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Causal attributions and perceived stigma for myalgic encephalomyelitis/chronic fatigue syndrome

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Abstract

Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) is a chronic disease with the hallmark symptom of post-exertional malaise. Evidence for physiological causes is converging, however, currently no diagnostic test or biomarker is available. People with ME/CFS experience stigmatization, including the perception that the disease is psychosomatic. In a sample of 499 participants with self-diagnosed ME/CFS, we investigated perceived stigma as a pathway through which perceived others' causal attributions relate to lower satisfaction with social roles and activities and functional status. Higher perceived attributions by others to controllable and unstable causes predicted lower health-related and social outcomes via higher perceived stigma.

Keywords

causal attributions, Chronic Fatigue Syndrome, ME/CFS, Myalgic Encephalomyelitis, stigma

People with Myalgic Encephalomyelitis or Chronic Fatigue Syndrome (we will use the acronym ME/CFS) have severe symptoms including profound exhaustion, cognitive impairment, sleep disturbances, and other symptoms including muscle weakness, pain, flu-like symptoms, and orthostatic intolerance (Carruthers et al., 2011; Fukuda et al., 1994). The hallmark symptom is post-exertional malaise (PEM; i.e. worsening of all symptoms after minimal exertion; Institute of Medicine, 2015). Usually, recovery from PEM is prolonged, lasting 24 hours or more. The etiology of ME/CFS is currently unknown. However, several physiological abnormalities have been identified, for example, impaired energy metabolism (Naviaux et al., 2016; Schreiner et al., 2020), cardiopulmonary abnormalities (Stevens et al., 2018; van Campen et al., 2020), and indicators of autoimmunity (e.g. Loebel et al., 2016; Sotzny et al., 2018). Research points to multi-faceted causes and to date no

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diagnostic biomarker or effective causal treatment has been identified (e.g. Bested and Marshall, 2015; Fischer et al., 2014; Wirth & Scheibenbogen, 2020).

People with ME/CFS are often severely impaired in their activity, creating barriers to education, employment, and social life, as well as low health-related quality of life (e.g. Collin et al., 2011; Falk Hvidberg et al., 2015; Rowe, 2019). In the United States, it is estimated that 1.09 million adults (0.42% of the population) and 0.40 million children (0.75%) are affected by ME/CFS (Jason and Mirin, 2021), and a meta-analysis of 46 studies in 13 countries showed a pooled prevalence of 0.39% for adults (Lim et al., 2020). In Germany, where the present study was conducted, a mean base rate of 0.4% would translate into 332,000 individuals affected by ME/CFS, including 54,000 adolescents and children.

ME/CFS and perceived stigma

To date, the etiology of ME/CFS is still poorly understood and the condition is largely unrecognized by health professionals and the public. There is currently no objective test or biomarker to verify the disease. People with ME/CFS are thus likely to experience delegitimizing experiences (e.g. others denying that ME/CFS is a "real" physiological condition; Dickson et al., 2007; Ware, 1992) and are at risk of being stigmatized (Green et al., 1999). *Stigma* is an "attribute that is deeply discrediting" and thus stigmatized individuals are "disqualified from full social acceptance" (Goffman, 1963). They are set apart from and perceived as inferior to others due to this attribute (Crabtree et al., 2010). Stigmatization thus involves stereotyping, discrimination, and status loss (Jones et al., 1984; Link and Phelan, 2001).

People with ME/CFS perceive being stigmatized by others based on their disease. Ware (1992) interviewed 50 ME/CFS patients, of which 90% reported delegitimizing experiences by physicians due to the fact that the disease is not visible (patients do not "look sick"), and that in light of the lack of objective diagnostic markers physicians often conclude that the disease is psychosomatic (there is "nothing physically wrong" with the patients, so their disease must "be in their head"; Ware, 1992). These findings were corroborated by an interview study with 12 women with ME/CFS, who perceived that their credibility and the veracity of their disease was questioned by others due to the absence of visible signs of disease (Åsbring and Närvänen, 2002).

The psychologization of ME/CFS symptoms and its relation to perceived stigma was further investigated in a questionnaire study with 39 ME/CFS patients (Green et al., 1999). A total of 95% of participants reported feeling estranged by the disease and 70% perceived psychological attributions. Looper and Kirmayer (2004) compared the experiences of stigma of people with ME/CFS, irritable bowel syndrome, and fibromyalgia (diseases characterized by yet medically unexplained symptoms) with a matched control group of people with well-known other severe chronic diseases. The 42 participants with ME/CFS reported higher levels of perceived stigma than all other participant groups. In sum, qualitative, and quantitative studies showed that people with ME/CFS perceive to be stigmatized, in part because symptoms are attributed to psychological causes.

