

# Master Thesis



**Universiteit Utrecht**

## **Lumpers Versus Splitters: Analysing the Effects of Soothers on Fibromyalgia and Chronic Fatigue Syndrome**

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## Abstract

The ‘Lumpers’ versus ‘Splitters’ debate has been central to the study of Functional somatic syndromes (FSS) for decades. Fibromyalgia (FM) and Chronic fatigue syndrome (CFS) share similar demographic and clinical characteristics; however, their aetiology and pathophysiology remain undetermined. Lumpers postulate that FM and CFS are simply different manifestations of a singular FSS, while ‘Splitters’ argue that they are separate FSSs. This study is part of a group project assessing the impacts of threats, soothers and drives on somatic symptoms. The current study focuses on the role of the soothing system, and investigates the potential ability of soothers to alleviate FM and CFS symptoms. Additionally, this study examines the experiential differences between these two FSSs, in order to add to the Lumpers versus Splitters debate. The sample consisted of FM and CFS patients ( $n = 804$ ) aged 18 and over. Participants answered three questionnaires relating to the impact of soothers on their somatic symptoms, mental wellbeing, physical functioning, and how the syndromes affect their quality of life and overall health. The findings tentatively support the splitters perspective. Results revealed significant differences between CFS and FM regarding fatigue severity (the central symptom of CFS), and also indicated that balancing activities and relaxation was more important within CFS treatment in comparison to FM treatment. There were also differences in overall symptom severity, with FM patients being the least affected and patients with both FM and CFS being the most affected. This study highlights the potential utility of soothers within FM and CFS treatment. The results tentatively indicate that FM and CFS should be considered separate FSSs and provides a platform for future study in this area.

*Keywords:* soothers, fibromyalgia, chronic fatigue syndrome, lumpers, splitters

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## 1.0 Introduction

*“Perhaps, psychology has focused more on repairing the negative and neglected the field of promoting the positive” (Overgaauw, 2020, p. 21).*

In the author’s opinion this conceptualization of psychology’s focus resonates with the study and treatment of Fibromyalgia (FM) and Chronic fatigue syndrome (CFS). FM and CFS are enigmatic illnesses with overlapping symptomatology and demographics; wide ranging symptom fluctuations and disability; high levels of psychiatric morbidity, yet poorly understood aetiologies (Friedberg & Jason, 2001; Mckay et al., 2021). Typically, the treatments for these syndromes aim to minimize stimuli that provoke negative symptoms and negative psychosocial repercussions, while ignoring the promotion of positive and soothing stimuli, which can lead to symptom reduction and adaptive coping. Gilbert’s affect regulation model (2014) identifies three key affect systems with regard to emotion regulation; the threat system, the drives system and the soothing system. ‘Soothers’ (e.g., social support) are comforting factors that may create feelings of calmness, wellbeing, safety or social connectedness. Soothers are associated with the soothing system and subsequent emotions which can mitigate symptoms in chronic physical conditions such as CFS (Overgaauw, 2020).

## 1.1 The Lumpers Versus Splitters Debate

The ‘Lumpers versus Splitters’ debate has long been a matter of deliberation. ‘Lumpers’ view various symptoms as different manifestations of a single underlying functional somatic syndrome (FSS). ‘Splitters’ claim these different manifestations represent important variations and potential alternative underlying FSSs that require different

assessments and treatments (Berenson, 2004; Fischer & Nater, 2014). Berenson (2004, p. 98) summarises these polarized perspectives; “Lumpers may look for a cure for cancer, while splitters may look for cures for a hundred different cancers”.

It is argued that FM, CFS, and associated FSSs are simply manifestations of a greater, singular syndrome (Ursin, 1997; Joustra, 2019). The pathophysiology of these FSSs remain uncertain, and due to similarities between them some posit that they share common pathophysiological mechanisms, namely central nervous system dysfunction (Gur & Oktayoglu, 2008). FM is characterized by chronic widespread musculoskeletal aches, pain, stiffness (Garcia-Campayo et al., 2007) and tenderness upon examination at specific, predictable anatomic sites (tender points) (Wolfe et al., 1990; Buskila & Cohen, 2007). CFS is characterized by intense disabling fatigue (lasting at least 6 months), accompanied by several rheumatological, infectious, and neuropsychiatric symptoms (Fukuda et al., 1994). Although, both of these definitions have been criticised (Natelson, 2019; Stewart et al., 2019). These FSSs are often diagnosed comorbidly together, with irritable bowel syndrome (IBS), sleep disorders, psychiatric disorders, neurological disorders and other somatoform disorders, therefore they require tailored treatment plans (Aaron et al., 2000; Afari & Buchwald, 2003; Buskila & Cohen, 2007; Carrillo de la Peña, et al., 2015; Jason et al., 2000; Natelson, 2019). 20%–70% of FM patients also meet the CFS criteria (Buchwald & Garrity, 1994; Hudson et al., 1992; White et al., 2000), and 35%–70% of those with CFS-like illnesses also have comorbid FM (Buchwald & Garrity, 1994; Goldenberg et al., 1990). Although there is plenty of research on these FSSs’ split categorisation, the debate remains unsettled.

Wessely and colleagues (1999) were some of the first to contest split categorisations with their meta-analysis on medically unexplained symptoms and syndromes including IBS and CFS. As Lumpers, they posit that the overlap of symptoms, non-symptom characteristics

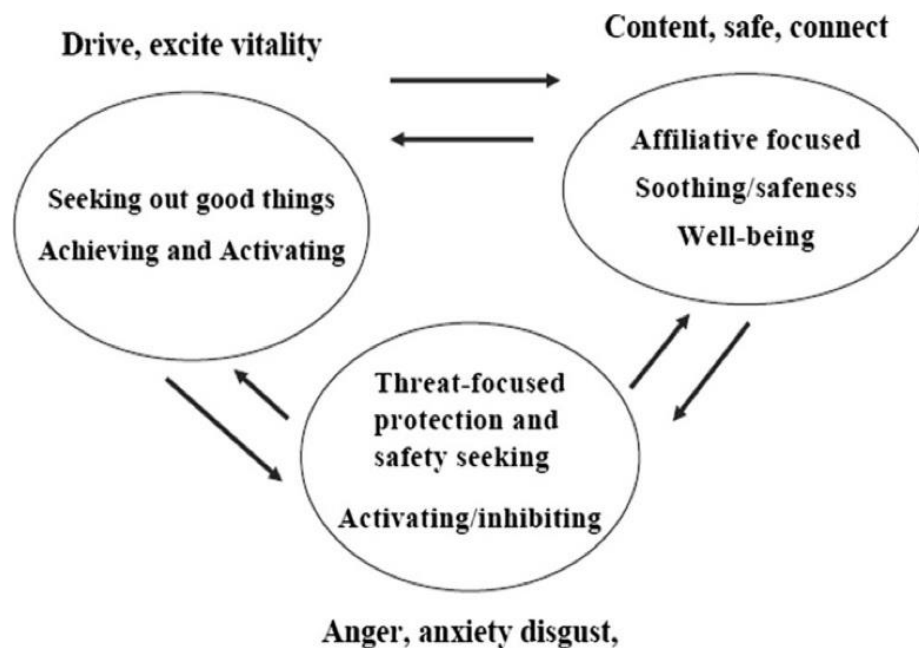
(e.g., Sex), and interventions for these FSSs (FM, CFS, IBS etc.) indicate that they are different dimensions of a greater singular syndrome (Geenen & Jacobs, 2003; Wessely et al., 1999). They concluded that differences between such syndromes were not as important as the similarities and that updated dimensional classifications were required. Congruently, recent research by Joustra (2019) indicated that all diagnostic symptoms of CFS, FM and IBS were connected directly or via other symptoms, and that the diagnostic overlap was significantly higher than what could be explained by chance.

Geenen and Jacobs (2003) acknowledge the considerable overlap between these syndromes, however they assert that we should not dispense with separate classifications. They contend that there are significant differences in neurochemical and brain structure activity, reactivity of the hypothalamic pituitary adrenal (HPA) axis and sympathetic nervous system (SNS) within FM and CFS, and therefore identical pathogenicity is unlikely. Similarly, Natelson (2019) emphasizes that distinct biochemical, physiologic, and genetic differences exist between the two syndromes. Research by Joustra (2019) notes quantitative differences such as higher general symptom and fatigue severity in CFS patients, and higher pain severity in FM patients. Moreover, Monden and colleagues (2020) found syndrome-specific predictors to be more common and relevant than shared predictors in their study of IBS, FM and CFS, indicating that shared aetiology is unlikely (see Appendix A for proposed pathophysiology of CFS and FM, and the apparent quantitative differences between patients). Geenen and Jacobs (2003) concluded that research into the similarities and differences between these syndromes will ultimately lead to more tailored management of each. Admittedly, there is evidence for both hypotheses, but while marked overlaps in symptoms and patients exist, these are not universal and the differences cannot be ignored (Kanaan et al., 2007).



## 1.2 Emotion Regulation Systems and the use of ‘Soothers’

Gilbert’s affect regulation model (2014) combines several approaches to emotion regulation. The model explains the interactions between the threat, drive, and soothing systems (Figure 1) which supposedly modulate the neural activity of somatic symptoms.



**Figure 1.** *Three types of affect regulation system. Obtained from Gilbert (2014).*

The threat system keeps us alert to threats and is characterized by anxiety focus, protection, safety seeking, activating and inhibiting, and is linked to feelings of anxiety, anger and sadness. Adrenaline, cortisol and noradrenaline are associated with this system. The drive system motivates us to obtain resources and rewards. It is characterized by resource focus, wanting, pursuing, achieving and consuming. This system is associated with feelings of excitement, joy and pleasure and the dopaminergic systems mediate its effects. The soothing system is characterized by safeness, connection and affiliative focus. Feelings of contentment and calmness are related to this system. This system is linked to the oxytocin and opiate systems’ functions, helping to abate the effects of the drive and threat systems, and to restore

energy (Depue & Morrone-Strupinsky, 2005; Gilbert, 2014). Research suggests that this system evolved in order to enable attachment and to provide soothing sentiments when affiliative signals arise, allowing for affiliation and attachment behaviours (Gilbert, 2014; Richardson et al., 2016). Moreover, Richardson and colleagues (2016) emphasise that the three systems regulate each other to produce blended affects. For instance, ‘affiliation’ is also associated with the drive system (e.g., excitement about a social relationship) and the threat system (e.g., anxiety when your wife becomes ill) (Richardson et al., 2016).

The label ‘soother’ is a recently coined term that is not yet commonly used within the literature. Research suggests that the assessment and treatment of CFS and FM should be multidimensional and patient-centred (Kwiatek, 2017; Castro-Marrero et al., 2017) and it appears that the soothers within Table 1 may be beneficial in this regard. Although the efficacy of current treatments is debated, there is some agreement that both FM (Macfarlane et al., 2016) and CFS (Roberts, 2018; Vink & Vink-Niese, 2018) treatment should focus on improving of quality of life, symptoms, and daily functioning.

**Table 1.***List of soothers used for this study*

- 
- 1) Mindfulness (living in the here and now and being non-judgmental)
  - 2) Aids (such as a wheelchair, jug, comfortable mattress or good pillows)
  - 3) Medication that reduces disease activity or symptoms
  - 4) Doing a fun thing with family or friends
  - 5) A leisure activity, such as reading, music, movie, dancing, drawing, painting or another hobby
  - 6) To be surrounded by lovely people (e.g., friends or family)
  - 7) Warm temperature (for example, bath, shower, infrared lamp, warm water bag)
  - 8) Having the freedom to do something in the way I want to do it myself
  - 9) Consistency and structure
  - 10) Something that cools me down (e.g., a cold shower, cold wind, cooling gels)
  - 11) Getting help from other people, such as kins or specialists
  - 12) Understanding my disease
  - 13) Getting a massage
  - 14) Supplements (vitamins, proteins, and so on)
  - 15) A calm surrounding such as nature, one's own house, pleasurable sound and light
  - 16) A good balance between activities and relaxation
  - 17) Having a good or positive conversation
  - 18) Healthy or good nutrition
  - 19) An alternative medicine such as osteopathy and reiki
  - 20) Seeing that people in my environment are happy and healthy
  - 21) A relaxation or breathing exercise such as yoga or meditation
  - 22) To take a rest or a break
  - 23) Intimacy
  - 24) Having a positive mindset (e.g., positive thinking, acceptance, being thankful)
  - 25) Talking to and sharing experiences with fellow sufferers
  - 26) Receiving physical affection such as being caressed or getting a hug
  - 27) Being accompanied by or caring for pets (e.g., horse, dogs, cats)
  - 28) Expressing myself to others and knowing that I'm not alone in all of this
  - 29) Professional help (for instance from physical therapists or psychologists)
  - 30) Taking a comfortable posture
  - 31) Being in a good mood
  - 32) A physical activity such as walking or cycling, gardening, Tai Chi or Qigong
  - 33) To be in a secure and trusted environment
  - 34) To remain within my limits or boundaries (e.g., saying 'no', or asking for help)
  - 35) Nice weather
  - 36) Sleeping
  - 37) Feeling recognized, understood, respected, loved, liked or important
  - 38) Drinking a yummy beverage (such as tea, cola or alcoholic drink), eating a treat (such as chocolate or sweets) or smoking a cigarette
  - 39) An activity in water (e.g., being in a pool, floating, hot tub)
  - 40) Performing a spiritual or religious activity such as going to church or praying
-

The FM literature reflects a preference for non-pharmacological soothers, but the integration of pharmacological soothers may also be effective. A number of soothers to be tested in this study have demonstrated mild to moderate efficacy within FM treatment. Examples include: Cognitive behavioural therapy (CBT) (e.g., education, goal-setting, cognitive restructuring) (Macfarlane et al., 2016; Aman et al., 2018, Pinto et al., 2020), exercise (e.g., tai chi, biofeedback, aqua aerobics, cardiovascular fitness regimens) (Aman et al., 2018; Busch et al., 2011; Kwiatek, 2017; Macfarlane et al., 2016; Offenbächer & Stucki, 2000; Pinto et al., 2020), Hydra/spa therapy/balneotherapy (Macfarlane et al., 2016; Nüesch, et al., 2013), patient education (Aman, 2018; Arnold, 2006; Kwiatek, 2017; Pinto et al., 2020) social support (e.g., social identification or the presence of a loved one) (Cameron et al., 2018; Montoya et al., 2004; Arnold, 2006) and Mindfulness/meditation (Aman, 2018; Adler-Neal & Zeidan, 2017).

