Distress and Coping in ME/CFS: Understanding the Role of Perfectionism and Self-compassion

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Reserved for Thesis Access Form

Declaration

I, the author, confirm that the thesis is my own work. I am aware of the University's Guidance on the Use of Unfair Means (www.sheffield.ac.uk/ssid/unfair-means). This work has not been previously presented for an award at this, or any other, university.

Structure and Word Counts

Section I: Literature Review

Excluding references and tables	6592
Including references and tables	10871

Section II: Empirical Project

Excluding references and tables	6859
Including references and tables	9180

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Lay Summary

Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) is a complicated long-term illness with symptoms that often restrict everyday activities and reduce quality of life. People with ME/CFS have a greater risk of experiencing mental health difficulties, including anxiety and depression. There is a lack of agreement on what causes ME/CFS and effective treatments. Therefore, it is important to find adaptive (i.e., helpful) ways of coping with this illness, in order to reduce mental health difficulties experienced. Additionally, research suggests traits like perfectionism and self-compassion (i.e., treating oneself with kindness) may affect coping and mental health for people with chronic illnesses. In ME/CFS, high levels of perfectionism are reported, with theories suggesting that perfectionism worsens coping and mental health difficulties. Self-compassion is linked to better coping and mental health in other illnesses. However, people who score highly on perfectionism struggle to treat themselves with kindness when faced with a past experience of perceived failure.

Firstly, this thesis investigated how different ways of coping relate to psychological distress (e.g., depression and anxiety) in ME/CFS. Section I includes a meta-analysis study which summarised past research looking into the relationships between different types of coping strategies and psychological distress in ME/CFS. Findings showed that using adaptive emotion-focused coping strategies were associated with lower levels of psychological distress. Adaptive emotion-focused coping strategies refer to changing one's emotional response to a stressful situation in a positive way, such as accepting life is now different from how it was pre-illness and coming to terms with this change. This result suggested that adaptive emotion-focused strategies may be helpful in managing mental health difficulties for people with ME/CFS. Study limitations are discussed when interpreting the results.

Section II includes a study that investigated whether asking individuals to respond self-compassionately to past experiences of perceived failure increases self-compassion, and if

perfectionism influences how able someone is to be self-compassionate. Participants with self-reported ME/CFS completed questionnaires measuring key factors including self-compassion, perfectionism and mental health difficulties in an online survey. Participants then completed a task where they recalled a past experience of perceived failure before being either prompted to write about the event while trying to be kind to themselves (the self-compassion group) or asked to recall more facts about the event (the control group). A self-compassion questionnaire was completed again after the task.

Findings showed that self-compassion was associated with lower levels of perfectionism, depression and anxiety, and higher confidence in their ability to cope with having ME/CFS. A significant increase in momentary self-compassion after the task was found in the self-compassion group only. Perfectionism did not influence the effectiveness of the self-compassion prompting task. These findings suggest that in ME/CFS, higher levels of self-compassion are associated with better coping and mental wellbeing, and that self-compassion can be momentarily increased by asking people to write self-compassionately about past experiences of perceived failure. As there is little research in this area, more studies are needed to explore whether compassion-focused strategies are helpful in lessening distress for people with ME/CFS.

Acknowledgements

First and foremost, my deepest gratitude goes to all the participants who kindly gave their time to take part in and support this study. Without you, this project would not have been possible. Thank you to my supervisor, Dr Fuschia Sirois, for her encouragement and guidance throughout the process. Many thanks also to Prof Glenn Waller for providing support and feedback on my thesis.

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Table of Contents

Section I: Literature Review

A meta-analysis of the associations between coping strategies and psychological distress in adults with Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS).

Abstract

Objectives: The ability to cope with challenges posed by a chronic illness predicts the degree of psychological distress that an individual experiences. This meta-analysis aimed to provide insights into how different coping strategies relate to psychological distress in individuals with Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS).

Methods: A systematic review of seven electronic databases were conducted. Searches yielded nine eligible studies (total $N = 1159$). Applying research and theory, separate meta-analyses were conducted to investigate the relationships between psychological distress and 1) adaptive emotion-focused coping strategies; 2) maladaptive emotion-focused coping strategies; 3) problem-focused coping strategies.

Results: A random effects meta-analysis revealed a significant negative medium effect for the association between adaptive emotion-focused coping strategies and psychological distress, $r_{avg} = -0.377$, CI [-0.544, -0.181], $p < .001$. A non-significant negative and small effect was found for the association between problem-focused coping strategies and psychological distress, $r_{avg} = -0.112$ [CI −0.28, 0.07], $p = .220$. Low study numbers meant it was not possible to examine the association between maladaptive emotion-focused coping strategies and psychological distress or conduct moderator analyses to probe sources of significant heterogeneity identified between-studies. Quality appraisal revealed risk of bias in numerous studies.

Conclusions: This meta-analysis offers preliminary insights in terms of our understanding of coping in ME/CFS and suggests that individuals with ME/CFS experiencing higher levels of psychological distress may benefit from learning to use adaptive emotion-focused coping strategies. More research is needed to elucidate how maladaptive emotion-focused coping strategies relate to mental health outcomes in ME/CFS.

Key words: Myalgic Encephalomyelitis, Chronic Fatigue Syndrome, coping, psychological distress

Practitioner Points

- Individuals with ME/CFS experiencing psychological distress may benefit more from psychological interventions that specifically foster the use of adaptive emotion-focused coping strategies.
- Directly assessing coping strategy use with standardised tools may be beneficial in helping identify maladaptive coping strategies alongside sources of strength/adaptation, when providing psychological support to individuals with ME/CFS.

Introduction

Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) is a long-term illness with a central symptom of extreme fatigue that endures for at least six months (Fukuda et al., 1994). This illness is often accompanied by other debilitating symptoms, including post-exertional malaise, cognitive difficulties, chronic pain and flu-like symptoms, with heterogeneity in the type and severity of symptoms experienced and duration of illness reported (National Institute for Health and Care Excellence [NICE], 2021). There are ongoing disputes regarding whether ME and CFS are discrete or hybrid diagnoses (Jason et al., 2014); however, this review utilised the term ME/CFS to reflect current clinical guidelines (NICE, 2021). ME/CFS has a significant impact on psychosocial and physical functioning, with 25% of sufferers reported to be housebound (Pendergrast et al., 2016). The significant reduction in functioning present in ME/CFS is associated with high rates of disability, educational drop-out and unemployment (Dimmock et al., 2016; Jason & Mirin, 2021). Reduction in attainment can place financial strain on individuals and their families (Anderson et al., 2014; Brenna et al., 2021) and generate a loss of identity in ME/CFS (Waite & Elliot, 2021; Whitehead, 2006). Disruptions to relationships and loss of social networks are also commonly reported by people with ME/CFS (Anderson et al., 2012). Overall, ME/CFS presents individuals with a wide variety of daily functional limitations alongside longer-term challenges.

Regarding the detrimental impact of ME/CFS on daily life, it is unsurprising that research indicates a higher prevalence of psychological distress in individuals with ME/CFS relative to the general population and other chronic illness cohorts (Hvidberg et al., 2015; Kingdon et al., 2018). Psychological difficulties include elevated levels of depression and anxiety (Cella et al., 2013; Jannseen et al., 2015), and increased risk of suicide (Jason et al., 2006; McManimen et al. 2016). Despite the prevalence of ME/CFS estimated to be around 0.2% in the United Kingdom (Nacul et al., 2011), there is no consensus on the cause of ME/CFS and no established cure for this illness (Friedman et al., 2021; NICE, 2021). The heterogeneity and fluctuation of symptoms, lack of universally accepted disease aetiology and view held by some that ME/CFS is a psychosomatic

illness have led to individuals with ME/CFS often feeling stigmatised by medical professionals and the public (Spander & Allen, 2018). Stigma and perceptions of not being believed about their condition are a significant source of distress for individuals with ME/CFS (Blease et al., 2016; Geraghty & Esmail, 2016).

Considering the various sources of distress that individuals with ME/CFS face, successful management of such challenges will require the use of adaptive coping strategies. Individuals are required to cope with the distress triggered by illness symptomology, alongside distress caused by the subsequent negative psychosocial and financial consequences of this illness, within systems that are often viewed to exacerbate distress through stigma. As ME/CFS is persistent and in the absence of clear medical treatments, it is crucial to determine adaptive ways of coping with this illness and decreasing psychological distress for these individuals. In order to ensure effective therapeutic interventions for people with ME/CFS, increased understanding for how individuals with ME/CFS cope with distress in needed. Understanding coping specifically in the context of chronic illness is important in regards to the way we view the adaptiveness or suitability of coping.

Coping with Distress

Coping strategies can be defined as cognitive and behavioural responses to manage the potential threat posed by a stressful situation (Folkman & Lazarus, 1985). One of the key models of coping and stress is Lazarus and Folkman's (1984) transactional model. This model proposes that coping responses are shaped by the individual's appraisal of the level of threat posed by a situation and the perceived resources available to them to effectively cope with the stressful situation. Within this model, coping strategies are separated into problem-focused strategies (i.e., to actively change the stressful situation in some way) and emotion-focused strategies (i.e., changing the individual's emotional response to the stressful situation) (Folkman & Lazarus, 1985).

There is no widespread agreement regarding which coping strategies may be most effective in reducing psychological distress. Emotion-focused coping strategies can be adaptive or

maladaptive depending on the situation faced by the individual (Lazarus, 1993). Yet, a dominant perspective in the stress and coping literature is that emotion-focused coping processes are maladaptive (Carver et al., 1989; Roth & Cohen, 1986). This view is based on research that proposes that emotion-focused coping strategies endorse avoidance of managing distress and distract from seeking out more active ways of coping (Baker & Berenbaum, 2007).

However, later theoretical accounts suggest that problem-solving strategies may not be adaptive in situations where the individual perceives their situation as being beyond their control (Conway & Terry, 1992; Finkelstein-Fox & Park, 2019). Considering the two-process model of perceived control (Rothbaum et al., 1982), coping is either aimed at eradicating the threat/stressor (primary control) or aimed at controlling factors like one's psychological response to the stressor as it is understood that some factors are not within their control (secondary control). Emotion-focused coping strategies may therefore be more helpful in managing distress in situations with low perceived controllability over the outcome and where individuals cannot engage in primary control processes (i.e., problem-focused strategies). Therefore, the utility of problem-focused coping strategies appears to rely on the individual's perceptions of controllability over the stressful situation which they are facing (Finkelstein-Fox & Park, 2019). Reliance on emotion-focused coping strategies may be necessary if problem-focused strategies are perceived as inaccessible.

Coping Strategies in Chronic Illness

Applying the transactional theory of coping to chronic illnesses, coping strategies involve the use of cognitive and behavioural responses to manage the potential threat posed by an illness. However, the picture remains unclear regarding which coping strategies are more adaptive for coping with distress in chronic illness populations. Emotion-focused coping strategies which involve acceptance, using social support and reframing the illness or perceived set-backs in a positive light, are adaptive in diverse chronic illness cohorts (Dunkel-Schetter et al., 1992; Krzemińska & Kostka, 2021; Voth & Sirois, 2009). Other emotion-focused coping strategies like

avoidance or self-blame are maladaptive and increase psychological distress in varying chronic illness samples (Bombardier et al., 1990; Carver et al., 1993; Sirois et al., 2006; Voth & Sirois, 2009; Vriezekolk et al., 2011). Regarding the application of problem-solving coping strategies in chronic conditions, evidence indicates that seeking information about the illness and planning increases adaptive coping and wellbeing (Ransom et al., 2005; Tuncay & Musabak, 2015). In accordance with theory (Conway & Terry, 1992; Finkelstein-Fox & Park, 2019), emotion-focused coping strategies are associated with lower distress in situations perceived as uncontrollable in chronic illness groups like diabetes, Human Immunodeficiency Virus and fibromyalgia (Johnson et al., 2014; Park et al., 2001; Santoro et al., 2014). In contrast, applying problem-focused strategies in uncontrollable situations has been related to increased distress in osteoarthritis and general population samples (Forsythe & Compas, 1987; Rivard & Cappeliez, 2007). Emotion-focused strategies may therefore be more relied upon (and potentially more adaptive) in the context of chronic illness where individuals have low controllability appraisals over illness symptoms and consequences.

Regarding how coping strategies reduce distress, it is suggested that adaptive coping skills may alleviate the emotional distress associated with illness management through promoting mastery/self-efficacy (Carver, 1998). Therefore, individuals with maladaptive coping skills may have low confidence in their ability to cope with illness-rated difficulties (i.e., coping efficacy), which may lead to higher levels of distress and illness burden.

Coping with ME/CFS

There has been limited empirical focus devoted to how different ways of coping relate to psychological distress in ME/CFS. Considering that ME/CFS is an illness with an undetermined aetiology, limited consensus on treatments and daily fluctuations in symptoms, this may leave individuals with low controllability over stressful situations encountered. Indeed, individuals with ME/CFS commonly describe feeling unable to have control over their illness (Malterud & Taksdal,

2007) with low coping efficacy regarding management of illness-rated difficulties reported (Doerr et al., 2017). Therefore, perceived controllability may play a key role in the coping strategies applied in ME/CFS. Considering that emotion-focused coping strategies are associated with lower distress in situations perceived as uncontrollable in other chronic illness populations, utilising emotion-focused strategies may be more helpful in managing distress in ME/CFS.