A study with 206 ME/CFS patients (Baken et al., 2018) investigated the consequences of perceived stigma for physical, mental, and social functioning. For the ME/CFS subsample, higher perceived stigma was associated with reduced physical, mental, and social functioning. Compared to normative populations with other neurological diseases, the ME/CFS sample showed higher stigma and lower functioning. McManimen et al. (2018) further showed that people with ME/CFS are at a greater risk of developing depressive symptoms and suicidal ideation. In a sample of 551 ME/CFS patients, participants who met the Beck Depression Inventory criteria for depression and suicidal ideation reported higher perceived stigma and unsupportive social interactions. Taken together, perceived stigma in ME/CFS can aggravate the already dire health-related and

social situation of many ME/CFS patients. However, the processes behind perceived stigma in ME/CFS are still unclear. Therefore, the present study draws on attribution theory to explain how perceived stigma relates to lower health and social functioning of people with ME/CFS.

The role of causal attributions for stigma in ME/CFS

In general, being affected by a chronic disease is associated with higher perceived stigma (e.g. Rao et al., 2009). However, Weiner et al. (1988) showed that different stigmatizing conditions were differently associated with causal attributions (i.e. perceived causes for the condition). According to attribution theory (e.g. Weiner, 1986), perceived causes for outcomes can be described on four dimensions, of which two are central to the current research: *Controllability* describes the extent to which the outcome is perceived to be controllable by an individual, whereas *stability* describes the extent to which the outcomes is perceived to be stable over time. Weiner et al. (1988) applied attribution theory to the domain of stigma (i.e. an unwanted outcome or negative effect eliciting attributions about its origin). They investigated how observers' reactions to social stigma (i.e. emotions and behavior towards the stigmatized person) were predicted by perceived causal attributions for the stigma. Physically-based stigmas (e.g. blindness, paraplegia) were perceived as uncontrollable (i.e. the respective person is not seen as responsible for its onset) and stable (i.e. the person is perceived as unlikely to recover). In contrast, the reverse pattern emerged for mental-behavioral stigmas (e.g. drug abuse, obesity), which were perceived as controllable and unstable. These attributional differences were associated with affective and behavioral consequences: Mental-behavioral stigmas were associated with observers' low pity and liking as well as high anger towards the stigmatized individual, and low intentions to help. In contrast, physically-based stigmas were associated with observers' high pity and liking as well as low anger towards the stigmatized individual, and high intentions to help (Weiner et al., 1988).

According to the International Consensus Criteria (Carruthers et al., 2011), the Centers for Disease Control and Prevention (2018), the ICD-10, as well as the upcoming ICD-11 (World Health Organisation, 2018), ME/CFS is classified as a neurological disease. However, the view that ME/CFS is a mental illness is widespread (Åsbring and Närvänen, 2002; Baken et al., 2018; Green et al., 1999; Terman et al., 2018). The main factor contributing to the perception of ME/CFS as a mental illness is likely the cognitive behavioral model of ME/CFS (Vercoulen et al., 1998). The deconditioning hypothesis proposed that fatigue and inactivity are aggravated by psychological factors like causal attributions (Vercoulen et al., 1998), which in turn has led to treatment recommendations of cognitive behavioral therapy (CBT) and graded exercise therapy (GET; e.g. Davinton et al., 2004; Wessely et al., 1991). The methodology applied by Vercoulen et al. (1998) was subsequently criticized, because the model could not be replicated for people fulfilling the ME/CFS case definitions (Sunnquist and Jason, 2018), and CBT and GET have been shown to have null effects or even aggravate ME/CFS symptoms (e.g. Kindlon, 2017; Wilshire et al., 2018). Nevertheless, the cognitive behavioral model has likely shaped the view of ME/CFS as a mental illness in the eyes of the public and among medical personnel.

Based on attribution theory we argue that ME/CFS patients perceive that their illness is associated with attributions of controllability (i.e. other people's belief that people with ME/CFS are responsible for their condition), and instability (i.e. other people's belief that ME/CFS is changeable and recovery is likely). The current research investigates how these social perceptions of ME/CFS (i.e. beliefs of people with ME/CFS about what others in their social environment think about the causes of their condition) relate to health-related and social outcomes. We expect that people with ME/CFS

perceive that others attribute their condition to controllable and unstable causes (*Hypothesis 1*). Building on Baken et al. (2018) and McManimen et al. (2018), we investigate the association of perceived causal attributions and perceived stigma with the functional status and satisfaction with social roles and activities of people with ME/CFS. We expect that perceived others' controllability and instability attributions predict higher perceived stigma, lower functional status, and lower satisfaction with social roles and activities (*Hypothesis 2*). To illuminate the process behind the negative consequences of stigmatization for people with ME/CFS, we investigate a path model including indirect effects: We expect the relationship of perceived others' attributions with functional status and satisfaction with social roles and activities to be mediated by perceived stigma (*Hypothesis 3*). The study was pre-registered prior to data collection on the Open Science Framework (OSF) [https://osf.io/spd9u/]. The pre-registration adheres to the disclosure requirements of the OSF.