CFS literature also indicates that both pharmacological and non-pharmacological strategies are used regularly. Seemingly, Graded exercise therapy (GET), CBT and Pacing are effective CFS treatments (Geraghty et al., 2019; Jackson, 2002; Rimes & Chalder, 2005; White et al., 2011), and these interventions also involve patient education (Castro-Marrero et al., 2017; Roberts, 2018; White et al., 2011). GET motivates patients to overcome their negative perceptions of themselves and their functional abilities through psychiatric assistance and gradually increasing the intensity of exercise (Kim et al., 2020). Pacing focuses on energy management through balancing rest and activity. Although these treatments have presented some positive outcomes, each have received criticism (Bjørkum et al., 2009; Cortes Rivera, 2019; Mengshoel et al., 2020; Sharpe et al., 2017; Twisk & Maes, 2009; Wilshire et al., 2017). Additionally, Kim and colleagues (2020) suggest that CBT is not a consistently effective treatment for CFS. Tentative evidence for nutritional supplements exists, however these findings have also been inconsistent (Castro-Marrero et al., 2017;

Rimes & Chalder, 2005; Sullivan, et al., 2009). Furthermore, social support was effective in some studies on CFS treatment (e.g., Jason et al., 2010), however several studies suggest it is not always important (e.g., LeGal et al., 2019; Prins et al., 2001; Prins et al., 2004; Whiting et al., 2001).

Both CFS and FM patients are most likely to benefit from non-pharmacological treatments, involving education, physical exercise, and psychological interventions. The evidence for pharmacological treatments remains limited (Castro-Marrero et al., 2017; Pinto et al., 2020; Rimes & Chalder, 2005). Non-pharmacological treatments produce fewer side effects and are able to target a broader range of symptoms compared to pharmacological treatments, and are generally recommended as first-line treatment (Kim et al., 2020; Thieme et al., 2017). Nevertheless, this does not mean that treatments should be identical, or that FM and CFS are different manifestations of a singular FSS, as subtle yet important differences clearly exist.

### **1.3 The Current Study**

This study is part of a project aiming to develop and validate an online questionnaire measuring the impacts of threats, soothers and drives on somatic symptoms. Previous research identified 40 threats, 40 soothers and 40 drives in patients with persistent physical symptoms (see Geenen et al., 2020), and these will be incorporated into the questionnaire. The current study focuses on the ability of some soothers or dimensions of soothers to alleviate symptoms of these FSSs. By addressing this gap in the literature, the author intends to add some evidence to the ‘Lumpers vs. Splitters’ debate. It is hypothesized that there will be significant differences in the perceived effectiveness of certain soothers for each illness. If the findings suggest that some soothers or soothers clusters are more effective at alleviating

CFS symptoms in comparison to FM symptoms, and vice versa, then this would represent a significant argument for the ‘Splitters’ perspective, while also informing future treatment.

#### **1.4 Hypotheses**

Based on the literature, the hypotheses are:

**H1:** Social activity, and in particular the soothers “Doing a fun thing with family or friends” and “Being surrounded by family or friends”, will be rated as more soothing by FM patients (Arnold, 2006; Cameron, et al., 2018; Cooper & Gilbert, 2017; Montoya et al., 2004; Pinto et al., 2020), compared to CFS patients (LeGal et al., 2019; Prins et al., 2001; Prins et al., 2004; Whiting, et al., 2001) – these studies indicate that social support is not always effective in CFS treatment. The analyses will also investigate whether the groups differ on Social Functioning (see RAND Short Form Survey), as this may also be related to social support.

**H2:** The soothers “Consistency and structure”, “A good balance between activities and relaxation” (Balance between activities), and “To remain within my limits or boundaries” (Remaining within my limits) will be rated as more soothing by CFS patients, compared to FM patients – because running out of energy is a more central problem for CFS patients (see links for ICD 10 criteria for CFS and FM in the reference section).

**H3:** FM patients will report higher pain severity compared to CFS patients – based on the nature of the syndrome itself.

**H4:** CFS patients will report greater fatigue severity compared to FM patients – based on the nature of the syndrome itself.

**H5:** Comorbid patients (those with both FM and CFS) will have poorer scores on mental wellbeing, physical functioning, daily functioning and somatic symptom severity – symptoms should be more severe for those experiencing both syndromes (Natelson, Lin, Lange, Khan, Stegner, & Unger, 2019).

## **2.0 Methods**

### **2.1 Participants**

Participants from the general population with FSSs or persistent physical symptoms were recruited to identify the threats, soothers and drives that may affect their symptoms. The current study focused on the impact of soothers on CFS and FM patients' somatic symptoms, mental wellbeing and physical functioning, and the differences between the two syndromes, on two self-report measures, the RAND Short Form Survey (SF-36), (VanderZee et al., 1996) and the Patient Health Questionnaire (PHQ-15) (Kroenke et al., 2002). Participants were 18 years or older. Data was collected for 804 individuals (Male = 50, Female = 753, Other = 1, left this question unanswered = 157). The 157 individuals who left this question unanswered clicked to participate but stopped when having read the information letter. The modal age group was 51-60 years old.

### **2.2 Procedure**

The project group asked patient associations to put a recruitment message on their websites (Facebook etc.). Participants could click on a link which brought them to the online questionnaire at Qualtrix. There they were presented with an information letter, informed consent, and the questionnaire. Participation was anonymous. The study was approved by the Ethics Committee of the Faculty of Social and Behavioural Sciences of Utrecht University (20-497).

### 2.3 Materials

An exploratory factor analysis of the 40 soother items was conducted to assess the relationship between the soothers and patient wellbeing. The scale asked participants to “Indicate how comforting the following circumstances are in your life” for each soother. An example item is “Doing a fun thing with family or friends”. The Likert-type scale responses included: 1 = “Not (comforting)”, 2 = “A little”, 3 = “Moderately”, 4 = “Very much”. The SF-36 was used – this 36-item questionnaire assesses Physical functioning, Role limitation due to physical health (Role physical health), Role limitation due to emotional problems (Role emotional problems), Energy/Fatigue, Social functioning, Emotional wellbeing, Pain, General health, and Health change of individuals. The SF-36 total scores were computed based on the RAND Corporation website’s scoring instructions (“36-Item Short Form Survey (SF-36) Scoring Instructions”, 2021). High scores on the SF-36 and each individual subscale are indicative of lower disability. The PHQ-15 was scored on a 3-point Likert-type scale, assessing the participants somatic symptom severity. This scale has strong construct validity and internal reliability (Cronbach’s  $\alpha = .8$ ) (Kroenke et al., 2002). The questionnaire has 15 items, each with the response options: 1 = “Not at all”, 2 = “A little”, 3 = “Very much”. The final scores are interpreted as follows: 1-4 = “Minimal (somatic symptom severity)”, 5-9 = “Low”, 10-14 = “Medium”, 15-30 = “High”.

### 2.4 Statistical Analysis

SPSS version 27.0 for Mac was used. A  $p$  value  $< .05$  was considered significant for each test. Principal axis factoring with oblimin rotation was used to derive the dimensions of the soothers section of the questionnaire for FM, CFS, and Comorbid patients. The number of factors was decided using the eigenvalue criterion  $>1$ , the eigen value scree-plot (Appendix B), the factor loading pattern (Field, 2009) and the contents of the factors. The final version



exclusion criteria for the items from the factor analysis was that the required factor loadings were  $<.40$  on any single factor (Comrey & Lee, 1992). The factor scores were computed by obtaining the mean scores of the items from each cluster. The internal consistency of the final dimensions of the soother clusters were calculated using Cronbach's  $\alpha$  coefficients (.10, .30 and .50 representing small, medium, and large correlations respectively) (Cohen, 1992).

The data distributions were screened and the majority of the score distributions did not exceed a skewness of -1.00 or 1.00, indicating normal distributions for these scales (e.g., Bulmer, 1979). Z-tests regarding the null hypothesis of normal distribution are often rejected in studies with large samples, as the standard error generally decreases in this scenario, even if the distribution does not substantially differ from normality. Therefore, adjusted critical values for rejecting the null hypothesis are required based on sample size. When the sample size exceeds 300, one must examine the histograms and the absolute values of skewness and kurtosis and  $z$ -values may be ignored. Absolute skewness values greater than 2 and absolute kurtosis values greater than 7 are acceptable as the critical values for non-normality in this case (Kim, 2013). Based on this, there were significantly skewed scores for CFS patients (3.59) and Comorbid patients (2.89) on Role physical health. There were also some kurtotic values for CFS patients (13.92) and Comorbid patients (8.31) on Role physical health (see Appendix C for skewness and kurtosis values). These non-normal distributions are somewhat understandable as the study participants have debilitating conditions that may significantly hinder their physical health, hence impacting their daily "roles".

### **3.0 Results**

In this section the outcomes of the factor analysis are reported first, followed by descriptive data, and finally the hypotheses are tested. The majority of the results were generated through a one-way ANOVA with a Tukey-HSD post-hoc examination. Levene's

assumption of the homogeneity of variances was violated on measures of Role physical health ( $p < .001$ ), Energy/Fatigue ( $p = .04$ ) and Pain ( $p = .01$ ), and the soother cluster “Comfort” ( $p = .04$ ), hence a Welch test and a Games-Howell post-hoc were also conducted. Additionally, due to several non-normal distributions, a Kruskal-Wallis and several Mann-Whitney U post-hoc examinations were performed.

### **3.1 Dimensions of soothing**

The factor analysis provided a score of .93 on the Kaiser–Meyer–Olkin measure of sampling adequacy, suggesting that the analysis contained acceptable and reliable factors (Field, 2009). The Bartlett’s test of sphericity was significant ( $\chi^2 = 8868.898, p < .001$ ), therefore we may reject the null hypothesis that the variables in the correlation matrix are uncorrelated (Field, 2009), and hence the factor analysis was appropriate. Table 2 displays the results of principal axis factoring for 9 soother-factors. The eigenvalue scree plot and factor loading patterns after rotation suggested nine factors, only seven of which presented item-factor correlations that contained more than one correlated item ( $>.40$ ) for each factor in the pattern matrix. 55% of the total variance was explained by the 9 extracted factors. Nineteen soother items were deleted, each with factor loadings of  $<.4$ . The remaining 21 items loaded on 7 factors (Table 3). Only 4 factors (subscales) had acceptable Cronbach’s  $\alpha$  coefficients ( $>.7$ ): Comfort ( $\alpha = .73$ ), Mindfulness ( $\alpha = .72$ ), Social activity ( $\alpha = .77$ ) and Balance, freedom and structure ( $\alpha = .73$ ). Professional care ( $\alpha = .44$ ), Coping with disease ( $\alpha = .51$ ) and “Me time” ( $\alpha = .37$ ) were omitted from the final questionnaire due to low internal consistency (Table 3).

