Author’s response to reviews

Title: Cost-effectiveness of cannabinoids for pediatric drug-resistant epilepsy: protocol for a systematic review of economic evaluations

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Author’s response to reviews:

Sue Harnan
Editor, Systematic Reviews

RE: Manuscript revision - Cost-effectiveness of cannabinoids for pediatric drug-resistant epilepsy: protocol for a systematic review of economic evaluations

Dear Dr. Harnan,

Please find attached our revised manuscript, “Cost-effectiveness of cannabinoids for pediatric drug-resistant epilepsy: protocol for a systematic review of economic evaluations

We thank you and the reviewers for the questions, helpful comments and for taking time to provide suggestions. We have considered each item individually, as described below.

We look forward to hearing from you at your earliest convenience.

Sincerely,
Reviewer 2:

- It would be useful for the authors to clarify whether a date limit will be included for the searches, and consider whether the inclusion of such date limit is appropriate. In particular, do the authors expect studies conducted 20 years ago to be applicable to the current context? The inclusion of a date limit may make the review more manageable.

Author response: In our review protocol, we did not propose to apply any date restrictions or limits for literature searching. Although we recognize that older studies may not be relevant to the current clinical context, we chose not to impose a limit because we anticipated a small number of eligible studies, given that most drug-resistant epilepsy syndromes are considered rare diseases. We have revised the text to explicitly state that no date restrictions were applied:

Page 7: “The search strategies will be adjusted to the individual database and will include a combination of controlled vocabulary (e.g., Medical Subject Headings) and keywords; no date restrictions will be applied.”

- the authors are planning to exclude non-English studies. Whilst this is commonly done, does this represent an important limitation given the focus appear to be on the Canadian setting, whereby both French and English are used (and possibly studies)?

Author response: We agree with Reviewer 2 that the exclusion of Canadian studies reported in French may exclude relevant Canadian data. We have revised the text to state that we will include studies performed in Canada that report data in French.

Page 7: “Conference abstracts and non-English language records will be excluded; however, cost and resource utilization data from studies performed in Canada but reported in French will be eligible.

- the authors are planning to exclude abstracts. This may be a limitation as a number of studies may only published in abstract form, as typically the case for utility and resource studies. The authors should consider whether conference abstracts should be included.
Author response: Although we considered including abstracts in our review, we feel that abstracts will not, for our purposes, provide sufficient information about the context, study design and methodology, and results, owing primarily to their low word count.

- In the method section, the authors state that "associated costs will be extracted only from studies performed in Canada". For completeness, and for the review to be used by researchers in other countries, the authors may want to extract costs irrespective of the country where the study was conducted.

Author response: As part of our final report, we will list any studies that provide cost data from countries other than Canada, such that interested researchers are able to extract the relevant data and apply it to their context. Because we will convert all reported costs to 2018 Canadian dollars, it would be inappropriate for researchers from other jurisdictions to directly use the values reported in our review and would require additional conversion to their local currency before use. As such, researchers from other jurisdictions will be better served by a list of resources where the original study data can be found.

- Finally, the authors are proposing to use the Drummond and CHEERs Checklist when assessing the quality of each study. Whilst these checklists are commonly used, these are limited to reporting. The authors may also want to consider using an additional checklist more focused on modelling, such as the Philips checklist (2014) if the aim is to assess the validity of the modelling, rather its the reporting

Author response: The Drummond and CHEERS checklists are commonly used in systematic reviews of economic evaluations. We believe that the Reviewer is referring to the article published by Philips in 2006, which proposes a framework for the quality assessment for decision-analytic models (Philips 2006). This framework covers similar aspects of decision model quality compared with Drummond and CHEERs checklists

Reference: