What matters to children with CFS/ME? A conceptual model as the first stage in developing a PROM

Roxanne Parslow, Aarti Patel, Lucy Beasant, Kirstie Haywood, Debbie Johnson, Esther Crawley

ABSTRACT

Background Paediatric chronic fatigue syndrome (CFS)/myalgic encephalomyelitis (ME) is relatively common and disabling. Research is hampered because current patient-reported outcome measures (PROMs) do not capture outcomes that are important to children with CFS/ME. Aim The aim of this study was to explore the aspects of life and health outcomes that matter to children with CFS/ME. Methods Twenty-five children with CFS/ME were interviewed (11 males, 14 females; mean age 12.9 years (SD 2.2), range 8–17). Twelve were trial participants interviewed during the trial and 13 were recruited as part of a follow-up qualitative study. Parents were present in 19 interviews with their children. Three mothers participated in a focus group. All the interviews and the focus group were audio-recorded and transcribed. Data were analysed thematically using techniques of constant comparison. NVivo was used to structure and categorise data in a systematic way. Results Children identified four key themes (health outcome domains): ‘symptoms’ that fluctuated, which caused an unpredictable reduction in both ‘physical activity’ and ‘social participation’ all of which impacted on ‘emotional well-being’. These domains were influenced by both ‘management’ and ‘contextual factors’, which could be positive and negative. The relationship between healthcare and school was considered pivotal. Conclusions Children’s descriptions helped to inform a conceptual model that is necessary to develop a new paediatric CFS/ME PROM. Doctors need to be aware of how children conceptualise CFS/ME; the relationship between healthcare and school is fundamental to ameliorate the impact of CFS/ME.

Trial registration number ISRCTN81456207.

INTRODUCTION

Chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) is characterised by severe disabling fatigue\(^1\) with additional symptoms including musculoskeletal pain, sleep disturbance, cognitive dysfunction, headaches, dizziness and sweats.\(^2\) Paediatric CFS/ME is relatively common with a prevalence of between 0.4% and 2.4%\(^3\)–\(^6\) in population studies and between 0.06% and 0.1%\(^7\)–\(^9\) in studies based in hospital settings. It is disabling\(^10\)–\(^11\) with a negative impact on quality of life.\(^12\)–\(^16\)

Despite this, research is hampered by the lack of high-quality patient-reported outcome measures (PROMs) for adults\(^17\) and children.\(^18\)

Well-developed PROMs enable clinicians and researchers to collect patient-centred evidence on outcome.\(^19\)–\(^20\) PROM development starts with a patient-derived conceptual framework that defines the key outcomes to be measured.\(^21\)–\(^23\) Qualitative exploration of the outcomes that really matter to children with CFS/ME is limited.\(^18\) This paper describes the combined results from two studies describing the aspects of life and health outcomes that really matter to children with CFS/ME.

METHODS

Participants

Children with CFS/ME were recruited into two separate studies through a large regional specialist CFS/ME service in South West England by their specialist clinician. Children were eligible if they were diagnosed with CFS/ME using the National Institute for Health and Care Excellence criteria,\(^4\)–\(^6\) mildly to moderately affected (not housebound) and able to understand the patient information sheets. Consecutive children were approached to take part in the Specialist Medical Intervention and Lightning Evaluation (SMILE) trial from September 2010 to December 2011 and PROMS study from...
June to August 2012 in assessment and follow-up clinics. If eligible, they were informed about the relevant study by their specialist clinician and given patient information sheets. Consent was obtained from the family to be contacted by a researcher to arrange an interview.

**SMILE trial**
Children (aged 12–18 years) and their parents were interviewed as part of the SMILE trial about whether PROMs used were suitable to assess the impact of CFS/ME and measure important outcomes.24 25 Children and their mothers were interviewed at three possible time points using a checklist of topics24 25 usually at the participants’ homes. Purposive sampling ensured interviews included a range of informants, in terms of age, sex and families from both intervention arms (maximum variation sampling).24 25

**PROMs study**
We then extended this work in a further qualitative study recruiting participants from a wider age range (aged 8–18 years) to explore the key health outcomes important in paediatric CFS/ME (PROMs study). Semistructured interviews in participants’ homes were used to explore how children with CFS/ME describe the impact of CFS/ME. A topic guide (see online supplementary appendix 1) informed by previous reviews17 26 was sent in advance to children and their parents before the interview. We conducted one focus group for parents (without their child) to discuss aspects of quality of life that they felt were important.

