BMJ Open Children's experiences of chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME): a systematic review and metaethnography of qualitative studies

Objective: To synthesis the qualitative studies of

myalgic encephalomyelitis (CFS/ME).

children's experiences of chronic fatique syndrome/

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

Methods: Studies exploring the experiences of

unpublished, using qualitative methods were eligible.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

Programme (CASP) checklist. Studies were synthesised

Results: Ten studies involving 82 children with CFS/ ME aged 8-18 were included. Our synthesis describes

children diagnosed with CFS/ME, published or

independently using the Critical Appraisal Skills

using techniques of meta-ethnography.

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

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ABSTRACT

involving children.

their illness.

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Conclusions: Physical, social, emotional and selfdimensions of life should be included when treating and measuring outcomes from healthcare in paediatric CFS/ME. There is a need for greater recognition and

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

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 Mappensis of childhood CFS/ME is common with a revelance between 0.06% and 2.4%¹⁻⁴ of a revelance between 0.06% and 2.4%¹⁻⁴ or a riginge⁸ and additional symptoms, including and is recognised as an important disabiling or and is recognised as an important is central to a fishore and additional symptoms, including and in other chronic paediatric or emo-pingainment is central to CFS/ME and higher and in other chronic paediatric or emo-pingainment is central to CFS/ME and higher and in other chronic paediatric or emo-pingainment is central to CFS/ME and higher and in other chronic paediatric or emo-pingainment is central to CFS/ME a

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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,¹⁴¹⁵ resulting in scepticism over its existence.¹⁶¹⁷ 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 narratives²⁶ 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 onwards³⁷ 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 openended 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 'qualitaand less developed database indexing.39 tive³⁸ 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 exploration⁴⁴ 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 transthe constructs to identify patterns of shared meaning **g** 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.

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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 $al^{\beta7}$ and National Institute for Health and Care Excellence (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 patients⁴⁷ 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 study⁴⁹ 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

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with a new restrictive body⁵³ 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 ŝ captures the changes in children's relationships with copyright, including for uses rel friends and family due to the isolating effect of CFS/ME.⁴ 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...⁴⁹

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 depression⁴⁷ 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

					Participant characteristics					
			CFS/ME	Number of	Age					
Study	Country	Setting	criteria	participants	range (years)	females	duration	Aim	Data collection	Data analysis
Jelbert <i>et al⁴⁷</i>	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 phenomenologica 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 phenomenologica analysis
Hareide <i>et al⁵⁴</i>	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 <i>et al⁵¹</i>	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 CES/ME	In-depth interviews	Phenomenologica hermeneutical design
Beasant <i>et al⁵⁵</i>	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 <i>et al</i> 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 analysi
Ashby <i>et al</i> 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

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Table 1 Continue	d									
		Setting	CFS/ME diagnostic criteria	Number of participants	Participant characteristics					
Study	Country				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-ord	er constructs	(developed by	the synth	nesis team)							
	Disruption and loss						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 <i>et al</i> 47		\checkmark		\checkmark	\checkmark	\checkmark		\checkmark		\checkmark	\checkmark	10
Fisher and	\checkmark	\checkmark	\checkmark	\checkmark		\checkmark	\checkmark	\checkmark	\checkmark	\checkmark		9
Crawley ⁴⁸												
Hareide <i>et al</i> ⁵⁴					\checkmark		\checkmark	\checkmark		\checkmark		8
Winger <i>et al⁵¹</i>	\checkmark	\checkmark		\checkmark		\checkmark				\checkmark		7
Beasant <i>et al⁵⁵</i>								\checkmark				9
Crix <i>et al⁴⁶</i>							\checkmark					6*
Ashby <i>et al</i> ⁴⁹							\checkmark					3*
Patel ⁵²	\checkmark	\checkmark	\checkmark	\checkmark		\checkmark			\checkmark		\checkmark	10
Williams-Wilson ⁵³	\checkmark	\checkmark	\checkmark	\checkmark	\checkmark	\checkmark	\checkmark	\checkmark	\checkmark			10
Lombard ⁵⁰	\checkmark	\checkmark	\checkmark	\checkmark					\checkmark			6*
*Weaker quality stud	V (CASP sc	ores <6) Includer	1 in a sensitivity a	analysis by	removing cons	tructs from the sv	nthosis					

