‘Perspectives of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis: A Q-methodology study’

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Declaration

I declare that the research in this thesis is my own work. It is submitted in part fulfilment of the degree of Doctorate in Clinical Psychology (DClinPsy). It has not been submitted for any other academic award. The thesis has been checked for completeness prior to submission.

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Thesis Abstract

Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome (ME/CFS) is characterised by fatigue, alongside many other symptoms. There is no clear aetiology. This has created uncertainty for stakeholders and led to polarisation in attitudes towards the condition, its causes, and interventions. People with ME/CFS and their families report experiencing disbelief. Healthcare professionals report often finding this a difficult condition to work with, particularly when their own viewpoints differ from the people they are working with.

Literature Review

Social support plays a significant role for people with chronic conditions. The impact on family members supporting people with ME/CFS is not well understood. A systematic review exploring the experiences of people with a family member with a diagnosis of ME/CFS was undertaken. Four databases were searched, and eight studies identified. A thematic synthesis was conducted which identified three themes: ‘Changes in relationships’ looked at how people relate to their relative in new ways. ‘Coping with an uncertain and misunderstood condition’ describes the challenges of negotiating stigma and uncertainty. ‘Loss’ is concerned with participants’ sense of losing the person who had become ill and of their own identities.

Research Report

Perspectives on ME/CFS were explored using Q-Methodology. Participants sorted statements about ME/CFS according to their relative level of agreement. A factor analysis was conducted and three factors extracted: Factor 1, ‘A debilitating physical health condition,’ placed emphasis on the physiological aspects of the condition, the severity of symptoms and the lack of understanding from others. Factor 2, ‘The mind affecting the body,’ placed more importance on the role of vulnerability to stress and emotional issues. Factor 3, ‘Management is key to recovery,’ stressed lifestyle management approaches. It is hoped that these findings can support all stakeholders in reflecting on the positions they and others hold and the similarities and differences, to support constructive dialogue.

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Part 1: Literature Review

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The experiences of relatives of people with Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome (ME/CFS): A meta-synthesis of qualitative studies

Abstract

Background

ME/CFS is a condition characterised by fatigue and many other symptoms. The cause is currently unknown and there is no diagnostic test, leading to much stigma being experienced by people with the diagnosis. The impact on relatives of people with ME/CFS has been relatively under-researched, and the present synthesis was undertaken to support understanding of relatives' experiences.

Method

A systematic review was conducted of primary research. Searches were carried out on PubMed, PsychINFO, CINAHL and Cochrane Library in July 2020. Studies were included that used a qualitative approach to exploring the experience of relatives of people with ME/CFS. A thematic synthesis was completed.

Findings

Eight studies were identified that met inclusion criteria. Participants experiences were explored in three superordinate themes. 'Changes in relationships' looked at the ways in which participants were required to support their family member, how this led to relating to them in new ways and how family life came to revolve around the person who was ill. 'Coping with an uncertain and misunderstood condition' describes the challenges of negotiating stigma, misunderstandings of ME/CFS, and uncertainty regarding prognosis. The final theme, 'Loss', is concerned with participants' sense of losing the person who had become ill, and also with loss of their own identities as a result of caring responsibilities.

Discussion

Across the heterogenous participants included in this study, common experiences were highlighted. When working with people with ME/CFS, consideration should be given to offering support to relatives. This needs to be done in a way that is sensitive to their experiences, which may or may not include defining themselves as a caregiver and might involve a very close relationship with the person who is ill, or a feeling of being shut out from their experience.

Background

ME/CFS

Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) is a condition characterised by fatigue that cannot be explained by other conditions. Over 20 different case definitions have been put forward over the last 30 years, with the Fukuda et al. (1994) definition (see Figure 1) being the most widely cited and most extensively validated (Brurberg, et al., 2014).

Figure 1: Fukuda et al. (1994) definition

1) Clinically evaluated, unexplained, persistent or relapsing chronic fatigue that is of new or definite onset (has not been lifelong); is not the result of ongoing exertion; is not substantially alleviated by rest; and results in substantial reduction in previous levels of occupational, educational, social, or personal activities; and

2) The concurrent occurrence of four or more of the following symptoms, all of which must have persisted or recurred during 6 or more consecutive months of illness and must not have predated the fatigue: self-reported impairment in short-term memory or concentration severe enough to cause substantial reduction in previous levels of occupational, educational, social, or personal activities; sore throat; tender cervical or axillary lymph nodes; muscle pain, multi-joint pain without joint swelling or redness; headaches of a new type, pattern, or severity; unrefreshing sleep; and post-exertional malaise lasting more than 24 hours.

Current guidelines from the National Institute for Health and Care Excellence (NICE) (National Institute for Health and Care Excellence, 2007) use a different, slightly broader set of criteria. Fatigue need only be present for four months (three months in under 16s), post exertional malaise lasting more than 24 hours is included in the definition of the nature of the fatigue. Only one of the additional symptoms need be present from a broader list which includes all those listed by Fukuda et al. with the addition of symptoms being worsened by physical or mental exertion, malaise/flu-like symptoms, dizziness/nausea or palpitations that do not have an identified cardiac pathology. There is not currently consensus within the research field as to the most accurate case criteria or most appropriate label. This review therefore includes all papers where the diagnostic label of 'ME/CFS' was explored.

There is much debate about the underlying physiology, with some research suggesting dysregulation in the autonomic nervous system and neuroendocrine pathways (Heim et al., 2009; Van Cauwenbergh et al., 2014). Other theories propose that symptoms are due to deconditioning of the muscles or maladaptive illness cognitions (Cox, et al., 2004). The lack of clear pathophysiology combined with a history of symptoms being attributed to psychiatric illness (Ware, 1992) has meant people with the diagnosis have experienced stigma and delegitamation of their experiences (Dickson et al., 2007).

Experience of the individual with ME/CFS

The issues surrounding lack of clarity in aetiology, diagnosis, and treatment lead to many difficulties for people with ME/CFS. A review by Anderson et al. (2012) found that both perceived and enacted stigmatisation were a common experience for people with ME/CFS. Their symptoms were often attributed to psychological processes, with a suspicion that symptoms were exaggerated. This was reflected in

findings by Drachler et al. (2009), who generated themes around 'The need for recognition of needs, respect and empathy from service providers' and 'The need for positive attitudes and support from family and friends'.

Social support has been identified as a factor in the perpetuation of illness in general (Cohen, 1988) and ME/CFS specifically (Prins et al., 2004). Partner attitudes have been found to impact the efficacy of interventions (Verspaandonk, et al., 2015) and the person's adaptation to illness (Heijmans et al.. Understanding the experience of relatives of people with this condition is therefore important for understanding, working with and improving the lives of people with ME/CFS.

Experience of relatives of people with chronic illness

Research has focussed on the impact of being a caregiver, known to impact physical and mental wellbeing (Hirst, 2004; Sullivan & Miller, 2015). The experience is complex, with individuals sometimes simultaneously experiencing burden and satisfaction from caregiving roles (Lawton et al., 1991), although spousal caregivers and adult-child caregivers were differentially affected. For spousal caregivers, in contrast with adult-child caregivers, satisfaction did not correlate with amount of care given, hypothesised to be due to any caring being viewed as an assumed part of the existing relationship. Many further factors affect the degree to which caregiving impacts the person, including social support, and socioeconomic status (Savage & Bailey, 2004). However many people giving care to close relatives (partners, parents, children) do not consider themselves 'carers' (O'Connor, 2007).

A smaller body of work has explored the impact of a chronic illness on non-caregiving members of the family. There is some evidence that the siblings of children with chronic conditions can be negatively impacted by this, although this varies depending on the type and severity of the condition (Sharpe & Rossiter, 2002; Vermaes, van Susante, & van Bakel, 2012). Kish et al. (2018) detailed the many personal challenges of having a chronically ill child on parents, such as balancing working life and the needs of the family.

The experience of the family unit when a member has a chronic illness has been explored through the lens of family life cycles (Rolland, 1987, 2013). The phase of life the family is in, particularly whether the family is in a centripetal, or inward facing, phase or a centrifugal, or outward facing phase, (Combrinck-Graham, 1985) at the time of the illness can affect the way in which the illness, itself a centripetal force, impacts the family (Sperry, 2012).

The issues affecting people with ME/CFS and their relatives have some themes in common with other conditions. This is particularly the case with other chronic conditions that also lack precise classification such as fibromyalgia. These patients and relatives may also experience disbelief from within and without the family as well as consequences such as the economic impact of chronic illness on the family (Borchers & Gershwin, 2015). This review focuses specifically on the relatives of people who have been given the

diagnostic label of ME/CFS as each diagnostic label carries its own set of assumptions, attributions and biases (Jason et al., 2002).

Aims and rationale

To date, the aforementioned reviews have been published exploring patients' experiences of ME/CFS (Anderson et al., 2012; Drachler et al., 2009) highlighting the impact of stigmatisation from others and the need for respect and empathy. There is a gap in the literature relating to the experience of relatives of people with ME/CFS. Given the importance of social support already identified for people with ME/CFS (Prins et al., 2004), and the known significant impacts on relatives of people with chronic health conditions (Hirst, 2004; Sullivan & Miller, 2015), it is clinically significant to review research considering the impact of living with someone who has ME/CFS, to provide insights into how patients and their systems can be supported.

The primary aim of this review was to take an inductive approach to explore all aspects of having a relative with a diagnosis of ME/CFS. This included all forms of relative (partner, parent, child, sibling etc.) as the intention was to gain an understanding of the ways in which having a relative with ME/CFS may impact the individual. Secondary aims were to consider the impacts of perceived and enacted stigma and to explore the relevance of social support for relatives of people with ME/CFS.

Method

Search strategy

A systematic search strategy was conducted to ensure comprehensive coverage of the literature. Key databases were searched for relevant studies (PyschINFO, PubMed, CINAHL and Cochrane Library) selected so that psychological, medical, and similar relevant journals were included.

Table 1 describes the search terms used. Scoping searches indicated these search terms were optimal. Limits placed on this search were for journal articles, written in English. No limits were placed on date of publishing, as studies from any period of time were felt to be relevant to the research question. Database searches were conducted on 21st July 2020. The first two sets of search terms were used in all databases. For one database (PubMed), this search strategy returned a large number of articles, so a third set of search terms was added (Table 1, Row 3).

Table 1: Search terms

Concept	Search terms
1.Relatives	Famil* OR relative OR relatives OR parent* OR child* OR partner* OR spouse* OR carer* OR mother* OR father* OR son OR daughter* OR brother* OR sister*
2.The condition	"Chronic fatigue syndrome" OR CFS OR "myalgic encephalomyelitis" OR "myalgic encephalitis" OR "myalgic encephalopathy" OR "post viral fatigue syndrome" OR "PVFS" OR "chronic fatigue and immune dysfunction syndrome" OR CFIDS OR "systemic exertion intolerance disease" OR SEID
3. Experiences (PubMed only)	experien* OR view* OR perception* OR perspective* OR attitude* OR stress OR carer- burden

In total, 2384 articles were returned. Duplicates were removed, and abstracts or full-text manuscripts were reviewed against the inclusion criteria outlined below. This resulted in a final sample of eight papers. The search process is illustrated in Figure 2, in line with the PRISMA Statement (Moher et al., 2009).

Inclusion criteria

Inclusion criteria were defined according to the SPIDER tool (Cooke et al., 2012) which advocates defining the search strategy in terms of Sample, Phenomenon of Interest, Design, Evaluation, Research type (see Table 2). As outlined in 1.2.1, the phenomenon of interest is the experience of those whose relatives have the diagnostic label of ME/CFS and the sense they make of this term. All studies using this term were therefore included, regardless of the specific definition of ME/CFS used. In addition, all studies were required to be written in English and either published in peer-reviewed academic journals or published books. No restrictions on date or country of publication were applied.

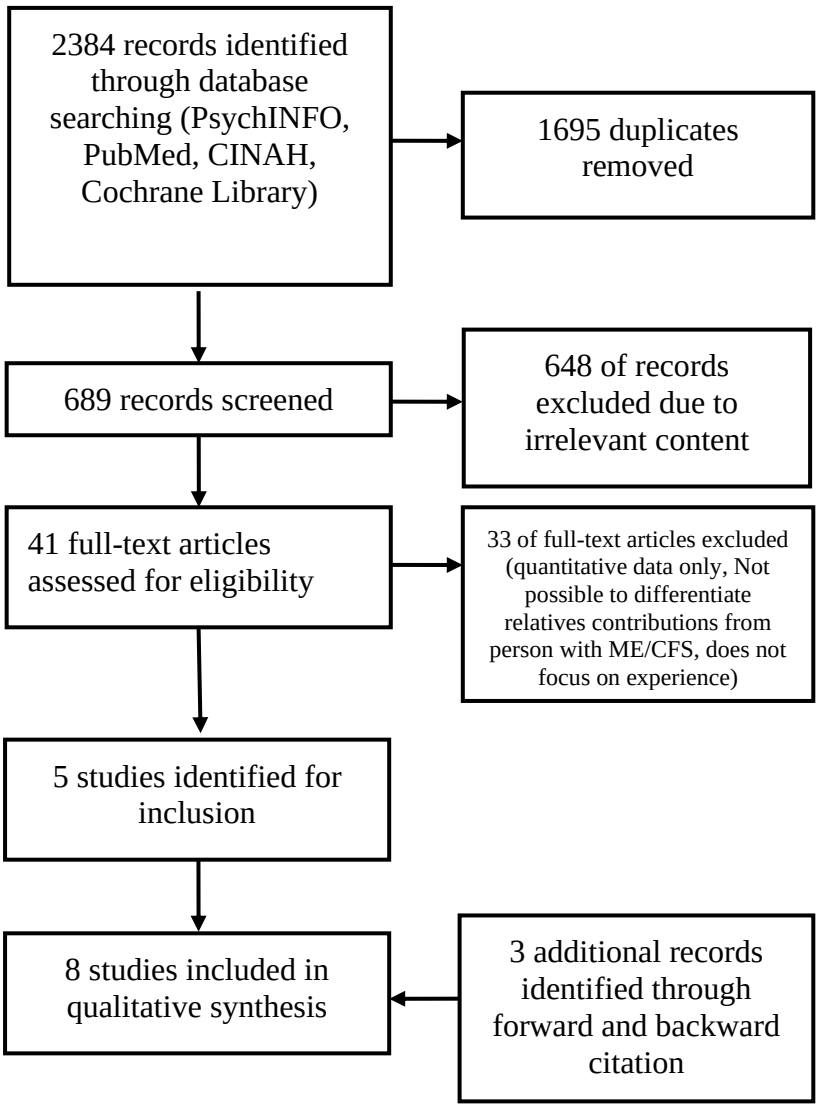
Table 2: Inclusion criteria based on SPIDER Tool

Sample	Relatives of people with ME/CFS
Phenomenon of Interest	Their experience of having a relative with ME/CFS
Design	Any were permitted
Evaluation	Any were permitted
Research type	Qualitative

Quality appraisal

The written report for each study was individually assessed, considering methodology, epistemological position of the paper and relevance to the current review. All eight identified studies were included on the basis of containing findings that were credible based on the described methodology and useful to the research question of this review (CASP, 2019) (see Appendix A).

Figure 2: Search strategy



Data extraction and analysis

NVivo 12 Pro software was used to support coding. An inductive thematic synthesis approach was taken, as described by Thomas and Harden (2008). This method was selected for its interpretive approach which fulfilled the aim of exploring experiences through the generation of new themes, rather than aggregating previous findings.

Thematic synthesis involved line-by-line coding of the 'results' and 'discussion' sections of all included studies. Codes were organised into descriptive themes, which were then used to develop analytical themes. This final stage allows for a level of interpretation that goes beyond the content of original source material through the generation of new interpretive constructs. Adapting approaches from meta-ethnography and grounded theory in this way allows key principles of systematic reviewing to be adhered to while addressing questions that are clinically relevant (Barnett-Page & Thomas, 2009). A subjective relativist epistemological position, as defined by Spencer et al. (2003), was adopted in accordance with the interpretive orientation of thematic synthesis.

Findings

This section comprises a summary of the eight studies included in this review, followed by the results from the thematic meta-synthesis.

Summary

Study characteristics and aims

Table 3 summarises the included studies. Studies were published between 2002 and 2019. Six were conducted in the UK and two in the USA. Studies had similar stated aims of exploring the experience of relatives of those with ME/CFS; some focussed on specific types of relatives, with one focussed specifically on maternal psychological health when a child has ME/CFS.

Table 3: Summary of included papers

Study		Methodology	Participants		Findings
Authors	Year	Approach to analysis	Total N	Relation to person who is ill	Aims
Ax, Gregg and Jones	2002	Thematic analysis	16	10 husbands/male partners, 2 wives, 1 parent, 1 child, 2 co-habiting friends	Improved understanding of carer's coping efforts, focussing on illness acceptance
Brooks, King and Wearden	2014	IPA	2	2 wives	Explore in depth beliefs and experiences
Catchpole and Garip	2019	IPA	7	1 wife 4 husbands 2 mothers	Improve understanding of carers by looking at their lived experience
Donalek	2009	Thematic analysis	12	8 families – 4 partners, 3 adult children, 1 parent, 4 teenage children	Describe responses of the family system
Horrocks and Ward	2015	Thematic analysis	7	4 husbands 2 wives 1 sister	To suggest possible ways of understanding, or imagining, how meanings associated with CFS/ME develop within intimate relationships
Mihelicova, Siegel, Evans, Brown and Jason	2016	IPA	19	12 mothers 7 fathers	The experiences of parents caring for people suffering from severe ME
Missen, Hollingworth, Eaton and Crawley	2011	Thematic analysis	8	8 mothers	Investigate psychological health of mothers of children with CFS/ME
Velleman, Collin, Beasant and Crawley	2016	Framework approach to thematic analysis	9	9 siblings (5 female, 4 male)	Understand impact on CFS/Me on siblings

Participant information

This review includes data gathered from a total of 80 participants, comprising 31 parents, 29 partners, 10 siblings, eight children (some adults) and two co-habiting friends.