Methods

Participants and procedure

A Monte Carlo power analysis for indirect effects (Hypothesis 3; Schoemann et al., 2017) with small effect sizes for the paths from perceived others' causal attributions to perceived stigma, functional status, and satisfaction with social roles and activities of $r = .15$, and an effect size for the path from perceived stigma to the outcome variables of $r = -.36$ (based on averaged correlations reported in Baken et al., 2018) and a power of .80 yielded a required sample size of $N = 332$. We pre-registered a minimum sample size of $N = 350$.

Participants with self-reported diagnosis of ME/CFS were recruited via mailing lists, homepages, and social media of the four largest patient organizations for ME/CFS in Germany. The online questionnaire took 30–45 minutes and was completed by 611 participants. We excluded participants who were younger than 18 years ($n = 7$) or did not consent to the inclusion of their data in the analyses ($n = 3$). Furthermore, we excluded participants who did not fulfil the Canadian Consensus Criteria for ME/CFS (Carruthers et al., 2003; $n = 30$; coded according to their responses to the DSQ-SF; Jason and Sunnquist, 2018). Finally, as Cotler et al. (2018) showed that a duration of PEM longer than 14 hours differentiated ME/CFS from other chronic diseases, we additionally excluded participants whose responses to the item "If you feel worse after activities, how long does this last" (item 9, DSQ-PEM, Jason et al., 2021) ranged between "1 hour or less" and "11–13 hours"; ($n = 72$). The final sample consisted of 499 participants (372 female, 125 male, two other), age ranged between 18 and 76 years ($M = 46.67$, $SD = 12.20$). Most participants were German (97%) and living in Germany (99%). A post-hoc power analysis showed that with the current sample, the hypothesized indirect effects could be detected with a power of .93.

Participants were informed about data protection and the aims of the study, and provided written consent in accordance with the EU General Data Protection Law and research ethics guidelines of the American Psychological Association. The study received IRB approval from the first author's institution. First, participants completed the DSQ-SF, the DSQ-PEM, the SF-36, measures of perceived others' causal attributions, perceived stigma, and satisfaction with social roles and activities. Then they provided information on demographics, medical history, and own causal attributions. Finally, participants were debriefed about the aims of the study and consented to their data being used for analyses. Scales for which no official translations were available were translated from English to German by the project team and back-translated by a professional translator. Materials, data, and analysis scripts are available on the OSF [https://osf.io/spd9u/].

Materials

Perceived others' controllability attributions were assessed with two items ("People see me as responsible for my illness," "People blame

me for my illness"), *perceived others' instability attributions* with one item ("People see my illness as changeable"; adapted from Weiner et al., 1988). Perceived others' attributions to psychological/ physiological causes were assessed with two items ("People see my illness as mental/ physical"). Responses ranged from 1 = not at all to 5 = very much. We also assessed participants' own causal attributions with three items ("I see myself as responsible for my illness," "I blame myself for my illness," and "I see my illness as changeable") as well as one item about perceived causes of the illness from the DePaul Symptom Questionnaire 2 (DSQ 2; "What do you think is the cause of your problem with fatigue/ energy/? 1 = definitely physical, 5 = definitely psychological"; Jason and Sunnquist, 2018). *Perceived stigma* was assessed with the Stigma Scale for Chronic Illnesses from the Neuro-QoL (SSCI-8; eight items; Molina et al., 2013; e.g. "Because of my illness, some people avoided me"), responses ranging from 1 = never to 5 = always. *Functional status* was assessed with the Short-Form Health Survey (SF-36; 36 items; Morfeld et al., 2011; Ware and Sherbourne, 1992). *Satisfaction with social roles and activities* was assessed with eight 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* were assessed with the De Paul Symptom Questionnaire Short Form (DSQ-SF, 14 items; Sunnquist et al., 2019) and the De Paul Symptom Questionnaire PEM (8 of 10 items assessed; Cotler et al., 2018). Furthermore, the questionnaire contained items on *demographics* and *illness history* from the DSQ 2 (items 3–11; 94–99; 111–114, 116; Jason and Sunnquist, 2018; demographics adapted to the German context).[1]

Data sharing statement

The current article includes the complete raw data-set collected in the study including the participants' data set, syntax file, and log files for analysis. Pending acceptance for publication, all of the data files will be automatically uploaded to the Figshare repository.

