

Perceived threatening influences on severity of somatic
symptoms in people with fibromyalgia: a concept
mapping study

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Master Thesis

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Date: May 24th 2021

Wordcount from *abstract* up to
and including *discussion*: 5628

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Abstract

Objective: It has been assumed that due to overactivation of the sensitive brain in individuals with fibromyalgia (FM), the threat detection system perceives threats in a more extreme way, leading to an increase in symptoms such as widespread musculoskeletal pain, fatigue and disturbed mood. In order to get a better idea of the perceived threatening influences on somatic symptoms in people with FM and to be able to adjust therapy accordingly, this study aims to obtain a hierarchical overview from the point of view of the individual suffering from FM.

Methods: 701 individuals with somatic symptoms participated in an online survey providing 40 threatening influences. These influences were sorted by 73 other individuals with somatic symptoms using a card sorting task based on similarity of meaning. Severity of somatic symptoms was assessed with the Patient Health Questionnaire 15 (PHQ-15). Hierarchical cluster analysis (squared Euclidean distances, Ward's method) was used to obtain a hierarchical overview of threatening influences. Differences in perceived importance of threats per category were checked using a repeated measure analysis of variance. The association between the threats of each category and symptom severity were examined using three linear regression analyses, correlating the total PHQ-15 with the scores of each category.

Results: The hierarchical cluster analysis outcomes showed three categories of threatening influences on somatic symptoms for people with FM: “symptoms and consequences of disease”, “(social) demands” and “negative thoughts and emotions”. The categories did not differ from each other in terms of relative importance. Nor were the categories related to somatic symptom severity.

Conclusion: This study found a comprehensive hierarchical structure for threats that influence somatic symptoms according to people with FM. This structure can be used to guide research and it can lead to tailor-made treatments, for example by means of a screening instrument that can provide direction for treatment based on perceived threats within the categories. This could lead to higher effectiveness of treatment.

1. Introduction

Fibromyalgia (FM) is a disease characterized by widespread musculoskeletal pain, tender points, fatigue, sleeping problems, and disturbed mood (van Middendorp et al., 2008). It is a chronic and disabling condition that affects about 3% of the adult population (Branco et al., 2010). The putative pathogenetic mechanism in FM is proposed to be rooted in persistent activation of the brain's salience network, over activating the threat-detection system and a lack of activation in the soothing system (Pinto et al., 2020a). This sensitivity of brain networks is assumed to be influenced by psychological factors, various threats can trigger the sensitive brain to respond with pain and other physical symptoms (Pinto et al., 2020a). To gain a better understanding of FM it is important that these systems are identified. Pinto et al. (2020a) suggest that the model of affect regulation by Gilbert (2016) provides guidance in this regard.

Gilbert's model of affect regulation states that there is an interaction between three emotion regulation systems, one for soothers, one for drives and one for threats (Gilbert, 2016). The *contentment, soothing focused system* (soothing system) enables a state of contentment, peacefulness, and openness. When enabled this system makes for an individual to be satisfied and experiencing a sense of well-being (Gilbert, 2016). The *drive, seeking and acquisition focused system* (driving system) enables the individual to pay attention and respond to stimuli indicating resources (Gilbert, 2016). The driving system provides motivation and makes it possible to pursue goals. The *threat and self-protection focused system* (threat detection system) enables the detection of and response to threats (Gilbert, 2016). Emotions such as anger, anxiety and disgust are associated with this system. When there is more knowledge about the specific threats that influence these negative affects and increase symptoms in people with FM, this knowledge could be used in the development of educational materials and therapies.

Examples of threats that are known to worsen disease activity in individuals with FM are social threats, non-social threats, and internal threats (Pinto et al., 2020a). An example of a social threat is social rejection. Social rejection occurs when an individual experiences exclusion from a social relationship or social interaction (Williams, Forgas & von Hippel, 2015). Perceived social rejection shares neural circuitry with physical pain, activating the threat detection system (Eisenberger, 2012; Perini et al., 2018). An instance of non-social threats are sensory inputs (Pinto et al., 2020a). Individuals with FM often experience multisensory hypersensitivity (Rivat et al., 2010, in Pinto et al., 2020a). Brain imaging studies

have shown that individuals with FM show brain activation in areas of the brain that modulate responses to pain for nonnoxious sensory stimuli, indicating distortion in sensory processing (Cook et al., 2004). Across multiple inputs such as taste, smell, auditory and tactile higher sensitivity has been found (Wilbarger & Cook, 2011). Examples of internal threats are physical pain and a negative thought style (Pinto et al., 2020a). People with FM display features of increased sensitivity to painful stimuli (Branco et al., 2010) making pain a catalyst for increase in disease behavior. A negative thought style, or “negativity filter” multiplies the perception of a threat as being more threatening increasing symptoms in people with FM (Rivat et al., 2010). Thus, multiple threats of FM are known. Nevertheless, a structured and encompassing overview of threats in FM is missing.

Such an overview could be used as a screening instrument or to develop or evaluate therapy in FM. In a meta-analysis by van Koulil and colleagues (2007) multiple Cognitive Behavioral Therapeutic (CBT) interventions have been compared such as cognitive restructuring, pain-coping skills, problem-solving techniques, goal setting, increasing activity levels, activity pacing, stress management and educational and relaxation components. Overall, the effects of these interventions in patients with FM has not proven to be effective in fully alleviating symptoms (Macfarlane et al., 2017). Pinto et al. (2020b), propose the usage of interventions that reinforce soothing perception, such as optimism, acceptance, compassion, mindfulness, valued living, social support, and connectedness (e.g., Mindfulness-based Stress Reduction, Acceptance and Commitment Therapy). These contextual cognitive-behavioral therapies (Hayes et al., 2011) show more promise by emphasizing to develop soothing personal skills, such as acceptance, embracing and non-judging (Pinto et al., 2020a). Threats however can knock out positive emotions, since pursuing positive resources in the face of high threat is maladaptive (Gilbert, 2016). In order to prevent threats from obstructing these interventions, it is important to identify these threats. This study aims to contribute to this knowledge by providing a broad overview of possible threats, measuring differences in terms of importance, and examining the relationship between threats and the severity of physical symptoms.

FM is a disease with many negative consequences that limit people in their daily life with currently few effective treatments. An overactive threats system contributes to the maintenance of symptoms such as fatigue, sleep problems, disturbed mood, and pain. Identifying threats and discovering underlying structures makes it possible to adapt and make therapies more effective. With this goal in mind, the following research questions were formulated.

The following research questions were examined:

1. What threats do people with fibromyalgia report and how are they structured in clusters and overarching categories?
2. What differences do people with fibromyalgia report in terms of perceived importance of these threat categories?
3. How do the observed categories relate to the severity of the somatic symptoms?

Due to the nature of this research being exploratory, trying to make fundamental findings in the research field, a hypothesis was only formulated for the first research question. This was as follows: It is expected that participants mention threats belonging to the categories of social threats (e.g., social rejection), non-social threats (e.g., sensory inputs), and internal threats (e.g., physical pain, negative thought style).

