Author’s response to reviews

Title: Patients' experiences of living with medically unexplained symptoms (MUS): A qualitative study.

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Author’s response to reviews:

The cover letter to the Editor with responses to both reviews has been included as a supplementary file.

RESPONSES TO REVIEWER 2

Main criticisms: There are some problems with the science and also some problems with the presentation, which I both will elaborate in my comments to the authors. Furthermore, I have some problems to see what this actual study adds to existing knowledge on this theme. Therefore, I would encourage the authors to strive for a more clear answer to the question: what does this study add? This could be done by some restructuring of the Discussion-part, I think.

Thank you very much for this valuable comment. We have restructured the Discussion section highlighting our main findings and addressing other research studies. The highlights include:

(1) The awareness of the psychological origin of the symptoms may help some patients to accept their symptoms, learn to live with them and to some extent control them;
(2) The majority of our patients did not reveal any expectations regarding medical services, but many admitted to visiting psychologists and psychiatrists privately;
(3) A video-filmed consultation with a follow-up analysis with the GP could turn out to be an important strategy of GP to treat patients with MUS, especially the most difficult group of chronic patients.
Major comments:

Overall, qualitative methods are appropriate to address the research questions being asked. However there are several methodological issues, which require clarification.

1. Reflexivity: The authors are almost "invisible" in the text and this is a major limitation, I think. A basic principle in qualitative research is to describe preconceptions as the research itself never can be "objective". What background and perspectives do the authors have?

Thank you for this remark. We agree with it. We have revised the Methods section by adding the following statements and our initials whenever appropriate:

"The patients agreed to participate in the study and to be filmed on condition that they would remain fully anonymous and would be interviewed by the doctor they had known well and whom they trusted. They refused to be interviewed by anyone else."

"The interviews were conducted by the second author (SC), according to the topic guide derived from the literature on MUS [30], shown in Table 3. To ensure the patients' understanding, the interview questions were pilot tested with two patients with MUS in 2014, before the project started."

"The interview data were subsequently transcribed verbatim by two research assistants, verified by one of the authors (AS), and coded using NVivo 10. The method applied aimed to establish a straight descriptive summary of the informational contents of the data [31,32]. The transcriptions were analyzed independently by the two authors, representing different disciplinary backgrounds: linguistics and health communication (AS) and primary care (SCz)."

2. Choice of method: I would encourage the authors to describe their rationale by choosing video-filmed interviews. Filming is more intrusive for the participants than an individual interview, and the method chosen for analysis is based on transcripts and not non-verbal communications, is it not?

Yes, that is right. This study is part of a larger ongoing research project on the role of MUS patients’ verbal and nonverbal behaviours in doctor-patient communication in a Polish primary care setting (the statement included in the Background section). The qualitative content analysis of the interviews was the first step of the research project and aimed to explore the patients' experiences of living with MUS. The other objectives of the study include the analysis of
narrative structure, self-touching behaviours and gestures, using ELAN software, for which video-filming was essential.

3. Theoretical framework: Again, "what kind of glasses" did the researchers wear during the analysis process is unclear. It does not seem like they have used an established theory to support their analysis. That is a limitation. What did they expect to find? What did they not expect to find? These conceptions will influence during the analysis process irrespectively of which method of analysis is being chosen.

The aim of the study was to understand and explore Polish patients' perspectives on living with MUS. To achieve the goal we conducted semi-structured interviews, based on a topic guide derived from the literature (Olde Hartman et al. 2013), which addressed five main dimensions of MUS: somatic, cognitive, emotional, behavioural, and social. The prompts were to help us to find the answer to the research question: What are Polish patients' experiences of living with MUS? We did not expect to find that most patients would stress how important the realization of the psychological explanation of symptoms was for them and that it enabled them to cope better with the symptoms.

The theoretical framework underpinning the study was content analysis, which aims to systematically organize data into a structured format (COREQ). In particular, we drew on the method presented in Graneheim and Lundman (2004) and in Sandelowski (2000), which aims to establish a straight descriptive summary of the informational contents of the data. The method assumes that every text has multiple meanings which should be described and interpreted. Yet, the level of interpretation is low-inference, if compared to Grounded Theory. Content analysis is a data-driven approach, thus the themes were derived from the data. Qualitative descriptive studies tend to draw from the general tenets of naturalistic inquiry, which implies only a commitment to studying something in its natural state, or as it is, to the extent that this is possible, without any a priori commitment to any one theoretical view of a target phenomenon (Sandelowski 2000).

