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of people with chronic fatigue syndrome/myalgic
encephalomyelitis and their support needs**

**Pilkington, K., Ridge, Damien T., Igwesi-Chidobe, Chinonso,
Chew-Graham, C., Little, P., Babatunde, O., Corp, N., McDermott,
C. and Cheshire, A.**

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A relational analysis of an invisible illness: A meta-ethnography of people with chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) and their support needs

Abstract

Chronic fatigue syndrome (CFS)/myalgic encephalomyelitis (ME) is indicated by prolonged, medically unexplained fatigue (amongst other symptoms), not alleviated by rest, and causing substantial disability. There are limited treatments on offer, which may not be effective and/or acceptable for all people, and treatment views are polarised. We, thus, aimed to take a step back from this debate, to explore more broadly preferences for formal and informal support among people with CFS/ME. We used a meta-ethnography approach to examine the substantial qualitative literature available. Using the process outlined by Noblit and Hare, and guided by patient involvement throughout, 47 studies were analysed. Our synthesis suggested that to understand people with CFS/ME (such as their invisibility, loss of self, and fraught clinical encounters), it was useful to shift focus to a ‘relational goods’ framework. Emotions and tensions encountered in CFS/ME care and support only emerge via ‘*sui generis*’ real life interactions, influenced by how social networks and health consultations unfold, and structures like disability support. This relational paradigm reveals the hidden forces at work producing the specific problems of CFS/ME, and offers a ‘no blame’ framework going forward.

Keywords: Chronic fatigue syndrome; myalgic encephalomyelitis; meta-ethnography; qualitative; relational good; social support; Users' Experiences

Introduction

Chronic fatigue syndrome (CFS) and/or myalgic encephalomyelitis (or encephalopathy) (ME) is marked by severe, medically unexplained fatigue, not helped by rest, lasting at least 4 months (Fukuda et al., 1994; National Institute for Health & Care Excellence, 2007). People can experience other symptoms like headaches, problems with sleep, concentration and memory, thus CFS/ME can be associated with substantial disability (Assefi et al., 2003; Bombardier & Buchwald, 1996; Collin et al., 2011). When severe, CFS/ME can lead to an individual becoming house, wheelchair or bed-bound, dependent upon carers for support with basic activities of daily living (Burley et al., 2007), with limited access to NHS or social care (XXX blinded for anonymous review). While a full recovery may only occur in about 5%, the statistics on improvement are better, with studies suggesting that anywhere between 8% and 63% of patients have reduced symptoms (Cairns & Hotopf, 2005). For many, the illness can last for decades, leaving individuals profoundly disabled and isolated. While causes are poorly understood, and may involve different processes, diagnosis is made by considering the patient's symptoms (patient history), and the elimination of other medical/psychiatric causes, with tests that come back normal (XXX blinded for anonymous review).

The prevalence of CFS/ME is 0.2% to 0.4% of the UK population, being higher in females, with all ages and ethnic groups affected (Bhui et al., 2011; Department of Health Independent Working Group, 2002; Ingman et al., 2016). The National Institute for Health and Care Excellence (NICE) encourages early diagnosis, but also recommends that advice on management of symptoms not wait for a diagnosis (National Institute for Health & Care Excellence, 2007). Patients report that receiving a diagnosis can be pivotal in managing their condition (Whitehead, 2006). Diagnosis is, however, delayed – the average time from symptoms to diagnosis is 3.6 years (XXX anonymised for blinded review), with late

diagnosis associated with severe CFS/ME (Pheby & Saffron, 2009). NICE recommends that advice be individualised to the specific symptoms, with a focus on minimising impact on daily living. The NICE guidelines also emphasise the importance of shared decision-making, recommending that ‘...therapeutic options [be given] to people with CFS/ME in ways that are suitable for the individual’. In addition to general management strategies, NICE recommends cognitive behavioural therapy (CBT) or graded exercise therapy (GET)/activity management programmes. Similarly, systematic reviews recommend their adoption to reduce symptoms in CFS/ME (Larun et al., 2019; Price et al., 2008). However, the role of GET and CBT as treatments for CFS/ME continue to be disputed (Lords Debates, 2013; The Lancet Editorial, 2011). For example, surveys of charity members show that almost half of respondents using GET report negative effects (Action for M.E., 2014). As a consequence, there is no accepted ‘gold standard’ treatment for CFS/ME. Additionally, because of polarised views on treatments, patients easily encounter confusing information.

A plethora of qualitative research has been conducted with people with CFS/ME, more aimed at understanding the experiences of individuals (i.e. aspects of the self) in context, rather than specifically focusing on interpersonal experiences, or using a relational framework for analysis. There have been a number of previous syntheses. In focusing on the individual with CFS/ME, Larun and Malterud (2007)’s meta-ethnography highlighted the way in which patients’ sense of identity was challenged by their symptom burden. The impact on identity was explored further by Anderson and colleagues (2012) who extended Larun and Malterud’s synthesis by inclusion of a larger selection of studies with greater breadth of methodologies and perspectives. Their analysis also revealed a significant effect of CFS/ME on the patients’ identity, suggesting that people experienced an evolving identity throughout their illness journey. Here, a reconstruction of identity, alongside cycles of health and ill-health, was reported. They also found reductions in function ‘across occupational,

education, personal, or social domains,' suggesting support is needed across all domains.

According to Pinxterhuis and colleagues (2015a), patients also undergo various psychological shifts to cope with their illness, further complicating any impact on identity.