2. Methods

2.1 Procedure and design

A research group was composed of six master's students clinical psychology from Utrecht University and prof. dr. Geenen. The study followed a concept mapping technique which makes it possible to interpret qualitative information using systematic analysis. The first step of this technique consists of an online survey with the aim of obtaining encompassing sets of threats, soothers and drives from people with chronic somatic symptoms. The treats reported in this survey were the primary concern of this study. In the second step, statements were chosen based on their representation using four criteria: definition, applicability, similarity, and specificity. In the third step a different group of participants sorted the previously mentioned statements on similarity in meaning and they had to categorize them to what extent they were affected by the statements. In the fourth step a hierarchical cluster analysis was used in order to obtain a structured representation of the card sorting task. The exact contents of the steps are further explained below. The studies (step 1 and 3) were approved by the ethical committee of the faculty of Social and Behavioral Sciences at Utrecht University (19-219). Informed consent was given by the participant prior to enrollment.

2.2 Participants

Participants had to be at least 18 years old and have chronic somatic complaints. Participants were recruited from different websites such as various patient associations (regarding Irritable Bowel Syndrome, Chronic Fatigue Syndrome, Fibromyalgia) (Fest, 2019; Nouwen, 2019; Prikkelbare Darm Syndroom Belangenorganisatie, 2019) and with the help of Facebook (Reerds, 2019).

A total of 701 participants filled out the online survey. Nationalities for these participants were notably Dutch ($n=407$), Belgium ($n=51$), Brazilian ($n=115$), Cyprian ($n=11$), Greek ($n=37$), Peruvian ($n=21$), and Portuguese ($n=30$). Of the participants forty-six were male and 655 were female, with a mean age of 45.5 ($SD = 11.93$).

The participants that participated in the card sorting task ($n=73$) were from the Netherlands. Forty-nine participants were diagnosed with FM. Four were male, sixty-nine were female with a mean age of 49.2 ($SD = 12.11$). These participants were also acquired using earlier mentioned websites, when given informed consent they would get the task by mail service which they could send back after finishing.

2.3 Measurements

The online survey first assessed demographic data, such as: country of residence, nationality, gender, age, relationship status, years of education from age six.

The second array of questions were regarding disease(s): whether and which rheumatic disease(s) the participant had, whether and which other disease(s) the participant had, and who diagnosed the disease(s).

The third array of questions were from the Patient Health Questionnaire 15 (PHQ-15; Kroenke, Spitzer, & Williams, 2002). This questionnaire assessed the severity of somatic symptoms using a three-point Likert scale ascending from zero to two on fifteen different symptoms during the past four weeks. Zero meaning “not bothered at all”, one meaning “bothered a little”, and two meaning “bothered a lot”. Score on the PHQ-15 is calculated by adding up the scores on each individual item. A combined score between zero and four is categorized as minimal severity of somatic symptoms, a score between five and nine as low severity of symptoms, between ten and fourteen as medium and fifteen and above as high. The internal consistency of the PHQ-15 is good with a Cronbach’s alpha of .80 (Kroenke et al., 2002).

The fourth and last array of questions in the online survey were regarding the threats,

soothers and drives. Each participant answered the three of questions. First, “This question is about ‘threats’ that may worsen your pain, fatigue or other physical symptoms. ‘Threats’ create experiences of danger, harm, damage or unsafety. Could you mention as many as possible ‘threats’ that may worsen your pain, fatigue or other physical symptoms?”. Second, “This question is about ‘comforting influences’ that may alleviate your pain, fatigue or other physical symptoms. They create feelings of calmness, well-being, safety or social connectedness. Could you mention as many as possible ‘comforting influences’ that may alleviate your pain, fatigue or other physical symptoms?” and “This question is about ‘drives’ that may alleviate your pain, fatigue or other physical symptoms. Drives are urges, ambitions and motivations that stimulate the person to pursue a specific activity or reach a goal. Third, Could you mention as many as possible ‘drives’ that may alleviate your pain, fatigue or other physical symptoms?”. After submitting their answers, the participants were thanked for participating.

2.4 Data collection and analyses

- Step 1: online questionnaire

The first part of the study was conducted anonymously with the use of LimeSurvey Version 3.20.0+191112 (LimeSurvey Project Team / Carsten Schmitz, 2020). Participants were asked to fill out the survey in either Dutch, English, Greek, Portuguese, Brazilian-Portuguese, or Spanish, starting with the informed consent, demographics, illness questions and PHQ-15. The last part of the survey focused on the main objective of threats, soothers, and drives.

- Step 2: Selection of threats, soothers and drives

All the threats, soothers and drives from the online questionnaire were translated into English and bundled in a spreadsheet. The researchers categorized the responses pairwise. For instance, “poor sleep”, “insomnia” and “little sleep” were categorized within the “sleep” category. A first selection of the responses was made into statements based on four criteria: definition, applicability, similarity, and specificity. The definition of a threat was: “something that creates experiences of danger, harm, damage or unsafety”. The criteria of applicability meant that the statement had to be applicable to the entire group, for example threats which only concerned females, or the elderly were discarded. Responses were checked on similarity, meaning that similar statements were combined and a single statement involving multiple threats was split into single ones. Another criterium for the statements was to neither be too specific or ambiguous and stick to the verbalization of the participant as close as possible,

with a sidenote that it should be understandable for common people. The statements were debated in project group meetings until consensus was reached. The last part of the selection procedure involved the research group members executing the card sorting task. Statements which were all put together on the same pile which overlapped in content were considered to be of no differentiative value, and therefore discarded. During the whole selection procedure, the threat-statements were reduced from 914 threats to 40 threats.

- Step 3: Card sorting task

Using again LimeSurvey (LimeSurvey Project Team / Carsten Schmitz, 2020) were recruited. Participants received an instruction booklet, questionnaire and the using mail service at their home. The questionnaire contained questions concerning demographics, illness history and severity of symptoms (PHQ-15; Kroenke et al., 2002).

The task consisted of two parts of card sorting for each of the facets (threats, soothers, drives). For the purpose of this study and convenience of explanation, it was decided only the threats are exemplified. However, note that the task was conducted thrice by the participants, once for threats, once for soothers and once for drives. Participant received an envelope containing 40 cards with statements concerning threats that influence their somatic symptoms. In the first card sorting task these cards had to be piled together with a minimum of 4 and a maximum of 12 piles according to similarity of meaning. Each pile could consist of minimally 2 and maximally 15 cards. After the sorting of the cards, participants had to label each of their piles for the similarity to which they categorized them.

In the second part of the task, the cards had to be sorted by to what extent the statement threatened to worsen their somatic symptoms. Cards had to be numbered from 1 (least threatening) to 5 (most threatening). Each pile had to contain 8 cards, and 5 piles had to be made not reusing cards and using all cards.

For both card sorting tasks, the participants wrote down their results and were asked to send these back within 10 days after finishing the task.

- Step 4: Analysis

Three analyses were conducted: hierarchical cluster analysis, repeated measures analysis and linear regression. For data analysis the 24th edition of IBM SPSS was used. Values were deleted for participants who had not performed the task as instructed. For the first sorting task, if a participant had forgotten to pile one or multiple cards, single cards were then put on

separate piles. This was also done if one card was piled on multiple piles.

The hierarchical cluster analysis was used to classify the statements that were individually sorted by the participants during the first card sorting task. In the cluster analysis, the cells of the input matrix of outcomes comprised the number of times that two outcomes were not sorted in the same pile. Squared Euclidean distances were computed between each pair of outcomes and Ward's method was used to derive the hierarchical structure of the threat-statements visualized in a dendrogram. Using the dendrogram a top-down interpretation was utilized, starting with two clusters adding a cluster until new content was no longer produced. Comparisons were made on the basis of content of adding or reducing the number of clusters until a final number of clusters was set. Per cluster a Cronbach's Alpha's was computed for reliability. This information was used to decide on the number of categories.

