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IMPACT OF SOCIAL SUPPORT ON THE RELATIONSHIP BETWEEN ILLNESS INVALIDATION AND SHAME AMONG INDIVIDUALS WITH MYALGIC ENCEPHALOMYELITIS/CHRONIC FATIGUE SYNDROME

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IMPACT OF SOCIAL SUPPORT ON THE RELATIONSHIP
BETWEEN ILLNESS INVALIDATION AND SHAME AMONG INDIVIDUALS
WITH MYALGIC ENCEPHALOMYELITIS/CHRONIC FATIGUE SYNDROME

A DISSERTATION APPROVED FOR THE
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BY

_____
Dr. Denise Beesley, Chair

_____
Dr. Melissa Frey

_____
Dr. Barbara Greene

_____
Dr. E. Laurette Taylor

_____
Dr. Nicole Campbell

This dissertation is dedicated to the light of my life, my son, Jonathan Caleb Kendrick. From the time I carried you under my heart, until today, when you stand taller than me, you have breathed inspiration into me to do more, to be better, to love deeper, and to reach for all that life has to offer.

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Abstract

The current study examined the relationship between illness invalidation and shame in a sample of 168 adults with myalgic encephalomyelitis/chronic fatigue syndrome. Based on review of existing qualitative research, it was hypothesized that there would be a significant positive relationship between perceived illness invalidation and experiences of state shame. Additionally, in light of the Stress Buffering Model, it was hypothesized that perceived social support would moderate the relationship between illness invalidation and shame. Results indicated that illness invalidation in the form of lack of understanding significantly correlated with experiences of shame. Hierarchical multiple regression revealed that perceived social support did not, however, significantly moderate the relationship between state shame and illness invalidation in the form of lack of understanding.

Chapter One

Overview

Myalgic encephalomyelitis (ME), also known as chronic fatigue syndrome (CFS), affects an estimated 2.2 million Americans (Bierl et al., 2004). The etiology of ME/CFS continues to be poorly understood and is likely complex (Bierl et al., 2004). Existing research suggests the involvement of inflammatory processes (Fulle et al., 2000; Maes, Mihaylova, Kubera, & Bosmans, 2007; Pall & Satterlee, 2001; Richards, Roberts, McGregor, Dunstan, & Butt, 2000), alterations in gene expression (Light et al., 2011), immune dysfunction (Maes, Twisk, Kubera, & Ringel, 2012; Masuda et al., 2002), involvement of the hypothalamic-pituitary-adrenal axis (HPA; Cleare, 2004; Johnson & DeLuca, 2005; Van Den Eede, Moorkens, Van Houdenhove, Cosyns, & Claes, 2007), and altered levels of progesterone (Pearson Murphy, Abbott, Allison, Watts, & Ghadirian, 2004). Patients experience debilitating fatigue, sleep dysfunction, post-exertional malaise, slowed cognitive processing, hypersensitivity to sensory stimulation, and other symptoms (Carruthers et al., 2003; Fukuda et al., 1994).

Despite physiological abnormalities in ME/CFS, the disorder is frequently met with challenge, doubt, and suspicion from medical and lay persons (Bayliss et al., 2014; Travers & Lawler, 2008). Individuals with ME/CFS have reported experiencing illness invalidation by having their illness experiences discounted, minimized, or dismissed by medical professionals and significant others (Johnson & Johnson, 2006; Larun & Malterud, 2007).

Further, individuals with ME/CFS have reported feelings of shame associated with invalidation of their symptoms (Larun & Malterud, 2007). In qualitative research,

patients have sometimes attributed their feelings of shame to medical professionals and others expressing the idea that ME/CFS was "all in their heads" (Clarke & James, 2003, p. 1390). Patients with ME/CFS have also indicated that experiences of illness-invalidation and shame led to choosing to isolate from their previous social circles (Clarke & James, 2003).

The Stress Buffering Model proposes that social support counteracts threats to self-concept during periods of elevated stress (Cohen & Wills, 1985). Existing research appears to support the Stress Buffering Model, with evidence of the emotional benefits of social support to patients with health concerns indicated across multiple studies (e.g., Demange et al., 2004; Kool et al., 2012; Nenova, DuHamel, Zemon, Rini, & Redd, 2013; Zabalegui, Cabrera, Navarro, & Cebria, 2011). Within the current study, the Stress Buffering Model provides a framework for examining the ameliorative role of social support in reducing the shaming effects of illness invalidation.

Significance of the Study

Despite qualitative studies indicating that individuals with ME/CFS commonly experience illness invalidation (Asbring & Narvanen, 2002; Dickson, Knussen, & Flowers, 2007; Dickson, Knussen, & Flowers, 2008), the construct is relatively new to consideration in health psychology, and little quantitative research has been conducted in this area. Additionally, the majority of research on illness invalidation has been limited to studies of patients with rheumatic diseases (Kool, van Middendrop, Lumley, Bijlsma, & Geenen, 2012). Therefore, this study aimed to elucidate the impact of illness invalidation and social support upon shame experiences among ME/CFS sufferers.

Chapter Two

Review of the Literature

Myalgic Encephalomyelitis/Chronic Fatigue Syndrome

Myalgic Encephalomyelitis has been classified in the World Health Organization's International Classification of Diseases as a neurological disease (Carruthers & van de Sande, 2005). While fatigue is often highlighted in ME/CFS, it is only one of multiple symptoms within the disorder. Individuals with ME/CFS experience post-exertional malaise, slowed cognitive processing, hypersensitivity to sensory stimulation, sleep dysfunction, myalgia, and other symptoms (Carruthers et al., 2003; Centers for Disease Control and Prevention, 1997; Fukuda et al., 1994; Furberg, Olarte, Afari, Goldberg, Buchwald, & Sullivan, 2005; Nisenbaum, Jones, Unger, Reyes, & Reeves, 2003).

The term *fatigue* does not adequately describe the debilitating exhaustion, malaise, feelings of heaviness, and lightheadedness that individuals with ME/CFS experience (Carruthers & van de Sande, 2005). Markedly different from ordinary fatigue, in which loss of energy is easily restored by rest, individuals with ME/CFS typically experience at least a 50% reduction in activity levels as a result of incapacitating fatigue (Carruthers & van de Sande, 2005), and the severity of fatigue is experienced as "highly disproportionate to the activities preceding them" (de Carvalho et al., 2011, p. 5). Research comparing patients with ME/CFS to patients with multiple sclerosis (MS) found that individuals with ME/CFS scored higher on measures of fatigue than individuals with MS (Taillefer, Kirmayer, Robbins, & Lasry, 2002).

Cognitive fatigue is often a component of ME/CFS, as well, with physical or cognitive demands resulting in slowed cognitive processing, decreased coherence, and struggles with retrieving information and/or words (Carruthers & van de Sande, 2005). This cognitive fatigue, often referred to as *cognitive fog*, is exhibited by impaired concentration, difficulties sustaining attention, forgetfulness, confusion, and slowed reaction time. Additionally, processing of complex information may be impaired.

In a meta-synthesis of 325 peer-reviewed qualitative studies of ME/CFS, the three symptoms most often reported by participants were severe fatigue, disabling pain, and cognitive difficulties (Anderson, Jason, Hlavaty, Porter, & Cudia, 2012). In contrast, a factor analysis of empirically-derived data from existing international datasets revealed that a five-factor model best explained ME/CFS (Hickie et al., 2009). The five domains were: (a) inflammation, (b) neurocognitive problems, (c) musculoskeletal pain and/or fatigue, (d) disturbed mood, and (e) disrupted sleep and/or fatigue. These factors were evident across cultures and throughout medical settings.

Prevalence

Research by the U.S. Centers for Disease (CDC) found that prevalence of ME/CFS among adults over a four-year period varied from 3.8 to 5.2 cases per 100,000, based on geographical location (CDC, 1997). Data from the U.S. Mid-Atlantic Twin Registry (N = 4,591) found a 2.7% lifetime prevalence for ME/CFS symptoms (Furberg et al., 2005). A stratified community study in Chicago revealed a point-in-time prevalence rate of 2.2% (Jason et al., 1999), while a Wichita, Kansas study found a weighted point prevalence of 235 per 100,000 individuals (Reyes et al., 2003). Natural

history research found that rates of ME/CFS remained approximately unchanged over roughly ten years (Jason, Porter, Hunnell, Rademaker, & Richman, 2010).

Diagnostic Challenges

One of the complexities of diagnosing ME/CFS lies in that distinctive laboratory tests are not yet available to identify the disorder, meaning that diagnosis is based upon symptoms, impaired functioning, and exclusion of other disorders that might explain symptoms (Reeves et al., 2003). Such exclusionary bases for diagnosis create difficulties in ascertaining the accuracy of diagnosis.

The current quest for an ME/CFS biomarker centers upon inflammatory models of the disorder (Arnett & Clark, 2012). A distinct challenge, however, is that many of the biomarkers of inflammatory processes that appear to be involved in ME/CFS are also found in other inflammatory disorders, making reliance on inflammatory biomarkers for diagnosis problematic. However, recent research by Light and colleagues (2011) has identified changes in the expression of seven specific genes that differentiated a large subgroup of ME/CFS patients from healthy controls with accuracy of .80. It has been suggested that lab tests of these gene expressions could be combined with behavioral assessments to increase the accuracy of ME/CFS diagnosis.

Diagnostic criteria: CFS Research Case Definition. The CFS Research Case Definition (Fukuda et al., 1994) is the most widely-utilized diagnostic criteria for ME/CFS (Brown et al., 2013). This diagnostic criteria requires a minimum of six consecutive months of sustained or recurrent fatigue that is of new onset, does not stem from exertion, is not resolved by rest, and leads to significant reductions in previous activities. In addition, at least four of the following symptoms must have occurred

concurrently throughout those six months: (a) diminished concentration or memory that impacts abilities to function in previous activities; (b) sore throat; (c) tender lymph nodes; (d) pain in muscles; (e) pain in multiple joints without redness or swelling; (f) headaches that differ in severity, pattern, or types from previous experience; (g) sleep that is not refreshing; and (h) malaise lasting for at least twenty-four hours following exertion. Furthermore, in order to arrive at diagnosis, clinicians must have completed: (a) a comprehensive medical and psychosocial history, (b) a mental status examination, (c) a complete physical examination, and (d) a battery of relevant laboratory screenings. When all other sources of symptoms are excluded, a diagnosis of CFS might be made.

Illness Invalidation

Illness invalidation has been defined as "a constellation of features that includes nonacceptance by others, misunderstanding, disbelief, rejection, stigmatization, and suspicion that the [health] problem is exaggerated or psychological" (Kool, Middendorp, Boeije, & Geenen, 2009, p. 1650). Illness invalidation may be particularly likely to occur when symptoms are invisible, ambiguous, and chronic.

Individuals with chronic ambiguous conditions, including ME/CFS, have reported that their families and their medical professionals discounted their symptoms and doubted their experiences (Dickson et al., 2007; Johnson & Johnson, 2006). Illness invalidation has also been reported by patients with other "invisible" conditions, such as fibromyalgia (Kool et al., 2009), rheumatic diseases (Kool & Geenen, 2012) and chronic pain (Newton, Soutball, Raphael, Ashford, & LeMarchand, 2013). In qualitative research, individuals with ME/CFS have identified invalidation as "a defining response of others to their illness" and as a "pervasive and enduring experience" (Travers &

Lawler, 2008, p. 318). Moreover, patients with ME/CFS reported belief that absence of visibly-apparent symptoms contributed to illness invalidation (Asbring & Narvanen, 2001; Dickson et al., 2007).

Perspectives of medical professionals. As Clarke and James (2003) noted, "…one of the distinguishing features of ME/CFS is that its reality is disputed by the medical profession" (p. 1393). Research involving general practitioners from the United Kingdom revealed that 28% did not believe that ME/CFS was an identifiable clinical diagnosis (Bowen et al., 2005). In the United States, 20% of physicians indicated belief that ME/CFS is "only in the patient's head" (Brimmer, Fridinger, Lin, & Reeves, 2010, p. 8). In a meta-synthesis of 325 peer-reviewed qualitative studies, physician-specific themes included skepticism that ME/CFS is a legitimate disorder and minimization of the ME/CFS illness experience (Anderson et al., 2012). Similarly, a meta-synthesis of studies spanning from 1988 to 2013 also echoed the finding that many medical professionals do not believe that ME/CFS is a real disorder, due to the limitations in pathological findings (Bayliss et al., 2014), current lack of objective tests to ascertain the disorder's presence, and the lack of clearly-identified etiological mechanisms (Asbring & Narvanen, 2003).

Medical doctors have been found to stereotype patients with ME/CFS, based on inadequate knowledge of the ME/CFS pathophysiology (Anderson et al., 2012). Physicians expressed perceptions of many ME/CFS patients as "illness focused, demanding, and medicalising" (Asbring & Narvanen, 2003, p. 711). In particular, physicians noted that individuals with ME/CFS do not act and appear the way someone who is ill "is expected to look and behave," and they indicated that their assessment of

physical appearance weighed heavily in their interpretation of the legitimacy of a patient's symptom description (Asbring & Narvanen, 2003, p. 714). Medical doctors made judgments about the veracity of their ME/CFS patients' reports and often questioned the morality of the patient, especially of the individual's work ethic. Similarly, Swedish physicians who treated ME/CFS patients also revealed skepticism about patient experiences, noting belief that the symptoms of ME/CFS were minor, that patients were exaggerating the severity of their illness experiences, and that these patients had particularly pessimistic attitudes about life (Asbring & Narvanen, 2003).

Factors significantly associated with medical professionals endorsing the legitimacy of ME/CFS were: (a) having a personal acquaintance with ME/CFS, (b) being male, and (c) having treated a greater number of ME/CFS patients within the previous year (Bowen et al., 2005). Physicians who had previously given an ME/CFS diagnosis were less likely to believe that the disorder was "in a patient's head" (Brimmer et al., 2010, p. 8).

Perspectives of patients. Patients with ME/CFS have reported having their symptoms met with skepticism, disbelief, trivialization, and dismissal by medical professionals (Cooper, 1997; Deale & Wessely, 2001; Gilje, Soderlund, & Malterud, 2008). ME/CFS patients have described finding their interactions with doctors to be more frustrating than helpful (Clarke, 1999) and perceiving that healthcare professionals did not listen to them (de Carvalho et al., 2011). Individuals with ME/CFS reported experiencing their physicians as doubting the legitimacy of their illness, disbelieving that the symptoms stemmed from organic causes (Clarke & James, 2003), and generally misunderstanding the nature of their sickness (Cooper, 1997; de Carvalho

et al., 2011). In one study, over half of participants with ME/CFS (N = 211) reported belief that medical specialists did not seriously consider their complaints (53%) and did not sympathize with their illness experiences (54%; Prins et al., 2000). Further, ME/CFS patients have reported "being treated with disdain" and as "an annoying irritation" by their physicians (Arroll & Senior, 2008, pp. 453-454).

In other research, patients indicated that the impact of ME/CFS symptoms was significant, and yet, review of their medical records found that symptoms of fatigue were rarely included in medical documentation (Evengard et al., 2005). These results provide convergent support for the discrepant perspectives of symptoms by patients and their physicians.

Impact of illness invalidation from medical professionals. In Western culture, medical professionals are culturally-sanctioned as having power to officially recognize an individual's symptoms as medically- and socially-legitimate (Cooper, 1997; Foucault, 1975; Hyden & Sachs, 1998; Woodward, Broom, & Legge, 1995). Foucault (1975) considered this power to be a form of medical policing, and others have interpreted it as "medical paternalism" (Finerman & Bennett, 1995, p. 2; Kirmayer, 1988). Within such a culture, some patients are not granted a medical sanction for their illness, particularly those individuals whose symptoms are difficult to diagnose or who suffer from ambiguous or chronic health conditions (Chrisman, 1977; Waxler, 1980). Without this medical sanction, illness invalidation from medical professionals may have a pronounced negative impact on patients.

Having medical professionals fail to validate symptoms has reportedly contributed to individuals with medically unexplained illnesses questioning themselves,

even when they knew their symptoms to be real (Nettleton, 2006). Patients with ME/CFS have reported feeling confused, disempowered, self-doubting, and vulnerable, as well as experiencing loss of identity, upon perceiving illness invalidation from medical professionals (Arroll & Senior, 2008; Woodward et al., 1995). Patients have also reported perceiving the lack of positive support from their doctors as leading to "deterioration in emotional well-being" (de Carvalho et al., 2011, p. 7). This type of interaction has been referred to as "psychogenic dismissal," a term used to describe an experience wherein a patient experiences iatrogenic psychological harm as a result of having their symptoms dismissed as being "all in the mind" (p. 202). Thus, it comes as no surprise that in a systematic literature review, a major area of expressed need for patient with ME/CFS was to receive empathy and respect from healthcare providers (de Lourdes Drachler, 2009).

In addition, patients indicated that, in their struggle to obtain a legitimate diagnosis, they often felt a great deal of pressure to demonstrate that they were genuinely ill and to appear to be "good" or "normal" patients (Cooper, 1997, p. 199). They noted experiencing the need to prove that, although their illness might be considered "deviant," they were not deviant themselves (p. 199). On the other hand, patients have reported withholding information about their symptoms, when interacting with medical professionals who were perceived as invalidating (Nettelton, 2006).

Individuals with ME/CFS noted that illness invalidation from medical professionals often contributed to invalidation from family, employers, and social networks and to decreased social support (Cooper, 1997; de Carvalho et al., 2011). As one patient described it:

> I was more or less bed bound for a couple of years, …and meanwhile the doctors were telling me there was nothing wrong with me. So I was under pressure from employers, family and everyone else to stop imagining that I was ill, and to get out of bed and get on with my life. (de Carvalho et al., 2011)

Illness invalidation from social networks. Despite having a diagnostic label to explain their symptoms, ME/CFS patients reported that the lack of visible symptoms contributed to illness invalidation from social networks (Fisher & Crawley, 2012). Patients reported having their symptoms trivialized and having others explain to them that "tiredness" is a normal experience (p. 564). Patients often reported that friends were skeptical of the legitimacy of their sickness, that they were judged and rejected by many in their previous social networks, and that they often lost their friendship networks (Clarke & James, 2003).

Adolescent patients reported that some friends and teachers invalidated their experiences of fatigue, doubting them, appearing to distrust their reported symptoms, dismissing their illness as malingering, and attributing their physical inabilities to laziness (Fisher & Crawley, 2012). Further, adolescents with ME/CFS noted having difficult experiences of teasing and bullying about their illness from peers.

Moreover, individuals with ME/CFS reported experiencing emotional pain as a result of illness invalidation from significant others, family members, friends, and the work environment (Asbring & Narvanen, 2002; Dickson et al., 2007; Larun & Malterud, 2007). The experiences of both illness invalidation and physical limitations often led individuals to believe that they were no longer entitled to their former

relationships, that they were now inferior in their interpersonal worth, and to a general sense of painful relational disconnection (Travers & Lawler, 2008).

In a meta-synthesis of 325 peer-reviewed qualitative studies of ME/CFS, a common theme among patients was the experience of reduced social connections and disrupted personal relationships in association with the illness (Anderson et al., 2012). ME/CFS patients often reported feeling socially isolated and as though they were "outsiders" (Clark & James, 2003, p. 1390). Biro (2012) described this process as follows:

> …[The ill] suffer just as much, if not more, because they feel isolated from others, because they feel alone. So the circle widens once again: the body unravels, the self unravels, and now our relationship with the world unravels. (p. 47)

Illness invalidation from family. In qualitative research, patients reported that few of their family members continued to support them and to validate their illness (Clarke & James, 2003). Invalidation of illness experiences by a spouse were considered particularly emotionally difficult (Dickson et al., 2007) and were negatively related to adaptive outcomes (Heijmans, DeRidder, & Bensing, 1999). As one patient described it,

> The difference with friends – if they're not helpful you don't have to talk to them, but with [family] you have to – you can't choose your family. So you learn what to say and what not to say. (Clarke & James, 2003, p. 1391).

Shame

Shame has been defined as an individual's global assessment of the self as inferior, deficient, and of diminished value (Woien, Ernst, Patock-Peckham, & Nagoshi, 2003). In qualitative research, adults have described shame as eroding positive self-concept, harming one's sense of connection with others, and ultimately, bringing about perceived loss of power (Van Vliet, 2008). Shame has been conceptualized as an emotional response that originates from interpersonal transactions (Tangney, Miller, Flicker, & Hill Barlow, 1996), evaluative social conditions, and threats to the social self (Dickerson, Gruenwald, & Kenemy, 2004; Dickerson, Gruenwald, & Kemeny, 2009). Shame is particularly likely to be evoked in situations in which a component of one's identity is, or has the potential to be, negatively evaluated by others (Dickerson et al., 2004).

While shame is considered a negative evaluation of oneself, it is believed to be linked to the social environment, to the belief that a critical "imagined other" is judging the self (Lewis, 1971), and to cultural expectations (Martens, 2005). Shame has also been posited as resulting from discrepancies between external expectations and assessment of internal realities, and as "an emotion which has incorporated the gaze and the voice of the other" (p. 404). Scheff (2000) described shame as a "social emotion" that arises from "threat to the social bond" (p. 97). Also, Dickerson and colleagues (2004) described shame as resulting "when perceptions of negative *social* evaluation are transformed into negative *self*-evaluation" (p. 1195).

Shame has also been conceptualized as an emotional response to low social standing and the associated risks of interpersonal rejection, exclusion, or maltreatment

(Balsamo et al., 2014; Dickerson et al., 2004). Research found that, among psychology students, a strong positive correlation existed between blaming oneself for having been socially criticized or "put-down" and the experience of shame (Gilbert & Miles, 2000, p. 768). Indeed, social rank and experiences of shame have been found to be highly correlated (Cheung, Gilbert, & Irons, 2004; Gilbert, 2000). The social aspect of shame has been reflected in qualitative research, as well. In a study consisting of nearly one hundred autobiographical narratives, results revealed that, in situations in which participants wrote shameful statements, the participants' writing focused upon how others might be negatively evaluating them (Silfver, 2007).

Illness and Shame. Anthropologists have noted that Western culture has shifted toward new explanatory models of illness that are "blame focused" and that place responsibility for disease processes and their outcomes upon the patients themselves, often ignoring the complexities of disease processes (Finerman & Bennett, 1995, p.1). These attributions of patient responsibilities for their illnesses may stem from increased lay and professional education about connections between lifestyle choices and disease. Such attributions ignore the reality that health difficulties are often induced by multiple causes and are frequently complex. However, the emphasis on patient accountability for lifestyle choices may translate into interpretations of sick individuals as blameworthy for their illnesses.

