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Children's experiences of chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME): a systematic review and meta-ethnography of qualitative studies

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ABSTRACT

Objective: To synthesis the qualitative studies of children's experiences of chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME).

Design: Systematic review and meta-ethnography.

Background: CFS/ME is an important disabling illness, with uncertain cause and prognosis. As a result, children with CFS/ME can find themselves living with greater uncertainty and stigma, exacerbating the impact of the condition. There is a growing body of qualitative research in CFS/ME, yet there has been no attempt to systematically synthesis the studies involving children.

Methods: Studies exploring the experiences of children diagnosed with CFS/ME, published or unpublished, using qualitative methods were eligible. MEDLINE, EMBASE, PsycINFO and CINAHL databases were searched as well as grey literature, reference lists and contacting authors. Quality assessment was done independently using the Critical Appraisal Skills Programme (CASP) checklist. Studies were synthesised using techniques of meta-ethnography.

Results: Ten studies involving 82 children with CFS/ME aged 8–18 were included. Our synthesis describes four third-order constructs within children's experiences: (1) disruption and loss: physical, social and the self; (2) barriers to coping: suspension in uncertainty, problems with diagnosis and disbelief; (3) facilitators to coping: reducing uncertainty, credible illness narratives, diagnosis and supportive relationships and (4) hope, personal growth and recovery. CFS/ME introduces profound biographical disruption through its effects on children's ability to socialise, perform school and therefore how they see their future. Unfamiliarity of the condition, problems with diagnosis and felt stigma prevent children from forming a new illness identity. Children adopt coping strategies such as building credible explanations for their illness.

Conclusions: Physical, social, emotional and self-dimensions of life should be included when treating and measuring outcomes from healthcare in paediatric CFS/ME. There is a need for greater recognition and diagnosis of childhood CFS/ME, specialist advice on activity management and improved communication between health and education providers to help children cope with their condition.

Strengths and limitations of this study

- To the best of our knowledge, this is the first systematic review and meta-ethnography of the qualitative literature of children's experiences of chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME).
- We included all published and unpublished studies from any language to avoid bias.
- The synthesis of studies from multiple contexts identified the main dimensions of life impacted, as well as barriers and facilitators to living with childhood CFS/ME.
- The findings from this synthesis could be used to inform healthcare practice and the development of outcome measures in paediatric CFS/ME.
- The majority of studies were conducted in western countries reducing the transferability of findings.

INTRODUCTION

Paediatric chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) is common, with a prevalence between 0.06% and 2.4%[1–6] and is recognised as an important disabling condition.[7–9] Children live with severe fatigue[8] and additional symptoms, including pain, sleep disturbance, cognitive dysfunction, headaches and dizziness.[9] Functional impairment is central to CFS/ME and higher than in other chronic paediatric or emotional disorders.[10] Loss of schooling occurs,

ranging from low attendance to extended periods of absence and some children can become bedbound.[11–13] CFS/ME is a complex condition with no visible symptoms and uncertain cause and prognosis,[14 15] resulting in scepticism over its existence.[16 17] General practitioners (GPs) have been found to be reluctant to diagnose CFS/ME and to hold negative attitudes towards patients with CFS/ME.[17–20] A recent meta-synthesis identified barriers to the diagnosis and management of adults with CFS/ME, including working within the biomedical model lead to scepticism over the existence of the illness, a lack of understanding and knowledge of specialist services resulted in failure on the part of GPs to validate and diagnose a patient's illness and further frustration on the part of patients.[21] The psychosocial experience of chronic illness is argued to be as important as its aetiology,[22] therefore, children with CFS/ME can find themselves living with greater uncertainty and stigma, exacerbating the impact of the condition.

Greater awareness of the experiences and priorities of patients with CFS/ME and their families is needed to facilitate better outcomes for children with this condition. The value of qualitative research for enhancing our understanding of patients' experiences of living with chronic illness is well recognised.[23–25] Qualitative research on the illness narratives[26] of those with chronic illness has given insights into the biographical disruption caused by chronic illness,[27] and profound impact on identity.[28] Such work can be used to frame our understanding of the illness experiences of children living with CFS/ME. There is a growing body of qualitative research in CFS/ME. Yet to date, these studies remain as individual 'islands of knowledge'[29] and need to be synthesised, in order to inform improvements to healthcare provision for children with CFS/ME, including better clinical measurement of outcomes that are meaningful to children and their families.[30] The synthesis of multiple qualitative studies with small purposefully selected samples has been advocated.[31–33] This can produce a more comprehensive understanding across different contexts, enhancing the generalisability of findings.[34] Syntheses of qualitative research on adults' experiences of CFS/ME have highlighted the impact on patients' identities and the limited understanding of the condition by health professionals.[21 35 36] To date, there has been no attempt to systematically review the qualitative literature on children with CFS/ME. The aim of this study was to synthesise children's experiences of living with CFS/ME in order to identify areas of life impacted by the condition, health outcomes valued by children, barriers and facilitators for positive adjustment and implications for healthcare provision.

METHODS

We registered the protocol with PROSPERO: (http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42014009896).

