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DOI:

[10.1016/j.ejpn.2016.06.004](https://doi.org/10.1016/j.ejpn.2016.06.004)

Document Version

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Citation for published version (APA):

Carroll, S., Chalder, T., Hemingway, C., Heyman, I., & Moss-morris, R. (2016). "It feels like wearing a giant sandbag." Adolescent and parent perceptions of fatigue in paediatric multiple sclerosis. *European Journal of Paediatric Neurology*. Advance online publication. <https://doi.org/10.1016/j.ejpn.2016.06.004>

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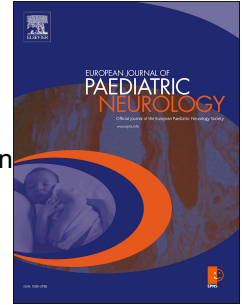
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Accepted Manuscript

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Susan Carroll, MSc, Trudie Chalder, PhD, Cheryl Hemingway, MBChB FRCPCH
PhD, Isobel Heyman, MBBS PhD, Rona Moss-Morris, PhD, Professor



PII: S1090-3798(16)30088-5

DOI: [10.1016/j.ejpn.2016.06.004](https://doi.org/10.1016/j.ejpn.2016.06.004)

Reference: YEJPN 2081

To appear in: *European Journal of Paediatric Neurology*

Received Date: 9 March 2016

Revised Date: 16 June 2016

Accepted Date: 19 June 2016

Please cite this article as: Carroll S, Chalder T, Hemingway C, Heyman I, Moss-Morris R, “It feels like wearing a giant sandbag.” Adolescent and parent perceptions of fatigue in paediatric multiple sclerosis, *European Journal of Paediatric Neurology* (2016), doi: 10.1016/j.ejpn.2016.06.004.

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“It feels like wearing a giant sandbag.” Adolescent and parent perceptions of fatigue in paediatric multiple sclerosis

Susan Carroll¹, MSc, Trudie Chalder², PhD, Cheryl Hemingway^{3,5}, MBChB, FRCPCH, PhD, Isobel Heyman^{4,5}, MBBS, PhD, Rona Moss-Morris¹, PhD

Affiliations: ¹Health Psychology Section, Institute of Psychiatry, Psychology and Neuroscience, King’s College London, UK; ²Department of Psychological Medicine, Institute of Psychiatry, Psychology and Neuroscience, King’s College London, UK; ³Department of Neurology, Great Ormond Street Hospital for Children, London, UK; ⁴Department of Psychological Medicine, Great Ormond Street Hospital for Children, London, UK; ⁵UCL, Institute of Child Health, Guildford Street, London, UK.

Address correspondence to: Professor Rona Moss-Morris, Health Psychology Section, Institute of Psychiatry, Psychology & Neuroscience, King’s College London, Guy’s Hospital Campus, London Bridge, London SE1 9RT, UK, rona.moss-morris@kcl.ac.uk, 00442071880180.

Abbreviations: caMS – children and adolescents with MS; CFS – Chronic Fatigue Syndrome; MS – multiple sclerosis

Word Count: 3342 + 241 word abstract

Abstract

Aim: Fatigue is one of the most common and disabling symptoms of paediatric MS, associated with depressed mood, impaired school performance and lower quality of life. Fatigue in children and adolescents with MS (caMS) is poorly understood, and effective treatments for fatigue are currently lacking. No qualitative studies have previously examined fatigue in caMS. Thus, the objectives were (a) to explore experiences of fatigue in paediatric MS and (b) to gain insight into how caMS and their parents respond to and manage fatigue.

Methods: In-depth semi-structured qualitative interviews were conducted with 15 caMS and 13 of their parents, either face-to-face or via telephone. Inductive thematic analysis was primarily used, incorporating elements of grounded theory.

Results: Five key themes were identified in the data. Participants described mentally and physically exhausting fatigue, which they perceived as uncertain and uncontrollable. Parents expressed concern about caMS' well-being and future because of fatigue, whilst caMS discussed the implications of disclosure or non-disclosure of fatigue to peers and teachers. An additional overarching theme addressed participants' emotional responses to fatigue. Participants stressed the need for better guidance on fatigue management.

Interpretation: This study provides a unique insight into the experience and management of fatigue in paediatric MS from the perspective of both caMS and parents. Some findings echoed experiences of fatigue in other populations, whilst new factors in paediatric MS also emerged. Fatigue management interventions involving caMS, parents and teachers should be developed and implemented in clinical practice.

Keywords: Multiple Sclerosis, Fatigue, Paediatrics, Adolescence, Qualitative

Highlights:

- Fatigue is a highly disruptive symptom for caMS and parents
- Parents and caMS experience fatigue as an uncertain, uncontrollable symptom
- Fatigue evokes concern in parents for caMS' well-being and future
- CaMS highlight the implications of disclosure or non-disclosure of fatigue
- Implementing fatigue management strategies in clinical practice is imperative

1. INTRODUCTION

Multiple Sclerosis (MS) is an inflammatory demyelinating condition of the central nervous system, usually diagnosed in adulthood. Although rare, paediatric MS can also occur, with a UK incidence rate of <1 in 100,000.¹ Sensory and motor impairments, bladder and bowel dysfunction and optic neuritis are among the most common physical symptoms, whilst cognitive deficits typically affect memory, language and attention and processing speed.²⁻⁴ Additionally, fatigue is one of the most common and disabling symptoms of MS, affecting up to 76% of children and adolescents with MS (caMS).^{5,6} The evidence to date suggests that fatigue in caMS is unrelated to clinical disease factors or disease modifying treatments, but is consistently associated with depressed mood, impaired school performance and lower quality of life, and may also be linked to impaired performance on some neurocognitive tasks.⁵

Despite its impact, fatigue in paediatric MS is poorly understood and under-researched. In adult MS, however, a wider body of evidence has supported a cognitive behavioural model of fatigue, which proposes that biological disease factors may initially trigger fatigue, which is then perpetuated by the interaction between biological factors and patients' cognitive, behavioural, and emotional responses.⁷ Cognitive factors such as perceiving fatigue as an uncontrollable, unpredictable symptom, and behavioural factors such as over-exertion and excessive rest have been associated with increased fatigue. Fatigue can also be exacerbated by its reciprocal relationship with emotional factors such as depression and anxiety.⁸⁻¹⁰ Similar models have been developed in adolescent Chronic Fatigue Syndrome (CFS) where fatigue is the dominant symptom of the illness.¹¹ These models have informed the development of effective cognitive behavioural therapy based interventions for fatigue,¹²⁻¹⁵ yet similar research in caMS is lacking.

A greater body of research on paediatric MS fatigue is warranted to inform the development of future interventions in this area.

Although cognitive behavioural models may provide some insight into fatigue-related factors in adult MS and adolescent CFS, we do not yet know how such factors manifest in paediatric MS. Previous studies have provided preliminary insights into the impact of fatigue in paediatric MS and its relationship to emotional factors such as anxiety and depression,^{4,16,17} but cognitive and behavioural responses to fatigue have not yet been explored. Until fatigue in caMS is better understood, factors to target in interventions or measure in future quantitative research on fatigue remain unclear. Qualitative research is paramount to developing an understanding of fatigue that is grounded in patient experiences, and generating future research questions that directly address factors that are associated with fatigue. As parental factors such as maternal distress have previously been associated with a higher risk of CFS in children, interviewing parents is also key to developing a comprehensive picture of fatigue.^{18,19} No qualitative studies have previously explored fatigue in paediatric MS. Thus, the objectives of this study were (a) to explore experiences of fatigue in caMS and their parents, and (b) to gain insight into how caMS and parents respond to and manage fatigue.

2. METHODS

2.1 Design

Qualitative methods were employed using in-depth semi-structured interviews. Purposive sampling was used to obtain a diverse sample of boys and girls of different ages.

2.2 Participants

Participants were included if they were aged 6-18, had MS, and self-reported significant fatigue. Parents of caMS with fatigue were also included. Non-English speakers were excluded from the study.

2.3 Procedure

Ethical approval was granted by the West London & GTAC NHS Research Ethics Committee (REC REF: 15/LO/0091). Participants were recruited through NHS specialist paediatric neurology clinics, MS charities and online support fora. Informed consent was obtained from all participants before interview. Separate interviews were conducted with caMS and parents by S.C. in person or via telephone, and lasted 20-45 minutes. Interviews followed semi-structured schedules (tables 1 and 2), which were reviewed by the research team prior to commencing interviews and were consistently followed throughout the study. Interviewing ceased when data saturation was achieved, indicating the point at which no new themes were observed in the data.²⁰

[INSERT TABLE 1]

2.4 Analysis

Interviews were audio-recorded and transcribed verbatim. Inductive thematic analysis followed Braun and Clarke's guidelines,²¹ incorporating elements of grounded theory to achieve a richer interpretation of the data that was grounded in participant experiences, rather than focusing on a purely narrative approach.²² Recordings were repeatedly listened to and transcripts repeatedly read to achieve familiarization with data. Transcripts were initially coded line by line, relying heavily on the verbatim. Similar codes were then grouped under the same label or a new label as new codes were identified. Themes were developed by collating similar codes into broader

themes and subthemes. Emerging themes were discussed amongst the research team throughout analysis, and constantly compared to the dataset to ensure accuracy.

