

# Quality of life related to residual snoring after adenotonsillectomy: a pilot study

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# **ABSTRACT**

Introduction: Few studies have addressed long-term quality of life related to residual snoring after adenotonsillectomy. The aim of this study was to compare scores from the OSA-18 questionnaire between children with residual snoring and non-snoring children two or more years after adenotonsillectomy. Material and Methods: The sample comprised 25 children divided into two groups, a group of 14 snoring children, and a control group of 11 non-snoring children. The OSA-18 questionnaire was applied to the volunteers. In the control group, it was completed by the caregivers of the children, while in individuals with residual snoring it was completed by the caregivers of children in the presence of a doctor or dentist. A statistical comparison was made using a generalized linear model. Results: The snorer group had a higher total OSA-18 score, and a higher score in all five domains compared to the control group. Conclusion: Children with residual snoring two or more years after adenotonsillectomy may have a worse quality of life compared to the control group.

Keywords: Sleep Apnea; Obstructive; Snoring; Indicators of Quality of Life; Tonsillectomy.

#### INTRODUCTION

In childhood, sleep-disordered breathing is associated with a myriad of health problems that reinforce the need for early diagnosis and treatment<sup>1</sup>. The prevalence of obstructive sleep apnea (OSA) in children over 3 years of age varies from 7.2% to 34.2%<sup>2</sup>, whereas habitual snoring has a prevalence of 5-35%<sup>3</sup>.

The most common otorhinolaryngological condition in children with OSA is adenotonsillar hypertrophy, with or without allergic rhinitis, and mouth breathing4. The goldstandard treatment for OSA in children is adenotonsillectomy, which normalizes polysomnography and improves quality of life<sup>5-7</sup>. Some patients still require additional treatment using alternative methods such as corticosteroids, rapid maxillary expansion8, myofunctional therapy9, behavioral measures for weight loss and diet10, or CPAP11. The orthodontic treatment for correction of maxillomandibular abnormalities or mandibular retrusion has been shown to improve OSA12. In addition, the sequelae of chronic oral breathing need to be corrected by the global treatment, which should involve a multidisciplinary team of sleep medicine that include: otorhinolaryngologist, speech and/or orthodontic therapy to restore normal breathing and craniofacial growth patterns<sup>13</sup>.

Studies have demonstrated that adenotonsillectomy can have a positive effect on a number of different aspects of the condition, such as improving behavior, cognition, and quality of life of children with adenotonsillar hyperplasia<sup>14-17</sup>. However, most studies assess any improvements immediately, or for only a few months, after surgery. Few studies have analyzed these results over the long-term, with periods equal to, or greater than one year. Lima Junior et al. (2008)<sup>18</sup> reported that a positive impact on quality of life was maintained for about 15 months after surgery. Significant improvement was observed in sleep-related QOL and behavioral problems in children with OSA during long-term follow-up after adenotonsillectomy<sup>19</sup>.

Another interesting finding was that, while short-term and long-term benefits of surgery were achieved, the score for the domain of physical symptoms was higher in the long-term, implying that caregivers perceive some recurrence of physical symptoms after surgery in their children despite long-term improvement in quality of life<sup>20</sup>.

Based on these findings, clinicians should note the potential for long-term changes in certain domains of quality of life; therefore, children with OSA require both short-term and long- term quality of life follow up after surgery. OSA can persist after adenotonsillectomy in 21% to 75%<sup>21,22</sup>. The possible factors that influence the failure of the surgery are patient comorbidities such as obesity, craniofacial anomalies, and nasal obstruction<sup>21,23</sup>.

Patients with residual OSA continue to have a low quality of life<sup>24</sup>. However, few studies have addressed quality of life in patients with only residual snoring after late adenotonsillectomy surgery. SDB (respiratory sleep disorder) is widely studied due

to its high prevalence, but it is not often considered in routine childcare visits<sup>25,26</sup>.

Failure to treat children with OSA can lead to a range of impairments that include behavioral problems<sup>27,28</sup>, increased cardiovascular dysfunction<sup>29,30</sup>, and impaired growth and development<sup>30</sup>. One of the complications of OSA in children is the increased prevalence of neurodevelopmental<sup>31,32</sup>. Children with primary snoring are also at risk of developmental disorders and behavioral disorders<sup>32,33</sup>. Persistent snoring and nocturnal enuresis can affect interpersonal relationships and quality of life<sup>34</sup>. Therefore, the objective of this study is to evaluate the quality of life by the OSA-18 of children with residual snoring 2 years after adenotonsillectomy surgery and a control group without snoring complaints.