**Data collection and analysis**

**Qualitative data**
For both studies, interviews were recorded and transcribed verbatim. Data analysis was an ongoing and iterative process. Interviews were stopped when we reached data saturation. We compared data collected from both studies for systematic differences in content before combining the data. Data were analysed thematically using techniques of constant comparison.27 Data items were systematically assigned codes using NVivo.28 Similar codes were cross-referenced and emerging themes were discussed with members of the research team to improve reliability. Two members of the research team independently analysed approximately 10% of the data to compare coding and enhance reliability. Descriptive accounts were produced, and theoretical explanations for behaviours, opinions and decisions developed.

**Quantitative data**
The service currently collects information routinely at assessment about fatigue, physical functioning, pain, anxiety and depression from the following inventories: Chalder Fatigue Scale,27 36-item short form (SF-36) physical function subscale,30 EQ-5D,31 visual analogue pain rating scale, Hospital Anxiety and Depression Scale32 and the Spence Children’s Anxiety Scale.33

**RESULTS**

**Participants**
Twenty-five children with CFS/ME were interviewed (11 males, 14 females; mean age 12.9 years (SD 2.2), range 8–17). Twelve (of the 13 approached) were interviewed during the SMILE trial and 13 (of the 17 approached) were recruited as part of the PROMs study. Parents were present with their child during 19/25 interviews. Twenty-five mothers were interviewed.

Table 1 compares those interviewed with those assessed in the specialist CFS/ME service in the same time period. Participants in the PROMs study were slightly younger. Participants were also less anxious and less likely to be female because we prospectively recruited male patients (38.5% compared with 72.6%, p=0.01). Initial clinical assessment data were available for 5/7 of the children who declined to participate. Analysis of these data shows that children who declined were broadly similar to those who participated in terms of the characteristics shown in table 1: mean (SD) age 14.4 (1.5) years; 80% (4/5) female; median (IQR) time to assessment 6 (6–10) months; median (IQR) Chalder Fatigue score 23 (22–25); median (IQR) SF-36 physical function 65 (60–80).

**Living with CFS/ME—‘feeling ill’**
We propose a conceptual model (figure 1) of being unwell with CFS/ME that suggests interactions between four key themes (health outcome domains) that contribute to how children understand their illness and ‘feeling ill’: ‘symptoms’, ‘physical activity’, ‘social participation’ and ‘emotional well-being’. These were influenced by two additional themes: ‘management’ and ‘contextual factors’. Children described a linear relationship between symptoms, impact on activity, participation and emotional well-being. When symptoms were worse, children felt this had a direct impact on their activity, “When I am worse it is mostly just resting”, and mood, “As I feel crash down I become a little bit less positive”. Children also described mood effects as a result of a lack of participation, “I could have been doing better and it is very frustrating”.

**Symptoms**
Most children described a range of symptoms: aches and pain, sleep disturbances, a lack of energy, tiredness and memory and concentration problems. Although the individual experience of symptoms differed, some described a collection of symptoms, “everything hurts”.

My legs felt like jelly, which was the cause of the … and um I got a sore throat, … um I got ear pain, headache, tummy ache, everything. (Female)

Several children described the cognitive impact; a reduced capacity to remember things, concentrate and a ‘brain fog’. This was often described as a ‘loss’ in ability to participate in academic activities:

I was just good at memorising stuff and now it’s completely gone. (Female)

**Fluctuation in symptoms**
Many children described the unpredictable fluctuation in symptoms and an associated lack of control, often describing a boom and bust cycle. This was particularly linked to their perceived levels of energy “ever changing energy level” and impacted their mood and confidence in returning to usual activities:

I’d like to do some certain sports again, but I am just worried about the energy levels…. (Male)