The Current Review

The current meta-analysis aimed to synthesize research examining the associations between coping strategies (both emotion- and problem-focused strategies) and psychological distress in ME/CFS. Based on theory and extant evidence, emotion-focused coping strategies were separated into adaptive and maladaptive strategies and examined independently. Separate meta-analyses were conducted to investigate the associations of 1) adaptive emotion-focused coping strategies; 2) maladaptive emotion-focused coping strategies; 3) problem-focused coping strategies, with psychological distress. It was expected that adaptive coping strategies would be more strongly associated with lower psychological distress than maladaptive coping strategies. Considering control theory (Rothbaum et al., 1982) and applying this to a condition like ME/CFS where low perceived controllability is reported (Malterud & Taksdal, 2007), it was expected that problem-focused coping strategies may not be used as frequently compared to emotion-focused coping strategies. Therefore, it was expected that stronger associations between adaptive emotion-focused coping strategies and psychological distress will be found, relative to the association between problem-focused coping strategies and psychological distress.

Moderators that might impact the magnitude of the associations between coping strategies and psychological distress were also considered. The following potential moderators were identified a priori based on previous research: type of psychological distress; coping strategy measure used; participant age. These variables were selected considering that there is disagreement in past research regarding how age may affect coping abilities in adults (Folkman et al., 1987; Trouillet et

al., 2009) and as there are a wide variety of coping strategy measures present in the coping research field (Kato, 2015). Different types of psychological distress (e.g., anxiety, depression) may also relate to coping strategies in diverse ways. For instance, maladaptive emotion-focused coping strategies like 'self-blame' and 'rumination' may be more relied upon for people experiencing depression compared to other forms of psychological distress, as these strategies act to reinforce symptoms of low mood according to the cognitive theory of depression (Beck, 1967).

To the author's knowledge, this is the first meta-analysis to statistically synthesise research on the association between coping strategies and psychological distress in adults with ME/CFS. Statistically pooling existing research means more robust conclusions can be drawn about how different ways of coping could have specific implications for psychological wellbeing in this clinical population. Elucidating the relationship between coping and psychological distress is particularly relevant when considering interventions for decreasing distress in individuals with ME/CFS and whether certain coping strategies may conflict with/ diminish the therapeutic elements of such interventions.

Method

The review protocol was registered with The Open Science Framework on 30/11/2021 and is accessible from https://osf.io/gqhu5/?view_only=02b7833533ce45f5979f655c7a80deee. This review was conducted in accordance with the Preferred Reporting Items of Systematic reviews and Meta-Analyses (PRISMA) checklist (Appendix A; Page et al., 2021).

Search Strategy

The following electronic databases were searched: CINAHL, Web of Science, PsycINFO, PubMed, Cochrane Library, Google Scholar (first 10 pages) and Scopus. A search for unpublished research was also conducted using OpenGrey (http://www.opengrey.eu/search/). The search was performed in November 2021, with forward and backward searches performed within selected studies in December 2021 and database auto alerts received from December 2021-April 2022.

Search terms were combined using the Boolean operators (AND / OR) and truncation (i.e., *), and adapted when searching PubMed to incorporate MeSH terms (Table 1). Search terms regarding coping strategies and psychological distress were constructed using identified coping strategies used within chronic illnesses in extant literature (Sirois et al., 2015) and prevalent mental health difficulties reported in ME/CFS (Cella et al., 2013).

Table 1.

Search terms used.

Concept 1: ME/CFS	Concept 2: Coping strategies	Concept 3: Psychological distress
"Chronic fatigue syndrome" OR CFS OR ME OR "Myalgic encephalomyelitis" OR CFS/ME OR "post viral fatigue syndrome" OR PVFS OR "chronic fatigue"	Cop* OR coping strateg* OR adaptive cop* OR maladaptive cop* OR self-blam* OR denial OR "social support" OR problem-focused OR emotion-focused OR self-car* OR pacing OR "disengag*" OR "escape avoidance" OR distancing OR confrontat* OR accept* OR "accepting responsibility" OR reappraisal	"Psychological distress" OR stress OR distress OR depression OR anxiety OR "mental health" OR "negative affect" OR "mood disorder" OR PTSD OR "post-traumatic stress disorder"

Note. *indicates that the specified term may form part of another word.

Inclusion & Exclusion Criteria

The inclusion and exclusion criteria are fully outlined in Table 2 using the PICO framework (*Population, Intervention, Comparator, Outcomes*; Higgins et al., 2019). Only studies that reported quantitative, cross-sectional data pertaining to the relationship(s) between coping strategies and psychological distress in ME/CFS samples were included. The following types of publications were excluded: papers not published in English, systematic reviews, and conference abstracts or posters. Unpublished theses were considered but no eligible studies were found.

Table 2.

Inclusion and exclusion criteria.

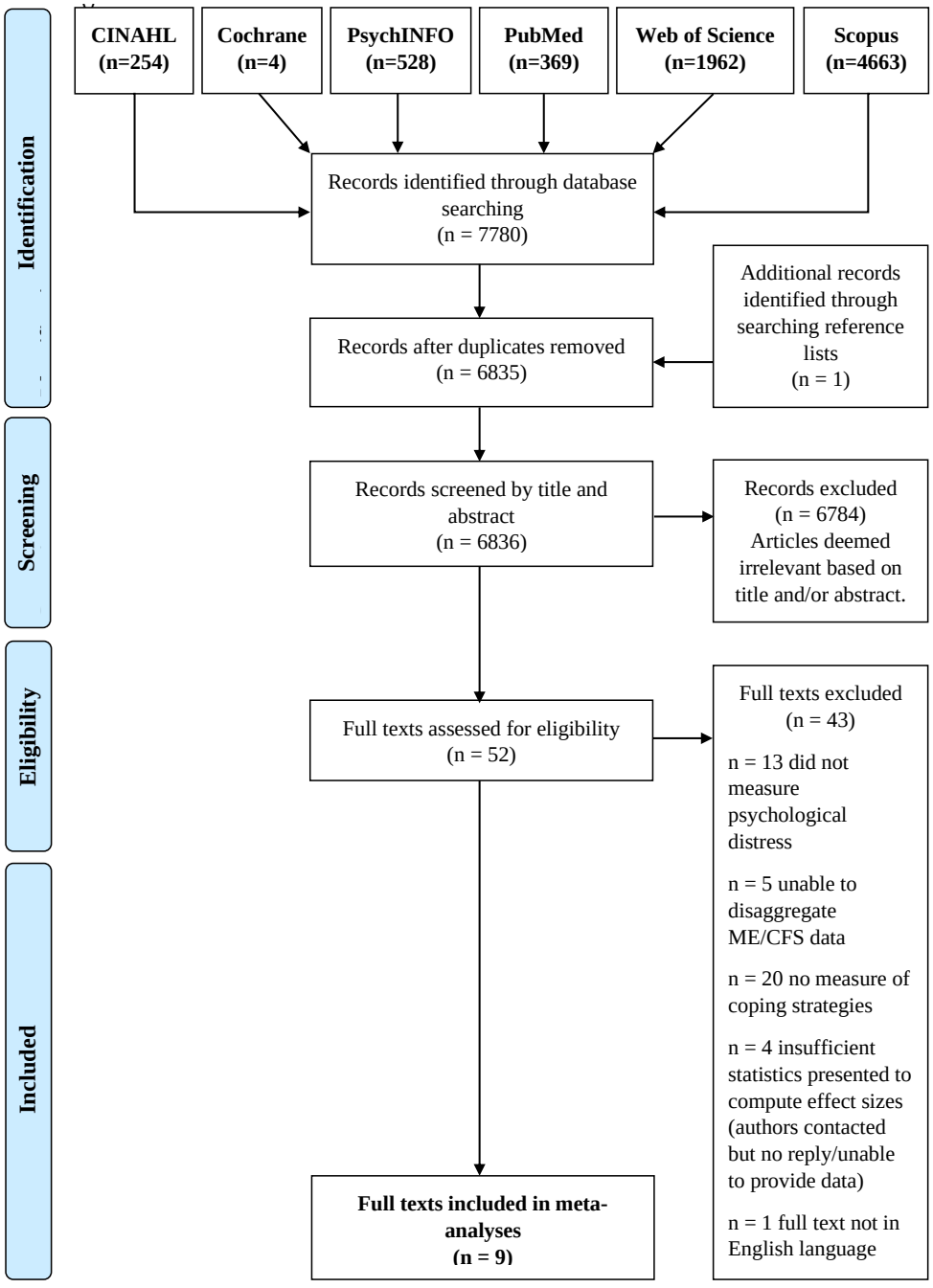
PICO domain	Inclusion criteria	Exclusion criteria
Population	Adults (>18 years old) with a current self-reported diagnosis of ME/CFS provided by a medical professional. No restriction on illness duration implemented.	Paediatric ME/CFS data that cannot not be disaggregated from adult ME/CFS data. Data from additional samples (e.g., other chronic health conditions or controls) that cannot be disaggregated from the ME/CFS participants.
Intervention	N/A.	N/A.
Comparator	N/A.	N/A
Outcomes	At least one quantitative measure of a psychological distress, e.g., psychometric measures of anxiety, depression or post-traumatic stress. At least one quantitative measure of a coping strategy, with coping strategies defined by existing theoretical accounts of coping and stress (Lazarus and Folkman, 1984).	Studies measuring outcomes other than those specified. This includes studies which measure wellbeing as opposed to distress (i.e., where higher scores on measures indicate better mental health outcomes).

Data Selection

Titles and abstracts of retrieved papers were screened against the inclusion criteria to identify relevant papers. Full text articles were subsequently assessed for eligibility. Searches generated a total of 7780 papers (CINAHL= 254, PsycINFO= 528, PubMed= 369, Scopus= 4663, Cochrane Library= 4, Web of Science= 1962). After de-duplication, 6835 papers remained. Forward and backward searches within selected studies yielded 1 paper. Initial screening and full text eligibility processes are presented in Figure 1. Nine studies were selected for meta-analyses.

Figure 1.

PRISMA diagram adapted.



Data Extraction

Data extraction was completed based upon the good practice data collection form by the Cochrane Effective Practice and Organisation of Care Group (EPOC, 2017; Appendix B). Relevant demographic, methodological and statistical data (including study design, participant numbers and

characteristics, measurement methods and associations between coping strategies and psychological distress) were extracted. Identified coping strategies were classified as 'emotion-focused' or 'problem-focused' dependent on how the study authors had categorised them and taking into consideration previous research. Within the category of emotion-focused strategies, these were classified as 'adaptive' or 'maladaptive' again based on how the study authors had defined them and considering previous theory and research within this field. If multiple analyses were reported in the same study, bivariate analyses examining associations between measures of coping and psychological distress were extracted. Where a univariate effect size was not reported, the study authors were contacted. When determining full text eligibility, four studies did not provide adequate statistics to compute effect sizes and were contacted by the researcher. Three authors did not respond and the one author that did respond was unable to provide the requested information. Therefore, these four studies were not included in the total number of included studies.

Quality Appraisal

The National Institute for Health and Care Excellence (NICE) quality appraisal checklist for quantitative studies reporting correlations and associations (NICE, 2012; Appendix C) was utilised. This tool was selected as it was anticipated that due to the nature of the research question, eligible studies would have observational study designs. Alternative tools do not appraise various observational study designs and are limited in the number of study designs they can evaluate, e.g. the Critical Appraisals Skills Programme (CASP) checklists (CASP, 2018), or only apply to intervention studies, e.g. the Downs & Black checklist (Downs & Black, 1998). While formal assessment of the reliability of the NICE (2012) tool has not been undertaken, it is based on the appraisal step of the Graphical Appraisal Tool for Epidemiological Studies (Jackson et al., 2006) - a validated tool with good interobserver reliability (Fitzgerald & Coop, 2011).

Risk of bias is computed in terms of internal validity (14 items), external validity (three items) and summary of validity (two items). Each study was given a rating for every item using the

following scoring system: ++ (none or minimal risks of bias exist); + (some sources of bias evident); - (significant sources of bias exist); NR (not reported); NA (not applicable). A second researcher (a trainee clinical psychologist at the University of Sheffield) independently reassessed one third of the papers (n= 3) selected at random. Following discussion, a 100% level of agreement per study was observed, compared to an 80.7% level of agreement pre-discussion.

Data Synthesis

Separate meta-analyses were conducted, to investigate the relationships between 1) adaptive emotion-focused coping strategies and psychological distress experienced and 2) problem-focused coping strategies and psychological distress experienced. As only two studies identified included measurement of maladaptive emotion-focused strategies, it was not possible to statistically analyse the association between maladaptive emotion-focused coping strategies and psychological distress.