An example of this phenomenon of focusing blame on patients for their illness, while ignoring complexities of diseases processes, is reflected in results from qualitative research in the United Kingdom. Research there found that individuals with lung cancer who had no prior history of smoking considered themselves as being unfairly blamed by

those around them for their disease (Chapple, Ziebland, & McPherson, 2004). Furthermore, some patients noted belief that media reports linking smoking to lung cancer may have contributed to the assessment that they were to blame for their cancer. Qualitative research involving patients visiting a Danish general practice revealed that patients who were not successfully managing their health acknowledged that they were failing to fulfill others' expectations and experienced shame (Guassora, Reventlow, & Malterud, 2014). Similarly, patients with obesity reported feeling shame in relation to messages of blame from the dominant culture (Kirk et al., 2014).

Multiple studies point to individuals with health difficulties experiencing shame pertaining to their physical condition and subsequently choosing to isolate from social circles. For example, patients with chronic back pain reported feelings of shame associated with relational contexts in which they perceived critical judgments from others (Smith & Osborn, 2007). These patients reported engaging in social withdrawal and avoidance in association with shame. Similarly, patients with irritable bowel syndrome (IBS) reported experiencing shame associated with intimate relationships, resulting in avoidance of situations where they might be socially embarrassed (Hakanson, Sahlberg-Blom, Nyhlin, & Ternestedt, 2009). Norwegian women who had weight-loss surgery and who experienced difficulties with chronic pain and fatigue afterward, or who began to regain the lost weight, were found to experience shame regarding others' perceptions of their choices and to restrict their social interactions (Groven, Raheim, & Engelsrud, 2010). Individuals with ME/CFS have reported shame in connection with failures to conform to social expectations, including the cultural norm that illnesses be physiologically observable, and in connection with experiences of

illness invalidation (Travers & Lawler, 2008). These experiences of shame were associated with social isolation and disconnection.

Shame has also been found to negatively correlate with psychological wellbeing, and this finding has been found to sustain across the lifespan (Orth, Robins, & Soto, 2010). Individuals living in Greece who were ashamed of past experiences of physical pain were found to have higher levels of psychopathology than individuals who had not experienced shame surrounding physical pain (Paschou, Damigos, Mavreas, & Gouva, 2010). Additionally, external shame (shame stemming from negative evaluations by others) has been found to correlate with experiences of depression and with submissive behaviors (Cheung et al., 2004; Kim, Thibodeau, & Jorgensen, 2011).

Research indicates that social-evaluative threat and shame may have negative effects on physical health, as well. Experiences of shame have been found to have a positive relationship with proinflammatory cytokine activity, suggesting that shame may result in immunological changes (Dickerson, Kemeny, Aziz, Kim, & Fahey, 2004). Additionally, a meta-analysis of 208 research studies found that stress stemming from uncontrollable social-evaluative threats was associated with significant increases in cortisol and adrenocorticotropic hormone (Dickerson & Kemeny, 2004). Further, in research conducted in Norway, higher reports of shame were associated with increased likelihood of protracted illness-related absences the following year (Knapstad, Overland, Henderson, Homgren, & Hensing, 2014). And, experiencing shame in association with medical care has been linked to avoidance of healthcare utilization or lying to doctors, in order to avoid repeated experiences of shame (Green et al., 2010).

Illness Invalidation and Shame

Discussing one's illness within the context of an invalidating environment may lead to internalized shame (Myers, 2004). Ware (1992) proposed that, when the subjective experience of one's illness is denied by the social context, shame of "being wrong in one's definition of reality" may occur (p. 347). Indeed, women with ME/CFS reported experiencing discrepancies between their own definition of their experience and the perceptions of their work environment and their physicians (Asbring & Narvanen, 2001).

Individuals with ME/CFS have expressed feelings of shame associated with illness invalidation and of experiencing their identity as questioned and no longer legitimate (Larun & Malterud, 2007). In qualitative research, participants sometimes attributed their experience of shame to physicians, members of the media, and other individuals having expressed the belief that ME/CFS was "all in their heads" (Clarke & James, 2003, p. 1390). Some individuals with ME/CFS even reported removing themselves from their former social lives out of feelings of shame over how others perceived them or after having others reject that their illness was legitimate (Clarke & James, 2003).

In research involving a large sample of adults (N = 915), half of the participants reported experiencing at least one interaction with a medical doctor in which they felt ashamed, with shame-provoking situations including those in which a physician engaged in illness invalidation of their health complaints (Harris & Darby, 2009). Similarly, a majority of Swedish patients who had experienced negative encounters within healthcare settings, including having their provider not believe them, doubt their

condition, or question their motivation to work, identified feelings of shame (Lynoe et al., 2013).

Social Support and the Stress Buffering Model

The Stress Buffering Model proposes that social support mitigates the negative effects of stress and promotes wellbeing (Cohen & Wills, 1985). Social support is conceptualized as manifesting across four support domains. *Esteem support* refers to communication that the individual is valued, worthy, and accepted. *Informational support* is operationalized as provision of coping support, advice, and guidance. *Social companionship* refers to time spent with others that is accompanied by a sense of belonging. Finally, *instrumental support* refers to assistance with tangible needs.

According to the Stress Buffering Model, esteem support counteracts threats to self-concept that may occur during stressful periods by communicating valuing and acceptance (Cohen & Wills, 1985). Informational support may promote cognitive reappraisals of stressful situations and/or may contribute to successful coping strategies. On the other hand, instrumental support and social companionship meet practical and affiliation needs during taxing times.

Existing research appears to support the Stress Buffering Model, with evidence of the emotional benefits of social support to patients with health concerns indicated across multiple studies. Among individuals with rheumatic diseases, social support has been found to significantly relate to improved mental health (Kool et al., 2012). Specifically, individuals with rheumatoid arthritis who received higher amounts of daily emotional support were more likely to report increased psychological wellbeing, decreased anxiety, fewer sleep difficulties, and less depression (Demange et al., 2004).

Similarly, research involving individuals with advanced cancer in Spain, revealed a significant moderate positive correlation between perceived social support and having a positive focus (Zabalegui, Cabrera, Navarro, & Cebria, 2011). Among survivors of hematopoietic stem cell transplant, emotional and instrumental social support predicted posttraumatic growth (Nenova, DuHamel, Zemon, Rini, & Redd, 2013). Furthermore, in a study of patients with HIV in Nepal, global satisfaction with social support significantly predicted quality of life, with the mediating effect of social support being hope (Yadav, 2010).

Social support has also been linked to health-related quality of life, as well. A literature review of 175 studies of social support and coping among prostate cancer patients revealed that the preponderance of research has indicated that perception of social support exerts a main effect upon health-related quality of life (Paterson, Jones, Rattray, & Lauder, 2013). Social support has also been linked to health-related quality of life in patients with heart failure (Bakan & Akyol, 2008). In addition, global satisfaction with social support predicted health-related quality of life among heart transplant recipients at five years post-transplant, and satisfaction with emotional aspects of social support predicted health-related quality of life at ten years post heart transplant (White-Williams et al., 2013). Among patients with diabetes in Turkey, perceived social support was positively correlated with quality of life as well (Goz, Karaoz, Goz, Ekiz, & Cetin, 2007).

In regard to the moderating effects of social support on health specifically, however, the Stress Buffering Model was not supported in a sample of Dutch and Belgian individuals with rheumatic diseases (Kool, Middendorp, Lumley, Bijlsma, &

Geenen, 2012). Thus, it may be that the buffering effects of social support are more likely to be found with regard to emotional wellbeing than actual physical health.

Individuals who lack medical explanations of illness symptoms have reported experiencing isolation and lack of social support (Nettleton, 2006). Patients with ME/CFS have expressed feelings of loneliness and separation from others, following perceived rejection by friends and significant others (Dickson et al., 2007), and adolescents with ME/CFS have noted perceiving that their inability to engage in activities as before had tested their relationships (Fisher & Crawley, 2012).

Many patients with ME/CFS noted belief that receiving social support and validation would have increased their quality of life and facilitated coping (Dickson et al., 2007). Additionally, individuals with moderate to severe ME/CFS indicated that social support was particularly important to them in practical areas of personal care, assistance with family responsibilities, and help with domestic chores (de Carvalho et al., 2011). In a systematic literature review, among the needs expressed by patients with ME/CFS was a strong desire for positive support from social networks (de Lourdes Drachler et al., 2009).

Research results have indicated that when members of patients' social networks understood that ME/CFS was seriously impacting their functional abilities, trust was increased in those relationships (Fisher & Crawley, 2012). Additionally, patients with ME/CFS who experienced continued social support, following illness onset, reported deeply valuing those relationships (Clark & James, 2003).

However, research on the ameliorative impact of social support among individuals with ME/CFS has been limited, further justifying the current study.

Illness Invalidation, Shame, and the Stress Buffering Model

Discussing one's illness within the context of an invalidating environment may lead to internalized shame (Myers, 2004). Indeed, individuals with ME/CFS have expressed feelings of shame associated with the invalidation of their symptoms and having their experiences discounted, minimized, or dismissed (Larun & Malterud, 2007).

According to the Stress Buffering Model, esteem support from social connection may help counteract threats to self-concept that may occur during stressful periods by communicating valuing and acceptance (Cohen & Wills, 1985). In fact, adults who considered themselves to have recovered from shame identified the following as helpful to their achieving healing from shame: (a) finding at least one or two individuals who supported them and offered unconditional acceptance, (b) connecting socially with others, (c) communicating with at least one person about their shame event and experiencing that person as listening, understanding, and maintaining belief in their positive attributes (Van Vliet, 2008).

Hypotheses

Although qualitative studies have indicated that individuals with ME/CFS commonly experience illness invalidation (Asbring & Narvanen, 2002; Dickson, Knussen, & Flowers, 2007; Dickson, Knussen, & Flowers, 2008), the construct is relatively new to consideration in health psychology, and little quantitative research has been conducted in this area. To date, no peer-reviewed quantitative studies have been published that evaluate experiences of illness invalidation in the ME/CFS population.

Therefore, this study aimed to elucidate the impact of illness invalidation and social support upon shame experiences among ME/CFS sufferers.

Based upon existing qualitative research, indicating connections between illness invalidation and shame (e.g., Asbring & Narvanen, 2002; Dickson, Knussen, & Flowers, 2007; Dickson, Knussen, & Flowers, 2008), it was hypothesized that there would be a positive and significant relationship between illness invalidation and state shame. From the theoretical foundation of the Stress Buffering Hypothesis (Cohen & Wills, 1985), it was further hypothesized that social support would moderate the relationship between illness invalidation and shame, with the relationship between illness invalidation and state shame changing significantly based on level of social support.

Chapter Three

Methods

Participants

Individuals were recruited to participate if they were between the ages of 18-64, identified as being ill with ME/CFS, and were not pregnant. Participants were recruited via ME/CFS and chronic illness support web sites, web sites for ME/CFS advocacy and research, and social networking sites. A snowball recruitment method was utilized, with participants encouraged to forward the survey to other potential participants.

One hundred eighty-five participants responded to the survey. Of those, ten recruits declined participation, three participants denied having ME/CFS, and four individuals agreed to participate, but subsequently, chose not to complete the survey. Thus, the final $N = 168$.

As indicated in Table 1 (Appendix A), the mean age of participants was 45.5 years ($n = 137$, $SD = 11.12$, range 18-64), with 8.5% between the ages of 18-30, 13.4% between the ages of 31-40, 26% between the ages of 41-50, 20.5% between the ages of 51-60, and 5.4% between the ages of 61-64. A relatively large percentage (25.9%, $n = 48$) of participants elected not to reveal their age. The self-identified gender of participants was 68.1% female, 6.5% male, and 0.5% other, with a number of participants (24.9%, $n = 46$) opting not to reveal their gender (see Appendix A, Table 1).

Participants identified their racial/ethnic background as 64.9% Caucasian, 0.5% Native American/American Indian, 0.5% African origin, 0.5% Asian origin, and 8.1%

Other (refer to Appendix A, Table 1). Approximately one-fourth (25.4%, *n* = 47) of participants did not identify their racial/ethnic background.

In regard to marital status, 37.3% identified as married, 18.4% as single, 11.4 % as divorced, 0.5% as widowed, and 7.6% as other (see Appendix A, Table 1). Roughly one-fourth of participants (24.9%) opted not to answer questions regarding marital status.

Nearly half of participants reported having completed a college degree (45.4%), with 7.6% having an associate's degree, 18.9% reporting a bachelor's or equivalent professional degree, 16.2% indicating a master's or equivalent professional degree, and 2.7% reporting a doctorate or equivalent professional degree. Of those who had not completed a college degree, 14.1% had completed some college, 7.6% had completed vocational training, 10.3% had completed high school, 1.6% had completed junior high/middle school, and 3.2% indicated their education as Other (refer to Appendix A, Table 1).

Despite being a relatively well-educated sample, 43.7% of participants indicated household incomes of $29,000 per year or less. Roughly half of participants (47.6%) reported being without employment, and 45.9% indicated disability status.

Regarding length of time since onset of ME/CFS symptoms, 2.7% of participants had experienced symptoms for less than one year, 10.8% for one to three years, 12.4% for four to six years, 15.1% for seven to 10 years, 18.9% for 11-20 years, 8.6% for 21-30 years, and 4.3% for over 30 years. Twenty-seven percent (27%) of participants did not answer questions regarding duration of symptoms. There was wide variability in length of time between symptom onset and diagnosis with ME/CFS, with

9.7 % being diagnosed within six months or less of symptom onset, 15.7% within six months to one year, 23.2% between two and four years, 8.1% within five to seven years, 7.6% between eight and 10 years, and 9.2% in 11 or more years. A large number of participants (*n* = 49; 26.5%), however, chose not to respond to this item.

Participants described their current symptom status as follows: intense symptoms and/or many problematic symptoms (39.5%), moderate symptoms (22.2%), mild symptoms (5.9%), and symptoms in remission (0.8%). As with previous demographic items, a large number of participants (*n* = 59; 31.9%) chose not to designate their current symptom status.

Participants were asked to rate, on a scale of 0-100, the severity of their ME/CFS symptoms, with "0" indicating total absence of symptoms and increasingly higher numbers indicating increased severity of symptoms. Participants provided ratings ranging from 28-100. The mean rating for ME/CFS symptom severity was 69.93 (*SD* = 15.57), with 3.7% of responding participants reporting severity between 28-40, 13.4% indicating limitation ratings of 41-60, 33.0% between 61-80, and 15.4% between 81-100. The percentage of participants who chose not to rate their symptom severity was 34.1% (*n* = 63).

Additionally, participants were asked to rate, on a scale of 0-100, how limited they were by their ME/CFS symptoms, with "0" indicating total absence of limitations and increasingly higher numbers indicating increased limitations by symptoms. Participants provided ratings ranging from 20-100. The mean rating for limitations was 70.36 (*SD* = 17.35), with 5.8% of participants rating their limitations between 20-40, 13.9% indicating limitations of 41-60, 25.6% between 61-80, and 22.6% between 81-

100. Finally, 31.4% (n = 58) elected not to rate their limitations from ME/CFS symptoms.

Roughly thirty five percent (35.1%) of participants reported being involved in a support group for individuals with ME/CFS.

Instruments

Demographic questionnaire. A demographic questionnaire was created by the author for use in this study (Appendix C). Demographic information was requested regarding gender, racial/ethnic background, highest level of education attained, employment, disability status, and income. Participants were also asked questions pertaining to time since ME/CFS symptom onset, time from symptom onset to diagnosis, symptom severity, and symptom-related limitations.

Illness Invalidation Inventory. The Illness Invalidation Inventory (I*3; Kool et al., 2010) is a 40-item self-report measure that assesses perceived illness invalidation from five sources: spouse, family, medical professionals, work environment, and social services (Appendix C). In factor analysis conducted by the measure's authors, I*3 statements were found to load on two factors of invalidation: Discounting and Lack of Understanding. Thus, the I*3 was divided into two subscales, Discounting and Lack of Understanding.

Each subscale of the I*3 contains eight statements, rated on a 5-point Likert scale (ranging from "never" to "very often"; Kool et al., 2010). A sample "Discounting" item is " My….finds it odd that I can do much more on some days than on other days." An example of a "Lack of Understanding" item is "My….takes me seriously" (R). A source subscale that does not apply (e.g., because the participant was not married or was

unemployed) is automatically skipped. Twenty-four items are reverse scored. Subscale scores are obtained by averaging scores for all items in the respective subscales.

In the current study, Cronbach's alpha for the overall I*3 measure was good, with (.91). Likewise, Cronbach's alphas were also good for the subscales, Lack of Understanding (.88) and Discounting (.85). (All reliability results may be found in Appendix A, Table 2.)

The State Shame and Guilt Scale. Measures of shame fall into two categories: (1) those that assess *trait* shame, and (2) those that measure *state* shame (Robins, Noftle, & Tracy, 2007). Measures of trait shame assess one's shame-proneness as an enduring aspect of the personality. Measures of state shame, on the other hand, assess whether one experiences shame at a certain point in time, and such instruments may be used to measure shame in response to situational factors. For the purposes of this study, it was preferable that shame be measured in regard to one's experiences of perceived illness invalidation, rather than in regard to one's personality. As a result, the State Shame and Guilt Scale (SSGS; Marschall, Sanftner, & Tangney, 1994), a measure of state shame, was selected (Appendix C).

The State Shame and Guilt Scale (Marschall et al., 1994) is a 15-item (five items in each of three subscales) self-report instrument developed to measure Shame (the self as intrinsically bad), Guilt (the self as having behaved poorly), and Pride (the self as valued; Marschall et al., 1994). Subscale items were reportedly developed in light of empirical and theoretical literature (Dearing & Tangney, 2002). Sample items include, "I feel worthwhile, valuable;" "I feel small;" and, "I feel worthless, powerless" (Marschall et al., 1994).

Participants indicate their agreement/disagreement with statements on a five-point Likert scale. All items are scored in a positive direction. Only the Shame subscale was utilized in data analysis, as it was the only SSGS subscale pertinent to the study's hypothesis. Shame subscale scores were calculated by obtaining mean scores. In the current study, Cronbach's alpha for the State Shame subscale was .80 (refer to Appendix A, Table 2).

The Medical Outcomes Study Social Support Survey. The Medical Outcomes Study Social Support Survey (MOS; Sherbourne & Stewart, 1991) is a 19-item self-report measure of social support that was designed specifically for use with patients with chronic illness (Appendix C). The MOS includes four subscales that measure various dimensions of social support, namely *emotional/informational*, *tangible*, *affectionate*, and *positive social interaction.*

The MOS (Sherbourne & Stewart, 1991) was developed in light of the Stress Buffering Hypothesis (Cohen & Wills, 1985) and current theory regarding social support outlined in the existing literature. In order to minimize participant fatigue, social support was measured without respect for the source (e.g., whether social support came from friends, community, religious circles, etc.). Items were found to discriminate from measures of loneliness, mental health, feelings of belonging, perceptions of current health, and other dimensions of family and interpersonal functioning. In the measure's development, internal-consistency reliability was found to be high for each subscale, with alphas ranging from .91 to .97, and one-year stability coefficients ranged from .72 to .78. In the current study, Cronbach's alpha = .96 for the overall scale (see Appendix A, Table 2).

The MOS Emotional/Informational Support subscale consists of eight items that assess participant experiences of having someone in their life who expresses positive feelings, empathy, understanding, and encouragement of expression of emotion, as well as someone who offers suggestions, guidance, information, or feedback. The Tangible Support subscale is comprised of four items that evaluate experiences of receiving material or physical assistance from others. The Positive Social Interactions subscale is composed of three items that evaluate the experience of relationships that include enjoyable experiences and activities to the participant. The Affectionate Support subscale contains three items that assess whether the participant has someone in their life who expresses positive regard, fondness, and love toward them. For each item, participants rate the availability of that particular support on a 5-point Likert scale, ranging from "None of the Time" to "All of the Time." Example items include: "Someone to confide in or talk to about yourself or your problems…;" "Someone to share your most private worries and fears…;" and, "Someone who understands your problems…."

All MOS items are scored in a positive direction (Sherbourne & Stewart, 1991). Subscale scores are calculated by averaging all scores on each respective subscale, and the Overall Support Index score is obtained by averaging the mean of all subscale scores and the additional item score (Rand Corporation, n.d.).

Results of principle components factor analysis supported the use of an overall total score (Overall Support Index; Sherbourne & Stewart, 1991). Multitrait and confirmatory factor analyses supported the use of MOS subscale scores, as well.

Further, the authors recommended the option of utilizing subscale scores in research, based upon their usefulness in testing theoretical hypotheses.

Procedure

The University of Oklahoma Institutional Review Board reviewed and approved this study (Appendix B). An online survey was created via Qualtrics, and was maintained via a secure server in the University of Oklahoma's Center for Educational Development and Research. Links to the study were posted on ME/CFS and chronic illness support web sites, web sites of ME/CFS advocacy and research organizations, and social networking sites. Participation was voluntary, without remuneration. Informed consent to participate was obtained electronically by providing an information sheet regarding the study and with participants clicking on whether they agreed to participate or not. If participants indicated that they did not wish to participate, they were immediately exited from the survey. Following consent, participants could skip questions or exit the survey at any time. No identifying information was collected.

Following informed consent, instruments were administered in the following order: I*3, MOS, SSGS, and Demographic Questionnaire.

Data Analysis

It was predicted that participants with ME/CFS would exhibit a significant positive relationship between their report of perceived illness invalidation and experiences of shame. Further, it was predicted that those who received more social support would evidence a significantly weaker positive relationship between illness invalidation and shame, due to the buffering effects of social support.

Preliminary analysis included assessment for violations of parametric assumptions, including review of the P-P plot and evaluation of skewness and kurtosis in the data. Additionally, Kolmogorov-Smirnov and the Shapiro-Wilk tests were utilized to assess for normality. After removing one item that contained an extreme outlier from the data set, no violations of normality were observed.

A hierarchical multiple regression was utilized to test the hypothesis that the MOS Overall Support Index would moderate the relationship between I*3 Lack of Understanding and SSGS Shame. Moderator variables are intended to affect the direction and/or strength of the relationship between a predictor variable and the outcome variable (Baron & Kenny, 1986), and hierarchical multiple regression is considered the preferred method for identifying the presence or absence of moderating effects when the predictor and/or moderator variables are measured on a continuous scale (Aiken & West, 1991). It is important to note that, in hierarchical regression, moderators may, or may not, be significantly related to the predictor or the outcome variable, and the predictor may or may not be correlated to the outcome (Frazier et al., 2004).

