The use of mixed methods in studying a chronic illness

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This article explores mixed-method approaches with an illness called myalgic encephalomyelitis (ME) and chronic fatigue syndrome (CFS). Qualitative and Quantitative data were used to investigate the epidemiology of this illness, as well as explore attributions based on the name of the illness, and finally treatment approaches. In each of the domains within the ME and CFS research reviewed, our analyses were richer and our findings ultimately more impactful when we integrated qualitative and quantitative research methods. The use of a multiphase mixed-methods research program provided our team unique vantage points for better understanding social and community issues involving this controversial chronic illness. Furthermore, this approach allowed us to implement the insights gained through an advocacy lens to change policy, recommend and evaluate treatments, and amplify voices within the patient population. In this way, we believe that the practice of methodological pluralism is especially applicable and effective to the study of chronic illness, and believe that other investigators will benefit from the use of these approaches with similar disenfranchised and unfairly treated populations.

Keywords: chronic fatigue syndrome; myalgic encephalomyelitis; advocacy; methodology; quantitative methods; qualitative research

1. Introduction

This article explores mixed-method approaches with an illness called myalgic encephalomyelitis (ME) and chronic fatigue syndrome (CFS). In this article, we use the terms CFS and ME, but we recognize that while the former name suggests the broader Fukuda et al. (1994) criteria definitions that might inadvertently include patients without core symptoms of this illness, the Canadian criteria (Carruthers et al., 2003), called ME/CFS and the ME-ICC (International Consensus Criteria) (Carruthers et al., 2011), delineate those patients with the key symptom such as “post-exertional malaise” which targets a more impaired and homogenous group of patients (Jason, Brown, Evans, Sunnquist, & Newton, 2013). Both labels are used in this article, as different research has used different criteria in patient selection. These illnesses are as debilitating as type II diabetes mellitus, congestive heart failure, multiple sclerosis, or end-stage renal disease (Anderson & Ferrans, 1997; Buchwald, Pearlman, Umali, Schmaling, & Katon, 1996). Yet, reaching a widely accepted operationalization of this condition has been difficult to achieve (Brown, Jason, Evans, & Flores, 2013). Research has delineated a number of obstacles that have contributed to the slowing of such progress and resistance from some professionals in the health-care community (Jason,
This article seeks to illustrate the need for applying flexible, robust mixed-method strategies when investigating the nature of the illness and the experiences of the patient community. Considerable patient and scientific activity in the ME and CFS field has focused on three areas: establishing estimates of the prevalence for the illness, addressing negative stigma toward patients due to the name of the illness, and the creation and evaluation of treatment strategies (Jason, 2012). This article will examine where research in these domains has faced limitations based on study design, and will reflect on the effects of methodological choices on the understanding of ME and CFS. Furthermore, we propose that moving to a responsive, community-based research paradigm that uses both quantitative and qualitative methods would open opportunities for more acute depictions of the factors contributing to epidemiology, perceptions of the name of this illness, and treatment issues.

In this article, we will focus on mixed methods, which involve the implementation of both quantitative and qualitative methods in one study design (Tebes, 2012). In brief, quantitative research is based in positivism, or the belief in a single reality accessible through scientific procedure. In contrast, qualitative studies are grounded in a constructivist paradigm (Ponterotto, Mathew, & Raughley, 2013), and rather than a single, universally shared reality, each participant has his or her own reality (Ponterotto et al., 2013). A number of scholars have increasingly defended critical multiplicity, or the belief that combining methods compensates for the errors inherent in either methodology (Tebes, 2005). Furthermore, it is suggested that methodological pluralism (the use of varied methodologies in research) is especially suitable for research in the pursuit of social justice for marginalized populations, because it is more sensitive to context, and is more likely to uncover power differentials and to give voice to participants (Ponterotto et al., 2013).

Furthermore, the level at which methodological pluralism is implemented can vary (Barker & Pistrang, 2012). For example, a pluralistic research discipline is one in which a balance of study designs and analysis techniques is used in publications (Barker & Pistrang, 2012). But mixed-methods research can occur within the confines of a single study, known as a pluralistic or mixed-methods study. Such a study incorporates both quantitative and qualitative data collection and analysis. At a different level, a pluralistic research program reflects pluralism across a program of studies, which is the focus of this article. In the following, we will illustrate some of the advantages of using a mixed-method approach emerging from a research group at DePaul University, and it first focused on estimating the prevalence of ME and CFS. We will suggest that quantitative procedures have the most power to appeal to collaborators in funding and policy, while qualitative studies are more likely to empower community members and gain insights into how to identify and work with participants, and that combining these methods can be most effective when undertaking community-based issues (Cokley & Awad, 2013; Lyons et al., 2013; Lyons, Bike, Johnson, & Bethea, 2012).