The patient-doctor relationship has also been scrutinised. Larun and Malterud (2007) aimed to provide an insight into the perspectives of doctors and the patient-doctor relationship. Their findings suggest that doctors struggle to maintain professional identities and authority in the face of opposing patient beliefs, and this impacts on the support that they are able to provide to patients. Thus, acknowledging the efforts patients make to understand and manage their condition is recommended as a valuable first step in bridging any divide. Bayliss and colleagues (2014) focused specifically on barriers to diagnosis and care of CFS/ME, exploring themes around illness models and the health professional-patient relationship. Taking a bio-psychosocial approach that remained flexible, building a relationship and collaborating with the patient to agree management were found to be markers of GPs perceived as supportive.

Bayliss and colleagues' synthesis also highlighted that it was essential to engage with family members; they may help or hinder management but without support from family, patients may feel alone and struggle to cope. Conversely, responses from significant others, particularly around legitimacy of their illness, also appear to impact on the identities of people with CFS/ME, with the result that they may feel 'blamed and dismissed' (Larun & Malterud, 2007). In synthesising the findings from 32 qualitative and quantitative studies, de Lourdes Drachler and colleagues (2009) highlighted the significant support needs and substantial help considered essential to rebuild lives of those with CFS/ME. The onus is on a wide range of people to understand - and respond to - patient needs. This reality may also explain why people with CFS/ME also appear to use a wide range of other coping strategies, self-care approaches and complementary therapies (Pinxterhuis et al., 2015a). Interestingly,

although not explored in any depth in the syntheses, these findings point to the need to understand more about the collective nature of the support required. Anderson et al. (2012) (p. 154) noted specifically that research was needed to better join up the varying elements of the experience, including “the members with the networks of people with ME/CFS, and the sociocultural environment in which we define and examine the illness”.

While these previous syntheses have highlighted relationships as important for CFS/ME support, relations have not been studied in-depth. Our meta-ethnography thus aimed to integrate the different components of the CSF/ME support experience using a relational framework, to reveal emergent needs and potential solutions hitherto overlooked. In the late 1980s, a kind of relational framework was outlined by Italian sociologist Pierpaolo Donati (2019) (p. 240) that was not a thing, nor a service, and which he called “relational goods”. Such goods were considered to create their own “ontological reality” with relations that (i) could not be reduced to dealings with others; (ii) were emergent in terms of the impacts on the people in relationship, (iii) had a “*sui generis* reality” i.e. a quality of distinctiveness of structure, dynamic and process; (iv) were created (and appreciated) by participants; (v) could produce benefits for contributors, as well as those who are able to contemplate the interactions from the outside, and (vi) no single person was able to appropriate these goods for themselves. While Donati focused on morally positive goods like trust, we extend the concept to encompass the production of more challenging goods like suspicion.

The aim of our meta-ethnography was to systematically synthesise available qualitative evidence on the informal and formal support that people experienced and preferred for CFS/ME. Here, we set out to examine the support patients with CFS/ME themselves wanted from professionals (formal), as well as significant others (informal). The large body of qualitative literature available, including further studies since previous meta-syntheses, provide a rich opportunity to address the collective gap(s) in knowledge. Here, we

applied a relational goods framework as described above. In recent years there has been a “relational turn” in a broad range of scholarly disciplines (Dépelteau, 2015, p. 46). In this way of anti-individualised thinking, “social worlds are not entities to be separated, but instead are comprised of mutually constituting relations” (Feldman & Worline, 2016, p. 308). Meaning is not innate to entities, instead it is emergent, and relies on human (and non-human) interactants, influenced by social structures and institutions (like the NHS). Thus, things and individuals only take on meaning via their connections to other things, precisely because human experience is itself “irreducibly relational” (Price-Robertson, Obradovic, & Morgan 2017, p. 108). Because previous syntheses of CFS/ME have focused more on the entities in context, rather than the relations specifically and in-depth, our paper applies the ‘relational goods’ framework to investigate the ways people with CFS/ME understand, perceive, experience and prefer their formal and informal support.

Methods

Three stages were involved in this review: systematic search, quality appraisal of included studies, and synthesis informed by Noblit and Hare’s (1988) seven-step meta-ethnography. The systematic review protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO) (ID: CRD42017081418). The PRISMA statement guidelines for reporting systematic reviews of studies that evaluate health care interventions were followed as far as was relevant (Liberati et al. 2009). The PRISMA statement guidelines for reporting systematic reviews of studies that evaluate health care interventions were followed as far as was relevant, with adaptations to suit the meta-ethnographic approach and journal requirements. Thus, a structured abstract was not included as specified in *Social Science & Medicine* guidelines. An adaptation of PICO and SPIDER was used to structure the research question as recommended for qualitative research (Cooke et al., 2012). Items relating to searching, selection and extraction of data are reported, and

risk of bias was replaced with quality assessment (as this approach is more suited to qualitative research). The process for synthesis is reported, but items relating to meta-analysis were omitted as not relevant to a meta-ethnography. The ENTREQ framework was also used as a guide to making the various stages of the synthesis (e.g. searches, selection of studies, quality appraisal, and synthesis methods) transparent (Tong et al.,2012).

Patient and Public Involvement and Engagement (PPIE)

Patient and public involvement and engagement (PPIE) in systematic reviews is considered good practice, although it is not always well reported in publications, nor in sufficient detail (Pollock et al., 2018). Nevertheless, PPIE has been successfully integrated into systematic reviews and qualitative synthesis research (Hyde et al., 2017). There are challenges, particularly in an area such as CFS/ME where there are strongly held and wide ranging perspectives to engage, as discussed previously. Our approach was pragmatic in seeking input from an existing PPI group (patient advisory group (PAG)), together with involvement of patient-researchers on the research team throughout the entire project. The study was informed by the PAG from the outset, including the development of the funding proposal, refining the research question, study design, data analysis and interpretation of findings.

