

## PEER REVIEW HISTORY

BMJ Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form (Error! Hyperlink reference not valid.) 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>	The familial experience of acute bacterial meningitis in children A transversal qualitative study using interpretative phenomenological analysis.
<b>AUTHORS</b>	Scanferla, Elisabetta; Gorwood, Phlip; Fasse, Léonor

### VERSION 1 – REVIEW

<b>REVIEWER</b>	Omoleke, Semeeh World Health Organisation, Birnin Kebbi
<b>REVIEW RETURNED</b>	11-Apr-2021

<b>GENERAL COMMENTS</b>	<p>The review comments have been shared with the Editor.</p> <p><u>Reviewer's comments</u></p> <ol style="list-style-type: none"><li>1. What is the occurrence of meningococcal meningitis in the study setting?</li><li>2. May want to insert additional reference to line 13 regarding the epidemic potential of the meningococcal infection. Recent studies from the tropics could be cited such as Omoleke et al. BMC Public Health 2018, 18(Suppl 4):1318 <a href="https://doi.org/10.1186/s12889-018-6196-9">https://doi.org/10.1186/s12889-018-6196-9</a>; Basse BE, Vaz RG, Gasasira AN, Braka F, Weldegriberiel G, Komakech W, et al. Pattern of the meningococcal meningitis outbreak in Northern Nigeria, 2009. Int J Infect Dis. 2016;43:62–7 &amp; Lingani C, Bergeron-Caron C, Stuart JM, Fernandez K, Djingarey MH, Ronveaux O, et al. Meningococcal meningitis surveillance in the African Meningitis Belt, 2004-2013. Clin Infect Dis. 2015;61:S410–5.</li><li>3. Line 31 needs to be referenced</li><li>4. Line 44 should be referenced (perhaps this may be useful: Greenwood B. Editorial: 100 years of epidemic meningitis in West Africa - has anything changed? Trop Med Int Heal. 2006;11(6):773–80; Current status of cerebrospinal meningitis and impact of the 2015 meningococcal C vaccination in Kebbi, Northwest Nigeria. <u>Vaccine Volume 36, Issue 11</u>, 7 March 2018, Pages 1423-1428</li></ol>
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	<ol style="list-style-type: none"> <li>5. Sentence starting from line 44- 48 should be referenced appropriately. Further, the sentence needs to be revised and properly linked to the preceding idea.</li> <li>6. Typo on line 23 should be corrected</li> <li>7. Typo between lines 28 and 30 should be corrected- specifically an omission.</li> <li>8. “dyadic coping”- can this be unpacked for readers who are not core social scientists?</li> <li>9. Typo on page 14, line 7, it should be “medical professional”</li> <li>10. Preferably number less than 10 should be written in full.</li> <li>11. I would rather use “infection” and not “contamination” on line 22 page 16</li> <li>12. Grammatical issues on page 18 under physical sequelae</li> <li>13. Typo on page 19 “relative”</li> <li>14. Page 30 line 35 “children” and not “child”</li> <li>15. Spelling mistake in page 32- “meaning -marking process”</li> <li>16. Persistently the physicians’ training and familiarity with the meningitis is in question in this study and similar study from the same setting. This may have implications on outcome of hospitalization and the disease sequelae. CPD and medical training should be designed to improve the understanding of medics undergoing training or exchange programme with Health Institutions in the Tropics can be considered to improve their familiarity with the disease evolution.</li> <li>17. Recommendations based on findings should be a separate subsection to emphasise areas of improvement in clinical management, clinical psychology service and recovery processes post-discharge administration, social protection administration to deal or ease with financial burden on parents and waiting time. Also, the relevance of parent’s association for meningitis survivors was well articulated by the study participants as it improves knowledge and allows experience sharing regarding the various forms the disease could take, signs and symptoms, the importance of early detection and presentation to the health system. As per the study finding, the value of social support within the immediate family – from the husband/partner and from the “extended” family is commendable and highly recommended. Financial cost, impact on professional life of parents, and the frustration of proving the legitimacy of the psychological burden and the burden of caring for the meningitis survivors were notable and should be addressed by an appropriate authority.</li> </ol>
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<b>REVIEWER</b>	Lawrence, David
<b>REVIEW RETURNED</b>	Botswana-Harvard AIDS Institute Partnership, Clinical Trials Unit 18-May-2021