Selection criteria

Studies were eligible for inclusion if they explored the experiences and/or perspectives of children (aged <18 years of age) diagnosed with CFS/ME; were English or non-English; reported published or unpublished studies from 1994 onwards[37] and used qualitative methods of data collection and analysis as either a stand-alone or part of a mixed methods study. Studies were excluded if they involved samples of patients with mixed chronic conditions and age groups (eg, >18 years of age); described outcomes reported by clinicians or parents alone; used methods such as open-ended survey responses or the full text of the paper was unobtainable.

Search and data sources

The search strategy was developed through scoping exercises and reviewed by specialist systematic reviewers. Search terms relating to the clinical topic (CFS/ME), population (children) and patient experience were combined by Boolean operators (see online supplementary appendix A). The following databases were searched from 1994 to July 2014: MEDLINE, EMBASE, PsycINFO and CINAHL. Identifying qualitative studies remains problematic due to the varied use of the term 'qualitative'[38] and less developed database indexing.[39] Therefore, no terms or filters were applied for qualitative research. Qualitative papers were extracted at the screening phase.[40] We examined reference lists and contacted first authors of all relevant studies. Key journals were individually searched using the journal's online search engine. Qualitative research is frequently published in books or theses,[41 42] therefore, electronic searches were carried out on grey literature databases for relevant conference proceedings, books, theses and dissertations. Google scholar was additionally searched.

Study selection

All titles and abstracts as well as full-text papers were double screened by three reviewers. Disagreements were resolved through discussion with two supervisory reviewers. Our search yielded 1432 studies after duplicates were removed (figure 1), 1354 were excluded through the abstract review. Of the remaining 78 studies, 68 were excluded. Exclusion reasons included CFS/ME diagnosis was unclear, adult or mixed age range population, quantitative methods, neither interview nor focus group used as the methodology, parental views only, non-research or abstract for an included study.

Critical appraisal

Quality assessment was done independently by two reviewers using the Critical Appraisal Skills Programme (CASP) checklist.[43] Each paper was scored out of 10 according to the total number of questions for which yes (or a positive answer) was obtained to give an indication of the reporting quality. Disagreements were resolved through discussion with a third reviewer. The checklist

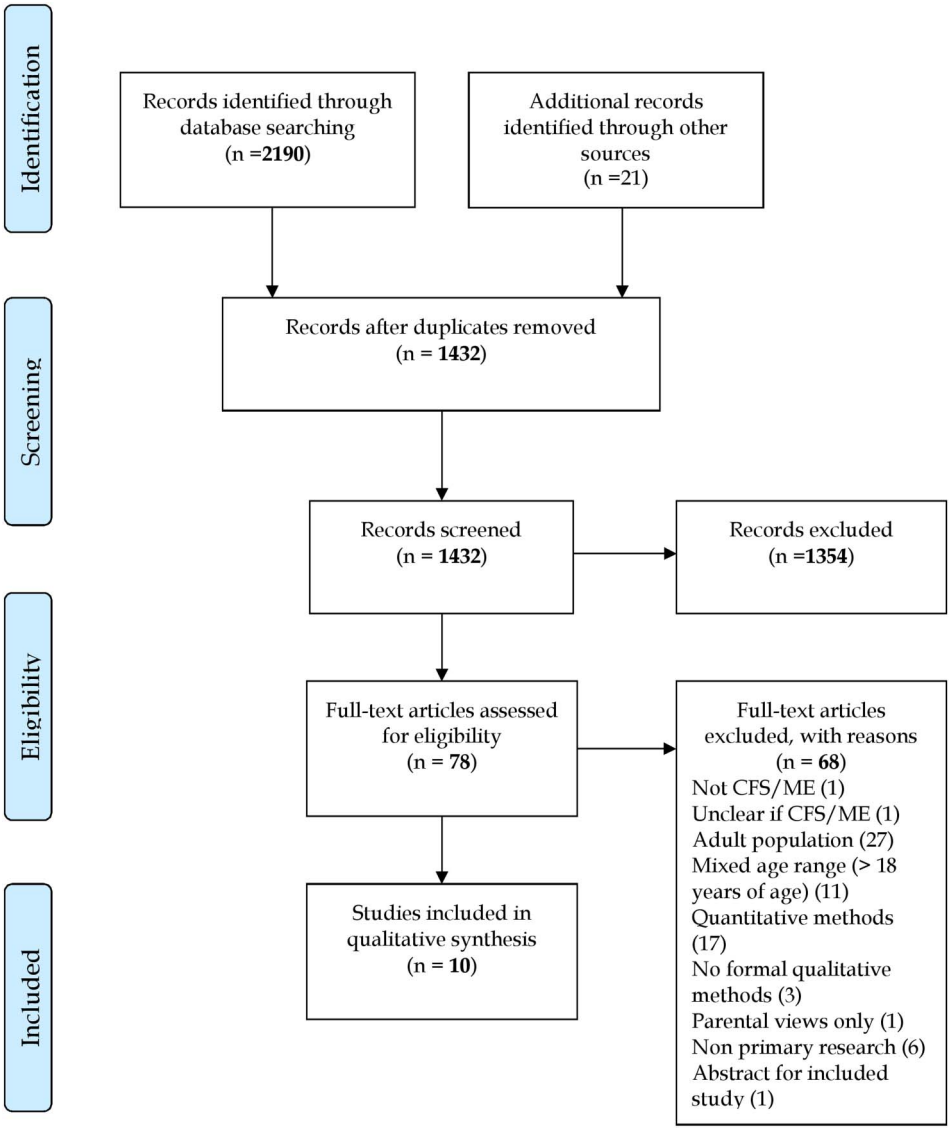

Figure 1 PRISMA flow diagram of systematic search.

was used as part of a process of exploration[44] and lower quality studies were reviewed to see if they altered the outcome of the synthesis in a sensitivity analysis.