2. Epidemiology

While fatigue is common in the general population, with an incidence between 19% and 28% (Kroenke, Wood, Mangelsdorff, Meier, & Powell, 1988), severe fatigue of six months or longer is less common, and an important question involved finding the percentage of these sufferers likely to be diagnosed with ME and CFS. The first widely publicized study of ME and CFS epidemiology was initiated by the Centers for Disease Control and Prevention (CDC) in the late 1980s (Gunn, Connell, & Randall, 1993). Investigators requested physicians in four cities to identify patients who might have this illness. Prevalence rates were found to range from 4.0 to 8.7 individuals per 100,000 cases (Reyes et al., 1997), suggesting that less than 20,000 people had this illness in the USA. The majority of identified cases were white, upper-
middle-class females, which contributed to the notion that the illness was a “Yuppie Flu-like” disease. This quantitative study underrepresented low-income and disadvantaged minorities who often do not have access to the health-care system, and thus were less likely to be counted in prevalence rates derived from physicians identifying possible cases at treatment sites (Richman, Flaherty, & Resplenda, 1994). In addition, this study probably underestimated the prevalence because the case ascertainment method depended upon health-care providers identifying possible patients from their catchment areas, but many physicians would not nominate patients as possibly having this illness if they were skeptical of its existence (Anderson, Jason, Hlavaty, Porter, & Cudia, 2012).

The early findings from the above prevalence study can be seen as an example of the pitfalls of positivism and the associated tendency to ignore the effect of one’s expectations and experiences on the design of one’s research. The CDC researchers who conducted these initial prevalence studies collected data which had serious potential biases, and it is possible that more mixed-method approaches might have offered a more accurate method (Creswell & Plano Clark, 2010; Greene, Caracelli, & Graham, 1989; Jason & Glenwick, 2012; Ponterotto et al., 2013).

We used qualitative approaches to gather information, and began informal conversations with critical gatekeepers about these findings from this CDC study. After talking with a number of patients and patient organizations, the consensus was that the CDC prevalence estimates were biased (Richman et al., 1994), and that there was a need to determine the prevalence with methods that did not rely on physician case ascertainment methods. Our group also found other converging data. We learned that the CDC was receiving thousands of phone calls each month from patients with severe fatigue (McCluskey, 1993, p. 288). In addition, the fact that there were thousands of patients who were members of the CFIDS (chronic fatigue and immune dysfunction syndrome) Association, the largest patient advocacy organization hinted at higher prevalence numbers. These informal data from a variety of sources suggested that the CDC estimates of less than 20,000 in the USA with ME/CFS were probably an underestimate.

Richman and Jason next formed a research team in Chicago, which included professionals from diverse areas, including epidemiology, psychology, psychiatry, medicine, immunology, sociology, and biostatistics, as well as patient representatives. The group approached the issue of how to contact patients without having a referral from a treatment setting or physician. To accomplish this, they first solicited input from patients. The team was provided the phone numbers of a group of patients with this illness, and then tried the use of phone calls to contact and interview them. Callers were able to successfully reach about 90% of a sample of people with ME and CFS by using telephones, and respondents mentioned that they thought telephone calling was an excellent way of reaching them (Jason, Fitzgibbon, Taylor, Johnson, & Salina, 1993). Our team now had a strategy to successfully reach people from the community with this illness, without needing the biased physician case ascertainment method. We also solicited feedback from members of the local Chicago Chronic Fatigue Syndrome Association. The patients suggested the inclusion of Spanish-speaking interviewers so that we could evaluate the many non-English-speaking individuals who lived in our area. Finally, the patients suggested that to increase rates of participation, we needed to provide transportation to those we brought in for medical and psychiatric examinations, and to provide babysitting money for those that would need someone else to look after their children. All of these recommendations and suggestions were incorporated into our plans to conduct a community-based prevalence study.

These strategies and considerations regarding ways to access this population involved conversations with professionals, patients, and patient groups, and was the first step in our mixed-methods strategy to find ways to increase the representativeness of our participants. This is known as participant enrichment, a rationale for mixed-methods implementation (Collins, Onwuegbuzie, & Sutton, 2006). Our experience above matches the use of this principle in
community psychology research as noted by Jason and Glenwick (2012): “… formative mixed methods studies can be instrumental in learning how to access and develop trusting relationships with different sectors of a community” (p. 53).

