Factors Affecting the Characterization of Post-Exertional Malaise Derived from Patient Input

Journal of Health Disparities Research and Practice

Volume 13 | Issue 2 Article 5

2020

Factors Affecting the Characterization of Post-Exertional Malaise Derived from Patient Input

Carly S. Holtzman , *DePaul University*, csholtzman@gmail.com

Claire Fisher , *DePaul University*, clairefisher524@gmail.com

Shaun Bhatia , *DePaul University*, sbhatia3@depaul.edu

Leonard A. Jason , *DePaul University*, ljason@depaul.edu

Abstract:
The National Institutes of Health/Center for Disease Control and Prevention (NIH/CDC) Common Data Elements (CDE) established a post-exertional malaise (PEM) workgroup with the task of describing PEM and recommending a standardized way of assessing it in patients with myalgic encephalomyelitis and chronic fatigue syndrome (ME/CFS). As a stigmatized group, patients with ME/CFS are in need of instruments which can properly describe their symptomatic experiences, which can help reduce the disparity between illness seriousness and appropriate attention from healthcare. The current study explored attitudes and preferences among 115 patients with ME/CFS who participated in the creation of a patient-driven instrument to measure PEM, the key symptom of the illness. Themes that emerged from the qualitative analyses of patient feedback focused on how their illness was experienced; their access to care; problems with physicians, researchers, and research methods; and expressions of gratitude for the collaborative process. Domains that were most important to the patient community were identified in the effort to create a comprehensive measure of PEM. Benefits of community-based action research are discussed.

Keywords:
post-exertional malaise; myalgic encephalomyelitis; chronic fatigue syndrome; participatory

research; patient advocacy

Factors Affecting the Characterization of Post-Exertional Malaise Derived from Patient Input

Cover Page Footnote

The authors would like to thank the patient community for their willingness to collaborate with us throughout this process.

Journal of Health Disparities Research and Practice
Volume 13, Issue 2, Summer 2020, pp. 51-64

School of Public Health
University of Nevada, Las Vegas

Factors Affecting the Characterization of Post-Exertional Malaise Derived from Patient Input

Carly S. Holtzman, DePaul University
Claire Fisher, DePaul University
Shaun Bhatia, DePaul University
Leonard A. Jason, DePaul University
**Corresponding Author:* Shaun Bhatia, sbhatia3@depaul.edu

ABSTRACT

The National Institutes of Health/Center for Disease Control and Prevention (NIH/CDC) Common Data Elements (CDE) established a post-exertional malaise (PEM) workgroup with the task of describing PEM and recommending a standardized way of assessing it in patients with myalgic encephalomyelitis and chronic fatigue syndrome (ME/CFS). As a stigmatized group, patients with ME/CFS are in need of instruments which can properly describe their symptomatic experiences, which can help reduce the disparity between illness seriousness and appropriate attention from healthcare. The current study explored attitudes and preferences among 115 patients with ME/CFS who participated in the creation of a patient-driven instrument to measure PEM, the key symptom of the illness. Themes that emerged from the qualitative analyses of patient feedback focused on how their illness was experienced; their access to care; problems with physicians, researchers, and research methods; and expressions of gratitude for the collaborative process. Domains that were most important to the patient community were identified in the effort to create a comprehensive measure of PEM. Benefits of community-based action research are discussed.

Keywords: post-exertional malaise; myalgic encephalomyelitis; chronic fatigue syndrome; participatory research; patient advocacy

INTRODUCTION

The National Institutes of Health/Center for Disease Control and Prevention (NIH/CDC) Common Data Elements (CDE) established a post-exertional malaise (PEM) workgroup with the task of describing PEM and recommending a standardized way of assessing it in patients with myalgic encephalomyelitis and chronic fatigue syndrome (ME/CFS; National Institute of Neurological Disorders and Stroke [NINDS] CDE Group, 2018). The recommendations from the NIH/CDC CDE PEM working group described PEM by noting the following characteristics: exacerbation of symptoms, increased disability, varying onset, extended and varying recovery

periods, and the relationship with exertion (NINDS CDE Group, 2018). However, it was beyond the scope of the working group initiative to provide a way of operationalizing these domains. Thus, these general descriptions lacked the precision necessary for researchers and clinicians to reliably assess the presence of PEM.

The NIH/CDC PEM working group also recommended the use of 5 operationalized items from the DePaul Symptom Questionnaire (DSQ; Jason et al., 2010a) which could be used as part of a patient screening process. For many patients seeking a more comprehensive diagnostic tool for assessing PEM, this screening tool was not deemed as sufficient. In part as a protest, an online poll was organized by the patient community, which indicated that patients preferred the general NIH/CDC PEM descriptions (Simon, 2018). But such general statements regarding PEM still needed to be operationalized if researchers were to have an instrument to comprehensively assess this symptom.