Research design and methodology

Four studies used thematic analysis, three studies used interpretive phenomenological analysis and one used content analysis. Two studies interviewed both the ill and unaffected partners in a couple, and one interviewed the family group, including an ill parent. Responses were reported separately from these family members so only those from the relatives are reported here.

Metasynthesis

Three superordinate themes were established (see Table 4 and Appendix B for sample audit trail). The first theme explores the ways in which a member of the family living with a chronic illness impacted the relationship the participants had with the person who became ill and how it impacted family dynamics. The second theme looks at the difficulties caused by ME/CFS being a condition that not much is known about and has significant perceived and enacted stigma attached to it. The third theme explored the different types of loss experienced by participants.

Table 4: Themes and subthemes

Theme	**Changes in relationships**	**Coping with an uncertain and misunderstood condition**	**Loss**
Subthemes	Supporting their family member Different ways of relating to the person Family life revolves around the person who is ill	Misunderstood and stigmatised Coping with uncertainty	Loss of the person they were before their illness Loss of identity

Changes in relationships

A relative's illness brought about many changes in family dynamics, in addition to practical changes. These are explored below as three subthemes. 'Supporting their family member' describes the ways in which participants provided support to their relative with ME/CFS and the emotional toll this took on them. 'Different ways of relating to the person' looks at changes in the relationship between participant and their ill family member, especially the shift towards caring responsibilities. 'Family life revolves around the person who is ill' explores the effects on family structure, including the ways ME/CFS resulted in greater closeness between some participants and the ill relative, whilst other relatives became pushed to the periphery of family life.

Supporting their family member. This subtheme is about the support that participants give their family member and the impact it can have on them. While dealing with their own emotional responses, relatives were often required to be the main source of emotional support to their ill relative.

> 'All of a sudden something will snap, and you'll go upstairs, and no-one will realise that she's … upstairs in a bed, lying down in bed crying, because she's emotional and exhausted. But I'm the one that picks up the pieces.' (Husband, Horrocks & Ward, 2015, p91)

Powerlessness was often referenced regarding how to help. Relatives felt they should be able to do more for their family member or that they were not giving enough support to other family members because of their caring responsibilities. Some felt guilt about having to prioritise other areas of life, such as running a business, meant they did not give the level of support they felt they should to their family.

> 'If you asked me whether I gave her enough support, the answer is probably no. Certainly not all the time... I mean it has to come in second place to what I'm doing. I've got a business to run, and it has to be done... So on occasions I probably don't support her as much as I ought to.' (Husband, Ax, Gregg & Jones, 2002, p39)

Many participants were involved in supporting the person to manage their illness and pace their level of activity. Many found that the 'role of "pacing patroller" was both welcomed and resented.' (Horrocks & Ward, 2015, p94), with many participants reporting that this led to friction as their efforts were perceived as 'nagging' (Brooks, King & Wearden, 2014, p11).

Different ways of relating to the person. The changes in the relationship between participants and the person with ME/CFS following the illness are explored in this subtheme. Prominent within the data was the taking on of caring responsibilities, although participants, particularly when they were partners of someone with ME/CFS, were very rarely quoted as having used the term 'carer' or 'caring'. It seemed that they identified primarily with their role within the family (mother, father, partner, sibling) and saw the additional caring duties as an extension of fulfilling these roles.

> ‘You know, we’re still husband and wife… I know that our relationship is not… it’s different to how it used to be’.’ (Catchpole & Garip, 2019, p7)

Relatives took on household tasks that had previously been done by the person who had become ill. For male participants especially, this could require them to take on roles that are seen as traditionally female. Some disliked needing to do tasks they felt they were not good at while others accepted that this ‘had to be done for the family to survive.’ (Donalek, 2009, p336). These new roles included practical tasks as well being an advocate for the ill person in healthcare and education settings.

Some siblings took on caring responsibilities for their ill sibling. For some children, where a parent was ill, this meant taking on household tasks and caring responsibilities themselves.

> ‘It was a gradual process, it gradually got worse. So, I kind of eased into doing more and more things, …. I did, like, the laundry and cooking and dishes, and I did my homework and everything, too.’ (Daughter, Donalek, 2009, p336).

For some spouses, maintaining a marital relationship was difficult. For one, this had been possible where a woman felt the condition was being managed well by professionals, allowing her to maintain an ‘identity as David’s wife and partner, rather than assuming a carer or advocate role’. Another woman had a very different experience, feeling that she had become ‘an unpaid servant’ (Two wives, Brooks, King & Wearden, 2014, p10, p13).

Physical relationships were very rarely addressed in these studies. It is possible that researchers and/or participants did not feel they had permission to discuss experiences that were sexual in nature or that participants did not feel comfortable disclosing intimate details. One couple reported a complete breakdown in their sex life. For another, spontaneity had had to be sacrificed.

> ‘It’s almost like a little space in our diary isn’t it now, that’s just to make sure we’ve got the energy for it’ (Husband, Horrocks & Ward, 2015, p93)

Parents of adults who were ill had to negotiate finding themselves returning to a caring role for their children. Fulfilling the caring role while trying to maintain their child’s independence was challenging.

> ‘We can’t revert her back to being that child, because she’s not, she’s an adult and she’s a person in her own right, but she’s dependent on us, which really, really upsets her.’ (Mother, Catchpole & Garip, 2019, p7)

For parents of children who were ill, not being able to show physical affection could be distressing.

> ‘As a mother, the most natural thing in the world is to gather your child in your arms and make everything better with a kiss and a cuddle. For too many years I was unable to do this for my daughter … Every part of her body hurt so much she couldn’t bear to be touched’ (Mother, Mihelicova et al., 2016, p2828)

Some had found ways to cope with this.

> ‘On a good day she would just “hold” my thumb, well I’d “rest” it, between her thumb and finger; it was all she could manage and nothing like the cuddle we both so desperately needed, but it was some level of contact; the contact, the comfort, she so desperately needed, as she felt so desperately ill.’ (Mother, Mihelicova et al., 2016, p2828)

Family life revolves around the person who is ill. This subtheme is concerned with changes that centred the needs of the person who was ill, sometimes resulting in closer dyadic relationships and sometimes pushing out other relatives. The person who was ill tended to become the focus of family life, where day to day life and future planning were based on their needs.

> ‘Life becomes a routine shared by all. One becomes used to the dark rooms to reduce light sensitivity, the lack of sound to avoid headaches, the changes to diet, the cost of supplements, the lack of a social life, the lack of family occasions, the removal of all things normal.’ (Husband, Mihelicova et al., 2016, p2832)

There was a strong sense of participants’ own emotional wellbeing being tied to the wellbeing of their family member. For example, when asked how things are at home, participants replied in relation to how the person with ME/CFS was doing.

> ‘They’re really good at the moment because John’s doing really well’ (Sibling, Velleman et al. 2016, p625)
>
> ‘I’ve started to, as she’s got better, I’ve got better’. (Husband, Catchpole & Garip, 2019, p8).

The demands of the illness led families to spend large amounts of time together at home. Many relatives became intimately attuned to the needs of the person who was ill. As many parents or partners had given up work, and socialising together was not possible, there was a sense of the family unit being cocooned together.

> ‘[speaking to his wife with ME/CFS] It’s okay, but everything is about you’ (Husband, Donalek, 2009, p337)
>
> ‘Couples sometimes seemed to be deeply immersed, together, in an unpredictable, all-encompassing illness… Couples often described social withdrawal as a shared retreat into an intimate space where partners seemed to be specially attuned to the ill person’s needs and where private understandings of illness were formed. This made the experience of CFS/ME seem like a closed world.’ (Horrocks & Ward, 2015, p104)

There were examples of siblings taking on new roles in relation to non-ill parents, by supporting them.

> ‘My sister can reduce my Mum to tears. . . and she [Mum] obviously has to talk to someone and Dad’s at work’ (Sibling, Velleman et al., 2016, p626).

There were exceptions to the centring of family life around the person who was ill. One experience was of feeling left out of the family. For some parents, being the one to continue working, while the other parent took on the majority of the caring responsibilities for an ill child, led them to feel that they did not know their child's needs and that the child developed a much closer bond with the other parent, to the exclusion of them.

> 'When [my daughter] is having a particularly bad day, the only person she wants is her mum. Sometimes this hurts but I know I have not been here much and I don't understand the illness or what she needs like my wife does. [My daughter] trusts her mum, they have been together day and sometimes night for the whole length of [my daughter's] illness; she knows what [my daughter] needs without her having to say and explain, I don't. I still have an awful lot to learn, but I'm trying.' (Father, Mihelicova et al. 2016, p2830)

Siblings noticed a similar change. Many felt that they were not communicated with about the illness.

> '…the siblings talked about negative, or lack of, communication within the family, for example, a change in the way the family communicated since their sibling became ill and a feeling of being unable to talk to their parents or siblings about their feelings: 'We used to have debates, the kind of, just jokey debates round the table; it's hard to remember that far back.' (Velleman et al., 2016, p625).

Some participants intentionally withdrew, for example choosing not to learn more about the illness. The demands of living with someone who was ill led some participants to want to retreat from family life.

> 'She often calls (at work) and asks me to come home early. I don't want to. Even if there isn't a lot of work, I just sit there having a cup of coffee. I usually need to stay at work till late, but I don't want to come home and bathe the children. In fact, in the evening I don't want to see the children.' (Husband, Ax et al., 2002, p37)

Coping with an uncertain and misunderstood condition

ME/CFS remains a poorly understood condition. This theme examines the ways this affected the participants. The impact is descried in two subthemes. 'Misunderstood and stigmatised' explores the ways in which misconceptions about the condition impacted the relatives. 'Coping with uncertainty' looks at the fluctuations and unclear prognosis that come with ME/CFS and ways participants responded to these.

Misunderstood and stigmatised. This subtheme is about challenges faced in relation to others outside the family not understanding the impact of ME/CFS on the person and the family. A widely reported experience was of ME/CFS being a difficult condition to understand.

> 'People talk about fatigue and I think sometimes there've been occasions in the past where I've had to say to myself, just, just leave it alone because people say, it's just a little bit like 'Oh yeah, I get tired', and it's kind of you kind of go, 'Ah okay. Yeah okay'.' (Wife, Catchpole & Garip, 2019, p4)

Some participants found that others were not interested in hearing about what it was like supporting a relative with ME/CFS.

> 'There's the way that after a while people— after weeks, months, certainly years—start to give off subtle and not-so-subtle signals that they really don't want to know about your daughter's continuing illness or its effects.' (Father, Mihelicova et al., 2016, p2829)

Others found that people misunderstood and either offered advice that was perceived as unhelpful, such as advice to increase exercise, or were told implicitly or explicitly that the condition was psychological or falsified. There were many ways in which participants felt that the condition was trivialised by others and in doing so, their experience was trivialised and dismissed

> 'My auntie...she always said 'well it's all in the head really isn't it?' (Mother, Missen et al., 2011, p509)

> 'I would have to explain it to people and they would say 'Yeah, but you know, if she just got up and did something surely you know, oh you know, her muscles might be bad, but they would be if she sat in bed all day, do you know what I mean? That sort of thing. And that's hurtful to her and me really.' (Husband, Catchpole & Garip, 2019, p4).

In addition to the limited medical understanding of the condition, the lack of understanding was sometimes felt to be due to the invisible nature of the condition. This was exacerbated by the fact that others saw the person with ME/CFS at moments when they were more well.

> 'We were only able to see them on [my son's] "good days" so of course they never saw that he paid for those days with weeks of "bad days" … There are people that struggle to believe [he] is unwell as they do not see him unless he's well enough to do so.' (Mother, Mihelicova et al., 2016, p2829)

Lack of understanding from healthcare professionals was also reported. Having a sympathetic GP was described as 'lucky' (Wife, Brooks et al., 2014, p10). Some felt that as carers/relatives, they were assumed not to have any medical knowledge and therefore were dismissed by healthcare professionals, who saw no benefit in explaining anything to them.

This scepticism and disbelief from friends, family and healthcare professionals was very upsetting to participants and added to the already difficult burden of their family member being chronically ill. They felt that their relative was being accused of malingering and some spoke about wanting to do something about it. They sometimes felt an obligation 'to defend the legitimacy of the illness' (Catchpole & Garip, 2019, p5). This was sometimes fuelled by anger.

> 'I was so angry, I felt like coming … and reading the riot act' (Wife, Horrocks & Ward, 2015, p98)

> ‘I try and tell two people a week who’ve got no idea about it’ (Mother, Catchpole & Garip, 2019, p6)

There were also times when participants had their own doubts about whether their relative was genuinely ill.

> ‘She might use her sickness or whatever as an excuse. Because I see how she acts with other things that she does. Things that matter to her, she can jump up and run and do this and be gone for 4 hours at a time while we’re watching the kids. And then when I’m going out, she’s like, “Well, you can’t do it”. And then it’s like a big deal. And it’s like, oh she can’t do this now. And maybe, like, maybe she’s just lying.’ (Son, Donalek, 2009, p336)

> ‘Well, I didn’t feel, like, sorry for him because he’s always been lazy, and I didn’t know if it was genuine or not’ (Sibling, Velleman et al., 2016, p625)

Coping with uncertainty. Coping with the uncertainty that living with a relative with ME/CFS brings is explored in this subtheme. There is uncertainty around diagnosis, prognosis and chances of recovery, and uncertainty in day-to-day life given that the condition fluctuates. This meant people felt like they were living ‘in a limbo balanced between previous aspirations and current realities.’ (Father, Mihelicova et al., 2016, p2831). At times, relatives were able to hold onto the hope that their family member’s health would improve or that they would completely recover. At other times, it felt as though things may remain difficult forever. Expectations of daily life and of the future were central.

> ‘Carers believed it was important to accept their day-to-day caring and were in various stages of acceptance, which appeared to involve acknowledging their situation, finding new routines and setting realistic expectations.’ (Catchpole & Garip, 2019, p8).

> ‘Several carers said that they stopped fantasising about the future but accepted whatever happened. These carers appeared to be content, as they did not have unrealistic aspirations.’ (Ax, Gregg, & Jones, 2002, p39).

It was not clarified what was meant by ‘realistic’ expectations in the above studies. Often, acceptance seemed to be a form of resignation and was associated with a loss of hope of improvement. For some, coping with this prospect was very difficult, and several people spoke about the severe impact on their mental health. This was not always easy for people to see in themselves, one participant only realised the impact it was having on him when he went to the GP.

> ‘She gave me a little booklet to look at and tick the boxes, if you tick four or more it’s very likely that you have stress and I ticked nearly thirty.’ (Husband, Horrocks & Ward, 2015, p95).

A variety of coping strategies were used by relatives. Some put a great deal of planning into ensuring they had a small amount of time to do their own activities. Knowledge and information were recognised as being key to dealing with daily life, alongside flexibility in responding to the needs of their family members. Often, it was the relationship with the person who was ill that helped the person cope.

> 'I tended to see us as a unit rather than (myself) as a unit and [name of sufferer] as a separate unit... We are a unit, the family is the unit... And what has to be done has to be done... Not really anything you can do. You struggle on and get through as best as you can.' (unspecified relative, Ax, Gregg & Jones, 2002, p37)

Participants were given very little information about the condition from healthcare professionals so sought information from patient support groups and online forums. While they found information here aimed at supporting the person with ME/CFS, specific information and support for relatives and carers was lacking. Through the information they found, couples or family units played a significant role in co-creating their own understanding of their experience.

> 'The important role that significant others play in helping the patient to make sense of their condition and in formulating an explanatory narrative to account for symptoms is clear in all participants' accounts.' (Brooks, King & Wearden, 2014, p9)

Loss

Loss was experienced in three main ways. The first subtheme, 'Loss of the person they were before their illness' explores the grief relatives experienced in response to the changes in their family member. The second subtheme of 'Loss of identity' looks at the losses the participants described in different roles in their lives. This included losing careers, losing futures they had hoped for and losing social lives.

Loss of the person they were before their illness. This subtheme explores participants' experience of feeling they had lost the person who had become ill. While they were still physically present, and in many cases able to engage in conversation and some activities, relatives felt the loss of who they had been prior to illness. Many described the 'real' person, as distinct from how they were whilst ill. The absence of the ill relative in participants' lives was described in terms of families not being able to do things together as a family in the way they had prior to illness.

> 'Patients and significant others clearly distinguished in their talk between the 'real' person (a healthy and active individual) and the current individual who is limited by their illness condition. The sense is that this person, currently limited by CFS/ME, is not who the patient really is.' (Brooks, King & Wearden, 2013, p11)
>
> 'The woman I married was very active. And we were both very active. We did a lot of walking, we did a lot of, um, well we were both Scout leaders, we did a lot of camps

> and things like that as well, and something like, everything that we used to do together that was us, has changed and gone.' (Husband, Catchpole & Garip, 2019, p7)

Many studies reported that this experience of loss was 'almost reminiscent of bereavement' (Brooks, King, & Wearden, 2014, p12). As with bereavement, this loss triggered many other responses. Relatives described the sadness and fear of seeing their family member so ill.