Along with helping to establish our next phase of study, these qualitative discussions and data helped to set our agenda for the development and submission of a quantitative community-based epidemiologic grant. But we soon learned that National Institutes of Health (NIH) reviewers felt that since the CDC had found only four people at most per 100,000 adult Americans had the illness, we would need to call a random sample of hundreds of thousands of people in order to find enough cases to conduct a community-based epidemiologic study, and federal officials informed us that such a large study would be too expensive to be funded. We needed to collect pilot data to deal with these NIH reviewer concerns. From January to May 1993, our research group telephoned a random sample of about 1000 people, and those with signs of this illness were interviewed by a psychiatrist and examined by a medical doctor. The CFIDS Association helped to fund this pilot study, and our pilot study’s findings suggested much higher prevalence rates than the prior estimates (Jason et al., 1995). In addition, we also assessed the prevalence of this illness among a national sample of nurses (Jason, Taylor et al., 1993), and the data from this study suggested even higher prevalence rates. This study also encouraged a nurse with the illness to form the organization called Medical Professionals/Persons with CFS (Dahlen, personal communication, March 31, 1998).

With these pilot data, and after several grant submissions, in June 1995, our large-scale community-based epidemiologic study was funded by the National Institute of Allergies and Infectious Diseases. In this study, we attempted to contact over the telephone a random sample of 28,673 households in Chicago. Of that sample, 18,675 individuals were screened using the telephone for signs of this illness. Based on the initial screening, participants with significant fatigue and other somatic symptoms were selected to receive a complete medical and psychiatric examination. Approximately 4% of the sample was determined to have the illness, and rates were higher among Latinos and African-Americans when compared to those who were white (Jason, Richman, et al., 1999). In addition, those in the higher socioeconomic income group did not have higher rates, and this finding challenged the “Yuppie Flu” past characterization of those having this illness. These findings suggested that there might be as many as 800,000 adults in the USA with this syndrome. Our recommendations for a community-based study, using a random sample and phone screenings, were ultimately accepted and integrated by the CDC into their research program (Reyes et al., 2003), and the study’s findings helped to set the agenda for more attention to this illness among policy and government officials.

Our community-based study identified patients who were not biased by access to the healthcare system, and thus provided us a richer and more diverse representation of people with ME/CFS. Both the experience with successfully seeking funding from the federal government, and the use of community partners, including the largest self-help organization, reflect the benefits of using mixed methods.

Having access to this community-based sample led to the next step in the “sequential-nested” sampling procedure. We examined a cohort 10 years following the completion of our epidemiologic, community-based study (Leech & Onwuegbuzie, 2010, p. 63). In this study, we collected data on those whose ME and CFS persisted over a 10-year period, those who developed this illness over this period of time, and those whose diagnosis remitted over the past decade (Jason, Porter, Hunnell, Rademaker, & Richman, 2011). Our quantitative data suggested that prevalence rates had remained somewhat comparable over this decade, thus helping to answer an important question: were illness rates increasing, decreasing, or staying the same? In addition to the quantitative methods involved in re-contacting the participants and giving them a complete medical and psychological evaluation, qualitative methods (Anderson, Jason, & Hlavaty, 2014).
were used to provide us with a deeper understanding of the interaction of the multiple systems involved in this chronic illness, as well as to hear the voice of the patients. A coding system was developed through a grounded theory framework in order to focus on the context-dependent and structural processes within the data (Glaser & Strauss, 1967; Strauss & Corbin, 1990). Based on the final coding system, two independent coders checked the reliability of the coding scheme with randomly selected portions of the transcripts and came to a 95% agreement.

The community response to the illness included themes centering around how members from the medical community as well as other related networks, such as support groups, understand and respond to the illness. As an example, many patients mentioned that they experienced negative attitudes such as physician minimization of their illness. Furthermore, many respondents told of their obstacles to securing disability in a climate where the illness encounters disbelief. Identifying the community response to the illness was an essential component of understanding the systems involved with the experiences of its sufferers. Qualitative methods allowed us to better understand the experiences of the patients who had been identified in the epidemiologic study, and thus provided us an opportunity to look at the connection of illness to the distribution of power and privilege within the medical community, as well as the social response to chronic illness and health care. Thus, we too found as Ponterotto et al. (2013) suggest, that mixed-method approaches can provide, “… multiple windows into the lives of the less empowered and historically silenced within our society” (p. 47).

