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"Functional somatic syndromes, one or many?": An answer by cluster analysis

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ABSTRACT

Objective: The aim of the present study was to address the lumper-splitter discussion on functional somatic syndromes by applying *k*-means cluster analyses on a heterogeneous sample of persons with unexplained somatic complaints. In favor of the lumper-side of the debate, clusters should differ only on the overall severity of the somatic complaints that were assessed. According to the splitters view, clusters should differ in symptom-specific patterns.
Methods: Three-hundred ninety four subjects with functional somatic symptoms were clustered based on their scores on 47 somatic symptoms. Three cluster solutions ($k = 2,3$, and 4 clusters) were compared on overall symptom severity, symptom patterns, and psychological distress.
Results: Results showed that in all three solutions the clusters were defined by increasing total symptom scores and increasing psychological distress. Cluster-specific symptom patterns where evident only when more clusters (three or four) were allowed. The best fit index was found for a 2-cluster solution.
Conclusion: The finding of symptom specific patterns in clusters which could not be differentiated on overall symptom severity is in favor of the splitters' view. The finding that all other clusters could be discriminated on overall symptom severity and that the 2-cluster solution had the best fit is in favor of the lumpers' view.

Introduction

When a person presents with somatic symptoms that cannot (fully) be explained by a known organic pathology, these symptoms will be labeled 'medically unexplained' or 'functional'. Often, more than one symptom is present and certain constellations of symptoms give way to a diagnosis of a specific functional somatic (FS) syndrome like for example chronic fatigue syndrome, fibromyalgia, or irritable bowel syndrome, with specific diagnostic criteria for each syndrome [1–3]. These FS syndromes have a high prevalence in our Western society [4,5].

The use of FS syndromes to diagnose persons with FS symptoms has been the topic of debate. The so-called splitters side of the debate defends the usefulness and even necessity to discriminate between syndromes as separate diagnostic categories [6,7]. Lumpers on the other hand argue that all of the syndromes represent one underlying common basic syndrome [8,9]. Arguments in favor of the latter position are as follows: a) the extensive overlap in core symptoms (e.g., fatigue, diffuse pain, general malaise); b) the fact that patients meeting criteria for one syndrome often meet criteria for other syndromes as well [8,9]; c) patients with different syndromes share non-symptom characteristics, like a history of stressful life events or a traumatic history [10,11]; and d) all syndromes share common psychiatric comorbidities (mainly anxiety disorders). Splitters argue that these arguments do not apply to all patients, and can thus not sufficiently explain the diversity and specificity of the syndromes. More recently, it has been suggested that both sides are true in that there is commonality as well as heterogeneity between (and within) FS syndromes in both onset-related factors and psychosocial and physiological patient characteristics [12].

Attempts have been made to solve the "splitters versus lumpers" debate on FS syndromes by statistical techniques such as principal component analysis that group FS symptoms to find specificities [13–15] or by latent class analyses to find communalities [16]. Some of the factor analytic studies have demonstrated multiple factor solutions with identifiable symptom groups per factor (e.g. gastrointestinal, musculoskeletal, cardiopulmonary) [13–16]. However, the symptom groups were found to differ between studies and the factors were inter-correlated [14,16], the most obvious reason being that patients often present symptoms from multiple factors. Therefore, grouping of symptoms across subjects may not be the appropriate statistical approach to solve the lumpers–splitters debate.

Of more relevance to the diagnosis of FS syndromes are techniques that try to categorize subjects in separate groups on the basis of the unique pattern of their symptoms. To date, only three studies have used this approach. Fink et al. [14] used latent class analyses to identify groups in their sample. These analyses yielded solutions with either two or three classes. In both results, classes could be distinguished by the *number* of symptoms and not by the *type* of symptoms. Gara et al. used hierarchical class analysis with a priori grouping of symptoms and found 11 patient clusters [17]. The clusters found in this study could be defined by "no symptom presentation", "presentation of one group of symptoms", "some groups of symptoms", or "all groups of symptoms". This partly

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confirms the findings by Fink et al. that persons can be clustered based on the number of symptoms (favoring a lumper's position), but also points to the fact that for a number of patients the symptoms that are experienced belong to one symptom group only (favoring a splitter's position). Finally, Kato et al. used latent class analysis of a sample of twins which yielded a five-cluster solution, with the clusters being different both in count of symptoms and in type of symptoms [18]; also supporting both the lumpers' and the splitters' side of the argument. Thus, these studies provide support for both sides of the argument, possibly implying that FS symptoms should be viewed from both a lumper's and a splitter's perspective.

Several methodological choices might have influenced the results in these studies. First, symptom presentation was dichotomized in most studies, thereby not taking into account symptom severity. This leads to giving an equal weight to vague side symptoms of a main symptom as to the main symptom itself. Second, the presence of physical symptoms was assessed over a relatively long retrospective period, either during the last two years [14] or during lifetime [17,18]. Symptom reports may therefore be strongly confounded by recall biases [19]. Third, predefined symptom groups were used in some studies [14,17], although the use of symptom groups is not strongly supported by factor analyses of the symptoms. Fourth, in two of the studies the data of healthy persons were combined with the data of patients in the same analyses [14,18]. As a result, the strongest differences within the sample are between the presence of *no* symptoms (i.e. the healthy persons) and the presence of *any* symptom (i.e. all the persons presenting with FS symptoms). This large difference between healthy subjects and patients may have masked a fine-grained cluster solution within patients. Based on these limitations it may be argued that clustering patients with FS symptoms needs further elaboration.