**Physical activity**
All participants described a reduction in physical activity and children often reflected on sport or hobbies that they had to give up, “used to be very sporty and active” (male). In some cases, symptoms stopped children doing activities at the time, in others, restricting activity led to a negative cycle of more and more limitation:
Children and their parents (box 1) reported anxiety caused by not keeping up with schoolwork. In some cases, children dropped subjects to reduce stress. In many cases, children were frustrated at not meeting their full potential:

I get really frustrated because then I think if I was there full time I could have been in more lessons and I could have been doing better. (Female)

**Impact on friendships**

Despite parental encouragement, being unwell with CFS/ME affected children’s ability and motivation to spend time with friends, often limited due to symptoms and reduced activities:

It was hard because like my friends were going out and leaving me at home because I just couldn’t do it. (Female)

**Impact on home life and family**

A few children reported that CFS/ME affected their ability to spend time with their relatives or siblings, going on family outings or on holiday. Parents emphasised the impact it had on

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Table 1 Comparison of characteristics of children at initial clinical assessment who were interviewed as part of the SMILE trial and PROMs study compared with children who were assessed and treated by the specialist service during the same period (2010–2012)

<table>
<thead>
<tr>
<th>Study sample</th>
<th>CFS cohort (n=511) unless otherwise shown</th>
<th>PROMs study</th>
<th>CFS cohort (n=530) unless otherwise shown</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>SMILE trial</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age (years)‡</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>14.5 (1.5)</td>
<td>14.0 (1.6)</td>
<td>0.21</td>
</tr>
<tr>
<td>Time to assessment (months)</td>
<td>12</td>
<td>12 (6–17)</td>
<td>9</td>
</tr>
<tr>
<td>Chalder Fatigue score (0–33)</td>
<td>13</td>
<td>26 (23–28)</td>
<td>13</td>
</tr>
<tr>
<td>SF-36 physical function (0–100)</td>
<td>13</td>
<td>50 (45–55)</td>
<td>11</td>
</tr>
<tr>
<td>Anxiety (SCAS) (0–90)</td>
<td>12</td>
<td>45 (21–58)</td>
<td>10</td>
</tr>
<tr>
<td>No. of symptoms (0–14)</td>
<td>13</td>
<td>9 (7–10)</td>
<td>13</td>
</tr>
<tr>
<td>Anxiety (HADS) (0–21)</td>
<td>13</td>
<td>12 (8–15)</td>
<td>8</td>
</tr>
<tr>
<td>Depression (HADS) (0–21)</td>
<td>12</td>
<td>9 (7–11)</td>
<td>8</td>
</tr>
<tr>
<td>Visual analogue pain (0–100)</td>
<td>12</td>
<td>42 (29–57)</td>
<td>12</td>
</tr>
<tr>
<td><strong>School attendance past week</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>None</td>
<td>0 (0.0%)</td>
<td>101 (21.2%)</td>
<td>4 (30.8%)</td>
</tr>
<tr>
<td>10%</td>
<td>2 (15.4%)</td>
<td>49 (10.3%)</td>
<td>0 (0.0%)</td>
</tr>
<tr>
<td>20%</td>
<td>2 (15.4%)</td>
<td>29 (6.1%)</td>
<td>1 (7.1%)</td>
</tr>
<tr>
<td>40%</td>
<td>1 (7.7%)</td>
<td>88 (18.5%)</td>
<td>0 (0.0%)</td>
</tr>
<tr>
<td>60%</td>
<td>3 (23.1%)</td>
<td>100 (21.0%)</td>
<td>3 (23.1%)</td>
</tr>
<tr>
<td>80%</td>
<td>3 (23.1%)</td>
<td>84 (17.6%)</td>
<td>4 (30.7%)</td>
</tr>
<tr>
<td>100%</td>
<td>2 (15.4%)</td>
<td>21 (4.4%)</td>
<td>1 (7.1%)</td>
</tr>
<tr>
<td>Not applicable</td>
<td>0 (0.0%)</td>
<td>5 (1.1%)</td>
<td>0 (0.0%)</td>
</tr>
</tbody>
</table>

*Characteristics included for one child whose mother was interviewed (the child was not interviewed).