Several PAG meetings were held: to contribute to the funding application; to agree our approach and priorities; to scope our definition of support; to discuss the emerging analysis and dissemination of findings. Together with the PAG group, we agreed on our concept of support to include positive and negative polarities of support as well as qualities and sources of support. We also presented our preliminary findings (key concepts) to the PAG group and asked each person to use these to tell their own stories within the group. These stories illustrated and helped contextualise and prioritise our key concepts, and acted as

an additional examination and critique of our attempts to interpret the findings. Finally, one of the authors presented the findings to a CFS/ME support group in North Staffordshire, and comments from participants in the group helped to finalise the findings. Members of the research team who had experience of living with CFS/ME, or caring for people with the illness, provided additional PPIE input, ensuring that we remained true to PPIE perspectives, even during the journal paper review process. The research team itself represented a diverse range of backgrounds, clinically (e.g. medicine, pharmacy, psychotherapy) and academically (e.g. sociology, psychology, primary care, public health, global health, complementary medicine). The team also encompassed positivist as well as more interpretive approaches to research. Several researchers had no previous experience with the field of CFS/ME research, while others had extensive clinical and research experience in this area.

Systematic search strategy and selection criteria

We searched the following databases using a combination of free text and index (MeSH or equivalent) terms: MEDLINE, PsycINFO, EMBASE, CINAHL-Plus, Social Care Online, ASSIA, AMED, British Library EThOS service, Web of Science (Science Citation Index (SCI), Social Science Citation Index (SSCI), Conference Proceedings Citation Index-Science (CPCI-S), Conference Proceedings Citation Index- Social Science & Humanities (CPCI-SSH)) and Sociological Abstracts from their inception to October 2017 (see Appendix 1 for full search strategy for OVID MEDLINE). The search strategy focused on sensitivity rather than specificity, and included relevant terms for CFS/ME informed by a strategy developed for a Cochrane review by Larun and colleagues (2017). These terms were combined with a qualitative search filter (DeJean et al., 2016). See Appendix 1 for further detail. We also carried out further searches for relevant studies including electronic and hand searches of conference proceedings, websites of expert societies and the reference lists of included studies.

We included studies of people aged at least 16 years with CFS/ME; support issues across different levels of severity of symptoms; and across different cultures acknowledging that culture and ethnicity overlap (Patsiurko et al., 2012). Outcomes (and focus) included experiences, perceptions, attitudes, emotions, views, behaviours. Inclusion criteria were based on a qualitative adaptation of the PICO and SPIDER techniques addressing Population, Phenomenon of Interest, Context, and Outcome (XXX blinded for anonymous review). All relevant studies (with a focus on informal and/or formal support) taking a qualitative approach to explore the experiences/perspectives/opinions of people with CFS/ME were included. Studies were not limited by location (country, healthcare or social setting), but only literature published in English was included (translation may affect the original meanings/interpretation of the findings), although we planned to note these studies and comment on the amount of evidence potentially lost through their exclusion. Within the heading of 'support' we sought to identify and differentiate between forms of intended support perceived as constructive and valued by people with CFS/ME, and those experienced as less than helpful or even detrimental. This point had been particularly emphasised by our PAG members. We also agreed on the scope of studies to include those that reported patients' perspectives, mixed-methods studies with a qualitative component, reports of primary data, and focused on support post-diagnosis rather than pre-diagnosis. We excluded studies focusing on fatigue in other medically unexplained symptoms (MUS), and those not focusing on support. We initially planned to include dissertations identified via the British Library EThOS service but decided retrospectively to exclude dissertations due to the large number of eligible studies for this review, many of which had also been published as articles.

Search results were exported into Endnote for manual removal of duplicate records. Following deduplication, two researchers (z and w) screened the titles and abstracts of the records to select studies that appear to meet criteria for inclusion. After initial screening, all

potentially relevant studies were obtained in full-text, and assessed as relevant or not by the two reviewers working independently. Assessments were then compared, and any conflicts worked out by discussion. In any cases where agreement could not be reached, a debate involving an extended project team took place (including XXX). A record was kept of the screening process, numbers included and excluded, and reasons for inclusion/exclusion documented.

Data extraction and quality assessment

Author (x) and Author (y) supported Authors (z and w) in designing and piloting the data extraction form. The data which were extracted included authors' names, year of publication, study aims, sample size, participant characteristics (age, gender, ethnicity), method of CFS/ME diagnosis, sampling method, data collection methods, setting (place of recruitment), data analysis methods, study quality, and key/recurring, first and second order concepts. Concepts were extracted from the results and discussion sections of the included studies. Data were extracted by two researchers (z and w) working independently. The quality of each study was also independently assessed by these two researchers using the 'QARI Critical Appraisal Checklist for Interpretive & Critical Research' (Joanna Briggs Institute, 2010). This assessment was based on the conduct of studies including validity and robustness, and the domains included overall research design, philosophical perspectives, reflexivity and ethical considerations. While structured quality assessment of qualitative research is widely debated (Carroll & Booth, 2015), using a formal checklist supported a consistent approach to studies included in the review. No study was excluded based on quality, however, the relative strengths and weaknesses of studies informed the process of synthesis and interpretations. Any discrepancies in data extraction and quality appraisal were resolved by discussion or, if necessary, by the wider research team arbitrating as a panel.