The correlation coefficient *r* was the effect size metric used in all included studies and was therefore the chosen metric for synthesising the effects. Comprehensive MetaAnalysis (CMA) was used to run a random effects model. CMA converts all effect sizes into Fisher's *z* (Hedges & Olkin, 1985) to calculate an integrated effect size. Applying recommendations by Card (2012), weighted averages were calculated using CMA where multiple effect sizes were reported in one study (e.g., where studies reported relationships between psychological distress and multiple types of coping strategies). Applying this method, an overall effect size was created for each category of coping strategy (i.e., adaptive emotion-focused; problem-focused). Cohen's (1992) guidelines were used when determining the strength of effect sizes in the meta-analyses, with effect sizes of $r = 0.10$ considered small, $r = 0.30$ considered medium and $r = 0.50$ considered large. An alpha value of <0.05 was utilised for determining statistical significance in accordance with convention (Borenstein et al., 2021) and data are presented regarding 95% confidence intervals of the effect size. Forest plots to visualise effect sizes and confidence intervals were generated.

Heterogeneity was assessed using the *Q* and I^2 statistic. An I squared value of 0% indicates no observed heterogeneity, 25% indicates low heterogeneity, 50% indicates moderate heterogeneity and 75% indicates substantial heterogeneity (Higgins et al., 2003). Cochran´s *Q* statistic was used to test whether the degree of heterogeneity between-studies was significant ($p < .05$). Moderation analyses were conducted where tests of heterogeneity were significant. Three potential moderators were identified a priori based on included study characteristics and previous research: type of psychological distress; coping strategy measure used; participant age. Sub-group moderation analyses were run where variables were categorical (coping strategy measure used; type of psychological distress) and were only performed if there were ≥ 3 studies per group (in accordance with Card, 2012). Meta-regression was considered for continuous moderators (i.e., participant age), but was not computed as there were <10 studies.

Studies with large effect sizes are more likely to be published which increases the likelihood of bias in studies included in meta-analysis, termed 'publication bias' (Quintana, 2015). A multi-pronged approach was taken for assessing publication bias as recommended by Card (2012). This approach included using a funnel plot (to visualise standard errors vs. effect sizes), with the trim-and-fill method (Duval & Tweedie, 2000) and Eggers' test (Egger et al., 1997) used where the funnel plot was asymmetrical. An unbiased sample of studies would consist of a collection of data points that is symmetric around the population effect size (Field & Gillett, 2010). The fail-safe *N* test (Rosenthal, 1979) was used to calculate the number of studies with nonsignificant findings necessary to make the overall effect size nonsignificant. Rosenthal (1979) proposed a fail-safe N value greater than $5k + 10$ as supporting absence of publication bias.

Results

Study Characteristics

Sample mean ages ranged from 32.13- 50.70 years and notably had a high proportion of female participants (68.70-93%; see Table 3). All of the included studies had a cross-sectional

design. Of the nine studies included, four were conducted in the United Kingdom (UK), with the remaining studies taking place in different Western countries across the world. All studies used self-report scales to measure coping strategies and psychological distress. The most frequently used measure of psychological distress was The Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983; $k = 5$). The measure of coping strategies used most often was The Illness Management Questionnaire (Ray et al., 1993) for problem-focused coping ($k = 3$) and The COPE (Carver et al., 1989) for emotion-focused coping ($k = 2$). However, a variety outcome measures were used. Two studies (Ray, 1992; Ray et al., 1993) used novel coping strategy measures, with the assessment of the psychometrics of these measures included in the papers.

Table 3.

Characteristics of included studies.

Study Characteristics			Participant Characteristics			Outcomes & Measures			
Authors & date	Location	n	Age (Mean; SD; Range)	Female (%)	Mean time since illness onset (SD)	Type & measure of psychological distress	Coping strategies measure	Emotion-focused coping strategies	Problem-focused coping strategies
Knussen & Lee (1998)	UK	81	Range = 15-65 years; mean and SD not reported	71%	5.3 years (SD= 4.4)	Emotional disturbance: [a]PFRS	The [b]IMQ	Not Assessed	1) Maintaining activity 2) Illness accommodation 3) Focusing on symptoms 4) Information seeking
Kraaij et al. (2019)	The Netherlands	30	32.13 years (SD= 10.65); range not reported	93%	53% reported having ME/CFS for over 5 years	Depression: [c]HADS	1) [d]CERQ - 9 subscales used. 2) [e]COPE - 2 subscales used.	CERQ: 1) Acceptance 2) Self-blame 3) Rumination 4) Catastrophising 5) Other-blame 6) Positive refocusing 7) Positive reappraisal 8) Putting into perspective. COPE: 1) Use of emotional support.	CERQ: 1) Refocus on planning. COPE: 1) Active coping

Lattie et al. (2013)	USA	117	50.7 years (SD and range not reported)	72%	Not reported	Emotional distress composite score computed by summing the z-scores of the following scales: [f]PSS; [g]CES-D; [h]POMS (depression-dejection and anxiety-tension	[i]MOCS	Not Assessed	Stress management skills
Poppe et al. (2012)	The Netherlands	117	38.05 years (SD= 9.97; ranging from 16-56 years)	86.30%	5.18 years (SD= 6.58)	Mental health quality of life: [j]SF-36	[k]ICQ	Accommodative coping - acceptance	Not Assessed
Ray (1992)	UK	207	39 years (SD and range not reported)	72%	4 years (SD not reported)	Depression & Anxiety: [c]HADS	Novel measure of social support created by authors.	Social support	Not Assessed
Ray et al. (1993)	UK	207	39.02 years (SD= 10.69). Range not reported.	72%	4 years (SD not reported)	Depression & Anxiety: [c]HADS	The [b]IMQ	Not Assessed	1) Maintaining activity 2) Illness accommodation 3) Focusing on symptoms 4) Information seeking

Ray et al. (1995)	UK	147	36.85 years (SD= 9.45). Range not reported.	68.70%	39.61 months (SD= 17.82)	Depression & Anxiety: [c]HADS	1) The [b]IMQ 2) [e]COPE - 4 problem-focused coping strategy subscales only.	Not Assessed	IMQ: 1) Maintaining activity 2) Illness accommodation 3) Focusing on symptoms 4) Information seeking. COPE: 1) Planning 2) Suppression of activities 3) Active coping 4) Restraint coping 5) Information support
Van Damme et al. (2006)	Belgium	97	40.06 years (SD= 8.36; range= 21-58 years)	80.41%	7.89 years (SD= 6.30; range= 1-26 years)	Overall emotional distress: [c]HADS	[k]ICQ	Acceptance	Not Assessed
Walker et al. (2009)	Australia	156	43.5 years (range= 18-72 years)	77.56%	Not reported	Depression: [l]CDS	[m]WOCQ	1) Seeking social support 2) Positive reappraisals 3) Confrontative 4) Self-controlling 5) Escape avoidance 6) Accepting responsibility	1) Distancing 2) Planful problem-solving

[a] = The Profile of Fatigue-related Symptoms (PFRS; Ray et al., 1992). [b] = The Illness Management Questionnaire (IMQ; Ray et al., 1993).[c] = The Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983). [d] = The Cognitive Emotion Regulation Questionnaire (CERQ; Garnefski et al., 2001). [e] = The COPE (Carver et al., 1989). [f] = The Perceived Stress Scale (PSS; Cohen et al., 1983). [g] = The Center for Epidemiologic Studies Depression Scale (CES-D; Radloff, 1977). [h] = The Profile of Mood States (POMS; McNair et al., 1971). [i] = The Measure of Current Status (MOCS; Carver, 2006). [j] = Short Form Health Survey (SF-36; Ware & Sherbourne, 1992). [k] = Illness Cognitive Questionnaire (ICQ; Evers et al., 2001). [l] = Cardiac Depression Scale (CDS; Hare & Davis, 1996). [m] = Ways of Coping Questionnaire (WOCQ; Folkman & Lazarus, 1986).

Associations between Coping Strategies and Psychological Distress

Extracted data and overall effect sizes for each study are presented in Table 4. There were six papers in the analysis testing the association between problem-focused coping strategies and psychological distress (*N* = 738; Figure 2) and five in the analysis testing adaptive emotion-focused coping strategies and psychological distress (*N* = 607; Figure 3). As noted above, there were only two studies testing maladaptive emotion-focused coping strategies and psychological distress (*N* = 186) and so this association was not meta-analysed. Figure 2 shows a non-significant negative and small effect size for the association between the use problem-focused coping strategies and psychological distress was found, r_{avg} = −0.112 [CI −0.28, 0.07], *p* = .220. However, Figure 3 shows a significant negative and medium sized effect between the use of adaptive emotion-focused coping strategies and psychological distress was found, r_{avg} = -0.377, CI [-0.544, -0.181], *p* < .001.

Table 4.

Meta-analysed effect sizes across studies.

Study	n	Psychological distress and adaptive emotion-focused coping strategies *R* [CI]	Psychological distress and problem-focused coping strategies *R* [CI]
Knussen & Lee (1998)	81	n/a	0.001 [-0.217-0.219]
Kraaij et al. (2019)	30	-0.254 [-0.563-0.117]	-0.176 [-0.504-0.197]
Lattie et al. (2013)	117	n/a	-0.460 [-0.592—0.304]
Poppe et al. (2012)	117	-0.590 [-0.697—0.457]	n/a
Ray (1992)	207	-0.250 [-0.374—0.118]	n/a
Ray et al. (1993)	207	n/a	0.100 [-0.037-0.233]
Ray et al. (1995)	147	n/a	-0.039 [-0.200-0.124]
Van Damme et al. (2006)	97	-0.540 [-0.668—0.382]	n/a
Walker et al. (2009)	156	-0.167 [-0.314—0.112]	-0.099 [-0.252-0.059]

Note. CI = confidence interval; n = participant numbers.

Figure 2.

Forest plot for the effect sizes between problem-focused strategies and psychological distress.

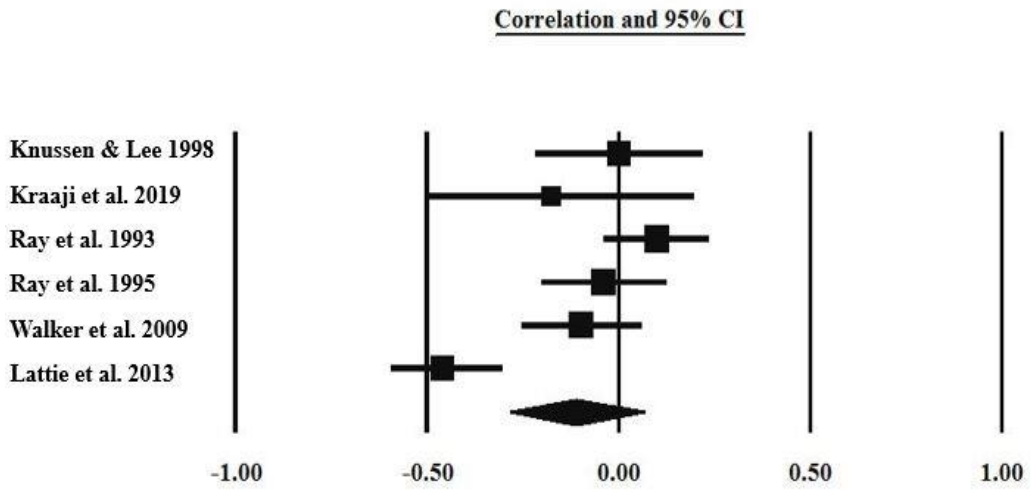
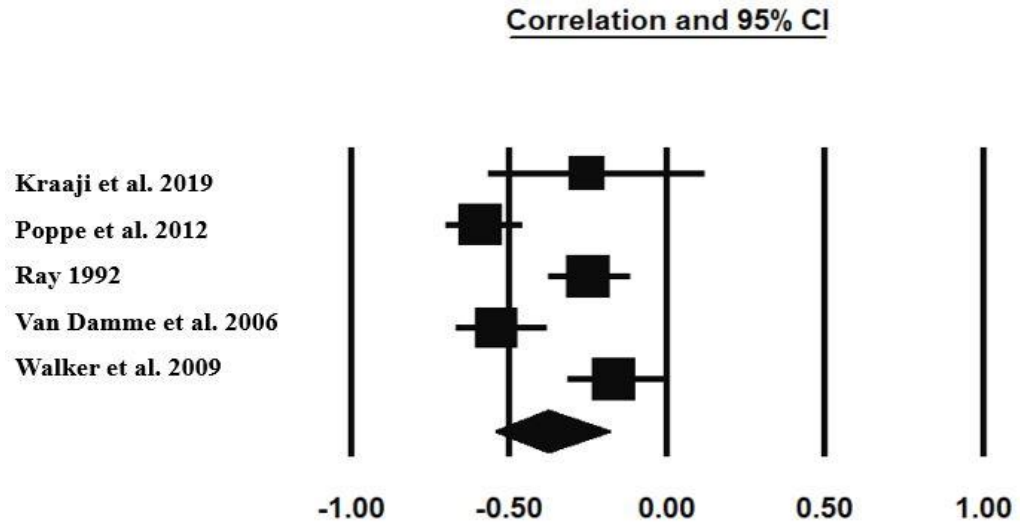


Figure 3.