3. The name and unintended stigma

The effects of stigmatization are often life-altering for persons with ME and CFS, as was evident in our qualitative study mentioned above. Most symptoms are not visibly apparent, which makes it difficult for others to believe in or understand the vast array of debilitating symptoms of patients. Shlaes, Jason, and Ferrari (1999) developed the CFS Attitude Test, as one way of assessing stigma, discrimination and attitudes toward individuals with this illness. They found a relationship between beliefs about the degree to which people with this illness are responsible for their illness, beliefs about the relevance and validity of the illness, and beliefs about the personality traits of patients. For example, if someone believes that people with ME and CFS are responsible for their illness, it is likely that they will also believe that people with this illness have negative personality characteristics, such as being compulsive or overly driven.

The names given to illnesses can cause stigma. For example, gay-related infectious disease was a stigmatizing name that fortunately was changed to AIDS. Many patient activists claimed that the term CFS contributed to health-care providers having negative attitudes toward those with this syndrome (Shlaes et al., 1999). The patient community has felt that the term CFS trivializes the seriousness of this illness, as fatigue is commonly experienced by many people in our society. The illness had been previously called ME before Holmes et al. (1988) at the CDC renamed it CFS in 1988. Quantitative and qualitative methods were also used to explore how the name selected to characterize an illness can contribute to stigma.

Many medical personnel and research scientists feel that if the name were to be changed, it would be best to have a scientific basis for the change. Unfortunately, in the 1990s, data had not been collected to help guide the process of revising the name. By the late 1990s, there were no studies providing concrete evidence that the name CFS might negatively influence attributions that others made toward patients. In 1998, a patient who had been trained as a social psychologist contacted Jason and suggested to him an experimental way to document how the name of this illness might influence attributions. This patient asked whether the DePaul research team might conduct such a study using quantitative methods. With this input, our research team embarked on a study to evaluate whether different names for this illness indeed influenced...
attributions regarding its cause, nature, severity, contagion, and prognosis. In this study, one case description of a patient with prototypic symptoms of this illness was given to a group of medical trainees. Participants were randomly assigned to different groups, with the only difference between groups being the type of diagnostic label given as the diagnosis (e.g. CFS, myalgic encephalopathy, etc.). Results of this study indicated that participants’ (Jason, Taylor, Plipolys, Stepanek, & Shlaes, 2002) attributions about the illness changed as a function of which diagnostic labels were used to characterize the person with this illness. For example, the more medically sounding term myalgic encephalopathy was associated with the poorest prognosis, and this term was more likely to influence participants to attribute a physiological cause to the illness.

The results of this study illustrate the value of being open and responsive to information from community members. Thanks to the one patient mentioned above, we were able to affirm negative attributions related simply to the name of the illness, giving direction to our further studies. These findings were widely disseminated, and in part due to this study, the first author was asked to be part of a name change working group, within a Health and Human Services federally sponsored Chronic Fatigue Syndrome Coordinating Committee. The federally appointed name change working group solicited opinions from patients and scientists as well as through various Internet websites and listservs (Jason, Eisele, & Taylor, 2001). Our qualitative data indicated that 92% of patients wanted the name changed, and they indicated a variety of preferred names including myalgic encephalopathy (ME), neuro-immune endocrine disorder, polyalgic asthenia (a name that attempts to describe key symptoms), Ramsay’s disorder, an eponym for one of the first physicians who had studied this illness, and Florence Nightingale disease (who many believe had this illness). Other open-ended surveys over time suggested other names as well including neuroendocrine immune disorder and ME.

Efforts at community building were made by seeking consensus among the patients and professionals, and a variety of other qualitative methods were used, including many informal polls and discussions among patients on listservs during this time.

The recommendations made by the Name Change Working Group ultimately were not approved by federal officials. But the momentum of this working group helped to spur further change, and ultimately different names were adopted by several patient organizations (e.g. the Patient Alliance for Neuroendocrineimmune Disorders Organization for Research and Advocacy, and the Myalgic Encephalomyelitis Society of America) and research/clinical settings (Whittemore Peterson Institute for Neuro-Immune Disease). Another strategy has been to adopt the term ME/CFS, which was thought to be a transition term, and one that might gain the support of both patients and scientists. A new ME/CFS clinical case definition was later developed in Canada that used the term ME/CFS (Carruthers et al., 2003). In addition, the organization of researchers called the International Association of CFS changed their name to the International Association of CFS/ME. The International Association of ME/CFS also organized an international task force and published guidelines for a new case definition for children and adolescents, with the name Pediatric ME/CFS (Jason et al., 2006). Ultimately, the National Institutes of Health adopted the term ME/CFS to replace the name CFS (see http://orwh.od.nih.gov/research/me-cfs/contact.asp). Efforts continue with this CFS name change issue.

