Author’s response to reviews

Title: The Diagnostic Pathway of Parkinson's Disease: a Cross-Sectional Survey Study of Factors Influencing Patient Dissatisfaction

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Version: 1  Date: 11 May 2017

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Manuscript Number: FAMP-D-16-00402

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Title: The Diagnostic Pathway of Parkinson's Disease: a Cross-Sectional Survey Study of Factors Influencing Patient Dissatisfaction

Dear editor and reviewers,

Thank you for the careful reviewing and constructive comments on our article entitled ‘The Diagnostic Pathway of Parkinson's Disease: a Cross-Sectional Survey Study of Factors Influencing Patient Dissatisfaction’. Please find enclosed our responses to each of the editor’s and reviewers’ comments and (if applicable) the referral to the revised section of the article (highlighted in the revised manuscript, figure 1 and additional file 2).
We sincerely hope that the revised version of our manuscript now merits publication in BMC Family Practice.

On behalf of all the authors,

Sincerely,

Annette OA Plouvier, MD

Editor and Reviewer Comments

Editor’s comment

Thank you for submitting your paper. I agree with the issues raised by Reviewer 1 and ask you to revise your paper and clarify the issues raised.

Reviewer 1

Thank you for the opportunity to review this paper which addresses an important aspect of care - patients' dissatisfaction with the diagnostic pathway for Parkinson's disease. The paper is well written and presented. However, I have a number of queries around the methodology, outlined below, which need to be addressed.

Background:

- "Patients' experiences during the pathway to a diagnosis can influence long-term care." In what way? This sentence is unclear, is it their satisfaction with, engagement with etc?

* We understand that reviewer 1 finds it unclear in what way long-term care is influenced. We aimed to use this sentence as an introduction for the following two paragraphs but we have apparently not succeeded in this. We therefore revised the first sentence of the Background.

Background was revised (line number 78-79)
Patients’ experiences during the pathway to a diagnosis can be negative and have long-term consequences.

Methods: the authors highlight they have published the qualitative paper previously but some more detail is necessary in the description of the methods in this current paper.

- Purposive sample of 52 essays was used. Based on what characteristics?

* We agree with reviewer 1 that more details can be provided on the characteristics used for purposive sampling in the preceding qualitative study. Purposive sampling was based on the collected demographic characteristics at the time of diagnosis: sex, age, highest level of education finished, employment status and civil status. We revised the Methods section to explicitly mention these criteria.

Methods were revised (line number 127-130)

Purposive sampling was based on the collected demographic characteristics at the time of diagnosis. We refer to the paper describing the qualitative analysis for more detailed information on recruitment, data collection and results.

- "The qualitative analysis results were used to create a format to examine the content of all essays. Details on the coding format are described in Additional file 1." In your content analysis, what was the coding agenda/rules for assigning satisfied or dissatisfied? "the overall feeling a patient expressed about the diagnostic pathway in his/her essay" isn't a specific definition and there are no further details in the appendix supplied. For instance, what if a participant expressed a mix of feelings - is that neutral?

* Thank you for giving us the opportunity to describe the rules for coding satisfaction in more detail. We based our decision for coding satisfaction/dissatisfaction on patients’ answers to the last question described in table 1: ‘Looking back on the diagnostic pathway, how do you feel about the timing of the diagnosis? Can you describe the consequences of this timing for you and your family?’ Explicit negative or positive remarks in other parts of the essay were used for coding satisfaction/dissatisfaction as well. A mix of feelings was indeed coded as neutral. In order to avoid misunderstandings, we revised the Methods section and included the coding rule on mixed emotions.

Methods were revised (line number 138-139)

All other cases, including the expression of mixed emotions, were coded ‘neutral’.
- Analysis is described as "multivariate logistic regression". This should be multivariable, the terms are often conflated. Multivariate describes analysis with more than one outcome variable. See: Hidalgo B, Goodman M. Multivariate or Multivariable Regression? American journal of public health. 2013;103(1):10.

* The terms multivariate and multivariable are often used interchangeably in the description of a logistic regression analysis. We have read the paper reviewer 1 suggests and agree that this paper describes a clear distinction between multivariate and multivariable. Following these distinct descriptions, multivariable would indeed be the suitable term to describe the type of logistic regression analysis we performed, an analysis with a single outcome variable. We therefore revised the Abstract, Methods section, Results and Discussion section, labeling of Table 4 and labeling of Additional file 2.