> 'You've got a beautiful person that you gave life to who is just wasting away here, in isolation, and it breaks your heart, it breaks your heart.' (Mother, Catchpole & Garip, 2019, p7).

> 'She was so still and her face so grey that her younger brother commented that she looked dead. How I feared she would die. How I prayed that every silent, shallow breath would not be her last.' (Mother, Mihelicova et al., 2016, p2832)

Relatives had to manage their feelings of loss finding a way to continue with their own lives.

> 'So it makes you kind of feel guilty, you know, every now and again. Like when I learnt to surf this summer, it was one of those things that we were going to do together.' (Brother, Velleman, 2016, p6271)

Relatives, therefore, were often stuck between wanting to hold on to who their family member had been prior to illness but also needing to continue in daily life with their family member's current functional ability.

> 'It seems that patients and significant others are treading a fine and often shifting line between protecting and honouring the 'true' identity of the patient, whilst at the same time coming to terms with the patient's current identity as defined in terms of their present abilities and limitations imposed by their illness condition.' (Brooks, King & Wearden, 2013, p12)

Loss of identity. The demands of caring for their relative often resulted in participants losing roles they had previously occupied. This subtheme looks at the loss of previously valued identities or hoped for identities. Participants lost roles they had previously had such as needing to give up employment. Relatives experienced loss of aspects of their own identity when caring responsibilities left less time for previous activities.

> 'I retrained and pieced together a second career that could be continued sporadically and if necessary, at home, but that too had to be abandoned as she was now too ill. I'd enjoyed listening to music, but that became impossible; the house must be quiet and I must be ever vigilant and attuned to respond to a faint call or a crash if she passed out' (Mother, Mihelicova, 2016, p2830)

Living with someone with chronic illness had a profound effect on the person's sense of identity. Many people perceived their lives very differently because of it, in the present and in the future.

> ‘I look at friends and colleagues around the office, and what they’re doing, they’ve got a totally different life to what I’ve got.’ (Husband, Catchpole & Garip, 2019, p6)
>
> ‘I always assumed I would have children, you know, I love being an uncle and that’s a great source of joy for me, you know, having nieces and nephews that I could spoil. But, you know, I never really had the choice to have children … um… I think that’s been a difficult thing.’ (Partner, Catchpole & Garip, 2019, p6).

Loss of social life occurred for two main reasons. Firstly, there were practical restrictions from being required to care for their family member and from their family member finding it difficult to tolerate having others in the house because of sensory sensitivities to noise and smells.

> ‘We cannot invite people back to our house because their everyday detergents and smells precipitate excruciating head-pain for [my daughter], but neither are we free to visit them. Our concerns cannot be shared by those who do not understand a situation that doesn’t make sense unless experienced and cannot be experienced by others without making her more ill. So we are met with disbelief and incomprehension.’ (Mother, Mihelicova, 2016, p2830)

Secondly, there was also the decision to withdraw from social contact because of the lack of understanding and negative judgement previously described.

> ‘None of our friends. None of our friends were supposed to know this, it was a secret. Nobody’s supposed to know that he has chronic fatigue. Because if that got out, we might lose clients.’ (Partner, Donalek, 2009, p336)

Participants would rather withdraw than be further exposed to the stigma they had experienced. This often meant they were dealing with the stressors discussed in preceding themes without the social support they had previously had available to them.

Discussion

A systematic review and metasynthesis was undertaken to explore the experience of living with someone with ME/CFS from the perspective of relatives. Findings were described in three superordinate themes. The first theme, 'Changes in relationships', was concerned with the ways in which relationships within family units often changed to centre the needs of the ill person, leading some relatives to have a very close relationship with the person who was ill while others felt pushed to the edges of family life. The second theme of 'Coping with an uncertain and misunderstood condition' explored the specific impact that comes from ME/CFS being a condition that continues to be contested. The third theme of 'Loss' emphasised that relatives of someone with ME/CFS experienced loss in multiple ways. Findings will now be considered in relation to existing literature, prior to consideration of review limitations and clinical implications.

Relationship to previous literature

The changes found to occur within family relationships are consistent with previous research into the impact of chronic illness on the family system. The reorganisation of the family structure to centre and accommodate the needs of the person who is ill echo the work of Rolland (1987, 2013). The centripetal force of a family member being ill had differential impacts on these participants, where some felt able to cope and others felt they had lost most of their identity. Further work could consider whether the developmental phase of the family in which ME/CFS occurs mediates the degree to which family members feel they have been able to cope.

There was an indication that taking on new roles within the household was a source of stress to some participants. It is possible, although as yet unexplored, that this is particularly challenging when the person is required to perform roles that they had not previously expected to undertake because of the role they had held within the family unit, traditional gender roles etc. Gender differences in coping among carers for people with dementia were explored in a review by Baker & Robertson (2008) although this review found that individual differences in coping styles may have more explanatory power. It would be useful for future work to explore how the experience of a member of the family being ill with ME/CFS differentially affects the individual based on their family role, gender, and coping strategies.

There is a significant body of literature relating to the impact of 'caregiving' and being a 'carer' for a chronically ill family member. The findings here relating to ME/CFS are reflective of findings relating to other diagnostic terms. Fulfilling these roles can affect a person's physical and mental health (Hirst, 2004). It is important to note, therefore, that very few of the studies included in this review reported people referring to themselves as 'carers'. There are several possible reasons for this. The process of coming to define oneself as a carer has been found to be a largely discursive process, where the position of 'caregiver' is constructed through contact with outside others (O'Connor, 2007). Given the number of families in these studies with reduced social contact, it is unsurprising that this process did not occur.

The emotional impact of living with a relative with ME/CFS reported in this review is consistent with previous work exploring caring for a relative with a long-term health condition. For example, Vassilev et al. (2013) consider the work of caring for a relative under three categories, derived from earlier work by Corbin and Strauss (1985). The first two are 'Illness (specific) work' of making appointments, managing medication regimens, etc., and 'Everyday work' that includes housekeeping, shopping, personal care. The third category of 'Emotional work' notes the work involved in comforting and supporting through anxious times, being a companion, and navigating changing expectations of self, relationships, and future. Those caring for a relative tend not to associate this third area of work as part of the caring role and so this forms another reason that they are less likely to consider themselves to be carers (Knowles et al., 2016). This effect may be amplified in a condition such as ME/CFS where there may be less 'Illness work' than with other conditions. Not identifying as a carer further impacts the wellbeing of people fulfilling caring roles as they are less likely to access services that support carers due to believing that these services are not for them (Brodaty, et al., 2005).

Strengths and limitations

This review aimed to explore the experiences of family members of people with ME/CFS. The in-depth analysis of qualitative research gives voice to this seldom heard group. The limited number of studies included in this review highlights that this is an area where there is much to be learned. The inherent bias in recruiting to studies must be considered when interpreting these studies. Most participants reported being broadly supportive of their family member, with very few describing feeling negative towards the person with ME/CFS. Those who query the veracity of the condition, do not want to engage in supporting the person with ME/CFS or who feel that they have not been impacted by it are less likely to choose to engage in research of this nature.

It is indicative of an endemic issue within current Western research that none of the studies stated the ethnicity or cultural background of the participants, or in the interest of reflexivity, of the authors (Appendix C contains information concerning my position as reviewer to support readers in contextualising my interpretations of the original studies). It is therefore not possible to state from this review whether these experiences are common across people from all backgrounds. Given that significant enacted stigma is still encountered in relation to ME/CFS, it may be useful to consider how the diagnosis intersects with other stigmatised or marginalised groups of people. As an example, LGBT people with a variety of (non-HIV related) long term health conditions reported feeling isolated from LGBT communities and discriminated against by healthcare professionals (Jowett & Peel, 2009). Further work is needed to understand how this impacts partners and family.

Clinical implications

The key finding of this review indicates the inclusion of the family system when working with people with ME/CFS. Family members are co-creators of explanatory narratives and key supporters in implementing

any interventions or management approaches. By including them in therapeutic approaches, their positive effects can be fully harnessed and any potentially negative impacts can be mitigated (Ward, 2012). This finding can be extended to many other conditions, particularly where the person and their family may experience invalidation from others and therefore support each other in developing their own narrative.

Including family members in the therapeutic process must be balanced with relatives' own needs. Support may be required in the form of practical or psychological help. Language should be used carefully when offering this support. Using terms such as 'carers' may lead many people to assume that the support is not for them. Instead, support should be offered on the basis of a family member having ME/CFS, as this review has demonstrated that this can be difficult for those involved in caring activities and for those who feel pushed out of the family. Family therapy may be appropriate in some cases where the family is trying to adapt to the new dynamics (Blazquez & Alegre, 2013).

Conclusion

This review has explored the experience of having a relative with ME/CFS. Key themes were found around the changes that occur within all relationships within the family, the difficulties that occur due to ME/CFS being frequently misunderstood and the many types of loss that relatives go through. These experiences are common to the different relatives represented in these studies, including spouses or children of people who are ill and parents and siblings of children who are ill. These findings demonstrate that the impact of ME/CFS on the family system surrounding the person who is ill is significant and requires attention from healthcare professionals and researchers.

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Part 2: Research Report

‘Perspectives of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis: A Q-methodology study’

‘Perspectives of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis: A Q-methodology study’

Abstract

Background

Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome (ME/CFS) is characterised by fatigue that is exacerbated by activity. The aetiology is currently unknown. Debate around the relative contribution of different causal factors is often presented as polarisation between those who hold that it is a predominantly physical condition and those who hold it is predominantly psychological. This polarisation extends to views on intervention, where there is ongoing debate about the efficacy of different approaches.

These uncertainties lead to difficult experiences for people with ME/CFS and their families who often experience stigma in relation to the diagnosis. Healthcare professionals and people with ME/CFS have both reported finding that the interaction can hold some tension, particularly when different views on the condition are held.

Method

This study used Q-Methodology as a means to explore the many viewpoints that are held about ME/CFS. A set of statements was developed with the aim of being representative of the many perspectives on the topic. Participants were purposively recruited to represent a range of views and included people with ME/CFS, people who have recovered, their friends and family and healthcare professionals who work with people with ME/CFS. The 40 participants were each asked to complete a Q-sort, placing these statements in a quasi-normal grid according to their relative level of agreement with the statements. A factor analysis was conducted to identify patterns within these responses. The resulting factors represent a shared viewpoint among those participants who load significantly onto each factor. These were interpreted using qualitative data collected during the Q-sorting process.

Findings

Three factors were identified in the data. Factor 1, ‘A debilitating physical health condition,’ placed emphasis on the physiological aspects of the condition, the severity of symptoms and the lack of understanding from others. Factor 2, ‘The mind affecting the body,’ placed more importance on the role of vulnerability to stress and emotional issues. Factor 3, ‘Management is key to recovery,’ stressed the value of lifestyle management approaches, supported by the healthcare professionals.

Discussion

Three distinct viewpoints on ME/CFS were identified within the data. However, many points of similarity existed between them including a belief in the legitimacy of the experience for the person with ME/CFS and a belief in the role of multiple factors, including physiological and psychological, in the condition. It is hoped that these findings can support all stakeholders in reflecting on the positions they and others hold and the similarities as well as differences between these so that this may support constructive dialogue.

Background

Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome (ME/CFS) is a long-term health condition. It is estimated to have a prevalence rate of between 0.2-0.4% (Nacul et al., 2011). It is characterised by fatigue that is exacerbated by activity and is not alleviated by rest or sleep. Fatigue can be accompanied by a wide variety of physical and cognitive symptoms, that may include post-exertional malaise lasting more than 24 hours, concentration or memory difficulties, sore throat, tender lymph nodes, muscle pain, multi-joint pain, headaches, and unrefreshing sleep. At least of one these symptoms, in addition to prolonged unexplained fatigue, must be present for at least four months (three months in children), for diagnostic criteria to be met (NICE, 2014). Diagnosis is usually made by a primary care physician, typically a GP, by the exclusion of other potential causes of symptoms. However ME/CFS remains a contested construct. There is debate as to whether the symptoms constitute a standalone syndrome, with some arguing that ME/CFS is better understood within the broader umbrella of persistent physical symptoms (Schröder, 2010).

The aetiology of ME/CFS is currently undetermined (Jason, Helgerson, Torres-Harding, Carrico, & Taylor, 2003). Over the last 40 years, there has been evidence put forward implicating the role of viral infection (e.g. Hickie et al., 2006; Mikovits et al., 2010), immune system dysfunction (Bradley, Ford, & Bansal, 2013), autonomic nervous system dysfunction (Nijs & Ickmans, 2013) and genetic risk factors (Dibble, McGrath, & Ponting, 2020), but no findings have been reliably replicated.

In parallel to biomedical research, a psychological perspective on the symptoms has been explored. There is some evidence that people with ME/CFS have cognitive biases in their interpretation of somatic information (Hughes, Hirsch, Chalder, & Moss-Morris, 2016), although it is unclear whether these develop as a coping strategy that function as a maintaining rather than precipitating factor. Having experienced childhood trauma, particularly sexual or emotional abuse, increases the risk of developing ME/CFS, with additional posttraumatic stress increasing the risk even further (Heim et al., 2009).There is overlap with presentations of depression and anxiety and ME/CFS which has led to people being given a psychiatric diagnosis when a diagnosis of ME/CFS would be more appropriate (Deale & Wessely, 2000). Anxiety and depression have been found to be present in around half of people with ME/CFS (Caswell & Daniels, 2018), a comparable prevalence to people with other chronic conditions (Campbell-Sills et al., 2013).

The uncertainties that exist about the aetiology of ME/CFS cause many difficulties for people with ME/CFS, their families and healthcare professionals working in the field. These difficulties and the many perspectives held on them are discussed below with reference to the interventions currently offered, the impact of uncertainty on people with ME/CFS and the impact on the relationship between them and healthcare professionals.

Intervention

Given the debates that exist about the construct and causes of ME/CFS, it is not surprising that debate also exists with regards the focus of intervention. UK guidelines (NICE, 2007) specify guidance for management

at primary and secondary levels. It is specified that general practitioners should 'acknowledge the reality and impact of the condition and the symptoms'. Primary care management should include symptom management, advice around healthy diet and sleep patterns and relapse planning. Pacing is also included as a self-management approach that many people with ME/CFS report to find useful, although there is not yet clear evidence of its utility. Referral to specialist care services should be offered within six months, or sooner if the person is severely affected or under 16 (ibid).

The guidelines state that interventions offered at secondary care level should be delivered by specialist services. Guidance relating to specific interventions is under review, with revised guidelines due to be published in August 2021. The three approaches recommended since the introduction of the guidelines (NICE, 2007) are Cognitive Behavioural Therapy, Graded Exercise Therapy and Activity Management. There is some evidence that CBT can improve symptoms, however the theoretical model for this is contested (Geraghty & Blease, 2018). One rationale for offering CBT is that it supports the person to accept their condition and may help associated low mood. Alternatively, those who propose illness beliefs as a perpetuating factor assert that CBT can directly improve symptoms. Graded Exercise Therapy is based on the deconditioning model of ME/CFS and uses 'gradual and planned increases in physical activity… encouraging the participant to extend their physical functioning beyond their current ability' (Bavinton, Darbishire, & White, 2004, p20). Some trials have found it to be moderately effective in improving symptoms but there has been much controversy around the validity of these findings (Vink, 2016). Activity management is described by NICE as a 'goal-oriented and person-centred approach' where physical cognitive and emotional activity is planned and balanced with appropriate levels of rest (NICE, 2007). There is evidence that some people with ME/CFS find this approach helpful (Pinxsterhuis et al., 2015).

Impact on individuals, relatives, and healthcare professionals

Research to date into the experiences of people living with ME/CFS, their relatives and healthcare professionals, has shown it has a multitude of impacts. Many qualitative studies have looked at the impact of the illness on the individual. It can be confusing for people with ME/CFS to learn how to understand their condition, as described by an article titled 'If the illness is not visible to others, does it exist?' (Winger, Ekstedt, & Helseth, 2013). In a metasynthesis by Anderson *et al.* (2012), a process of disruption of self-perception was described. This challenge to the person's identity results in the need to build a new identity, incorporating the new limitations (Arroll & Howard, 2013). A systematic review by Drachler et al. (2009) looked at the expressed needs of people with ME/CFS. One of the first priorities for people in this study was the need to make sense of their symptoms and receive a diagnosis. Drachler et al. stated the importance of feeling respect and empathy from healthcare providers and receiving information about the condition and strategies for managing it.

The ambiguity around the aetiology of ME/CFS can lead to difficult experiences for people with these conditions. This can include friends and relatives questioning the validity of their symptoms and scepticism

from professionals (Dickson et al., 2007). Lian and Robson (2017) found that many patients had to fight for healthcare professionals to believe that they felt ill. One participant reported that her doctor believed she had a mental health condition and therefore refused to investigate her symptoms.

The impact of the condition is felt by those around them, with increased levels of distress among significant others (Harris et al., 2016). Family members who become carers report a similar process to the person who is ill, also developing a new identity for themselves in relation to the condition (Mihelicova et al., 2016). Participants described the disbelief they were met with from the medical community, friends, and relatives. This resulted in feeling socially disconnected. They also reported uncertainty around diagnosis and therefore uncertainty in prognosis and symptom management. Brooks, King and Wearden (2014) found that for couples where one partner had ME/CFS, interacting with outside others was a key issue where they often experienced attitudes as trivialising of the condition and felt that responsibility was attributed to personal factors such as laziness.

