Long-term Impact of Adenotonsillectomy on the Quality of Life of Children with Sleep-disordered breathing

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Abstract

Introduction Adenotonsillectomy is the first-line treatment for obstructive sleep apnea secondary to adenotonsillar hypertrophy in children. The physical benefits of this surgery are well known as well as its impact on the quality of life (QoL), mainly according to short-term evaluations. However, the long-term effects of this surgery are still unclear.

Objective To evaluate the long-term impact of adenotonsillectomy on the QoL of children with sleep-disordered breathing (SDB).

Method This was a prospective non-controlled study. Children between 3 and 13 years of age with symptoms of SDB for whom adenotonsillectomy had been indicated were included. Children with comorbities were excluded. Quality of life was evaluated using the obstructive sleep apnea questionnaire (OSA-18), which was completed prior to, 10 days, 6 months, 12 months and, at least, 18 months after the procedure. For statistical analysis, *p*-values lower than 0.05 were defined as statistically significant.

Results A total of 31 patients were enrolled in the study. The average age was 5.2 years, and 16 patients were male. The OSA-18 scores improved after the procedure in all domains, and this result was maintained until the last evaluation, done 22 ± 3 months after the procedure. Improvement in each domain was not superior to achieved in other domains. No correlation was found between tonsil or adenoid size and OSA-18 scores.

Conclusion This is the largest prospective study that evaluated the long-term effects of the surgery on the QoL of children with SDB using the OSA-18. Our results show adenotonsillectomy has a positive impact in children's QoL.

Keywords
- sleep-disordered breathing
- quality of life
- tonsillectomy
- adenoidectomy
- OSA-18
- child

Introduction

Sleep-disordered breathing (SDB) affects from 7 to 17% of the pediatric population, and the symptoms range from primary snoring to obstructive sleep apnea (OSA).[1,2] The nocturnal symptoms related to SDB are snoring, witnessed apnea, sialorrhea, restless sleep, and enuresis. The daytime symptoms are hyperactivity, attention deficit disorder, daytime naps,

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excessive daytime sleepiness, behavioral disorders (including aggressivity, anxiety, and depression). Physical impact is also described, including muscular and skeletal changes. Untreated disease is believed to affect emotion, cognitive function, and neurobehavior of both children and family members that may contribute to a poor quality of life (QoL).[2,3]

Treatment of SDB is associated with an increase in health care utilization and cost. Children with SDB, as compared with controls, have a significantly higher rate of antibiotic use, 40% more hospital visits, and an overall elevation of 215% in health care usage, mostly from increased respiratory tract infections.[4–7] Failure to thrive is reported in 27 to 62% of pediatric OSA cases, and up to 40% of children with SDB exhibit behavioral problems.[8]

Though SDB is a common condition with a high impact on physical, developmental, cognitive, and behavior aspects in childhood, data from pediatrician knowledge, training and abilities on treating children with SDB shows there is a lack of knowledge and misinformation about this subject.[9] One study that evaluated 112 pediatricians demonstrated that the majority of them (62%) consider that information about SDB during medical school and residency are scarce, albeit children with SDB are frequently attended at the pediatric office.[10]

Adenotonsillectomy (AT) is the first line of treatment for OSA secondary to adenotonsillar hypertrophy in children.[2] Data, in 1993, from the National Hospital Discharge Survey noted a decrease of 0.50% in inpatient tonsillectomy rates from 1977 to 1989,[11] and similar reports from 1978 to 1986 showed that the rate of tonsillectomy for treatment of throat infections declined; however, the frequency of SDB as the primary indication for the procedure increased, especially in children under 3 years of age.[12] Although the current indication for ATis well established in literature, misconceptions about the surgery concerning to risks, benefits and impact on the immunological system are still present and lead some pediatricians to discourage the caregivers to accept AT. This is a secure surgery, and published data reported mortality rates for tonsillectomy from 1 per 2,360 (in inpatient settings) to 1 per 56,000 patients.[13–15] Previous studies show there would, therefore, appear to be a therapeutic advantage to removing recurrently diseased tonsils because they are no longer able to function adequately in local protection. However, some studies demonstrate minor alterations of immunoglobulin concentrations in the serum and adjacent tissues following tonsillectomy.[16,17] Nevertheless, there are no studies to date that demonstrate a significant clinical negative impact of tonsillectomy on the immune system.