**Table 2.***Principal axis factor loadings: Pattern Matrix of the Soother items*

<b>Soother Items</b>	<b>1</b>	<b>2</b>	<b>3</b>	<b>4</b>	<b>5</b>	<b>6</b>	<b>7</b>	<b>8</b>	<b>9</b>
Taking a comfortable posture	<b>0.629*</b>	-0.058	0.032	0.106	0.232	0.05	-0.023	0.107	-0.107
To remain within my limits or boundaries	<b>0.589*</b>	0.075	-0.034	-0.101	0.074	-0.097	0.099	-0.038	0.153
Being in a good mood	<b>0.438*</b>	0.009	-0.039	0.277	-0.03	0.012	0.007	0.162	0.1
Sleeping	<b>0.417</b>	0.027	0.185	-0.109	0.034	-0.096	0.103	0.121	0.006
To take a rest or a break	0.398	0.284	-0.031	-0.005	0.067	-0.034	0.167	-0.101	0.059
A physical activity	0.356	0.109	0.022	0.203	-0.186	0.191	0.056	0.054	-0.081
Having a positive mindset	0.353	0.132	0.022	0.306	-0.096	0.004	-0.072	0.018	0.173
To be in a secure and trusted environment	0.313	-0.006	-0.269	0.049	0.014	0.122	0.238	0.267	0.184
A relaxation or breathing exercise such as yoga or meditation	-0.017	<b>0.802*</b>	0.104	-0.082	-0.063	0.062	0	0.036	0.016
Mindfulness	0.01	<b>0.697*</b>	-0.065	0.066	0.076	-0.045	0.078	-0.1	-0.001
Performing a spiritual or religious activity	0.04	0.24	-0.024	0.081	0.103	0.164	-0.076	0.018	0.02
Getting a massage	0.041	0.005	0.656	0.024	-0.006	0.041	-0.026	0.044	0.098
Warm temperature	0.03	0.129	0.334	0.13	0.158	-0.256	0.217	0.124	-0.111
An alternative medicine such as osteopathy and reiki	-0.018	0.231	0.33	-0.073	-0.077	0.298	0.067	0.101	0.01
Professional help	0.067	-0.012	0.282	0.062	0.229	0.199	-0.081	0.061	0.153
Doing a fun thing with family or friends	0.001	-0.114	0.126	<b>0.797*</b>	0.062	0.005	-0.022	-0.032	0.031
To be surrounded by friends and family	-0.035	0.082	-0.028	<b>0.656*</b>	0.027	-0.078	0.039	0.093	0.126
Leisure activity	0.006	0.124	-0.078	<b>0.589*</b>	0.024	-0.004	0.174	0.062	-0.088
Having a positive conversation	0.07	0.146	-0.044	0.315	-0.017	0.138	0.166	0.116	0.251
Seeing that people in my environment are happy and healthy	0.123	0.077	-0.141	0.313	-0.074	0.171	0.017	0.176	0.141
Aids	0.098	0.184	-0.037	0.012	<b>0.499*</b>	-0.116	-0.002	0.064	0.039
Medication	0.032	-0.034	0.012	0.009	<b>0.479*</b>	0.095	-0.024	-0.025	0.003
Something that cools me down	-0.032	0.022	0.002	-0.012	0.049	0.39	0.017	-0.02	-0.012
Supplements	0.014	0.053	0.282	0.012	0.062	0.331	0.214	0.017	-0.031
Having the freedom to do something in the way I want to do it myself	0.104	0.032	0.196	0.2	-0.042	-0.046	<b>0.534*</b>	-0.009	0.051
Consistency and structure	0.029	0.032	-0.095	0.012	0.021	0.115	<b>0.448*</b>	0.106	0.026
A good balance between activities and relaxation	0.298	0.164	0.126	0.102	-0.073	-0.126	<b>0.407*</b>	-0.111	0.096
A calm surrounding such as nature, one's own house, pleasurable sound and light	0.182	0.197	-0.059	0.011	-0.024	0.091	<b>0.403*</b>	0.154	0.065
Healthy or good nutrition	0.16	0.194	-0.012	0.145	-0.065	0.187	0.273	0.043	0.016
Drinking a yummy beverage, eating a treat, or smoking a cigarette	-0.001	-0.068	-0.092	0.102	-0.014	0.074	0.051	<b>0.474*</b>	0.029
An activity in water	0.024	0.189	0.221	0.025	0.117	-0.088	-0.17	<b>0.452*</b>	-0.074
Nice weather	0.133	-0.051	0.165	0.131	-0.013	-0.083	0.092	0.387	-0.052
Being accompanied by or caring for pets	0.027	-0.02	0.075	-0.116	-0.031	-0.017	0.085	0.231	0.149
Understanding my disease	-0.02	0.018	0.025	0.048	0.192	-0.084	0.236	-0.003	<b>0.457*</b>
Physical affection	0.127	0.062	0.168	0.253	-0.068	-0.034	-0.114	0.125	<b>0.439*</b>
Getting help from other people, such as kin or specialists	0.002	-0.052	0.037	0.085	0.382	0.149	0.125	-0.067	<b>0.415*</b>
Feeling recognized, understood, respected, loved, liked or important	0.114	0.093	-0.051	0.026	0.03	-0.096	0.096	0.385	0.391
Expressing myself to others and knowing that I'm not alone in all of this	0.2	0.083	-0.006	0.057	0.144	0.044	0.015	0.136	0.385
Intimacy	0.24	0.14	0.174	0.216	-0.177	0.001	-0.177	-0.01	0.365
Talking to and sharing experiences with fellow sufferers	-0.03	0.115	0.07	0.137	0.116	0.12	-0.043	0.114	0.274

\*  $p > .4$

**Table 3.***Soother subscales based off factor loadings*

<b>Comfort</b>	<b>Mindfulness</b>	<b>Social activity</b>	<b>Balance, freedom and structure</b>	<b>* Professional Care</b>	<b>* Coping with disease</b>	<b>* “Me time”</b>
1. Taking comfortable posture	1. Relaxation exercise	1. Doing a fun thing with family or friends	1. Having freedom to do things the way I want	1. Medication	1. Understanding my disease	1. Drinking a yummy beverage, eating a treat, or smoking a cigarette
2. Remain within limits	2. Mindfulness	2. Being surrounded by family or friends	2. Consistency and structure	2. Aids	2. Physical affection	
3. Being in a good mood		3. Leisure activity	3. Balance between activities		3. Getting help from others	2. An activity in water
4. Sleeping			4. Calm surroundings			

*Note: The subscales marked with an ‘\*’ were omitted from the final analyses as they had low internal consistency*

**Table 4.***Descriptive statistics on FM, CFS and Comorbid patients*

Measure	FM ( <i>n</i> = 357)		CFS ( <i>n</i> = 48)		Comorbid ( <i>n</i> = 82)		<i>F</i> (2, 484)	$\eta_p^2$
	M	SD	M	SD	M	SD		
<b>Comfort</b>	3.28	.62	3.24	.56	3.24	.72	.19	.001
<b>Mindfulness</b>	2.42	.99	2.43	.89	2.44	.92	.02	< .001
<b>Social activity</b>	3.05	.76	2.92	.90	3.03	.76	.67	.003
<b>Balance, freedom and structure</b>	3.32	.64	3.44	.61	3.28	.70	1.01	.004
<b>Physical functioning</b>	54.37	20.15	44.69	24.37	40.43	22.20	17.24**	.066
<b>Role physical health</b>	19.96	30.78	4.69	14.27	10.37	23.55	8.65**	.035
<b>Role emotional problems</b>	50.98	44.80	57.64	47.01	36.59	43.39	4.39*	.018
<b>Energy/fatigue</b>	36.04	15.80	26.15	15.24	27.01	11.83	18.18**	.07
<b>Social functioning</b>	52.94	22.61	25.52	22.91	36.28	22.87	42.84**	.15
<b>Emotional wellbeing</b>	62.03	17.40	63.25	18.17	58.98	18.50	1.22	.005
<b>Pain</b>	41.56	16.86	49.69	23.18	32.77	17.35	14.88**	.058
<b>General health</b>	39.10	16.74	29.17	14.04	28.54	14.79	19.45**	.074
<b>Health change</b>	41.95	24.43	34.38	23.98	32.93	23.53	5.89**	.024
<b>Total SF-36</b>	46.15	14.34	38.73	13.49	35.41	13.41	22.53**	.085
<b>Total PHQ-15</b>	13.32	3.90	14.62	4.41	15.49	3.83	11.23**	.044

\*  $p < .05$ , \*\*  $p < .01$ 

*Note: ANOVA test descriptive statistics are in black font, Welch test descriptive statistics are highlighted in grey*

### 3.2 Descriptive Statistics

The descriptive statistics reveal that the ANOVA and Welch test  $\eta_p^2$  (partial eta squared) effect sizes for the soother clusters were quite small (Table 4). FM patients' mean scores indicated higher levels of general functioning and wellbeing compared to CFS and Comorbid patients'. Although FM patients' mean scores were higher on Social activity and lower on Balance, freedom and structure, these differences did not reach significance. Total SF-36 exhibited a notably large effect size and results revealed that FM patients scored significantly higher than CFS and Comorbid patients on the Total SF-36, indicative of lower disability. Finally, FM patients scored significantly lower than Comorbid patients, but not CFS patients on Total PHQ-15, indicative of superior overall health (Table 4).

### 3.3 Social Activity and Social Functioning

H1 predicted that Social activity, and in particular, the soothers "Doing a fun thing with family or friends" and "Being surrounded by family or friends" would be more soothing for FM patients compared to CFS patients. No significant differences existed between the groups for any measures related to Social activity ( $p < .05$ ), however, the one-way ANOVA revealed significant differences between the groups on Social functioning [ $F(2, 484) = 42.84, p < .001$ ]. The Tukey-HSD post-hoc revealed that FM patients' mean score ( $M = 52.94, SD = 22.61$ ) was significantly higher than CFS ( $M = 25.52, SD = 22.91$ ) and Comorbid patients' ( $M = 36.28, SD = 22.87$ ), indicating that FM patients' Social Functioning was significantly better than the other two groups'. Nonetheless, based on these findings one may accept the null hypothesis of H1.

### 3.4 Balance, Structure and Limits

H2 speculated that the soothers “Consistency and structure”, “Balance between activities”, and “Remaining within my limits” would be more soothing for CFS patients’ symptoms compared to FM patients’. Several Mann-Whitney U tests were conducted as nonparametric analyses of these soothers. This causes an inflation in the Type I error rate, hence a Bonferroni adjustment was conducted by dividing the alpha level of significance (.05) by the number of comparisons to be made (3). The new alpha level for the Mann-Whitney U test was .0167. The only significant result was that CFS patients ( $Mdn = 4.00, n = 48$ ) rated having “Balance between activities” significantly more soothing than FM patients ( $Mdn = 4.00, n = 357$ ),  $U = 6904.00, z = -2.50, p = .01, r = .12$ . Consequently, one may mostly accept the null hypothesis of H2.

### 3.5 Pain Severity

H3 postulated that FM patients would report higher pain severity compared to CFS patients. The Welch test revealed a significant effect for syndrome type on the Pain subscale of the RAND-36 [ $F(2, 96.38) = 12.52, p < .001$ ]. The Games-Howell post-hoc found that FM patients ( $M = 41.56, SD = 16.86$ ) did not score significantly lower (which would have indicated higher pain severity) compared to CFS patients ( $M = 49.69, SD = 23.18$ ) ( $p = .058$ ). Comorbid patients ( $M = 32.77, SD = 17.35$ ) however, scored significantly lower than both CFS and FM on this subscale ( $p < .001$ ). Due to non-normal distributions (based on histograms and Shapiro Wilk values) a Kruskal-Wallis test was conducted to analyse PHQ-15 items related to pain (Table 5). Several significant differences were found between the groups, hence Mann-Whitney U post-hoc tests were performed (Table 6). Compared to CFS patients, FM patients had significantly higher mean ranks on I2,  $U = 6668.00, z = -5.01, p < .001, r = -.23$  and I3,  $U = 7392.00, z = -5.67, p < .001, r = -.27$ . Comorbid patients had

higher mean ranks on all pain related items, however, only I1,  $U = 13512.00$ ,  $z = -3.41$ ,  $p = .001$ ,  $r = -.16$ , and I5,  $U = 13572.00$ ,  $z = -3.44$ ,  $p = .001$ ,  $r = -.16$ , reached statistical significance. Finally, Comorbid patients scored significantly higher on I2,  $U = 1944.00$ ,  $z = -1.95$ ,  $p < .001$ ,  $r = -.16$ , and I3,  $U = 1568.00$ ,  $z = -4.73$ ,  $p < .001$ ,  $r = -.40$ . Based on this one may, one may reject the null hypothesis of H3.

**Table 5.**

*Results of the Kruskal-Wallis test examining PHQ-15 items related to pain and fatigue*

Measure	FM (n = 390)	CFS (n = 54)	Comorbid (n = 88)	$\chi$	H	Asymp. Sig.
	Mean rank	Mean rank	Mean rank			
I1 – Stomach pain	256.59	262.50	312.86	8.39	11.58	.003**
I2 – Back pain	272.78	175.47	294.55		30.17	<.001**
I3 – Pain in your arms, legs or joints, knees, hips, elbows	273.15	192.26	282.59		37.21	<.001**
I4 – Headache	258.76	276.61	294.61	8.79	5.14	.077
I5 – Chest pain	255.85	270.02	311.52	9.71	11.88	.003**
I10 – Pain or problems with sexual contact	263.01	281.40	272.81	1.27	1.09	.581
I13 – Fatigue or low energy	254.29	293.55	304.00		25.75	<.001**

\*  $p < .05$ , \*\*  $p < .01$

*Note: According to SPSS, median tests could not be performed on items 2, 3, and 13 as all values were less than or equal to the median. Hence there are no Chi-Square ( $\chi$ ) statistics for these items.*



**Table 6.**

*Results of Mann-Whitney U test comparing FM, CFS and Comorbid patients on PHQ-15*

*items related to pain and fatigue*

<b>1. Measure</b>	<b>FM (n = 390)</b>	<b>CFS (n = 54)</b>	
	<b>Mean rank</b>	<b>Mean rank</b>	<b>Z-Value</b>
<b>I1 – Stomach pain</b>	221.95	226.50	-.27
<b>I2 – Back pain</b>	232.40	150.98	-5.01**
<b>I3 – Pain in your arms, legs or joints, knees, hips, elbows</b>	230.71	163.22	-5.67**
<b>I4 - Headache</b>	220.77	235.03	-.852
<b>I5 – Chest pain</b>	221.05	232.94	-.725
<b>I10 – Pain or problems with sexual contact</b>	220.65	235.83	-.921
<b>I13 – Fatigue or low energy</b>	218.53	251.18	-2.697*
<b>2. Measure</b>	<b>FM (n = 390)</b>	<b>Comorbid (n = 88)</b>	
	<b>Mean rank</b>	<b>Mean rank</b>	<b>Z-Value</b>
<b>I1 – Stomach pain</b>	230.15	280.95	-3.41**
<b>I2 – Back pain</b>	235.87	255.57	-1.43
<b>I3 – Pain in your arms, legs or joints, knees, hips, elbows</b>	237.94	246.41	-.925
<b>I4 - Headache</b>	233.49	266.13	-2.25
<b>I5 – Chest pain</b>	230.30	280.27	-3.44**
<b>I10 – Pain or problems with sexual contact</b>	237.86	246.77	-.617
<b>I13 – Fatigue or low energy</b>	231.26	276.00	-4.41**
<b>3. Measure</b>	<b>CFS (n = 54)</b>	<b>Comorbid (n = 88)</b>	
	<b>Mean rank</b>	<b>Mean rank</b>	<b>Z-Value</b>
<b>I1 – Stomach pain</b>	63.50	76.41	-1.95
<b>I2 – Back pain</b>	51.99	83.47	-4.98**
<b>I3 – Pain in your arms, legs or joints, knees, hips, elbows</b>	56.54	80.68	-4.73**
<b>I4 - Headache</b>	69.08	72.98	-.595
<b>I5 – Chest pain</b>	64.57	75.75	-1.72
<b>I10 – Pain or problems with sexual contact</b>	73.06	70.54	-.392
<b>I13 – Fatigue or low energy</b>	69.87	72.50	-1.81

\*  $p < .0167$ , \*\*  $p < .00167$

*Note: FM and CFS comparisons are labelled 1, FM and Comorbid comparisons are labelled*

*2, and CFS and comorbid comparisons are labelled 3.*

### 3.6 Fatigue Severity

H4 predicted that CFS patients would report greater fatigue severity compared to FM patients. The Welch test reported a significant difference between the groups on the Energy/Fatigue subscale of the SF-36 [ $F(2, 108.58) = 21.74, p < .001$ ]. The Games-Howell post-hoc revealed that FM patients ( $M = 36.04, SD = 15.80$ ) scored significantly higher on Energy/Fatigue (indicative of lower fatigue) compared to CFS ( $M = 26.15, SD = 15.24$ ) and Comorbid patients ( $M = 27.01, SD = 11.83$ ). There were no significant differences between CFS and Comorbid patients on this subscale. Due to non-normal distributions (based on histograms and Shapiro Wilk values) a Kruskal-Wallis test was completed to analyse the PHQ-15 item related to fatigue (Table 5). The Mann-Whitney U post-hoc examination (Table 6) showed that compared to FM patients ( $Mdn = 2.00, n = 390$ ), both CFS patients ( $Mdn = 2.00, n = 54$ ),  $U = 8981.50, z = -2.70, p = .007, r = -.13$ , and Comorbid patients ( $Mdn = 2.00, n = 88$ ),  $U = 13948.00, z = -4.40, p < .001, r = -.2$ , had significantly higher mean ranks on I13. Although Comorbid patients did have higher mean ranks compared to CFS, the difference was not significant  $U = 2288.00, z = -1.81, p = .07, r = -.15$ . After interpreting these results, one may reject the null hypothesis of H4.

### 3.7 Are Comorbid Patients More Severely Affected?

H5 postulated that Comorbid patients would have lower scores on the SF-36 and higher scores on the PHQ-15, and would generally be more severely affected by their symptoms. The ANOVA revealed significant differences for Total SF-36 [ $F(2, 484) = 22.53, p < .001$ ] and Total PHQ-15 [ $F(2, 484) = 11.23, p < .001$ ]. The Tukey-HSD pot-hoc indicated that FM patients scored significantly higher compared to CFS and Comorbid patients on the Total SF-36 ( $M = 46.15, SD = 14.34$ ). FM patients scored significantly lower than Comorbid patients (but not CFS patients) on Total PHQ-15 ( $M = 13.32, SD = 3.90$ ). Comorbid patients reported

scores indicative of poorer wellbeing on all subscales compared to FM, and poorer than CFS on 8 of 15 subscales. Therefore, one may partially reject the null hypothesis of H5.

#### **4.0 Discussion**

The current study examined the potential effects of certain soothers on the symptoms of FM, CFS and Comorbid patients, and the symptomatic differences between these FSSs. This exploratory study intended to present tentative associations that may be further examined in future studies, while also aiming to add to the Lumpers versus Splitters debate. The aetiological and pathophysiological origins of these FSSs are still largely unclear, however the results suggest that there are some potentially important differences between these syndromes, despite their overlapping symptomology. Based on these findings, there is tangible evidence to suggest that fatigue is a more prominent feature of CFS than FM. Moreover, compared to FM patients, CFS patients seem to be more severely affected across the board. The study's key findings suggest that Comorbid patients are more severely affected than FM patients and slightly more affected than CFS patients across the measures. Only one significant difference was found between the three groups regarding the soother clusters or individual soothers. The findings highlight both the differences and similarities between FSSs, which does not completely align with the splitter argument.

#### **4.1 Accounting for the Findings**

H1 posited that Social activity, and in particular, the soothers "Doing a fun thing with family or friends" and "Being surrounded by family or friends" would more soothing for FM patients' symptoms compared to CFS patients'. The results did not reflect this. Perhaps these soothers may not encapsulate the phenomenon of 'social support'. For example, it is possible to spend time with loved ones and enjoy oneself, but to also not feel supported. Studies stress

the importance of ‘perceived’ social support in CFS patients (e.g., McManimen et al., 2018; Jason, Witter, & Torres-Harding, 2003), and this may be interesting to investigate in future research. The aforementioned soothers were generally rated “A little” to “Moderately” comforting (see Appendix D), indicating that they may be useful within CFS and FM treatment. FM patients’ Social Functioning was significantly better than the other two groups, which may be related to FM patients’ seemingly less severe symptoms (as reflected by the current findings), however future research must investigate this postulation.

H2 predicted that soothers such as “Consistency and structure”, “Balance between activities”, and “Remaining within my limits” would be rated as more affective for alleviating CFS symptoms compared to FM symptoms. Only “Balance between activities” was significantly more soothing for CFS patients – this ties in with the concept that energy management is more important for CFS and should be investigated in future studies. No other significant differences were found, which may again be due to these soother titles’ inability to encapsulate the concept of energy management, or because energy management is equally important for FM patients (Huijnen et al., 2015). These soothers were generally rated as “Moderately” soothing judging by the mean scores (Appendix D), indicating that they may be useful for treating both these FSSs.

H3 (relating to pain severity) was rejected and H4 (relating to fatigue severity) was accepted based on the current findings. The outcomes of H3 may relate to the failure of the measures to encapsulate the concept of widespread pain (the main diagnostic feature of FM) In terms of H4, Cortes Rivera and colleagues (2019) highlighted metabolic imbalances and compromised mitochondrial functioning in CFS patients which does not appear to be a factor within FM. More research is needed in this area, because it is hard to distinguish between cause and consequence in pain research.

Comorbid patients reported poorer wellbeing on all subscales compared to FM and poorer than CFS on 8 of 15 subscales. In the authors opinion, it was surprising how severe CFS patients rated their symptoms. CFS patients displayed higher symptom severity in general compared to FM patients, which may be related to the slightly more restricted and rigid CFS diagnostic criteria (Natelson, 2019). CFS patients exhibit more severely disturbed energy levels which may have larger consequences for wellbeing and functioning compared to FM patients' widespread pain. There are no known biomarkers for these FSSs, hence diagnosis solely depends on clinical criteria. One distinct discrepancy between these two diagnoses is that the presence of any medical cause of severe fatigue excludes patients from receiving a CFS diagnosis, however this is not the case with FM diagnoses. Alternatively, patients who do not exhibit any other cause of body-wide pain are ascribed a primary FM diagnosis, and individuals presenting coexisting rheumatologic diagnoses are assigned a secondary FM diagnosis (Natelson, 2019). This diagnostic discrepancy is responsible for a 10-fold difference in prevalence with CFS in approximately 0.3% of the population and FM in approximately 4% (Jason, et al., 1999; White et al., 1999, as seen in Natelson, 2019).

## **4.2 Implications**

In particular, H2 provided tentative evidence that “Balance between activities” (which coincides with the principles of Pacing therapy) could be more beneficial within CFS treatment compared to FM treatment, and future research should explore this theory. No significant differences between the groups on any other measures related to H1 and H2, however these results are still useful. The soothers tested were generally rated “A little” to “Moderately” comforting, indicating that they may be beneficial in the treatment of both syndromes, and future research should also investigate this possibility. Lumpers may argue that this indicates that CFS and FM are manifestations of a greater FSS, but this is not solid

evidence of identical pathophysiology. Many therapies and therapeutic techniques are effective across a range of disorders, diseases and syndromes. For instance CBT methods are used to treat numerous FSSs, psychological disorders, and psychogenic movement disorders (Cuijpers et al., 2016; Gupta & Lang, 2009; Henningsen, Zipfel, & Herzog, 2007). Evidently, the key to maximising these therapies' efficacy is by adapting them to the patient's needs (Secker, & Brown 2005, Erickson, & Newman, 2005; Johansson et al., 2012). The fact that so little is known of the origins of these FSSs is an extra incentive to offer patients specifically tailored treatments in order to provide optimal care (Carrillo de la Peña, et al., 2015, Kwiatek, 2017; Castro-Marrero et al., 2017).

The author reiterates that the differences between the two illnesses are more important than the similarities. The findings relating to fatigue and general symptom severity highlight differences in the symptomatic elements of these FSSs. Although widespread pain is documented as the primary symptom of FM, and fatigue as the defining symptom of CFS, these criteria have been challenged because both syndromes involve debilitating pain and fatigue (McKay et al., 2019). In accordance to these concerns, Natelson (2019) worked with the Centre for Disease Control and Prevention to improve and operationalize the assessment of fatigue severity in CFS by asking patients to rate their reduction in activity, and the duration and burden of several other symptoms (sore throat, unrefreshing sleep etc.). Additionally, patients are asked whether minimal physical or mental exertion cause these other symptoms to occur. Moreover, the American College of Rheumatology have devised an operational definition of widespread pain within FM: "pain on the left and right sides of the body, above and below the waist, and accompanied by pain in the cervical spine, anterior chest, thoracic spine, or lower back" (Natelson, 2019, p. 614). With these updated diagnostic criteria, the status of widespread pain as the primary diagnostic symptom for FM and fatigue as the primary diagnostic symptom for CFS may become more accurate.

Clearly there is substantial overlap in symptoms and treatment methods regarding these FSSs, but subtle differences such as those highlighted by the outcomes of H2 and H4 could be particularly important to consider for future research on the pathophysiological basis of these FSSs and the individualisation of their treatments. Similar to this study, Joustra (2019) observed higher general symptom and fatigue severity within CFS patients, and even found higher pain severity within FM patients. Based on the current findings and the literature, CFS treatment should focus more on improving and maintaining energy levels, and that CFS and FM are rightly classified as separate.

Potentially the most valuable finding of this study was that Comorbid patients generally reported scores indicative of poorer wellbeing compared to CFS and in particular FM. The outcomes coincide with those of Natleson's (2019) review which strongly indicated that comorbidity (FM and CFS) is not simply a more severe manifestation of a singular FSS, but a categorically different syndrome to CFS. Comorbid patients' increased symptom severity indicates that CFS and FM diagnosed together form a separate comorbid condition, as do the results indicating that CFS patients were generally more severely affected by their symptoms compared to FM patients. These findings emphasise the importance of labels, a view complemented by Noble and colleagues (2018). They highlighted that despite the potential negative effects of labelling (e.g., psychological distress), it may also provide positive effects (e.g., validating patients' experiences) (Noble et al., 2018; Ward & Horrocks, 2015). Paradoxically, several studies indicate that labelling may actually be useful in scenarios where the patient is experiencing a disease that is not particularly well understood (e.g., FM and CFS) (Huibers & Wessely 2006; de Lourdes Drachler et al., 2009). Patients often worry that without a concrete diagnosis they will not be taken seriously (Åsbring & Närvänen, 2004), and the sense of living with a mystery illness, lacking legitimacy, further affects their participation in several areas such as family-life, work, and social relationships.

Consequently, many also find it difficult to decide who to talk to about their condition (de Lourdes Drachler et al., 2009; Whitehead, 2006). This too is true for FM – Goldenberg (2009) accentuates that together, diagnosis and patient education can lower the risk of psychosocial factors developing into maladaptive illness behaviours. If explained correctly, diagnostic labels can reassure patients and healthcare providers that there is a rational explanation for their symptoms, leading to fewer referrals, tests, or invasive therapies (Goldenberg 2008; Goldenberg, 2009). Therefore, labelling these FSSs as separate syndromes is practical for patients and practitioners alike.

### **4.3 Limitations and Future Research**

This study presented several limitations. Firstly, the study did not examine possible risk factors such as education, gender, age, and disease onset (Monden et al., 2020; Fitzcharles et al., 2014; Hempel et al., 2008; Rusu et al., 2015). For example FM is more prevalent in women (Heidari et al., 2017). Women also report more somatization and negative affect, and are more concerned with physical sensations than men (Hazemeijer & Rasker, 2003). The broad nature of the topic, word count restrictions, and the time-frame for this study meant that it focused more on the potential differences between soothers and symptoms regardless of these factors. Therefore, future research should investigate these factors.

Another limitation relates to the paucity of literature surrounding soothers with respect to FSSs. This research is in its infancy and a substantial body of literature may need to be built before any significant patterns are uncovered. Future research should investigate the use of soothers within multicomponent therapy (MT). There is tentative evidence to suggest the effectiveness of MT, which combines several therapy methods (e.g., educational, psychological and exercise components), but this requires further research (Nüesch et al., 2013; Loades et al., 2016; Serrat et al., 2020a). The multifaceted nature of these FSSs



suggests that they should incorporate individualised treatments (Carrillo de la Peña, et al., 2015; Hassett & Gevirtz, 2009; Cortes Rivera et al., 2019), and soothers could possibly aid in this individualisation.