<p><b>GENERAL COMMENTS</b></p>	<p>General  This is a nicely written article and the authors are commended for starting to fill a large, important gap in the literature. There are a few important limitations to the study but these have been acknowledged throughout. The paper should be published after some minor changes and clarifications as outlined below.</p> <p>Abstract - perhaps elaborate more on the results to then give greater support to the conclusion</p> <p>Introduction  P4 L20 - closed bracket missing after 'under twenty-five years'.  P5 L30 - should this read 'in the mid and long term'?  There is no mention of research conducted with patients who survived and would be worth mentioning (if any)</p> <p>Methods  The methods were clear and nicely summarised the key principles of analysis  Can you please share the call for testimonies as a supplementary material? I think this is important as it may help the reader understand the motivation for taking part and therefore appreciate any potential bias in the sample.</p> <p>Results  You refer to the difference in sequelae as a limitation in the discussion and allude to the different outcomes with your included quotes but perhaps you could add something to the start of the results about the different type of sequelae (if any) that the children were left with.  In addition, what about the different location/hospital setting during the initial episode? Was there any difference between locations or level of hospital? I Did all of those have access to emergency psychological support for example?  P10 L5 - no space between 10 and mothers  The authors have left out the first theme - meningitis disease, but this could contextualise the illness within each of these experiences and the authors could consider including this unless they can justify that these data are not essential and/or the core findings are covered within over themes e.g knowledge/ignorance  P15 L50 - I would consider reviewing 'compensated for the passive attitude of healthcare professionals' and perhaps changing to '....perceived passive attitude....'  P16 L40 - Can you further contextualise this quote? I do not fully understand. Was this a case where a patient was given a terminal diagnosis but survived?  P21 L8 - 'If his...' - should this be 'If her...'?</p> <p>Discussion  As you have identified a significant mis-match between patients/relatives and healthcare professionals, could you comment further on that and the potential for further research and/or intervention to address this? Including from the perspective of the healthcare worker.  In terms of the benefits of family open visitation - was this not the case at the centres where your informants were attending? I think</p>
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	this is quite widely adopted for paediatric hospitals (where I work in the UK at least). So I am a bit surprised to see it as one of the primary recommendations. Perhaps the situation is quite different in France.
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## VERSION 1 – AUTHOR RESPONSE

Reviewer(s)' Comments to Authors:

Reviewer 1:

Dr. Semeeh Omoleke, World Health Organisation, Birnin Kebbi,

Comments to the Author(s):

1. What is the occurrence of meningococcal meningitis in the study setting?

Concerning the bacterial agent causing the disease, among the 11 participants in this study, 3 participants reported meningococcal, 3 reported pneumococcal, and 4 reported streptococcal bacterium. For one participant, the causative bacteria was not identified, although a meningitis diagnosis had been made by medical staff. For more details, please see Table 1.

2. May want to insert additional reference to line 13 regarding the epidemic potential of the meningococcal infection. Recent studies from the tropics could be cited such as Omoleke et al. BMC Public Health 2018, 18(Suppl 4):1318 <https://doi.org/10.1186/s12889-018-6196-9>; Bassey BE, Vaz RG, Gasasira AN, Braka F, Weldegriebriel G, Komakech W, et al. Pattern of the meningococcal meningitis outbreak in Northern Nigeria, 2009. Int J Infect Dis. 2016;43:62–7 & Lingani C, Bergeron-Caron C, Stuart JM, Fernandez K, Djingarey MH, Ronveaux O, et al. Meningococcal meningitis surveillance in the African Meningitis Belt, 2004-2013. Clin Infect Dis. 2015;61:S410–5.

Thank you for your valuable comment. The suggested references [Omoleke (2018) and Lingani (2015)] have been included in the revised manuscript.

3. Line 31 needs to be referenced

Thank you. As per your suggestion, we added the reference to the WHO's report on epidemic diseases. [https://www.who.int/gho/epidemic\\_diseases/meningitis/suspected\\_cases\\_deaths\\_text/en](https://www.who.int/gho/epidemic_diseases/meningitis/suspected_cases_deaths_text/en). May 2020

4. Line 44 should be referenced (perhaps this may be useful: Greenwood B. Editorial: 100 years of epidemic meningitis in West Africa - has anything changed? Trop Med Int Heal. 2006;11(6):773– 80; Ajibola et al., Current status of cerebrospinal meningitis and impact of the 2015 meningococcal C vaccination in Kebbi, Northwest Nigeria. Vaccine Volume 36, Issue 11, 7 March 2018, Pages 14231428

R.: Thank you for your comment. The suggested references have been added in the manuscript.