Results

Descriptive analyses

Multi-item constructs were averaged to scales. The SF-36 was scored according to Ware et al. (1993). For the DSQ-SF, per item the frequency and severity ratings were averaged and then multiplied by 25 to create a scale ranging from 0 to 100. Then all items were averaged to single scales per measure (Jason and Sunnquist, 2018). Fulfillment of case definition was computed based on the DSQ-SF, the DSQ PEM, and the SF-36 following Jason and Sunnquist (2018). Descriptive statistics and internal consistencies are displayed in Table 1, correlations in Table 2. Missing data was <2% for all variables. We employed listwise deletion for *t*-tests and FIML estimation for path modeling (Lüdtke et al., 2007).

Perceived causal attributions for ME/ CFS

Single-sample *t*-tests showed that in line with Hypothesis 1, perceived others' instability attributions were higher than the scale midpoint of 3, $M = 3.44$, 95% CI [3.33; 3.55]; SE = 0.05, $t(492) = 8.27$, $p < 0.001$, $d = 0.37$. Unexpectedly, however, perceived others' controllability attributions were lower than the scale midpoint, $M = 2.38$, 95% CI [2.28; 2.49]; SE = 0.06, $t(492) = 11.01$, $p < 0.001$, $d = 0.50$. Perceived others' attributions to physiological causes did not differ from the scale midpoint, $M = 2.95$, 95% CI [2.83; 3.05]; SE = 0.05, $t(492) = 1.08$, $p = 0.282$, $d = 0.04$, whereas perceived others' attributions to psychological causes were above the scale midpoint, $M = 3.28$, 95% CI [3.16; 3.40]; SE = 0.06, $t(492) = 4.78$, $p < 0.001$, $d = 0.22$.

In exploratory analyses, we investigated the mean levels of participants' own causal attributions for ME/CFS (i.e. how patients themselves viewed the controllability and instability of

Table 1. Descriptive statistics and internal consistencies.

	Scale	*M* [95% CI]	SE	Cronbach's α
Causal attributions				
Controllability	1–5	2.39 [2.27; 2.50]	0.06	0.85
Instability	1–5	3.44 [3.33; 3.54]	0.05	–
Physiological	1–5	2.93 [2.83; 3.03]	0.05	–
Psychological	1–5	3.28 [3.17; 3.39]	0.06	–
Perceived stigma	1–5	2.77 [2.69; 2.85]	0.04	0.84
Functional status (SF-36)				
Physical functioning	0–100	30.94 [29.14; 32.21]	0.99	0.90
Role physical	0–100	2.81 [2.05; 3.68]	0.42	0.37
Bodily pain	0–100	30.03 [28.06; 32.21]	1.06	0.72
General health	0–100	22.29 [21.11; 23.47]	0.59	0.50
Vitality	0–100	14.00 [12.83; 15.16]	0.61	0.62
Social functioning	0–100	18.00 [16.09; 19.87]	0.95	0.67
Role emotional	0–100	64.55 [60.62; 68.43]	2.02	0.93
Mental health	0–100	56.70 [54.85; 58.47]	0.92	0.85
Satisfaction with social roles and activities	1–5	1.70 [1.64; 1.76]	0.03	0.81

For constructs assessed with single items, no consistency is displayed. For constructs assessed with two items (controllability attributions, social functioning, bodily pain), Spearman's rho is displayed.

their disease) as well as the interrelations between perceived others' and own causal attributions. We averaged the two items measuring own controllability attributions (Spearman's $rho = 0.69$; $p < 0.001$). Own controllability attributions were lower than perceived others' controllability attributions ($M = 1.52$, 95% CI [1.45; 1.59], SE = 0.03, $t(494) = 14.46$, $p < 0.001$, $d = 0.65$). Own instability attributions were also lower than perceived others' instability attributions ($M = 2.45$, 95% CI [2.37; 2.54], SE = 0.05, $t(494) = 13.74$, $p < 0.001$, $d = 0.62$). Furthermore, participants themselves rather attributed their illness to physical than psychological causes (on a scale of 1 = completely physical to 5 = completely psychological; $M = 1.57$, 95% CI [1.51; 1.63], SE = 0.03).

Own controllability attributions were positively related to own instability attributions ($r(493) = 0.13$, 95% CI [0.03; 0.22], $p = 0.005$) as well as to perceived others' controllability and instability attributions (controllability: $r(493) = 0.20$, 95% CI [0.10; 0.29], $p < 0.001$; instability: $r(493) = 0.13$, 95% CI [0.05; 0.21], $p < 0.003$). In contrast, own instability attributions were negatively related to perceived others' controllability attributions ($r(493) = -0.11$, 95% CI [−0.20; −0.02], $p = 0.013$) and unrelated to perceived others' instability attributions ($r(493) = -0.03$, 95% CI [−0.12; 0.06], $p = 0.573$).

