

Quality of life of children with sleep-disordered breathing treated with adenotonsillectomy

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Abstract

Objective: To assess the quality of life of UK children with sleep-disordered breathing undergoing adenotonsillectomy, by using the Obstructive Sleep Apnoea 18 questionnaire and determining score changes and effect sizes.

Design: Prospective, longitudinal study.

Setting: The otolaryngology department of a university teaching hospital in Northern England.

Participants: Twenty-eight children for whom adenotonsillectomy was planned as treatment for sleep-disordered breathing, and who had either a clinical history consistent with obstructive sleep apnoea or a polysomnographic diagnosis.

Main outcome measure: The Obstructive Sleep Apnoea 18 questionnaire, a previously validated, disease-specific quality of life assessment tool; changes in questionnaire scores and effect sizes were assessed.

Methods: The Obstructive Sleep Apnoea 18 questionnaire was administered to each child's parent pre-operatively, then again at the follow-up appointment. Questionnaire scores ranged from 1 to 7. Score changes were analysed using the paired *t*-test; effect sizes were calculated using 95 per cent confidence intervals.

Results: Complete data were obtained for 22 children (mean age, 61 months). Ten had undergone pre-operative polysomnography. Twenty-one children underwent adenotonsillectomy (one underwent tonsillectomy). Median follow up was eight weeks (interquartile range, six to 11 weeks). Following surgery, the overall mean score improvement was 2.6 ($p < 0.0001$) and the mean effect size 2.4 (95 per cent confidence interval 1.9 to 2.8). There were significant improvements in each of the individual questionnaire domains, i.e. sleep disturbance (mean score change 3.9, $p < 0.0001$), physical suffering (2.2, $p < 0.0001$), emotional distress (2.0, $p = 0.0001$), daytime problems (1.8, $p = 0.0001$) and caregiver concerns (2.6, $p < 0.0001$).

Conclusion: In these children with sleep-disordered breathing treated by adenotonsillectomy, Obstructive Sleep Apnoea 18 questionnaire results indicated significantly improved mean score changes and effect sizes across all questionnaire domains, comparing pre- and post-operative data.

Key words: Tonsillectomy; Adenoidectomy; Sleep-Disordered Breathing

Introduction

Sleep-disordered breathing covers a number of conditions, from simple snoring to obstructive sleep apnoea (OSA).¹ The predominant clinical feature of sleep-disordered breathing is habitual snoring. A number of other symptoms can be present which may be more indicative of OSA; these include witnessed apnoea and fragmented sleep.² It has been proven that symptoms alone do not reliably differentiate OSA from primary snoring.³ Severe OSA, left untreated, can cause right heart failure and cor pulmonale. Mild to moderate OSA can result in behavioural and neurocognitive problems.⁴

In the UK, the decision to perform adenotonsillectomy on a child presenting with habitual snoring is usually made on clinical grounds, without undertaking a sleep study.⁵ In children with multiple problems, very severe symptoms or an uncertain diagnosis, a sleep study may be requested to clarify the diagnosis. While polysomnography has limitations, it is the most accurate diagnostic tool currently available.² However, polysomnography does not measure the physical and emotional suffering of a child, their daytime problems or their parents' concerns.

There has been increasing interest in the measurement of the quality of life of children with OSA, in

order to help rectify these problems. Disease-specific quality of life measures include the Obstructive Sleep Apnoea 18 and the Obstructive Sleep Disorders 6 questionnaires.^{6,7} In the UK, two studies have been published on the quality of life of children with sleep-disordered breathing.^{8,9} Both these studies used the Child Health Questionnaire, a generic quality of life assessment tool.

Patient-reported outcome measures are becoming increasingly important in measuring the quality of service delivery, and they have been used to justify clinical practice.

The most widely used tool to measure the quality of life of children with sleep-disordered breathing is the Obstructive Sleep Apnoea 18 questionnaire (see Appendix 1).^{6,10} This survey measures quality of life in five domains: sleep disturbance, physical suffering, emotional distress, daytime problems and caregiver concerns. It has been psychometrically validated and shown to be reliable and responsive. The questionnaire includes all the points covered when taking a clinical history for sleep-disordered breathing.

In this study, we aimed to use the Obstructive Sleep Apnoea 18 questionnaire to assess the quality of life of UK children with sleep-disordered breathing treated with adenotonsillectomy, in order to determine whether their quality of life improved post-operatively (as assessed by score changes and effect sizes).