Considerable alienation has occurred between the ME/CFS patient community and the NIH due to being repeatedly excluded from major decisions regarding the name of the illness as well as how to measure and treat it. However, there have been instances in which involvement of the patient community in the characterization of their illness has resulted in change. For instance, criticism from patients regarding the first ME/CFS Primer resulted in revisions by the International Association of Chronic Fatigue Syndrome/Myalgic Encephalomyelitis (Friedberg et al, 2014). However, despite the inclusion of patient representatives on CDE PEM workgroups, many patients were still left feeling excluded and marginalized with the recent PEM recommendations.

In an effort to adopt a more participatory action process, the authors and the patient community collaborated on developing a PEM measure that had the direct approval and input from the patient community (details of the survey's development were reported by Jason, Holtzman, Sunnquist, and Cotler [2018a] and the instrument can be retrieved at https://journals.sagepub.com/doi/suppl/10.1177/1359105318805819). This project is one example of action research, as the nature of the efforts between researchers and the patient community fall in line with the core features of action research: the focus on partnership and participation, as well as the contribution to action research theory/practice (Bradbury-Huang, 2010). We believed that by having patients work collaboratively in the creation of the survey, we would be able to establish a more valid instrument, as well as a trusting and reciprocal relationship with the patient community (Balcazar, Keys, Kaplan, & Suarez-Balcazar, 1998). This type of participatory, action-oriented research has infrequently occurred within the ME/CFS arena (Jason, 2012).

Qualitative research is a way of gathering information that is particularly appropriate with patient groups that have been stigmatized and left out of the decision making process. Qualitative studies within the ME/CFS literature have explored issues related to the context of illness, onset of illness, and illness management (Anderson, Jason, Hlavaty, Porter, & Cudia, 2012; Anderson, Jason, & Hlavarty, 2014). Additional qualitative studies have provided a rich, descriptive perspective of the ME/CFS experience (Bennett, Goldstein, Friedlander, Hickie, & Lloyd, 2007; Stormorken, Jason, & Kirkevold, 2017). However, no qualitative study with patients having ME/CFS has focused specifically on PEM or the process of creating a patient-driven survey.

The current study involved participatory action by including the patient community, using Facebook as a medium to conduct these collaborative, interactive discussions. The study used qualitative methods to assess the patient perspective, and how it informed the development of a new, patient-driven PEM questionnaire. By engaging in this process, it was the hope of the authors to highlight the need for instruments which can properly describe ME/CFS symptomatic

experiences, which can help reduce the disparity between illness seriousness and appropriate attention from healthcare.

METHODS

Design

The majority of communication between researchers and the patient community took place on Facebook.com, a popular social media platform. The use of Facebook in research has been growing in recent years, though most studies utilizing the social media site focus on descriptive analysis of users, motivation for use, its impact on social interactions, issues related to privacy, and its effectiveness at recruiting research participants (Wilson, Gosling, & Graham, 2012; Thornton et al., 2016). The goal of the current study was to explore aspects of PEM that were discussed via Facebook during the development of a PEM-specific questionnaire for patients with ME/CFS. We used a qualitative approach to analyze data from multiple Facebook posts and the resulting conversations. Specifically, we used an inductive thematic analysis grounded in the essentialist perspective (Braun & Clarke, 2006).

The authors did not require Institutional Review Board approval for this study, as all the data originated from a public profile page on Facebook.com. The researchers indicated to participants that the products of this participatory action work would be documented and published for the larger scientific and public policy oriented community to view.

Participants

Eligible participants were individuals with a self-reported diagnosis of ME and/or CFS who commented on the nine Facebook posts by the last author (LAJ). In total, 115 patients responded to these posts, and their responses were included in analysis. While it is unusual to utilize a sample size this large in qualitative research (e.g. Anderson, Jason, & Hlavarty, 2014; N = 19; Devendorf, Jackson, Sunnquist, & Jason, 2017; N = 10), we decided to include all participants, as the majority only made one comment over the course of the nine distinct Facebook discussions, and we wanted to ensure proper saturation of the data. Due to the public nature of Facebook data, we were able to determine the sex of the participants who had that information publicly available (see Table 1).

Table 1: *Participant information (N = 115)*

Sex	% (N)
Female	52.17 (60)
Male	9.57 (11)
Not determined	38.26 (44)
Number of Comments Made	% (N)
1	45.2 (52)
2	24.4 (28)
3	8.70 (10)
4	3.48 (4)
5	7.83 (9)
6	0.87 (1)
7	0.87 (1)
8	0.87 (1)
9	2.61 (3)
10	0.87 (1)
12	1.74 (2)
17	0.87 (1)
23	0.87 (1)
92	0.87 (1)

Note. Sex information was gained from participants who had public Facebook profiles and information was available.

Procedures

LAJ initially spoke with several leading patient activists regarding the patient poll as well as the overall NIH/CDC CDE PEM recommendations. A decision was collectively made to try to develop a comprehensive PEM questionnaire which would begin with posting an operationalized version of the NIH/CDC CDE description of PEM (which was operationalized by LAJ, with input from several patients). Twenty-eight patients responded to the first posting of this PEM questionnaire, and when the suggestions were incorporated into the revised PEM questionnaire, a second posting occurred, garnering 62 comments. This process continued for a total of nine postings for the construction of a PEM questionnaire, which was a dynamic process involving a patient driven effort of continually revising and updating the new instrument. After nine postings, the majority of comments were favorable, and little new information about changes to be made in the PEM questionnaire were being obtained. Therefore, a decision was made to end the data collection phase of the study, and patients were in agreement with this decision. The Facebook posts along with the patient comments were downloaded into NVivo 12 software for qualitative coding.

