Our experience of a paediatrician-led clinic for the medical management of children with obstructive sleep disordered breathing.

Haytham Kubba¹, Katherine Rowe², Gideon Pinczower², Robert Sloan², Michelle Waterham³, Emma Weiss³, Rachel Harwood³.

¹Otolaryngologist, Department of Otolaryngology, Royal Children's Hospital, Parkville, Victoria, Australia 3052

²Paediatrician, Department of General Paediatrics, Royal Children's Hospital, Parkville, Victoria, Australia 3052

³Registrar, Department of General Paediatrics, Royal Children's Hospital, Parkville, Victoria, Australia 3052

Correspondence to:

Haytham Kubba

Consultant Paediatric Otolaryngologist

Royal Hospital for Children

1345 Govan Road

Glasgow G51 4TF

Scotland

Email: hkubba@nhs.net

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ABSTRACT

Background: Obstructive sleep disordered breathing (OSDB) is very common in children. Adenotonsillectomy is usually curative, but there is emerging evidence that topical nasal steroids can also be effective for some children, and may avoid the need for surgery. The number of children referred for assessment of OSDB is increasing and in some departments, waiting times are long. We established a paediatrician-led clinic for assessment and initial medical management of OSDB and in this study we report the proportion of children who avoided the need for surgery.

Method: Referral letters to the otolaryngology department were screened and those with suspected OSDB and no significant co-morbidities were diverted to the paediatrician-led clinic. We recorded data for a consecutive series of children seen in this clinic with suspected OSDB over a 3-month period. Parents completed a 5-item subset of questions from the OSA-11 questionnaire before and after treatment with 6 weeks of topical nasal steroids.

Results: In the 3-month study period, 103 children were seen, with a mean age of 6 (range 1-16). Six (5.8%) had improved spontaneously before clinic attendance. Of the 97 children who were still symptomatic, 17 (17.5%) were referred directly for surgery on the basis of the severity of their symptoms, or because of other coexisting conditions that required surgical treatment (such as recurrent tonsillitis or otitis media). 3 declined intranasal steroids. 77 had a trial of intranasal steroids, of whom 34 (35%) reported enough improvement to avoid

surgery, and 29 (28%) failed to improve and were referred for surgery. 14 (14%) failed to attend after the trial of steroids: of these, 5 (5%) were contactable by phone and confirmed improvement after topical steroids. OSA-5 scores were significantly improved following intranasal steroids.

Discussion: A paediatrician-led clinic can be an effective way to ease the workload of an over-stretched otolaryngology service, and judicious use of topical nasal steroids can help around 40% of children with OSDB avoid surgery.

Key words: Sleep Disordered Breathing, Obstructive Sleep Apnoea, Children, Paediatrics, Follow up, Medical management

INTRODUCTION

Obstructive sleep disordered breathing (OSDB) is characterised by prolonged partial airway obstruction and/or intermittent complete pharyngeal obstruction which disrupts normal ventilation during sleep and sleep continuity (1). OSDB was initially described in 1880 (2) and encompasses the more severe form of OSDB known as obstructive sleep apnoea (OSA).

Obstructive sleep apnoea (OSA) is a common condition in childhood, affecting 1-5% of children (3). It occurs most commonly in children aged 2-8 years of age (4) and declines in prevalence thereafter, although estimates of prevalence have been as high as 2-3% in older children (5,6). Many more children snore habitually without having OSA (7,8). Allergic rhinitis (9,10) and asthma (11) are increasing in prevalence and both allergic and non-allergic rhinitis are often associated with adenotonsillar hypertrophy and OSDB (12,13).