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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 lives⁵³ 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 fatigue⁴⁸ 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 the children psychological agency to prove to others that they are not responsible for their condition. Hareide *et al*^{\bar{p}^4} 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-Wilson⁵³ 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

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3 Development of third of	rder constructs	
-order constructs loped by the synthesis		Studies that include the
	Second-order constructs (original author themes)	second-order construct
otion and loss: physical—	Physical experience of CFS/ME	Fisher and Crawley ⁴⁸
ness	The body, the illness and me	Winger <i>et al</i> 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
	Intransychic subsystem: general cognitive dysfunction	Lombard ⁵⁰
	Intrapsychic subsystem: general cognitive dystanction	Lombard ⁵⁰
tion and loss: social	Superordinate theme activity	Potol ⁵²
f a parmal adalaaaant lifa	Limiting and limited activity	Potol ⁵²
r a normal addrescent life	Limiting and interacts	Patel ⁵²
	Hobbles and Interests	Palei
	Stories of loss	Seiber and Crowley ⁴⁸
	Social loss and adjustment	Fisher and Crawley
	The loss of normal adolescent life	Fisher and Crawley
	On the side of life—locked in and shut out	Winger <i>et al</i> ²
	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	Williams-Wilson ³³
	due to peer segregation	
	Overarching theme—impact of feeling unwell	Patel
	Superordinate theme—social life	Patel ⁵²
	Friends	Patel ⁵²
	Isolation and Ioneliness—a demise in peer relationships	Williams-Wilson ⁵³
	Ecological subsystem: socialising	Lombard ⁵⁰
otion and loss: social—	The need for adjustments to family relationships	Fisher and Crawley ⁴⁸
sed dependence	Superordinate theme—family life	Patel ⁵²
	Adolescent CFS experienced as living with changes in	Williams-Wilson ⁵³
	family relationships and member's life experiences	
	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

self

A cause of friction within parent-adolescent relationships Ecological subsystem: family relationships Lombard⁵⁰ Williams-Wilson⁵³ Feeling confused, guilty, fearful and powerless Fisher and Crawley⁴⁸ Disruption and loss: change in Increased worries about school work A major cause of academic disruption Williams-Wilson⁵ Jelbert et al47 The difficult emotional experience Increased emotionality Fisher and Crawley⁴⁸ Superordinate theme-emotional well-being Patel⁵² Patel⁵² Anxiety and mood Lombard⁵⁰ Intrapsychic subsystem: depression Lombard⁵⁰ Intrapsychic subsystem: personality changes Williams-Wilson⁵³ The forced need to adapt to constraints of diminished enerav Williams-Wilson⁵³ Needing to relinquish extracurricular activities and hobbies The vulnerable self- internal, individual experience of Fisher and Crawley⁴⁸ CFS/ME Fisher and Crawley⁴⁸ Identity confusion The body, the illness and me Winger et al Uncertainty about the future Fisher and Crawley⁴⁸