Data extraction

For each study, three types of data were extracted: (1) descriptive data about the studies, (2) first-order constructs (participants' quotes) and (3) second-order constructs (author themes) in the results and discussion sections. A standardised prepiloted data extraction form was used by two reviewers to independently extract the data. Variations in second-order constructs extracted between reviewers were discussed and agreement reached.

Synthesis

We used techniques of meta-ethnography originally developed by Noblit and Hare.[45] Following detailed reading of the full texts, the majority of studies focused broadly on children's experiences of CFS/ME, therefore, it was decided to synthesise the studies as a whole. The final agreed second-order constructs were entered into an excel chart; second-order construct labels were in the original authors' own words with little reinterpretation. A description of each second-order construct was added to preserve the original terminology. First-order constructs (quotes) were examined next to the second-order constructs (author themes) to provide context. To translate second-order constructs across studies, RP compared the constructs to identify patterns of shared meaning where authors used varied language to label the same phenomenon. In collaboration with members of the synthesis team (AA, AS and EC), the translated second-order constructs were reinterpreted to develop new overarching third-order constructs. The final third-order constructs were established prior to looking at psychological theories to explain the constructs.[32] We undertook a reciprocal translation of third-order constructs across the studies resulting in a line of argument synthesis.

RESULTS

Included studies

In total, 10 studies involving 82 children aged 8–18 were included (table 1). Half of the studies did not specify the CFS/ME diagnostic criteria and half used the Centre for Disease Control and Prevention (CDC) Fukuda *et al*[37] and National Institute for Health and Care Excelllence (NICE)[9] criteria. Nine studies were published in English and one in Afrikaans. Seven of the ten studies were based in the UK, two in Norway and one in South Africa. One study employed a family interview,[46] all others used individual interviews (in-depth and semistructured). Two studies included specific populations: recovered patients[47] and those with high anxiety.[48]

Critical appraisal

There was good agreement (74%) on the CASP responses for the studies by the two reviewers. The CASP scores ranged from 3 to 10 with only one study[49] scoring below 5 (table 2). We undertook a sensitivity analysis and removed constructs from three studies with the lowest CASP scores (<6)[46 49 50] from the synthesis. The constructs emerged as supportive as they were also reported in other studies. Therefore, these studies did not alter the synthesis findings but resulted in less support for the 'credible illness narratives' construct. We also explored whether the results changed if we only included the studies where it was clear that children were diagnosed using the CDC or NICE criteria. We found that exclusion of studies with no clear reporting of diagnostic criteria did not change the results of the synthesis, as the themes reported in the excluded studies simply supported those identified in the included studies.

Synthesis

Table 3 shows the translation of second-order constructs across the studies and the resultant third-order constructs developed by the synthesis team. Our synthesis describes four third-order constructs within children's experiences of CFS/ME: (1) disruption and loss: physical, social and the self; (2) barriers to coping: suspension in uncertainty, problems with diagnosis and disbelief; (3) facilitators to coping: reducing uncertainty and disbelief, credible illness narratives, diagnosis and supportive relationships and (4) hope, personal growth and recovery.

Disruption and loss: physical, social and self

Physical: learning to accommodate a new restrictive body

This construct describes the disruption children experience to their bodies. They can have an array of debilitating symptoms, including tiredness, lowered energy levels, pain, headaches, sore throat, memory loss, sleep deprivation and sensory overload.[48 50–53] The predominant symptom is relentless fatigue unresolved by rest; this can be physical, mental and/or emotional and can lead to a lack of motivation.[53] Children have to learn to live with a new restrictive body[53] and they can no longer be impulsive; constantly thinking about what their body is capable of. This creates barriers between them and things they want to do.[51]

> B2: I was suddenly very tired, and had energy for nothing other than lying in bed.[54]

Social: loss of a normal adolescent life and increased dependence

The social implications of CFS/ME were very evident in this synthesis, demonstrated by the most second-order constructs across studies. This is best described by loss, which captures the changes in children's relationships with friends and family due to the isolating effect of CFS/ME.[48] Long periods spent unable to get out of bed and out of the house, detaches children from normal social experiences. They feel left out and different from friends.[48 51] This leads to loss of social norms, loneliness and rejection from peers due to lack of understanding.[48 50 52 53]

> I lost contact with some of my friends, I became more distant from them.[47]

The natural growth in independence is disrupted as children with CFS/ME become more dependent, relying on their family for emotional and practical support.[48 53] Families have to plan to consider the extra needs of the ill child[50 52 53] and guilt can develop due to the extra burden that children are aware they place on their families.[53]

> Cause my sisters had to stop swimming and piano 'cause it costs too much, and I feel a bit guilty for that…[48]

Change in self: emotional vulnerability and uncertainty

The third-order construct captures how a change in self can occur as a result of CFS/ME. Dealing with a restrictive body can lower children's self-confidence and bring a sense of fragility and vulnerability.[48] A number of undesirable emotions are described across the studies, including irritability, sadness, worry, anxiety and depression[47 48 50 52 53] and this can add further to the negative experience of the illness.

