

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	WORKING-AGE ADULTS' PERSPECTIVES ON LIVING WITH PERSISTENT POSTURAL-PERCEPTUAL DIZZINESS: A QUALITATIVE EXPLORATORY STUDY
AUTHORS	Sezier, Ann; Saywell, Nicola; Terry, Gareth; Taylor, Denise; Kayes, Nicola

VERSION 1 - REVIEW

REVIEWER	Andreas Sprenger University Luebeck, Department of Neurology, Luebeck, Germany
REVIEW RETURNED	13-Jul-2018

GENERAL COMMENTS	<p>Persistent postural-perceptual dizziness (PPPD) is a quite common among somatoform dizziness patients. The study presented by the authors sheds some light on the social and psychological aspects of PPPD using Thematic Analysis method. In clinical practise it would be helpful if patients, non-patients and health insurances would accept the notion that socio-psychotherapeutic treatment is helpful and essential. Therefore the topic of the study is important to enhance the treatment of patients with dizziness. The introduction is concise, the methods are described appropriately. In the results some information is missing. Furthermore I have some comments on the discussion.</p> <p>Major points</p> <p>Results</p> <p>1) It would be helpful to see results of the clinical examination of the patients, e.g. quantitative head impulse test (VOR gain), oVEMPs, cVEMPs, caloric irrigation results, posturography, subjective visual vertical, etc. I do trust in the clinical expertise of the authors but this might be helpful for reader to build an own image of the patients.</p> <p>2) Qualitative research is not quite common in the vertigo community. I know that qualitative analysis is a hard job and time consuming. The authors might think about adding (as a supplement?) an example of analysing the interview, e.g. of one sentence. I appreciate the frequent examples of patients' statements but this does not reflect the amount of work that has been done. My idea is to convince quantitative vertigo researchers that the reported information is valid.</p> <p>Discussion</p> <p>1) 23 subjects did not participate in the study. The authors might speculate why there was such a large rate of non-responders.</p> <p>2) In the last para of the discussion the authors shortly mention possible effective treatment of PPPD. The authors should think about an enlargement of the points because it is about the impact of the study on this topic. The interline message is that the authors</p>
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	<p>have some ideas about changing the current treatment of PPPD. For me it would be worth to show them point by point to increase the discussion on socio-psychologic support for the patients.</p> <p>Minor points Page 11, line 20: This sentence of Ethan is cited twice. The authors might refer to the first citation or should point out why it is important to cite this sentence twice.</p>
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REVIEWER	Floris L Wuyts University of Antwerp, Belgium
REVIEW RETURNED	24-Aug-2018

