

## PEER REVIEW HISTORY

BMJ Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form (<http://bmjopen.bmj.com/site/about/resources/checklist.pdf>) and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

### ARTICLE DETAILS

<b>TITLE (PROVISIONAL)</b>	How Congenital Zika Virus impacted my child's functioning and disability: a Brazilian qualitative study guided by the ICF
<b>AUTHORS</b>	Campos, Taynah; Schiariti, Veronica; Gladstone, Melissa; Melo, Adriana; Tavares, Jousilene; Magalhães, Adriana; Longo, Egmar

### VERSION 1 – REVIEW

<b>REVIEWER</b>	Tracey Smythe London School of Hygiene & Tropical Medicine, UK.
<b>REVIEW RETURNED</b>	16-May-2020

<b>GENERAL COMMENTS</b>	<p>In this study the authors use qualitative research methodology, through the use of focus groups, to identify outcome measures that are important to parents of children with Congenital Zika Syndrome (CZS). The study aims to inform methods of evaluation that can be used to assess the impact of CZS on a child's function and disability.</p> <p>Participants were parents of children with CZS and the study approach uses the International Classification of Functioning, Disability and Health (ICF) as a framework for analysis. The authors report parental concern included environmental barriers in addition to themes of functioning. The topic of the article is relevant and contributes to the literature on the Zika virus, as well as the development of outcome measures for children with developmental disabilities in general.</p> <p>This article benefits from the qualitative research approach and the in-depth responses that were gained from participants. This study has the potential to provide important evidence toward understanding the use of outcomes measures from a caregiver perspective. However, the authors could further strengthen this manuscript by a more in-depth discussion of the study population and participants, detailed methods and analysis and a critical review of use of language to effectively communicate their research findings. My specific revision suggestions are outlined below:</p> <p>Please review language and sentence structure, for example:</p> <ol style="list-style-type: none"><li>1. P 3 Line 10 -14</li><li>2. P4 Lines 49 – 60 would benefit from being two sentences to assist the reader</li><li>3. P5 Line 5 – 6 please include more information about COMET in the main text – such as the institutions that are involved and main aim of the trials</li><li>4. P5 line 14 – clarification of what is meant by 'in this context' is required for the reader to understand whether this relates to the COMET trials, this study or the context of parental participation.</li></ol>
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	<p>5. P5 Line 36 – the title ‘Zika Cos and congenital infections:’ is unclear. Consider clarifying ‘Cos’ – my understanding is that this may be an acronym that relates to ‘Core Outcome Set’</p> <p>6. P 9 Line 35 – 40</p> <p>7. P11 Line 18 – 23</p> <p>8. P14 Line 16</p> <p>The study would be strengthened through using the Consolidated Criteria for Reporting Qualitative Studies (COREQ) in the study text – although the study flow chart is detailed in Figure 1 and the study notes that the COREQ guidelines are considered (P 7 Line 25), important additional details are required in the methods in order to assess the rigor of analysis and credibility of the findings, and these should be included in the text of the paper, such as:</p> <p>9. Page 6 Line 41 - 48: Who were the participants purposively selected by? What was the method of approach? What are the characteristics of the respective rehabilitation services in Rio Grande do Norte and Paraíba? P7 Line 29 – where was the sociodemographic questionnaire undertaken? (Q9-16 of COREQ guidelines) L14, was only one physiotherapist involved or was this the primary physiotherapist of the child? When did the physiotherapist classify the child’s motor abilities with GMFM? Detail of research team and reflexivity (Q1-8 of COREQ guidelines) is required. How many research assistants?</p> <p>10. Was the questionnaire piloted prior to use?</p> <p>11. Greater explanation of ‘fun fishing’ is required P7 Line 57 – eg what it is, what the purpose of it is and when it was encouraged to be used.</p> <p>12. P8 were the interviews in Brazilian Portuguese? If so, were they translated?</p> <p>13. P9 subheading on line 15 – consider including this as a sentence to clarify exactly what patients were not involved in.</p> <p>With regards the results,</p> <p>14. P 9 Line 42 it is unclear why personal factors were not considered given the use of the ICF. Please include justification of this.</p> <p>15. P11 Line 7 – 16 The text notes that a father desired more access to rehabilitation services, however the illustrative quote is from Mother 20.</p> <p>16. Also, the discussion section should not include results but instead discuss them according to the literature on the subject, which is partially done. There may be other relevant studies that could enrich the discussion of the results.</p> <p>17. Please provide the ethical approval numbers/IDs of ethical approvals on p6 line 15-28</p> <p>18. The abstract would benefit from a revision to provide succinct detail of methods and results.</p>
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<b>REVIEWER</b>	Ashley Walker Georgia Southern University USA
<b>REVIEW RETURNED</b>	17-Jun-2020