> [I felt] stressed and depressed, 'cos I was like a sporty person and I couldn't do it.[47]

CFS/ME takes away who children 'used to be' as enjoyable hobbies are increasingly lost until there is nothing. School, a significant feature of children's lives, is disrupted. Missing school can cause stress due to falling behind and be a setback to their ideals and aspirations.[48 53] Areas of achievement in the past such as academic attainment and peer popularity are lost and this leads to a sense of failure and identity confusion.[53] Children with CFS/ME reflect on themselves as changed.[51]

Table 1 Table of included studies

Study	Country	Setting	CFS/ME diagnostic criteria	Number of participants	Participant characteristics			Aim	Data collection	Data analysis
					Age range (years)	Males/ females	Illness duration			
Jelbert *et al*[47]	UK	Outpatient clinic	None specified. Clinical diagnosis of CFS/ME	5	13–18	1:4	1.5–2 years	Recovered adolescent experiences of CFS/ME	Semistructured interviews	Interpretative phenomenological analysis
Fisher and Crawley[48]	UK	Outpatient clinic	None specified. Clinical diagnosis of CFS/ME. Above the 90th percentile cut-off on SCAS Scale	11	12–18	2:9	NS	Anxious young people's experiences of CFS/ME	Interviews	Interpretative phenomenological analysis
Hareide *et al*[54]	Norway	Hospital	Modified version of the CDC criteria—3 rather than 6 months duration of fatigue	9	12–17	NS	2.5 years	Illness beliefs and coping strategies among adolescents with CFS/ME	Semistructured interviews	Thematic analysis
Winger *et al*[51]	Norway	Hospital and primary care	3 months of unexplained fatigue (RCPCH and NICE)	17	12–18	5:12	NS	Experience of being an adolescent with CFS/ME	In-depth interviews	Phenomenological hermeneutical design
Beasant *et al*[55]	UK	Specialist CFS/ME service	NICE 2007. Mild to moderately affected	12	12–18	3:9	9–18 months	Experiences of adolescents and families accessing a specialist service	In-depth interviews	Thematic analysis
Crix *et al*[46]	UK	Hospital	None specified. Clinical diagnosis of CFS/ME	1	16	0:1	1–2 years	How members of one family define and understand a contested diagnosis through talk	Family interview	Discourse analysis
Ashby *et al*[49]	UK	CAMHS	None specified. Clinical diagnosis of CFS/ME	10	8–16	3:7	3 months–2 years	Service users' perceptions of the treatment they received	Semistructured interviews	None specified
Patel[52]	UK	Specialist CFS/ME service	NICE 2007, mild to moderately affected (not housebound)	7	8–16	5:2	NS	Illness domains that are important to young people with CFS/ME and their parents	Semistructured interviews Focus group with three mothers	Thematic analysis

Continued

Table 1 Continued

Study	Country	Setting	CFS/ME diagnostic criteria	Number of participants	Participant characteristics: Age range (years)	Males/ females	Illness duration	Aim	Data collection	Data analysis
Williams-Wilson[53]	UK	Specialist CFS/ME service	Clinical diagnosis of CFS/ME	8	11–18	2:6	NS	Personal experiences of young people with CFS/ME	Open-ended interviews	Thematic analysis
Lombard[50]	South Africa	Through medical doctors	CDC	2	17	2:0	NS	Description of living with CFS/ ME to create guidelines	Interviews, document analysis and observation	Phenomenology

CAMHS, child and adolescent mental health service; CFS/ME, chronic fatigue syndrome/myalgic encephalomyelitis; NS, not stated; RCPCH, Royal College of Paediatrics & Child Health; SCAS, Spence Children's Anxiety Scale.

Table 2 Distribution of second-order constructs across studies and CASP scores

	Third-order constructs (developed by the synthesis team)											
	Disruption and loss				Barriers		Facilitators					
Studies	Physical —the illness	Social—loss of a normal adolescent life	Social— increased dependence	Change in self	Problems with diagnosis	Uncertainty, disbelief and stigma	Credible illness narratives	Diagnosis, advice and increasing awareness	Supportive relationships	Personal growth and hope	Recovery	CASP scores 0–10
Jelbert *et al*[47]		✓		✓	✓	✓		✓		✓	✓	10
Fisher and Crawley[48]	✓	✓	✓	✓		✓	✓	✓	✓	✓		9
Hareide *et al*[54]					✓		✓	✓		✓		8
Winger *et al*[51]	✓	✓		✓		✓				✓		7
Beasant *et al*[55]								✓				9
Crix *et al*[46]							✓					6*
Ashby *et al*[49]							✓					3*
Patel[52]	✓	✓	✓	✓		✓			✓		✓	10
Williams-Wilson[53]	✓	✓	✓	✓	✓	✓	✓	✓	✓			10
Lombard[50]	✓	✓	✓	✓					✓			6*

*Weaker quality study (CASP scores <6). Included in a sensitivity analysis by removing constructs from the synthesis.