The gold standard for the diagnosis of OSA is a polysomnography (PSG). However, PSG is expensive, requires time and a trained professional, and is often unavailable at many institutions where children with SDB are treated.[2,18] Therefore, most cases of SDB are treated based on clinical assessments and physical exams, with no objective testing. Albeit it may not be an ideal situation, children without comorbities may the treated without PSG.[19,20] Though PSG is ideal for evaluating the presence and severity of apneas, it fails to measure the impact of SDB on children's QoL in general, as well as in terms of its effect on behavior and emotion.[19,21] Because of this, the use of questionnaires to evaluate QoL has become popular in recent years.[22–24]

Recent articles have evaluated the impact of surgery on QoL among children with SDB through questionnaires on physical, emotional, behavioral, and familial factors.[3,21,25–27] There are different methods for evaluating postadenotonsillectomy QoL among children, and the obstructive sleep apnea questionnaire (OSA-18) is one of the most frequently used questionnaires worldwide. It has been validated in a variety of languages.[28–31] Most of the studies evaluated children for 6 months or less and rarely the patients were reassessed over 1 year after AT.[19,32–34]

Objective

The present study aimed to describe the QoL in children with SDB and the long-term impact of AT on this disease.

Method

This was a prospective, non-controlled study that was approved by the research ethics committee of the institution (decision no. 1.429.954).

This study included patients between 3 and 13 years of age, of both genders, in ASA classes 1 and 2 who were treated in the otorhinolaryngology department between January 2017 and January 2018 and whose symptoms included snoring, witnessed apneas, noisy breathing, mouth breathing, and restless sleep. All of the patients were treated by the same pediatric otolaryngologist. Patients with symptoms consistent with rhinitis were treated using nasal corticosteroids and/or antihistamines, as recommended for 3 months, and were then reevaluated.[35] Once the child presented complete recovery of the symptoms after clinical treatment, surgery was discarded. Tonsil size was evaluated using the Brodsky grading scale.[36] Adenoid size (in percentage of obstruction) was calculated through the skull lateral-view X-ray.[37]

The children were weighed and measured by the principal investigator. Patients with overweight, obesity, underweight, craniofacial malformations, genetic syndromes, neuromuscular diseases, or developmental delays were excluded, as were patients who required a concomitant procedure (including inferior turbinate surgery).

The patients selected for surgery were informed of the study and were officially included once the guardians signed the informed consent form, and the children signed the informed consent form when possible. The OSA-18 was administered by the hospital social worker 1 week before, and then again 10 days, 6 months, and 12 months after the procedure; another assessment was done in July 2019. The first two assessments were in person, and the other evaluations were made by phone.

The OSA-18 is a validated questionnaire that contains 18 questions divided into 5 domains: sleep disturbance, physical suffering, emotional suffering, daytime problems, and caregiver concerns. Each domain receives a score from 1 to 7; higher

scores reflect a worse QoL. Total scores lower than 60 suggest that the sleep disorder has little impact on QoL; scores between 60 and 80 indicate a moderate impact, and scores above 80 reflect a high negative impact of the sleep disorder on QoL.

The statistical analyses were performed using the IBM SPSS software version 22.0 (IBM Corp., Armond, NY, USA). The normality of the samples was evaluating using the Kolmogorov-Smirnov test, which allowed for the use of parametric tests in the data analysis. The Chi-squared test and analysis of variance (ANOVA) were used to evaluate the data, and *p*-values lower than 0.05 were considered statistically significant.