Adenotonsillectomy benefits most children with OSA (14), however the number of referrals to some specialist services can exceed their capacity. This results in long waiting lists and potential delay in treatment. In addition, there is evidence that supports conservative management of milder cases of OSA with a period of observation (14) and topical nasal anti-

inflammatories (15). Inflammatory mediators such as leukotrienes and prostaglandins are elevated in the upper airways of children with OSA (15). Montelukast (16,17) and/or intranasal corticosteroids (18-20) show benefit in randomised, placebo-controlled studies for patients with OSDB and/or OSA. A reduction in the severity of OSA symptoms was seen after anti-inflammatory treatment with either montelukast and/or intranasal budesonide. There was a sustained reduction in OSA symptoms for a further 8 weeks after a 6 week trial of intranasal budesonide when compared to placebo (21).

Our hospital provides specialist paediatric services to a large and diverse population, and the otolaryngology department has been struggling to cope with the volume of referrals for the management of routine conditions. Referrals of children for assessment of OSDB alone were occurring at more than 150 per month. A paediatrician-led clinic was commenced to assist with assessing referrals for OSDB or OSA or other relatively straightforward conditions like epistaxis, recurrent sore throat and recurrent ear infections. The clinic consisted of 4 paediatricians, 3 supervised paediatric trainees and an otolaryngologist. Referrals for audiology and home pulse oximetry were coordinated with the clinic.

Our study is an audit of consecutive children age 1-18 years referred during a 3-month period from August 2017 to October 2017 for assessment of OSDB in a paediatrician-led clinic where intranasal steroids would be offered. The outcomes were measured by parent report and before-and-after questionnaires. Our primary aim is to describe the proportion and characteristics of children who reported sustained benefit from intranasal corticosteroids (or who improved spontaneously) and therefore did not require surgical review, and our secondary aim is to describe the proportion and characteristics of children who were referred for surgery.

METHODS

Referral letters to the otolaryngology service were examined initially by senior clinicians to identify children who would be suitable to divert to the paediatrician-led clinic. Those with identified syndromes, neurological conditions, chronic respiratory disease, or obesity documented in the referral were excluded. Consecutive children between 1 and 18 with suspected OSDB were considered suitable for this study.

At the clinic, routine history and examination were performed. Audiological testing and tympanometry were available for those with coexisting ear disease. Children were assessed and either given a trial of intranasal steroids for 6 weeks and reviewed, or if there was concern about severe OSA based on history, examination and video record, they were referred directly for an opinion from the otolaryngologist present at the clinic.

We collected the following data for each child seen at the clinic: original referral date, allergic history (presence of asthma, eczema or allergic rhinitis), family history and day care/school attendance, course of symptoms, presence of obesity and other relevant medical /developmental history. Tonsillar size, status of the turbinates, nasal obstruction and aural examination including tympanometry were also recorded.

For this study we used five questions taken from the OSA-11 questionnaire (22). This questionnaire was derived from OSA-18 (23-26). Unlike OSA-18, OSA-11 uses a 4- rather than 7-point Likert scale as Rasch modelling has confirmed that parents can only reliably differentiate 4 categories (27). The four categories were 'None of the time', 'Some of the time', 'Most of the time', or 'All of the time', scored from 0 to 4. Only 5 items from OSA-11 have been shown to be reliably related to the presence of OSA on polysomnography (22). These items were: *During the past 4 weeks, how often has your child had...* 'Loud snoring'; 'Breath holding spells or pauses in breathing at night'; 'Choking or made gasping sounds while asleep'; 'Mouth breathing because of a blocked nose'; or 'Breathing problems during sleep that made you worried that they were not getting enough air'? This 5-item subset of OSA-11 will be hereafter referred to as OSA-5 for the purpose of this study.

For those prescribed intranasal steroids, a short video was shown to the child and parent demonstrating correct technique for administering intranasal spray (28) and instructions were given for the parent/caregiver to compare observation of the child sleeping currently and just prior to returning for review. The steroid used was mometasone furoate, 50 micrograms per spray, in a dose appropriate for the age and weight of the child. Parents were encouraged, if possible, to video record their child sleeping. If considered appropriate, home overnight pulse oximetry was organised towards the end of the trial period to help assess severity for surgical triage. This decision was taken at the clinician's discretion and not every child underwent overnight pulse oximetry. For this reason, we are not reporting pulse oximetry findings in detail in this study. No child in this study underwent polysomnography.