4. Discussion of the method: This is the weakest part of this study, I think.

First, as already mentioned, there is a lack of focus on reflexivity. Second, it is not correct to state that recruitment from more than one practice would have enhanced the external validity of this study: I am sure that the authors know that their choice of method never could cover all relevant patient-experiences from living with MUS irrespectively of N participants.
Thank you for this remark. We agree with this comment and we have revised the Discussion section by adding the following sentences in Strengths and Limitation subsection: "The patients in our study were recruited from a single GP practice, and most of them were interviewed by their own GP. This may have introduced a bias in the sample reflecting experiences from a selected patient group and a special doctor-patient relationship. On the other hand, this allowed us to explore personal and sensitive themes and elicit often intimate accounts of the patients' experimentum of MUS, which they would not have revealed to anyone else. It should also be noted that all patients in our study were diagnosed with F45, thus were at a later stage of their illness, which may have possibly restricted the variation of our findings. Yet, the selection of the group seems justified and shows that experiences of living with MUS differ depending on the stage of the diagnostic and treatment process and the level of insight the patients gained."

5. Third, the last sentence does not give any meaning at all - it is not a limitation that some of the findings are consistent with previously studies on MUS!

We have removed the sentence.

6. Fourth, there is a lack of discussion of internal validity.

We have revised the Discussion section and presented our findings against other research studies. We have also added Strenghts and Limitation subsection addressing potential biases. Yet, we think that the criterion of internal validity is not the essence of qualitative research as it is in quantitative studies.

7. Discussion of results: Needs to be re-structured. For me, the findings that the participants had realized that their symptoms were "psychological" are surprising. Also that they had to visits psychologists or psychiatrists private. This should be further discussed as it opposes findings from others studies on patients with MUS.

Thank you for this valuable suggestion. We have re-structured the Discussion section, highlighting our main findings and discussing them against research findings from other studies.
8. Furthermore, the finding that the participants seemed to avoid doctors contrasts the authors' claims in the Background, namely that 25% of all consultations in general practice are about MUS. This need to be highlighted in some way. Either by the use of another reference about prevalence in PHC (which have operationalized MUS in a more chronic/disabling way) or by highlighting this contrast in their Discussion.

We have addressed this issue indicating the following: "Although this seems to contradict a common perception of patients with MUS as high utilisers, it may be possibly explained by the sample reflecting patients who have already been through a long diagnostic and treatment process, and are thus at a later stage of their illness [4,5]."

Minor comments:

Background:

1. I would suggest the authors to be consistent in referring to patients with MUS as in the first sentence throughout the whole manuscript, instead of mix the use of patients with MUS and MUS patients. This may seem pedantic, but for me the latter has some negative connotations /may be more stigmatizing.

Thank you for bringing this to our attention. We agree with it and we have changed the reference according to the suggestion throughout the manuscript.

2. The authors have operationalized patients with MUS in this way: diagnose F45 + at least six months timespan suffering from their symptoms. Is this correct? Isn’t so that according to ICD-10, the diagnosis F45 is used for symptoms that cause clinically significant distress or impairment in social, occupational, or other areas of functioning - and have lasted for several years? Or am I wrong?
Thank you for the remark. We have checked it and corrected it. All patients in our study were diagnosed with distinctive somatoform disorders (F45), and complained about the symptoms for at least 2 years.

3. Statement: There are no satisfactory scientific theories explaining the outbreak of such symptoms. Isn’t that misleading to claim? There are several theories that try to unexplain the unexplained symptoms.

Yes, we agree with this remark. We revised the statement in the following way, adding some references: "The existing scientific theories explaining the outbreak of medically unexplained symptoms such as, for example, somatization, which assumes the presence of bodily symptoms as indirect markers of psychological distress, or somatosensory amplification, explaining MUS as a result of stress-related physiological arousal, are not fully satisfactory or definite [6, 8, 11]"

4. Statement: MUS cause heavy financial burden. This is substantiated by the use of two references which I don’t think actually shows any costs of MUS. Need to be changed.

