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Published in:
General Hospital Psychiatry

DOI:
[10.1016/j.genhosppsy.2016.10.008](https://doi.org/10.1016/j.genhosppsy.2016.10.008)

IMPORTANT NOTE: You are advised to consult the publisher's version (publisher's PDF) if you wish to cite from it. Please check the document version below.

Document Version
Publisher's PDF, also known as Version of record

Publication date:
2017

[Link to publication in University of Groningen/UMCG research database](#)

Citation for published version (APA):

Janssens, K. A. M., Houtveen, J. H., Tak, L. M., Bonvanie, I. J., Scholtalbers, A., van Gils, A., Geenenc, R., & Rosmalen, J. G. M. (2017). A concept mapping study on perpetuating factors of functional somatic symptoms from clinicians' perspective. *General Hospital Psychiatry*, 44, 51-60.
<https://doi.org/10.1016/j.genhosppsy.2016.10.008>

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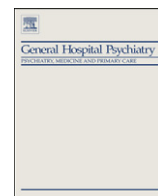
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A concept mapping study on perpetuating factors of functional somatic symptoms from clinicians' perspective



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ARTICLE INFO

Article history:

Received 6 May 2016

Revised 15 October 2016

Accepted 24 October 2016

Keywords:

Functional somatic symptoms

Hierarchical cluster analysis

Ontology

Perpetuating factors

ABSTRACT

Objective: The aim of this concept mapping study was to identify the structure and alleged importance of perpetuating factors of functional somatic symptoms (FSS) from the perspective of professionals. Further, we examined to which extent these factors have been addressed in scientific literature.

Methods: Ninety-nine perpetuating factors were sorted with respect to content using a card-sorting task by 61 experienced clinicians (62.3% psychologists, 75.4% female, mean age: 45.7 [SD: 10.6] years, mean duration of experience in treating FSS patients: 10.5 [SD: 7.6] years). These factors had been derived from in-depth interviews with 12 clinicians, with extensive experience in treating FSS. Thirty-eight clinicians rated the importance of the 99 factors on a scale ranging from 1 ('not important at all') to 10 ('extremely important').

Results: Hierarchical cluster analysis revealed three overarching domains of perpetuating factors: 'Hypochondria', 'Social and relational problems' and 'Symptom-related emotions and habits'. These domains comprised 16 clusters, which were rated on importance between 6.1, 'Adverse physical factors and counterproductive lifestyle', and 7.8, 'Frustration and despair regarding the symptoms'. All clusters have been addressed in scientific literature.

Conclusions: This study revealed an encompassing hierarchical structure of somatic, emotional, cognitive, behavioral, and social factors of importance in the perpetuation of FSS based on expert opinions. This structure will guide the development of personalized treatment of FSS.

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1. Introduction

Functional somatic symptoms (FSS), i.e. symptoms not fully explained by a medical condition, are common in general practice [1,2]. Although often perceived as innocent, these symptoms can become persistent and impairing and have large individual, societal and economic consequences [3,4]. The origin of FSS is still largely unclear, but there is consensus that their perpetuation is the result of a complex interaction between biological, psychological and social factors [5,6].

FSS constitute of a wide range of symptoms, from mild and occasionally occurring somatic symptoms to severe disabling chronic health conditions. Obviously, not all these conditions need the same treatment intensity, and therefore it has been recommended to customize treatment to the individual, e.g. by taking a stepped-care approach [7,8]. For mild symptoms, education and self-help interventions can be

offered [9,10]. More severe symptoms can be treated by more intensive treatments, like psychotherapy [11].

A problem with current treatment of FSS is that the perpetuating factors that are targeted depend mainly on the received type of treatment, rather than on the factors that are important for a specific patient. For example, cognitive behavioral therapies mainly focus on challenging unhelpful thoughts [12], graded-exercise therapy on increasing activity level [13], and mindfulness-based and acceptance and commitment therapy on helping patients to pay attention to their symptoms with curiosity and without judgment, and to be committed to live a valued life [14,15]. Other factors like doctor-patient relationship and relational problems have been found to play a role in the perpetuation of FSS as well [16,17], but are often not the focus of treatment.

Treatment of FSS might become more effective, when therapeutic targets are tailored to factors that play a role in individual patients. Research on the effects of personalized treatment of patients with FSS is currently lacking, but has been done and be found promising for other disorders [18]. In order to tailor the treatment to individual patients with FSS, an overview is needed of factors that perpetuate FSS. Such an overview helps in screening such factors and to identify the

Abbreviation: FSS, functional somatic symptoms.

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appropriate therapeutic targets for individual patients. Such a screening procedure is likely to be especially useful in self-help programs when no therapist is available to sort out the perpetuating factors that play a role in a specific patient.

Therefore, the goal of this study was to provide an overview of perpetuating factors of FSS from the perspective of experienced clinicians, with a special focus on factors that are treatable with self-help interventions. Further, we aimed to examine the underlying hierarchical structure and relative importance of these factors. Finally, we examined to what extent perpetuating factors of FSS according to clinicians are indicated to be relevant in scientific literature. The perspective of experienced clinicians on perpetuating factors of FSS was examined using a combined qualitative and quantitative concept mapping procedure consisting of interviews, a card sorting task (for hierarchical cluster analysis), and importance ratings.

2. Methods

2.1. Procedure

The development of the encompassing overview of perpetuating factors of FSS treatable with self-help interventions consisted of five steps. First, health care providers were interviewed about perpetuating factors of FSS. Second, the 662 statements obtained during the interviews were deduced to 99 perpetuating factors. Third, these 99 factors were sorted during an offline or online card sorting task. Fourth, these 99 factors were rated on importance. Fifth, a systematic literature search to examine the scientific evidence for the identified perpetuating factors was performed. These separate steps are explained in more detail below.

