ORIGINAL RESEARCH

Adenotonsillectomy and obstructive sleep apnea in children: A prospective survey

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OBJECTIVE: Prospective survey of children up to 14 years of age with OSA submitted to adenotonsillectomy.

METHODS: Clinical evaluation, with questionnaires and clinical scales evaluating facial structures including tonsils and Mallampati scales and otolaryngologic evaluation; nocturnal polysomnography and repeat evaluation three to five months postsurgery. **RESULTS:** Of 207 successively seen children, 199 had follow-up polysomnography, and 94 had still abnormal sleep recording. Multivariate analysis indicates that Mallampati scale score 3 and 4, retro-position of mandible, enlargement of nasal inferior turbinates at +3 (subjective scale 1 to 3), and deviated septum were significantly associated with persistence of abnormal polysomnography (with high 95% CI for Mallampati scale and deviated septum).

CONCLUSION: Mallampati scale scores are resultant of several facial factors involving maxilla, mandible, and oral versus oral breathing but add information on risk of partial response to adenotonsillectomy.

SIGNIFICANCE: Adenotonsillectomy may not resolve obstructive sleep apnea in children.

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A denotonsillectomy is the treatment of choice for obstructive sleep apnea (OSA) in children. A prospective survey was conducted during a three-year period in children between the ages of 18 months and 14 years. All children were nonsyndromic and had no chronic illness, an exclusion criterion of the study. All children were seen at a multidisciplinary sleep clinic and were evaluated by the same sleep specialist and otolaryngologist. Adenotonsillectomy was performed by different otolaryngologists; selection of the surgeon was determined by either the insurance plan or the referring pediatrician. The importance of postoperative follow-up was emphasized to the parents as well as the referring pediatricians. Variables examined included the anatomic findings at the initial evaluation, the results at three to five months following surgery, and the anatomic features related to the persistence of sleep-disordered breathing following surgery.

METHOD

Children were referred for variable symptoms (Table 1). Most of them presented a history of some degree of snoring, independent of other complaints. All children had a private pediatrician, but 15% of the parents initiated the consult themselves. Information was obtained from private pediatricians. All children had pediatric evaluations including body mass index, a sleep/wake and sleep medicine history, and the Pediatric Sleep Questionnaire.¹ Parents were asked to fill out a seven-day sleep diary indicating sleep periods and timing.

All of the children underwent a systematic airway examination with the use of subjective scales. This examination followed a standardized approach with investigation of external and internal nasal valves conducted based on frontal examination to determine whether there was depression on

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Table 1 Complaints and causes of referral

Symptoms	Ν	%
Noisy breathing or snoring	174	84
Agitated sleep or disrupted nocturnal		
sleep	163	79
Nocturnal awakenings, insomnia	47	23
Daytime fatigue	95	46
Daytime sleepiness with regular		
napping	19	9
Morning headache	31	15
Sleep terrors	27	13
Sleepwalking (with and without sleep		
terrors)	41	20
Persistence of bed-wetting	26	12.5
Poor eater or failure to thrive, growth		
problems	13	6
Delayed puberty	6	3
Hard to wake up in the morning	59	28.5
Delayed sleep phase syndrome	22	10.6
Learning difficulties	78	38
Abnormal daytime behavior	49	23.7
a. aggressiveness		
b. hyperactivity		
c. inattention		
d. daytime fatigue		
Abnormal shyness/ depression	17	8
Repetitive URI	39	19
Dental problems appreciated by		
dentist	77	37
a. Cross-bite		
b. Malocclusion (Class II or III)		
c. Small jaw with overcrowding of		
teeth		

the lateral wall at the internal valve region. Clinical examination for presence of deviated septum or enlargement of turbinates by nasal speculum and headlight illumination was used because it allowed for direct visualization of the nasal airway patency in regard to nasal septum morphology as well as the size of the inferior turbinates using a scale from 1 to 3. The nasal nare and region of the external valve located at the lower lateral cartilage region was examined for constriction of the external valve.