Finally, this study fails to examine the effects of pharmacological soothers and supplements. Pharmacological treatments for both FSSs have provided inconsistent results (Collatz et al., 2016; Lacourt et al., 2013; Schmidt-Wilcke & Diers, 2017), however several studies highlight their potential benefits (Clauw, 2009; Collatz et al., 2016; Goldenberg et al., 2004; Macfarlane et al., 2016; Schmidt-Wilcke & Diers 2017). Research tentatively suggests that dietary supplements may be beneficial within FM and CFS treatment (Castro-Marrero et al., 2021; Pagliai et al., 2020; Rimes & Chalder, 2005; Werbach, 2000; Whiting et al., 2001), although many studies also claim that supplementation is not effective (Joustra et al., 2017). Pharmacological and supplementary treatments may only target one or two symptoms alone, however they may be more effective when used alongside non-pharmacological treatments within MTs (Monro & Puri, 2018; Schmidt-Wilcke & Diers 2017; Serrat et al., 2020b).

## **5.0 Conclusions**

Evidently, FSSs are incredibly complex and poorly understood concepts. Schadt and Björkegren (2012) highlight the complexity of the matter by affirming how health and disease patterns generally involve complex networks of interaction between genes, environment, diet, lifestyle, and social environment (Bland, 2017), while patients' perceptions, attributions, and coping skills, may also lead to the perpetuation of the illness (Afari & Buchwald, 2003; Cedraschi et al., 2013). This study's findings are marginally in favour of the splitters perspective. The differences between CFS and FM on the central (CFS) symptom of fatigue support the Splitter argument. Moreover, CFS patients were generally more negatively affected by their symptoms than FM patients, and Comorbid patients were

generally more negatively affected than both groups, which is even more convincing for the Splitter argument. This indicates that FM and CFS are different FSSs and that a combination of the two is a comorbid condition with increased symptom severity, which is in line with the findings of Natelson (2019). Furthermore, the science of Psychology as a whole tends to focus largely on healing or fixing what is ‘wrong’ or maladaptive in human beings, while neglecting the opportunity to promote and cultivate the positive qualities they may already possess (Seligman & Csikszentmihalyi, 2014).

This study’s findings: (1) suggest that Gilbert’s affect regulation model (2014) could help us to better understand these FSSs and may also inform future treatment (much like Pinto et al., 2020), (2) tentatively indicates that FM and CFS should be considered separate FSSs, and (3) provided tentative indications that soothers may be useful in the treatment of both these FSSs, while providing a platform for future studies to examine this. Nevertheless, this study by no means disproved the Lumper argument, as many similarities between these FSSs were clear throughout, therefore further research is needed in this area.

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## References

36-Item Short Form Survey (SF-36) Scoring Instructions. (2021). Retrieved 9 March 2021, from [https://www.rand.org/health-care/surveys\\_tools/mos/36-item-short-form/scoring.html](https://www.rand.org/health-care/surveys_tools/mos/36-item-short-form/scoring.html).

2021 ICD-10-CM Diagnosis Code M79.7: *Fibromyalgia*. Icd10data.com. (2021). Retrieved 27 April 2021, from <https://www.icd10data.com/ICD10CM/Codes/M00-M99/M70-M79/M79-/M79.7>.

2021 ICD-10-CM Diagnosis Code R53.82: *Chronic fatigue, unspecified*. Icd10data.com. (2021). Retrieved 27 April 2021, from <https://www.icd10data.com/ICD10CM/Codes/R00-R99/R50-R69/R53-/R53.82>.

Aaron, L. A., Burke, M. M., & Buchwald, D. (2000). Overlapping conditions among patients with chronic fatigue syndrome, fibromyalgia, and temporomandibular disorder. *Archives of internal medicine*, *160*(2), 221-227.

Abbi, B., & Natelson, B. H. (2013). Is chronic fatigue syndrome the same illness as fibromyalgia: evaluating the 'single syndrome' hypothesis. *QJM: An International Journal of Medicine*, *106*(1), 3-9.

Adler-Neal, A. L., & Zeidan, F. (2017). Mindfulness meditation for fibromyalgia: mechanistic and clinical considerations. *Current rheumatology reports*, *19*(9), 1-9.

- Afari, N., & Buchwald, D. (2003). Chronic fatigue syndrome: a review. *American Journal of Psychiatry*, *160*(2), 221-236.
- Aman, M. M., Yong, R. J., Kaye, A. D., & Urman, R. D. (2018). Evidence-based non-pharmacological therapies for fibromyalgia. *Current pain and headache reports*, *22*(5), 1-5.
- Arnold, L. M. (2006). Biology and therapy of fibromyalgia. New therapies in fibromyalgia. *Arthritis research & therapy*, *8*(4), 1-20.
- Åsbring, P., & Närvänen, A. L. (2004). Patient power and control: A study of women with uncertain illness trajectories. *Qualitative Health Research*, *14*(2), 226-240.
- Berenson, R. (2004). Lumpers And Splitters: Different Approaches To Understanding Variations Research. *Health Affairs*, *23*(Suppl2), VAR-98-VAR-103.  
<https://doi.org/10.1377/hlthaff.var.98>.
- Bjørkum, T., & Wang, C. E. (2009). Patients' experience with treatment of chronic fatigue syndrome. *Tidsskrift for den Norske laegeforening: tidsskrift for praktisk medicin, ny raekke*, *129*(12), 1214-1216.
- Bland, J. (2017). Defining function in the functional medicine model. *Integrative Medicine: A Clinician's Journal*, *16*(1), 22.

- Buchwald, D., & Garrity, D. (1994). Comparison of patients with chronic fatigue syndrome, fibromyalgia, and multiple chemical sensitivities. *Archives of Internal medicine*, *154*(18), 2049-2053.
- Bulmer, M. G. (1979). *Principles of statistics*. New York: Dover.
- Busch, A. J., Webber, S. C., Brachaniec, M., Bidonde, J., Dal Bello-Haas, V., Danyliw, A. D., Overend, T. J., Richards, R. S., Sawant A., & Schachter, C. L. (2011). Exercise therapy for fibromyalgia. *Current pain and headache reports*, *15*(5), 358.
- Buskila, D., & Cohen, H. (2007). Comorbidity of fibromyalgia and psychiatric disorders. *Current pain and headache reports*, *11*(5), 333-338.
- Cameron, J. E., Voth, J., Jaglal, S. B., Guilcher, S. J., Hawker, G., & Salbach, N. M. (2018). “In this together”: Social identification predicts health outcomes (via self-efficacy) in a chronic disease self-management program. *Social science & medicine*, *208*, 172-179.
- Carrillo-de-la-Peña, M. T., Triñanes, Y., González-Villar, A., Romero-Yuste, S., Gómez-Perretta, C., Arias, M., & Wolfe, F. (2015). Convergence between the 1990 and 2010 ACR diagnostic criteria and validation of the Spanish version of the Fibromyalgia Survey Questionnaire (FSQ). *Rheumatology international*, *35*(1), 141-151.

- Castro-Marrero, J., Zaragoza, M. C., López-Vílchez, I., Galmés, J. L., Domingo, J. C., Maurel, S., & Alegre-Martin, J. (2021). Effect of Melatonin Plus Zinc Supplementation on Fatigue Perception in Myalgic Encephalomyelitis/Chronic Fatigue Syndrome: A Randomized, Double-Blind, Placebo-Controlled Trial.
- Castro-Marrero, J., Sáez-Francàs, N., Santillo, D., & Alegre, J. (2017). Treatment and management of chronic fatigue syndrome/myalgic encephalomyelitis: all roads lead to Rome. *British journal of pharmacology*, *174*(5), 345-369.
- Cedraschi, C., Girard, E., Luthy, C., Kossovsky, M., Desmeules, J., & Allaz, A. F. (2013). Primary attributions in women suffering fibromyalgia emphasize the perception of a disruptive onset for a long-lasting pain problem. *Journal of psychosomatic research*, *74*(3), 265-269.
- Cohen, J. (1992). A power primer. *Psychological Bulletin*, *112*(1), 155–159.
- Collatz, A., Johnston, S. C., Staines, D. R., & Marshall-Gradisnik, S. M. (2016). A systematic review of drug therapies for chronic fatigue syndrome/myalgic encephalomyelitis. *Clinical therapeutics*, *38*(6), 1263-1271.
- Comrey, A. L., & Lee, H. B. (1992). *A first course in factor analysis* (2nd ed.). Hillsdale: Lawrence Erlbaum Associates Inc.

- Depue, R. A., & Morrone-Strupinsky, J. V. (2005). A neurobehavioral model of affiliative bonding: Implications for conceptualizing a human trait of affiliation. *Behavioral and Brain Sciences*, 28(3), 313-349.
- Field, A. P. (2009). Chapter 17: Exploratory factor analysis. In *Discovering statistics using SPSS* (3rd ed., pp. 627–685). SAGE Publications.
- Fischer, S., & Nater, U. M. (2014). Functional somatic syndromes: asking about exclusionary medical conditions results in decreased prevalence and overlap rates. *BMC public health*, 14(1), 1-7.
- Fitzcharles, M. A., Rampakakis, E., Ste-Marie, P. A., Sampalis, J. S., & Shir, Y. (2014). The association of socioeconomic status and symptom severity in persons with fibromyalgia. *The Journal of rheumatology*, 41(7), 1398-1404.
- Friedberg, F., & Jason, L. A. (2001). Chronic fatigue syndrome and fibromyalgia: clinical assessment and treatment. *Journal of clinical psychology*, 57(4), 433-455.
- Fukuda, K., Straus, S. E., Hickie, I., Sharpe, M. C., Dobbins, J. G., & Komaroff, A. (1994). The chronic fatigue syndrome: a comprehensive approach to its definition and study. *Annals of internal medicine*, 121(12), 953-959.
- Garcia-Campayo, J., Pascual, A., Alda, M., & Ramirez, M. T. G. (2007). Coping with fibromyalgia: usefulness of the Chronic Pain Coping Inventory-42. *Pain*, 132, S68-S76.



Geenen, R. Overgaauw, E., Papi, R., Reerds, Z.E.C., Schelwald, R., Pino, T., Anagnostaki, E., & Prent, Y. (2020). Bedreigende en kalmerende invloeden bij fibromyalgie [Threatening and soothing influences in fibromyalgia]. *FES Magazine*, 35(190), 26-29.

Geenen, R., & Jacobs, J. W. G. (2003). Fibromyalgie, chronisch vermoeidheidssyndroom en andere somatische klachtensyndromen. De reactie van 'splitters' op 'lumpers'. *Gedrag & Gezondheid*, 31, 113-121.

Geraghty, K., Hann, M., & Kurtev, S. (2019). Myalgic encephalomyelitis/chronic fatigue syndrome patients' reports of symptom changes following cognitive behavioural therapy, graded exercise therapy and pacing treatments: Analysis of a primary survey compared with secondary surveys. *Journal of health psychology*, 24(10), 1318-1333.

Giovengo, S. L., Russell, I. J., & Larson, A. A. (1999). Increased concentrations of nerve growth factor in cerebrospinal fluid of patients with fibromyalgia. *The Journal of rheumatology*, 26(7), 1564-1569.

Gilbert, P. (2014). The origins and nature of compassion focused therapy. *British Journal of Clinical Psychology*, 53(1), 6-41.

Goldenberg, D. L., Burckhardt, C., & Crofford, L. (2004). Management of fibromyalgia syndrome. *Jama*, 292(19), 2388-2395.

- Goldenberg, D. L. (2008). Multidisciplinary modalities in the treatment of fibromyalgia. *Journal of Clinical Psychiatry*, 69(Suppl 2), 30-4.
- Goldenberg, D. L. (2009). Diagnosis and differential diagnosis of fibromyalgia. *The American journal of medicine*, 122(12), S14-S21.
- Goldenberg, D. L., Simms, R. W., Geiger, A., & Komaroff, A. L. (1990). High frequency of fibromyalgia in patients with chronic fatigue seen in a primary care practice. *Arthritis & Rheumatism: Official Journal of the American College of Rheumatology*, 33(3), 381-387.
- Gupta, A., & Lang, A. E. (2009). Psychogenic movement disorders. *Current opinion in neurology*, 22(4), 430-436.
- Gur, A., & Oktayoglu, P. (2008). Central nervous system abnormalities in fibromyalgia and chronic fatigue syndrome: new concepts in treatment. *Current pharmaceutical design*, 14(13), 1274-1294.
- Harris, R. E., Clauw, D. J., Scott, D. J., McLean, S. A., Gracely, R. H., & Zubieta, J. K. (2007). Decreased central  $\mu$ -opioid receptor availability in fibromyalgia. *Journal of Neuroscience*, 27(37), 10000-10006.
- Hassett, A. L., & Gevirtz, R. N. (2009). Nonpharmacologic treatment for fibromyalgia: patient education, cognitive-behavioral therapy, relaxation techniques, and complementary and alternative medicine. *Rheumatic Disease Clinics*, 35(2), 393-407.

