mean I'm going to get my scar removed.''**INT#4.F***

It is unclear whether she was aware that surgery would not be able to remove the scar completely or whether she was talking about enhancing the look of the scar and make it more subtle. This was not followed up in the interview.

Non physical outcome expectation was also highlighted about confidence. There was an expectation of improved confidence following treatment.

> *'' Outcome, hopefully will be a bit more ... I'll have a bit more confident ... I got confident but have a bit more confidence without me''**INT#13.F***

> *'' Have a bit more confidence in myself, where ... that's it really. Have a bit more confidence. That's all I really want, a bit more confidence to stand out a bit more. Yeah, that's really. Nothing else like that.''* **INT#14.M**

It is unclear whether the increased confidence would be from improved aesthetics and/or function in this case.

Context/Process Focused Expectations

The second set of responses were about expectations related to the treatment process or context rather than the outcomes of the treatment. Expectations about the treatment process itself were often influenced by previous experiences. One participant, for example explained that negative experiences from previous surgery influenced her expectations for the future procedure by adding a negative element to it:

> *''I also, sorry I forgot to mention that, I had my wisdom teeth removed here at [name of hospital] as well, all four of them, yeah and I was quite worried about the jaw surgery only because back home I had three unsuccessful bone grafts with my hip and [name of surgeon] the Surgeon said that its very likely that he's going to have to do a bone graft again and that was the only reason I was worried and a bit panicky about it because I knew it didn't work out three times, why would it work out now but he didn't have to do it, there was enough bone in my mouth to get it stitched up somehow without having to do a bone graft which is fantastic. ……… I think I had to come in a week after the surgery because one of the elastics broke and me being very paranoid about everything and the progress of how things were going, decided to come in to get one fixed, apparently it wasn't a big deal, but in my head it was and yeah it was all pretty much plain sailing considering the surgery was so serious and it included seven screws in my face and I was quite confident and quite, I was able to trust the Doctors here at [name of hospital] really, so yeah it was much easier than I thought it will be.''* **INT#7.F**

The negative experiences of treatment effects related to friends or family also effected expectations, particularly around the potential pain of orthodontic treatment which is a far more common treatment to receive:

> *'' I think like my friends told me that having brace will really hurt but…it's not been too bad settling that might be like less painful than other people get…I don't know…''* **INT#4.F**

This can mean that there is trepidation about the treatment prior to the start but may also mean that the actual process is less painful that expected as was the case here.

There were issues around timing related to treatment processes, both in relation to the time taken to complete treatment and also the amount of information participants had access to prior to the treatment to help shape their expectations in this regard. As having braces is quite common, participants tend to compare themselves with other people and develop the expectations based on what they see around them. Participants suggested that having braces was seen as a more positive process in line with what would 'normally' happen particularly if friends were having orthodontic treatment around the same time. This meant, however, that expectations around the timing of orthodontic treatment, and the length of time that the treatment will take may be based on the more routine orthodontic treatments undertaken by peers without CLP, leading to disappointment when treatment lasted considerably longer that for peers without CLP.

> '' I got it in when I had GCSEs like a year after like you know people only have for year year and a half so you know there is lots of teen who having brace will it been great to for like a year a year and a half ..after that you like I want it off my face you know...so I guess after that I really keen to get it off ..so I think because it took so long 8 years is a long time to have braces on...so I think the time I had it on..I really wanted it off...you know''**INT#1.M**

Or the treatment did not finish when expected:

> " It got quite frustrating when I realised that I was going to have it longer than what I was initially expected to, so I suppose that's quite frustrating, the fact that I thought I was going get off sooner than what I have done, so maybe not telling people when they're going to get it off because it could change and then they could be disappointed,"**INT#18.M**

This raises questions about the amount of information that participants received and/or recalled about the length of time the different stages of treatment would take.

The importance of information about the treatment was highlighted by a number of participants in relation to shaping expectations of treatment. Alongside information about the length of treatment itself, the issue of recovery times was raised:

> '' yeah .. I do think it was..I think in terms of my timing with the operation I think it could have been better ..actually I don't think that there is anything that I could have done apart.. I mean there is never a good time to be off for 6 weeks 8 weeks it's never a good time.. so yeah timing is always going to be bad so yeah ..so I think it was a good thing.. I just need to make sure that the next one is at a good time that fit with my work because I work at a place where I'm pretty much worked for two years to get into a place like this..so if I'm going to have a worry..it's going to be like number one is it going to look good and number two it needs to fit well at work because if work is happy ..and I'll take all the time that I need but pretty much saying i want 2 months off 8 weeks off ...it's hard..its hard 2 month off work you know''**INT#1.M**

This was important not just in relation to patient expectations but also in enabling participants to organise work and home commitments and fit their treatment into the rest of their lives. This ties in with the results from the previous chapter, highlighting the importance of understanding the treatment of CLP within the context of daily life. Expectations are shaped not just by clinical outcomes but also by outside, contextual factors such as experiences of stigma or timing and planning issues.

Expectations around treatment also covered other aspects of the organisation of treatment, such as the content of appointments. This may cause some distress if the patient was expecting more than just ten minutes follow up and this could be even more accentuated in case of travel and distance was an issue:

> *''… but even with a, the guy I just had, the, my first appointment with him, I waited in the waiting room for an hour, I went in there, he saw me for about thirty seconds and said okay, yeah we'll do all this treatment next time. That's a bit annoying but it's not really a, it's just a, in the context of the day, he might be running late and you can't really hold it against anyone for waiting...It was, yeah it was to see [name], just a, I think he was just going to see what the problems were and I was expecting more. It was probably because I was expecting him to do something.''* **INT#11.M**

No expectation/A Laisse-faire Approach

The last group stated that they did not have expectations about their treatment, or were unwilling/did not feel the need to tie down their expectations.

The data provided some possible explanations for this lack of expectations. When looking at expectations for orthodontic treatment, for example, a lack of specific expectations could be related to the fact that this type of treatment was common amongst teenagers and so not seen as a particular concern.

> *'' so when I'll have my braces on I wasn't thinking about the end goal I was thinking … I didn't think about it ……..I was like its braces cool like ill put it on and go back to school.''* **INT#1.M**

> *''I don't really know. I never really thought about orthodontic treatment an awful lot, partly because braces are such a common thing among people my age. Also I suppose it wasn't really as important as the surgery which was much bigger, especially in my eyes. Yeah, I suppose I have just been going really quite regularly for a very long time. I suppose it's a just ... I suppose it's a bit like a dentist's, just a bit more advanced. I think*

it sort of felt like going to the dentist and putting braces on, just doing it a bit differently, but it didn't really feel like a sort of hard appointment, it was just something I did.''INT#3.F

These two examples reflect a more Laissez faire approach, rather than a specific tactic employed to avoid expectations.

Expectations about individual stages within the pathway may also be influenced by the overall journey that is being undertaken, both in relation to the care pathway as a whole, and also across time. Children with CLP may be less concerned about having expectations but this changes over time as expectations are laid out.

'' So at first, I wasn't quite sure what the expectations were. I had this kind of childish philosophy of, "Oh well, someday it'll all be alright. Don't know quite when that day will be but someday it'll happen." And when I started to get a more comprehensive timeframe of thinking, "Okay, well when you get to sixteen this'll happen, when you get to eighteen this'll happen, when you get to twenty-ish that'll happen," my expectations became more coherent and more precise. I sort of knew roughly by which stages I should be experiencing certain things.''INT#17.M

What is interesting here is the suggestion that expectations have been set by other people, and that this becomes clearer over time. Expectations in this context take on the form of a timetable of treatment leading towards an overall goal. The same participant also related expectations to the broader context in which treatment was being undertaken, tying in with the previous chapter where it was suggested that CLP care cannot be understood without understanding the wider context.

'' I think I see what you mean. It's sort of, in some ways, when I first think about it, it was kind of in waves. Like sometimes, it would be a case of thinking, "Okay, so we have no idea what we're getting ourselves into, we'll find out." And then other times I'd be like, "Yeah, I've got a pretty good feeling that this is going to have a good outcome." And then sometimes I'd be like, "Oh God, I'm terrified, I don't want to go under the knife again." It would be up and down. I think sometimes it would depend upon what day of the week it was, really……….I mean, it kind of needs to be put into a broader context of other things going on in my life, like social situations at the time, schoolwork and worrying about the future and that kind of thing. And when I think of all of that, I sort of think that … sometimes it would not be on my mind at all. Because I would be worried about whether I was going to pass my 'A' Levels, or whether my girlfriend at the time was going to get jealous of me because I was talking to somebody that I knew who she didn't like, something like that.''INT#17.m

It is evident in the data that expectation can be influenced by multiple factors and can change with time. What is also clear from the quote above is the presence of uncertainty.

This ties in with one of the key themes in the wider sociological literature on long-term conditions.

One participant explained they simply did not know what to expect:

> *''I didn't really know what to expect. I was very happy with the outcome because they were a lot nicer than when I went in, my teeth but I don't know, I suppose they were never going to be perfect because obviously I'm cleft palate, they've, I'm missing a couple of teeth and stuff, so they've had to fill in gaps and things, but yeah I was happy with the outcome. I wouldn't, obviously you could always have nicer teeth and I'd like mine to be a bit nicer. They were harder to clean with the braces and stuff, so they're a little bit discoloured, a little bit crooked because if I don't wear retainers all the time they'll loosen up a bit and twist, but on the whole they've, it could have been so much worse. Yeah, I'm pretty impressed with what they've done with just a bit of metal and plastics, pretty incredible really, it's good.''INT#11.M*

Another participant used a lack of expectation as a specific strategy to avoid the disappointment of a treatment outcome that did not match their expectation.

> *''Well if you think about what you want to happen in a very detailed way you can then be easily disappointed because you can then, you can completely imagine and I'm sure many people do, they think that they're going to be fixed to the, and their look, they're going to look as any other normal person without a cleft which is never going to be the case. So I was quite aware that this and that was going to happen really, so.''INT#7.F*

In this case the strategy developed was related to an idea of how a 'normal' face should look and an acceptance that this was not, in fact, an achievable outcome. This also relates to language used in a number of the interviews which takes the idea of 'normal' aesthetics as a comparator, either in relation to stigma, bullying and self-esteem, or in relation to the endpoint of treatment – something that is explored further in the section on the definitive orthodontic/orthognathic treatment. It also reflects the uncertainty about how the face will end up looking at the end of the treatment process.

A lack of expectations or a lack of expressed concern about expectations can, then, reflect uncertainty about outcomes.

> *'' when they were talking about what may happen after the surgery in terms of …eating and talking they said ohhh well it's a possibility that might make your speech slightly better because of the repositioning but there is also a slight possibility that it can make it worse I'm not unlikely more used to be ..or..but it didn't make a lot of difference in the end and I got to think they anticipated it won't make a lot of difference one way or another there*

*might be a slight change but it wouldn't been a significant impact upon the way I spoke…and as I said in the end there wasn't any that I noticed"**INT#6.M***

As this quote shows, this uncertainty may come from clinicians themselves when they are unable to offer certainties about treatment, again linking with the wider literature on uncertainty in relation to long-term conditions. Uncertainty emerged in relation to expectations, as shown above. It was also raised as a specific concern by some participants when asked to expand on how they felt about the different treatment they received within the care pathway. This is explored further in the next section.

6.1.2.3 Concerns
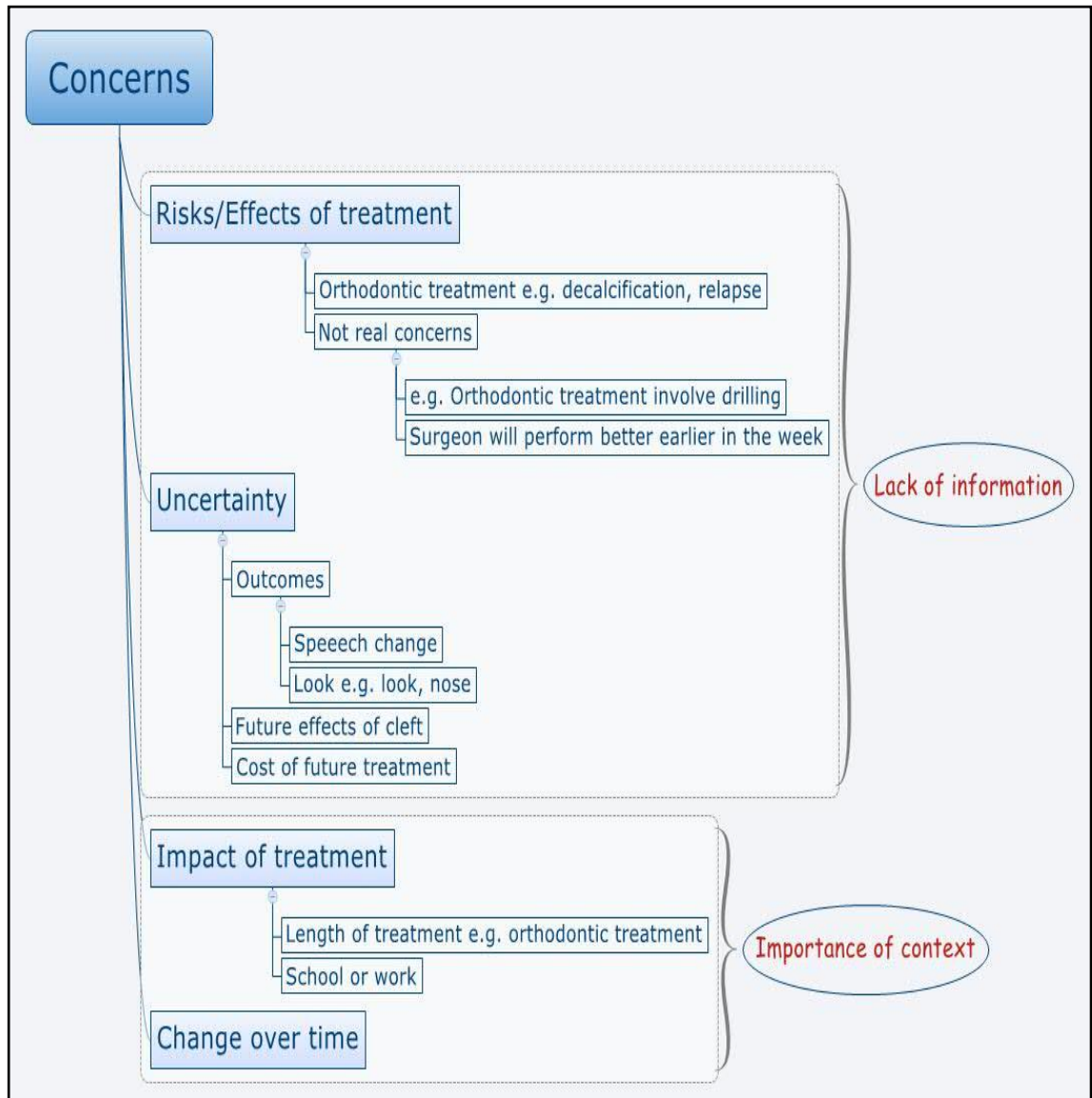
Figure 13: Concerns related to treatment

When asked to talk about experiences of treatment a number of concerns were raised. These included risks associated with particular courses of treatment, uncertainty about current and future treatment, the impact of treatment on daily life and a lack of information about treatment processes were all cited as concerns. (Figure 13).

Effects or risks associated with particular aspects of treatment may cause a concern. For example, concerns associated with the effects of orthodontic treatment and the risk associated.

> '' With any treatment, there's always risks, and you're always concerned about the possible risks, due to my cleft and palate, I have short roots, I have root resorption and

*that's a risk, so there's always risks in the back of your mind, but again, like the orthodontists are so good that you're not really concerned, and I've been quite happy with everything that's gone on."**INT#21.F***

*'' Yeah how I would look after, how I would be speaking, about my brushing, being hurt like with the wires and that. They were the main questions that I asked after I had braces like the day I had braces I was like can I eat normal food and can I brush my teeth normally. I was told that it was going to hurt a bit and yeah. It wasn't a nice experience, but it had to be done to make my teeth look straight."**INT#27.F***

One participant talked more generally about the risk associated with the day of the surgery when her surgery was scheduled for a Friday as she was concerned that usually people are not at their best on the last day the week:

*'' The only thing that has concerned me before my surgeries is the fact that they're done on a Friday because I think people's productivity and concentration is always lowest on a Friday because you had a busy week and it might be weird, but I always thought, I really trusted in the Surgeons and people, but I thought I know how I am on a Friday and I think I perform better on a Wednesday or a Tuesday than on a Friday, well I start thinking about the weekend and I had a busy week, long hours at work and I think that was the only thing that ever concerned me that the surgeries were done on a Friday. It might be the most bizarre answer you get in this research project but that was my concern."**INT.7.F***

Finally, in relation to risk, the risk of relapse once orthodontic treatment had been successfully completed was also raised as a concern:

*'' I do worry about my teeth moving back because I have to wear my retainer at night times and my teeth already move in the day so when I put my retainer in its still really tight and I have had that for so long now that I don't … I can feel them still moving that is another thing that worries me if they start telling me to wear it once a week, are they all going to move back and then I will have to put the thing on and they will be moving too much. That is what I worry about. I do sometimes worry that I will have to have further operations on my teeth if I have got any that they don't know about because I am sure I must have teeth still left up there that are just sitting there, they worry me a little bit."**INT#8.F***

This last concern relates to uncertainty around the long term outcome of treatment rather than risks associated with the treatment *per se.*

Uncertainty about the outcome of treatment was also expressed. One participant talked, for example, about the potential impact of jaw surgery on her speech:

*"My general concerns were just like before I had the jaw surgery I was worried if my speech was going to be different or my lips or if anything on my face was going to change apart from my jaw, like when my face was swollen I thought is anything else going to change apart from my jaw and it didn't. But there is further more surgery that I would like to have."***INT#27.F**

Whilst another talked more generally about whether the outcome would be sufficiently positive to make the pain worthwhile.

*'' Just about really all my operations, what's it going to look like in the end, is it going to be worth it, going through all the pain for it, and whether it'll be how I want it to be, because if it's not, then I'm going to be upset, and I won't want to go out, but nothing majorly [ph]."***INT#9.F**

In this example potential benefits and disadvantages of treatment are being weighed up in a context where outcomes are uncertain. The concerns expressed are general rather than specific.

Furthermore, uncertainty about the future can be another source of concern. One participant was not sure how the CLP will affect them in the future.

*'' Is it going to affect me at all or if there was anything wrong that would happen over time. Or if nothing would happen over time and I can just stop going to doctors' appointments or …"***INT#2.F**

Whilst another participant was concerned about future treatment and whether it would be covered by the NHS or not if it was needed:

*'' I am a little bit worried about in the future if anything happens regarding my teeth especially ones that haven't came down in the right place, if I am going to be able to get treatment for that on the NHS or if I am going to have to pay for it or whether anything will be able to be done at all about it."***INT#8.F**

What is clear about these concerns is that the uncertainty is caused by a lack of information, but it is information that clinicians cannot give – they cannot say with certainty what will happen in the future and can only give answers based on the best evidence to date.

Two participants talked more generally about concerns they had about treatment in relation to how it would impact on how people saw them. One participant, for example, became concerned when she continued wearing braces while everyone else had had their braces removed.

'' I didn't really have any concerns about them, I just was a bit concerned about me growing up, this was when I was younger, about me growing up and people wondering why I still have braces on, this day and age, people wondering why I'm eighteen and still have braces but.'' **INT.21**

It was not the length of time that the braces were to be worn that was the cause of concern but the perceptions of other people about an older teenager still wearing braces. This ties in with a response from another female participant who highlighted both the fact that her concerns were related to perceptions of her, both aesthetically and in conversation, but also that these concerns changed over time:

'' I think that in the beginning quite a lot of it was, well for me I was more concerned about the physical aspect and how I looked, but more recently I've been more concerned, I don't know, more hopeful that they'll be changing my speech, just because I do keep meeting lots of people and it does sometimes get difficult when they don't quite understand'' **INT#3.F**

The change from concern about looks to concern about communication could indicate a growing confidence with age but it could also reflect the change in context from a school environment where aesthetics was very important to a work environment where communication becomes more central to how people are judged.

The importance of seeing CLP care in context was also highlighted by another participant who talked about the difficulty of juggling exams with treatment:

'' It's purely because I'm a bit forgetful but these two years have been massively – at school it's been really hard. We've had our GCSEs and all that and so when I go to a doctor, I just feel like I need to get the appointment over and done with, just get the information or get the treatment and go home, go back to work or whatever. Now that I've finished that massive piece of work the questions are all starting to get to me now. Now that the treatment is over, in terms of braces, I feel like I should have asked these questions before.'' **INT#2.F**

In this example the end result was a series of questions needing answers and concern about a lack of information.

A lack of information – or the inability to recall information that has been given – remained a problem for many participants, exacerbating fears. For example, although orthodontic treatment is quite common, there may still be confusion over what the treatment entails. One participant reported that he was concerned that orthodontic treatment would include drilling his teeth.

> *'' I thought they were going to drill holes in my teeth to put in the braces themselves, but they said oh no, they're going to glue it on, I was okay thank you, that's what I was worried about.''***INT.23.M**

This type of concern can easily be rectified by ensuring that information provided to patients is clear and is understood. Giving clear information, where possible, can lessen concerns related to treatment. This is understandable as information may help to reduce uncertainty which might cause concerns. One participant explained her lack of concerns is this way:

> *'' Not really because it was all explained to me, so I didn't really think that anything could go majorly [ph] wrong with it, I was told everything, so I didn't really have any concerns.''***INT#9.F**

A number of participants also reported that they had no concerns about treatment at all:

> *'' No, no concerns what so ever actually, no concerns.''***INT#23.M**

What is clear from this is that most of the concerns raised centre around a lack of information, either about risks or due to clinical uncertainty about the implications of treatment for individual patients. The remaining concerns reflect the importance of context and understanding how treatment fits in to peoples' daily lives, both practically, personally and interpersonally.

This first section of results has focused on issues that related to the context in which treatment was being undertaken, predominantly before treatment was started (although it is worth noting that some of the concerns were relating to post treatment outcomes). The next section focuses in more detail on what happens during the treatment process itself.

6.1.3 During treatment

This second section focuses in some detail on experiences of the treatment process itself. This incorporates the environment and staff as well as exploring how decisions were made and practical issues around pain, managing time off school or work and so forth. The data is split into six sections:

- Hospital environment
- Staff
- Information
- Decision/Choice
- Effects of treatment (orthodontic/orthognathic treatment)
- Coping

The first two themes relate to the wider context of the care environment, the second two then focus in on information and decision making processes and the final two look at the impact of treatment on individual lives. Each sub-theme is presented in turn and illustrated with quotes from the interviews.

6.1.3.1 Hospital environment

The clinical or hospital environment may influence the patient experience. It can provide a pleasing atmosphere which may be related to how clinics look or to how efficient they are. A positive or negative environment can affect how people see and experience their treatment and potentially could affect patient satisfaction levels even before treatment has been undertaken. (Figure 14).
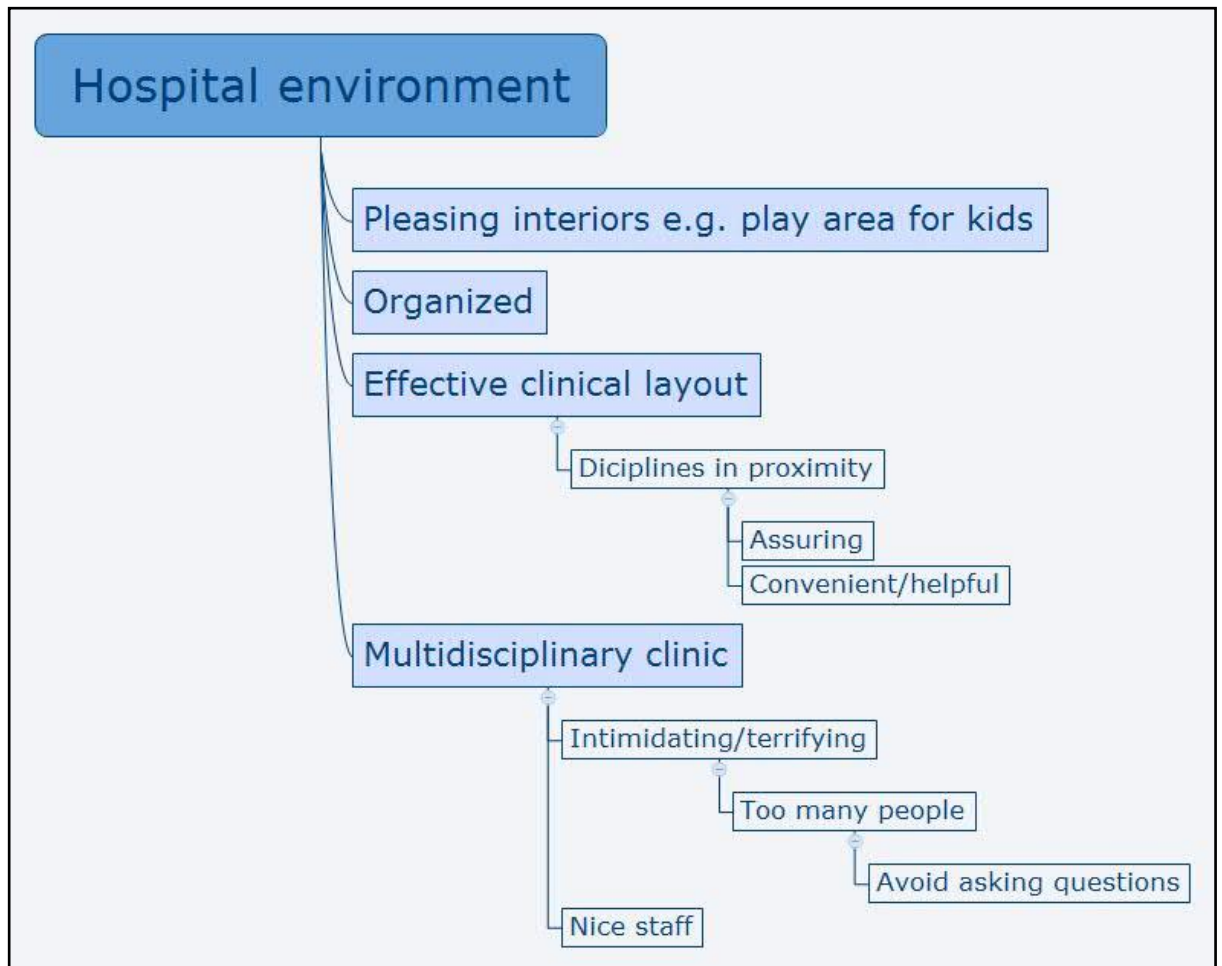
Figure 14: Hospital environment

In relation to the early childhood surgeries, participants often only remembered moments around the surgery time. These were often memories related to the hospital environment. As the following example shows, making the experience at hospital as pleasing as possible could be the only positive memory that a patient can maintain from the surgery.

> '' it just a little things I remember like there been like a kitchen somewhere and there is nice toast my mum used to make it for me so it was really nice [laugh] I remember it was like a chocolate milkshake like nesquick but its little flashes that's all I can remember''**INT#1.M**

A child friendly interior clinical environment may also help the patient to think more positively about their treatment. One participant mentioned this and referred to the waiting area with toys as good.

> '' Yeah, well that would have been I suppose when I started going to floor twenty two, Orthodontics, that would have been my first brace. I remember it was quite good, they had the waiting room with all the toys and stuff, so it wasn't too bad, I'd chuck around some books and stuff and look out the window across London, that was cool and so I had so many different braces, I was coming in here all the time but it was alright…...''**INT#11.M**

The same patient thought that the waiting room layout made the reception staff work more efficiently.

> "…… you had to buzz your way in so they knew you were there straightaway and it had a, it was nicely done for the kids, nice colours and stuff and then they had a couple of rooms, so it was quite efficient, they knew you were there and if you did have to wait there was always someone explaining why instead of it being a bit like a cattle market and you're all just in and out, it was a bit better …"**INT#11.M**

This quote describes the situation once a clinic had changed location and the positive difference the change in location had made. This was contrasted with a previous location which was deemed more chaotic.