[INSERT TABLE 2]

3. RESULTS

3.1 Participants

Fifteen caMS and thirteen of their parents were interviewed. Participant characteristics are outlined in table 3.

[INSERT TABLE 3]

3.2 Themes

[INSERT TABLE 4]

Five key themes were identified (table 4). Three themes, “the lived experience of fatigue and impact on daily activities”, “uncontrollability and uncertainty of fatigue” and “finding a balance” were common to both caMS and parents, while the theme “concern” was unique to parents and “social support and disclosure” was unique to caMS. An additional overarching theme “emotional reactions to fatigue and its impact” addressed participants’ emotional responses to fatigue (figure 1). Feelings of anger, sadness, fear and frustration were pertinent to all other themes, so will be discussed within their context as they are subsequently described. Additional quotes are shown in table 5.

[INSERT FIGURE 1]

3.2.1 The Lived Experience of Fatigue and Impact on Daily Activities

CaMS experienced physically and mentally exhausting fatigue. Physical fatigue was described as a complete loss of energy and heaviness that felt “*like wearing a giant sandbag*”, whilst cognitively, fatigue felt “*like looking through a haze*”. Fatigue varied in severity, but consistently interfered with school, social and family life. Both CaMS and parents emphasised that fatigue was particularly problematic in school. In addition to reduced attendance, they felt that learning was disrupted by fatigue-related impairments in memory, concentration, and difficulty maintaining attention throughout a full school-day. Although stressful, CaMS often pushed themselves to avoid falling behind on lessons.

“It’s hard to concentrate because I’m just so tired that when the teacher speaks, it goes in one ear and out the other. I have to push myself to work because I don’t want to fail.” (CaMS-115)

Parents stressed the impact of fatigue on family life, as they felt they had to “*plan everything around it*”. CaMS expressed greater concern about their social lives, and sometimes felt upset and angry when their recreational activities were limited by fatigue.

“It makes me feel like I’m missing out on life.” (CaMS-110)

“It makes me angry when I can’t do things.” (CaMS-108)

Additionally, CaMS discussed poor sleep patterns because of fatigue. Following a full day of school or activities, they would be completely “*wiped out*” and take long naps to recover, which would then disrupt night-time sleeping. Even when CaMS managed to get adequate sleep, they reported feeling unrested and having poorer sleep quality.

“It does mess up my sleep pattern because when I get back from school or somewhere, I usually just fall asleep.” (CaMS-101)

“The quality of sleep isn’t as great as it could be.” (CaMS-106)

3.2.2 Uncontrollability and Uncertainty of Fatigue

3.2.2.1 Uncontrollability of Fatigue

Participants perceived fatigue as an uncontrollable symptom. Some caMS expressed frustration at their lack of control over fatigue and felt disheartened after numerous failed attempts to manage fatigue. Parents also struggled with this and wished that they could help to alleviate their child’s symptoms.

“I’ve tried diet, exercise, rest – nothing works. It’s a fact of life at this point... it’s frustrating and it sometimes affects me emotionally, like I get down.” (CaMS-111)

“Sometimes you feel helpless. I try to make it as easy for her as I can. There’s not much else I can do.” (Parent-209)

Contrastingly, others felt that fatigue was just another part of MS that couldn’t be changed, so accepted it as part of life rather than attempting to alleviate it in any way.

“We’ve sort of taken it for granted... it’s just become acceptable, rather than trying to find another way of not feeling so tired.” (Parent-201)

3.2.2.2 Uncertainty and lack of knowledge

Parents spoke about their uncertainty about causes of fatigue. Parents of teenagers sometimes found it difficult to decipher between normal teenage fatigue and MS-related fatigue,

and all parents desired better knowledge of fatigue triggers. Parents' uncertainty appeared to be exacerbated by a lack of available information on paediatric MS, which hindered their ability to optimally manage their children's fatigue. Parents stressed a need for tailored information and guidance on managing fatigue.