On review, parents were asked to complete the OSA-5 questionnaire a further time and an assessment made whether there was clinical and reported improvement. If improvement had occurred, the child and parent were instructed in how to identify when intranasal steroids were needed. If there was no change or insufficient change in sleep disordered breathing or other conditions needing attention the child was referred for an opinion on surgery from the otolaryngologist present at the clinic. Associated conditions that might require surgery in addition to OSDB included persistent mild to moderate conductive hearing loss or significant recurrent tonsillitis consistent with the American Association Otolaryngologists- Head and Neck Surgeon's (AAO-HNS) Guidelines (29). Children who failed to attend for follow up were contacted to ascertain the reason and a further appointment provided if requested.

Data were recorded regarding the number of children referred for surgery directly, the number who required surgery after a trial of intranasal steroids, those who had reported resolution of symptoms following the trial as well as those who refused medical treatment or did not return for follow up. Analyses were conducted using Statistica 13 (Statsoft – TIBCO California). Pre and post intervention mean scores on OSA-5 were analysed using student's t-test and one-way ANOVA to compare the baseline means between groups.

Ethical considerations

Our institution's ethical review board does not require specific ethical approval for a study such as this which consists solely of an evaluation of a clinical service development.

RESULTS

Of the 132 children referred with OSDB during the 3 month period, 20 failed to attend (FTA) and requested an appointment to be rescheduled outside the assessment period, and were not included in the study. The most common reason given was related to logistic issues in keeping the appointment rather than lack of concern about the condition. An additional nine failed to attend the first appointment and were not rescheduled. Of these: 2 had had surgery elsewhere, 3 improved without intervention (referred 4 months, 6 months and 2 years prior), 3 were unable to be contacted and 1 was overseas.

This resulted in 103 children who attended the clinic (Figure 1). The mean age was 6.3 years (range 1.2-16.3 years, sd 3.4), with 52% males. The length of time from referral until appointment was <1 year for 58% of the children, with 21% waiting 1-2 years, and 19% 2-3 years and 2% waiting 3-4 years. The range was from 2 months to 3.7 years.

Six (5.8% of those attending clinic) improved without treatment but attended the appointment for reassurance. Including the 3 who failed to attend, 9 (8.1%) of 111 referrals reported improvement prior to the appointment.

OSA-5 questionnaires were completed for 78 of the 97 children seen at the clinic with OSDB (80%). Language difficulties in non-native English speakers were the main reason for non-completion.

Of the 97 children who were still symptomatic on attendance, 17 (17.5%) were referred to surgery without a trial of medical treatment on the basis of severity of symptoms or the presence of coexisting conditions that require surgery (Table 1). Twenty nine (29.9%) were referred for surgery after a failed trial of medical treatment (Table 2). Thirty four (35%) reported improvement with medical treatment. Three reported 'no change' but they did not try intranasal steroids as they felt that their child would not tolerate the spray and the symptoms were tolerable. Fourteen (14.4%) failed to attend follow up appointment at 6 weeks. Of these, 9 (9.3%) were lost to follow up and could not be contacted and 5 (5.2%) reported marked improvement and did not keep the follow up appointment. Based on intention to treat there is a significant difference in outcome scores following the 6 week trial of intranasal steroids. (Figure 2).

Figure 3 indicates the significant difference in reported ratings following a trial of intranasal steroids for those that did not proceed to surgery compared with those who showed no change and proceeded to surgery (Figure 4). Of those who came for a second opinion after having seen an otolaryngologist, external to the hospital, who recommended surgery for OSA, 4 of the 5 did not proceed to surgery after a trial of intranasal steroids.

With regard to the 29 children referred for surgery after an unsuccessful trial of topical nasal steroids, the mean age was 5.8 years (range 1.6-16.1, s.d. 4). 62% were <6 years and 75% were male. All had significant adenotonsillar hypertrophy confirmed at surgery.