Some have argued that centering of predictor variables is no longer considered a necessary step in hierarchical regressions, as centering has reportedly been found to change the intercept, without affecting the shape of the moderation results (Jose, 2013). However, as a precaution, variables were centered in the current study.

Since correlation analyses revealed significant relationships between select demographic variables (i.e., income and length of time since symptom onset) and the outcome variable (i.e., SSGS Shame), these demographic variables were entered into

the first step of the regression model, in order to control for their effects on SSGS Shame. Subsequently, I*3 Lack of Understanding was entered into the second step, and the MOS Overall Support Index was entered into the third step. Finally, the interaction between I*3 Lack of Understanding and the MOS Overall Support Index was entered into the fourth step.

Chapter Four

Results

Preliminary Analyses

Preliminary examination of the data revealed one extreme outlier. After removal, no violation of the assumptions of normality, linearity, multicollinearity, and homoscedasticity were observed.

Means and standard deviations of the predictor and criterion variables included in the overall model are given in Table 2 (Appendix A). Pearsons's bivariate correlational analyses of relationships between the variables of interest are summarized in Table 3 (Appendix A).

Demographic variables that significantly correlated with SSGS Shame included income, marital status, and length of time since symptom onset. Annual income of participants evidenced a significant negative relationship with SSGS Shame (r = -.19, p < .05), indicating a trend toward individuals with lower income reporting higher levels of state shame. Length of time since symptom onset was also significantly and negatively related to SSGS shame (r = -.20, p < .05), such that those who were newer to the symptom experience were more likely to experience shame.

Race evidenced a significant negative relationship with the Lack of Understanding subscale of the I*3 (r = -.24, p < .01), such that participants of color reported higher levels of illness invalidation in the form of Lack of Understanding. Further, participant ratings of the severity of their ME/CFS symptoms were significantly positively related to their experiences of illness invalidation in the form of discounting (I*3 Discounting), r = .15, p < .05, indicating that participants who reported

greater symptom severity also experienced greater levels of illness invalidation in the form of discounting.

Income also evidenced significant relationships with the MOS Overall Support and with all of the four MOS subscales. MOS Overall Support was significantly positively correlated with income ($r = .26$, $p < .01$), such that increases in income corresponded to increases in overall social support. Positive Social Interactions subscale scores were also significantly positively correlated with income ($r = .33$, $p < .01$), as were Affectionate Social Support ($r = .27$, $p < .01$), Emotional/Informational Support ($r = .21$, $p < .05$), and Tangible Social Support ($r = .17$, $p < .05$).

In regard to the variables of interest, illness invalidation in the form of lack of understanding (I*3 Lack of Understanding) was significantly correlated with state shame (SSGS Shame), $r = .15$, $p < .05$. However, illness invalidation in the form of discounting (I*3 Discounting) was not significantly related to shame (SSGS Shame), $r = .11$, $p = .10$.

Overall perceived social support (MOS Overall Support Index) evidenced a significant negative relationship with state shame (SSGS Shame), $r = -.17$, $p < .05$, such that lower perceived social support was indicative of higher reported state shame. When social support was further broken down into subscale components, perceived Emotional/Informational Support (MOS Emotional/Informational) evidenced a significant negative relationship with shame ($r = -.20$, $p < .01$), as did perceived Positive Social Interactions ($r = -.21$, $p < .01$) and perceived Affectionate Support ($r = -.18$, $p < .05$). Of the MOS subscales, only the subscale measuring Tangible Social Support

(MOS Tangible) did not exhibit a significant negative correlation with shame (SSGS Shame).

Hierarchical Multiple Regression Analysis

A hierarchical multiple regression model was utilized to examine the variance in state shame (i.e., SSGS Shame subscale) accounted for by illness invalidation in the form of lack of understanding (i.e., I*3 Lack of Understanding subscale). Since correlation analyses revealed significant relationships between select demographic variables (i.e., income and length of time since symptom onset) and the criterion variable (i.e., SSGS Shame), these demographic variables were entered into the first step of the regression model in order to control for their effects on SSGS Shame. Subsequently, I*3 Lack of Understanding was entered into the second step, and the MOS Overall Support Index was entered into the third step. Finally, the interaction between I*3 Lack of Understanding and MOS Overall Support Index was entered into the fourth step.

As noted in Table 4 (Appendix A), the total variance in scores explained by the overall regression model was not significant. Annual income and length of time since symptom onset explained 6.8% of the variance in state shame at Step 1 of the model, achieving significance, F (2, 111) = 5.13, $p < .01$, and evidencing a small effect size (Cohen, 1988). Illness invalidation in the form of Lack of Understanding (I*3 Lack of Understanding subscale) did not significantly explain variance in the second step, and the MOS Overall Support Index did not have a significant main effect on SSGS Shame scores in the third step. The interaction term (I*3 LOU x MOS Overall Support Index) was entered at Step 4, but was not significant.

Chapter Five

Discussion

Qualitative studies have indicated that individuals with ME/CFS frequently experience illness invalidation (Asbring & Narvanen, 2002; Dickson, Knussen, & Flowers, 2007; Dickson, Knussen, & Flowers, 2008), with associated feelings of shame and of experiencing their identities as questioned and no longer legitimate (Larun & Malterud, 2007). Prior to this study, however, there have been no known peer-reviewed quantitative research studies measuring the relationship between illness invalidation and shame among individuals with ME/CFS.

Based on existing qualitative research, the current study hypothesized that there would be a positive and significant relationship between perceived illness invalidation (i.e., I*3) and experiences of state shame among individuals with ME/CFS (i.e., Shame subscale of SSGS). Data analysis partially supported this hypothesis, in that illness invalidation in the form of lack of understanding exhibited a significant positive correlation with experiences of shame. However, illness invalidation in the form of discounting was not significantly related to shame.

The correlation between illness invalidation in the form of lack of understanding and experiences of shame is in alignment with existing qualitative research indicating that discussing one's illness within the context of an invalidating environment may lead to internalized shame (Myers, 2004), particularly in relation to the perception of "being wrong in one's definition of reality" (Ware, 1992, p. 347). Qualitative research has found that individuals with ME/CFS experienced particular difficulties stemming from others not understanding the fluctuations in ME/CFS symptoms or the physical and social ramifications of managing the illness (Larun & Malterud, 2007). Similarly,

patients have reported that their partners did not understand the consequences of their symptoms, such as limitations in being able to predict whether they would feel well enough to participate in planned activities (Dickson, Knussen, & Flowers, 2007). Further, individuals with ME/CFS have revealed challenges associated with others not understanding that pacing themselves and conserving energy on days when they felt better was a necessary component of managing the disorder.

In searching for potential ameliorative factors to counter experiences of illness invalidation and shame, the Stress Buffering Model (Cohen & Willis, 1985) was reviewed as a possibly helpful theoretical framework. The Stress Buffering Model suggests that social support moderates the emotional effects of negative experiences and facilitates increased wellbeing (Cohen & Wills, 1985). Existing research appears to support the Stress Buffering Model, with evidence across multiple studies indicating the emotional benefits of social support to patients with health concerns (e.g., Kool et al., 2012; Nenova, DuHamel, Zemon, Rini, & Redd, 2013; Zabalegui, Cabrera, Navarro, & Cebria, 2011).

In keeping with the Stress Buffering Model, qualitative research has indicated the benefits of social support in counteracting the emotional impact of illness invalidation (Travers & Lawler, 2008). Specific social supports that were indicated as useful included: receiving help, acquiring information, and experiencing interpersonal safety. Additionally, individuals with ME/CFS described these social experiences as contributing to the reclamation of valuable aspects of the self.

Thus, the second hypothesis of the study predicted that social support would moderate the relationship between illness invalidation and state shame, with increases in

social support leading to decreases in shame. A hierarchical regression was utilized to evaluate the relationships between variables. However, results of the overall model were not significant. Thus, the hypothesis was not supported in this sample.

In assessing the seeming discrepancies between the results of the current study and existing qualitative research on experiences of illness invalidation, shame, and social support among individuals with ME/CFS, it is important to note that the relational aspect of qualitative research may create a more inviting milieu in which to divulge one's experiences of illness invalidation and shame than that found in quantitative research conducted online. Thus, it may be that the format in which research is conducted contributes to differences between qualitative and quantitative research participants' willingness to reveal experiences of illness invalidation and shame.

While this sample was relatively well-educated, nearly half of participants (42.7%) reported annual incomes of $29,000 per year or less. Roughly half of participants (47.6%) reported being without employment, and 45.9% indicated disability status. It may be that the symptom composite of ME/CFS contributes to impaired ability to work, which may lead to unemployment and/or disability status. The impact of loss of employment and/or disability status may lead to lower income, despite educational qualifications.

In the current study, income evidenced a significant positive relationship with overall social support, as well as with the majority of the social support subcomponents, including positive social interactions, affectionate support, emotional/informational

support, and tangible support. Thus, as income increased, these components of social support, as well as overall social support, also increased.

The indications that income and overall social support were positively related may reflect broader classist ideas within the culture that place greater value on individuals with higher financial worth than upon individuals of lower socioeconomic status (SES). This may account for individuals with lower income receiving less social support in the form of affectionate support, positive social interactions, and emotional/informational support. Additionally, it may be that individuals from lower SES have less financial resources for engaging in positive social interactions (e.g., less money to hire a babysitter to go to events with friends, less funds for going out to restaurants with others, etc.). Further, in regard to informational support, it may be that individuals with higher incomes have increased access to educated and knowledgeable individuals who can offer them helpful information, as well as better access to advanced medical care and specialists. Finally, individuals with greater income may have increased financial capacity to pay for individuals to provide tangible support services, such as housekeepers, home health care, individuals to run errands, etc.

The results of this study also indicated a significant negative relationship between shame and income among individuals with ME/CFS, indicating that participants with higher income were more likely to report lower levels of state shame. Or, stated another way, the trend was that individuals with lower income were more likely to report increased levels of shame. These results may point to the intersectionality of illness-related shame and class-related experiences of shame. This

intersectionality may be summed up in the words of Scambler (2006), who wrote the following regarding health-related shame:

> …the disadvantage sometimes accruing to those regarded as shameful through stigmatization is more often than not mixed in with, even secondary to, exploitation and oppression. It is empirically rare for an individual to be *simply* stigmatized *or* exploited *or* oppressed…. (p. 292)

An additional finding of this study was that symptom severity was significantly and positively related to experiences of illness invalidation in the form of discounting. These results may reflect trends found in other research, indicating that chronicity, ambiguity, and invisibility of symptoms may be particularly related to illness invalidation (Dickson, Knussen, & Flowers, 2007; Johnson & Johnson, 2006; Kool, Middendorp, Boeije, & Geenen, 2009). This relationship between symptom severity and illness invalidation in the form of discounting is likely to produce particular difficulties for persons needing to negotiate necessary illness-related adjustments in work, familial, or social roles. As Chrisman (1977) noted, "Illness-related shifts in role behavior imply a 'bargaining' process in which modified rights and obligations are established with others in the social environment" (p. 357). The lack of visible symptoms in ME/CFS may create increased difficulties for patients who are attempting to maintain modifications to their previous roles, while experiencing discounting of their illness experience from the environment.

In keeping with other studies involving individuals with ME/CFS, the overwhelming majority of participants in this study were female, supporting the idea that females appear to be at greater risk of developing ME/CFS. Previous research has

indicated that females in the United States are at least two to three times more likely to experience ME/CFS symptoms than males (e.g., Bierl et al., 2004; Furberg et al., 2005; Jason et al., 2009), and females constituted the majority of participants in large studies in Sweden (Sullivan et al., 2005) and Iceland (Lindal et al., 2002), as well.

A valuable question that might be raised pertains to whether a link exists between the gendered nature of ME/CFS and its association with illness invalidation. Mik-Meyer (2011) noted the gender biases in medical care as follows:

> In this case a man's illness story appears normal whereas a woman's illness story is an exotic mystery. The thinking seems to be that she might not, like ill men, have the pain she describes. (p. 35)

Indeed, consideration of the gendered nature of ME/CFS, as well the prolific participant reports of illness invalidation across multiple qualitative research studies, raise important questions as to the political role of gender in association with this illness.

The majority of participants in this study identified as Caucasian, which appears consistent with previous research indicating that ME/CFS symptoms have been found to occur at a significantly higher rate in Caucasians than in individuals of other racial backgrounds (Bierl et al., 2004). Indeed, CDC prevalence results indicated that 98% of individuals with ME/CFS were Caucasian (CDC, 1997).

The current study makes a meaningful contribution to the scholarly literature, in that the construct of illness invalidation is relatively new to consideration in health psychology, and little quantitative research has been conducted in this area. It appears that, to date, there have been no peer-reviewed quantitative studies published that evaluate experiences of illness invalidation in the ME/CFS population.

An additional beneficial outcome of the current study was that the Illness Invalidation Inventory (Kool et al., 2010) was found to be a reliable measure for use with individuals with ME/CFS. Use of the I*3 had previously been limited to populations having rheumatic disorders. However, the results of the current study suggest that the instrument may be utilized in research with individuals who have ME/CFS, as well.

Limitations

Limitations of the current study include use of a convenience snowball sampling method to recruit participants. While a strength of this method is that it allowed for recruitment from a wide pool of individuals with ME/CFS, including those who may not currently participate in medical or counseling services, use of this method limits the generalizability of results. Additionally, posts of the survey link to internet sites sometimes prompted online discussion of the study by participants, and it is possible that such discussions may have biased responses of subsequent participants.

Additionally, it is possible that participant response bias played a role in the current results. Although the I*3 contains both positively and reverse-scored items, all positively-scored items load on the Discounting subscale, while all reverse-scored items load on the Lack of Understanding scale. Given that only the Lack of Understanding subscale correlated with SSGS Shame, it is conceivable that systematic response bias contributed to the results.

Another potential limitation is that individuals with more severe or limiting symptoms may have elected not to begin the survey. For example, individuals with more severe or limiting symptoms may have been less likely to participate in the survey, due to experiences of malaise, debilitating fatigue, or cognitive fog. Thus, there

may be a subset of the ME/CFS population whose experiences are not adequately reflected in this study.

Recommendations for Future Research

Future online research involving individuals with ME/CFS should include demographic questions regarding country of residence, in order to better delineate the generalizability of results. Given that lack of understanding evidenced a significant correlation with state shame, while discounting did not, future research should focus on the specific components of illness invalidation that may contribute to emotional distress for individuals with ME/CFS.

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Appendix A

Tables

Table 1

Demographic Variables

Variable	*M*	*SD*	*Range*	*%*	*N*
Age	45.5	11.12	18-64	--	137
Gender					
Female	--	--	--	68.1	126
Male	--	--	--	6.5	12
Other	--	--	--	.5	1
No Response	--	--	--	24.9	46
Race/Ethnic Identity					
Caucasian	--	--	--	64.9	120
Native American/ American Indian	--	--	--	0.5	1
African Origin	--	--	--	0.5	1
Asian Origin	--	--	--	0.5	1
Other	--	--	--	8.1	15
No Response	--	--	--	25.4	47
Marital Status					
Married --	--	--	37.3	69	
Single	--	--	--	18.4	34
Divorced	--	--	--	11.4	21
Widowed	--	--	--	.5	1
Other	--	--	--	7.6	14
No Response	--	--	--	24.9	46
Education					
College Degree --	--	--	45.5	84	
Some College	--	--	--	14.1	26
Vocational Training	--	--	--	7.6	14
High School	--	--	--	10.3	19
Junior High/ Middle School	--	--	--	1.6	3
Other	--	--	--	3.2	6
No Response	--	--	--	17.3	32

Table 2

Reliability, Means, and Standard Deviations Table for Variables

Variable	α	*M*	*SD*	*N*
1. I*3 LOU	.88	3.19	.64	159
2. I*3 Discounting	.85	2.94	.75	159
3. SSGS Shame	.80	11.83	4.97	138
4. MOS OSI	.96	2.94	.91	144
5. MOS Emot/Inform	.93	2.83	.97	144
6. MOS Affectionate	.92	3.43	1.25	144
7. MOS PSI	.89	2.93	.99	144
8. MOS Tangible	.87	2.81	1.11	144

Note. I*3 LOU = Illness Invalidation Inventory, Lack of Understanding Subscale – higher scores suggest higher levels of perceived illness invalidation in the form of lack of understanding. I*3 Discounting = Illness Invalidation Inventory, Discounting Subscale – higher scores suggest higher levels of perceived illness invalidation in the form of discounting. SSGS Shame = State Shame and Guilt Scale, Shame Subscale – higher scores suggest higher levels of state shame. MOS OSI = Medical Outcomes Study Social Support Survey, Overall Support Index – higher scores suggest higher levels of perceived overall social support. MOS Emot/Inform = Medical Outcomes Study Social Support Survey, Emotional/ Informational Support Subscale – higher scores suggest higher levels of perceived emotional/ informational social support. MOS Affectionate = Medical Outcomes Study Social Support Survey, Affectional Support Subscale – higher scores suggest higher levels of perceived affectionate social support. MOS PSI = Medical Outcomes Study Social Support Survey, Positive Social Interaction Subscale – higher scores suggest higher levels of perceived positive social interactions. MOS Tangible = Medical Outcomes Study Social Support Survey, Tangible Support subscale – higher scores suggest higher levels of perceived tangible social support.

Table 3

Pearson's Bivariate Correlations of Variables

Variable	1	2	3	4	5	6	7	8	9	10
1. Income	---	.07	-.19*	.07	.00	.26**	.21*	.27**	.33**	.17*
2. Time Symptomatic		---	-.20*	.13	.04	.00	.03	-.02	-.01	-.05
3. SSGS Shame			---	.11	.15*	-.17*	-.17*	-.18*	-.20*	-.01
4. I*3 Discounting				---	.61**	--.41**	-.40**	-.43**	-.23**	-.35**
5. I*3 Lack of Understanding					---	-.49**	-.52**	.53**	-.305**	-.369**
6. MOS Overall Support Index						---	.93**	.86**	.82**	.822**
7. MOS Emotional/Informational							---	.73**	.68**	.71**
8. MOS Affectionate								---	.75**	.60**
9. MOS Positive Social Interaction									---	.54**
10. MOS Tangible										---

Note. Income = Annual Income; Time Symptomatic = Length of time since symptom onset; SSGS Shame = State Shame and Guilt Scale, Shame Subscale – higher scores suggest higher levels of state shame. I*3 Discounting = Illness Invalidation Inventory, Discounting Subscale – higher scores suggest higher levels of perceived illness invalidation in the form of discounting. I*3 Lack of Understanding = Illness Invalidation Inventory, Lack of Understanding Subscale – higher scores suggest higher levels of perceived illness invalidation in the form of lack of understanding. MOS, Overall Support Index = Medical Outcomes Study Social Support Survey, Overall Support Index – higher scores suggest higher levels of perceived overall social support. MOS Emotional/Informational = Medical Outcomes Study Social Support Survey, Emotional/Informational Support Subscale – higher scores suggest higher levels of perceived emotional/informational social support. MOS Affectionate = Medical Outcomes Study Social Support Survey, Affectional Support Subscale – higher scores suggest higher levels of perceived affectionate social support. MOS Positive Social Interaction = Medical Outcomes Study Social Support Survey, Positive Social Interaction Subscale – higher scores suggest higher levels of perceived positive social interactions. MOS Tangible = Medical Outcomes Study Social Support Survey, Tangible Support subscale – higher scores suggest higher levels of perceived tangible social support. *$p < .05$. **$p < .01$.

Table 4

Hierarchical Multiple Regression Analysis for Variables

IV	Step	R^2	ΔR^2	*F* Change	*df*	*B*	*SE B*	ß
Income	1	.085	.068	5.128**	(2, 111)	-.066	.036	-.169
Symptom Duration	1	.085	.068	5.128**	(2, 111)	-.061	.024	-.228**
I*3 LOU	2	.097	.073	1.546	(1, 110)	.012	.010	.114
MOS OSI	3	.105	.072	.982	(1, 109)	-.006	.006	-.096
I*3 LOU X MOS OSI	4	.106	.065	.100	(1, 108)	.000	.001	.753

Note. Income = Annual Income; Symptom Duration = Length of time since symptom onset; I*3 LOU = Illness Invalidation Inventory, Lack of Understanding; SSGS Shame = State Shame and Guilt Scale, Shame Subscale. MOS OSI = Medical Outcomes Study Social Support Survey, Overall Support Index.

**$p < .01$.

Appendix B

IRB Approval Letter

The UNIVERSITY of OKLAHOMA

Institutional Review Board for the Protection of Human Subjects

Initial Submission – Exemption Approval

Date: March 24, 2015

IRB#: 5231
Approval Date: 03/23/2015

To: Denise Beesley, PhD

Study Title: Impact of Social Support on the Relationship between Illness Invalidation and Shame Among Sufferers of Myalgic Encephalomyelitis/Chronic Fatigue Syndrome

Collection/Use of PHI: Yes

Exempt Criteria: Exempt Category 2

On behalf of the Institutional Review Board (IRB), I have reviewed the above-referenced research study and determined that it meets the criteria for exemption from IRB review. Study documents approved for this submission are located on page 2 of this letter. To access the study submission form and study documents approved for this submission, open this study from the *My Studies* option, click to open this study, under *Protocol Items*, click to open/access the current approved *Application*, *Informed Consent*, or *Other Study Documents*.

If this study required routing through the Office of Research Administration (ORA), you may *not begin your study yet*, as per OUHSC Institutional policy, until the contract through ORA is finalized and signed.

As principal investigator of this research study, you are responsible to:

- Conduct the research study in a manner consistent with the requirements of the IRB and federal regulations 45 CFR 46 and/or 21 CFR 50 and 56.
- Request approval from the IRB prior to implementing any/all modifications as changes could affect the exempt status determination.
- Maintain accurate and complete study records for evaluation by the HRPP Quality Improvement Program and, if applicable, inspection by regulatory agencies and/or the study sponsor.

In addition, it is your responsibility to obtain informed consent and research privacy authorization using the currently approved, stamped forms and retain all original, signed forms, if applicable.

If you have questions about this notification or using iRIS, contact the IRB @ 405-271-2045 or irb@ouhsc.edu.