<b>GENERAL COMMENTS</b>	A nice application of the ICF to help contextualize the perspectives of caregivers. I would recommend one quick review to identify minor formatting issues in the manuscript.
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<b>REVIEWER</b>	Raphael Ximenes
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	University Health Network, Canada
<b>REVIEW RETURNED</b>	26-Jul-2020

<b>GENERAL COMMENTS</b>	<p>This study aims to understand the views and perceptions of parents with regard to the needs of their children with CZS framed within the perspective of the ICF.</p> <p>Even during the COVID-19 pandemic, it is very important that we do not forget other diseases like Zika.</p> <p>This work shows the relevance of continuing research on Zika and the impact caused by the 2015/16 epidemic in South American countries, mainly in Brazil.</p> <p>I recommend publishing the manuscript with minor adjustments.</p> <p>Page 6:</p> <p>“The inclusion criteria included being; parents/caregivers of children with confirmed diagnosis of CZS by polymerase chain reaction (PCR) or presumed diagnosis based on obstetric ultrasound, transfontanellar ultrasound, computed tomography (CT) and magnetic resonance imaging (MRI), who were from zero to six years old and who lived in the area covered by the study (states of Rio Grande do Norte and Paraíba) and who attended the respective rehabilitation services.”</p> <p>If the Zika cases happened in 2015, why do the inclusion criteria include children up to 6 years of age?</p> <p>Same question about the Zika Focus Group, in the Supplementary material 1. Why did you include children from 0-10 years and not 0-5 years old only?</p> <p>Page 7:</p> <p>Please, make sure all acronyms are defined when you use it at first.</p> <p>You only define CP on page 16 but you’re using it on page 7. “This is an age-specific scheme designed for children with CP based on five levels”</p> <p>Same thing for COREQ on page 7.</p> <p>Consolidated criteria for reporting qualitative studies (COREQ).</p> <p>Page 11</p> <p>“In the following verbatim, it is possible to observe the father’s desire to have access to a rehabilitation service in his own city.”</p> <p>“Also that in Alagoas state has been the same possibilities that here in Campina, to don’t need to move from one place to other, it’s so complicated spend 3 months here, searching treatment that there it should have. I don’t know they can’t leave this treatment there, to every</p>
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	<p>states have, for us don't need to move a lot to do a treatment. For them could at least sit, maintain their trunk, catch and walk." Mother 20.</p> <p>You wrote "father's desire" but the sentence cites Mother 20. Is that an error or are you using Mother as a general reference? If yes, why not to use parent/caregivers?</p> <p>Page 29- Fix typo - Participants were invited via phone call or in person</p>
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<b>REVIEWER</b>	Michel Counotte Institute of Social and Preventive Medicine, University of Bern, Bern, Switzerland
<b>REVIEW RETURNED</b>	27-Jul-2020

<b>GENERAL COMMENTS</b>	<p>Campos et al describe the need for a patient/care-taker centred approach to improve the core outcome sets for studies conducted in children growing up with congenital zika syndrome. They conduct focus group interviews to identify their perspectives on relevant areas of functioning and disability. The work is important and conducted well, however the reporting and discussion needs clarification.</p> <p><b>Abstract:</b></p> <ul style="list-style-type: none"> <li>* the authors start with a percentage, could they first provide the total number of included participants?</li> <li>* the abbreviation GMFCS is not explained</li> </ul> <p><b>Methods:</b></p> <ul style="list-style-type: none"> <li>* Can the authors be more explicit about the enrolment, how many people were approached, how many refused?</li> <li>* Given the age distribution of the children reported in figure 2, it seems this study was conducted some time ago. Can the authors be more explicit when the focus groups were conducted.</li> </ul> <p><b>Results:</b></p> <ul style="list-style-type: none"> <li>* Since a main objective seems to be to identify priorities, it is unclear how 'frequency' translates into 'priority'. Does the ranking in table 1 reflect priority, or frequency of mentioning? And is mentioning this the same as having difficulties within this category? Can the authors be more clear/explicit about this (which are barriers and which are facilitators?)?</li> <li>* The English translation of some of the verbatim text quoting the parents/caretakers is somewhat confusing, would the authors consider improving these translations (possibly back/forth translation to ensure the meaning is not lost)</li> <li>* Figure 2+3: Even though I appreciate the effort the authors put in Figure 2+3, it reports an overview of the characteristics of the study population. A structured table would be more appropriate, reporting the number, the denominator and the percentages. Rounded percentages are sufficient for these small numbers (n=32).</li> </ul> <p><b>Discussion:</b></p> <ul style="list-style-type: none"> <li>* Would the authors consider starting the discussion with the most important findings?</li> <li>* Which factors do the authors recommend to be included as part of the COS?</li> </ul>
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	<p>* This is a relatively small sample, the author do hint briefly about the representativeness, can the authors discuss this in more detail, how their sample might be generalized?</p> <p>* Can the authors comment on the potential shift of priorities as the children age? Now they all seem to be still young, care needs/priorities will likely shift in the future.</p>
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### VERSION 1 – AUTHOR RESPONSE