Data Analysis

Qualitative analyses in the ME/CFS field have primarily followed the grounded theory perspective (Anderson, Jason, Hlavaty, Porter, & Cudia, 2012). However, due to the nature of data collection taking place via a public online forum, it was not possible to re-contact participants to continue data collection, which is a key component of grounded theory (Corbin & Stauss, 1990). Instead, we adhered to a thematic analysis under an essentialist perspective, as our goal was to

provide a rich description of the experiences and reality of participants. Our approach was a data-driven inductive process, similar to grounded theory analysis. Analysis was conducted in six stages, as proposed by Braun and Clarke (2006). Thematic coding was conducted by two of the authors.

In stage 1, coders read all of the Facebook comments to familiarize themselves with the content of the discussions that took place. In stage 2, CSH searched for patterns by re-reading all Facebook comments, and developed codes with definitions and guidelines for coding. CSH and CF then coded the entire dataset and took thorough notes for future discussions. Initial interrater reliability was established for the first three Facebook postings and was found to have moderate agreement (K= 0.78; McHugh, 2012). In stage 3, points previously made in memos were discussed until a consensus was reached. We then began organizing emerging themes in stage 4. Visual representations such as mind maps and charts were used throughout the process to determine relationships between themes. In stage 5, we continued to review, further define, and name themes (see Figure 1). Final interrater reliability is reported in Table 2. In stage 6, we selected excerpts from the data to represent results.

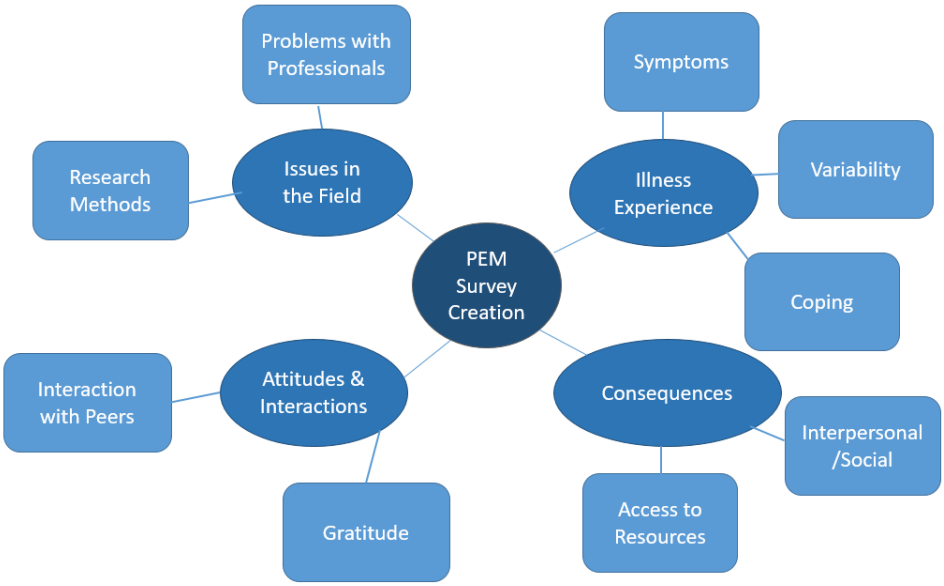

Figure 1. Conceptual map of themes and subthemes

Table 2: *Interrater reliability*

Themes	Range of Kappa	Mean Kappa	Weighted Mean Kappa
Illness Experience	0.76-1	0.89	0.84
Symptoms	0.50-1	0.78	0.65
Variability	0.50-1	0.85	0.79
Coping	0.50-1	0.81	0.66
Consequences	0.50-1	0.86	0.67
Interpersonal/Social	0.50-1	0.74	0.41
Access to Resources	0.74-1	0.94	0.54
Issues in the Field	0.70-1	0.90	0.81
Problems with Professionals	0.90-1	0.97	0.91
Research Methods	0.64-1	0.86	0.71
Attitudes & Interactions	0.49-1	0.86	0.84
Interactions with Peers	0.50-1	0.81	0.82
Gratitude	0.82-1	0.94	0.88

RESULTS

The international online sample included 115 individuals who commented on at least one of the nine Facebook posts by LAJ. As described in Table 1, the majority of participants were female (52.17%). Most individuals made 1 or 2 comments, (45.2% and 24.4%, respectfully).

Interrater reliability metrics between the two coders were established using Kappa scores. Because data was distributed across nine Facebook postings, NVivo output provides nine Kappa scores for each theme and subtheme. In order to provide the most accurate reliability scores, we report the range of Kappa scores across the nine Facebook posts, the mean Kappa score, and the weighted mean Kappa score which takes into account the size of each Facebook post (at the character unit). Interrater reliability for all themes and subthemes fell in the moderate to excellent range ("Coding comparison query," n.d.).

Four themes emerged from the Facebook discussions regarding PEM: (1) *illness experience*; (2) *consequences*; (3) *issues in the field*; (4) *attitudes and interactions*. We explain the components of each theme and illustrate how patients' Facebook comments were taken into consideration during the participatory process of creating a PEM questionnaire.

Theme 1: Illness Experience

The first major challenge of this study was to determine how to describe the patient experience of PEM. Historically, patients have been outspoken about past descriptors of PEM being overly simplified and not capturing the many nuances of the symptomatology (Jason, McManimen, Sunnquist, & Holtzman, 2018b). For example, one participant said:

> *It is not getting tired. It is so much more. The whole body just feels like it stops functioning properly. Everything aches, I can't sleep, I am cold and can't control my body temperature, I can't think, I have problems making decisions, even easy decisions like if I want to eat or drink something. And then, for days, I am feeling weak and worse than normal. With horrible headaches.*

Topics of conversations also discussed the varying triggers that could prompt PEM. It is a popular opinion among patients that possible triggers extend further than physical or cognitive exertion to include an array of exposures such as:

> *Chemicals we are sensitive to; foods we're intolerant of; emotional event's (good or bad); light; noise; vibration; sensory overload; drugs; supplements. It just seems that the hardest things to manage and to gain help with are being left out.*

Patients were also concerned about capturing the amount of variability that exists in the experience of PEM. Based on the current study, it seems that there is variability both between and within patients. For example, one participant explained how the onset of PEM varies after being exposed to a trigger, and how the fluctuations cause confusion:

> *For me, some crashes are delayed, some aren't, and how long they are delayed varies, as does how long any crash lasts… [And] since I generally do not recover back to my previous level of functioning, it's sometimes hard to tell if I am still crashing, or if that's just my new normal.*

Another patient commented:

> *Someone who has lived with the illness for many years will have a different answer [than] someone recently diagnosed.*

The quote above lends support that variation exists both within and between patients, highlighting how someone who has been ill for a long time will have a different experience than someone who has not been sick for as long. The excerpt also suggests that, as the illness progresses, the experience of PEM may change or fluctuate.

Coping with symptom complications was also a discussion narrative. The most common management technique is the pacing strategy (Goudsmit, Nijs, Jason, & Wallman, 2012; Jason et al., 2013), which encourages patients to reserve energy to ensure they don't go beyond their energy limit. However, the majority of patients rate pacing as only mildly to moderately effective at reducing the level of severity of symptoms (Holtzman, Bhatia, Cotler, & Jason, 2019). The following quote from a participant highlights the need for further research of possible treatment for patients with ME/CFS.

> *Even though I pace, it doesn't always stop symptom exacerbation; and many patients need to pace a long time (months) to reach a degree of stability whereby they start to experience pacing benefits.*

In order to address these issues in the development of the questionnaire, we included a list of 32 symptoms related to PEM for patients to endorse. We also included a list of 14 possible triggers beyond physical or cognitive exertion. To accommodate the variability in the course of PEM, we included Likert-type scales to assess the frequency of symptom exacerbation, items assessing varying onsets of PEM, as well as possible correlates that affect the onset of PEM, such as types of triggers that elicit a PEM response and which symptoms are typically exacerbated. There were also items that assess pacing and its effectiveness in reducing symptoms.

Theme 2: Consequences

The second theme that emerged from the data related to the consequences of PEM. The following selected quotes illustrate how the experience of PEM affects daily life for those with ME/CFS in interpersonal, social, and financial domains. When discussing the impact of severe periods of PEM, one participant commented on the social isolation that many patients experience:

I find walking less debilitating than having a phone conversation, which is one of the most exhausting activities. This is very unfortunate because I find the isolation absolutely brutal and spend much of my time just trying to manage my anguish.

Another area that is affected by PEM is a patient's access to care, such as being able to see a physician/specialist regularly, or accessing disability benefits. Many patients are not able to work due to the severity of their symptoms, so most rely on disability payment. However, qualifying for disability with an invisible illness is difficult and often involves physical tests that cause patients to become sicker, as described below:

It is really horrible that proving you are disabled makes many of us much sicker, to the point where if I have to go through a major review again, I am literally too sick to prove I am sick.

In order to gauge the level of disability in participants, we asked about past exercise tests the participant completed, and about the types of activities a participant is able to accomplish. While we tried to include as many patient recommendations as possible, the decision was made among our research team not to include questions about social support for two reasons: (1) we did not want to add to the length of the questionnaire, and (2) items of that nature are typically associated with mental illnesses such as depression, and we did not want to perpetuate the common misconception that ME/CFS is a psychological disorder.