Healthcare professionals also report uncertainty when working with people with ME/CFS (Marks, Huws, & Whitehead, 2016). While all participants reported a belief that there are both physiological and psychological contributing factors, some participants placed greater emphasis on an as yet undiscovered physical cause, while others emphasised the function of symptoms, for example avoidance of activity. Chew-Graham et al. (2009) found a variety of views held by nurses regarding the aetiology of the condition. While some held pejorative views towards patients, most viewed it as a multi-faceted condition and had sympathy for patients. Several studies highlight that healthcare professionals feel they lack knowledge and experience of the condition, for example Peters et al. (2011) found that practice nurses being asked to work with this patient group for the first time were impacted by feeling that they were not experts, particularly given that this group of patients have typically developed a high level of knowledge in their condition and often question the expertise of the professionals they are working with.

Research Focus

The many perspectives held about ME/CFS, on aspects including aetiology and intervention, has led to experiences and interactions that can be uncomfortable for all involved. This study set out to explore different ways of conceptualising ME/CFS and the way these attitudes group together.

Aims and rationale

Previous research demonstrates that the many ways that this condition can be conceptualised has significant impact on the individual with the condition, friends, family, and healthcare professionals. It was hoped that deepening our understanding of ways of thinking about this condition can inform the debate around it. The chosen methodology (described in 3.1) is well suited to identifying areas of commonality between different sets of attitudes and allows conflicting sets of attitudes to be presented in positively connoted ways. Thus the outputs may be used to support more constructive dialogue within a subject area in which debates are often highly polarised (Goudsmit & Howes, 2017).

Previous research indicates that there are a range of attitudes and beliefs held about ME/CFS. There is also evidence indicating that beliefs held by others impacts on the people with the condition and on interactions between people with ME/CFS and healthcare professionals. This research therefore aimed to;

- Develop understanding of how these attitudes and beliefs group together
- Further dialogue about the condition between different stakeholders and between people who hold different views
- Support healthcare professionals in reflecting on their interactions with people living with ME/CFS and their families.

The key research question is therefore

- What are the ways in which ME/CFS is conceptualised?

Method

Design

Q-Methodology was selected as an appropriate methodology for addressing this question. It is a mixed methods approach, sometimes described as the 'quantification of qualitative data' (Shemmings, 2006, p147). First devised by Stephenson in 1935 (described in 1953), Q-methodology sets out to scientifically study human subjectivity. Its purpose is to 'reveal subjective structures, attitudes and perspectives from the standpoint of the person or persons being observed.'(Brown, 1996). It has two distinctive features, the 'Q-sort' and a 'by-person' factor analysis, described below. The Q sorting process is distinct from other data gathering processes, such as interview, in the bounded nature of responses that participants are invited to give. This research took a critical realist approach (see Appendix C).

Q-methodology is conducted in five stages, outlined below. Ethical approval was granted by the University of Leicester prior to beginning development interviews (see Appendix D).

Developing the Q-set

The Q-set comprises a set of statements that are representative of the concourse of ways of speaking about a particular phenomenon, in this case, perceptions of ME/CFS. These statements were derived from extracting statements from published literature (peer-reviewed research, publications from charities, and media articles) and through interviews with people with ME/CFS (n=2), people who had recovered from ME/CFS (n=3), someone with family members with ME/CFS (n=1) and healthcare professionals specialising in working with people with ME/CFS (n=4). Interviewees were recruited through the researcher's personal network and healthcare professionals were recruited via a private chronic fatigue syndrome clinic. After receiving the participant information sheet (see Appendix E) and completing a consent form (see Appendix F), interviews were conducted based on a semi-structured interview and then a review of the Q-set, which was developed iteratively in parallel with interviews. Through this process, a large number of statements were generated and the set refined based on feedback from these interviews, until a point of saturation was reached where few suggested changes arose from the later interviews. Thematic grouping was used to ensure a balanced coverage of relevant topics and to remove statements with similar or inverse meaning (see Appendix G for full description of this process). This resulted in a Q-set of 64 statements (see Table 2).

Participants

Participants were required to be over 18 and fluent in English. They were required to have some knowledge of ME/CFS in at least one of the following ways: (a) having CFS/ME, (b) being recovered from CFS/ME, (c) being friends, relatives, or partners of people with CFS/ME, (d) being a professional who works with or has worked with people with CFS/ME.

Strategic sampling was used as the aim in Q-methodology is to gain the input of a wide variety of viewpoints. This is because in Q-methodology, the participants function as variables rather than as a

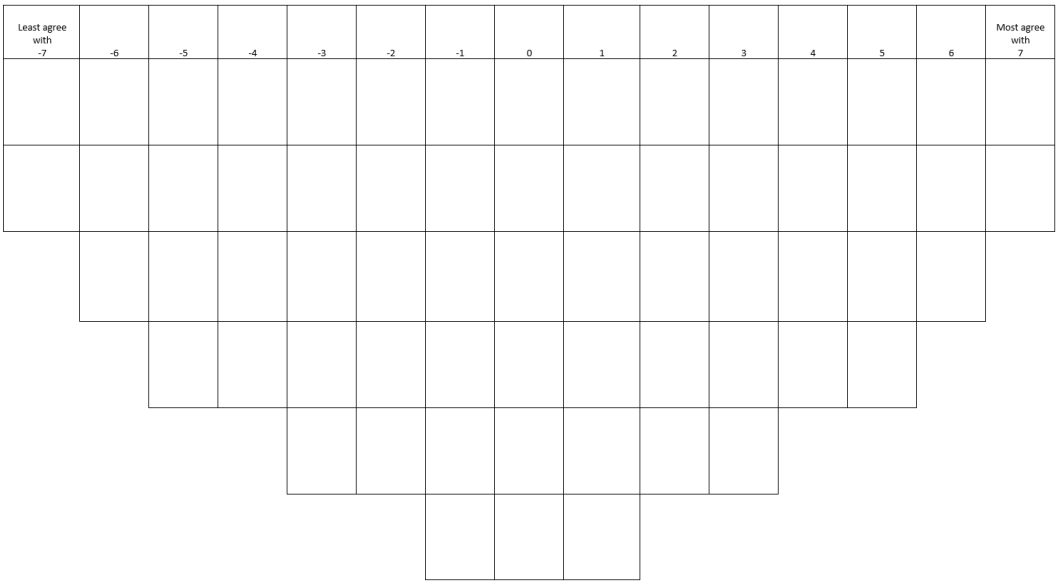
representative sample as they would in more traditional research methods. In addition to inviting those who had already taken part in development interviews, the following methods were used for recruitment.

- Adverts via charities that support people with ME/CFS and professionals that work with them.
- Research groups with an interest in ME/CFS
- Complementary and alternative medicine practitioners who work with people with ME/CFS
- Snowball sampling, where participants were invited to refer others with target viewpoints to the study
- The researcher's personal network

Q-sorting

All Q-sorts were conducted online using the VQMethod website (Nazariadli, Morais, Supak, Baran, & Bunds, 2019). Each participant was given the opportunity to read the participant information sheet (see Appendix H) and completed an online consent form ahead of participation (see Appendix I). Nineteen sorts were conducted during a live video call with the researcher to allow for ongoing conversation during the sort. These conversations were audio recorded and used to aid factor interpretation (see 3.2.5). 21 were conducted by the participant without the (virtual) presence of the researcher. The website took the participant through three stages of the sort. Participants were first asked to place statements into three groups – 'agree', 'disagree' or 'neutral'. In the second stage, the statements were placed on to a forced-choice quasi-normal grid with 64 spaces (see Figure 1), where statements they agreed with most were on the right and statements they agreed with least were on the left, from -7 to +7. Finally, participants were asked to comment on their responses to statements and their reasoning for the order they had chosen. Three participants who completed the sort without the researcher, who were later found to be factor exemplars, were interviewed by the researcher at a later date to gather further qualitative data to support interpretation.

Figure *1*: Grid used for Q-Sort



Factor analysis and interpretation

Q-methodology uses by-person factor analysis to identify patterns in the arrangements of the participants' Q-sorts. This analysis results in 'factors'. Each participant's association with each factor is calculated by their correlation with that factor, known as the factor loading. Participants with similar patterns of how the statements were arranged are identified. These patterns are represented by an idealised Q-sort, calculated using weighted averages of the significantly loading sorts. Analysis was conducted using the KenQ software package (Banasick, 2019).

These factors were interpreted following the procedure outlined by Watts and Stenner (2005). For each factor, the idealised Q-sort was assessed and statements ranked highest and lowest identified. Distinguishing statements, where the average ranking for that factor is significantly different to any other factor, are also identified. A holistic examination of these statements was conducted, with reference to the additional qualitative data from exemplars (participants loading significantly onto a given factor only), to develop a written summary of the viewpoint represented by each extracted factor.

Findings

Summary of participants

Forty-three participants completed the Q-sort. For three of these, data did not record correctly, and it was not possible to correct this, so they were excluded from analysis. Demographic information as collected to situate the sample. One participant did not disclose demographic information. Of the remaining 39 participants, 31 were female and eight were male. Ages ranged from 27 to 73 years (mean= 50.1 years, SD=14.6). Fifteen were healthcare professionals, 12 were ill with ME/CFS, six were recovered from ME/CFS and six were relatives of someone with ME/CFS. Thirty-five participants identified as White British. Nineteen sorts were completed via video call with the researcher and 21 were completed in the participants' own time. Details can be found in Table 1.

Table *1*: Participant demographics

Participant code	Relationship to ME/CFS	Factor 1	Factor 2	Factor 3
1	Recovered	**.47**	.19	**.49**
2	Recovered	**.34**	.29	**.62***
3	Recovered	**.58**	.29	**.45**
4	Currently ill	**.36**	.16	**.48**
5	Currently ill	**.46**	.28	.05
6	Relative	.19	.11	.20
7	Recovered	**.37**	.17	**.41**
8	Healthcare Professional	.19	.26	**.58***
9	Currently ill	**.56**	-.05	**.41**
10	Healthcare Professional	**.37**	**.37**	**.41**
11	Healthcare Professional	**.70***	-.23	.29
12	Healthcare Professional	.19	**.54**	**.58**
13	Healthcare Professional	**.34**	.05	**.51**
14	Currently ill	**.67***	.28	.22
15	Healthcare Professional	**.44**	.28	.27
16	Healthcare Professional	.23	**.53***	.28
17	Currently ill	-.06	-.01	**.52***
18	Healthcare Professional	**.34**	.09	**.61**
19	Currently ill	**.50**	-.07	**.57**
20	Currently ill	**.49**	.29	**.39**

21	Recovered	**.56**	.16	**.34**
22	Relative	**.51**	.06	**.75**
23	Currently ill	**.68***	.19	.32
24	Healthcare Professional	.30	**.45**	**.40**
25	Relative	**.56**	.16	**.40**
26	Recovered	**.48**	.11	**.66**
27	Currently ill	**.73***	.31	.18
28	Currently ill	**.63***	.12	.25
29	Healthcare Professional	.30	.24	**.73***
30	Healthcare Professional	**.57**	.02	**.48**
31	Currently ill	**.50**	-.09	**.61**
32	Relative	**.34**	**.39**	.28
33	Healthcare Professional	-.03	**.57***	.17
34	(not disclosed)	**.49**	-.10	**.53**
35	Healthcare Professional	.14	.25	**.43**
36	Relative	**.53**	.23	.13
37	Healthcare Professional	.25	**.68***	.30
38	Currently ill	**.36**	.18	**.52**
39	Healthcare Professional	**.41**	**.33**	.22
40	Relative	-.01	.25	**.48**

Significant loadings ($p<0.01$) are indicated in bold. Exemplar sorts are indicated by '*'

Factor analysis

The 40 Q-sorts were intercorrelated and a Horst Centroid Factors analysis was conducted, extracting seven factors (Brown, 1980). Five factors were kept for rotation, based on the Kaiser-Guttman criterion of an Eigenvalue greater than one (Guttman, 1954; Kaiser, 1960). A varimax rotation was conducted, followed by hand rotation to maximise exemplar loadings (see Appendix J). Three factors were retained for interpretation, following the criteria of at least two sorts significantly loading at the 0.01 level. These factors accounted for a total of 48% variance. Table 2 shows factor loadings for all participants' sorts.

Table 2: Q-Set and rankings

Statement Number	Statement	Factor 1	Factor 2	Factor 3	Consensus statements
1	Getting a diagnosis helps the person to manage the	4	0	5	

	symptoms of ME/CFS				
2	People who've had ME/CFS are always worried that they will get ill again	2	2	3	C*
3	People with ME/CFS should be advised to just rest	-2	-5	-3	
4	It can be challenging for people with ME/CFS to put lifestyle management techniques into practice	4	1	3	c
5	People with ME/CFS should be encouraged to push through the fatigue	-5	-3	-6	c
6	ME/CFS requires a more holistic approach than most other conditions	0	2	6	
7	Very little is known about ME/CFS	3	-1	-2	
8	There isn't anything that can be done to help people recover from CFS/ME	-2	-7	-7	
9	I sometimes feel I negatively judge people with ME/CFS	-3	-3	-3	C*
10	People with ME/CFS often blame themselves for their symptoms	6	-2	-2	
11	People tend to feel sympathy towards people with ME/CFS	-3	-4	-1	c
12	People with ME/CFS should be offered cognitive behavioural therapy	0	0	1	C*
13	People with ME/CFS often exaggerate their symptoms	-6	-5	-4	C*
14	When people with ME/CFS describe their symptoms, they are usually believed by others.	-6	-5	0	
15	Employers should make adjustments for people with ME/CFS	5	6	7	C*
16	Many people with ME/CFS are significantly disabled by it	4	3	-1	
17	It is better to call the symptoms 'unexplained' than have a diagnosis of ME/CFS	-3	-3	-6	c
18	People with ME/CFS experience fatigue because they are focussed on their symptoms	-3	-1	-5	
19	ME/CFS can be managed with medication	0	-6	-4	
20	People with ME/CFS experience stigma associated with the condition	5	7	5	C*
21	People with ME/CFS should be offered graded exercise therapy	-5	2	-4	
22	Most people with ME/CFS struggle with anxiety	2	5	2	c

23	People with ME/CFS tend to have been very busy people before they became ill	0	5	5	
24	Having ME/CFS is a mindset	-6	-4	-7	c
25	People with ME/CFS tend to need more emotional support than people with other conditions	-1	0	1	C*
26	ME/CFS can provide an identity for the person that is helpful in some ways	1	3	-1	
27	People with ME/CFS tend to do too much	1	1	4	c
28	ME/CFS can be managed by making changes to diet	-2	-6	2	
29	People with ME/CFS are motivated to recover	4	0	6	
30	People with ME/CFS retain a strong interest in other aspects of their lives	3	-2	4	
31	People with ME/CFS are often inappropriately given psychiatric diagnoses	5	-1	0	
32	People with ME/CFS are open to psychological explanations of their symptoms	-1	-2	1	c
33	ME/CFS occurs in people who are vulnerable to the impact of stress	-1	7	-1	
34	Most people with ME/CFS are depressed	0	1	-2	c
35	People stay ill with ME/CFS because of the situation they are in	-1	-3	1	
36	People stay ill with ME/CFS because they are not managing the condition well	-3	-1	3	
37	People with ME/CFS should be offered support with activity management from a healthcare professional	1	1	7	
38	People with ME/CFS show improvement when they are able to change their beliefs about symptoms	-4	6	2	
39	The fatigue experienced by people with ME/CFS is different from a healthy person's fatigue	7	4	6	C*
40	Low mood is a factor that stops people with ME/CFS getting better	-1	1	-2	C*
41	Getting the diagnosis of ME/CFS makes people more likely to behave like they are ill	-5	2	-5	
42	People with ME/CFS tend to set very high standards for themselves	1	4	5	
43	ME/CFS is likely caused by a virus	3	-1	2	

44	People with ME/CFS tend to fear activity	2	0	1	C*
45	ME/CFS symptoms are likely due to weakened muscles	-1	-4	-5	
46	There is likely a physiological cause for ME/CFS	6	-2	4	
47	Most people with ME/CFS believe they will never get better	1	5	0	
48	ME/CFS is an umbrella term that likely refers to a number of conditions	3	-2	-1	
49	People with ME/CFS should be offered counselling	0	-1	3	
50	People with ME/CFS are more likely to have experienced childhood trauma	-4	2	-2	
51	ME/CFS is likely caused by a combination of factors in each person	5	5	3	C*
52	Onset is triggered by a stressful event	-4	0	0	
53	I don't know what causes ME/CFS	3	-5	-3	
54	Emotional issues are likely to be a significant causal factor in ME/CFS	-2	3	0	
55	ME/CFS is likely a mental health condition	-7	-3	-6	
56	My beliefs about the potential causes of ME/CFS often change	1	-6	-3	
57	It is difficult for people with ME/CFS to describe their symptoms	6	6	0	
58	It matters what name is used for the condition	2	1	-1	
59	ME/CFS is difficult to understand for people who haven't experienced it	7	4	4	
60	ME/CFS should be regarded as a 'phenomenon' not an illness	-4	3	-5	
61	CFS/ME affects mostly middle-class people	-5	-7	-4	C*
62	The term 'chronic fatigue syndrome' is the best term for this condition	-2	3	1	
63	Most people experience tiredness that could be called ME/CFS at some point in their lives	-7	-4	-3	
64	People with ME/CFS can make a full recovery	2	4	2	C*

Consensus statements significant at $p<0.05$ are connoted by 'c'. Consensus statements significant at $p<0.01$ are connoted by 'C*'

Table 3 indicates that all factors were positively correlated, with the highest correlation being between Factors 1 and 3.

