Causal Attributions and Perceived Stigma for Myalgic Encephalomyelitis/ Chronic Fatigue Syndrome

Introduction

People with Myalgic Encephalomyelitis (ME) or Chronic Fatigue Syndrome (CFS) have severe symptoms including profound exhaustion, muscle weakness and fatiguability, pain, cognitive dysfunction, sleep disturbance, flu-like symptoms and orthostatic intolerance (Carruthers et al., 2011; Fukuda et al., 1994). The hallmark symptom of the illness is post-exertional malaise (i.e., the worsening of all symptoms after minimal exertion; Institute of Medicine, 2015). Usually, the recovery from post-exertional malaise is prolonged, taking 24 hours or more. The etiology of ME/CFS is currently unknown. However, several physiological abnormalities have been identified, e.g. an impaired energy metabolism (Fluge et al., 2016; R. K. Naviaux et al., 2016), a reduced anaerobic threshold and maximal oxygen uptake (VO2 max; B. A. Keller, Pryor, & Giloteaux, 2014; S. Stevens, Snell, Stevens, Keller, & VanNess, 2018), as well as several indicators of autoimmunity (e.g., Bradley, Ford, & Bansal, 2013; Loebel et al., 2016). Research points to multi-faceted causes of this acquired illness and to date no diagnostic biomarker or effective causal treatment has been successfully identified (e.g. Bested & Marshall, 2015; Fischer et al., 2014; Scheibenbogen et al., 2017).

People with ME/CFS are often severely impaired in their activity, creating barriers to their education, employment and social life, as well as low health-related quality of life (e.g., Collin et al., 2011; Falk Hvidberg, Brinth, Olesen, Petersen, & Ehlers, 2015; Lattie et al., 2013; Rowe, 2019). In the United States, it is estimated that one million adults are affected by ME/CFS (e.g., Jason et al., 1999), and estimations for other countries show comparable base rates varying between 0.2 and 0.4 percent of the population (e.g., Nacul et al., 2011). In Germany, where the present study will be conducted, it is estimated that 240,000 individuals are affected by ME/CFS (Scheibenbogen et al., 2014).
To date, the etiology of ME/CFS is still poorly understood and the condition is largely unrecognized by health professionals and the public. People with ME/CFS are thus likely to experience delegitimizing experiences (e.g., physicians or family members denying that ME/CFS is a “real” physiological condition; Dickson, Knussen, & Flowers, 2007; N. C. Ware, 1992) and are at risk of being stigmatized (Green, Romei, & Natelson, 1999). Stigma can be defined as an “attribute that is deeply discrediting” and thus the stigmatized individual is “disqualified from full social acceptance” (Goffman, 1963). Stigmatized individuals are set apart from and perceived as inferior to others due to this marked attribute (Crabtree, Haslam, Postmes, & Haslam, 2010). Stigmatization thus involves processes of stereotyping, discrimination, and status loss of the individual associated with the stigma (Jones et al., 1984; Link & Phelan, 2001). Stigmatization in ME/CFS involves questioning the veracity and accuracy of symptoms (e.g., Åsbring & Närvänen, 2002) as well as attributing the symptoms to psychological rather than physiological causes (Baken, Harvey, Bimler, & Ross, 2018; Green et al., 1999; Looper & Kirmayer, 2004). Recent studies showed that perceived stigma in ME/CFS is related to reduced physical, mental and social functioning as well as depression and suicidal ideation (Baken et al., 2018; McManimen, McClellan, Stoothoff, & Jason, 2018). The level of perceived stigma was higher for people with ME/CFS compared to healthy controls or people with other chronic neurological conditions (e.g., epilepsy, Parkinson’s disease, multiple sclerosis; Baken et al., 2018).

In general, being affected by a chronic illness is associated with stigma (e.g., Rao et al., 2009). However, Weiner, Perry, and Magnusson (1988) showed that different stigmatizing conditions were systematically associated with different causal attributions (i.e., perceived causes for the condition). Attributions along the dimensions of controllability (i.e., perceived responsibility for the condition) and stability (i.e., perceived reversibility of the condition) varied for physically-based stigmas (e.g., blindness, Alzheimer’s disease, paraplegia) and mental-behavioral stigmas (e.g., drug abuse, obesity). Whereas physically-based stigmas were
perceived as uncontrollable and stable, mental-behavioral stigmas were perceived as controllable and unstable. These differences in attributions were in turn associated with affective and behavioral consequences: Mental-behavioral stigmas were associated with low pity and liking, high anger, and low intentions to help the stigmatized individual. In contrast, physically-based stigmas were associated with high pity and liking, low anger and high intentions to help the stigmatized individual (Weiner et al., 1988). According to the International Consensus Criteria (Carruthers et al., 2011) and the Centers for Disease Control and Prevention (2018), ME/CFS is classified as a neurological disease with physiological symptoms. However, the view that ME/CFS is a mental illness is widespread (Åsbring & Närvänen, 2002; Baken et al., 2018; Green et al., 1999; Terman, Awsumb, Cotler, & Jason, 2018). We thus argue that ME/CFS will be associated with attributions of controllability (i.e., the belief that people with ME/CFS are responsible for their condition), as well as attributions of instability (i.e., the belief that people with ME/CFS can change their condition). We will further investigate the associations of these attributions with perceived stigma of ME/CFS. In turn, we will investigate the association of perceived causal attributions and perceived stigma with central physiological and psychological outcome variables, that is, the functional status and satisfaction with social roles and activities of people with ME/CFS.