Following the suggestion, we have changed the references.

Methods

1. To diminish the selection bias all patients were asked to fill in 4 DSQ. Why? Those patients who scored low in all dimensions were excluded. What is the rationale for this?

The selection of 16 patients with high somatization was dictated by further objectives of the research project, most notably the analysis of the patients' non-verbal behaviours. However, we have decided to include and analyze the remaining four transcriptions. We coded them in NVivo following the same method to check whether they introduce new perspectives. The analysis did not actually reveal any differences between the patients' accounts. We have added minor details in the Results section to reflect the other four participants' views. Yet, essentially, their experiences overlapped with the other patients' accounts.
2. A clarification is needed about the excluded patients. 20 were recruited, but 16 were filmed. What happened to the four remaining then? Did I understand it correct that one did not want to be filmed, and the other remaining three scored low on all dimensions of 4 DSQ? I am not sure.

Twenty patients were recruited and all were filmed, only one refused to participate in the study. Twenty video-filmed consultations were transcribed verbatim. Patients were also asked to fill in the 4DSQ test. The 4DSQ test results of 20 patients are presented in Table 2. We have added the following explanation in the Methods section: "Some patients scored low on the somatization dimension, which indicates that they did not present any symptoms in the week when the interview was conducted, which, in turn, may suggest they coped well with the symptoms. Much as the 4DSQ test results were relevant for the subsequent stages of the research project, which involved the selection of patients with high somatization to explore their nonverbal behaviors, they did not, however, affect the present study and qualitative content analysis of the interviews."

3. I do not understand the rationale by describing that "three patients suffered from chronic conditions: ......". Does this mean that the rest (thirteen) did not suffer from any other comorbid illnesses or diseases? How was this information gathered?

Information has been gathered based on medical records accessible in the GP practice and only three patients suffered from chronic conditions.

4. It is only said that the interview questions were pilot tested, but how was this actual done?

We have added the following information on pilot testing: "To ensure the patients' understanding, the interview questions were pilot tested with two patients with MUS in 2014, before the project started."

5. I do not understand the following sentence: "After switching off the cameras most patients agreed that combining the interview and video-filming helped to understand better their medical problems". Does this mean that the participants found that the video filming itself had an additive effect? Why and how? Needs some clarification.
The patients analysed the videos with their GP after the interviews had finished. We hope we have clarified this in the Methods section by adding the following statements: "After switching off the cameras, the patients analysed the videos with the GP. Most patients agreed that combining the interview and video filming helped them to understand the relationship between what they experienced, their emotions and their symptoms. It can be stated that the patients found that the video registration had an additive effect to control their own behaviour, including their symptoms. They did not visit their GP for the next three months, and did not change the primary care setting."

Results
1. I would suggest more stringence when it comes to the quantification. Personally, I find a few/many/most etc. better than the use of distinct number of participants.

We agree with this comment and revised the manuscript accordingly.

2. The result in line 51: does not this belong more to the foregoing result about coping/awareness of their psychological origin?

We disagree with it.

Discussion
1. The use of references 16, 17 and 18 in the beginning of the discussion-part do not make sense as the authors there sum up findings from their study. I would recommend to omit these references here.

We have omitted these references.
2. It may be confusing for readers with the new acronym MUPS instead of MUS. In this research field, without any consensus on terminology, I think it is OK for the authors to claim that both reference 17 + 23 are about patients with MUS.

Thank you. We agree with the remark and corrected the sentence.

3. Statement: There are no studies on MUS patients’articulation of emotions. Is that true? Maybe this statement should be less bastant?

We have removed the sentence altogether.

4. Statement: Our findings seem to prove that patients with MUS can be offered an audiovisuel interview combined with a follow-up analysis with the GP to manage MUS. First, this is not proven by your study! Change wording.

Thank you for this remark. We have corrected the statement.

5. Second, the authors should use video-film instead of a the new term audiovisual interview.

We agree with this remark and we have corrected the statement.

6. Third, the authors should use patients with MUS instead of only MUS.

Following the suggestion, we have corrected it.