PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	Pilot Study of an Integrated Model of Sleep Support for Children: A Before and After Evaluation
AUTHORS	Elphick, Heather; Lawson, Candi; Ives, Ann; Siddall, Sue; Kingshott, Ruth; Reynolds, Janine; Dawson, Victoria; Hall, Lorraine

VERSION 1 – REVIEW

REVIEWER	Reviewer name: Peter Flom Institution and Country: Peter Flom Consulting USA Competing interests: None
REVIEW RETURNED	19-Jul-2019

GENERAL COMMENTS	I confine my remarks to statistical aspects of this paper. I have some concerns that need to be addressed before I can recommend publication. p 6 The authors seem to have data on multiple time points. Why did they then do a pre-, post-, analysis? This is a very weak design and subject to fairly severe regression to the mean effects, esp. since most of the measures are self-report and therefore subject to bias. what does the last sentence in the 2nd paragraph mean? line 52 ff What if the parents don't know when the child woke or how long he/she too to go to sleep? p 7 LOCF is not a great method (multiple imputation is better) but the data are likely to be missing not at random, so nothing really works very well. At least the authors did a per protocol analysis. What kind of t-tests were used? Paired or independent? And some of the measures are not appropriate for a t-test. Why not use a multilevel model, which would let you explore for independent variables (like age, sex, etc). Table 1 - there seems to be a typo for the MD for time to sleep in the bottom panel.

REVIEWER	Reviewer name: Dr Desaline Joseph Institution and Country: Children's Sleep Department Evelina London Children's Hospital St Thomas' Hospital London England United Kingdom Competing interests: None
REVIEW RETURNED	14-Aug-2019

GENERAL COMMENTS	This is a well written piece in an under researched but clinically important area. Work regarding sleep interventions is important and this paper in particular is attractive as it shows work across agencies for high risk groups of children and young people. The authors are to be congratulated. On page 4 (line 4-8) at start of the methods is it possible to make it clearer what the study design is. Is it an observational study? With the absence of a control group or 'waiting list controls' how was the sample size determined? Was there any bias in selection of the participants? How was that managed? Can this be expanded/explained in the discussion? Was the recruitment serial? Was it from a ADHD clinic or a LAC list? Is this group representative of the denominator population? Were the participants known to the sleep practitioners before hand? The exclusion criteria was not entirely clear - how were 'other issues taking priority over sleep problems' defined and standardised? Who made that decision and how? How was the screening done for pre-existing medical and specific sleep disorders? If a child had for example a history of sleep walking, night terrors or nightmares where they excluded or included? In terms of the split in cases between clinical and Looked After Children (LAC) recruitment (p 9 line 20,21 results) - 42 had ADHD and 14 were looked after children. Was that the intended ratio? Can the authors in discussion make any comments about the much smaller percentage of children and young people that were in the LAC group? What were the barriers to recruitment from this source? Were the children with ADHD sourced via the Local Authority or were they from a clinical NHS setting? The paper reads as though the entire study is set in the Local Authority rather than the NHS. Maybe this can be highlighted more in the discussion as an achievement. On page 9 line 31-38- results "All 45 participants that started the intervention completed it. Six participants dropped out after completing the intervention and did not complete the follow-up evaluation. One carer was not able to follow the bedtime routine and so dropped out of the project. The other five families disengaged without giving a reason. " Later you state 39 completed final evaluation. Is the intervention the clinic/workshop only or does it include the follow up telephone support ? This section could be slightly clearer. page 10 line 17-24 - What were mean/median scores and range for SDQ? Did any of the children or young people screen indicate that they had any emotional/mental health issues? Is there a reason this has been excluded from the reporting of the results? Did any onward referrals need to be made? Was a mental health issue not part of the exclusion criteria? Why or why not if other conditions were excluded? Also were the baseline diagnoses of ADHD confirmed in any way at start of recruitment? the results table on page 11 line 10 onwards (Table 1) very clear. page 12 line 53,54 - The inclusion of the implementation model is very welcome.