Theme 3: Issues in the Field

An additional theme that emerged spoke to the participants' challenges with medical professionals and government officials. Sub-themes included the challenges participants expressed about receiving a correct diagnosis and the terminology used in describing the illness. One participant expressed the difficulties of advocating for oneself to physicians and having to relay information on case definitions:

I was first diagnosed with CFS by my Infectious Diseases Specialist then [diagnosed with] ME by [a] ME doctor. Three years later, I gave the same Infectious Diseases Specialist a copy of the CCC and ICC [case definitions] and after reading them he changed [the diagnosis] to ME. He used to be our provincial head epidemiologist, so I would not make that change without careful consideration. Like most MDs, he had never heard of the CCC or ICC.

Participants also expressed the lack of precision in identifying the unique presence of PEM to diagnose ME/CFS, as described in the quote below:

Yes, I agree, which is why it is so important the PEM section really does select for PEM and not just tired after activity or fatigue like is seen in cancer and other diseases since almost all diseases have chronic fatigue. I think we all agree this is not just a form of fatigue that is resolved by rest.

Standardizing terminology to describe an illness is an important step in creating strong diagnostic guidelines. One participant explained the lack of specific language to describe issues related to PEM, as well as confirm the presence of PEM:

Language is a massive problem. Unless you have experienced PEM, you have no need of language for it. So we are constantly trying to describe our experience using words that cannot directly describe our actual experience. PEM is not just more or worse of something other people experience. It is a unique experience without unique language to describe it. That makes it hard for us. And makes it very hard for people trying to identify who has PEM and who does not.

One of the most contested debates was regarding the term "PEM," because the word "malaise" trivializes the severity of patient's symptoms and level of disability. Instead, we used the term "abnormal response to physical and/or cognitive exertion," as this term elicited a more positive response from the patient community. It also emphasizes that the symptom the questionnaire is trying to assess is a reaction to exertion, regardless of what "exertion" means to the individual participant.

Theme 4: Attitudes and Interactions

Participants often interacted with each other during the Facebook discussion. They replied to one another in a largely respectful and proactive manner, whether they were agreeing or disagreeing with each other's statements. One participant expressed validation for the illness experience that s/he found in another's comment, which further facilitated the discussion:

> *Love your commentary… your points hit the nail on the head for my more severe periods. This poll currently reflects my "relatively moderate months" as I call them. It's not a bad survey, at all – just missing some of the picture needed to full paint PEM.*

Finally, many participants expressed opinions during the Facebook discussion that showed gratitude for the research process, and the work being done by the DePaul research group. Expressions of gratitude were exemplified by the comment below:

> *The ME/CFS patient community has long been recognized for its almost unprecedented degree of proactive involvement, but it is only in partnership with skilled scientists and investigators that the greatest benefit can be gained. My thanks.*

DISCUSSION

The objective of this study was to use community-based action research using qualitative analyses to explore the themes surrounding the development of a patient-driven, comprehensive PEM questionnaire. The recommendations of a federal working group elicited considerable patient anger, and the university-patient collaboration described in this article emerged from this controversy. Brydon-Miller, Greenwood, and Maguire (2003) claim action research is "embedded within a system of values and promotes some model of human interaction," (p. 11). The current project adhered to these tenets in order to give a voice to the patient community. In some instances, direct input of patients has been ignored in major policy decisions that have affected this community (Holtzman et al., 2019; Jason, Evans, So, Scott, & Brown, 2015b; Jason, et al., 2018a). It was our hope that by working democratically with the patient community, we might reduce some of the alienation that has occurred among patients with ME/CFS, and thus shrink the disparity between illness seriousness and appropriate attention from healthcare. The process of working collaboratively with the patient community allowed for the expansion of knowledge and the development of a comprehensive PEM instrument (Jason et al., 2018a) , and this article documents qualitative thematic analysis and four key themes that emerged during that process.

The first theme, *illness experience*, consisted of three subthemes: *symptoms*, *variability*, and *coping*. The *symptoms* subtheme, which characterized comments regarding specific aspects of symptomatology, has been reflected in past literature of PEM. For example, many patients suggested items assessing the frequency of experiencing symptoms such as fatigue, cognitive deficiencies, and flu-like symptoms, which have been assessed in past measures of ME/CFS (Jason et al., 2010a). Patients also offered suggestions for new items to add to the current survey that have not been previously assessed, such as "decreased heart rate," "premenstrual symptoms," "paralysis," and "severe burning sensation all over skin." Another main concern of the patient

community was how to capture the level of *variability* in symptom presentation. Recent research has attempted to tap into this domain; for example, Chu (2018) found variation between and within individuals in terms of onset and duration of PEM symptoms when observed over time after an exercise challenge. In an attempt to further characterize variability, based on patient commentary, we added additional items asking about the frequency of delayed and immediate onset, as well as questions assessing associations between PEM and the type, intensity, frequency, and duration of the exertion or exposure to a trigger. In regards to *coping*, participants mentioned pacing as the key strategy in managing PEM symptoms, and discussed how their ability to cope may influence their answers to some items. The pacing approach encourages patients to stay as active as possible within their limits, which requires the patient to determine the level at which they can function without exacerbating their symptoms (Goudsmit et al., 2012; Jason et al., 2013). Participants discussed different methods of pacing, including trigger-specific pacing based on their symptomatic reactions and pacing by monitoring changes in heart rate. Unfortunately, only 7.6% rate pacing as "very effective" at reducing the severity of PEM symptoms (Holtzman et al., 2019), which highlights the need for research into more effective coping strategies to lessen the burden caused by PEM.