Methods

At the James Cook University Hospital, a university teaching hospital in Northern England, the majority of children with sleep-disordered breathing presented to a single consultant team with a paediatric interest. Of these children, we identified those who were listed for adenotonsillectomy due to either a clinical diagnosis of sleep-disordered breathing or a sleep study proving OSA. The single consultant team recruited consecutive patients between November 2007 and April 2008.

Children were excluded if they had undergone previous adenoid or tonsillar surgery, or if they had craniofacial abnormalities, syndromic diagnoses or comorbidity likely to contribute to respiratory difficulties (e.g. severe asthma).

On the day of surgery, demographic data were recorded, and one of the parents or guardians was given the Obstructive Sleep Apnoea 18 questionnaire to complete. Surgery was conducted by a single consultant and their team.

Post-operatively, a routine follow-up appointment was made for six weeks' time. If the family failed to attend, an invitation for a replacement follow-up review was sent. If this appointment was not kept, then the Obstructive Sleep Apnoea 18 questionnaire was sent in the post.

At the follow-up appointment, the same parent or guardian was asked to again complete the Obstructive Sleep Apnoea 18 questionnaire.

The Obstructive Sleep Apnoea 18 questionnaire has five domains, each with three to four individual items (Appendix 1). Each of the 18 items gives a score from 1 (ie the lowest) to 7 (ie the highest). The mean scores for each domain can be obtained by averaging the scores of the items within the domain. The overall mean score is obtained by averaging the 18 items. Using Sohn and Rosenfeld's definitions, a change of less than 0.5 in the 7 point scale was classified as trivial, 0.5 to 0.9 as small, 1.0 to 1.4 as moderate, and 1.5 or greater as large.⁶ Detection of a trivial level of change in a single sample, with 80 per cent power, would require 26 patients.

Statistical analysis was performed using Stata software (StataCorp, College Station, Texas, USA). Pre- and post-adenotonsillectomy differences were analysed using the paired *t*-test.

A request for study approval was submitted to the local research and ethics committee, which concluded that this study was an exercise in practice evaluation, with no randomisation or change in treatment, and that formal ethical approval was therefore not required.

Results

Twenty-eight children were recruited into the study. Six were excluded due to inadequate follow-up data (five patients) or the involvement of different raters (one patient). Twenty-two patients were thus included in the analysis: 15 boys and seven girls, with a mean age of 61 months. Twenty-one patients underwent adenotonsillectomy and one tonsillectomy alone. The mean time to follow up was eight weeks (interquartile range six to 11 weeks).

Ten of the patients had undergone a sleep study. Of this group, seven were diagnosed with mild OSA, one with moderate OSA and two with severe OSA.

The other 12 patients had a clinical diagnosis of sleep-disordered breathing.

The patients' Obstructive Sleep Apnoea 18 questionnaire score changes are shown in Table I (the score change being the difference between the pre- and post-operative scores). These represent the amount of change in the seven-point scale. The statistical significance of the score changes were calculated using the paired *t*-test; the resulting *p* values are shown in Table I. All of the domains and the overall score showed a large degree of change, as defined by Rosenfeld's classification (i.e. a change of more than 1.5 in the seven-point scale).⁶ The largest change was seen in scores for sleep disturbance, followed by caregiver concern.

The effect size (also termed the standardised response mean) was calculated by dividing the score change by its standard deviation. An effect size of greater than 0.8 was considered large. By this definition, all the calculated effect sizes were large. The largest effect sizes were for sleep disturbance and

TABLE I
OSA-18 QUESTIONNAIRE RESULTS

Domain	Mean score		Mean score change (SD)	<i>p</i> *	Effect size (95% CI) [†]
	Pre-op	Post-op			
Sleep disturbance	5.4	1.5	3.9 (1.3)	<0.0001	3.0 (2.6–3.5)
Physical suffering	4.1	1.9	2.2 (1.6)	<0.0001	1.4 (1.0–1.9)
Emotional distress	4.2	2.2	2.0 (2.0)	0.0001	1.1 (0.6–1.5)
Daytime problems	3.4	1.6	1.8 (1.7)	0.0001	1.0 (0.6–1.5)
Caregiver concerns	4.0	1.4	2.6 (1.6)	<0.0001	1.7 (1.2–2.1)
Overall	4.3	1.7	2.6 (1.1)	<0.0001	2.4 (1.9–2.8)

*Paired *t*-test. [†]Standardised response mean. OSA-18 = Obstructive Sleep Apnoea 18; pre-op = pre-operative; post-op = post-operative; SD = standard deviation; CI = confidence interval

caregiver concern, while the smallest were for emotional distress and daytime problems.