	Is there room in the discussion to comment on what barriers to implementation may be and any suggestions on how to overcome them? Page 15 line 21,22 "Caution is needed when interpreting the results of this uncontrolled before and after study. " Is there a more scientific term to be used than "uncontrolled before and after study"? page 15 line 41-48 " Analysis taking into account withdrawals was therefore also carried out based on the assumption that baseline data would remain unchanged without the intervention and using imputed values for the final outcome measure by carrying forward the baseline value for research participants lost to follow- up. " Is this a recognised technique or form or analysis to include data from those that dropped out? Can you add reference if so? I would be interested to hear a statistician's comment on this approach as being valid or invalid. p16 line 19,20 "Aligning this process and different organisational cultures and expectations was more challenging..." - further comment on this maybe of interest to the reader as to what challenges were and how study would be redesigned in future. This is a unique and novel aspect to this work and may be worth expanding as the authors have significant insight and experience in this area.

REVIEWER	Reviewer name: Dr Anna Weighall Institution and Country: School of Education, University of Sheffield Competing interests: None. I know Prof Elphick, Dr Kingshott and Ms Dawson in a professional capacity and I am aware of their work. However, I have not been involved with the research reported here at any stage.
REVIEW RETURNED	08-Sep-2019

GENERAL COMMENTS	This is an important and much needed piece of work which is of timely importance locally nationally and worldwide. The modelling for inter-agency working and delivery plan is to be commended. I enthusiastically recommend this paper for swift publication.

VERSION 1 – AUTHOR RESPONSE

Reviewer 1 (Comments to the Author):

p 6 The authors seem to have data on multiple time points. Why did they then do a pre-, post-, analysis? This is a very weak design and subject to fairly severe regression to the mean effects, esp. since most of the measures are self-report and therefore subject to bias.

Time points varied from patient to patient depending on the complexity of the case and the number of contacts needed for each individual family. It was therefore decided that, rather than analysing according to non-standardised time points, that only pre- and post-evaluation time points would be analysed. The post-evaluation time point was defined as the point at which both the parent/child and practitioner considered that the intervention had been either successful or unsuccessful (see below). We accept that this is a weak aspect of the study design, but the nature of the intervention did not allow fixed time points.

We have included some text to explain this in the limitations section of the discussion.

what does the last sentence in the 2nd paragraph mean?

"….a level that was considered to be a successful or unsuccessful intervention by the parent/young person and practitioner" was the point at which no further input from the practitioner was deemed beneficial, ie the primary goal score was no longer improving. This point was not strictly pre-defined but was decided by the same two practitioners throughout the study and was therefore as consistent as possible within the limits of clinical practice.

We have added some clarification to the text.

line 52 ff What if the parents don't know when the child woke or how long he/she too to go to sleep?

We accept that this measure is subjective and may be inaccurate. However, in asking the same parent to assess their child before and after the intervention it was deemed to be as accurate as possible in the absence of an objective measure. Our experience of objective measurements of sleep duration such as actigraphy is that children's sleep is disturbed by the presence of the monitor and although this method was considered, it was not feasible within the scope of the study and therefore subjective measures were used.

We have added clarification to the text in the discussion.

p 7 LOCF is not a great method (multiple imputation is better) but the data are likely to be missing not at random, so nothing really works very well. At least the authors did a per protocol analysis.

Thankyou, we considered this a reasonable approach to accommodate the missing data.

What kind of t-tests were used? Paired or independent? And some of the measures are not appropriate for a t-test. Why not use a multilevel model, which would let you explore for independent variables (like age, sex, etc).

We used independent-tests and have clarified this in the text. We used simple statistical methods as we did not feel that more complex methods were justified, given the missing data and the weaknesses in the study design.

Table 1 - there seems to be a typo for the MD for time to sleep in the bottom panel.