Consequences of perceived others' causal attributions for ME/CFS

To test Hypotheses 2 and 3, we computed a path model in Mplus version 7 (Muthén and Muthén, 2008–2015). The fully identified model included all direct and indirect paths from perceived others' causal attributions to perceived stigma, functional status, and satisfaction with social roles and activities. In line with Hypothesis 2, perceived others' controllability and instability attributions positively predicted perceived stigma. In turn, perceived stigma predicted lower functional status (all subscales except vitality) and lower satisfaction with social roles and activities (Table 3 and Figure

Table 2. Bivariate correlations (*r*, [95% CI]).

	(1)	(2)	(3)	(4)	(5)	(6)	(7)	(8)	(9)	(10)	(11)	(12)	(13)
(1) Controllability attributions	–												
(2) Instability attributions	0.62*** [0.56; 0.67]	–											
(3) Physiological attributions	−0.48*** [−0.55; −0.41]	−0.42*** [−0.50; −0.33]	–										
(4) Psychological attributions	0.66*** [0.61; 0.71]	0.58*** [0.51; 0.63]	−0.70*** [−0.76; −0.64]	–									
(5) Perceived stigma	0.57*** [0.50; 0.63]	0.49*** [0.42; 0.56]	−0.33*** [−0.42; −0.25]	0.47*** [0.40; 0.53]	–								
(6) Physical functioning	−0.09* [−0.18; −0.01]	−0.12** [−0.21; −0.02]	−0.04 [−0.13; 0.05]	−0.02 [−0.11; 0.07]	−0.29*** [−0.37; −0.20]	–							
(7) Role physical	−0.02 [−0.11; 0.07]	−0.04 [−0.12; 0.05]	0.02 [−0.06; 0.10]	−0.06 [−0.15; −0.02]	−0.17*** [−0.24; −0.09]	0.15** [0.04; 0.24]	–						
(8) Bodily pain	−0.01 −0.11; 0.09]	−0.02 [−0.11; 0.07]	0.04 [−0.06; 0.14]	−0.05 [−0.14; −0.05]	−0.20*** [−0.27; −0.11]	0.34*** [0.25; 0.43]	0.12* [0.02; 0.20]	–					
(9) General health	−0.14** [−0.24; −0.05]	−0.17*** [−0.26; −0.09]	0.07 [−0.03; 0.15]	−0.13** [−0.22; −0.05]	−0.26*** [−0.35; −0.18]	0.23*** [0.14; 0.32]	0.13** [0.01; 0.24]	0.25*** [0.16; 0.34]	–				
(10) Vitality	−0.10* [−0.18; −0.01]	−0.14** [−0.22; −0.05]	0.11* [0.02; 0.19]	−0.18*** [−0.26; −0.09]	−0.12** [−0.21; −0.03]	0.23*** [0.13; 0.31]	0.03 [−0.05; 0.12]	0.19*** [0.10; 0.27]	0.30*** [0.21; 0.39]	–			
(11) Social functioning	−0.14** [−0.22; −0.05]	−0.20*** [−0.29; −0.10]	0.02 [−0.08; 0.11]	−0.12** [−0.22; −0.03]	−0.32*** [−0.40; −0.23]	0.40*** [0.31; 0.47]	0.13** [0.03; 0.23]	0.19*** [0.10; 0.28]	0.25*** [0.16; 0.33]	0.24*** [0.14; 0.33]	–		
(12) Role emotional	−0.03 [−0.12; 0.06]	−0.01 [−0.10; 0.08]	0.16*** [0.07; 0.25]	−0.16** [−0.24; −0.06]	−0.19*** [−0.28; −0.10]	−0.02 [−0.12; 0.07]	0.06 [−0.01; 0.13]	0.20*** [0.12; 0.28]	0.06 [−0.04; 0.14]	0.13** [0.04; 0.21]	0.06 [−0.03; 0.14]	–	
(13) Mental health	−0.13** [−0.23; −0.05]	−0.07* [−0.15; −0.02]	0.16*** [0.08; 0.24]	−0.23*** [−0.31; −0.15]	−0.34*** [−0.42; −0.26]	0.07 [−0.03; 0.16]	0.04 [−0.06; 0.14]	0.21*** [0.12; 0.29]	0.25*** [0.16; 0.33]	0.29*** [0.21; 0.36]	0.24*** [0.15; 0.31]	0.53*** [0.46; 0.59]	–
(14) Satisfaction social roles and activities	−0.19*** [−0.27; −0.11]	−0.23*** [−0.31; −0.14]	0.14** [0.04; 0.23]	−0.24*** [−0.33; −0.15]	−0.37*** [−0.45; −0.29]	0.31*** [0.22; 0.40]	0.20*** [0.09; 0.30]	0.20*** [0.10; 0.30]	0.34*** [0.25; 0.42]	0.31*** [0.23; 0.39]	0.43*** [0.34; 0.52]	0.16** [0.08; 0.25]	0.31*** [0.23; 0.38]

*$p < 0.05$. **$p < 0.01$. ***$p < 0.001$.

