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A Review and Meta-Synthesis of Qualitative Studies on Myalgic Encephalomyelitis/Chronic Fatigue Syndrome

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Abstract

Objective—To review and synthesize findings across qualitative studies on Myalgic Encephalomyelitis/chronic fatigue syndrome (ME/CFS).

Methods—Articles were systematically reviewed and analyzed within a meta-analytic framework. Analyses included a multi-perspective (e.g., individual, physician, familial) examination of ME/CFS, as well as a comparative analysis of ME/CFS versus other chronic conditions.

Results—Thirty-four (34) qualitative studies on ME/CFS were identified and included. Findings include three substantive thematic areas that focus on studies concerning: (1) experiences of people with ME/CFS, (2) experiences of physicians, and (3) themes that intersect both of these groups. For patients, illness development influenced identity, reductions in functioning, and coping. Physician-specific themes described lack of awareness about the illness and recommended improvement in medical education. Themes that intersected expressed issues with diagnosis which creates tensions and fuels the stigmatization of ME/CFS.

Conclusions—Findings indicate multilayered, context-specific experiences and ways in which both people with ME/CFS, as well as the people involved in their lives, such as family or the medical community, understand their illness. Future qualitative studies should coalesce the various facets of the ME/CFS experience, the network members of people with ME/CFS, and the sociocultural environment through which the illness is understood.

Practice implications—Health care professionals can gain unique insight from patient experiences, allowing for more accurate diagnoses and treatment recommendations.

Keywords

chronic fatigue syndrome; Myalgic Encephalomyelitis; qualitative methods; review; meta-analysis

1. Introduction

Chronic fatigue syndrome (CFS) is a highly complex medical condition and has been referred to as postinfectious or postviral fatigue syndrome, myalgic encephalomyelitis (ME), Royal Free disease, chronic mononucleosis, and chronic Epstein-Barr virus [1,2,3,4]. We will refer to this illness as Myalgic Encephalomyelitis/chronic fatigue syndrome (ME/CFS). As characterized by the most widely used case definition, ME/CFS is disabling fatigue for six or more months and four out of eight core symptoms including: impaired memory or concentration, headaches, sore throat, lymph node pain, muscle pain, joint pain, unrefreshing sleep, and post-exertional malaise [5]. A clinical case definition of ME/CFS was developed in Canada [6,7]. The prevalence of ME/CFS has been estimated to be .42% among adults in the United States [8]. Unfortunately, ME/CFS is often either inaccurately or underrepresented in major sources of medical education for health care providers [9].

Over the course of the past two decades, approximately 35 studies have been published worldwide that used qualitative methods in attempt to understand ME/CFS. This article provides a systematic review and analysis of qualitative ME/CFS studies within a meta-analytic framework. Qualitative meta-analysis, also referred to as a meta-ethnography, synthesizes interpretive research by analyzing the relationships among the studies, translating the findings, and expressing the findings in a solidified, written format [10].

The aim of this review is to concretize the major thematic findings of these previous studies and make recommendations for future qualitative work. Recently, Larun and Malterud [11] published a meta-analytic review, overlapping with some of the studies included in this analysis. The present article differs from the review by Larun and Malterud in that our study includes 34 empirical articles whereas their study only included 20 studies published up until 2006. What particularly differentiates this meta-review is the inclusion of qualitative studies that take a multi-perspective examination (e.g., physician, families), as well as includes comparative studies (e.g., experiences of people with ME/CFS versus people with fibromyalgia). While it is certainly important to focus on the experiences of people with ME/CFS, it is also critical to examine these other perspectives in order to better understand the context of ME/CFS. Thus, this article reviews a larger selection of studies than that of Larun and Malterud, as well as includes a greater breadth of qualitative methodologies while examining these multiple perspectives.

2. Methods

This analysis provides a comprehensive review of studies on ME/CFS that, broadly defined, utilize qualitative methods. Qualitative methods aim to identify interrelated themes across cases and analyze the themes through a scientific process that is both creative and multilayered [12,13]. This review includes studies of different populations of interest including physicians or other health care professionals, experiences of people with ME/CFS, a comparison of people with ME/CFS versus other chronic conditions such as FM or cancer-related fatigue, and family system responses.