Table 3: Factor correlations

	Factor 1	Factor 2	Factor 3
Factor 1	1	0.4349	0.6066
Factor 2		1	0.5532
Factor 3			1

Factor descriptions

Factor descriptions for the three factors are given below, provided in this order for narrative purposes. Rankings of relevant statements are provided, such that (#24, +5) would indicate that statement number 24 was ranked at +5 in the idealised factor array for that factor. Statements marked with '*D' are distinguishing statements for that factor indicating that the statement was ranked significantly differently for that factor than for the other two factors. Comments made by participants are included so that the meaning of these rankings can be understood and placed in the context of the broader viewpoint.

Factor 1: A debilitating physical health condition

Factor 1 explains 20% of the study variance. Five participants are significantly associated with this factor. Four were people who were ill with ME/CFS (P14, P23, P27, P28). The fifth was a healthcare professional who worked with people with ME/CFS and had a relative who was ill with ME/CFS (P11). Figure 2 shows the idealised Q-sort for this factor.

From this viewpoint, ME/CFS is a physical health condition (#24, -6; #46, +6; #55, -7), although the exact aetiology is not known (#43, +3; #45, -1*D; #53, +3*D).

'The symptoms couldn't be explained by anything else apart from a physiological cause.' (P11)

There is a greater emphasis placed on the debilitating nature of the symptoms than the other factors. The fatigue is qualitatively different from the tiredness experienced by healthy people (#39, +7; #63, -7*D).

> ‘ME/CFS debilitating fatigue is nothing like the tiredness other people experience… I wish I could give them all a dose so they could experience it because they cannot see anything wrong with you. They usually say, “Oh yes, I get very tired too!”’ (P14)

ME/CFS can be difficult to describe (#57, +6) and difficult for others to understand as it is not something most people experience (#59, +7*D). People are often dismissed and disbelieved when they try to explain the severity of the condition to others (#14, -6) and there has been much stigma associated with the diagnosis (#20, +5).

> ‘You always hear people talking about yuppy flu. I think there’s almost a sense that it’s a made-up condition.’ (P28)

> ‘My experiences of trying to explain my daughter's condition to other people has generally been met with bafflement or assumption she is mentally ill.’ (P32)

There is not a strong opinion about what the condition should be called (#58, +2), although there are some concerns that the term ‘Chronic Fatigue Syndrome’ does not communicate the range and severity of symptoms that can be experienced (#62, -2*D).

> ‘[My] preferred name is ‘ME’ rather than ‘chronic fatigue’ because chronic fatigue doesn’t cover everything of the symptoms that they experience, it just implies that it’s a fatigue thing.’ (P11)

Figure 2: Idealised Q-Sort for Factor 1

-7	-6	-5	-4	-3	-2	-1	0	1	2	3	4	5	6	7
55. ME/CFS is likely a mental health condition	13. People with ME/CFS often exaggerate their symptoms	5. People with ME/CFS should be encouraged to push through the fatigue	38. People with ME/CFS show improvement when they are able to change their beliefs about symptoms	18. People with ME/CFS experience fatigue because they are focussed on their symptoms	54. Emotional issues are likely to be a significant causal factor in ME/CFS	32. People with ME/CFS are open to psychological explanations of their symptoms	34. Most people with ME/CFS are depressed	26. ME/CFS can provide an identity for the person that is helpful in some ways	44. People with ME/CFS tend to fear activity	53. I don't know what causes ME/CFS	4. It can be challenging for people with ME/CFS to put lifestyle management techniques into practice	31. People with ME/CFS are often inappropriately given psychiatric diagnoses	46. There is likely a physiological cause for ME/CFS	59. ME/CFS is difficult to understand for people who haven't experienced it
63. Most people experience tiredness that could be called ME/CFS at some point in their lives	24. Having ME/CFS is a mindset	21. People with ME/CFS should be offered graded exercise therapy	60. ME/CFS should be regarded as a 'phenomenon' not an illness	36. People stay ill with ME/CFS because they are not managing the condition well	3. People with ME/CFS should be advised to just rest	35. People stay ill with ME/CFS because of the situation they are in	49. People with ME/CFS should be offered counselling	47. Most people with ME/CFS believe they will never get better	58. It matters what name is used for the condition	7. Very little is known about ME/CFS	1. Getting a diagnosis helps the person to manage the symptoms of ME/CFS	20. People with ME/CFS experience stigma associated with the condition	10. People with ME/CFS often blame themselves for their symptoms	39. The fatigue experienced by people with ME/CFS is different from a healthy person's fatigue
	14. When people with ME/CFS describe their symptoms, they are usually believed by others.	61. CFS/ME affects mostly middle-class people	52. Onset is triggered by a stressful event	9. I sometimes feel I negatively judge people with ME/CFS	8. There isn't anything that can be done to help people recover from CFS/ME	40. Low mood is a factor that stops people with ME/CFS getting better	6. ME/CFS requires a more holistic approach than most other conditions	27. People with ME/CFS tend to do too much	2. People who've had ME/CFS are always worried that they will get ill again	30. People with ME/CFS retain a strong interest in other aspects of their lives	29. People with ME/CFS are motivated to recover	51. ME/CFS is likely caused by a combination of factors in each person	57. It is difficult for people with ME/CFS to describe their symptoms	
		41. Getting the diagnosis of ME/CFS makes people more likely to behave like they are ill	50. People with ME/CFS are more likely to have experienced childhood trauma	17. It is better to call the symptoms 'unexplained' than have a diagnosis of ME/CFS	28. ME/CFS can be managed by making changes to diet	33. ME/CFS occurs in people who are vulnerable to the impact of stress	23. People with ME/CFS tend to have been very busy people before they became ill	37. People with ME/CFS should be offered support with activity management from a healthcare professional	22. Most people with ME/CFS struggle with anxiety	48. ME/CFS is an umbrella term that likely refers to a number of conditions	16. Many people with ME/CFS are significantly disabled by it	15. Employers should make adjustments for people with ME/CFS		
				11. People tend to feel sympathy towards people with ME/CFS	62. The term 'chronic fatigue syndrome' is the best term for this condition	45. ME/CFS symptoms are likely due to weakened muscles	19. ME/CFS can be managed with medication	56. My beliefs about the potential causes of ME/CFS often change	64. People with ME/CFS can make a full recovery	43. ME/CFS is likely caused by a virus				
						25. People with ME/CFS tend to need more emotional support than people with other conditions	12. People with ME/CFS should be offered cognitive behavioural therapy	42. People with ME/CFS tend to set very high standards for themselves						

Dark red indicates distinguishing statements at p<0.01, pale red indicates distinguishing statements at p< 0.05, green indicates consensus statements at p<0.05

As with other physical health conditions, it is possible that mood can be affected by the experience of living with a chronic physical health condition (#22, +2; #34, 0) but low mood or anxiety are not causal factors of ME/CFS.

> 'If you've got cancer, motor neurone disease, or M.S. you're maybe sometimes going to get depressed and that's going to influence your symptoms too so that's maybe a psychological overlay. If you're depressed, that can affect your appetite for example, so you know, it's no different from any other illness.' (P11)

From this viewpoint, past trauma, current emotional issues, and stress are not felt to be related to symptom onset (#33: -1; #50: -4; 52: -4*D; 54: -2).

> 'I think there are plenty of people who have emotional issues who don't have ME and people who don't [have emotional issues] who have ME.' (P27)

While people loading onto the other factors believed there was a tendency towards some personality traits being associated with ME/CFS, for this factor tendencies like being very busy and having very high standards are regarded as just as likely in people who do not have ME/CFS as in those who do (#23, 0*D; #42, +1*D).

The approach to managing the condition needs to be much like any other physical health condition (#6, 0). It is possible to use medication to help some symptoms (#19, 0*D) but ultimately it is a difficult condition to recover from (#64, +2). 'Most people are able to manage it [ME/CFS] themselves without a healthcare professional' (P11), input from whom can sometimes be unhelpful (#4, +4; #37, +1). Graded Exercise Therapy is harmful and should be avoided (#5, -5; #21, -5).

> 'It is so detrimental to our health as it uses energy we just don't have … Bear in mind the heart is a muscle, and if we have no energy left for our heart to function ...' (P23)

Factor 2: The mind affecting the body

Factor 2 explains 8% of the study variance. Three participants were exemplars of this factor. They were all professionals who worked with people with ME/CFS. One is a health coach (P37), and the other two are healthcare professionals in the NHS (P16 and P33). Figure 3 shows the idealised Q-sort for this factor.

This viewpoint holds that ME/CFS cannot be accurately described as solely physiological (#46, -2*D). ME/CFS occurs in people who are vulnerable to the impact of stress (#33, +7*D). Anxiety is common in people with ME/CFS (#22, +5*D).

> 'I believe anxiety is a major perpetuating factor for people with a diagnosis of ME/CFS as the majority of my patients report symptoms and feelings of anxiety.' (P16)

> ‘In my dealings with people with ME it has become clear to me that there are nearly always discernible predisposing factors in a person’s life - stress in all its forms, especially previous unprocessed emotional/psychological stresses or multiple stressors occurring within a short time of each other just prior to onset of CFS.’ (P24)

There is greater emphasis on the role that emotional issues may play (#54, +3*D) than either of the other factors. People with ME/CFS are believed to be more likely to have experienced childhood trauma (#50, +2*D).

> ‘I do think it’s trauma. Not with a big capital ‘T’ trauma necessarily, but something traumatic being brought up… All of that stuff is very prominent with the people that I’m working with.’ (P37)

The condition is recognised as hard to describe and understand (#57, +6; #59, +4). ‘Chronic Fatigue Syndrome’ is considered the best name for the condition out of the terms that currently exist (#62, +3).

> ‘I like ‘Chronic Fatigue Syndrome’ because that’s what I see. I see someone that’s got enduring fatigue, lower energy levels, so I quite like that.’ (P16)

There is significant stigma associated with the diagnosis (#20, +7). The symptom of fatigue is acknowledged by this viewpoint as being different from a healthy person’s fatigue (#39, +4; #63 -4).

> ‘We all get tired and fatigued at various times in our lives, but we do not experience the additional symptoms that contribute to a diagnosis of ME/CFS.’ (P16)

This factor is the only one that found the term ‘phenomenon’ a potentially useful term (#60, +3*D), as many other terms misrepresent what is being referred to.

> ‘I don’t like the word disease because for me I feel a disease is something pathological… phenomenon is a big umbrella term, so I quite like that one.’ (P16)

It is possible that giving someone the diagnosis of ME/CFS could make them more likely to behave as though they are ill (#41, +2*D), a statement agreed with significantly more than the other two viewpoints. Many people with ME/CFS are perceived as believing they will never get better (#47, +5*D). While the other two factors emphasise that people with ME/CFS *are* motivated to get better, this viewpoint is neutral on this point (#29, 0*D).

Figure 3: Idealised Q-sort for Factor 2

-7	-6	-5	-4	-3	-2	-1	0	1	2	3	4	5	6	7
61. CFS/ME affects mostly middle-class people	19. ME/CFS can be managed with medication	53. I don't know what causes ME/CFS	11. People tend to feel sympathy towards people with ME/CFS	35. People stay ill with ME/CFS because of the situation they are in	48. ME/CFS is an umbrella term that likely refers to a number of conditions	43. ME/CFS is likely caused by a virus	29. People with ME/CFS are motivated to recover	27. People with ME/CFS tend to do too much	2. People who've had ME/CFS are always worried that they will get ill again	26. ME/CFS can provide an identity for the person that is helpful in some ways	59. ME/CFS is difficult to understand for people who haven't experienced it	23. People with ME/CFS tend to have been very busy people before they became ill	15. Employers should make adjustments for people with ME/CFS	20. People with ME/CFS experience stigma associated with the condition
8. There isn't anything that can be done to help people recover from CFS/ME	28. ME/CFS can be managed by making changes to diet	14. When people with ME/CFS describe their symptoms, they are usually believed by others.	63. Most people experience tiredness that could be called ME/CFS at some point in their lives	17. It is better to call the symptoms 'unexplained' than have a diagnosis of ME/CFS	46. There is likely a physiological cause for ME/CFS	49. People with ME/CFS should be offered counselling	1. Getting a diagnosis helps the person to manage the symptoms of ME/CFS	37. People with ME/CFS should be offered support with activity management from a healthcare professional	6. ME/CFS requires a more holistic approach than most other conditions	54. Emotional issues are likely to be a significant causal factor in ME/CFS	42. People with ME/CFS tend to set very high standards for themselves	51. ME/CFS is likely caused by a combination of factors in each person	38. People with ME/CFS show improvement when they are able to change their beliefs about symptoms	33. ME/CFS occurs in people who are vulnerable to the impact of stress
	56. My beliefs about the potential causes of ME/CFS often change	3. People with ME/CFS should be advised to just rest	24. Having ME/CFS is a mindset	5. People with ME/CFS should be encouraged to push through the fatigue	30. People with ME/CFS retain a strong interest in other aspects of their lives	18. People with ME/CFS experience fatigue because they are focussed on their symptoms	44. People with ME/CFS tend to fear activity	4. It can be challenging for people with ME/CFS to put lifestyle management techniques into practice	50. People with ME/CFS are more likely to have experienced childhood trauma	16. Many people with ME/CFS are significantly disabled by it	39. The fatigue experienced by people with ME/CFS is different from a healthy person's fatigue	22. Most people with ME/CFS struggle with anxiety	57. It is difficult for people with ME/CFS to describe their symptoms	
		13. People with ME/CFS often exaggerate their symptoms	45. ME/CFS symptoms are likely due to weakened muscles	9. I sometimes feel I negatively judge people with ME/CFS	10. People with ME/CFS often blame themselves for their symptoms	36. People stay ill with ME/CFS because they are not managing the condition well	25. People with ME/CFS tend to need more emotional support than people with other conditions	34. Most people with ME/CFS are depressed	41. Getting the diagnosis of ME/CFS makes people more likely to behave like they are ill	62. The term 'chronic fatigue syndrome' is the best term for this condition	64. People with ME/CFS can make a full recovery	47. Most people with ME/CFS believe they will never get better		
				55. ME/CFS is likely a mental health condition	32. People with ME/CFS are open to psychological explanations of their symptoms	7. Very little is known about ME/CFS	52. Onset is triggered by a stressful event	58. It matters what name is used for the condition	21. People with ME/CFS should be offered graded exercise therapy	60. ME/CFS should be regarded as a 'phenomenon' not an illness				
						31. People with ME/CFS are often inappropriately given psychiatric diagnoses	12. People with ME/CFS should be offered cognitive behavioural therapy	40. Low mood is a factor that stops people with ME/CFS getting better						

Dark red indicates distinguishing statements at p<0.01, pale red indicates distinguishing statements at p< 0.05, green indicates consensus statements at p<0.05

People who hold this viewpoint do believe that recovery is possible (#64, +4). One component of that is that changing someone's beliefs about their symptoms could help (#38, +6*D).

> 'If you change your way of being and the way you view your illness and the way you view yourself… it's going to make everything so much easier.' (P37)

Medication or diet are not helpful in managing the condition (#19, -6; #28, -6*D). While people associated with this factor believe that it is unhelpful to encourage people to push through their fatigue (#5, -3), this viewpoint is distinct from the others in that Graded Exercise Therapy is cautiously recommended as a potentially helpful intervention (#21, +2*D), if used appropriately.

> 'I like to talk about it as a method of introducing more physical movement. I'm always very honest to say that some people love it, some people absolutely hate it … I don't use it in its pure form at all – much, much, more informally.' (P16)

Factor 3: Management is key to recovery

Factor 3 explains 20% of the study variance. Four participants were significantly associated with this factor. Two were healthcare professionals working in speciliast ME/CFS services (P29 and P8). One was someone who was recovered from ME/CFS (P2) and another was ill with ME/CFS (P17). Figure 4 shows the idealised Q-sort for this factor.

This viewpoint considers ME/CFS to be a physical health condition (#24, -7; #46, +4; #55, -6).

> 'The body is physiologically running out of energy.' (P29)

There is a focus on management as a means to recovery. This viewpoint, more than the other two, holds that getting a diagnosis is felt to be very important as this then enables the person to put management techniques into place (#1, +5; #17, -6*D).

> 'I think it's easier to accept how you feel if you have a diagnosis and you know what you're dealing with. And you can look at ways to either live with it or improve it. Whereas if it's unexplained, you're in limbo.' (P2)

There are no concerns that receiving the diagnosis makes the person more likely to behave as though they are ill (#41, -5), nor do symptoms arise because the person is focussing on them (#18, -5*D). Living with a chronic physical health condition can be difficult and may result in low mood or depression but this is not a cause of the symptoms (#34, -2*D). Symptoms are not seen as particularly difficult to describe (#57, 0*D), as this viewpiont has more of a sense of it being possible to understand and help people with the condition. This viewpoint does not see the condition as being as disabling as the other two (#16, -1), as there is more of a focus on recovery and learning to live well.

It is strongly believed that things can be done to help people recover (#8, -6). Activity management is a key approach to enable people with ME/CFS to improve (#37, +7*D). It is essential that this approach is holistic, more so than with many other conditions (#6, +6*D).

> ‘If people are given the opportunity for early intervention and guidance in managing their condition, they are more likely to make changes in their lifestyle and have better health and therefore continue with their daily tasks and work/education.’ (P29)

> ‘I think it does need a more holistic approach, because you need to look at sleeping patterns, your diet, your environment, what type of exercise you can do, so yeah I think that you do need [that approach], particularly with this, because that’s what’s going to help you to recover.’ (P17)

This approach includes the person learning how to get quality rest in balance with the amount of activity they do (#3, -3). There is a perception that people with ME/CFS often do too much activity (#27, +4*D), and have tendency to have been very busy people before they were ill and set high standards for themselves (#23, +5; #42, +5).