# MATERIAL AND METHODS

In this pilot study, the sample comprised 2 groups, one with snoring children at least 2 years after adenotonsillectomy surgery, and a control group without snoring. Four hundred caregivers of patients who had undergone adenotonsillectomy at Hospital São Paulo were interviewed. Children who continued to have snoring symptoms were recruited into the study. The inclusion criteria were children, boys and girls, aged between 5 and 12 years old. The exclusion criteria were children with cardiac, pulmonary, neuromuscular diseases, and chromosomal syndromes. This study was approved by the research ethics committee of the Universidade Federal de São Paulo, number 0698011806/2017. The clinical trial registration number is RBR-463byn.

# Snoring group

The group comprised 14 snoring children who were selected from patients, attending the Outpatient Clinic of the Pediatric Otorhinolaryngology Department at the Escola Paulista de Medicina, Universidade Federal de São Paulo, who had previously undergone adenotonsillectomy (a minimum period of 2 years) and had a severe snoring complaint on most nights, or every night, over the previous four weeks (questionnaire OSA-18). All children in this group underwent an otorhinolaryngological clinical examination, nasofibroscopy, and polysomnography.

# Control group

The control group were selected from the Escola Paulistinha de Educação, an elementary school of education for Unifesp workers, and comprised 11 children selected at random who did not snore, according to the results of the OSA-18 questionnaire.

# **Procedures**

OSA-18 Questionnaire

The OSA-18 is a questionnaire (Supplementary Table) designed to be used with children, validated in Portuguese<sup>34</sup>, that assesses the quality of life in children with apnea<sup>35</sup>.

OSA-18 consists of 18 questions divided into 5 domains: sleep disorder, physical symptoms, emotional symptoms, daytime function and caregiver concerns, each item has a score of 7 points (1 - "never" to 7 - "always")<sup>34-36</sup>. Values less than 60 have a mild impact on quality of life, 60 to 80 have a moderate impact on quality of life, and above 80 is considered a serious impact on quality of life.

In healthy volunteers, the caregivers completed the questionnaire, while in individuals with residual snoring it was carried out by the children's caregivers, in the presence of an otorhinolaryngologist or orthodontist.

#### Polysomnography

Only patients with residual snoring underwent an overnight polysomnography exam. The polysomnography considered the following values: children with an obstructive apnea/hypopnea index (OAH) >2/hour, or an obstructive apnea index (OAI) >1/hour, were considered abnormal, in addition to percutaneous oxyhemoglobin saturation (SpO<sub>2</sub>) <92%  $^{37}$  (Table 1).

Table 1. Snorer sample: polysomnography characteristics.

1 1 7 8	1 7
Variables	Snorer
Sleep efficiency	80.7 % ± 13.3
Sleep latency	$45.1\% \pm 40.4$
Arousal index	$7.8 \% \pm 3.7$
АНІ	$1.4 (n^{\circ}/h) \pm 1.3$
Obstructive AHI	$1.1 (n^{\circ}/h) \pm 1.2$
Central AHI	$0.3  (n^{\circ}/h) \pm 0.4$
RDI	$1.5 (n^{\circ}/h) \pm 1.4$
Baseline SpO <sub>2</sub>	96.6 % ± 1.1
Mean SpO <sub>2</sub>	95.7 % ± 1.4
$\operatorname{Minimum} \operatorname{SpO}_2$	91% ± 3.3

Notes: AHI: Apnea-hypopnea index; RDI: Respiratory disturbance index; SpO2: Oxygen saturation.

#### Statistical analysis

Shapiro-Wilk tests were used to evaluate the distribution of the variables. Descriptive data were presented as means and standard deviations or absolute frequency, and data were analyzed using a generalized linear model (GzLM) test. For the categorical variables, a GzLM with binary logistic distribution was used, and gamma distribution was used for continuous variables. A *p*- value of ≤0.05 was considered significant.

# **RESULTS**

The sample comprised 25 children, 14 snorers after adenotonsillectomy, and 11 controls. The evaluation of the OSA-18 questionnaire was controlled by age values. There was no difference between groups in respect of the anthropometric absolute and z score, and sex (Table 2). However, the snorers were older than the control group. All domains of quality of life in the OSA-18 were worse in the snorer group than in the control group. As a result, the

snorer group had the highest total OSA-18 score, as well as the highest score in all 5 domains compared to control. The overall average score for OSA-18 was 82.5 (severe impact), which was statistically significant compared to the control group (p>0.05). In the descriptive assessment of polysomnography, snorers had an average obstructive apnea/hypopnea index of  $1.1\pm1.2$  and a minimum saturation of  $91\% \pm 3.3$  and a  $95.7\% \pm 1.4$  (Table 3).