The face was assessed via systematic frontal and lateral photographs to determine any asymmetrical development. Several elements were used to evaluate the development of the maxilla and the mandible. Based on prominence of the chin and retro-maxillary or retro-mandibular position,² the facial contour on profile was assessed to determine if there was a C or D facial presentation. The face (frontally examined) was subdivided in three thirds following the established landmarks and presence of an abnormally elongated lower third of the face was noted.² Children were studied for the presence of a high arched and narrow hard palate (scored from 1 to 3). Information was obtained regarding indentation on the lateral tongue border, position of the relaxed tongue, the presence of overlapping teeth, and oc-

clusal patterns. Particularly, subjects were evaluated for possible indicators of a narrow maxilla and/or a narrow mandible, which include an over-jet (antero-posterior distance in millimeters between upper and lower teeth in a relaxed, supine position), over-bite (vertical coverage in millimeters of lower incisors), or cross bite.

Results of these evaluations were summarized on a preestablished standardized form, where the evaluator gave a score to the specific categories (Table 2).

The presence of mouth breathing was assessed during the evaluation, from parental interview, and while the child was distracted and not speaking. Tonsil size was evaluated using the Friedman et al classification.³ The upward extension of tonsils and adenoid size was studied via fiberoptic scope to correct the lateral size score. Both scores were further corrected based on maxillary and mandibular scores. The presence of enlarged pharyngeal walls was scored independently from tonsils on a scale from 0 to 4. Evaluation of the position of the uvula tip compared with the base of the tongue was calculated using the criteria described by Mallampati et al in adults⁴ (Table 2). During assessment, the patient is instructed to open the mouth as wide as possible, while protruding the tongue. Class I is defined as soft palate and entire uvula visible; Class II, soft palate and portion of uvula visible; Class III, soft palate visible (may include base of

Table 2

Results of clinical evaluation using subjective scoring scales

	n	%
Tonsils 0	0	0
1+	3	1.4
2+	75	36.2
3+	97	47
4+	32	15.4
Mallampati score 1	11	5.3
2	21	10.1
3	105	50.7
4	70	33.8
Soft palate redundancy 0	7	3.4
1+	47	22.7
2+	101	48.8
3+	52	25.1
Hard palate 1	9	4.3
2	29	14.0
3	169	81.6
Inferior nasal turbinates 0	13	6.3
1+	24	11.6
2+	97	46.9
3+	73	35.6
Evidence of allergic rhinitis - no	82	39.6
- yes	125	60.4
Nasal septum deviation - no	81	39.1
- yes	126	60.9
Retro-position of mandible - no	112	54.1
- yes	95	45.9
Long face presentation - no	97	46.8
- yes	110	53.2

uvula); Class IV, soft palate not visible. Fiberoptic evaluation was performed several times including by all surgeons before performing surgery. The presence of allergies, particularly upper airway allergies, was questioned. If the status was unknown, atopic status was determined by a skin prick test with a panel of 17 aeroallergens and food allergens (ALK America, Round Rock, TX). The test was administered by an allergist; a positive response was defined by the presence of a wheal that was at least 3 mm greater than the wheal of a control saline prick.

The operations were performed by different surgeons. Information was not sufficient to compare the different techniques, and thus techniques were not controlled. Each child underwent an in-laboratory polysomnography with lights out and total recording time guided by sleep diaries. The following variables were systematically monitored: EEG (C3/A2, C4/A1, Fpz/A1-A2, O1-O2), right and left electro-oculogram, chin and leg EMG, ECG (modified V2 lead), position sensor, nasal-cannula-pressuretransducer system, mouth thermistor, chest and abdominal piezo-electric bands, neck microphone, and finger pulseoximetry. If questions on presence or severity of abnormal breathing during sleep were raised, the clinical evaluation protocol called upon the possibility to perform a second nocturnal polysomnogram with addition of esophageal pressure (Pes) monitoring to accurately determine the presence, degree, and duration of abnormal respiratory effort during sleep. All variables were calibrated before sleep onset. There was continuous video monitoring during total recording time.

Recording time was based on sleep diaries but was a minimum of 7½ hours. Three to five months postsurgery, children had a follow-up evaluation and repeat polysomnography. Performance of clinical survey research was approved by an internal review board and a parent-signed approved informed consent for anonymous usage of the clinical data of their children in research.