†Age for the SMILE study and the CFS cohort is age at first assessment. Age for the PROMs study is age at interview. This is because children were recruited from follow-up visits not at clinical assessment.

CFS, Chronic Fatigue Syndrome; HADS, Hospital Anxiety and Depression Scale; PROMs, patient-reported outcome measures; SCAS, Spence Children’s Anxiety Scale; SMILE, Specialist Medical Intervention and Lightning Evaluation.
family life with the family having to work around the fluctuating symptoms:

There aren’t a lot of family days out… I really miss them. (Female)

**Emotional well-being**
Children and their parents described a fear and anxiety associated with having the illness. This was often related with the fluctuation and uncertainty about the duration of periods of feeling ill:

…I was so scared of it lasting a long time again. (Female)

Activity restrictions often led directly to feelings of low mood and frustration. Parents noted that the loss of social interaction affected their child’s morale.

aaah, it’s a bit annoying and stressful, um I’m not always able to do what I want to do. (Male)

**Management (self and therapeutic)**
Children and their parents described a variety of management strategies for their CFS/ME. Some of these were described as strategies they had learnt ‘naturally’ or on their own through experience (self):

I actually kind of naturally did quite a good balance between doing enough but not doing too much. (Female)

In other cases, children described managing activity using similar descriptions as used in the specialist service (therapeutic):

…I just have to save a bit of energy up for it it’s like money you have to save a bit, spend a bit. (Male)

Parents reinforced that important activities such as having a job were often adapted to minimise the impact of the illness. Where children had managed to continue with some activity, these were sometimes prioritised if they were particularly enjoyable although often limited to prevent an increase in symptoms.

**Contextual factors**
Contextual factors, such as support and understanding from family, friends, school and healthcare providers, influenced their ability to manage their illness. Friends were often willing to undertake less physical activities indoors, “We just invite them in and watch a movie or whatever”. However, CFS/ME was often perceived as illegitimate and participants talked about the negative attitudes and comments they received.

I am more happy to be in the sanctuary of my own home where my parents understand me than people at school who don’t really have a clue. (Male)

Parents were more concerned about the impact on their child’s emotional well-being than children themselves. Some parents talked about a lack of understanding from extended family members, “Grandparents they don’t understand”. Some children reported significant support from schools enabling them to manage their symptoms:

…they have offered the best help they can get me as well, time off resting, if I am feeling ill I can go straight to the nurses’ office and lie down. (Male)

However, others reported difficulties in having their condition recognised and appropriately supported by educational authorities. Parents and children described the importance of getting a diagnosis and being understood, “Relief really to be understood”. The interaction between healthcare and educational institutions was fundamental to acknowledge the condition and
manage the impact. A mother and daughter described the interaction between a delayed diagnosis, lack of support from schools and impact on the daughter:

Mother: ...I was trying to get her off from school and they wouldn’t let me take her from school.

Child: it was really annoying.

Mother: ...she was really struggling and then it wasn’t, until she was diagnosed that they said that you can only do so many hours. (Mother and daughter)

**DISCUSSION**

CFS/ME was described as an unpredictable and fluctuating illness with a pervasive impact. Children described a linear relationship between four key outcomes: symptoms impacted on physical activity, school and social activities, which affected mood and emotional well-being. These can be ameliorated (or not) by contextual factors such as social support from family, friends, school and healthcare. The relationship between healthcare and school was considered pivotal.

**Strengths and weaknesses**

This is a large qualitative study (n=25), and participants were representative of those assessed in a specialist service. We are unable to generalise results for patients seen in primary care, those who are severely affected or younger than 8 years old. Most children chose to be interviewed with parents, which may have caused them to conceal problems. To ensure the child’s voice remained paramount, quotes from parents have been included where they have enriched their child’s viewpoints, which may mean we have not included areas that are important to parents. Eleven children were recruited from a trial. Trial participants can differ from those that do not take part in a trial. In addition, those who took part in the PROM study received the topic guide before the interview, which may have influenced their results; however, these groups and the data they generated were similar to the clinic population and to each other.