5. Sentence starting from line 44- 48 should be referenced appropriately. Further, the sentence needs to be revised and properly linked to the preceding idea.

As per your suggestion, we revisited the sentence and referenced it as follow:

[If major physical sequelae of childhood meningitis have been studied extensively, this is not the case with regard to the familial experience of bacterial meningitis in children. When tracking empirical studies dedicated to the familial experience of this disease, we note that a certain number of articles are devoted

to the issue of vaccination and the identification of potential barriers to it [7-9] ...]

6. Typo on line 23 should be corrected

The typo was corrected.

7. Typo between lines 28 and 30 should be corrected- specifically an omission.

Le sentence was corrected as follow:

[The psychological adjustment of parents after meningococcal disease was also quantitatively studied in the mid and long terms].

8. “dyadic coping”- can this be unpacked for readers who are not core social scientists?

Thank you for your comment. Le sentence was corrected as follow:

[(...) dyadic coping” in the context of illness, concept that refers to the stress management process in the context of a couple relationships, (...)]

9. Typo on page 14, line 7, it should be “medical professional”

Le sentence was corrected as follow:

[Therefore, they often felt discredited by the medical professional and expressed their frustration].

10. Preferably number less than 10 should be written in full.

Thank your suggestion. We wrote in full all numbers inferior to 10 appearing in the manuscript.

11. I would rather use “infection” and not “contamination” on line 22 page 16

We agree with your comment. Le sentence was corrected as follow:

[Therefore, many of the participants needed to acquire some form of expertise on the disease to learn the means and modalities of infection].

12. Grammatical issues on page 18 under physical sequelae

Le paragraph was modified as follow:

[Most of the interviews revealed the negative impact of meningitis on their family children and grandchildren health, and in particular the significant physical and cognitive sequelae associated with meningitis including partial paralysis, heart problems, hearing and visual impairments. These impairments significantly limited the survivors’ ability to function in an autonomous manner, and caused daily distress and frustration for the whole family].

13. Typo on page 19 “relative”

The typo was corrected.

14. Page 30 line 35 “children” and not “child”

The typo was corrected.

15. Spelling mistake in page 32- “meaning -marking process”

The typo was corrected.

16. Persistently the physicians’ training and familiarity with the meningitis is in question in this study and similar study from the same setting. This may have implications on outcome of hospitalization and the disease sequelae. CPD and medical training should be designed to improve the understanding of medics undergoing training or exchange programme with Health Institutions in the Tropics can be considered to improve their familiarity with the disease evolution.

Thank you for your thoughtful comment. After consultations, we added the mentioned need for the physicians’ training in the section “Clinical implication” section. Le paragraph was completed as follow:

[The value of the intervention of experts in the field of infectious diseases in the Continuing Professional Development of physicians, which would improve not only their familiarity with meningitis and its treatment, but also their capacity to interact with carers in the most appropriate and efficient way (18)].

17. Recommendations based on findings should be a separate subsection to emphasise areas of improvement in clinical management, clinical psychology service and recovery processes postdischarge administration, social protection administration to deal or ease with financial burden on parents and waiting time. Also, the relevance of parent's association for meningitis survivors was well articulated by the study participants as it improves knowledge and allows experience sharing regarding the various forms the disease.

Thank you for your suggestions. The role of parent's association was stressed. Regarding the recommendations, for more clarity, the authors decided to list them as follows:

[The findings in this study highlighted several main dimensions that could be of relevance:

- The mis-match between family members and healthcare professionals during the onset and course of the meningitis, as well as its management in health care facilities that should be given attention. A promising avenue for improvement, both from a research perspective and from a clinical standpoint, is the integration of expert patients into care procedures.
  - The value of the intervention of experts in the area of infectious diseases as part of the Continuing Professional Development of physicians, particularly of private general practitioners, which would improve not only their familiarity with the meningitis and its treatment, but also their capacity to interact with carers in the most appropriate and efficient way [14].
  - The role of patient and carer associations play in this regard, providing families emotional and psychological understanding and support. (...).
  - The implementation of experimental initiatives related to the family and siblings' visits in emergency care settings and intensive care units (ICU). (...)
  - The need for continuing efforts to consider and monitor the emotional burden of the disease on the child's family ascendants, the psychological adjustment process and families' trajectories over time (...).
- These findings seem to be of central importance for both clinical practice and research, demonstrating the importance of direct involvement of relatives of meningitis survivors in identifying key aspects of care and areas of improvement in clinical management, clinical psychology services and recovery processes post discharge].