GENERAL COMMENTS	<p>The concept of elaborating on the experiences suffered by PPPD patients is very valuable. It adds to the way history taking can be refined. Particularly the stress and anxiety that is present among the patients is to be studied further on.</p> <p>My main concern about this paper, which I think deserves to be published, is that the description given by the patients with PPPD could as well be given by patients with for example Mal de Debarquement Syndrome, bilateral vestibulopathy, vestibular migraine or vestibular paroxysmia. These vestibular disorders are unfortunately very much unknown to the clinicians and cause very similar sensations like the three themes that are described in this paper.</p> <p>To my opinion, and based on my experience in a specialized vestibular clinic, many patients complain of very similar sensations as described in the paper. Little attention has been given to these descriptions so far in the literature and therefor I think this paper is very valuable. But it should be elaborated that these themes are not only for PPPD. Reading the patients' testimonies as described in the book Dizzy Me, by T. Stadsbader and F. Wuyts, Kugler Publications, 2017 corroborates this. If the authors are convinced that this is not the case, then they should interview also a similar group of patients with for example Mal de Debarquement Syndrome, to show that the described themes are unique for PPPD. I'm convinced that this is however not the case and that a huge overlap exists. But that should not be a big issue for the publication. It should just be clearly mentioned that the themes are not unique for PPPD. To be clear: Neuritis, BPPV or Meniere's disease may give indeed different themes than PPPD, although fear and anxiety is also very prominent in Meniere disease patients, not knowing when the next attack will come, thus e.g. restricting social life.</p> <p>It may be helpful to interview also the other types of patients and extract the typical themes for the different disorders. This way, this approach could help differentiate the different diagnoses, when added with for example a systematic history taking such as the SO STONED method, published by Wuyts, Maes et al, Frontiers of Surgery, 2016.</p> <p>Specific comments:</p> <ul style="list-style-type: none"> - You should refer to table 1, and thus the latest Barany criteria, already in the beginning of the manuscript where you describe PPPD. - In the data analysis it is not clearly stated that all subjects were asked the same questions.
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	<p>- The method of describing the sentences by the patients is pretty confusing. The identification (.) suggests pauses in speech. However, even with introducing pauses, these sentences make not so much sense in certain cases, and thus confusing.</p> <p>- Regarding the aspect of the 'invisibility' of PPPD, this can be stated for almost all vestibular disorders. Even worse, when walking like a drunk, it looks as if somebody has had too much alcohol early in the morning, leading to shame and embarrassment.</p> <p>- The impact on work of the disorder can be likewise difficult to cope with for episodic disorders too, next to the chronic ones.</p> <p>- The 'ignorance' or not being well informed of the clinical staff is also the case for several other disorders, such as MdDS, Vestibular paroxysmia, Vestibular Migraine, Superior Canal dehiscence (which has as a key symptom that the patient hears his or her own eyeballs roll!!! – "I'm I crazy?"). It is true that most of these disorders were not part of the education of all clinical staff that graduated before 2012 approximately, except perhaps for SCD which is ±10 year older as clinical diagnose. Thus the great majority of clinicians that is older than roughly 40 years have not learned about these diagnoses. This contributes to the frustration of the patients not being recognized with their disease, neither by the health professionals, neither by their surrounding colleagues, family and friends.</p> <p>Good luck with the revision</p>
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REVIEWER	Brayan V. Seixas Fielding School of Public Health University of California, Los Angeles USA
REVIEW RETURNED	09-Dec-2018

GENERAL COMMENTS	<p>The paper addresses a relevant gap in the literature and utilizes an appropriate methodological design to do so. But it would be beneficial for the reader to fully understand the work done and its limitations if the following changes were made:</p> <p>1) In the results section of the abstract, only plain codes emerged from the work are presented. I recommend the inclusion of a brief contextualizing narrative (a few sentences) connecting these codes, apart from the implications already discussed in the conclusion section of the abstract.</p> <p>2) In regard to methods, it is stated that a single author carried out the actual coding process but in dialogue with the other authors. Is it the same of having multiple coders? What is the difference between having multiple coders and having a single coder that develops her work discussing the findings with colleagues? This point would be welcomed in the discussion section.</p> <p>3) Participants were selected on the basis of a single clinic. How does that impact on the "generalizability" (given the applicability of this concept within the realm of qualitative studies) of findings for PPPD patients outside this community? How can we assure that the problems of these patients are similar to the overall population of PPPD patients in New Zealand. It would be enlightening to have authors' views on that in the discussion section too.</p> <p>It is important to highlight that these comments are mere suggestions. The reviewer is satisfied with the current version for publication, but strongly encourages these changes.</p>
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VERSION 1 – AUTHOR RESPONSE