Therefore following medical assessment and a clinical trial of intranasal steroids, 40.2% improved sufficiently to avoid the need for surgery. This figure includes the 5.2% that reported improvement but did not keep the follow up appointment. Forty-seven per cent (47%) of the children underwent surgery, 5 of whom required surgery for other conditions (chronic tonsillitis, conductive hearing loss) that had not improved in conjunction with the improvement in OSDB. One eighth (12.5%) had no medical trial or were lost to follow up.

There is a suggestion that those referred directly to surgery had more severe symptom scores compared with who avoided surgery (Table 3). However, there is no significant difference between those who responded to a trial of medical treatment and those who did not. (Figure 5). Total score is possibly predictive of likely need for direct referral to surgery. Younger subjects (<6 years) who proceeded to surgery tended to have higher scores on the OSA-5 and to have additional reasons for surgery such as persistent conductive hearing loss, or recurrent pustular tonsillitis.

DISCUSSION

This study indicates that a paediatrician-led clinic can be a very efficient and effective way to manage referrals for OSDB. This study was not intended to demonstrate the effectiveness of non-surgical interventions in children with suspected OSDB: this has been established in randomised controlled trials already (16-21). Rather, our aim was to demonstrate in a prospective observational study that a worthwhile proportion of children, even those under 6 years of age, can be managed in such a way as to avoid surgery. This is good for children and families, and relieves a significant burden of work from an over-stretched otolaryngology department, allowing surgeons to provide more timely intervention for the children who need it most and who are most at risk of the long-term cognitive effects of OSDB. The collegiate approach of paediatricians and surgeons working in the same clinic, addressing upper airway concerns in this way can provide patient benefit and general benefit in reducing long hospital waiting lists. Based on history and examination, the otolaryngologist involved in the clinic confirmed that the overwhelming majority of children seen would have been recommended to have adenotonsillectomy at their first visit had they been seen directly by otolaryngologists rather than paediatricians.

In our hospital, the limited availability of otolaryngologists is a major barrier to children accessing timely intervention. This clinic has enabled us to use the skills, experience and

judgement of paediatricians to give children speedier access to diagnosis and treatment, and to allow the otolaryngologist to focus their time on the subset of children with the most severe symptoms and therefore the most to gain from surgery.

The study further highlights that the use of a relatively straightforward medical intervention may prevent subjects from requiring surgery. The use of intranasal steroids before considering surgery can potentially reduce the number of children with suspected OSA having surgery by 40%, which is a clinically important reduction. In addition, those with rhinitis might well be the ones most likely to have residual symptoms after adenotonsillectomy and it is particularly important to avoid unnecessary or ineffective surgery in this group.

Even when a trial of intranasal steroids is not successful in relieving symptoms, parents are more accepting of the need for risks and the discomforts of surgery, having seen that non-surgical options have been tried and failed.

OSA-5 is a quick and easy assessment tool based on a validated subset of questions from OSA-11. Based on the previous study, scores above 5 on those 5 items suggest possible mild OSA whereas scores about 10 indicate that significant OSA is likely. We have found OSA-5 to be more useful than the original OSA-11, not just because it is faster and easier to complete, but because it includes only those questions that have been shown to predict the presence of OSA on polysomnography (22). This shortened version has primarily been designed as a screening and diagnostic tool, hence the emphasis on the limited number of elements that correlate most strongly with OSA on polysomnography. We have shown here that the OSA-5 scores correlate well with clinical assessment of symptom severity at baseline, and that the scores are responsive to change after both medical and surgical intervention. While it could be argued that losing a number of questions on wider quality-of-life issues means we are losing a lot of potentially useful information, the fact remains that this shortened version of the questionnaire performs well in a clinical research setting and we would like to see it used more widely.