2.2. Interviews

Twelve health care providers who had extensive experience with treating persons with FSS were interviewed by four researchers (AS, JH, JR, KJ). Health care providers throughout the Netherlands were selected in such a way that a diversity of professions was covered, e.g. five psychologists, three general practitioners, two psychiatrists, one rehabilitation specialist and one physiotherapist were interviewed. The health care providers were also chosen based on their expertise with different patient groups, e.g. one therapist specialized in treating male patients, one in treating non-Caucasian patients, and one in treating elderly patients were included. Also one expert on mindfulness-based therapies and one on acceptance commitment therapy were interviewed. These health care providers were asked in a semi-structured interview about factors that according to them perpetuated FSS. The open question of the interview was 'In your opinion or experience which factors perpetuate FSS?' To make statements more concrete, health care providers were asked to provide examples of perpetuating factors in specific patients they had treated in the past. Guided by the biopsychosocial perspective, the interviewers asked in open questions for somatic, cognitive, emotional and social factors. The interviews took between 20 and 60 min. All interviews were verbatim transcribed. Three researchers (KJ, JH, AS, IB, AvG or JR) independently selected all statements of perpetuating factors from the transcribed interviews and discussed these selections in a consensus meeting. Statements that were ambiguous or abstract were removed, overlapping statements were combined, and statements including multiple assertions were divided. These selections led to the ultimate statements about perpetuating factors for each interview.

2.3. Card sorting task

Participants completed the card sorting task either offline or online. Offline participants received a set of 99 cards, each containing one of the perpetuating factors derived from the interviews. Participants were

asked to sort these cards thematically into different piles. In line with previous studies [19,20], the following rules applied to this sorting task: 1) all of the statements had to be placed in a pile, 2) each statement could be placed in one pile only, 3) a minimum of 4 and a maximum of 15 piles had to be formed, and 4) each pile could contain 2 to 25 statements. When finished sorting, participants were asked to place each pile in an envelope and to write down a name of the overarching theme on the envelope. In total, 38 participants completed the offline card sorting task. Twenty-three participants completed the online card sorting task, using the program *Optimal Sort*. In this program, participants could drag the statements that they believed belonged together to the same column and give that column a name reflecting the overarching theme. The four rules of the offline card sorting task also applied to participants of the online card sorting task. Offline and online participants who put more than 10% of the cards into a category 'other' or 'not sorted' were excluded from the analyses ($n = 2$). In case of some participants ($n = 3$), grouping less than 10% of the cards in a miscellaneous category, each card in this category was considered a separate pile in order to prevent non-thematic correlations. Participants who performed the card sorting task offline, did so in our attendance. Therefore, we were able to provide support if needed and took care that participants performed the task carefully. Most offline participants completed the task in 60 min. Online participants took on average 57 min (range 19 to 138 min) to complete the task.

2.4. Questionnaire about importance

After completing the card sorting task, participants were asked to rate how important each of the 99 factors was for the perpetuation of FSS in their patients. The statements could be rated on a scale ranging from 1 (= not important at all) to 10 (= extremely important). Thirty-eight participants completed this questionnaire; participants who had time left after completing the offline card sorting task completed the questionnaire offline ($n = 7$), others ($n = 31$) completed the questionnaire online, see [Table 2](#).

2.5. Analysis

Hierarchical cluster analysis in SPSS version 22 was used to create a hierarchical clustering of the 99 perpetuating factors of FSS [21]. Ward's statistics and squared Euclidean distances were used. With hierarchical cluster analyses, the factors that were frequently grouped together by individual participants are grouped together in clusters. The hierarchical structure also shows the domains that categorizes the clusters. Since no rules exist to determine the number of clusters, the number of clusters was determined by the investigators. The main criteria were that the clusters had to contain meaningful distinguishable information and had to consist of three or more statements. The hierarchical cluster solution was depicted in a dendrogram. Based on the importance questionnaire, the average importance score with standard deviations of each of the 99 statements and the average importance scores and standard deviation of the clusters were calculated.

3. Literature search

To examine to what extent perpetuating factors that were mentioned by clinicians are indicated to be relevant in scientific literature, we searched PubMed using the search strategy presented in [Appendix A](#) for each of the identified clusters. We restricted our findings to studies performed in adults. We searched for meta-analyses, systematic reviews, and narrative reviews. If no meta-analyses or reviews were found, we examined whether clinical studies examined the alleged perpetuating factors as summarized in the clusters.

4. Results

4.1. Interview statements

The interviews yielded 662 perpetuating factors; between 21 and 102 factors per interview. These apparently high numbers of perpetuating factors reflect the specificity of the factors reported. For example, 'thinking low about oneself', 'high self-criticism', 'unable to accept oneself', 'not loving oneself', 'low self-confidence', 'feeling less worthy than others', 'wish to be someone else' and 'low self-compassion' were specific factors reported by one health-care provider. These factors were combined into the single item 'a negative self-image' after several consensus meetings. Combining overlapping factors this way greatly reduced the number of factors (i.e. about 80% reduction). Since the goal of the study was to obtain an overview of perpetuating factors of FSS for identifying targets for self-help, statements that were not treatable with self-help interventions were not selected (e.g. statements about traumas during childhood or severe psychiatric comorbidity). Statements were further reduced in several consensus meetings, in which ambiguous statements or statements that were only applicable to small specific subgroups (e.g. 'Not trusting white skinned doctor') were deselected. Six authors participated in these consensus meetings. They have different backgrounds: AvG, and IB are medical doctors, JH is physiological psychologist, JR is a medical biologist and social psychologist. KJ is a developmental psychologist with a medical background, and LT is a psychiatrist. This ultimately resulted in 99 perpetuating factors; see [Appendix A](#) for the factors.

4.2. Sorting task

Characteristics of participants of the card sorting task can be found in [Table 1](#). Participants were mostly female, had an average age of 45.7 years, and had ample experience (average over ten years) with treatment of FSS patients. Most of them were psychologists or psychiatrists. The number of piles formed by the participants ranged from four to seventeen. The overarching themes that were named by ten or more participants were: 'Acceptance', 'Anxiety', 'Avoidance', 'Bodily perception', 'Coping', 'Dysfunctional cognitions', 'Illness gain', 'Lifestyle', 'Negative cognitions', 'Personality factors', 'Problems in the social network', and 'Somatic attribution'.

4.3. Hierarchical card sorting solution

The hierarchical card sorting solution is depicted in [Fig. 1](#). The dendrogram showed that perpetuating factors of FSS can be subdivided in

three main domains, namely 'Hypochondria', 'Social and relational problems', and 'Symptom-related emotions and habits'. Sixteen clusters were identified at the lowest level of the hierarchy. The cluster 'Hypochondria' comprised the clusters 'Non-acceptance of the symptoms', 'Preoccupation with the symptoms', 'Excessive concerns about the symptoms', 'Somatic fixation', 'Excessive use of care facilities'. The cluster 'Social and relational problems' consisted of the clusters 'Dysfunctional interaction with friends and relatives', 'Bad relationship with healthcare professionals', 'Problems fulfilling roles' and 'Feeling too little acknowledgement of suffering or restrictions'. Finally, the cluster 'Symptom-related emotions and habits' consisted of the clusters 'Making unrealistic demands on yourself', 'Difficulty asking for help', 'Sense of shame and failure due to the symptoms', 'Frustration and despair regarding the symptoms', 'Difficulty perceiving and interpreting emotions and physical processes', 'Adverse physical factors or counterproductive lifestyle' and 'Avoidance behavior'. [Table 2](#) shows the names, number of statements, an example statement of the clusters, and their average importance rate.

4.4. Number of clusters

Ward linkage gave an indication of the number of clusters that represented the data best, but the exact number of clusters was guided by interpretation of the content of the given clusters. A cluster solution consisting of seventeen clusters resulted in subdivision of the cluster 'Dysfunctional interactions with friends and relatives' into two clusters, one consisting of only two items: 'Belief that friends and relatives will not or cannot help you' and 'Over-sensitivity to other people's opinions of the cause of the symptoms'. We considered this not to be a self-contained cluster, and therefore preferred a sixteen over a seventeen cluster solution. A cluster solution containing fifteen clusters resulted in a combination of the clusters 'Preoccupation with symptoms' and 'Non-acceptance of the symptoms', which we considered to be both meaningful and distinguishable clusters in the perpetuation of FSS. Therefore, we decided that a hierarchical cluster solution with sixteen clusters represented the data best.

4.5. Importance of perpetuating factors

Thirty-eight participants ([Table 1](#)) completed a questionnaire about the importance of the 99 factors. The mean importance scores ranged from 5.0, 'Deviant or restricted diet (e.g. gluten free, raw foods)', to 8.3, 'Letting the symptoms play a major part in your life' (see [Appendix B](#)). The range of average importance scores of the sixteen clusters was small (6.1–7.8). The clusters that were found most important were 'Frustration and despair regarding the symptoms' with an average score of 7.8 (SD = 0.8) and 'Preoccupation with the symptoms' with an average score of 7.7 (SD = 0.8). The clusters that were considered least important were 'Adverse physical factors or counterproductive lifestyle', 'Difficulty asking for help', and 'Sense of shame and failure towards the symptoms', which had average scores of 6.1 (SD 1.2), 6.2 (1.5) and 6.2 (SD 1.3), respectively.

4.6. Literature search

The meta-analysis, reviews and clinical studies found for the different clusters are presented in [Table 3](#). Proof for a relation between FSS and the following clusters was found in systematic reviews: 'Excessive concerns about the symptoms' [22–24], 'Avoidance behavior' [25,26], 'Frustration and despair' [23,27], 'Difficulty perceiving and interpreting emotions and physical processes' [28,29], 'Adverse physical factors or counterproductive lifestyle' [30–32], 'Dysfunctional interactions with friend and narratives' [33,34], 'Somatic fixation' [35], 'Excessive use of care facilities' [36], and 'Problems fulfilling roles' [37]. A systematic review was also performed for 'Bad relationship with healthcare professionals', however this review only retrieved four studies of which

Table 1
Characteristics of participants in the card sorting task or the importance questionnaire.

	Card sorting task (n = 61)	Importance questionnaire (n = 38)
Female	46 (75.4%)	29 (76.3%)
Age	45.7 (SD 10.6)	43.3 (SD 10.5)
Years of work experience	10.5 (SD 7.6)	9.8 (SD 6.7)
Number of patients treated each year ^a	53.5 (SD 47.0)	41.1 (SD 30.8)
Profession		
Psychologist	38 (62.3%)	28 (73.3%)
Psychiatrist	10 (16.4%)	6 (15.8%)
Physiotherapist	4 (6.6%)	1 (2.6%)
Psychomotoric therapist	3 (4.8%)	2 (5.2%)
Nurse	2 (3.2%)	0 (0.0%)
Rehabilitation therapist	1 (1.6%)	1 (2.6%)
General practitioner	1 (1.6%)	0 (0.0%)
Neurologist	1 (1.6%)	0 (0.0%)
Psychosomatic therapist	1 (1.6%)	0 (0.0%)
Online	23 (37.8%)	31 (81.6%)

^a The data entry of the general practitioner on number of patients was exceptionally high and therefore considered invalid.