Most studies used a grounded theory approach to the analysis of data. Grounded theory as a methodological framework is rooted in phenomenology and the goal is to identify central, context-dependent psychological and structural processes [14, 15]. The grounded theory and constant comparison analysis process, as outlined by Glaser and Strauss, involves reading all interviews, identifying and categorizing themes, re-reading and revising, making comparisons, reviewing the codes, and reflecting. Table 1 provides a complete overview of every article used in this analysis. The table includes the specific make-up of the population

studied, the case definition used, the methods the researchers used for inclusion and classification of participants, the methodological strategies, and tools used in the analysis.

Article selection was initiated by searching for peer-reviewed qualitative studies on ME/CFS up until May 2010 within the following major scientific databases: ProQuest, PubMed, PsycInfo, Web of Science, and Google Scholar. We searched topic and subject headings with the following keywords: qualitative, ethnography, narrative, grounded theory, chronic fatigue syndrome, Myalgic Encephalomyelitis, Myalgic Encephalopathy, ME/CFS, CFS, ME, post-viral fatigue syndrome, chronic mononucleosis, and chronic fatigue immune dysfunction syndrome. The search produced over 1,000 articles and 325 articles were originally screened for this review. Of those 325 articles, two were articles retrieved via personal communication with an author. Seventy-five (75) articles were assessed from this pool. Studies were assessed by identifying salient methodological aims, sampling and design, analytic strategy, theoretical grounding, and a clear discussion of research implications. Forty (40) articles were further excluded from this sample. Studies that were not methodologically rigorous or did not include this level of detail were excluded from the synthesis. As well, studies that did not primarily examine ME/CFS were excluded. The final synthesis included 34 empirical qualitative studies and one previous meta-analysis. See Flowchart 1 for the number of studies included and excluded throughout the identification, screening, and eligibility processes using the PRISMA standards [16].

The analysis phase began with reading each of the articles. The researchers then proceeded to analyze each of the articles based on three substantive areas. First, the authors identified major theoretical differences among the articles. Major theoretical differences might include the focus and scope of the study, composition of the population of interest in the study, or the theory of thought about the illness to which the researcher subscribed. Second, major themes and findings of each study were identified. Third, implications and recommendations for future research were identified. The authors then synthesized the theoretical differences, major findings, and recommendations. This analysis is organized in Table 2 and further explained in the results section.

3. Results

The synthesis of themes is broken down into three sections: (1) themes that are specific to studies focusing on people with ME/CFS, (2) themes that intersect studies including both patients and physicians, and (3) themes that are specific to studies focusing on physicians. It should be noted that these themes and groups are neither mutually exclusive nor exhaustive.

3.1. Patient-specific themes

The group of patient-specific themes includes an analysis of illness phases in connection to chronicity and identity changes, reductions or losses experienced due to ME/CFS, and descriptions of coping mechanisms. Each of these areas also includes subthemes.

3.1.1. Chronicity and identity changes—A number of studies found that people with ME/CFS experience a change in identity throughout the course of their illness [17]. These changes can lead to a profound shift in the relationship between the body and the mind [18]. This is consistent with Whitehead's qualitative work [19], who found that people with ME/CFS experience a disruption in and reconstruction of their identity when living with ME/CFS. In addition to disruption in identity, Clarke and James [20] found that people with ME/CFS expressed a loss of confidence and self-esteem. Disruptions in self-perception and loss of identity in the development and progression of ME/CFS have been noted in other qualitative studies [21, 22, 23]. Donalek's [13] qualitative research investigated the ME/CFS experience within a multilevel family system framework (individual level, family level, and

across families) and found an identity disruption at the family-unit level. This disruption was largely implicated in the transformation of family roles (e.g., children taking on additional responsibilities).

Another emergent theme centered around various stages of change, consistent with findings in other studies on disruption in identity in people living with chronic illnesses. This fits well with Fennell's [24] four-phase theory examining the phases of ME/CFS illness experience. The Fennell phases include periods of crisis, stabilization, resolution, and integration, and empirical support has documented these stages [25, 26]. The qualitative ME/CFS literature supports the existence of distinct illness phases as well. Whitehead [19] outlines three distinct phases in identity reconstruction. First, there is an acute phase where there is a disruption in identity. Whitehead uses the example of adopting the "sick role" in the acute phase. The second phase, called the medium term, is when the person accepts being sick. Finally, in the long term phase the person experiences a reconstruction in identity and develops coping strategies [19]. People with ME/CFS are constantly in search of self-renewal through their experiences with loss, healing, and evolution [27].