Thankyou for pointing out this error. The correct values should be MD 1.43; 95% CI 0.95-1.91 and the table has been changed accordingly.

Reviewer 2 (Comments to the Author):

This is a well written piece in an under researched but clinically important area. Work regarding sleep interventions is important and this paper in particular is attractive as it shows work across agencies for high risk groups of children and young people. The authors are to be congratulated.

Thankyou

On page 4 (line 4-8) at start of the methods is it possible to make it clearer what the study design is. Is it an observational study? With the absence of a control group or 'waiting list controls' how was the sample size determined? Was there any bias in selection of the participants? How was that managed? Can this be expanded/explained in the discussion? Was the recruitment serial? Was it from a ADHD clinic or a LAC list? Is this group representative of the denominator population?

Were the participants known to the sleep practitioners before hand? The exclusion criteria was not entirely clear - how were 'other issues taking priority over sleep problems' defined and standardised? Who made that decision and how? How was the screening done for pre-existing medical and specific sleep disorders? If a child had for example a history of sleep walking, night terrors or nightmares where they excluded or included?

Thankyou for these comments. We have responded to each and made changes to the document. We hope that our responses will clarify the methodology.

- It was an observational study – added to methods section.

- Sample size is explained on p7. We hope this is sufficient explanation.

- Participants were selected sequentially on referral from an ADHD clinician or key worker dealing with LAC who felt that the child/family would benefit from the intervention. Inclusion criteria were checked by the research team prior to recruitment. This has been added to the recruitment section in the methods.

- The study sample was not representative of the denominator population in terms of all children with ADHD or LAAC; children were referred on the basis of clinical need.

- Participants were not known to sleep practitioners before referral to the project and only had contact for the duration of the project. This has been added to the recruitment section in the methods.

- Exclusion criteria – children were excluded if it was felt that the sleep disturbance had a medical basis that should be prioritised over the sleep support intervention – this was screened for by the practitioners at initial assessment and discussed with the consultant (HE) as necessary. HE made the final decision as to whether or not they were included. “Issues taking priority over sleep” were factors such as clinical (physical or mental health) or social life-events that would have interfered with the implementation of the sleep intervention within the time-frame of the project. We therefore did include children with other sleep problems such as sleep walking etc if it was considered that the child may still benefit from the sleep support intervention with no other concerns identified. This has been added to the recruitment section in the methods.

We appreciate these comments, we hope that the description of the recruitment process is now clearer for the reader.

In terms of the split in cases between clinical and Looked After Children (LAC) recruitment (p 9 line 20,21 results) - 42 had ADHD and 14 were looked after children. Was that the intended ratio? Can the authors in discussion make any comments about the much smaller percentage of children and young people that were in the LAC group? What were the barriers to recruitment from this source? Were the children with ADHD sourced via the Local Authority or were they from a clinical NHS setting?

We did not specify an intended ratio of children with ADHD:LAAC in the study design. Recruitment was opportunistic and relied on an initial approach and referral to the project by a member of the child’s clinical team (for ADHD patients) or key worker (for LAAC). The recruitment of participants was therefore dictated by the referral rate possible within the timeframe of the project. A large amount of time was devoted at the beginning of the project to visit the appropriate agencies with information about the project and recruitment process. Barriers to recruitment were largely centred around the availability and engagement of the referring staff with some expressing a great deal of enthusiasm for the project and others citing lack of time and changes in management structure as barriers to engagement.

We have added some text to the discussion to explain these barriers in more detail, thankyou.

The paper reads as though the entire study is set in the Local Authority rather than the NHS. Maybe this can be highlighted more in the discussion as an achievement.

Thankyou. The project intervention was delivered by practitioners in the local authority but is easily transferrable to NHS services. We have added some text to this effect in the discussion.

On page 9 line 31-38- results

"All 45 participants that started the intervention completed it. Six participants dropped out after completing the intervention and did not complete the follow-up evaluation. One carer was not able to follow the bedtime routine and so dropped out of the project. The other five families disengaged without giving a reason. " Later you state 39 completed final evaluation. Is the intervention the clinic/workshop only or does it include the follow up telephone support ? This section could be slightly clearer.

