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CLINICAL EXPERIENCE

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Caregiver acceptability of a UK trial for paediatric sleep disordered breathing: A qualitative interview study

1 | INTRODUCTION

Paediatric sleep disordered breathing, a general term for breathing difficulties during sleep, can range from frequent loud snoring to obstructive sleep apnea (OSA), a condition where part, or all, of the airway is blocked repeatedly during sleep.[1] Paediatric sleep disordered breathing is usually caused by large adenoids and tonsils, and the commonest treatment in the UK is adenotonsillectomy.[2] While serious complications from adenotonsillectomy are low, bleeding and return to theatre are relatively common.[3] Mortality is very rare, but can occur in up to 1:26683 children.[4] Twelve percent of 4–5 year olds snore and up to 2% have OSA in the UK.[1] Cross sectional and longitudinal data highlight significant quality of life issues for both children and their families.[5] Remarkably, given the impact of the paediatric sleep disordered breathing spectrum on quality of life, and the need to balance benefits of this treatment with the risks of intervention, worldwide there have been only two large scale randomised controlled trials.[6,7] However, these were skewed samples confined to polysomnography proven OSA.[6,7] There is little evidence about the most effective and safest way to treat children with sleep disordered breathing[8] or whether treatment is always necessary. We aimed to investigate the acceptability of performing a trial for paediatric sleep disordered breathing in the UK in terms of whether caregivers would hypothetically participate.

2 | MATERIALS AND METHODS

Approval was obtained from: South Central—Hampshire B Research Ethics Committee (ref number 18/SC/0378); Health Research Authority (IRAS ID: 239892); Newcastle Upon Tyne Hospitals NHS Foundation Trust (R&D number: 8841). This study is reported according to the consolidated criteria for reporting qualitative research.

A clinical secretary screened ENT appointments each month to identify eligible patients aged 2–9 years with sleep disordered breathing symptoms referred to the clinic. Appointment letters included a study information sheet. At appointment parents of patients consented to the collection of a small amount of clinical data about their child and to complete validated questionnaires consisting of: T14, a 14-item parent-reported outcome questionnaire for paediatric throat disorders; OSA-18, an 18-item parent-reported evaluation of sleep disordered breathing; PedsQL, a 28-item assessment of a child's quality of life. Patients with serious comorbidities or existing health conditions were excluded. With consent, parent contact details were passed to the study team who invited them to participate in an in-depth qualitative interview.

Parents were purposively sampled to ensure variation in regard to gender and age of child as presentation of sleep disordered breathing differs by these characteristics. Gender also influences decisions to participate in trials. GPs who might refer children to ENT and hospital doctors involved in treating children with sleep disordered breathing were approached by the study team through known contacts as a convenience sample. Judgements about sample size and data saturation in thematic analysis are subjective, and therefore could not be determined (wholly) in advance of analysis but based on previous research experience it was estimated that around 20 interviews would provide sufficient data.

All participants were sent a letter confirming their interview with an information sheet and a consent form signed prior to the interview. A study identification number was allocated to each participant. Parent participants received a £10 high street shopping voucher to compensate for expenses. Interviews were carried out either face-to-face or over the telephone at the convenience of the participant and were not repeated. Interviews lasted up to 1 h and were semi-structured based on flexible topic guides (see Supporting Information). Piloted topic guides, based on literature and discussion among the study team, explored symptoms, effects, management and treatment of paediatric sleep disordered breathing (reported elsewhere[9]) plus views about data collection tools and willingness to be randomised into a surgical trial (reported here). Interviews, conducted between September 2018 and April 2019 by a trained and experienced female social scientist and qualitative researcher unknown to the participants, were audio recorded, transcribed verbatim, anonymised and coded (managed using NVivo). Field notes were not made. Coding was then discussed as a team to develop themes. Participants did not provide feedback on transcripts or findings.

3 | RESULTS

Eleven parents (out of 23 approached) participated (three declined, six unable to be contacted, three lost contact). Five parents were

TABLE 1 Parent participants.

Number	ID code	Parent	Child gender	Child age (years)
1	P1	Mother	Male	9
2	P3	Father	Male	5
3	P4	Mother	Male	3
4	P6	Mother	Female	4
5	P8	Mother	Female	7
6	P9	Mother	Male	9
7	P10	Mother	Female	4
8	P11	Mother	Female	9
9	P12	Mother	Female	4
10	P13	Mother	Male	8
11	P15	Mother	Female	6

TABLE 2 Health professional participants.

Number	ID code	Role	Gender
1	GP1	GP, ad-hoc primary care GP sessions	Male
2	GP2	Senior partner in GP practice	Female
3	GP3	Locum GP	Female
4	GP4	Practice GP	Female
5	GP5	Practice GP	Male
1	HD1	General and respiratory paediatrician	Female
2	HD2	Consultant ENT surgeon	Male
3	HD3	Respiratory paediatrician	Female
4	HD4	Consultant ENT surgeon	Male
5	HD5	Consultant in respiratory paediatrics	Male
6	HD6	Consultant ENT surgeon	Male

interviewed face-to-face and six over the telephone (see Table 1). Five GPs and six hospital doctors also participated, most were interviewed over the telephone, with two face-to-face interviews (see Table 2).

Themes are presented below with illustrative quotations.