Travers and Lawler [27] found that people with ME/CFS go through cycles of health and unhealthiness and that there are transformative aspects of the chronic illness experience. Travers and Lawler suggested this is likely related more to the chronic nature of the condition rather than an ME/CFS-specific experience. Chronicity, as it relates to phases of illness, contributes the cycle of being overwhelmed by and eventually learning to live with the illness. Travers and Lawler reviewed the historical context of contested illnesses where previous studies have described ME/CFS in a way that makes patients accountable for the cause of their illness due to a psychosomatic explanation [28, 29]. Shifting accountability from the medical system to the individual patients is one way in which the societal response blames the victim [30].

3.1.2. Development of the illness and etiology—Six of the qualitative studies specifically addressed etiology and factors related to illness development. Stressful conditions and life events have been implicated as factors in the development of various chronic health conditions including ME/CFS [31, 32, 33, 34]. Lovell [33] examined aid workers who developed ME/CFS while overseas and found that the participants were very healthy prior to their onset, which was initiated by stressors and physical causes. One possible contributor to the development of ME/CFS was overwork and strenuous living conditions. Similarly, Clements and colleagues [31] established that almost all of the participants, drawn from primary, secondary, and tertiary care settings, believed that physical factors and disease were implicated in the course of their illness. More than half of the participants believed that stress, including social stressors and over-activity, played a role in the development of their illness. Whereas all of the participants in Whitehead's [35] study believed that they had the flu or an acute virus at illness onset. Donalek [13] described a similar process in which participants with ME/CFS attributed the development of their symptoms to the flu, migraines, stress, or overwork.

Findings also indicated that etiologic differences in ME/CFS were found for men and women [36, 37, 38]. Men and women experienced the same symptoms, but have different etiological pathways; men described their illness onset as related to working with chemicals, whereas women described their illness onset as related to stress and surgery [37].

Horton-Salway [39] interviewed a woman with ME/CFS and her partner to identify how ME/CFS is constructed as a physical illness through the use of attributional and identity narratives; descriptions of pre-ME/CFS life provided grounding for the current explanation of their illness. For example, Horton-Salway analyzes part of the conversation where both

the participant and her partner discuss an event—exposure to a virus—as a potential origin. These causal links provide insight into the organic genesis of her illness rather than a psychological attribution. This suggests that researchers should be less concerned with “true” descriptions, but rather with the interactional contexts in which illness narratives are developed. These narrative analyses function as a way to make general storytelling patterns within a community recognizable [40]. Overall, the qualitative literature looking at patient illness perceptions focuses on the organic, physiological development of ME/CFS. This will be discussed further in the intersecting themes section under symptoms.

3.1.3. Reductions across physical, social, and economic domains—Current case definitions of ME/CFS refer to substantial reductions in functioning for individuals across occupational, education, personal, or social domains [5, 41, 42, 43]. The qualitative literature supports this as an important domain of ME/CFS. In particular, people with ME/CFS describe social and economic reductions, as well as personal losses and disruptions, in addition to the physical reductions experienced in everyday life.

Economic and occupational reductions are a salient experience in the lives of people with ME/CFS [13, 35, 44]. People with ME/CFS suffer from profound and multiple losses concerning their jobs and finances, including major disruptions to their careers and financial instability [21, 22, 23]. Ware [45] studied role constriction in employment and found that people with ME/CFS tend to experience difficulties at work, particularly due to cognitive impairments, where 50 percent of the participants reported unemployment as a direct result of ME/CFS.

Another dimension of substantial reductions concerns the loss of social roles and major disruptions in personal relationships [22, 23, 44, 46]. People with ME/CFS described reductions in the form of a loss of stamina and ability to develop future plans [21]. Additionally, people with ME/CFS described changes in relationships, activities and social networks because of the illness. ME/CFS has a profound negative impact on the social lives of people that are affected with the illness [20, 47].

3.1.4. Coping and balancing activity—Qualitative studies differentiated the emotional and coping responses based upon which symptoms are present and the severity of the symptoms among patients with ME/CFS [22]. For example, people who developed ME/CFS used various coping mechanisms such as seeking support from others, religion, and using selective comparisons (“there are other people worse off than me”) to deal with their new illness [33]. In particular, Whitehead [35] noted that it was common for people with ME/CFS to use alternative medications and therapies.