The Abstract was revised (line number 53-58)

The χ2 test and a multivariable logistic regression analysis were performed to assess the relation between dissatisfaction and sex, level of education, duration of the pathway, communication with the general practitioner (GP) and the neurologist, the number of healthcare providers involved, whether or not a second opinion had taken place (including the person who initiated it) and diagnostic delay (taking into consideration who caused the delay according to the patient).

The Methods section was revised (line number 176-178 and 183-184)

A multivariable logistic regression analysis was performed to assess the independent association between dissatisfaction and sex, level of education, duration of the diagnostic pathway, the number of different healthcare providers involved, second opinion and experienced delay.

Therefore, we added interaction terms of sex with the other variables to the multivariable regression model.

The Results and Discussion section was revised (line number 207-209)

The multivariable analysis showed that low-educated patients were more likely to be dissatisfied than medium and high-educated patients (OR 0.45; CI 0.2-0.9 and OR 0.46; CI 0.2-0.9, respectively).

Labelling of table 4 was revised (line number 428-429)
Table 4. Multivariable logistic regression of factors influencing patient dissatisfaction with the diagnostic pathway of Parkinson’s disease

Labelling of Additional file 2 was revised

Additional file 2. Multivariable logistic regression of factors influencing patient dissatisfaction with the diagnostic pathway of Parkinson’s disease, including interaction terms of sex with other variables

- A justification needs to be given for the inclusion of the covariates - based on literature or just what was available in the dataset?

* Indeed we based our selection of the covariates both on literature, the results of the qualitative analysis and expert opinion. From literature it is known that both sex and level of education can influence patient dissatisfaction. From the qualitative analysis we knew that duration of the diagnostic pathway, the experienced communication with the general practitioner and neurologist, and the experience of delay had an impact on patient dissatisfaction. Based on expert opinion we also included the number of health care providers involved and the request for a second opinion. In order to more explicitly describe the selection criteria for the inclusion of covariates in the analysis, we revised the Methods section.

The Methods section was revised (line number 155-160)

We wanted to assess the relation between dissatisfaction with the diagnostic pathway and a selection of factors. These factors were selected based on literature [5, 18], the results of the preceding qualitative analysis [16] and expert opinion. The χ2 test was used to assess the relationship between dissatisfaction and the demographic variables sex and level of education, the latter divided into low (primary school/vocational education), medium (secondary school) and high (higher professional education/university).

- Some of the categorical variables are problematic in the model. Particularly the "Experienced delay" variable as it has 5 levels and I suspect that is why you are reporting the very large OR of 38.78. Dummy variables may be more appropriate here.

* We agree with reviewer 1 that the OR of 38.78 is striking. Perhaps we do not interpret the reviewer’s suggestion of adding dummy variables to the multivariable logistic regression
model correctly, as this would not lead to different results: SPSS 22.0 automatically includes dummy variables into the analysis. In absolute and relative numbers there is also a remarkable difference in patient dissatisfaction between the different categories of experienced delay (Table 3). For instance, in the ‘no delay’ group only 3.2% of the patients is dissatisfied, whereas in the group that feels the healthcare provider caused the delay 61.0% is dissatisfied. Doubts or confusion about the large OR might perhaps be avoided if we had described the results of the univariable analysis of the relationship between patient dissatisfaction and experienced delay more explicitly. We therefore revised the Results and Discussion section.

The Results and Discussion section was revised (line number 203-206 and 257-259)

Of the patients who felt the healthcare provider caused delay, more than half were dissatisfied (n=111; 61.0%), whereas less than 5% of the patients, who did not experience delay or felt delay was due to themselves, were dissatisfied (n=15; 3.2% and n=2; 2.6%, respectively).

Our study shows that patients who feel that their healthcare provider is responsible for delay in the diagnostic pathway are far more likely to be dissatisfied than patients who do not describe delay.

Results & Discussion

- The results will need to be reinterpreted in light fitting the model with dummy variables.

* In order to understand why we did not revise our results, we would like to refer you to our response to the reviewer’s suggestion to add dummy variables.

- Table 3. should be labeled univariable, not Univariate.

* We agree with reviewer 1 that in light of the earlier mentioned article concerning the difference between multivariate and multivariable regression analysis, labeling of table 3 should also be revised.

Labeling of table 3 was revised (line number 422-423)
Table 3. Univariable analysis of factors influencing patient dissatisfaction with the diagnostic pathway of Parkinson’s disease

Reviewer 2

Reviewer 2 has no comments that need to be answered or require revision of the manuscript.

* We would like to thank reviewer 2 for the positive feedback on our manuscript.