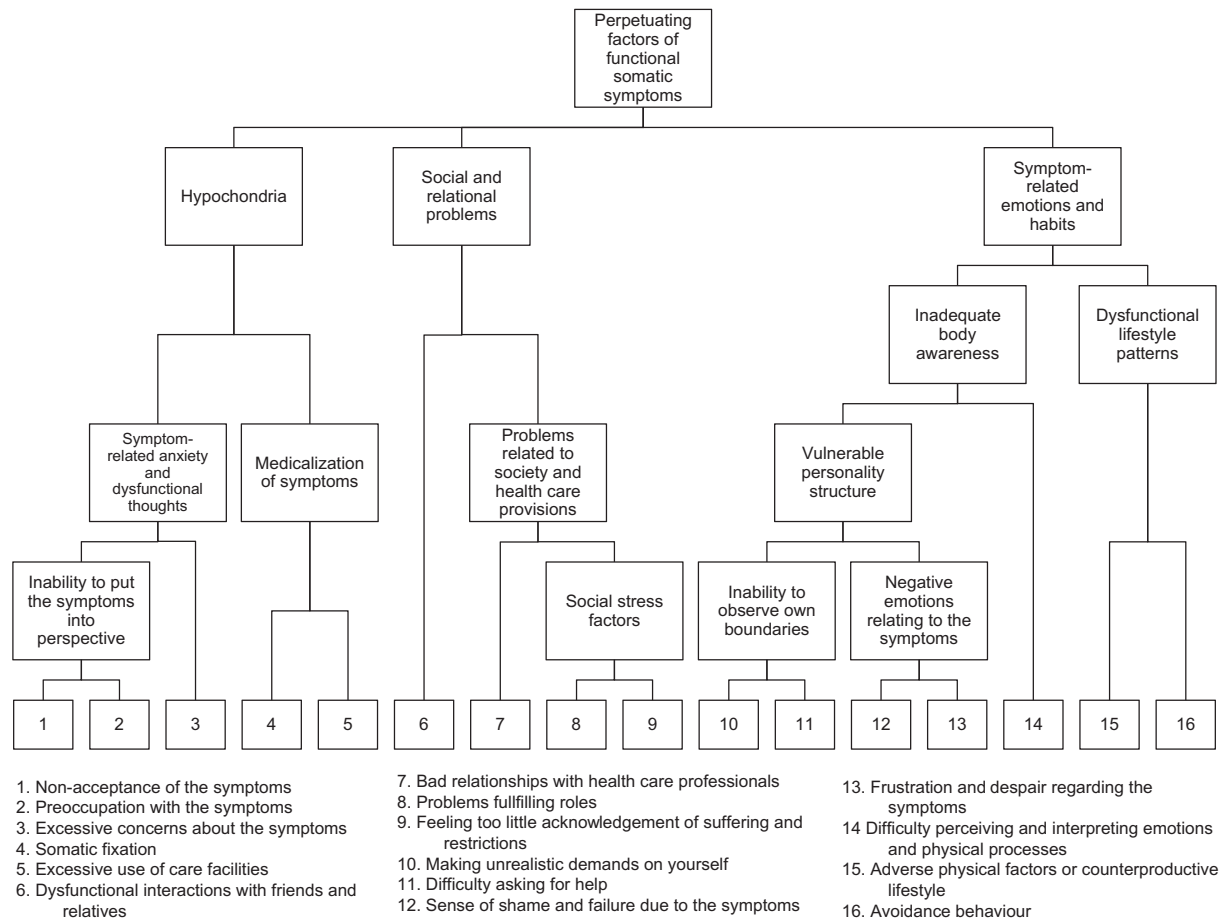


Fig. 1. Hierarchical structure of perpetuating factors of functional somatic symptoms identified with a hierarchical cluster analysis using Ward's statistics and squared Euclidian distances.

three were qualitative [38]. 'Preoccupation with symptoms' was mentioned in several narrative reviews [5,39,40], but a meta-analysis on selective bias to pain-related words showed little evidence that such a bias was specific for pain patients [41]. A role for the clusters 'Feeling too little acknowledgement of suffering and restrictions' [42] and 'Making unrealistic demands on yourself' [43–45] was suggested in narrative reviews. Clinical studies indicated that the clusters 'Difficulty asking

for help' [46,47], 'Sense of shame and failure' [48] and 'Non-acceptance of the symptoms' [45] are related to FSS.

5. Discussion

This concept mapping study revealed an overview of perpetuating factors of FSS according to clinicians. These factors consisted of the

Table 2
Main clusters in the perpetuation of FSS and mean importance score.

Cluster	Number of statements	Examples of statements	Importance ^a
Non-acceptance of the symptoms	5	Dwelling on what you used to be capable of, and what other people can still do	7.5 (1.0)
Preoccupation with the symptoms	6	Letting the symptoms play a major part in your life	7.7 (0.8)
Excessive concerns about the symptoms	10	Catastrophic interpretation of physical symptoms	7.5 (0.8)
Somatic fixation	8	Attributing the symptoms to a chance finding in the diagnostic process	7.2 (1.0)
Excessive use of care facilities	4	Seeking reassurance for somatic symptoms repeatedly	7.2 (0.9)
Dysfunctional interaction with friends and relatives	10	Friends and relatives who pay too much attention to the symptoms	6.8 (1.1)
Bad relationship with healthcare professionals	4	Anger towards healthcare professionals	6.8 (0.9)
Problems fulfilling roles	4	Lack of activities that boost confidence (e.g. work, hobbies) due to symptoms	7.2 (1.1)
Feeling too little acknowledgement of suffering or restrictions	6	Need to prove that you are ill to gain social recognition	7.1 (1.2)
Making unrealistic demands on yourself	6	Heightened sense of responsibility	7.2 (1.3)
Difficulty asking for help	4	Difficulty asking friends and relatives for help	6.2 (1.5)
Sense of shame or failure due to the symptoms	4	Feeling ashamed of having the symptoms	6.2 (1.3)
Frustration or despair regarding the symptoms	3	Frustration about the symptoms	7.8 (0.8)
Difficulty perceiving and interpreting emotions and physical processes	11	Paying insufficient attention to the emotions evoked by the symptoms	7.0 (1.1)
Adverse physical factors or counterproductive lifestyle	7	Insomnia (e.g. problems falling asleep, staying asleep)	6.1 (1.2)
Avoidance behaviour	7	Avoidance of physical exertion	7.5 (0.9)

^a Average score (and SD) of importance attributed to perpetuating factors in a specific cluster; scores could range from 1 (= not important at all) to 10 (= extremely important).

Table 3

Evidence in scientific literature for the relevance of the sixteen clusters of perpetuating factors as identified by experienced clinicians.