Discussion

This is the first UK study to use a disease-specific quality of life tool to assess children with sleep-disordered breathing. We studied children diagnosed with sleep-disordered breathing (based either on clinical grounds or sleep study analysis) who were treated with adenotonsillectomy, and we found a substantial post-operative improvement in quality of life (as measured by the Obstructive Sleep Apnoea 18 questionnaire). This questionnaire was easy to administer, and has been proven to be a valid measure of the effect of operative intervention.

The score changes and effect sizes were large and statistically significant, for all the questionnaire domains and also the overall scores. As expected, the biggest change was seen in sleep domain scores, reflecting the effects of obstructed breathing noises and disturbed sleep patterns. The second biggest change was seen in the caregiver concern domain. The daytime problems and emotional distress domains also showed large changes in mean score and effect size; however, the degree of change was not as large as for the sleep and caregiver concern domains, and the lower end of the 95 per cent confidence interval for effect size was in the realm of moderate rather than large change.

This study is an accurate representation of our clinical practice and will reflect the practice of many units. In our department, children are treated based on a combination of clinical and polysomnographic assessment. Resource implications mean that it would not be possible to perform polysomnography on every patient. In the present study, we found a large improvement in our patients' post-operative quantitative quality of life measures. If quality of life measures are not assessed and polysomnography not undertaken, it is difficult to quantify patients' post-operative improvement, even if qualitative improvement is reported at follow up.

The Obstructive Sleep Apnoea 18 questionnaire has been psychometrically validated for a North American patient sample.¹⁰ As this questionnaire had good face

validity, and no problems were found with application to UK parents, revalidation was not undertaken.

A number of studies have compared the quality of life of patients with sleep-disordered breathing, both before and after adenotonsillectomy, using the Obstructive Sleep Apnoea 18 questionnaire.^{6,11–18} All of these studies, bar two focussing on specific subgroups with severe OSA¹⁷ and obesity,¹⁸ have been analysed in a recent meta-analysis.¹⁹ The seven studies included assessed a total of 369 children. This group comprised a mixture of clinical diagnoses^{6,11,16} and polysomnographic diagnoses.^{12–15} The mean patient age was higher than that of our group, at 6.3 years. The mean score change values for each of the questionnaire domains were 2.9 for the sleep domain, 2 for physical suffering, 1 for emotional distress, 1.4 for daytime problems and 2.3 for caregiver concern; the overall mean score change was 2. All of these changes were highly statistically significant on paired *t*-test analysis. As can be seen, large changes were seen for each domain score and for the overall score.⁶ The largest changes were seen in the same domains as in our study, namely sleep disturbance and caregiver concern, followed by physical suffering. The only difference in results between this meta-analysis and our own study was our finding of greater change in the emotional distress domain compared with the daytime problems domain. With regard to the magnitude of change, there was a large difference in sleep disturbance domain improvement, comparing our group (mean score change 3.9) and the meta-analysis group (mean score change 2.9), and also a difference in overall score change (with a mean score change of 2.6 in our group versus 2.0 in the meta-analysis group). As we found greater improvement in our patients, this raises the question of whether our patients had more severe symptoms. Such an interpretation would be difficult to explain, as there was no patient selection, and those patients who had undergone sleep studies had predominantly mild OSA. Indeed, the meta-analysis authors commented that a number of their patients came from tertiary referral centres, so one might expect larger score changes in such patients compared with patients from non-teaching hospitals.¹⁹

In our study, the necessary exclusions meant that the final sample size of 22 patients failed to match the 26 patients specified by the power calculation. However, the statistically significant results for each domain mean that there was no type two error, and that there was thus a true difference between pre- and post-operative scores.

As all of the enrolled patient cohort underwent surgery, our study had no control group. Therefore, it is not possible to state conclusively that the observed changes occurred purely as a result of adenotonsillectomy, even though this is very likely to be the case. However, the effect of parents consenting to an adenotonsillectomy and their child undergoing a surgical procedure could also lead to biased reporting of the degree of post-operative improvement. It should be remembered that the observed quality of life improvement was based on a proxy measurement by the parent rather than a direct report by the child.