Data synthesis

Whilst various methods are available for meta-synthesis of qualitative research, we chose meta-ethnography, due to its well-developed methodology, fidelity with primary studies, and focus on developing new conceptual insights (France et al., 2015). A seven-step process was used based on the Noblit and Hare model, an approach that has been widely used in health-related meta-synthesis (Noblit & Hare, 1988). The seven stages included: '1. Getting started, 2. Deciding what is relevant to the initial interest, 3. Reading the studies, 4. Determining how the studies are related, 5. Translating the studies into one another, 6. Synthesising translations, and 7. Expressing the synthesis.' We considered whether it would be appropriate to carry out any form of subgrouping of papers as the number identified was large. We decided, however, that based on our initial discussions with the PAG, that the emphasis in the synthesis should be on an overall, holistic view of support, drawing all the facets together, but focused on the patient's perspective. We read and re-read the included studies to identify the key /recurring concepts related to support issues. First, second and third order constructs were identified. First order constructs refer to participant reported perspectives, beliefs and experiences, while second order constructs pertain to author interpretations of participant accounts, and third order constructs are those generated by the current review team (Atkins et al., 2008). Quotes from participants in the original studies (as opposed to quote pertaining to the researchers' interpretations) are identified as such in the findings.

In the first and second order analyses, XX and XX identified recurring concepts from patient quotes (first order constructs) and the corresponding publication author interpretations (second order constructs). We explored the links between the key concepts from different articles by creating a comprehensive grid using Microsoft Excel software as recommended by France et al. (2015), in which we included the key concepts from each paper with the corresponding patient quotes and author interpretations and explanations. The studies were

represented in columns and the concepts were in rows which helped to establish the relationship between studies – i.e. the translation of studies into one another.

In order to synthesize the translations, we considered the nature of the relationship between studies. We looked for refutations within the studies as well as identifying the reciprocal relationships from which an argument could be developed. Third order (current review team constructs) interpretations were identified inductively and iteratively, and via entire research team analytical sessions following methods developed by Malpass (2009) and colleagues. The grid comparing study details and all concepts generated was used to track and document the process across the three orders to arrive at a final systematic analysis and synthesis output. Interpretations were presented to the wider research team and PPIE groups for debate and identification of congruence and priority of the interpretations. We drafted and re-drafted a diagrammatic model until we were satisfied the model represented the key/recurring concepts, and the relationship between these and the third order interpretations. For the qualitative sensitivity analysis, we explored the impact of removing lower rated studies (those judged by both researchers not to meet key criteria) from the analysis, but no changes in findings were discovered.

Study characteristics and quality

Figure 1 illustrates how final studies were selected, while Table 1 shows the characteristics of the 47 studies included in this review. Studies were published between 1997 and 2017, involved over 1,028 people living with CFS/ME, of whom over half were women. The studies were conducted in UK= 28; Norway= 7; USA= 7; Australia= 4 (1 of which also included participants from Canada) and Belgium= 1. Data were collected via semi-structured or unstructured interviews (n= 32); focus groups (n= 4); written accounts (n= 4); mixed interviews and written accounts (n= 3); mixed focus groups and written accounts (n= 2); mixed participant observation and interviews and focus groups (n= 1); mixed interviews and

focus groups (n= 1). Most of the studies were of high quality, meeting at least 7 out of 10 in terms of JBI criteria. The most common criteria which studies were judged not to meet were: ‘Is there a statement locating the researcher culturally or theoretically?’ and ‘Is the influence of the researcher on the research, and vice versa, addressed? These shortcomings may be at least partly due to word limits imposed by journals.

Results

Table 2 outlines the key/recurring concepts and their definitions from the translation across the 47 studies. Here, for each of the 16 key/recurring concepts, an illustrative quote is presented for the first order concepts (primary quotes from participants in the studies) and second order concepts (authors’ interpretations from the studies). A full list of all studies in which the concept was identified is also included. Figure 2 summarises the CFS/ME support model developed from our interpretations, and below, we summarise our main findings. The model shows the relationships between the key/recurring concepts and our overarching third order interpretations. It consists of the background or the ‘scenery’ against which the search for support takes places (e.g. invisibility), key points in the illness trajectory (e.g. first health encounters), and ongoing psycho-social processes that run alongside this trajectory (e.g. reconstructing a social circle and identity).

1. ‘Invisibility’ in relationships– gaining support for ‘an invisible illness’

CFS/ME is rendered invisible by the kind of sociality encouraged by the condition(s). The description of ‘invisible illness’ or similar constructs were frequently encountered in studies (Best & Butler, 2013; XXX blinded for anonymous review; Dickson, Knussen, & Flowers, 2007; Edwards, Thompson, & Blair, 2007; Hannon et al., 2012; Ware, 1992; Williams, Christopher, & Jenkinson, 2019). As a kind of “relational good” (or perhaps “relational evil” as Donati (2019) might put it) (p. 249), invisibility of CFS/ME emerges in relation to other people (e.g. family, friends and professionals), and the condition is mostly

only visible to those who have it. This overarching theme (third order interpretation) incorporates not only the invisibility of the illness in society where there are no specific signs of disease available (Key/recurring concept 1), but also the invisibility of the person as they are socially constituted: People with CFS/ME avoid places, situations and others perceived to negatively impact on their illness (Lian & Rapport, 2016). Although people may 'come out' to others, many others 'hide' the illness (Key/recurring concept 2).