Cluster	Reviews	Type of study	Conclusion	
1	Non-acceptance of the symptoms	Van Damme et al., 2006 [4]	Cross-sectional observational study	Acceptance has a positive effect upon fatigue and psychological aspects of well-being. More specifically, acceptance is related to more emotional stability and less psychological distress, beyond the effects of demographic variables, and fatigue severity.
2	Preoccupation with the symptoms	Crombez et al., 2013 [5]	Meta-analysis	Individuals who experience chronic pain display an attentional bias towards pain-related words or pictures, but this bias was of a small effect size and did not differ from that in control groups
		Kirmayer et al., 1994 [2]	Narrative review	Absorption may make individuals more liable to focus attention on symptoms and more vulnerable to suggestions that induce illness anxiety.
		Rief and Broadbent, 2007 [6]	Narrative review	Modern neuroimaging techniques show that the expectation of symptoms leads to the activation of brain areas corresponding to symptom perception, while distraction from symptoms reduces brain activity in perception areas.
3	Excessive concerns about the symptoms	Knoop et al., 2010 [7]	Narrative review	The focusing on fatigue might be involved in the perpetuation of fatigue.
		Lukkahatai and Saligan, 2013 [8]	Systematic review	An association exists between catastrophizing and fatigue.
		Burke et al., 2015 [9]	Meta-analysis	Individuals with chronic pain are more likely to experience physically focussed psychological problems than other psychological problems. Fear of pain is intrinsically tied to the chronic pain experience.
		Edwards et al., 2006 [10]	Systematic review	Catastrophizing is positively related, in both cross-sectional and prospective studies across different musculoskeletal conditions, to the reported severity of pain, affective distress, muscle and joint tenderness, pain-related disability, poor outcomes of pain treatment, and, potentially, to inflammatory disease activity.
4	Somatic fixation	Douzenis and Seretis, 2013 [1]	Systematic review	Somatic attribution style in somatoform disorder has acceptable descriptive but insufficient predictive validity. This confirms that the overlap between somatic and psychological attributions is often substantial.
		Kirmayer et al., 1994 [2]	Narrative review	Attribution style can discriminate between somatoform disorder patients with and without comorbidity.
5	Excessive use of care facilities	Puri and Dimsdale, 2011 [3]	Systematic review	More proximate influences on the selective amplification of somatic symptoms include repressive style, somatic attributional style, and alexithymia; however, data in support of these factors are scant.
6	Dysfunctional interaction with friends and relatives	Leonard et al., 2006 [27]	Systematic review	While there is no standard criterion for high utilization, regardless of definition, evidence supports over-utilization, particularly in outpatient visits. However, no unique pattern of utilization exists that could identify somatizers within a broader group of high utilizers.
		Band et al., 2015 [28]	Systematic review	Overall, a consistent positive relationship was demonstrated between solicitous and distracting spouse responses and pain severity.
7	Bad relationship with healthcare professionals	Dhaliwal and Hunt, 2004 [29]	Systematic review	Significant other beliefs attributing patient responsibility for the onset and ongoing symptoms of chronic fatigue syndrome are associated with increased patient distress. Increased symptom severity, disability, and distress are also associated with both solicitous and negative significant other responses.
8	Problem fulfilling roles	Duenas et al., 2016 [30]	Narrative review	Some irritable bowel syndrome patients in primary care experience dissatisfaction and negative attitudes in general practitioner interactions.
		Annemans et al., 2009 [31]	Systematic review	In addition to the serious consequences on the patient's life, chronic pain has a severe detrimental effect on patients' social and family environment, as well as on health care services.
		Hakanson, 2014 [32]	Narrative review	The patient burden of fibromyalgia is very high in comparison with many other conditions.
9	Feeling too little acknowledgement of suffering and restrictions	Newton et al., 2013 [33]	Narrative review	The limitations of persons with irritable bowel syndrome manifested as lack of ability to move about freely, fulfill ambitions or commitments at work, maintain social activities, uphold or develop close and/or sexual relationships and parenting, and live a life with spontaneity.
		Clarke and Iphofen, 2005 [34]	Systematic review (but no studies retrieved)	The experience of stigma can occur in a number of ways. It may be through actual or perceived encounters with others; it can be through the use of psychologic explanations of pain; it can come through a perceived challenge to one's integrity and subsequently affect an individual's identity; and such stigma may be influenced by negative female stereotypes.
10	Making unrealistic demands on yourself	Abbey, 1993 [13]	Narrative review	There are no studies that look exclusively at the effects of having a health professional believe a patient's reported pain experience; however, within the literature there are several references to the need to examine this phenomenon.
		Hasenbring et al., 2014 [16]	Narrative review	Psychological and psychodynamic constructs such as depressive vulnerability in individuals with chronic fatigue syndrome depend upon achievement for the maintenance of self-esteem and euthymic mood, perfectionism, and helplessness.
				By using the avoidance-endurance model of pain, a concept is provided that elucidates a range of responses to pain, fear, and anxiety that mediate the transition from acute to chronic pain.

(continued on next page)

Table 3 (continued)

Cluster	Reviews	Type of study	Conclusion
	Van Damme and Kindermans, 2015 [17]	Narrative review	A self-regulation perspective does not consider avoidance and persistence behaviour to be intrinsically adaptive or maladaptive, but argues that their effects on disability and well-being rather depend on the goals underlying these behaviours.
11	Lackner and Gurtman, 2005 [18]	Cross-sectional observational in irritable bowel syndrome patients and healthy controls	The interpersonal profile of irritable bowel syndrome patients was characterized by difficulties with assertiveness and, to a lesser extent, social inhibition.
	Yücel et al., 2002 [19]	Cross-sectional observational in headache patients and healthy controls	Compared with healthy controls, the subjects with headache had significantly higher scores on measures of depression, automatic thoughts, and alexithymia, and lower scores on assertiveness.
12	Turner-Cobb et al., 2015 [12]	Cross-sectional observational study in patients and healthy controls	Significantly greater levels of shame, guilt, fear of negative evaluation, and mental defeat were observed in chronic pain patients compared to controls. In the pain group, self-conscious emotions significantly predicted affective pain intensity; only mental defeat was significantly related to disability.
13	Fernandez and Turk, 1995 [11]	Systematic review	Chronic pain patients experience anger but this may be underestimated because of denial.
	Burke et al., 2015 [9]	Meta-analysis	The chronic pain group reported experiencing significant problems in a range of psychological domains (depression, anxiety, somatization, anger/hostility, self-efficacy, self-esteem and general emotional functioning), with the largest effects observed for pain anxiety/concern and somatization; followed by anxiety and self-efficacy; and then depression, anger/hostility, self-esteem and general emotional functioning.
14	Waller and Scheidt, 2006 [20]	Systematic review	Overall, somatoform disorders are linked to a diminished capacity to consciously experience and differentiate affects and express them in an adequate or healthy way.
	De Gucht and Heiser, 2003 [21]	Systematic review	A small to moderate relationship was found between general alexithymia and somatic symptom reporting. The alexithymia dimension measuring difficulty in identifying feelings showed the strongest association with symptom reports.
15	Bjurstrom and Irwin, 2016 [24]	Systematic review	Whereas a consistent pattern of objective sleep disturbances is not identified, alterations of sleep continuity are commonly reported. Alterations of sleep architecture such as increases in light sleep or decreases in slow-wave sleep are less commonly reported and inconsistent.
	Jackson and Bruck, 2012 [25]	Systematic review	Polysomnographic and other objective measures of sleep have observed few differences in sleep parameters between patients with chronic fatigue syndrome and healthy controls, although some discrepancies do exist. This lack of significant objective differences contrasts with the common subjective complaints of disturbed and unrefreshed sleep by patients with chronic fatigue syndrome.
	Shephard, 2005 [26]	Narrative review	Overtraining, a negative energy balance, excessive physical or environmental stress, disorders of personality and affect, dysfunction of the hypothalamus-pituitary adrenal axis, hormonal imbalance, nutritional deficits, immune suppression or activation, and chronic infection have all been proposed as factors precipitating chronic fatigue syndrome, but none of these precipitants are observed consistently. Impairments of peak aerobic power and muscle strength, together with many functional disturbances, seem related to patient- or physician-imposed inactivity.
16	Nijs et al., 2013 [22]	Systematic review	Fear of movement and avoidance behaviour towards physical activity is highly prevalent in both the chronic fatigue syndrome and fibromyalgia population. In addition, it is related to various clinical characteristics of chronic fatigue syndrome and fibromyalgia, including symptom severity and self-reported quality of life and disability.
	Vlaeyen and Linton, 2000 [23]	Systematic review	Pain-related fear and avoidance appear to be an essential feature of the development of a chronic problem for a substantial number of patients with musculoskeletal pain.