Thankyou, we have added some text which we hope will clarify this for the reader. 45 completed the intervention (including telephone support). 6 dropped out between completion of the intervention and the final evaluation. Reasons for this were given as unable to continue with the routine (1) or not given (5). 39 completed the final evaluation.

page 10 line 17-24 - What were mean/median scores and range for SDQ? Did any of the children or young people screen indicate that they had any emotional/mental health issues? Is there a reason this has been excluded from the reporting of the results? Did any onward referrals need to be made? Was a mental health issue not part of the exclusion criteria? Why or why not if other conditions were excluded? Also were the baseline diagnoses of ADHD confirmed in any way at start of recruitment?

The reason for exclusion of these results was based on word count limitations. As there was no significant change it was considered to be of less interest to the reader. Results of our SDQ analysis are attached as a separate document with median scores and range as requested by the reviewer. We have not included this in the paper, but are happy to include as supplementary material if required.

Mental health issues were only excluded if they were felt to be influencing the ability to take part in the intervention. One child did develop significant mental health problems and was unable to participate in the intervention despite completing the baseline evaluation. No further issues that raised concern arose or were identified on screening.

ADHD referrals were received from the clinical team and confirmation of diagnosis was not sought.

the results table on page 11 line 10 onwards (Table 1) very clear.

Thankyou

page 12 line 53,54 - The inclusion of the implementation model is very welcome. Is there room in the discussion to comment on what barriers to implementation may be and any suggestions on how to overcome them?

Thankyou for this suggestion. There was a great deal learned about the implementation of both the intervention within families and of the service model, as well as the logistics of a combined research approach within Local Authority and NHS services. There is insufficient room to allow a detailed discussion of all of these points and they will be reported in a separate paper. A brief summary has been included in the discussion as follows (if the editors are happy to allow the extended word count):

Barriers to the implementation of the intervention to families included engagement of the young person (usually around negotiating removal of technology), finding the optimum time at which to introduce the programme around other events at home or other therapies taking priority, parental tiredness and mental state and logistics such as other children in the home or lack of support for the parent. Parental motivation was another factor as many felt they had tried sleep support before or believed that other issues such as the ADHD diagnosis would prevent the intervention from being helpful. The skill needed to motivate parents and young people beyond their initial beliefs is a requirement of a sleep practitioner as well as a knowledge of sleep. A consistent and whole household approach is crucial, along with appropriate timing.

Barriers to the implementation of the service model were twofold - workforce and training resources and engagement of services and individual staff. However, since oral dissemination of the results in our region has taken place some of these barriers are being overcome.

Page 15 line 21,22 "Caution is needed when interpreting the results of this uncontrolled before and after study. " Is there a more scientific term to be used than "uncontrolled before and after study"?

Thankyou, we have changed this to “observational”

page 15 line 41-48 " Analysis taking into account withdrawals was therefore also carried out based on the assumption that baseline data would remain unchanged without the intervention and using imputed values for the final outcome measure by carrying forward the baseline value for research participants lost to follow- up. " Is this a recognised technique or form or analysis to include data from those that dropped out? Can you add reference if so? I would be interested to hear a statistician's comment on this approach as being valid or invalid.

Please refer to the comments of reviewer 1 (statistics expert) and our responses.

p16 line 19,20 "Aligning this process and different organisational cultures and expectations was more challenging..." - further comment on this maybe of interest to the reader as to what challenges were and how study would be redesigned in future. This is a unique and novel aspect to this work and may be worth expanding as the authors have significant insight and experience in this area.

Thankyou, we agree and will expand further in a separate paper. A brief summary has been included in the discussion, as summarised in the point above regarding barriers to implementation.

Thankyou once again to the reviewers for these insightful comments. We look forward to learning the editorial decision on the manuscript.