Analysis

Scales and questionnaires were scored following published recommendations or previously published data. Most of the subjective scales were Likert scales from 1 to 3 or 0 to 3 or 4, with 0 or 1 usually indicative of "small," "absent," and normal.

Polysomnograms were scored following published criteria. Sleep-wake was scored following the international manual, as were short EEG arousals.⁵⁻⁷ Apneas and hypopneas were also defined following international recommendations. Events were subdivided into central, mixed, and obstructive and were tabulated when longer than two breaths. Using a nasal cannula, hypopneas were scored when the nasal flow curve decreased by 30% compared to prior normal nasal breathing, and apneas were scored when the nasal flow curve was less than 80% of normal nasal breathing curve (see Table 3). The number of oxygen saturation (SaO_2) drops of 3% or more was also tabulated, as was the lowest SaO₂. The scoring of these events based on total sleep time allowed calculation of an apnea-hypopnea index (AHI). The definition of an abnormal AHI was based on the International Classification of Sleep Disorders at greater than one

Term	Definition
Apnea	Absence of airflow at nose and mouth for longer than two breaths, independent of desaturation or change in EEG. Subdivision in central, mixed, or obstructive based on airflow and thoraco-abdominal movements and Pes recording.
Hypopnea	Reduction by at least 30% in nasal flow signal amplitude for longer than two breaths. Scored independently from SaO ₂ drop or EEG arousal. Often but not always associated with snoring.
Abnormal respiratory effort	Reduction in nasal flow of less than 30% with flattening of nasal cannula signal (flow limitation) and decrease in the mouth signal (thermistor). Often seen with snoring and increased effort shown on Pes signal defined as:
 Pes Crescendo^{9,10} 	Sequence of four or more breaths that show increasingly negative peak end inspiratory pressure. May be seen with flow limitation on nasal cannula.
 Continuous sustained effort,^{9,10} 	Repetitive, abnormally negative peak end inspiratory pressures, ending at same negative inspiratory pressure without a crescendo pattern. Associated with discrete flow limitation on nasal cannula/pressure transducer signal, with "flattening" of the breath signal curve for at least four successive breaths.
Pes reversal ^{9,10}	Termination of abnormal increase in respiratory effort with abrupt switch to a less negative peak end inspiratory pressure.
Tachypnea	Increase in respiratory rate, above that seen during quiet unobstructed breathing, by minimum of three breaths/minute in NREM sleep, or four breaths/minute in REM sleep, for 30 seconds or more. Tachypnea is score only if respiratory rate is ≥20 breaths/minutes in children two years or older. No changes in oxygen saturation, Pes, or EEG are required. ^{9,10}

Table 3		
Abnormal	broothing	natto

event/hour.⁸ AHI did not cover all breathing abnormalities. Flow limitation was determined from published criteria and calculated from the nasal flow curve. It was calculated when there was an abnormal flow curve on three successive breaths with "flattening" of the flow-curve or its modification with reduction between 2% and 29% of the normal amplitude of the curve.^{9,10} Breathing was studied for abnormal patterns, such as an abrupt, short peak at the beginning of inspiration followed by an immediate drop of the curve or peak of the curve in the middle of inspiration with flattening at the beginning and end of inspiration. If Pes monitoring to detect respiratory effort was added, two previously described patterns (Pes crescendos and sustained continuous effort) were also scored (Table 3).^{9,10} These events were added to the AHI to give a respiratory disturbance index (RDI), which was calculated based on total sleep time (with abnormal RDI > 1.5 event/hour).¹¹ Finally, children may have an increase in breathing frequency as an important indication of abnormal breathing during sleep. Increase in breathing frequency (Bf) is a physiological defense mechanism when tidal volume decreases to maintain minute ventilation, following the formula: Bf \times tidal volume = minute ventilation. Tachypnea was considered present when the breathing frequency was over 20 breaths/minute after two years of age during sleep, and a tachypnea time was calculated based on the number of 30-second epochs with tachypnea present in the recording.9,10

Children were identified by a number, and all data were placed in a research database that utilized the number as the unique identifier. The same three trained scorers scored all polysomnograms, which were also identified only by a number. Scorers were blind to identify, age, and condition of the child. The interscorer reliability for sleep scoring was 89% and 95% for abnormal breathing identification.