Reviewer's comments	Authors' responses
Reviewer 1	
<p>It would be helpful to see results of the clinical examination of the patients, e.g. quantitative head impulse test (VOR gain), oVEMPs, cVEMPs, caloric irrigation results, posturography, subjective visual vertical, etc. I do trust in the clinical expertise of the authors but this might be helpful for reader to build an own image of the patients.</p>	<p>Thank you for your suggestion. We thought carefully about the level of clinical detail necessary to describe our sample. However, given: a) one of the main characteristics of PPPD, as described by the committee for the Classification of vestibular Disorders of the Bárány Society, is that people have normal ENT and audiometric test results and normal or near normal balance function; and b) our focus on lived experience and therefore how PPPD is experienced by participants regardless of objective clinical indicators, we decided this level of detail would add little value.</p> <p>However, we have clarified in the manuscript that ALL participants fulfilled ALL FIVE criteria proposed by the Bárány Society (see P5 line-101 and 108) i.e. all clinical examinations and tests, such as CT scans, VEMPs, head impulse test and posturography and other balance tests were negative.</p> <p>Further, as outlined in the paper, all participants were purposefully sampled from a clinic specialising in the assessment and treatment of vestibular and balance disorders. What perhaps was not so clear was that all patients had been referred to the clinic and by their general practitioner (GP) or specialist consultant and had been thoroughly assessed by a team of experienced and vestibular trained otorhinolaryngologists, audiologist and physiotherapists. All participants had undergone a battery of medical and vestibular tests (including those named by the reviewer) as a result of their referral to the clinic. We have added further clarification on P5 - line 97-99, which we hope will make this clearer for the reader.</p>
<p>The authors might think about adding (as a supplement?) an example of analysing the interview, e.g. of one sentence. I appreciate the frequent examples of patients' statements, but this does not reflect the amount of work that has been done. My idea is to convince quantitative</p>	<p>Thank you. We have carefully considered this suggestion. However, we argue that adding an excerpt of the analysis is problematic as codes only make sense within their context and themes were constructed across the entire data set rather than on the basis of an isolated excerpt. Indeed, we may be at risk of minimising the work, nuance and complexity of</p>

<p>vertigo researchers that the reported information is valid.</p>	<p>the analysis process in offering an excerpt in this way. However, most certainly a rigorous process of data analysis was followed, as outlined in the manuscript and the reporting checklist for qualitative studies based on the SRQR guidelines was followed (submitted to the editor). As such, in lieu of including an excerpt, we have expanded on the process that was followed and included further information to demonstrate the rigour of our analytical process on P7- Line 126-138. We sincerely hope that this applied methodological rigour is sufficient to convince any researcher of its validity.</p>
<p>23 subjects did not participate in the study. The authors might speculate why there was such a large rate of non-responders.</p>	<p>As described on page 7, thirty-three patients from the clinic database were invited of which nine responded. This response rate of 27.2%, which we consider to be acceptable, was not a concern for us as it was a qualitative study where a much larger sample size may have quickly become unmanageable. Indeed, consistent with Interpretive Descriptive methodology, we were not seeking a representative sample per se. Rather, our interest was in seeking a diversity of perspectives. We did not collect data from non-responders regarding reasons for not taking up the invitation and as such, we prefer not to speculate.</p>
<p>In the last para of the discussion the authors shortly mention possible effective treatment of PPPD. The authors should think about an enlargement of the points because it is about the impact of the study on this topic. The interline message is that the authors have some ideas about changing the current treatment of PPPD. For me it would be worth to show them point by point to increase the discussion on socio-psychologic support for the patients.</p>	<p>Thank you for your reflection. This research aimed to gain deeper insight in the experience of PPPD and to advance our understanding of how this condition impacts on those living with the condition. In doing so we hoped to identify potential gaps in current clinical practice and highlight areas where health professionals could have greater impact. It is not our intention to suggest a change in treatment modalities per se, but rather to highlight the importance of the way we work and what we pay attention to as potentially formative for how care is experienced and indeed outcome. As such, we instead make suggestions on ways to enhance existing treatments. However, in response to this suggestion we have expanded the last paragraph in the discussion. See P21 - L478-480.</p>
<p>Page 11, line 20: This sentence of Ethan is cited twice. The authors might refer to the first citation</p>	<p>Thank you for pointing out this repetition. We have now replaced the quote on page 11 with</p>