The proximity of departments and specialists was also discussed. Having the disciplines together or close to each other so the individual patient cases could easily be discussed was seen as an advantage:

> '' And I think the key to success in that was that the Orthodontist and the Surgeon were working together on this which was another problem I had back home where my Orthodontist and the Surgeon knew each other very well but they were about three hundred kilometres apart and they saw each other about once a year if, and I think it was very disjointed where here I realised that [names] know each other very well and they work together and they look at patients' cases together which was the case and that has given me a lot of confidence that."**INT#7.F**

In addition, having the departments in close proximity or at the same place was helpful for the patient.

> '' … and that is, that's even better still because now they've got the photography and everything on one floor because again another problem was if you had to get photographs you'd wait maybe forty minutes for your orthodontic appointment and deal with the work then say right you had to go for photographs which is on floor four or whatever, so you'd have to go down to floor four, then wait another hour because it was a, just a, the whole

hospital used it and then you'd have to take them back up and it just added so long to the day, but I imagine now its going to be a lot more streamlined which is good, ten years too late but yeah. So good for everyone else that uses it. "INT#7.F

What these quotes suggest is that the physical environment, organisation and atmosphere in the clinical area can all affect the patient experience.

Multidisciplinary clinic

One of the key areas of organisation which came up frequently in the interviews was the multidisciplinary clinic. Patients and their families were expected to attend on a regular basis and this was the source of concern and distress for some participants. It was described as a terrifying experience because there were so many clinicians present. This intimidating environment led some patients to stay silent and not raise any issues or ask questions, exacerbating problems related to lack of information and uncertainty highlighted in the previous section.

" and it was always a room full of people and I found it terrifying. I hated that, that was horrible because there were so many faces all looking at you and all ... one person talking to you but the rest all scribbling something down and asking questions. And someone would ask you to say count to 20 and then someone would be looking. And then there will be other medical students in the room. And I wouldn't say anything. I would just sit there and just be completely silent until my mum basically answered all the questions."INT#10.F

" It's quite blurry if I'm honest, you know I just remember being taken up to Guy's and coming and sitting in a room like this and have like a circle, a cornucopia of people around me sort of each asking me questions based on their individual areas of expertise."INT#24.F

Whilst many participants recognised the importance of the multidisciplinary meeting for communication, only one described the experience as positive:

"Everyone was nice and they were explaining everything what's, I used to go in there, they wanted to all see how my process was going ahead, and just talking about options I could have and saying what I can improve, things I need to change or work on, anything, it was just more like an open discussion, the whole team wanting to see how things were going with the dental treatment."INT#13.F

The descriptions which participants gave, in relation to the clinical environment, show the importance of this for shaping positive and negative experiences, regardless of the

treatment itself. As the data showed, a young child may only remember the play area in the hospital where he had his operation but not the operation itself. This shows how the hospital environment played an important and hidden role in transforming the operation experience into a pleasing memory for the child. In contrast, the intimidating feel of the multidisciplinary meetings can exacerbate problems around information leading to uncertainty and potentially a more negative experience, which again is unrelated to the actual treatment itself.

As the clinical environment can play a role in the participant experience, the people working within the clinic can have an important role in determining the participant experience. This is explored in the next section.

6.1.3.2 Staff

A large component of the environment described in the previous section relates to the people working within it. Expertise, continuity of care, good communication skills, trust and empathy were all seen as positive attributes whilst lack of communication (between patients and staff and between clinicians), lack of expertise and poor organisation were seen to have negative impact. These factors are illustrated in (Figure 15).
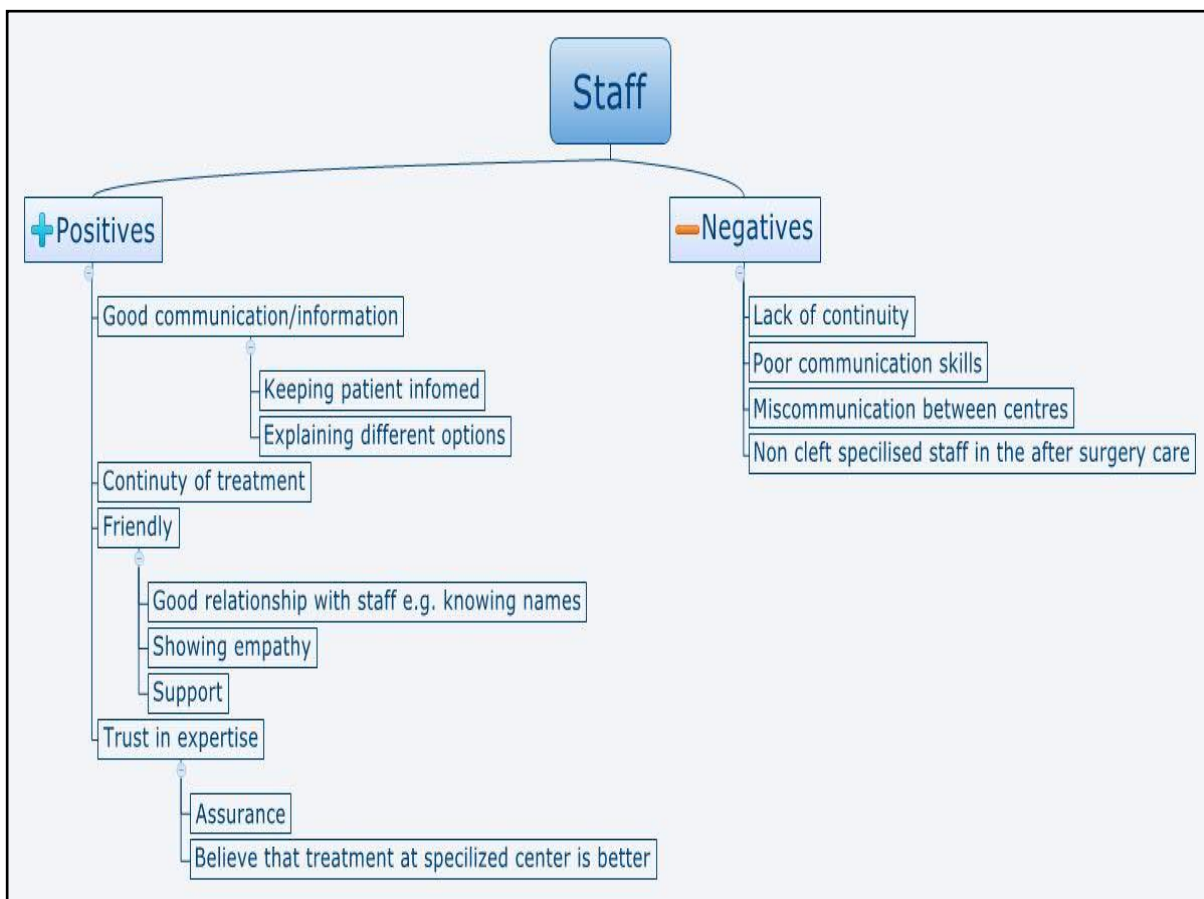
Figure 15: Staff

Good communication/information was at the heart of descriptions of positive relations with clinic staff. This appears to promote trust and an environment in which the patient feels able to ask questions and make decisions:

> " Yeah, I think it was good, I really liked my orthodontist, so that helped, he was really nice and he used to talk me through everything that they were doing, so that made it better, I can imagine some of them not really going into detail about it, I think you need to be told every little detail about it, because it just makes you feel a lot happier with it…. "INT#9.F

Explaining treatment options and outlining risks and benefits increased the likelihood that the patient would have a positive experience and reduced the likelihood of uncertainty:

*" Positive, I think definitely that I was very much involved in the process of the treatment in many different ways and I was given choice and options of what I think is better, what do I want to get out of this more than we're going to do A, B, C and D, it was more of a joint decision and I was always very well informed, I can always ask questions and stuff like that."***INT#7.F**

*" …the orthodontist would always make sure that I knew exactly what was going to happen every time they were trying to do something. And every time a brace broke, or every time that I was in any serious pain, I could always be seen by them. So it always felt very comfortable with them, and I always felt like I was well looked-after"***INT#17.M**

This quote ties in good communication with trust and continuity of care. The importance of continuity of care and trust can be seen when a care pathway extends over 2 decades and was an integral part of the life a person with CLP throughout this time.

Friendly and empathetic care was seen as a reflection of an ongoing relationship.   For example, recognising people and asking them how they were when they attended the clinics was seen as an important aspect of care. This was perceived by participants as an important factor in making the treatment experience positive:

*`I've had a positive experience……Like I know some of the people here I've seen throughout my appointment years and you know they make you feel like they know you and that you're welcome here so I don't feel very alienated, I come here knowing that I feel like people know me here, I feel like they're doctors and they know me here you know and that they actually care. That's the feeling I get from it the way they interact like they genuinely care."*I**NT#20.F**

Again the care is emphasised here, not just from clinicians but also from the support staff:

*" that I think it's really good like the people actually care...I guess that's why they come to the NHS ... it is really good that you can form a relationship with people who work in the office because they are the same people...I generally like it I enjoy coming in like to see everyone feel is not like a circle people in and out you come here you have a chat with people...people know about your life so they care I like it I think it's really good way of doing things really..."***INT#1.M**

*"So it was more than just being a number and more than just being their patient. It felt like there was a very nice kind of bond between us, all these people. I genuinely felt like they had my best interests at heart, and always acted with my best interests."***INT#17.M**

The length of treatment makes the CLP care pathway a significant part of the lives of the people within it. The development of good, trusting, long term relationships between staff and patients was seen by all participants as important. One patient even stated that he felt sad after he finished with his treatment because he had become used to coming into the clinic on a regular basis and would miss contact with the staff.

> " …I wasn't afraid of Dentists, I don't have that fear of the Orthodontist Department and I, to some extent I felt a bit sad, I'm not going to be coming here any longer, once a month or every six weeks during lunch hour, so it felt a bit more sentimental than I thought really, but yeah I was a bit disappointed with myself but it wasn't such a big deal after all."**INT#7.F**

Alongside the more human side of relationships demonstrated through care and empathy, there was the belief that CLP team staff were experts and a huge amount of trust was placed in them. Receiving treatment within a specialised care pathway was seen as very positive.

> " apparently it wasn't a big deal, but in my head it was and yeah it was all pretty much plain sailing considering the surgery was so serious and it included seven screws in my face and I was quite confident and quite, I was able to trust the Doctors here at [this hospital] really, so yeah it was much easier than I thought it will be."**INT#7.F**

In this example, trust made a difficult and serious surgical procedure seem more manageable and was linked with good communication between the surgeon and the patient but also between the clinicians themselves:

> " And I think the key to success in that was that the Orthodontist and the Surgeon were working together on this which was another problem I had back home where my Orthodontist and the Surgeon knew each other very well but they were about three hundred kilometres apart and they saw each other about once a year if, and I think it was very disjointed where here I realised that [names] know each other very well and they work together and they look at patients' cases together which was the case and that has given me a lot of confidence that."**INT#7.F**

The examples here show that positive, long term relationships with staff based on good communication and trust can have a positive impact on the experiences of people within the CLP care pathway.

Unsurprisingly, the lack of these characteristics was highlighted by those who had a less positive experience. A lack of continuity, for example, was seen as having a negative impact on treatment experiences:

*" I've seen quite a few people over the years and so obviously as people move on it's hard to ... in a sense it's hard to keep consistency,..."***INT#26**

*" It wasn't ideal because we preferred some to others but I suppose I've been going through the system since I was born, there's always going to be changes but there are a few that we didn't know about, we'd turn up and it would be someone different, they were always professional and decent but some of them we, they were a bit nicer and we preferred them and when they disappeared it was a bit upsetting but."***INT#11.M**

This highlights the importance of building up a relationship over time and of interpersonal skills. It was recognised that attending a busy clinic and the length of time spent within the pathway would make continuity difficult, good communication skills could overcome some of this whilst a lack of communication skills could exacerbate the problem.

*"… but even with a, the guy I just had, the, my first appointment with him, I waited in the waiting room for an hour, I went in there, he saw me for about thirty seconds and said okay, yeah we'll do all this treatment next time. That's a bit annoying ..."***INT#11.M**

This quote suggests both poor communication skills and a lack of relationship with the patient which exacerbates the experience of waiting a long time within a busy clinic. Better communication could have helped make the experience a more positive one.

Poor communication was also highlighted as a problem during treatment itself:

*" …sometimes I was laying there with two hands in my mouth and they were prodding and stuff and I didn't really know when it was going to end. So yeah to have a bit more reassurance maybe especially as a young kid, but then again that's probably down to the Nurse. We've had a few more serious ones that don't really want to chat and just get the work done and leave and then we've, our favourite ones are the ones that explain it and have a laugh about it and yeah."***INT#11.M**

Particularly when treatment was being shared across two sites, a lack of communication here could lead to both reduced continuity of care and also inconvenience for the patient and their families.

*" In the beginning they could probably have worked on the communication between [the other site] and here, because I keep on switching between going to [the other site] and going here and when there's people not around at [the other site] who should have been around …it can make getting things done and organising things difficult because there was a ... what felt like a lack of communication between them at times which meant that I'd go up there and they'd say the same thing again despite having heard it the last time and stuff like that. "***INT#26.M**

In addition to the above, one participant highlighted the importance of expertise through an experience of receiving non-expert care following surgery outside the CLP care pathway:

> *" I was in intensive care for I don't know about a day and a half and then I was put over to the ears, nose and throat ward......I don't know if they hadn't read my notes or you know one nurse came up to me in intensive care, I'd just had all this surgery, I could hardly speak and she was asking me what I'd had done and why I'd had it done you know surely it was on my notes ...... and when I got transferred over to the ward the doctor that came into check me asked me to open my mouth wide and I'd just had jaw surgery you know and it was just silly little things like that that just didn't give me the confidence ....* *"INT#12.F*

Whilst this is not directly related to the pathway, it highlights the importance of the pathway itself and suggests the need for greater awareness beyond the CLP pathway.

The fact that the positive and negative experiences highlighted here relate to the presence or absence of key interpersonal skills and the organisation of clinics to enhance continuity , demonstrates the importance of these factors in the overall experiences of patients within the care pathway. Whilst these factors could be considered peripheral to the clinical care provided within the pathway it is clear that they were integral to the overall experience and satisfaction of the person making their way along the pathway.

6.1.3.3 Information

One of the key concepts that has emerged throughout the interviews relates to information. Information has been discussed in relation to what people understand about CLP and its causes, it has been discussed in relation to combatting stigma and bullying and a lack of information was raised as a concern for participants and as a cause of uncertainty. In this section participants were asked specifically about the information that they received in relation to the treatment they were undergoing. Patients were happy about the amount and quality of information they received, however, there were specific gaps in information that were raised in relation to the organisation of treatment and aftercare. Again the importance of interpersonal skills was highlighted with noticeable differences between the communication skills of different members of the team. (Figure 16).
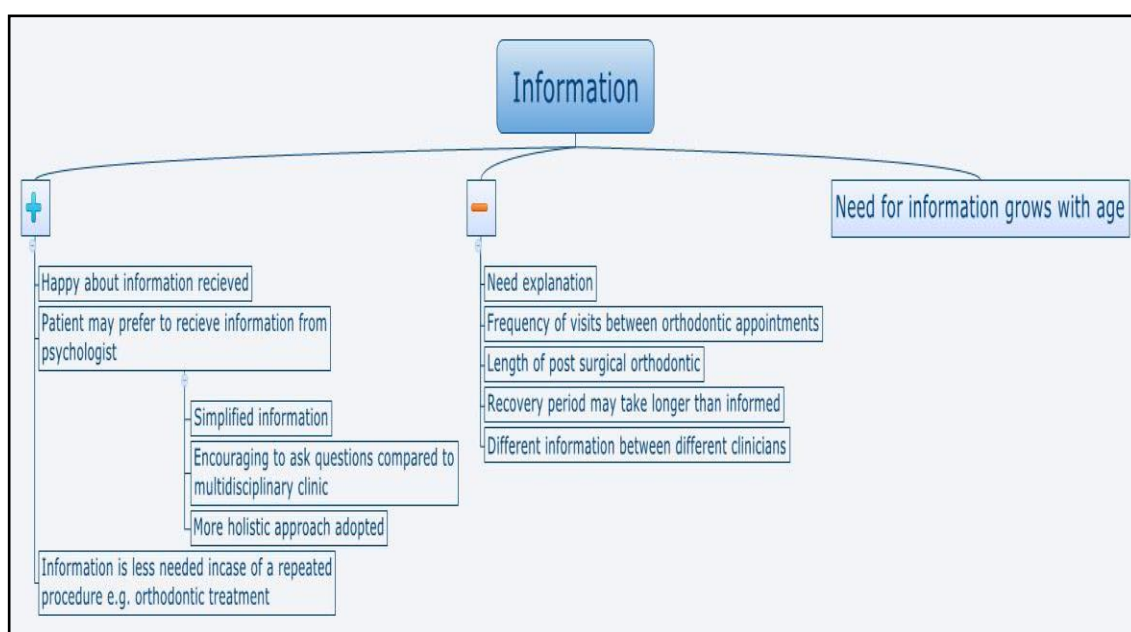
Figure 16: Information

Some participants were aware of a lack of information and the need for more explanation:

> '' At the time when I was – yes, I think I would like more because, as you may be able to tell, I don't really understand much and that might be more because of me. I got loads of information in one go and then I would come back in four weeks and that information has gone because of school. I think I would have liked a few more little random appointments in-between those times to discuss how things were going.''**INT#2.F**

> ''The only complaint I have about the whole treatment would be a lack of – not lack of information, lack of understanding of the information and that's on my behalf. I would

*have liked to have visited the psychiatrist more or discussed with a doctor more. At the end of every appointment they would say "Okay, you're going to come back here in four to five weeks, we are going to tighten the brace, how do you feel?" And it would be the same response every time, "Yeah, that's fine, I'm good." I think if I could go back, I would ask for a better understanding which is what I am lacking right now. Apart from that, the treatment overall was very good."***INT#2.F*

This suggests the need to ensure not just that information was provided but that it was understood and that patients were given the time and space to absorb it and ask questions. In some cases, however, participants were unaware of missing information and this only became apparent when issues were raised which demonstrated this lack of information. One participant, for example talked about the inconvenience of having to wait in between orthodontic appointments:

*'' I mean the only thing I would say the time to get from one appointment to another yeah that what I would say the timing so if... I can be seen in 2 weeks you know I have to delay for another 2 weeks so it be 4 weeks until I can be seen again because so many people that's the only thing I would change so if I change my brace and it only takes me a week before I have to come back I would like to be seen in a week as opposed to 3 weeks because that would mean the rate of the process is quicker..."***INT#1.M*

It is clear that the participant has either not been made aware of the need to leave time for movement to occur in between appointments, or their understanding has not been checked after information has been provided. Either way the lack of information has led to avoidable dissatisfaction. The same can be seen in the following quote from a participant who felt that she was not given enough information about the length of treatment she would be taking on:

*'' The timing of it. I just ... only in that I wasn't told after the operation how long I would still be seeing the orthodontist for."***INT#10.F*

A further respondent felt that the full ramifications of her surgery were not explained beforehand:

*'' Yeah, I was in a lot of pain, I was on loads of tablets and medicine a day, so I didn't think that really helped my moods, but I would not advise people to have it done, well I would, but I wouldn't be, like me and my mum were talking about it and we didn't really know the whole of what the recovery would be before I had the operation, we didn't realise how long, how hard it would be, because we never really got told, but I'd advise people, if someone was having the same operation that I would, I'd advise them to know literally every single thing to make sure they want to go through it first, because it is quite, because obviously, it's the biggest operation really and it's quite hard."***INT#9.F*

This led her to suggest other people should be far more informed prior to surgery, not just in relation to what is involved but also the length of time needed for recovery. Again this ties in with the need to think about treatment within the wider context of daily life.

Even where information was provided there was no guarantee that it would be helpful to the patient. Previous examples suggest a potential lack of understanding about information that was provided, but some participants also highlighted problems brought about by receiving different information from different clinicians. This was seen as a particular problem in a care pathway like this one where a large multidisciplinary team is involved and may cause confusion and uncertainty.

> *'' I'd say sometimes there can be a sort of ... it's quite interesting, a hierarchy when you're in the consultation room and … the speech therapist and the nurses and things. And I know when I've been to the hospital with my mum quite a lot over the last couple of years we keep finding that we're going to consultation, and we come out being told something different. It has been a bit challenging for us. I think we try to adapt and expect something to come up or something new to be suggested, but it still can be a little bit difficult. Yeah.''* **INT#3.F**

Receiving information about different aspects of treatment from the CLP psychologist in conjunction with the clinicians was found to be positive and a way of helping to ensure information was understood. It was suggested that the psychologist could break down the clinician loaded information from clinics and re-tell the information in a more patient friendly way and a less pressured environment.

> *'' I think that the psychiatrist would be a bit more relaxed. It would be one to one, it wouldn't be a panel of doctors talking to me, it would be one person talking to me, making sure that I understand, asking me questions instead of me asking a doctor questions, if that makes sense?''* **INT#2.F**

Another possible benefit was that psychologists may have been able to adopt a more holistic approach giving consideration into the context, whilst clinicians may have been more likely to provide information that was relevant to the clinical procedures.

> *'' I think psychiatrists are more social in the way they talk and doctors are more precise and medical. I feel that talking to psychiatrists would be easier for me. Like I said, I get these big meetings with the doctors and they are very spaced apart like they say "I saw you two years ago" or "I saw you a year ago, a few months ago" and that information has left me. I know that is because they have loads of appointments to go to and whatever doctors do, but I know that psychiatrists are more accessible. I think it will be easier for me as well as the doctors if I went to them more often.''* **INT#2.F**

This highlights the importance of the multidisciplinary team with different, but complimentary, skills.

A final issue highlighted in relation to information was that information needed to change over time. The need to know about treatment tended to grow with time and develop as the individual became older.

> *'' I don't know a lot about it actually because as a child, sort of forty five years ago we were just put in hospital and not really told what we were having done so I just grew up to accept it and we weren't sort of given counselling or explained a lot really and I've never felt that I really needed to know much about it. I mean obviously I've looked into it in later years but it just shows how ignorant when you asked me what type I had I've just not really thought too much about it, I've got it but yes, you know I just know that part of my lip and my palate didn't form properly and I've just had surgery to repair it throughout the years.''***INT.12.F***

In childhood, professionals usually tended to provide the information to the parents rather than the child possibly because of the legal obligations towards the responsible person who was going to sign a consent form. This meant, however, that by the time the young people reached the age where they were expected to take an active part in decision making they often had little, or only a sketchy, understanding of their condition and the treatment that they had received so far.

> *'' ... it's when you get more older you start to learn about it so the way you could do it is if you could ... it would be a bit better yeah if you talked more about it to the patient than to the carer, yeah.''***INT#14.M***

This lack of information could make an informed decision difficult, a theme that is explored in more detail in the next section.

6.1.3.4 Decision/Choice

Increasingly patients are expected to take an active role in their care and the new rhetoric around patient centred care focuses on patients making informed decisions about the kind of care they want to receive. Participants in this study gave data about the role that they played in making decisions about treatment. The key findings are presented in (Figure 17) and can be distinguished between those who felt able to make decisions relating to both place and type of treatment and those who felt they were given little choice. The factors affecting decision making and choice were then explored in more detail.
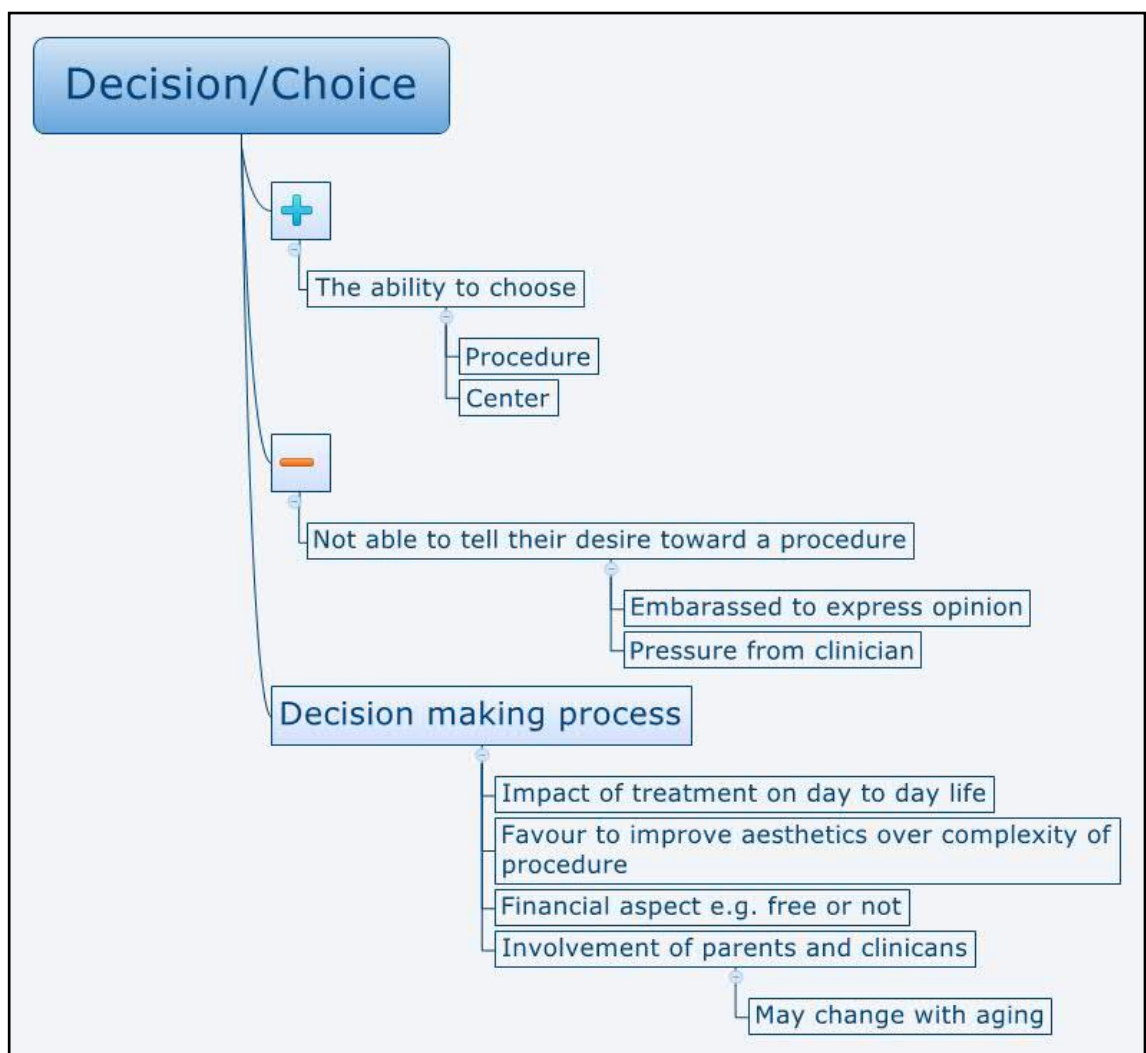
Figure 17: Decision/Choice

The ability to choose was perceived as a positive feature of the care. For example, a participant felt that she was able to make informed decisions about the type of care that she received:

*"Positive, I think definitely that I was very much involved in the process of the treatment in many different ways and I was given choice and options of what I think is better, what do I want to get out of this more than we're going to do A, B, C and D, it was more of a joint decision and I was always very well informed, I can always ask questions and stuff like that."***INT#7.F**

And the place in which the care would be provided:

*" I was getting the braces to start with, we decided to ... we had the option of coming here or going to the local orthodontist ... and it will probably be like ... they would all just start of a bit quicker if we just stayed here because my sister had to wait quite a long time because she had to have chains I think, so her whole braces thing took a long time so we just thought it would be quicker if we stayed here"***INT#15.F**

This had a positive impact on how they felt about the care they were receiving. The way that decisions were made and the amount of information needed prior to making decisions would depend on the type of treatment being considered.