1). In line with Hypothesis 3, all indirect effects from perceived others' attributions to functional status (except vitality) and satisfaction with social roles and activities via perceived stigma were also significant (Table 3).[2] Results were robust when controlling for whether participants answered the attention check correctly ($n = 463$, 92.8% correct).[3] In additional exploratory analyses, we excluded item eight from the stigma scale ("Some people act as though it was my fault I have this illness") due to conceptual similarity to perceived others' controllability attributions. Results were unchanged (see OSF).

Discussion

ME/CFS is associated with stigmatization due to its unclear etiology, which in turn relates to lower physical and social functioning (e.g. Baken et al., 2018). Based on attribution theory (Weiner, 1986), we investigated the relations of perceived others' causal attributions for ME/CFS, perceived stigma, and functional status, as well as satisfaction with social roles and activities in a sample of 499 participants with self-reported ME/CFS (fulfilling the Canadian Consensus Criteria and reporting >14 hours of PEM after exertion) in Germany.

Attributional patterns for ME/CFS

On average, participants perceived that others in their social environment viewed their disease as changeable, but uncontrollable. Perceived others' attributions to physical causes were moderate, however, perceived attributions to psychological causes were high. These results partly support Hypothesis 1. In line with Weiner et al. (1988), we expected perceived others' attributions for ME/CFS to be consistent with mental-behavioral stigma (high controllability and instability attributions). However, results point to a mixed perception of ME/CFS as a physically-based stigma (low controllability and moderate physiological attributions) and a mental-behavioral stigma (high attributions to unstable, psychological causes). People with ME/CFS might perceive a shift in others' attributions for their illness as less controllable and more physical due to current research substantiating that ME/CFS is a physical condition. However, notions that ME/CFS is psychological still persist. In addition to psychologization of ME/CFS due to the lack of visibility and diagnostic tests (e.g. Green et al., 1999; Ware, 1992), the cognitive behavioral model of ME/CFS (Vercoulen et al., 1998) likely contributes to these enduring psychological attributions. The deconditioning hypothesis proposed that fatigue and inactivity are aggravated by psychological factors like causal attributions (Vercoulen et al., 1998), which in turn has led to treatment recommendations of CBT and GET (e.g. Davinton et al., 2004; Wessely et al., 1991). Although these recommendations have later been shown to be ineffective or even harmful (Kindlon, 2017; Wilshire et al., 2018), the current study underlines that the conception that psychological factors cause or primarily contribute to ME/CFS symptoms continues to negatively affect people with ME/CFS.

Interestingly, there was a discrepancy between the causes participants ascribed to their disease (own attributions) and the attributions they thought others make (perceived others' attributions). Participants' own attributions painted a clear picture corresponding to attributions for physically-based stigma (Weiner et al., 1988). Participants themselves viewed their disease as uncontrollable, stable, and physiological. Own controllability attributions and perceived others' controllability attributions corresponded (i.e. individuals who thought that they are not responsible for their disease also reported that others do not view them as responsible). In contrast, own and perceived others' instability attributions were unrelated, which might indicate that people with ME/CFS are more aware of variability in their symptoms than the people around them.

The role of perceived stigma for ME/CFS

In line with Hypotheses 2 and 3, higher controllability and instability attributions were related to lower functional status and lower satisfaction

Table 3. Direct effects (upper part of table) and indirect effects (lower part) in path model.