<p>or should point out why it is important to cite this sentence twice.</p>	<p>a quote from Thea, which conveyed a similar meaning.</p>
<p>Reviewer 2</p>	
<p>To my opinion, and based on my experience in a specialized vestibular clinic, many patients complain of very similar sensations as described in the paper....It should just be clearly mentioned that the themes are not unique for PPPD It may be helpful to interview also the other types of patients and extract the typical themes for the different disorders. This way, this approach could help differentiate the different diagnoses.</p>	<p>We agree with the reviewer that many patients visiting a specialised vestibular clinic complain of similar sensations and share the experience of having a relative invisible condition. The constructed themes may as such not necessarily be unique to PPPD. As suggested, we have now made this more explicit. P19-L435-448. We agree that better differentiation between vestibular diagnoses would be extremely helpful and this is a significant topic of discussion within the vestibular research community. However, our aim was not to compare and contrast experiences of a range of vestibular conditions and so doing so is outside the scope of this specific project. We argue the exclusive focus on PPPD to be a strength of this paper given the lack of current research available exploring the experiences of this population, particularly working age adults. Indeed, having a more in-depth understanding of the specific and unique experiences of PPPD may aid future work aiming to compare and contrast their experiences with other related conditions.</p>
<p>You should refer to table 1, and thus the latest Barany criteria, already in the beginning of the manuscript where you describe PPPD.</p>	<p>This study draws on the five main characteristics of PPPD as described by the Bárány Society in their pre-publication consensus paper, given it commenced prior to PPPD being admitted as a vestibular disorder to the International Classification of Disorders (ICD-11) in early 2017. As such, while we are aware of the updated, expanded version of the Bárány criteria, it is more accurate to report on the earlier criteria. In response to your suggestion we have cited the ICD-11 and a recent publication reporting on the most up to date characteristics of PPPD as described by the Bárány Society in the introduction P4 – L68 and 69.</p>
<p>In the data analysis it is not clearly stated that all subjects were asked the same questions.</p>	<p>Participants took part in a semi-structured interview as described in the data collection section P6-L115. Inherent to this approach, guiding questions (page 7) are commonly used to start the conversation followed by an iterative approach to questioning, meaning that consecutive questions follow from the points in</p>

	<p>the conversation that are relevant to the research question and topic of discussion. These guiding questions are described in table 2 (p7). Although all interviews started with similar guiding questions, they subsequently varied for each participant consistent with a semi-structured interview approach. An explanation is provided on P6- Line120-122.</p>
<p>The method of describing the sentences by the patients is pretty confusing. The identification (.) suggests pauses in speech. However, even with introducing pauses, these sentences make not so much sense in certain cases, and thus confusing.</p>	<p>It is common in qualitative reporting to use quotes taken directly from the participants transcripts to illustrate and strengthen the findings. We acknowledge that some of the quotes often do not entirely make sense when seen in written form. However this incoherence perfectly demonstrates the participants' inability to concisely articulate their experience of PPPD with many participants expressing their own frustration in not being able to draw that cohesive picture health professionals and researchers often aim to capture. Retaining the narrative verbatim is important to demonstrate the points we are trying to make in the paper. (.) suggests a natural pause in the participant's speech and were not added by the authors. For clarity we have added that information to P8-L157.</p>
<p>Regarding the aspect of the 'invisibility' of PPPD, this can be stated for almost all vestibular disorders. Even worse, when walking like a drunk, it looks as if somebody has had too much alcohol early in the morning, leading to shame and embarrassment.</p>	<p>We appreciate this reflection and agree with the reviewer.</p>
<p>The impact on work of the disorder can be likewise difficult to cope with for episodic disorders too, next to the chronic ones.</p>	<p>We totally agree with the reviewer.</p>
<p>The 'ignorance' or not being well informed of the clinical staff is also the case for several other disorders, such as MdDS, Vestibular paroxysmia, Vestibular Migraine, Superior Canal dehiscence (which has as a key symptom that the patient hears his or her own eyeballs roll!!! – "I'm I crazy?"). It is true that most of these disorders were not part of the education of all clinical staff that graduated before 2012 approximately, except perhaps for SCD which is ±10 year older as clinical diagnose. Thus the great majority of clinicians that is older than roughly 40 years have not learned about these diagnoses. This contributes to the frustration of</p>	<p>We appreciate this reflection and the reviewer's engagement in making sense of our findings with this context. We hope that diagnostic advances and research have contributed to a greater awareness and knowledge of a range of conditions, including those named by the reviewer. Interestingly, as discussed in our findings, participants seemed able to accept that health professionals may not know everything there is to know about their condition and even accepted that they may not have all the answers or power to do something about the condition. However, participants were particularly disappointed and</p>