“Is it too much activity, sleeping, diet? Is it the colour of the sky? It's just so completely random.” (Parent-213)

“There's not a lot of information to give out on paediatric MS... So that's challenging.” (Parent-205)

3.2.3 Finding a balance

CaMS struggled to find a balance between doing too much and resting too much. Some were reluctant to slow down when fatigued as they didn't want to allow fatigue to interfere with their lives.

“Normally I can power through. I'd just do what I normally do and ignore feeling tired.” (CaMS-102)

However, caMS often suffered after pushing too much as they would be even more exhausted afterwards, and less able to function.

“If she does too much, she pays the price for it.” (Parent-210)

Others lost motivation to do things when fatigued, as they felt there was no point if they were unable to achieve as much as they could when not fatigued. Resting was often seen as an easier option.

“I’d just be so tired it’s like, well why bother even going if I don’t even have the energy to do it.” (CaMS-111)

Some caMS discussed the process of learning to balance activity, and finding the line between too little and too much activity took time.

“It’s sort of just a trials and errors thing, where you have to push yourself a little too far every now and then so you know how far is too far.”
(CaMS-106)

Parents also struggled to find the right balance when managing their child’s fatigue. They feared that too much activity could cause a relapse, so were often inclined to be cautious and encourage caMS to rest.

“She’ll push really hard and that seems to make her MS flare. It’s hard to get her to push pause on those activities... that’s a battle we fight.”
(Parent-205)

However, parents were less inclined to encourage caMS to do activities when they were perhaps resting too much. Some felt that their children had already lost so much control when diagnosed with MS that they wanted to give them as much freedom as possible to manage fatigue whatever way they liked.

“I just let him rest and do as little as possible. If he wants to just sit and watch TV all day and do absolutely nothing, that’s the way it’s got to be until he’s got his energy back.” (Parent-203)

3.2.4 Concern

3.2.4.1 Concern about well-being

Parents, but not children, aired concerns about the adverse effect of fatigue on mental health. They worried that caMS were reluctant to talk about how much fatigue really impacted their mood, either to avoid upsetting parents, or to maintain privacy.

“She doesn’t really go on much about it, but I can just see how much it affects her.” (Parent-202)

Parents also raised concerns about their children feeling left out or inferior to others when their activities were limited by fatigue.

“...he’s realizing there are things that MS is stopping him from doing and that’s really tough for him. I don’t want him to think he’s not as great as everybody else because he is. It’s just this horrible condition he’s got to grow up with.” (Parent-203)

3.2.4.2 Concern about future

Parents expressed concern about their children’s future ability to manage fatigue. They felt that fatigue was easier to accommodate now when caMS have fewer responsibilities, and worried that it would become increasingly problematic when managing an independent adult life.

“What worries me is the future, like for work when it’s more of a 9 to 5, five days a week... and then having a life where you’re working and juggling bills. How would he cope with that?” (Parent-201)

Finally, parents worried that caMS would miss out on some life experiences because of fatigue and regretted that they had to experience such a debilitating symptom at a young age.

“...you can just see that she’s going to miss out on a lot of things. So yeah, I’d love to just say “Give me the illness. I’ve lived my life”. There’s nothing you can do.” (Parent-202)

3.2.5 Social Support & Disclosure

CaMS differed in their decisions to disclose MS and fatigue to others. Those who disclosed fatigue largely experienced positive responses, and felt that friends reacted in a supportive and understanding manner.

“I would just say to my friends, “do you guys mind if I take a rest for a little while, I feel tired” and they would perfectly understand why.” (CaMS-103)

In school, disclosure benefitted caMS as teachers responded by helping to manage schoolwork. In some cases, provisions were made to adjust individual timetables for caMS to enable them to optimally manage their time and workload:

“School is really supportive of me.” (CaMS-109)

“I’ve had my timetable adjusted.” (CaMS-112)

In contrast, some caMS chose not to disclose fatigue as they were concerned about others’ perceptions, and didn’t want to be treated differently to their peers. Choosing not to disclose fatigue sometimes meant that caMS became even more exhausted by over-exerting themselves to keep up with friends.

“I hate drawing attention to myself... so I just have to just get on with it.”