Reviewer: 1

Reviewer Name: Tracey Smythe

Institution and Country: London School of Hygiene & Tropical Medicine, UK.

Dear Dr Smythe, thank you for all suggestions, they made possible an important improvement of our paper.

P 3 Line 10 -14 (Please review language and sentence structure)

Answer: The second author, native in English, carried out an extensive review of the writing style.

P4 Lines 49 – 60 would benefit from being two sentences to assist the reader (Please review language and sentence structure)

Answer: We made this change.

P5 Line 5 – 6 please include more information about COMET in the main text – such as the institutions that are involved and main aim of the trials (Please review language and sentence structure)

Answer: We made this change by providing this information.

P5 line 14 – clarification of what is meant by ‘in this context’ is required for the reader to understand whether this relates to the COMET trials, this study or the context of parental participation. (Please review language and sentence structure)

Answer: We made this change by providing this information.

P5 Line 36 – the title ‘Zika Cos and congenial infections:’ is unclear. Consider clarifying ‘Cos’ – my understanding is that this may be an acronym that relates to ‘Core Outcome Set’ (Please review language and sentence structure)

Answer: We made this change by providing this information.

P 9 Line 35 – 40 (Please review language and sentence structure)

Answer: We edit the text.

P11 Line 18 – 23 (Please review language and sentence structure)

Answer: We edit the text.

P14 Line 16 (Please review language and sentence structure)

Answer: We edit the text.

Page 6 Line 41 - 48: Who were the participants purposively selected by? What was the method of approach? What are the characteristics of the respective rehabilitation services in Rio Grande do Norte and Paraíba?

Answer: We edit the text, providing this information (participants with important common characteristics were purposively selected by members of the research team, through active search by

phone call or direct personal approach. These rehabilitation centers are public services linked to research institutions and higher education in the region).

P7 Line 29 – where was the sociodemographic questionnaire undertaken?

Answer: We edit the text, providing this information (all parents responded to a sociodemographic questionnaire, applied by the researchers before the realization of the focus groups, at rehabilitation services).

L14, was only one physiotherapist involved or was this the primary physiotherapist of the child? When did the physiotherapist classify the child's motor abilities with GMFM? Detail of research team and reflexivity (Q1-8 of COREQ guidelines) is required. How many research assistants?

Answer: We edit the text, providing this information.

Was the questionnaire piloted prior to use?

Answer: We edit the text, providing this information (this questionnaire was applied to the first group and observed whether the questions were well understood by the respondents. Two research assistants were involved in the data collection).

Greater explanation of 'fun fishing' is required P7 Line 57 – eg what it is, what the purpose of it is and when it was encouraged to be used.

Answer: We edit the text, providing this information (in order to thank the parents for participating in the study. They were encouraged by the professional team to use it as one therapeutic toy for stimulation of their children at home and promote fun).

P8 were the interviews in Brazilian Portuguese? If so, were they translated?

Answer: We edit the text, providing this information.

P9 subheading on line 15 – consider including this as a sentence to clarify exactly what patients were not involved in

Answer: We edit the text, providing this information (families were not involved in the design, recruitment or conduct of the study. However, the results will be presented to families, professionals and managers at each participating center, with a view to discussing strategies to meet the needs of children with CZS and their families).

P 9 Line 42 it is unclear why personal factors were not considered given the use of the ICF. Please include justification of this.