A repeated measures analysis was computed to examine the relative importance of each cluster from the second card sorting task, controlling for age, gender, education level and marital status.

A linear regression analysis was conducted between the threat categories and the severity of symptoms, controlling for covariation of the participants education level to assess an association between a category and symptom severity controlling for education level.

3. Results

3.1 Participants

To have sufficient statistical power and from a practical point of view, it was decided to use all participants with somatic symptoms for this study. In the online survey 701 people from Europe and South America participated. The 73 participants in the card sorting task were from the Netherlands. Scores from participant 34 was discarded from the analysis because the task was not understood properly. Table 1 shows the characteristics of the participants in both studies.

Table 1*Characteristics of Participants*

	Online survey (<i>N</i> =701)	Card sorting task (<i>N</i> =73)
Sex		
Female, <i>n</i> (%)	655 (93.4%)	69 (94.5%)
Male, <i>n</i> (%)	46 (6.6%)	4 (5.5%)
Age, mean (min-max)	45.4 (18-80)	49.2 (22-68)
Country of origin, <i>n</i> (%)		
The Netherlands	407 (58.1%)	73 (100%)
Belgium	51 (7.3%)	
Brazil	115 (16.4%)	
Cyprus	11 (1.6%)	
Greece	37 (5.3%)	
Peru	21 (3.0%)	
Portugal	30 (4.3%)	
Other	29 (4.1%)	
Relationship status, <i>n</i> (%)		
Having a relationship	534 (76.3%)	49 (67.1%)
Not in a relationship	166 (23.7%)	24 (32.9%)
Education level		
Low or middle		34
High		39
total PHQ-15, mean (SD)		12.9 (4.5)

Notes. For relationship status there was one missing value in the online survey, with a participant having skipped the question. .

In the card sorting task, low or middle levels of education were defined as the highest obtained diploma being elementary school, pre-vocational secondary education or secondary vocational education. Higher educational level was defined as the highest obtained diploma being senior general secondary education, pre-university education, higher professional education, or university education.

Abbreviation: PHQ-15, patient health questionnaire 15.

3.2 Online survey: hierarchical cluster analysis

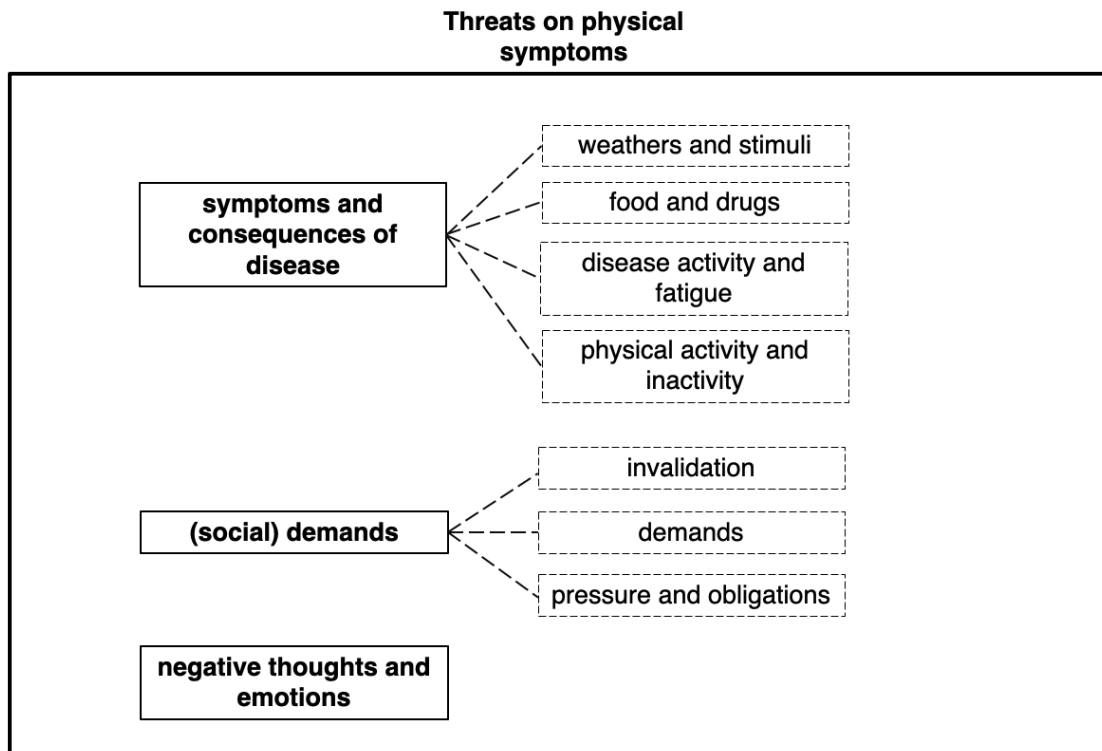
With the use of the dendrogram, made with the hierarchical cluster analysis, three categories were constructed, with a total of eight clusters. This is schematically shown in figure 1. The three categories constructed were: “symptoms and consequences of disease”, “(social) demands” and “negative thoughts and emotions”. The first category was divided into the clusters “weather and stimuli”, “food and drugs”, “disease activity and fatigue” and “physical activity and inactivity”. The second category was divided into the clusters “invalidation”, “(social)demands”, and “pressure and obligations”. A supplementary table and figure with all the items are found in Appendix A and Appendix B.

The eight cluster solution was chosen over nine or more clusters because adding in a ninth cluster would have divided “physical activity and inactivity”. The split would mean there would be a cluster with “physical effort (22)”, “common physical activity (36)” and “task work household (30)” and a cluster with “physically inactive (11)”, “doing nothing (31)” and “posture for long (3)”. If split, “posture for long” would not match the other two items in its cluster, being more an item of physical activity rather than inactivity. If another split would have occurred, the item would have been a cluster of its own, however, a criterion in the creation of clusters was to include a minimum of two items.

The highest reliability was acquired for a cluster solution with eight clusters, having three overarching categories. The eight cluster solution was chosen over a seven or less clusters because decreasing the amount of clusters would have meant that the clusters “disease activity and fatigue” and “physical activity and inactivity” would have to be combined, whereas self-contained these clusters cover more of a defined category. The items within the cluster “physical activity and inactivity” are focused on something that the individual performs himself rather than something that happens to the individual, for example performing a physical motion or being idle. The cluster “disease activity and fatigue” relies more on a consequence, which can be being out of energy, a physical symptom or disease activity.

Figure 1

Schematic view of the hierarchical categorization of threats.