*"Yeah on the operations that are more important to me like the operations like having my teeth pulled out from the root because of the palate obviously, it is just a few teeth it's not really going to bother me. But with something big like having nose surgery then obviously I'm going to ask more about it and what the outcome is going to be. It depends if the operation is going to be big or not. I mean if it's like a big deal then I'll look on the internet and look at more pictures and look at what the improvement is on some people that do have it and that do take it"***INT#27.F**

This is a good example of a participant who took a very active role in their treatment when they felt it necessary to do so.

There is not a uniform response to decision making, however, and where people did make active decisions about the treatment they were going to undergo they did so for a range of different reasons which may or may not have been related to clinical considerations or outcomes. One participant, for example, explained that they made their decision based on which treatment would have less impact on day to day life:

*" I chose the orthodontic route…I was kind of for I going to have orthodontic anyway with the surgery…and obviously I was at that point where I had a job as well…so committing to a time to have the surgery as such as well…because if I have the surgery but then he said the pain you kind of be feeling from it will last a good few weeks…he said I wouldn't be able to eat like solid food even for a good month or so…it just would been a commitment that I was prepared to do…but then when I found out there is the other option that didn't require that commitment I just thought that made more sense to go for that option and take the orthodontic route…"***INT#5.M**

Conversely, however, other patients talked about choosing the treatment option most likely to have the best aesthetic result despite it being the most invasive:

> *'Well I guess always when you have an option in life of doing something else, definitely you should go for looks always better, I think that more than anything else, I knew that the whole treatment would be more lengthy and probably painful and time consuming but in effect it was the better solution long term. So yeah I wasn't happy about my braces but I think what helped was that, pardon me, I got the lower brace a few months before the upper brace, so I wasn't just given train tracks on both of my jaws.'''* **INT#7.F**

As found in the section looking at reasons why people decided to undergo treatment, aesthetics was the single biggest motivator for treatment choices.

> *'' I saw my friends or my family and if there was anyway that I could look a little bit more like those people then I was going to do it and if it meant that I had to miss a bit of school here and there or give up three or four days to have an operation and I was going to be knocked out anyway then it didn't matter too much to me. So I wanted to look more like them so that is why I did it really.''* **INT#8.F**

> *'' Because as part of my cleft and palate, I have a bad bite, so it messes up my teeth, so I have to get that sorted but I didn't have to go orthognathic surgery, I didn't have to go with it, but it's more of an, it's not compulsory it was my choice and I chose it because if I have an option to look better, then you're obviously going to take it so.''* **INT#21.F**

> *'' So the decision was very easy for me. It is daunting because it's an operation and it is a major thing and your cleft palate is a big part of your life but I think it is important to go through the operations. Even though you look in the mirror and you can see the swelling and you can see the different facial features and you kind of lose your identity of who you are ….''* **INT#28.M**

The quotes here show the importance placed on aesthetics by some participants, tying in with the findings in the previous chapter around stigma, identity and low self-esteem from looking or feeling different. Treatment could be seen here as a way of alleviating this difference.

One final reason participants gave for taking an active part in decision making was more practical and related to finances. Some cosmetic treatments were only available whilst people with CLP remained within the care pathway and so there was a pressure to get all expensive treatment completed within this time frame.

> *'' … because after twenty one it costs, so I agreed to it before I was twenty one and then I had it done in June this year, I was twenty one in January, had it done in June and that*

*was good, that was a day surgery but I stayed in overnight and had a weeks rest or recuperation where its blurred a bit, then I was back.''***INT#11.M**

One older participant, treated initially prior to the development of the pathway, also talked about being referred back in to the pathway to receive treatment that had not previously been available.

*'' Yes, like I say I initiated it, it was probably about six years ago I went to my dentist and I asked them to refer and said about having my teeth straightened and he gave me a really high quote and said it would cost me thousands and I sort of just said well is it not because of a medical condition that you know that I need to have my teeth straightened. So he then actually referred me to [cleft] services and I suppose because I was seeking the treatment I can only say it's been a very positive experience,''***INT#12.F**

Again here financial factors played a key part in how treatment decisions were made, rather than simply clinical outcomes.

Whilst many of the participants in this study felt able to make key decisions about the treatment that they underwent, some felt less able to do so. Whilst not suggesting that the clinicians themselves were intimidating, a number of participants highlighted the difficulty they found in asking questions or challenging treatment options laid out by the expert:

*"Yeah. I don't think I've ever felt, oh, I'm being forced to do this. But it can be quite hard sometimes to sort of ... I don't know. I think I'm thinking more of surgery here but it can be quite hard to sort of go against what the surgeon is saying or the orthodontist is saying, because they do know their stuff, they do know that they can do really good things and they can change how you look or how you eat or speak. … I suppose it can occasionally feel like you're sort of just a cog in the machine and you just have to keep going along with it. And I suppose there comes a point where it would be odd to stop half way through when they can do so much more and it can make such a big difference. I don't know. ''***INT#2.F**

This kind of unintentional intimidation was experienced by several participants:

*''Yeah, that was offered to me a while ago and I didn't really want it and I was never, I was never really sure I wanted it but they kept convincing me, they weren't telling me to do it, they said well you could do this, we could do that …''***INT#11.M**

This suggested a problem related to communication and rapport was influencing the ability of the respondents to participant actively in their care, alongside the power relation between a patient and clinician.

One participant also highlighted the fact that many decisions within the pathway were taken by parents rather than the patients themselves, particularly when the children are very young which is often when the big surgical interventions were undertaken.

> *"I think it's more of a ... I think my parents would probably have made a lot of the decision making when I was younger whereas obviously as I've got older I've made a lot more of the decisions where there's been a decision to make based on my own thought rather than them. Like a lot of the surgery happened when I was young and so obviously I'm too young to decide whether I need like bone grafts and all that sort of stuff, whereas the jaw surgery was my choice, they offered it and I chose to go with it, so that's probably the most amount of choice I've had. ."***INT#26.M***

What is clear from this is that there is a lot of variation both in the active decisions that different patients made and in whether they felt able, and enabled, to make those decisions. Furthermore, even when they were making active decisions, they did so for a range of different reasons, few of which were based solely on clinical outcomes. Understanding the reasons why patients do and do not get actively involved in their care, along with motivations and enablers (information, rapport, communication skills) will help clinicians to improve the care pathway and patients experiences of it as well as improving patient satisfaction.

6.1.3.5 Effects of treatment

This section of the results explores the effects of treatment on participants. Focussing specifically on the definitive orthodontic treatment and/or the orthognathic treatment. A detailed description of the definitive orthodontic treatment is presented using participants' voices and incorporating immediate effects of treatment such as pain, or the impact of the treatment on meal times and eating. These immediate effects relate specifically to the nature of the treatment itself and are qualitatively different from the data presented in the previous chapter which focused on the impact of treatment on day to day life, exploring how the individual`s life commitments were affected as a consequence of treatment.

Orthodontic treatment

Whilst much of the data collected for this study was broad in remit, seeking to contextualise participants` experiences of the cleft care pathway within their everyday lives, a set of detailed questions were asked relating specifically to the definitive orthodontic phase of treatment. As previously explained, this reflects the significance of this part of the treatment both as the likely endpoint of the pathway and of treatment itself. In this section the data collected from the detailed questions about the definitive orthodontic stage of treatment are presented (Figure 18). The data are arranged chronologically to reflect the treatment process being described:

- Prior to the fitting of braces
- At the time of fitting and whilst the braces are *in situ*.
- After the removal of the braces
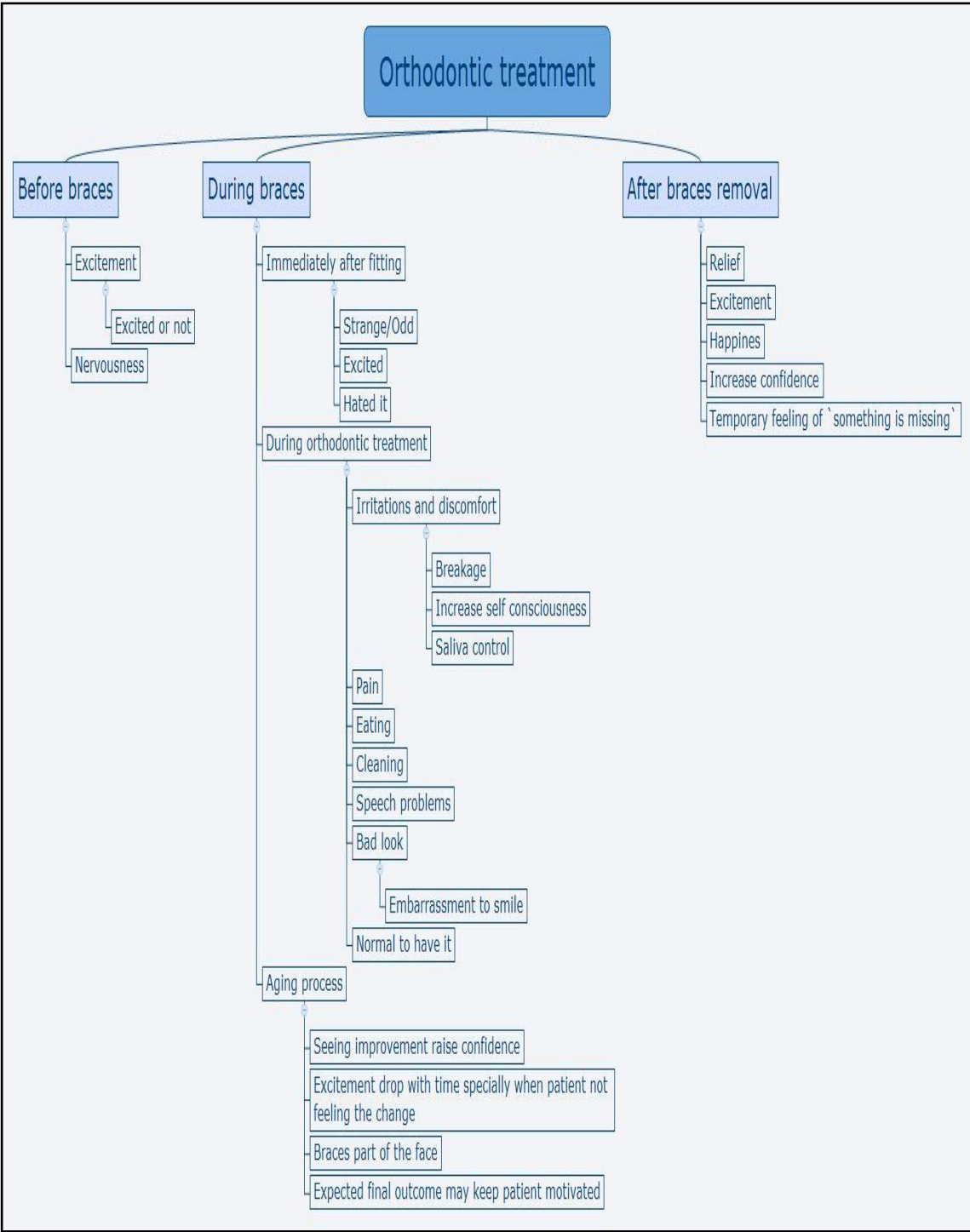
Figure 18: Orthodontic treatment

Prior to the fitting of braces

When asked how they felt about the definitive orthodontic stage of treatment participants expressed a range of positive and negative emotions. The idea of having braces fitted was approached with a mixture of excitement and trepidation by many participants.

Excitement at reaching this stage of treatment and at the expectation of finally having 'normal' teeth was expressed:

> ''I felt excited, because I know that these braces are going to straighten my teeth and I'm going to have straight teeth.''**INT#23.M**

This was often alongside some trepidation about the process itself:

> '' I was a little bit anxious a little bit, just because you know they don't look particularly great, they look, and you hear all these stories about being, they were quite painful and stuff so a little bit of anxiety but not too much.''**INT#24.M**

And about the potential outcome and whether it would meet expectations:

> '' A little bit nervous just because I didn't know what I was going to look like, what it was going to feel like, how would it affect me but they always told me what I can and cannot do so they just sort of, before the treatment they always informed me what they were going to do, what it was going to look like and just, there was a lot of preparation before putting the braces in.''**INT#22.M**

Unlike other aspects of the pathway, orthodontic treatment was relatively common and many of the participants' friends also had braces. Whilst this normalised this stage of the treatment process, it also led to some trepidation about the potential effects of living with the brace itself. Information gathered from friends about orthodontic treatment lead to some concerns, particularly about pain and the look of the braces themselves:

> '' No. I wasn't even excited either, no. [Why?] Knowing that I was going to have braces, I don't know, the pain, looking funny. But I know quite a few people that have got braces and they don't care anymore, but it got to the point where I hated it. I felt embarrassed and a bit silly but now they are off it feels great.''**INT#27.F**

In this example the anticipation of discomfort and pain was realised in the treatment itself and in the length of the treatment. This is looked at in more detail in the next section. The quote above puts the discomfort experienced in the context of a positive outcome, and it is worth noting that the accounts presented here are from people who have completed their treatment and been debonded or at the finishing stages of treatment, so will be shaped by the perceived success of the treatment as well as the experience itself.

At the time of fitting and whilst the braces are *in situ*

Effects outlined during the period when participants were fitted with, and wore, braces included how they felt when they first had their braces fitted and the emotional and physical effects of the braces themselves.

Perhaps unsurprisingly, many participants talked about the initial impact of having strange material in their mouths:

> *'' It felt a bit odd at first, I didn't, I weren't sure what to do, but I remember when I had it off, I remember going, when we went to go into McDonalds it was the worse idea ever because I got a cold drink and I didn't realise that the cold stuff would really affect and it really hurt my gums and my teeth, made them feel a lot colder than what they normally did, normally do, when I have a cold drink, sometimes when I used to have hot food or hot drinks, everything used to tingle on my teeth but after a while, it faded away but sometimes hot and cold things did hurt a bit whenever I used to eat or drink, but I think it's because it's metal and it's reacting to the metal so it, but.''***INT#13.F***

Whilst some participants got used to the braces, however, for at least one participant it was an extended period of misery:

> *''Bloody uncomfortable, horrendous (laughter), I hated them so much''***INT#17.M***

They went on to explain:

> *''But when something went wrong with my train-track, something went wrong big time, and it was always very painful. And there'd be instances where the rings round my molars would come out, or something would crack. ……… So they'd break quite a lot and I'd have to have them repaired quite a lot. It led to a level of frustration where I'm thinking, "Oh, for God's sake, they've broken again, I hate these things. I can't drink … coke, it hurts to brush my teeth at night, I have to take Paracetamol pills, there are these metal things in my mouth and they're scraping the side of my jaw, and sometimes they get caught." I hated them, absolutely despised them.'''***INT#17.M***

In this example the misery was cause by a combination of factors exacerbated by discomfort.

How participants felt when they had their braces fitted was shaped not just by the physical experience however, but also by the wider expectations of the treatment and of 'fitting in'.

> *''it didn't really impact my life having a brace that much because so many people have braces now, it's just normal, even if I didn't have a cleft, people just say wear loads of braces,''***INT#7.F***

> *''I was so excited, I really was, because all my friends had train tracks and I was getting them on, because I thought I was so cool, because they all had train tracks''***INT#21.F***

In the second account the feeling of normality was tempered by the extended length of treatment, but the excitement at the end was more obvious possibly because of this:

> *''I was in the days where people were getting train tracks and braces, but when everyone was getting them off, I had to still have them on, so that was quite annoying, but I was really excited when I was getting them on.''***INT#21.F***

Orthodontic treatment may help participants during their treatment to fit with 'normal'. However extended treatment length can be viewed as 'out of normal'. Having orthodontic treatment within the CLP care pathway felt to be normal for CLP participants as this sort of treatment is quite common among teenagers at that age. Orthodontic treatment is one of the few parts of the cleft care pathway where treatment reflects experiences common to many teenagers without cleft as well as those with it. Whist the timing may differ and the braces often have to be on for longer periods, the feeling of 'normality' is widely seen as positive. The data shows that participants' initial reaction to receiving braces differed according to a range of factors including the experiences of others as well as their own physical experiences and expectations. During the orthodontic treatment participants experienced some direct physical effects caused by the fixed appliance treatment. These effects were almost entirely negative and included discomfort, pain and speech problems, along with aesthetic and psychological effects related to the look of the braces themselves, embarrassment when smiling and increased levels of self-consciousness.

-Pain

Experiences of pain were consistent in the data from the first fitting of the braces. As the data will show, this could influence eating and cleaning behaviour. The pain associated with the braces could be described as episodic. It started when the braces were first fitted and lasted for a few days to a couple of weeks. Then it dropped down gradually until it faded completely. The pain then returned again when the wire was changed or tightened and this pain again then lasted for a few days until it faded again. (Figure 19).

> *'' Painful, because obviously braces are painful, they're not really like comfortable to have, so when they're getting changed, they're painful, except from that, it helped, it's helped my teeth to get to how they are now, and yeah, it's been fine, the only weird thing I can say about braces is that they're painful. ……Like when they get changed and tightened, anyone will tell you, when they've had braces, have you had braces, exactly, they're painful, when they get tightened, when you come out of having braces, it's very*

*uncomfortable, but when you've had them for a while, it's fine, but it's just having them changed, it's a painful process."***INT#19.M**

Fixed appliance treatment can cause discomfort and pain to any individual of any age undergoing orthodontic fixed appliance treatment. One participant suggested, however, that their experience of orthodontic treatment was more painful as an adult than as a teenager.

> *'' The actual orthodontic treatment I mean was a pain as a teenager obviously having braces but I found it even more of a pain as a forty odd year old having braces, it annoyed me far more this time round than first time round and the jaw surgery as a teenager"***INT#12.F**

It is not clear from this, however, whether it was the physical pain or the disruption which were most disruptive.
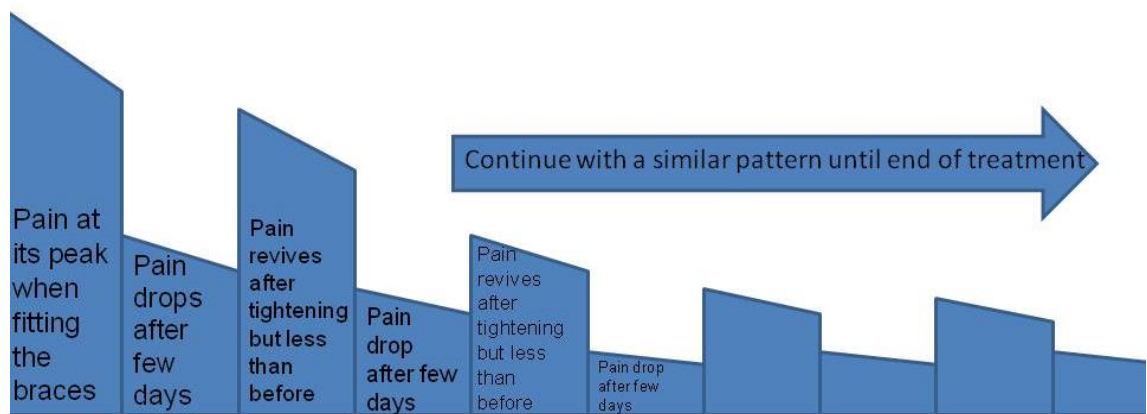
Figure 19: The nature of pain associated with orthodontic fixed braces

Whilst pain was a universal effect of orthodontic treatment, participants managed their pain differently, using a combination of changes in behaviour, mind over matter and analgesics. One participant, for example, found that drinking warm soup helped to reduce the pain.

> *'' Yeah. I mean, it was primarily that and sometimes I'd have to be … I'd drink soup quite a lot to pacify the pain. Like, if I was having a particularly terrible day, I'd say to my parents, "Look, I just want soup tonight, just give me soup, I do not want to have to use my mouth at all." So they'd make me some homemade soup to compensate for the fact of that. So there wasn't just discomfort and pain; it prevented me from eating properly for a while. But that, primarily, was a side-effect of the pain and discomfort."***INT#17.M**

Other participants used psychological techniques such as thinking about the end result and the reason for enduring the pain:

> '' Only when it comes to pain wise, I'd want it off, like I'd go in to, for them to change the wire to tighten it up, and that's when I'm like, I can't handle this anymore, just take it off, I want to take it off, but I reassured myself in my head that I need this on to keep me having straight teeth, I just need it on and that's what drove me through the procedure.''**INT#23.M**

Whilst yet another participant used mind over matter in a different way, to confront the pain:

> '' ahh…not really no…having them tightened..make obviously a couple of days after having them tightened would make it feel a little more painful but I find that a bit…weirdly nice like quite..whenever I could feel it getting tighter…I actually quite…not enjoy it but I didn't mind that pain…ahhm…because to me it wasn't a pain is such like I was aware that it wasn't exactly comfortable …but no I don't know when I feel the pain I just like bite down on my teeth even more in a way to make it more excruciating to a point that I didn't actually mind it…and that was only for a couple of days anyway… after having it tightened…and so that's obviously every few months…mmm…they didn't hurt and that's was literally just a general discomfort that I would probably make it worse than it was by deciding to bite down on them..which I really don't know why I did that but…I think in a weird way I quiet liked the pain…I really don't know why…but there we go…''**INT#5.M**

Alongside behavioural and psychological coping mechanisms, painkillers were regularly used and seen to be helpful:

> '' because of the medication that they gave me, the type of medication they gave me to help me ease through the pain,''**INT#23.M**

Whilst there was universal agreement that braces were painful and disruptive, however, it is also worth noting that there was wide agreement that it was an important treatment process with valuable results.

-Eating

One of the quotes above highlighted the way in which warm liquid could be used to ease pain. There was also a more widespread acknowledgement that undergoing orthodontic treatment affected eating behaviour more generally. Some foods were seen as more difficult to eat, often causing pain. This was often due to problems biting in to hard food:

*" I remember they put like a band to push one of my teeth forward..so I remember getting it and I remember biting into it like a baguette which is like the French bread and I remember I couldn't do it because it hurt my teeth...but it didn't feel like it hurt it hurted but when just I bit into it really hurt..I felt my teeth is going to fall out...then I started to pick the inside of the sandwich the lettuce..I couldn't even eat the lettuce..I remember that very well like I couldn't eat my food..and then people like ohh yeah it hurts you can't really eat it...so that's what I remember yeah.."***INT#1.M***

But could also be due to heat and the effect of hot and/or cold food on sensitive teeth.

*" but I remember when I had it off, I remember going, when we went to go into McDonalds it was the worse idea ever because I got a cold drink and I didn't realise that the cold stuff would really affect and it really hurt my gums and my teeth, made them feel a lot colder than what they normally did, normally do, when I have a cold drink, sometimes when I used to have hot food or hot drinks, everything used to tingle on my teeth but after a while, it faded away but sometimes hot and cold things did hurt a bit whenever I used to eat or drink, but I think it's because it's metal and it's reacting to the metal so it, but."***INT#13.F***

*" I mean, like, sometimes I'd just have soup. On particularly bad days I would avoid things that were sensitive to my teeth, so ice cream would go out the window. Even though ice cream was very simple to eat, the temperature of ice cream would inflame my mouth and cause my mouth to hurt. So I wouldn't have ice cream."***INT#17.M***

Particular types of food were chosen to reduce the likelihood of pain and the inconvenience of broken appliances.

One participant suggested further, that the inconvenience and pain associated with difficulties in eating could have knock on effects such as causing tiredness:

*" I mean, like, sometimes I'd just have soup. On particularly bad days I would avoid things that were sensitive to my teeth, so ice cream would go out the window. Even though ice cream was very simple to eat, the temperature of ice cream would inflame my mouth and cause my mouth to hurt. So I wouldn't have ice cream. I stopped eating apples because I once bit into an apple and actually broke my brace. And then another time, a bit of the apple got stuck in my brace whilst at school and I couldn't get it out, and that wasn't particularly good fun. So some kinds of fruit I sort of gave up for a while. I didn't lose any weight, I've always been quite slim, but there were certainly periods where I felt tired because I wasn't eating. Like in the morning, say, I'd have a cup of tea and, if I could manage it, some toast – I wouldn't eat cornflakes because, again, the milk would inflame my teeth. I'd avoid it."***INT#17.M***

But it was noted that these effects were transitory and eating habits reverted to a more normal pattern in the weeks after fitting or tightening.

*'' yeah because of the pain yeah..but after that you needed for while eating yoghurt and that and then after a week you eat like bread soft bread and last you just go back to normal..but you don't really think about it you just do what you felt...you just follow you body if it hurts you don't do it..alright you do it ..and then after a while you a little bit there it's a little bit painful but you still eat it..and then you are fine.."***INT#1.M**

One participant explained that eating habits change not only because of the braces, whilst their habits had changed over time, this was more to do with the aging process than the specific influence of the orthodontic treatment.

*'' ahhmm…no I wouldn't say the train tracks who did such I think it was just my general growing up mentality that changed that…specially my age having them…yeah I've got them a week before my 18th birthday and had them off the week after my 20th so…obviously that kind time I do a lot of growing up anyway so I've changed a lot of my general diet…around that time just as a thing well I don't need pizza for dinner every night….obviously you start going out for food with your friends…ahhh…to restaurant and stuff so…a lot of I was always quiet a fussy eater so obviously experiencing more food like now I'll eat anything…so I wouldn't say the train tracks had any impact on that its purely my mental health and own taste buds that changed that…I wouldn't say the train tracks had any bearing really.."***INT#5.M**

This may suggests that the impact of the braces was temporary, both in relation to pain and food.

-Cleaning

The other practical issue that was raised by participants when asked specifically about the effects of orthodontic care related to the need to keep the appliance and the mouth clean. The fact that braces have a tendency to retain food because of their irregular shape was acknowledged by one participant:

*'' but I felt comfortable wearing them but whenever I was eating, I always felt like I had to go and brush my teeth straightaway after I'd eaten, because I didn't like the fact that whenever I'd eaten something, it used to get stuck in my brace, but that's the only problems I had."***INT#13.F**

Whilst another participant talked about the difficulty, and importance of maintaining oral hygiene. Poor oral hygiene was a well known risk of braces and can cause decalcified and decayed teeth:

> *'' so I was trying to copy in a way and I used to brush my teeth quite a lot, but it never seemed to make any difference, it still looked the same as what it was before, so my teeth did become quite a bit stained, because I weren't able to get in the places where I needed to get, and even though I used those little dents in brushes and still it weren't doing as much as what it should have done, but.''***INT#13.F***

Both the difficulty of maintaining oral hygiene and the acknowledgement of the effects of failing to do so can be seen in this quotes and reflects the widespread understanding of the importance of maintaining oral hygiene that emerged through the data.

Some people made no changes to their usual cleaning routines,

> *Just brush your teeth, that's all, you've just got to brush your teeth, I didn't really find it hard, just brush them like you normally do without braces,''***INT#19.M***

Others acknowledged an awareness of the fact that they felt they probably should have done:

> *'' you know what I heard that you had to get like a little ..I did the [...] normal tooth brush was probably isn't ideal but [laugh] yeah ''***INT#1.M***'

Most, however, talked about the need for a more rigorous routine. One of the effects of braces on cleaning is that cleaning could take longer than usual.

> *'' It's fine, it takes a bit longer but it's, it's fine.''***INT#15.F***

And involve the use of different cleaning aids:

> *''... a special brush and a normal brush. But I do use a normal''***INT#15.F***

Having braces may change the cleaning behaviour for the better. Spending more time to clean and/or increasing the frequency of brushing was acknowledged to have a positive effect:

> *'' The braces really didn't affect me in any way, not really. I still ate the same food. Just my personal hygiene improved, that's all.''***INT#2.F***

The data suggest that cleaning regimes could be affected but were not seen as a problem but rather a minor inconvenience at most.

One participant, however, admitted that he became obsessed with cleaning after he was fitted with braces. He increased his brushing frequency and duration.