- Hazemeijer, I., & Rasker, J. J. (2003). Fibromyalgia and the therapeutic domain. A philosophical study on the origins of fibromyalgia in a specific social setting. *Rheumatology*, 42(4), 507-515.
- Heidari, F., Afshari, M., & Moosazadeh, M. (2017). Prevalence of fibromyalgia in general population and patients, a systematic review and meta-analysis. *Rheumatology international*, 37(9), 1527-1539.
- Hempel, S., Chambers, D., Bagnall, A. M., & Forbes, C. (2008). Risk factors for chronic fatigue syndrome/myalgic encephalomyelitis: a systematic scoping review of multiple predictor studies. *Psychological medicine*, 38(7), 915-926.
- Henningsen, P., Zipfel, S., & Herzog, W. (2007). Management of functional somatic syndromes. *The Lancet*, 369(9565), 946-955.
- Hudson, J. I., Goldenberg, D. L., Pope Jr, H. G., Keck Jr, P. E., & Schlesinger, L. (1992). Comorbidity of fibromyalgia with medical and psychiatric disorders. *The American journal of medicine*, 92(4), 363-367.
- Huibers, M. J., & Wessely, S. (2006). The act of diagnosis: pros and cons of labelling chronic fatigue syndrome. *Psychological medicine*, 36(7), 895-900.
- Huijnen, I. P., Verbunt, J. A., Meeus, M., & Smeets, R. J. (2015). Energy expenditure during functional daily life performances in patients with fibromyalgia. *PainPractice*, 15(8), 748-756.

- Jackson, E. (2002). An overview of chronic fatigue syndrome (Rehabilitation). *Nursing standard, 17*(13), 45-56.
- Jason, L. A., Richman, J. A., Rademaker, A. W., Jordan, K. M., Plioplys, A. V., Taylor, R. R., Taylor, R. R., McCready, W., Huang, C. F., & Plioplys, S. (1999). A community-based study of chronic fatigue syndrome. *Archives of internal medicine, 159*(18), 2129-2137.
- Jason, L. A., Roesner, N., Porter, N., Parenti, B., Mortensen, J., & Till, L. (2010). Provision of social support to individuals with chronic fatigue syndrome. *Journal of clinical psychology, 66*(3), 249-258.
- Jason, L. A., Taylor, R. R., & Kennedy, C. L. (2000). Chronic fatigue syndrome, fibromyalgia, and multiple chemical sensitivities in a community-based sample of persons with chronic fatigue syndrome-like symptoms. *Psychosomatic Medicine, 62*(5), 655-663.
- Jason, L., Witter, E., & Torres-Harding, S. (2003). Chronic fatigue syndrome, coping, optimism and social support. *Journal of Mental Health, 12*, 109 – 118. <https://doi.org/10.1080/09638230021000058346>.
- Johansson, R., Sjöberg, E., Sjögren, M., Johnsson, E., Carlbring, P., Andersson, T., Rousseau, A., & Andersson, G. (2012). Tailored vs. standardized internet-based cognitive behavior therapy for depression and comorbid symptoms: a randomized controlled trial. *PloS one, 7*(5), e36905.

- Joustra, M. L., Minovic, I., Janssens, K. A., Bakker, S. J., & Rosmalen, J. G. (2017). Vitamin and mineral status in chronic fatigue syndrome and fibromyalgia syndrome: A systematic review and meta-analysis. *PloS one*, *12*(4), e0176631.
- Joustra, M. L. (2019). *Similar but different: Implications for the one versus many functional somatic syndromes discussion*. [Doctoral dissertation, Rijksuniversiteit Groningen], <https://research.rug.nl/en/publications/similar-but-different-implications-for-the-one-versus-many-functi>.
- Kanaan, R. A., Lepine, J. P., & Wessely, S. C. (2007). The association or otherwise of the functional somatic syndromes. *Psychosomatic Medicine*, *69*(9), 855.
- Kim, D. Y., Lee, J. S., Park, S. Y., Kim, S. J., & Son, C. G. (2020). Systematic review of randomized controlled trials for chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME). *Journal of translational medicine*, *18*(1), 1-12.
- Kim, H. Y. (2013). Statistical notes for clinical researchers: assessing normal distribution (2) using skewness and kurtosis. *Restorative dentistry & endodontics*, *38*(1), 52.
- Kroenke, K., Spitzer, R. L., & Williams, J. B. (2002). The PHQ-15: validity of a new measure for evaluating the severity of somatic symptoms. *Psychosomatic medicine*, *64*(2), 258-266.
- Kwiatek, R. (2017). Treatment of fibromyalgia. *Australian prescriber*, *40*(5), 179.

- Lacourt, T., Houtveen, J., & van Doornen, L. (2013). "Functional somatic syndromes, one or many?": An answer by cluster analysis. *Journal of psychosomatic research*, 74(1), 6-11.
- Lao-Villadoniga, J. I., Poca-Dias, V., & Fernandez-Sola, J. (2008). Identification of differential genetic profiles in severe forms of fibromyalgia and chronic fatigue syndrome/myalgic encephalomyelitis: a population-based genetic association study. *Journal of Clinical Research*, 11, 1-24.
- LeGal, P., Rhéaume, A., & Mullen, J. (2019). The long-term effects of psychological demands on chronic fatigue. *Journal of nursing management*, 27(8), 1673-1681.
- Loades, M. E., Sheils, E. A., & Crawley, E. (2016). Treatment for paediatric chronic fatigue syndrome or myalgic encephalomyelitis (CFS/ME) and comorbid depression: a systematic review. *BMJ open*, 6(10), e012271.
- Macfarlane, G. J., Kronisch, C., Dean, L. E., Atzeni, F., Häuser, W., Fluß, E., Choy, E., Kosek, E., Amris K., Branco J., Dincer, F., Leino-Arjas, P., Longly, K., McCarthy, G. M., Makri, Perrot, S., Sarzi-Puttini, P., Taylor, A., & Jones, G. T. (2016). EULAR revised recommendations for the management of fibromyalgia. *Annals of the rheumatic diseases*, 76(2), 318-328.
- Maes, M., & Twisk, F. N. (2010). Chronic fatigue syndrome: Harvey and Wessely's (bio) psychosocial model versus a bio (psychosocial) model based on inflammatory and oxidative and nitrosative stress pathways. *BMC medicine*, 8(1), 1-13.

Mckay, P. G., Martin, C., Walker, H., & Fleming, M. (2019). Chronic fatigue syndrome (CFS)/Myalgic Encephalomyelitis (ME) and Fibromyalgia (FM): the foundation of a relationship. *British Journal Of Pain, 15*(1), 26-39.

<https://doi.org/10.1177/2049463719875164>.

Mckay, P. G., Walker, H., Martin, C. R., & Fleming, M. (2021). Exploratory study into the relationship between the symptoms of chronic fatigue syndrome (CFS)/myalgic encephalomyelitis (ME) and fibromyalgia (FM) using a quasiexperimental design. *BMJ open, 11*(2), e041947.

McManimen, S. L., McClellan, D., Stoothoff, J., & Jason, L. A. (2018). Effects of unsupportive social interactions, stigma, and symptoms on patients with myalgic encephalomyelitis and chronic fatigue syndrome. *Journal of community psychology, 46*(8), 959-971.

Mengshoel, A. M., Helland, I. B., Meeus, M., Castro-Marrero, J., Pheby, D., & Bolle Strand, E. (2020). Patients' experiences and effects of non-pharmacological treatment for myalgic encephalomyelitis/chronic fatigue syndrome—a scoping mixed methods review. *International journal of qualitative studies on health and well-being, 15*(1), 1764830.

Monden, R., Rosmalen, J. G., Wardenaar, K. J., & Creed, F. (2020). Predictors of new onsets of irritable bowel syndrome, chronic fatigue syndrome and fibromyalgia: the lifelines study. *Psychological medicine, 1-9*.

- Monro, J. A., & Puri, B. K. (2018). A molecular neurobiological approach to understanding the aetiology of chronic fatigue syndrome (myalgic encephalomyelitis or systemic exertion intolerance disease) with treatment implications. *Molecular neurobiology*, *55*(9), 7377-7388.
- Montoya, P., Larbig, W., Braun, C., Preissl, H., & Birbaumer, N. (2004). Influence of social support and emotional context on pain processing and magnetic brain responses in fibromyalgia. *Arthritis & Rheumatism*, *50*(12), 4035-4044.
- Natelson, B. H., Lin, J. M. S., Lange, G., Khan, S., Stegner, A., & Unger, E. R. (2019). The effect of comorbid medical and psychiatric diagnoses on chronic fatigue syndrome. *Annals of medicine*, *51*(7-8), 371-378.
- Natelson, B. H. (2019). Myalgic encephalomyelitis/chronic fatigue syndrome and fibromyalgia: definitions, similarities, and differences. *Clinical therapeutics*, *41*(4), 612-618.
- Noble, S., Bonner, C., Hersch, J., Jansen, J., McGeechan, K., & McCaffery, K. (2019). Could disease labelling have positive effects? An experimental study exploring the effect of the chronic fatigue syndrome label on intended social support. *Patient Education And Counseling*, *102*(3), 486-493.  
<https://doi.org/10.1016/j.pec.2018.10.011>.



- Nüesch, E., Häuser, W., Bernardy, K., Barth, J., & Jüni, P. (2013). Comparative efficacy of pharmacological and non-pharmacological interventions in fibromyalgia syndrome: network meta-analysis. *Annals of the rheumatic diseases*, 72(6), 955-962.
- Offenbächer, M., & Stucki, G. (2000). Physical therapy in the treatment of fibromyalgia. *Scandinavian journal of rheumatology*, 29(113), 78-85.
- Overgaauw, E. (2020). Comforting factors that influence the severity of fatigue and other somatic symptoms from the perspective of individuals with chronic fatigue syndrome: A concept mapping study (Master's thesis).  
<https://dspace.library.uu.nl/handle/1874/397594>.
- Pagliai, G., Giangrandi, I., Dinu, M., Sofi, F., & Colombini, B. (2020). Nutritional Interventions in the management of fibromyalgia syndrome. *Nutrients*, 12(9), 2525.
- Pinto, A. M., Geenen, R., Palavra, F., Lumley, M. A., Ablin, J. N., Amris, K., Branco, J., Buskila, D., Castelo-Branco, M., Crofford, L. J., Fitzcharles, M., Luís, M., Reis Marques, T., Rhudy, J. L., Uddin, L. Q., Castilho, P., Jacobs, J. W. G., da Silva, J. A. P. (2020). An Updated Overview of the Neurophysiological and Psychosocial Dimensions of Fibromyalgia – A Call for an Integrative Model. doi: 10.20944/preprints202007.0224.v1.
- Porges, S. W. (1995). Orienting in a defensive world: mammalian modifications of our evolutionary heritage: a polyvagal theory. *Psychophysiology*, 32, 301–318.

- Porges, S. W. (2007). The polyvagal perspective. *Biological Psychology*, 74(2), 116–143.
- Porges, S. W. (2009). The polyvagal theory: new insights into adaptive reactions of the autonomic nervous system. *Cleveland Clinic Journal of Medicine*, 76 (Suppl 2), S86.
- Prins, J. B., Bleijenberg, G., Bazelmans, E., Elving, L. D., de Boo, T. M., Severens, J. L., van der Wilt, G., Spinhoven, P., & Van der Meer, J. W. (2001). Cognitive behaviour therapy for chronic fatigue syndrome: a multicentre randomised controlled trial. *The Lancet*, 357(9259), 841-847.
- Prins, J. B., Bos, E., Huibers, M. J. H., Servaes, P., Van Der Werf, S. P., Van Der Meer, J. W. M., & Bleijenberg, G. (2004). Social support and the persistence of complaints in chronic fatigue syndrome. *Psychotherapy and psychosomatics*, 73(3), 174-182.
- Richardson, M., McEwan, K., Maratos, F., & Sheffield, D. (2016). Joy and calm: How an evolutionary functional model of affect regulation informs positive emotions in nature. *Evolutionary Psychological Science*, 2(4), 308-320.
- Rimes, K. A., & Chalder, T. (2005). Treatments for chronic fatigue syndrome. *Occupational medicine*, 55(1), 32-39.
- Roberts, D. (2018). Chronic fatigue syndrome and quality of life. *Patient related outcome measures*, 9, 253.

- Rusu, C., Gee, M. E., Lagacé, C., & Parlor, M. (2015). Chronic fatigue syndrome and fibromyalgia in Canada: prevalence and associations with six health status indicators. *Health Promotion and Chronic Disease Prevention in Canada: Research, Policy and Practice*, 35(1), 3.
- Sarchielli, P., Di Filippo, M., Nardi, K., & Calabresi, P. (2007a). Sensitization, glutamate, and the link between migraine and fibromyalgia. *Current pain and headache reports*, 11(5), 343-351.
- Sarchielli, P., Mancini, M. L., Floridi, A., Coppola, F., Rossi, C., Nardi, K., Acciaresi, M., Pini, L. A., & Calabresi, P. (2007b). Increased levels of neurotrophins are not specific for chronic migraine: evidence from primary fibromyalgia syndrome. *The Journal of Pain*, 8(9), 737-745.
- Schadt, E. E., & Björkegren, J. L. (2012). NEW: network-enabled wisdom in biology, medicine, and health care. *Science translational medicine*, 4(115), 1-8.
- Schmidt-Wilcke, T., & Clauw, D. J. (2011). Fibromyalgia: from pathophysiology to therapy. *Nature Reviews Rheumatology*, 7(9), 518.
- Schmidt-Wilcke, T., & Diers, M. (2017). New Insights into the Pathophysiology and Treatment of Fibromyalgia. *Biomedicines*, 5(4), 22.  
<https://doi.org/10.3390/biomedicines5020022>.