Results

Fifty-one patients were evaluated, out of whom 8 were excluded because they presented complete recovery of the symptoms after treating rhinitis. From the 43 children initially included in the study, only 31 completed all phases of research. The average age was 5.1 ± 2.9 years, and 16 (51%) of the patients were male. The demographic data and clinical characteristics of the patients are described in ►**Table 1**.

The most common clinical findings were snoring (which was an inclusion criterion) followed by restless sleep, chocking noise, frequent awakening, sialorrhea, witnessed apnea, and enuresis, as shown in ►**Table 2**.

The mean preoperative OSA-18 scores were 79.9 ± 12. The scores of all the evaluated patients improved after surgery ($p < 0.05$), with mean reduction of 40 ± 8 in the first postoperative evaluation (10 days after surgery). Mean total OSA-18 score after surgery remained under 37 in all assessments. The last evaluation, done in 2018 July, was done 22 ± 3 months after surgery (see ►**Table 3**). None of the patients presented increased values of OSA-18 scores after surgery, neither in total scores nor in each domain separately.

The reduction in the OSA-18 scores was not different between one domain in relation to another ($p > 0.05$). Both male and female experienced improvement of OSA scores before and after the surgery, without gender effect (►**Fig. 1**).

Tonsil size and adenoid size were correlated with OSA-18 scores before surgery. No correlation was found between questionnaire scores and the degree of tonsil size, as detailed

Table 1 Demographic data and clinical characteristics of the patients

Age in years (mean ± SD)	5.2 ± 2.9
Male	51%
Height in cm (mean ± SD)	119 ± 18
Weight in kg (mean ± SD)	18.4 ± 7
Adenoid size (%)	79.1 ± 9
Presence of rhinitis symptoms (n/%)	7/ 22%
Tonsil size (Brodsky grade)	Grade 2—17%
	Grade 3—66%
	Grade 4—17%

Abbreviation: SD, standard deviation.

Table 2 Distribution of sleep-disordered breathing symptoms

Symptoms	Frequency (%)
Snoring	100%
Restless sleep	92%
Choking	80%
Frequent awakening	73%
Sialorrhea	65%
witnessed apnea	9%
Enuresis	6%
Hyperactivity	3%
Poor school performance	3%
Aggressive behavior	0%

Table 3 Scores of the obstructive sleep apnea questionnaire before and after surgery

OSA-18 scores		Mean ± SD	*p*-value*
Sleep disturbance	Before	16.3 ± 5	< 0.001
	10 days	7.9 ± 4	
	6 months	8.0 ± 3	
	12 months	6.9 ± 3	
	Late follow-up	5.7 ± 2	
Physical suffering	Before	21.6 ± 3	< 0.001
	10 days	8.9 ± 4	
	6 months	9.0 ± 5	
	12 months	8.5 ± 4	
	Late follow-up	8.3 ± 3	
Emotional suffering	Before	11.5 ± 6	< 0.001
	10 days	4.1 ± 2	
	6 months	3.9 ± 2	
	12 months	4.0 ± 2	
	Late follow-up	5.0 ± 1	
Daytime problems	Before	9.2 ± 4	0.007
	10 days	6.0 ± 3	
	6 months	6.9 ± 4	
	12 months	6.6 ± 4	
	Late follow-up	6.5 ± 4	
Caregiver concerns	Before	21.8 ± 5	< 0.001
	10 days	9.0 ± 3	
	6 months	8.1 ± 4	
	12 months	7.6 ± 4	
	Late follow-up	7.2 ± 4	
Total	Before	79.9 ± 12	< 0.001
	10 days	36.8 ± 13	
	6 months	35.3 ± 14	
	12 months	34 ± 13	
	Late follow-up	32.9 ± 14	

Abbreviation: OSA-18, obstructive sleep apnea questionnaire; SD, standard deviation.
*ANOVA.