Invisibility also relates to difficulties in receiving a CFS/ME diagnosis, which is reportedly time consuming. Challenges of diagnosis are compounded by the perception that CFS/ME is 'an illness that you couldn't really explain' to others, to gain their support (Edwards et al., 2007) (p207). Even when diagnosed, the condition continues to promote inconspicuousness. For instance, when symptoms are at their worst, CFS/ME can prevent people leaving their houses for work, socialising or consulting with professionals. Thus, indications of illness which might be more recognisable are less prone to be witnessed, with participants likely to be seen by others on relatively symptom free days (Hannon et al., 2012) (p6). Additionally, people may actively try to hide their condition, due to the perceived stigma of having a condition that is not widely accepted, and which may even be seen by some as a source of shame (Travers & Lawler, 2008). This means that at a societal level, the version of CFS/ME being produced for public display 'is not a true reflection of the illness' (XXX blinded for anonymous review) (p343), with all the subsequent lost opportunities for recognition and help. Those who do disclose their condition are taking a risk, as significant others and/or professionals respond variably, with reactions ranging from disbelief (Cooper, 1997; Dickson et al., 2007; Travers & Lawler, 2008), through to recognition and acceptance (XXX blinded for anonymous review; Pinxsterhuis, Hellum, Aannestad, & Sveen, 2015b). Financial support, via disability benefits needed by people with severe symptoms, is perceived as difficult to obtain, due to problems with legitimisation from government

institutions. CFS/ME has serious hidden financial implications due to the cost of treatments, potential loss of employment, not to mention the cost of obtaining practical help (Key/recurring concept 5. Illness at a cost). Such costs are compounded by the limited access to employment or disability benefits, as well as many workplaces reportedly not being well set up to adapt to the less severe, albeit variable symptoms, of workers with CFS/ME. Those with CFS/ME tend to be excluded (and rendered invisible) by work and financial considerations.

2. Relationship with health professionals: The first health encounters

This third order interpretation relates to the social processes of legitimisation and validation (key/recurring concept 3) and psychologisation (key/recurring concept 4). In initial consultations with health services, people with CFS/ME want to be taken seriously, and have their symptoms recognised as a genuine illness. However, many people with CFS recount (especially initial) consultations where their symptoms and/or condition are dismissed or 'trivialised' by professionals (Gilje, Soderlund, & Malterud, 2008; Taylor, 2005; Travers & Lawler, 2008; Ware, 1992). Participants give an impression that in consultations, professionals try to prevail with their particular interpretations of symptoms as psychological e.g., "...[he said] 'Oh you women, that's all you ever say...you're depressed'" (participant, Cooper, 1997, p. 198). Subsequently, participants, alert to the risk that professionals might not take them at face value, can become defensive about hints of psychologisation (Ax, Gregg, & Jones, 1997; Gilje et al., 2008; Guise, Widdicombe, & McKinlay, 2007; McCue, 2004). Participants experience CFS/ME foremost in physical ways, where psychological symptoms tend to be considered secondary. Thus, they wanted their physical symptoms – which are the ones that are critical for them – prioritised. Nevertheless, some participants said that having comorbid psychological issues like depression recognised and treated could be helpful, especially where emphasis is placed on psychological aspects as a *consequence* of

illness, rather than a cause (Brown, Huszar, & Chapman, 2017; Picariello, Ali, Foubister, & Chalder, 2017; Wheeler, 1992).

Lack of recognition and perceived support in medical consultations (e.g. “I’d been rejected here, there and everywhere and *made* [our emphasis] to feel like an idiot”) (participant, Edwards, Thompson & Blair, 2007, p. 208), highlights the creation of difficult relational goods (e.g. shame). This means some participants turn to complementary or alternative therapies for recognition and help (Key/recurring concept 8: Searching for alternatives). These latter consultations typically involve greater legitimisation of the whole person, and in some cases, there were perceived beneficial effects (e.g. “...acupuncture ...gave me a sort of energy boost...”) (Ax, Gregg & Jones, 1997) (participant, p. 253). Positive results were not assured, however, and the significant costs involved could exacerbate the financial burden associated with CFS/ME (Key concept 5: Illness at a cost) (Ax et al., 1997; de Carvalho Leite et al., 2011; Hannon et al., 2012). Some participants turn away from complementary or alternative therapies due to “high costs and minimal benefit” (J. S. Anderson & Ferrans, 1997) (p. 363). Participants noted that peers with CFS/ME “became important alternative sources of information and support” when professional routes failed (Brooks, King, & Wearden, 2014) (p. 9).

3. Person-centred and relational care

Person-centred care – increasingly recognised as essential to quality healthcare – involves patients working together with professional carers to plan care, where carers (health and social) attempt to understand patients and their communities as a whole, in essence giving them as much control of their care as possible, and then bringing together the services that best ensures the outcomes valued by patients, not to mention better equity in care for patients [XXX blinded for anonymous review]. This overarching theme (third order interpretation) particularly reflects the Key/recurring concepts 6 (Seeking patient-centred

care) and 13 (Support that is flexible, adaptable and understanding). It also incorporates the ideas of Ongoing support (Key/recurring concept 15) and equity in support (Key/recurring concept 14). In relation to professionals, a deep need for relational goods like recognition, acceptance and validation emerged from the studies. Participants wanted their clinicians to share in their understanding of CFS/ME, as a means to begin to offer “a route out of this cycle of fatigue” (XXX blinded for anonymous review) (p. 575). It is when participants trust that the relationship with a professional is safe and respectful, that participants are freer to explore their more complex feelings about CFS/ME, and impacts on their lives, including the psychology. Participants expect health professionals to provide a foundation of understanding, from which individualised management strategies and a range of options emerge (Gladwell, Pheby, Rodriguez, & Poland, 2014; Peters et al., 2011). One participant put it this way, ‘her [health professional’s] empathic nature, was her greatest skill, anything else for me came secondarily’ (Peters et al., 2011) (p9). In relationship to professionals (and more widely), people want to be listened to, taken at face value, have their feelings understood, and feel valued. Here, they want a sense that their clinicians know something of what it is like to experience CFS/ME (V. R. Anderson et al., 2014; Peters et al., 2011; Ryckeghem et al., 2017; Ward, Hogan, Stuart, & Singleton, 2008). It should be noted that some participants pointed out that women (and Black or ethnic minorities) can have heightened problems eliciting empathy from professionals (Arroll & Senior, 2008; Cooper, 1997; de Carvalho Leite et al., 2011). In general, the relational approach requested of clinicians involves flexibility in terms of adapting to unique patient circumstances, including their choices, preferences, and limitations. Practical support, flexibility in frequency and type of appointments, is also valued (Broughton, Harris, Beasant, Crawley, & Collin, 2017). Participants need support for the stage and type of condition they have, e.g. to accept that their condition may be long-standing, or that there is hope for improvement and/or recovery.