(CaMS-109)

This was often quite a lonely experience for caMS, as they felt that their friends wouldn't enjoy spending time with them as much when they were less able to participate in social events.

“Nobody understands what it's really like for me.” (CaMS-113)

“My friends just think I'm boring and that I find them boring, but I don't.

It makes me sad.” (CaMS-107)

When teachers were unaware of fatigue, caMS also faced the challenge of falling behind on schoolwork without having adequate support in place to help them to catch up. CaMS expressed concern that their fatigue was perceived as laziness.

“Teachers just think I'm being lazy and stuff. I miss out on my schoolwork and it's just hard. It's frustrating.” (CaMS-107)

Notably, even when others were understanding of fatigue, caMS expressed feeling guilty when they felt that their fatigue placed limitations on their friends.

“It makes me feel guilty sometimes because it's like I'm making them stop doing what they want to do to accommodate me. If it wasn't for my MS or for me, that wouldn't happen.” (CaMS-106)

[INSERT TABLE 5]

4. DISCUSSION

This study provides a novel insight into the experience of fatigue in paediatric MS from the perspective of both caMS and parents. Our findings highlight the extremely disabling nature of fatigue, and its impact on daily functioning and psychological well-being. Fatigue was considered a debilitating, uncontrollable and uncertain symptom, which evoked deep concern in parents for their children's well-being and future. Interestingly, although fatigue undoubtedly impacted their mood at times, caMS did not share their parents' level of concern about the issues. Rather, they were more focused on day-to-day functioning in school and with friends, and considered fatigue to be "annoying", "frustrating" or "a nuisance", rather than a deeply concerning symptom. However, the decision to disclose or not disclose fatigue seemed to have a greater impact on caMS, as those who disclosed fatigue in school and social contexts appeared to benefit from support in helping them manage fatigue.

Some themes echoed findings of prior research on adult MS and adolescent CFS, particularly those addressing beliefs about symptoms and behavioural responses to fatigue. For example, perceiving fatigue to be uncontrollable has previously been linked to higher fatigue levels.^{8,9} However, as noted by parents, uncertainty about optimal fatigue management was heightened by the lack of available information on paediatric MS, highlighting the need to address this gap in clinical practice. CaMS also discussed patterns of over-exertion and excessive rest in response to fatigue, both of which have been linked to greater fatigue levels in other populations.^{7,23} Educating caMS and parents about behavioural patterns that contribute to fatigue, and helping them to establish a balanced routine and sleeping pattern may comprise an important part of a future intervention to alleviate fatigue.

The present study also identified some factors unique to paediatric MS, such as the issue of disclosure and social support. CaMS differed in the extent to which they shared their

experience of MS and fatigue, partly because they wanted to live a normal life in the same way as their peers, and partly because they worried about others' perceptions and understanding of fatigue. Indeed, one of the barriers to disclosing fatigue in school was caMS feeling that teachers confused fatigue with laziness, and wouldn't understand the complexity of MS and fatigue if they attempted to explain it. As caMS also felt that fatigue interfered with their cognitive functioning and school performance, a lack of awareness of fatigue in schools meant that they sometimes missed out on much-needed support. Future interventions would benefit from incorporating information and guidance for teachers on how best to support caMS with fatigue.

Considering the parallels drawn above between fatigue in paediatric MS, adult MS and adolescent CFS, it may be useful to examine some of the well-established contributing factors of fatigue in adult MS and adolescent CFS within a paediatric MS context. It may also be interesting to compare fatigue in caMS to fatigue in children with other chronic illnesses to explore experiences of fatigue across different conditions. Future quantitative studies in this area may enhance our understanding of fatigue in paediatric MS and facilitate the development of tailored interventions for fatigue, incorporating elements specific to caMS.

As cognitive behavioural therapy (CBT) has been effective in reducing fatigue in adult MS^{13,24} and adolescent CFS,^{12,15,23,25} it may be useful to investigate its efficacy within a paediatric MS context. CBT in this context helps patients first to understand how their thoughts and behaviours may contribute to fatigue, and subsequently to adapt their cognitive and behavioural responses that may perpetuate or exacerbate fatigue. CBT based interventions may be effective in targeting factors discussed by parents and caMS in the present study, such as perceiving fatigue as an uncertain, uncontrollable symptom, and engaging in behavioural patterns of over-exertion and excessive rest. In adolescent CFS, adopting a family based

approach to CBT has been effective in helping families to address beliefs about fatigue, such as seeing fatigue as a sign of damage, and to establish a balanced sleeping and activity pattern, rather than engaging in “all or nothing” behaviour.²³ As social support and disclosure was a key theme for caMS, helping caMS with challenges relating to social support and disclosure should also be incorporated into future interventions. As caregivers are likely heavily involved in helping caMS to manage their symptoms, involving caMS, families and teachers in future fatigue management interventions is paramount. Finally, as attending further appointments may be burdensome for caMS with fatigue, self-management treatments may provide an accessible and cost-effective approach to CBT.