Two subthemes emerged from the second observed theme, *consequences*: *access to resources* and *interpersonal/social consequences*. Both of these topics have been described in previous ME/CFS literature. Sunnquist, Nicholson, Jason, and Friedman (2017) found that over half of patients have never seen a specialist for their illness, noting geographic scarcity and financial resources as barriers to accessing this type of care. Additionally, over half of patients with ME/CFS in the United States report dissatisfaction with the medical care they do receive, and note that it can take two years or longer to receive a diagnosis (Tidmore et al., 2015). The personal experiences shared by the patient community corroborated these findings, noting the difficulties in accessing appropriate care. Participants also discussed how social interactions often cause PEM, but how the lack of social interaction negatively affects their mood. This is troubling, given that perceived social support has been found to decrease fatigue and increase vitality in patients with ME/CFS (Jason, Roesner, Portern, Parenti, Mortensen, & Till, 2010c).

The third theme, *issues in the field*, consisted of two subthemes: *problems with professionals* and *research methods*. As mentioned above, patients with ME/CFS often have negative experiences with health care professionals, especially with those who may endorse a psychogenic view of the illness. Jason, Taylor, Plioplys, Stepanek, and Shlaes (2002) found that 37% of medical students believed that patients with CFS' primary illness was major depressive disorder. In fact, patients who experience unsupportive interactions have an increased risk for both depression and suicidal ideation, suggesting that the stigmatization associated with ME/CFS causes feelings of depression, but clinical depression is not the cause of their symptoms (McManimen, McClellan, Stoothoff, & Jason, 2018). The lack of visibility experienced from medical professionals may be compounded by the lack of accessible information about ME/CFS in the general medical literature, where less than half of medical textbooks have a mention of ME/CFS, and only 21% discuss diagnostic criteria (Jason, Paavola, Porter, & Morello, 2010b). Additionally, the patient community was concerned about the research methods commonly used in the ME/CFS field. For instance, some patients were concerned about the specificity of terminology that is used to describe symptom experience (for example, "feeling tired"), as certain phrases describe patients with other illnesses, such as fibromyalgia, cancer, or depression. Also, certain terms are associated with stigma (for example, "minimal exercise making you feel

physically tired"), which has been noted in the past literature (Jason et al., 2018b). Patients also expressed concern about whether this survey was meant for patients with ME, CFS, or both. This patient reaction is rooted in a fundamental controversy concerning the illness: whether ME and CFS is the same disease, or distinct diseases with varying etiologies (Jason, Evans, Brown, Sunnquist, & Newton, 2015a). Further research is necessary to identify possible biomarkers to settle the debate as to if they are distinct illnesses or the same.

The fourth theme, *attitudes and interactions*, relates to the second research question, of how participants utilized Facebook as a means for collaboration between patients and researchers. There were two main types of interaction that took place: participants expressing *gratitude* for being involved in this innovative process, and *interactions between peers* relating to each other's experiences. Research on gratitude has been a growing field in recent years. Expressing gratitude, especially for those suffering from chronic illness, can be a protective factor against depression, and it is associated with more positive coping and enhanced quality of life (Sirois & Wood, 2017). Interacting with other patients could also provide beneficial effects, as there has been evidence of perceived social support protecting against poor long-term outcomes. For example, Jason et al. (2010c) found that patients with CFS who received social support had decreased fatigue and increased vitality over time. Future research using social media should continue exploring the protective role social support plays among those with ME/CFS.

The data used in this study were instrumental in creating a comprehensive assessment of PEM (Holtzman et al., 2019; Jason, Holtzman, Sunnquist, & Cotler, 2018). Over 1,500 patients filled out the new measure within a short period, indicating enthusiasm among the patient community for the instrument that they shaped and designed. Our qualitative exploration of the major themes has expanded our knowledge of the nuances of PEM.

Several limitations should be noted in the current study. First, there was an observed, uneven distribution of comments, where one participant accounted for 22% of the overall number of comments. During the analyses of comment data, this reality was salient for the authors, and as such, comments described in this study were specifically selected so that equal thematic representation of all participants was ensured.

Second, the opinions of participants in our sample may not be generalizable to the entire patient community due to the nature of the convenience sampling that was used. Though aggregated demographic information of Facebook users is available (Statistica, 2020), the distribution of Facebook users with ME/CFS is not known. As such, future studies may examine the distribution and usage patterns of patients with ME/CFS who are active on Facebook and other social media platforms to help inform sampling methodology. Further, future studies utilizing these mediums should be wary of increasing privacy restrictions, and may want to use a more active mode of recruiting participants (Wilson et al., 2012).

The third limitation concerns the lack of patient demographics. Because participants were not surveyed directly, the authors were not able to obtain demographic information other than what was publicly available on their Facebook profiles. There was also no way to confirm a diagnosis of ME/CFS in our patient sample. Future studies qualitatively analyzing patient perspectives on PEM should also include a survey to assess diagnostic criteria and demographic characteristics.