Subsequently, positive relations can be internalised by participants, e.g. “I feel that I’m having more consideration for myself than ever before...” (Pinxsterhuis et al. 2015b) (participant, p. 121). Participants prefer support that has continuity, so that they have a “safety net” to fall back on when things are difficult (Broughton et al., 2017) (participant, p. 7). When participants had “flown the nest”(Broughton et al, 2017) (participant, p.7) in terms coming to the end of a helpful kind of support (e.g. by being discharged from a specialist health service), there can be feelings of abandonment. Thus, reassurance that they could return for – or call on – support, if struggling in the future, was comforting for such participants.

4. Wide ranging support needed for all aspects of daily living

Donati (2019, p. 255) is clear that it is the “social fabric that produces the [relational goods] and is both enabled and constructed by them.” People who are more severely affected by CFS/ME need to call on their relationships with an especially wide range of people including significant others, professionals (health and social care), families, colleagues and friends (J. S. Anderson & Ferrans, 1997; de Carvalho Leite et al., 2011; Edwards et al., 2007; L. Larun & Malterud, 2011; C. XXX blinded for anonymous review; Stormorken, Jason, & Kirkevold, 2015; Ware, 1998). As one participant put it, they required the “Full Monty”¹ of support (Key/recurring concept 16), reflecting the fact that she needed a range of support (de Carvalho Leite et al. 2011) (participant, p. 8). Another participant explained, he “...needed complete rest and somebody [else] to do the shopping, to do the cooking ...” (de Carvalho Leite et al., 2011) (participant, p. 8). With increased severity of illness, help was needed to cover increasing facets of people’s lives, including medical/health, practical, social, and

¹ According to the Cambridge Dictionary, the Full Monty refers to “...the most or [best](https://dictionary.cambridge.org/dictionary/english/full-monty) that you can have, do, get, or [achieve](https://dictionary.cambridge.org/dictionary/english/full-monty), or all that you [want](https://dictionary.cambridge.org/dictionary/english/full-monty) or need.”
<https://dictionary.cambridge.org/dictionary/english/full-monty>

financial. [XXX blinded for anonymous review] and colleagues (2011) (p. 575) found that “practical pressures from work or family, escalating worry about how to cope, increased strain on relationships and worsening physical and mental exhaustion” combine to create a particularly “vicious circle”. Given that professional support costs are prohibitive for most, informal supporters may be asked to take over daily functions of the person, e.g. ensuring health care needs are met, helping with social participation, or covering expenses (Donalek, 2009; Reynolds & Vivat, 2010; Williams et al., 2019). Significant others thus play a role in supporting the functionality of people with CFS/ME throughout the course of their illness, from pre-diagnosis onwards. However, a ‘Catch 22’, (key/recurring concept 7) was identified by some participants. Severely affected people can find it exhausting to do things by themselves, but equally, having to explain and justify needs in order to gain help from others could also contribute to fatigue. Williams and colleagues (2016) (p. 7) reported that participants found “informing [people] of how, when and why is almost as exhausting as completing the task themselves”. Yet, without such help, daily living could become difficult or impossible.

5. Social circles and identities

People with CFS/ME describe how their social circles shrink over time, due to the limitations set by their illness and the difficulties they had coping with symptoms (like fatigue), preventing them from fully engaging with friends and family (J. S. Anderson & Ferrans, 1997; Best & Butler, 2013; Reynolds & Vivat, 2010; Travers & Lawler, 2008). CFS/ME exerts a strain on relations that sorts out who is (and is not) going to be supportive, e.g. “people I thought I could count on weren't there for me...people who I thought were mere acquaintances turned out to be my real friends” (Anderson & Ferrans, 1997) (participant, p364). Reportedly, friends can find it difficult to accept symptoms, understand the illness, as well as cope with the loss of activities that initially formed the basis of

friendship. In turn, participants report being unable to give back to friends as they once did, creating a loss of reciprocity. Importantly, although people with CFS/ME may need assistance to do so, participants report forming new relations and friendship networks post-illness with peers who have similar experiences e.g. “I’m developing another circle of friends through CFS” (Travers & Lawler, 2008, participant, p. 322). Others sought activities more suited to their current capabilities and found that these led to new networks: “...it’s budding, it’s opening up again” (Travers & Lawler, 2008) (participant, p322). In many cases, new friendships were perceived as more accepting, supportive and/or sustainable, and fitted within the limits created by the illness (J. S. Anderson & Ferrans, 1997) (p364). Where people had recovered enough, ‘giving back’ by providing support to others more severely affected by CFS/ME could be rewarding (Broughton et al., 2017). For example, appreciation about the “limited information and advice about recovery, prompted [participants] to feel responsible for advising others about this process (of recovery)” (Brown et al., 2017) (p. 705).