Forest plot for the effect sizes between adaptive emotion-focused strategies and psychological distress.



Heterogeneity Analysis

Tests of heterogeneity of the effect sizes were significant for both problem-focused coping strategies–psychological distress, $Q(5) = 27.53$, $p < .001$; $I^2 = 81.84$, and adaptive emotion-focused

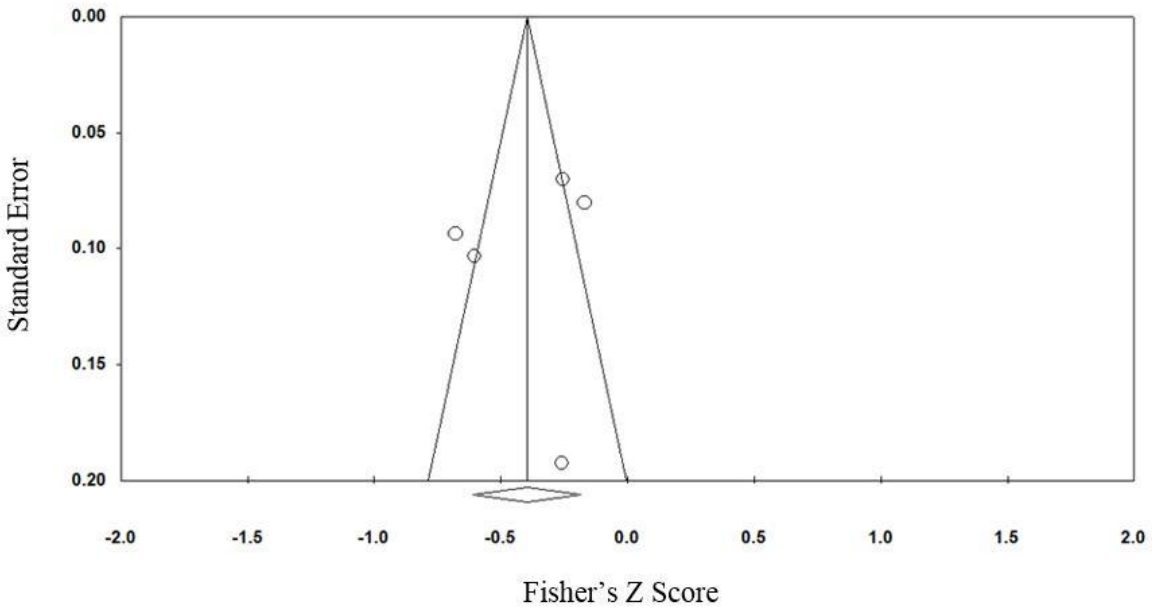
coping strategies-psychological distress, $Q(4) = 25.28$, $p < .001$; $I^2 = 84.18$. The I^2 values for both analyses of coping strategies with psychological distress were above 75%, suggesting substantial between-study heterogeneity. As there were fewer than three studies per sub-group, moderation tests were not feasible and therefore were not conducted.

Analysis of Publication Bias

Examining adaptive emotion-focused coping strategies with psychological distress, the fail-safe *N* analysis indicated that 102 studies with null results would need to be added to the analysis to yield a statistically non-significant ($p < 0.05$) overall effect. This was well above the threshold value of 35 studies (Rosenthal; 1979). The funnel plot was fairly symmetrical (Figure 4), although, 2 studies fell outside of the funnel area. The trim-and-fill test resulted in no studies needing to be trimmed and the Egger's test was also non-significant, (t (3) = 0.508, p = .646). Together, these results suggested the absence of publication bias within studies included in the meta-analysis of the association between emotion-focused coping strategies and psychological distress.

Figure 4.

Funnel plot for emotion-focused coping strategies and psychological distress.



Risk of Bias

Results of quality appraisal are presented in Table 5. Internal and external validity were both problematic regarding bias. Two studies received an overall rating of 'significant bias' for both internal validity (Ray, 1992; Ray et al., 1995). Six studies received an overall rating of 'significant bias' for external validity (Knussen & Lee, 1998; Kraaij et al., 2019; Ray, 1992; Ray et al., 1993; Ray et al., 1995; Van Damme et al., 2006). The core justifications for receiving poorer ratings of internal validity were no/minimal effort to minimise selection bias, inadequate reporting of statistics and consideration of whether sufficient power had been achieved. Studies generally failed to consider or account for confounding variables, e.g., illness duration/severity. Studies that received poor ratings on external validity had failed to outline clear inclusion/exclusion criteria and provided insufficient information regarding their recruitment process, meaning that it was difficult to ascertain if the eligible population was representative of the source population.

Strengths noted across almost all studies included the use of multiple explanatory variables in the analyses, providing a strong theoretical rationale for predictor and dependent variables, adequate sample demographics, and using validated outcome measures. Notably, Walker et al. (2009) received ratings of minimal risk of bias for both internal and external validity. Ratings of 'Not Applicable' were assigned to all studies for three items which probed methodological factors not relevant to the study designs of included papers, e.g., examining follow-up periods and between-group comparisons.

Table 5.

Quality appraisal judgements.

	External Validity						Internal Validity										Summary	
Study	Sample Description	Sample Generalisability	Sampling Bias	Selection Bias	Theoretical Basis for Variables	Contamination Bias	Confounders	Study Setting Applicability	Outcome Measure Reliability	Outcome Measure Completion	Outcomes Assessed	Assessment of Follow-up Phases		Statistical Power	Variables Analysed	Data Analysis Methods	Statistics Presented	Overall Internal Validity	Overall External Validity
1. Knussen & Lee (1998)	-	+	-	+	++	NA	+	++	+	-	+	NA	NA	-	++	+	-	+	-
2. Kraaij et al. (2019)	+	-	-	-	++	NA	-	-	++	-	+	NA	NA	-	++	+	-	-	-
3. Lattie et al. (2013)	+	+	-	-	++	NA	+	-	++	+	+	NA	NA	+	++	++	+	+	+
4. Poppe et al. (2012)	+	+	-	-	++	NA	++	-	++	++	+	NA	NA	+	++	++	-	+	+
5. Ray (1992)	-	NR	NR	NR	-	NA	NR	++	-	NR	+	NA	NA	+	+	+	-	-	-
6. Ray et al. (1993)	+	-	-	NR	+	NA	NR	++	-	+	+	NA	NA	+	+	+	-	+	-
7. Ray et al. (1995)	+	-	+	-	++	NA	-	++	+	++	+	NA	NA	+	+	-	-	-	-
8. Van Damme et al. (2006)	+	-	-	NR	++	NA	+	+	+	+	+	NA	NA	-	++	+	+	+	-
9. Walker et al. (2009)	++	++	+	+	++	NA	++	++	++	+	+	NA	NA	+	++	++	+	++	++

Note. ++ =no or minimal risks of bias exist; + =some sources of bias evident; - =significant sources of bias exist; NR =not reported; NA =not applicable.

Discussion

This is the first study to statistically synthesise research on the association between coping strategies and psychological distress in adults with ME/CFS. A significant medium negative association was found between psychological distress and adaptive emotion-focused coping strategies. This finding suggests that greater use of adaptive emotion-focused coping strategies is linked to lower levels of psychological distress in the included samples. In contrast, problem-focused coping strategies were not significantly associated with psychological distress. These findings therefore offered support for the study hypothesis in that stronger associations between adaptive emotion-focused coping strategies and psychological distress were found, relative to the association between problem-focused coping strategies and psychological distress.

Collectively, these findings are consistent with existing theories of control and coping in chronic illness. Applying Lazarus and Folkman's (1984) transactional model, coping responses are influenced by the individual's appraisal of the level of threat posed by an illness and the perceived resources available to them to enable them to effectively cope with the stressful situation. The 'goodness-of-fit' hypothesis (Finkelstein-Fox & Park, 2019) extended this theoretical account further in proposing that an individual's appraisal of the level of controllability they have over their illness/situation plays a fundamental role in determining which coping strategies are used. Therefore, adaptive emotion-focused coping strategies, like acceptance and seeking social support, may be more beneficial for individuals in situations where they have perceived low levels of control compared to problem-focused strategies (Lazarus & Folkman, 1984). Indeed, Moss-Morris et al. (1996) found that individuals with ME/CFS who strongly believed their condition was outside of their control reported using more emotion-focused coping strategies. In contrast, individuals who felt that they had more control over their illness reported using more problem-focused coping strategies. Applying this previous literature and theoretical perspectives to the results of the current study, perhaps adaptive emotion-focused coping strategies are more helpful in managing

psychological distress in ME/CFS due to the low perceived controllability that often accompanies this illness (making it more challenging to engage in problem-focused coping strategies).

Additionally, problem-focused coping strategies by nature involve the individual actively changing a stressful situation that they face (Folkman & Lazarus, 1985). In the context of a chronic illness like ME/CFS where there is perceived or actual low control over situations related to one's illness, problem-focused strategies may not be realistically possible. Applying the two-process model of perceived control (Rothbaum et al., 1982), there are argued to be two coping approaches where coping is either aimed at eradicating the threat/stressor (primary control) or aimed at controlling factors like one's psychological response to the stressor as it is understood that some factors are not within their control (secondary control). Relating this theory to ME/CFS, individuals may be left with relying on secondary control processes (like managing emotional responses to stress) as primary control processes are not possible, e.g., it is not possible to eradicate their illness symptoms. The findings of the current study align with this theory as there was a significant association between emotion-focused coping strategies and psychological distress, which was not observed for problem-focused coping strategies. Therefore, perhaps individuals with ME/CFS are having to engage in more secondary control processes relative to primary control processes, with secondary control processes relying more heavily on emotion-focused coping strategies. If individuals with ME/CFS are unable to take direct action to eradicate problems, i.e., they are unable to control their symptoms of fatigue in order to use a problem-focused strategy, this may mean that the only factor that they are able to control is their emotional response to the problem.

Furthermore, adaptive emotion-focused coping strategies identified in the current study were also consistent with strategies characterized as adaptive in previous chronic illness research, such as 'social support' and 'acceptance' (Dunkel-Schetter et al., 1992; Krzemińska & Kostka, 2021). The current study highlighted that emotion-focused strategies like acceptance and social support may be beneficial in alleviating psychological distress in people with ME/CFS. Acceptance in chronic illness has been defined as an individual acknowledging that their life is now different from their

pre-illness situation and coming to terms with this change (Hayes et al., 1999). Qualitative research has demonstrated that individuals with ME/CFS who reported greater acceptance of the reality of their condition felt better able to engage with fatigue management strategies like pacing, felt more confident in rebuilding their pre-illness identities, and experienced greater perceived control over their lives (Pinxsterhuis et al., 2015; Wilson et al., 2011). Therefore, 'acceptance' may be highly adaptive in ME/CFS due to their promoting psychological adjustment and perhaps subsequently allowing individuals to engage with problem-focused strategies. Research has also found that ME/CFS is often accompanied by a reduction in social networks and an increased experience of loneliness (Boulazreg & Rokach, 2020; Schweitzer et al., 1995), with social isolation well documented as a prevalent maintaining factor for mental health difficulties (Leigh-Hunt et al., 2017). Therefore, 'seeking social support' may be a highly relevant coping strategy in ME/CFS, increasing social connectedness and subsequently reducing psychological distress.

Notably, there was significant substantial heterogeneity found between studies for both meta-analyses. Low study numbers meant it was not possible to test hypotheses regarding possible sources of the heterogeneity observed. Sources of between-study heterogeneity can be hypothesized from visible differences between-studies and considering theoretical perspectives and past research. Regarding possible sources of this heterogeneity, there were evident differences between-studies in sample size and characteristics. Several studies recruited participants from hospital outpatient settings while others recruited more widely via online settings, which may have led to variability in ME/CFS symptom severity across samples. Notably, there is a large degree of heterogeneity in symptoms and symptom severity in this population (White, 2019). Heterogeneity in illness severity results in differences in how in control of their illness individuals feel in ME/CFS (Arroll, 2009). Therefore, individuals with less severe impairment may be better able to actively make changes to stressors (i.e., engage in problem-focused coping), compared to individuals with more severe impairment. Heterogeneity in ME/CFS symptoms could also explain variation in effect sizes across

included studies and the consequent absence of an association between psychological distress and problem-focused coping strategies in this meta-analysis.

Moreover, there was wide variety in measures and types of coping strategies assessed. As outlined in the introduction when a priori moderators were identified, distinct strategies within each branch of coping strategy (emotion-focused; problem-focused) may relate to psychological difficulties in different ways. Furthermore, the construct of psychological distress included measurement of depression, anxiety, and generic emotional distress. Therefore, differences between studies in outcome variables may also account for the heterogeneity found across included studies.

Strengths and Limitations

The current findings should be considered in light of the following strengths and limitations. A strength of the present study is that the meta-analyses were conducted in accordance with PRISMA guidance (Page et al., 2021), to enhance study methodology and reporting quality. Additional study strengths include that a formal data extraction form was used, and quality appraisal judgments were conducted using a validated tool with inter-rater reliability verified. Using standardised forms and tools improved the validity and reliability of the systematic review process in the current study.

Regarding study limitations, included studies were all cross-sectional design. Therefore, the direction of causality between coping strategies used and psychological distress cannot be determined. Furthermore, only one researcher was involved in the data screening and extraction process. Having additional researchers to determine eligible studies and perform data extraction would have reduced the risk of any bias and human error. Quality appraisal revealed substantial risk of bias for internal and external validity within studies, indicating that the results of the current study should be tentatively interpreted.

As highlighted in the quality appraisal, included studies often failed to report detailed sample demographics and recruitment strategy. The majority of included studies were also >15

years old, with ME/CFS diagnostic classification and clinical guidance having changed considerably since these studies were published (NICE, 2021). This reduces the generalisability of the current study's results to wider samples. Additionally, the location of studies may have impacted on the coping strategies available to individuals. Over half of included studies were conducted outside of the UK in countries where residents are required to pay for medical care and have different social care systems. Therefore, the financial impact of having ME/CFS and potentially being unable to maintain employment may be greater for individuals in these non-UK samples. Financial stress may mean that certain coping strategies are less accessible to these individuals. For example, problem-focused coping strategies like 'illness accommodation' and 'suppression of activities' may be more difficult to use if an individual's circumstances mean they must persevere with employment despite their illness. Therefore, again results found in the current meta-analyses may have limited generalizability beyond the specific samples included.

Notably, there was a total of only nine studies included in this review. Between-study differences could not be statistically tested and therefore remain undetermined. Averaging the effect sizes and selecting one effect size per study to be included is consistent with commonly applied methods for conducting meta-analysis (Card, 2012). However, this may have been problematic in the current study where studies reported multiple relationships between psychological distress and different strategies within the branches of emotion-focused and problem-focused coping strategies.

Considering study limitations, the results of the meta-analyses were therefore cautiously applied when considering clinical implications and avenues for future research. Nonetheless, testing the associations between psychological distress and coping strategies in ME/CFS helps to build an evidence base in a particularly understudied area within chronic illness research. It is valuable to conduct small-scale meta-analyses to give preliminary insight into understudied research topics (Cumming, 2012).

Clinical Implications

This review highlights the utility in more routine measurement of coping strategies within healthcare services when working with individuals with ME/CFS. Directly assessing coping strategy use with standardised tools may be beneficial in helping identify maladaptive coping strategies alongside sources of strength/adaptation. This information may enhance psychological assessment and formulation, meaning that interventions to manage psychological distress could be more person-centred to the current coping capabilities of the individual.

In the current study, adaptive emotion-focused coping strategies were found to be negatively associated with psychological distress. This finding suggests that individuals with ME/CFS experiencing higher levels of psychological distress may be the most in need of learning to use adaptive emotion-focused coping strategies. The results indicated no relationship between distress and problem-focused coping, which suggests problem-focused coping strategies may be less useful in ME/CFS. Interventions fostering the use of adaptive emotion-focused strategies measured within included studies, like 'acceptance' and 'positive reappraisal', may therefore be more beneficial in reducing psychological distress in ME/CFS. Clinical guidelines recommend using CBT for managing psychological distress in ME/CFS (NICE, 2021). Considering how identified emotion-focused coping strategies align with this therapeutic approach may be helpful in ensuring treatment efficacy and person-centred intervention. Using alternative therapeutic approaches that more acutely tap into adaptive emotion-focused coping strategies may be beneficial in reducing psychological distress for these individuals. Alternative interventions could include Acceptance and Commitment Therapy (ACT; Hayes et al., 2004) which specifically fosters coping through acceptance or Compassion-Focused Therapy (CFT; Gilbert, 2009) which promotes coping through self-compassionate reappraisal (Gilbert, 2005; Harris, 2006).

Future Research Directions

The current study highlights the limited empirical evidence on how specific coping strategies may be important for cultivating psychological wellbeing in adults with ME/CFS. More

research is needed to further elucidate the associations between coping strategies and psychological distress in ME/CFS, to offer clarification for which emotion-focused strategies may be maladaptive specifically in ME/CFS. Obtaining a greater understanding of the adaptability of different emotion-focused coping strategies in ME/CFS may be beneficial for clinicians offering mental healthcare to these individuals, in informing psychological formulation and intervention. Equally, studies are needed which experimentally test whether interventions focused on increasing the use of adaptive emotion-focused coping strategies may be helpful in reducing psychological distress in ME/CFS.

It is likely that other factors besides controllability appraisals of stressors may influence whether coping strategies may be adaptive in managing psychological distress in chronic illnesses like ME/CFS. Evidence suggests that individual traits and qualities like self-compassion may influence which coping strategies are accessed and the efficacy of such strategies in managing distress (Allen & Leary, 2010). Self-compassion refers to adopting a kind, accepting, and non-judgmental stance towards oneself in times of failure and difficulty (Neff, 2003). This trait is suggested to foster adaptive as opposed to maladaptive coping strategies and reduce stress by enabling people to self-regulate negative emotions that may be triggered by unexpected or uncontrollable events (Neff et al., 2007). In research using path analysis models, self-compassion was associated with using more adaptive- and less maladaptive coping strategies, which in turn were linked to greater coping efficacy and lower perceived stress in people with arthritis and inflammatory bowel disease (Sirois et al., 2015). Notably, high prevalence of perfectionistic traits is reported in ME/CFS populations (Deary & Chalder, 2010), with perfectionism proposed to have a negative relationship with self-compassion through promoting self-criticism (Stoeber et al., 2020). Recent research demonstrated that participants who scored highly on perfectionism held greater negative beliefs about self-compassion and subsequently had difficulties in enacting self-compassion (Biskas et al., 2022). Therefore, while self-compassion may be a mechanism that promotes the use of adaptive coping in other illness populations, perhaps traits prevalent in

ME/CFS like perfectionism may interfere with this process. Future research is needed regarding how individual traits may influence coping and stress in ME/CFS.

Conclusions

This small-scale meta-analysis found that adaptive emotion-focused coping is negatively related to psychological distress in adults with ME/CFS. The use of problem-focused coping strategies was not associated with psychological distress. Together, these results contribute to theoretical accounts and previous research that suggest emotion-focused coping strategies may be more frequently used in illness cohorts where there is often low perceived controllability over their condition. These results offer preliminary insights in terms of our understanding of coping in ME/CFS and suggest that individuals with ME/CFS experiencing higher levels of psychological distress may be the most in need of learning to use adaptive emotion-focused coping strategies. More research is needed to elucidate how maladaptive emotion-focused coping strategies relate to mental health outcomes in ME/CFS. Directing greater empirical focus to this understudied research area is recommended, to ensure that mental health support can best meets the needs of individuals coping everyday with this chronic condition.

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Appendices

Note Appendices removed for copyright purposes.

Section II: Empirical Project

The role of perfectionism and self-compassion in psychological outcomes in individuals with Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS).

Abstract

Background: Perfectionism is associated with poor coping and distress in Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS). While self-compassion is linked to better coping and wellbeing in other illness cohorts, individuals high on perfectionism struggle to engage in self-compassion. This study investigated whether prompting individuals to respond self-compassionately to past challenges increases state self-compassion, and if this is moderated by perfectionism.

Method: Individuals with self-reported ME/CFS (n = 147) completed measures of self-compassion, perfectionism, coping efficacy, fatigue and psychological distress in an online survey. Participants then recalled a past event of perceived failure before randomly assigned to one of two groups. The self-compassion group were prompted to write about the event self-compassionately. The control group were asked to recall further details of the event. Both groups completed a measure of state self-compassion post-task.

Results: Trait self-compassion was negatively associated with perfectionism and psychological distress, and positively associated with coping efficacy. A significant increase in state self-compassion was noted in the self-compassion group ($p < .001$) but not in the control group. Perfectionism did not moderate the effect of self-compassionate prompting on state self-compassion.

Conclusions: Findings suggest that higher levels of self-compassion are associated with better coping efficacy and less psychological distress in individuals with ME/CFS and provide preliminary evidence that state self-compassion can be increased by prompting individuals to write self-compassionately about past experiences of perceived failure. More research is needed regarding whether compassion-focused strategies are beneficial for alleviating psychological distress and to further elucidate whether perfectionism influences self-compassion processes in ME/CFS.

Key words*:* Myalgic Encephalomyelitis; Chronic Fatigue Syndrome; self-compassion; perfectionism

Practitioner Points

- Self-compassion and perfectionism may be important factors for clinicians to consider during the assessment, formulation and intervention for psychological distress in people with ME/CFS.
- Directly assessing levels of perfectionism and self-compassion with standardised tools may be beneficial in helping identify self-critical cognitions and maladaptive behaviours that maintain distress in ME/CFS.
- Psychological interventions that specifically foster the use of self-compassion may be beneficial in alleviating psychological distress in this population.

Introduction

Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) is a long-term complex health condition with the central symptom of extreme fatigue (Fukuda et al., 1994; National Institute for Health and Care Excellence [NICE], 2021). Illness symptoms and severity are heterogenous, with there being no clear medical consensus on illness causes and treatments (Friedman et al., 2021). ME/CFS has a significant impact on psychosocial and physical functioning (Brittain et al., 2021), with stigma frequently experienced by individuals with ME/CFS regarding medical and public perceptions of the legitimacy of this illness (Baken et al., 2018). Thus, it is unsurprising that having ME/CFS is associated with elevated psychological distress, including depression and anxiety (Hamilton et al., 2009; Janssens et al., 2015). In the current study, psychological distress is operationalised as severity of depression and anxiety experienced, as these are the most commonly reported mental health difficulties in people with ME/CFS.

The daily functional limitations often experienced by individuals with ME/CFS require different coping strategies to successfully manage such challenges. Research indicates that ME/CFS patients use more maladaptive coping strategies, particularly self-blame, than healthy controls (Nater et al., 2012) and individuals with arthritis/fibromyalgia (Sirois & Molnar, 2014). One method of examining coping in chronic illness is coping efficacy, i.e. appraisals of how successfully one is coping with illness-related stressors (Gignac et al., 2000). Low confidence in one's ability to cope with illness-rated difficulties is prevalent in individuals with ME/CFS (Doerr et al., 2017).

As ME/CFS is a chronic illness with a lack of clear medical treatments, it is vital to determine adaptive ways of coping with this illness and decreasing psychological distress for these individuals. Cognitive Behaviour Therapy (CBT) is the recommended intervention for managing anxiety and depression in ME/CFS (NICE, 2021). However, there is disagreement

in the literature regarding the efficacy of CBT in ME/CFS (Geraghty, 2017; Vink, 2016; White et al., 2011). Some professionals who maintain that ME/CFS has psychosomatic origins have assumed that CBT can alleviate physical symptoms, as opposed to managing distress caused by this illness (Sharpe & Greco, 2019). While this issue is explicitly clarified in the updated NICE guidelines (2021), this assumption is noted to have elicited reluctance in some ME/CFS communities to undertake CBT (Freidberg, 2016). Increased understanding is needed of what factors may influence how individuals with ME/CFS cope with distress, in order to offer alternative therapeutic options alongside ensuring currently recommended psychological interventions like CBT are effective in this population.

Perfectionism & Coping in ME/CFS

Research indicates that individual differences play a role in coping and psychological distress in ME/CFS. A robust evidence base indicates high prevalence of perfectionism in individuals with ME/CFS (Deary & Chalder, 2010), with this trait implicated in poor adjustment and mental wellbeing specifically in ME/CFS (Kempke et al., 2011; Sirois & Molnar, 2014). Perfectionism is widely conceptualised as a multidimensional construct (Frost et al., 1990; Hewitt & Flett, 1991). This trait is commonly categorised as perfectionistic strivings (striving for flawlessness and excellence) and perfectionistic concerns (concerns over failure and other's expectations) (Stoeber & Otto, 2006). Hewitt & Flett's (1991) model of perfectionism offers a method of conceptualising and measuring these categories. Self-oriented perfectionism (holding self-imposed very high and unrealistic standards) is proposed to map onto and measure perfectionistic strivings. Socially prescribed perfectionism (viewing very high and unrealistic standards as being imposed by others) is proposed to relate to perfectionistic concerns (Hewitt & Flett, 1991).

Within ME/CFS populations, greater perfectionistic concerns are associated with maladaptive coping (Sirois & Molnar, 2014), and increased severity of fatigue and depression

symptoms (Kempke et al., 2011; Wright et al., 2021). Positive associations between perfectionistic concerns and perfectionistic strivings are observed in ME/CFS (Kempke et al., 2011). Perfectionistic strivings are not adaptive when coupled with perfectionistic concerns in this population, as attempts to maintain high standards associated with perfectionistic strivings are extremely challenging when faced with self-criticism from perfectionistic concerns and the debilitating symptoms of ME/CFS (Deary & Chalder, 2010).