4. Treatment

The third area we will review concerns treatment issues. Twemlow, Bradshaw, Coyne, and Lerma (1997) found that individuals with this illness reported that they were made worse by their healthcare workers 66% more often than general medical patients. Mixed research could be used to help identify reasons for the anger among patients with current ME and CFS treatment protocols and could be used to encourage the development of alternative treatment approaches.
While cognitive behavior therapy (CBT) has been applied to many medical problems, from epilepsy to cancer, its application to ME and CFS has been more controversial. CBT with graded exercise constitutes a dominant form of treatment found in the literature for ME and CFS. This approach to treatment challenges patients’ attributions of their symptoms as resulting from physical disease, such as viral or immunological problems (Sharpe et al., 1996), and it encourages patients to attribute their symptoms to social and psychological factors. Patient groups are critical of CBT because they feel that the attribution of their symptoms to social and/or psychological factors demeans and/or ignores the biological nature of their illness, and they claim that increased exercise has led to a worsening of their health (The ME Association, 2010). In addition, when patients with cancer or heart disease are offered CBT, it represents only one treatment approach within a larger medical effort involving a number of pharmacological interventions. However, for patients with ME and CFS, this CBT approach has often been the only treatment offered.

Patient surveys confirm that many patients with this illness are dissatisfied with CBT- or graded exercise-based interventions. For example, one patient survey found that among patients who had received graded exercise therapy, 33.1% felt “much worse” and 23.4% judged themselves to be “slightly worse” (The ME Association, 2010). In addition, Price, Mitchell, Tidy, and Hunot (2008) reviewed 15 studies of CBT with a total of 1043 CFS participants. At treatment’s end, 40% of people in the CBT group showed clinical improvement in contrast to only 26% in usual care, but changes were not maintained at a one- to seven-month follow-up when patients who had dropped out were included.

As we reviewed patient reactions to CBT, we decided to solicit patient input into considering other potential interventions that might help them better cope with their illness. In 1996, our research team distributed to patients a brief survey of open-ended items designed to assess their utilization, of and preference for, a variety of services (Jason, Ferrari, Taylor, Slavich, & Stenzel, 1996). One preferred service involved a volunteer caregiver system to provide assistance with daily chores and errands. These data were instrumental in our group developing a service program involving volunteer buddies to provide assistance to patients. In collaboration with the Chicago Chronic Fatigue Syndrome Association, Shlaes and Jason (1996) developed a comparison study within which people with ME and CFS either received a volunteer buddy and a mentor who had this illness, or no treatment. The buddy was an individual in the community who agreed to spend one hour per week conducting home visits with a patient. Buddy–participant matches were made based upon need and interest assessments completed by the participants and buddies. Mentors were individuals with the illness who were willing and able to engage in two hours of phone contact each month with the participants. Participants who received the buddy/mentor intervention experienced significant decreases in fatigue severity, while the control group experienced significant increases in fatigue severity (Shlaes & Jason, 1996).

Over time, with the help of patient feedback, our team began developing what we now call the Envelope theory (Jason, Melrose, et al., 1999). The actual term was provided to us by a patient. This approach does not challenge patients’ beliefs in a medical cause for the illness. Instead, envelope theory recommends that patients pace their activities according to their available energy resources. In this approach, the phrase, “staying within the envelope”, is used to designate a comfortable range of energy expenditure in which an individual avoids both overexertion and under-exertion, maintaining an optimal level of activity over time. If a comfortable level of activity is maintained over time, the health status of individuals with may slowly improve, and patients may find themselves able to engage in increasing levels of activity. Findings indicated that when the patients’ perceived limits and expended energy levels were maintained close to the boundaries (within the envelope), the patients experienced decreases in fatigue over time (Jason et al., 2013).
We have now incorporated our Energy Envelope approach into our buddy program with patients (Jason, Roesner et al., 2010). In our last study, 30 patients with this illness were randomly assigned to either a four-month buddy intervention or a control condition. Those who received the buddy intervention had significantly greater reductions in fatigue severity and increases in vitality. Once again, we found that helping patients to both monitor and stay within their energy boundaries led to important improvements.