3.3 Order of importance

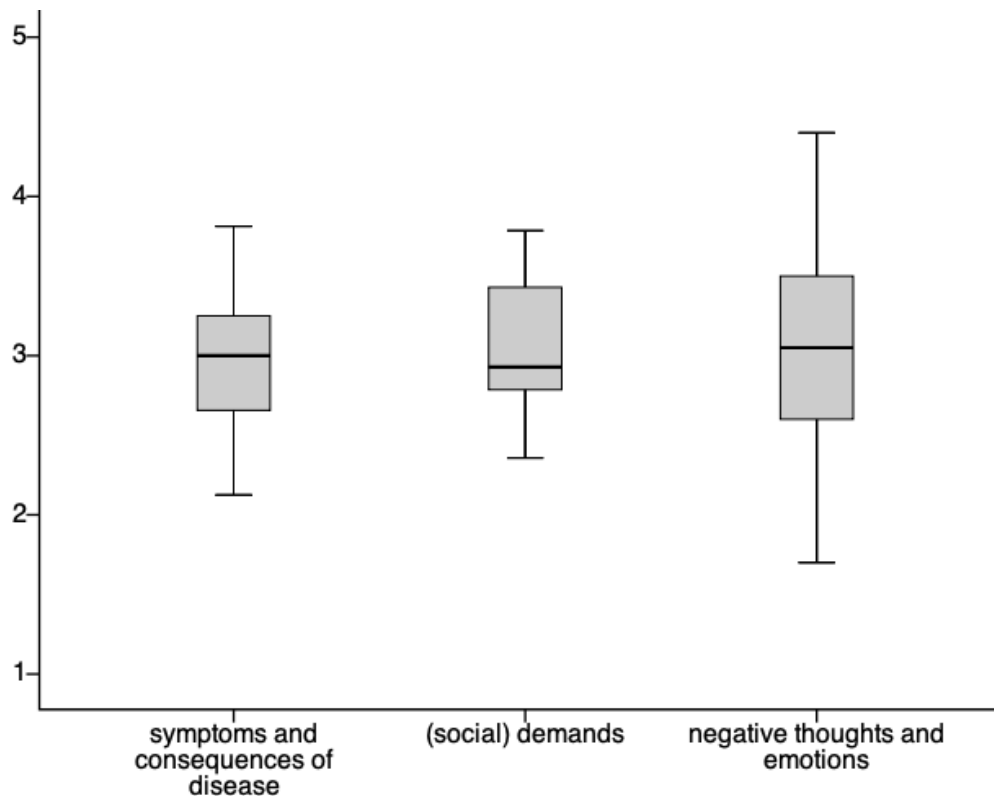
Bivariate Pearson's correlation coefficients (r) were calculated to check whether the age, gender, education or marital status correlated with the perceived importance of the threat categories. Because none of the correlations were significant, none were used as a covariate. Then a repeated measure analysis of variance was performed using a General Linear Model to check whether there was a significant difference in perceived importance of threats per category.

In figure 2 a boxplot is shown with the distributions of scores on perceived importance for the threatening categories of somatic symptoms. Shapiro-Wilk statistics indicated that the assumption of normality was supported in all categories: $F_{\max}=0.0740$ demonstrating homogeneity of variance; and Mauchly's test indicated that the assumption of sphericity was not violated. The results showed that there is no difference in perceived importance of the threat per category $F(2, 132) = 1.849, p = .155$ partial $\eta^2 = .028$. Also, pairwise comparisons revealed no further differences. The category for negative thoughts and emotions shows a wide range in the boxplot. The big spread in scores shows that perceived importance for

participants differs a lot in this category. In the supplementary table listed in Appendix C, the importance scores are shown for all the categories and separate items.

Figure 2

Distribution of scores of the perceived importance values per threat category for people with chronic somatic symptoms.



Note. The least threatening items got a lowest score of 1, where the most threatening items got the highest score of 5. The bottom of the box shows the 25th percentile of the scores on the category. The top shows the 75th percentile and the line in between shows the median. The minimum and maximum values are show with the bars outside of the boxed.

3.4 Card sorting task: relation between somatic symptom severity and threat categories

Bivariate Pearson's correlation coefficients (r) were calculated to check whether the age, gender, education, or marital status correlated with the total score on the PHQ-15. The bivariate correlation between education and the PHQ-15 was negative and small, $r(76) = -.253$, $p = .036$. Therefore, it was included as a covariate in the regression analysis. Other demographic variables were not significant, therefore will not be used as covariate in the

regression analysis. Prior to calculating r , the assumptions of normality, linearity and homoscedasticity were assessed, and found to be supported.

To examine the association between the threats of each category and symptom severity, three regression analyses were used correlating the total PHQ-15 with the scores of each category. The participants' education levels were included as a covariate, to partial out the effects of participants' education level. For the category "symptoms and consequences of disease" no significant regression equation was found ($F(1,65) = .178, p = .675$), with an R^2 of .003. Nor was there for the category "(social) demands" ($F(1,64) = .027, p = .870$), with an R^2 of .000. Nor was there for the category "negative thoughts and emotions" ($F(1,64) = 2.110, p = .151$), with an R^2 of .032. Concluding, no significant regressions were found meaning that the three threat categories have no association with the experienced severity of somatic symptoms.

4. Discussion

The aim of this study was to identify and structure threats to the symptoms of people with FM, examining the perceived importance between threat-categories and the relationship between those categories and severity of symptoms. Three categories of threats were found: "symptoms and consequences of disease", "(social) demands" and "negative thoughts and emotions". The category "symptoms and consequences on diseases" consisted of threats concerning weather and stimuli, food and drugs, disease activity and fatigue and physical activity and inactivity. The category "(social) demands" was structured around threats concerning invalidation, demands and pressure and obligations. The threats in the category "negative thoughts and emotions" were all concerning negative thoughts and emotions such as being stressed or tense, having worries, being angry or feeling lonely. The importance of each category did not differ between the categories, meaning that the three categories were on average perceived as to be of even importance. None of the categories were associated with the severity of the presented symptoms.

It was hypothesized that the threats would belong to categories for social-, non-social-, and internal threats. Social threats reported were a lack of understanding from others, getting negative judgments or comments from others, social pressure, and social invalidation, getting visitors at home, not being able to keep up in a group activity and other social activities outside of the house. These threats were categorized amongst other threats in the category for "(social) demands". Non-social threats encountered were as hypothesized different stimuli

such as noises, scents, bright lights, or radiation. Also influences of weather such as temperature, humidity or abrupt changes in weather conditions are mentioned as threatening for somatic symptoms. Other reported threats closely related are the use of medication, particular food and substance abuse such alcohol, cigarettes, or soft drugs. In terms of internal threats, the physical threats found concerned disease activity, fatigue, physical activity, and inactivity. An example of disease activity could be inflammation, infection, or flu. As to physical activities threatening somatic symptoms there were mentions of physical effort, common activity such as walking or cycling, but also work tasks, household activities and administrative tasks. Holding a certain posture for long, doing nothing and being physically inactive can all lead to increase in symptoms as well. Not hypothesized another category was found regarding negative thoughts and emotions. The content of thoughts is of great importance in the process of catastrophization (Malin & Littlejohn, 2015). This is the phenomenon of expecting or worrying about major negative consequences from a situation (Turner et al., 2002). This phenomenon is associated with the intensity of pain and psychological distress therefor possibly being harmful to somatic symptoms for patient with FM. Negative emotions can be a result of negative thought, having found to modulate neurophysiological responses relevant to the pathophysiology of FM (Malin & Littlejohn, 2015). In conclusion, categories were found that were in accordance with the hypothesis. Beyond expectations, more threats were found with cognition and emotions that made sense. In hindsight, these can be found in literature as well.

Part of this research was to explore what perceived differences people with FM report for importance. Mean scores on importance were calculated for each category, comparison showed that the means did not differ significantly between categories of threats. Meaning that there is no difference in importance for threats based on symptoms and consequences of disease, threats based on (social) demands and threats based on negative thoughts and emotions. However, it is important to note here that many differences between the participants were reported, especially in the category for negative thoughts and emotions. The influence of the threats in this category are very important for some of the participants and for some very unimportant, making that the average is ultimately in between. The spread is somewhat smaller in the other categories. An important conclusion that can be drawn from this is that the importance of threats differs greatly from individual to individual. This is recognized in literature, not having found a specific FM personality that drives the FM mechanism, Malin and Littlejohn (2012) propose that the individual differences in terms of experiencing the disease, coping with psychological stress, and physical responses differs as

much between people with FM as personality itself. This indicates the importance of tailor-made treatment.