> *'' this I started while having the braces yeah but I got just In a habit of it now…but I consider it a good habit because it's not..ok it makes the general teeth brushing go from*

*2 minutes to 5 minutes…it's not a great deal of time…ammm… I tend to when I'm brushing my teeth once I actually got the second brush in my hand and I'm going around with the tooth paste..I tend to liker have my phone in hand or something anyway and I'm messaging someone or whatever…so in a way that even can make the time go for longer because I'm distracted whilst I was doing it…obviously I got the tooth brush in one hand brushing the teeth and then phone in the other…I can then like realise I've been brushing my teeth for about 4 minutes and so obviously then think…right probably I should get rid of this mint off my mouth before it burns me…brush to the sink and that's it really…and I've definitely got in that habit of taking a long time with my teeth brushing but I don't think I'm the only one to that I've known a lot of people who had braces and they do the same kind of thing…do a little brush first without anything on to get rid of any food or whatever that might be stuck…and then go and do a proper clean…so I don't think I'm the only one to do that but…I have to say it's a good habit"***INT#5.M***

Even in this case, the increased time taken was seen to have an overall benefit. Only one participant spoke of difficulties with cleaning that actually prevented him from keeping his mouth clean. In this case it was the pain that proved the biggest problem rather than the time and inconvenience.

*'' it hurts to brush my teeth at night, I have to take Paracetamol pills"***INT#17.M***

*'' It was very difficult to clean my teeth sometimes because, one, the braces would physically get in the way, so it was a hard time keeping one's teeth clean. And because they were discomforting, I'd sometimes not want to touch my teeth, I'd be like, "I'm not brushing my teeth, I'm not sticking that in there, it'll just make them even worse." So there'd be periods where I wouldn't actually brush my teeth.''***INT#17.M***

The results here suggest that pain and inconvenience in relation to cleaning and eating were widespread but accepted as a part of the treatment process, and whilst pain was made bearable because of the time limited nature of it, the increased attention given to cleaning was actually seen as a positive in the longer term.

-Speech

Difficulties associated with speech and pronunciation because of the brace were present in the data. One participant explained that they developed a lisp which affected their speech. This could make communication more difficult and challenging, particularly for those whose speech had already been affected by the cleft itself. This was seen as a particularly difficult aspect in employment:

'' It was more, when I was at college, it didn't affect me as much, because I weren't talking as much as what I used to, but once I got to, when I started my job, it, I was more interacting with customers and my colleagues, trying to talk to them, and some of the

customers that come in, fine, but when you get certain customers that try to tell you everything and you're trying to explain something to them in detail, that's when I found it more harder because I was talking more and then once I'd finished talking, my mouth just started drying up, so I had to go and get water, so it was just more." **INT#13.F**

Again this example illustrates the fact that treatment, and the effects of treatment need to be understood in context. It is the employment context that the participant was in at the time of treatment which exacerbated the negative impact of the treatment.

-Appearance

In addition to the practical issues raised above, participants highlighted the aesthetic impact of wearing braces. This affected how people perceived themselves as well as well as how they were seen, or thought they might be seen, by others.

'' It's like ... they're not the nicest things in photos and stuff like that and it's just that it'd be much easier because if you didn't have time for the appointments and didn't have to wear elastics and stuff like that, it's just ... yeah, maybe ... it will be nice when they come off because sometimes they hurt as well so, it's just ..." **INT#15.F**

It also influenced behaviour. The participant can also feel embarrassed to show their teeth because of the look of the braces. This feeling may have some influence for the patient to stop smiling:

'' I don't smile. Well, I smile at this but I don't smile so people see my teeth. If I'm smiling I'll always do that, I will never do that." **INT#26.M**

What is not clear here is whether the visual impact of the braces caused people to be seen or treated differently, whether it resulted in felt and/or enacted stigma, or whether it simply impacted on self satisfaction with appearance.

-Discomfort and irritation

Fixed appliance breakage is common with treatment which may be annoying and frustrating for the patients because they have to go to clinics and fix it. This includes bands or brackets. Also, significant amount of irritation can be caused by the braces to participants soft tissues.

''But when something went wrong with my train-track, something went wrong big time, and it was always very painful. And there'd be instances where the rings round my molars would come out, or something would crack. I remember I was at a … in the last year of

having train-tracks I was at a concert, and in the beast of the crowd where everyone's jumping up and down, and somebody whacked me in the face by accident – total accident, didn't mean to do it. And he knocked one of my train-tracks out. And that set me back three weeks, where I had to have that repaired and we had to wait for the damage to be corrected because, obviously, my tooth stopped moving in that time. So they'd break quite a lot and I'd have to have them repaired quite a lot. It led to a level of frustration where I'm thinking, "Oh, for God's sake, they've broken again, I hate these things. I can't drink … coke, it hurts to brush my teeth at night, I have to take Paracetamol pills, there are these metal things in my mouth and they're scraping the side of my jaw, and sometimes they get caught." I hated them, absolutely despised them.'"INT#17.M

One participant reported that self-consciousness was higher as an adult wearing fixed appliance braces. This was particularly annoying when eating in a social context like going out with friends.

'' Well I wouldn't eat, for example, during the day when I was at my house or in public, I wouldn't necessarily eat either cous cous or food that is quite small that can get stuck between your teeth, I guess carrots if it has some parsley in it it can get stuck between your teeth as well but that was more for aesthetic reasons than anything else''INT#7.F

Saliva control was an issue. Spitting was experienced as an effect of braces and this made it initially difficult for communication.

'' You get this sort of lisp thing and the spitting as well you know but it's one of those things that I was, I didn't really get too frustrated about that because it's just one of those things that everybody knows if you have braces on then that's what happens so, you know if I was to spit or people could hear a lisp or whatever then it would just say it's down to the braces and they would understand, so that wasn't really a problem.''INT#24.M

The end point

The final, but potentially most significant, point to emerge about participants` experiences of orthodontic treatment related to position of this treatment within the pathway and the sheer length of time that the treatment took. Average treatment time is between 2 to 3 years and it can be considerably longer where there is a CLP with other craniofacial disorders. The definitive orthodontic treatment was also often the very last process undertaken within the pathway. What was interesting; however, was how this manifested itself. Participants talked about being able to see a physical and aesthetic change in their teeth as the treatment progressed.

> *'' When you see improvement your feeling changes, when you start seeing an improvement with your teeth, you start feeling more confident and that's it.''***INT#19.M**

Where the change was visible this impacted positively on both motivation to continue and also self-confidence. At this stage treatment adherence was often high as the motivation was to finish the treatment as soon as possible.

> *'' Like I was looking forward to getting them off but other than that there's not really much that needs to be motivated by, I just ... like they do the work, I wear the bands, I don't find that I need to motivate myself to do that, it's just what needs to get done. There's not really much motivation required, I just carry on going until I have to stop going. ''***INT#26.M**

The expected final outcome could help the patient to be motivated to continue and finish treatment.

As prolonged orthodontic treatment can cause frustration and annoyance, taking the braces off was described as a relief.

> *'' But you know it's a relief to have braces off, it's a good feeling knowing that you haven't got to struggle anymore to brush your teeth and with what to eat and things like that.''***INT#27.F**

Although for some the happiness was mixed with a strange sense of something missing.

> *'' because after such a long time of having braces and being comfortable it felt like something was missing when I took them out.''***INT#20.F**

Happiness and excitement were stated after removal of the braces. For example, looking in the mirror and showing people how the teeth look after braces. In addition, the person may feel more confident to smile. However, this feeling may not last for long.

> *'' It felt weird at first, I must say [laughing] because they didn't feel like my teeth at all when they came off, they were really shiny and I could feel my gums again, I hadn't been able to feel them for a long time……… I had a lot of people going, oh come on, I want to see what our teeth look like, you've got them off, not, well off, but you had the braces off on my teeth [laughing], they would keep getting me to smile all the time, and it's died off now, but at first it was oh let me have a look at your teeth, oh you haven't got your braces any more, they all just kept wanting to see what my teeth looked like without them.''* **INT#13.F**

This final look of the teeth may have helped the individual to reach 'normality'. This ties in with the experience of stigma and feeling different and the importance of aesthetic reasons of treatment.

> *'Knowing that it was going to come off in the end and knowing that I'd look different and it's how I'd wanted to look, if that makes sense, look more normal to other people.''* **INT#9.F**

> *'' At the end of the day what do you want? Do you want to feel a bit of pain or are you going to walk around with bent teeth? Because I know surely I'd prefer to have a little bit of pain in my teeth knowing the end result's going to end up straight.''* **INT#16.M**

Participants talked about their orthodontic treatment as the endpoint, the point at which they would become 'normal'. This stage of treatment potentially had been predetermined many years ago and talked about as a given throughout the treatment. This magnifies the potential importance of this stage of treatment, and, as will be seen when we look at satisfaction, can impact on how they feel about the end result of their treatment.

In this section, participants gave a detailed longitudinal description of their definitive orthodontic treatment with all positives and negatives. The next section will provide some insight into the jaw surgery procedures that some CLP participants may have it as part of their treatment.

Orthognathic surgery

This patient cohort included participants that underwent jaw surgery as part of their definitive orthodontic treatment, so in addition to looking at the definitive orthodontic phase of treatment, it is possible to use the available data to gain some insight and understanding of the experience of orthognathic surgery.

Orthognathic surgery involves a combination of fixed orthodontic braces and jaw surgery. It is a sort of treatment approached when orthodontics alone cannot correct the dentofacial problem which may indicate a more severe problem.
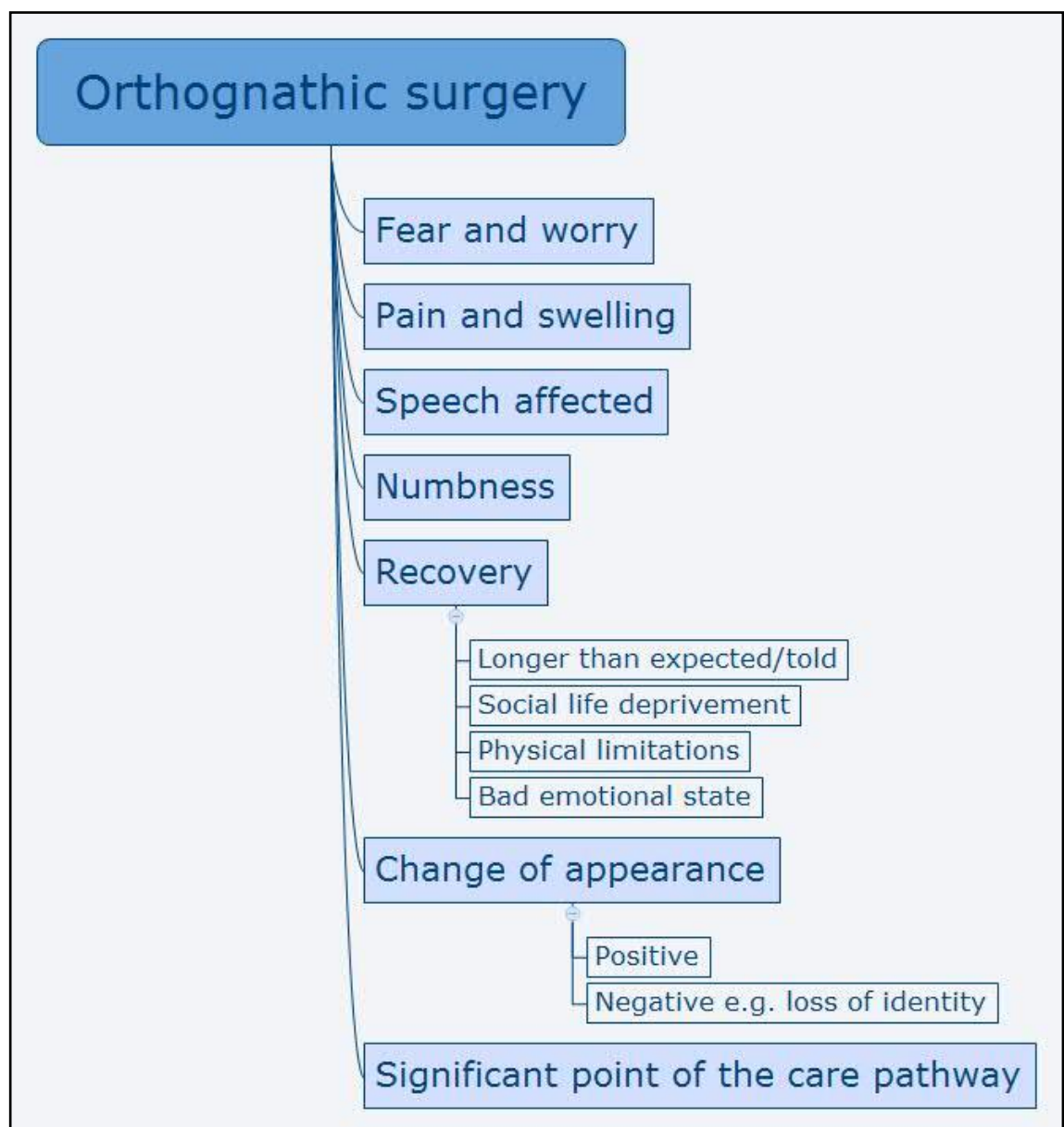
Figure 20: Orthognathic treatment

Participants highlighted a number of issues about their orthognathic surgery (Figure 20). Perhaps unsurprisingly prior to the surgery the participants were worried, not just about the process itself, but also about whether it would be successful and what would happen if it went wrong:

> '' its happening this isn't great but that just like a fear, like a fear which harms, you know '' **INT#1.M**

> '' And I was very nervous about going up on the train, I was thinking, "What if I wake up in the morning and I find that I can't speak, and I'll never be able to speak again," something like that. There were always fears bubbling from my subconscious that I was thinking, "Oh, what if this happens or what if that…"**INT#17.M**

Although the primary sensation after jaw surgery is numbness, as the data will show, pain can be experienced after orthognathic surgery. This may be as a result of the osteotomy cuts or the pain can be experienced in other parts of the body due to the need for bone grafts from the hip bone. This can limit movement and mobile, and widens the impact of the surgery substantially:

> '' and after the operation it was painful I couldn't even like walking as well but I mean that's part of the operation''**INT#1.M**

The pain was exacerbated in one case by the fact that the intensity of the pain was unexpected:

> '' We didn't know how, because they were saying it wouldn't be that painful, but it was the worst pain I've ever had in my whole life, and I was on so many tablets and maybe it was our fault for not asking more details but we definitely said if we knew, we would have gone back and made sure we knew everything, because being inside for over a month, your emotions, and it's not just being in pain, it's your emotions as well, so I had the psychologist from the cleft team had to come to my house and talk to me because I was really upset, so but it was quite nice that they came out to my house.'**INT#9.F**

Recovery from orthognathic surgery can also be problematic. The experience in itself is unpleasant and the length of recovery seemed, it was explained in cases to be significantly longer than originally suggested.

The extended recovery period post surgery was difficult not just because of the duration, but also the discomfort and wider physical effects of the recovery process.

> '' It was, I was housebound for a good two months, I couldn't do anything physical because they took a bone graft out also and I was able to walk, able to walk upstairs and do physical activities, but I was exhausted much, much quicker then I would be if I wasn't

173

*outside the surgery so I was tired, I was, sometimes, the physical activities that I do make me in pain''**INT#23.M**

*'' In terms of the recovery period, the first month was horrible. I was bedridden a lot of the time and doped up on a lot of drugs – morphine primarily – sleeping for long periods of the day, in a lot of discomfort, couldn't really get out of bed, if I got out of bed I'd run the risk of potentially falling over. It would hurt to speak. I would avoid funny films because I found that if I laughed it would actually hurt, which was really rubbish. (Laughs). And my entire mouth was so swollen that I couldn't actually move it, no matter how hard I tried. And over the course of about three weeks I found that I could speak again. And about two weeks after the surgery I ventured out of my house for the first time and went into a pub, and I was drinking, I think, Sprite with a straw because I couldn't open my mouth still to pour the liquid into my throat, that sort of thing.''**INT#17.M**

*'' I lost a lot of weight obviously because I wasn't eating properly.''**INT#26.M**

Negative experiences which were related to the recovery period included being unable to move and the need to stay in bed, pain and discomfort, being unable to speak, unable to laugh, the taking of medicines, eating only liquids and isolation. Committing to a more liquid diet for a relatively long time was seen as unpleasant, and in some cases resulted in weight loss. It may have been several months before a person could eat solid food post surgery and this had a big impact on daily life.

Again, context was deemed very important and the negative impact of the surgery and recovery period was exacerbated where there was a clash with other aspects of life. For one participant the timing of the surgery was less than ideal:

*'' you know say right now still learning I'm still developing so right now it will be a bad time of having an operation like I've learned from my last time...the last time I recently started the job I had the operation I was still recovering when I went back...they didn't really know me that well so it was all a bit of nightmare really..it was a bit of train crash that whole experience so I want to learn from that and so yeah and sort of adjust myself a little bit so ill have a good time for myself you know because at the end of the day work is work I don't work to live you know..no... I don't live to work..I work to live ..so I want to do the operation at a good time for myself but not having a bad effect for my job...so I take time off ..I come back and start working again''**INT#1.M**

Also, returning to work too soon made the recovery period more stressful:

*'' I went back to work too early...I was a bit..You know I can barely talk... you know and you work in advertising so you need to talk''**INT#1.M**

Speech was one of the areas that could be affected by the orthognathic surgery and the immediate effects of the surgery included the inability to speak because of the pain, numbness and swelling. Also, the numbness may remain long after the surgeries.

> '' I think I can't really feel the inside of my lip I think but I can feel the outside..so I think I'll never feel the inside of my lip again..but I think it's alright yeah"**INT#1.M**

As recovery progressed changes in speech quality were noted and participants talked about the difficulty they experienced adapting the tongue to the new position of the teeth and the jaw. This process of adaptation took some time.

In spite of the pain, extended recovery period and interference with everyday life, the surgery was seen to have a hugely significant effect on the appearance of the face and mouth. This was summed up by one participant in the sense of excitement that surrounded this stage of their treatment:

> '' Oh yeah, of course, just the teeth, yeah. I knew that. (Laughs). So that was kind of treated as a bit of a big day moment. There was a lot of … it was hyped up quite a lot by both people from the NHS but also people that I knew – relatives and close friends, that sort of thing – as being like a sort of looming day that everyone was very excited about for me. And I was quite looking forward to it as well. But then, in another sense, it was a very kind of scary, looming day and I'm thinking, "Okay, alright, you're going to have the surgery done and then it's going to take about four months to recover. So you're basically going to be out for most of the spring/summer time, which is going to be a bit weird. And you're going to have to take things very carefully."**INT#17.M**

The sense of ambivalence and the balance between positive and negative feelings was also summed up by the respondent quoted above in relation to pain. She had experienced significant pain and psychological distress post surgery and thought aloud as she answered what she had experienced:

> "It wasn't worth, I don't know if it was worth, no it was worth it from that outcome that I got in the end, because I love how I look now but during the time of it, it wasn't worth it at all, and it was a lot of my mum and dad had to watch me go through it and they didn't exactly like it. '**INT#9.F**'

This sense of ambivalence may stems from the anticipation and physical reality of the surgery as well as the significant visual result.

Interestingly, whilst the impact was enormous, responses were tempered by the sheer visual difference it made to the face. As one participant explained:

" I think we did, benefits are that you just, you don't have to wonder what, you don't have the same jaw as you did before because you are, for me my bottom jaw was in front of my top jaw which is why I had the whole jaw surgery. So not having that is one less part of the identity of not having a cleft palate any more. Even though that was not the major thing, it's more to do with the exterior and what you can see from like the nose being swift and the lips and the scar and that is more what it is. But moving the jaw back does also help the lips as well because it moves back the lip a little bit. But they are like the two benefits, you don't have that anymore and move the lip a little bit. Other than there aren't really, it's not benefits, it's not really a positive thing, it is just something that you feel like you've got to do because I've had this for like 20 years going up and down to different hospitals. It's just something that you do, as part of natural progression."INT#28.M

Whilst the surgery was acknowledged as having moved the jaw, and lips into a more usual/normal position this was not seen as a positive outcome in and of itself. There is the suggestion that this change removed a visible component of the cleft face and that there was an assumption that this was a good thing. This raised an interesting question about ideas or normality and where they fitted within the pathway. It also echoed questions raised earlier in the analysis about choice within the pathway and the extent to which future treatment was seen as inevitable and unavoidable rather than an active choice.

''it's not really a positive thing, it is just something that you feel like you've got to do"INT#28.M

'' amm...I would say that… the braces...and then the operations that happened..and even when I was young I knew that I was going to have this operation when I was 18 ...I always had that..they said you will have treatment and when you are 18 you will have an operation...and you'll be fine. you know...so obviously I was 22 or something when I actually had it done ...but I always knew that it's going to happen...''INT#1.M

The final point to note here is the importance of the face in relation to identity and the impact of radically changing the look of the face on the way that the individual sees themselves as well as how they are seen by others.

This section together with the orthodontic treatment effects provided an insight into the patient experiences. It also showed that participants struggled with the effects of treatment in their day to day life. The next section will show some treatment aspects that some participants felt had a higher importance to them.

Treatment Significance

When asked about which parts of their treatment they felt were more important one participant highlighted the braces and the orthognathic surgery alongside the very early operation that happened when he was a baby as the major parts of their CLP care. One explanation for this was that these were the parts of treatment that had an immediate impact on look and function. This may reflect the importance of aesthetics and associates with findings in the previous chapter about the impact of aesthetics on self-esteem and confidence as well as on social relations and experiences of stigma. Orthodontic and orthognathic surgery treatment could be viewed as part of the overall treatment but as two distinct stages within CLP care.

> *'' amm...I would say that… the braces...and then the operations that happened..and even when I was young I knew that I was going to have this operation when I was 18 ...I always had that..they said you will have treatment and when you are 18 you will have an operation...and you'll be fine. you know...so obviously I was 22 or something when I actually had it done ...but I always knew that it's going to happen...but I didn't quite know what it was until they said ok this is what's going to happen...so yes I think apparently the braces on it actually made my teeth straight...and then having the operation which made my bite better...those the 2 big things I would say...ohh yeah I mean obviously like the old baby and sorting out the cleft and palate probably be the most important thing and then after that that process of braces and operation...so I would say three big things..''***INT#1.M*

Nasal surgery was also given importance beside the orthodontic treatment. This may also reflect the importance of correcting the look.

> *'' My braces they were really important because without them I wouldn't have my straight teeth and I wouldn't have my teeth in the positions they are in. And my most recent operation is probably the most important because it has re-aligned my nose and got rid of all the scar tissue in my mouth which means now I sound a little bit less funny when I speak. And my nose is on the outside its one of the first things people see and you are not meant to judge someone on their looks but you do don't you. And now because my nose is not like this, it's like this. So that was important to me to look more centralised definitely I would say they were the two most important bits of it''***INT#8.F*

The initial surgery in the first year of a child's life may be perceived as important because these surgical procedures may correct most of the problem of the CLP.

> *'' When I had the operation when I was a baby''***INT#25.F*

A different view was that orthodontic treatment was perceived simply as another part of the process, and was seen within the pathway as an overall journey. A participant stated

that orthodontic treatment was part of the CLP care and she didn't feel that it was separate.

> '' so it all just tying into one really the orthodontic work and cleft stuff in the past I don't really consider it separate so..''**INT#5.M**

Understanding the effects of such major treatments will certainly help clinicians to improve their approach of care. By updating patient information or showing more effort to minimise the negative effects of treatment to participants. This may help participant to have a better experience of their treatment. Also, clinicians should understand the importance that participants may place on treatments that change the look and make them ''normal''.

In this section, the data presented the experience of the definitive orthodontic treatment including the orthognathic treatment that may be involved. The data showed how these aspects of treatment could be problematic and needed participants to manage with these treatments. This connects with the long term illness literature theme 'managing medical regimen' presented in the literature review. CLP participants understandably go through a long and complex treatment that may be exhausting and daunting. The next section of the findings will show some aspects on how participants cope with their treatment.

6.1.3.6 Coping

People living with cleft have to manage their feelings about having a condition and the meaning of coping is anything that helped to lessen the effects or impact of living with CLP and its treatment. Living with CLP can be problematic. Also, CLP participants go through a long and complex treatment. In this section of the findings, factors that helped participants to cope with living with CLP or to cope with treatment will be presented (Figure 21).

These factors can be grouped to include:

- Maintaining a positive outlook/perspective

- Comparison with those worse off

- Involvement into CLP social network or making links with other people with cleft

- Professional and lay support networks

- The quest for a 'good' outcome and 'normailty'
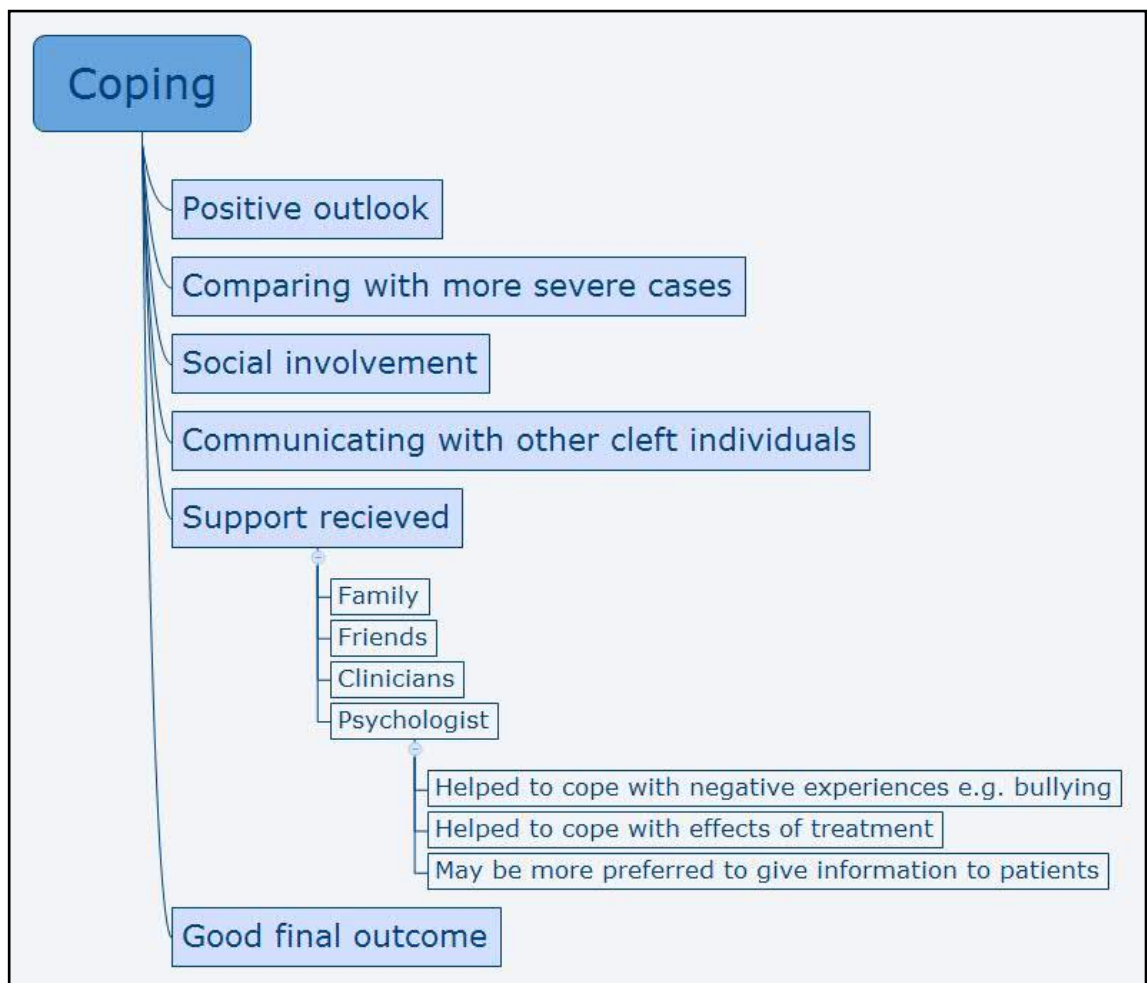
Figure 21: Coping with treatment

How patients perceive their CLP and their treatment may influence coping ability. Maintaining a positive outlook and seeing treatment as a tool to make them better or 'normal' may be more helpful than looking at the treatment journey as a burden. The development of these positive beliefs depend upon many factors from the individual background which are beyond the scope of this thesis. However, the data here helped to identify that coping ability may be related to how the individual was brought up which clearly influences the way of thinking.

