**Author’s response to reviews**

**Title:** Measuring the Quality of Care in Nursing Home Residents with Early-onset Neurodegenerative Diseases: A scoping review

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**Author’s response to reviews:**

Maastricht, January 31, 2020

Dear Dr. Dylan Smith,

Thank you for the opportunity to revise our manuscript ‘Measuring the Quality of Care in Nursing Home Residents with Early-onset Neurodegenerative Diseases: A scoping review’ (PCAR-D-19-00156). We were pleased that our manuscript is potentially acceptable for publication in BMC Palliative Care after carrying out the revisions suggested by the reviewers.

We appreciate the careful review and constructive suggestions. It is our belief that the manuscript has been improved substantially after making the suggested edits.

In this letter we show the reviewers’ comments together with our responses, including how and where the text has been modified. Also a clean version of our manuscript is submitted.

The revision has been developed in consultation with all co-authors, and each author has given approval to the final form of this revision.

We would like to thank you for allowing us to submit a revised version of the manuscript and we hope that the revised manuscript will be acceptable for publication in BMC Palliative Care. Thank you for your consideration.

Yours sincerely,
On behalf of all co-authors,

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Reviewer reports:

PCAR-D-19-00156

Measuring the Quality of Care in Nursing Home Residents with Early-onset Neurodegenerative Diseases: A scoping review

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BMC Palliative Care

Reviewer # 1

Comment 1:

Search criteria. I fear that limiting it to electronic database could cause the loss of important data. To my knowledge there are studies exploring the unmet needs of people with neurodegenerative conditions that were not included in the reference list, so not mentioned in the paper, that should have been considered. Some are a little older than 2008 but still very informative (eg Kristjanson LJ, Aoun SM, Oldham L. Palliative care and support for people with neurodegenerative conditions and their carers. Int J Palliat Nurs. 2006 Aug;12(8):368-77.) Other may be published in not indexed elect DB but still interesting and easy to retrieve (eg S. Veronese, G. Gallo, A. Valle, C. Cugno, A. Chiò, A. Calvo, C. Rivoiro &amp; D. J. Oliver (2015) The palliative care needs of people severely affected by neurodegenerative disorders: A qualitative study, Progress in Palliative Care, 23:6, 331-342, DOI: 10.1179/1743291X15Y.0000000007). I was astonished by the absence of recent assessments of MS patients' and carers' needs (Borreani C, Bianchi E, Pietrolongo E, Rossi I, Cilia S, Giuntoli M, Giordano A, Confalonieri P, Lugaresi A, Patti F, Grasso MG, de Carvalho LL, Palmisano L, Zaratin P, Battaglia MA, Solari A; PeNSAMI project. Unmet needs of people with severe multiple sclerosis and their carers: qualitative findings for a home-based intervention. PLoS One. 2014 Oct 6;9)
Response 1:

We understand the fear of the reviewer with regards to missing important data. However, we chose to follow the mostly used search strategy as performed in electronic databases in combination with a check of the reference lists of included articles. We focused on a time frame fitting in the rapidly changing health care sector.

Amendment 1: (Methods, Line 132, Page 6)

Finally, the reference lists of included articles were screened for additional studies.

Changed into:

Finally, the reference lists of included articles were screened for additional studies to ensure that no relevant publications were missed.

Response 1:

Furthermore, we had a look at the three articles suggested by the reviewer. We agree they are informative from the perspective of quality of care from a broader perspective than the target groups of nursing home residents or institutional long-term care residents. However, as we are specifically interested in assessing quality of care in nursing home residents with neurodegenerative diseases, we explicitly chose to focus our scoping review on this setting and target group, because we are solely interested in how patients experience the care in the nursing home. In addition, the focus was not on palliative care (needs).

We do believe it is very informative to compare our results with the results of Borreani et al., who looked at unmet needs of patients with severe MS living at home. Therefore, we included a comparison to the discussion (page 11, line 236)

Amendment 1: (Discussion, Line 236, Page 11)

Added to the discussion:

A study of Borreani et al. (2014) (21) focused on needs, views and experiences, perceived by adults with severe MS living at home, their carers and health professionals. The quality of care items found in this study (21) are highly similar to the items found in our review. However, the setting of the study is community care, whereas the setting of our study is institutional long term care. Also, the instruments described in this review do not include the domain ‘health and social policies’ (with categories ‘rights’, ‘culture’ and ‘patient organizations’) found in the study of Borreani et al. (21). Whereas, the domain ‘expertise’ included in our review was not described in the study of Borreani (21).
Response 1:

A number of sentences describing the study of Peters et al. in the discussion have been shortened to ensure readability.