CONCLUSION

Overall, analyzing Facebook comments related to the experience of PEM has led to new insights, which in turn allowed us to create a comprehensive, patient-driven questionnaire

assessing PEM. Our goal was to identify the most important aspects of PEM, as well as how the patient community interacted with each other via Facebook. This action-oriented process has led us to believe that it is crucial to collaborate with the patient community which can result in a more insightful, accurate and valid perspective of the illness. This qualitative analysis using community-based action research has made substantial contributions to the study of PEM, and ME/CFS in general. It is our hope to provide a model of how scientists and patients in this area can work together in the development of methods and instruments to better assess this illness.

REFERENCES

Anderson, V. R., Jason, L. A., & Hlavarty, L. E. (2014). A qualitative natural history study of ME/CFS in the community. *Health Care for Women International, 35*(1), 3-26. doi:10.1080/07399332.2012.684816

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

Balcazar, F. E., Keys, C. B., Kaplan, D. L., Suarez-Balcazar, Y. (1998). Participatory action research and people with disabilities: Principles and challenges. *Canadian Journal of Rehabilitation*, 12, 105-112.

Bennett, B., Goldstein, D., Friedlander, M., Hickie, I., & Lloyd, A. (2007). The experience of cancer-related fatigue and chronic fatigue syndrome: a qualitative and comparative study. *Journal of Pain and Symptom Management, 34*(2), 126-135. doi:10.1016/j.jpainsymman.2006.10.014

Bradbury-Huang, H. (2010). What is good action research? Why the resurgent interest? *Action Research, 8(1)*: 93-109. doi: 10.1177/1476750310362435

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77-101.

Brydon-Miller, M., Greenwood, D., & Maguire, P. (2003). Why action research? *Action Research, 1(1),* 9–28. doi: 10.1177/14767503030011002

Chu, L., Valencia, I. J., Garvert, D. W., & Montoya, J. G. (2018). Deconstructing post-exertional malaise in myalgic encephalomyelitis/chronic fatigue syndrome: A patient centered, cross-sectional survey. *PLoS ONE, 13*(6), e0197811. doi: 10.1371/journal.pone.0197811

Corbin, J., & Stauss, A. (1990). Grounded theory research: Procedures, Canons and Evaluative Criteria. *Zeitschrift fur Soziologie, 19*(6), 418-427.

Devendorf, A. R., Jackson, C. T., Sunnquist, M., & Jason, L. A. (2017). Approaching recovery from myalgic encephalomyelitis and chronic fatigue syndrome: Challenges to consider in research and practice. *Journal of Health Psychology*, 1-13. doi:10.1177/1359105317742195

Friedberg, F., Bateman, L., Bested, A. C., Friedman, K. J., Gurwitt, A., Jason, L. A., ... & Vallings, R. (2014). ME/CFS: A primer for clinical practitioners. *Chicago, USA: International Association for Chronic Fatigue Syndrome/Myalgic Encephalomyelitis.*

Goudsmit, E. M., Nijs, J., Jason, L. A., & Wallman, K. E. (2012). Pacing as a strategy to improve energy management in myalgic encephalomyelitis/chronic fatigue syndrome: a consensus document. *Disability and Rehabilitation, 34*(13), 1140-1147. doi:10.3109/09638288.2011.635746

Holtzman, C. S., Bhatia, S., Cotler, J., & Jason, L. A. (2019). Assessment of Post-Exertional Malaise (PEM) in Patients with Myalgic Encephalomyelitis (ME) and Chronic Fatigue Syndrome (CFS): A Patient-Driven Survey. *Diagnostics (Basel), 9*(1). doi:10.3390/diagnostics9010026

Jason, L. A. (2012). Small wins matter in advocacy movements: Giving voice to patients. *American Journal of Community Psychology, 49*, 307–316. doi: 10.1007/s10464-011-9457-7

Jason, L. A., Brown, M., Brown, A., Evans, M., Flores, S., Grant-Holler, E., & Sunnquist, M. (2013). Energy Conservation/Envelope Theory interventions to help patients with chronic fatigue syndrome. *Fatigue: Biomedicine, Health, & Behavior,* 1, 27-42. doi: 10.1080/21641846.2012.733602

Jason, L. A., Evans, M., Brown, A., Sunnquist, M., & Newton, J. L. (2015a). Chronic fatigue syndrome versus sudden onset myalgic encephalomyelitis. *Journal of Prevention and Intervention in the Community, 43(1),* 62-77. doi: 10.1080/10852352.2014.973233

Jason, L. A., Evans, M., So, S., Scott, J., & Brown, A. (2015b). Problems in defining post-exertional malaise. *Journal of Prevention and Intervention in the Community, 43*(1), 20-31. doi:10.1080/10852352.2014.973239

Jason, L. A., Evans, M., Porter, N., Brown, M., Brown, A., Hunnell, J., Anderson, V., Lerch, A., De Meirleir, K., & Friedberg, F. (2010a). Development of a revised Canadian myalgic encephalomyelitis chronic fatigue syndrome case definition. *American Journal of Biochemistry and Biotechnology, 6*(2), 120-135. doi: 10.3844/ajbbsp.2010.120.135