One theory developed to understand how perfectionism may impact on coping and wellbeing in ME/CFS is the Stress and Coping Cyclical Amplification Model of Perfectionism in Illness (SCCAMPI; Sirois & Molnar, 2016). The SCCAMPI model suggests that perfectionism may intensify the stressful effects of adjusting to and managing chronic illness (e.g., through low perceived control and self-criticism, contributing to greater risk for poor health outcomes). Additionally, other researchers propose that perfectionism is a central etiological factor in ME/CFS, through negative self-evaluation associated with perfectionism actively generating and maintaining chronic stress responses in the body (Kempke et al., 2016; Luyten et al., 2006; 2011). Together, these theories suggest that reducing the negative impact of perfectionism in ME/CFS may be effective in treating poor coping efficacy and psychological distress frequently experienced with this illness.

The Interplay between Perfectionism, Self-Compassion and Coping

Self-compassion may be a promising approach for managing the harmful self-criticism associated with perfectionism and poor psychological outcomes in ME/CFS. Self-compassion, as defined by Neff (2003), refers to adopting a kind, accepting, and non-judgmental stance towards oneself in times of failure and difficulty. Self-compassion is proposed to be beneficial in chronic illness care through encouraging kind as opposed to self-critical responses to perceived setbacks and subsequently reducing perceived and physiological stress (Sirois & Rowse, 2016). Indeed, greater self-compassion has been linked

to more adaptive responses to perceived failures (e.g., engaging in positive reframing) in inflammatory bowel disease and arthritis (Sirois et al., 2015), higher self-rated health in ME/CFS (Sirois, 2020), and better mental health outcomes and coping efficacy in diabetes (Ferrari et al., 2017) and Human Immunodeficiency Virus (Brion et al., 2014). Recent systematic reviews which have synthesized the effects of compassion-related therapies on psychological distress in individuals with chronic health conditions reported significantly increased self-compassion with medium to large effect sizes (Hughes et al., 2021; Kılıç et al., 2021).

Critically, research has demonstrated that self-compassion mediates the relationship between perfectionism and psychological distress in healthy controls (James et al., 2015; Stoeber et al., 2020) and clinical cohorts (e.g., depression - Ferrari et al., 2018). Specifically, greater self-compassion weakens the positive relationship between perfectionism and psychological distress. Interventions which seek to increase self-compassion may therefore be beneficial in reducing the negative impact of perfectionism on coping and mental wellbeing. Studies using interventions where individuals were instructed to reframe past experiences of perceived failure in a self-compassionate light found increased state self-compassion and psychological wellbeing across varying cohorts, including chronic pain (Ziemer et al., 2015), breast cancer (Przezdziecki & Sherman, 2016), parents regarding challenging parenting events (Sirois et al., 2019), and university students regarding shame (Johnson & O'Brien, 2013). Therefore, increasing self-compassion may be effective for improving coping and psychological wellbeing in ME/CFS via reducing self-criticism, which is commonly found among individuals with elevated perfectionism.

Despite the benefits of prompting individuals to respond to past challenges with self-compassion outlined above, a recent study by Biskas et al. (2022) which utilised this task found that perfectionistic concerns may interfere with an individual's ability to engage in

self-compassion. In this study, healthy participants were less likely to reframe past experience of failure in a self-compassionate light if they held negative beliefs about self-compassion and experienced difficulties enacting self-compassion. Specifically, participants who scored highly on perfectionistic concerns held greater negative beliefs about self-compassion and expressed difficulties in enacting self-compassion. Regarding why individuals high in perfectionism may hold negative beliefs about self-compassion, research reports that some individuals feel taking a self-compassionate stance will lower their personal standards, decrease their motivation for personal growth, and mean that they will fail to attain goals (Kelly et al., 2021). These are all core characteristics of perfectionism (Hewitt & Flett, 1991). Equally, high levels of self-criticism (another key element related to perfectionism; Békés et al., 2015) have been linked to fearing self-compassion as this may feel like an alien concept for these individuals (Gilbert & Procter, 2006). Considering this body of literature, it could be theorised that while self-compassion may help to negate the harmful effects of perfectionism on wellbeing, it may be challenging for individuals who score highly on perfectionism to engage in self-compassion.

The Current Study

Considering theory and research on the positive impact of self-compassion on wellbeing in individuals with chronic illness, and the risk of perfectionism for distress in people with ME/CFS, the current study aimed to provide further insights into the relationships between self-compassion, perfectionism, psychological distress and coping in ME/CFS. The relationships between these constructs were first examined using correlational analysis.

Applying an experimental task used in previous research (Sirois et al., 2019), this study also investigated whether prompting individuals to write self-compassionately about a past event of perceived failure is effective in increasing state self-compassion in individuals

with ME/CFS. This aimed to examine whether state self-compassion could be experimentally increased in this clinical population. Considering the high prevalence of perfectionism in individuals with ME/CFS (Deary & Chalder, 2010) and that individuals who score high on perfectionism find it difficult to engage in self-compassion (Biskas et al., 2022), this research also aimed to examine whether perfectionism interferes with the effectiveness of self-compassion prompting in individuals with ME/CFS.

Hypotheses

Based on previous research, the following hypotheses were made:

1. Trait self-compassion will be positively associated with coping efficacy, and negatively associated with perfectionism (both perfectionistic strivings and concerns) and psychological distress in individuals with ME/CFS.
2. Prompting individuals with ME/CFS to respond to past challenges with self-compassion will be effective in increasing state self-compassion, compared to individuals with ME/CFS who do not receive such prompting (control group).
3. Perfectionism will moderate the impact of self-compassion prompting on state self-compassion, in that those with higher scores on both perfectionistic concerns and strivings will be less responsive to the prompt.

Method

Design

Participation involved the completion of an anonymous online study via the software platform Qualtrics. A quantitative experimental study was conducted. The independent variable was condition assigned for the experimental task (self-compassion; control). Both groups were measured on all outcomes of interest at baseline (pre-task), followed by a measure of state self-compassion again immediately after the experimental task (post-task).

Change in state self-compassion from pre- to post-task was the within-subject dependent variable. Moderators examined were perfectionistic strivings and perfectionistic concerns.

Ethics

Ethical approval was obtained from the University of Sheffield Ethics Committee (Appendix A). All participants provided informed consent to take part in the current study (Appendix B). The study may have evoked distress as participants were asked to consider topics like coping with chronic illness, psychological difficulties and recalling an incident whereby ME/CFS prevented them from achieving something that was important to them. Participants were made aware of this prior to participation to ensure informed consent, signposted to support services and advised to contact the researcher if needed. Considering the impact of potential fatigue and burden on participants, the outcome measures utilised were specifically selected due to having lower numbers of questionnaire items. This was to minimise any potential harm to participants and ensure that the study procedure is ethical for use within a population where fatigue may already play a large factor in study engagement and performance. Participants who completed the survey were given the opportunity to enter a prize draw for one of two £25 retail vouchers as gratuity for their time. The voucher amount was considered proportionate for the amount of participation time and was not deemed to coerce participation (British Psychological Society, 2014). All data were stored within encrypted password protected files, with participant email addresses stored separately and deleted after the prize draw was completed to preserve anonymity.

Sample Size Calculation

A priori power analysis was conducted using G*Power to determine the sample size required to prevent type II errors. This was conducted based upon the most stringent statistical analysis used: Analysis of Variance (ANOVA) with between-subject effects. This analysis related to hypothesis two. Assuming a medium effect size of f = 0.25 (Cohen, 1992;

due to the limited past research in this specific field), a significance level of alpha = 0.05, with two groups (self-compassion; control) and two measures specified (pre- and post-task state self-compassion), a total sample size of 82 participants was required to achieve 80% power.

Participants and Recruitment

An opportunity sampling method was used. Participants were recruited using online study advertisements from national and international ME/CFS research groups, support groups and charities via social media and organisation websites (Appendix C). Eligible participants were aged ≥18 years and self-reported a diagnosis of ME/CFS that had been provided by a medical professional. Participants were required to have sufficient understanding of the English language and not have any significant comorbid physical health conditions or memory impairment that could act as confounding variables. These inclusion criteria were explicitly stated on the study advert and participant information sheet (Appendices D-E), and specific questions probed these criteria within the online survey (Appendix F). Data collection took place from April 2021- March 2022. Participants who had started the survey but had failed to submit their final data were excluded if they had completed fewer than 80% of the measures across the survey.

A total of 147 participants were included in the final data set (Table 1). Of these, 87.8% were female and ages ranged from 19-73 years. 54.4% of participants were based in the United Kingdom. All participants had a current self-reported diagnosis of ME/CFS provided by a medical professional, with illness duration ranging from <1-43 years. Regarding employment, 38.1% were currently unemployed or on sick leave.

Table 1.

Summary of participant characteristics.

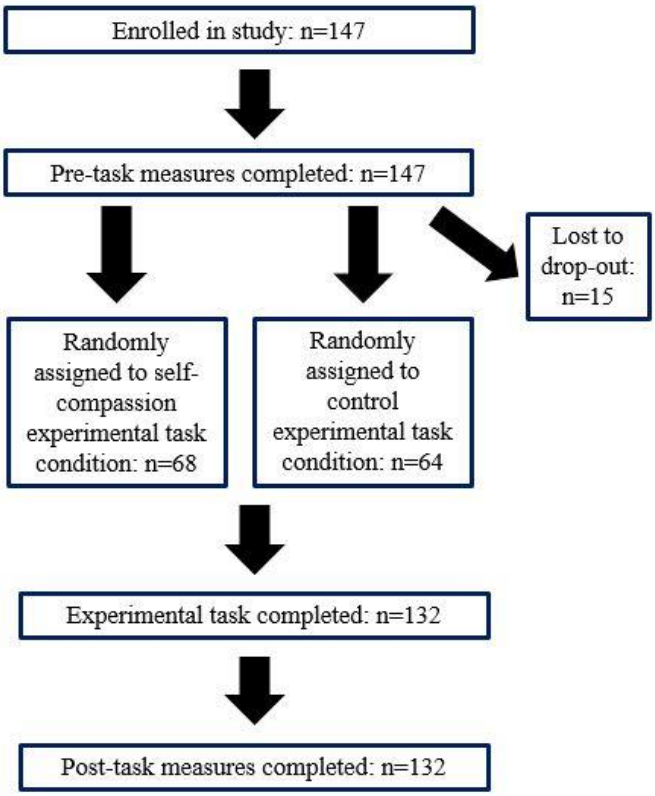
Variable	n (%), Mean (*SD*)
Age in years	40.98 (*13.29*)
Gender	
Male	14 (9.5%)
Female	129 (87.8%)
Non-binary	4 (2.7%)
Relationship Status	
Married/Living with intimate partner	82 (55.8%)
Separated/Divorced	21 (14.3%)
Never married/Single	43 (29.3%)
Widowed	1 (0.7%)
Ethnicity	
Black	1 (0.6%)
White	129 (87.8%)
Mixed	8 (5.4%)
Other	8 (5.4%)
Education Level Obtained	
Some high school	7 (4.8%)
High school graduate	11 (7.5%)
College/University undergraduate	87 (59.1%)
Postgraduate school	42 (28.6%)
Illness duration in years	11.63 (*9.08*)
Currently receiving treatment	83 (56.5%)
Co-morbid physical health conditions	62 (42.2%)

Note. SD = standard deviation.

Of these participants, n= 15 did not complete the post-task measure and so were not included in statistical analysis to examine hypotheses two and three. See Figure 1 for participant flow through the study.

Figure 1.

Participant flow through the study.



Materials

Outcome Scale Reliability Checks

Reliability of the outcome measures within the study sample was calculated using Cronbach's alpha (α). Criteria set by George and Mallery (2003) was applied when interpreting Cronbach's alpha reliability coefficients (see Table 2).

Table 2.

Criteria for assessing outcome scale internal consistency.

Cronbach's Alpha (α)	Level of Reliability
>0.90	Excellent
0.80-0.89	Good
0.70-0.79	Acceptable
0.60-0.69	Questionable
0.50-0.59	Poor
<0.50	Unacceptable

Perfectionism

Perfectionistic strivings and perfectionistic concerns were measured using the Multi-dimensional Perfectionism Scale Short (MPS Short; Hewitt et al., 2008; Appendix G). The MPS Short consists of 15 items in which participants rate their response on a Likert scale from 1 (strongly agree) to 7 (strongly disagree). A score for perfectionistic strivings was computed from five items which measure self-orientated perfectionism. A score for perfectionistic concerns was computed from five items which measure socially prescribed perfectionism. Higher scores indicated greater perfectionism. The MPS Short has been found to correlate highly with the longer 45-item MPS Scale (Hewitt & Flett, 1991; Stoeber, 2018), which is reported to have good internal consistency and construct validity (Lo et al., 2020). In the current study, internal consistency was good for both perfectionistic strivings ($\alpha = 0.89$) and perfectionistic concerns ($\alpha = 0.84$).