Amendment 1: (Discussion, Line 244, Page 11)

Furthermore, Peters et al. (21) investigated patients’ experiences of care services for long-term neurological conditions (LTNC). A survey was carried out on MND, MS and PD patients (n=2563), who were members of patient associations and due to their illness, required lifelong support from health and social care services. The questionnaire that was used consisted of six dimensions, divided into 35 items, and was based on relevant quality requirements, guidelines, scientific articles and expert opinions (21). Of the six dimensions (‘diagnosis’, ‘treatment’, ‘patient-centered care’, ‘health and social care professionals understanding about LTNC’, ‘treated with respect and dignity’ and ‘personal care and support’) the majority correspond with the eight domains found in this review. The differences between this study and our review can be found in the dimensions ‘diagnosis’ and ‘general practitioner’, which are present in the instrument of Peters et al. (21) and not in the domains in our review. These differences in the dimensions can be explained by differences in the setting: diagnosis is less frequently discussed in the nursing home setting than in other settings and in addition, ‘general practitioner’ involvement in nursing homes differs among countries. Furthermore, the content of the dimension ‘personal care and support’ in the study of Peters et al. (21) is more focused on ‘obtaining financial support and help with housework’ whereas in this study the focus is more on ‘nursing’ and ‘medical care’. These differences can therefore also be explained by differences in setting.

Changed into:

Furthermore, Peters et al. (22) investigated patients’ experiences of care services for long-term neurological conditions (LTNC) in MND, MS and PD patients (n=2563) living at home. The questionnaire consisted of six dimensions, divided into 35 items, and was based on relevant quality requirements, guidelines, scientific articles and expert opinions (22). The majority of the dimensions in this study correspond with the eight domains found in this review. However, the dimensions ‘diagnosis’ and ‘general practitioner’, in the instrument of Peters et al. (22) are not present in the domains of our review. One explanation for this may be the studies are carried out in different settings. Furthermore, the involvement of the general practitioner in nursing homes differs among countries. Finally, the content of the dimension ‘personal care and support’ in the study of Peters et al. (22) is more focused on ‘obtaining financial support and help with housework’ whereas in this study the focus is more on ‘nursing’ and ‘medical care’.

Comment 2:
The inclusion criteria were too strict to be adequately informative. I'm sure that the place of care is important, because that's where you want the QoC to be measured, but the assessing tools may be very valid in picking up the needs even if were not specifically developed in NH. I think that for example Patient Reported Outcome Measures (PROMs) such as the Integrated Palliative Outcome Scale (IPOS) or its predecessor POS (with the some specific physical needs assessment scales for conditions like PD or MS), had to be considered.

The exclusion of dementia "as this neurodegenerative disease usually has a much later onset in life and shows no physical disability" I think is really a poor exclusion criteria: first it's not true that there is no physical disability in demented patients, and physical symptoms are often overlooked, patients become progressively dependent in all their daily living activities. Second dementia can occur very early even in neurodegenerative conditions other than "dementia", consider for example Parkinson's plus syndromes like PSP or Lewy body dementia, or some MS forms with primary psychiatric symptoms. In NH demented patients should be assessed in terms of quality of care considering their unmet needs (see for example the IPOS-DEM https://journals.plos.org/plosone/article?id=10.1371/journal.pone.0200240)

Response 2:

The reviewer is correct that tools which were not developed in nursing home patients may still be valid to pick up needs in this population. Therefore, we included both tools developed in nursing home patients and tools applied in nursing home patients (the last ones might have been developed elsewhere). This is clarified in our methods section (methods, data extraction, page 7, line 151).The reason why we did not find and include PROMs is that we are specifically interested in what tools are available/used in the nursing home sector and PROMs appear to not have been used in our population and setting.

Amendment 2: (Discussion, Line 276, Page 12)

Added to the discussion:

Examples are Patient Reported Outcome Measures (PROMs) that could in time be an option to use within our target group, possibly in adapted form. These instruments will be taken into account in future development of a QoC assessment tool for our target group.

Response 2:

Secondly, excluding nursing home residents with dementia was done to exclude the larger dementia population in nursing homes that are on average older in age and mainly limited in their cognitive functioning compared to residents with early-onset neurodegenerative diseases. Our rationale behind this will be more explicitly explained in the manuscript (page 6, line 116).

Parkinson’s plus syndromes, like PSP or forms of MS, are not excluded; early stage dementia in these diseases are a symptom of a disease and not a pure form of dementia.
Amendment 2: (Methods, Line 116, Page 6)

Furthermore, studies focusing on dementia were excluded, as this neurodegenerative disease usually has a much later onset in life and shows no physical disability.

Changed into:

Furthermore, studies focusing on isolated dementia were excluded, as we were specifically interested in other target groups in the nursing home setting with early-onset neurodegenerative disorders with cognitive as well as physical deterioration. In addition, pure dementia usually has a much later onset in life and only shows physical disability in the end stage of the disease.