> *'' I mean I know I'm aware obviously of what I've got but I didn't need to research it for ages and find out other people's experiences, it doesn't really matter to me, I think my mum brought me up to be well that's it you just deal with it you know you don't need to go into things too deeply"***INT#12.F***

As discussed previously, a change of environment and ageing may have some influence on how CLP participants perceive CLP and its care. Also, psychological support may influence the way the individual perceives things in general.

> *'' And the reason why there was such a large chunk that came from me was because the psychological treatment basically taught me that, "We can only give you advice and support you, which we're happy to do. But if you want to make these changes, they have to come from you. Because, fundamentally, it's your internal psyche that you need to redesign and reassess through our advice, in order to get to a better place." So I would say that was about 40% input."***INT#17.M***

Comparing themselves to other CLP individuals with a more severe form of CLP or other condition was used as a way to cope and to appreciate that their CLP maybe less severe than others. This can work both ways as it can be more of a negative influence if others were individuals with milder cases.

> *'' Thank you to the NHS, yeah it's been a pretty painless experience really. Its only a minor thing really a cleft palate compared to proper disabilities and diseases, but they treat it with respect and fix it yeah with minimal fuss and I cant really complain, its been great, yeah."***INT#11.M***

> *'' Yeah...like I said I can't think of anything...nothing to me would come to mind as a negative...ieah its not...obviously I can think oh maybe there is things that I haven't had but I wouldn't be aware that the fact that I have not had them I mean..I think the fact that the severity of my cleft lip I think probably as not as bad as other people...amm...that's...I've not had to really experience it that much a would say it's been a casual thing throughout my life really I know I've got it so it's just been casual so the treatment I've had I don't think I've missed out on anything ah hi think it's all been positive and...top care really"***INT#5.M***

Furthermore, a good social life and friendship network may also provide some help to lessen the focus on the CLP condition.

> *'' Socially at school I wouldn't say by the time I got to, I mean it wasn't really affected like I say amongst my friends you know I had lots of friends, I was very popular at school, you used to get the odd person shouting things at you, you know like I say name calling but I was never bullied or teased in any really bad manner that it would affect me and normally if I was with my friends and someone, some lad two years higher in school would say a horrible remark my friends would just have a go at them anyway, it never affected me socially. I mean obviously I was self-conscious and meeting new people I was always a bit wary, I would never push myself forward in a crowd if I was meeting new people but amongst my group of friends it never affected me at all.''INT#12.F*

Building on the same point, being involved in social activities may be helpful. For example, a participant found being involved in playing music was helpful to cope.

> *'I got really involved in music. I was in the choir. I was in most of the sports teams. I just was busy all the time.''INT#10.F*

To meet and speak to other individuals with CLP was found to be helpful. This was helpful because patients could see that they were not the only ones and could share experiences of care. Also sharing personal experiences of treatment helped as they could generate a feeling of a greater degree of understanding and they could relate themselves to those with similar experiences. It was also reported that talking to other CLP patients who went for a specific procedure could be helpful for the patient who would be going for the same procedure.

Some CLP services arrange a day for CLP patients to meet, get to know each other and discuss their experiences. That has been reported as a very positive experience for the patient and indeed they requested to have this experience more often within the duration of their care.

> *'' I would like to meet people my age a little bit more often than I did, I only met once with other people from the area with a cleft and that was when I was about 13 or 14 I met with them. I would like to be able to … sort of not every year but every other year meet with other teenagers with a cleft as well because I think that would be good to keep in contact with them. Because I thought that was a really good day and everybody else agreed that it was really good, it was just a shame that there was never another one really……….*
>
> *It helped because I was sitting in a room where I wasn't the only one with a cleft for once and everybody could share their problems, their backgrounds it was a good way to make new friends. ………And I met people that had double clefts as well because obviously I*

have only got it one side they had it both and I wouldn't say I felt sorry for them but I felt lucky that I only had half a cleft really. Yeah it was a good day and I would like to do it again. ''**INT#8.F**

Some participants had experienced this type of social support but others clearly had not but would have liked to have that kind of support.

*'' I know as soon as I leave I'll think of something. I don't, do you… like take the patients of cleft palates who are younger, like probably 13/14 just coming to teens and sit them all in a room? And so understand each other's stories, I think that would be very positive …… if you could sit all your patients down at the same age who are going through the similar thing and let them just discuss what they feel, instead of discussing now one on one. It will give them greater understanding of their journey and others' journeys and being able to compare some of the questions that you asked me. I had no comparison to another person, they've gone through their things. I think they should sit them in a room like this and just ask them all the same questions. As if it's like an AA meeting in a way. And that will give them greater strength that they are not the only ones going through it, one, two, greater understanding of their own personal journey and knowing how to relate it to someone else, is it that bad, is it worse than theirs, is it good, how they've gone about this situation, can I apply that to mine so on and so on. So I think a lot of people in, who have cleft palates are on a lonesome journey in a way. It is very much on your own to deal with, it's your own burden but it is not, I don't think you should look at it as a burden but it is because other people make it a burden. So it is very much on your own because it is not, you don't really know much about it and still don't' know why people are born with cleft palates. If you were to allow them to sit in a room and actually discuss for the, what they are about to go through I think it'd give them greater strength to overcome the fact of how painful jaw surgery is and things like that, being more aware, I think that's a…''***INT#28.M***

Another major support that helped participants to cope was the support that came from the family. This may have come from parents and/or siblings. Parents do their best to help their affected child to avoid any negative experiences. Participants realise their parents supported them even if they do not remember it.

*'' If I didn't come in, then the treatment would have stopped, and I didn't want it to stop because I'd only had half the work done, so I wanted to finish this off, that's what motivated me, to keep coming, plus I have family members, kick me out of the house to get me into the dentist.''***INT#23.M***

'' I've coped okay from everything, everything has gone like good and bad really. I mean I've got lots of support behind me like family and friends that have helped me stay strong and operations and surgery and treatment it all gets a bit upsetting but I've had to live

*with it. But it makes me happy knowing that I've got people that are there for me to make sure that I'm okay.''**INT#27.F**

*'' Yeah, well you have your breathing problems and your ears pop more than the average person and you wear a brace for a number of years and you have a couple of surgeries but its, yeah I think it might have been worse for my mum having to look after a child with a cleft and going through surgeries than myself really.''**INT#7.F**

In addition, if there were other family members with CLP, this was helpful and could provide some support in relation to understanding of the experience and providing the correct levels of information.

*"Because my mum had it, it was just something, because my mum was the first one, there's no other records in the family of anyone else having a cleft lip and palate and because my mum had it, I just followed in her footsteps in a way, because she was telling me her experiences through having it, so I never really thought of looking it up."**INT#13.F**

Friends were also found to be a source of support. Sometimes they may have been the only support available. For example one patient, because she lived alone in London and all of the care and support around surgery time that helped her to cope with treatment was provided by her friends.

*'' I guess because in UK by myself and not my family and I was reliant on my friends to bring me here, pick me up, look after me for the first few days and I had really, really good friends, it was never really an issue or a worry that something's going to go wrong there. I think I wasn't, I think I was more tired after the surgery than I thought I will be because I'm usually quite energetic and I thought it would be fine but I must have lost about ten kilos in the first week after the surgery.''**INT#7.F**

Also, support that was provided by the clinicians and staff was found to be helpful and made the patient reassured.

*'' It's been very, very positive, definitely. I mean I haven't the comparison, what I experienced before, it has been pretty much plain sailing. My surgeon was [name], well teaching the fellow Surgeon and then also been quiet, very attentive, very much involved and inform me about anything and explain during my visits what is the issue, how they can address it, giving me options of treatment which was never the case back home and that made me feel much more relaxed, and also the additional services of the Psychologist which I didn't actually need but the knowledge, that it was there, available for me was fantastic and also my restorative Dentist, [name] who I think isn't part of cleft, really both are related …''**INT#11.M**

The support provided by the psychologist within the CLP care was substantial to some participants to help them to cope.

*'' On a short-term level, I'm not ashamed to admit it, on a short-term level I think it probably stopped me from killing myself at the time. Because I was really, really, really miserable at that point. So it was really helpful.''***INT#17.M**

This support received from psychologists was provided in many different forms. For example, their support that was provided to cope with negative experiences related to the CLP such as bullying.

*'' At that point my parents referred me, or rather the NHS referred me to a psychiatrist, who actually operated at this particular hospital a lot of the time. So I had psychiatric help, which was very important at the time because I was quite shaken and, on the wire, shall we say, a familiar phrase, and not really sure about who I was, why I was being treated this way and feeling very different and very isolated. And there was a lot of mixed feelings. Like, a lot of the time I kind of thought to myself, "Oh, this is something I can't control but I'm being treated by both adults and children as if it's different, and therefore that I should be treated different. I'm really no different from anybody else. What's going on here? Why am I being treated that way?" It wasn't exactly the most pleasant period. ''***INT#17.M**

Surprisingly, although there is some confusion of the role of the psychologist; it was felt beneficial to receive information related to treatment from the psychologist as a support to information received from the clinicians.

*'' I only ever go the top brace, never the bottom. They gave me a psychiatrist just to check … I didn't get many meetings with her. I think I would have liked a few more, just to discuss the situation. I think that would have been a bit more beneficial to me having a psychiatrist discuss what's going on and how I feel about that. That's all I remember really, just the braces.''***INT#2.F**

In one case, external support was sought to cope with an unpleasant time at school.

*'' I went to see a counsellor at one stage. That was in year 11. But that wasn't necessarily ... maybe it was. I was very unhappy at the time. The girls I was with or the friends that I had, I didn't ... I don't know if it was bullying, I'm not sure. But I had just had very, very little confidence. And so my house mistress suggested I go and talk to someone about it. So I did. I went to a counsellor who came into the school once a week and I went to see her I don't know how long for, I can't remember. And she was just someone completely neutral that I could just talk to and she gave me advice on how to present yourself and body language and just talk through what I'd done that week and little things about not making everything into such a big deal. And I guess, I think it did help. So yeah I saw her for a bit but I don't know how long for.''*INT#10.F

Also, the desire to achieve the final outcome may have played an important role to keep participants motivated to come to treatment appointments. To be specific, outcomes related to the look and function were identified as relevant. This was in line with the need to become 'normal'.

> '' The motivation was just because I knew the outcome would improve my speech, my teeth so that was the reason why I came down every time they wanted me because I just realised that the outcome would improve me, improve my teeth and improve my speech sort of thing.''**INT#22.M**

> '' So that it improves the way I look in my face obviously and obviously I want it to change even if it's only a little bit. But I take all surgery that is offered to me, to make myself look better and feel better about myself.''**INT#27.F**

> '' My motivation is the outcome really if I'm going to go in and they're going to do something that will have like an obvious outcome, like coming here for my retainers there's the outcome of having my retainers and having them fit, so I think that's the motivation coming in and also knowing that everything is okay that reassurance, that okay I'm fine I don't have to go through anything any other treatment, you know to know that everything is okay, everything should be how it is and there's no complication, no issue I think that's reassuring and a motivation to come in and know what's happening with me.''**INT#20.F**

The data show that CLP patients, without doubt, need support. This was because of what CLP could do to their living experience. The support could be from the family, friends or the CLP service. This emphasizes the importance of support CLP care staff give and especially the input of the psychologist.

As the data showed, the idea of achieving a good final result may have been a reason that helped a participant to cope with complex CLP care. This will place an importance of how participants think about aspects that come after they completed their treatment such as the outcomes and the benefits of treatment. This will be presented in the next section of the findings (6.1.4).

6.1.4 After treatment

In the previous parts of the findings, the data presented issues related to different aspects of before and within treatment. In this part of the findings, the data presents the 'after treatment' related concepts including the benefits of CLP care and the satisfaction as perceived by participants.

This last section presents the data related to:

- Benefits of treatment
- Satisfaction with care

Each sub-theme is presented in turn and illustrated with quotes from the interviews.

6.1.4.1 Benefits of treatment

All participants had found the CLP care to be beneficial to different degrees. Experiences ranged from just improving the look of the teeth to positive complete changes of life.
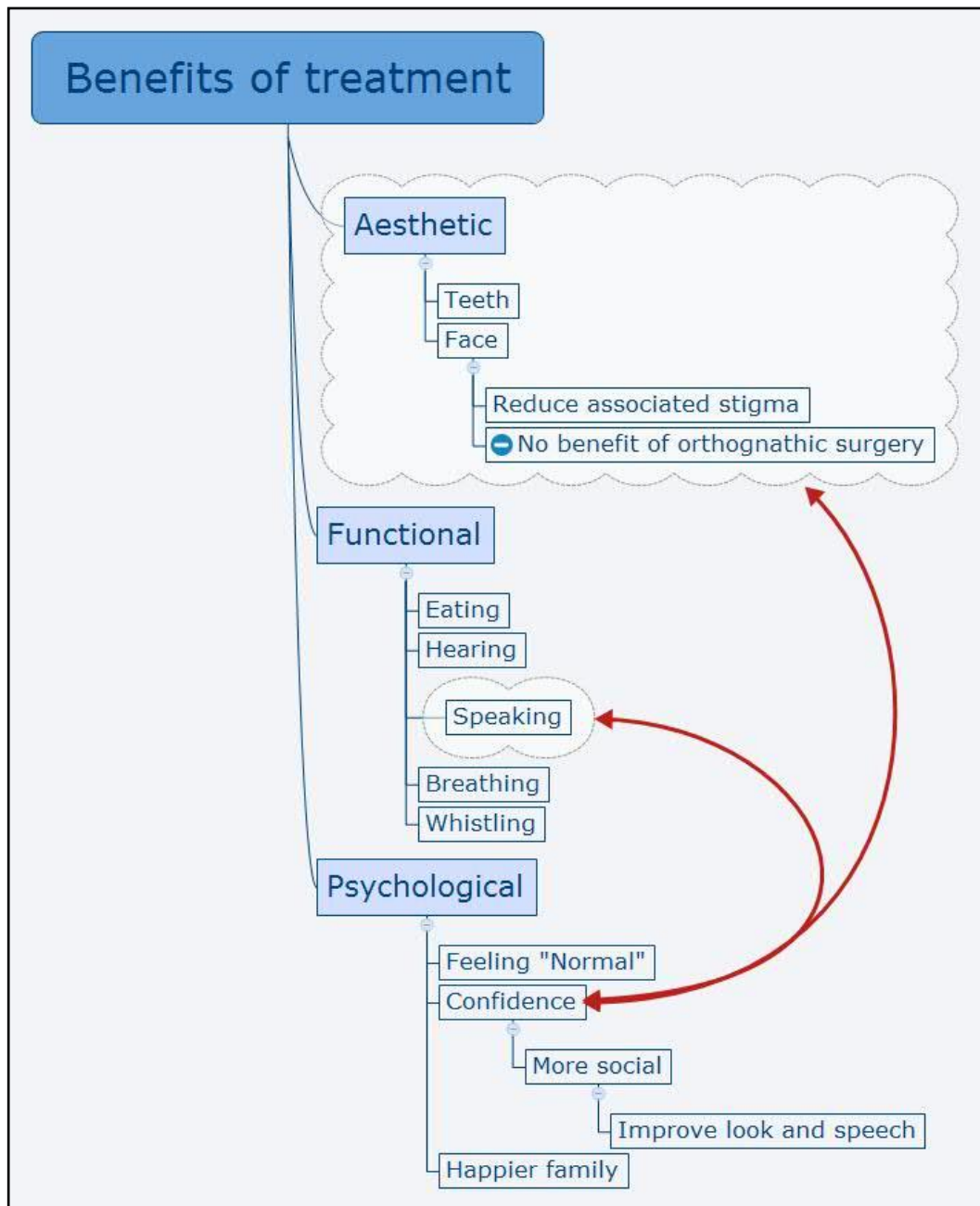
Figure 22: Benefits of treatment

It is important to note that the benefits of treatment were not necessarily immediate. CLP patient were on a long care pathway that required complex longer procedures which in turn sequentially built up on one another in order to achieve a final end result. Patients may or may not appreciate this at the start of treatment.

Treatments that were placed near the end of the CLP care were probably when patients would recognise the benefits and could see them more clearly. In particular, the benefit of having optimum aesthetics might have been considered as the major aspect.

Benefits as realized by participants could be divided into: aesthetic, functional and psychological (Figure 22). Aesthetic benefits related to the look of the teeth or the face. Functional benefits included eating, hearing, speaking and breathing. Psychological benefits were as a consequence of improving aesthetics or in function. These improvements may have helped the patient to experience less stigma or possibly to eradicate it totally. This may be an addition to the feeling that they were becoming 'normal' and not feeling 'odd' anymore.

Improvement in the appearance of either the teeth or the face was one of the benefits that CLP participant received from treatment. This was mainly as a result of the patient proceeding with orthodontic treatment with or without orthognathic surgery. In addition, improvement of mastication and eating was also identified as another benefit.

> *" Well my teeth are a lot straighter than they were and now with the jaw surgery I can, my bite is far better, before I had the surgery I couldn't, say I was eating a sandwich that had like sliced tomato in it I couldn't actually bite the tomato because my teeth didn't meet so I had a gap so I couldn't actually bite, my bite didn't meet. So I can now bite properly and I think it has improved my profile so I think before my face was even flatter and so it's brought the top part of my face forward a bit."INT#12F*

Gaining more confidence was also reported as a benefit together with smiling more often which helped the participant to be more social. This may also have been a consequence of having improved aesthetics.

> *"Like I said I feel amazing, to be offered this surgery, it's just amazing to make me feel good about myself, obviously they are doing it for me. I was born with it so they are offering me the treatment and it makes me feel happy, glad. As I've got older and had more surgery I am feeling a little bit more confident about myself like I go out there and meet new friends. I don't care what anybody has to say about my appearance and my looks. It's their opinion, everybody is entitled to their own opinion. But at the end of the day, I don't care, I've got a life to lead, it doesn't bother me anymore. It did, but it doesn't."INT#27.F*

> *" It makes me feel confident that I can smile normally and yeah that's about it."INT#27.F*

Speech improvement was also reported as a benefit. Speech therapy classes helped some patients to improve their speech. This may have increased self-confidence and helped the patient to be more socially active; CLP speech difficulties may have acted as a barrier.

> *" It's straightened my teeth dramatically, it's made me feel a lot more confident about my teeth I think, I think I've noticed a small amount of speech improvement you know prior to*

*having them on, yes more confident, small amount of speech stuff and yes definitely, I think the confidence thing was quite a big thing because my teeth weren't great before having the braces on, the braces I personally don't think they help really in confidence but having them off you know and with nice teeth yes it really sends your confidence sky rocketing."***INT#24.F**

*" I'm half way there now, and from what I was like a few years ago, it's really made a huge impact, from what I said earlier, remember how I said that no one would talk to me because of my speech, I think that's changed, I still have a lisp but people are still understanding me so that's a good thing."***INT#21.F**

Improvement in hearing and breathing were also reported as benefits.

*" My hearing was the worst bit because, obviously, with the internal movement of the mouth it disrupts the ear tubes to the rest of one's face. So I have a bit of a hard time hearing. I'm normally leaning in like this and asking people to repeat themselves. Like earlier, when you met me down at reception, I asked you to repeat something straightaway because I couldn't hear you. Things like that. I had hearing classes for that, I had grommets in my ears, God knows what else. And that's kind of got to about as good as it gets and I've sort of come to accept that my hearing's fine, I just need to pay attention more often. (Laughs)."***INT#17.M**

*" It never affected my sight, never ever. And in terms of my smell, for about 20 years I couldn't breathe through one of my nostrils. And when I had the operation on my nose, one of the surgeons, [Name], he did the work for my nose and I found that I was able to breathe out of both my nostrils for the first time ever. I'm not entirely sure quite how he managed to do that – it remains a mystery to me, it was never explained. But I found myself being able to breathe out of both my nostrils for the first time. And I was just like, "This is really weird, what is going on here?" (Laughs). Things like that. But I don't breathe through my nostrils, I always breathe completely through my mouth because, having spent so long with only one working nostril, you have a hard time breathing. So I normally breathe through my mouth."***INT#17.M**

Improvement of the aesthetics may have reduced the stigma associated with CLP. Consequently, the affected individual may have experienced an increase in confidence levels. The end result would positively reflect on self-confidence and perceptions about CLP and treatment. This appeared in the data specifically after orthognathic surgery as this was described as a life changing event to some patients.

*" Well people changed around me, so as I got older, people, things got better, the treatment got better, so it improved my looks, so therefore, people changed around me, so therefore, my self esteem got higher because people were changing their views around me, so things have got better in the long run."***INT#21.F**

" And then around about nineteen, I had a pretty serious operation on my jaws, because my bite was all out of proportion. So my upper jaw was brought forward, so, to align it to look like that. And that was kind of like, there's a before and after Mark, in terms of how I felt about the situation. After that operation, it's fair comment to say my attitude about things completely changed. I felt far more confident about things, far more comfortable. People were no longer looking in my direction and thinking, "That guy's jaw's a bit weird, has somebody punched him in the mouth, say?" Things like that." **INT#17.M**

Interestingly, one participant found that he will be able to whistle and he was happy about it.

" About a year ago, I found I could whistle again and I'm thinking, "Oh, I haven't been able to do that for about three years. What on earth?" So it feels like there are still things going on." **INT#17.M**

It was also stated that becoming 'normal' was a benefit. A participant`s reason for treatment to become 'normal' was finally achieved and considered as a benefit.

" Because I think I look better and I look more normal, and I'd like to look normal in my own eyes. " **INT#26.M**

Other benefits were actually related to people's perceptions and behaviours around the affected individual. Having a good end result shown outwards to their affected family members would reflect positively on them and could make them happier.

" the benefits have been gigantic, absolutely gigantic. Not just for me as an individual, but also for the people that I have grown up with, the people that I've lived with, the people who've known me from about the age of ten, right up until present day, who can recall various different stages in my life where it was fanatically different. Even my parents, my father's often saying to me, "You know, I'm very proud of you." And I often can't figure out why and I have to ask him, "Why are you proud of me? I'm no different from most kids my age. I have a degree, I've done a bit of travelling, I have a job. What's there to be proud of, Dad?" And he's just like, "Well, you've gone through pretty serious surgery and you never really complained about it. Sure, you'd be quite miserable sometimes but that was understandable. You never really complained about it. You just got on with it like, well, and you sort of exceeded our expectations of how you were going to handle it." **INT#17.M**

One final point to note was that not everyone should be happy about every aspects of care. One participant reported that there were minimal benefits to undergoing orthognathic surgery. This was mainly because he wasn't happy with the results and his new look.

*" I think we did, benefits are that you just, you don't have to wonder what, you don't have the same jaw as you did before because you are, for me my bottom jaw was in front of my top jaw which is why I had the whole jaw surgery. So not having that is one less part of the identity of not having a cleft palate any more. Even though that was not the major thing, it's more to do with the exterior and what you can see from like the nose being swift and the lips and the scar and that is more what it is. But moving the jaw back does also help the lips as well because it moves back the lip a little bit. But they are like the two benefits, you don't have that anymore and move the lip a little bit. Other than there aren't really, it's not benefits, it's not really a positive thing, it is just something that you feel like you've got to do because I've had this for like 20 years going up and down to different hospitals. It's just something that you do, as part of natural progression."***INT#28.M***

The data show that patients benefit from treatment in different forms: aesthetic, functional and psychological. The results in this section come strongly in line with the reasons patients' state when seeking treatment and the experience of living with CLP. Treatment could correct functional problems and correct patients' deformity and make them more 'normal'. As a result, they may suffer less stigma and gain more confidence to speak, smile and become more social and able to live like everyone else. This, as presented in the data, showed how treatment could change CLP patients' lives.

Understanding the benefits may provide a reasonable explanation for understanding participants' satisfaction of their care. However, the next section will provide several reasons which build on previous findings that may explain their satisfaction.

6.1.4.2 Satisfaction with care

Participants gave positive responses when they were asked if they were happy and satisfied with the overall CLP care they received.
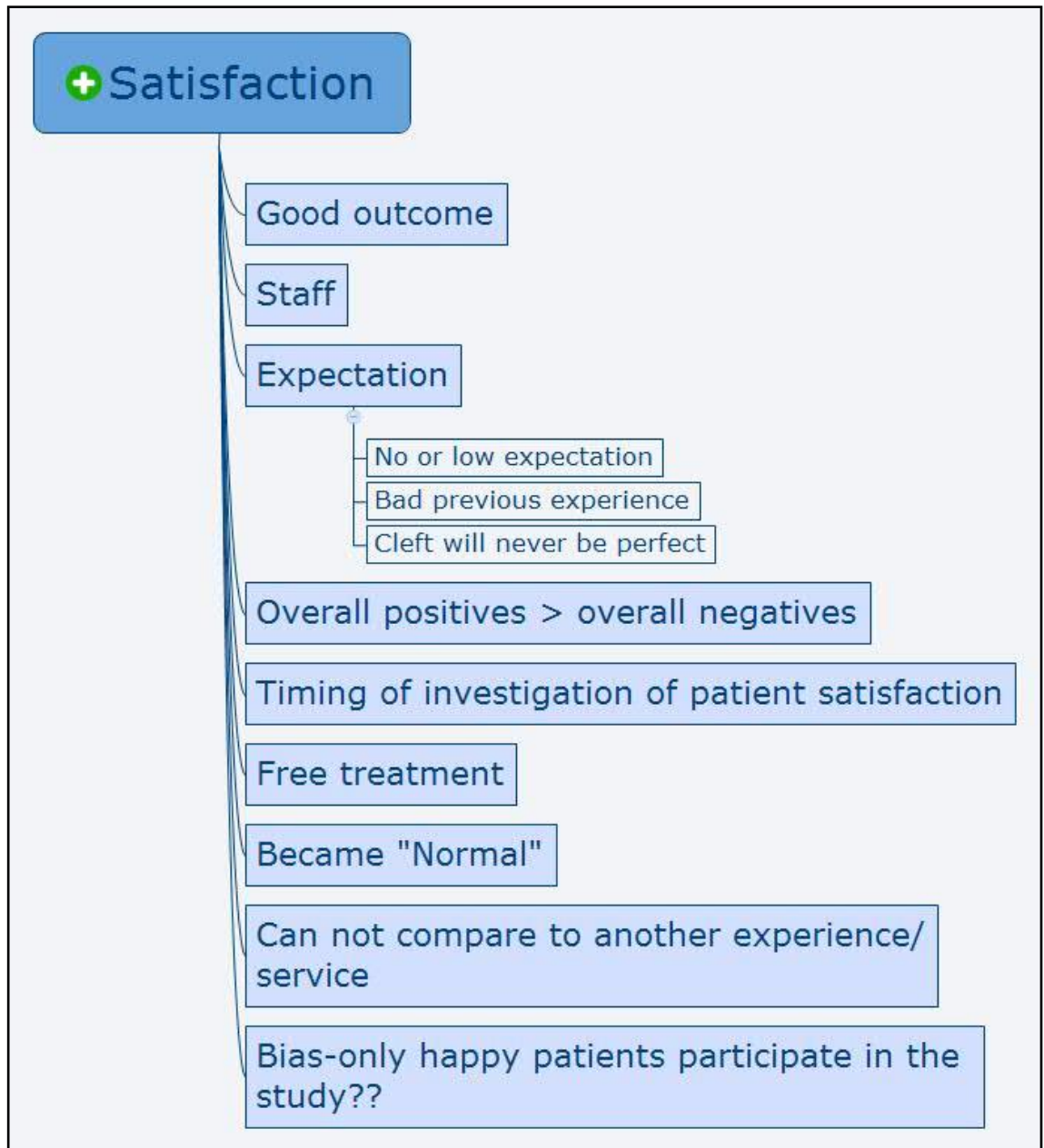

Figure 23: Satisfaction with care

Despite some participants having experienced some difficulties and distress which related to different aspects of CLP care. The data suggest that participants may have been isolating these negative experiences when they recalled their overall satisfaction with the care. Their responses about their final satisfaction did not coincide with some

negative experiences that were related to the care. This suggesting that the overall good outweighed the bad.