Sincerely,

Karen Beckman, MD
Chairperson, Institutional Review Board

1000 Stanton L Young Blvd, LIB 176, Okla City, OK 73117 (FWA 00007961)

Study documents associated with this submission:

Study Document			
Title	**Version Number**	**Version Date**	**Outcome**
De-identified PHI Form	Version 1.0	03/09/2015	Approved
Permission to use the MOS Social Support Inventory	Version 1.0	02/15/2015	Noted
Permission to use Illness Invalidation Inventory	Version 1.0	02/15/2015	Noted
State Shame and Guilt Scale	Version 1.0	02/15/2015	Approved
MOS Social Support Survey	Version 1.0	02/15/2015	Approved
Illness Invalidation Inventory	Version 1.0	02/15/2015	Approved
Recruitment Script for Web Sites	Version 1.0	02/15/2015	Approved
Protocol	Version 1.0	02/15/2015	Approved
Script for contacting administrators	Version 1.1	02/15/2015	Approved

Study Consent Form			
Title	**Version Number**	**Version Date**	**Outcome**
Informed Consent	Version 1.3	02/15/2015	Approved

Appendix C

Instruments

Demographic Questionnaire

1. Do you suffer from chronic fatigue syndrome/myalgic encephalomyelitis?
   a. Yes b. No

   [If No, proceed to Exit Page. If Yes, proceed to Question 2.]

Page 1

In order to complete this study, I would like to know more about you. The information that you provide will not be used to identify you in any way.

2. Gender: a. Female b. Male c. Other

3. Age: *[drop-down with ages from 18-64]*

4. What is the highest level of education that you have **completed**?
   a. Junior high/middle school
   b. High school
   c. Some college
   d. Vocational training
   e. Associate's degree
   f. Bachelor's degree or equivalent professional degree
   g. Master's degree or equivalent professional degree
   h. Doctorate degree or equivalent professional degree
   i. Other (please specify): _____

5. Marital Status: a. Single b. Married c. Divorced d. Widowed e. Other: _____

6. How would you describe your race/ethnicity?
   a. African origin
   b. American Indian/Native American
   c. Asian origin
   d. Biracial or Multiracial

e. Caribbean origin
f. Caucasian
g. Latino/a
h. Middle Eastern origin
i. Native Hawaiian or Pacific Islander origin
j. Other (please specify) _____

7. What is your annual household income?
   a. Under $20,000
   b. $20,000-$29,000
   c. $30,000-39,000
   d. $40,000-$49,000
   e. $50,000-$59,000
   f. $60,000-$69,000
   g. $70,000-$79,000
   h. $80,000-$89,000
   i. $90,000-$99,000
   j. $100,000 +

8. Do you have children?
   [If Yes, proceed to Question 9. If No, proceed to Question 11.]

Page 2

9. How many children do you have? *[drop-down with numbers 1-10+]*

10. How many of your children live with you? *[drop-down with numbers 1-10+]*

Page 3

11. Are you disabled? a. Yes b. No
    [If Yes, proceed to Question 12. If No, proceed to Question 13.]

Page 4

12. What diagnosis or diagnoses led to your disability status?
    _____

Page 5

Please answer the following questions about your experiences with chronic fatigue/myalgic encephalomyelitis (ME/CFS).

13. How long has it been since you first began suffering from symptoms of chronic fatigue syndrome/ myalgic encephalomyelitis?
    a. Less than 1 year
    b. 1 year
    c. 2 years
    d. 3 years
    e. 4 years
    f. 5 years
    g. 6 years
    h. 7 years
    i. 8 years
    j. 9 years
    k. 10 years
    l. 11-20 years
    m. 30+ years

14. Were you diagnosed with chronic fatigue syndrome and/or myalgic encephalomyelitis by a medical care provider?
    a. Yes b. No

[If Yes, proceed to Question 15. If No, proceed to Question 18.]

Page 6

15. Who diagnosed you with chronic fatigue syndrome/ myalgic encephalomyelitis?
    a. General medical doctor/Family medical doctor
    b. Medical doctor who is a specialist in a specific area of medicine
    c. Physician's assistant
    d. Nurse practitioner
    e. Other (please specify) _____

    [If "Medical doctor who is a specialist in a specific area of medicine," proceed to Question 16. If any other response, proceed to Question 17.]

Page 7

16. What was the specific specialty area of the medical doctor who diagnosed you with CFS/ME? _____

Page 8

17. How long did you suffer from the symptoms of CFS/ME before receiving a diagnosis? (Please list in years and months) _____

18. Are you currently under the care of a medical doctor who is a specialist in a specific area of medicine?

    a. Yes
    b. No

    [If "Yes," proceed to Question 19. If "No," proceed to Question 21.]

Page 9

19. What was the length of time from the onset of your symptoms to your receiving care from a medical doctor who is a specialist? (Please list in years and months) _____

20. What is the specific specialty area of the medical doctor who is currently providing care for your ME/CFS? _____

Page 10

21. How limited are you by your ME/CFS symptoms? (Note: 0 indicates absence of limitations, and increasingly higher numbers indicate increased limitations by symptoms)

    [response on slider from 0-100]

22. How would you rate the severity of your current ME/CFS symptoms? (Note: 0 indicates absence of symptoms, and increasingly higher numbers indicate increased severity of symptoms)

    [response on slider from 0-100]

23. How would you describe your current symptom status?

    a. Intense symptoms and/or many problematic symptoms
    b. Moderate symptoms
    c. Mild symptoms
    d. Symptoms in remission

24. What have you found most helpful in managing your physical symptoms? _____

25. What have you found most helpful in emotionally coping with your illness? _____

26. If you were to give advice to someone else with ME/CFS, what advice would you share? _____

27. Are you involved in a support group for individuals with ME/CFS?

    a. Yes
    b. No

    [If "Yes," proceed to Question 28. If "No," proceed to Question 29.]

28. Please describe the type of support group in which you are involved. _____

Illness Invalidation Inventory (3*I)

We are interested in how others react to people who have health problems or an illness. Each of the sections below refers to different people in your life. We would like you to rate **how often during the past year** each person or category of people reacted toward you in the way described. After each statement, circle the number between 1 (never) and 5 (very often) to indicate how often they reacted toward you that way.

The questionnaire has five sections, and you will rate the same reactions a number of times, but referring to different people. If a particular section does not apply to you, you may skip that part of the questionnaire and go on to the next section. Remember, rate the items with respect to how others reacted toward you **as a person who has health problems or an illness.**

Section 1: Spouse or partner

If you are single (not married, a widow/widower, or without a steady partner) then skip Section 1 and go directly to Section 2.

My spouse or partner……	**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
1.1 ….finds it odd that I can do much more on some days than on other days.	1	2	3	4	5
1.2 ….thinks I should be tougher.	1	2	3	4	5

1.3	….takes me seriously.	1	2	3	4	5
1.4	….gives me unhelpful advice.	1	2	3	4	5
1.5	….understands the consequences of my health problems or illness.	1	2	3	4	5
1.6	….makes me feel like I am an exaggerator.	1	2	3	4	5
1.7	….thinks I can work more than I do.	1	2	3	4	5
1.8	….gives me the chance to talk about what is on my mind.	1	2	3	4	5

Section 2: Family

For example, children, parents, brothers, sisters, uncles, aunts, grandparents, in-laws.

My family……		**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
2.1	….finds it odd that I can do much more on some days than on other days.	1	2	3	4	5
2.2	….thinks I should be tougher.	1	2	3	4	5
2.3	….takes me seriously.	1	2	3	4	5
2.4	….gives me unhelpful advice.	1	2	3	4	5
2.5	….understands the consequences of my health problems or illness.	1	2	3	4	5
2.6	….makes me feel like I am an exaggerator.	1	2	3	4	5
2.7	….thinks I can work more than I do.	1	2	3	4	5
2.8	….gives me the chance to talk about what is on my mind.	1	2	3	4	5

Section 3: Medical professionals

For example, your primary care physician, medical specialist, physical therapist, and other medical professionals. (Do not include your employer's company physician).

Medical professionals	**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
3.1 ….find it odd that I can do much more on some days than on other days.	1	2	3	4	5
3.2 ….think I should be tougher.	1	2	3	4	5
3.3 ….take me seriously.	1	2	3	4	5
3.4 ….give me unhelpful advice.	1	2	3	4	5
3.5 ….understand the consequences of my health problems or illness.	1	2	3	4	5
3.6 ….make me feel like I am an exaggerator.	1	2	3	4	5
3.7 ….think I can work more than I do.	1	2	3	4	5
3.8 ….give me the chance to talk about what is on my mind.	1	2	3	4	5

Section 4: Work environment

For example, your co-workers and boss. (Do not include your employer's company physician).

If you did not have paid or unpaid employment in the past year, then skip this Section and go directly to Section 5.

People at work…….	**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
4.1 ….find it odd that I can do much more on some days than on other days.	1	2	3	4	5
4.2 ….think I should be tougher.	1	2	3	4	5
4.3 ….take me seriously.	1	2	3	4	5
4.4 ….give me unhelpful advice.	1	2	3	4	5
4.5 ….understand the consequences of my health problems or illness.	1	2	3	4	5
4.6 ….make me feel like I am an exaggerator.	1	2	3	4	5
4.7 ….think I can work more than I do.	1	2	3	4	5
4.8 ….give me the chance to talk about what is on my mind.	1	2	3	4	5

Section 5 : Social services

For example, your employer's company physician, work-reintegration or vocational rehabilitation staff, unemployment and other government agencies, organizations for care at home, general government workers and health insurance companies.

If you did not have any interactions with these providers, you may skip this Section.

People in social services…….	**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
5.1 ….find it odd that I can do much more on some days than on other days.	1	2	3	4	5
5.2 ….think I should be tougher.	1	2	3	4	5
5.3 ….take me seriously.	1	2	3	4	5
5.4 ….give me unhelpful advice.	1	2	3	4	5
5.5 ….understand the consequences of my health problems or illness.	1	2	3	4	5
5.6 ….make me feel like I am an exaggerator.	1	2	3	4	5
5.7 ….think I can work more than I do.	1	2	3	4	5
5.8 ….give me the chance to talk about what is on my mind.	1	2	3	4	5

The State Shame and Guilt Scale

(Marschall, Sanftner, & Tangney, 1994)

The following are some statements which may or may not describe how you are feeling **right now**. Please rate each statement using the 5-point scale below. Remember to rate each statement based on how you are feeling **right at this moment**.

	Not feeling this way at all	Feeling this way somewhat	Feeling this way very strongly
1. I feel good about myself.	1 ------- 2 ------- 3 ------- 4 ------- 5		
2. I want to sink into the floor and disappear.	1 ------- 2 ------- 3 ------- 4 ------- 5		
3. I feel remorse, regret.	1 ------- 2 ------- 3 ------- 4 ------- 5		
4. I feel worthwhile, valuable.	1 ------- 2 ------- 3 ------- 4 ------- 5		
5. I feel small.	1 ------- 2 ------- 3 ------- 4 ------- 5		
6. I feel tension about something I have done.	1 ------- 2 ------- 3 ------- 4 ------- 5		
7. I feel capable, useful.	1 ------- 2 ------- 3 ------- 4 ------- 5		
8. I feel like I am a bad person.	1 ------- 2 ------- 3 ------- 4 ------- 5		

9. I cannot stop thinking about something bad I have done. 1 ------- 2 ------- 3 ------- 4 ------- 5

10. I feel proud. 1 ------- 2 ------- 3 ------- 4 ------- 5

11. I feel humiliated, disgraced. 1 ------- 2 ------- 3 ------- 4 ------- 5

12. I feel like apologizing, confessing. 1 ------- 2 ------- 3 ------- 4 ------- 5

13. I feel pleased about something I have done. 1 ------- 2 ------- 3 ------- 4 ------- 5

14. I feel worthless, powerless. 1 ------- 2 ------- 3 ------- 4 ------- 5

15. I feel bad about something I have done. 1 ------- 2 ------- 3 ------- 4 ------- 5

Medical Outcomes Study Social Support Survey

(Sherbourne & Stewart, 1991)

Next are some questions about the support that is available to you.

People sometimes look to others for companionship, assistance, or other types of support. How often is each of the following kinds of support available to you if you need it?

None of the time	A little of the time	Some of the time	Most of the time	All of the time
1	2	3	4	5

1) Someone to help you if you were confined to bed….
2) Someone you can count on to listen to you when you need to talk….
3) Someone to give you good advice about a crisis….
4) Someone to take you to the doctor if you needed it….
5) Someone who shows you love and affection….
6) Someone to have a good time with….
7) Someone to give you information to help you understand a situation….
8) Someone to confide in or talk to about yourself or your problems….
9) Someone who hugs you….
10) Someone to get together with for relaxation….

11) Someone to prepare your meals if you were unable to do it yourself….

12) Someone whose advice you really want….

13) Someone to do things with to help you get your mind off things….

14) Someone to help with daily chores if you were sick….

15) Someone to share your most private worries and fears with….

16) Someone to turn to for suggestions about how to deal with a personal problem….

17) Someone to do something enjoyable with….

18) Someone who understands your problems….

19) Someone to love and make you feel wanted….

Appendix D

Prospectus

Impact of Social Support on the Relationship between Illness Invalidation and Shame Among Sufferers of Myalgic Encephalomyelitis/Chronic Fatigue Syndrome

Elizabeth Kendrick, M.S.

University of Oklahoma

Abstract

The current study will examine the relationship between illness invalidation and shame in a sample of adults with myalgic encephalomyelitis/chronic fatigue syndrome. It is expected that this relationship will be moderated by perceived social support, particularly in the form of esteem support. Results are expected to support the Stress Buffering Model.

Chapter One

Introduction

Myalgic encephalomyelitis (ME), also known as chronic fatigue syndrome (CFS), is a complex and poorly understood disorder that affects an estimated 2.2 million Americans (Bierl et al., 2004). ME/CFS appears to involve dysregulation of the immune system (e.g., Gaab et al., 2005; Panerai et al., 2002; White et al., 2010), the endocrine system (e.g., Cleare, Blaire, Chambers, & Wessely, 2001; White, Light, Hughen, VanHaitsma, & Light, 2012), the neuroendocrine system (e.g., Antoni, et al., 2010; Maloney, Boneva, Nater, & Reeves, 2009), and the central nervous system (e.g., de Lange et al., 2004; Siemionow, Fang, Calabrese, Sahgal, & Yue, 2004). Patients experience debilitating fatigue, sleep dysfunction, post-exertional malaise, slowed cognitive processing, hypersensitivity to sensory stimulation, and other symptoms (Carruthers et al., 2003; Fukuda et al., 1994).

Despite physiological abnormalities in ME/CFS, the disorder is poorly understood and is frequently met with contest, doubt, and suspicion from medical and lay persons (Bayliss et al., 2014; Travers & Lawler, 2008). Individuals with ME/CFS have reported experiencing illness invalidation by having their illness experiences discounted, minimized, or dismissed by medical professionals and significant others (Johnson & Johnson, 2006; Larun & Malterud, 2007). Further, individuals with ME/CFS have reported feelings of shame associated with invalidation of their symptoms (Larun & Malterud, 2007). The Stress Buffering Model, with its proposal that social support counteracts threats to self-concept during periods of elevated stress

(Cohen & Wills, 1985), provides a rationale that social support may play an ameliorative role in reducing shaming effects of illness invalidation.

Despite qualitative studies indicating that individuals with ME/CFS commonly experience illness invalidation (Asbring & Narvanen, 2002; Dickson, Knussen, & Flowers, 2007; Dickson, Knussen, & Flowers, 2008), the construct is relatively new to consideration in health psychology, and little quantitative research has been conducted in this area. Additionally, the majority of research on illness invalidation has been limited to studies of patients with rheumatic diseases (Kool, van Middendrop, Lumley, Bijlsma, & Geenen, 2012). Therefore, this study aims to elucidate the impact of illness invalidation and social support upon shame experiences among ME/CFS sufferers.

Chapter Two

Literature Review

Myalgic Encephalomyelitis/Chronic Fatigue Syndrome

Myalgic Encephalomyelitis has been classified in the World Health Organization's International Classification of Diseases as a neurological disease (Carruthers & van de Sande, 2005). While fatigue is often highlighted in ME/CFS, it is only one of multiple symptoms within the disorder. Individuals with ME/CFS experience post-exertional malaise, slowed cognitive processing, hypersensitivity to sensory stimulation, sleep dysfunction, and other symptoms (Carruthers et al., 2003; Fukuda et al., 1994). Centers for Disease Control and Prevention (CDC; 1997) surveillance results found that 95% of patients reported neurocognitive complaints, 81% reported sleep disturbance, 80% reported fever and/or chills, and 78% reported myalgia. The most frequently reported ME/CFS symptom found in data from the Mid-Atlantic Twin Study (Furberg, Olarte, Afari, Goldberg, Buchwald, & Sullivan, 2005) and in the Wichita, Kansas population-based study was unrefreshing sleep (Nisenbaum, Jones, Unger, Reyes, & Reeves, 2003).

The term *fatigue* does not adequately describe the debilitating exhaustion, malaise, feelings of heaviness, and lightheadedness that individuals with ME/CFS experience (Carruthers & van de Sande, 2005). Markedly different from ordinary fatigue, in which an individual experiences loss of energy that is easily restored by rest, individuals with ME/CFS typically experience at least a 50% reduction in activity levels as a result of incapacitating fatigue. These levels of fatigue may require an individual to become homebound and/or dependent upon others for care.

Research comparing ME/CFS patients with multiple sclerosis (MS) patients found that ME/CFS patients scored higher on measures of fatigue than individuals with MS (Taillefer, Kirmayer, Robbins, & Lasry, 2002). While both groups reported similar scores on physical functioning, ME/CFS patients scored significantly higher on measures of the impact of their illness on their employment and daily activities.

Cognitive fatigue is often a component of ME/CFS as well, with physical or cognitive demands resulting in slowed cognitive processing, decreased coherence, and struggles with retrieving information and/or words (Carruthers & van de Sande, 2005). This cognitive fatigue is often referred to as *cognitive fog*, exhibited by impaired concentration, difficulties sustaining attention, forgetfulness, confusion, and slowed reaction time. Processing of complex information may be impaired, and during incidences of over-fatigue, individuals may display dyslexia.

Individuals with ME/CFS also report experiencing hypersensitivity to sensory stimuli within their environment (Carruthers & van de Sande, 2005). Sounds, lights, temperature extremes, and other sensory experiences may become overwhelming. When multiple sources of sensory input are present (e.g., concurrent visual and auditory stimuli; physical activity combined with cognitive activity; unclear situations or hurried environments), patients may experience difficulties with focus. Emotional overload may also result in a "crash," in which the individual becomes debilitated by fatigue and has difficulty recovering.

Debate over the Diagnostic Label

The label *chronic fatigue syndrome* has been commonly associated with this disorder; however, patient advocate groups believed that use of *fatigue* in the label

emphasizes only one dimension of a complex disorder (Carruthers et al., 2011) and contributes to public perception that the disorder is entirely psychological in nature (Arnett & Clark, 2012). As research has begun to increasingly elucidate the physiological pathology underlying the disorder, it has been considered more appropriate and accurate to call the illness *myalgic encephalomyelitis* (Carruthers et al., 2011). This label is believed to be more consistent with the World Health Organization's classification of the illness as a neurological condition.

The change in the disorder's name has not come without debate, however. Fukuda and colleagues (1994) asserted that no name change should be made, due to the public and medical awareness associated with the existing label and due to confusion that might result from changing the name. Further, Fukuda and colleagues argued that no label change should occur until the etiology and processes of the disorder were better understood. Huibers and Wessely (2006) maintained that *chronic fatigue syndrome* was preferable because this term does not infer etiology and because it allows for a multifactorial explanation.

Others have argued that chronic fatigue syndrome and myalgic encephalomyelitis are separate, but related, disorders, with divergent etiologies (Jason, Helgerson, Torres-Harding, Carrico, & Taylor, 2003; Maes, Twisk, & Johnson, 2012). Some research indicated that post-exertional malaise and markers of inflammatory/ infectious processes differentiated ME from CFS, despite their shared characteristics (Maes, Twisk, & Johnson, 2012). As such, the argument has been made that the two diagnoses should not be subsumed under one label, but should be distinguished from

each other. Furthermore, Jason et al. (2003) posited that ME/CFS may, in fact, represent a cluster of distinct disease processes that have been inappropriately grouped together.

Given the current debate over the diagnostic label, many authors prefer to simply combine the labels into one (e.g., ME/CFS; Arnett & Clark, 2012). In the current study the disorder will be referred to as *ME/CFS.*

Etiology

The etiology of ME/CFS continues to be poorly-understood and is likely complex (Bierl et al., 2004). Existing research suggests the involvement of inflammatory processes (Fulle et al., 2000; Maes, Mihaylova, Kubera, & Bosmans, 2007; Pall & Satterlee, 2001; Richards, Roberts, McGregor, Dunstan, & Butt, 2000), alterations in gene expression (Light, , 2011), immune dysfunction (Maes, Twisk, Kubera, & Ringel, 2012; Masuda et al., 2002), involvement of the hypothalamic-pituitary-adrenal (HPA; Cleare, 2004; Johnson & DeLuca, 2005; Van Den Eede, Moorkens, Van Houdenhove, Cosyns, & Claes, 2007), and altered levels of progesterone (Pearson Murphy, Abbott, Allison, Watts, & Ghadirian, 2004).

Inflammatory processes. In the 2000s, indications began to arise from research that inflammatory processes were at play in ME/CFS (Arnett & Clark, 2012). For example, studies found elevated oxidative stress (Fulle et al., 2000; Pall & Satterlee, 2001; Richards et al., 2000) and decreased antioxidant power (Jason et al., 2011) in patients with ME/CFS. *Oxidative stress* occurs when there is a disruption in the equilibrium between the production of reactive oxygen intermediates (commonly known as *free radicals*) and antioxidant defenses, such as cellular production of enzymes that detoxify free radicals and repair the damage they have incurred

(Betteridge, 2000; Storz & Imlay, 1999). Oxidative stress results in a state of chronic inflammation (Jason et al., 2011). Furthermore, oxidative stress and reduced antioxidant capability have been found disruptive to the hypothalamic-pituitary-adrenal (HPA) axis (Epel et al., 2004).

Research has found that elderly individuals with ME/CFS experienced increased histone deacetylase-2 (HDAC-2) activity (Jason et al., 2011). Elevated HDAC-2 activity is typically associated with neuronal death stemming from oxidative stress, and HDAC-2 elevations are found in many neurological disorders. On the other hand, decreased HDAC-2 activity protects against neuronal death induced by oxidative stress.