> I feel like I have changed as a person, and I am not as energetic and outgoing and stuff… I don't really understand what I have kind of turned into…[48]

Additionally, the unclear aetiology, treatment and prognosis of the disease introduce profound uncertainty into children's lives[53] making them question their future.[48]

> Thinking of CFS there's an image, big scary monster, big black tunnel where you don't know where you're going or when its going to end.[47]

Barriers to coping: suspension in uncertainty and disbelief

Problems with diagnosis

This construct describes how children are suspended in uncertainty, as they struggle to get a diagnosis and as a result are unable to construct a new illness identity. Negative medical encounters were reported in several studies, including feeling unsupported by family doctors, diagnostic delays and misdiagnosis.[47 53 54] This can leave families feeling isolated from the medical community.[53] A lack of medical advice led to too much rest or overextension making children feel worse.[54]

> B1: [The doctor] transformed into a psychologist, and started asking whether I had attempted suicide and that sort of thing. This made me angry…[54]

Disbelieved and stigmatised

Children with CFS/ME can experience stigma due to the uncertainty surrounding the illness and lack of understanding from others, which can impact on how they feel about themselves. Even when a diagnosis is achieved, this can lead to disappointment as it is not accepted as a 'proper illness'.[51] The lack of medical and visible physical signs of illness make it difficult to explain;[48] many of the studies reported that children were not believed about their fatigue[48 51 53] and this introduced difficulties into relationships with children's own families and friends, as well as relationships outside of their home.[47] A lack of understanding from schools makes managing the illness as well as reintegration difficult.[47 50 52]

> G2: 'The worst thing was not to be believed; that I was forced to go to school and that I was pushed. It was horrible'.[54]

Some of the studies reported how children with CFS/ME can feel self-conscious in public places, due to concerns that strangers are commenting on them in a negative way.[53] A study that included children with high levels of anxiety found that children were distressed about being distrusted by others,[48] whether by strangers or by those known to them, which impacted their sense of credibility.

> They make like little jokes about it like; 'O no, he cannot go and get his racquet… No that takes energy'… It's not even funny…[48]

Facilitators to coping: reducing uncertainty and disbelief

Building credible illness narratives

Half of the studies examined how children understood CFS/ME and what had caused it. The synthesis revealed that children develop narratives of physical and psychological attributions to gain legitimacy. Most children attribute physical reasons such as infection as a key factor in developing CFS/ME,[49 53 54] some children have a multicausal understanding of their condition as physical and psychological in origin. Psychological difficulties, such as experiencing stressful events, were perceived by children as causing their condition.[46 49 53 54]

> I had glandular fever before it so, I think that was like where CFS came from.[47]

> G3: 'Both my mom and I think that, if I have this disease. . .that it [a traumatic event] might have triggered it'.[54]

Crix *et al*[46] discourse analytic study found that family discourses about CFS/ME were divided. Two family members constructed CFS/ME as a 'genuine illness' using medical discourse, whereas two constructed the illness as 'laziness' used intentionally for advantage. This can add to the strain already experienced in families due to the illness.

> 50 Mother: …you got a viral illness and hh(1.8) you just sort of turned from being a really strong (3.7) healthy person, to into someone who couldn't do anything didn't you? 53 Daughter : yeah em[46]

Forming coherent explanations for their illness gave children psychological agency to prove to others that they are not responsible for their condition. Hareide *et al*[54] identified a 'simple illness profile' in some children with CFS/ME. These children have an outer attribution for the cause (physical causes— not being responsible for their condition) and an inner attribution of control (having psychological agency). This helped to decrease their experience of helplessness. Those with a 'complex illness profile' added psychological attributions to the cause of their condition and were able to integrate difficult feelings in their self-understanding to cope with their condition.

> G1: 'I think that I will get well. I hope so. I do not intend to do nothing the rest of my life'.[54]

Diagnosis, advice and increasing awareness

Our synthesis revealed that reducing uncertainty through diagnosis, advice on management and validating the illness within children's social networks helped children cope with the condition. Williams-Wilson[53] found children to report a sense of relief following diagnosis. A study of children with CFS/ME attending a specialist service emphasised that recognition of the condition by specialists, along with advice on