Direct effects

Outcomes	Controllability attributions			Instability attributions			Perceived stigma		
	β [95% CI]	SE	*p*	β [95% CI]	SE	*p*	β [95% CI]	SE	*p*
Perceived stigma	0.43 [0.35; 0.52]	0.04	<.001	0.23 [0.14; 0.31]	0.05	<.001	–	–	–
Functional status									
Physical functioning	0.11 [−0.01; 0.23]	0.06	.053	−0.02 [−0.13; 0.09]	0.06	.763	−0.34 [−0.44; −0.24]	0.05	<.001
Role physical	0.11 [−0.01; 0.22]	0.06	.085	0.02 [−0.10; 0.13]	0.06	.783	−0.24 [−0.34; −0.13]	0.05	<.001
Bodily pain	0.13 [0.01; 0.24]	0.06	.037	0.05 [−0.06; 0.16]	0.06	.393	−0.30 [−0.40; −0.19]	0.05	<.001
General health	0.05 [−0.07; 0.17]	0.06	.420	−0.08 [−0.19; 0.03]	0.06	.174	−0.25 [−0.36; −0.15]	0.05	<.001
Vitality	0.02 [−0.10; 0.14]	0.06	.754	−0.11 [−0.22; 0.01]	0.06	.060	−0.08 [−0.18; 0.03]	0.06	.174
Social functioning	0.11 [−0.01; 0.22]	0.06	.064	−0.10 [−0.24; 0.01]	0.06	.070	−0.33 [−0.43; −0.23]	0.05	<.001
Role emotional	0.08 [−0.04; 0.19]	0.06	.217	0.07 [−0.04; 0.19]	0.06	.197	−0.28 [−0.38; −0.17]	0.05	<.001
Mental health	0.04 [−0.08; 0.15]	0.06	.520	0.11 [0.01; 0.22]	0.05	.040	−0.41 [−0.51; −0.31]	0.04	<.001
Satisfaction with social roles and activities	−0.09 [−0.20; 0.01]	0.05	.085	0.08 [−0.03; 0.19]	0.06	.156	−0.37 [−0.47; −0.27]	0.05	<.001

Indirect effects

Outcomes	Controllability attributions via perceived stigma			Instability attributions via perceived stigma		
	ab [95% CI]	SE	*p*	ab [95% CI]	SE	*p*
Functional status						
Physical functioning	−0.15 [−0.20; −0.09]	0.03	<.001	−0.08 [−0.11; −0.04]	0.02	<.001
Role physical	−0.10 [−0.15; −0.05]	0.03	<.001	−0.05 [−0.09; −0.02]	0.02	.001
Bodily pain	−0.13 [−0.18; −0.08]	0.03	<.001	−0.07 [−0.10; −0.03]	0.02	<.001
General health	−0.11 [−0.16; −0.06]	0.03	<.001	−0.06 [−0.09; −0.02]	0.02	.001
Vitality	−0.03 [−0.08; 0.02]	0.02	.179	−0.02 [−0.04; 0.01]	0.01	.190
Social functioning	−0.14 [−0.19; −0.09]	0.03	<.001	−0.07 [−0.11; −0.04]	0.02	<.001
Role emotional	−0.12 [−0.17; −0.07]	0.03	<.001	−0.06 [−0.10; −0.03]	0.02	<.001
Mental health	−0.18 [−0.23; −0.12]	0.03	<.001	−0.09 [−0.14; −0.05]	0.02	<.001
Satisfaction with social roles and activities	−0.16 [−0.21; −0.11]	0.03	<.001	−0.08 [−0.12; −0.04]	0.02	<.001