Based on the data, there are many factors that may have provided insight to explain why all patients studied were satisfied (Figure 23). It is also important to note that these possible influencers or factors will be listed without significant levels of data as all of these points have been previously presented but scattered throughout the data. These are related to good outcome and/or staff, having low expectations, realising the overall positives, the timing of interviewing, no cost of treatment, becoming 'normal', no other service to compare with and the possibility that only satisfied patients will actually accept to take part in this study.

Firstly, as the outcome of the treatment was predominantly good and pleasing, the patient could see the benefit which made them happy about it. The patient could now enjoy the benefits of the improved aesthetics and function.

> '' … yeah, so I felt far more satisfied, far more happy as well. Knowing that things were being corrected and things were finally being taken care of was always a good confidence boost, always led to a positive feeling. Because I always knew that I was getting better all the time.''**INT#17.M**

> '' . Everything that needed to be done is done. Let's draw a line; this is corrected; the nose is corrected; the speech is fine; the hearing is about as good as it's ever going to be. I don't think there's anything more we need to worry about.''**INT#17.M**

> '' the outcome and the orthodontists are all really good, because I'm with [name], I'm under [name] care and she's so good, because I was with one of the other orthodontists, and I can't remember his name, but then I moved to [name] and she's just as good and the cleft centre is really good in a way that they're not slow, they're all, I have monthly updates, not monthly updates, my appointments will be on time, so but yeah, the experience is good.''**INT#21.F**

Secondly, the staff within the care were seen as good. Therefore, patients appreciated what the staff did for them and they were pleased. The final result was that they could report that they were satisfied with their overall care.

> '' INT: so talking about the care pathway so far overall..are you happy going to the care pathway?
>
> RES: yeah 100% to be honest I think the people I've met on the way I think they all really care they are all very good you know you don't go to school like 6 years [...] you doing it like you know you really care about what you are doing..so I..it shows..because you are dealing with people all the time you really do take the time to know them because I've

*been coming to here since I was a baby ..I was born here in this hospital so I've been coming here obviously..even today so you know.."**INT#1.M***

*'' no..the entire thing that I've got from the experience was just it was all very good…I was always very happy and very informed ..everything was very clear everyone is very helpful ..so I never had any problems what so ever throughout the treatment.."**INT#6.M***

Thirdly, patient expectation could play a significant role in determining overall patient satisfaction. Some patients had no or low expectations at the start of treatment, whilst others had expectations that were influenced by some bad previous treatment. When this was compared to their current CLP care, it meant that patients were very happy. Also, the understanding that a CLP patients` outcome would never be perfect might prepare the participants to be satisfied with any outcome. In addition, looking at CLP as 'normal' compared to more serious conditions may have influenced their satisfaction.

*''I didn't really know what to expect. I was very happy with the outcome because they were a lot nicer than when I went in, my teeth but I don't know, I suppose they were never going to be perfect because obviously I'm cleft palate, they've, I'm missing a couple of teeth and stuff, so they've had to fill in gaps and things, but yeah I was happy with the outcome. I wouldn't, obviously you could always have nicer teeth and I'd like mine to be a bit nicer. They were harder to clean with the braces and stuff, so they're a little bit discoloured, a little bit crooked because if I don't wear retainers all the time they'll loosen up a bit and twist, but on the whole they've, it could have been so much worse. Yeah, I'm pretty impressed with what they've done with just a bit of metal and plastics, pretty incredible really, it's good."I**NT#11.M***

*'' Thank you to the NHS, yeah it's been a pretty painless experience really. Its only a minor thing really a cleft palate compared to proper disabilities and diseases, but they treat it with respect and fix it yeah with minimal fuss and I cant really complain, its been great, yeah."**INT#11.M***

Fourthly, when the overall positives were overwhelming the negatives the patient may think that the treatment was worth it.

'' Yeah. I think there have been a few more negative times, but not more than good times, but they aren't very many, and I think ... well I think there was one which was quite distressing, I think it was the consultation where we were discussing whether to cut my pharyngoplasty or not, and yeah, that was quite a difficult one, ... and I left that consultation feeling quite distressed. And I think it is more sort of immediately after it you can feel the most strong emotions, just because it often is quite sudden or a bit out of the blue when they tell you sometimes or suggest something. Yeah, and I suppose something my mum and I discussed that day was the fact that you can often be strong in the room in front of the surgeons and everyone else, but they don't see what goes on

*later on or when you're at home and you're trying to think about what to do, and it's very confusing and you just don't know. But in the end it actually turned out really quite well."**INT#3.F***

Fifthly, the timing of investigating patient satisfaction may have had an influence. For example, it was stated that if participants were interviewed just after the surgery then they would have a far more negative view than after full recovery and settling into a routine life following the surgery. A point worthy of note was that this study reviewed patients at or near the end of their care pathway when levels of satisfaction would be likely to be high. There may have been different responses if interviewed earlier on in the pathway.

*'' . It entirely brought forward my jaw and just that in turn affected the profile of my cheeks and my nose [ph] even [ph] a little bit. So while I was and I still do have a fairly flat profile it has changed a lot and become a lot more prominent. And especially my chin as well. So yeah, while it wasn't nice [inaudible-47:24] at the time, in the long run I was very happy with the results."**INT#3.F***

*'' Just my operations, with my distraction I didn't go out of the house for over a month, I was just inside and then just all the operations have so much recovery time, so, and when I'm not recovering, I don't really want to see even my friends, I don't want to see, because I just look a mess, and just better when I look normal, and I don't know, I think it's weird how I've, how my looks have changed, when I look at old pictures, I look like a different person, everyone always says to me in my old pictures that I look completely different, so my family had to come to terms with me looking different to what I once used to look like, so I don't think it, it wasn't stressful but it was hard, because I didn't like it at first because I wasn't used to it, but now I love it, so."**INT#9.F***

Sixthly, the patient was happy because the services are free on the NHS.

*'' Not really, I've just been pretty impressed with it. Yeah, there's always the slight hiccup here and there with waiting times and losing notes and stuff but that's to be expected from a giant hospital Trust with notes going about everywhere, they're going to misplace it from time to time. No, I'm pretty happy with how it's gone really. I imagine there will be people that will say they weren't but we're pretty forgiving and its not, treated me well, it's been free, I can't really complain, it's been good."**INT#11.M***

Seventhly, the possible patient feeling that treatment made him/her more 'normal'.

*'' they don't have a clue so me, I hate to say, not normal but by me having my face and lying not normal ... but obviously I'm not saying I'm not normal but I am normal, it will give me that courage to more talk about it with people."**INT#14.M***

Eighthly, it was also reported that the patient was happy however they could not compare their care to other care so they would not know another experience.

> *'' Decent I suppose, I've only lived this one life with these treatments, I wouldn't know in comparison to someone else. You would have to sit me down with someone who has the same thing as me, probably got a worse off hand. And I am very fortunate to have what I have, to be able to sit here and discuss it. So I suppose it's been decent. But I could not possibly measure it compared to others.''INT#28.M*

Another possible reason is that only happy participants agreed to take part in this study. It would be sensible to consider that patients may become satisfied if the treatment outcomes were good. However, the data in this study showed that many factors or influencers may have participated to shape the patient's perception of their satisfaction of the care they received. Patients may have experienced some negatives influences about the care however it appears that these minor negative issues had less influence on their satisfaction. This in part provided some possible explanations to patient satisfaction. The data so far presented the participants` perspectives on different aspects of their treatment. The next section will overview overarching concepts that came out across the data.

6.2 Overarching themes

Within the data, some concepts/themes appeared repeatedly. These will be presented in this section. These themes are drawn out to provide links between the different parts of the results and between the different chapters. This will hopefully help us to understand the 'big picture'.

1. The CLP care experience could be better understood within participants' lives: this links the experiences of living with CLP with the participants` reasons for treatment and with the treatment benefits realized by participants. This also supports the consistent finding that participants need to feel 'normal' as well as the participants' perception about treatment being influenced by their day to day life experiences.

2. The CLP pathway was a temporal pathway - over time and across ages: this concept was explained previously. The ageing and changes that occur as patients become older may impact all aspects of care, from information and treatment choices, to satisfaction and outcome. What is perhaps most interesting about this group of patients was that they grew up within the care pathway and the end point was often set with very little input from them but became a focal point for their treatment and the lives that build up around the treatment. Treatment episodes were happening at different times in their lives and that the end point was when they grew up. This subliminally became an accepted fact of their childhood although this also raised their motivation levels at later treatment milestones. This suggests that a person with CLP has little of the background of the early treatment and may have little control over the end point as this was often set before they reached adulthood. Furthermore, many experiences were temporal and tended to resolve with ageing such as bullying, stigma etc.

3. The end point of the treatment is normality: based on the data, the ideas of wanting to be normal and making comparisons with others were evident. This concept was present in a positive form as feeling better when comparing themselves to more severe cases and in a negative form as they felt that they needed to be 'normal'. There was an end point but the parameters of that end point were largely decided before the patient was old enough to fully contribute. In terms of aesthetics and function, the end result may not have been absolute but may have been closer than before. Treatment was part of a CLP patient's life, and orthodontic/orthognathic treatment in particular was seen as the end point, the final stage of treatment. This may place more importance on treatment closer to the end of the care pathway.

6.3 Summary
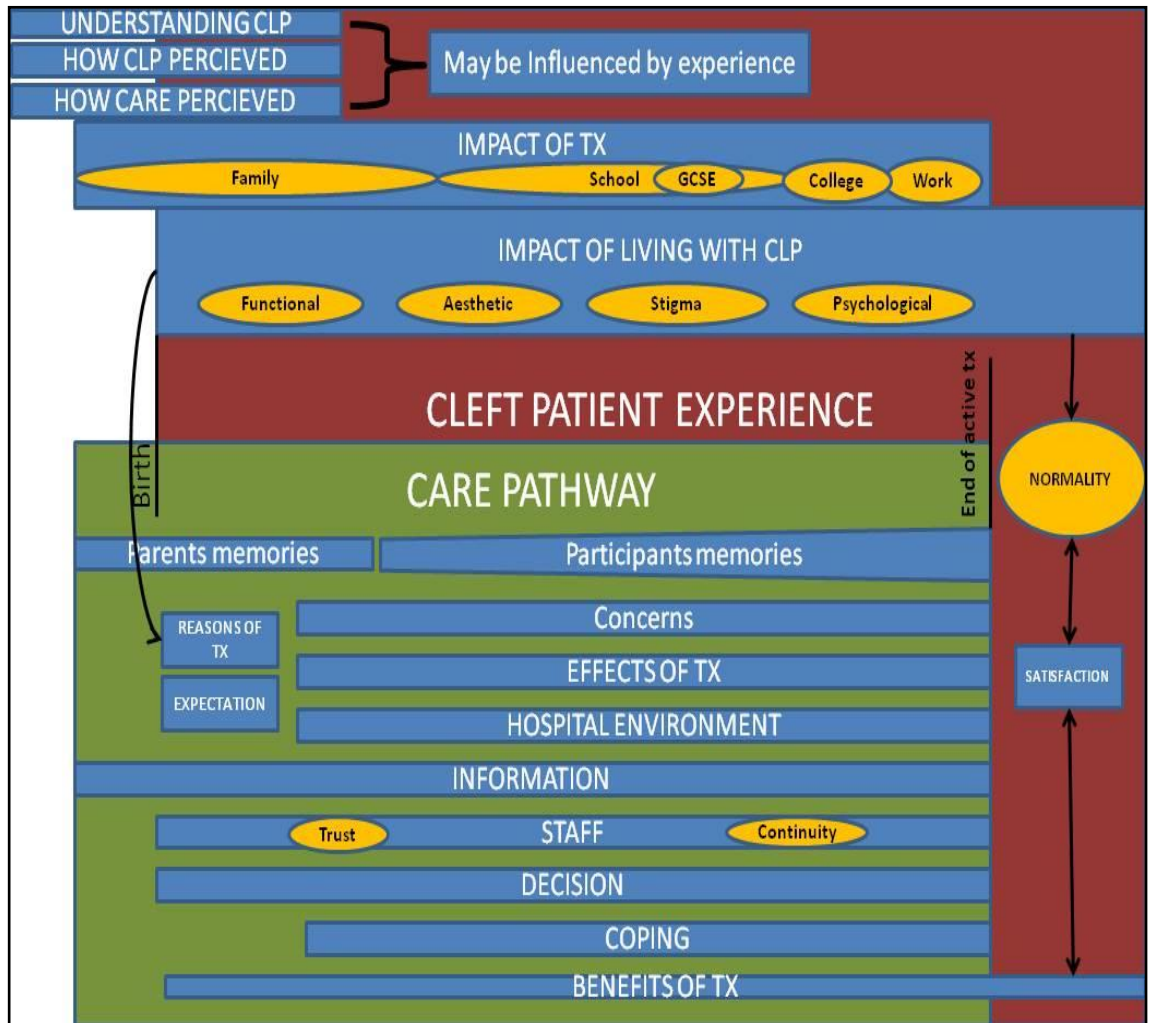
Figure 24: Summary of chapter 2

In this chapter, the findings demonstrated the different aspects of the CLP patients' experience which were related closely to different treatment episodes. Together with the previous chapter, the data provides a rich understanding of the CLP patients' experience that cover not only treatment; but also provide an insight into the broader context within which care is experienced and understood.

7. DISCUSSION

This chapter brings together the results of this study which has investigated the experiences of people living with CLP and how the CLP care pathway can be understood within the context of daily life. Particular attention is paid to definitive orthodontic and/or orthognathic stages of treatment as they can be seen to represent the end of the care pathway and the end of active treatment for patients born with an orofacial cleft. The research findings have been presented in some detail in the previous two chapters and will be addressed here both in relation to the aim and research questions that the study sought to address and also in relation to the wider literature around CLP care and around the evidence from more general long-term condition research.

The chapter starts by placing the aim of this study within the wider literature before taking each of the research questions in turn. The first research question looks at the experiences of living with CLP on a daily basis, drawing out the fact that experiences of CLP are inextricably tied in with experiences of the care pathway and treatment. The second research question explores perceptions and experiences of the care pathway specifically, looking at various aspects of treatment as well as at expectations and experiences before focussing in on definitive orthodontic and/or orthognathic treatment as the end point of care and exploring the implications of this. The fourth part of this chapter then looks at CLP within the context of wider literatures around the sociology of long term conditions and what we can learn from, and add to, this literature. The limitations of the current study are then outlined.

7.1 Aim of the study

To understand experiences of the CLP care pathway within the everyday life experiences of young people and adults living with CLP, with a particular focus on definitive orthodontic and/or orthognathic treatment at the end of the care pathway.

Sharif et al., (2013) identified the paucity of qualitative evidence in their review in relation to the experience of people living with CLP. This also includes the experience and perception of the long term treatment of CLP. Understanding the perspective of people living with CLP may help to understand issues related to treatment such as levels of adherence and satisfaction. CLP care in the UK was reorganised after the critical report

of The Clinical Standards Advisory Group in 1998 (Sandy et al., 1998). Little qualitative work has been done, however, with a focus on understanding the experiences of people using the newly centralized CLP services following the reorganisation. Qualitative studies which involve interviewing parents or carers are present in the literature (Nelson et al., 2012a, Nelson et al., 2012b) but there are fewer studies involving the participation of children (Hall et al., 2012, Hall et al., 2013).

The focus on the experience of definitive orthodontic and/or orthognathic treatment is justified for several reasons. The first is that it is placed at the end or near end of the CLP care; participants at that stage have experienced a great deal of diagnosis dependant CLP care and therefore they can reflect on the entire process. Another reason is that orthodontic/orthognathic treatment involves the major contact hours between participants and clinicians as orthodontic treatment can take up to 3 or 4 years. Furthermore, there is a gap in the literature about people with cleft's experiences of their orthodontic treatment within the care pathway and within their day to day life.

7.2 Findings and clinical implications of the study

- **What Is The Experience Of Living With Cleft Lip and/or Palate and what Impact Treatment Have On The Daily Lives Of Participants?**

There is a widespread belief that we should build up a picture of the views, expectations and understanding of an individual in order to understand their particular needs and experiences. This idea has been most eloquently argued in debates around the provision of care. As far back as 1847 the Danish philosopher Soren Kierkegaard suggested that care could only be provided through understanding the 'position' of the person that is receiving the care:

> ''If you really want to help somebody, first you must find him where he is. This is the secret of caring. If you cannot do that, it is only an illusion if you think you can help another human being. Helping somebody implies you understand more than he does, but first of all you must understand what he understands. (Kierkegaard, 1847)

Building on this idea, the first research question looks at the experiences of living with CLP on a daily basis, drawing out the fact that experiences of CLP are inextricably tied

in with experiences of the care pathway and treatment. Widespread pre-natal testing in the UK at the 20 week anomaly scan means that the majority of the participants in this study were already within the care pathway when they were born. Cleft lip is now commonly identified on antenatal scans although cleft palate remains a rarer diagnosis at this point but should be diagnosed in advance of discharge from the maternity unit. This can make it difficult to separate out treatment as this becomes part of daily life, particularly in childhood where decisions about treatment, appointments, timings etc. are taken by parents rather than the participants themselves. This also raises interesting questions about the extent to which experiences of CLP and the care pathway are shaped by both the physical and psychological ageing process. Participants grow up within the care pathway and so are likely to experience different issues and have different needs at different stages. This has implications for the provision of information at different stages of care and in different formats and for interactions within the CLP team as well as for issues such as stigma which appear to be tied in to particular time points within the course of life.

The main themes that emerged from the analysis are presented below and synthesised with the wider literature.

Understanding of cleft lip and/or palate

The literature is lacking any research focussing on what CLP participants understand about their condition. The results of this study suggest that there are a broad range of interpretations and explanations of CLP amongst those living with the condition, with explanations ranging from the failure of the palate to close prior to birth, to the mother being bitten by a dog during pregnancy. What is clear, however, is that there is no consistent, shared understanding of the condition and how it came about amongst people living with CLP. Who have grown up within the care pathway. One possible explanation for this is that information is provided to parents when the children are very young and it is left to them to explain to their children. The children are then reliant on what parents have understood, remember, or choose to tell them. This supports work carried out by Chapados (2000) and Patel and Ross (2003) who emphasise the importance of giving appropriate information to people with CLP. Adopting a more systematic approach to providing information with facilitated information sessions or giving leaflets supported with explanations followed by clarity insurance would be a potential way forward. Information should also be tailored to different age groups including children. It would be interesting to find out what parents understand about CLP and how they decide what information to pass on to their children.

Some participants in this study were not keen to know anything more about their CLP. There were several possible reasons for this, some participants felt that they already had sufficient information, some felt that they were happy with the current situation and did not need any more information, and others felt that, as there were few definitive answers, there was no point in learning more until there was something worth learning. Customising information to individual needs would ensure that information was provided in the right format at the right stage to maximise the chances of patients within the pathway having the information they need at the point at which they need it. Also, all participants must have the option to get more information if they want it, and should know where they can access that information. As life stories start early in cleft participants (Hall et al., 2013), this emphasizes the need to provide information about cleft earlier on in the treatment journey. This may help to combat bullying and stigma that might be faced by participants at particular points in their journey. The need to understand why they had CLP is evident in the data and this emphasize on the importance of the current large ongoing multicentre research to answer the question, 'why do I have a cleft'.

Impact of living with cleft lip and/or palate

The data from this study showed consistently that aesthetics was seen to have impact on daily life for participants living with CLP. Functions such as speech and eating were also affected. Participants highlighted the impact of living with a face which looks 'different'. This ties in with the work of Hunt et al. (2005) who stated that CLP participants have low facial satisfaction. Also the findings of Chetpakdeechit et al. (2009) may confirm this as they reported that females use make-up to hide any visible defects. Participants in this study linked visual difference in their faces to low self-confidence, again building on previous work (Turner et al., 1997) which found that the majority of young adults think that their self-confidence is affected by having a cleft. It is also reported that cleft may affect social engagement (Tiemens et al., 2013) which again is reflected in the results of this study. This study also explains the change of some behaviour due to the effect on physical appearance like being less social because of teeth appearance.

One particularly interesting finding in our study was the use of the word 'normal' by participants, who talked about wanting to look 'normal'. This is an important finding and it raises questions about why participants chose to refer to themselves in this way and how they came to perceive themselves as abnormal. One potential explanation as to how the term normal may be adopted is in response to particular incidents within daily life. Participants in this study highlighted a range of stigmatising experiences or events within their lives that they linked to looking, or sounding abnormal. This ties in with the

CLP literature, which shows that affected individuals faced different forms of stigma (Chapados, 2000, Chetpakdeechit et al., 2009, Sharif et al., 2013). It also builds on, and expands, work by Tiemens et al., (2013) which found that girls living with cleft were likely to experience stigma. This study supports this finding and adds that both genders may be affected. There is also the suggestion that people with less severe CLP and less obvious visual differences may be less likely to experience stigma, but this needs to be explored further as it was beyond the scope of this study. This is confirmed by the findings of (Hall et al., 2013) who note that cleft does not necessarily cause biographical disruption but this study emphasizes that disruption may be caused by how severe the case is and any related experiences.

The feeling of 'abnormality' expressed by participants in this study may be a response to the way in which society acts towards them. This supports the findings from other studies which show that cleft participants seek normality (Havstam et al., 2011) and want to be like everyone else (Chetpakdeechit et al., 2009). Perceptions of having a lower chance of gaining employment (as presented in the data) or of making meaningful relationships work alongside felt and enacted stigma to boost feelings of difference or of feeling 'not normal'. This is particularly interesting in relation to the definitive orthodontic/orthognathic treatment (as will be explored in more detail later) as this possibly becomes seen as the endpoint of treatment, the point at which they would become 'normal' or as close to it as possible.

The role that school environments play in the fostering or combating of stigma and bullying is also worth highlighting. Stigma was experienced by most participants at some point and the most common times to feel stigmatised were at school. Surprisingly, and somewhat disappointingly, participants suggested that their schools adopted a passive response to enacted stigma toward CLP individuals. This corroborates the findings of (Chapados, 2000) that participants who faced teasing related to their cleft had little support from teachers. This is a disappointing finding as children spend a significant part of their life at school. Bullying and stigma can have serious consequences, particularly in relation to self-esteem, as can be seen from the accounts of the participants in this study. Perhaps, therefore, a more active approach to dealing with bullying is necessary at schools and other organizations that deal with children. Chapados (2000) findings support this and suggested that nurses can also play a key role here by providing information to teachers.

A related issue within this study was the importance of the CLP clinical psychologist as a key part of the team that can provide support for children and adults dealing with the issues of self-esteem, self-confidence and stigma highlighted above. Early assessment

of CLP participants by the psychologist has been written into the current clinical service specification for CLP services in the UK. This echoes the findings of (Turner et al., 1997). The group of participants that presented with more severe forms of CLP and those who had experienced felt and/or enacted stigma, indicated to the need for more psychological support. The participants in this study suggested that stigma reduced over time. The provision of age appropriate, personalised information on CLP for affected individuals and schools and ongoing support from the psychologist within the team are positive ways of addressing the negative impacts of CLP.

In addition to this the amount of time spent attending hospital appointments can also have the effect of making people with CLP feel different from peers, a finding backed up by Chetpakdeechit et al. (2009). That found that the length of stay in hospitals made participants feel different.

It may be that organising appointments to minimise disruption could facilitate a less disruptive experience for the CLP patients and their families. Both Havstam et al. (2011) and Alansari et al., (2013) made the link between the impact of cleft and cleft care on daily life and the satisfaction levels of cleft patients. We would go further to suggest that CLP care is not just affected by daily life but cannot be understand as separate from the day to day lives of participants. To reiterate a point made previously, children with CLP are born into the CLP care pathway, it is a part of their lives from the start and it would seem counterintuitive to separate the two.

The impact of treatment on the day to day life of cleft participants

Clinical treatment is understood as part of the journey that is taken by children with CLP and the results of this study suggest that it is difficult to separate this out from daily life. Attendance at appointments, for example, may affect school attendance, resulting in difficulty keeping up with school work, and by association this will also affect the parent or carer who also needs to take repeated time off to attend appointments. This is compounded by the fact that the CLP care pathway is a long and continuous system, requires regular attendance at clinics and is funded in the UK health system from 0-21years of age. In addition to that some procedures may require long recovery times and this can also cause disruption to the participant`s life, and to that of their parents or carers where the treatment involves children (Nelson et al., 2012a). It is important to note, however, that participants in this study viewed these impacts as relatively small when analysing the expected overall benefits from their treatment.

The data in this study showed the impact of participants' treatment on schools and on the role that schools can play in minimising or maximising the effects of treatment on education. Participants suggested that some schools are not understanding and adaptive either cooperative and in relation to attendance at multiple appointments within the school day or in providing support in combating preventing bullying. This may create a conflict between parents and the affected child's school. This reflects the findings of Chapados (2000), suggesting that schools need to be more aware of the need for flexibility and pastoral support for students with CLP. It would also be helpful if communication extends between care providers and the school to make this 'official' and maybe this can be more helpful for parents as they do not need to explain and give excuses every time their child goes for treatment. However participants' carers' permission must be granted.

Previous studies have identified the negative impact of CLP on educational achievement (Hunt et al., 2005) and this study also showed that education may be affected by the long term commitment to treatment as part of the CLP care pathway and also the lowered levels of self-esteem and self-confidence identified previously. It is important to note, however, that not all participants in this study were adversely affected and that the ability to catch-up with missed work and the associated resilience was seen as a positive result.

Simplifying access to, and flexibility of, appointments would be one way to begin to address this issue. This would both reduce the impact of treatment on education and may also improve patient attendance reducing failed appointments. Evening and weekends appointments would potentially make treatment more family friendly as it would also reduce the amount of time taken off work by parents. This should be considered within the organization, resource allocation and management of care delivery. The same flexibility is needed in relation to treatments which require significant recovery periods. As much of the CLP care provided is elective, chosen by the patient and/or their family (depending on age) more information about recovery times and the degree of flexibility available for pre and post operative appointments will help patients to plan their care and their education/employment around one another, potentially improving both satisfaction and the likelihood of adherence.

Following the centralisation of CLP services in the UK after the CSAG report, CLP care has become focused on high volume centres. Whilst acting as central points for expertise, these centres may well not be geographically convenient for all patients (Sandy et al., 1998). Issues around travel times and cost were also raised in this study. This might be a downside of reducing the CLP treating centres as a response to the CSAG report. The findings of the Eurocleft study support that treatment may affect

participant`s school and social life (Semb et al., 2005a). This study also shows that the financial cost of that travel could be a problem to some families.

One final point to highlight when looking at the impact of treatment on daily life is the reliance on memory and the recall of participants around treatment that was often undertaken many years previously and/or when they were young children. The data from this study suggest that participant's early treatment memories are constructed from what parents or carers tell them and from partial memories of particular aspects of treatment. This is similar to the findings of Hall et al. (2013) and Havstam et al. (2011) and this highlights the importance of giving clear and detailed information to parents as a significant part of this information will be passed on to the child receiving the treatment. Again this also highlights the importance of providing tailored information to children throughout the pathway.

The data supports the, perhaps obvious, suggestion that participants tended to remember most accurately the most recent part of their treatment. This would suggest that more recent treatment would highly influence overall participant satisfaction. In addition, it would be advisable to assess satisfaction with a specific procedure shortly after that procedure has been undertaken so participants can recall it more clearly. The data within this study demonstrated that participants tend to recall much about their orthodontist and their experiences within that specialty when compared to other disciplines. This might be because orthodontic treatment requires a long and protracted treatment time and this means that the orthodontic team may have the most direct contact hours with participants. Also participants tend to be older when receiving orthodontic care and so will be more likely to recall that phase of their treatment – clearly they are unlikely to recall anything to do with their primary surgery as they were so young at this time. Another explanation may be the influence of this study's' introduction and the information sheet given that explains to participants that we wanted to understand their perspective on and experiences of orthodontic treatment.