Table 3 Development of third order constructs

Third-order constructs (developed by the synthesis team)	Second-order constructs (original author themes)	Studies that include the second-order construct
Disruption and loss: physical—the illness	Physical experience of CFS/ME	Fisher and Crawley[48]
	The body, the illness and me	Winger *et al*[51]
	Superordinate theme—feeling unwell	Patel[52]
	Symptoms	Patel[52]
	Physical changes	Patel[52]
	Adolescent CFS experienced as having to adapt to debilitating physical symptoms	Williams-Wilson[53]
	Being constantly exhausted	Williams-Wilson[53]
	Some level of cognitive disruption	Williams-Wilson[53]
	Learning to accommodate the boom bust cycle	Williams-Wilson[53]
	Physical subsystem: physical exhaustion	Lombard[50]
	Physical subsystems: sleep disturbances	Lombard[50]
	Intrapsychic subsystem: general cognitive dysfunction	Lombard[50]
	Intrapsychic subsystem: neurological signs	Lombard[50]
Disruption and loss: social—loss of a normal adolescent life	Superordinate theme—activity	Patel[52]
	Limiting and limited activity	Patel[52]
	Hobbies and interests	Patel[52]
	Stories of loss	Jelbert *et al*[47]
	Social loss and adjustment	Fisher and Crawley[48]
	The loss of normal adolescent life	Fisher and Crawley[48]
	On the side of life—locked in and shut out	Winger *et al*[51]
	Adapting to a life put on hold	Williams-Wilson[53]
	Feeling life has been put on hold	Williams-Wilson[53]
	A loss of social knowledge regarding norms and mores due to peer segregation	Williams-Wilson[53]
	Overarching theme—impact of feeling unwell	Patel[52]
	Superordinate theme—social life	Patel[52]
	Friends	Patel[52]
	Isolation and loneliness—a demise in peer relationships	Williams-Wilson[53]
	Ecological subsystem: socialising	Lombard[50]
Disruption and loss: social—increased dependence	The need for adjustments to family relationships	Fisher and Crawley[48]
	Superordinate theme—family life	Patel[52]
	Adolescent CFS experienced as living with changes in family relationships and member's life experiences	Williams-Wilson[53]
	Needing to alter family life to accommodate one member's physical limitations	Williams-Wilson[53]
	A cause of friction within parent–adolescent relationships	Williams-Wilson[53]
	Ecological subsystem: family relationships	Lombard[50]
	Feeling confused, guilty, fearful and powerless	Williams-Wilson[53]
Disruption and loss: change in self	Increased worries about school work	Fisher and Crawley[48]
	A major cause of academic disruption	Williams-Wilson[53]
	The difficult emotional experience	Jelbert *et al*[47]
	Increased emotionality	Fisher and Crawley[48]
	Superordinate theme—emotional well-being	Patel[52]
	Anxiety and mood	Patel[52]
	Intrapsychic subsystem: depression	Lombard[50]
	Intrapsychic subsystem: personality changes	Lombard[50]
	The forced need to adapt to constraints of diminished energy	Williams-Wilson[53]
	Needing to relinquish extracurricular activities and hobbies	Williams-Wilson[53]
	The vulnerable self- internal, individual experience of CFS/ME	Fisher and Crawley[48]
	Identity confusion	Fisher and Crawley[48]
	The body, the illness and me	Winger *et al*[51]
	Uncertainty about the future	Fisher and Crawley[48]

Continued

Table 3 Continued

Third-order constructs (developed by the synthesis team)	Second-order constructs (original author themes)	Studies that include the second-order construct
Barriers: problems with diagnosis	Seeking understanding	Jelbert *et al*[47]
	Negative medical encounters	Hareide *et al*[54]
	Dealing with ignorance from 'gate keepers' of further medical assistance	Williams-Wilson[53]
	Rest also increased fatigue	Hareide *et al*[54]
	Overextension made it worse	Hareide *et al*[54]
Barriers: uncertainty, disbelief and stigma	Uncertainty of the validity of CFS/ME: feeling disbelieved	Fisher and Crawley[48]
	Feeling uncertain about how to explain CFS/ME	Fisher and Crawley[48]
	Adolescent CFS experienced as feeling misunderstood and judged	Williams-Wilson[53]
	Feeling self-conscious in public places	Williams-Wilson[53]
	Negative psychosocial influences	Jelbert *et al*[47]
	School. Negative:	Patel[52]
	Difficult reintegration	Jelbert *et al*[47]
	Friendships were put to the test	Fisher and Crawley[48]
	Enduring teasing and misunderstanding from classmates	Williams-Wilson[53]
	Emotional bullying	Patel[52]
	If the illness is not visible to others, does it exist?	Winger *et al*[51]
	Introduction of uncertainty and unpredictability	Fisher and Crawley[48]
Facilitators: credible illness narratives	Attribution: psychological or somatic? Initial somatic attributions	Hareide *et al*[54]
	Additional psychological attributions	Hareide *et al*[54]
	Triggered by some physical condition, although these vary greatly	Williams-Wilson[53]
	Understanding of CFS, including factors important in its development	Ashby *et al*[49]
	Psychological stress discourse used to account for the development of the illness	Crix *et al*[46]
	Simple illness profile	Hareide *et al*[54]
	Complex illness profile	Hareide *et al*[54]
	Individual differences	Fisher and Crawley[48]
	Content of anxiety	Fisher and Crawley[48]
	Onset of anxiety	Fisher and Crawley[48]
	The construction of a 'genuine illness' account	Crix *et al*[46]
	The construction of the illness as 'intentionally used for advantage'	Crix *et al*[46]
	The negotiation of CFS/ME's status as a genuine physical illness	Crix *et al*[46]
Facilitators: diagnosis, advice and increasing awareness	Experiencing a sense of relief on achieving a diagnosis	Williams-Wilson[53]
	Recognition and progress—taking the next steps	Beasant *et al*[55]
	Influences on the illness	Jelbert *et al*[47]
	Positive psychosocial influences	Jelbert *et al*[47]
	Coping: activity or rest? Rest experienced as beneficial	Hareide *et al*[54]
	Contributions towards recovery	Fisher and Crawley[48]
	Investigating alternative therapies and medications	Williams-Wilson[53]
	Awareness of CFS/ME	Fisher and Crawley[48]
Facilitators: supportive relationships	School Positive (support from schools):	Patel[52]
	Ecological subsystem: management of schooling	Lombard[50]
	Good relationships	Fisher and Crawley[48]
	Feeling reassured when in contact with others in a similar situation	Williams-Wilson[53]
Hope and personal growth	Personal growth	Jelbert *et al*[47]
	Sharing experience and knowledge	Jelbert *et al*[47]
	Hope	Fisher and Crawley[48]
	Most informants used a flexible coping strategy	Hareide *et al*[54]
		Hareide *et al*[54]