**Results in context with previous literature**

Child-derived measures specific to CFS/ME do not exist. The identified themes underpin a conceptual model of the lived experience of paediatric CFS/ME, defining the health outcomes that should be assessed in children, and enhancing the relevance and acceptability of a new CFS/ME-specific PROM. This study supports and extends existing (largely quantitative) evidence of the impact of CFS/ME on children’s schooling, emotional function and social activities, contextual factors, management and emotional well-being. These can be ameliorated (or not) by contextual factors such as social support from family, friends, school and healthcare. The relationship between healthcare and school was considered pivotal.

**Symptoms**

"She’s given up what was her best subject English, because you know, there’s an awful lot of reading and she can’t focus to read”. (Mother of female)

**Physical activity**

"She used to be active she used to run around all time". (Mother of female)

**Impact on school**

"So obviously there was apprehension from her point of view going back to school, a different peer group because she’s in a different group”. (Mother of female)

"She’s ended up dropping one of the subjects just to stop the stress". (Mother of female)

**Impact on friendships**

"It is very difficult keeping friends on board because obviously they don’t want to deal with the illness and they don’t want to be around it”. (Mother of male)

**Impact on family**

"We’ve also tried to prioritise so yes she does still go to Stagecoach on Saturday. Even if that means that the whole of the rest of the family have to work around that’s when her energy is used”. (Mother of female)

**Emotional well-being**

"I also see how by her not going out, how that affects her morale as well”. (Mother of female)

**Contextual factors**

"…she’d also got herself a job. Um, again just 12 hours a week, so 4 hours, um, stint, um, in the evenings spaced out every other day because we were thinking about her activity levels". (Mother of female)

"If friends come over for two hours it’s a really positive experience if friends stay for two and a half hours it turns into a negative experience”. (Mother of male)

**Quotes from parents to support themes**

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**Management**

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"If friends come over for two hours it’s a really positive experience if friends stay for two and a half hours it turns into a negative experience”.

**Contextual factors**

"He’s had all sort of comments whilst he’s been at school about being a skiver and all the rest of it so he’s had to deal with that”.

"I mean I… I don’t feel very supported by the school. Yeah. Um, at all really. I mean they basically just let her come and go as she likes. Um, with very little sort of thought about how she could be helped”.

**Box 1 Quotes from parents to support themes**

**Symptoms**

"She’s given up what was her best subject English, because you know, there’s an awful lot of reading and she can’t focus to read”. (Mother of female)

**Physical activity**

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"We’ve also tried to prioritise so yes she does still go to Stagecoach on Saturday. Even if that means that the whole of the rest of the family have to work around that’s when her energy is used”. (Mother of female)

**Emotional well-being**

"I also see how by her not going out, how that affects her morale as well”. (Mother of female)
Implications
Paediatricians need to be aware that from a child’s perspective CFS/ME is more than just fatigue; it impacts on activity, school attendance, socialising and mood. Contextual factors are important in how children experience symptoms and disability. Doctors need to develop strong relationships with schools. Current PROMs used for CFS/ME measure fatigue, disability and mood. A new PROM needs to collect data on symptoms, activity and socialising. Most of these themes are not currently assessed by PROMs used in paediatric CFS/ME. Future research should explore which outcomes are most important to children to include in a new PROM. The conceptual model is informative for clinicians and researchers who often have different views from children on what is important to measure in research and healthcare settings.47

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Contributors
EC developed the idea for the SMILE study, EC and KLH developed the idea for the PROMs study. AP, LB and DJ conducted the qualitative interviews and focus group. AP, LB and RP conducted the data analysis. KLH contributed to the analyses. All authors contributed to the interpretation of results and to drafting this paper.

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Disclaimer
The views expressed in this publication are those of the authors and not necessarily those of the NHS, the National Institute for Health Research or the Department of Health.

Competing interests
EC is a medical advisor for the Association for Young people with ME (AYME) and the Sussex and Kent ME/CFS society.

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