Answer: We edit the text, providing this information (as personal factors were expressed only minimally, they are not shown in the Figure).

P11 Line 7 – 16 The text notes that a father desired more access to rehabilitation services, however the illustrative quote is from Mother 20.

Answer: this was a mistake, thanks for the observation, it was corrected in the text.

Also, the discussion section should not include results but instead discuss them according to the literature on the subject, which is partially done. There may be other relevant studies that could enrich the discussion of the results.

Answer: we edited the text of the discussion and included new and relevant studies.

Please provide the ethical approval numbers/IDs of ethical approvals on p6 line 15-28

Answer: we edit the text, providing this information.

The abstract would benefit from a revision to provide succinct detail of methods and results.

Answer: we edit some sentences of the abstract.

Reviewer: 2  
Reviewer Name: Ashley Walker  
Institution and Country:  
Georgia Southern University  
USA

I would recommend one quick review to identify minor formatting issues in the manuscript.  
Dear Dr Walker, thank you, we edit the style and format of the paper.

Reviewer: 3  
Reviewer Name: Raphael Ximenes  
Institution and Country: University Health Network, Canada

Dear Dr Ximenes, thank you very much indeed, we believe that the paper is much better with the suggested changes.

Page 6:

“The inclusion criteria included being; parents/caregivers of children with confirmed diagnosis of CZS by polymerase chain reaction (PCR) or presumed diagnosis based on obstetric ultrasound, transfontanellar ultrasound, computed tomography (CT) and magnetic resonance imaging (MRI), who were from zero to six years old and who lived in the area covered by the study (states of Rio Grande do Norte and Paraíba) and who attended the respective rehabilitation services.”

If the Zika cases happened in 2015, why do the inclusion criteria include children up to 6 years of age?

Same question about the Zika Focus Group, in the Supplementary material 1. Why did you include children from 0-10 years and not 0-5 years old only?

Answer: thank you very much for realizing this, we adjust in all sessions to age 0-5 years.

Page 7:

Please, make sure all acronyms are defined when you use it at first.

You only define CP on page 16 but you're using it on page 7.

“This is an age-specific scheme designed for children with CP based on five levels”

Same thing for COREQ on page 7.

Consolidated criteria for reporting qualitative studies (COREQ).

Answer: we made these adjustments to specific parts of the text.

Page 11

“In the following verbatim, it is possible to observe the father’s desire to have access to a rehabilitation service in his own city.”

“Also that in Alagoas state has been the same possibilities that here in Campina, to don’t need to move from one place to other, it’s so complicated spend 3 months here, searching treatment that there it should have. I don’t know they can’t leave this treatment there, to every states have, for us don’t need to move a lot to do a treatment. For them could at least sit, maintain their trunk, catch and walk.” Mother 20.

You wrote “father’s desire” but the sentence cites Mother 20. Is that an error or are you using Mother as a general reference? If yes, why not to use parent/caregivers?

Answer: this was a mistake, thanks for the observation, it was corrected in the text.

Page 29-

Fix typo - Participants were invited via phone call or in person ...

Answer: we edit the text.

Reviewer: 4

Reviewer Name: Michel Counotte

Institution and Country: Institute of Social and Preventive Medicine, University of Bern, Bern, Switzerland

Dear Dr Counotte, we were very happy with your suggestions, thank you very much for contributing to improve our article.

the authors start with a percentage, could they first provide the total number of included participants?

Answer: we edit the text.

the abbreviation GMFCS is not explained

Answer: we edit the text (Gross Motor Function Classification System - GMFCS).

Can the authors be more explicit about the enrolment, how many people were approaches, how many refused?

Answer: yes, we edit the text, providing this information (of the 36 parents identified and invited to the study, 32 agreed to participate).



Given the age distribution of the children reported in figure 2, it seems this study was conducted some time ago. Can the authors be more explicit when the focus groups were conducted.

Answer: we edit the text, providing this information (the focus groups were carried out between September / 2018 and January / 2019).

\* Since a main objective seems to be to identify priorities, it is unclear how 'frequency' translates into 'priority'. Does the ranking in table 1 reflect priority, or frequency of mentioning? And is mentioning this the same as having difficulties within this category? Can the authors be more clear/explicit about this (which are barriers and which are facilitators?)?

Answer: we provide this information in the methods and we also make it clear in the results which aspects were considered barriers or facilitators.