Alterations in gene expression. Research that evaluated differences in gene expression between individuals with ME/CFS and controls, following moderate exercise, found alterations in gene expression among ME/CFS patients, following physical activity (Light et al., 2011). Prior to exercise, gene expression did not differentiate patients from controls, but following exercise, ME/CFS patients evidenced larger increases in mRNA for seven genes. These dysregulated genes are directly implicated in the signaling of fatigue, as well as being involved in immunological activity, cellular energy, and the cardiovascular system. Patients with the highest illness severity evidenced the greatest increase in gene expression, and those with the lowest severity displayed the least increase in gene expression. The mRNA increases in patients were apparent within 30 minutes of engaging in exercise and remained evident for at least 48 hours. Even the most functional ME/CFS patients differed from healthy controls in gene expression.

Light and colleagues (2011) proposed that the results "might indicate that upstream transcription factors common to all of these genes are dysregulated and control these downstream genes in a pathological fashion" in ME/CFS (p. 77). This suggestion is particularly interesting given that all seven genes whose expression differed in ME/CFS patients share common transcription factors. However, it was alternatively noted that alterations in gene expression might result from viral infections. This is a relevant consideration, given evidence suggesting that ME/CFS often develops following viral infections, such as Epstein-Barr infection (Hickie et al., 2006; Lerner, Beqaj, Deeter, & Fitzgerald, 2004).

Other research found that, compared to controls, ME/CFS patients evidenced significant reduction in the median expression of six genes in isolated CD19+ proteins on B cells (a type of white blood cell; Aspler, Bolshin, Vernon, & Broderick, 2008). The genetic expression of these cells is preferentially up-regulated in healthy individuals. CD19+ proteins on B cells play a role in immune function, and previous research found that CD19+ mutations were associated with antibody-deficiency syndrome (van Zelm et al., 2006). Two CD19+ genes (PTPRK and TSPAN3) that are linked with the development and adhesion of immune cells evidenced the most suppression (Aspler et al., 2008). PTPRK is known to be suppressed by the Epstein-Barr virus (Flavell et al., 2007), a virus linked to the onset of ME/CFS in some patients (Hickie et al., 2006; Lerner et al., 2004). Additionally, the CD14+ monocyte gene set of ME/CFS patients was found to share significant co-expression with CD19+ B cell genes, indicating an immunological interaction consistent with chronic inflammation (Aspler et al., 2008).

Overall, the co-expression of immune system gene sets indicates that patients with ME/CFS experience immune signaling processes consistent with chronic inflammation mechanisms (Aspler et al., 2008). It should be noted, however, that research involving the Swedish Twin Registry found no significant correlation between ME/CFS symptoms and zygosity of twin pairs (Evengard et al., 2005).

Immune dysfunction. Research evidence has also suggested that inflammatory pathways and cell-mediated immunity (CMI) play a key role in the pathology of ME/CFS (Maes et al., 2012). When five immune biomarkers (plasma inerluekin -1, tumor necrosis factor- α, PMN-elastase, serum neopterin, and lysozyme) were evaluated, all immune biomarkers were significantly elevated in ME/CFS patients, when compared to controls. These results suggested that ME/CFS is associated with persistent low-grade inflammation.

The immune biomarker, PMN-elastase, has been found to be significantly increased in ME/CFS, compared to controls (Maes et al., 2012). This is of interest since PMN-elastase regulates inflammatory responses, via a feedback mechanism that may result in cyclical inflammation (Doring et al., 1986). Furthermore, PMN-elastase has been linked to decreased daily functioning and diminished capacity for exercise (Meeus et al., 2008; Nijs & de Meirleir, 2005; Nijs et al., 2005).

Intensive research over the past two decades has evaluated the role of cytokine activity in the pathphysiology of ME/CFS (Arnett & Clarks, 2012). Cytokines are proteins, synthesized by cells, that play a role in the regulation of immunological responses to infection, inflammation, and injuries (Dinarello, 2000; Lyall, Peakman, & Wessely, 2003). Cytokines are manufactured by T cells and other cells of the immune

system, following immunological insult (Lyall et al., 2003). Cytokines, which serve as signals between cells of the immune system and other cells in the body (Lyall et al., 2003), are involved in interactions between the immune system and the central nervous system, and serve to alert the brain of disease (Kapsimalis et al., 2008).

Some cytokines assist with healing (anti-inflammatory), while others increase disease processes (pro-inflammatory; Dinarello, 2000). Additionally, some pro-inflammatory cytokines, such as tumor necrosis factor, trigger manufacture of additional inflammatory cytokines (Chu, 2013). Pro-inflammatory cytokine-mediated illness behaviors consist of symptoms that include fatigue, slowed psychomotor activity, hyperalgesia (increased pain sensitivity), anhedonia, changes in circadian patterns that alter sleep, and depressive symptoms (Dinarello, 2000). When administered to humans, some pro-inflammatory cytokines, such as interleukin (IL)-1 and tumor necrosis factor- α, induce fever, destruction of tissue, inflammation, myalgia, and sometimes shock and death (Dinarello, 2000; Janik et al., 1996).

The majority of cytokine genes are never expressed, unless aversive events trigger the stimulation of their expression (Dinarello, 2000). Examples of cell stressors that may trigger cytokine expression include exposure to ultra-violet light, thermal shock, infection, and inflammation, to name a few. Incidentally, the inflammation that results from pro-inflammatory cytokine mediation can lead to expression of other previously unexpressed cytokine genes, resulting in "a cascade of gene products usually not produced in healthy persons" (p. 504).

Arnett and Clark (2012) presented an excellent literature review of existing research on the inflammatory pathophysiology, neurological responses to inflammation,

cytokine-mediated illness behavior, and potential neuroimmunological interactions in ME/CFS. The authors proposed that ME/CFS results from a pathological variation of pro-inflammatory cytokine-mediated illness behavior comparable to the experiences of fatigue that are found in many other inflammatory disorders. The authors proposed that ME/CFS results from immunological responses that are "inappropriately robust or are of an inappropriate duration," resulting in a symptom picture reflecting a "deranged, maladaptive permutation" of cytokine-mediated illness behavior (p. 304).

Arnett and Clark (2012) also noted that the overlap between inflammatory cytokine-induced illness behaviors and the symptom picture of ME/CFS is pronounced and may point to a shared underlying mechanism. For example, cytokine-mediated illness behaviors may consist of symptoms that include fatigue, slowed psychomotor activity, hyperalgesia (increased pain sensitivity), anhedonia, changes in circadian patterns that alter sleep, and depressive symptoms. These cytokine-mediated illness behaviors share remarkable similarities with salient symptoms of ME/CFS. It is noteworthy that numerous studies appear to support Arnett and Clark's (2012) hypothesis. Other authors have noted that the flu-like malaise that is frequently found in ME/CFS may reflect sickness behaviors induced by the elevated cytokines found in ME/CFS patients (Maes et al., 2012). In fact, some have proposed that consideration should be given to the prospect of treating ME/CFS with anti-inflammatory compounds that target cytokine processes, such as the prescription drug Anikinra (Arnett & Clark, 2012; Maes et al., 2012).

In related research, post-exertional malaise among individuals with ME/CFS was found to be significantly related to inflammatory and cell-mediated immune

biomarkers (Maes, Twisk, & Johnson, 2012). The cytokine network and the neuroendocrine system appear to act together in the regulation of sleep-wake cycles, with their interactions appearing to play a role in the pathogenesis of some sleep difficulties, such as those accompanying illness (Kapsimalis et al., 2008). Multiple studies indicating that sleep disorders are a prominent and problematic component of ME/CFS (e.g., Carruthers et al., 2003; CDC, 1997; Fukuda et al., 1994; Hickie et al., 2009) add support to the premise that cytokine-mediated illness symptoms may be at play in ME/CFS.

Natural killer (NK) cell activity has also been found to be suppressed in individuals with ME/CFS, when compared to healthy controls (Masuda et al., 2002). NK cells serve the immunological function of destroying infectious cells within the body. Compared to controls, ME/CFS patients were found to evidence increased expression of a set of four natural killer (NK) cell genes, with greatest increase in expression of NKG2A/C (Aspler et al., 2008). NKG2A/C plays a role in cytotoxic activities. (Note: *Cytoxicity* refers to destroying of cells, e.g., destruction of infected cells by lymphocytes.). Therefore, these findings were consistent with suppression of cytotoxic activity in ME/CFS. Patients who developed ME/CFS following an identifiable infection (e.g., Epstein-Barr virus infection, cytomegalovirus infection, etc.) had the greatest suppression of NK cell activity (Masuda et al., 2002).

Additionally, elevated nuclear factor κB (NFκB) has been found in ME/CFS (Maes, Mihaylova, & Bosmans, 2007; Maes, Mihaylova, Kubera, & Bosmans, 2007). Nuclear factor κB (NFκB) plays a role in cellular reactions to stress, cytokines, and antigens (Brasier, 2006; Gilmore, 20006). Dysregulation of NFκB has been linked to

inflammatory processes, autoimmune disorders, and faulty immune system development. Increased NFκB prompts manufacture of pro-inflammatory cytokines (Morris & Maes, 2012).

The research linking dysfunctional immunological response to ME/CFS has not been conclusive, however. In a systematic review of ME/CFS studies published from 1996 to 2000, Lyall and colleagues (2003) found that deviations in the number, function, and activation markers of T cells were found in the highest-rated studies, but atypical levels of cytokines and abnormal numbers and functions of NK cells were typically not found in the highest-rated studies. The authors noted, however, that their results did not eliminate the potential that ME/CFS is caused, at least partially, by immune system dysfunction.

HPA-axis and immune function. An increasing accumulation of research evidence supports the hypothesis that dysregulation of the hypothalamic-pituitary-adrenal (HPA) axis response to stress plays an important role in ME/CFS (Cleare, 2004; Johnson & DeLuca, 2005; Van Den Eede et al., 2007). The HPA axis is responsible for regulating the functioning of the endocrine and autonomic nervous systems, as well as multiple pathways of the immune system (McEwen & Stellar, 1993). Research indicates that the HPA axis provides regulation of the immune system through regulation of corticoseteriod secretion and directly involves inflammatory feedback, with pro-inflammatory cytokines modulating the hypothalamus' release of corticotrophin releasing factor (Cupps & Fauci, 1982; Hench, Slocum, Polley, & Kendal, 1950).

Progesterone. Mean levels of progesterone and all of its metabolites have been found to be higher in patients with ME/CFS than in age-matched healthy controls

(Pearson Murphy et al., 2004). Some have suggested that ME/CFS symptoms may stem directly from elevated progesterone levels. Freeman et al. (1993) found that administering micronized progesterone to healthy women resulted in increases in fatigue and confusions, as well as slowing of verbal recall and of ability to copy symbols.

Prevalence

An estimated 2.2 million adults in the United States suffer from ME/CFS symptoms (Bierl et al., 2004). Research by the U.S. Centers for Disease (CDC) found that prevalence among adults over a four-year period varied from 3.8 cases per 100,000 people to 5.2 per 100,000, based on geographical location within the country (CDC, 1997). Data from the U.S. Mid-Atlantic Twin Registry (N = 4,591) found a 2.7% lifetime prevalence for ME/CFS symptoms (Furberg et al., 2005). A stratified community study in Chicago revealed a point-in-time prevalence rate of 2.2% (Jason et al., 1999), while a Wichita, Kansas study found a weighted point prevalence of 235 per 100,000 individuals (Reyes et al., 2003). Prevalence rates for ME/CFS in Iceland were found to be 1.4% (Lindal, Stefansson, & Bergmann, 2002). Natural history research found that rates of ME/CFS remained approximately unchanged over roughly ten years (Jason, Porter, Hunnell, Rademaker, & Richman, 2010).

Demographics

Race. Research on the prevalence of ME/CFS, based on race, has yielded mixed results. ME/CFS symptoms have been found to occur at a significantly higher rate in Caucasians than in individuals of other racial backgrounds (Bierl et al., 2004), with CDC results indicating that 98% of patients were Caucasian (CDC, 1997). A stratified

community study in Chicago, however, found that Native Americans and Latinos had the highest prevalence rates (5.4% and 3.9%, respectively), while Asians and Caucasians had the lowest prevalence rates (1.0% and 1.4%, respectively; Jason et al., 1999).

African Americans and Latinos with chronic fatigue have reported significantly higher levels of fatigue following exertion than Caucasians (Jason et al., 2002). Latinos have also indicated significantly higher scores on cognitive difficulties associated with fatigue than Caucasians.

Gender. Females appear to be at the highest risk of developing ME/CFS. Females in the U.S. have been found to be two to three times more likely to experience ME/CFS symptoms than males (Bierl et al., 2004; Furberg et al., 2005; Jason et al., 2009). Women in Wichita, Kansas experienced 4.5 times higher rates of ME/CFS than males (Reyes et al., 2003). The U.S. CDC (1997) surveillance study found that 85% of individuals with the disorder were female.

Research utilizing data from the Swedish Twin Registry also found that the majority of participants exhibiting symptoms of ME/CFS were female (64.17%; Sullivan et al., 2005). Women in the Swedish Twin Registry were 3.92 times more likely to have a 6-month prevalence of ME/CFS symptoms than men (Evengard, Jacks, Pedersen, & Sullivan, 2005). Further, the majority (78%) of participants with ME/CFS in an Icelandic prevalence study (N = 4,000) were female (Lindal et al., 2002).

Gender differences were found to become more pronounced as definitions of fatigue became more stringent (Furberg et al., 2005) and as symptoms became more severe (Reyes et al., 2003). Females were found to develop significantly more

associated symptoms and at significantly younger ages than males (Furberg et al., 2005). Women were also found to score significantly higher on problems pertaining to memory, concentration, and information-processing than men (Jason et al., 2002).

Explanations for the association between female gender and ME/CFS remain unknown. Some have conjectured that the relationship is likely complex and may include a number of factors, ranging from genetic composition to sociological issues related to being female (Evengard et al., 2005). For example, some have proposed that a risk factor associated with being female may be that employment is often combined with being responsible for the majority of housework and childcare at home, which may lead to increased stress, diminished rest, and increased susceptibility to ME/CFS (Jason et al., 1999).

Age. ME/CFS appears to primarily affect adults, with reported point prevalence for adolescents being only 0.11% (Nijhof et al., 2011). However, research results have been mixed regarding associations between ME/CFS and particular adult age subsets. Some research has found that ME/CFS symptoms were reported at a significantly higher rate among individuals ages 40-69, with the greatest risk occurring between ages 40-49 (Bierl et al., 2004; Jason et al., 1999). Other research has determined that the median age for onset for females was 31, while the median onset age for males was 33.5 (Furberg et al., 2005). CDC (1997) surveillance results indicated that the mean age of onset was 34 years. A weighted point-prevalence study in Wichita, Kansas found that prevalence increased with age among white females, peaking between ages 50-59, and subsequently decreasing in ages 60 to 69 (Reyes et al., 2003). In a study involving

participants from the Swedish Twin Registry, no significant association was found between age and ME/CFS symptoms (Evengard et al., 2005).

Education. Previous research has provided conflicting evidence pertaining to typical levels of education among ME/CFS participants. Some U.S. research found that ME/CFS symptoms were most likely among individuals who had only high school education or less (Bierl et al., 2004; Jason et al., 1999), while other research found that the majority of ME/CFS sufferers (63%) had attained education beyond high school (Nisenbaum et al., 2003). CDC (1997) surveillance results from four U.S. cities (Atlanta, Wichita, Grand Rapids, and Reno) found that the majority of individuals with the disorder had attended college (although not all had completed a degree). Research conducted via the Swedish Twin Registry, however, found no differences in educational attainment, when comparing fatigued and non-fatigued participants (Evengard et al., 2005).

Income. Results have been mixed regarding typical income attainment of individuals with ME/CFS. Some research has found that ME/CFS symptoms have the highest prevalence among individuals with annual incomes of $40,000 or less and that lower education and less income were the strongest predictor of symptoms (Bierl et al., 2004). When compared to individuals from middle and high socioeconomic status (SES), individuals with chronic fatigue who are from lower SES scored significantly higher on measures of cognitive difficulties and of worsened fatigue following physical exertion (Jason et al., 2002). In the CDC (1997) surveillance study, individuals earning $20,000-$50,000 annually composed the largest group (45%) of patients. Community-based research in Wichita, Kansas, on the other hand, found that the highest rates of

ME/CFS were found in individuals earning greater than $40,000 per year (Nisenbaum et al., 2003).

Cultural background. ME/CFS does not appear to be culture-bound. Multivariate analyses of epidemiological and clinical datasets from twenty-one countries having widely-varying cultures (including European and non-European, as well as English- and non-English-speaking) found that symptom profiles for ME/CFS remained consistent, regardless of culture (Hickie et al., 2009). When datasets from three English-speaking countries (the United States, the United Kingdom, and Australia) were utilized, the results were comparable to those found when using the entire dataset. Thus, the ME/CFS symptoms composite does not appear to be culturally- or medically-constructed.

Symptom Domains

In a meta-synthesis of 325 peer-reviewed qualitative studies of ME/CFS, the three symptoms most often reported by participants were severe fatigue, disabling pain, and cognitive difficulties (Anderson, Jason, Hlavaty, Porter, & Cudia, 2012). In contrast, a factor analysis of empirically-derived data from existing international datasets revealed that a five-factor model best explained ME/CFS (Hickie et al., 2009). The five domains were: (a) inflammation, (b) neurocognitive problems, (c) musculoskeletal pain and/or fatigue, (d) disturbed mood, and (e) disrupted sleep and/or fatigue. These factors were evident across cultures and throughout medical settings.

Factor analysis of responses provided by individuals from Wichita, Kansas who had varying levels of ME/CFS symptom manifestations revealed that a three-factor model explained the symptom picture (Nisenbaum et al., 2004). The three factors

included musculoskeletal symptoms, infection symptoms, and cognitive-mood-sleep symptoms.

A random sample of 18,675 individuals from Chicago, Illinois found that 780 (4.2%) reported experiencing chronic fatigue, although severity of fatigue varied (Jason et al., 2002). Factor analysis of participants' responses to questions regarding their fatigue revealed that four factors explained 38% of total variance, namely: (a) lack of energy (weakness, tiredness, and fatigue), (b) physical exertion resulting in worsened fatigue, (c) problematic cognitive functioning (difficulties with concentration, memory, and information-processing), and (d) whether fatigue was relieved by rest.

In data obtained from the Swedish Twin Registry, the most-commonly reported ancillary symptoms were unrefreshing sleep (79.87%), impaired memory/concentration (48.48%), muscle pain (37.26%), and joint pain (37.26%; Sullivan, Pedersen, Jacks & Evengard, 2005). The four least-frequently reported ancillary symptoms were tender lymph nodes (6.89%), headaches of a new type (11.11%), sore throat (11.99%), and post-exertional malaise (15.76%).

Finally, cluster analyses revealed that participants with ME/CFS fall into three differing subcategories of fatigue patterns, making the ME/CFS population a heterogeneous group (Jason et al., 2010). These subcategories are: low, moderate, and severe, with categorization based upon severity of postexertional fatigue, wired fatigue, brain fog, diminished energy, and flu-like fatigue.

Course of Illness

Review of existing literature revealed that the majority of patients with ME/CFS engaged in lifestyles that were active and healthy, prior to becoming ill (Carruthers &

van de Sande, 2005). During the year prior to ME/CFS onset, individuals typically evidenced elevated levels of stress (Masuda, Munemoto, Yamanaka, Takei, & Tei, 2002). Individuals with ME/CFS have also often described experiencing the onset of illness as being comparable to a severe form of influenza, and patients have reported that, when the "flu" did not improve, they experienced emotional distress and believed they had lost their identity (Clarke & James, 2003). In qualitative research, individuals often reported that they were "catapulted from a period of excessive busyness to a period of complete immobility" (Clarke & James, 2003, p. 1390).

Research focusing on illness onset has yielded mixed results. For example, Nisenbaum et al. (2003) found that 75% of participants described the onset of their illness as gradual. The CDC (1997) surveillance study also found that the majority of patients experienced gradual onset of their illness. On the other hand, in qualitative research with participants from Canadian support groups, onset was typically sudden (Clarke, 1999; Clarke & James, 2003).

A meta-synthesis of qualitative studies found that individuals with ME/CFS tended to experience extensive variability in both symptom severity and frequency (Andersen et al., 2012). Significant levels of impairment were reported by the majority (60.54%) of symptomatic individuals in the Swedish Twin Registry (Sullivan et al., 2005). Individuals with ME/CFS who were tracked for up to three years were found to display an illness course characterized by intermittent periods of relapse and remission and by an overall decrease in symptoms over time (Nisenbaum et al., 2003).

Research regarding the prognosis for ME/CFS symptom resolution has also yielded mixed results. CDC (1997) surveillance results found that the mean duration of

illness was slightly over six years. In a longitudinal study, at three-year follow-up, 20-33% of participants continued to meet criteria for ME/CFS, 56.9% were in partial or total remission, 10% had sustained remission, and 23.1% had received substitute diagnoses (Nisenbaum et al., 2003). Higher total number of symptoms and increased fatigue severity scores were negatively related to achieving remission. Having had an illness duration of two years or less was a significant predictor of maintaining sustained remission. Systematic review of literature on the prognosis of ME/CFS found that few patients (6-15%) reported complete symptom resolution (Cairns & Hotopf, 2005; Pheley, Melby, Schenck, Mandel, & Peterson, 1999).

Patients who developed ME/CFS following infection with an identifiable infectious agent (e.g., Epstein-Barr virus, cytomegalovirus, etc.) experienced a shorter illness duration than those who developed ME/CFS in the absence of an identifiable infection (Masuda et al., 2002). Individuals who developed ME/CFS following infection with *Giardia duodenalis* were found to resolve ME/CFS symptoms within three-to-five years (Morch et al., 2013).

Diagnostic Challenges

Medical professionals have reported having an inadequate understanding of ME/CFS (Bayliss et al., 2014). One of the complexities of diagnosing ME/CFS lies in that distinctive laboratory tests are not yet available to identify the disorder, meaning that diagnosis is based upon symptoms, impaired functioning, and exclusion of other disorders that might explain symptoms (Reeves et al., 2003). For example, general practitioners in the United Kingdom were found to exclude the following disorders, prior to diagnosis: thyroid disorders (75%), anemia (73%), mood disorders (47%),

diabetes (38%), Epstein-Barr virus (19%), kidney disorders (17%), and liver disorders (10%; Bowen, Pheby, Charlett, & McNulty, 2005). Such exclusionary bases for diagnosis create difficulties in ascertaining the accuracy of diagnosis.