- Secker, D. L., & Brown, R. G. (2005). Cognitive behavioural therapy (CBT) for carers of patients with Parkinson's disease: a preliminary randomised controlled trial. *Journal of Neurology, Neurosurgery & Psychiatry*, 76(4), 491-497.
- Seligman, M. E., & Csikszentmihalyi, M. (2014). Positive psychology: An introduction. In *Flow and the foundations of positive psychology*, (Vol. 10, pp. 279-298). Dordrecht: Springer.
- Serrat, M., Almirall, M., Musté, M., Sanabria-Mazo, J. P., Feliu-Soler, A., Méndez-Ulrich, J. L., Luciano, J. V., & Sanz, A. (2020a). Effectiveness of a multicomponent treatment for fibromyalgia based on pain neuroscience education, exercise therapy, psychological support, and nature exposure (NAT-FM): A pragmatic randomized controlled trial. *Journal of clinical medicine*, 9(10), 3348.
- Serrat, M., Sanabria-Mazo, J. P., Almirall, M., Musté, M., Feliu-Soler, A., Méndez-Ulrich, J. L., Sanz, A., & Luciano, J. V. (2020b). Effectiveness of a Multicomponent Treatment based on Pain Neuroscience Education, Therapeutic Exercise, Cognitive Behavioural Therapy, and Mindfulness in Patients with Fibromyalgia (FIBROWALK study): A Randomized Controlled Trial, 1- 22. <https://doi.org/10.31234/osf.io/kj8sp>.
- Sharpe, M., Chalder, T., Johnson, A. L., Goldsmith, K. A., & White, P. D. (2017). Do more people recover from chronic fatigue syndrome with cognitive behaviour therapy or graded exercise therapy than with other treatments?. *Fatigue: Biomedicine, Health & Behavior*, 5(1), 57-61.

Shepherd C (1999) *Living with ME*. London, Vermilion.

Stewart, J. A., Mailler-Burch, S., Müller, D., Studer, M., von Känel, R., grosse Holtforth, M., ... & Egloff, N. (2019). Rethinking the criteria for fibromyalgia in 2019: the ABC indicators. *Journal of pain research*, *12*, 2115.

Sullivan, Å., Nord, C. E., & Evengård, B. (2009). Effect of supplement with lactic-acid producing bacteria on fatigue and physical activity in patients with chronic fatigue syndrome. *Nutrition journal*, *8*(1), 1-6.

Thieme, K., Mathys, M., & Turk, D. C. (2017). Evidenced-based guidelines on the treatment of fibromyalgia patients: Are they consistent and if not, why not? Have effective psychological treatments been overlooked?. *The Journal of Pain*, *18*(7), 747-756.

Torpy, D. J., Bachmann, A. W., Grice, J. E., Fitzgerald, S. P., Phillips, P. J., Whitworth, J. A., & Jackson, R. V. (2001). Familial corticosteroid-binding globulin deficiency due to a novel null mutation: association with fatigue and relative hypotension. *The Journal of Clinical Endocrinology & Metabolism*, *86*(8), 3692-3700.

Twisk, F. N., & Maes, M. (2009). A review on cognitive behavioral therapy (CBT) and graded exercise therapy (GET) in myalgic encephalomyelitis (ME)/chronic fatigue syndrome (CFS): CBT/GET is not only ineffective and not evidence-based, but also potentially harmful for many patients with ME/CFS. *Neuro endocrinology letters*, *30*(3), 284-299.

- Ursin, H. (1997). Sensitization, somatization, and subjective health complaints. *International journal of behavioral medicine*, 4(2), 105-116.
- Ward, C. D., & Horrocks, M. (2015). The self and others in CFS/ME: reinterpreting research evidence. In *Meanings of ME: Interpersonal and Social Dimensions of Chronic Fatigue*, Palgrave Macmillan, London, 106-128.
- Werbach, M. R. (2000). Nutritional strategies for treating chronic fatigue syndrome. *Alternative Medicine Review*, 5(2), 93-108.
- Wessely, S., Nimnuan, C., & Sharpe, M. (1999). Functional somatic syndromes: one or many?. *The Lancet*, 354(9182), 936-939.
- White, K. P., Speechley, M., Harth, M., & Ostbye, T. (1999). The London Fibromyalgia Epidemiology Study: the prevalence of fibromyalgia syndrome in London, Ontario. *The Journal of rheumatology*, 26(7), 1570-1576.
- White, K., Speechley, M., Harth, M., & Ostbye, T. (2000). Co-existence of chronic fatigue syndrome with fibromyalgia syndrome in the general population: a controlled study. *Scandinavian Journal of Rheumatology*, 29(1), 44-51.

- White, P. D., Goldsmith, K. A., Johnson, A. L., Potts, L., Walwyn, R., DeCesare, J. C., Baber, H. L., Burgess, M., Clark, L. V., Cox, D. L., Bavinton, J., Angus, B. J., Murphy, G., Murphy, M., O'Dowd, H., Wilks, D., McCrone, & P., Sharpe, M., on behalf of the PACE Trial Management Group. (2011). Comparison of adaptive pacing therapy, cognitive behaviour therapy, graded exercise therapy, and specialist medical care for chronic fatigue syndrome (PACE): a randomised trial. *The Lancet*, 377(9768), 823-836. [https://doi.org/10.1016/s0140-6736\(11\)60096-2](https://doi.org/10.1016/s0140-6736(11)60096-2).
- Whitehead, L. C. (2006). Quest, chaos and restitution: Living with chronic fatigue syndrome/myalgic encephalomyelitis. *Social science & medicine*, 62(9), 2236-2245.
- Whiting, P., Bagnall, A. M., Sowden, A. J., Cornell, J. E., Mulrow, C. D., & Ramírez, G. (2001). Interventions for the treatment and management of chronic fatigue syndrome: a systematic review. *Jama*, 286(11), 1360-1368.
- Wilshire, C., Kindlon, T., Matthees, A., & McGrath, S. (2017). Can patients with chronic fatigue syndrome really recover after graded exercise or cognitive behavioural therapy? A critical commentary and preliminary re-analysis of the PACE trial. *Fatigue: Biomedicine, Health & Behavior*, 5(1), 43-56.
- World Health Organization. (2016). R53.82 *International statistical classification of diseases and related health problems* (10<sup>th</sup> ed.). <https://www.icd10data.com/ICD10CM/Codes/R00-R99/R50-R69/R53-/R53.82>.

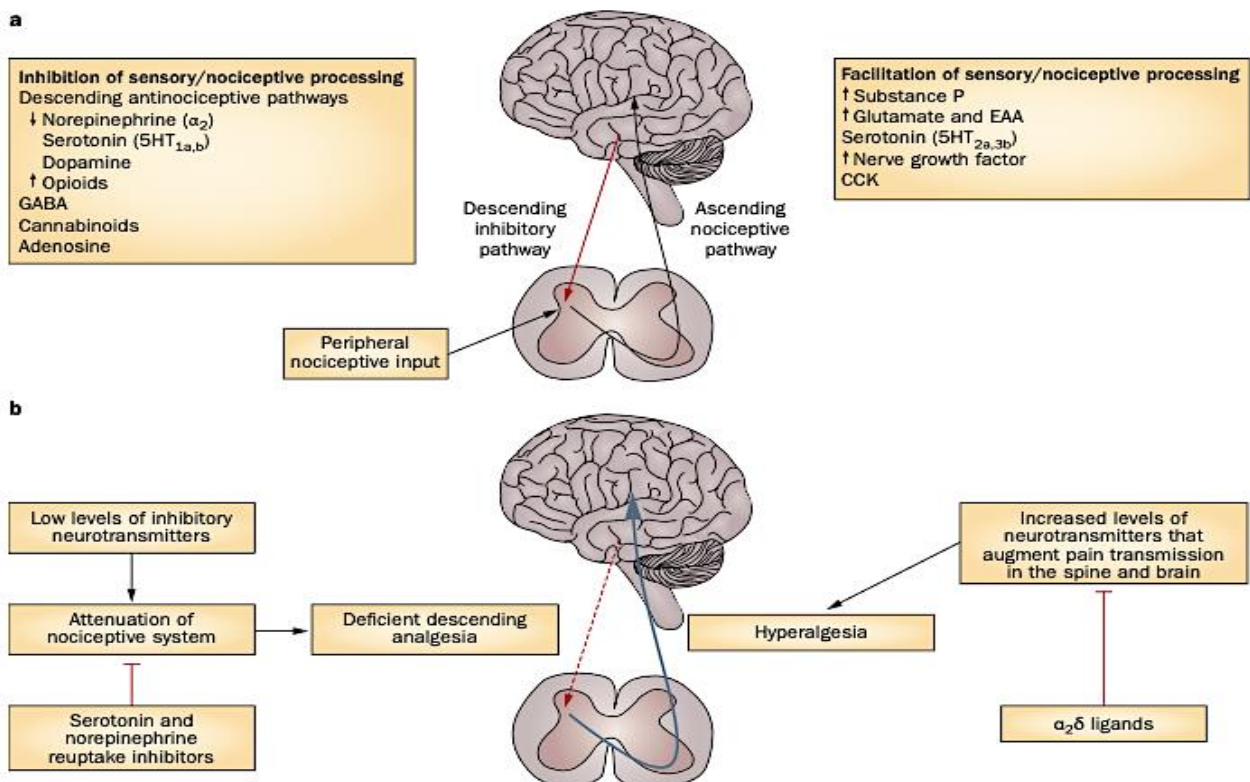
- Wolfe, F., Smythe, H. A., Yunus, M. B., Bennett, R. M., Bombardier, C., Goldenberg, D. L., Tugwell, P., Campbell, S. M., Abeles, M., Clark, P., Fam, A. G., Farber, S. J., Fiechtner, J. J., Franklin, C. M., Gatter, R. A., Hamaty, D., Lessard, J., Lichtbroun A. S., Masi, A. T., ... & Sheon, R. P. (1990). The American College of Rheumatology 1990 criteria for the classification of fibromyalgia. *Arthritis & Rheumatism: Official Journal of the American College of Rheumatology*, 33(2), 160-172.
- Vander Zee, K. I., Sanderman, R., Heyink, J. W., & de Haes, H. (1996). Psychometric qualities of the RAND 36-Item Health Survey 1.0: a multidimensional measure of general health status. *International journal of behavioral medicine*, 3(2), 104-122.
- Vink, M., & Vink-Niese, A. (2018). Graded exercise therapy for myalgic encephalomyelitis/chronic fatigue syndrome is not effective and unsafe. Re-analysis of a Cochrane review. *Health psychology open*, 5(2), 2055102918805187.



## Appendix

### Appendix A

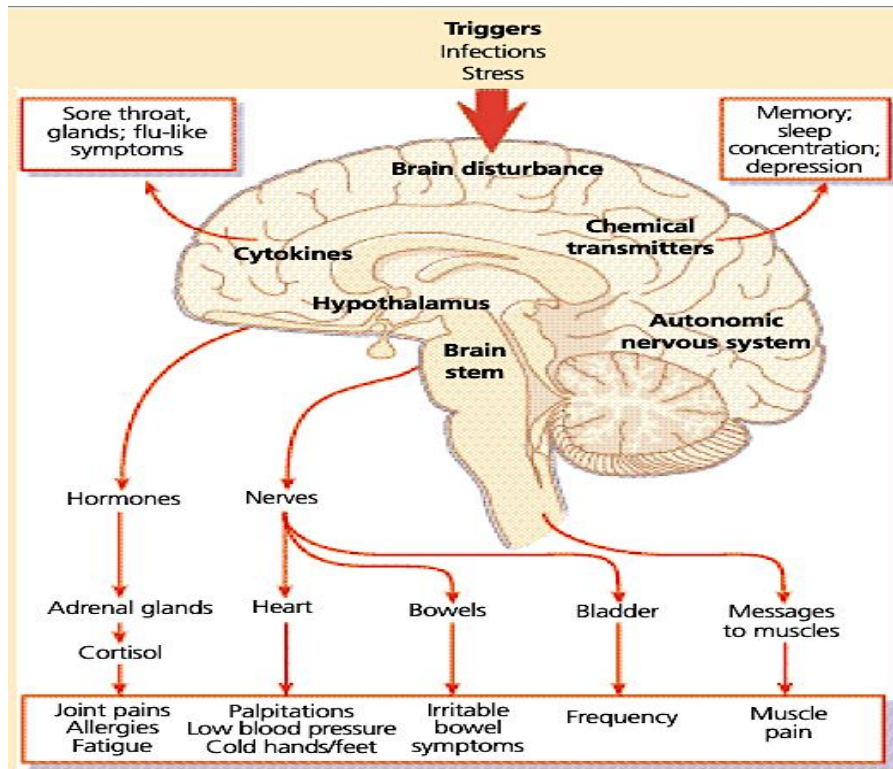
Appendix A looks at the differences in proposed pathophysiology of both syndromes and the quantitative differences reported in the literature. While there are many similarities between FM and CFS, there are also some qualitative (e.g., self-reported fatigue) and quantitative differences (e.g., spinal fluid substance P, reported to be elevated in FM patients, but unremarkable in CFS patients) that must not be ignored (Abbi & Natelson, 2019). The current study generally looked at the qualitative differences, but is also important mention the quantitative differences.