Reviewer 2:

Dr. David Lawrence, Botswana-Harvard AIDS Institute Partnership, London School of Hygiene and Tropical Medicine Department of Clinical Research

Comments to the Author:

General

This is a nicely written article and the authors are commended for starting to fill a large, important gap in the literature. There are a few important limitations to the study but these have been acknowledged throughout. The paper should be published after some minor changes and clarifications as outlined below.

Thank you for your supportive comments.

1. Abstract - perhaps elaborate more on the results to then give greater support to the conclusion

Thank you for your thoughtful comment. Le paragraph was modified as follow:

[Six superordinate themes (Meningitis disease; Healthcare services and professionals; Knowledge /

ignorance; Repercussions of the meningitis experience - "Life afterwards"; Sick child attitudes / behaviour; Siblings attitudes / behaviour) and two main meaning-making processes in relation to the participants' experience of meningitis were identified: (1) the sick child becoming a "hero" – comparison with other children; (2) engaged action/attitude: finding the "positive" of the traumatic experience and engaged action to improve the care system. These two processes underpin the psychological adjustment to the meningitis and its consequences].

#### Introduction

2 - P4 L20 - closed bracket missing after 'under twenty-five years'.

The typo was corrected.

3- P5 L30 - should this read 'in the mid and long term'?

Yes, sure. The sentence was completed.

4- There is no mention of research conducted with patients who survived and would be worth mentioning (if any)

Thank you for your thoughtful comment. We referred to this point at the end of the introduction, citing the study of survivors' experiences described in a previous study of the author published by BMJ Open. This study is included in the references (Scanferla, 2020, e037168.full.pdf (bmj.com). Reference n. 18.

#### Introduction

[(...) A better understanding of the parents' subjective experience would make it possible to offer more assistance to the parents and satisfactory care for the children. Indeed, we already underlined the paucity of the empirical literature regarding the long-term adjustment of meningitis survivors during adolescence or adulthood in a previous research. Not only did we do so in terms of presence or absence of symptoms but also in terms of emotional reactions, feelings, memories, and meaning-making process in the mid and/or long term (18)].

#### Methods

The methods were clear and nicely summarised the key principles of analysis

5 - Can you please share the call for testimonies as a supplementary material? I think this is important as it may help the reader understand the motivation for taking part and therefore appreciate any potential bias in the sample."

Thanks for your comment. We have included the "call for testimonies" hereunder in appendix and submitted it as a supplementary material, as suggested.

#### Results

6.- You refer to the difference in sequelae as a limitation in the discussion and allude to the different outcomes with your included quotes but perhaps you could add something to the start of the results about the different type of sequelae (if any) that the children were left with.

In addition, what about the different location/hospital setting during the initial episode? Was there any difference between locations or level of hospital? Did all of those have access to emergency psychological support for example?

Thank you for your thoughtful remark. After reflection and consultation with the authors, we have withdrawn this sentence. Indeed, the clinical differences related to the participants' experiences (for example severity of the sequelae presented by their children, hospital setting ...) are not limits. On the contrary, in IPA, we are looking for diversity between participants having the same experience.

Le paragraph was completed as follow:

[(..) Thus, this study might not be representative of the wider spectrum of close family members who faced the meningitis disease. In future studies, a more diverse sample, including male participants, recruited from a plurality of treatment facilities and setting contexts of care, would be preferred and

provide a complete map of the meanings attached to subjective experiences and the generalisation of the study findings].

7.- P10 L5 - no space between 10 and mothers  
The mistake was corrected.

8.- The authors have left out the first theme - meningitis disease, but this could contextualise the illness within each of these experiences and the authors could consider including this unless they can justify that these data are not essential and/or the core findings are covered within over themes e.g knowledge/ignorance

Thank you for your comments. Please note that the core findings are covered within over the themes described in the manuscript. The theme missing (“meningitis disease”) was not essential in the present manuscript; nevertheless it was largely detailed in a paper referring to the same study/data and previously published by the authors. This study is included in the references (Scanferla, 2020, e037168.full.pdf (bmj.com). Reference n. 18.