The study aimed to explore how the categories of threats found relate to the severity of the somatic symptoms. None of the categories have been found to have any association with the experienced severity of somatic symptoms. This could partly be explained by the individual differences in patients with FM as mentioned in the previous paragraph, each patient has vastly different experiences regarding specific threats making it difficult to find a trend among them. Also, due to the fact that the cards in this sorting task had to be divided evenly among five stacks of eight, someone who might have experienced all threats as serious needed to score just as high across all cards as someone who did not experience any threat. Another side note is that the PHQ-15 is an instrument that asks for physical complaints, where people with FM also have complaints related to cognition and emotion. This could explain why no link was found.

With the results of this study come clinical implications. It is proposed to compile a screening list based on the threats found. This list could be used to distinguish what is of interest to an individual regarding their somatic symptoms. The use of the screening list also allows to adapt the treatment to a certain category of threats the patient must face. If the screening list shows that there is a high score for threats from the category for "symptoms and consequences of disease", Mindfulness based interventions can offer relief for FM symptoms (Pinto et al., 2020a). Adler-Neal and Zeidan (2017) propose that mind-body interventions, particularly Mindfulness-Meditation, attenuate pain by improvements in mood and cognitive flexibility. The pain relief is established by developing sustained attention to arising sensory, affective and cognitive events, recognizing such events as momentary and fleeting and softening the judgment to these events. The interventions ensure that catastrophization stops and the perceived threat values decreases (Adler-Neal & Zeidan, 2017). When threats in particular affect the patient from the "(social) demands" category, interventions aimed at increasing self-efficacy and pain acceptance can help (Cameron et al., 2018). Cameron et al. (2018) suggest that combined occurrence of high self-efficacy and high acceptance is associated with lower lack of understanding. Aimed at changing the content of irrational thoughts, the premise of cognitive therapy is that emotions and behavior can be changed by changing underlying thoughts (Ellis & Maclaren, 2021). By promoting helpful thoughts and behavior Cognitive Behavioral Therapy (CBT) aims to increase coping skills such self-efficacy, strengthening the personal belief that one can successfully achieve a goal by performing particular behavior (Bandura, 1977, in Cameron et al., 2018; Beck, 1997; Keefe,

Abernethy & Campbell., 2005) this confirmation can make a person less prone to threats of invalidation, demands, pressure and obligations. Acceptance and Commitment Therapy has been found effective helping patients to cope with chronic disease and commit to daily life (Hayes, et al., 2006). If the screening list shows that someone is particularly troubled by threats from the category "negative thoughts and emotions", they could possibly thrive with Emotion Awareness and Expression Therapy (EAET). This treatment integrates different techniques of trauma-related and emotion-focused therapies such as experiential therapy, intensive psychodynamic therapy, prolonged exposure therapy, expressive writing therapy, and therapeutic rescripting therapy (Lumley et al., 2017). The therapy aims to teach FM patients to attribute their somatic symptoms to emotionally activated central nervous system mechanisms and become aware of, experience, and adaptively express their emotions, mood from adversity, trauma, or conflict (Lumley et al., 2017). EAET shows promising results, with lower FM symptoms and widespread pain than CBT (Lumley et al., 2017). It is important to note that Lumley et al. (2017) report in their study that differences were found on pain-related outcomes and research needs to be conducted in order to check what particular patients would benefit from what particular treatment. In future research, the screening list can distinguish patients who experience many threats in the category "negative thoughts and emotions" to see if they benefit more from the EAET or CBT.

The current study presented a strength in the form of its procedure. By asking participants open questions in the first phase of the study, all responses were completely based on information provided by patients not directing them in any direction, adding to the explorative nature of the study. Also, by providing a large sample of participants and them having different nationalities during the acquisition of threats lead to a great diversity of threats. This diversity contributed to a large representative framework of threat factors for people with FM. However, a limitation can be found in the generalization of the study. With it having largely been female participants, the results make to be mainly generalizable for women. Also, data was used for participants with different somatic disorders (e.g., irritable bowel syndrome, migraine, osteoarthritis) possibly influencing the external validity.

Regarding future research, it is recommended to implement the identified threat categories in a screening list and to have it validated by patients with FM. The effectiveness of the treatments mentioned after using the screening list could also be examined.

Using the perspective of people with FM this research contributes to scientific knowledge in that it provides a structured and compassing overview of threats that overactivate the threat-detection system in people with FM. As a result, this research can be

used for screening, intakes and to put together a tailor-made treatment with the aim of making this disabling condition more bearable.

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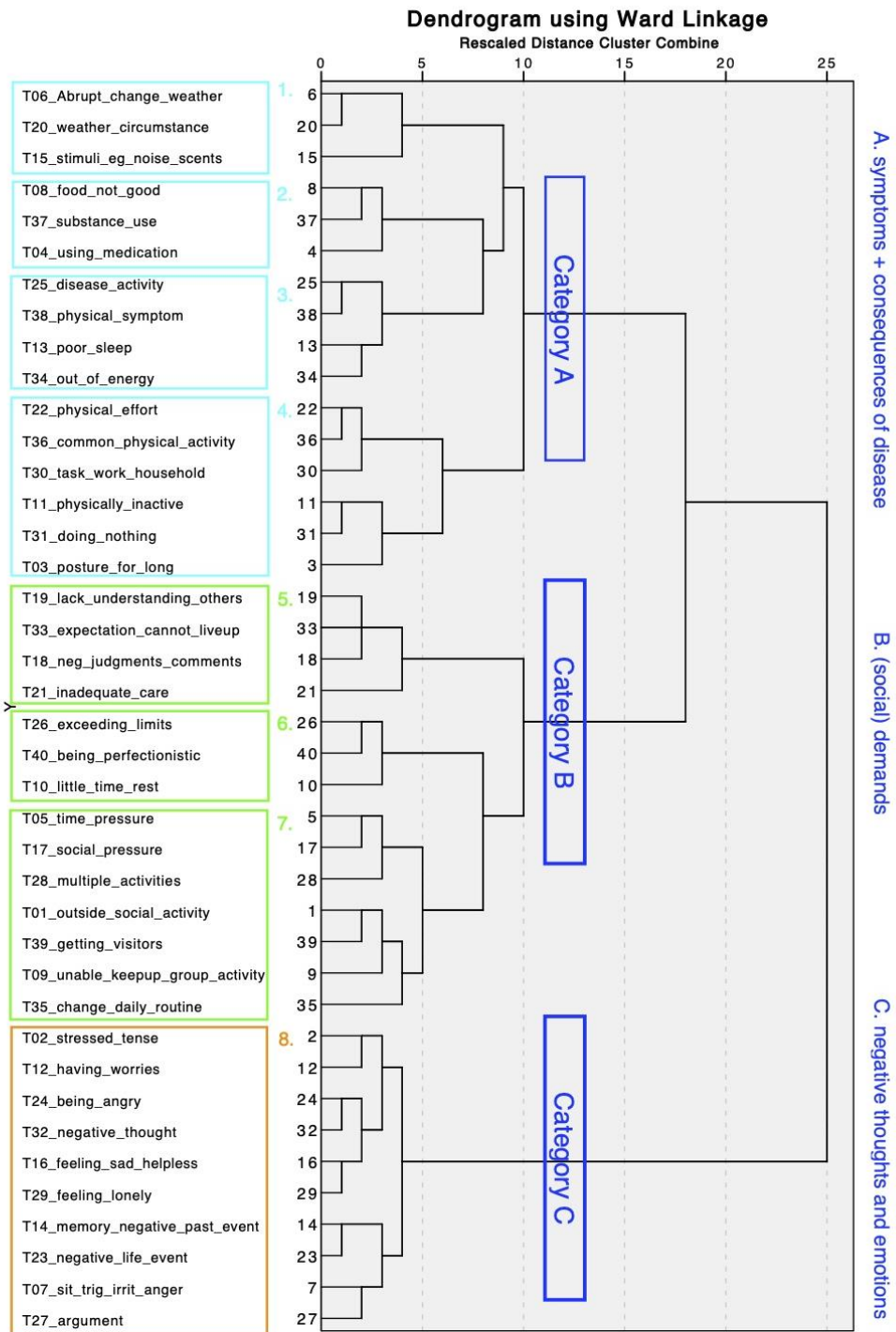
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6. Appendices