Table 2. Total sample characteristics.

Variables	Control	Snorers	p
Males, n(%)	11 (79)	14(56)	0.30
Age, mean(SD)	$7.9 \pm 1.2$	$9.7 \pm 1.8$	< 0.01
BMI z-score, mean (SD)	$2.0 \pm 2.5$	$1.7 \pm 2.8$	0.71

Notes: GzLM test; SD: Standard deviation.

Table 3. Total sample: OSA-18 characteristics.

Variables	Control	Snorers	p
Sleep disturbance	5 ± 1.6	19.6 ± 5.6	< 0.01
Physical symptoms	$8.4 \pm 6.8$	$18.2 \pm 5.1$	< 0.01
Emotional symptoms	$4.9 \pm 2.8$	$13.4 \pm 4.5$	< 0.01
Daytime function	$6 \pm 3.7$	$12.8 \pm 6.0$	< 0.01
Caregiver concerns	$6.3 \pm 3.2$	$19.2 \pm 7.9$	< 0.01
Total	$30.5 \pm 12.4$	$83.1 \pm 20.7$	< 0.01

Note: GLZM test, adjusted for age.

#### **DISCUSSION**

All domains of quality of life evaluated by the OSA-18 were worse in the snoring group after adenotonsillectomy compared to healthy individuals.

The highest mean scores of the OSA-18 were observed for the domains "sleep disturbance", followed by "caregiver concerns", and "physical symptoms", which involve questions about the most common aspects of apnea, namely snoring, choking, restless sleep, mouth breathing, upper airway infection, difficult feeding, concern of the parents about the child's health. Lee et al. (2015)<sup>20</sup> in their study also scored for the domain of physical symptoms was higher in the long run, implying that caregivers perceive some recurrence of physical symptoms after surgery in their children despite the long-term improvement in quality of life. However, Mitchell et al. (2004)<sup>38</sup> found results similar to our study also in the domains sleep disorders and physical suffering were significantly lower (*p* .005) in the short-term than in the long-term.

This study is relevant, because the impact of residual snoring on the quality of life of children who had undergone adenotonsillectomy 2 or more years previously is of great importance. It was observed that in all domains of quality of life evaluated by the OSA-18 were worse in the snoring group after adenotonsillectomy compared to healthy individuals. These results should alert all professionals who deal with patients with similar profiles to check for the presence of snoring in the long-term postoperative period and to seek treatment alternatives.

The data found in this study can contribute to the reevaluation and retreatment of these children to improve their quality of life. It is interesting to note that there are many studies that show that children with primary snoring present changes in their quality of life and more behavioral problems when compared to the control groups<sup>39-43</sup>. However, our study uses a sample of children at least 2 years after the adenotonsillectomy who have signs and symptoms of OSA. Also a systematic review demonstrated the effectiveness of adenotonsillectomy in improving the quality of life of pediatric patients with OSA44. All studies involving shortterm follow-up (≤6 months) have shown improvements in quality of life scores after adenotonsillectomy compared to preoperative values and studies involving long-term follow-up (>6 months) have shown mixed results. We must consider the low educational level and socioeconomic level of parents or guardians and their difficulty in accessing specialized services of the Brazilian health system (SUS). Children who continued to have snoring symptoms were recruited into the study.

Residual snoring should be thoroughly investigated in clinical practice, especially after adenotonsillectomy, as quality of life may be compromised in these children.

Quality of life has been increasingly seen as an important health outcome. OSA-18 is an easy and fast test, with high reliability and consistency, to assess the subjective aspects of quality of life<sup>7</sup>. This questionnaire, when combined with other clinical and objective parameters, allows professionals to better assess the impact of OSA on children and their family life, and to select the best type of treatment<sup>24</sup>. Being a subjective tool, it should be used as an adjunct to clinical examination to improve the diagnosis of pediatric OSA and not a replacement for pediatric polysomnography<sup>7</sup>.

The presence of nasal disuse and breathing through the mouth are abnormal functions that cause developmental and functional changes that are associated with the development of DRS. The impact of mouth breathing promotes changes in craniofacial growth patterns and how these changes lead to impaired developmental functions and the consequent persistence of DRS. Understanding the dynamics that lead to the development of DRS and recognizing the factors that affect craniofacial growth and the resulting functional impairments, allows for an adequate multidisciplinary treatment planning for sleep apnea in childhood. The increase in lymphoid tissue may actually be a consequence and not a cause of these initial dysfunctions<sup>45</sup>.

