Author’s response to reviews

Title: Prevalence and factors associated with sleep disorders among children with cerebral palsy in Uganda; a cross-sectional study.

Authors:

Kisughu Munyumu (kisughumunyumu@gmail.com)
Richard Idro (ridro1@gmail.com)
Catherine Abbo (cathyabo180@gmail.com)
Mark Kaddumukasa (kaddumark@yahoo.co.uk)
Elly Katabira (katabira@infocom.co.ug)
EZEKIEL MUPERE (mupez@yahoo.com)
Angelina Kakooza-Mwesige (angelina_kakooza@yahoo.co.uk)

Version: 1 Date: 10 Jul 2017

Author’s response to reviews:

8/07/2017

Editor,

BMC Pediatrics ,

Thank you, so much for the review aimed at improving our manuscript, we have responded to the editor’s comments as shown below in italics.

Reviewer reports:

Reneva Petersen, M. D. (Reviewer 1): Thank you for submitting this article for review. There are a few things that need to be addressed for the paper to be ready for publication.

1. There are some grammatical errors and I would suggest maybe having someone formally edit the document for you to look at phrasing etc, to improve the flow.

Grammatical errors have been corrected throughout the text.
2. Abstract: Results: Look at phrasing for GMFCS. I know you mean the highest GMFCS score but this could be interpreted as the highest functional level. Could you look at rephrasing this to make it less ambiguous?

This has been rewritten as “lowest levels of gross motor function V (p = 0.001); OR:(95%CI), 7.980: (2.4 – 26.8) and IV (p = 0.025); OR:(95%CI), 7.607: (1.3 – 44.6) respectively and lowest manual ability (p<0.001) ; OR:(95%CI), 13.031: (3.1 – 55.2)”, see lines 42 - 45.

3. Background: This does not flow very well- you start with risk factors, then allude to importance of sleep but then come back to risk factors. May I suggest organising your background along themes of? Introduction: lines 51-54 as is (correct grammar) Importance of sleep and consequences of disrupted sleep evidence reviewed: health, QOL, family Factors in literature that have been found to contribute to sleep disorders Other review of literature you feel relevant like epidemiology of sleep disorders in other low income/African countries or lack of data. The sentence about sleep disturbances and mortality might be better in the latter section of the document: where you look at deaths during sleep.

The background has been rewritten and re-organized as suggested see lines 53 – 73.

4. Methodology: Setting: it might be nice to add? Population size that the hospital serves or the number of children reviewed in the service per annum to give the audience a sense of how representative this sample is of your total population. Line 100- 103: needs rephrasing, it does not read well Sample size: fine There is some duplication about consent: if you describe consent in study procedures, you do not have to mention it under study participants which will save some on your word count. ( line 99). You could possibly include a description of the sleep disturbances scale here: how many items, domains and cut off scores and validity etc.

The methods section has been rewritten, limiting the redundant text and including more descriptions of the scales used. See lines 92 – 159.

5. Results: Line 135: this has no meaning unless you explain under setting what the main population in your setting consists of and what other cultural groups might be seen here.

This has been rewritten as “The majority of participants 69% (93/135) were Baganda who are the largest tribe in Uganda.” See lines 175 – 176.
6. Discussion:

Line 166: this sentence is incomplete. I think you need to review your discussion for grammar and flow. Line 168: there must be a reference for this statement

When you report your prevalence and quote other settings try to summarize: our findings showed a higher prevalence as compared to other low income settings (Malaysia + reference) as well as high income settings like Europe. Line 170: this sentence is not clear, state what their reported prevalence is, and then your reasons for your higher prevalence (if your study had more severely disabled children-then state that in the European studies: x% vs your study were GMFCS IV and V to make the point)

This section of discussion has been rewritten as “This study set out to determine the prevalence and factors associated with sleep disorders among children with cerebral palsy in Uganda. The study found that, one-third of children with cerebral palsy attending Mulago have sleep disorders. Sleep disorders among this population were associated with severe gross or fine motor function level involvement or disability. This high prevalence of sleep disorders reported at 32%, is higher than what has been described in earlier studies such as Malaysia and several European countries, where the reported prevalence is between 10-25% (12). In Italy, the prevalence was reported as 13% (13) while in Ireland, a prevalence of 22.5% was reported (3). This might be due the differences in the patient populations, as patients in Mulago had more severe cerebral palsy compared to the other countries. Mulago which serves as a national referral tends to receive more seriously ill patients compared to other hospitals and this can influence the results.

The most common type of sleep disorders was Disorder in Initiating and Maintaining Sleep (DIMS) 27%. Others were Sleep-Wake Transition Disorder (SWTD) 13%, sleep hyperhydrosis 10%, Sleep Breathing Disorder (SBD) 10%, Disorder of Excessive Somnolence (DOES) 8% and then Disorder of arousal (DA) 4%. A similar pattern was found also in both the Malaysian and other studies mentioned above (3, 12, 14). Several co-morbid problems may affect the initiation of sleep including posture (increased risk of painful reflux), concurrent breathing problems, pain and constipation which are all common in children with CP.