Statistical Analysis

As data were not normally distributed, Mann-Whitney U test was used to compare presurgery and postsurgery data. The percentage was analyzed with χ^2 statistics. We performed a multiple variate regression analysis with AHI greater than 1 as the dependent variable and presence/absence of allergy, presence/absence of narrow and high hard palate (score of 3 vs 1 and 2), presence/absence of retro-position of mandible, enlargement of nasal inferior turbinates (score of 3 vs 1 and 2), Mallampati scale score (3 and 4 vs 1 and 2), and presence/absence of long face presentation as defined in Method section.

RESULTS

Two hundred and seven children (94 girls) were involved in this prospective survey. The age range was 19 months to 14 years; the mean age was 7.3 ± 2.3 and the median age was 6.7 years. One hundred seventy-four (84%) children were known snorers and 126 (61%) were known to have allergies or tested positive with the skin test. Only 17 (8%) were considered overweight based on body mass index (BMI), and only one was considered obese. Per entry criteria, none were syndromic, had a chronic medical illness, or were receiving medications on a chronic basis. Table 1 presents complaints and causes of referrals. The anatomic findings extracted from the subjective evaluation scales are presented in Table 2.

Follow-up investigation occurred 15.3 ± 2.7 (range 12-20) weeks after surgery. A total of 202 children (97.6%) had follow-up clinical evaluation, 199 patients (96%) had follow-up polysomnography after clinical evaluation, and three children (1.4%) had a cardio-respiratory monitoring, not full polysomnography, due to insurance plans. Nineteen surgeons were involved in the treatment of the children.

Isolated adenoidectomy or tonsillectomy was performed only in 19 children out of 202 (9.4%), but treatment of nasal inferior turbinates and nasal valves with adenotonsillectomy was performed by only one surgeon, who performed that surgery on 17.4% of the total group. Different surgical approaches were used for the surgery.

Parents reported improvement of health problem in 187/ 202 (92.6%) children, but their completion of the same scales and questionnaires as at entry showed a discrepancy with interviews, as 101 children (48.8 %) had indication of some persistent problems. Ninety-three percent of these individuals were in the group with elevated AHI and RDI postsurgery. The listed problem was incomplete treatment of the initial complaint, more particularly symptoms of fatigue, nocturnal sleep disruption, and parasomnia, even if all family and patients indicated clear decrease in severity and frequency of occurrence of the problem. The symptoms were often sufficiently intermittent not to be emphasized at a general interview.

Results of the polysomnograms at entry indicated a mean AHI of 8.1 \pm 5 with a range of 2 to 35 events/hour and a median of 7.6 events/hour. The mean RDI was 12 \pm 10 with a range of 6 to 48 and a median of 14 events/hour. Post-surgery (n = 199), the mean AHI was 3.1 (*P* = 0.0001) with a range of 0.2 to 22 and a median of 3.8; the mean RDI was 6 \pm 6.5 (*P* = 0.001) with a range of 0.3 to 27 and a median of 6.9. Polysomnography or cardio-respiratory monitoring (n = 202) indicated that the lowest SaO₂ was improved in all cases. The mean lowest SaO₂ was 90.1% \pm 2.6% at entry (range 85%-94%), and it was 93% \pm 3.1% (range 99%-89%) postsurgery (*P* = 0.0001).

In summary, surgery had a beneficial effect in all cases in our children. But 92/199 (46.2%) children had an AHI greater than or equal to one postsurgery (and 94/202, or 46.5%, if we include the cardio-respiratory investigated children). When the RDI was calculated, the number of affected children was higher with 101/199 (50.7%), demonstrating a RDI greater than or equal to 2. The relationship between subjective scale scores at entry and persistence of elevated AHI and RDI postsurgery are shown in Table 4. Results of the multivariate regression analysis are presented