9- P15 L50 - I would consider reviewing 'compensated for the passive attitude of healthcare professionals' and perhaps changing to '....perceived passive attitude....'

Thank you. We reviewed the sentence as suggested.

10- P16 L40 - Can you further contextualise this quote? I do not fully understand. Was this a case where a patient was given a terminal diagnosis but survived?

Thank you for your relevant comment. Yes, the mentioned young person survived. We think that this quote underlines that some diagnoses are shocking and that physicians should be cautious when they announce it to families. As diagnosis might be contradicted by the favorable evolution of the disease, as in the case of this mother's daughter.

After consultation with the authors, we have modified this sentence, as follow:

[The experience of a lack of responsiveness on the part of healthcare professionals caused frustration and feelings of helplessness in the patients' families. They also stress that some diagnoses are shocking and that physicians should be cautious in the way they phrase them as their diagnosis might be contradicted by the favourable evolution of the disease (...)].

11. - P21 L8 - 'If his...' - should this be 'If her...'?

Yes, thank you. The pronoun was corrected.

## Discussion

12.- As you have identified a significant mis-match between patients/relatives and healthcare professionals, could you comment further on that and the potential for further research and/or intervention to address this? Including from the perspective of the healthcare worker.

Thank you for this comment. Indeed this significant mismatch could be improved. After consultation, the authors decided to add to the “Clinical Implications” section the following paragraph:

[The mis-match between family members and healthcare professionals during the onset and course of the meningitis, as well as its management in health care facilities that should be given attention. A promising avenue for improvement, both from a research perspective and from a clinical standpoint, is the integration of expert patients into care procedures (...)].



13- In terms of the benefits of family open visitation - was this not the case at the centres where your informants were attending? I think this is quite widely adopted for paediatric hospitals (where I work in the UK at least). So I am a bit surprised to see it as one of the primary recommendations. Perhaps the situation is quite different in France.

Thank you for your thoughtful comment. After reflection and consultation with the authors, we have modified this sentence, as follow:

[Several authors have emphasized the positive impact of family open visitation and parental presence for the care of their child, especially in emergency care settings and intensive care units, even in extremely difficult contexts, such as the one experienced with the Covid pandemic (26, 27, 28)].

## Appendix

### Call for testimonies

#### Meningitis survivors and families wanted for telephone interviews

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“Petit Ange” is an association participating in a working group\* aiming to describe the long-term effects of meningitis. For that purpose the association is looking for people with meningitis and/or their family members.

To date publications have described the medical consequences of meningitis. However, impact of the disease on people's daily life, relationships with family and others, and professional career are not sufficiently known nor taken into account.

To this end the working group is looking for meningitis survivors and their families to interview either by phone or face-to-face in Paris.

Would you be interested in taking part in a phone or face-to-face interview in Paris, run by “Edusanté” and lasting approximatively 45-60 min,?

A 50-euro compensation is given for the interview.

The content of these interviews will of course remain anonymous and the association “Petit Ange” will share with you the overall results of the study, should you choose so.

(\*) - The working group consists of representatives from two associations, “Petit Ange” and “Méningite France”, a pediatrician, a psychiatrist, a sociologist, and an organization dedicated to patient education and support – “Edusanté”.

- The working group is independent and aims to improve knowledge about consequences of meningitis on the affected families. This research is funded by a pharmaceutical company, GSK.

### VERSION 2 – REVIEW

<b>REVIEWER</b>	Lawrence, David Botswana-Harvard AIDS Institute Partnership, Clinical Trials Unit
<b>REVIEW RETURNED</b>	21-Jun-2021
<b>GENERAL COMMENTS</b>	Thanks to the authors for the response letter, modifications and additional supplementary material.

	<p>I am sorry but I cannot see some of the changes outlined in the response letter in the modified document. These are queries 6, 10 and 12.</p> <p>In addition, the proposed response to 6 does not address the query about the different types of healthcare facility and the response to 13 does not address the query about what is the norm in France.</p>
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## **VERSION 2 – AUTHOR RESPONSE**

Thank you for sharing the comments of Dr. David Lawrence, reviewer 2.

As per his request, we have made visible in the modified document the following changes related to queries 6, 10 and 12.

In addition, we detailed the response to querie 6 (types of healthcare facility and the response) and 13 (norm in France).

For further details, please see the attached file "The subjective experience of meningitis Families - Responses to reviewers 21 6 21" v2 – Please contact the publisher for this file.

We stay at your disposal for any further comments and queries.