> ‘I have noticed that all my patients are "fast-lane" people. Busy "Go-getters" "high-achievers" with high expectations for themselves.’ (P35)

It is important not to encourage the person to push through the fatigue (#5, -6). Graded Exercise Therapy is seen as unhelpful (#21, -4).

There is no strong opinion about what term is used for the condition (#58, -1; #62, +1). Referring to it as a ‘phenomenon’ is not seen as helpful (#60, -5) as this can perpetuate a dismissive attitude towars it.

> ‘I prefer [Chronic Fatigue Syndrome] to ME… I don’t like that [the term phenomenon]. No, it’s an illness in my opinion.’ (P17)

People with ME/CFS are believed to be motivated to recover (#29, +6).

> ‘[The symptoms are] so awful, they ARE motivated,’ (P2)

Figure 4: Idealised Q-sort for Factor

-7	-6	-5	-4	-3	-2	-1	0	1	2	3	4	5	6	7
8. There isn't anything that can be done to help people recover from CFS/ME	5. People with ME/CFS should be encouraged to push through the fatigue	45. ME/CFS symptoms are likely due to weakened muscles	61. CFS/ME affects mostly middle-class people	56. My beliefs about the potential causes of ME/CFS often change	40. Low mood is a factor that stops people with ME/CFS getting better	33. ME/CFS occurs in people who are vulnerable to the impact of stress	47. Most people with ME/CFS believe they will never get better	35. People stay ill with ME/CFS because of the situation they are in	64. People with ME/CFS can make a full recovery	36. People stay ill with ME/CFS because they are not managing the condition well	59. ME/CFS is difficult to understand for people who haven't experienced it	42. People with ME/CFS tend to set very high standards for themselves	6. ME/CFS requires a more holistic approach than most other conditions	15. Employers should make adjustments for people with ME/CFS
24. Having ME/CFS is a mindset	17. It is better to call the symptoms 'unexplained' than have a diagnosis of ME/CFS	41. Getting the diagnosis of ME/CFS makes people more likely to behave like they are ill	19. ME/CFS can be managed with medication	9. I sometimes feel I negatively judge people with ME/CFS	10. People with ME/CFS often blame themselves for their symptoms	11. People tend to feel sympathy towards people with ME/CFS	31. People with ME/CFS are often inappropriately given psychiatric diagnoses	44. People with ME/CFS tend to fear activity	43. ME/CFS is likely caused by a virus	4. It can be challenging for people with ME/CFS to put lifestyle management techniques into practice	30. People with ME/CFS retain a strong interest in other aspects of their lives	20. People with ME/CFS experience stigma associated with the condition	29. People with ME/CFS are motivated to recover	37. People with ME/CFS should be offered support with activity management from a healthcare professional
	55. ME/CFS is likely a mental health condition	60. ME/CFS should be regarded as a 'phenomenon' not an illness	21. People with ME/CFS should be offered graded exercise therapy	63. Most people experience tiredness that could be called ME/CFS at some point in their lives	34. Most people with ME/CFS are depressed	48. ME/CFS is an umbrella term that likely refers to a number of conditions	52. Onset is triggered by a stressful event	25. People with ME/CFS tend to need more emotional support than people with other conditions	38. People with ME/CFS show improvement when they are able to change their beliefs about symptoms	51. ME/CFS is likely caused by a combination of factors in each person	46. There is likely a physiological cause for ME/CFS	23. People with ME/CFS tend to have been very busy people before they became ill	39. The fatigue experienced by people with ME/CFS is different from a healthy person's fatigue	
		18. People with ME/CFS experience fatigue because they are focussed on their symptoms	13. People with ME/CFS often exaggerate their symptoms	3. People with ME/CFS should be advised to just rest	7. Very little is known about ME/CFS	26. ME/CFS can provide an identity for the person that is helpful in some ways	14. When people with ME/CFS describe their symptoms, they are usually believed by others.	12. People with ME/CFS should be offered cognitive behavioural therapy	22. Most people with ME/CFS struggle with anxiety	2. People who've had ME/CFS are always worried that they will get ill again	27. People with ME/CFS tend to do too much	1. Getting a diagnosis helps the person to manage the symptoms of ME/CFS		
				53. I don't know what causes ME/CFS	50. People with ME/CFS are more likely to have experienced childhood trauma	16. Many people with ME/CFS are significantly disabled by it	57. It is difficult for people with ME/CFS to describe their symptoms	32. People with ME/CFS are open to psychological explanations of their symptoms	28. ME/CFS can be managed by making changes to diet	49. People with ME/CFS should be offered counselling				
						58. It matters what name is used for the condition	54. Emotional issues are likely to be a significant causal factor in ME/CFS	62. The term 'chronic fatigue syndrome' is the best term for this condition						

Dark red indicates distinguishing statements at p<0.01, pale red indicates distinguishing statements at p< 0.05, green indicates consensus statements at p<0.05

Discussion

In this study, Q-methodology was used to explore viewpoints that exist towards ME/CFS. Three ways of conceptualising ME/CFS were found that showed significant differences but also points of consensus. Factor 1, 'a debilitating physical health condition' conceptualised ME/CFS as a physiological condition that is hard for those who have not experienced it to understand. Factor 2, 'the mind affecting the body', focussed on the role of stress and anxiety in the onset and maintenance of symptoms. Factor 3, 'Management is key to recovery', highlighted the importance of receiving a diagnosis to enable the person to access a holistic management approach. There was consensus across all factors, in that they conceptualised the experiences of people with the diagnosis as legitimate. Participants across the three viewpoints conceptualised ME/CFS fatigue as qualitatively different from what is experienced by healthy people, that there is stigma associated with the diagnosis and that employers should make adjustments. The key differences in viewpoints are discussed below with reference to the relationships between beliefs regarding aetiology and perspectives on intervention.

This research was undertaken in the context of previous literature suggestion polarisation in viewpoints regarding the aetiology of ME/CFS (Kean, 2010). The three factors represented viewpoints with different perspectives on this, with Factor 2 holding that the person's vulnerability to stress is a significant causal factor. Factors 1 and 3 placed stronger emphasis on physiological causes. Both factors 2 and 3 were neutral about whether a specific stressful event tends to trigger symptoms, while the factor 1 viewpoint posited that stress was not related to onset.

It is of interest that viewpoint in Factor 1 tended to be held by people who were currently ill with ME/CFS. Factor 2 was a viewpoint that tended to be held by some healthcare professionals and Factor 3 was a viewpoint that was held by a mix of people (healthcare professionals, someone currently ill and someone recovered). Q Methodology does not claim to be representative of any population. These findings are relevant to future work that may be interested in further exploring the derivation of different viewpoints.

These beliefs about aetiology could be linked with the beliefs about intervention held by each viewpoint. The Factor 1 viewpoint was neutral on whether it was possible to aid recovery and held the strongest views against graded exercise therapy, with grave concerns about the impact on the body and pathophysiology believed to be present. Factor 2 is the only viewpoint where Graded Exercise Therapy had some acceptability. In interview, participants did not explicitly link this with beliefs about aetiology so inferences about the link are made tentatively. However, it could be inferred that placing less emphasis on physiological factors leads to a stronger belief in this approach. For Factor 3, a viewpoint that did not particularly emphasise physiological and

psychological factors regarding aetiology, the approaches to intervention that were most favoured related to professionals supporting with a holistic activity management programme, where all physiological and psychological components are addressed and supported.

It is useful to consider these findings within the framework of Levanthal's Common Sense Model of Self Regulation (Levanthal et al.,1997) in which illness representations are a key construct. The differences between the three factors can be seen with reference to the five central illness representations in this model. The first, Identity, is more central in Factor 1, where defining the condition as physical and having that label understood by others was ranked of higher importance than for the other factors. Each factor held beliefs about Cause, the second illness representation, but there were subtle differences in the emphasis placed on physical causes and the role of psychological precipitants. The Illness representation, Timeline, was addressed in this study via beliefs about the possibility of recovery, with Factor 2 holding the strongest belief that recovery is possible. Beliefs about the Consequences of the condition included the consensus statement that people with ME/CFS do experience stigma. The statement that it is a significantly disabling condition was more strongly agreed with by participants loading on Factors 1 and 2, whereas from the point of view of Factor 3 it is possible to learn to live well with the condition. Beliefs about Controllability, the fifth central illness representation, related to beliefs about locus of control.

Health-related locus of control refers to beliefs about the degree to which a person is in control of their health (internal locus of control) or external factors are in control (external locus of control) (Wallston, Wallston, & DeVellis, 1978). An internal locus of control is associated with better health outcomes across conditions (Náfrádi, Nakamoto, & Schulz, 2017), and has been found to have a tendency to be low in people with ME/CFS (Van De Putte et al., 2005). Many of the statement rankings for Factor 1 relate to a low internal locus of control, such as not believing anything can be done to help people recover, that changing beliefs does not improve symptoms and that medication may be useful in management of the condition. The other two factors hold that people with ME/CFS tend to have been busy people who hold high standards. Factor 1 does not ally itself with these beliefs, again placing less agency within the individual. It is interesting therefore to note that this is the only factor that holds that people with ME/CFS tend to blame themselves. There are a number of possible interpretations for this, such as a sense that people unduly blame themselves for symptoms they have no control over or that the self-blame that some people feel results in the need to more strongly reject suggestions that are perceived as blaming of the individual.

There is more evidence of personal agency in the other two factors. Factor 2 holds that a person changing their beliefs about the condition will lead to an improvement in symptoms. From this viewpoint, people with ME/CFS often do not believe it is possible to recover and are not necessarily

motivated to recover. The implication here is that people with ME/CFS are perceived to have an internal locus of control, reflecting the viewpoint found in Factor 1. From the Factor 3 viewpoint, it is strongly believed that the individual has agency to affect their health by implementing management strategies.

It is worth noting that, in this study, the participants who loaded significantly onto Factor 1 were people with ME/CFS and participants who loaded significantly onto Factor 2 were healthcare professionals, while Factor 3 was a mix of both. These differences in perceived locus of control need to be further explored as a potential cause of some of the difficulties that are present in the patient-practitioner relationship (Dickson et al., 2007).

Limitations

There is a potential bias in recruitment in that people who volunteer to take part in research on ME/CFS are much more likely to hold a perspective that validates its existence and empathises with those who have it. Other research has evidenced that many people do not believe in that ME/CFS should exist as a separate construct and would prefer to understand these experiences as an expression of distress. This view is not represented within these findings.

A further bias in recruitment is acknowledged in the predominance of white British participants and under representation of people from racialised groups. Issues of intersectionality, where a person belongs to two or more stigmatised groups, have been explored in relation to people who have ME/CFS and are from racialised groups (Bayliss et al., 2016), indicating that negative stereotypes that exist towards both groups exacerbate attitudes towards people who are in both. Further work into the impact of this type of intersectionality is required.

The correlations between factors, particularly factors 1 and 3 are noted to be high. However it is noted that 28 sorts did not load exclusively onto a single factor. Only one sort loaded onto all three factors and one sort loaded onto no factors. 17 sorts loaded on to factors 1 and 3, reflecting the high correlation between these factors. They met statistical criteria for distinct factors and a qualitative review of each factor found a meaningful difference between the factors.

It is acknowledged that the researcher has personal experience of the issues raised in this study. The epistemological position taken for this research does not attempt to separate person from context, so personal experience may be seen as informative. It can however also be a potential source of bias. Reflexivity was attended to throughout the research process by the researcher using a reflective diary (see Appendices B and J).

Clinical implications

For healthcare professionals working clinically with people with ME/CFS, it may be useful to reflect on the different viewpoints that are present in the interaction. The viewpoint held by the

professional will influence how they discuss management approaches. For example, they may emphasise one aspect of management over another. This may change at different times and with different service users. The viewpoint held by the service user is very relevant in this interaction as it will affect how any recommendations are received and the likelihood of the service user implementing these. It is also important to consider how each person in this interaction perceives the other's viewpoint and the influence this is having on communication. Where the degree of difference between positions held appears large, it may be useful to return to beliefs that are shared. These included the belief that the fatigue is different to healthy fatigue and that the belief ME/CFS is likely caused by a combination of factors. From this point, it may become easier to observe and respect any differences in emphasis that may be present. These findings may be useful to healthcare professionals, such as clinical psychologists, who support patient facing clinicians, such as general practitioners, in reflecting on their work and interaction. The three factors outlined here can be used as a basis for discussion, or raised as points for personal reflection.

Conclusion

This study used Q-methodology to explore the ways in which beliefs about different aspects of ME/CFS group together. Three viewpoints were established. They have some key areas of disagreement but many areas of consensus. It is hoped that this examination of similarities and differences can be used to support future discussion on this highly polarised topic.

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Appendices

* indicates mandatory appendices

Appendix A: Quality appraisal guided by CASP checklist

Study	Was there a clear statement of the aims of the research?	Is a qualitative methodology appropriate?	Was the research design appropriate to address the aims of the research?	Was the recruitment strategy appropriate to the aims of the research?	Was the data collected in a way that addressed the research issue?	Has the relationship between researcher and participants been adequately considered?	Have ethical issues been taken into consideration?
Ax, Gregg and Jones, 2002	Improved understanding of carer's coping efforts, focussing on illness acceptance	Yes	Yes - Semi-structured interviews, thematic analysis	Yes – CFS support group members asked to invite carers.	Yes	Not reported	Not reported
Brooks, King and Wearden, 2014	Explore in depth beliefs and experiences	Yes	Yes – semi-structured interviews and IPA	Yes – via clinic	Yes	yes	yes
Cathchpole and Garip, 2019	Improve understanding of carers by looking at their lived experience	Yes	Yes, semi-structured interviews and IPA	Yes – advertising through two main charities	Yes	Yes – in depth reflection on personal experience's impact on interviews and analysis	yes
Donalek, 2009	Describe responses of the family system	Yes	Yes, semi-structured interviews, thematic analysis	Yes – CFS support groups	Yes	Yes - researcher kept diary, commented on the personal experience of interveiwing	Yes
Horrocks and Ward, 2015	Suggest possible ways of understanding, or imagining, how meanings associated with CFS/ME develop within intimate relationships	Yes	Yes, semi-structured interviews, thematic analysis	Not reported	Yes	Some consideration to influence of clinical experience	Not reported
Lingard and Court, 2014	Investigate strategies used by couples	Yes	Yes - Semi-structured interviews and content analysis	Unclear – professional agencies, medical practices and personal contacts.	Yes	No	Can't tell
Mihelicova et al., 2016	The experiences of parents caring for people suffering from severe ME	Yes	Yes - IPA	Yes - From the book Lost Voices from a Hidden Illness. Not stated how these were originally recruited.	Yes	No – less relevant as did not meet them	No
Missen et al., 2011	Investigate psychological health of mothers of children with CFS/ME	Yes	Yes - Semi-structured interviews, thematic analysis	Yes - From specialist service	Yes	No	Yes

Velleman et al., 2016	Understand impact on CFS/Me on siblings	Yes	Yes – semi-structured interviews and thematic analysis	Yes – via specialist paediatric service	Yes	No	Yes

Appendix B: Example extracts, codes and sub-themes for papers included in meta-synthesis

Example Extract	Code(s)	Related Sub-theme
We are cocooned as a family. There are no family holidays, as [my daughter] is too ill and needs care 24 hours a day. So, she never gets a break from her environment. One of the saddest things for me to watch has been [my daughter's] friends losing contact one by one. This was one of the cruellest blows. (Mihelicova et al., 2016)	changing dynamics loss of family life witnessing distress loss	Family life revolves around the person who is ill Loss of the person they were before the illness
there's no-one I can't talk to, but there's lots of people who don't listen. So I don't bother talking to them. You start to talk and you withdraw from it. Because you feel they are not listening or they infer they are listening but they are not really, and people within the wider social networks that I'm part of, don't really get it. (Horrocks and Ward, 2015)	loss of social support misunderstood	loss of identity Misunderstood and stigmatised
never … felt the pain for myself, … I've only ever felt it for her. So, I think … ultimately it has probably brought us closer together. Definitely supporting each other a lot more now even than before and I think we used to support each other quite a lot previously…. I think … we've developed as it were as a couple and we just, we now work on the basis of 'This is what we've got and let's make the most of what we've got' don't we? (Horrocks and Ward, 2015)	witnessing distress changing dynamics	Different ways of relating to the person
People talk about fatigue and I think sometimes there've been occasions in the past where I've had to say to myself, just, just leave it alone because people say, it's just a little bit like 'Oh yeah, I get tired', and it's kind of you kind of go, 'Ah okay. Yeah okay'. (Liz) (Catchpole and Garip, 2019)	misunderstood	Misunderstood and stigmatised
"If you asked me whether I gave her enough support, the answer is probably no. Certainly not all the time... I mean it has to come in second place to what I'm doing. I've	changing dynamics holding on to identity	Family life revolves around the person who is ill (example of exception) Loss of identity (example of exception)

got a business to run, and it has to be done... So on occasions I probably don't support her as much as I ought to." (Ax, Gregg and Jones. 2002)	guilt	
Bette: He gets quite low and everything I do drives him up the wall and I just don't know what to do. Just in general, he has mood swings and he's not talkative, so when he's really fed up I don't know what triggers it. (Brooks, King and Wearden, 2013)	offering advice losing the relationship	Supporting their family member Loss of the person they were before their illness
All of the siblings talked about some level of restriction that having a child with CFS/ME in the family caused, for example, limiting going out as a family, limiting family activities, such as holidays, and limiting activities inside the house, such as TV and friends coming over: 'That's another thing that's changed because of Sam's illness, is that before we were big campers, big walkers, big cyclers and now we just can't do that'. (Velleman et al., 2016)	stopping family activities limited social contact different family identity	Family life revolves around the person who is ill Loss of identity Loss of identity
Getting her, the rest of the family to understand, that was very difficult. (Mother 1) Erm,my auntie...she always said 'well it's all in the head really isn't it?'. . . (Mother 2) (Missen et al., 2011)	extended family not understanding psychologised beliefs	Misunderstood and stigmatised
Mother: Because in the nature of our work, that we're freelance, we can't really tell anybody. None of our friends, none of our friends were supposed to know this, it was a secret. Nobody's supposed to know that he has chronic fatigue. Because if that got out, we might lose clients. (intently addressing her son) You knew that, right? Son: Yeh, yeh, yeh. I know. (Donalek, 2009)	secrecy lack of social support Fear	Misunderstood and stigmatised

Appendix C: Epistemological position taken by the researcher *

A critical realist position was adopted by the researcher. Based on the work of Bhaskar, critical realism employs elements of constructivism and realism, while providing an alternative to these (Denzin & Lincoln, 2011). It holds that human knowledge can only capture part of reality, or that what is real cannot be reduced to only what can be empirically known (Fletcher, 2017) and what we can know limited by our individual experience. From this perspective, there is a real social world of which this study aims to gain an understanding.