Variable	N before surgery	N after surgery	% of before surgery	χ^2 (2 ways)
Allergic rhinitis (yes)	125	60	48	
(no)	82	34	41.4	
Inferior nasal turbinate score 3+	73	51	68	0.000
Score 0, 1+, and $2+$	134	43	32	
Deviated septum (yes)	126	52	41.3	0.01
(no)	81	42	51.85	
Mallampati scale score 3 and 4	175	63	36	0.000
score 1 and 2	32	31	9.7	
Retro-position of mandible (yes)	95	45	47.3	
(no)	112	49	43.75	
Hard palate +3 score	169	72	42.6	
+1 and +2 score	38		57.8	

in Table 5. As can be seen, presence of deviated septum, enlarged inferior turbinates (ie, abnormal nasal breathing), retroposition of the mandible, and Mallampati scale scores 3 and 4 were significant predictive variables. The most interesting were the presence of deviated septum and the Mallampati scale score that had very important 95% confidence interval, indicating the strength of the finding. The Mallampati scale scores are resultant of different factors such as limited growth of maxilla, limited growth of mandible, and limited growth of maxilla and mandible (that will lead to a class I dental occlusion). It will be seen with chronic mouth breathing and secondary impact on facial growth.

DISCUSSION

Table 4

Our prospective survey without intervention confirms prior findings but on a larger scale. It also indicates for the first time in children that a simple visual examination with performance of a simple maneuver by the child can indicate risk of persistence of abnormal breathing during sleep postsurgery. Adenotonsillectomy has a beneficial effect on obstructive sleep apnea in children. The most important effect is probably the improvement in lowest oxygen saturation during sleep. It must be emphasized that we only had one obese child in our group.

The SaO₂ drop associated with REM sleep atonia, due to the secondary chest bellows impairment related to physiologic muscle tone change, was not present in the overwhelming majority of our children. It confirms another finding, namely that adenotonsillectomy may not be sufficient to completely eliminate abnormal breathing problems during sleep. In many cases, there are associated anatomical problems that contribute heavily to the sleep-disordered breathing (SDB). The fact that persistent elevated AHI was seen postsurgery had been suggested on a small scale by two studies in 1980 and 1990, which looked at very longterm follow-up.^{12,13} This finding was recently re-emphasized in a shorter follow-up.^{14,15} Our study is one of the largest prospective studies to date with systematic collection of data.

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Significant factors associated with subjects with elevated AHI postsurgery

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Variables*	Wald	Odds ratio (95% CI)	<i>P</i> value
Inferior nasal turbinate score 3+ Deviated septum Mallampati scale score 3 and 4 Retro-position of mandible	21.82 14.14 17.137 5.447	0.004 (0-0.04) 141.93 (10.72-1878.71) 187.74 (15.74-2239.02) 4.89 (1.29-18.54)	0.000 0.000 0.000 0.020

*Data derived from multivariate logistic regression (forward elimination used AHI > 1 as dependent variable). Note the important 95% confidence interval (CI) for Mallampati scale score and deviated septum.

The ubiquitous presentation of abnormal breathing during sleep has been reviewed and clinical complaints of our children, from pseudo ADHD, learning difficulties, parasomnias, daytime abnormal behavior, and diverse types of sleep disorders, are similar to what has been previously reported in association with abnormal breathing during sleep.⁹ Nasal allergy as a cause of SDB is known, and its association with other anatomical changes is not surprising. The development of the anatomical risks for occurrence of OSA in children can involve the presence of chronic nasal allergies from early childhood.¹⁶ Congenital nasal septum deviation, chronic nasal allergies, and familial traits involved in maxillo-mandibular growth have been suggested as important risk factors in the development of SDB in children during early infancy, particularly with the knowledge that 60% of the adult face is already built by four years of age.¹⁷ These different factors, their potential role, and the lessons learned from animal models with abnormal nasal resistance have been previously reviewed and well summarized.^{18,19} Allergies are known to be frequent in the climatic conditions of the San Francisco Bay Area. The allergic status of a child with SDB should be investigated. It also raises the question of treating enlarged inferior nasal turbinates more aggressively when seen at examination: impairment of nasal breathing with increased nasal resistance has a well-documented negative impact on the early childhood maxillo-mandibular development, a concerning element considering the speed at which growth occurs. Due to the impact on facial growth, no direct treatment of nasal septum deviation can be performed early, but radio-frequency treatment of nasal inferior turbinates will reestablish better nasal breathing and decrease nasal resistance much faster than nasal steroidal medications, which may take months for an appropriate response.^{10,19} Consideration should be given to using both approaches simultaneously.