The Obstructive Sleep Apnoea 18 questionnaire is a useful adjunct to polysomnography, offering a practical way to formalise history-taking and to quantify quality of life improvement. A high score is not diagnostic of sleep apnoea, in the same way that clinical history is unreliable;³ if there are concerns about severity or uncertainty about diagnosis then a sleep study should still be undertaken. There is evidence that lack of post-operative improvement in Obstructive Sleep Apnoea 18 questionnaire score can indicate residual sleep apnoea, and this can be useful in the post-operative context.²⁰

The present study had a relatively short follow-up period of eight weeks. It is not possible to conclude that the benefit from the procedure lasts longer than this, on the basis of our results; however, other studies have shown that the benefit is maintained.^{15,16}

Our patients were selected pragmatically based on clinical and polysomnographic diagnoses; therefore, it is not possible to state that all the children included had OSA, as 12 were diagnosed clinically. The study was not sufficiently well powered to detect differences between the patients who were clinically and polysomnographically diagnosed, so this comparison was not made.

The Obstructive Sleep Apnoea 18 questionnaire can be easily integrated into clinical practice, and provides some evidence that improvements in symptoms and quality of life can be gained by performing adenotonsillectomy. In our experience, the use of this questionnaire has helped to simplify the history-taking and review processes. The questionnaire also provides quantifiable data on symptomatic improvement in a group of patients few of whom receive pre- or post-operative polysomnography.

To overcome the limitations we have discussed, the Obstructive Sleep Apnoea 18 questionnaire would need to be assessed in the context of a randomised, controlled trial. There is currently no published prospective, randomised trial of adenotonsillectomy versus

conservative treatment for the management of patients with mild OSA. Should such a trial be conducted, Obstructive Sleep Apnoea 18 questionnaire scores would be a useful outcome measure, alongside polysomnographic data. However, there would still be a significant potential for bias, as the caregiver who completed the questionnaire would obviously not be blinded to the surgery.

- **Sleep-disordered breathing is common in the UK population**
- **Affected children can suffer neurobehavioural consequences**
- **North American and European studies have used disease-specific quality of life scores to demonstrate symptomatic improvement in patients treated for sleep-disordered breathing**
- **The current study represents the first UK application of a disease-specific quality of life tool to children treated for sleep-disordered breathing**
- **Large changes in quality of life scores were evident for children with sleep-disordered breathing treated with tonsillectomy**

Conclusion

The Obstructive Sleep Apnoea 18 questionnaire can easily be incorporated into clinical practice. This study found large, statistically significant score changes and effect sizes across all domains of this questionnaire, following adenotonsillectomy undertaken for children with sleep-disordered breathing. Quality of life is an important outcome measure in patients with sleep-disordered breathing, and should be assessed in future prospective, randomised investigations.

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Appendix 1. Obstructive Sleep Apnoea 18 questionnaire

Instructions. For each question below, please circle the number that best describes how often each symptom or problem has occurred during the last four weeks (or since last survey if sooner). Thank you.

Date:

Patient name:

Date of birth:

Completed by: Mother Father Guardian

	None of the time	Hardly any of the time	A little of the time	Some of the time	A good bit of the time	Most of the time	All of the time
Sleep disturbance							
During the past 4 weeks, how often has your child had...							
...loud snoring?	1	2	3	4	5	6	7
...breath-holding spells or pauses in breathing at night?	1	2	3	4	5	6	7
...choking or gasping sounds while asleep?	1	2	3	4	5	6	7
...restless sleep or frequent awakenings from sleep?	1	2	3	4	5	6	7
Physical suffering							
During the past 4 weeks, how often has your child had...							
...mouth-breathing because of nasal obstruction?	1	2	3	4	5	6	7
...frequent colds or respiratory infections?	1	2	3	4	5	6	7
...nasal discharge or runny nose?	1	2	3	4	5	6	7
...difficulty in swallowing foods?	1	2	3	4	5	6	7
Emotional distress							
During the past 4 weeks, how often has your child had...							
...mood swings or temper tantrums?	1	2	3	4	5	6	7
...aggressive or hyperactive behaviour?	1	2	3	4	5	6	7
...discipline problems?	1	2	3	4	5	6	7

Continued

	None of the time	Hardly any of the time	A little of the time	Some of the time	A good bit of the time	Most of the time	All of the time
<u>Daytime problems</u>							
During the past 4 weeks, how often has your child had...							
...excessive daytime drowsiness or sleepiness?	1	2	3	4	5	6	7
...poor attention span or concentration?	1	2	3	4	5	6	7
...difficulty in getting out of bed in the morning?	1	2	3	4	5	6	7
<u>Caregiver concerns</u>							
During the past 4 weeks, how often have the above problems...							
...caused you to worry about your child's general health?	1	2	3	4	5	6	7
...created concern that your child is not getting enough air?	1	2	3	4	5	6	7
...interfered with your ability to perform daily activities?	1	2	3	4	5	6	7
...made you frustrated?	1	2	3	4	5	6	7