Quest for Biomarkers. The current quest for an ME/CFS biomarker centers upon inflammatory models of the disorder (Arnett & Clark, 2012). A distinct challenge, however, is that many of the biomarkers of inflammatory processes that appear to be involved in ME/CFS are also found in other inflammatory disorders, making reliance on inflammatory biomarkers for diagnosis problematic. However, recent research by Light and colleagues (2011) has identified changes in the expression of seven specific genes that differentiated a large subgroup of ME/CFS patients from healthy controls with accuracy of .80. It has been suggested that lab tests of these gene expressions could be combined with behavioral assessments to increase the accuracy of ME/CFS diagnosis.

Competing Diagnostic Models. An additional complexity of diagnosis is that various diagnostic criteria for ME/CFS exist, including: the CFS Research Case Definition (Fukuda et al., 1994), the Centers for Disease Control and Prevention Recommendations (Reeves et al., 2003), the Canadian Consensus Document (Carruthers & van de Sande, 2005), and the International Consensus Criteria (Carruthers et al., 2011). Anderson and colleagues (2012) noted that this lack of diagnostic consensus has led to tensions between the various players associated with ME/CFS, with qualitative research indicating that these tensions detrimentally impact patients.

Models of Diagnostic Criteria

As noted previously, multiple diagnostic criteria for ME/CFS exist, and the lack of consensus on criteria has led to dissension within the field (Anderson et al, 2012). Among the diagnostic models mentioned above, the CFS Research Case Definition (Fukuda et al., 1994) diagnostic criteria persists as the most widely-utilized diagnostic criteria (Brown et al., 2013), but debate continues as to the appropriate criteria to be utilized in diagnosing ME/CFS.

CFS Research Case Definition. Fukuda et al. (1994) developed a set of diagnostic guidelines designed to distinguish CFS from neuropsychiatric disorders and from other fatiguing physiological conditions. (Note: Fukuda et al. opposed changing the disorder's name from *CFS* to an alternative label. Therefore, in discussing the views of Fukuda et al., this study will refer to the disorder as *CFS*.). Noting that psychiatric disorders, such as somatoform disorder, major depressive disorder, anxiety disorders, etc., were sometimes confounded with CFS, Fukuda et al. (1994) sought to establish clarity in diagnostic delineation. The primary motivation for establishing the Fukuda et al. (1994) criteria was to increase the validity of research on CFS.

The Fukuda et al. (1994) diagnostic criteria requires a minimum of six consecutive months of sustained or recurrent fatigue that is of new onset, does not stem from exertion, is not resolved by rest, and leads to significant reductions in previous activities. In addition, at least four of the following symptoms must have occurred concurrently throughout those six months: (a) diminished concentration or memory that impacts abilities to function in previous activities; (b) sore throat; (c) tender lymph nodes; (d) pain in muscles; (e) pain in multiple joints without redness or swelling; (f)

headaches that differ in severity, pattern, or types from previous experience; (g) sleep that is not refreshing; and (h) malaise lasting for at least twenty-four hours following exertion. Furthermore, in order to arrive at diagnosis, clinicians must have completed: (a) a comprehensive medical and psychosocial history, (b) a mental status examination, (c) a complete physical examination, and (d) a battery of relevant laboratory screenings. When all other sources of symptoms were excluded, a diagnosis of CFS might be made.

Criticisms of the Fukuda et al. (1994) criteria have centered upon its polythetic approach, meaning that not all symptoms must be present for diagnosis (Jason et al., 2012). Since four of eight symptoms are required for diagnosis, using the Fukuda et al. criteria, individuals may be diagnosed without evidencing symptoms that are considered core to CFS (e.g., post-exertional malaise; Brown et al., 2013).

An additional concern with the Fukuda et al. (1994) criteria has to do with research not justifying the cut-point requiring that four out of eight ancillary symptoms be present (Sullivan et al., 2005). By establishing a cut-point in number of symptoms, the Fukuda et al. (1994) criteria assumed that there were notable differences between patients reporting zero-to-three ancillary symptoms and those reporting four-to-eight (Sullivan et al., 2005). However, research utilizing data from the Swedish Twin Registry found that the diagnostic cut-point of four ancillary symptoms was not clinically-meaningful. Rather than demarcating between normalcy and pathology, no important differences existed between patients reporting three symptoms and those reporting four symptoms.

Yet another critique of the polythetic approach utilized by the Fukuda et al. (1994) criteria surrounds the inherent supposition that “the ancillary symptoms are

fundamentally interchangeable and thus similar in their impact on the probability of CFS" (Sullivan et al., 2005, p. 1344). Data from the Swedish Twin Registry revealed that this assumption was not supported, as prevalence of symptoms reported ranged from ~7% for tender lymph nodes to ~80% for unrefreshing sleep. In fact, the eight ancillary symptoms were not equivalent in their individual ability to predict the disorder.

As a result of research indicating that post-exertional malaise is a component of the CFS symptom picture, the Fukuda et al. (1994) criteria have also been critiqued for failing to include this symptom (Maes, Twisk, & Johnson, 2012). An additional criticism of the Fukuda et al. (1994) criteria is its requirement that fatigue be present for over six months (Brown et al., 2013).

Centers for Disease Control and Prevention Recommendations. The Centers for Disease Control and Prevention (CDC) Recommendations arose out of concerns that the existing CFS Research Case Definition (i.e., Fukuda et al., 1994) contained ambiguities that led to inconsistencies in diagnosis (Reeves et al., 2003). Between May, 2000 and May 2002, the CDC sponsored structured workshops with international ME/CFS experts from the fields of infectious disease, epidemiology, immunology, endocrinology, neurology, psychiatry, psychology, and biostatistics, as well as CDC staff and patient advocates. The purpose of the workshops was to evaluate the existing CFS Research Case Definition (i.e., Fukuda et al., 1994), to review existing scientific research literature, to discuss clinical experiences, and to brainstorm solutions to the existing diagnostic difficulties. From these sessions, summaries were compiled, analyzed, and drafted into formal recommendations. The final document resulted in a

series of recommended modifications to the CFS Research Case Definition presented by Fukuda and colleagues.

Among the CDC diagnostic modifications was the development of a specific list of exclusionary diagnoses, divided into three domains (Reeves et al., 2003). These categories were: (1) permanent medical exclusions (e.g., systemic lupus, Sjogren's syndrome, multiple sclerosis); (2) temporary medical exclusions (e.g., untreated diabetes mellitus, sepsis, morbid obesity); (3) and permanent psychiatric exclusions (e.g., schizophrenia, bipolar disorder, organic brain disorders). The CDC criteria further clarified that short-term psychiatric conditions that had been resolved for over five years should not be considered in exclusionary diagnosis. CDC Recommendations included suggestions of standardized assessments that might be utilized in diagnosis (e.g., the CDC Symptom Checklist).

The CDC Recommendations also noted that the Fukuda et al. (1994) criteria required that fatigue associated with CFS not be related to continued exertion (Reeves et al., 2003). This stipulation presumably sought to distinguish ME/CFS from the normal levels of fatigue experienced by typical persons who engage in excessive activities. However, this Fukuda et al. (1994) requirement created diagnostic difficulties, in that individuals with ME/CFS often do experience extreme fatigue and malaise following activities that were previously tolerated well. Thus, the CDC Recommendations proposed that fatigue should be assessed in light of whether it stemmed from over-exertion that would fatigue a healthy adult.

CDC Recommendations also addressed the Fukuda et al. (1994) criterion that fatigue not be substantially relieved by rest (Reeves et al., 2003). CDC experts noted

that most individuals with ME/CFS do experience some relief from fatigue upon resting, although rest does not bring about the level of pre-illness physiological or cognitive stamina. It was also pointed out that incorporating rest periods into activities is often helpful to some patients with ME/CFS. Therefore, the CDC Recommendations proposed that rest as a therapeutic measure should not be used to exclude an individual from an ME/CFS diagnosis.

Recent research has revealed problems with both the sensitivity and the specificity of the CDC criteria (Jason et al., 2010). In a study involving participants diagnosed with ME/CFS and participants diagnosed with Major Depressive Disorder (MDD), the CDC criteria failed to differentiate between ME/CFS and MDD (Jason, Najar, Porter, & Reh, 2008). In fact, 38% of the individuals previously diagnosed with MDD were inaccurately classified as having ME/CFS, when the CDC criteria were used.

Jason and colleagues (2008) speculated that aspects of the CDC criteria that may have contributed to lack of specificity might include the CDC criteria requiring that symptoms be present for one month (as compared to the six months required by the Fukuda et al. [1994] criteria). This reduction in length of time that symptoms are present might allow some symptoms to be included as part of the ME/CFS diagnosis that were actually a part of a transient physical ailment such as influenza.

Further, the CDC criteria do not differentiate between symptoms critical to ME/CFS diagnosis, such as sleep disturbance, cognitive difficulties, and postexertional malaise, and other less-definitional symptoms (Jason et al., 2008; Jason et al., 2012). Since all symptoms are given equal value in the CDC criteria, "a participant reporting

severe and frequent headaches is given the same value as a participant reporting severe and frequent postexertional malaise" (Jason et al., 2008, p. 6). The CDC criteria has also been criticized as targeting broad areas, such as social functioning, difficulties fulfilling roles, and activity issues (Jason et al., 2012).

The Canadian Consensus Document. The Canadian Consensus Document arose from the efforts of the National Myalgic Encephalomyelitis/Fibromyalgia Action Network of Canada to develop expert consensus on the diagnostic criteria for ME/CFS (Carruthers & van de Sande, 2005). A questionnaire was sent to physicians across Canada, requesting input regarding what they believed to be most helpful in diagnosing ME/CFS patients. Using these responses, a panel of ten ME/CFS experts drafted an illness definition and diagnostic criteria.

The Canadian Consensus Document defined ME/CFS as "an acquired organic, pathophysiological, multi-systemic illness that occurs in both sporadic and epidemic forms" (Carruthers & van de Sande, 2005, p. 1). Diagnostic criteria included:

1) Fatigue - Significant physical and mental fatigue that is unexplained, chronic, and notably reduces activity.
2) Post-Exertional Malaise and/or Fatigue – Excessive reduction in physical or mental stamina; rapid onset of fatigue; and/or, post-exertional malaise, fatigue and/or pain.
3) Sleep Dysfunction – Sleep disturbance or unrefreshing sleep.
4) Pain – Significant myalgia that may be experienced in muscles and joints and that may migrate and/or headaches "of new type, pattern or severity" (p. 2).

5) Neurological/Cognitive Manifestations – Two or more of the following symptoms are present: poor concentration and short-term memory; confusion; disorientation; impaired information processing, retrieval of words, and categorization; disturbances of perception and sensory experiences; and/or, cognitive, sensory, and/or emotional overload.
6) At least one symptom from two of the following:
    a. Autonomic Symptoms – Orthostatic intolerance, lightheadedness, significant pallor, palpitations, neurally-mediated hypotension, postural orthostatic tachycardia syndrome, and/or exertional dyspnea.
    b. Neuroendocrine Symptoms – Repeated episodes of feeling feverish, marked weight change, and/or increased symptom severity associated with stress.
    c. Immune Symptoms – Repeated flu-like symptoms, general malaise, lymph node tenderness, repeated sore throat, and/or new sensitivity to medications, foods, and/or chemicals.
7) In an adult, the illness continues for six or more months (more than three months, if diagnosing a child).

In addition, the Canadian Consensus Document (Carruthers & van de Sande, 2005) requires that (a) symptoms must have begun or become more pronounced after the onset of the disorder, (b) clusters of symptoms may fluctuate and vary over time, and (c) active disorders that might explain the majority of symptoms should be excluded, prior to diagnosis with ME/CFS.

When the CDC criteria were compared to the Canadian Consensus criteria; however, the CDC criteria was not as sensitive in discriminating between those with ME/CFS and those without the disorder (Jason et al., 2012). Symptoms typically considered cardinal traits of ME/CFS, including postexertional malaise and neurocognitive difficulties, did not predict diagnosis in the CDC criteria, but they did predict diagnosis using the Canadian criteria.

International Consensus Criteria. The International Consensus Criteria arose out of significant modifications to the Canadian Consensus Criteria (Carruthers & van de Sande, 2005; Carruthers et al., 2011). In developing the International Consensus Criteria, a panel of researchers, clinicians, educators, and an unaffiliated patient advocate collaborated in drafting, reviewing, and revising the diagnostic criteria. The panel was free of sponsorship and consisted of experts from thirteen countries and from a range of specializations.

The International Consensus Panel was motivated by a desire to develop a set of diagnostic criteria that would better assist in differentiating ME/CFS from other illnesses (Carruthers et al., 2011). In particular, there were concerns that the Fukuda (1994) criteria failed to differentiate ME/CFS from depressive disorders. Additionally, the CDC Criteria were believed to be too broad and inclusive to adequately identify patients with ME/CFS. Further, the panel wished to improve upon the Canadian Consensus Criteria. To that end, the International Consensus Criteria focused on the following changes: (a) reducing the six-month waiting period for diagnosis, (b) replacing the term *fatigue* with *postexertional neuroimmune exhaustion*, and (c) better clarifying and operationalizing symptoms (Carruthers et al., 2011).

Thus, according to the International Consensus Criteria, an individual must meet the following criteria to be diagnosed with ME/CFS: (a) symptoms of postexertional neuroimmune exhaustion; (b) one or more symptom of neurological impairment; (c) one or more symptom of either immune, gastro-intestinal, or genitourinary difficulty; and (d) one or more symptom of impaired energy metabolism or energy transport (Carruthers et al., 2011). Furthermore, to be diagnosed with ME/CFS, symptoms must be so severe that the individual's premorbid activity level is significantly reduced.

Postexertional neuroimmune exhaustion was described by the International Consensus Criteria as a "cardinal feature" of the disorder and was defined as "a pathological inability to produce sufficient energy on demand with prominent symptoms primarily in the neuroimmune region" (Carruthers et al., 2011, p. 329). Manifestations of postexertional neuroimmune exhaustion included: (a) profound and rapid physical and/or cognitive fatigue following exertion (including minimal levels of exertion associated with daily tasks); (b) postexertional exacerbation of symptoms (e.g., malaise, pain, intensification of other symptoms); (c) postexertional exhaustion; (d) prolonged recovery period (i.e., typically greater than 24 hours); and (e) substantial reduction in physical and mental stamina that leads to marked reduction of premorbid activity.

Neurological impairments were divided into four categories: (a) neurocognitive impairments (i.e., impaired information processing and/or loss of short-term memory); (b) experience of pain (headaches and/or noninflammatory pain); (c) disturbances to sleep (i.e., disrupted sleep patterns and/or unrefreshing sleep); and (d) neurosensory, perceptual, and motor disturbances (e.g., impaired visual focus, hypersensitivity to

sensory stimuli, disturbed depth perception, difficulties with coordination; Carruthers et al., 2011).

Immune, gastro-intestinal, and genitourinary impairments consisted of five categories: (a) recurrent or chronic flu-like symptoms that usually activate or become worse with exertion; (b) increased susceptibility to viral illnesses with lengthy recovery; (c) gastro-intestinal tract symptoms (e.g., irritable bowel syndrome, nausea, abdominal discomfort); (d) genitourinary symptoms (e.g., increased urinary frequency or urgency); and (e) sensitivity to chemicals, medications, foods, or smells (Carruthers et al., 2011).

Impairment of energy production/transport included at least one of the following symptoms: (a) cardiovascular (e.g., orthostatic intolerance, palpitations, light-headedness, dizziness); (b) respiratory (e.g., labored breathing, shortness of breath); (c) loss of thermostatic stability (e.g., recurrent sensations of feverishness, episodes of sweating); and (d) intolerance of temperature extremes (Carruthers et al., 2011).

The International Consensus Criteria also clarified levels of impairment, ranging from mild to very severe, noting that symptom severity might fluctuate (Carruthers et al., 2011). Additionally, the criteria provided increased clarification for pediatric diagnosis. Furthermore, the diagnostic criteria allowed for an alternative diagnosis of *atypical myalgic encephalomyelitis* for individuals who experience postexertional neuroimmune exhaustion, but exhibit two fewer symptoms than is required for full diagnosis of ME/CFS.

Research comparing the International Consensus Criteria (Carruthers et al., 2011) with the Fukuda et al. (1994) criteria found that individuals who met the International Consensus Criteria exhibited more pronounced impairment in their

functional abilities and greater numbers of physical, cognitive, and mental difficulties than those diagnosed using the Fukuda et al. criteria (Brown et al., 2013). Further, the Fukuda et al. criteria resulted in a much larger number of diagnoses than the International Consensus Criteria, indicating increased specificity in the International Consensus Criteria.

Finally, Maes, Twisk, and Johnson (2012) expressed that the International Consensus Criteria should be modified to include ME as a subset of CFS. This assertion was based upon research revealing that 45.8% of participants with ME/CFS (according to Fukuda et al. [1994] criteria) could be differentiated by their experience of postexertional malaise and by clinical indications of increased inflammatory/infectious processes.

Difficulties with Obtaining a Diagnosis

Individuals with ME/CFS have often reported that obtaining a diagnosis was an arduous process (Clarke & James, 2003; Cooper, 1997; Ware, 1992). Patients frequently endeavored to find a physician who could recognize, explain, and treat their symptoms (Clark & James, 2003), and they often saw multiple physicians in their quest for a diagnosis (Clarke, 1999; Cooper, 1997). Patients were found to have visited between three and four hospitals, complaining of their symptoms, prior to receiving diagnosis (Masuda et al., 2002). Despite medical examinations, the absence of organic abnormalities often led to lack of diagnosis. Patients frequently went undiagnosed for several years (Cooper, 1997), and many symptomatic individuals were never able to obtain a medical diagnosis (Clark & James, 2003). Individuals often described the

difficulties in obtaining diagnoses from their doctors as reducing them to "rock bottom" and as leading them to pessimism over the future (Cooper, 1997, p. 196).

The likelihood of being formally diagnosed with ME/CFS appears to be based upon individual dynamics, such as access to medical care, willingness to pursue a protracted and vigorous quest for services, determination to obtain a diagnosis, and the attitudes of the treating physicians (Huibers & Wessely, 2006). Even when finally diagnosed, however, many individuals reported that their physicians expressed some doubt about whether ME/CFS was a legitimate disorder (Cooper, 1997).

"It must be in your head." Both health professionals and laypersons tend to view illnesses in a dualistic manner, namely that the source of the disorder is either physiological or psychological (Chalder, 2005). Substantial research suggests that when medical explanations are not readily available, medical professionals may default to psychological explanations (e.g., Nettleton, 2006). Given the current lack of a fully understood medical model of ME/CFS, and given the current absence of biomarker assays, ME/CFS patients are sometimes diagnosed with a psychological explanation for their symptoms (Anderson et al., 2012).

Providing a psychological explanation for illness often implies that the patient is somehow to blame for their difficulties, while providing a physical explanation for difficulties typically does not (Nettleton, O'Malley, Watt, & Duffey, 2004). Furthermore, within the modern realm of biomedicine, psychological explanations are typically devalued as less "legitimate" than those that are organically explained (p. 47). In a meta-synthesis of qualitative studies, a salient theme among the accounts of those exhibiting ME/CFS symptoms was that of experiencing stigmatization by family,

friends, and medical professionals in association with having their illness experience labeled as psychosomatic (Anderson et al., 2012).

Qualitative research has revealed that, for some patients who lack a medical explanation for their symptoms, pragmatic difficulties arise from attributing their symptoms to psychological sources (Nettleton et al., 2004). Namely, patients have reported that medical professionals and family members no longer proffered support, as they believed symptoms were "explained away" (p. 55). Furthermore, the psychological attributions posed difficulties in engaging additional professionals to procure a medical diagnosis, as the psychological explanation became a part of the medical record that followed the patient "like a criminal record" (p. 55). The combination of these challenges made it increasingly difficult for the ill person to seek help.

Dimsdale (2011) pointed out several concerns about applying psychiatric diagnoses to symptom composites that are not yet fully understood or explained by the medical profession. First, the quality of medical evaluations should be considered, as doctors vary in their individual knowledge repertoires, in the thoroughness of their examinations, in the amount of time they spend on each patient's case, and in their ability to synthesize information. Additionally, the current state of medical knowledge may have not yet identified the etiology, underlying processes, or existence of a disorder.

Dimsdale (2011) described this concern as follows:

> …diagnoses are shaped by the state of medical knowledge at the time when the patient is evaluated. One 'sees' what one is prepared to see or understand. If one has no tools for recognizing hepatitis C, for instance, one will not make that diagnosis until very late in the progression of the infection. New diseases are constantly arising, either totally new diseases or, more commonly, disease that have previously not been well understood. (p. 511)

An additional consideration posed by Dimsdale (2011) is that many illnesses present with nonspecific symptoms that are difficult to diagnose until medical technologies, such as bioinstrumentation and assay development, progress to the point of allowing for detection.

In similar fashion, Anderson and colleagues (2012) argued that attributing ME/CFS symptoms to psychosomatic sources is a form of victim blaming, in which patients may be blamed for the lack of inadequate scientific knowledge of ME/CFS. Horton-Salway (2002) described it as the construction of "a narrative associated with *low* accountability for doctors and *high* accountability for patients" (p. 416). Given that women are more likely to suffer from ME/CFS (Bierl et al., 2004; Furberg et al., 2005; Jason et al., 2009), Richman and Jason (2001) argued that the failures of the medical community to identify biological markers for ME/CFS has been used to reinforce social ideas that link female gender to psychiatric explanations of illness.

Furthermore, qualitative research has revealed that the psychological explanation for unexplained illness symptoms was found to contribute to social isolation. Nettleton and colleagues (2004) described this social dilemma, as follows:

> She is in a double bind; if she reveals that she feels 'down' or 'depressed' this would only reinforce those psychological interpretations. She has therefore to work hard at her presentation of self. To get angry, anxious, depressed, unhappy, or suicidal would serve to confirm an identity that she has to work hard to dispel. (p. 56)

The significance of diagnosis to patients. The importance of diagnosis of ME/CFS for patients lies, in part, in the need for health-related shifts in roles and behaviors, as a result of illness symptoms. As Chrisman (1977) noted, "Illness-related shifts in role behavior imply a 'bargaining' process in which modified rights and

obligations are established with others in the social environment" (p. 357). The lack of visible symptoms in ME/CFS creates increased difficulties for patients who are attempting to achieve modifications to their previous roles. Chrisman explained,

> Unambiguous acute symptoms place the individual in the strongest position for attaining the fullest extent of modifications in role behaviors and place upon him the strongest obligations to get well. On the other hand, ambiguous or chronic problems are not nearly so compelling. (p. 357)

Given the ambiguous and chronic nature of ME/CFS, the need to obtain a diagnosis is particularly pronounced among patients who are attempting to bargain for adjustments in work, familial, or social roles. According to Clarke and James (2003), when ill individuals obtain a diagnosis, they are granted a "transitional identity as a person whose body is temporarily in poor functioning order, a person in a 'special' medically legitimated place in the social order" (p. 1393). When these individuals are not medically or socially permitted to take on the sick role, they "[lack] a place in the social order and [have] no obvious way of returning to his/her previous place" (p. 1393). Further,

> The lack of a clear and generally accepted diagnosis in the case of CFS means that the self is without the legitimating discourse from the powerful medical profession. Because most people have lost jobs and friends, they also lack the legitimacy offered by a place in the economic and social structural orders. (Clarke & James, 2003, p. 1393)

Qualitative research with ME/CFS sufferers has produced similar outcomes, with patients reporting experiencing difficulties with both employers and with family in association with lack of diagnosis. Cooper (1997) described these difficulties as follows:

> By not being allowed full and decisive entry into the sick role, sufferers found that their social position was eroded, their social identity devalued and

stigmatized, whilst they found it difficult to obtain legitimate absence from work or disability benefit. (p. 196).