Please note that this overview is non-exhaustive and meta-analyses on therapies for FSS that might influence these clusters are not presented in this table, but are mentioned in the discussion of the article.

overarching domains 'Hypochondria', 'Social and relational problems' and 'Symptom-related emotions and habits', which could be further subdivided into sixteen clusters. This hierarchical overview will help to screen for perpetuating factors in individual FSS patients, and subsequently will aid development of adequate personalized self-help support programs and more intensive forms of treatment.

All identified clusters have been addressed in the literature [5,22–47, 48–52]. Most evidence for the potential relevance of the clusters was found for: 'Excessive concerns about the symptoms', 'Avoidance

behavior', 'Frustration and despair', 'Difficulty perceiving and interpreting emotions and physical processes', 'Adverse physical factors or counterproductive lifestyle', 'Dysfunctional interactions with friend and narratives', 'Somatic fixation' and 'Excessive use of care facilities' and 'Problems fulfilling roles'. Least evidence was found for the relevance of the clusters: 'Sense of shame and failure', 'Difficulty asking for help', and 'Non-acceptance of the symptoms'. It should be noted that most research concerned cross-sectional and observational studies and it is therefore still unclear whether those factors are really

perpetuating FSS or are just co-occurring with FSS, or are predominantly consequences of FSS.

Randomized controlled trials are needed to test the alleged perpetuating role of the clusters. For example, the effectiveness of acceptance and commitment therapy and mindfulness-based therapies for FSS might indicate a role of 'Non-acceptance of the symptoms' and of 'Difficulty perceiving and interpreting emotions and physical processes'. The effectiveness of graded-exercise therapy might indicate perpetuating roles of 'Adverse physical factors or counterproductive lifestyle' or 'Avoidance behavior'. Effects of assertiveness training on FSS might indicate a role of 'Difficulty asking for help' and 'Making unrealistic demands on yourself'. The effectiveness of psychoeducation in treatment of FSS might indicate a role for 'Difficulty perceiving and interpreting emotions and physical processes', and 'Excessive use of care facilities'. The effectiveness of cognitive therapy might indicate a role of 'Excessive concerns about the symptoms', 'Somatic fixation' and 'Preoccupation with the symptoms'. These treatments have found to be beneficial for the treatment of FSS (e.g., 14), but the underlying mechanisms of these treatments should be identified to examine whether they truly influence the proposed perpetuating factors. Based on the identified clusters of the card sorting task, treatments that focus on patient-doctor interaction, interpersonal problems, feelings of shame, feelings of anger, or sense of too little recognitions for the symptoms might be helpful additions to those existing therapies.

Strong points of this study are that qualitative input of an extensive number of respondents was analyzed with quantitative methods to shed light on perpetuating factors of FSS and their hierarchical structure. Advanced cluster analysis techniques were performed, using Ward's statistics and squared Euclidian distances. Moreover, the clinicians who were interviewed and those who performed the card-sorting task were highly experienced covering a wide number of professions and treatment expertise.

With respect to external validity, it is important to note that the performers of the card sorting task were predominantly psychologists. Although we noticed during the interviews that psychologists had a differentiated, detailed and broad view on the persistence of FSS, including mostly psychologists may have colored the importance rates of the different statements. For example, physical factors, especially factors concerning food intake, were on average considered least important, although these factors were often mentioned in the interviews by medical doctors or physiotherapists. Another challenge to external validity is that the study was performed in one country and the design is inherently susceptible to subjective choices. However, our literature search revealed that all clusters identified with the card sorting task have been addressed in scientific literature. Nevertheless, we cannot make sure that some aspects are not covered by the sixteen clusters. Furthermore, although we had 61 participants of the card sorting task, which is considered ample [53], we noticed that the structure slightly changed by reanalyzing the data while excluding some participants. This was especially true for the grouping of clusters in domains, probably due to using squared Euclidian distances. Therefore, we focused our results and discussion mainly on the sixteen clusters. Further the overview of perpetuating factors focused mainly on factors that were treatable with self-help interventions. Although we believe that the use of this overview could be important for personalizing more intensive therapies as well, it should be kept in mind that not all factors (such as psychiatric comorbidity or attachment problems) are captured in the overview. Finally, although we attempted to systematically search the literature, due to practical reasons only a selection of reviews has been included.

To get another perspective on factors in perpetuation of FSS, the study could be repeated in a sample of patients with FSS. Although several of the domains found in our study were also found to be important by patients in previous qualitative studies [e.g., [22–27]], a replication of our study in patients might identify perpetuating factors of FSS that went unnoticed by clinicians. It should be noted that the card sort approach has been performed by both clinicians and patients in a previous

study on body-relatedness [54], and it turned out that patients' and clinicians' perspective on body-relatedness were quite similar.

Further, our research only focused on factors that perpetuate FSS, and thus on vulnerability factors. It would be interesting to also examine resilience factors for FSS [55]. Resilience factors of FSS might consist of positive emotions, optimism, acceptance of FSS, and social resilience [56]. These resilience factors might be important intervention targets for treatment of patients with FSS over and above perpetuating vulnerability factors [55].

The findings of the current study can be used to develop a screening tool which helps to systematically identify perpetuating factors in individual patients with FSS in clinical practice. This will facilitate choosing optimal treatment personalized to a particular patient. Especially, specific self-help interventions can be developed based upon the items and corresponding clusters found in the current study. It is still unclear how acceptable such self-help interventions will be to patients. Self-help support programs for FSS have recently been found to be effective, and drop-out in the groups who did receive and those who did not receive self-help interventions were comparable, which might be an indication of acceptability [57]. Another remaining question is whether personalized treatment of FSS is more efficient than non-personalized treatment. The concept mapping approach as applied in this study has been used before to develop a self-management program for patients with rheumatic diseases [58]. The data collection of the RCT is ongoing.