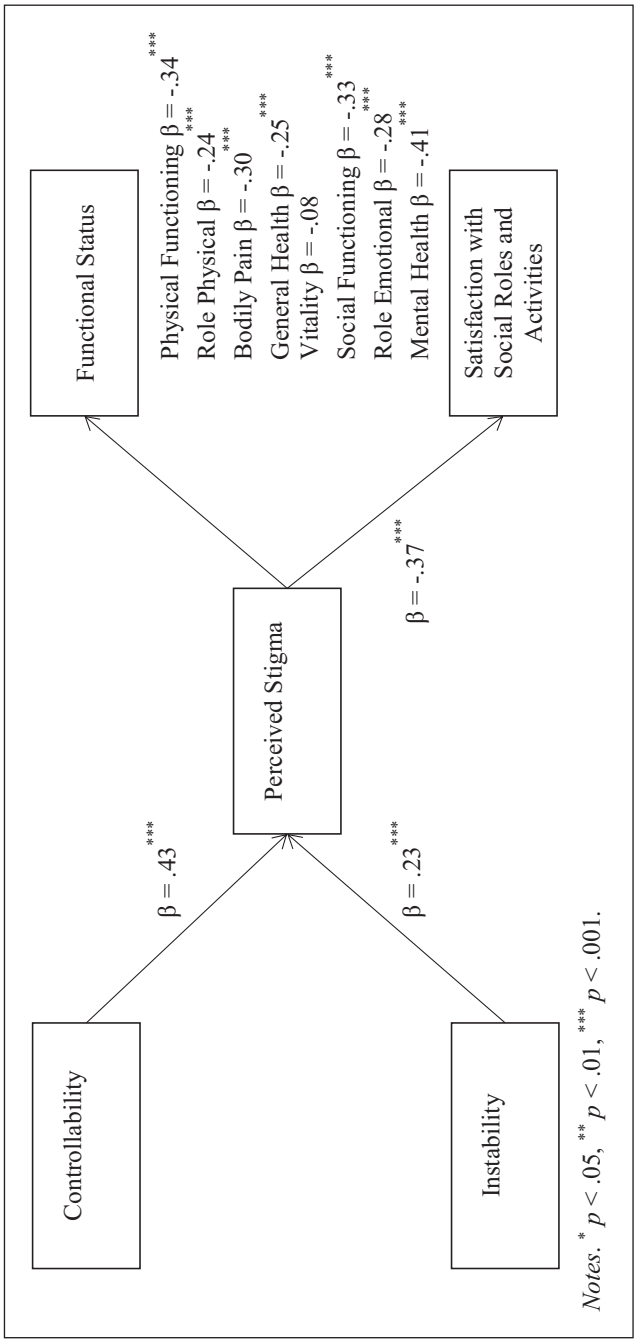

Figure 1. Results of the path model according to hypotheses (results for complete model are displayed in Table 3).

with social roles and activities via increased perceived stigma. This means that health-related quality of life outcomes were worse for people with ME/CFS who thought that others in their social environment viewed them as responsible for their condition and viewed the condition as likely to change. In turn, when people with ME/CFS perceived this pattern of others' attributions, they also felt more stigmatized by these other people. Results were consistent except for non-significant direct and indirect effects for vitality. This might be explained by the fact that vitality items measure energy and exhaustion, which are closely tied to ME/CFS symptoms.

The perception that others view the illness as controllable and changeable indirectly predicted lower health-related quality of life for people with ME/CFS. This result highlights that perceived stigma is a relevant process in ME/CFS: In line with basic stigma definitions (e.g. Crabtree et al., 2010; Goffman, 1963), ME/CFS is perceived as discrediting (e.g. participants reported feeling embarrassed because of their disease) and leads to social exclusion (e.g. participants reported being left out and avoided because of their disease). In turn, perceived stigma was not only related to lower functional status, but participants also reported being less satisfied with their social roles (e.g. being bothered by their limitations to socialize and meet the needs of family and friends) and activities (e.g. not being satisfied with the amount of household and leisure activities they can do). Consequently, the social perceptions of ME/CFS are relevant to patients' health and relationships. Findings point to pathways to improve the health-related and social situation of ME/CFS patients: Further education of health practitioners and the public about physiological causes of ME/CFS could improve the situation for patients. A widespread, evidence-based conception of ME/CFS as a physical condition could also increase the fit between illness perceptions by patients and their social environment, which in turn could reduce the negative consequences associated with ME/CFS (e.g. unsupportive social interactions, suicidal ideation; McManimen et al., 2018).

Limitations and future research directions

The current cross-sectional research can only provide correlational evidence for the mediating role of perceived stigma. Moreover, it included people with self-reported ME/CFS (all participants included in the analyses fulfilled case definitions but nothing was known about differential diagnostics). Further, completing longer questionnaires might induce PEM for ME/CFS patients, we thus kept the survey short. Therefore, the number of items with which the attribution constructs were measured was comparatively low. Future research should investigate cohorts with medically documented ME/CFS diagnosis, analyze causal relations in longitudinal studies, compare the role of perceived attributions and stigma for other illnesses of unknown etiology (e.g. fibromyalgia), increase the number of items to measure causal attributions, and evaluate the effect of educational programs about ME/CFS on perceived attributions and other patient outcomes.

Conclusion

The unknown etiology and ongoing search for diagnostic tests and biomarkers for ME/CFS poses a challenge for practitioners, patients, and their relatives. Views of ME/CFS as controllable and unstable aggravate the challenge of managing the condition and maintaining health-related quality of life. The current study provided new perspectives on the interplay of social perceptions of people with ME/CFS and health-related patient outcomes.

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Data availability statement

The data that support the findings of this study are openly available on the Open Science Framework at [https://osf.io/spd9u/].

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Notes

1. Further items included the DSQ-PEM (Cotler et al., 2018), access to medical care (adapted from Sunnquist et al., 2017; Thanawala and Taylor, 2007), and satisfaction with medical care (Sunnquist et al., 2017). Detailed analyses of demographics and the medical care situation of people with ME/CFS in Germany are reported in Froehlich et al. (2021).
2. We also tested a reversed model in which functional status/ satisfaction with social roles and activities predicted attributions, which in turn predicted perceived stigma. However, indirect effects were only significant for satisfaction with social roles and activities, but not for functional status.
3. We pre-registered computing case definitions according to the International Consensus Criteria, but we used the Canadian Consensus Criteria for comparability with previous research.

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