Comment 3:

Results: the review found out 8 domains: 'emotional support', 'physical support', 'social support', 'care', 'care content', 'expertise', 'communication' and, finally, 'organization of care'. They are all obviously reasonable. What strikes me is the absence of Advanced Care Planning (ACP), since in most studies addressing the quality of life, preferences and fears of both patients and carers this aspect is very common.

Response 3:

We agree with the reviewer that it is striking that ACP was not explicitly found in the literature we included. Some of the items found in our review (e.g. ‘promotion of autonomy’ and ‘getting to know the patient is essential to understanding their wishes and needs’ in the domain ‘emotional support’) are related to the topic of ACP. The focus of our search was experienced QoC, without a specific focus on End of Life Care, what might explain the absence of ACP as domain. To denote this absence an amendment was made in the discussion.

Amendment 3: (Discussion, Line 255, Page 12)

Added to the discussion:

Early-onset neurodegenerative diseases such as PD and HD in an advanced stage require a palliative care approach (23, 24) and therefore it is common for care providers to address aspects of advance care planning or end-of-life planning. However, the domain ‘advance care planning’ or ‘end-of-life planning’ was not explicitly found in this review.

Comment 4:

In conclusion I think that this review adds important information about some important aspects of the quality of care evaluation in people with NDC, but there are some methodological choice
that limit the validity of the results. Before concluding that another Quality of care assessment tool is needed, I would consider the role of others already in use and test if can be of help

Response 4:

We are happy that the reviewer thinks the review adds important information. As mentioned in earlier comments, our research question specifically focused on what tools are available and used in nursing home residents with early-onset neurodegenerative diseases. Even though we agree that there might be tools available in other settings and for other target groups, this search was not part of this scoping review. Therefore, we will more clearly address the generalizability of our results in the discussion.

Amendment 4: (Discussion, Line 272, Page 12)

Added to the discussion:

To answer our research question, the search was explicitly limited to instruments developed and/or used to measure QoC in nursing home residents with early-onset neurodegenerative diseases. We are aware of the fact that there might be instruments developed and/or used to measure QoC in other settings or target groups (e.g. younger residents with dementia) which were not included in this review. Examples are Patient Reported Outcome Measures (PROMs) that could in time be an option to use within our target group, possibly in adapted form. These instruments will be taken into account in future development of a QoC assessment tool for our target group.

Response 4:

A number of sentences in the discussion have been shortened to ensure readability.

Amendment 4: (Discussion, Line 264, Page 12)

Published research in this topic is scarce. This demonstrates the current gap in the literature and the need for more research. The conclusions drawn in this review are based on a few studies that represent currently existing information. The quality of the studies ranged from moderate to outstanding. This makes comparison of the findings difficult. Still, since this is a scoping review, the gaps in methodological quality can be taken into account when reflecting on the data (14).

Changed into:

Published research in this topic is scarce and therefore this study adds important information. However, this study also demonstrates the current gap in the knowledge and the need for more research. The quality of the studies included in this review ranged from moderate to outstanding.
and this makes comparison of the findings difficult. It is recommended to take the differences in methodological quality into account when interpreting the data.

Response 4:

A number of sentences in the discussion have been shortened to ensure readability.

Amendment 4: (Discussion, Line 270, Page 12)

However, due to the subject of our review, and the amount of included studies, the size of the sample is less important than the content.

Changed into:

However, due to the subject of our review, and the number of included studies, the size of the sample is less important than the content of the instrument.

Reviewer # 2

Comment 5:

Basic assumption for the research question is that this special group of patients "needs more complex care" than "regular" residents in nursing homes. However, this statement is not very well explained except for describing living circumstances in younger ages. What does this mean for the quality of care and how should this be represented when assessing QoC. This should be discussed, mainly because the results seem to hide any differences between the focused residents and "regular" ones.

Response 5:

To explain this statement more clearly, extra information is added in the background section.

Amendment 5: (Background, Line 63, Page 3)

Sentence added:

In comparison with isolated dementias, such as Alzheimer’s disease, the neurodegenerative diseases at stake require more specific activities, with regard to the physical, psychological, social and environmental support, mainly because of the early onset and its related impact on daily life and prognosis.
Comment 6:

Please start with the information that you refer to neurodegenerative diseases other than pure dementia.

Response 6:

We inserted the words ‘Other than pure dementia’ in the first sentence in the background section.

Amendment 6: (Background, Line 49, Page 3)

Patients suffering from neurodegenerative diseases, such as..

Changed into:

Patients suffering from early-onset neurodegenerative diseases other than pure dementia, such as..

Comment 7:

The keywords should include early onset… or the most common disease in this group.

Response 7:

The keyword ‘early-onset’ is included now.

Amendment 7: (Abstract Keywords, Line 46, Page 3)

Neurodegenerative diseases

Changed into:

Early-onset neurodegenerative diseases