Ray, Weir, Stewart, Miller, and Hyde [48] developed a model of four coping strategies to manage ME/CFS including maintenance of activity, accommodating to the illness, focusing on symptoms, and information seeking. The most common of these themes across the studies was the description of patient efforts to balance activity. Consistent with the various camps of thought about the nature of ME/CFS, there are also conflicting suggestions about what is best for patients when it comes to balancing activity as a treatment or coping mechanism. Participants described that they are somewhat able to control symptoms by reducing activity [31]. The response to ME/CFS is complex and many people with ME/CFS practice “living within [their] limits” by monitoring and/or self-initiating the restriction of activities [27]. Ware [45] explored the experience of role constriction in employment due to ME/CFS and effective ways to resist the constriction. For example, people cope with ME/CFS by reducing activities and accommodating to their illness.

Findings from qualitative literature stress the importance of pacing oneself [20, 35]. ME/CFS may hinder one's ability to perform activities or cause them to avoid activities altogether, which may in turn reinforce ME/CFS symptoms [17]. Gray and Fossey [17] implied that activities are crucial to maintaining well-being. Lombaard and Mouton [18] concluded that the distress caused by ME/CFS can be repaired when the self makes efforts to actively listen to the body and balance activity and restriction. This model of balancing activity is supported by the envelope theory [49, 50], which posits that people with ME/CFS should try to balance their perceived and expended energy levels, thus staying within their energy envelope [51].

3.2. Intersecting themes

The intersecting category includes themes addressing symptoms of ME/CFS, issues involving diagnosis, and patient-physician power dynamics. For example, physicians and patients both explain symptomatology but there may be variance in their descriptions or general ideology. Nonetheless, it is important to address overlapping areas as well as the dichotomies.

3.2.1. Symptoms—The three of the most common ME/CFS symptoms described by participants in the studies reviewed included extreme fatigue, disabling pain, and cognitive problems [37, 44]. In another study, the participants described intense cognitive dysfunction as a key part of their illness experience [21]. Soderlund, Skoge, and Malerud [47] recruited people with ME/CFS from a local support organization. Many of the themes Soderlund and colleagues found focused on symptom experience. For example, participants provided descriptions of severe exhaustion and pain, problems with auditory perceptions and sensitivity to sound, and deterioration in concentration. In Lovell's [33] study, participants provided in-depth descriptions of their physical symptoms including "debilitating fatigue; unrefreshing sleep; muscle and joint pain; headaches; dizziness; solid, swollen or throbbing glands; sore throats; eyesight unsteadiness, and concentration and memory problems" (p. 18).

Comparative qualitative studies have focused on ME/CFS experiences in relation to other chronic or fatigue-related illnesses. For example, Bennett, Goldstein, Friedlander, Hickie, and Lloyd [52] identified a similar core set of symptoms and consequences in people with cancer-related fatigue and people with ME/CFS. However, participants with ME/CFS reported more musculoskeletal pain and influenza-like manifestations than those with cancer-related fatigue. ME/CFS is described in multiple studies as a serious, enigmatic illness that is not psychological in nature and should be understood as a physiological illness mediated by immunological responses [47, 53, 54].

3.2.2. Issues with operationalizing ME/CFS—We also found that people with ME/CFS use descriptive and linguistic devices to portray their illness as well as explain what their illness is *not* [54]. Hart and Grace [38] provided a critical examination of how fatigue is not well operationalized. The major themes in the study include difficulties with cognition and communication, low energy and difficulty with mobility. The findings of Hart and Grace's [38] study examine fatigue as a lack or an absence of something, which makes it difficult to define in biomedical terms.

3.2.3. Variability—A number of studies indicated that people with ME/CFS experience wide variability in their symptom presentation with regards to both frequency and severity [21]. In addition, people with ME/CFS experience variation in fatigue type [17]. Horton-Salway [55] interviewed general practitioners in the United Kingdom and established that physicians construct and justify patient illness experiences as psychosomatic through the use

of the biopsychosocial model [56]. These findings also supported the use of narrative analysis in illness construction as a basis for scene-setting and understanding context, previously described in detail in Reissman [57]. Horton-Salway [55] suggested that the rhetoric of physicians and researchers may be used to shift blame from the medical field, in which clinicians, investigators, and science do not yet understand the full nature of the condition. Consistent with victim blaming theory [30], the psychosomatic approach is a way to manage blame within a lack of scientific understanding, as Horton-Salway [55] describes it as a way to allow for a “low accountability for doctors and high accountability for patients” (p. 416). This is compatible with other theorists such as Richman and Jason [58] who have described a paradigmatic shift in which this failure of the medical system to find biological markers in people ME/CFS reinforces social and psychiatric explanations.