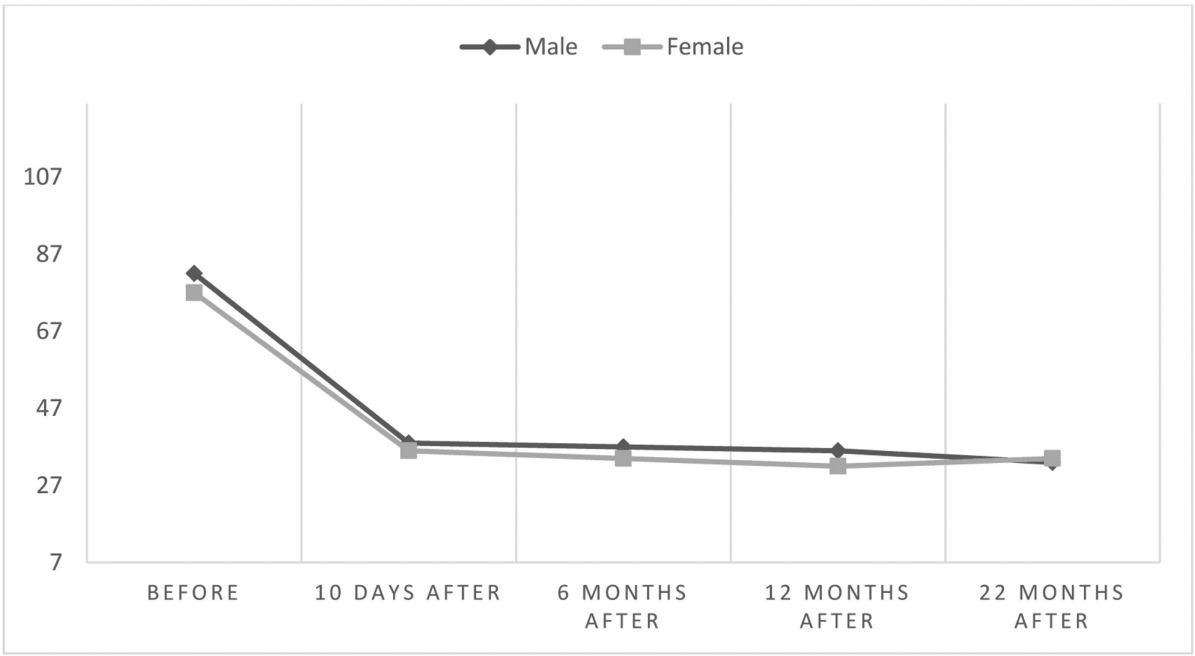

Fig. 1 Obstructive sleep apnea (OSA-18) questionnaire scores before and after surgery regarding to gender. Analysis of variance. *P*-value = 0.55.

Table 4 Correlation between tonsil and adenoid size and total obstructive sleep apnea questionnaire score

Tonsil size	Corr (r)	17.2%
	p-value*	0.95
Adenoid size	Corr (r)	28.1%
	p-value*	0.02

*Pearson correlation.

in ►**Table 4**. A correlation was found between adenoid size and OSA-18 scores ($p = 0.02$), but it was a weak correlation that may be discharged.

Discussion

The present study had described the characteristic of pediatric SDB and long-term impact of AT on the QoL in an adequate and objective manner.

Preoperatively, only a small percentage of patients presented with symptoms related to emotional, behavior, and diurnal complaints. All participants were referred for snoring, and the majority of them for nocturnal symptoms. Previous studies had demonstrated that snoring and sleep disturbances are the hallmarks for pediatric SDB.[12] Only a small portion of pediatric SDB presented with emotional and diurnal disorders.[3] This characteristic is recognized as one of the significant differences between adult and pediatric SDB.[3,38]

Sleep-disordered breathing can negatively affect children's memory and attention levels, likely because of hypoxia, which is also considered to be responsible for daytime symptoms such as irritability, aggression, and hyperactivity.[39,40] Furthermore, SDB is associated with enuresis, metabolic problems, and with an increased risk of metabolic and cardiovascular abnormalities, decreasing children's QoL.[18,41]