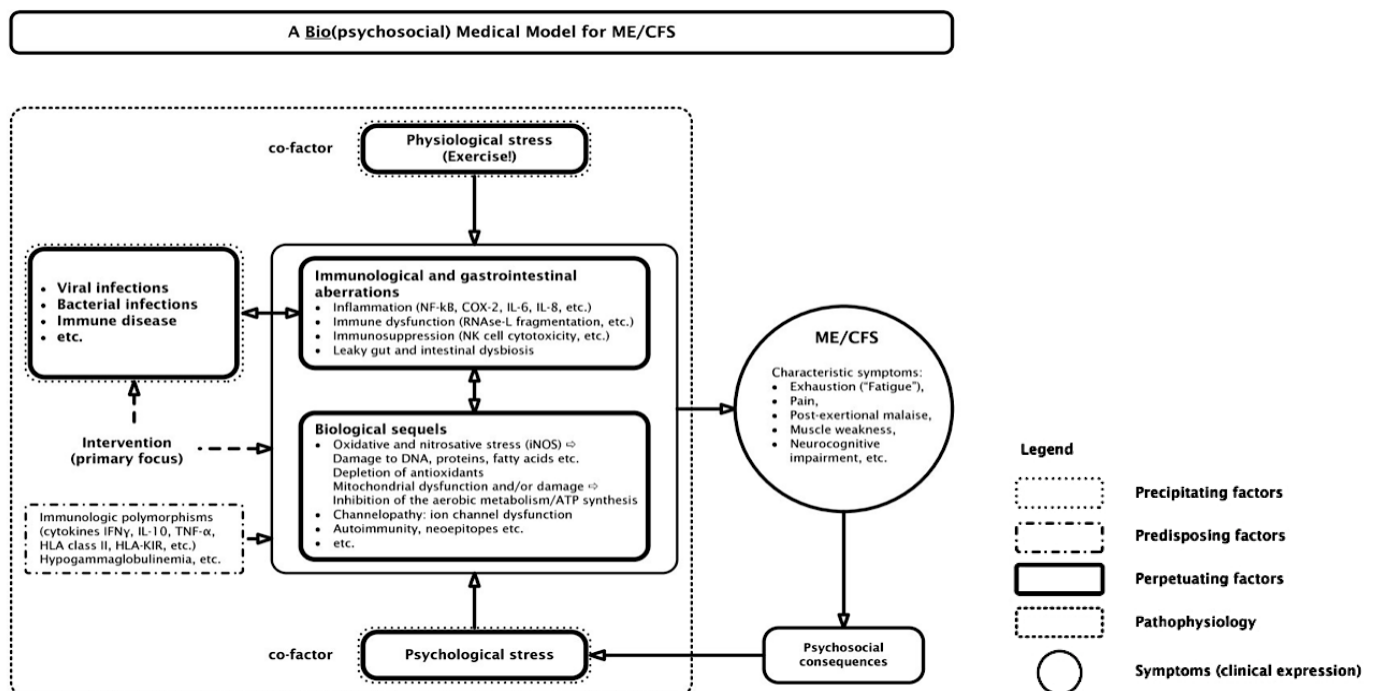
**Figure.** Diagram of the proposed pathophysiology of fibromyalgia: adapted from Schmidt-Wilcke & Clauw (2011). (Abbreviations: 5-HT, 5-hydroxytryptamine; CCK, cholecystokinin; CNS, central nervous system; DNIC, diffuse noxious inhibitory control; EAA, excitatory amino acids; GABA,  $\gamma$ -aminobutyric acid).

*Note: This figure depicts the pain processing pathways involved in chronic pain and FM: (a) These CNS neurotransmitters facilitate or inhibit sensory/pain transmission – the arrows specify whether these neurotransmitters are decreased or increased in FM patients. (b) FM patients exhibit imbalances across several neurotransmitters that control the level and frequency of pain experienced. This may result in augmented pain processing. For example, low levels of inhibitory neurotransmitters can lead to diminished antinociceptive system function (which acts to block the detection of painful stimuli through sensory neurons). Serotonin–norepinephrine reuptake inhibitors can have the opposite effect, increasing antinociceptive activity.*

The pathophysiology of FM appears to involve multiple underlying mechanisms. There are several prevalent risk factors which are linked to the development of FM, these include; stress, genetic polymorphisms and familial predisposition (Schmidt-Wilcke & Clauw, 2011). Glutamate (an excitatory neurotransmitter) also appears to play a role in FM, as studies have revealed elevated cerebrospinal fluid – glutamate levels and altered glutamate levels in the insula of FM patients (Sarchielli et al., 2007b; Harris et al., 2007). Nerve growth factor is another example of a neurotransmitter which has been observed in higher concentrations (in the cerebrospinal fluid) of FM patients (Giovengo, Russell, & Larson, 1999; Sarchielli et al., 2007a; Sarchielli et al., 2007b). These are prime examples of pain augmenting neurotransmitters which are present in higher concentrations within FM and research indicates that this is likely associated with increased activity in ascending pain transmission pathways (Schmidt-Wilcke & Clauw, 2011).



**Figure.** Suggested mechanism for symptoms of chronic fatigue syndrome: Obtained from Jackson (2002), adapted from Shepherd (1999).



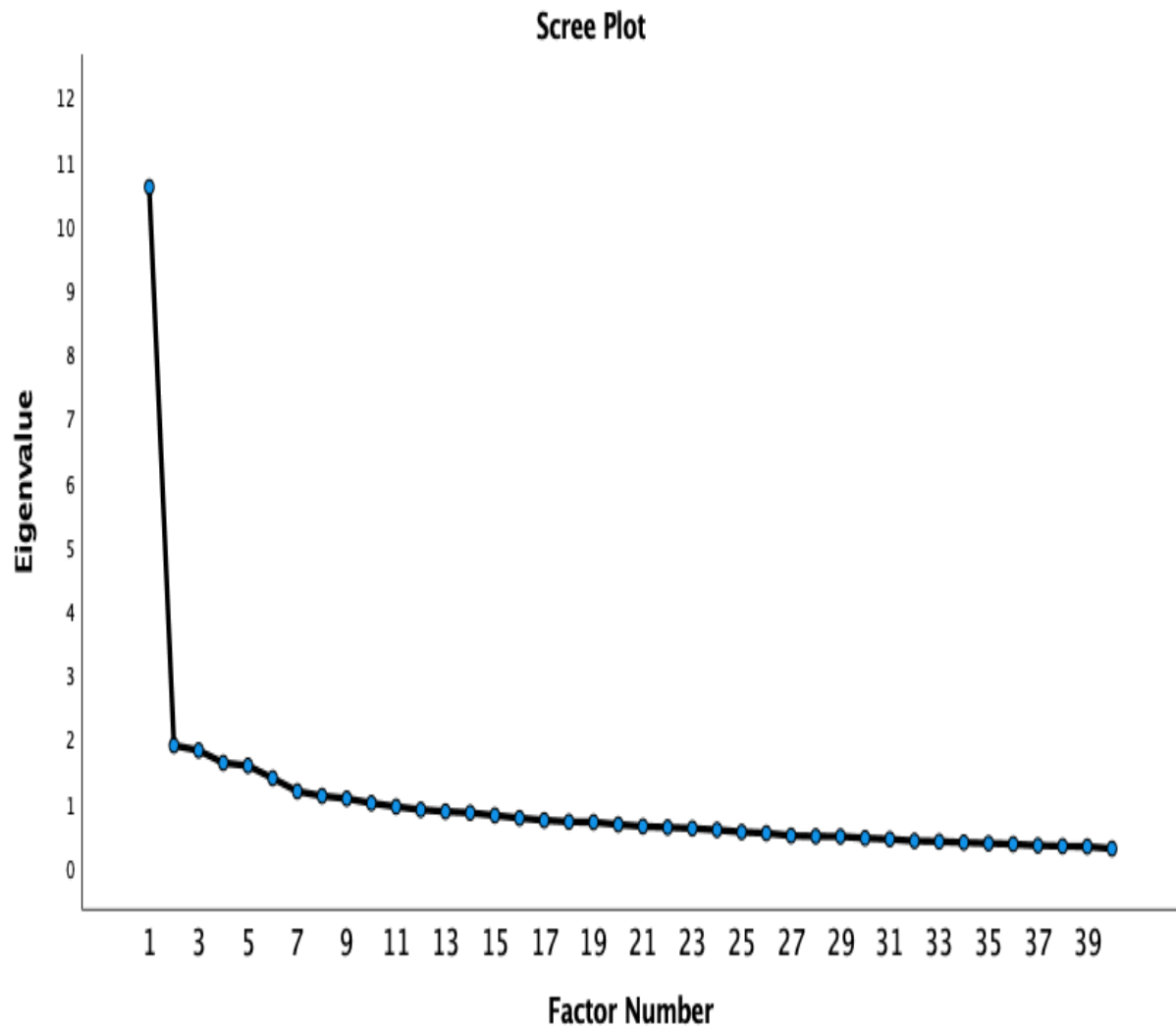
**Figure.** Proposed (bio)psychosocial model for chronic fatigue syndrome: Obtained from Maes & Twisk (2010).

*Note: The inflammatory and oxidative and nitrosative (IO&NS) pathophysiology of myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS). COX-2 = cyclo-oxygenase 2; iNOS = inducible nitric oxide synthase; PUFA = polyunsaturated fatty acids; NFκB = nuclear factor κB.*

The pathophysiology of CFS remains unclear, however some research postulates that the presence of pathophysiological abnormalities within CFS which indicate that the syndrome is heterogeneous. One particular family study on CFS discovered a genetic mutation that disrupts globulin production, which is a protein that is involved in transporting of cortisol in the blood (Torpy et al., 2001). Interesting research by Dantzer and colleagues (2014) posits that inflammation plays a critical role in the development and persistence of fatigue in patients suffering from physical illness. Research indicates that infectious, immunological, neuroendocrine, sleep, and psychiatric mechanisms are involved in predisposing, precipitating and perpetuating CFS (Afari & Buchwald, 2003; Cortes-Rivera et al., 2019)

Finally, Abbi and Natelson (2019) acknowledged the overlapping symptomatology of CFS and FM in their systematic review, and despite several shortcomings within their review they maintain that it still tentatively points to differing pathophysiology for CFS and FM. They mainly highlighted quantitative differences between the FSSs. In particular they highlighted differences in the genetic profiles (e.g., single-nucleotide polymorphisms) (Lao-Villadoniga et al., 2008), mechanisms of autonomic dysfunction (e.g., blood pressure), and biomarkers (e.g., fatty acid composition and in the membrane fluidity of the muscle) related to CFS and FM. While there is much research yet to be completed on the differences between these FSSs, there are tentative indications that FM and CFS should remain as separate diagnoses.

## Appendix B



**Figure.** *Scree plot of factor eigenvalues*

## Appendix C

**Table.** *Skewness, Kurtosis and Z values for each syndrome group*

Factor	Fibromyalgia				Chronic Fatigue				Both (Comorbid)			
	Skewness	Kurtosis	Z value Skewness	Z value Kurtosis	Skewness	Kurtosis	Z value Skewness	Z value Kurtosis	Skewness	Kurtosis	Z value Skewness	Z value Kurtosis
<b>Comfort</b>	-.74	.08	-5.70	.27	-.37	-.13	-1.08	-.20	-.82	-.39	-3.08	-.74
<b>Mindfulness</b>	.13	-1.13	1.01	-4.38	.21	-.79	.60	-1.18	.18	-.89	.69	-1.68
<b>Social activity</b>	-.55	-.48	-4.28	-1.87	-.49	-.79	-1.43	-1.18	-.60	-.21	-2.27	-.40
<b>Balance between freedom and structure</b>	-.89	.25	-.87	.95	-1.72	4.59	-5.01	6.81	-1.09	.73	-4.09	1.39
<b>Physical functioning</b>	-.09	-.54	-.71	-2.1	.37	-.63	1.07	-.93	.42	-.48	1.57	-.91
<b>Role limitation due to physical health</b>	1.39	.66	10.77	2.56	3.59	13.92	10.47	20.65	2.89	8.31	10.85	15.8
<b>Role limitation due to emotional problems</b>	-.02	-1.79	-.14	-6.98	-.3	-1.87	-.88	-2.77	.57	-1.47	2.15	-2.79
<b>Energy/fatigue</b>	.29	-.13	2.27	-.51	.68	.75	1.97	1.12	.35	-.57	1.32	-1.08
<b>Social functioning</b>	-.23	-.05	1.81	-.19	.59	-.46	1.71	-.68	.38	-.58	1.44	-1.10
<b>Emotional wellbeing</b>	-.46	-.006	-3.53	-.02	-.12	-.88	-.034	-1.31	-.52	.13	-1.96	.25
<b>Pain</b>	-.15	-.29	-1.14	-1.14	.05	-.33	.15	-.49	-.05	-.8	-.17	-1.52
<b>General health</b>	.42	-.45	3.26	-1.77	.76	1.14	2.22	1.69	.19	-.22	.73	-.43
<b>Health change</b>	.48	-.18	3.74	-.72	-.08	-.99	-.023	-1.47	.50	.24	1.89	.45
<b>Total RAND-36</b>	.23	-.27	1.77	-1.05	.85	1.35	2.49	2.00	.53	-.23	1.99	-0.44
<b>Total PHQ-15</b>	.31	-.29	2.40	-1.12	-.27	-.85	-.8	-1.26	-.12	.08	-.05	.14

## Appendix D

**Table.** Descriptive statistics for H1 and H2 soother items

Measure	FM ( <i>n</i> = 357)		CFS ( <i>n</i> = 48)		Comorbid ( <i>n</i> = 82)		<i>F</i> (2, 484)	$\eta_p^2$
	M	SD	M	SD	M	SD		
<b>Balance between activities</b>	3.39	.82	3.65	.79	3.34	.89	2.28	.009
<b>Consistency and structure</b>	3.10	.93	3.21	.87	2.98	1.03	1.01	.004
<b>Remain within my limits</b>	3.20	.91	3.46	.71	3.37	.87	2.49	.01
<b>Being surrounded by family and friends</b>	3.17	.90	3.12	1.08	3.23	.99	.22	.001
<b>Doing a fun thing with family or friends</b>	2.88	.91	2.63	1.02	2.78	1.02	1.78	.007