3.2.4. Stigmatization—Asbring and Narvanen [59] compared two groups of participants, one group comprised of people with ME/CFS and the other group comprised of people with fibromyalgia and found that ME/CFS was more trivialized and psychologized than fibromyalgia. As well, Clarke [37] found a gendered response in terms of treatment; women with ME/CFS were more likely than men to be sent to a psychiatrist, as it seems that some physicians have a proclivity to explain women’s ME/CFS illness experience as psychosomatic.

People with ME/CFS described the stigmatizing aspects of their illness including the psychologizing of symptoms by doctors, family members, and friends [13, 35]. For example, some physicians felt that people with ME/CFS or fibromyalgia experience symptoms that can be lived with and that patients tend to exaggerate the severity of their symptoms [60]. Clarke and James [20] described the disputed nature of the condition by the medical community and the alienation experienced by people with ME/CFS as one of the “distinguishing features” of the illness (p. 1393). Other studies have indicated that stigmatizing aspects of the name CFS lead to experiences of marginalization by the medical community [61].

3.2.5. Diagnosis—We found that patients as well as physicians and other health care professionals discussed diagnostic issues in ME/CFS. The diagnosis of ME/CFS has been identified by patients as both a relief and a burden [59] as well as the difficulty in receiving and accepting the diagnosis [33], and subsequently receiving care [13]. The route to receiving a diagnosis also has implications in illness outcomes. For example, Whitehead [35] found that people with a medically led diagnosis presented a shorter chaos narrative, for example, “life will never get better” or “no one is in control,” than those who had to search for a diagnosis (p. 2238). Similarly, Woodward, Broom, and Legge [23] found that obtaining a diagnosis was the single most helpful event in the search for social and medical legitimacy during the course of their illness.

Gilje, Soderlund, and Malterud [62] noted how the lack of illness acknowledgement from physicians, family members, and friends can be worse than the actual experience of associated symptoms. People with ME/CFS tend to feel labeled rather than receiving a legitimized diagnosis [44]. People with ME/CFS expressed concern about the ME/CFS label, whereas referring to the illness as depression is less controversial [63]. Taylor [64] noted that there is a lack of identification among people with ME/CFS within the disability community. From a physician perspective, recent studies indicate that general practitioners lack confidence in diagnosing ME/CFS because of no known cure and concern of the consequences produced by the ME/CFS label [65].

3.2.6. Power dynamics—Asbring and Narvanen [59] interviewed people with ME/CFS as well as fibromyalgia and defined the construct of “patient power,” which is gaining

knowledge about one's illness, as an important pathway in developing a sense of control over the illness. Strategies for handling the stigma associated with a ME/CFS diagnosis included withdrawal and approach strategies in interactions with caregivers [36]. Power strategies include learning about the cause of their illness and various treatments, and utilizing strategies in interactions with physicians in order to influence and control the health care process (e.g., exiting, noncompliance, confrontation, persuasion, making demands, distancing). Power is seen as a resource and the cultural and social capital of the patient is important in the interactions with health care providers. Another theme Denz-Penhey and Murdoch [53] explored was a need-to-know; people with ME/CFS seek information about and explanations of their illness. Power strategies may also be seen as coping mechanisms. Actively seeking knowledge to gain control over the illness is compatible with Ray et al.'s [48] theory of information seeking as part of the coping process. Horton-Salway [63] derived an empirical investigation of power by examining a focus group of people with ME/CFS from a local support group to explore the dynamics in physician-patient interactions through an "expertise versus experience" paradigm. Tensions in the power struggle between physicians and patients can cause a barrier that is detrimental to the giving and receiving of proper health care [66].

3.3. Physician-specific themes

Physician-specific themes include (1) skepticism of the illness and physician minimization, and (2) analysis of knowledge sources and educational initiatives on ME/CFS.

3.3.1. Skepticism and minimization—A general skepticism exists among physicians as to whether ME/CFS or fibromyalgia are considered diseases or illnesses [60]. Considered a scientific ideal, an illness can be classified as a disease when objective biological markers and a cause have been established. This illuminates that physicians cast moral judgments on the illness and their patients rather than utilizing biomedical knowledge. In addition, Schoofs et al.'s [44] study suggested that many physicians did not believe in ME/CFS. Physician minimization is common in that they believe people with ME/CFS or fibromyalgia do not behave or look the way a sick person is expected [60]. Taylor [64] found that patients experience both minimization and mistrust of their illness by others. Denz-Penhey and Murdoch [53] recognized the need for doctors to legitimize symptom control and proposed a call to not completely dichotomize psychological and physiological illnesses. Much of the qualitative literature supports the notion that ME/CFS is too psychologized and trivialized by physicians and other health care professionals.