Appendix A: Final threats used for the card sorting task

1. A social activity outside the home
2. Being stressed or tense
3. Holding a certain posture for long
4. Using medication
5. Time pressure
6. An abrupt change in weather
7. A situation that triggers irritation or anger
8. Food that is not good for me
9. Being unable to keep up in a group activity
10. Little time to rest
11. Being physically not active
12. Having worries
13. Poor sleep
14. Memory of a negative past event
15. Stimuli, such as noises, scents, bright lights or radiation
16. Feeling sad or helpless
17. Social pressure and invalidation
18. Getting negative judgments or comments
19. Lack of understanding from others
20. A weather circumstance, such as temperature or humidity
21. Getting inadequate care
22. Physical effort
23. A negative life event
24. Being angry
25. An inflammation, infection, flu or other disease activity
26. Exceeding my limits
27. An argument
28. Having multiple activities scheduled
29. Feeling lonely
30. A task at work or in the household, or an administrative task
31. Doing nothing
32. A negative thought
33. An expectation that I cannot live up to
34. Being out of energy
35. A change in daily routine
36. A common physical activity such as walking or cycling
37. Substances such as alcohol, cigarettes or soft drugs
38. A physical symptom such as pain, fatigue or stiffness
39. Getting visitors at home
40. Being perfectionistic

Appendix B: Dendrogram



- 1. weather and stimuli
- 2. food and drugs
- 3. disease activity and fatigue
- 4. physical activity and inactivity
- 5. invalidation
- 6. demands
- 7. pressure and obligations
- 8. negative thoughts and emotions

Appendix C: Supplementary file

This table shows the mean importance rating (and standard deviation) of each individual outcome and the overarching cluster. An example for an item would be: “An abrupt change in weather creates experiences of danger, harm, damage or unsafety” The lowest possible score is 1 (least important) and the highest possible score is 5 (most important).

	Importance	
	Mean	Standard deviation
<u>Cluster 1 – Symptoms and Consequences of disease</u>	<u>2.98</u>	<u>0.44</u>
Weather and stimuli	2.71	0.98
An abrupt change in weather (6)	2.17	1.24
A weather circumstance, such as temperature or humidity (20)	2.80	1.56
Stimuli, such as noises, scents, bright lights or radiation (15)	2.44	1.37
Food and drugs	2.30	0.94
Food that is not good for me (8)	2.83	1.43
Substances such as alcohol, cigarettes or soft drugs (37)	2.16	1.54
Using medication (4)	1.89	1.30
Disease activity and fatigue	3.95	0.64
An inflammation, infection, flu or other disease activity (25)	3.69	1.36
A physical symptom such as pain, fatigue or stiffness (38)	4.17	1.06
Poor sleep (13)	4.13	1.16
Being out of energy (34)	3.80	1.23
Physical activity and inactivity	2.90	0.72
Physical effort (22)	3.21	1.38
A common physical activity such as walking or cycling (36)	2.56	1.34
A task at work or in the household, or an administrative task (30)	2.49	1.24
Being physically not active (11)	3.00	1.49
Doing nothing (31)	2.49	1.58
Holding a certain posture for long (3)	3.66	1.27
<u>Cluster 2 – (social) Demands</u>	<u>3.16</u>	<u>0.39</u>
Invalidation	3.08	0.71
Lack of understanding from others (19)	3.17	1.12
An expectation that I cannot live up to (33)	3.01	1.09
Getting negative judgments or comments (18)	3.14	1.37
Getting inadequate care (21)	2.99	1.44
Demands	3.88	0.70
Exceeding my limits (26)	4.56	0.79
Being perfectionistic (40)	3.04	1.36
Little time to rest (10)	4.06	1.05
Pressure and obligations	2.62	0.64
Time pressure (5)	3.04	1.24
Social pressure and invalidation (17)	2.85	1.29

Having multiple activities scheduled (28)	3.43	1.29
A social activity outside the home (1)	2.49	1.19
Getting visitors at home (39)	2.11	1.08
Being unable to keep up in a group activity (9)	2.42	1.62
A change in daily routine (35)	2.13	1.19
<u>Cluster 3 – Negative thoughts and emotions</u>	<u>3.07</u>	<u>0.64</u>
Being stressed or tense (2)	4.01	1.02
Having worries (12)	3.56	1.12
Being angry (24)	2.59	1.18
A negative thought (32)	2.66	1.07
Feeling sad or helpless (16)	3.39	1.24
Feeling lonely (29)	2.72	1.47
Memory of a negative past event (14)	2.62	1.20
A negative life event (23)	3.20	1.27
A situation that triggers irritation or anger (7)	2.86	1.16
An argument (27)	3.01	1.27

Appendix D: Syntax for hierarchical cluster-analysis

* Encoding: UTF-8.

* RUN THE FIRST STEP OF THE CLUSTERANALYSIS.

DATASET DECLARE D0.7933626911670203.

PROXIMITIES T01_outside_social_activity

T02_stressed_tense

T03_posture_for_long

T04_using_medication

T05_time_pressure

T06_Abrupt_change_weather

T07_sit_trig_irrit_anger

T08_food_not_good

T09_unable_keepup_group_activity

T10_little_time_rest

T11_physically_inactive

T12_having_worries

T13_poor_sleep

T14_memory_negative_past_event

T15_stimuli_eg_noise_scents

T16_feeling_sad_helpless

T17_social_pressure

T18_neg_judgments_comments

T19_lack_understanding_others

T20_weather_circumstance

T21_inadequate_care

T22_physical_effort

T23_negative_life_event

T24_being_angry

T25_disease_activity

T26_exceeding_limits

T27_argument

T28_multiple_activities

T29_feeling_lonely

T30_task_work_household

T31_doing_nothing

T32_negative_thought

T33_expectation_cannot_liveup

T34_out_of_energy

T35_change_daily_routine

T36_common_physical_activity

T37_substance_use

T38_physical_symptom

T39_getting_visitors

T40_being_perfectionistic

/MATRIX OUT(D0.7933626911670203)

/VIEW=VARIABLE

/MEASURE=SEUCLID

/PRINT NONE

/STANDARDIZE=VARIABLE NONE.

*Now I have a file containing how many times card have been piled together.

*In the next step we make them in to Squared Euclidian Distances.