Conclusion:

This audit suggests that a paediatrician-led clinic held in conjunction with an otolaryngologist can increase efficiency and potentially reduce the need for surgery in a significant proportion of referrals for OSDB. The screening questionnaire (OSA-5) was helpful in identifying severity and recording change.

Disclaimers

This study was done without any external funding.

None of the authors has any relevant conflict of interest, financial or otherwise.

The authors are happy to comply with any reasonable request to supply the original data upon which this study is based.

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Figure 1: Flow chart for patients with OSDB seen in Upper Airways Clinic during a 3 month period.

Figure 2: Box and whisker plot: t-test of means of OSA-5 scores before and after 6-week trial of intranasal steroids .(Intention to treat) (Means 8.40 sd 3.5, 5.38 sd 3.6, $t=3.42$ df 63 $p=0.001$)

Figure 3: Box and whisker plot for pre and post trial of intranasal steroids in those that did not proceed to surgery (mean score before 8.6, mean score after 4.44. $t= 3.62$ df 34 $p=0.0009$)

Figure 4: Pre and post trial of intranasal steroids in those who subsequently had surgery

Mean score 8.07 before trial medication - mean 7.31 after (t= 0.54 df 25 p=0.60)

Figure 5: ANOVA of score on OSA-5 by group- direct to surgery, improved after course of intranasal steroid and requiring surgery after course of intranasal steroid. ($F(2, 60)=6.2367$, $p=.00346$)

Table 1: Features of the 17 referred directly to surgery with OSDB without a trial of medical treatment (F:M 8:9 and mean age 4.86 (1.2-14.2years) with 18%<2 years. All had significant adenotonsillar hypertrophy and 5 had ventilation tubes inserted.

Clinical diagnosis	Mean score on OSA-5 (range)	<6 years	>6 years	Total
Clinical OSDB with stertor at rest and/or significant destaurations on oximetry	12.8 (11-15)	6	1	7
OSDB and tonsillitis	12	2	1	3

Persistent mild/ mod conductive hearing loss with OSDB	11.2 (8-14)	5		5
Recurrent tonsillitis with OSDB	8		2	2
Total	11.8 (8-15)	13 (76%)	4	17

Table 2: Features of the 29 referred for surgery following no or minimal improvement following trial of medical treatment.

	Mean score OSA-5 (range)	<6years	>6years	total
No improvement	9 (3-15)	5	1	6
Some improvement with reduced turbinate size but still reduced nasal flow	10 (7-13)	5		5
Persistent OSDB and associated mild to moderate conductive hearing loss	7.3 (2-12)	6	1	7
Improved OSDB but persistent recurrent pustular tonsillitis	7 (6-8)		3	3
Improved OSDB but significant history of tonsillitis and recurrent otitis media	8 (7-9)		2	2
Persistent severe OSDB/stertor at	11 (7-15)	1	3	4

rest				
Persistent OSDB and Tonsillitis	14 (13-15)	1	1	2
	9.2 (2-15)	18	11	29

Table 3: OSA-5 Questionnaire data at first visit for the based on outcome

Group	Outcome	N	Mean score on 5 items (sd)	range
1	Direct to surgery	13	11.5 (2.3)	8-15
2	Surgery post trial	25	9.2 (3.73)	2-15
3	No surgery post trial	25	7.5 (3.33)	2-14
4	No treatment	3	4.3 (1.52)	3-6
5	Lost to FU	5	7.2 (3.03)	2-12
6	Improved prior to clinic	5	6.4 (4.04)	2-12
7	No change	2	5.5 (2.12)	4-7
	Total	78	8.4 (3.65)	2-15

ABBREVIATIONS

OSDB – obstructive sleep disordered breathing

OSA – obstructive sleep apnoea

FU- follow up

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coa_13479_f1.tiff

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coa_13479_f4.tiff

coa_13479_f5.tiff

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Author/s:

Kubba, H;Rowe, K;Pinczower, G;Sloane, R;Waterham, M;Weiss, E;Harwood, R

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