Summary

The experience of living with CLP can only be understood within the wider context of daily life. What is apparent here is that understanding of the condition and what it means is socially constructed. Information about CLP is often obtained indirectly through parents rather than members of the care team, at least initially, and is subject to the interpretation, and explanations that parents provide. The impact of CLP on daily life takes two forms. The first form is biomedical and depends on the severity of the condition and how it manifests both aesthetically and functionally (particularly in relation to speech). The second form is social and depends on the responses of those around the individual to their CLP, manifesting itself in experiences of stigma and bullying. What is clear from the accounts given in this research, backed up by findings from other studies, is that it is the social impact that becomes the focus of people's accounts of living with CLP (the focus on difference and the move through treatment towards 'normality' is explored further in the next section). Understanding the impact of CLP on daily life provides a more nuanced understanding of the reaction of participants to some aspects of the care pathway such as the underlying reasons for seeking elective treatments and satisfaction with treatment. These issues and are explored in more detail in relation to the second research question and are discussed below. A final point to make here is that the impact of time emerged very strongly in the accounts in this study as a way of making sense of the experience of living with CLP. Underlying understanding about the condition, social experiences and the impact of treatment is the concept of time/ageing and the fact that participants have grown up within the care pathway. The journey starts before the participants in this study were born and it is a journey over time that encompasses childhood and beyond into adulthood. It is within this context that CLP and CLP care needs to be understood.

- **How do participants perceive/experience the treatment care pathway including the definitive orthodontic/orthognathic?**

This second research question explored perceptions and experiences of the care pathway specifically, focussing on the different aspects of treatment as well as exploring expectations and experiences. Again the link between expectation, experience and the wider context in which the care is taking place comes across clearly.

Definitive orthodontic and/or orthognathic treatment was highlighted as the potential endpoint of most planned CLP treatments and the point at which participants left the care pathway. After this point they are able to return to the service if new clinical concerns arise but for the purposes of this research this was taken to represent the final experience of the care pathway. What is interesting here is that the endpoint of treatment was seen by some participants as the point at which they were going to become 'normal' or as close to it as they were able to be. This was the point that they had been working towards since before they were able to have a say in treatment options and, for many, the endpoint was a given that had been established by the decisions that were taken many years previously and what they had been told since early childhood.

The results from answering this research question add to the literature about the experiences of patients receiving CLP treatment and can be grouped into three sections:

1. Treatment reasons/rationale/expectations
2. Inside the care pathway
   a. The care environment and process
   b. Experiences of definitive orthodontic/orthognathic care
3. The end of the care pathway to 'Normality'

1. Treatment reasons/rationale/expectations

Participants chose to seek or indeed not to seek treatment for many different reasons. Participants also may not actively seek treatment at all but might simply remain on the journey started by their parents / carers when they were babies. The results suggest that aesthetic reasons drive patients to seek care most consistently. One possible reason for this is that functional problems are usually corrected earlier within the CLP care pathway. The majority of treatments in the earlier years are related to functional problems and these are dealt with at the beginning of the care pathway. At that stage, decisions about seeking treatment come from the parents/carer's rather than the people living with CLP, due to age, and as such it was not possible to explore this within the current study.

When asked about treatment decisions participants talked about the need to correct their aesthetic concerns to minimise the stigma associated with looking different. This ties in clearly with the consistently expressed desire to become 'normal' with CLP affecting their aesthetics. It also explains why participants are willing to undergo complex and possibly high risk procedures solely to improve aesthetics. This ties in with the finding of another study Havstam et al. (2011) that found that cleft participants tend to stop having treatments when they feel that treatment can not improve their look. This raises interesting questions about who imposes aesthetic values and why aesthetics are so very important in current times.

In relation to treatment expectations, there is no previous work that has used a qualitative approach to focus on the expectations of people living with CLP. The results from this study suggest that participants may not be entirely clear about their expectations of treatment, and in some cases participants stated that they do not have any expectations. This may reflect the fact that treatment is a journey started before the participants were actively involved in decision making and so participants find themselves on a pathway that is, or may seem, predetermined. Interestingly, other group of participants responded to the research question about expectations of treatment by talking about the nature of the treatment rather than the outcomes. This may be because they know about different aspects of treatment from previous information they received so have less uncertainty when undertaking treatment. The drift of answers from outcomes to treatment itself raises questions about whether participants clearly know what outcomes they can expect and why they are choosing not to think about outcomes. One possible explanation for this drift is that expectations becomes blurred as participants have received treatment over such a long time period that they become detached from the process and became passive. Some participants also talked about uncertainty, either because of a paucity of information that they have received or because information received emphasises the uncertainty, for example where the clinician has explained that they are uncertain of the precise outcome of some procedures when carried out for an individual patient.

Where expectations were outlined, a participant talked about scar removal, an outcomes which were not clinically feasible. This raises concerns about unrealistic expectations and may cause significant distress for the participant and the clinician providing that care when the expectations are not met at the end of the treatment process. The uncertainty and variability of expectations expressed highlight the importance of clarifying to patients what treatment can do for them, the risks and benefits of treatment and of having no treatment. Understanding should be checked and a rigorous and thorough informed consent process should be adhered to. Within the care pathway patients should be given as much time and support as possible and it is clear that speaking to the psychologist

and to other patients who have received the same treatment is beneficial. Going into a surgery expecting to 'remove the scar' can be severely disappointing if the outcome is to 'improve the look of the scar' rather than removing it, so the language used needs to be very clear, the use of simulation and images and involving patients / parents / carers in the planning is vital. The key here is to provide detailed and appropriate information tailored to the age and needs of each individual patient. This should help to reduce uncertainty.

2. Inside the Care Pathway:

A significant section of the answers to the second research question focused on the actual treatment process, incorporating the physical, clinical and interpersonal environment in which treatment occurs.

a) The care environment and process

Hospital environment

The precise make up of the clinical setting may play a role in an improved participant experience. Ironically the play area may be the only memory that sticks in a child's mind when coming to the hospital for a major operation. The same concept applies with adults but in a different form. The results of this study have demonstrated that an organized and effective clinical layout can be perceived as better than a chaotic environment which may appear disorganised. An integrated service with different clinical departments in the same area was perceived as reassuring. This should be the aim for any service reorganisation or update and this study, suggests that patients would prefer an integrated environment. Interestingly, this may have a crucial influence in participant satisfaction with the service. This suggests that care providers can implement some modifications in the clinical settings to help for a better participant experience.

Multidisciplinary clinic environments are widely seen as beneficial but multidisciplinary team meetings were sometimes perceived in a negative way. A CLP team may include many different specialties and their trainees. These meetings could be seen as intimidating if too many people attended the joint clinic meetings. This could make it less likely that patients will ask the questions they want answered about their care and so acts as a barrier to adequate communication. Some participants in this study stated a preference for smaller meetings. This reflects the findings in studies by Hall et al. (2012)

and Chapados (2000), who both found that children may refrain from raising questions in the multidisciplinary clinic. If patients and/or their families feel unable to ask the questions they need answered they may leave the clinic with uncertainty about the condition or treatment and dissatisfaction with the process. One potential way to address this would be to seek permission first from patients and/or parents/carers to allow extra people to be present at the meetings. This could be arranged in their previous appointment to avoid embarrassment on the day of the joint clinic. Alternatively the clinicians may clear the room of all non essential staff for at least part of each meeting. One to one meetings with psychologists were also seen as particularly helpful by participants in addition to more general meetings.

Staff

Patel and Ross (2003) found, perhaps unsurprisingly, that participant's quality of life was improved by compassionate and competent care from the multidisciplinary team. This was also seen in the data from this study, where the importance of time and care were highlighted. Participants in this study placed a large amount of trust in the clinicians involved in their care and stated that they were largely happy to base their decisions about treatment on the expertise of clinicians with whom they had often developed relationships with over a period of years. This continuity of care was also seen as both a positive and important factor in raising levels of satisfaction and trust, tying in to the work of Semb et al., (2005b) who found that continuity of care and good communication improve patient satisfaction with treatment. Clinical and administrative staff avoid 'if possible' overbooking clinics as this may affect the quality of communication and influence a patient's experience.

It has been suggested in the literature that participants may prefer to get information orally from the CLP team specialists (Semb et al., 2005b). This supports the finding that participants seem to place a large amount of trust in clinicians. Clearly an open relationship with the clinical team is desirable for good patient communication but they must be given time and space to make their own choices as a stressful or pressured clinical environment may lead to the participants blindly following what clinicians suggest without fully evaluating and engaging with establishing what they want to do. Participant choice is all important. Continuity of treatment with the same clinicians within the team also appears to be important to participants. Therefore, it is advisable to let the same orthodontist to see the same participants throughout treatment. This may also augment patients' contentment and act as an aid to improving treatment satisfaction (Semb et al., 2005b).

The importance of multidisciplinary expert care was highlighted when contrasted with non-specialist care received by some participants. Examples of care received pre and post surgical recovery suggests that non CLP specialist nurses may not have a full understanding of the care pathway that a patient is on and the implications of the treatment within the bigger picture of ongoing care. Participants highlighted episodes where nursing staff had not been made aware that a patient would have a wired jaw post-operatively and require a liquid diet, for example. This can be upsetting for patients and introduce difficulties if some of the treatment is delivered away from the core team. Whilst information should be shared between the centre and all sites where CLP care is delivered across a clinical network, ideally via a fully integrated electronic system of notes, in reality this is clearly not happening in all cases. The availability of a specialist CLP nurse post operatively to check on patient care would be one way to address at least part of the problem encountered.

Information

In terms of information and sharing, there is much information shared with patients around the CLP care pathway and this information is channelled to different ages and understanding levels. This is reinforced and refreshed at many opportunities throughout the process. Chapados (2000) emphasized the importance of patient information and the need to provide appropriate explanation to CLP participants. Sharing of information is also evident in support networks, CLAPA, family, friends and from clinical staff and all this information is of paramount importance in assisting a patient in negotiating their way through the care pathway. The results from this study suggest, however, that not all of the information provided is seen as helpful or in fact remembered by participants. The previous section exploring the findings in relation to the first research question highlighted the fact that participants may not have sufficient information about CLP and about how and why it occurred and what could be done to treat the symptoms of CLP. The difficulties around the provision of information for patients of different ages and the cross over between parental and patient decision making were also touched upon. All of this has implications for the provision of information.

Some participants in this study were confused about treatment times and recovery times at different points in their care. Confusion over the length of time needed for post-operative recovery, for example, and the implications of this for planning when elective treatments should be undertaken and also organizing time off work/study was highlighted. Participants felt that they did not receive enough information about the nature of the treatment that they would be undergoing. One participant was not aware,

for example, prior to starting orthodontic treatment, that they would be required to attend the clinic every 6 weeks and that this was for clinical / biological reasons and not due to a lack of appointment availability. Clearly it is crucial for patients to understand not only what treatment they are likely to receive but also how that care is delivered. As highlighted in the previous section, treatment needs to be integrated into daily life and this can only be done if patients are aware of the potential impact that each course of treatment may have. The data showed that insufficient information could be a source of concern for patients.

The findings of this study suggest that, where possible, clinicians should give standard and customized information based on participant`s need and that information should be age appropriate and provided throughout the care pathway, with specific information made available even to young children, information should be explained and patient understanding should be tested along with time and space for questions.

The CLP care pathway is long and complex and involves a large number of specialists within the multidisciplinary team. This means that information can be received from a number of different sources at the same time and raises the possibility that patients may receive conflicting information from different clinicians. This may be confusing. This stresses the need for a standardized set of information to be given to participants where possible, or at least for members of the team to communicate with one another prior to communicating with the patient. It has been reported in the CLP literature that confused or unclear information may be a cause of dissatisfaction (Semb et al., 2005b). This reinforces the findings of this study.

Decision/Choice

Participants' state that they need to be involved in their treatment and need to be listened to and this is reinforced by the literature (Semb et al., 2005b), tying in to current rhetoric around patient centred care and patient choice. There are times in the CLP care pathway where this is difficult, however. Early in the pathway choice about treatment may be limited as the primary surgeries are deemed mandatory to restore anatomy and try to ensure the best operating function. The lack of choice, or expectation that certain treatments would be undertaken was clear in the data, but it is important to note that this was not always seen in a negative way. Expectations can be that certain treatments would be undertaken at certain times and this was taken as pre-determined, simply because participants had been told from a young age that this was what would happen. For others there was an expectation that a trusted expert would inform them of what

needed to happen and when. Neither of these paths was seen as particularly problematic but accepted as the way things were and had always been. These patients grew up in, and were socialised into, the care pathway. This said, there is evidence that some CLP patients would prefer to be more involved in decisions making, mirroring the findings of Chapados (2000) in an earlier study who found that participants do not feel involved in decision making process.

One possible recommendation is that it would be also helpful for participants to not feel pressured into making decisions at the time of clinic and defer their opinion until a further appointment where they feel more comfortable. By doing so, participants may discuss their issues at home, with the psychologist or with other patients in a more comfortable one to one environment and may be encouraged to raise their concerns and/or questions subsequently at a second appointment. The clinician must be completely sure that participants are happy and convinced to go for the discussed procedure.

The literature suggests that the decision making process around treatment planning should involve patients, parents and clinicians. The precise construction of that process varies across time but there is wide agreement that the process should be a joint one. This is supported by the work carried out by Hall et al., (2012) who suggest that decision making involve family members and differ according to treatment type. The findings from this study add to understanding in this area by demonstrating the importance of aesthetics as an outcome and the need for treatment choices and planning to be understood within the wider context of daily life.

Coping

In relation to coping mechanisms, psychological assessment should be undertaken as early as possible within the program of care. This was also suggested by (Turner et al., 1997). In addition, the data shows that support from other CLP participants can be perceived as a strongly positive tool and helpful for patients when considering coping with the effects of treatment. However, it seems that these meetings do not happen that often and therefore care providers could introduce structures to facilitate more meetings of this type for their patients. Also, being involved in a social task and not being isolated may help participants to cope with the condition. This places an important responsibility on the affected individual`s family as well as their clinical teams and charitable CLP organizations such as CLAPA who do facilitate face to face as well as social activities over the internet.

Finally, the fact that the clinical teams main aim through the overall treatment plan is to help improve final functional and aesthetic appearance - to remove the stigmas of orofacial clefting and to re-establish the familial facial features may aid the patient cope with treatment. This is similar to the finding of (Havstam et al., 2011) where treatment was tolerated with the perception that the care provided was hoped to 'normalize' the participants' facial form. This ties in to the consistent theme of 'the restoration of participants' to produce 'normality'. This is explainable as many participants had suffered in their life experiences to date from stigma related to their appearance.

Patient Satisfaction

The benefits of CLP care are clearly stated with some of them obvious and some of them concealed. CLP treatment can change the life of the average CLP participant. In the literature, there is an emphasis on treatments involving reconstructive surgeries to improve aesthetics which, in turn, improves confidence (Tiemens et al., 2013). Speech improvement also improves quality of life (Patel and Ross, 2003) and participants of this study have made this clear. Developments and improvements in the delivery of CLP care in the developing world and an increasing sense of benevolence in the general population along with wide scale advertising campaigns in cities all over the world has placed a greater importance on international CLP organizations to support actions that provides CLP care pathways, often via charitable organizations, in developing parts of the rest of the world.

When asked about satisfaction with care, overall satisfaction was high amongst the sample in this study. Whilst there was dissatisfaction with aspects of care along the way, the overall feeling was positive. This ties in with work by other researchers in the field who suggest that the notion of participant satisfaction is multi-factorial and rarely hinges on an individual event (Semb et al., 2005b, Sitzia and Wood, 1997). This is particularly the case where care has been undertaken over an extended period of time across the life course. Participants in this study took the whole experience as a package and this means that if one aspect of care dropped below beyond a certain standard, another aspect may compensate and the overall satisfaction level remained positive. This said it is worth being aware that self-selection bias may have occurred in this study, in that only happy/satisfied people agreed to participate, as is possible with any study of this kind. Alternatively, it has been suggested that participants tend to give socially desirable responses in such studies (Sitzia and Wood, 1997) as they aim to please, answering the research questions positively, damping down any possible negativity. Also when considering memory bias, and to avoid the influence that different treatment aspects may

place on one another, it would be advisable to assess participant satisfaction about an individual procedure soon after the results of the effect it has are evident. By doing this, we may reduce memory bias and we may get a more genuine representation of satisfaction with that particular procedure.

The findings around the treatment process within the CLP care pathway suggest that a multidisciplinary environment is seen as positive but care needs to be taken when involving patients in multidisciplinary team meetings as these may be intimidating and stifle patient involvement. Trust and continuity of care were seen as extremely important and the need for a specialist team was emphasised. Information tailored to the age and needs of individual patients and their families was needed but simply providing written information without checking understanding is not sufficient. In addition it was noted that the nature of the CLP care pathway means that patients felt that they had little choice over treatment (at least in relation to major treatment options) as there was an expectation that certain treatments were to be undertaken at certain times. Patients were brought up within the pathway and socialised into these expectations so this did not affect overall positive levels of satisfaction with care.

b) Experiences of definitive orthodontic/orthognathic care

One of the foci of this study was definitive orthodontic care within the pathway. Definitive orthodontic/orthognathic treatment needs to be understood as part of a long process that people with CLP are born into. Previous studies on orthodontic treatment have used numerical measures to assess the different components of the experience. Factors such as pain and satisfaction and the impact on daily life have been evaluated (Fleming et al., 2009, McNair et al., 2009, Mandall et al., 2006). Whilst some studies have used telephone interviews or focus groups (Bennett et al., 2001, McNair et al., 2006) this study is the first to use in depth qualitative interviews to gain the perspectives of CLP participants and to evaluate their thoughts in a longitudinal manner, describing the impact of their orthodontic treatment within the wider context.

This allowed for the development of an understanding of how orthodontic care fits within the bigger picture. Also, the longitudinal description of the process allowed the exploration of how the ageing process may influence thoughts about different packages of orthodontic treatment. For example, there is an increase in self-confidence as the teeth become straighter but this excitement and motivation may reduce with time especially if the participant cannot see significant improvements themselves. This is evident with CLP

participants as usually they require a more complex and longer orthodontic treatment plan which may take several years to complete.

The detailed description of a patient's experience can provide relevant facts that could be useful to update information leaflets. Adding specific examples provided by those actually undergoing care would offer a 'closer to reality' perspective and would offer more convincing information for participants. From the data, it appears that the length of treatment and how often they need to be seen should be clearly understood by participants and is another item that should be explained clearly by clinicians. Therefore, a complete and comprehensive variety of patient information would be helpful. Participants should be told in full about all the possible beneficial effects and the risks of treatment and should be involved in their treatment planning.

Orthodontic treatment is normalised to the extent that non-cleft peers may well be having treatment at the same time. However, participants may reduce their levels of excitement with time and especially if they don't see positive changes. Towards the end of orthodontic care, once the first and very visible phase of aligning the teeth is completed, space closure and finishing phases which are less immediately visible, may be perceived by participants as 'no changes phase'. Therefore, clinicians should be aware of this and encourage different techniques to keep motivating their participants. For example, using images / photos of similar clinical situations of how their teeth should look the end of their treatment plan can often continue a patients' motivation. Participants may be kept motivated until the end of treatment especially if they are aware of the likely treatment time. Where the length of treatment relies on the participant, for example when elastics are required to improve the bite in orthodontic care, the clinician may 'encourage' the participant that their commitment to this treatment adjunct may speed the treatment time. Knowing this in advance may possibly help participants to closely follow instructions and will also be likely to empower them to engage with their treatment such as wearing elastics. For orthognathic surgery patients, similar principles apply. Inaccurate information about the estimated recovery period appears to be problematic and the data enclosed also, suggests that during the recovery period, psychological support is of prime importance.

Due to the fact that orthodontic/orthognathic treatment is positioned towards the end of the care pathway and the fact that there are likely to be changes of appearance; it may be considered that this is the point where they are having all their remaining CLP stigma removed or camouflaged and 'becoming normal'. As participants describe that orthognathic surgery is a significant part of their overall treatment may support this. However, the change of the appearance could be unwanted and potentially

psychologically damaging if the participant was not prepared for it. Therefore, clear information and usage of the available contemporary, prediction softwares that can morph images to guide the team and the participant on the possible outcomes is essential. Although these prediction softwares are not always totally accurate, they at least provide an envelope for participants to have a reasonable and fair idea of their proposed aesthetic outcome.

3. The end of the care pathway and 'Normality'

Babies born with CLP are born in to the CLP care pathway, the appointments, specialist team and clinic itself become a part of their childhood. This means that it is difficult to separate experiences of CLP or its treatment from everyday life. The end of the care pathway can be seen then to represent both the end of treatment but also the end of a key stage of life. What was interesting in this study was that it showed how some participants may perceive treatment as 'the road to normality'. Treatments placed around the end of care such as the definitive orthodontic/orthognathic stage are final steps in becoming 'normal'. Participants may well have been aiming, throughout the whole of their lives, to get to this point, and some spoke of this final stage of treatment as something that had been discussed throughout their treatment as the endpoint, tying in to the earlier discussion about a lack of choice and predetermined treatment plans.

The desire to become 'normal' is clear and evident from the findings of this study and much of the associated literature. This provides an explanation that connects participants' experiences of stigma related to their CLP and their focus on the importance of aesthetics. Therefore, orthodontists working with CLP patients should understand the importance that CLP patient may place on this aspect of the care pathway and should provide accurate information to help to shape expectation.

Summary

As the data presented shows, many aspects of CLP care treatment are influenced by day to day life experiences. Understanding these experiences will allow us to understand treatment need, treatment expectations and treatment satisfaction. Participants may feel that the treatments received at the end of the care pathway are a final step on the road to becoming 'normal'. Orthodontic and/or orthognathic treatment can provide participants with a big step towards what they perceive as 'normality'. The results also demonstrate that actually everything may be connected within the participant perception of treatment. The participant's life experiences influence some aspects of treatment. Also, expectations of treatment proved a strong uncertainty. Furthermore, the end point of treatment is important as participants can see results and probably because they barely remember the first stages of their treatment, treatments that they will not recall that were consented for in infancy by parents or carers. Also, the data showed the similarities between the CLP experience and the sociological literature on living with long term conditions. This is expanded upon in the next section.

Understanding cleft lip and/or palate as a long term condition

In the literature review in chapter two the sociological literature on living with long term conditions was outlined along with the contention that CLP can be understood as a long term condition and that there may be similarities in the experiences of people living with CLP and other long term conditions. To this end a long term condition was defined as any condition that lasted for an extended period of time and affected multiple areas of daily life. The case was made that CLP could be understood as a long term condition as the symptoms and treatment extend over a period of at least 2 decades and affect many areas of daily life including education and social relations. Analysis of the transcripts from this study showed that the key themes identified within the sociological literature also appear in the experiences of people living with CLP.

One interesting difference between CLP and other long term conditions which needs to be noted at the outset is that unlike most other long term conditions which are degenerative, orofacial cleft improves over time with treatment. Orofacial clefting starts with the 'worst' clinical picture and as treatments are undertaken, CLP related aspects improve throughout the CLP care pathway. The primary presentation is usually with the newborn baby with a variation of orofacial clefting and as a result of the various treatment interventions the child starts a journey towards restoring what could be considered `normal` aesthetics and function as time elapses. This is in stark contrast to many other long term conditions, which often worsen with time and are often progressively disabling. Although for long term conditions treatments may aim to address the symptoms of the condition, treatments for orofacial clefting aim to fully restore form and function.

Based upon the data presented, all of the six themes presented by (Conrad, 1987, Scambler, 2008) that emanate from the long term condition and sociology literature can be picked out from the data in this study. In the literature review chapter the evidence is set out to see whether CLP can be understood as long term condition in a way that has not been done before and we draw parallel by using research on long term conditions to provide a potential context. The results showed that this hypothesis; if used carefully; is valid and hopefully will allow a better understanding of CLP. The themes of uncertainty; stigma; family involvement; management of the medical regimen; information and sharing; and the rethinking of self can all be identified within this study and are briefly outlined below.

Uncertainty is very clearly highlighted throughout the results of this study. Participants repeatedly state that they do not know how treatment will conclude, what the results of the treatment will be and what they are going to look like at the end of their care. They have all kinds of hopes for an ideal but they do not know what is ultimately going to

happen. This ties in with the literature (Scambler, 2008, Scambler, 2012) from studies of other long term conditions where there is uncertainty about the long term outcomes and the impact of treatment. Uncertainty is also evident in terms of the information patients are given during their care and this ties into the whole process by which information is provided at different stages and for different reasons. Clinical teams need to produce information for patients and parents and in an age appropriate way. CLP treatment starts with care of the infant and as the participant ages, the consent process switches from parent to patient. This transition can sometimes be difficult for the family to manage – often there will be a broad knowledge that a particular treatment will happen at a particular time but until the patient is deemed suitably mature, they will have little say on this matter.

The results from this study also identifies that ***stigma*** (Goffman, 1986) was experienced and this was discussed in the first results chapter. The experiences of participants in this study suggest that stigma is most likely to be experienced at specific times within the lives of participants, coinciding with points at which they are in formal social situations. The stigma experienced was both felt and enacted and most often experienced whilst participants were at school. Stigma experienced was most likely to be linked to unclear speech and the aesthetic appearance of the face. The literature talks about managing information in relation to difference and this is also evident in the accounts of participants who highlighted the fact that a better understanding of the condition and how and why it happens would better enable them to control information in a positive way and combat stigma.

The ***family*** is involved in supporting the patient in a huge number of ways. Due to pre-natal testing a baby is born within the CLP care pathway and so all of the initial decisions about treatment are made by the parents. From this starting point CLP impacts on the family in several ways. The family, both collectively and as individuals, is involved in the care of their participant and there is a sense that the family shapes the direction of the treatment and may initially act as a gate keeper for the information shared at the start of the care pathway. In addition the family negotiates access for the treatment. This is evident in the way the data shows their concerns, for example, about missing time from school and how committing to treatment may affect parents' and carers own employment situation. Family involvement appears to reduce with time rather than increase as the individual takes control of the direction of their treatment. However the burden on the family is evident especially in the early years of treatment.

Management of the biomedical regimen becomes evident when participants report the effects of treatment, for example attending any CLP appointment, having to keep an

orthodontic appliance clean, the wearing of orthodontic elastics and the different types of food that all orthodontic patients are advised they can or cannot eat. In a sense, around the differing treatment times, their whole lifestyle becomes dictated by a biomedical regime. This is as clinical teams are committed to helping their patients maximise the quality of the outcome and to engage the patient to shape the direction the treatment is going and to ensure overall that treatment is working and is likely to be a success. This is similar to the experience demonstrated in the literature when considering long term conditions (Locker, 2008, Pinder, 1998).

The importance of, and need for ***information*** throughout the care pathway, age appropriate and personalised to the needs of the individual patient and/or their parents has been demonstrated throughout the study. The results highlight the need to ensure that information is being channelled to patients at different ages and different understanding levels throughout the care pathway. Sharing is evident in the CLP support networks, in the family, among friends and the clinical staff and all are crucially important as all are committed to reaching a satisfactory end point.

Finally, rethinking the work that talks of *self* (Bury, 1988), the impact of CLP on identity can be seen most clearly in the comments that participants make about how they look and feel and their experiences of stigma and bullying in school. Participants make clear links between the way that they look and levels of self-confidence and self-esteem.

This suggests that people working within the CLP pathway could learn from the experiences of those working with people with a range of long term conditions. The key point is that CLP, as with any long term condition, affects multiple areas of daily life and treatment can only be understood within this wider context. The CLP care pathway is only one aspect of life with CLP and is experienced and interpreted as part of this bigger picture rather than as an entity in its own right. Understanding this emphasises the importance of members of the care team working with CLP patients and their families to put together a treatment pathway that fits into the context in which it is being experienced.