RECODE T01_outside_social_activity
T02_stressed_tense (10=25)
T03_posture_for_long (12=36)
T04_using_medication (14=49)
T05_time_pressure (16=64)
T06_Abrupt_change_weather (18= 81)
T07_sit_trig_irrit_anger (20=100)
T08_food_not_good (22=121)
T09_unable_keepup_group_activity (24=144)
T10_little_time_rest (26=169)
T11_physically_inactive (28=196)
T12_having_worries (30=225)
T13_poor_sleep (32=256)
T14_memory_negative_past_event (34=289)
T15_stimuli_eg_noise_scents (36=324)
T16_feeling_sad_helpless (38=361)
T17_social_pressure (40=400)
T18_neg_judgments_comments (42=441)
T19_lack_understanding_others (44=484)
T20_weather_circumstance (46=529)
T21_inadequate_care (48=566)
T22_physical_effort (50=625)
T23_negative_life_event (52=676)
T24_being_angry (54=729)
T25_disease_activity (56=784)
T26_exceeding_limits (58=841)
T27_argument (60=900)
T28_multiple_activities (62=961)
T29_feeling_lonely (64=1024)
T30_task_work_household (66=1089)
T31_doing_nothing (68=1156)
T32_negative_thought (70=1225)
T33_expectation_cannot_liveup (72=1296)
T34_out_of_energy (74=1369)
T35_change_daily_routine (76=1444)
T36_common_physical_activity (78=1521)
T37_substance_use (80=1600)
T38_physical_symptom (82=1681)
T39_getting_visitors (84=1764)
T40_being_perfectionistic (86=1849)
(2=1) (88=1936)
(4=4) (90=2025)
(6=9) (92=2116)
(8=16) (94=2209)

(96=2304)
(98=2401)
(100=2500)
(102=2601)
(104=2704)
(106=2809)
(108=2916)
(110=3025)
(112=3136)
(114=3249)
(116=3364)
(118=3481)
(120=3600)
(122=3721)
(124=3844)
(126=3969)
(128=4096)
(130=4225)
(132=4356)
(134=4489)
(136=4624)
(138=4761)
(140=4900)
(142=5041)
(144=5184)
(146=5329)
(148=5476)
(150=5625)
(152=5776)
(154=5929)
(156=6084)
(158=6241)
(160=6400)

(162=6561)
(164=6724)
(166=6889)
(168=7056)
(170=7225)
(172=7396)
(174=7569)
(176=7744)
(178=7921)
(180=8100)
(182=8281)
(184=8464)
(186=8649)
(188=8836)
(190=9025)
(192=9216)
(194=9409)
(196=9604)
(198=9801)
(200=10000)
(202=10201)
(204=10404)
(206=10609)
(208=10816)
(210=11025)
(212=11236)
(214=11440)
(216=11664)
(218=11881)
(220=12100).

EXECUTE.

*This is the cluster analysis.

CLUSTER

```
/MATRIX IN(D0.7933626911670203)
/METHOD WARD
/PRINT SCHEDULE CLUSTER(4,12)
/PLOT DENDROGRAM VICICLE.
Dataset Close D0.7933626911670203.
```

Appendix E: Syntax for clusters and regressions

* Encoding: UTF-8.

* QUESTION 2.

*1) CHECK FOR ERRORS AND RECODE

*In our dataset (N=99) we have two participants who have not filled in our threats; therefore, they will be deleted from the dataset.

*ELIMINATE SUBJECT 76

FILTER OFF.

USE ALL.

SELECT IF NOT (Participant = 76).

EXECUTE.

*ELIMINATE SUBJECT 94

FILTER OFF.

USE ALL.

SELECT IF NOT (Participant = 94).

EXECUTE.

*we also eliminate subject 35, for this subject misinterpreted the research, therefor being an outlier in the study

FILTER OFF.

USE ALL.

SELECT IF NOT (Participant = 35).

EXECUTE.

*Incidental missing values of six participants were put on separate piles.

IF (participant=10) T37_substance_use=10.

IF (participant=11) T04_using_medication=6.

IF (participant=17) T34_out_of_energy=8.

IF (participant=22) T06_Abrupt_change_weather=8.

IF (participant=25) T10_little_time_rest=6.

IF (participant=25) T13_poor_sleep=7.

IF (participant=25) T15_stimuli_eg_noise_scents=8.

IF (participant=75) T27_argument=6.

IF (participant=75) T40_being_perfectionistic=7.

EXECUTE.

*One importer accidentally noted a 55 instead of a 5 on T_value13, we correct for this with

IF (participant=3) T_value13=5.

EXECUTE.

*RELATIONSHIP STATUS

* Relationship status is recoded to having a relationship or not.

RECODE MARITALSTATUS (1=1) (2=2) (3=1) (4=1) **INTO**
RECODED_MARITAL_STATUS.
EXECUTE.

* Moreover, the “other” relationship status should be added.

* The following syntax is suggested.

IF (Marital_Other = "Lat relationship") **RECODED_MARITAL_STATUS** =2.
IF (Marital_Other = "Lat relationship") **RECODED_MARITAL_STATUS** =2.
IF (Marital_Other = "lat-relatie") **RECODED_MARITAL_STATUS** =2.
EXECUTE.

* EDUCATION.

* Two “other education” can be used recoded to a given education level.

IF (Educ_other = "option 3 and 5") **Education**=5.
IF (Educ_other = "prop. HBO") **Education**=5.
EXECUTE.

* RECODE OF EDUCATION INTO LOWER OR MIDDLE VS. HIGHER LEVEL.

RECODE Education (1=1) (2=1) (3=1) (4=1) (5=2) (6=2) (7=2) **INTO**
RECODED_EDUCATION.
EXECUTE.

*2) COMPUTE CRONBACH ALPHA FOR CATEGORIES(3) TO KNOW WHICH ITEMS SHOULD BE INCLUDED IN THE CALCULATION OF CATEGORY SCORES.

*A. Symptoms and Consequences for Disease: T_Symptoms_Con_Disease

RELIABILITY

/VARIABLES=T06_Abrupt_change_weather T20_weather_circumstance
T15_stimuli_eg_noise_scents
T08_food_not_good T37_substance_use T04_using_medication T25_disease_activity
T38_physical_symptom
T13_poor_sleep T34_out_of_energy T22_physical_effort T36_common_physical_activity
T30_task_work_household T11_physically_inactive T03_posture_for_long
/SCALE('ALL VARIABLES') ALL
/MODEL=ALPHA
/STATISTICS=DESCRIPTIVE SCALE CORR ANOVA
/SUMMARY=TOTAL MEANS VARIANCE.