It is striking that the number of clusters found in the previous studies differs strongly, ranging from two to eleven. The number of clusters to search for is a matter of choice, and thus the subtlety of the solution (which depends on the number of clusters within the solution) is a consequence of this a-priori choice. Examining multiple cluster solutions within one sample could give some insight into the effects of the number of clusters on cluster structure.

In the current study, a sample of subjects with heterogeneous self-reported FS complaints was clustered on self-reported severity of 47 symptoms in the past seven days. The aim was to address the lumper–splitter discussion by examining cluster solutions on symptom severity and symptom patterns. In favor of the lumper-side of the debate, clusters should differ only on the overall severity of the complaints that were assessed. In contrast, according to the splitter-view, clusters should differ in symptom-specific patterns (i.e., syndromes). We chose to use a *k*-means clustering technique that allows for multiple cluster solutions as this technique allows for setting the number of clusters in a solution a priori and assigns each person into one cluster only (as opposed to other much used cluster techniques, such as latent class analysis). This way, it was possible to examine whether an a priori choice in the number of clusters in the solution would influence the explanation the cluster solution offers for the lumper–splitter debate. For further interpretation of the cluster solutions, the solutions were examined on between-cluster differences in total symptom scores, cluster-specific symptom patterns, and psychological distress. Psychological distress was included to incorporate the lumpers' position that the number of symptoms would be a function of the level of anxiety and depression.

Methods

Subjects

The source population consisted of subjects with heterogeneous FS complaints. Eligible participants were recruited through the internet, by placing links to the questionnaire on FS syndrome patient sites (i.e., for patients with chronic fatigue syndrome; fibromyalgia; irritable bowel syndrome; hyperventilation syndrome; and unexplained chronic pain). This way, it was ensured that only persons who consider themselves to have FS symptoms were exposed to the call. Only respondents who completed the survey were retained in the sample. The survey was started 653 times and completed 466 times. Fourteen respondents completed the survey twice and for these persons the second response was deleted, resulting in a sample of 452 unique respondents. Two respondents were younger than 18 years of age at the time of responding and were deleted from the dataset. After exclusion of respondents who reported having used soft- or hard drugs in the last week (n = 5), having either an autoimmune disorder (n = 12), thyroid disorder (n = 29), or a disorder which leads to severe pain complaints (hernia, scoliosis, spondylosis, arthritis; n = 10), a sample of 394 respondents remained. Mean age at the time of responding was 48.4 years (range: 18–84) and the majority of the respondents was female (76.1%).

Procedure

The link on FS syndrome patient sites led to a homepage on which information regarding the study was posted. Persons who agreed to participate could click on a questionnaire-link on the homepage which automatically directed them to the information letter, entailing information about purpose and length of the questionnaire and storage of information. Informed consent was obtained for all respondents before entering the questionnaire. Respondents could give informed consent by checking the box below the information letter that said "I have read the information and agree to participate in this study". The questionnaire was presented using NetQuestionnaires, version 6.5.

Measures

General descriptive information was obtained for gender, age, education, body length and body mass. Also, items were included concerning use of recreational drugs in the last week and presence of a chronic disease (reportedly diagnosed by a physician). It was also assessed whether a diagnosis of a FS syndrome was made in the past.

Somatic complaints were assessed with a 47-item symptom list specifically created for the current study (see Appendix). The list was primarily based on the Bodily Sensations Questionnaires [20] to which additional symptoms were added from several other somatic symptoms lists. Respondents could indicate for each symptom to what extent they had experienced this symptom in the last seven days on a five-point Likert scale (1 = not; 2 = a little; 3 = quite a bit; 4 = quite a lot; 5 = highly). The list included four gastrointestinal symptoms, six cardiac symptoms, five respiratory symptoms, six physical fatigue symptoms, six musculoskeletal symptoms, six cognitive symptoms, and fourteen 'other' symptoms.

The Dutch translation of the Hospital Anxiety and Depression Scale (HADS) [21] was used to assess psychological distress. The questionnaire contains 14 items on emotional state during the past week which have to be answered on a four-point scale. Because somatic symptoms of anxiety and depression do overlap with somatic symptoms of a disease (or with FS symptoms), regular mood questionnaires are often not valid in a somatic population. The HADS is specifically designed for assessment of anxiety and depression levels in persons with somatic symptoms. Reliability of the Dutch translation of the questionnaire is acceptable with cronbach's coefficient alpha 0.81–0.84 for the anxiety subscale and 0.71–0.86 for the depression subscale [21].

Cluster analyses

Respondents were clustered on their scores on the 47 somatic symptoms with a *k*-means cluster analysis, using SPSS for Windows, version 20.0.0. The *k*-means cluster analysis is an iterative partitioning method in which the number of desired clusters is set a-priori. After an initial