7.3 Methods and limitations

As with any research there are certain limitations within this study that need to be acknowledged. The first limitation is that the researcher in this study is a dentist and orthodontist. This raises the possibility of bias both in the way that questions are framed and in the way that the data is interpreted. To counter this, a trial interview was conducted and observed and transcripts were reviewed at regular intervals through the data collection process. In addition the data was blind double and triple coded by non-clinical and clinical members of the research team to ensure that analysis was consistent and unbiased. The potential for the researcher to impact on the research was also a concern in relation to the interaction between the participants and the interviewer and a reflexive approach was adopted throughout the research process to ensure awareness of potential bias. This is standard practice within qualitative research and there is an expectation that interviewers influence the data that they collect. Reflexivity allows for this to be acknowledged and incorporated into the understanding of the data.

A further limitation in this study relates to the retrospective nature of the study as it explores participant experience across a number of years. This makes it likely that memory bias may be present, but this is addressed within the analysis where the nature of events remembered is considered, and is a problem common to any study which asks participants to reflect on previous events or experiences.

Participants' bias may also be present which means only happy or satisfied people agreed to participate. There is also a possibility of the study introduction bias which places a greater importance on the orthodontic treatment. This could influence participants to respond with answers that relate to orthodontic treatment.

A consistent criticism of qualitative studies is that sample sizes are small and that the results cannot, therefore, be generalizable.

28 participants were recruited and interviewed before data saturation was reached. This is within standard parameters for a qualitative study, and the large amounts of rich data generated from 28 in-depth interviews allow us to develop a good understanding of experiences of CLP and CLP care. The purposive sampling strategy adopted within this study could also be considered a limitation as participants were approached at certain stages in the care process. Participants who had reached the end of the care pathway and experienced the definitive orthodontic/orthognathic treatment involved as part of their care were specifically targeted to allow for an overview of the whole process to be obtained along with detailed accounts of specific experiences along the journey. The age range of participants is broad because; depending on the case, the definitive orthodontic

treatment can be completed any time from 15 years upwards and orthognathic treatment can be completed from 17 years. Beside the diversity in participants' ages, participants with different types of CLP were purposively invited to participate and equal numbers of male and female participants were invited to participate to incorporate as much variation as possible. This necessarily means that a small number of participants were interviewed within each group, but this is consistent with the aim of the study which was to explore broad experiences of CLP care within the context of daily life. Although 'generalization' of the findings is not intended, this study focused on 2 CLP centres which follow the CLP care pathway that other central services follow in the United Kingdom. Therefore, it is possible to hypothesize that CLP participants in other CLP services may share similar experiences.

8. CONCLUSIONS AND RECOMMENDATIONS

8.1 Conclusions

The aim of the present research was to undertake a qualitative study to explore the CLP care pathway from the perspective of people living with CLP and with a particular focus on the definitive orthodontic/orthognathic treatment. In meeting this aim, the first research question explored the experiences of people living with CLP and the impact of treatment on daily life. The second research question explored experiences and expectations of the CLP care pathway with a focus on definitive orthodontic/orthognathic treatment as the end point of the pathway.

In addressing these research questions six key findings emerged, these are outlined below:

1. Experiences of the care pathway cannot be separated from the daily lives of people living with CLP and need to be understood within this wider context.

   o The need for information, and the type of information needed will be influenced by the context in which CLP is being experienced.
   o How participants perceive CLP and its care pathway was largely influenced by their experiences of living with the condition and how the CLP and its treatment affected/impacted their day to day life.
   o The participants in this study suggest that CLP impacts negatively on daily life including aesthetic, functional, psychological and social impacts.
   o Experiences of stigma, the severity of the CLP and participants perceptions of their own CLP influenced the degree to which CLP impacts on daily life.
   o Participants sought treatment to correct aesthetic and functional effects of their CLP.
   o The data suggests that the most recent treatment are likely to be remembered best. The data also emphasised the importance of providing adequate information to parents for early years' procedures and information targeted at children throughout the process.

2. Building on this, the second key finding is that people with CLP are born within, and grow up in the care pathway. Therefore, there needs to be acknowledgement of this ageing process within the pathway. This can be understood in relation to the change in who makes decisions, the need for information, and the impact of

particular stages of the life course on CLP and treatment – i.e. schooling/stigma/orthodontics etc.

- Ageing influences various aspects of care, such as information needs at different stages, treatment choices that are made, expectations, concerns and so forth.
- The people involved in the decision making process change over time as the patient ages and takes on responsibility for their own treatment choices.

3. The third related key point is that whilst the participants in this study are adults, for large parts of the pathway they have had little say over the treatment they are undertaking – either because of age or a lack of information.

- Participants in this study highlighted a lack of understanding of information at some points.
- There was a perception that treatment choices had been made a long time ago, prior to the participants reaching adulthood, and that there was little chance to change the preordained process.

4. There are specific learning points around the organisation of the care pathway.

- Staff can play a significant role influencing participants' experiences of their care. Empathy, expertise, the provision of information and good communication skills were all highlighted along with continuity and the chance to build trusting relationships over time.
- Interdisciplinary clinics may be intimidating to some patients. This may influence their information seeking behaviour. Some participants welcomed the opportunity to questions different members of the team whilst others felt intimidated and unable to ask questions or seek clarification.
- The CLP care pathway may be perceived as a burden which negatively affects daily activities, such as the need to take regular time off school, and/or work, both for the affected child/adult and their family or carer.
- CLP and the care pathway affect the lives of both the affected person and their wider family, both through practical aspects of care (transport, time off etc) but also by the way that the condition is understood and perceived by others.

- o The data suggest that participants found it helpful to arrange major life interrupting treatment procedures around significant time points such as taking exams, starting college/university or before starting a new job.

5. Definitive orthodontic/orthognathic treatment might be of symbolic significance within the pathway as the endpoint. Participants characterised this as the point at which they would become 'normal' and it was much anticipated, often for many years, as the journey through the pathway was undertaken.

    - o Orthodontic care was normalised and accepted by participants as many peers also underwent orthodontic treatment at around the same time. This meant that the treatment was perceived to have less impact on daily life.
    - o The CLP care pathway may be perceived as a journey towards 'normality' with orthodontic care as the end point of this journey.
    - o The benefits of treatment were particularly apparent at the end of the pathway.

6. Finally, this study shows that there is a strong similarities between CLP participants' experiences and the common themes in the long term illness literature.

    - o This related to uncertainty about cause and outcome, the impact on the wider family, the need to fit treatment into daily life, the experiences of stigma, and the impact of CLP on self-esteem, confidence and the importance of information.

    - o The main difference with other long term conditions lay in the positive trajectory of CLP and the finite nature of the pathway.

Overall this thesis has provided rich data to help clinicians to understand the experiences and perspectives of people with CLP on their treatment which may help to inform the CLP care pathway.

8.2 Recommendations

The findings of this study provide valuable insight into the experiences of people who have travelled along the CLP care pathway. From this insight a series of recommendations can be made with a focus on the provision of information, aspects of clinical practice and the CLP service and in relation to the role of schools. These recommendations are presented below.

8.2.1 The Provision of Information

- Standardised, age appropriate, comprehensive, personalized and accurate information should be provided about different aspects of treatment (treatment choices, processes, risks and outcomes) to parents and affected children/adults. People with CLP and their carers should know where to get this information from and be given the opportunity to ask questions. This will help them to make informed choices and plan treatment around everyday life commitments.

- Effective communication is essential and children/adults with CLP and their families need time and space to make sense of the information that they are given and ask questions. Continuity of care and building trust over time are important in fostering a relationship where patients and their families feel that they can ask questions and make decisions about their care.

- Current information leaflets need updating to address specific issues raised in this study. For example, the reason for long intervals between orthodontic appointments should be added to the information leaflet and explained to patients committing to orthodontic treatment.

- It was suggested in this study that information can be used as a tool to combat stigma and bullying but many participants felt that they lacked the information needed to explain their condition and its causes. Information should be developed to provide a basic outline of CLP and its implications that could be used to raise awareness in schools, workplaces and more widely as needed. This could be done in association with organizations such as *CLAPA*. Other resources are also available for individuals, parents and schools, through organisations such as Changing Faces (www.changingfaces.org.uk › Get Support › Anti-bullying Info).

8.2.2 Cleft care and the Care Pathway

- Care providers need to understand how care fits into daily life and the impact that multiple appointments can have on education and work for both patients and their families. All efforts should be made to provide flexibility in relation to appointment times and locations, possibly incorporating evening and weekend appointments.

- The importance of psychological support was highlighted within this study, particularly at specific points within the pathway (when starting or moving schools, or starting work) when participants were most likely to experience stigma and bullying. Patients should be assessed by a psychologist as a standard procedure to identify early those patients in need of further support, particularly around coping strategies, and self-esteem.

- It is essential that clinicians understand the reasons why patients choose to undergo treatment and their expectations, as these may not mirror those of the clinician and this could affect patient satisfaction.

- Mulitdisciplinary team meetings, whilst useful, can be intimidating for patients and their families and make it less likely that they will ask questions or offer opinions on the treatment options being discussed. Clinicians should offer their patients in advance the option of a smaller meeting with only essential staff in attendance.

- In addition, care providers are encouraged to promote continuity of care by the same clinicians/team throughout treatment.

- CLP international organizations should support actions that aid providing a CLP care pathway in other areas of the world. Care assessors should understand the complex nature of patients' perspective and should adopt a more holistic approach and consider all factors and influencers of patient satisfaction. This study provided an insight to understand CLP patients' experiences.

8.2.3 Schools

- It is important that teaching staff within schools are aware of the implications of treating a child with CLP, the need for frequent appointments and the likelihood that time off will be needed within the school term. Parents and schools should work together to minimise the impact of repeated absences.

- It is essential that schools are aware of the specific experiences of children with CLP in relation to stigma, bullying and low self-esteem and put support in place within schools to tackle and bullying that does occur.

- Direct communication between care providers and schools would support parents and children in ensuring that the needs of the child are met educationally, clinically and psychologically. This could come in the form of a leaflet specifically designed for schools (primary and secondary) outlining the clinical implications of CLP, stages of treatment, experiences of children living with CLP and recommendations.

8.2.4 Recommendations for Future Research

Due to the inductive nature of this study, the findings can inspire many disciplines to continue research in this area. There are several recommendations for researchers:

- One is to recruit patients treated at different sites in the UK to see if their experiences are the same or different.
- For future research, it is advisable to include patients and their parents/or clinicians to see how well their experiences and views match up.
- Future researchers might consider a broader range of different care pathways other than the UK model to understand the strength and weaknesses of different care pathways.
- A large scale quantitative study would be useful to look at the issue of stigma and the numbers of people who encounter felt or enacted stigma that they attribute to their CLP. This would be particularly useful if it was able to link experiences of stigma to particular points in the life course.
- There were a number of suggestions made about aspects of the care pathway that could be changed. For example, a qualitative study to explore participants' perspectives of two different styles of the multidisciplinary clinic could be undertaken. It was also be useful to look at ways of providing information targeted at different age groups and specific points in the life course - such as school transitions - across the pathway.
- It would be both interesting and potentially useful to conduct a further qualitative study with healthcare professionals working within the cleft care pathway to follow up on the issues raised in this study about the pathway and experiences of it and identify any ways in which current practices could be changed to better meet the needs of patients.

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10. APPENDICES

-Ethical approval

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06 August 2012

Dr Sasha Scambler
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Dear Dr Scambler,

Study title:	Patient and parent experience of orthodontic treatment for cleft lip and palate patients (CLP).
REC reference:	12/LO/1022
Protocol number:	N/A

Thank you for your letter of 01 August 2012, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information was considered in correspondence by a sub-committee of the REC. A list of the sub-committee members is attached.

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.

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission 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 sites

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

-All related documents

Reference number: 12/LO/1022
Version number: 2
23-07-2012

KING'S College LONDON

LETTER TO PARTICIPANT

Dear Parent/Carer/Guardian/Participant

RE: A research project entitled 'Patient and parent experience of orthodontic treatment for cleft lip and palate'

We are writing to ask for your kind help in participation in a research project. Please read the information sheet enclosed. This information sheet is to inform you about a research project that is being carried out by a postgraduate research student, Raed Abualfaraj, at Kings' College London.

As part of his PhD, Raed is assessing patient and parent/carer/guardian experience of orthodontic treatment for cleft lip and palate. The project aims to discover how you may feel about the orthodontic treatment involved as part of ongoing management for cleft lip and palate.

Please read the enclosed information sheet and if you decide to participate or if you need further information please contact Raed on 020 7848 5145 or email: raed.abualfaraj@kcl.ac.uk

Also, if you are interested to know more about this study. Raed will be available in the clinical area at the day of your appointment and he is more than happy to answer all your questions.

Thank you very much for your time in considering this matter.

Yours Sincerely,

Professor Fraser McDonald
Professor of Orthodontics and Head of Orthodontic Department
Kings College London

Cc Raed Abualfaraj, Postgraduate Student

King's College London

LETTER TO PARTICIPANT

Dear Parent/Carer/Guardian/Participant

RE: A research project entitled 'Patient and parent experience of orthodontic treatment for cleft lip and palate'

We are writing to ask for your kind help in participation in a research project. Please read the information sheet enclosed. There are two different information sheets (one for the parent/carer/guardian, the other one for the child). This information sheet is to inform you about a research project that is being carried out by a postgraduate research student, Raed Abualfaraj, at Kings' College London.

As part of his PhD, Raed is assessing patient and parent/carer/guardian experience of orthodontic treatment for cleft lip and palate. The project aims to discover how you and your youngster may feel about the orthodontic treatment received as part of ongoing management for cleft lip and palate.

Please read the enclosed information sheet and if you decide to participate please tick (I am happy to take part in this study) and write your details into the reply form enclosed, and then post it back to us on the enclosed stamped envelope.

Or if you need further information please contact Raed on 020 7848 5145 or email: raed.abualfaraj@kcl.ac.uk

Thank you very much for your time in considering this matter.

Yours Sincerely,

Professor Fraser McDonald
Professor of Orthodontics and Head of Orthodontic Department
Kings College London

RaedAbualfaraj
Postgraduate Student

Reply Form

-I am not happy to take part in this study ☐

-I am happy to take part in this study and here are my contact details ☐

My contact details:

Name:

Contact number:

Email:

Please tick on the box and place this form into the enclosed stamped envelope and post it back to us.

Thank you.

ASSENT FORM
For Participation In Research (14-16 years old)

Title of Study: <u>PATIENT AND PARENT EXPERIENCE OF ORTHODONTIC TREATMENT FOR CLEFT LIP AND PALATE PATIENTS</u>

REC Reference: 12/LO/1022

Please Initial box

1. I confirm that I have read and understand the information sheet dated *06.06.2012* for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

   ☐

2. I consent to my interview being recorded and transcribed verbatim.

   ☐

3. I understand that I can decide at any time in the research to stop participation and to have my data withdrawn without giving any reason. <u>I understand also that if I decide not to participate or to withdraw at any time, it will not affect the standard of care I receive.</u>

   ☐

4. I understand that relevant sections of my dental notes and my personal information may be looked at by individuals from the research team or, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.

   ☐

Participant's Statement:

I -_____
Agree that the research project named above has been explained to me to my satisfaction and I agree to take part in the study. I have read both the notes written above and the Information Sheet about the project, and understand what the research study involves.

Signed .... **Date**............................

Investigator's Statement:

I -_____
Confirm that I have carefully explained the nature, demands and any foreseeable risks (where applicable) of the proposed research to the volunteer.

Signed.. **Date**............................

248

CONSENT FORM
For Participation In Research (16 years and older)

Title of Study: PATIENT AND PARENT EXPERIENCE OF ORTHODONTIC TREATMENT FOR CLEFT LIP AND PALATE PATIENTS

REC Reference: 12/LO/1022

Please
initial box

1. I confirm that I have read and understand the information sheet dated *06.06.2012* for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I consent to my interview being recorded and transcribed verbatim. ☐

3. I understand that I can decide at any time in the research to stop participation and to have my data withdrawn without giving any reason. I understand also that if I decide not to participate or to withdraw at any time, it will not affect the standard of care I receive. ☐

4. I understand that relevant sections of my dental notes and my personal information may be looked at by individuals from the research team or, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records. ☐

Participant's Statement:

I -
_____
Agree that the research project named above has been explained to me to my satisfaction and I agree to take part in the study. I have read both the notes written above and the Information Sheet about the project, and understand what the research study involves.

Signed .... Date.............................

Investigator's Statement:

I -
_____
Confirm that I have carefully explained the nature, demands and any foreseeable risks (where applicable) of the proposed research to the volunteer.

Signed... Date.............................

CONSENT FORM
For Participation In Research (Parent/Carer/Guardian)

Title of Study: <u>PATIENT AND PARENT EXPERIENCE OF ORTHODONTIC TREATMENT FOR CLEFT LIP AND PALATE PATIENTS</u>

REC Reference: 12/LO/1022

Please Initial box

1. I confirm that I have read and understand the information sheet dated *06.06.2012* for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I consent to my interview or my child/youngster interview will be recorded and transcribed verbatim.

3. I understand that I can decide at any time in the research to stop participation and to have my data withdrawn without giving any reason. <u>I understand also that if I decide not to participate or to withdraw at any time, it will not affect the standard of care my child/youngster receive.</u>

4. I understand that relevant sections of my child/youngster dental notes and personal information may be looked at by individuals from the research team or, where it is relevant to my taking part in this research. I give permission for these individuals to have access to these records.

Parent's/Carer's/Guardian's Statement:

I -

Agree that the research project named above has been explained to me to my satisfaction and I agree to take part in the study. I have read both the notes written above and the Information Sheet about the project, and understand what the research study involves.

Signed .... Date.............................

Investigator's Statement:

I -

Confirm that I have carefully explained the nature, demands and any foreseeable risks (where applicable) of the proposed research to the volunteer.

Signed... Date............................

250

What if I want to take more time to think or if I want to ask more questions about this research?

If you would like any further information please do not hesitate to contact me by:

phone on 020 7848 5145 or alternatively by e-mail; raed.abualfaraj@kcl.ac.uk

Thank you for reading this sheet – please ask any questions if you need to.

KING'S College LONDON

PATIENT AND PARENT EXPERIENCE OF ORTHODONTIC TREATMENT FOR CLEFT LIP AND PALATE PATIENTS

Participants information sheet (14-16 years old)

What do you think of...

Orthodontic treatment?

Who are you?

My name is Raed Abualfaraj and I am looking into orthodontic experiences as part of my study at Kings College London University.

What are you doing?

My research is trying to find out about what patients with cleft think about their orthodontic treatment.

What you will do?

I would like to talk to you and ask you a few questions about what do you think of the orthodontic treatment you have received. There are no right or wrong answers. Your views are important and you can say anything you want. We can meet wherever you like at any time that is helpful for you; at your house or at the cleft centre .I will sit and talk to you and I will record what we both say with a voice recorder to help me remember our conversation. No one will know what you said other than myself. A nurse will be with us when I meet you. I hope that you will find this fun.

Why you invited me?

Because I am interested to know what patients with your tooth condition think about their orthodontic treatment.

What we should do next if I want to participate?

If you decide to take part we will ask you to sign a consent form. Then we will arrange a time for the interview. The interview is a set of questions asking about your experience and what did you expect from the orthodontic treatment. The interview will allow you to talk freely about your experience and also will help us to understand what you think.

What if I don't want to participate?

You are totally free to choose to participate or not. Your treatment will go on as normal and nothing will be affected. I would really be happy if you decided to join and I will appreciate you participation.

Even if you decided to join and then change your mind, there will be no problem with withdrawing from the study.

Is there any danger or advantage for me to participate?

There is no any danger related with this. We will just sit and talk.

The main advantage for the information you provide is to help us to understand cleft patients more so we maybe able to do things differently with future patients.

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PATIENT AND PARENT EXPERIENCE OF ORTHODONTIC TREATMENT FOR CLEFT LIP AND PALATE PATIENTS

Participants information sheet (16 years and older)

Researchers: Sasha Scambler
Blánaid Daly
Fraser McDonald
Raed Abualfaraj

We are pleased to invite you to take part in a research project. It is important for you to understand the reason for this research and what will be involved before you decide. Please take your time to read the following information carefully. Ask us if there is anything you need to be described in more detail and we will be more than happy to explain.

Thank you for reading this and we would appreciate your participation

1. What is the purpose of the study?

The project aims to explore the experience and the expectation of the orthodontic phase of treatment of cleft lip and/or palate patients.

2. Why I have been invited?

This study is to find out about the experience of cleft patients who have recently finished orthodontic treatment. We hope to involve parents/carers/guardians as well as patients. In order to do this, we need a group of people who have cleft lip and palate and their parents/carers/guardians who are willing to discuss their orthodontic treatment with us. .

3. Do I have to take part?

It is up to you to decide whether or not to take part. If you decide not to take part this will not affect the treatment you receive.

4. What will happen to me if I take part?

If you decide to take part we will ask you to sign a consent form. Then we will arrange for a private interview with both you and then your parent/carer/guardian. The interviews will follow a set of questions asking about your experiences and expectations of the orthodontic

treatment. The interview will allow you to talk freely about your experience and also will help us to understand what you think.

For your convenience, the interviews will be held in a non-clinical setting according to your preference either in a vacant room at The Cleft Centre at Guys' Hospital or at your home. The researcher will be accompanied by a nurse where interviews are conducted in your home. Interviews will be held separately between patient and parent/carer/guardian and the contents will be anonymous to both of you.

All interviews will be recorded with a voice recorder and transcribed verbatim into Microsoft Office Word document. Then it will be analyzed.

5. What are the possible risks to take part?

We believe that there are no risks to taking part in this project.

6. What are the possible benefits of taking part?

We believe that there are no direct benefits to you in taking part in this project. However, participating might help us to understand your perspective of the treatment and after consideration, this could benefit future planning of care for patients.

7. Will my taking part in this study be kept confidential?

Your responses and all information which is collected about you for this project will be kept confidential. Your name and personal details will be anonymised.

8. What will happen to the results of the study?

The results will be used by Dr Abualfaraj as part of his PhD project. The research will be published in academic journals and conferences and we will send you copies if you wish.

9. Who has reviewed the study?

This project was reviewed by the London - City & East Research Ethics Committee.

10. What will happen if I don't want to carry on with the study?

You can withdraw from the study at any time. Information collected may still be used depending on what you tell us.

11. Contact for further information?

If you would like any further information please do not hesitate to contact Dr Abualfaraj by phone on 020 7848 5145 or alternatively by e-mail; raed.abualfaraj@kcl.ac.uk.

If you are unsure feel free to discuss this with any member of the research team.

Thank you for considering taking part and for taking time to read this sheet – please ask any questions if you need to.

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Reference number: 12/LO/1022
Version number: 2
23-07-2012

PATIENT AND PARENT EXPERIENCE OF ORTHODONTIC TREATMENT FOR CLEFT LIP AND PALATE PATIENTS

Parent/Carer/Guardian information sheet

Researchers: Sasha Scambler
Blánaid Daly
Fraser McDonald
Raed Abualfaraj

We are pleased to invite you and your child to take part in a research project. It is important for you to understand the reason for this research and what will be involved before you decide. Please take your time to read the following information carefully. Ask us if there is anything you need to be explained in more detail and we will be more than happy to assist you.

Thank you for reading this and we would appreciate your participation

1. What is the purpose of the study?

The project aims to explore the experiences and the expectations of the orthodontic phase of treatment for cleft lip and/or palate patients and their parents/carers/guardians.

2. Why I have been invited?

This study is to find out about the experiences and expectations of children with cleft lip and palate and their parents/carers/guardians with regard to the children's completed orthodontic treatment. In order to do this, we need to speak to a group of children with cleft lip and palate and to their parents/carers/guardians.

Do I have to take part?

It is up to you to decide whether or not to take part. If you decide not to take part this will not affect the treatment your child is receiving.

3. What will happen to me if I take part?

If you decide to take part we will ask you to sign a consent form. Then we will arrange for a confidential one to one interview with me. The interviews will follow a set of questions asking about the experiences and expectations of the orthodontic treatment as part of

ongoing management of cleft lip and/or palate. The interview will allow you to talk freely about your experience and also will help us to understand what you think.

For your convenience, the interviews will be held in a non-clinical setting according to your preference either in a vacant room at The Cleft Centre or at your home. The researcher will be accompanied by a nurse where interviews are conducted in your home. Interviews will be held separately between child and parent/carer/guardian.

All interviews will be recorded with a voice recorder and transcribed verbatim into Microsoft Office Word document. Then it will be analysed.

4. What are the possible risks to take part?

We believe that there are no risks to taking part in this project.

5. What are the possible benefits of taking part?

We believe that there are no direct benefits to you taking part in this project. However, participating might help us to understand your perspective of the treatment and after consideration, this could benefit future patients.

6. Will my taking part in this study be kept confidential?

Your responses and all information which is collected about you for this project will be kept confidential. Your name and personal details will be anonymised.

7. What will happen to the results of the study?

The results will be used by Dr Abualfaraj as part of his PhD project. The research will be published in academic journals and conferences.

8. Who has reviewed the study?

This project was reviewed by the London - City & East Research Ethics Committee.

9. What will happen if I don't want to carry on with the study?

You can withdraw from the study at any time up to the data analysis phase. Information collected may still be used dependent on your wishes.

10. Contact for further information?

If you would like any further information please do not hesitate to contact Dr Abualfaraj by phone on 020 7848 5145 or alternatively by e-mail; raed.abualfaraj@kcl.ac.uk

If you are unsure feel free to discuss this with any member of the research team.

Thank you for considering taking part and for taking time to read this sheet – please ask any questions if you need to.

Reference number: 12/LO/1022

Version number: 2

25/07/2012

Topic Guide

This aim of this interview is to explore patients' or parents'/carers'/guardians' perspectives regarding the definitive orthodontic treatment as part of cleft lip or and palate (CLP) management.

Introduction:

- Reintroduce myself
- Reassure Confidentiality
- Reassure that either taking part or not will not affect their treatment care
- explain that there is no right or wrong answer and to say whatever they want.
- Explain the study briefly: Title, why we doing this study including aim and objectives
- Remind that the interview will be recorded and explain the reason why
- Remind that they are free to say anything they want and all responses will remain confidential

Personal information:

- Name, age, sex and ethnicity?
- Education
- Social status (works or studies ..)
- Other family member with CLP?
- Other family member who had orthodontic treatment?

Body of interview:

- Tell me what do you know about CLP?
- Tell me about your experience with CLP?
- Reassure that all responses will be anonymised and confidential

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- What do you think of the care involved? How did it work?
- Tell me about your comprehensive orthodontic treatment. (What do you think of it?)
- What are the reasons do you think you want for orthodontic treatment? (Or your youngster want to orthodontic treatment)
- What you were told about the orthodontic treatment?
- Was the information you had about orthodontic helped you enough?
- Tell me about the experience of the orthodontic treatment? Including any negative or positive experiences and concerns before or during orthodontic treatment?
  - Can you give me any examples of situations you encountered where you consider it was bad/negative?
  - Can you give me any examples of situation you encounter where you consider it was good/positive?
- Is there anything in relation to the orthodontic treatment you want to talk about?
- What was your expectation of the orthodontic intervention?
  - What was your expectation before starting the treatment?
  - What was your expectation in the middle of the treatment?
  - What was your expectation near the end of the orthodontic treatment?
- How does the experience correspond with the expectations?
  - (After the response) Why?
- How satisfied are you after orthodontic treatment?
  - (After the response) Why?
  - How satisfied were you during orthodontic treatment?
- What impact does orthodontic treatment have on the daily lives of you/your child?
  - What impact does orthodontic treatment have on the daily lives of you/your child during the treatment?
  - What impact does orthodontic treatment have on the daily lives of you/your child after the treatment finished?
- was the orthodontic treatment beneficial?
- what benefits you (or your youngster/child) gained from orthodontic treatment?
- Do you think it was worth it?

Ending:

- Is there anything you think we should have asked you about that we have not covered?
- Is there anything you want to add? Or if you want to add anything later you can call me..
- Reassure confidentiality of all responses
- Thanking for participation