Trait Self-Compassion

Trait self-compassion was measured using the Self-Compassion Scale-Short Form (SCS-SF; Raes et al., 2011; Appendix H). The SCS-SF consists of twelve items in which participants rate their response on a Likert scale from 1 (almost never) to 5 (almost always). This measure assesses the three main components of self-compassion: self-kindness, common

humanity, and mindfulness (Neff, 2003). Higher scores indicated greater self-compassion. The SCS-SF has been found to have good internal consistency ($\alpha \geq 0.86$) and a near perfect correlation with the long form SCS ($r \geq 0.97$; Raes et al., 2011). In the current study, internal consistency was good ($\alpha = 0.80$).

State Self-Compassion

State self-compassion was assessed using the State Self-Compassion Scale-Short form (SSCS-S; Neff et al., 2020; Appendix I). The SSCS-S consists of six items in which participants rate their response on a Likert scale from 1 (not true at all) to 5 (very true for me) while prompted to think about a situation they are experiencing right now that is painful or difficult. Higher scores indicated a high level of self-compassion. The SSCS-S has been found to have high internal reliability ($\alpha = .86$) and is strongly correlated with the longer form of the State Self-compassion Scale ($r = 0.957$; Neff et al., 2020). In the current study, internal consistency was good ($\alpha = 0.83$).

Psychological Distress

Depression was measured using the nine-item Patient Health Questionnaire (PHQ-9; Kroenke et al., 2001; Appendix J). Anxiety was measured using the seven-item Generalised Anxiety Disorder-7 (GAD-7; Spitzer et al., 2006; Appendix K). Both scales require participants to rate their response for each item using a Likert scale from 0 (not at all) to 3 (nearly every day). Higher scores indicated greater anxiety and depression symptoms. Both measures are routinely used in mental health services and well-validated in clinical samples (Kroenke et al., 2010). These measures have been found to have good internal consistency ($\alpha = 0.87$-0.89) and construct validity (Kocalevent et al., 2013; Löwe et al., 2008). In the current study, internal consistency was good for the PHQ-9 ($\alpha = 0.80$) and excellent for the GAD-7 ($\alpha = 0.91$).

Coping Efficacy

Coping efficacy was measured using the Coping Efficacy Scale (CES; Gignac et al., 2000; Appendix L). The CES consists of three items in which participants rate their response on a Likert scale from 1 (strongly disagree) to 5 (strongly agree) regarding how well participants feel they are coping with their chronic illness. Higher scores indicated greater coping efficacy. The CES has been used with various chronic illness populations in previous research (Falter et al., 2003; Sirois et al., 2015) and has demonstrated good internal reliability (α =.79; Gignac et al., 2000). In the current study, internal consistency was good (α = 0.86).

ME/CFS Fatigue Severity

Fatigue severity was measured using the Fatigue Severity Scale (FSS; Krupp et al., 1989; Appendix M). The FSS consists of nine items in which participants rate their response on a Likert scale from 1 (strongly disagree) to 7 (strongly agree) regarding how often participants experience fatigue and whether fatigue interferes with their lives. Higher scores indicated greater severity of fatigue. This scale has been used with participants with ME/CFS (Jason et al., 2011) and is reported to have good internal consistency (α =.88), test-retest reliability and concurrent validity (Krupp et al., 1989). The FSS has been found to be more sensitive to identifying fatigue in ME/CFS compared to other illness cohorts (Pepper et al., 1993) and alternative fatigue measures (Jason et al., 2011). In the current study, internal consistency was good (α = 0.89).

Procedure

The survey was accessed through a web link in the online study advertisement. Participants were provided with a participant information sheet and consent form before proceeding. Participants first completed all pre-task outcome measures (60 items in total) with the ordering of these measures randomised by Qualtrics.

Participants were then presented with the experimental task after being randomly assigned via Qualtrics to either the self-compassion or control condition (Appendix N).

Participants were instructed to recall an event where they struggled to achieve a goal due to an aspect of ME/CFS and felt disappointed in themselves. This type of event was selected as individuals who score highly on perfectionism commonly set excessively high standards for their own performance, which if not met are accompanied by harsh self-criticism (Frost et al., 1990). Therefore, the type of event was highly relevant when investigating the impact of perfectionism on the subsequent ability to view this event in a self-compassionate light.

In the self-compassion condition, participants were then prompted to write about this event in a more self-compassionate light, writing about the event with self-kindness, common humanity and mindfulness. Participants assigned to the control condition were instructed to write further details about the event that were solely factual in content, e.g., what the weather was like during that event. All participants then completed the state self-compassion measure again post-task.

Finally, a mood repair task was completed which aimed to induce positive emotions and ease any distress that may have been generated by the experimental task (Appendix O). Participants were asked to recall a time when they successfully met a personal goal which made them feel happy. When this task was previously used in research, it was found to be effective in mood neutralisation for individuals scoring highly on perfectionism that had been asked to recall past experiences of failure (Sirois et al., 2010).

Participants who completed the survey were forwarded to a debrief form (Appendix P). The debrief sheet contained details of the self-compassion writing task alongside signposting participants to self-compassion related resources, to mitigate any disadvantages of being assigned to the control group.

Consultation

Prior to recruitment, consultation was sought from ME/CFS charities/organisations to obtain feedback on the study procedure, specifically regarding the study advertisement and

length of study participation. Feedback was obtained via email from two service users. This resulted in the formatting of the study information sheet being adjusted so that it was presented within one screen (as opposed to this information originally being spread across several screens). It was also brought to the author's attention that within the study documents there were points where the term 'chronic fatigue syndrome' had been used as opposed to ME/CFS. This was also amended to ensure consistency with current NICE guidance (NICE, 2021).

Data Analysis

Data Screening & Normality Checks

Data analysis was conducted using Statistical Package for the Social Sciences (SPSS; Version 26). All data were screened for errors, missing data, duplicate entries and consistent middle scorers. Skewness and kurtosis statistics and inspection of histograms was used to assess the assumption of normally distributed data (Field, 2009). Where appropriate, non-parametric statistical tests were used to correct for any violations of this assumption.

Hypothesis 1: Relationships between trait self-compassion, perfectionism and psychological outcomes

Descriptive statistics were calculated for all outcome variables. To examine the bivariate relationships between trait self-compassion, perfectionism (strivings and concerns), coping efficacy, anxiety and depression in ME/CFS, a correlation matrix was computed using Spearman Rho tests. An alpha level of $p = .01$ was applied when interpreting statistical significance to reduce the likelihood of Type 1 errors.

Hypothesis 2: Can state self-compassion be increased through self-compassionate prompting?

Randomisation checks

Between-group comparisons in key demographics (age and gender) and the pre-task outcome variables were made as a check of the randomisation process. A series of Mann Whitney U tests and chi-squared tests were then used to test whether there were significant differences in demographic or baseline outcomes between participants in the self-compassion and control group. Again, an alpha level of $p<.01$ was adopted to correct for multiple comparisons.

Group comparisons – state self-compassion

Within- and between-group comparisons were made in order to investigate whether providing a prompt to engage in self-compassionate writing was effective in increasing state self-compassion in individuals with ME/CFS. A mixed 2x2 Analysis of Variance (ANOVA) was used to determine the effects of group (self-compassion; control) on state self-compassion (pre-task score; post-task score) and explore the interaction between these variables. Paired t-tests were then conducted to compare within-group differences between pre- and post-task state self-compassion.

Hypothesis 3: Does perfectionism moderate the effectiveness of self-compassion prompting?

To test whether perfectionism is moderating the effectiveness of prompting individuals to write self-compassionately in improving state self-compassion, moderation analysis was performed using the PROCESS macro add-on for SPSS (version 4.1; Hayes, 2022). The outcome variable was post-task state self-compassion. The predictor variable was pre-task state self-compassion. This analysis was conducted separately for perfectionistic strivings and perfectionistic concerns as the moderating variables. Post-hoc analysis was conducted using Spearman rho tests, to determine the nature of relationships between the perfectionism dimensions and pre- and post-task self-compassion in the self-compassion group.

Results

Data Screening & Normality Checks

Regarding missing data screening, two participants were excluded for having <80% of the pre-task outcome measures completed. No errors were identified, so it was assumed that data reflected true scores (Field, 2009). Using the skewness and kurtosis statistics and inspecting histograms (see Appendix Q), data for some of the pre-task outcome measures was found to violate assumptions of normality. Scores on measures of anxiety, depression and coping efficacy were negatively skewed. Fatigue scores were positively skewed. Therefore, non-parametric statistical tests were used when analysing relationships and group differences on pre-task outcome variables.

Hypothesis 1: Relationships between trait self-compassion, perfectionism and psychological outcomes

Table 3 presents descriptive statistics and the bivariate associations for all baseline (pre-task) outcome variables. Notably, trait self-compassion was negatively associated with perfectionistic strivings, perfectionistic concerns, anxiety and depression. Trait self-compassion was positively associated with coping efficacy. Collectively, these findings offer support for hypothesis one.

Table 3.

Means, standard deviations and correlations for the pre-task outcome variables.

Variable	*M*	*SD*	1	2	3	4	5	6
1. Trait Self-compassion	2.89	0.62						
2. Perfectionistic Strivings	21.99	6.24	-.26*					
3. Perfectionistic Concerns	21.46	4.44	-.43**	.48**				
4. Anxiety	9.70	5.85	-.49**	.09	.30**			
5. Depression	14.36	5.89	-.42**	.08	.30**	.65**		
6. Coping Efficacy	8.18	2.96	.43**	-.08	-.24*	-.35**	-.47**	
7. Fatigue Severity	57.23	7.85	-.02	-.06	.19	.19	.31**	-.25**

Note. M and *SD* are used to represent mean and standard deviation, respectively. * indicates $p<.01$. ** indicates $p<.001$.

Hypothesis 2: Can state self-compassion be increased through self-compassionate prompting?

Randomisation checks

As presented in Table 4, Mann Whitney U tests and chi-squared tests revealed no significant differences in demographic or baseline outcomes between participants assigned to the self-compassion group and participants assigned to the control group.

Table 4.

Between-group comparisons for age, gender and pre-task outcome variables.

Variable	Self-compassion Group (n=68)	Control Group (n=64)	Statistic (U or X^2)	*p* value
Gender	Female n= 58 (85.3%) Male n= 8 Non-binary n= 2	Female n= 57 (89.1%) Male n= 5 Non-binary n= 2	0.580	.748
Mean age in years	39.53 (*12.46*)	41.73 (*13.38*)	2009.0	.447
Trait Self-compassion	2.83 (*0.56*)	2.94 (*0.67*)	1969.5	.346
Perfectionistic Strivings	21.84 (*6.26*)	22.55 (*6.26*)	2009.5	.448
Perfectionistic Concerns	21.60 (*4.33*)	21.58 (*4.58)*	2095.5	.713
Pre-task State Self-compassion	2.74 (*0.77)*	3.00 (*0.85*)	1735.0	.044
Anxiety	10.76 (*6.14*)	8.47 (*5.61*)	1686.5	.026
Depression	15.15 (*5.23*)	13.28 (*6.13)*	1770.5	.064
Fatigue Severity	56.22 (*10.02*)	58.00 (*5.19)*	2051.0	.567
Coping Efficacy	8.03 (*2.80*)	8.45 (*2.86*)	1968.0	.567

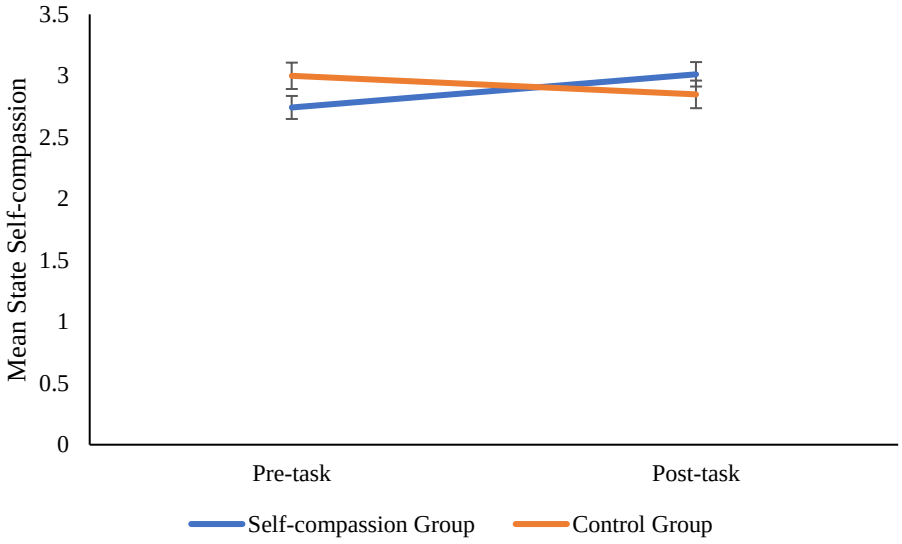
Note. Standard deviations (*SD*) presented in brackets.

Group comparisons – state self-compassion

Using a mixed 2x2 ANOVA, main effects of time (F (1, 130) = 1.641, p=.202, partial η2 = 0.012) and group (F (1,130) = .116, p=.734, partial η2 = 0.01) were not observed. However, there was a significant interaction between time and group on state self-compassion scores (F (1,130) = 20.660, p < .001, partial η2 = 0.137; see Figure 3).