Our group thought that qualitative research on the self-perceptions of the people with this illness could provide us a way to better understand the experiences of marginalization. In addition, qualitative studies might provide us with context-specific experiences of people with this illness and those involved in their lives (e.g. family or the medical community). To accomplish this, Anderson et al. (2012) reviewed 34 qualitative studies of patients with this illness. 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.

These qualitative studies had themes that enhanced our understanding of symptom severity and variability of symptoms, differential viewpoints of patients versus physicians, as well as the power dynamics that emerged. For example, we found that patients experience stigmatization from healthcare professionals, family, and friends. A key theme was that physicians lacked an awareness of, as well as an understanding of, the pathophysiology of the illness, and this contributed to the stereotyping of their patients as having a psychogenic illness. There were repeated examples of victim-blaming tendencies within the medical community, which attributes psychiatric and psychosocial causes to this illness. For both our volunteers who were implementing our buddy program and for other health-care professionals, the themes that emerged from these qualitative studies allowed us to have a more accurate understanding of the patients’ experience with this illness and ultimately, a better appreciation of the types of non-stigmatizing treatments that are needed for patients. Combined with the quantitative work, our experience is similar to conclusions offered by Leech and Onwuegbuzie (2010), who assert that using mixed methods in the evaluation of treatments and interventions can help gain access to more insightful and nuanced assessment when compared to mono-method designs, while providing important tools to facilitate navigation through a challenging issue within a community setting.

5. Discussion

The present article illustrates how a multiphase mixed-methods research program provided our team unique vantage points for better understanding social and community issues involving controversial chronic illnesses. Our research teams involved over time diverse investigators, and this helped us to flexibly use a variety of context sensitive methodological approaches. In each of the three domains within research reviewed in this article, our analyses were richer and our findings ultimately more impactful when we intertwined both qualitative and quantitative research methods.

Regarding the first domain we reviewed, determining the prevalence of ME and CFS has been a challenging task for the scientific field. We concluded that drawing cases from physician-based or community-based samples did have important influences on prevalence rates. In our work, qualitative methods were used to help us develop better quantitative methods to study epidemiology, using community-based samples. The sample generated was subsequently used to probe qualitative features of the illness experiences of patients. Quantitative research provided us data on the magnitude of this illness, whereas the qualitative methods allowed us to better understand the unique challenges and stigma that the patients had experienced from their families, friends, and treatment professionals. These data were ultimately used to provide better...
appreciations of the magnitude of the effects of this illness on patients, as we used these data to estimate the economic costs of this illness to our nation (Jason, Benton, Johnson, & Valentine, 2008).

The next set of studies involved the issue of the name given to this illness, and our review suggested that the term CFS did influence the medical legitimacy of the illness. It is of importance to note that technically, the medical legitimacy is not influenced by the name (as medical legitimacy is invariable and inviolate). It is the human perception of medical legitimacy that is influenced by the name. Despite being dismissed by many physicians, many, if not most, patients know how sick they really are, and know that there is an organic cause to their illness; it is not all in their head. The medical legitimacy exists; it is not always recognized.

The integration of community members alongside researchers guided this research. By following the recommendation of a patient and conducting a quantitative study with medical trainees, we found that considerable stigma occurred with the term CFS. Using quantitative methods to document negative CFS attributions alongside qualitative approaches to generate alternative names has proven useful in the 20-year, ongoing effort to rename this illness.

In our third domain, which we reviewed in this article, we suggested that CBT interventions that challenge basic patient illness beliefs do not build an empathic and trusting patient–therapist relationship. We found considerably more support for interventions that used non-stigmatizing approaches, such as the envelope theory. Envelope theory provides an explanation for the limited ability of patients to do work and/or participate in activities based upon an abnormally limited amount of energy available to these patients. That abnormality is physiologically or metabolically based, that is, is organic in origin, and, therefore, much more acceptable (palatable) to patients who do not believe that their illness is “all in their heads”.

We also learned from key qualitative feedback that there is a need for the general public and the medical community to become better educated about the problems and difficulties associated with this illness. Quantitative approaches were used to validate the effectiveness of the buddy/mentor program. Some patients may need assistance from relatives, caregivers, buddies, or other well persons in order to complete daily living tasks, and others might just want someone to help them deal with their isolation. Our buddy programs might be helpful in fulfilling those needs. Qualitative studies helped us to better appreciate that comprehensive, non-stigmatizing treatment programs need to be developed that are uniquely tailored to the medical needs of patients.

