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SCIENTIFIC INVESTIGATIONS

Prevalence of malocclusions and oral dysfunctions in children with persistent sleep-disordered breathing after adenotonsillectomy in the long term

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Study Objectives: To evaluate the prevalence of craniofacial/orthodontic abnormalities and oral dysfunctions in a population of children with persistent sleep-disordered breathing despite adenotonsillectomy.

Methods: Medical charts of 4,000 children with sleep-disordered breathing operated on in a tertiary hospital were retrospectively reviewed. Patients reporting persistent sleep-disordered breathing symptoms were invited to an orthodontic/myofunctional evaluation following the Sleep Clinical Score), followed by a 1-night ambulatory type III sleep study.

Results: One hundred nonsyndromic symptomatic patients were examined (mean age 8.8 ± 3.5 years), from 1 to 12 years after surgery (mean 4.6 ± 3.1 years); 24% were overweight/obese; 69 had a sleep study. Although prevalent, oronasal abnormalities and malocclusions were not specifically associated with pathological sleep parameters (cartilage hypotonia 18%, septal deviation 5%, short lingual frenulum 40%). Malocclusions were associated with a higher respiratory event index in children under 8 years only, whereas an impaired nasal dilator reflex and tongue immaturity were associated with an increased obstructive respiratory event index in all patients (1.72 ± 2.29 vs 0.72 ± 1.22 events/h, $P = .011$) and Respiratory Event Index, respectively (3.63 ± 3.63 vs 1.19 ± 1.19 events/h). Male sex, phenotype, nasal obstruction, oral breathing, and young age at surgery (< 3 years) were significantly related to higher respiratory event index. Using the Sleep Clinical Score > 6.5 cut-off, patients with persistent sleep apnea were significantly distinct from chronic snoring (2.72 ± 2.67 vs 0.58 ± 0.55, $P < .01$).

Conclusions: Oronasal anatomical and functional abnormalities were quite prevalent and various in persistent sleep-disordered breathing after adenotonsillectomy. Nasal disuse and tongue motor immaturity were associated with a higher obstructive respiratory event index in the long term, whereas craniofacial risk factors might have a more pronounced impact at younger age.

Keywords: sleep-disordered breathing, obstructive sleep apnea, child, adenotonsillectomy, adenoidectomy, craniofacial, malocclusions, functional assessment

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BRIEF SUMMARY

Current Knowledge/Study Rationale: Long-term studies of adenotonsillectomy or sole adenoidectomy suggest an incomplete resolution of sleep-disordered breathing in certain populations or its possible relapse with time. The aim of this study was to evaluate the prevalence of craniofacial, oronasal abnormalities, and dysfunctions in nonsyndromic children and adolescents with persistent sleep-disordered breathing after surgery.

Study Impact: In our sample of mild to moderate persistent sleep-disordered breathing, several clinical phenotypes were found, including a prevalence of 24% in children and adolescents who were overweight/obese. Oral breathing impaired nasal dilator reflex and tongue motor immaturity were more associated with increased apnea-hypopnea indexes than were craniofacial features or malocclusions. The Sleep Clinical Record (when score > 6.5) could adequately discriminate persistent obstructive events from persistent chronic snoring.

INTRODUCTION

Sleep-disordered breathing (SDB), a spectrum of disorders affecting both pediatric and adult populations, arises from repetitive episodes of partial or complete obstruction of the upper airways during sleep. SDB has various degrees of severity,[1] starting with primary snoring, affecting approximately 12% of children, to obstructive sleep apnea (OSA), which affects from 1.2 to 5.7%.[2] Left untreated, OSA has been related to significant morbidity in children: growth failure, neurocognitive and behavioral abnormalities, reduced academic performance, and also cardiovascular effects.

SDB conditions share a common pathophysiology, ie, the narrowing of upper airways, resulting from an increased soft tissue content, a volumetric reduction of the facial skeletal frame, an alteration of its neuromuscular tone, or a combination of those factors.