4.1 Limitations

As interviews were conducted at one time point, findings cannot account for changes in fatigue or its management over time at various stages in the disease course. Exclusion of non-English speakers from the study may limit its cross-cultural applicability.

5. CONCLUSIONS

Fatigue is a disruptive and debilitating symptom of paediatric MS, which substantially impedes functioning and impacts on the psychological well-being of caMS and parents. Fatigue is perceived as an uncertain, uncontrollable symptom for which adequate information and treatment strategies are lacking. Future interventions should aim to improve fatigue self-management strategies to enable caMS to fully engage with school and recreational activities, and should provide accessible, tailored psycho-educational information to caMS, families and teachers. Interventions should be informed by further research which expands upon this study and quantitatively explores fatigue-related factors in a larger sample.

Acknowledgements

We would like to sincerely thank the young people and parents who took part in this research. We would also like to thank Dr Evangeline Wassmer at Birmingham Children's Hospital and Dr Ming Lim at the Evelina London Children's Hospital for their input with recruitment. This study was funded by the MS Society UK and supported by the National Institute for Health Research Biomedical Research Centre at Great Ormond Street Hospital for Children NHS Foundation Trust and University College London.

References

1. Absoud M, Lim M, Chong WK, et al. Paediatric acquired demyelinating syndromes: incidence, clinical and magnetic resonance imaging features. *Mult Scler.* 2013; 19(1): 76-86
2. Compston A, Coles A. Multiple sclerosis. *Lancet.* 2008; 372(9648): 1502-1517
3. MacAllister WS, Christodoulou C, Milazzo M, et al. Paediatric multiple sclerosis: what we know and where are we headed? *Child Neuropsychol.* 2013; 19(1): 1-22
4. Goretti B, Portaccio B, Ghezzi A, et al. Fatigue and its relationships with cognitive functioning and depression in paediatric multiple sclerosis. *Mult Scler.* 2012; 18(3): 329-334
5. Carroll S, Chalder T, Hemingway C, Heyman I, Moss-Morris R. Understanding fatigue in paediatric multiple sclerosis: a systematic review of clinical and psychosocial factors. *Dev Med Child Neurol.* 2016; 58(3): 229-239
6. Braley TJ, Chervin RD. Fatigue in multiple sclerosis: mechanisms, evaluation, and treatment. *Sleep.* 2010; 33(8): 1061-1067
7. van Kessel K, Moss-Morris R. Understanding multiple sclerosis fatigue: a synthesis of biological and psychological factors. *J Psychosom Res.* 2006; 61(5): 583-585
8. Knoop H, van Kessel K, Moss-Morris R. Which cognitions and behaviours mediate the positive effect of cognitive behavioural therapy on fatigue in patients with multiple sclerosis? *Psychol Med.* 2012; 42(1): 205-213
9. Jopson NM, Moss-Morris R. The role of illness severity and illness representations in adjusting to multiple sclerosis. *J Psychosom Res.* 2003. 54(6): 503-511
10. Skerrett TN, Moss-Morris R. Fatigue and social impairment in multiple sclerosis: the role of patients' cognitive and behavioral responses to their symptoms. *J Psychosom Res.* 2006; 61(5): 587-593
11. Lievesley K, Rimes KA, Chalder T. A review of the predisposing, precipitating and perpetuating factors in Chronic Fatigue Syndrome in children and adolescents. *Clin Psychol Rev.* 2014; 34(3): 233-248
12. Knight SJ, Scheinberg A, Harvey AR. Interventions in paediatric chronic fatigue syndrome/myalgic encephalomyelitis: a systematic review. *J Adolesc Health.* 2013; 53(2): 154-65
13. Moss-Morris R, McCrone P, Yardley L, van Kessel K, Wills G, Dennison L. A pilot randomised controlled trial of an Internet-based cognitive behavioural therapy self-management programme (MS Invigor8) for multiple sclerosis fatigue. *Behav Res Ther.* 2012; 50(6): 415-421
14. van Kessel K, Moss-Morris R, Willoughby E, Chalder T, Johnson MH, Robinson E. A randomized controlled trial of cognitive behavior therapy for multiple sclerosis fatigue. *Psychosom Med.* 2008; 70(2): 205-213
15. Knoop H, Stulemeijer M, de Jong LWAM, Fiselier TJW, Bleijenberg G. Efficacy of cognitive behavioral therapy for adolescents with chronic fatigue syndrome: Long-term follow-up of a randomized controlled trial. *Pediatrics.* 2008; 121(3): e619-e625

