Reviewer’s report

Title: Children’s and Adolescents Adjustment to Parental Multiple Sclerosis. A Systematic Review

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Reviewer: Jesper Hagemeier

Reviewer’s report:

“Children’s and Adolescents Adjustment to Parental Multiple Sclerosis. A Systematic Review” is a collection of literature looking into the psychological wellbeing of children who have at least one parent diagnosed with MS. There are several methodological limitations (such as selection bias) that cannot be dismissed, however, what this review highlights is that the current literature on the subject matter is sorely lacking quality studies and that further research should be conducted into quantitative, reproducible metrics of psychosocial wellbeing of children of parents with MS.

Major:

1. Accuracy of data abstraction was cross-checked and confirmed randomly for 10 studies using a 2nd reviewer (R.N.). Is this based on 10 out of 70 articles (as mentioned in the Literature Search section: “Seventy articles underwent full-text review”)? Or 10 out of an even larger (or smaller) group of papers? This is not clear. Out of those 10, what was the level of “agreement” between the two reviewers (on data, final score etc.).

2. The authors use the GATE framework as a method to evaluate the selected studies. To this reviewer’s knowledge, this is a somewhat basic method to evaluate epidemiological studies, mostly catered to students (e.g. see Baker et al. 2010). Granted, that does aid in assessing the literature quicker than other methods may allow. How confident are the authors of the GATE method, even when evaluating several works that are not purely epidemiological? Furthermore, GATE does not assign a strict “score” or “grade” to the evaluations. How confident are the authors that their (somewhat subjective) “low”, “medium” and “high” scores are reliable and reflect that actual work?

3. None of the 18 studies examined specified which MS diagnostic criteria were used (and only few reported disability measures). Since diagnostic criteria have changed over time, it would be of importance to know which criteria were used. Only 8 out of 18 had a control group, and 4 studies were not quantitative. Furthermore, the authors highlight that 16 studies had either “moderate” or “low” quality scores. All these factors highlight that most studies conducted so-far are of mostly low quality and more importantly drawing conclusions on a population level is difficult. To enhance the difficulty even further there does not appear to be any standard for assessing children in these households, as a wide range of measures were used to measure psychosocial wellbeing: anxiety, depression,
peer relations, caregiving responsibility, family cohesion, body image, parent-child interaction, and hopefulness.

Furthermore, out of 1114 studies, 1044 were excluded for not satisfying study inclusion criteria. Then, another 52 were excluded while doing more extensive review. Only 18 studies remained in the review. Of those remaining 18, only a handful scored “high” in study quality. Originating from such a large initial sample, the fact that only 2 studies are of high quality is surprising, even more so considering the wide range of psychosocial outcomes that was included across studies. This does highlight the importance of future research in this subject matter. However, this does also limit the possibility of drawing meaningful conclusion from this review, as more research is needed. Before more research has been carried out, the authors should refrain from drawing conclusions and need to further downplay findings of the reported studies. Please provide recommendations for future research on how studies should be standardized and improved.

4. It is mentioned several times throughout the manuscript, including in the abstract, that “having a parent with MS was typically reported to have negative psychosocial effects…” However, the results appear rather dispersed and unclear, for example: “8 found a negative association between exposure to parental MS and adjustment in their offspring. Five studies did not find an association and 5 studies found both positive and negative effects of caring for a parent with MS.” The authors should highlight that results vary widely and no consensus can be reached at this moment.

5. Please also expand in the abstract and discussion/conclusion (not only in the limitation section) on the overall weakness of the literature investigated and that conclusion are difficult to make from the present literature.

6. Finally, the authors should on the importance of investigating the psychosocial wellbeing of children with parents with MS as compared to other chronic illnesses. Especially considering that in MS disability is limited in earlier disease stages and with low EDSS scores, and lifespan may not be affected as with other chronic diseases.

Minor:

7. “Families are the primary source of experience for most children”. Consider rephrasing “experience” as it is ambiguous.

8. The first sentence makes reference to “Western societies” and a range of 4 to 12% of Children with parents living in households with a parent with a chronic illness. The cited paper is part of the (north) Germany Hamburg Health Survey, and find a 4.1% prevalence rate. The authors should add additional references substantiating their claim or alter the sentence.

9. In the study description (Results section) it would be helpful to elaborate on other possible factors influencing the wellbeing of children of MS parent(s). For example, is the divorce rate or percentage of single-parent households higher
among these families?

10. Results are summarized purely descriptively. It is hard to draw conclusions when no information is known about (study specific) statistical measures such as effect sizes and p-values, although this is most likely an inherent problem of the source studies.

11. Regarding line 161, 162 and 163. It should be stressed that the children who are 3x as often perceived by their parent(s) (with MS) to have psychological problems, might well be due to not the child’s actual psychological well being, but due to for example the parents’ perceived feeling of guilt.

12. The limitations of the review process are adequately described. Consider also discussing the GATE method as limitation as well.

**Level of interest:** An article whose findings are important to those with closely related research interests

**Quality of written English:** Acceptable

**Statistical review:** No, the manuscript does not need to be seen by a statistician.

**Declaration of competing interests:**

I declare that I have no competing interests