In conclusion, this study revealed an overview of factors involved in the perpetuation of FSS based on expert opinions, consisting of physical, cognitive, emotional and societal factors. This overview will aid future research on FSS and guide the development of personalized treatment in patients with FSS.

Disclosure

The authors do not declare any conflict of interest.

Acknowledgement

The authors are thankful for all clinicians who participated in this research. This study received no direct funding. AvG, JHH and JGMR were sponsored by a grant from the Dutch Healthcare Insurers Innovation Foundation during the conduct of the study. This funder had no role in the study design, collection, analysis, and interpretation of data, writing of the manuscript, or the decision to submit the manuscript for publication. The authors do not have any conflicts of interest.

Appendix A. Search strategy

Pubmed

(Somatoform Disorders[MeSH] OR Fatigue Syndrome, chronic[Mesh] OR Irritable Bowel Syndrome[MeSH] OR Somatoform Disorder*[tiab] OR Fatigue Syndrome[tiab] OR Irritable Bowel Syndrome[tiab] OR Fibromyalgia[tiab] OR CFS[tiab] OR IBS[tiab] OR psychosomatic disorder*[tiab] OR somatoform pain disorder*[tiab] OR somatisation disorder*[tiab] OR chronic headache[tiab] OR 'chronic limb pain'[tiab] OR 'chronic back pain'[tiab] OR 'chronic neck pain'[tiab] OR 'chronic musculoskeletal pain'[tiab] OR 'chronic abdominal pain'[tiab] OR 'chronic pain'[tiab] OR 'chronic fatigue'[tiab] OR functional somatic[tiab] OR unexplained symptom*[tiab] OR unexplained complaints[tiab] OR unexplained illness*[tiab] OR 'unexplained physical symptoms'[tiab] OR 'unexplained somatic symptoms'[tiab] OR 'unexplained physical complaints'[tiab] OR 'unexplained somatic complaints'[tiab] OR somatoform symptoms[tiab] OR psychosomatic symptom*[tiab] OR psychosomatic complaints[tiab] OR psychosomatic illness*[tiab] OR FSS[tiab] OR FSSs[tiab] OR MUS[tiab] OR MUPS[tiab] OR 'chronic benign pain'[tiab] OR functional pain[tiab] OR psychogenic pain[tiab] OR longstanding pain[tiab] OR idiopathic pain[tiab] OR

'recurrent abdominal pain'[tiab] OR RAP[tiab] OR FAP[tiab] OR 'functional abdominal pain'[tiab] OR 'functional gastrointestinal symptoms'[tiab] OR functional gastrointestinal disorder*[tiab] OR functional headache[tiab] OR 'non specific headache'[tiab] OR 'nonspecific headache'[tiab] OR tension headache[tiab] OR 'idiopathic musculoskeletal pain'[tiab] OR 'chronic widespread pain'[tiab] OR 'nonspecific musculoskeletal pain'[tiab] OR 'non specific musculoskeletal pain'[tiab] OR 'non cardiac chest pain'[tiab] OR 'noncardiac chest pain'[tiab] OR 'hyperventilation syndrome'[tiab] OR persistent fatigue*[tiab] OR prolonged fatigue*[tiab] OR somatization disorder[tiab] OR musculoskeletal complaints[tiab] OR 'Chronic abdominal complaints'[tiab] OR 'Chronic abdominal symptoms'[tiab] OR unexplained complaint*[tiab] OR somatoform pain[tiab] OR somatoform complaint[tiab] OR idiopathic headache[tiab] OR 'nonspecific musculoskeletal complaints'[tiab] OR 'non specific musculoskeletal complaints'[tiab] OR 'Nonspecific musculoskeletal symptoms'[tiab] OR 'Non specific musculoskeletal symptoms'[tiab] OR Nonspecific musculoskeletal disorder*[tiab] OR Non specific musculoskeletal disorder*[tiab] OR 'tension-type headache'[tiab] OR 'tension type headache'[tiab] OR 'tension-type headache'[tiab] OR 'recurrent headache'[tiab])

AND

'somatic attribution' OR
 'health care utilization' OR
 'acceptance' OR
 'attentional bias' or 'selective attention' OR
 'catastrophizing' OR
 'anger' OR
 'shame' or 'guilt' OR
 'perfectionism' OR
 'assertiveness' OR
 'alexithymia' OR
 'avoidance behavior' or 'avoidance behaviour'
 'sleep', 'diet' or 'physical condition'
 'social support' or 'significant other'
 'doctor' AND 'interaction or communication'
 'stress' or 'quality of life'
 'stigma'

Appendix B. Overview of 99 statements of perpetuating factors of functional somatic symptoms and their importance rating

Statements	Importance score [mean (SD)]
<i>Cluster: Non-acceptance of the symptoms</i>	
Belief that you have no influence on the cause of, and solution to, your symptoms	7.8 (1.6)
Belief that your perception is reality	7.3 (1.4)
The notion that you have a right to a healthy body without symptoms	6.6 (1.6)
Dwelling on what you used to be capable of, and what other people can still do	7.8 (1.4)
Inability to accept the symptoms	7.8 (1.2)
<i>Cluster: Preoccupation with the symptoms</i>	
Greater focus on your limitations than your possibilities	7.9 (1.4)
Being unaware of the moments when you do not experience the symptoms	7.4 (1.4)
No appreciation of the good things in life beyond health	7.1 (1.2)
Over-identification with the body and symptoms	8.1 (1.0)
Letting the symptoms play a major part in your life	8.2 (1.2)
Perception of body dominated by symptoms	7.5 (1.1)

Appendix B. (continued)