Factors associated with sleep disorders among children with cerebral palsy

In this study, we found that bilateral, gross motor classification of level V or IV, manual ability of level V and presence of epilepsy were associated with sleep disorders. These patients had more severe functional motor limitation often characterized by bilateral spasticity, experiencing stiffness and contractures suggesting that severe disability is associated with sleep difficulties. Indeed, in the Italians study by Romeo et al, 48% of children with level V on the GMFCS and work reported by Sandella et al, shows that GMFCS predicted sleep problems (13, 15). The 10%
prevalence of hyperhidrosis in the present cohort, a marker of autonomic involvement also suggests a more severe injury and disease”. See lines 227 – 271.

7. Line 174- 176- you need a reference to base this statement on. Although sleep disordered breathing is a part of sleep disorders in children with CP: your findings did not include a high number of this, so how can you ascribe your higher prevalence to this?

This statement has been deleted from the text.

8. Line 177-180: include the percentages here to make it clear the difference between the types of sleep disorders.

The percentages have been added see lines 238 -266.

9. Line 181: you mention comorbid problems which have been found in other studies that affect initiation of sleep but you do not include any justification for why you did not look at this/ why this was not addressed in your study or whether you are planning to consider this in future research to address the issue.

A sentence proposing to study this has been included in the text see line 278 -279.

10. Line 192: I am not happy with your postulation that because the seizures were controlled this explains the lack of association. You need to support epilepsy which comes out so strongly in other studies is not associated in yours.

We have reanalyzed the data and rewritten this as shown on line 272.

11. Study limitations: I would have included lack of control population- it always helps to compare prevalence with local normal population. Your findings are also based at one institution and may not be generalizable to the rest of the country.

These have been rewritten as “The major limitation of this study is that a screening tool (Sleep Disturbance Scale for Children) was used to determine the prevalence of sleep disorders in this population. The Sleep Disturbance Scale for Children (SDSC) was also not validated for this community and there was no concurrent assessment for pain, a risk for sleep disturbance in
children with CP. Other limitations are lack of control population and findings based at one institution and may not be generalizable to the rest of the country.” see lines 284 – 289.

12. Conclusions

I do not think you can conclude that the prevalence of SDB for Uganda.

You have to specify that the prevalence in a cohort at a tertiary or teaching hospital in Uganda was …..

This has been rewritten as “Approximately one third of children with cerebral palsy have disorders of sleep in a cohort at a national referral and teaching hospital in Uganda. The most common type of sleep disorders in children with cerebral palsy was disorders in the initiation and maintenance of sleep (DIMS). Severe disability and presence of epilepsy were associated with Sleep disorders among children with cerebral palsy.” see lines 294 -298.

Kandavel K. Thennarasu (Reviewer 3):

1. The sample size calculation part needs more information on confidence level, precision of estimation (relative/absolute)

The sample size estimation has been rewritten as “A total sample size of 135 participants was estimated using the Kish Leslie (1965) formula for finite populations, based on a prevalence of sleep disorders in children with cerebral palsy of 22.5% by Newman CJ et al (3). The formula for the sample size of surveys i.e. the Kish Leslie (1965) formula below was used (adjusted for available population): sample size = n/1+n/N, where n =z2 x p (1-p)/e2.” See lines 159 – 163.

2. Since overall p value under bivariate analysis is not provided, the variables entered in the multivariable model to be stated explicitly.

This has been rewritten to show the cut offs for the included variables see lines 191 – 192.

3. Table 1: Grouping of children based on the age to be clearly stated. (Could be rounded off to closest year) and unit of measure to be mentioned

Table 1 has been corrected, see table 1 lines 388 – 389.
4. Table 2: What is a pathological total sleep score? It can be named as pathological sleep group based on total score and group name to be mentioned rather than score. This may go in to methodology.

Table 2 has been corrected see table 2 lines 405.

5. Table 3: care has not been taken for column headings.

These have been added see table 3. Line 415.

a. The columns of Sleep Disorder YES and NO seem to be named wrongly except for the Epilepsy.
b. Overall p values are to be given for all the variables. And in line no:151 it was mentioned that, all factors in table 3 and table 4 with a p-value < 0.2 on bivariate analysis were entered in the model.

c. GMFC - OR for level III is not reported.
d. Reference category for Epilepsy should have been NO category.

These has been corrected see table 3 to reflect the suggested changes.

6. Table 4:

a. AED data differ from the numbers mentioned in Epilepsy data of Table 3.
b. Caregiver educational level: The data does not tally to the total number. Seems to be entered manually,

Table 4 has been corrected to reflect the suggested changes.

7. Table 5:

a. Overall p value should be given for all the variables.
b. Bed sharing all night: Reference category needs to be changed.
This has been changed

8. Confidence interval for prevalence estimate could've been given. These have been added to the tables.

9. Line No 186: The word "independently" may be removed. This has been removed from the text.

10. Line no: 155, multivariate analysis can be replaced by multivariable model. This has been replaced.

Thank you,

Corresponding author

Kisughu Munyumu.