Although the “buddy” system appears to be helpful, it is probably too weak an intervention by itself to permanently succeed in helping this disenfranchised group of patients. However, in the future, these types of interventions might be paired perhaps with other more biological interventions (e.g. a short-term trial of immune/neuroendocrine or other modulator). However, what is critical is changing medical professionals/family/community attitudes toward this illness. In the field of medical education, there have been some preliminary attempts to request that medical schools add ME and CFS to their curriculums (Jason, Porter, et al., 2010).

Over the past three decades, a series of key decisions were made concerning methods for gathering prevalence data, the name, and treatment approaches for ME and CFS. Many of these decisions were formulated within a societal and political context in which this illness was assumed to be a psychologically generated problem. In general, physicians have also regarded fatigue as one of the least important presenting symptoms (Lewis & Wessely, 1992). In such controversial community contexts, the employment of a purposeful multiphase mixed-methods program coupled with the values of community psychology can be a tool especially suited to provide patients, health-care workers, scientists, and government officials less stigmatizing ways of understanding this illness. This approach more accurately represents the illness to family, friends and health-care professionals, and, thereby, has the possibility of improving the
services to those affected. Such integrated and value-based approaches are useful for the purpose of advocacy research that works toward the understanding and empowerment of other, similar groups who may experience negative social perceptions and dismissive treatment.

Science as a discipline attempts to discover the truth, which should be immutable and withstand the tests of time. Is the failure of Science to do so, in some instances, a failure of Science as a discipline or its procedures? Or, are the failures of Science the failures, the human failures, of its practitioner? Science does not prevail as a discipline and an activity unaltered and unswayed by the human practitioners who guide it and do the work. The ability of Science to yield altered truths or distortions of it can result in scientific imprecision and worse. A key question is: how then to correct Science and what are appropriate methods of corrections? These are the questions addressed in this article. It would seem that the process of correcting Science when it strays is a science or involves several other sciences. It is a side of Science that needs the telling in rigorous, dispassionate tone and that is what we tried to accomplish in this article for an illness that affects millions worldwide. It is important to get the Science of this illness right.

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References
Anderson, J. S., & Ferrans, C. E. (1997). The quality of life of persons with chronic fatigue syndrome. *The Journal of Nervous Mental Disorders, 185*, 359–367.
Anderson, V. R., Jason, L. A., & Hlavaty, L. E. (2014). A qualitative natural history study of ME/CFS in the community. *Health Care for Women International, 35*, 3–26.
Anderson, V. R., Jason, L. A., Hlavaty, L. E., Porter, N., & Cudia, J. (2012). A review and meta-synthesis of qualitative studies on myalgic encephalomyelitis/chronic fatigue syndrome. *Patient Education and Counseling, 86*(2), 147–155.
Barker, C., & Pistrang, N. (2012). Methodological pluralism: Implications for consumers and producers of research. In L. A. Jason & D. S. Glenwick (Eds.), *Methodological approaches to community-based research* (pp. 33–50). Washington, DC: American Psychological Association.
Brown, A. A., Jason, L. A., Evans, M. A., & Flores, S. (2013). Contrasting case definitions: The ME international consensus criteria vs. the Fukuda et al. CFS criteria. *North American Journal of Psychology, 15*(2), 103–120.
Buchwald, D., Pearlman, T., Umali, J., Schmaling, K., & Katon, W. (1996). Functional status in patients with chronic fatigue syndrome, other fatiguing illnesses, and healthy individuals. *The American Journal of Medicine, 101*, 364–370.
Carruthers, B. M., Jain, A. K., De Meirleir, K. L., Peterson, D. L., Klimas, N. G., Lerner, A. M., … Van de Sande, M. I. (2003). Myalgic encephalomyelitis/chronic fatigue syndrome: Clinical working case definition, diagnostic and treatments protocols. *Journal of Chronic Fatigue Syndrome, 11*, 7–115.
Carruthers, B. M., van de Sande, M. I., De Meirleir, K. L., Klimas, N. G., Broderick, G., Mitchell, T., … Stevens, S. (2011). Myalgic encephalomyelitis: International consensus criteria. *Journal of Internal Medicine. doi:10.1111/j.1365-2796.2011.02428.x*
Cokley, K. O., & Awad, G. H. (2013). In defense of quantitative methods: Using the “master’s tools” to promote social justice. *Journal for Social Action in Counseling and Psychology, 5*(2), 26–41.
Collins, K. M., Onwuegbuzie, A. J., & Sutton, I. L. (2006). A model incorporating the rationale and purpose for conducting mixed methods research in special education and beyond. *Learning Disabilities: A Contemporary Journal, 4*(1), 67–100.
Leech, N. L., & Onwuegbuzie, A. J. (2010). Guidelines for conducting and reporting mixed research in the field of counseling and beyond. *Journal of Counseling & Development, 88*, 61–69.