The English translation of some of the verbatim text quoting the parents/caretakers is somewhat confusing, would the authors consider improving these translations (possibly back/forth translation to ensure the meaning is not lost)

Answer: we took great care in that regard. The content of the focus groups in Portuguese was translated into English by a Brazilian pediatrician who at the time of the study was doing research at the University of Liverpool with Dr Gladstone. After this translation, the authors held a skype meeting to discuss with him possible doubts, to ensure that the participants' expressions were not lost. Upon your request, the second author, a native of the English language, made an extensive review of the citations.

Figure 2+3: Even though I appreciate the effort the authors put in Figure 2+3, it reports an overview of the characteristics of the study population. A structured table would be more appropriate, reporting the number, the denominator and the percentages. Rounded percentages are sufficient for these small numbers (n=32).

Answer: We appreciate your suggestion, but we would really like to keep the presentation of the data with this visual information, as this is an innovative and attractive way of presenting our sample data. This trend has been growing and holds more attention from the reader. I hope you understand.

Would the authors consider starting the discussion with the most important findings?

Answer: we edit the text, providing this information.

Which factors do the authors recommend to be included as part of the COS?

Answer: we edit the text, providing this information.

This is a relatively small sample, the author do hint briefly about the representativeness, can the authors discuss this in more detail, how their sample might be generalized?

Answer: we edit the text, providing this information.

Can the authors comment on the potential shift of priorities as the children age? Now they all seem to be still young, care needs/priorities will likely shift in the future.

Answer: we edit the text, providing this information.

### VERSION 2 – REVIEW

<b>REVIEWER</b>	Raphael Ximenes University Health Network, Canada
<b>REVIEW RETURNED</b>	11-Sep-2020

<b>GENERAL COMMENTS</b>	The authors made the changes suggested by me in my first review. That way I don't have any more editing/correction recommendations.
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<b>REVIEWER</b>	Michel Counotte Institute of Social and Preventive Medicine (ISPM), Bern, Switzerland
<b>REVIEW RETURNED</b>	08-Sep-2020

<b>GENERAL COMMENTS</b>	<p>The authors have sufficiently addressed most of the issues raised previously.</p> <p>However the point below has not been ignored:</p> <p>"The English translation of some of the verbatim text quoting the parents/caretakers is somewhat confusing, would the authors consider improving these translations (possibly back/forth translation to ensure the meaning is not lost)</p> <p>Answer: we took great care in that regard. The content of the focus groups in Portuguese was translated into English by a Brazilian pediatrician who at the time of the study was doing research at the University of Liverpool with Dr Gladstone. After this translation, the authors held a skype meeting to discuss with him possible doubts, to ensure that the participants' expressions were not lost. Upon your request, the second author, a native of the English language, made an extensive review of the citations."</p> <p>The authors claimed to have reviewed the quotes, but have not made any corrections. The verbatim text often remains confusing. For example this quote:</p> <p>"For E* which facilitates activities it's on first the glasses, because I take her glasses and the vision been worse, put the glasses and she animates." Mother 06.</p> <p>Could the authors translate these quotes into language the reader understands? I assume the child here does not see without glasses, but does interact when wearing glasses?</p>
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## VERSION 2 – AUTHOR RESPONSE

Reviewer 1 previously requested that more information about COMET is presented in the main text – such as the institutions that are involved and main aim of the trials (Please review language and sentence structure). This revision does not appear to have been carried out.

Answer: Corrections in the language were made by Dr. Gladstone and the requested information was added to the text.

The Core Outcomes Measures in Effectiveness Trials (COMET) is an initiative aimed at identifying and creating a core set of outcomes for any clinical health situation. This is often conducted through a process of systematic reviews of outcomes measured, consensus work as well as the involvement of families who support the development of these outcomes in order to ensure that researchers consider outcomes that are most relevant and appropriate to the patient's needs

Reviewer: 4

Reviewer Name: Michel Counotte

Institution and Country: Institute of Social and Preventive Medicine (ISPM), Bern, Switzerland

The English translation of some of the verbatim text quoting the parents/caretakers is somewhat confusing, would the authors consider improving these translations (possibly back/forth translation to ensure the meaning is not lost)

Answer: One of the English authors (Dr. Gladstone) made changes to the text and corrected the English.

Reviewer: 3

Reviewer Name: Raphael Ximenes

Institution and Country: University Health Network, Canada

Please leave your comments for the authors below

The authors made the changes suggested by me in my first review.

That way I don't have any more editing/correction recommendations.