16. Ketelslegers IA, Catsman-Berrevoets CE, Boon M, et al. Fatigue and depression in children with multiple sclerosis and monophasic variants. *Eur J Paediatr Neurol*. 2010; 14(4): 320-325
17. MacAllister WS, Christodoulou C, Troxell R, et al. Fatigue and quality of life in pediatric multiple sclerosis. *Mult Scler*. 2009; 15(12): 1502-1508
18. Rimes KA, Goodman R, Hotopf M, Wessely S, Meltzer H, Chalder T. Incidence, prognosis, and risk factors for fatigue and chronic fatigue syndrome in adolescents: a prospective community study. *Paediatrics*. 2007; 119(3): 603-609
19. Collin SM, Tilling K, Joinson C, et al. Maternal and childhood psychological factors predict chronic disabling fatigue at age 13 years. *J Adolesc Health*. 2015; 56(2):181-187
20. Mason M. Sample Size and Saturation in PhD Studies Using Qualitative Interviews. *Forum Qualitative Sozialforschung / Forum: Qualitative Social Research*. 2010; 11(3).
21. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol*. 2006; 3(2): 77-101
22. Glaser BG, Strauss AL. The discovery of grounded theory: Strategies for qualitative research: Transaction Publishers; 2009
23. Chalder T, Deary V, Husain K, Walwyn R. Family-focused cognitive behaviour therapy versus psycho-education for chronic fatigue syndrome in 11-to 18-year-olds: a randomized controlled treatment trial. *Psychol Med*. 2010; 40(8): 1269-1279
24. van Kessel K, Moss-Morris R, Willoughby E, Chalder T, Johnson MH, Robinson E. A randomized controlled trial of cognitive behavior therapy for multiple sclerosis fatigue. *Psychosom Med*. 2008; 70(2): 205-213
25. Stulemeijer M, de Jong LW, Fiselier TJ, Hoogveld SW, Bleijenberg G. Cognitive behaviour therapy for adolescents with chronic fatigue syndrome: randomised controlled trial. *BMJ*. 2004; 330(7481): 14

Table 1. Interview Questions and Related Probes (CaMS Interview)

Key Questions	Probes
Tell me about your fatigue.	How does it feel physically/mentally? How often do you feel fatigue? Understanding of causes/triggers
How does fatigue affect the things you do?	School life/school work Relationships with friends/family Activities & Social Life How does fatigue affect your sleep?
Can you tell me about some of the things you do when you feel fatigue?	Things that are helpful Things that are unhelpful Social support
Is there anything else you'd like to say about your fatigue?	

Table 2. Interview Questions and Related Probes (Parent Interview)

Key Questions	Probes
Tell me about your child's fatigue?	How does it affect him/her physically? How does it affect school/social/family life? How do you tell when your child is fatigued? How do you understand your child's fatigue? Causes & triggers
How do you respond to your child's fatigue?	How do you feel when he/she is fatigued? How does it affect your life? What do you find helpful/unhelpful in managing your child's fatigue?
What does your child do when he/she is fatigued?	Examples of things he/she does when fatigued What does he/she find helpful/unhelpful?
When you are feeling fatigued, what do you do?	What triggers your fatigue? What do you find helpful/unhelpful in managing your fatigue?
Is there anything else you'd like to say about your experience of your child's fatigue?	

Table 3. Demographic Characteristics of Participants

Variable	caMS (<i>n</i> = 15)	Parents (<i>n</i> = 13)
Age <i>M</i> (range)	15.2 (9-18)	46.8 (32-52)
Gender		
Female <i>n</i> (%)	8 (53)	11 (85)
Male <i>n</i> (%)	7 (47)	2 (15)
Relationship to Child		
Mother <i>n</i> (%)		11 (85)
Father <i>n</i> (%)		2 (15)
Nationality		
British <i>n</i> (%)	8 (53)	8 (62)
American <i>n</i> (%)	5 (33)	5 (38)
Belgian <i>n</i> (%)	1 (7)	
Russian <i>n</i> (%)	1 (7)	
Ethnic Group		
White <i>n</i> (%)	11 (73)	12 (92)
Black <i>n</i> (%)	2 (13)	
Mixed <i>n</i> (%)	1 (7)	
Hispanic <i>n</i> (%)	1 (7)	1 (8)
Age at MS Onset <i>M</i> (range)	10.6 (4-17)	
Disease Duration Years <i>M</i> (range)	2.9 (1-11)	
Relapses in Past Year <i>M</i> (range)	1.8 (0-5)	

Table 4. Summary of Main Themes and Subthemes Identified in the Data

Themes	Subthemes
Emotional Reactions to Fatigue and its Impact	
The Lived Experience of Fatigue and Impact on Daily Activities	
Uncontrollability and Unpredictability of Fatigue	Uncontrollability of fatigue Uncertainty and lack of knowledge
Finding a Balance	
Concern	Concern about well-being Concern about future
Social Support and Disclosure	

Table 5. Additional Quotes from CaMS and Parent Interviews

Theme	CaMS	Parents
The Lived Experience of Fatigue and Impact on Daily Activities		
	<p>“I used to be really sporty, but as the tiredness came in I struggled more and more with PE and even doing anything outside school which I enjoyed. So I kind of had to drop it all.”</p> <p>“My life’s been arranged around fatigue.”</p> <p>“I just feel really stressed and like I can’t do it (schoolwork). It just makes me feel like I can’t do anything really.”</p>	<p>“Fatigue has definitely changed our lifestyle – mostly keeping the schedule from getting too crazy.”</p> <p>“We have to put the needs of his tiredness first.”</p> <p>“She’ll lose half of what the teacher’s saying. She’ll just struggle to keep up with what she’s learned for the day.”</p>
Uncontrollability & Uncertainty of Fatigue		
<i>Uncontrollability of Fatigue</i>	<p>“Well I know that until there’s like a cure for MS, the fatigue is going to be there.”</p> <p>“I thought it would go away, but now I know that it won’t.”</p>	<p>“I suppose with that kind of fatigue, it’s uncontrollable tiredness.”</p> <p>“I don’t think it will ever get better.”</p>
<i>Uncertainty & lack of knowledge</i>	<p>“It just kind of came on and there wasn’t really anything I could think of that could have caused it.”</p>	<p>“Are there things that I should be doing? I don’t know what the answers are. I just don’t.”</p>
Finding a balance		
	<p>“Sometimes I do overwork my body, like try to do a million things in the day and not take a break.”</p>	<p>“He’s got to miss out on some things and there’s nothing else we can do, because if he doesn’t rest he will go into a relapse. Obviously it’s dangerous, so he needs to shut down and take some time</p>

out.”

“The problem is trying to do too much at once and then you stress and then you just sleep to get rid of the stress.”

“I don’t pressure her with her exams or anything because I just think, you know, you’ve gone through so much over the last few years. Just do what you can and that’s it.”

Concern

Concern about well-being

“They don’t want to be spending half the day thinking “I can’t wait to get back to bed” and the other half thinking “I’ve got to get out of bed” because life’s passing us by and we’re just tired all the time.”

“I think that the fatigue has made her depressed, on top of her diagnosis. I think that it has had a big impact on her mental health.”

Concern about future

“It’s really hard. He’s missing out on his whole life.”

“People are going to be going out and doing things and not coming back ‘til late, and I just can’t see that happening.”

Social Support and Disclosure

“They (friends) know I’m fatigued and they do understand. They know about the condition and the limitations and they’re fine with it.”

“I don’t want to talk to anyone about it.”

“I feel like they think I get special consideration and it’s embarrassing. Most of my friends don’t even know I have MS.”

“If I’m with friends, I feel like I’m holding them back.”

“When I play football, I get tired very quickly and I still want to play but I physically can’t play and I’m putting all

my team down.”

“It just really pulls you down because you feel bad for being moody, but you can’t help it because you’re just so tired. It does make you feel awful to know you’re being rude when you shouldn’t be.”

Figure Legends

Figure 1. Thematic map showing themes common to primary caregivers and caMS, and themes specific to each group