Lewis, G., & Wessely, S. (1992). The epidemiology of fatigue: More questions than answers. *Journal of Epidemiology and Community Health, 46*, 92–97.

Lyons, H. Z., Bike, D. H., Johnson, A., & Bethea, A. (2012). Culturally competent qualitative research with people of African descent. *Journal of Black Psychology, 38*, 153–171.

Lyons, H. Z., Bike, D. H., Ojeda, L., Rosales Meza, R., Johnson, A., & Flores, L. Y. (2013). Qualitative research as social justice practice with culturally diverse populations. *Journal for Social Action in Counseling and Psychology, 2*, 10–25.

McCluskey, D. R. (1993). *Pharmacological approaches to the therapy of chronic fatigue syndrome*. New York, NY: John Wiley.

The ME Association. (2010). *Managing my M.E.: What people with ME/CFS and their carers want from the UK’s health and social services*. Gawcott: Author. Retrieved from http://www.meassociation.org.uk/wp-content/uploads/2010/09/2010-survey-report-lores10.pdf

Ponterotto, J. G., Mathew, J. T., & Raughley, B. (2013). The value of mixed methods designs to social justice research in counseling and psychology. *Journal for Social Action in Counseling and Psychology, 5*(2), 42–68.

Price, J. R., Mitchell, E., Tidy, E., & Hunot, V. (2008). Cognitive behaviour therapy for chronic fatigue syndrome in adults. *The Cochrane Database of Systematic Reviews, 3*, 1–55. doi:10.1002/14651858.CD001027.pub2

Reyes, M., Gary, H. E., Jr., Dobbins, J. G., Randall, B., Steele, L., Fukuda, K., … Reeves, W. C. (1997). Descriptive epidemiology of chronic fatigue syndrome: CDC surveillance in four cities. *Morbidity and Mortality Weekly Report Surveillance Summaries, 46*(SS2), 1–13.

Reyes, M., Nisenbaum, R., Hoaglin, D. C., Unger, E. R., Emmons, C., Randall, B., … Reeves, W. C. (2003). Prevalence and incidence of chronic fatigue syndrome in Wichita, Kansas. *Archives of Internal Medicine, 163*, 1530–1536.

Richman, J. A., Flaherty, J. A., & Rospendra, K. M. (1994). Chronic fatigue syndrome: Have flawed assumptions derived from treatment-based studies? *American Journal of Public Health, 84*, 282–284.

Sharpe, M., Hawton, K., Simkin, S., Surawy, C., Hackmann, A., Klimes, I., … Seagroatt, V. (1996). Cognitive behaviour therapy for the chronic fatigue syndrome: A randomized controlled trial. *British Medical Journal, 312*, 22–26.

Shlaes, J. L., & Jason, L. A. (1996). A buddy/mentor program for people with chronic fatigue syndrome. *The CFIDS Chronicle, 9*, 21–25.

Shlaes, J. L., Jason, L. A., & Ferrari, J. (1999). The development of the chronic fatigue syndrome attitudes test: A psychometric analysis. *Evaluation and the Health Professions, 22*, 442–465.

Strauss, A., & Corbin, J. (1990). *Basics of qualitative research: Grounded theory procedures and techniques*. Newbury Park, CA: Sage.

Tebees, J. K. (2005). Community science, philosophy of science, and the practice of research. *American Journal of Community Psychology, 35*(3–4), 213–230.

Tebees, J. K. (2012). Philosophical foundations of mixed methods research: Implications for research and practice. In L. A. Jason & D. S. Glenwick (Eds.), *Methodological approaches to community-based research* (pp. 33–50). Washington, DC: American Psychological Association.

Twemlow, S. W., Bradshaw, S. L., Jr., Coyne, L., & Lerma, B. H. (1997). Patterns of utilization of medical care and perceptions of the relationship between doctor and patient with chronic illness including chronic fatigue syndrome. *Psychological Reports, 80*, 643–658. doi:10.2466/pr0.1997.80.2.643