

PEER REVIEW HISTORY

BMJ Paediatrics Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Paediatric Neurodisability and Sleep Disorders: Clinical pathways and management strategies
AUTHORS	Joseph, Desaline; McDonald, Aoife

VERSION 1 – REVIEW

REVIEWER	Reviewer name: William Whitehouse Institution and Country: University of Nottingham, UK Competing interests: No competing interests to declare
REVIEW RETURNED	24-Oct-2018

GENERAL COMMENTS	<p>A powerful and useful paper. I think it should be published, with minor revisions. Congratulations.</p> <p>It highlights the importance of paediatric Sleep Medicine, while acknowledging the lack of expertise in the UK and patchy service provision. To many paediatricians and managers this is an "Unknown Unknown" which this paper will help transform into a tractable "Known Unknown".</p> <ol style="list-style-type: none"> Box 6 leave out all the details of the costs of melatonin. It is not a fair comparison as you do not include costs of other drugs or behavioural interventions. Key is to recommend withdrawing it when it fails to help adequately. Once published open access it will be read by many whose first language is not English and who have never worked in the UK. So spell out each abbreviation in full the first time it is used and then put the abbreviation in brackets after that. Every subsequent use of the term should be only as the abbreviation. Make the tables and boxes understandable in isolation so add a legend after each with the abbreviations explained. As there are so many abbreviations ask the Editor if you can include a "List of Abbreviations" to help unfamiliar readers. Use capital letters correctly, i.e. for proper names. The Editor can decide if e.g. they want "Sleep Medicine" or "sleep medicine" but be consistent. <p>Minor corrections:</p> <ol style="list-style-type: none"> Dr Joseph has MRCPCH not RCPCH, and is a "consultant in...." not "consultant..." Delete the extra ", ." on page 2 line 25. Change "seizure disorder" to "epilepsy" page 2 line 28. Change "...been..." to "...being..." page 4 line 19. Change "...or..." to "...are..." page 4 line 25. Change "...child..." to "...children..." page 5 line 8. Tidy the bottom of box 1, page 5 line 52. Change to "...improve the management of..." page 6 line 3. Close brackets
-------------------------	--

	<p>10. Change "...seen..." to "...disorder..." Box 5 line 7.</p> <p>11. Change "...measure ..." to "...measures..." and make it "...considered..." Box 6 line 35.</p> <p>12. Delete "...which..." page 13 line 31.</p> <p>13. Delete "...around the crisis..." page 13 line 41.</p> <p>14. Change "...cannot be underestimated..." to "...is vital...".</p> <p>Something of very high value or esteem "cannot be over estimated"; but something worthless or of low esteem "cannot be under estimated". Conversely something of very high value or esteem "must not be under estimated"; and something worthless or of low esteem "must not be over estimated". Better to avoid this use of language and keep it simple.</p>
REVIEWER	<p>Reviewer name: Cathy Hill</p> <p>Institution and Country: University of Southampton/Southampton Children's Hospital, UK</p> <p>Competing interests: None. Have received no obligation loan of oximeter for prior research from Masimo Inc</p>
REVIEW RETURNED	08-Nov-2018
GENERAL COMMENTS	<p>Well done to addressing a really important topic!</p> <p>The case studies are an excellent way of leading the reader through a diagnostic approach</p> <p>I have some detailed comments about the paper which I will submit to the editor by way of a track-changed/comments pdf - some of these relate to phrasing or grammar -some are more factual. Please contact me if you do not receive these (cmh2@soton.ac.uk)</p> <p>My principle suggestions relate to the following:</p> <ol style="list-style-type: none"> 1. In tables you use the term sleep disorders but then list a mix of symptoms and disorders 2. The use of the terms neurodevelopmental disorder/neurogenetic disorder and neurodisability are used inter-changeably - I would suggest either consistency or clear definitions early in the manuscript. 3. The table linking neurodevelopmental disorders to sleep disorders is great but it is odd that the largest groups of children we see (ADHD/ASD and CP) do not feature - it may be that you wanted to focus on the neurogenetic disorders here but if so I suggest you are clearer in your intro that ASD and physical disabilities e.g. CP numerically are most likely to present - indeed you use these as your case studies. Also there are some notable syndromes missing e.g. Williams syndrome and I was not clear for your rationale for including some incredibly rare disorders most DGHs are extremely unlikely to see - there is a risk that this makes the article less rather than more accessible and paediatricians can readily look these up themselves 4. I struggled with the ASD case as it was presented as a non-24 hour circadian rhythm disorder which would be very rare in practice - I suggest you re-frame this as a more typical DSWPS case - also be careful about the use of the term chronotherapy. Be careful to define acronyms and explain different sleep studies for the uninitiated - a text box with simple definitions would be helpful - also be careful to refer to ERJ European guidelines for OSA diagnosis - see comments again <p>With some revisions this will make a really helpful article for the general reader</p>

VERSION 1 – AUTHOR RESPONSE

Cathy Hill Comments:

1. Introduction paragraph 2: Missing reference–Reference added to list of contributing factors for sleep issues in neurodisability.
2. Introduction paragraph 2 :Neurodisability/neurogenetic/neurodevelopmental used interchangeably - 6 replacements made with term neurodisability to make consistent
3. Introduction paragraph 2: Missing definition for acronym OSA – OSA defined as Obstructive Sleep Apnoea
4. Table 1 state rationale for using these conditions – the initial rationale was to include less common conditions seen in neurodisability practice that may have a non -behavioural component to sleep problem (rarer and more specialist conditions). However we have taken on board reviewer's comments and added in the more common neurodisability conditions as well as adding in more non respiratory presentations to give a broader scope to what the table includes which may be used as a quick reference guide in clinic for paediatricians in secondary care. Suggestions regarding labelling columns in the table and modifying the number and nature of the conditions featured have been taken on board. More common neurodisability conditions have been included and much rarer conditions omitted. We have created separate columns for respiratory and non-respiratory sleep conditions and have tried to add very brief notes for some conditions that need special consideration.
5. Title of table 1 – use of term neurodevelopmental too general – changed to neurodisability
6. Table 1 content: grammatical adjustments made
7. Baseline knowledge in sleep medicine page 4 paragraph 1: 'our training' changed to 'paediatric training;' 'for undergraduate' removed from 2nd sentence para 1;
8. Under same heading page 4 paragraph 3: added location and time of local survey (Evelina paediatricians) 2018; ' knowing what sleep studies is' changed to 'knowing what sleep studies are'
9. Table 2 Title: changed 'physician' to 'paediatricians'
10. Subheading in main text (bottom page 4) 'Improving access....': changed to 'Improving access to services....'
11. Para 1 page 5 under heading 'Improving access to services': 'sleep physiologist or sleep practitioners' – the term 'or' was removed and sleep practitioners defined; 'clinical' added to psychology; 'psychology' changed to psychologists'
12. Box 1 page 5: Title 'Training and support for parents and allied professionals' changed to 'Training and Support in Sleep medicine'
13. Box 1 page 5: incorrect link/web address for Southampton course removed and course changed to 'four day' course plus names of courses amended
14. Box 1 page 5: amendments made regarding listed questionnaires and descriptions of sleep investigations added
15. Case 1 page 6 2nd paragraph: comma added after reflux
16. Case 1 page 7 1st paragraph: the family 'was' going into crisis changed to 'is' going into crisis

17. Box 3 page 8 : info regarding sunrise lamp added as well as use of bedtime passport as reward/incentive system
18. Box on melatonin moved to box 4
19. Box on clonidine moved to box 5
20. Box 5 page 9 removed hypnic jerks from indication for clonazepam ; also removed mention of imipramine and carbamazepine for hypnic jerks
21. Page 10 paragraph 2: HPC second sentence amended as suggested 'This following a traumatic incident in school that he 'put himself to bed' in order to avoid schooling.'
22. Page 11 Q1: changes made as follows C. Sleep 'related' Movement disorder D. Obstructive sleep apnoea
23. Case 2: It was suggested that possible this case should be changed or simplified. We have decided to slightly simplify the language but to keep the core message of the case the same. This is a rarer case but is seen and knowledge around the difficulties surrounding the case are important for Paediatrics to know and understand. We have asked some junior trainees to proof read and they have been able to follow and learn from the case. Our desire is that the reader will be able to generalise the skills of using a diary to fine tune the sleep diagnosis, liaising with school and using general principles of management of a circadian disturbance will be a useful educational outcome. We hope this is acceptable.
24. Box 5 changed to Box 6: Circadian rhythm disturbance - content changed in line with suggest made regarding social withdrawal/anxiety rather than pure DSPS
25. Box on melatonin: added ASD as listed condition for indication for melatonin; added Neurim reference and defined DSPS as circadian rhythm disorder (now box 4)
26. Conclusion page 13 paragraph 3: 'Psychology is key' changed to 'access to psychological and behavioural interventions is key'

William Whitehouse comments:

1. Box 6: We value the opinion of the reviewer regarding the cost of melatonin. We acknowledge that cost of other sleep medications have not been included. Melatonin is regularly the drug of choice and is used judiciously by many paediatricians. Highlighting cost may help with business case planning and support local trusts to dialogue with stake holders for modest investment in sleep services including the training of sleep practitioners and supporting paediatricians' sleep education. We have added the following sentence – 'Cost benefit should be considered and the above figures may be used to help support business case development for local sleep services including trained sleep practitioners' Exact costs of melatonin spend have been removed.
2. Once published open access it will be read by many whose first language is not English and who have never worked in the UK. So spell out each abbreviation in full the first time it is used and then put the abbreviation in brackets after that. Every subsequent use of the term should be only as the abbreviation. Make the tables and boxes understandable in isolation so add a legend after each with the abbreviations explained. As there are so many abbreviations ask the Editor if you can include a "List of Abbreviations" to help unfamiliar readers. This has been completed, if it would be useful to add.

3. Use capital letters correctly, i.e. for proper names. The Editor can decide if e.g. they want "Sleep Medicine" or "sleep medicine" but be consistent. Sleep Medicine listed with capital letters and proper nouns have been checked.

Minor changes:

Minor corrections:

1. Dr Joseph has MRCPCH not RCPCH, and is a "consultant in...." not "consultant...". DONE
2. Delete the extra ", ." on page 2 line 25. DONE
3. Change "seizure disorder" to "epilepsy" page 2 line 28. DONE
4. Change "...been..." to "...being..." page 4 line 19. DONE
5. Change "...or..." to "...are..." page 4 line 25. DONE
6. Change "...child..." to "...children..." page 5 line 8. DONE
7. Tidy the bottom of box 1, page 5 line 52. DONE
8. Change to "...improve the management of..." page 6 line 3. DONE
9. Close brackets
10. Change "...seen..." to "...disorder..." Box 5 line 7. SENTENCE REMOVED
11. Change "...measure ..." to "...measures..." and make it "...considered..." Box 6 line 35. DONE
12. Delete "...which..." page 13 line 31. DONE
13. Delete "...around the crisis..." page 13 line 41. DONE
14. Change "...cannot be underestimated..." to "...is vital...". DONE