Continued

(developed by the synthesis team)	Second-order constructs (original author themes)	Studies that include second-order constru	the uct
Barriers: problems with	Seeking understanding	Jelbert <i>et al</i> 47	
diagnosis	Negative medical encounters	Hareide <i>et al⁵⁴</i>	
Ū	Dealing with ignorance from 'gate keepers' of further medical assistance	Williams-Wilson ⁵³	
	Rest also increased fatigue	Hareide <i>et al⁵⁴</i>	
	Overextension made it worse	Hareide <i>et al⁵⁴</i>	
Barriers: uncertainty, disbelief	Uncertainty of the validity of CFS/ME: feeling disbelieved	Fisher and Crawley ⁴⁸	
and stigma	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 <i>et al</i> 47	
	School, Negative:	Patel ⁵²	
	Difficult reintegration	Jelbert <i>et al</i> 47	
	Friendships were put to the test	Fisher and Crawlev 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 <i>et al⁵¹</i>	
	Introduction of uncertainty and unpredictability	Fisher and Crawlev ⁴⁸	
acilitators: credible illness	Attribution: psychological or somatic? Initial somatic attributions	Hareide et al ⁵⁴	
	Additional psychological attributions	Hareide <i>et al⁵⁴</i>	
	Triggered by some physical condition, although these vary greatly	Williams-Wilson ⁵³	
	Understanding of CFS, including factors important in its development	Ashby <i>et al</i> ⁴⁹	
	Psychological stress discourse used to account for the development of the illness	Crix <i>et al</i> ⁴⁶	
	Simple illness profile	Hareide <i>et al⁵⁴</i>	
	Complex illness profile	Hareide <i>et al⁵⁴</i>	
	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 <i>et al</i> ⁴⁶	
	The construction of the illness as 'intentionally used for advantage'	Crix <i>et al</i> ⁴⁶	
	The negotiation of CFS/ME's status as a genuine physical illness	Crix <i>et al</i> ⁴⁶	
Facilitators: diagnosis, advice	Experiencing a sense of relief on achieving a diagnosis	Williams-Wilson ⁵³	
and increasing awareness	Recognition and progress-taking the next steps	Beasant <i>et al⁵⁵</i>	
	Influences on the illness	Jelbert <i>et al</i> 47	
	Positive psychosocial influences	Jelbert <i>et al</i> 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	School Positive (support from schools):	Patel ⁵²	
elationships	Ecological subsystem: management of schooling	Lombard ⁵⁰	
	Good relationships Feeling reassured when in contact with others in a similar	Fisher and Crawley ⁴⁸ Williams-Wilson ⁵³	
	situation		
Hope and personal growth	Personal growth	Jelbert <i>et al</i> 47	
	Sharing experience and knowledge	Jelbert et al47	
	Норе	Fisher and Crawlev ⁴⁸	
	Most informants used a flexible coping strategy	Hareide <i>et al⁵⁴</i>	

Third-order constructs (developed by the synthesis			Studies that include the
team)	Second-order constructs (original a	uthor themes)	second-order construct
Recovery	Hope, meaning and learning as a part coping Handling life while hoping for a better Superordinate theme—feeling well Doing more Feeling different How I am now: personal growth, caution Positive changes in recovery	of psychological future on and optimism	Winger <i>et al</i> ⁵¹ Patel ⁵² Patel ⁵² Patel ⁵² Jelbert <i>et al</i> ⁴⁷ Jelbert <i>et al</i> ⁴⁷
CFS/ME, chronic fatigue syndrome/r	nyalgic encephalomyelitis.		
management reduced uncerta of structure and normality ba Children reported improven manage activity wisely to symptoms. ⁵⁴	inty and brought a sense Many ck into children's lives. ⁵⁵ CFS/M nents after learning to learnin cope with fluctuating new per for oth thesis a	y of the studies d IE can experien ig how to manag erspective on life ters and wanting t also highlighted t	lemonstrated how children ce personal growth, incl ge their energy levels; hav c; developing more comp to raise awareness. ^{47–54} Th the changes in children f

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 isolation⁵³ 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.⁵²

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

<text><text><text><text><text><text>

coping. Individuals need to work out how to explain the illness to themselves and others²⁶ 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 Frank²⁸ described illness as a vehicle for self-transformation. In our synthesis, symptoms and a loss of the ability to carry out activities reflected Frank's²⁸ 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 by copyright, 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 Health⁶⁸ text 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 nol 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 system⁷¹ 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

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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} Whitehead⁷⁶ 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 literature¹⁶ 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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