3.1 | Acceptability of a trial for paediatric sleep disordered breathing

Despite not seeing many children with sleep disordered breathing, all GPs reported willingness to refer eligible children into a surgical trial of sleep disordered breathing:

> "…if there are trials going on that can improve the health and wellbeing of the population, and contribute to new research, I don't have a problem with referring unless I have significant ethical concerns about the study, or I think the science is dubious." (GP1)

All hospital doctors were supportive of a trial although they acknowledged the inherent difficulties such as cost, parental preference, eligibility criteria, length of follow up, objective outcome measures, seasonable effects and generalisability. However not all hospital doctors were in clinical equipoise:

> "I appreciate that there's no evidence, but we all believe it works, or we wouldn't be referring children for it." (HD3)

While most parents were willing for their child to be randomised into a trial there were four parents who would be reluctant due to the risk that it would either delay surgery or that it would force them into surgery that they did not want:

> "…just for the sake of getting them [tonsils] out and seeing if it works, I wouldn't do it. I would rather wait." (P11)

Key points

1. There is no conclusive evidence about the safest and most effective way to treat children with sleep disordered breathing.
2. Some parents are reluctant for their child to be randomised into a trial where surgery could be delayed.
3. General Practitioners are willing to refer children to a trial for paediatric sleep disordered breathing.
4. Hospital doctors may not be in equipoise regarding the most effective way to treat children with sleep disordered breathing.
5. A partially randomised patient preference trial may be the best way to establish the most effective way to treat children with sleep disordered breathing.

3.2 | Acceptability of T14, OSA-18 and PedsQL

Almost all parents reported the T14, OSA-18 and PedsQL as acceptable. Most parents reported the questions easy to understand, straightforward and brief to complete while they waited for their appointment:

> "The questions were all-I liked the way they were all worded and set out. Very clear to understand them. I think there is nothing worse than having a question and reading it and thinking, "Well I'm not sure. What does this mean, or do you mean this," but no I found it all very clear. Very understandable" (P1)

In contrast, one parent did report that the questionnaires were drawn out and difficult to complete but this may have been because they completed the questionnaires after their appointment and reported finding it difficult to keep their children entertained while they filled them in. Some parents commented on how relevant the questions were "*because I was going through it and it was so raw when doing the*

questionnaire" (P8) while others found the questionnaires highlighted a much wider spectrum of symptoms than some parents had initially associated with sleep disordered breathing:

> "Really, really useful, to be honest…because I had no idea all of those things were linked to just having massive tonsils…lack of sleep, lack of concentration, mood swings, poor appetite." (P12)

One parent reported confusion over how certain questions were relevant to sleep disordered breathing in particular questions about feeling afraid and those relating to social functioning.

3.3 | Suggested outcomes measure for a trial for paediatric sleep disordered breathing

GPs found it difficult to suggest what outcomes measures might be most appropriate for a surgical trial for sleep disordered breathing but suggested a variety including sleep scores, academic performance, cardiac pressures, qualitative parental views of child's sleep, snoring volume, parental stress, and child daytime tiredness. GPs were concerned with identifying an objective outcome measure as parents would not be blind to their child's intervention:

> "But that could be skewed…by bias in terms of the parents obviously would know if their child had an operation or not." (GP4)

Hospital doctors suggested sleep studies as the primary objective outcome measure which could be potentially used in a trial but also mentioned academic performance, growth, or a snoring score. They also suggested other more subjective outcome measures such as quality of sleep, quality of life, parental views/satisfaction, self-reported chest infections/cough. Parents reported preferring qualitative methods of data collection to quantitative:

> "Yes. I mean, you can fill in a form, that's fine, but sometimes…it's not yes or no. It's this grey area in the middle that might not suit everybody. So, it is nice to be able to sit and talk to somebody face-to-face" (P1)

4 | DISCUSSION

While most parents were willing for their child to be randomised, some parents would be reluctant for their child to be randomised into a trial of sleep disordered breathing. GPs were willing to refer children into such a trial however hospital doctors may not be in equipoise. Randomised controlled trials are the gold standard to provide unbiased data however when patients have a treatment preference, randomisation may influence participation and outcomes. A recent meta-analysis assessed the influence of patients' preference in randomised controlled trials by analysing partially randomised patient preference trials; a randomised controlled trial and preference cohort combined.[10] Findings revealed that patients' preference led to a substantial proportion of a specific patient group refusing randomisation, while it did not influence the primary outcome within a partially randomised patient preference trial. Therefore, partially randomised patient preference trials could increase external validity without compromising the internal validity compared with randomised controlled trials.[10] While there is still a need to establish the most effective way to treat paediatric sleep disordered breathing[8] a partially randomised patient preference trial may be the most suitable way forward. However, this study was carried out in only one geographical location and the sample of parents included only one father. Further research is needed within other regions and with a greater variation in sample.

AUTHOR CONTRIBUTIONS

Catherine Haighton made substantial contributions to the design of the study, analysis and interpretation of the data. Rose Mary Watson made substantial contributions to the acquisition, analysis and interpretation of the data. Janet A. Wilson made substantial contributions to the design of the study and interpretation of the data. Steven Powell made substantial contributions to the conception and design of the study and interpretation of the data. All authors contributed to drafting the work, revising it critically for important intellectual content and approved the final version. All authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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CONFLICT OF INTEREST STATEMENT

Janet A. Wilson was previously, and Steven Powell is currently employed by Newcastle upon Tyne Hospitals NHS Foundation Trust.

PEER REVIEW

The peer review history for this article is available at https://www.webofscience.com/api/gateway/wos/peer-review/10.1111/coa.14125.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

Participants provided written informed consent to participate in this study. In line with the terms of consent to which participants agreed,

the data are not publicly available. There are ethical restrictions on sharing the de-identified data set. The data contain potentially identifying and sensitive participant information and we do not have participant consent to share this dataset. Data requests may be sent to South Central—Hampshire B Research Ethics Committee (ref number 18/SC/0378).

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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