3.3.2. Knowledge of the illness and recommendations—Chew-Graham, Cabill, Dowrick, Wearden, and Peters [67] interviewed both people with ME/CFS and physicians from primary care settings and found that physicians use social and cultural knowledge instead of biomedical knowledge in diagnosing ME/CFS. This lack of physician knowledge can lead to patient maltreatment [62]. Raine et al. [66] ran focus groups with general practitioners in the UK and asked them about their perceptions of people with ME/CFS in comparison to people with FM and their beliefs on the management of these illnesses. Physicians stereotyped people with ME/CFS based upon a lack of understanding of pathophysiology in ME/CFS, the continual reclassification of the syndrome, and the transgression of social roles [66].

Finally, many of the ME/CFS qualitative studies recommended better education of health care providers. Chew-Graham and colleagues [65] recommended better training for physicians and educational initiatives. In turn this training will help with making accurate diagnoses and managing ME/CFS symptoms as well as advocating for legitimization of the illness [64, 67].

4. Discussion and Conclusions

4.1 Discussion

This analysis demonstrated that multiple perspectives and sources of information align with our understanding of ME/CFS. The use of a meta-analytic framework, also referred to as a meta-ethnography or meta-synthesis, to analyze qualitative studies has been demonstrated as a powerful tool in systematically integrating a range of interpretive data [68]. Across patient, physician, and comparative qualitative studies on ME/CFS, the authors identified themes specific to each group as well as themes central to both patients and physicians in their experiences with ME/CFS. The major patient-specific themes include the issue of chronicity and illness phases, reductions due to the illness, and coping mechanisms. The physician-specific themes focused on skepticism and the bases of knowledge of ME/CFS among health care professionals. Themes that intersected, whether congruent or contested among patients and physicians, looked at the scope of symptom experience, the diagnosis of ME/CFS, and patient-provider power dynamics.

This review supports conclusions from the Larun and Malterud [11] review including the occurrence of changes in self-identity and dealing with multiple people in the patients' network questioning the legitimacy of the illness. As well, this review also found similar strategies for coping including seeking out more information about the condition and learning their limits in terms of balancing activity. In addition to these supported findings, we found that many of the qualitative studies emphasized the etiological and developmental processes and, in particular, the gendered experience of ME/CFS. The synthesis also expanded upon our understanding of the reductions people with ME/CFS face across physical, social, and economic domains. These findings have important implications for the understanding of health care professionals working with people with ME/CFS and potential treatment options in clinical practice.

Further, our analysis expanded upon those results by finding additional themes across patient and physician perspectives. The qualitative literature enhances our understanding of symptom severity and variability of symptom presentation in ME/CFS populations, the differential viewpoints of patients versus physicians, and the inevitable power dynamics that come into play. The lack of an operationalized definition of the illness also creates tensions with the various groups of people that ME/CFS affects. For example, the lack of a concretized definition creates constraints around the diagnosis of ME/CFS, which the qualitative literature has shown to have detrimental effects on patient populations.

Finally, the synthesis addressed findings in terms of physician-specific themes, adding a richer perspective to our knowledge base of health care professionals. In particular, the findings indicated that physicians stereotyped people with ME/CFS based on lacking understanding of ME/CFS pathophysiology. As well, other reviews and qualitative research have not taken into account the literature on experiences of ME/CFS within the family system [13, 46].

4.1.2 Limitations—The authors recognize limitations within the search strategy, given that there are some methodological constraints on comprehensively identifying all literature on ME/CFS that is considered qualitative. Therefore, there may well be other studies tapping into similar realms that the authors did not identify in their search. Similarly, there is large variation in research design among qualitative studies making it difficult to objectively synthesize results across every study. As well, the variation in ME/CFS case definition utilized in each study may contribute to differences in study outcomes and the overall synthesis. Finally, a meta-synthesis does not necessarily provide for a re-analysis of raw data

as quantitative research can do, but rather an attempt of systematic interpretation of other interpretive studies.