It is important to evaluate children's QoL because it reflects individual sentiments toward physical, emotional, and social factors in their life and how these factors relate to their health.[3,21] The OSA-18 is an evaluative, discriminative, and validated instrument to assess QoL in children with SDB, and it is the most widely used evaluation tool worldwide.[19,25,31]

An analysis of the literature revealed that AT is the treatment of choice for children with SDB caused by adenotonsillar hypertrophy. As a result, most of the studies that evaluate QoL in this group of patients do not have a control group.[31,32,41] Studies that compare the surgery to watchful waiting have shown that children who undergo AT exhibit a significant improvement in QoL when compared with children in the watchful waiting control group.[7,42] Our study found a significant improvement in all of the domains assessed by the OSA-18, similarly to the results found in other studies.[23,32,43] This improvement can be measured not only by the numerical decrease in the scores for each domain of the OSA-18, but also through the changes from total scores reflecting high or moderate impacts on QoL (scores above 60) to total scores reflecting low impact on QoL, which occurred in the current study and in prior research.[7,38,44] Our finding that this outcome is maintained for at least 6 months is consistent with other reports in the literature.[31,34,42,45] Long-term follow-up studies after AT can be difficult to be performed, since most patients do not need further medical evaluation after improvement of the symptoms is observed. This is especially noticeable in children without comorbidities, since the surgical success

rate in this group is high. Only 2 studies that evaluated QoL evaluated children for more than 1 year after AT, and one of them was prospective. The present study followed the children for 24 months after the procedure.[33,46] This is the largest prospective study that evaluated the long-term effects of the surgery on the QoL of children with SDB using the OSA-18.

Some studies describe reappearance of symptoms during the follow-up, mainly in females with normal weight and older than 6 years, which was attributed to an incomplete therapeutic response and due to a change the parental perception of the symptoms.[32,44] In our study, none of the patients presented worsening of the OSA-18 values after surgery. Although allergic rhinitis is considered a risk factor for AT failure, it was not found in our study.

Tonsil and adenoid sizes are a reason for concern not only for caregivers, but also for many physicians who are involved in these patients' care. Some studies have found that there is an association between tonsil size and OSA as well as between tonsil size and PSG findings, but most of these studies have low levels of evidence.[47] Well-designed studies have shown, as our study found, that tonsil and adenoid size is not correlated with either SDB severity or patient QoL.[19,38,47,48]

Our study did not find differences between OSA-18 scores and gender, which is similar to the findings of other studies. The analysis of previous studies that found a higher prevalence of OSA in males shows that it is probably influenced by age, once this difference was found only in older boys. It could be explained by hormonal changes due to puberty, because OSA is more prevalent in male adults.[49,50] Since our study group was limited to 13-year-old patients, they probably were not under hormonal effects yet.

One advantage of our study is that all steps were performed by a single team during a long-term follow-up. But there are some limitations, including the fact that many caregivers believe that surgery is the best treatment for the child, particularly after agreeing to surgery under general anesthesia. This belief may influence caregivers' answers in the postoperative period.[32,51] Another limitation of this study is the lack of the preoperative PSG. This exam is the gold standard in evaluating children with SDB and is particularly useful for children without comorbities. However, the difficulty in implementing this exam on a large scale leads to use other exams or clinical evaluation alone to diagnose and treat these patients.[30,38,52] Finally, considering that SDB is a prevalent condition, a large, diverse subject population must be surveyed to determine whether AT leads to improvement in the QoL of children,.

Conclusion

This study concludes that SDB in children leads to a poor QoL and that AT improvement remains for at least 22 months after the surgery. The positive impact of surgery occurs both in nocturnal and daytime symptoms. Since pediatricians are the medical professionals with the greatest contact with children with SDB, their knowledge of the subject is fundamental to these patients' care.

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