The multiple regression analysis indicates that nasal problems and facial structures can predict incomplete response to adenotonsillectomy. The most interesting outcome from this analysis was the demonstration of the validity of the Mallampati scale. This scale has been well used in adults⁴ but not in children. We grouped scores 3 and 4 together and consider these abnormal, as is done in adults with OSA. Our group of children had a mean and median age around seven years and there is no problem having individuals in this age range undergo the maneuver described in Mallampati et al to assess tongue base airway. With parental cooperation, good participation of children three years and older was obtained. Also, as the growth of the face is very rapid the position of the hyoid bone is not an issue when children are performing the maneuver and adult definitions can be used. The position of the tip of the uvula compared to the base of tongue is related to different factors and retroposition of mandible, narrow maxilla, and mouth breathing and its consequences on the growth of the cranio-facial skeleton play a role in the score. It can be seen with a class I dental

occlusion seen with a combined maxillary and mandibular growth limitation.

Determination of the position of the tip of the uvula compared to the base of tongue is easy to determine during a clinical evaluation and may lead the examiner to question why there is such presentation. As mentioned, the important 95% confidence interval at the multivariate analysis indicates the strength of the finding and the importance of determining this score when possible during clinical examination.

There are limitations to our survey. The patients studied were mostly from a middle-class socioeconomic background. As mentioned, only 17 overweight children and one obese child were included in the survey; thus, the role of weight cannot be extracted from this group of primarily nonobese children. Also, there are many ways to perform adenotonsillectomy. Many surgeons have different techniques and views on the surgical approach, and the approach can play a role in the short-term (three to four months) outcome of surgery for SDB in children. Data on surgical techniques in our children are lacking. Persistence of abnormal AHI may be related to this technical issue as shown previously,¹¹ and we acknowledge this limitation. Technical factors, such as associated usage of radio-frequency treatment of inferior nasal turbinates as performed by one surgeon, are unaccounted for in our analyses. OSA is often mentioned to be recognized at an earlier age than the mean and median age of seven years of our group. Despite the fact that school difficulties and behavioral problems are less tolerated in school than home, these issues are strong incentive for parents to seek treatment. Despite the number of children involved, this is a case control survey and not a general population study, and we acknowledge this factor.

One of the obvious findings of our study is the need to perform follow-up sleep evaluation post-adenotonsillectomy. Tonsils may appear enlarged not due to enlargement of the lymphoid tissues but rather due to the narrow bone structures that many children with SDB present.^{15,19} Also, the tonsils may have a significant upward extension rather than a lateral one. Adenotonsillectomy may be performed because the soft tissues occupy space; their elimination will allow better air exchange, but this simple surgery will not resolve other anatomic problems that may be present in children with OSA. Many of the children seen had a narrow face or an abnormal antero-posterior facial development. It is unknown if the abnormal nasal resistance, which may have been present since birth in some cases, had a role in the current clinical presentation. Considering the known impairment of cognitive functions and personality disorders with persistence of abnormal breathing during sleep, it is imperative to completely address the anatomic impairment and residual abnormal breathing. Orthodontic approaches have been shown to be useful in children with persistent SDB post-adenotonsillectomy. These approaches (aggressive treatment of allergies including usage of leukotriene receptor antagonist medication at bedtime, orthodontic treatments if needed) may not resolve all problems but may further improve the residual SDB. Clear enlargement of inferior nasal turbinates, a known consequence of upper airway allergy, was one of the significant factors in the multivariate analysis, despite its low 95% CI. Sometimes, no solution other than nasal CPAP usage will be possible as a stop-gap before considering orthognathic surgery²⁰ during teenage years. But recognition of those children who need further attention is crucial. Given that the structural problems seen in many children with SDB will not be fully resolved with adenotonsillectomy, the risks of slow but progressive recurrence of full obstructive sleep apnea syndrome warrant postsurgical testing. The possibility of treatment beyond adenotonsillectomy should be explained to the parents.

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