<p>the patients not being recognized with their disease, neither by the health professionals, neither by their surrounding colleagues, family and friends.</p>	<p>frustrated by what they perceived as a lack of interest in the condition once they identified that there is not much known about the condition or that there is no remedial action to take (as demonstrated by Elvy's quote on P11-L227) or in the way it affected the participants life. Some participants spoke extremely favourably about health professionals who found the time to listen (as demonstrated by Thea on P11-Line 233), acknowledge the impact the condition had on their lives and were interested in helping them find ways in which they could live a meaningful life with the condition.</p>
<p>Reviewer 3</p>	
<p>In the results section of the abstract, only plain codes emerged from the work are presented. I recommend the inclusion of a brief contextualizing narrative (a few sentences) connecting these codes, apart from the implications already discussed in the conclusion section of the abstract.</p>	<p>We would have liked to expand on the findings in the abstract. However, there is an editorial restriction on the word count for the abstract that prevented us from doing so.</p>
<p>In regard to methods, it is stated that a single author carried out the actual coding process but in dialogue with the other authors. Is it the same of having multiple coders? What is the difference between having multiple coders and having a single coder that develops her work discussing the findings with colleagues? This point would be welcomed in the discussion section.</p>	<p>Although the coding was lead by AS, co-authors did code randomly chosen transcripts; with GT (co-author specialised in Thematic Analysis) taking a greater role in the coding and theme generation process. This has now been clarified in the data analysis section on P7- L126-138</p>
<p>Participants were selected on the basis of a single clinic. How does that impact on the "generalizability" (given the applicability of this concept within the realm of qualitative studies) of findings for PPPD patients outside this community? How can we assure that the problems of these patients are similar to the overall population of PPPD patients in New Zealand. It would be enlightening to have authors' views on that in the discussion section too.</p>	<p>As qualitative studies do not seek to establish generalisability in the same way as quantitative studies do, our findings are not intended to make claims about the PPPD population as a whole. Instead, our aim was to create insight in the experience of PPPD by a range of people to create a deeper understanding, highlighting commonalities as well as contextual differences. Further, we have adopted a range of strategies to enhance trustworthiness (as noted in our response above) which can aid transferability (versus generalisability) of our findings to other contexts. However, it is important to note that one of our key messages is the ideographic nature of the experience of PPPD and therefore the need to consider the unique and specific needs of individual patients (Smith, B.</p>

	(2018).Brett Generalizability in qualitative research: misunderstandings, opportunities and recommendations for the sport and exercise sciences, <i>Qualitative Research in Sport, Exercise and Health</i> , 10:1, 137-149, DOI: 10.1080/2159676X.2017.1393221)
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VERSION 2 – REVIEW

REVIEWER	Andreas Sprenger Andreas Sprenger, PhD Senior Researcher Department of Neurology University of Luebeck Germany
REVIEW RETURNED	28-Jan-2019

GENERAL COMMENTS	The authors replied to my remarks appropriately. I would have loved to see an example of sentence coding, but I can go with the authors' argumentation of the context in the interviews. I appreciate this study in order to bring PPPD nearer to GPs and Neurologists. PPPD is a quite frequent syndrome in our specialist ambulance (20-50%). Working on PPPD, a common statement I hear is: "PPPD is simply an anxiety disorder, nothing more". I think the authors here clearly show that PPPD is more than this. Therefore, this qualitative study is of high importance.
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REVIEWER	Brayan V. Seixas University of California, Los Angeles USA
REVIEW RETURNED	12-Feb-2019

GENERAL COMMENTS	I appreciate the authors' considerations on major and minor comments from the reviewers.
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