The authors recognize that many of these studies may be biased due to factors related to patient selection. As seen in Table 1, the majority of these studies recruited patients from tertiary care settings, ME/CFS support groups, and patient organizations. Therefore, many of the patients included in these studies are individuals with access to the medical and health care system. There are virtually no qualitative studies that assess the ME/CFS experience with a community-based sample. Future research should look at more diverse groups of people with ME/CFS by using more epidemiological samples. Given that qualitative studies do not require representative samples nor do they seek external validity, there is less concern regarding selection bias in this meta-synthesis. However, it is important to note that these studies may represent a more homogenous subset of people affected by ME/CFS.

4.2 Conclusion

Findings revealed multifaceted and interdependent themes across patients and physician accounts. Both patients and health care professionals struggle to understand and manage ME/CFS, which can fuel stigmatization and tensions. The authors suggest that future qualitative research should aim to integrate the various components within the ME/CFS experience, the members with the networks of people with ME/CFS, and the sociocultural environment in which we define and examine the illness. Finally, the authors call to challenge the dominant sociocultural narratives around researchers' and clinicians' beliefs about ME/CFS as well as account for their victim-blaming tendencies. Not upholding the accountability of all stakeholders is detrimental to the patient community as well as moving research and practice forward.

4.3 Practice implications

From a contextual, feminist perspective Richman and Jason [58] identified the dichotomy where a significant portion of the medical community still attributes psychiatric and psychosocial causes, while people with ME/CFS largely attribute their illness to a currently unknown biological etiology. Historically, illnesses primarily affecting women are chronicled as disproportionately explained through psychosocial causes, as seen in the case of multiple sclerosis [58]. In addition, it has been well-documented that people with ME/CFS experience stigmatization from healthcare professionals, family, and friends [69].

Given these factors, the call for more qualitative studies on the self-perceptions of the people with ME/CFS as a mechanism to understand experiences of marginalization is needed [16, 27]. Health care professionals can gain important insight into patient experiences through qualitative research. Synthesizing findings allows for more a more accurate description of the illness and can lead to more accurate diagnosis and appropriate recommendations for treatment.

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Flowchart 1.

Identification and Selection of Qualitative Studies on ME/CFS

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Table 1

Overview of Qualitative ME/CFS Literature

Study	Population	Recruitment	Case Definition	Methods/Analysis
Anderson & Ferrans, 1997	CFS (n=22)	Recruited from support groups and CFS newsletters and online forums (US)	Holmes	Mixed method; qualitative portion utilized semi-structured interviews
Asbring & Narvanen, 2002	CFS (n=12) and Fibromyalgia (n=13)	Tertiary care setting (Sweden)	CFS- Fukuda FM- Wolfe	Semi-structured interviews
Asbring & Narvanen, 2003	Physicians (n=26)	Selected based on information from a patient study; various types of physicians	CFS – Fukuda FM- Wolfe	Semi-structured interviews
Asbring & Narvanen, 2004	CFS (n=12) and Fibromyalgia (n=13)	Tertiary care setting (Sweden)	Fukuda	Semi-structured interviews
Bennett, Goldstein, Friedlander, Hickie, & Lloyd, 2007	CRF (n=16) and CFS (n=12)	Tertiary care setting (Australia)	Fukuda	Audio-taped focus groups (separate ones for both group)
Chew-Graham, Cabill, Dowrick, Weardan, & Peters, 2008	ME/CFS (n=38) 24 CFS, 14 physicians	Primary-based randomized controlled trial from the (UK)	Oxford	Semi-structured interviews
Chew-Graham, Dowrick, Wearden, Richardson, & Peters, 2010	General Practitioners (n=22)	GPs working in practices (UK)	Fukuda	Semi-structured interviews
Clarke, 1999	CFS (n=59)	Local support groups (Canada)	Not specified	Open-ended interviews via telephone
Clarke & James, 2003	CFS (n=59)	Local support groups (Canada)	Not specified	Open-ended interviews via telephone
Clements et al., 1997	CFS (n=66)	Primary, secondary, and tertiary care settings (UK)	Oxford	Open-ended interviews
Denz-Penhey & Murdoch, 1993	CFS (n=10)	Recruited from general practitioners (New Zealand)	Lloyd	Interviews, field notes, journal entries, questionnaires
Donalek, 2008	CFS families	Local CFS community	Fukuda	Individual and collective family semi-structured interviews
Donalek, 2009	CFS families (n=8)	Local CFS community (US)	Fukuda	Individual and collective family semi-structured interviews
Gilje, Soderlund, & Malterud, 2008	CFS (n=12) 10 women, 2 men	Local patient organization (Norway)	Fukuda	Group interviews and questionnaires and follow-up meeting one year later
Gray & Fossey, 2003	CFS (n=5)	Advertisement put into ME/CFS newsletter	Fukuda	Semi-structured interviews and Occupational Performance History Interview-II
Guisse, Widdicombe, & Mckinlay, 2007	ME/CFS (n=56)	Local ME support groups (UK)	Fukuda	Open-ended interviews and e-mail communication with support groups
Hart & Grace, 2000	CFS (n=11)	ME support group and general practitioners (New Zealand)	Lloyd	Open-ended, in-depth interviews
Horton-Salway, 2001	ME/CFS (n=1)	Unknown	Unknown	Interview with a woman with ME/CFS and her partner
Horton-Salway, 2002	General practitioners (n=10)	Recruited by referral from patient support groups (UK)	Unknown	Unstructured interviews
Horton-Salway, 2004	ME (n=20)	Local ME support group (UK)	Unknown	Focus group at the support group's monthly meeting.