Not surprisingly, the quest for a diagnosis was often linked by patients to a need to have their suffering named, to find potential relief from symptoms, to generate meaning out of the confusing constellation of symptoms, to experience validation of their sickness, and to attain the sick role (Cooper, 1997). In their qualitative research with individuals who struggled to find a diagnosis and explanation for the symptoms, Nettleton and colleagues (2004) noted that an additional concern is that these patients experience difficulties in making "sense" of or reconstructing "explanatory models" of their symptoms, since they cannot yet connect their illness to medical theory (p. 47). In light of this, these undiagnosed patients "hover precariously in a void between illness and disease" (p. 47), facing the specter of "no diagnosis, no prognosis" (p. 52).

Additionally, lack of diagnosis means that one does not have a "categorical identity," limiting the patient from joining group membership with others who share the illness or from seeking the support of others who face the same situation (Nettleton et al., 2004). Thus, lack of diagnosis leads to difficulties in obtaining both formal and informal sources of assistance and support.

Interestingly, participants often reported having self-diagnosed, following discussion in support groups and self-assessment of symptoms (Clarke, 1999; Clark & James, 2003) or after discovering articles in periodicals that explained ME/CFS (Cooper, 1997). These approaches to self-diagnosis are in keeping with Chrisman's (1977) Health-Seeking Process Model, a medical anthropological model that describes

how patients often consult with others in seeking to identify an illness, for suggestions in treatment, and for processing health professionals' responses to their illness.

For those who were able to obtain a formal medical diagnosis, the process often involved locating a physician who was known to consider ME/CFS to be legitimate or one who was open to learning from patients about ME/CFS (Clark & James, 2003). Physicians who provided a diagnosis and support became sources of hope to patients (Cooper, 1997). Whereas not having a diagnosis had contributed to uncertainty and confusion, when patients finally obtained a label for the symptoms and an idea of the course of illness, they reported experiencing an increase in feelings of hopefulness (Fisher & Crawley, 2012).

As might be predicted, obtaining diagnosis was often viewed as a symbolic milestone, and many reported psychosocial improvements as a result of identification of their illness (Cooper, 1997). Many experienced diagnosis as having "finally found some respite from the chaos and anarchy of their illness" (p. 196).

Concerns of medical professionals regarding diagnosis. Huibers and Wessely (2006) described ME/CFS as "a cultural phenomenon and metaphor of our times" (p. 897) that has been promoted by support groups, online information, self-help sources, and the media. The authors proposed that diagnosing the physical symptoms of ME/CFS as a legitimate illness may reinforce illness behaviors in patients, may result in self-validation and self-fulfilling prophesies, may contribute to negative outcomes, may lead to "transgression into the sick role," and may lead to "development of an illness identity and the experience of victimization" (p. 898).

Posing the question, "…should we tell [a patient they have ME/CFS] or not?" Huibers and Wessley (2006) recommended that individuals who are in the acute or early stages of ME/CFS should not be informed of their diagnosis "because the label may stimulate chronicity" (p. 899). They proposed that physicians should only share the news of diagnosis with individuals who have suffered with ME/CFS for an extended period of time, who are in an advanced stage of illness, and who evidence only low chances of recovery. The rationale for providing a diagnosis to those with advanced ME/CFS was that receiving a diagnosis may provide "relief, acceptance and the preservation of self-esteem to the experience of illness" (p. 899).

Interestingly, Huibers and Wessley (2006) did acknowledge that their advice to physicians regarding diagnosis was not without problems. In fact, their recommendation to provide symptomatic individuals a diagnosis, only after they had reached an advanced and prolonged state of illness, would, no doubt, be particularly difficult for individuals seeking validation of their symptoms. As Huibers and Wessley (2006) noted of ME/CFS sufferers:

> …the contrast between normal appearance and far from normal feeling, and the lack of objective or medically accepted disease verification, continues to leave the sufferer stranded uncomfortably between illness and disease. (p. 899)

Qualitative research with Swedish physicians noted that they, too, refrained from providing diagnoses to ME/CFS patients during the early stages of their illness, as a strategy "for hindering the patient from becoming too focused on his/her diagnosis and problems" (Asbring & Narvanen, 2003, p. 717). Furthermore, these medical doctors

reported that they did not provide their ME/CFS patients with medical papers necessary for exemption from work, during the early stages of illness, for the same reason.

Illness Invalidation

Illness invalidation has been defined as "a constellation of features that includes nonacceptance by others, misunderstanding, disbelief, rejection, stigmatization, and suspicion that the [health] problem is exaggerated or psychological" (Kool, Middendorp, Boeije, & Geenen, 2009, p. 1650). Illness invalidation may be particularly likely to occur when symptoms are invisible, ambiguous, and chronic.

Individuals with chronic ambiguous conditions, including ME/CFS, have reported that their families and their medical professionals discounted their symptoms and doubted their experiences (Dickson et al., 2007; Johnson & Johnson, 2006). Illness invalidation has also been reported by patients with other "invisible" conditions, such as fibromyalgia (Kool et al., 2009), rheumatic diseases (Kool & Geenen, 2012) and chronic pain (Newton, Soutball, Raphael, Ashford, & LeMarchand, 2013). Moreover, individuals with ME/CFS reported belief that absence of visibly-apparent symptoms contributed to illness invalidation (Asbring & Narvanen, 2001; Dickson et al., 2007).

Perspectives of medical professionals. As Clarke and James (2003) noted, "…one of the distinguishing features of CFS is that its reality is disputed by the medical profession" (p. 1393). Research involving general practitioners from the United Kingdom revealed that 28% did not believe that ME/CFS was an identifiable clinical diagnosis (Bowen et al., 2005). In the United States, 20% of physicians indicated belief that ME/CFS is "only in the patient's head" (Brimmer, Fridinger, Lin, & Reeves, 2010, p. 8). In a meta-synthesis of 325 peer-reviewed qualitative studies, physician-specific

themes included skepticism that ME/CFS is a legitimate disorder and minimization of the ME/CFS illness experience (Anderson et al., 2012). Similarly, a meta-synthesis of studies spanning from 1988 to 2013 also echoed the finding that many medical professionals do not believe that ME/CFS is a real disorder, due to the limitations in pathological findings (Bayliss et al., 2014), current lack of objective tests to ascertain the disorder's presence, and the lack of clearly-identified etiological mechanisms (Asbring & Narvanen, 2003).

Medical doctors have been found to stereotype patients with ME/CFS, based on inadequate knowledge of the ME/CFS pathophysiology (Anderson et al., 2012). Physicians expressed perceptions of many ME/CFS patients as "illness focused, demanding, and medicalising" (Asbring & Narvanen, 2003, p. 711). In particular, physicians noted that individuals with ME/CFS do not act and appear the way someone who is ill "is expected to look and behave," and they indicated that their assessment of physical appearance weighed heavily in their interpretation of the legitimacy of a patient's symptom description (Asbring & Narvanen, 2003, p. 714). In other words, physicians often doubted whether ME/CFS patients were legitimately ill or were attempting to obtain the sick role, procure time off from work, etc. (Asbring & Narvanen, 2003). Medical doctors made judgments about the veracity of their ME/CFS patients' reports and often questioned the morality of the patient, especially of the individual's work ethic. Similar research conducted with Swedish physicians who treated ME/CFS patients also reported physician skepticism about patient experiences, noting belief that the symptoms of ME/CFS were minor, that patients were exaggerating

the severity of their illness experiences, and that these patients had particularly pessimistic attitudes about life (Asbring & Narvanen, 2003).

Physicians also frequently expressed the opinion that "it was not desirable to have too many of these patients, as it could prove difficult to put up with them psychologically" (Asbring & Narvanen, 2003, p. 715). Medical doctors, particularly males, reported consciously avoiding and distancing themselves from their ME/CFS patients. In research involving general practitioners in the United Kingdom, only 12% indicated that they enjoyed seeing patients with ME/CFS (Bowen et al., 2005). Those who accepted the legitimacy of ME/CFS were over twice as positive about working with these patients.

Interestingly, many of the stereotypically negative behaviors of ME/CFS patients might be considered a normal part of the health-seeking process when viewed through the lens of medical anthropology. Based upon anthropological studies of health-related behaviors in the U.S., Chrisman (1977) developed the Health Seeking Process Model, in which the stages of illness typically consist of gaining a definition of symptoms, obtaining shifts in roles to accommodate the illness, lay consultation and referral, treatment behaviors, and adherence. Because individuals with ME/CFS often experience difficulties obtaining diagnoses (Clarke & James, 2003; Masuda et al., 2002), it would seem likely that these patients might become more persistent with their doctors, as diagnosis is often the first step in receiving cultural permission to shift work, familial, and social roles to accommodate the illness (Chrisman, 1977).

Research has also suggested that doctors treating ME/CFS patients often did not share patients' understanding of terms used to describe symptoms (Cooper, 1997).

Cooper (1997) described this as a "breakdown of a shared meaning system, where the same term signifies different experiences" (p. 197). For example, patients who used the term *fatigue* often later explained a different meaning for the term than is commonly understood.

> To sufferers, 'fatigue' means being so tired they cannot brush their hair or even sit up in bed. To doctors 'fatigue' may simply mean a term to describe a common occurrence in the general population as a result of modern-day stress. (Cooper, 1997, p. 197)

As such, symptoms that were experienced as severe by the patients were trivialized by their physicians.

In a more recent study, patients indicated that the impact of ME/CFS symptoms was significant, and yet, review of their medical records found that symptoms of fatigue were rarely included in medical documentation (Evengard et al., 2005). These results provide convergent support for the discrepant perspectives of symptoms by patients and their physicians.

In a meta-synthesis of qualitative studies, the theme of obtaining diagnosis was found to intersect with the theme of patient-physician power dynamics (Anderson et al., 2012). Physicians reported perceptions of individuals with ME/CFS as being doubtful of the doctor's knowledge of ME/CFS and of the doctor's likelihood of assisting them (Asbring & Narvanen, 2003). On the other hand, physicians often reported questioning their own professional knowledge as they worked with ME/CFS patients, and many expressed beliefs that there had been gaps in their medical education (Asbring & Narvanen, 2003). In another study, less than 30% of physicians reported having enough

information about making an ME/CFS diagnosis (Brimmer et al., 2010). Furthermore, doctors noted experiencing difficulties associated with being unable to provide causal explanations to patients, with having limited treatment options to offer, and with having inadequate answers to patients' questions (Asbring & Narvanen, 2003; Brimmer et al., 2010). Feelings of incompetence and helplessness, combined with patient dissatisfaction, appeared to threaten professional identity and contributed to physicians' experiencing negative opinions of treating patients with ME/CFS (Asbring & Narvanen, 2003).

Additionally, a study involving 811 general practitioners in the United Kingdom found that 48% lacked confidence in diagnosing ME/CFS, and 41% lacked confidence in treating these patients (Bowen et al., 2005). Physicians who accepted the legitimacy of ME/CFS were roughly three times more confident about their ability to diagnose the disorder. Finally, other factors significantly associated with medical professionals endorsing the legitimacy of ME/CFS were: (a) having a personal acquaintance with ME/CFS, (b) being male, and (c) having treated a greater number of ME/CFS patients within the previous year. Physicians who had previously given an ME/CFS diagnosis were less likely to believe that the disorder was "in a patient's head" (Brimmer et al., 2010, p. 8).

Perspectives of patients. Patients with ME/CFS have reported having their symptoms met with skepticism, disbelief, trivialization, and dismissal by medical professionals (Cooper, 1997; Deale & Wessely, 2001; Gilje, Soderlund, & Malterud, 2008). In qualitative research, most ME/CFS patients reported finding their interactions with doctors to be more frustrating than helpful (Clarke, 1999). Individuals with

ME/CFS reported experiencing their physicians as doubting the legitimacy of their illness, disbelieving that the symptoms stemmed from organic causes (Clarke & James, 2003), and generally misunderstanding the nature of their sickness (Cooper, 1997). In one study, over half of participants with ME/CFS (N = 211) reported belief that medical specialists did not seriously consider their complaints (53%) and did not sympathize with their illness experiences (54%; Prins et al., 2000). Further, ME/CFS patients have reported "being treated with disdain" and as "an annoying irritation" by their physicians (Arroll & Senior, 2008, pp. 453-454).

Other patients reported that doctors misrepresented their symptoms as stemming from depression, rather than understanding that their depression was a consequence of living with a severe physiological illness (Cooper, 1997). Female patients, in particular, reported having their symptoms dismissed and doubted by their physicians, as well as being labeled as "malingerers" and "bored housewives" (p. 198).

One participant reported:

> I had got a list of symptoms …one written down was mild depression. Having to give up work and be ill at the same time was bound to cause a slight depression…. Of course having the depression written down on a piece of paper he said, 'Oh you women, that's all you ever say…you're depressed.' 'Bored Housewife Syndrome' that's what he called it. (Cooper, 1997, p. 198)

Patients from this study also reported perceiving the lack of positive support from their doctors as reducing them to "rock bottom" and to a "bleak" outlook (Cooper, 1997, p. 196). This type of interaction has been referred to as "psychogenic dismissal," a term used to describe an experience wherein a patient experiences iatrogenic psychological harm as a result of having their symptoms dismissed as being "all in the mind" (p. 202). In addition, patients indicated that, in their struggle to obtain a

legitimate diagnosis, they often felt a great deal of pressure to demonstrate that they were genuinely ill and to appear to be "good" or "normal" patients (Cooper, 1997, p. 199). They noted experiencing the need to prove that, although their illness might be considered "deviant," they were not deviant themselves (Cooper, 1997, p. 199).

Some ME/CFS patients have reported that their physicians appeared to resent their active involvement in the diagnostic process and seemed to perceive the involvement as a threat to their medical knowledge and their position (Cooper, 1997). Other patients expressed that, in trying to demonstrate that they were "good" patients, they did not "provoke" their doctors by sharing information they had learned about ME/CFS from other sources (p. 199). These patterns are in keeping with other qualitative research involving patients with unexplained medical illnesses who reported that they worried that, if they were not perceived as "working" to get well, they would be suspected of malingering (Nettleton, 2006). These same patients also reported withholding information about their symptoms, when interacting with medical professionals who were perceived as invalidating.

Furthermore, the difficulties in obtaining a diagnosis often challenged patients' "underlying myth" that physicians are symbols of authority and symbols of healing (Cooper, 1997). As doctors disappointed patients by failing to address their health needs, patients often felt frustration and contempt. These patients described reaching a point where they rejected their internalized myth of the doctor as healer and authority figure and came to trust more deeply in their own ability to gather information about their illness. Despite this "demystification" of physicians' authority and increase in trust of self, these patients usually either confronted their existing doctor or changed medical

providers until a trusted physician could be found (p. 203). In qualitative research with patients who had unexplained illness, most understood that medical answers are not always straightforward and forthright; however, what these patients longed for was to have their medical professionals validate their illness experiences as "genuine" (Nettleton, 2006, p. 1170).

Impact of illness invalidation from medical professionals. In Western culture, medical professionals are culturally-sanctioned as having power to officially recognize an individual's symptoms as medically- and socially-legitimate (Cooper, 1997; Foucault, 1975; Hyden & Sachs, 1998; Woodward, Broom, & Legge, 1995). Foucault (1975) considered this power to be a form of medical policing, and others have interpreted it as "medical paternalism" (Finerman & Bennett, 1995, p. 2; Kirmayer, 1988). Within such a culture, some patients are not granted a medical sanction for their illness, particularly those individuals whose symptoms are difficult to diagnose or who suffer from ambiguous or chronic health conditions (Chrisman, 1977; Waxler, 1980). Without this medical sanction, illness invalidation from medical professionals may have a pronounced negative impact on patients.

Patients with ME/CFS have reported feeling confused, disempowered, self-doubting, and vulnerable, as well as experiencing loss of identity, upon perceiving illness invalidation from medical professionals (Arroll & Senior, 2008; Woodward et al., 1995). Moreover, illness invalidation from medical professionals has been reported as contributing to lack of validation from family and employers (Cooper, 1997) and as potentially undermining social support that would typically accompany the sick role (Deale & Wessely, 2001). Furthermore, having medical professionals fail to validate

symptoms has reportedly contributed to individuals with medically-unexplained illness questioning themselves, even when they knew their symptoms to be real (Nettleton, 2006).

Illness invalidation from social networks. In qualitative research, individuals with ME/CFS reported experiencing emotional pain as a result of illness invalidation from significant others, family members, friends, and the work environment (Asbring & Narvanen, 2002; Dickson et al., 2007; Larun & Malterud, 2007). In a meta-synthesis of 325 peer-reviewed qualitative studies of ME/CFS, a common theme among patients was the experience of reduced social connections and disrupted personal relationships in association with the illness (Anderson et al., 2012).

Patients often reported that friends were skeptical of the legitimacy of their sickness, that they were judged and rejected by many in their previous social networks, and that they often lost their friendship networks (Clarke & James, 2003). Adolescent patients reported that some friends and teachers invalidated their experiences of fatigue, doubting them, appearing to distrust their reported symptoms, dismissing their illness as malingering, and attributing their physical inabilities to laziness (Fisher & Crawley, 2012). Further, adolescents with ME/CFS noted having difficult experiences of teasing and bullying about their illness from peers. Other ME/CFS patients often reported feeling socially isolated and as though they were "outsiders" (Clark & James, 2003, p. 1390). Further, patients often found it difficult to meet new friends, due to physical limitations on activities.

Despite having a diagnostic label to explain their symptoms, ME/CFS patients reported that the lack of visible symptoms contributed to continued illness invalidation

from social networks (Fisher & Crawley, 2012). Patients reported having their symptoms trivialized and having others explain to them that "tiredness" is a normal experience (p. 564).

On the other hand, ME/CFS patients who experienced continued social support following illness onset reported deeply valuing those relationships (Clark & James, 2003). When members of patients' social networks understood that ME/CFS was seriously impacting their functional abilities, trust was increased in those relationships (Fisher & Crawley, 2012).

Illness invalidation from family. In qualitative research, patients reported that few of their family members continued to support them and to validate their illness (Clarke & James, 2003). Invalidation of illness experiences by a spouse were considered particularly emotionally difficult (Dickson et al., 2007) and were negatively related to adaptive outcomes (Heijmans, DeRidder, & Bensing, 1999). As one patient described it,

> The difference with friends – if they're not helpful you don't have to talk to them, but with [family] you have to – you can't choose your family. So you learn what to say and what not to say. (Clarke & James, 2003, p. 1391).

Shame

Shame has been defined as an individual's global assessment of the self as inferior, deficient, and of diminished value (Woien, Ernst, Patock-Peckham, & Nagoshi, 2003). Shame has been conceptualized as an emotional response that originates from interpersonal transactions (Tangney, Miller, Flicker, & Hill Barlow, 1996), evaluative social conditions, and threats to the social self (Dickerson, Gruenwald, & Kenemy,

2004; Dickerson, Gruenwald, & Kemeny, 2009). Shame is particularly likely to be evoked in situations in which a component of one's identity is, or has potential to be, negatively evaluated by others (Dickerson et al., 2004).

While shame is considered a negative evaluation of oneself, it is believed to be linked to the social environment or to the belief that a critical "imagined other" is judging the self (Lewis, 1971). Cooley (1902) explained this as follows, "…there is no sense of 'I,' in pride or shame, without its correlative sense of you, or he, or they" (p. 182). As Dickerson and colleagues (2004) described it, "…shame results when perceptions of negative *social* evaluation are transformed into negative *self*-evaluation" (p. 1195).

Shame has also been conceptualized as an emotional response to low social standing and the associated risks of interpersonal rejection, exclusion, or maltreatment (Balsamo et al., 2014; Dickerson et al., 2004). Research found that, among psychology students, a strong positive correlation existed between blaming oneself for having been socially criticized or "put-down" and experience of shame (Gilbert & Miles, 2000, p. 768). Furthermore, social rank and experiences of shame have been found to be highly correlated (Cheung, Gilbert, & Irons, 2004).

The social aspect of shame has been reflected in qualitative research, as well. In a study consisting of nearly one hundred autobiographical narratives, results revealed that, in situations in which participants wrote shameful statements, the participants' writing focused upon how others might be negatively evaluating them (Silfver, 2007).

Shame and Illness. Across history, there has been a social tendency to treat individuals with health problems as deficient or blame-worthy (Hodgkins & Baility).

One hypothesized motivation for this is that individuals with health difficulties serve as "reminders" to healthy individuals of "the dead body" and, as such, [are] repressed and repulsed" (p. 226). In Leviticus 21:16-24 of *The Bible*, individuals with health difficulties were relegated to an equivalent status as prostitutes and were classified within society as being unclean (Stiker, 1999). Hodgkins and Baility (2009) noted that "the dominant view of the disabled body [is] as a natural tragedy, rather than as a matter of political oppression and human diversity" (p. 214).

Anthropologists have noted that Western culture has embraced explanatory models of illness that are "blame focused" and that place responsibility for diseases and their outcomes upon the patients themselves, often ignoring the complexities of disease processes (Finerman & Bennett, 1995, p.1). These attributions of patient responsibilities for their illnesses may stem from increased lay and professional education about connections between lifestyle choices and disease. Such attributions ignore the reality that health difficulties are often induced by multiple causes and are frequently complex. However, the emphasis on patient accountability for lifestyle choices may translate into interpretations of sick individuals as blameworthy for their illnesses.