Finally, a sense of identity (prior to illness) emerged from participant work roles and social networks. However, at some point in their illness, their identity undergoes a kind of ‘crisis’ linked to the loss of such affiliations (Dickson, Knussen, & Flowers, 2008), e.g. “It was like a death. You had to grieve for that...” (Travers and Lawler, 2008) (participant, p. 321). In particular, participants tended to lose aspects of their previous identities when no longer supported via relations (e.g. best friend), and the loss of self can initially seem complete, e.g. “I felt for a while that I had no identity apart from just being a sick, non-person” (Reynolds & Vivat, 2010) (participant, p. 70). However, participants sought new social activities and networks, which could in turn support the development of a valued sense of self. Here, sharing personal vulnerabilities and stories openly with others, could help create supportive networks and ways of understanding their condition, as well as reassure and promote a sense of coping with the illness, e.g. “[realising um] that other people were

experiencing exactly the same thing” (Broughton et al., 2017) (participant, p. 6). Thus, it is supportive social circles, with the generation of relational goods (like feeling helpful towards others, feeling seen by others) that helps reinvigorate wellbeing and self among those with CFS/ME.

Discussion

This is the first meta-ethnography designed to integrate the different components of the CFS/ME experience of support (e.g. invisibility) using a relational approach. Specifically, we used Donati’s (2019) relational goods framework to take the focus off individuals, and instead direct attention to the “irreducibly relational” nature of support – or lack thereof. While Donati (2019) uses the term ‘relational good’ to focus on experiences that are morally desirable, other writers use the term in less moral ways, so that goods can be more or less positive or negative (Ulhaner, 1989). While retaining the full sociality of Donati’s approach, we use the term in this latter way. Importantly, our framework showed that while dependent on social networks, such goods (e.g. recognition) did not themselves belong to individuals *per se*. Rather, these feeling-based qualities are better thought of as being created socially, and circulated in and around the actors involved. Additionally, such goods are not possible to generate outside of sociality, whether informal or personal-professional in nature. Our approach to synthesis revealed how the key bits of meaning related to CFS/ME are circulating communally, dependent on the social fabric available to participants for coping with CFS/ME. Thus, our interpretation suggests it is now timely to move the conversation away from the current emphases on individualised interpretations of CFS/ME (e.g. personal recovery journeys, identity loss and reconstruction) towards a high level of sociality. For better or for worse, these relational goods are constructed and shaped by social structures available, including lay understandings of fatigue that circulate in society, medical training, social institutions involved (like NHS healthcare), and even the non-human things in the

equation (like financial resources) (Donati, 2019; Dépelteau, 2015). Here, it is important to note the relative lack of power of actors with CFS/ME, and how formidable the structures that promote the conditions to allow useful social goods to flourish (e.g. via mores supportive social networks and person-centred care), or less useful goods to be established (e.g. via top down authoritative structures), can be.

As illustrated in Figure 2, and as highlighted in previous research in this area (Best & Butler, 2013; XXX blinded for anonymous review; Dickson, Knussen, & Flowers, 2007; Edwards, Thompson, & Jenkinson, 2019), we uncovered invisibility as a key, complex emergent good produced out of ME/CFS experiences. Clearly, invisibility is not unique to CFS/ME, as it features in conditions like depression, back pain and diabetes, and even multiple sclerosis (Joachim & Acorn, 2000; Glenton, 2003; Methley et al., 2016). However, we emphasised that invisibility has a specific multifaceted and emergent pattern in CFS/ME, dependent on the way patients, significant others, professionals and healthcare organisations behave. Invisibility was variously promoted by: the way that there were no clear observable signs nor diagnostic markers that professionals could observe in patients; the particular difficulties patients had in having their symptoms recognised and taken seriously from the very first health consultations; substantial delays in receiving a diagnosis; lack of readily available treatments (e.g. back pain and depression has medication available for treatment); current dangers that the value of person-centred care is dismissed for CFS/ME; the kinds of social stigma that encouraged participants and their families to hide CFS/ME from view; as well as the varying severity of the condition, resulting in participants becoming more socially isolated when disabled.

The findings of previous meta-syntheses (e.g. Anderson et al., 2012; Larun and Materud, 2008; Pinxsterhuis et al., 2015) revealed that participants understood health encounters as potentially hazardous to navigate, with a range of risks involved for patients,

including being discredited by professionals; not experiencing the empathy and the understanding they yearn for; having the condition trivialised or psychologised in a manner that did not fit with their current understanding of their illness; and not receiving a timely diagnosis that could legitimise and give social recognition to their experiences [XXX anonymised for blinded review]. This risk dynamic can set up consultations as a battle, whereby both parties try to prevail in terms of their interpretations. The stakes in this battle can be high for patients, as doctors are seen as gatekeepers to legitimation and valuable specialist health services. Here, patients face the potential of having their experiences dismissed, and of being rejected, and in ways that can be experienced as especially traumatic (Baruch, 1981). Such lack of recognition has wide ranging impacts (Gladwell et al., 2014), including on the ways participants relate to themselves (negative relations are internalised), others (e.g. feeling blamed and abandonment), as well as their finances (including difficulties in gaining disability benefits). Alternatively, positive interactions, empathy, and continuity of such relations can be internalised by participants and lead to validation and wellbeing. Validation that a health condition is genuine can be particularly important to patients experiencing conditions like CFS/ME that are stigmatised, given the potential for CFS/ME patients to be cast as somehow bad and/or malingering (Dickson et al. 2007; Dickson et al. 2008). For some patients, the potential for discrimination may reinforce peoples' beliefs that their illness is largely biological in origin, with outside causes and serious corollaries (Moss-Morris, 2005), partly as a way of side-stepping stigma.