Study	Population	Recruitment	Case Definition	Methods/Analysis
Larun & Malterud, 2007	Meta- ethnography of studies (n=20)	Searched for qualitative studies on CFS in major scientific databases	N/A	Meta-ethnography (see Noblit & Hare, 1998)
Lombaard & Mouton, 2005	CFS (n=4)	Physician and personal referrals as well as various media advertisements	Fukuda	In-depth interview and autobiographical sketch (questionnaire)
Lovell, 1999	CFS (n=12)	Travel health clinics overseas aid workers (UK)	Oxford	Open-ended interviews and self-report questionnaires
Raine, Cater, Sensky, & Black, 2004	General Practitioners (n=46)	Randomly selected sample of clinicians (UK)	Unknown	Focus groups
Schoofs, Bambini, Ronning, Bielak, & Woehl, 2004	CFS and Fibromyalgia (n=16)	Three local support groups (US) & one international web-based support group	Fukuda	Mixed Method; Qualitative portion conducted telephone interviews using a guided questionnaire.
Soderlund & Malterud, 2005	CFS (n=8)	Support organization (Norway)	Fukuda	Semi-structured interviews
Sodurlund, Skoge, & Malerud, 2000	CFS (n=12)	Local ME patient organization	Fukuda	Mixed method; qualitative portion utilized focus groups
Taylor, 2005	CFS (n=47)	Local CFS organizations, physician referrals, advertisements in CFS newsletter, website, and local media (US)	Fukuda	Focus groups, questionnaires, progress notes, and program evaluation questionnaire.
Travers & Lawler, 2008	CFS (n=19)	Recruited through a CFS newsletter and referrals (Australia)	Fukuda	Semi-structured interviews
Tuck & Wallace, 2000	CFS (n=22)	Support groups & recruited from a teleconference	Fukuda	Mixed method study, qualitative portion used questionnaires
Ware, 1998	CFS (n=66)	Tertiary care sample (US)	Fukuda	Longitudinal three-year study including semi-structured interviews and questionnaires
Whitehead, 2006	ME/CFS (n=17)	Local tertiary care setting, support group and referrals (UK)	Not Specified	Longitudinal study with up to three in-depth interviews
Whitehead, 2006	ME/CFS (n=17)	Local tertiary care setting, support group and referrals (UK)	Not specified	Longitudinal study with up to three in-depth interviews
Woodward, Broom, & Legge, 1995	CFS (n=50) and Physicians (n=20)	Recruited from local college of general practitioners (Australia)	Holmes	Interview with general practitioners and three interviews over a period of two years with participants with CFS

* CFS, ME, or ME/CFS terminology used in the table is the way that the authors described the illness their studies.

Table 2

Content Analysis of Major Themes from Qualitative ME/CFS Studies

Patient-Specific Themes	Intersecting Themes	Physician-Specific Themes
I. Identity, illness phases, and chronicity II. Illness development and etiology III. Physical, social, and economic reductions IV. Coping mechanisms V. Balancing activities	I. Description of symptoms II. Operationalizing CFS III. Illness variability IV. Stigmatization V. Diagnostic issues VI. Power dynamics	I. Skepticism and minimization II. Bases of knowledge III. Education and recommendations