*B. (social) demands: T_Press_Soc_Dem

RELIABILITY

```
/VARIABLES=T19_lack_understanding_others T33_expectation_cannot_liveup  
T18_neg_judgments_comments  
T21_inadequate_care T26_exceeding_limits T40_being_perfectionistic T10_little_time_rest  
T05_time_pressure T17_social_pressure T28_multiple_activities T01_outside_social_activity  
T39_getting_visitors T09_unable_keepup_group_activity T35_change_daily_routine  
/SCALE('ALL VARIABLES') ALL  
/MODEL=ALPHA  
/STATISTICS=DESCRIPTIVE SCALE CORR ANOVA  
/SUMMARY=TOTAL MEANS VARIANCE.
```

*C. Negative thoughts and emotions: T_Neg_Thoughts_Emo

RELIABILITY

```
/VARIABLES=T02_stressed_tense T12_having_worries T24_being_angry  
T32_negative_thought  
T07_sit_trig_irrit_anger T14_memory_negative_past_event T16_feeling_sad_helpless  
T23_negative_life_event T27_argument T29_feeling_lonely  
/SCALE('ALL VARIABLES') ALL  
/MODEL=ALPHA  
/STATISTICS=DESCRIPTIVE SCALE CORR ANOVA  
/SUMMARY=TOTAL MEANS VARIANCE.
```

*3) COMPUTE THE CATEGORY VARIABLES

*A. Symptoms and Consequences for Disease: T_Symptoms_Con_Disease

```
COMPUTE T_Symptoms_Con_Disease=(T_value06 + T_value20 + T_value15 + T_value08  
+ T_value37 + T_value04 + T_value25 + T_value38 + T_value13 + T_value34 + T_value22  
+ T_value36 + T_value30 + T_value11 + T_value31 + T_value03) / 16.  
EXECUTE.
```

*B. (social) demands: T_Press_Soc_Dem

```
COMPUTE T_Press_Soc_Dem=(T_value19 + T_value33 + T_value18 + T_value26 +  
T_value40 + T_value10 + T_value05 + T_value17 + T_value28 + T_value01 + T_value39 +  
T_value09 + T_value21 + T_value35) / 14.  
EXECUTE.
```

*C. Negative thoughts and emotions: T_Neg_Thoughts_Emo

```
COMPUTE T_Neg_Thoughts_Emo=(T_value02 + T_value12 + T_value24 + T_value32 +  
T_value16 + T_value29 + T_value14 + T_value23 + T_value07 + T_value27) / 10.  
EXECUTE.
```

* 4) COMPARE THE MEANS OF THE THREAT CATEGORIES.

```
GLM T_Symptoms_Con_Disease T_Press_Soc_Dem T_Neg_Thoughts_Emo  
/WSFACTOR=Threatcategories 3 Polynomial  
/METHOD=SSTYPE(3)  
/PLOT=PROFILE(Threatcategories)  
/EMMEANS=TABLES(Threatcategories) COMPARE ADJ(BONFERRONI)  
/PRINT=DESCRIPTIVE ETASQ  
/CRITERIA=ALPHA(.05)  
/WSDESIGN=Threatcategories.
```

*The threat categories don't differ from each other significantly.

* 5) CHECK WHETHER AGE, GENDER OR RECODED_EDUCATION
RECODED_MARITAL_STATUS ARE CORRELATED WITH ONE OR MORE OF THE 3
CATEGORIES.

*A. Symptoms and Consequences for Disease

CORRELATIONS

```
/VARIABLES=T_Symptoms_Con_Disease RECODED_EDUCATION  
RECODED_MARITAL_STATUS Gender Age  
/PRINT=TWOTAIL NOSIG  
/STATISTICS DESCRIPTIVES  
/MISSING=PAIRWISE.
```

*B. (social) demands

CORRELATIONS

```
/VARIABLES=T_Press_Soc_Dem RECODED_EDUCATION  
RECODED_MARITAL_STATUS Gender Age  
/PRINT=TWOTAIL NOSIG  
/STATISTICS DESCRIPTIVES  
/MISSING=PAIRWISE.
```

*C. Negative thoughts and emotions

CORRELATIONS

```
/VARIABLES=T_Neg_Thoughts_Emo RECODED_EDUCATION  
RECODED_MARITAL_STATUS Gender Age  
/PRINT=TWOTAIL NOSIG  
/STATISTICS DESCRIPTIVES  
/MISSING=PAIRWISE.
```

* IF SO, ADD THE COVARIATES IN THE ANALYSIS BELOW.

```
GLM T_Symptoms_Con_Disease T_Press_Soc_Dem T_Neg_Thoughts_Emo WITH Age  
Gender RECODED_EDUCATION RECODED_MARITAL_STATUS  
/WSFACTOR=Threatcategories 3 Polynomial  
/METHOD=SSTYPE(3)
```

```

/PLOT=PROFILE(Threatcategories)
/EMMEANS=TABLES(Threatcategories) WITH(Age=MEAN Gender=MEAN
RECODED_EDUCATION=MEAN RECODED_MARITAL_STATUS=MEAN)COMPARE
ADJ(BONFERRONI)
/PRINT=DESCRIPTIVE ETASQ
/CRITERIA=ALPHA(.05)
/WSDESIGN=Threatcategories
/DESIGN=Age Gender RECODED_EDUCATION.

```

*No significant covariates were found

```

EXAMINE VARIABLES=T_Symptoms_Con_Disease T_Press_Soc_Dem
T_Neg_Thoughts_Emo
/COMPARE VARIABLE
/PLOT=BOXPLOT
/STATISTICS=NONE
/NOTOTAL
/MISSING=LISTWISE.

```

*QUESTION 3.

*COMPUTE TOTAL_phq FIRST.

```

COMPUTE TOTAL_PHQ =
15*MEAN.10(PHQ01,PHQ02,PHQ03,PHQ04,PHQ05,PHQ06,PHQ07,PHQ08,
PHQ09,PHQ10,PHQ11,PHQ12,PHQ13,PHQ14,PHQ15).
EXECUTE.

```

*CHECK WHETHER COVARIATES (THE FOUR) ARE CORRELATED WITH THE DEPENDENT VARIABLE (total_phq).

CORRELATIONS

```

/VARIABLES=TOTAL_PHQ RECODED_EDUCATION
RECODED_MARITAL_STATUS Gender Age
/PRINT=TWOTAIL NOSIG
/STATISTICS DESCRIPTIVES
/MISSING=PAIRWISE.

```

* RECODED_EDUCATION correlates with TOTAL_PHQ.

*THUS, INCLUDE RECODED_EDUCATION AS COVARIATE IN REGRESSION ANALYSIS.

* WE DO THE ANALYSIS SEPRATELY FOR EACH OF THE THREE CATEGORIES.

*A. Symptoms and Consequences for Disease: T_Symptoms_Con_Disease

REGRESSION

```

/MISSING PAIRWISE
/STATISTICS COEFF OUTS R ANOVA CHANGE
/CRITERIA=PIN(.05) POUT(.10)
/NOORIGIN

```

```
/DEPENDENT TOTAL_PHQ  
/METHOD=ENTER T_Symptoms_Con_Disease  
/METHOD=ENTER RECODED_EDUCATION  
/SCATTERPLOT=(*ZRESID ,*ZPRED).
```

*B. (social) demands: T_Press_Soc_Dem

REGRESSION

```
/MISSING PAIRWISE  
/STATISTICS COEFF OUTS R ANOVA CHANGE  
/CRITERIA=PIN(.05) POUT(.10)  
/NOORIGIN  
/DEPENDENT TOTAL_PHQ  
/METHOD=ENTER T_Press_Soc_Dem  
/METHOD=ENTER RECODED_EDUCATION  
/SCATTERPLOT=(*ZRESID ,*ZPRED).
```

*C. Negative thoughts and emotions: T_Neg_Thoughts_Emo

REGRESSION

```
/MISSING PAIRWISE  
/STATISTICS COEFF OUTS R ANOVA CHANGE  
/CRITERIA=PIN(.05) POUT(.10)  
/NOORIGIN  
/DEPENDENT TOTAL_PHQ  
/METHOD=ENTER T_Neg_Thoughts_Emo  
/METHOD=ENTER RECODED_EDUCATION  
/SCATTERPLOT=(*ZRESID ,*ZPRED).
```