Jason, L. A., Holtzman, C. S., Sunnquist, M., & Cotler, J. (2018). The development of an instrument to assess post-exertional malaise in patients with myalgic encephalomyelitis and chronic fatigue syndrome. *Journal of Health Psychology*, 1359105318805819. doi:10.1177/1359105318805819

Jason, L. A., McManimen, S. L., Sunnquist, M., & Holtzman, C. S. (2018b). Patient perceptions of post exertional malaise. *Fatigue: Biomedicine, Health & Behavior, 6*(2), 92-105. doi:10.1080/21641846.2018.1453265

Jason, L. A., Paavola, E., Porter, N., & Morello, M. L. (2010b). Frequency and content analysis of chronic fatigue syndrome in medical text books. *Australian Journal of Primary Health, 16*(2), 174-178. doi: 10.1071/PY09023

Jason, L. A., Roesner, N., Porter, N., Parenti, B., Mortensen, J., & Till, L. (2010c). Provision of social support to individuals with chronic fatigue syndrome. *Journal of Clinical Psychology, 66*(3), 249-258. doi:10.1002/jclp.20648

Jason, L. A., Taylor, R. R., Plioplys, S., Stepanek, Z., & Shlaes, J. (2002). Evaluating attributions for an illness based upon the name: chronic fatigue syndrome, myalgic encephalopathy and Florence Nightingale disease. *American Journal of Community Psychology, 30*(1), 133-148. doi:10.1023/A:1014328319297

National Institute of Neurological Disorders and Stroke Common Data Elements Group. Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) Post-Exertional Malaise Subgroup: Statement of Purview. (2018). Retrieved from https://www.commondataelements.ninds.nih.gov/sites/nindscde/files/Doc/MECFS/PEM_Subgroup_Summary.pdf

NVivo 12. Coding comparison query. (n.d.). Retrieved from https://help-nv.qsrinternational.com/12/win/v12.1.71-d3ea61/Content/queries/coding-comparison-query.htm

McHugh, M. L. (2012). Interrater reliability: the kappa statistic. *Biochemical Medicine (Zagreb), 22*(3), 276-282.

McManimen, S. L., McClellan, D., Stoothoff, J., & Jason, L. A. (2018). Effects of unsupportive social interactions, stigma, and symptoms on patients with myalgic encephalomyelitis and chronic fatigue syndrome. *Journal of Community Psychology, 46*(8), 959-971. doi:10.1002/jcop.21984

Simon, M. Results of the Poll to Inform the NIH/CDC's Definition of PEM in All Their Future ME/CFS Research [msg#1]. Retrieved from https://www.s4me.info/threads/results-of-the-poll-toinform-the-nih-cdc%E2%80%99s-definition-of-pem-in-all-their-future-me-cfs-research.2221

Sirois, F. M., & Wood, A. M. (2017). Gratitude uniquely predicts lower depression in chronic illness populations: A longitudinal study of inflammatory bowel disease and arthritis. *Health Psychology, 36*(2), 122-132. doi:10.1037/hea0000436

Statistica. (2020). Distribution of Facebook users ub tge United States as of May 2020, by gender. Retrieved from https://www.statista.com/statistics/266879/facebook-users-in-the-us-by-gender/#:~:text=Facebook%20user%20share%20in%20the%20United%20States%202020%2C%20by%20gender&text=Facebook%20is%20the%20most%20popular,percent%20of%20users%20were%20male

Stormorken, E., Jason, L. A., & Kirkevold, M. (2017). From good health to illness with post-infectious fatigue syndrome: a qualitative study of adults' experiences of the illness trajectory. *BMC Fam Pract, 18*(1), 49. doi:10.1186/s12875-017-0614-4

Sunnquist, M., Nicholson, L., Jason, L. A., & Friedman, K. J. (2017). Access to medical care for individuals with Myalgic Encephalomyelitis and Chronic Fatigue Syndrome: A call for Centers of Excellence. *Modern Clinical Medicine Research, 1*(1), 28-35. doi:10.22606/mcmr.2017.11005

Thornton, L., Batterham, P. J., Fassnacht, D. B., Kay-Lambkin, F., Calear, A. L., & Hunt, S. (2016). Recruiting for health, medical or psychosocial research using Facebook: Systematic review. *Internet Interventions, 4*, 72-81. doi:10.1016/j.invent.2016.02.001

Tidmore, T. M., Jason, L. A., Chapo-Kroger, L., So, S., Brown, A., & Silverman, M. C. (2015). Lack of knowledgeable healthcare access for patients with Neuro-endocrine-immune Diseases. *Frontiers in Clinical Medicine, 2*, 46–54.

Wilson, R. E., Gosling, S. D., & Graham, L. T. (2012). A Review of Facebook Research in the Social Sciences. *Perspectives on Psychological Science, 7*(3), 203-220. doi:10.1177/1745691612442904