Figure 3.

Group differences in state self-compassion scores.



Note. Error bars indicate standard error of mean.

A significant increase in state self-compassion was found between pre-task (mean= 2.74; SD= .772) and post-task (mean = 3.01; SD = .819) in the self-compassion group (t (67) = -4.069, p < .001, Hedges's *g* = 0.34). In contrast, a significant decrease in state self-compassion was found between pre-task (mean= 3.00; SD= .857) and post-task (mean = 2.85; SD = .898) in the control group (t (63) = 2.346, p = .022, Hedges's *g* = 0.17). Overall, hypothesis two was supported as prompting individuals to engage in self-compassionate writing was found to be effective in increasing state self-compassion in ME/CFS.

Hypothesis 3: Does perfectionism moderate the effectiveness of self-compassion prompting?

Two moderator regression analyses were conducted to assess whether baseline scores of perfectionistic strivings and perfectionistic concerns moderated changes to state self-compassion pre- to post-task within the self-compassion group. Perfectionistic concerns did not moderate the

relationship between pre- and post-state self-compassion (B = .0038, F (1, 64) = .597, p = .44, CI = -.15-.04). Equally, perfectionistic strivings did not moderate the relationship between pre- and post-state self-compassion (B = .000, F (1, 64) = .002, p =. .96, CI = -.02-.05). Overall, hypothesis three was not supported as perfectionism did not moderate the relationship between pre- and post-state self-compassion in the self-compassion group.

Post-hoc correlational analysis found that perfectionistic concerns were strongly negatively correlated with both pre-task (r_s = -.501, p<.001) and post-task (r_s = -.475, p<.001) state self-compassion. Perfectionistic strivings were not significantly correlated with either pre-task (r_s = -.149, p = .225) or post-task (r_s = -.180, p = .142) state self-compassion.

Discussion

To the author's knowledge, this is the first study to examine whether prompting individuals to respond to past challenges with self-compassion is effective in increasing state self-compassion in people with ME/CFS, and whether perfectionism interferes with the efficacy of such prompting. This research also investigated the relationships between perfectionism, self-compassion, psychological distress and coping efficacy in this illness group. In support of study hypotheses, trait self-compassion was negatively related to perfectionism and psychological distress, and positively associated with coping efficacy. These findings suggest that higher levels of self-compassion are associated with better coping efficacy and less psychological distress in individuals with ME/CFS. For participants who were prompted to engage in self-compassionate writing, a significant increase in state self-compassion was noted, which was not found in the control group. This finding offered support for the study hypothesis and provides evidence that prompting individuals to engage in self-compassionate writing is effective in increasing state self-compassion in people with ME/CFS. In contrast with the study hypothesis, perfectionism did not moderate the effects of self-compassion prompting in increasing state self-compassion.

Study results are consistent with previous research which found a negative relationship

between perfectionism and self-compassion in general population samples (Stoeber et al., 2020). The results also aligned with past findings that greater levels of perfectionism are implicated in poorer mental wellbeing in both ME/CFS and other chronic illnesses (Kempke et al., 2011; Sirois & Molnar, 2014; Wright et al., 2021). The positive relationship found between trait self-compassion and coping efficacy also corresponds with research which found links between higher levels of trait self-compassion and the use of more adaptive coping strategies which in turn explained greater coping efficacy in chronic illnesses (Sirois et al., 2015). Considering these results collectively, the current study highlighted that self-compassion is related with better psychological outcomes in ME/CFS. In contrast, perfectionism is related to poorer psychological outcomes in ME/CFS.

This is the first study to demonstrate that state self-compassion can be induced by prompting individuals with ME/CFS to write about past experiences of perceived failure in a self-compassionate light. This finding contributes to the evidence base which has used this self-compassion writing task in different illness and non-clinical populations and found it to be an effective strategy at increasing state self-compassion alongside other constructs related to mental wellbeing (Przezdziecki & Sherman, 2016; Sirois et al., 2019; Ziemer et al., 2015).

It is acknowledged that this increase was not substantial enough for post-task state self-compassion scores to be significantly different from those of participants assigned to the control group who did not have this prompt. A recent study found that individuals who score highly on perfectionism are more likely to hold negative beliefs about self-compassion and subsequently find it difficult to engage in self-compassion strategies (Biskas et al., 2022). It is noted that mean scores on the measures of perfectionism were high in the current sample. Applying the findings of Biskas et al. (2022), participants assigned to the self-compassion group may hold negative beliefs about self-compassion which could have suppressed their ability to engage with the self-compassion writing task. Equally, participants may not have had many past opportunities to practice engaging in self-compassion if high perfectionism interferes with their ability to do so. As changes in state self-compassion were examined after engaging in the task once, this may not have been sufficient to

elicit more prominent changes in state self-compassion post-task.

Regarding the significant reduction in state self-compassion post-task found in the control group, this may be explained by the nature of the experimental task. The task was specifically used as it involved recalling a time of perceived failure which may therefore tap into difficulties associated with scoring highly on perfectionism, such as setting excessively high personal standards which if not met result in self-criticism (Frost et al., 1990). Considering the high mean scores on the perfectionism dimensions in the current sample, it may be that in the absence of having the prompt to engage in self-compassionate writing, participants in the control condition may have experienced self-criticism and consequently felt less self-compassionate after the task as a result.

Notably, the current study did not find that perfectionism moderated the relationship between pre- and post-task self-compassion in the self-compassion group. Thus, perfectionism did not appear to weaken the effects of engaging in self-compassionate writing as predicted. This finding appears to contrast with theoretical accounts of how perfectionism (via harsh self-criticism) contributes to poorer psychological outcomes in ME/CFS and chronic illnesses (Kempke et al., 2016; Sirois & Molnar, 2016). Equally, this result is not consistent with research that suggests high perfectionism is a vulnerability factor for engaging in self-criticism (Békés et al., 2015) which subsequently interferes with the ability to engage in self-compassion (Biskas et al., 2022).

When considering methodological factors in the current study which may have influenced study findings, it is noted that perfectionistic concerns were strongly negatively correlated with both pre-task and post-task state self-compassion. The mean scores on both perfectionistic strivings and concerns were high in this sample, consistent with previous reports of high levels of perfectionism present in ME/CFS samples (Deary & Chalder, 2010). Moderation analysis compares high levels of a construct with low levels of a construct in order to determine whether the strength of the relationship between the predictor and outcome variables changes as the levels of this moderator construct change (Holmbeck, 1997). This may explain why moderation effects were not identified if there were fewer instances of low levels of perfectionism present in the sample when performing

the moderation analysis. Alternatively, despite perfectionism scores being high, there was some variation in perfectionism scores observed (Appendix Q). Therefore, perhaps the degree of perfectionism in the current sample may have been lower than levels observed in other research samples. This could have diluted the moderation effects of perfectionism in the current study.

Strengths & Limitations

The current findings should be considered in light of the following study strengths and limitations. Regarding study strengths, a priori power analysis indicated that the sample size acquired was sufficient for adequate power for statistical analysis. Additionally, the methodology and outcome measures utilised were specifically selected to reduce participant burden as much as possible, with consultation sought from ME/CFS charities and groups on study materials.

However, there are notable limitations of the current study. Considering the heterogeneity in symptom severity in ME/CFS, it should be acknowledged that the sample may be biased to participants with less severe illness symptoms. Individuals with more debilitating symptoms, e.g., extreme fatigue and cognitive impairments, will be less able to engage with this research, despite efforts to limit participant burden. Equally, the majority of the sample were White females who had achieved high educational attainment. Therefore, the generalisability of the results may be limited to wider ME/CFS populations. However, this appears to be a consistent profile of individuals with ME/CFS who take part in research (Lim et al., 2020). Furthermore, the higher proportion of White females within the current sample is also similar to demographics reported for individuals with ME/CFS accessing specialised clinical services in the United Kingdom (Collins et al., 2011; 2016; Nacul et al., 2019).

While consultation with individuals with ME/CFS was beneficial in adjusting how materials were presented to participants, there was limited interest from approached organisations in providing feedback and/or assistance with recruitment, with a common reason being that the current study does not align with wider agendas in ME/CFS communities around increasing the production of biomedical research in this population (Spandler & Allen, 2018). Obtaining opinions on pre-

developed materials and study methodology could be considered tokenistic lived experience consultation. Considering models of co-production in health and social care (National Co-production Advisory Group, 2021), future research could focus on increasing co-production with people with lived experience in this population, to increase engagement of ME/CFS communities in empirical studies and ensure that research aligns with the needs of this community as much as possible. Considering the opposition to psychological research and intervention held by many in the ME/CFS community (Spandler & Allen, 2018), this may also mean that the included sample is biased to individuals who may be more open to and interested in considering the psychological impact of having this physical health condition. The study results may therefore be biased if the sample contains individuals who may be more motivated to engage in self-compassionate and psychological thinking relative to the wider ME/CFS population, consequently reducing the generalisability of the findings.

It may have been more beneficial to use a longer-term follow-up, in order to determine if the increases in state self-compassion observed within the group who were prompted to write self-compassionately about a past challenging event would be sustained or would increase. However, a longitudinal design was not deemed to be appropriate due to the extra burden this may place on participants who are already experiencing extreme levels of fatigue as a result of their illness. Equally, designs involving follow-up phases may have resulted in a high attrition rate due to the 'boom-and-bust' cycle that is frequently experienced by individuals with ME/CFS (NICE, 2021). 'Boom-and-bust' refers to a common pattern of alternation between periods of excessive physical or cognitive activity (when individuals are feeling better) and consequent periods of feeling extremely fatigued/having symptom flare-ups and having to rest for longer periods of time (Burgess, 2019). Using a longitudinal study design could mean that participation may be understandably compromised when individuals are experiencing a flare-up or in the 'bust' stage of the cycle.

Clinical Implications

The current findings highlighted that self-compassion and perfectionism may be important factors to consider in the assessment, formulation and intervention for psychological distress in people with ME/CFS. Directly assessing levels of perfectionism and self-compassion with standardised tools may be beneficial in helping identify self-critical cognitions and maladaptive behaviours that maintain distress in these individuals. This information may enhance psychological assessment and formulation, through obtaining a clearer picture of how psychological difficulties develop and are maintained in ME/CFS.

The findings of this study suggest that engaging in writing about past experiences of perceived failure in a self-compassionate manner increased state self-compassion for people with ME/CFS. The results also found links between lower levels of trait self-compassion with higher levels of perfectionism, anxiety and depression in this sample. Previous research suggests higher levels of self-compassion are associated with better psychological wellbeing including reduced distress in chronic illness populations (Brion et al., 2014; Ferrari et al., 2017; Sirois et al., 2015). Therefore, using psychological interventions that specifically adopt the use of self-compassion strategies may be more beneficial in alleviating psychological distress in this population, e.g., Compassion-Focused Therapy (CFT; Gilbert, 2009). Using regular outcome measures of self-compassion and perfectionism throughout intervention is recommended to generate practice-based evidence, but also to offer clarity as to whether high perfectionism may impact upon the efficacy of compassion-focused therapies for individuals with ME/CFS.

Future Research

The current study suggests state self-compassion can be experimentally increased in individuals with ME/CFS. Further research is needed regarding the application of compassion-focused strategies in ME/CFS and whether these are helpful in alleviating psychological distress experienced. Considering that the current study was the first to examine the use of self-compassionate writing in ME/CFS, replication of these findings in more diverse samples is needed,

to determine if the benefits of prompting individuals to engage in self-compassion are evident in wider ME/CFS populations where there may be more heterogeneity in symptoms and severity.

Considering the limited evidence base regarding potential benefits of self-compassion in chronic health condition populations (Sirois & Rowse, 2016), future research utilising qualitative methodology is recommended in order to widen understanding of how individuals with ME/CFS perceive self-compassion and to identify potential barriers to engaging in self-compassionate thinking and behaviour. These data may help to tailor extant psychological approaches, increasing person-centred care provided for this clinical population. By examining beliefs about self-compassion in a population like ME/CFS, where elevated perfectionism is common, researchers may be able to develop a better understanding of how these traits relate and whether there are individuals who may find it more challenging to adopt a self-compassionate stance.

Conclusions

Overall, the current study found that higher levels of self-compassion were associated with better coping efficacy and less psychological distress in individuals with ME/CFS. This research also provided preliminary evidence that state self-compassion can be increased by prompting individuals to write about past experiences of perceived failure in a self-compassionate light. Further research is needed to offer greater clarification on whether the high prevalence of perfectionism typically observed in this clinical cohort may relate to the utility of compassion-focused approaches in ME/CFS. Considering that the current study was the first to examine the use of self-compassionate writing in ME/CFS, replication of these findings in wider samples is needed, to determine if the benefits of prompting individuals to engage in self-compassion are evident in broader ME/CFS populations. This research highlights that self-compassion and perfectionism may be important factors for clinicians to consider during the assessment, formulation and intervention of psychological distress in people with ME/CFS.

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Appendices

Note Appendices removed for copyright purposes.