The researcher took the position that her own and the participants' perceptions exist within a social and cultural context and interpretations of reality are shaped by all aspects of personal history and cultural contexts. Q-methodology fits with this epistemology in that it gives a systematic approach to exploring these different perceptions. Critical realism acknowledges that understanding is constructed by the person within their context. For this study, the researcher's context of having been ill with ME/CFS and since recovered is something that was recognised and taken into consideration when interpreting findings, rather than attempting a pretence of objectivity.

Denzin, N. K., & Lincoln, Y. S. (2011). The SAGE handbook of qualitative research. Sage.

Fletcher, A. J. (2017). Applying critical realism in qualitative research: methodology meets method. International Journal of Social Research Methodology, 20(2), 181–194.

Appendix D: Letter of approval from University of Leicester Ethics Committee *

UNIVERSITY OF LEICESTER

24/08/2020

Ethics Reference: XXXXXXXXXXXXXXXXXXXXXXXXXX

TO:

Name of Researcher Applicant: Anna Rickard

Department: Psychology

Research Project Title: Attitudes towards people with a diagnosis of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis: A Q methodology study

Dear Anna Rickard,

RE: Ethics review of Research Study application

The School of Psychology Research Ethics Committee has reviewed and discussed the above application.

1. Ethical opinion

The Committee grants ethical approval to the above research project on the basis described in the application form and supporting documentation, subject to the conditions specified below.

2. Summary of ethics review discussion

The Committee noted the following issues:

The amendment does not pose any ethics concerns, and the revised documents have been uploaded accordingly.

3. General conditions of the ethical approval

The ethics approval is subject to the following general conditions being met prior to the start of the project:

As the Principal Investigator, you are expected to deliver the research project in accordance with the University's policies and procedures, which includes the University's Research Code of Conduct and the University's Research Ethics Policy.

If relevant, management permission or approval (gate keeper role) must be obtained from host organisation prior to the start of the study at the site concerned.

4. Reporting requirements after ethical approval

You are expected to notify the Committee about:

- Significant amendments to the project
- Serious breaches of the protocol
- Annual progress reports
- Notifying the end of the study

5. Use of application information

Details from your ethics application will be stored on the University Ethics Online System. With your permission, the Committee may wish to use parts of the application in an anonymised format for training or sharing best practice. Please let me know if you do not want the application details to be used in this manner.

Best wishes for the success of this research project.

Yours sincerely,

XXXXXXXXXXXXXXXXXXX

Chair

Appendix E: Development Interview participant information sheet*

Participant Information Sheet

Development interviews for future study titled:

Attitudes towards people with a diagnosis of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis: A Q methodology study

Researcher introduction

My name is Anna Rickard and I am a Trainee Clinical Psychologist. I would like to invite you to take part in a research study I am completing as part of my Doctorate qualification in Clinical Psychology at the University of Leicester. To help you decide if you would like to participate, I would like you to understand why the research is being done and what it would involve for you.

I will go through the information sheet with you and answer any questions you have.

Brief summary of the research

We are seeking to explore the different ways that people think about people with a diagnosis of CFS/ME. It is hoped that, in identifying and making sense of a range of ways of understanding, the results will support more constructive dialogue which is often characterised in terms of strong opinions and disagreement. The first stage of this research is development interviews to gain insight into the many different attitudes and opinions that exist currently about people with CFS/ME.

Why have I been invited?

I am looking to speak to people who have some experience of CFS/ME. This may be
(a) having CFS/ME
(b) being recovered from CFS/ME
(c) being friends, relatives, or partners of people with CFS/ME
(d) being a professional who works with or has worked with people with CFS/ME

Do I have to take part?

No. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with your relatives/friends if you wish. You are free to decide whether or not to take part in this study. Please feel free to ask any questions or for more information.

What will happen if I decide to take part?

If you do decide to participate, we will arrange a mutually convenient time to speak via video call. We ask that you find a place and time where you are unlikely to be interrupted. During this interview, I will ask you about different aspects of CFS/ME and your opinions about those. I will then show you a set of statements about CFS/ME and ask you for feedback on whether they are clear and whether you feel there are any areas that are not

covered by the statements. I expect this to take around an hour. If needed, we can take breaks during this process.
I would like to record our conversation so that I can listen back to it. Only the clinical psychology research team will listen to this recording. It is still possible to take part in this interview if you do not wish to be recorded, in which case I will make written notes of our conversation.

It is your choice whether or not you take part in the study. You have the right to withdraw your involvement at any point during the interview. After completion, you have the right to request the withdrawal of your data at any point without giving a reason, however, this will not be practically possible during the later stages of the study after data has been analysed.

If you would like to take part, I will ask you to complete a consent form to say that you understand and agree to participate. Please feel free to ask questions at any point through the research process. At the end I will ask if you would like to take part in the second stage of this research, where you will be asked to sort the statements in order of how much you agree with them. There is no obligation to do this. If you might like to take part, I will email you again at a future date with further information about what this will involve. I will also ask you if you would like to see a summary of the research findings. If so, I will ask for your email address, which will be stored securely and separately to other personally identifiable information.

Are there any benefits or risks to taking part?
Your involvement will be voluntary and there are no financial benefits to doing so. It is hoped that the study will add to our understanding of this condition and how we can work with those who are diagnosed with it, and that this deepened understanding can support an ongoing dialogue about the condition.

There are no direct risks to being involved in the study, but it is worth being aware that some people find thinking about their experience CFS/ME can be emotive. Unfortunately, the Researcher is not able to provide any emotional support if this is the case but will provide space for a brief debrief after completion of the interview.

What if I am harmed by the study?
It is very unlikely that you would be harmed by taking part in this type of research study. However, if you wish to complain or have any concerns about the way you have been approached or treated in connection with the study, you should ask to speak to myself, Anna Rickard as I am the main researcher (my contact details are below), or you can speak with my Research Supervisor, Dr Gareth Morgan (their contact details are below) and we will do our best to answer your questions. If you remain unhappy and wish to address your concerns or complaints on a formal basis, you should contact:

Professor Noelle Robertson, Programme Director (DClinPsy), nr6@le.ac.uk

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for legal action for compensation against the University of Leicester but you may have to pay your legal costs.

What will you do with the information I provide, and will it be safe?
We will be using information from you in order to undertake this study and will act as the data controller for this study. All information will be stored in line with GDPR guidance. This means that we are responsible for looking after your information and using it properly. The University of Leicester will keep identifiable information about you for 10 years after the study has finished.
Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally identifiable information possible.

We will follow ethical and legal practice and all information about you will be handled in confidence and stored securely and the only people that would have access to your personal information would be myself, Anna Rickard, the main researcher and my Research Supervisor, Dr Gareth Morgan.

All your information will be stored securely. The thesis and any subsequent publications may include quotes from our discussion, but these will be anonymised. After the study has been submitted to the University, it may also be submitted for dissemination in academic publications or conferences.

If you provide any information during the interview that makes us believe that you or someone else is in danger, we are obliged to make this known to relevant authorities. We would discuss this with you before doing so unless this were not possible.

What will happen with the results of the study?
The study will form part of my doctoral thesis and as such will be published online by the University of Leicester. It may also be submitted for dissemination in an academic journal or conference.

Who has reviewed this study?
This study has been reviewed and given favourable opinion by the University of Leicester Research Ethics Committee.

What happens now?
If you have any questions or would like to discuss this further with me, you can contact me on the below details. If you would like to take part, please email me to arrange a mutually convenient time for us to complete the study. Please be aware that, unfortunately, I am not

able to provide support and the contact details can only be used for matters related to the study.

Thank you for taking the time to read this information

Anna Rickard aw426@le.ac.uk
Trainee Clinical Psychologist
Main Researcher

Contact details: 0116 223 1639 c/o University of Leicester, Clinical Psychology, Centre for Medicine, Leicester, LE1 7HA

Supervisor: Dr Gareth Morgan, gsm23@le.ac.uk

PARTICIPANT INFORMATION SHEET
Version number 1
Dated 30/06/2020

Appendix F: Development interview consent form*

Consent Form

Development interviews for future study titled:

Attitudes towards people with a diagnosis of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis: A Q methodology study

Please initial box

1. I can confirm that I have read and understood the participant information sheet for the study and have had the opportunity to ask questions.	
2. I understand that my participation is voluntary and I am free to withdraw at anytime without penalty by emailing the researcher. However, I will not be able to withdraw once analysis is underway (expected to be January 2021).	
3. I understand that my personal data will be kept by the University of Leicester for 5 years for the purposes explained in the information sheet, but identifiable information will not be shared with any external organisation.	
4. I understand that any personal information I provide during this study will remain confidential unless there are any serious concerns about my safety or the safety of others. I understand that the researcher will firstly speak to me before making any disclosures.	
5. I would like to be invited to take part in the second stage of this research. Please contact me on this email address:	
6. I give permission for the interview to be recorded and for the interviewer to take notes during the interview.	
7. I give permission for quotes during the interview to be used verbatim in publications, but I understand that I will not be identifiable from quotes.	
8. I agree to take part in the above study.	

If you would like to receive a report of the findings, please write your email address below:

……..

Name of participant ……Signed …… Date ……

Name of researcher …… Signed …… Date ……

Consent form
Version 1
Dated 30/06/2020

Appendix G: Development of the Q-set*

Stage 1 – Statements extracted from the following published works as well as media articles. This process generated over 180 statements. These were then reviewed and statement with similar meaning removed.

Anderson, V. R., Jason, L. A., Hlavaty, L. E., Porter, N., & Cudia, J. (n.d.). A review and meta-synthesis of qualitative studies on Myalgic Encephalomyelitis/chronic fatigue syndrome. https://doi.org/10.1016/j.pec.2011.04.016

Anderson, V. R., Jason, L. A., Hlavaty, L. E., Porter, N., & Cudia, J. (2012). A review and meta-synthesis of qualitative studies on myalgic encephalomyelitis/chronic fatigue syndrome. *Patient Education and Counseling*, *86*(2), 147–155. https://doi.org/10.1016/j.pec.2011.04.016

Arroll, M. A., & Howard, A. (2013). 'The letting go, the building up, [and] the gradual process of rebuilding': Identity change and post-traumatic growth in myalgic encephalomyelitis/chronic fatigue syndrome. *Psychology & Health*, *28*(3), 302–318. https://doi.org/10.1080/08870446.2012.721882

Baker, K. L., & Robertson, N. (2008). Coping with caring for someone with dementia: Reviewing the literature about men. *Aging and Mental Health*, *12*(4), 413–422. https://doi.org/10.1080/13607860802224250

Banasick, S. (2019). KADE: A desktop application for Q methodology. *Journal of Open Source Software*, *4*(36), 1360. https://doi.org/https://doi.org/10.21105/joss.01360

Barnett-Page, E., & Thomas, J. (2009). Methods for the synthesis of qualitative research: a critical review. *BMC Medical Research Methodology*, *9*, 59. https://doi.org/10.1186/1471-2288-9-59

Bayliss, K., Riste, L., Band, R., Peters, S., Wearden, A., Lovell, K., … Chew-Graham, C. A. (2016). Implementing resources to support the diagnosis and management of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis (CFS/ME) in primary care: A qualitative study. *BMC Family Practice*, *17*, 66. https://doi.org/10.1186/s12875-016-0453-8

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Bradley, A. S., Ford, B., & Bansal, A. S. (2013). Altered functional B cell subset populations in patients with chronic fatigue syndrome compared to healthy controls. *Clinical and Experimental Immunology*, *172*(1), 73–80. https://doi.org/10.1111/cei.12043

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Brooks, J., King, N., & Wearden, A. (2014). Couples' experiences of interacting with outside others in chronic fatigue syndrome: a qualitative study. *Chronic Illness*, *10*(1), 5–17. https://doi.org/10.1177/1742395312474478

Brown, S. R. (1980). *Political Subjectivity: Applications of Q Methodology in Political Science*. *Journal of Marketing Research* (Vol. 19). https://doi.org/10.2307/3151542

Brown, S. R. (1996). Q Methodology and Qualitative Research. *Qualitative Health Research*, *6*(4), 561–567. https://doi.org/10.1177/104973239600600408

Brurberg, K. G., Fønhus, M. S., Larun, L., Flottorp, S., & Malterud, K. (2014). Case definitions for chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME): A systematic review. *BMJ Open*. https://doi.org/10.1136/bmjopen-2013-003973

Campbell-Sills, L., Stein, M. B., Sherbourne, C. D., Craske, M. G., Sullivan, G., Golinelli, D., … Roy-Byrne, P. (2013). Effects of medical comorbidity on anxiety treatment outcomes in primary care. *Psychosomatic Medicine*, *75*(8), 713–720. https://doi.org/10.1097/PSY.0b013e31829def54

Caswell, A., & Daniels, J. (2018). Anxiety and depression in chronic fatigue syndrome, prevalence and effects on treatment: A systematic review and metaregression. In *researchportal.bath.ac.uk*.

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Stage 2 – Ten development interviews conducted. Each interview comprised a semi-structured interview from which further potential statements were extracted and a review of the Q-set as it existed at the time of the interview. This review of the Q-set included checking for clarity, and coverage of major topics. Statements were grouped into broad categories to assist the development process. The categories were:

- Describing the symptoms
- Aetiology
- Maintenance
- Mental health related
- Impact on person
- Views of others
- Intervention
- Recovery

Stage 3 – Final review by researcher and supervisor resulting in 64 statements.

Below is a sample of the Q set in development. This sample was created in the first stage where statements were taken from published works. Comments were added during the first four development interviews. Please note that other statements were later as a result of the development interview process.