Continued

Table 3 Continued

Third-order constructs (developed by the synthesis team)	Second-order constructs (original author themes)	Studies that include the second-order construct
	Hope, meaning and learning as a part of psychological coping	
	Handling life while hoping for a better future	Winger *et al*[51]
Recovery	Superordinate theme—feeling well	Patel[52]
	Doing more	Patel[52]
	Feeling different	Patel[52]
	How I am now: personal growth, caution and optimism	Jelbert *et al*[47]
	Positive changes in recovery	Jelbert *et al*[47]

CFS/ME, chronic fatigue syndrome/myalgic encephalomyelitis.

management reduced uncertainty and brought a sense of structure and normality back into children's lives.[55] Children reported improvements after learning to manage activity wisely to cope with fluctuating symptoms.[54]

> When it first happened, I felt sort of like lost. I didn't really feel myself, but then after [the hospital appointment], after knowing what I had, I had like a plan to get through it…[48]

The important role of communication between healthcare and schools to reduce disbelief and uncertainty was highlighted in the synthesis.[48]

> If the school hadn't been telling all my friends, I don't think I would be where I am now recovering…[48]

Supportive relationships

Supportive relationships in which friends, family and teachers provide practical help, such as giving lifts or short visits help children feel understood and considered.[47 48 52] Reaching out to other children with CFS/ME (eg, through Action for Youth with ME (AYME)), can give a sense of legitimisation and lessen feelings of isolation[53] and being part of a community of others with CFS/ME brings a sense of sharing, being valued and becoming credible.

> It's nice to have people going through the same thing as you. It's nice to be able to say —I'm feeling really bad today and have one of your friends say —Oh, me too.[53]

Hope, personal growth and recovery

The final construct in the synthesis is hope, personal growth and recovery. Although children's future plans may have been altered, our synthesis revealed an expressed need to keep hopeful. Finding meaning in small activities such as spending time with friends created a balance with managing a difficult condition.[48 51 53]

> When I'm dancing or singing then it's like I'm in another world…I feel free! Especially now, when I'm ill…[51]

Many of the studies demonstrated how children with CFS/ME can experience personal growth, including learning how to manage their energy levels; having a new perspective on life; developing more compassion for others and wanting to raise awareness.[47 54] This synthesis also highlighted the changes in children feeling better[52] or recovered.[47] When children with CFS/ME feel better, they report 'feeling different' and having more energy allowing them to feel like 'doing more'.[52] Getting back to a 'normal' adolescent life, including seeing friends and returning to hobbies led to positive hopes for their future.[47] Children with CFS/ME can have a shift in their self-concept; a new appreciation for life and knowing themselves better.

> I feel like I've benefited from having it, I know my personal boundaries, I know what I can and cannot do. . .I take advantage of everything.[47]

Line of argument

We have brought the constructs together into a final line of argument. The physical and social loss and increased emotionality experienced by children with CFS/ME can be understood through Bury's[27] concept of biographical disruption. Chronic illness represents continuing disruption that has an impact on the self. Fluctuating symptoms in CFS/ME present children with a new restrictive body; daily life is more difficult and there is a focus on this disruption to the body. Most widely accepted definitions of the 'self' consider it to be constructed through interaction with others.[56] Therefore, the loss of a normal adolescent social life has a significant impact on the self. In our synthesis, school is disrupted; children with CFS/ME become more distant from peers and dependent on their parents. This results in a shift from a perceived normal trajectory of academic achievement and independence to one that is uncertain,[27] and children begin to question plans they had for the future. The biography that children with CFS/ME construct about their lives past, present and future is interpreted and changed as a result of the illness.

The unfamiliarity of the illness and problems with diagnosis and disbelief from others act as barriers to

coping. Individuals need to work out how to explain the illness to themselves and others[26] and complete knowledge given from healthcare with their total biography.[57] Children with CFS/ME develop explanations for their illness in order to gain legitimacy and allow them to cope. Illness representations are patients' own commonsense beliefs about their illness that guide coping efforts.[58]

Finally, our synthesis revealed that children with CFS/ME can have a new appreciation for life and experience personal growth. Disruption in chronic conditions has been noted to create a redefinition of the self.[59] Frank[28] described illness as a vehicle for self-transformation. In our synthesis, symptoms and a loss of the ability to carry out activities reflected Frank's[28] chaos narrative. This was exacerbated by problems with diagnosis and feeling disbelieved by others. Chaos was alleviated in part through a diagnosis of CFS/ME. Finally, reflecting Frank's quest narrative, children with CFS/ME have a new appreciation for life and know themselves better achieving a new self that draws on the experience of having suffered.