An example of this phenomenon of focusing blame on patients for their illness, while ignoring complexities of diseases processes, is reflected in results from qualitative research in the United Kingdom. Research there found that individuals with lung cancer who had no prior history of smoking considered themselves as being unfairly blamed by those around them for their disease (Chapple, Ziebland, & McPherson, 2004). Furthermore, some patients noted belief that media reports linking smoking to lung cancer may have contributed to the assessment that they were to blame for their cancer.

Shame has also been found to negatively correlate with psychological wellbeing, and this finding has been found to sustain across the lifespan (Orth, Robins, & Soto, 2010). Individuals living in Greece who were ashamed of past experiences of physical pain were found to have higher levels of psychopathology than individuals who had not experienced shame surrounding physical pain (Paschou, Damigos, Mavreas, & Gouva, 2010). Additionally, external shame (shame stemming from negative evaluations by others) has been found to correlate with experiences of depression and with submissive behaviors (Cheung et al., 2004; Kim, Thibodeau, & Jorgensen, 2011).

Research also indicates possible gender differences in external shame. To illustrate, women have been found to score higher on measures of external shame than men (Cheung et al., 2004; Orth et al., 2010).

Illness Invalidation and Shame

Discussing one's illness within the context of an invalidating environment may lead to internalized shame (Myers, 2004). Ware (1992) proposed that, when the subjective experience of one's illness is denied by the social context, shame of "being wrong in one's definition of reality" may occur (p. 347). Indeed, women with ME/CFS reported experiencing discrepancies between their own definition of their experience and the perceptions of their work environment and their physicians (Asbring & Narvanen, 2001).

Individuals with ME/CFS have expressed feelings of shame associated with illness invalidation and of experiencing their identity as questioned and no longer legitimate (Larun & Malterud, 2007). In qualitative research, participants sometimes related this experience to physicians, other individuals, and members of the media,

expressing the belief that ME/CFS was "all in their heads" (Clarke & James, 2003, p. 1390). Some individuals with ME/CFS even reported removing themselves from their former social lives out of feelings of shame over how others perceived them or after having others reject that their illness was legitimate (Clarke & James, 2003).

Social Support and the Stress Buffering Model

The Stress Buffering Model proposes that social support mitigates the negative effects of stress and promotes wellbeing (Cohen & Wills, 1985). Social support is conceptualized as manifesting across four support domains. *Esteem support* refers to communication that the individual is valued, worthy, and accepted. *Informational support* is operationalized as provision of coping support, advice, and guidance. *Social companionship* refers to time spent with others that is accompanied by a sense of belonging. Finally, *instrumental support* refers to assistance with tangible needs.

According to the Stress Buffering Model, esteem support counteracts threats to self-concept that may occur during stressful periods by communicating valuing and acceptance (Cohen & Wills, 1985). Informational support may promote cognitive reappraisals of stressful situations and/or may contribute to successful coping strategies. On the other hand, instrumental support and social companionship meet practical and affiliation needs during taxing times.

Numerous research studies appear to support the Stress Buffering Model, with evidence of the emotional benefits of social support to patients with health concerns indicated across multiple studies. Among individuals with rheumatic diseases, social support has been found to significantly relate to improved mental health (Kool et al., 2012). Specifically, individuals with rheumatoid arthritis who received higher amounts

of daily emotional support were more likely to report increased psychological wellbeing, decreased anxiety, fewer sleep difficulties, and less depression (Demange et al., 2004). Similar results among individuals with advanced cancer in Spain, revealed a significant moderate positive correlation between perceived social support and having a positive focus (Zabalegui, Cabrera, Navarro, & Cebria, 2011). Among survivors of hematopoietic stem cell transplant, emotional and instrumental social support predicted posttraumatic growth (Nenova, DuHamel, Zemon, Rini, & Redd, 2013). Furthermore, in a study of patients with HIV in Nepal, global satisfaction with social support significantly predicted quality of life, with the mediating effect of social support being hope (Yadav, 2010). A Japanese study discovered a significant buffering effect of social support on depressive symptoms, but the effect was found only in males, and not in females (Takizawa et al., 2006).

Social support has also been linked to health-related quality of life, as well. A literature review of 175 studies of social support and coping among prostate cancer patients revealed that the preponderance of research has indicated that perception of social support exerts a main effect upon health-related quality of life (Paterson, Jones, Rattray, & Lauder, 2013). Social support has also been linked to health-related quality of life in patients with heart failure (Bakan & Akyol, 2008). In addition, global satisfaction with social support predicted health-related quality of life among heart transplant recipients at five years post-transplant, and satisfaction with emotional aspects of social support predicted health-related quality of life at ten years post heart transplant (White-Williams et al., 2013). Among patients with diabetes in Turkey,

perceived social support was positively correlated with quality of life as well (Goz, Karaoz, Goz, Ekiz, & Cetin, 2007).

Individuals who lack medical explanations of illness symptoms have reported experiencing isolation and lack of social support (Nettleton, 2006). Patients with ME/CFS have expressed feelings of loneliness and separation from others, following perceived rejection by friends and significant others (Dickson et al., 2007), and adolescents with ME/CFS have noted perceiving that their inability to engage in activities as before had tested their relationships (Fisher & Crawley, 2012). Many patients with ME/CFS noted belief that receiving social support and validation would have increased their quality of life and facilitated coping (Dickson et al., 2007). However, research on the ameliorative impact of social support among individuals with ME/CFS has been limited, further justifying the current study.

Illness Invalidation, Shame, and the Stress Buffering Model

Discussing one's illness within the context of an invalidating environment may lead to internalized shame (Myers, 2004). Indeed, individuals with ME/CFS have expressed feelings of shame associated with the invalidation of their symptoms and having their experiences discounted, minimized, or dismissed (Larun & Malterud, 2007). According to the Stress Buffering Model, esteem support from social connection may help counteract threats to self-concept that may occur during stressful periods by communicating valuing and acceptance (Cohen & Wills, 1985). Thus, the following research questions appear worthy of investigation.

Research Questions

This study will seek to ascertain the following:

1) Is there a relationship between illness invalidation and shame among ME/CFS sufferers?
2) Does social support moderate the relationship between illness invalidation and shame among ME/CFS sufferers?

Chapter Three

Method

Participants

Participants will be 300 adults who suffer from ME/CFS and who are not pregnant. Participants will be recruited via ME/CFS and chronic illness support web sites, web sites of ME/CFS advocacy and research organizations, and social networking sites.

Instruments

Demographic information. Demographic information will be requested of participants, including information regarding gender, marital status, parenting status, income, sexual orientation, ethnicity, and highest level of education attained. Participants will also be asked questions pertaining to time since ME/CFS symptom onset and time since diagnosis.

Illness Invalidation Inventory. The Illness Invalidation Inventory (I*3; Kool et al., 2010) appears to be the only self-report instrument available, to date, that specifically measures perceived illness invalidation. The I*3 is a 40-item self-report measure that assesses perceived illness invalidation from five sources: spouse, family, medical professionals, work environment, and social services. Each subscale contains eight statements, rated on a 5-point Likert scale (ranging from "never" to "very often"). Twenty-four items are reverse scored.

The structure of illness invalidation was established by employing a pilot study in which adult patients with fibromyalgia were interviewed regarding their everyday experiences of illness invalidation (Kool, Middendorp, Boeije, & Geenen, 2009). Semi-

structured interviews, utilizing open-ended questions followed by probes, were conducted until no new themes emerged across two consecutive interviews. Interviews were then transcribed to identify statements regarding illness invalidation. Four members of the research team independently evaluated the statements, according to the following criteria: (a) statements not made by at least two participants were removed; (b) statement that were vague, abstract, or were not generalizable to other patients were deleted; and, (c) statements were edited for language and grammar, while retaining the participants' meaning. Participants then performed two separate Q-sort tasks of the 94 statements. Mean scores were calculated for each statement, in order to rate how characteristic each statement was of illness invalidation. A hierarchical cluster analysis revealed two higher-order aspects of illness invalidation, namely discounting and understanding, and 15 lower-order clusters.

In construction of the I*3, 45 of the most characteristic statements from the pilot study (three statements from each of the 15 lower-order components) were evaluated, including scoring and commenting, by thirty patients with rheumatic diseases (Kool et al., 2010). Additionally, participants identified the five sources of validation/invalidation that they considered most important to them, namely: spouses, family, medical professionals, work environments, and social services. Items for the instrument were selected according to the following criteria: (a) only one item for each original cluster; (b) factor loading of greater than 0.45 following factor analysis; and, (c) items should be applicable to all five sources. Eight items met these criteria and were utilized in the final instrument.

In factor analysis, I*3 statements were found to load on two factors of invalidation: Discounting and Lack of Understanding (Kool et al., 2010). A sample "Discounting" item is " My….finds it odd that I can do much more on some days than on other days." An example of a "Lack of Understanding" item is "My….takes me seriously" (R). Cronbach's alphas for both Discounting and Lack of Understanding subscales were found to be > .83 in a large New Zealand sample of individuals with rheumatic diseases.

For the purposes of this study, the total score for each individual will be utilized in data analyses as an indicator of global experience of illness invalidation.

The State Shame and Guilt Scale. Measures of shame fall into two categories: (1) those that assess *trait* shame, and (2) those that measure *state* shame (Robins, Noftle, & Tracy, 2007). Measures of trait shame assess one's shame-proneness as an enduring aspect of the personality. Measures of state shame, on the other hand, assess whether one experiences shame at a certain point in time, and such instruments may be used to measure shame in response to situational factors. For the purposes of this study, it was preferable that shame be measured in regard to one's experiences of perceived illness invalidation, rather than in regard to one's personality. As a result, the State Shame and Guilt Scale (SSGS; Marschall, Sanftner, & Tangney, 1994), a measure of state shame, was selected.

The State Shame and Guilt Scale (Marschall et al., 1994) was originally designed as a manipulation check of the initiation of shame in an experimental study of shame and empathy (Marschall, 1996). The SSGS is a 15-item (five items in each of three subscales) self-report instrument developed to measure Shame (the self as

intrinsically bad), Guilt (the self as having behaved poorly), and Pride (the self as valued; Marschall et al., 1994). Subscale items were reportedly developed in light of empirical and theoretical literature (Dearing & Tangney, 2002). Sample items include, "I feel worthwhile, valuable;" "I feel small;" and, "I feel worthless, powerless" (Marschall et al., 1994). Participants indicate their agreement/disagreement with statements on a five-point Likert scale. All items are scored in a positive direction. Subscale reliability coefficients ranged from .82 to .89 in a large sample of undergraduate students from an east-coast university. For the purpose of the current study, only the Shame subscale scores will be utilized in data analyses.

The Medical Outcomes Study Social Support Survey. The Medical Outcomes Study Social Support Survey (MOS; Sherbourne & Stewart, 1991) is a 19-item self-report measure of social support that was designed specifically for use with patients with chronic illness. The MOS includes four subscales that measure various dimensions of social support; namely, emotional/informational, tangible, affectionate, and positive social interactions. For each item, participants rate the availability of that particular support on a 5-point Likert scale, ranging from "None of the Time" to "All of the Time." Example items include: "Someone to confide in or talk to about yourself or your problems…;" "Someone to share your most private worries and fears…;" and, "Someone who understands your problems…." All items are scored in a positive direction.

In developing the MOS, Sherbourne and Stewart (1991) generated 50 potential items based upon a review of existing measures of social support and in light of the Social Buffering Hypothesis (Cohen & Wills, 1985) and current theory of social support

outlined in existing literature. Items were drafted to measure multiple dimensions of support, to be as comprehensive as feasible, and to be concise enough to limit participant fatigue. In order to minimize participant burden, social support was measured without respect for the source (e.g., whether social support came from friends, community, religious circles, etc.). Items were designed to distinctly differentiate from similar measures of loneliness, emotional health, family functioning, and limitations on social activity.

In order to assess face validity of the MOS, six behavioral scientists assigned each item to a social support category. Items that were difficult to categorize were discarded (Sherbourne & Stewart, 1991). The remaining thirty-eight items were utilized in a pilot study with patients at a rural medical clinic in Southern Illinois. Items that were not internally consistent with their hypothesized dimension of social support and that did not discriminate between social support and other aspects of health and health-related behavior were eliminated, resulting in the final instrument consisting of 19 items. Items were hypothesized to measure the following dimensions of social support: (a) *emotional support* (expression of positive feelings, empathy, understanding, and encouragement of expression of emotion); (b) *informational support* (offering suggestions, guidance, information, or feedback); (c) *tangible support* (providing material or physical assistance); (d) *positive social interactions* (providing enjoyable experiences and activities); and, (e) *affectionate support* (expression of positive regard, fondness, and love).

In order to evaluate the psychometric properties of the MOS, a large sample of patients (N = 2,987) from three research sites (Boston, Chicago, and Los Angeles) and

from three different healthcare systems (health maintenance organizations, large multispecialty groups, and private fee-for-service practice) completed the measure (Sherbourne & Stewart, 1991). In order to establish discriminant validity of social support items and to evaluate the construct validity of the MOS, participants also completed numerous health and wellbeing instruments.

Because multitrait scaling revealed notable overlap between emotional support and informational support items in the MOS, these items were combined into a single emotional/informational subscale (Sherbourne & Stewart, 1991). All items evidenced high correlation with their hypothesized subscales ($r \geq .72$), surpassing convergent validity criterion of $r = .30$. Item-subscale correlations ranged from $r = .72$ to .90. Internal-consistency reliability was high for each subscale, with alphas ranging from .91 to .97, and one-year stability coefficients ranged from .72 to .78.

All items in the four subscales of the MOS evidenced discriminant validity (Sherbourne & Stewart, 1991). Additionally, items were found to discriminate from measures of loneliness, mental health, feelings of belonging, perceptions of current health, and other dimensions of family and interpersonal functioning. The MOS subscales were found to be highly correlated ($r = .69$ - .82), as might be anticipated if they measure dimensions of the shared higher order factor of social support. However, multitrait and confirmatory factor analyses supported the use of subscales for scoring. Further, the authors recommended utilization of subscale scores in research, based upon their usefulness in testing theoretical hypotheses. Therefore, the current study will utilize combined scores from the *emotional/information support* subscale and the *affectionate support* subscale, as these subscales seem to best represent the construct of

esteem support (i.e., communication that the individual is valued, worthy, and accepted), as outlined in the Stress Buffering Model (Cohen & Wills, 1985).

Procedure

University of Oklahoma Institutional Review Board review and approval will be obtained prior to commencing this study. A snowball convenience sampling method will be utilized. An online survey will be created via Qualtrics, and will be maintained via a secure server in University of Oklahoma's Center for Educational Development and Research. Links to the study will be posted on ME/CFS and chronic illness support web sites, web sites of ME/CFS advocacy and research organizations, and social networking sites. Participation will be voluntary, without remuneration. Following consent, participants may skip questions or exit the survey at any time. No identifying information will be collected. Contacts and participants will be encouraged to forward the link to other adults with ME/CFS.

Hypotheses

It is hypothesized that there will be a positive and significant relationship between illness invalidation (I*3) and shame (SSGS). It is further hypothesized that social support (MOS) will moderate the relationship between illness invalidation and shame, with increases in social support leading to decreases in shame.

Statistical Analyses

A hierarchical regression model will be utilized to evaluate the hypotheses.

Chapter Four

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Appendix A

Illness Invalidation Inventory (3*I)

We are interested in how others react to people who have health problems or an illness. Each of the sections below refers to different people in your life. We would like you to rate **how often during the past year** each person or category of people reacted toward you in the way described. After each statement, circle the number between 1 (never) and 5 (very often) to indicate how often they reacted toward you that way.

The questionnaire has five sections, and you will rate the same reactions a number of times, but referring to different people. If a particular section does not apply to you, you may skip that part of the questionnaire and go on to the next section. Remember, rate the items with respect to how others reacted toward you **as a person who has health problems or an illness.**

Section 1: Spouse or partner

If you are single (not married, a widow/widower, or without a steady partner) then skip Section 1 and go directly to Section 2.

My spouse or partner……	**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
1.1 ….finds it odd that I can do much more on some days than on other days.	1	2	3	4	5

1.2	….thinks I should be tougher.	1	2	3	4	5
1.3	….takes me seriously.	1	2	3	4	5
1.4	….gives me unhelpful advice.	1	2	3	4	5
1.5	….understands the consequences of my health problems or illness.	1	2	3	4	5
1.6	….makes me feel like I am an exaggerator.	1	2	3	4	5
1.7	….thinks I can work more than I do.	1	2	3	4	5
1.8	….gives me the chance to talk about what is on my mind.	1	2	3	4	5

Section 2: Family

For example, children, parents, brothers, sisters, uncles, aunts, grandparents, in-laws.

My family……		**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
2.1	….finds it odd that I can do much more on some days than on other days.	1	2	3	4	5
2.2	….thinks I should be tougher.	1	2	3	4	5
2.3	….takes me seriously.	1	2	3	4	5
2.4	….gives me unhelpful advice.	1	2	3	4	5
2.5	….understands the consequences of my health problems or illness.	1	2	3	4	5
2.6	….makes me feel like I am an exaggerator.	1	2	3	4	5
2.7	….thinks I can work more than I do.	1	2	3	4	5

2.8 ….gives me the chance to talk about what is on my mind.	1	2	3	4	5

Section 3: Medical professionals

For example, your primary care physician, medical specialist, physical therapist, and other medical professionals. (Do not include your employer's company physician).

Medical professionals	**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
3.1 ….find it odd that I can do much more on some days than on other days.	1	2	3	4	5
3.2 ….think I should be tougher.	1	2	3	4	5
3.3 ….take me seriously.	1	2	3	4	5
3.4 ….give me unhelpful advice.	1	2	3	4	5
3.5 ….understand the consequences of my health problems or illness.	1	2	3	4	5
3.6 ….make me feel like I am an exaggerator.	1	2	3	4	5
3.7 ….think I can work more than I do.	1	2	3	4	5
3.8 ….give me the chance to talk about what is on my mind.	1	2	3	4	5

Section 4: Work environment

For example, your co-workers and boss. (Do not include your employer's company physician).

If you did not have paid or unpaid employment in the past year, then skip this Section and go directly to Section 5.

People at work…….	**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
4.1 ….find it odd that I can do much more on some days than on other days.	1	2	3	4	5
4.2 ….think I should be tougher.	1	2	3	4	5
4.3 ….take me seriously.	1	2	3	4	5
4.4 ….give me unhelpful advice.	1	2	3	4	5
4.5 ….understand the consequences of my health problems or illness.	1	2	3	4	5
4.6 ….makes me feel like I am an exaggerator.	1	2	3	4	5
4.7 ….think I can work more than I do.	1	2	3	4	5
4.8 ….give me the chance to talk about what is on my mind.	1	2	3	4	5

Section 5 : Social services

For example, your employer's company physician, work-reintegration or vocational rehabilitation staff, unemployment and other government agencies, organizations for care at home, general government workers and health insurance companies.

If you did not have any interactions with these providers, you may skip this Section.

People in social services…….	**Never**	**Seldom**	**Some-times**	**Often**	**Very often**
5.1 ….find it odd that I can do much more on some days than on other days.	1	2	3	4	5
5.2 ….think I should be tougher.	1	2	3	4	5
5.3 ….take me seriously.	1	2	3	4	5
5.4 ….give me unhelpful advice.	1	2	3	4	5
5.5 ….understand the consequences of my health problems or illness.	1	2	3	4	5
5.6 ….make me feel like I am an exaggerator.	1	2	3	4	5
5.7 ….think I can work more than I do.	1	2	3	4	5
5.8 ….give me the chance to talk about what is on my mind.	1	2	3	4	5

Appendix B

The State Shame and Guilt Scale

(Marschall, Sanftner, & Tangney, 1994)

The following are some statements which may or may not describe how you are feeling **right now**. Please rate each statement using the 5-point scale below. Remember to rate each statement based on how you are feeling **right at this moment**.

	Not feeling this way at all	Feeling this way somewhat	Feeling this way very strongly
1. I feel good about myself.	1 ------- 2 ------- 3 ------- 4 ------- 5		
2. I want to sink into the floor and disappear.	1 ------- 2 ------- 3 ------- 4 ------- 5		
4. I feel remorse, regret.	1 ------- 2 ------- 3 ------- 4 ------- 5		
4. I feel worthwhile, valuable.	1 ------- 2 ------- 3 ------- 4 ------- 5		
5. I feel small.	1 ------- 2 ------- 3 ------- 4 ------- 5		
6. I feel tension about something I have done.	1 ------- 2 ------- 3 ------- 4 ------- 5		
7. I feel capable, useful.	1 ------- 2 ------- 3 ------- 4 ------- 5		
8. I feel like I am a bad person.	1 ------- 2 ------- 3 ------- 4 ------- 5		
10. I cannot stop thinking about something			

bad I have done. 1 ------- 2 ------- 3 ------- 4 ------- 5

10. I feel proud. 1 ------- 2 ------- 3 ------- 4 ------- 5

11. I feel humiliated, disgraced. 1 ------- 2 ------- 3 ------- 4 ------- 5

12. I feel like apologizing, confessing. 1 ------- 2 ------- 3 ------- 4 ------- 5

13. I feel pleased about something I have done. 1 ------- 2 ------- 3 ------- 4 ------- 5

14. I feel worthless, powerless. 1 ------- 2 ------- 3 ------- 4 ------- 5

15. I feel bad about something I have done. 1 ------- 2 ------- 3 ------- 4 ------- 5

Appendix C

MOS Social Support Survey

(Sherbourne & Stewart, 1991)

Next are some questions about the support that is available to you.

People sometimes look to others for companionship, assistance, or other types of support. How often is each of the following kinds of support available to you if you need it?

None of the time	A little of the time	Some of the time	Most of the time	All of the time
1	2	3	4	5

20) Someone to help you if you were confined to bed….

21) Someone you can count on to listen to you when you need to talk….

22) Someone to give you good advice about a crisis….

23) Someone to take you to the doctor if you needed it….

24) Someone who shows you love and affection….

25) Someone to have a good time with….

26) Someone to give you information to help you understand a situation….

27) Someone to confide in or talk to about yourself or your problems….

28) Someone who hugs you….

29) Someone to get together with for relaxation….

30) Someone to prepare your meals if you were unable to do it yourself….

31) Someone whose advice you really want….

32) Someone to do things with to help you get your mind off things….

33) Someone to help with daily chores if you were sick….

34) Someone to share your most private worries and fears with….

35) Someone to turn to for suggestions about how to deal with a personal problem….

36) Someone to do something enjoyable with….

37) Someone who understands your problems….

38) Someone to love and make you feel wanted….