Statements	Importance score [mean (SD)]
('becoming' the symptoms)	
<i>Cluster: Excessive concerns about the symptoms</i>	
Fear of a serious underlying disorder	7.6 (1.5)
Fear that the symptoms will progress and become worse	7.3 (1.3)
Feeling no control over the symptoms	7.7 (1.4)
Lack of trust in your own body	7.5 (1.1)
Fear of movement	7.5 (1.3)
Belief that movement will exacerbate the symptoms	7.4 (1.4)
Worrying about the symptoms and the implications	7.4 (1.2)
Notion that you are unable to live with the symptoms	7.4 (1.4)
Belief that the symptoms will never go away	7.6 (1.3)
Catastrophic interpretation of physical symptoms	7.8 (1.1)
<i>Cluster: Somatic fixation</i>	
Belief that the symptoms are a sign of a physical injury	7.4 (1.5)
Belief that syndrome diagnoses point to an underlying disease process	7.3 (1.5)
Belief that the solution depends on finding the cause of the symptoms	7.7 (1.8)
Unreasonable expectations of medical diagnostics	7.2 (1.7)
Belief that the medical world can solve everything	7.4 (1.6)
Incorrectly attributing the symptoms to an explained comorbid disorder	6.4 (1.3)
Attributing the symptoms to a chance finding in the diagnostic process	5.9 (1.6)
Fixation on finding a somatic cause for the symptoms	8.0 (1.5)
<i>Cluster: Excessive use of care facilities</i>	
Excessive internet searching for information about diseases or symptoms	7.0 (1.3)
Inefficient, frequent consultations with regular and alternative healthcare professionals	7.2 (1.4)
A constant need for reassurance regarding the symptoms	7.2 (1.2)
Chronic use of medication and drugs to ease symptoms	7.2 (1.4)
<i>Cluster: Dysfunctional interaction with friends and relatives</i>	
Over-reliance on social network	6.9 (1.2)
Lack of social network	7.1 (1.5)
Demanding too much attention, support or reassurance from those around you	7.2 (1.1)
Not experiencing enough practical help from friends and relatives	6.3 (1.6)
Not experiencing enough emotional support from friends and relatives	7.0 (1.3)
Belief that friends and relatives will not or cannot help you	6.3 (1.9)
Friends and relatives who pay too much attention to the symptoms	7.0 (1.4)
Friends and relatives who regularly take over	7.1 (1.5)
Over-sensitivity to other people's opinions of the cause of the symptoms	6.1 (1.7)
Friends and relatives who play down the symptoms or think you are exaggerating	6.9 (1.7)
<i>Cluster: Bad relationship with healthcare professionals</i>	
The idea that healthcare professionals do not take you seriously	7.6 (1.5)
Lack of trust in healthcare professionals	7.2 (1.4)
Anger towards healthcare professionals	7.2 (1.3)
No longer seeing healthcare professionals	5.4 (2.0)
<i>Cluster: Problems fulfilling roles</i>	
Current psycho-social stress factors (e.g. debts, conflict at work)	7.4 (1.2)
Feeling the pressures of work	7.4 (1.3)
Adverse physical working conditions	6.5 (1.8)
Lack of activities that boost confidence (e.g. work, hobbies) due to symptoms	7.6 (1.3)
<i>Cluster: Feeling too little acknowledgement of suffering and restrictions</i>	
Lack of sympathy for the symptoms at work or school/college/university	7.2 (1.5)
Feeling let down by your employer	6.8 (1.6)
Feeling under pressure to return to work or resume studies	7.3 (1.5)
Material benefits of the symptoms (e.g. sickness benefits, damages for injury)	7.2 (1.8)
Immaterial benefits of the symptoms (e.g. attention from others, status of being ill)	7.3 (1.8)
Need to prove that you are ill to gain social recognition	7.0 (1.9)

Appendix B. (continued)

Statements	Importance score [mean (SD)]
<i>Cluster: Making unrealistic demands on yourself</i>	
Physical over-exertion	7.8 (1.8)
Making high demands on own body	7.5 (1.7)
Perfectionism	6.7 (1.5)
Heightened sense of responsibility	7.1 (1.5)
A negative self-image	6.8 (1.7)
Problems setting boundaries	7.4 (1.7)
<i>Cluster: Difficulty asking for help</i>	
Difficulty asking friends and relatives for help	6.8 (1.7)
Not wanting to bother other people with the symptoms	6.3 (1.7)
Hiding the symptoms from other people	5.9 (1.8)
Difficulty asking healthcare professionals for help	5.9 (1.8)
<i>Cluster: Sense of shame and failure due to the symptoms</i>	
A sense of failure about having developed the symptoms	6.3 (1.9)
A sense of failure about having to ask for help	6.1 (1.8)
Feeling ashamed of having the symptoms	6.0 (1.4)
Fear of losing face if the symptoms recede before a cause has been found	6.4 (1.6)
<i>Cluster: Frustration and despair regarding the symptoms</i>	
Desperation and despondency	7.9 (1.2)
Feeling powerless with regard to the symptoms	8.2 (1.1)
Frustration about the symptoms	7.4 (1.0)
<i>Cluster: Difficulty perceiving and interpreting emotions and physical processes</i>	
Difficulty expressing emotions	7.0 (1.5)
Difficulty dealing with other people's emotions	6.1 (2.1)
Failure to recognize own normal physical, emotional and stress reactions	7.7 (1.4)
Inability to feel whether the body is tense or relaxed	7.4 (1.4)
Inability to live in the here-and-now	6.7 (1.5)
Cognitive need for control (emphasis on thinking at the expense of feeling)	6.7 (1.7)
Paying insufficient attention to the emotions evoked by the symptoms	6.9 (1.7)
Heightened sensitivity to physical sensations	7.5 (1.1)
Inability to relax	7.4 (1.5)
Lack of understanding of anatomy and physiology	6.3 (1.9)
Failure to understand how own behaviour and thinking affect the symptoms	7.8 (1.2)
<i>Cluster: Adverse physical factors or counterproductive lifestyle</i>	
Disturbed circadian rhythm	6.6 (1.5)
Insomnia (e.g. problems falling asleep, staying asleep)	6.8 (1.5)
No physical stamina, decreased physical stamina	6.8 (1.9)
Restricted mobility	6.6 (1.5)
Obesity	5.8 (1.8)
Lack of regular eating pattern	5.5 (1.8)
Deviant or restricted diet (e.g. gluten-free, raw foods)	5.0 (1.6)
<i>Cluster: Avoidance behaviour</i>	
Avoidance of social activities	7.6 (1.2)
Stopping work or education	7.3 (1.7)
Spending too little time on pleasant or relaxing activities	7.5 (1.3)
All-or-nothing pattern of behaviour	8.1 (1.3)
Avoidance of physical exertion	7.8 (1.3)
Avoidance of mentally challenging activities (e.g. reading, crosswords)	6.9 (1.8)
Dysfunctional behaviour to ease symptoms (e.g. scratching, bad posture)	7.0 (1.4)

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