DISCUSSION

Our synthesis highlights the physical and social loss experienced by children with CFS/ME that has a profound impact on their sense of self. Children are suspended in a state of emotional vulnerability managing debilitating symptoms yet are unsure if they will ever recover, disrupting their aspirations and ideal trajectory. Unfamiliarity of the condition results in problems with diagnosis and stigma preventing children from forming a new credible illness identity. However, children with CFS/ME can gain a new appreciation for life and integrate their experiences into a new identity. Facilitators to help children cope include reducing uncertainty and disbelief through better diagnosis and legitimisation of their illness by health professionals and improved understanding and acceptance within their social network.

Strengths and limitations

We undertook a comprehensive systematic search and aimed to include all published and unpublished studies from any language to avoid bias. Multiple reviewers screened the studies, extracted the data and identified second-order constructs. This helped to ensure consistency.[32] RP led on the development of third-order constructs; however, we incorporated the views of others in the team to enrich the synthesis. We were interested in the views of children (<18 years of age) and excluded studies with mixed age ranges (including children and adults). Therefore, we may have missed important results; however, we could not be sure which themes had been derived from children or adults. We were also unable to describe age differences because the majority of the data (quotations) did not indicate age. We did not exclude studies based on quality as methods for critically appraising qualitative research are still emerging, and there is ongoing debate about exclusion.[34 60 61] Some argue that weak studies should be excluded,[60 62 63] however, this may discount important conceptual insights.[44] Campbell *et al*[64] do not recommend 'abandoning appraisal' altogether. We used the CASP checklist in a sensitivity analysis by removing studies considered to have weaker quality (lowest CASP scores <6).[46 49 50] The constructs emerged as supportive as they were also reported in other studies and this was a valuable way to use the critical appraisal. Similarly, removal of studies with no clear reporting of diagnostic criteria did not alter the results. Most studies explored the experiences of children who were currently ill. In a condition with no physiological marker of recovery, future research is needed to understand how children define recovery.

Previous research

Feeling disbelieved was a key construct in this synthesis and 'social loss' had the most second-order constructs across studies. The physical and social limitations of children living with CFS/ME are similar to those with juvenile idiopathic arthritis, chronic kidney disease and cystic fibrosis who also experience loss of control over their bodies and social isolation.[65–67] However, in this synthesis, the disbelief and stigma that surround CFS/ME act to exacerbate the social isolation children experience due to their physical limitations. The International Classification of Functioning, Disability and Health[68] regards stigma as a key factor limiting participation that go beyond the activity limitations resulting from physical impairment. Social isolation was also prolonged for children in this synthesis due to the lack of understanding from schools making reintegration difficult. Our synthesis revealed that children use illness narratives of physical or psychological attributions to legitimatise their illness experience and cope with the condition, and previous accounts of CFS/ME suffers have been found to position themselves as 'legitimately ill'.[69]

While previous research has described increased rates of psychiatric comorbidity in young people with CFS/ME,[70] our synthesis demonstrated how the high emotional burden of CFS/ME along with the unclear prognosis of the disease can lead to identity confusion. Children may be unable to perform at school, their aspirations are disrupted and as the course of the illness and recovery is unclear, the future remains uncertain. Disbelief from others has been found to jeopardise a patient's sense of identity in the synthesis of qualitative research in adults with CFS/ME.[35 36] Childhood is a time of developmental growth influenced by peers, family and the education system[71] and similarly in this synthesis, as children with CFS/ME experience scepticism from others, this acts as a key barrier to forming a coherent identity. Acceptance has been found to be important for adjusting to a life with CFS/ME.[72] Moreover, this synthesis revealed that biographical disruption was not only negative but could be positive; children with CFS/ME can experience a new appreciation for life, personal growth and a positive shift

in hopes and expectations for their future. Positive reinterpretation and illness gains in identity have also been found in adults with CFS/ME.[56 73–75] Whitehead[76] identified three phases in changes in identity in CFS/ME: the sick role, accepting being ill and finally a reconstruction of identity.

Problems with diagnosis were a key construct in this synthesis. Diagnosis is important for an individual's interpretation and management of an illness.[77–79] Our findings align with the CFS/ME literature[16 21 72 80 81] and reviews of studies in adults with CFS/ME: diagnosis problems fuel stigmatisation,[36] for patients, getting a diagnosis is necessary for recovery, whereas doctors are reluctant towards the diagnosis.[35] However, this synthesis also revealed that simply getting a diagnosis may not be enough as it is still not considered a 'proper illness' and stigma remains. Postdiagnosis, good communication between healthcare providers and schools is an important facilitator in which key individuals and settings in the child's social network can be educated about the condition, to enable them to support children to cope with living with CFS/ME. In addition to general support from GPs, children and their families require specialist management and advice on activity from health professionals to help them manage their condition and function in the different spheres of their lives.

Policy and practice implications

Clinicians and researchers should consider physical, social, emotional and self-dimensions of life when treating and measuring outcomes from healthcare in paediatric CFS/ME. There is a need for better recognition and diagnosis of CFS/ME and advice on activity management by healthcare professionals, including those working in primary care. Improved public awareness and understanding of the condition may enable more acceptance of children with CFS/ME within their social networks. Our synthesis highlights the benefits of peer support from other patients with CFS/ME, where children and their families can use access support groups (eg, AYME).

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