Statement	Comments pre development interview	Comments from first four development interviews
Describing the symptoms		
1. The fatigue experienced by people with CFS/ME is different from a healthy person's fatigue		
2. Most of us experience tiredness that could be called CFS/ME at some point in our lives		
3. The term 'chronic fatigue syndrome' is misleading with regards to what the condition is really like		
4. CFS/ME affects mostly middle-class people		
5. The symptoms of CFS/ME are difficult to describe		
6. It matters what term is used for the condition		
7. CFS/ME is poorly understood by people who haven't [lived with it]		
8. CFS/ME is not as serious as other conditions		Too broad
Aetiology		
9. Unresolved psychological distress is likely to be a significant factor in the onset and maintenance of CFS/ME	Reword for ease of comprehension? - e.g. People get ill with CFS/ME because of emotional issues	
10. CFS/ME is probably a neurological condition		'neurological condition' not a well-defined or understood concept. Not needed as similar to other statements
11. My beliefs about the potential causes of CFS/ME often change		
12. There is probably an undiscovered physical cause for CFS/ME		
13. CFS/ME is likely caused by different things for different people		
14. CFS/ME is likely caused by multiple factors in each		

person 15. People with CFS/ME have a greater probability of having experienced childhood trauma 16. The aetiology of CFS/ME is a controversial topic 17. Stress is often a factor in onset Maintenance 18. People with CFS/ME experience fatigue and pain as a result of hypervigilance to bodily sensations 19. Giving the diagnosis of CFS/ME makes people more likely behave like they are ill 20. Low mood is a factor that stops people with CFS/ME getting better 21. People with CFS/ME stay ill because of the beliefs they hold about their symptoms 22. Addressing unhelpful beliefs about their illness can be helpful for people with CFS/ME 23. Having CFS/ME symptoms help the person psychologically in some way 24. It is harder to adjust to the illness when there are lots of fluctuations in symptoms Mental health-related 25. People with CFS/ME are likely to also have depression 26. People with CFS/ME are likely to also have anxiety 27. CFS/ME occurs in people who are prone to the impact of stress 28. Suggesting to a person with CFS/ME that their illness might be psychological can have a negative impact upon them 29. People with CFS/ME prefer a physical explanation of their symptoms to a psychological explanation 30. People with CFS/ME are open to psychosocial explanations	Use interviews to check understanding of phrasing	change to 'more likely' for ease of comprehension Stating the obvious? Aetiology not always understood change to 'focussed on symptoms' 'unhelpful beliefs' not always understood, duplicates previous statement Too vague Not related to research question? change to low mood to avoid issues around diagnostic terms and criteria change to psychological as 'psychosocial' not

of their symptoms		always understood
31. People with CFS/ME are often inappropriately given psychiatric diagnoses		
32. People with CFS/ME tend to retain a strong interest in other aspects of their lives		
33. People with CFS/ME don't tend to have much motivation to recover		rephrase to avoid use of the negative
34. Many people with a diagnosis of CFS/ME would be better served with a diagnosis of depression	A diagnosis of depression would be more accurate for many people with CFS/ME	duplicates previous statement
35. Treatment should focus on depressive symptoms		duplicates previous statement
36. People with CFS/ME gain material advantages from having the diagnosis		unclear phrasing, misinterpreted
37. People with CFS/ME tend to avoid activity		
38. People with CFS/ME tend to do too much/be overactive		'too much' is fine
39. People with CFS/ME tend to go to the doctors repeatedly with a lot of symptoms		original meaning (going to the GP too much) is lost in the phrasing
40. People with CFS/ME tend to use health services intensively relative to their needs		unnecessary
41. No specific personality profile predisposes people to the risk of developing CFS/ME		duplicates other statements
42. It is important to understand CFS/ME in the context of the person's life		unclear
43. CFS/ME can provide an identity for the person that is helpful in some way		
44. CFS/ME symptoms can be understood as expressions of psychological problems	Very similar to 5 but ok?	
45. People with CFS/ME tend to have higher emotional needs than most people		
46. People with CFS/ME tend to set very high standards for themselves		
Impact on person		
47. People with CFS/ME experience stigma associated with the condition		
48. People's quality of life is significantly affected by CFS/ME		likely to be agreed with by most
49. CFS/ME impacts on the self-esteem of people with the		not relevant to research question

condition		
50. Being given a diagnosis of CFS/ME can be a relief		not relevant to research question
51. Having a diagnosis of CFS/ME is better for people than having unexplained symptoms		
52. Feeling that they have social support is important for people with CFS/ME		not relevant to research question
53. People with CFS/ME experience loss as consequence of being ill		not relevant to research question
54. Having CFS/ME affects how the person feels about their own identity		
55. Other people can be very judgemental of people with CFS/ME		meta level of presupposing others' attitudes
56. CFS/ME can cause conflict in personal relationships		not relevant to research question
57. CFS/ME profoundly affects personal relationships		not relevant to research question
58. People with CFS/ME are often misunderstood		duplicates previous statements
59. It can hurt to not be believed		not relevant to research question
Views of others		
60. People with CFS/ME tend to need to demonstrate their illness is real		too ambiguous – at least two clear interpretations
61. People with CFS/ME tend to ignore the professional advice they have been given		too ambiguous – at least two clear interpretations
62. Other people tend to hold different views from me on the cause of the condition		meta level of presupposing others' attitudes
63. People with CFS/ME tend to be defensive		not always understood
64. People with CFS/ME want their experience to be recognised and validated		too easy to agree with
65. People with CFS/ME struggle to get others to acknowledge their symptoms		
66. Some people with CFS/ME exaggerate their symptoms		
67. I tend to find it hard to have sympathy for people with CFS/ME		
68. People with CFS/ME often blame themselves for their symptoms		
69. I sometimes feel I negatively judge people with CFS/ME		

70. People with CFS/ME want to feel their symptoms are being taken seriously		duplicates others symptoms
Intervention		
71. CFS/ME requires a more holistic approach than other conditions		
72. A great deal can be done to improve the quality of life for people with CFS/ME		overstated, needs rephrasing
73. People with CFS/ME need encouragement to push through the fatigue		
74. People with CFS/ME would benefit from working with mental health specialists		too ambiguous?
75. People with CFS/ME place too much emphasis on getting the diagnosis of CFS/ME		
76. Getting a diagnosis helps the person to manage the symptoms of CFS/ME		
77. It can be challenging for people with CFS/ME to put lifestyle management techniques into practice		
78. Graded exercise therapy is the therapy most likely to lead to recovery		rephrase more broadly?
Recovery		
79. The person themselves makes the biggest difference with regards to how quickly they get better		
80. Recovery is the responsibility of the person with CFS/ME		
81. Symptoms of CFS/ME are perpetuated by the person not managing the condition well		rephrase for ease of comprehension
82. It is essential to have collaboration between professionals and the person with CFS/ME for them to recover from CFS/ME		
83. People shouldn't expect a full recovery from CFS/ME		rephrase to avoid use of the negative
84. People with CFS/ME need to rest enough in order to recover	Both rest and activity have a role to play in recovery – this item from literature has been expanded into two items.	rephrase
85. People with CFS/ME need to become active again in order to recover		

Attitudes towards people with a diagnosis of Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome: A Q methodology study

Researcher introduction

My name is Anna Rickard and I am a Trainee Clinical Psychologist. I would like to invite you to take part in a research study I am completing as part of my Doctorate qualification in Clinical Psychology at the University of Leicester. To help you decide if you would like to participate, I would like you to understand why the research is being done and what it would involve for you.

I will go through the information sheet with you and answer any questions you have.

Brief summary of the research

We are seeking to explore the different ways that people make sense of ME/CFS. It is hoped that, in identifying and making sense of a range of ways of understanding, the results will support more constructive dialogue, which has often been characterised in terms of strong opinions and disagreement.

Why have I been invited?

I am looking to speak to people who have some experience of ME/CFS. This may be
(a) having ME/CFS
(b) being recovered from ME/CFS
(c) being friends, relatives, or partners of people with ME/CFS
(d) being a professional who works with or has worked with people with ME/CFS

Do I have to take part?

No. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with your relatives/friends if you wish. You are free to decide whether or not to take part in this study. Please feel free to ask any questions or for more information.

What will happen if I decide to take part?

If you do decide to participate, we will arrange a mutually convenient time to speak via video call. We ask that you find a place and time where you are unlikely to be interrupted. Using online software, I will show you a set of approximately 70 statements relating to people who have ME/CFS. I will ask you to order the statements with regards to how strongly you agree with each item, in a predetermined configuration. We can discuss your reasoning while we do this if you choose. I expect this to take around an hour. If needed, we can take breaks during this process. This is known as a 'Q sort'. It is part of a method known as 'Q-Methodology', which is an approach used to make sense of subjective ways of thinking about a given phenomenon. I would like to record this conversation so I can refer back to it.

Only the clinical psychology research team will listen to this recording. If you would prefer not to be recorded, I will make written notes as we speak.

It is your choice whether or not you take part in the study. You have the right to withdraw your involvement at any point during the interview. After completion, you have the right to request the withdrawal of your data at any point without giving a reason, however, this will not be practically possible during the later stages of the study after data has been analysed.

If you would like to take part, I will ask you to complete a consent form to say that you understand and agree to participate. Please feel free to ask questions at any point through the research process. I will also ask you if you would like to see a summary of the research findings. If so, I will ask for your email address, which will be stored securely and separately to other personally identifiable information.

Are there any benefits or risks to taking part?
Your involvement will be voluntary and there are no financial benefits to doing so. It is hoped that the study will add to our understanding of this condition and how we can work with those who are diagnosed with it, and that this deepened understanding can support an ongoing dialogue about the condition.

There are no direct risks to being involved in the study, but it is worth being aware that some people find thinking about their experience ME/CFS can be emotive. Unfortunately, the Researcher is not able to provide any emotional support if this is the case but will provide space for a brief debrief after completion of the interview.

What if I am harmed by the study?
It is very unlikely that you would be harmed by taking part in this type of research study. However, if you wish to complain or have any concerns about the way you have been approached or treated in connection with the study, you should ask to speak to myself, Anna Rickard as I am the main researcher (my contact details are below), or you can speak with my Research Supervisor, Dr Gareth Morgan (their contact details are below) and we will do our best to answer your questions. If you remain unhappy and wish to address your concerns or complaints on a formal basis, you should contact:

Professor Noelle Robertson, Programme Director (DClinPsy), nr6@le.ac.uk

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for legal action for compensation against the University of Leicester but you may have to pay your legal costs.

What will you do with the information I provide, and will it be safe?
We will be using information from you in order to undertake this study and will act as the data controller for this study. All information will be stored in line with GDPR guidance. This means that we are responsible for looking after your information and using it properly.

University of Leicester will keep identifiable information about you for 10 years after the study has finished.
Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally identifiable information possible.

We will follow ethical and legal practice and all information about you will be handled in confidence and stored securely and the only people that would have access to your personal information would be myself, Anna Rickard, the main researcher and my Research Supervisor, Dr Gareth Morgan.

All your information will be stored securely. The thesis and any subsequent publications may include quotes from our discussion, but these will be anonymised. After the study has been submitted to the University, it may also be submitted for dissemination in academic publications or conferences.

If you provide any information during the interview that makes us believe that you or someone else is in danger, we are obliged to make this known to relevant authorities. We would discuss this with you before doing so unless this were not possible.

What will happen with the results of the study?
The study will form part of my doctoral thesis and as such will be published online by the University of Leicester. It may also be submitted for dissemination in an academic journal or conference.

Who has reviewed this study?
This study has been reviewed and given favourable opinion by the University of Leicester Research Ethics Committee.

What happens now?
If you have any questions or would like to discuss this further with me, you can contact me on the below details. If you would like to take part, please email me to arrange a mutually convenient time and place for us to meet and complete the study. Please be aware that, unfortunately, I am not able to provide support and the contact details can only be used for matters related to the study.

Thank you for taking the time to read this information

Anna Rickard aw426@le.ac.uk
Trainee Clinical Psychologist
Main Researcher

Contact details: 0116 223 1639 c/o University of Leicester, Clinical Psychology, Centre for Medicine, Leicester, LE1 7HA

Supervisor: Dr Gareth Morgan, gsm23@le.ac.uk

PARTICIPANT INFORMATION SHEET
Version number 3
Dated 16/10/2020

Appendix I: Q Sort online consent form*

Consent form

Page 1: Page 1

1. I can confirm that I have read and understood the participant information sheet for the study and have had the opportunity to ask questions.

- Yes

2. I understand that my participation is voluntary and I am free to withdraw at anytime without penalty by emailing the researcher. However, I will not be able to withdraw once analysis is underway (expected to be January 2021).

- Yes

3. I understand that my personal data will be kept by the University of Leicester for 5 years for the purposes explained in the information sheet, but identifiable information will not be shared with any external organisation.

- Yes

4. I understand that any personal information I provide during this study will remain confidential unless there are any serious concerns about my safety or the safety of others. I understand that the researcher will firstly speak to me before making any disclosures.

- Yes

5. I give permission for the interview to be recorded and for the interviewer to take notes during the interview.

- Yes

6. I give permission for quotes during the interview to be used verbatim in publications, but I understand that I will not be identifiable from quotes.

- Yes

7. I agree to take part in the above study.

- Yes

8. If you would like to recieve results of this study, please write your email address below

9. Name:

10. Date:

Dates need to be in the format 'DD/MM/YYYY', for example 27/03/1980.

(dd/mm/yyyy)

Appendix J: Analysis history

Step	Action	Rationale
1	Data loaded to KenQ as Excel Type 1 file	
2	Horst Centroid Factors Extracted: 7 No convergence: 300 iterations	7 factors appropriate starting point (Brown, 1980).
3	Number of factors selected for rotation: 5	Using Kaiser-Guttman criterion of an Eigenvalue greater than one (Guttman, 1954; Kaiser, 1960).
4	Rotation Varimax rotation applied Factor 1 and Factor 2 rotation: 12 degrees Factor 1 and Factor 3 rotation: 4 degrees Factor 1 and Factor 4 rotation: 7 degrees Factor 2 and Factor 3 rotation: 4 degrees Factor 2 and Factor 4 rotation: 3 degrees Factor 3 and Factor 4 rotation: 6 degrees Factor 4 and Factor 5 rotation: 5 degrees Factor 1 and Factor 2 rotation: 1 degrees Factor 1 and Factor 3 rotation: -4 degrees Factor 1 and Factor 4 rotation: -2 degrees Factor 2 and Factor 3 rotation: -1 degrees Factor 3 and Factor 4 rotation: 11 degrees	Varimax rotation initially applied followed by hand rotation, applied iteratively to establish optimal loadings as per Watts and Stenner (2005). All rotations are orthogonal.
5	Sorts flagged to be included in idealised factor sort 18 sorts auto-flagged 17 sorts unflagged One sort reflagged	$p < 0.05$ and a majority of common variance was required. Removed sorts that loaded more than 0.3225 ($p<0.01$) on more than one factor (Brown, 1980) variance explained higher for one factor than others
6	Three factors retained for interpretation	Criterion: two or more sorts significantly loading

Appendix K: Reflective Piece

I have used exerts from the research diary that I kept through the research process to reflect on personal learning points. The study is close to home for me in many ways. I have been a 'sufferer' of ME/CFS, I am recovered from it (a status many believe is not possible), I have had many personal interactions with healthcare professionals on the topic, some which were hugely helpful and supportive and some which felt the opposite. I have attempted to understand these experiences and the impact they have on me as a researcher, using this diary as a tool in considering the influence of my personal history on the research.

> 28/4/2020 – 'I will need to consider whether it is reasonable for many participants to be people I know personally. Being transparent about this in the write up may make it ok but I will need to consider the potential for this to bias the findings. On a similar note, I will need to consider how to recruit people with a range of viewpoints.'

In this exert, I was grappling with the process of recruitment. In Q-Methodology the need is for participants to hold a broad range of views rather than for the participants to be representative of any particular group of people. As I have many friends and friends of friends who would meet recruitment criteria, I was aware that if I went down the route of advertising via personal social media, I could meet the approximate number of participants. I was concerned of the various ways this could bias the research, as participants would have more homogenous backgrounds than a more general recruitment strategy and would be more likely to share much more similar perspectives on the condition. I therefore recruited via national organisations and was able to recruit more than enough participants via this method, without the temptation to cope with time pressures on recruitment by using personal channels.

> 1/9/2020 – 'Completed the first pilot interview with someone who has been ill themselves but also had family members and friends who were ill. It was quite emotional at times. It also brought up for me so many memories I hadn't thought of for a long time.'

I took time to process memories with friends and family afterwards about parts of my own experience. I tried to pay particular attention to when I found myself internally agreeing or disagreeing with things that participants said, in order to address where my own perceptions and preconceptions lie. In this sense, it was very useful to complete the Q-sort myself and at the analysis stage, see exactly how my own viewpoint loaded onto the different factors. My sort was associated almost equally with factors 1 and 3 and not at all with factor 2. I therefore paid particular attention to how factor 2 was reported, attempting to embody the purpose of the research – to facilitate people of different viewpoints in understanding each other.

Appendix L: Chronology of the research process*

January 2018	Research proposal preference form submitted Research supervisor allocated
February – May 2018	Research idea in development
May 2018	Research proposal submitted
June 2018	Review meeting for research proposal
July – December 2018	Development of research proposal
February 2019	University peer review process completed
March 2019	Service User review completed
March -June 2019	Development of Q-set
June 2019 – April 2020	Research on pause due to maternity leave
May 2020	Review and adaptation of research focus due to COVID19 pandemic
June 2020	Research proposal resubmitted to peer review
July 2020	Literature review begun
August 2020	Research proposal peer review process completed Service User review completed Application to the University of Leicester Ethics Committee submitted and approved
September 2020	Development interviews completed
October 2020	Literature review first draft submitted
November 2020 – March 2021	Data collection
February 2020	Literature review second draft submitted
March – April 2021	Data analysis and interpretation
April – May 2021	Writing up period
May 2021	Short talk given at conference of British Association of CFS/ME on research findings Thesis submitted to University of Leicester
July 2021*	Viva examination
July - September 2021*	Dissemination of findings through poster presentation and submission of research paper for publication

* intended activities

Appendix M: Guidelines to authors for the journalS targeted for literature review and research report*

Target journal for literature review: Fatigue: Biomedicine, Health & Behavior

Guidelines for authors can be found on the website:

https://www.tandfonline.com/action/authorSubmission?show=instructions&journalCode=rftg20

Target journal: British Journal of Health Psychology

Guidelines for authors can be found on the website:

https://bpspsychub.onlinelibrary.wiley.com/hub/journal/20448287/homepage/forauthors.html

Appendix N: Checklist to ensure anonymity of clients/services*

	Checked in Executive Summary/Abstract/ Overview (if included in assignment)	**Checked in main text**	**Checked in appendices**
Pseudonym or false initials used	x	x	X
Reference to pseudonym/false initials as a footnote	NA	NA	NA
Removed any reference to names of Trusts/hospitals/clinics/services (including letterhead if including letters in appendices)	x	X	x
Removed any reference to names/specific dates of birth/specific date of clinical appointments/addresses/ location of client(s), participant(s), relatives, caregivers, and supervisor(s). [For research thesis – supervisors can be named in the research thesis "acknowledgements" section]	x	x	x
Removed/altered references to client(s) jobs/professions/nationality where this may potentially identify them. [For research thesis – removed potential for an individual research participant to be identifiable (e.g., by a colleague of the participant who might read the thesis on the internet and be able to identify a participant using a combination of the participants specific job title, role, age, and gender)]	x	x	x
Removed any information that may identify the trainee (consult with course staff if this will detract from the points the trainee is making)	x	x	x
No Tippex or other method has been used to obliterate the original text – unless the paper is subsequently photocopied and the trainee has ensured that the obliterated text cannot be read	NA	NA	NA
The "find and replace" function in word processing has been used to check the assignment for use of client(s) names/other confidential information	x	x	x