This study is unique for investigating long-term (>2 years) quality life of children submitted to adenotonsillectomy and the comparison with a control group. However, is has some limitations, including small sample size, limited spectrum of the condition, and a non-surgical control group.

As the effect size of the difference between groups were huge, the small sample size did not affect the final results.

Were choose to have a healthy control group, instead of a post-surgical, in order to have a purer comparison. Yet, a study comparing children post-adenotonsillectomy with and without residual snoring still need to be done. Confirmatory results are essential, and the repetition of studies by other researchers, replicating the conditions described, will produce the evidence for the advancement of science.

#### **CONCLUSION**

The data suggest that snoring patients two or more years after adenotonsillectomy surgery have worse quality of life compared to the control group in long-term follow-up.

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OSA-18 for children with OSAS - English version.	
OSAS Quality of Life Survey (OSA-18) Date: _ /_	/_

For each question below, please circle the number that best describes how often each symptom or problem has occurred during the past 4 weeks. Please circle only one number per question. Thank you.

	None of the time	Rarely 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	the time	of the time	the time	the time	or the time	the time	the time
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 made gasping sound while asleep?	1	2	3	4	5	6	7
restless sleep or frequent awakenings from sleep?	1	2	3	4	5	6	7
Physical Symptoms							
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 upper respiratory infections?	1	2	3	4	5	6	7
nasal discharge or a runny nose?	1	2	3	4	5	6	7
difficulty in swallowing foods?	1	2	3	4	5	6	7
Emotional Symptoms							
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 behavior?	1	2	3	4	5	6	7
discipline problems?	1	2	3	4	5	6	7
Daytime Function							
During the past 4 weeks, how often has your child had							
excessive daytime sleepiness?	1	2	3	4	5	6	7
aggressive or hyperactive behavior?	1	2	3	4	5	6	7
discipline problems?	1	2	3	4	5	6	7
Caregiver Concerns							
During the past 4 weeks, how often have the problems described above.							
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

Port	uguese	version	of OSA-1	8 (OSA-	18-pv)						
OSA	-18 Ver	são Port	uguesa (C	OSA-18- <sub>1</sub>	ov) Da	ta: _ / _ /	_				
Non	ne:										
172							,	 1.	,	- 11	

Em cada uma das questões seguintes, faça por favor um círculo à volta do número que melhor descreve a frequência de cada sintoma ou problema nas últimas 4 semanas. Assinala apenas um número por questão. Obrigado.

	Nunca	Quase Nunca	Poucas Vezes	Algumas Vezes	Bastantes vezes	Quase Sempre	Sempre
Distúrbio do Sono							
Nas últimas 4 semanas, com que frequência o seu filho teve							
ressonar alto?	1	2	3	4	5	6	7
paragens na respiração durante a noite?	1	2	3	4	5	6	7
engasgos ou respiração ofegante enquanto dormia?	1	2	3	4	5	6	7
sono agitado ou despertares frequentes do sono?	1	2	3	4	5	6	7

Continue...

# Continuation...

Sintomas Físicos							
Nas últimas 4 semanas, com que frequência o seu filho teve							
respiração bucal por obstrução nasal?	1	2	3	4	5	6	7
resfriados ou infecções das vias aéreas superiores?	1	2	3	4	5	6	7
secreção e congestão nasal?	1	2	3	4	5	6	7
dificuldade em engolir alimentos?	1	2	3	4	5	6	7
Problemas emocionais							
Nas últimas 4 semanas, com que frequência o seu filho teve							
alterações do humor ou acessos de raiva?	1	2	3	4	5	6	7
comportamento agressivo ou hiperactivo?	1	2	3	4	5	6	7
problemas disciplinares?	1	2	3	4	5	6	7
Problemas do quotidiano							
Nas últimas 4 semanas, com que frequência o seu filho teve							
sonolência diurna excessiva?	1	2	3	4	5	6	7
episódios de falta de atenção ou concentração?	1	2	3	4	5	6	7
dificuldade ao levantar da cama de manhã?	1	2	3	4	5	6	7
Opinião do Informante							
Nas últimas 4 semanas, com que frequência os problemas acima descritos							
causaram preocupação com a sua saúde?	1	2	3	4	5	6	7
preocuparam-no pelo seu filho não poder respirar ar suficiente?	1	2	3	4	5	6	7
interferiram com as suas actividades diárias?	1	2	3	4	5	6	7
deixaram-no frustrado?	1	2	3	4	5	6	7