**Hypotheses**

**H1**: People with ME/CFS perceive that others attribute their condition to controllable and unstable causes.

**H2**: Perceived controllability and instability attributions predict higher perceived stigma, lower functional status and lower satisfaction with social roles and activities.

**H3**: The relationship of attributions with functional status and satisfaction with social roles and activities is mediated by perceived stigma.
Sampling Plan

Participants and Procedure

Participants with self-reported diagnosis of ME/CFS will be recruited to complete an online questionnaire via patient organizations, mailing lists, and social media. After receiving information on data protection, the aims of the study and providing written consent, participants will provide demographic information and will complete the DSQ-SF, the DePaul Post-Exertional Malaise Questionnaire, and the SF-36. Then they will complete measures of perceived causal attributions, perceived stigma, as well as satisfaction with social roles and activities. Last, they will complete demographics and items on illness history from the DSQ-2. Additional measures include access to and satisfaction with medical care, these measures will be analyzed in a separate research project. Finally, participants will be debriefed about the aims of the study. The study received approval by the first author’s institutional ethics commission. Materials were translated from English to German by the project team and back-translated by a professional translator. For the stigma and satisfaction with social roles and activities scales official translations of the Neuro QoL are used.

Sample Size

We aim at collecting a sample of at least $N = 350$ participants to be able to conduct path analysis to investigate H2 and H3. We conducted a Monte Carlo power analysis for indirect effects for the mediational model (H3) with an R web application (Schoemann, Boulton, & Short, 2017). With small effect sizes of the a and c paths of $r = .15$, and an estimated effect size for the b path of $r = -.36$ (based on the averaged correlations between stigma with global physical/mental health and ability to participate in social roles and activities reported in Baken et al., 2018) and a power of $1 - \beta = .80$, this yielded a required sample size of $N = 332$. 
**Materials/ Variables**

*Responsibility attributions* will be assessed with two items (“People see me as responsible for my illness”, “People blame me for my illness”), *stability attributions* will be assessed with one item (“People see my illness as changeable”; adapted from Weiner et al., 1988). Furthermore, attributions to psychological/physiological causes will be assessed with two items (“People see my illness as mental/physical.”). Responses will be provided on a Likert scale ranging from 1 = not at all to 5 = very much. Furthermore, participants’ individual attributions will be assessed (e.g., “I see myself as responsible for my illness”).

*Perceived stigma* will be assessed with the Stigma Scale for Chronic Illnesses from the Neuro-QoL (SSCI-8; 8 items; Molina, Choi, Cell, & Rao, 2013; e.g., “Because of my illness, some people avoided me”), responses will be provided on a Likert scale ranging from 1 = never to 5 = always. *Functional status* will be assessed with the Short-Form Health Survey (SF-36; 36 items; J. E. Ware & Sherbourne, 1992). *Satisfaction with social roles and activities* will be assessed with 8 items from the Neuro-QoL (e.g., “I am bothered by limitations in my regular activities with friends; Baken et al., 2018) on a scale ranging from 1 = not at all to 5 = very much. *ME/CFS symptoms* will be assessed with the De Paul Symptom Questionnaire Short Form (DSQ-SF, 14 items; Sunnquist, Lazarus, & Jason, 2019) and the DePaul Post-Exertional Malaise Questionaire (DSQ-PEM; 10 items; Cotler, Holtzman, Dudun, & Jason, 2018). Furthermore, the questionnaire will contain items on *access to medical care* (adapted from Sunnquist, Nicholson, Jason, & Friedman, 2017; Thanawala & Taylor, 2007), *satisfaction with medical care* (Sunnquist et al., 2017), *demographics* and *illness history* from the DSQ-2 (items items 3 – 11; 94 – 99; 111 – 114, 116; Jason & Sunnquist, 2018; demographic items adapted to the German context).
Analysis Plan

Transformations

For constructs measured with more than one item (i.e., responsibility attributions, perceived stigma, satisfaction with social roles and activities), responses will be averaged (if Cronbach’s alpha ≥ .70). Responses to the SF-36 will be scored according to Jason, Brown, Evans, Sunnquist, and Newton (2013). Responses to the DSQ-SF will be scored according to Sunnquist et al. (2019). Responses to the DSQ-PEM will be scored according to Cotler et al. (2018).

Statistical Models

H1: We will test whether participants perceive that their condition is attributed to controllable and unstable causes with one-sample t-tests testing whether the means differ from the scale mean.

H2: We will compute a path model with controllability/stability attributions as predictors and perceived stigma, functional status, and satisfaction with social roles and activities as outcomes.

H3: We will include indirect effects in the path model with controllability/stability attributions as predictors (X), perceived stigma as the mediator (M) and functional status and satisfaction with social roles and activities as outcomes (Y).

In an exploratory analysis, we will investigate whether severity of ME/CFS symptoms moderates the paths. In an exploratory analysis, we will control for whether patients fulfill the ICC case definition for ME/CFS (0 = no, 1 = yes) and for whether patients correctly responded to an attention check question. In an exploratory analysis, we will rerun the models without participants who indicate a depressive disorder as a comorbidity and/or indicate taking anti-depressant medication. In an exploratory analysis, we will investigate the correlation between perceived and individual causal attributions for the condition.
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