In terms of health interactions, the notion of person-centred care has been emergent since the 60s, elevating the importance of patient expertise, as well as the primary role of personal relationships in care, not to mention the importance of considering wider social contexts affecting patients (Santana et al., 2018). Not surprisingly, participants appreciated CFS/ME specialist services which were considered person-centred. However, these services

were not always available, accessible or affordable. Some participants reported receiving person-centred care, but more commonly reported situations where professionals appeared uncomfortable with patient expertise. Additionally, participants report needing to draw on a range of practical, social and financial help beyond the clinic, and it can be challenging to explain the need for such support to others. While flexible and adaptable support is sought, the reality is that many of newly diagnosed people will experience social isolation with loss of contact with friends and work, although they may build more supportive and understanding social networks in time (e.g. via peers who themselves have experienced CFS/ME). People's identities also undergo transitions as they lose previous work and social roles that are not initially replaced, they may even come to feel like a "non-person". They may have to grieve for the "death" of their former self that once emerged from (or was supported by) previous affiliations. However, new supportive and validating networks can be established (e.g. "it wasn't all in my head") (Broughton et al., 2017, participant, p. 6). Here, better coping can be explored (e.g. not over-identifying with the condition); and a new valued identity may emerge (e.g. as someone who can help others with CFS/ME).

The involvement of the PAG in the process of designing and conducting the meta-ethnography added a crucial dimension to our work. The scope of the concept 'support' was extended based on discussions with the group. This both increased the validity (and scope) of our interpretation of support and, correspondingly, the number of qualitative studies that therefore might contribute to the synthesis. Nevertheless, a broad approach was requested by the PAG. The meeting at which the participants used our key concepts to tell their own stories was particularly insightful and valuable; in some cases concepts were prioritised based on relevance, referred to in a chronological order, or organised into categories to reflect different aspects of the person's experiences. Overall, this process helped to emphasise which interpretations to prioritise.

Limitations

Although we examined beliefs and perspectives of service user participants in relation to others, relationships cannot be fully explained by the views of one side. While beyond the scope of this meta-ethnography (as too many papers would have been retrieved to do the analysis justice), a future relational analysis from the perspectives of healthcare professionals and other carers is important. Additionally, we excluded papers written in a non-English language. While we initially planned to identify studies in languages other than English, we determined that translation may affect the original interpretation of the findings. We also realised that to fully assess relevance of studies, they would require translation, and this was not feasible within the resources of our study. While this is a limitation, the studies we included represented views of patients from a number of countries with widely varying health care systems (Australia, Canada, Belgium, Norway, UK, USA). Ethnicity was not reported in the majority of studies, but a range of ethnic groups were represented in the studies, and the potential impact of ethnicity on support did arise as a component of the key concept 'equity in support' (see Table 2). However, including additional, foreign language papers might potentially revealed culturally mediated CFS/ME perspectives we have not considered in this review, thus influencing the overall interpretation of findings. Similarly, we did not carry out extensive searching of the grey literature nor included theses which may also have revealed additional insights. While the aim was for a comprehensive search for relevant literature, the large number of citations retrieved meant that including further studies through extensive searches of the grey literature was not feasible.

With the large number of diverse studies included, we could have carried out a number of different sensitivity analyses. However, we focused on comparing findings over time, as well as the effect of the quality of the studies. We also focused on the patients' perspectives, and we did not extract findings related to other categories of participant.

Severity of the condition was raised by patients as an issue and was an aspect we considered during the analysis. We did not carry out an analysis on the sources of recruitment of patients, or investigate in detail the extent of any variation in definitions of CFS/ME. However, these elements are presented in the Table of Characteristics (Table 1), and might usefully be the focus of future studies. Finally, the focus of this meta-ethnography was on support, and support needs, rather than participants' illness beliefs, although this is an important dimension of the CFS/ME experience, and the attitude of others' towards people symptoms was mentioned within the PAG. Attitudes of - and support from - others is included in our model. Further qualitative investigation (either by meta-synthesis or primary data generation) may provide more in-depth understanding on how beliefs influence and interact with perceived support needs.

Conclusion

Early on, it became clear it was impossible to interpret the range of concerns facing participants with CFS/ME that had been elicited in the included studies (like the risk of the clinical consultation), without focusing on how the CFS/ME experience was largely a product of relationships. Our analysis showed that a focus on the entities produced by relationships, like individualised identities, risked missing the bigger picture. Without developing the field beyond individualistic journeys and frameworks, the multifaceted and relationally sustained realities of CFS/ME remain hidden from view. Doctors admit that their medical education fails to equip them with the skills necessary for managing the complexities of patients with CFS/ME (Bayliss et al., 2014). Our synthesis suggests that health professionals may be assisted by viewing solutions for those with CFS/ME as emergent out of interactions (however conflicting) with patients, producing predictable experiences e.g. both patients and professionals feeling unheard. Importantly, professionals do find ways to collaborate with patients to reach agreement on symptom management and good self-care (Anderson et al.,

2012). People with CFS/ME long for such partnerships with health professionals who are not threatened by patients' (many times differing) knowledge about CFS/ME, where conflict with medical knowledge is inevitable. Our research suggests a relational paradigm shift is needed to address the dynamics that construct professionals and people with CFS/ME in particular ways, e.g. as blameworthy. Previous work has been done to try to achieve a paradigm shift in the management of people with CFS/ME in the UK, but clearly this remains a considerable challenge (Bayliss et al., 2016). Nevertheless, we end by noting that by shifting the focus away from individuals (and blame), and instead focusing on a framework about the co-creation of relational goods, novel ways of addressing CFS/ME are possible. Such an approach – of maximising valued relational goods, and minimising less helpful goods – might be more acceptable to a range of stakeholders. Dissemination of the results and framework from this study to the primary care community, along with focussed training developed in collaboration with the PAG, could shift clinician (and societal) attitudes towards greater understanding and receptivity towards the experiences and needs of patients with CFS/ME.

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