In children, the most described cause of upper airway obstruction is the hypertrophy of adenoids and palatal tonsils[3] (part of Waldeyer's lymphoid ring), potentially aggravated by a nasal obstruction and associated with mild abnormalities of the facial skeleton, such as a narrow maxilla and/or a retruded, steep mandible.[4] The asynchronism of lymphoid growth velocity compared with skeletal growth[5] explains the peak prevalence of

SDB, clinically observed between 3 and 5 years of age[6], but some patients might be affected with obesity, similar to the most frequent type of OSA in adults. Syndromic patients constitute another phenotype, associated with major alterations of craniofacial growth or neuromuscular tone and they display early and complex forms of OSA.

In children with symptoms, polysomnography (PSG) is the gold standard method to score respiratory events. There is agreement on the definition of pathological indices in children, with a pathological limit of obstructive apnea-hypopnea index > 1.5 events/h: an index < 5 defines a mild OSA, an index between 5 and 10 is a moderate OSA, and an index exceeding 10 is a severe OSA.

For patients whose treatment cannot be postponed, especially those with a high apnea-hypopnea index, adenotonsillectomy (A&T) is the first-line treatment of OSA. The CHAT (Childhood Adenotonsillectomy Trial) study prospectively evaluated OSA outcomes after A&T and reported an overall success of 79%, defined as an apnea-hypopnea index < 2 events/h and an obstructive apnea index < 1 events/h. Nevertheless, several studies report the persistence of SDB in 20 to 40% of patients after A&T,[7,8] the success rate being significantly worse when residual OSA is defined as an apnea-hypopnea index >1 events/h on postoperative PSG rather than reported symptoms.[9]

Data on long-term outcomes of A&T and sole adenoidectomy are scarce,[10,11] but it has been suggested that there is an incomplete resolution of SDB in certain populations, such as patients with craniofacial abnormalities, especially syndromic (such as in Down syndrome[12]), patients with obesity, or children with underlying asthma or allergic rhinitis.[13] SDB could also possibly relapse with time.[14]

Therefore, the aim of the present study was to assess the long-term prevalence of persistent SDB symptoms following adenoid and/or tonsil surgery and to evaluate the contribution of various risk factors, such as obesity, craniofacial nonsyndromic features[4] (transverse deficiency of the jaws, maxillary and/or mandibular sagittal deficiencies—retrognathia—and/or excessive or deficient vertical growth, with associated malocclusions) and oronasal dysfunctions, that can persist and worsen beyond early childhood.

METHODS

Study design

This is a prospective observational study. The study protocol was approved by the Ethical Committee of St Justine Hospital, Montreal, Canada (# 2016-1033), in accordance with the 1964 Helsinki declaration and its later amendments. Consent was obtained from the children's legal guardians, and patients themselves when able to understand and sign the informed consent forms, for each section of the study.

Participants

Participants were recruited from the Ear Nose and Throat unit of a pediatric tertiary hospital, who had previously undergone surgery of adenoids and/or tonsils between January 2000 and March 2016, with the necessary report of chronic snoring or witnessed apneas in their preoperative clinical charts. They were invited to participate in this study, which included 3 separate visits (V1–V3). During V1, participants completed the hierarchic severity clinical scale (HSCS)[15,16] to assess self-reported persistent SDB symptoms; these results were previously published.[17] Participants who reported chronic snoring (HSCS > 0) or who scored positive for suspected OSA (HSCS > 2.72) in absence of craniofacial syndrome were invited to complete the second (V2) and third (V3) study visits (see **Figure 1**). V2 consisted of a physical evaluation to assess craniofacial morphology, oronasal function, and body mass index (BMI); V3 consisted of a home sleep apnea test (HSAT). Participants and their parents could choose to complete all or only 1 of these study visits. All study data were collected and managed using a secure electronic data capture tool for research, the RedCap platform (Research Electronic Data Capture), hosted at the Université de Montréal.

Physical examination

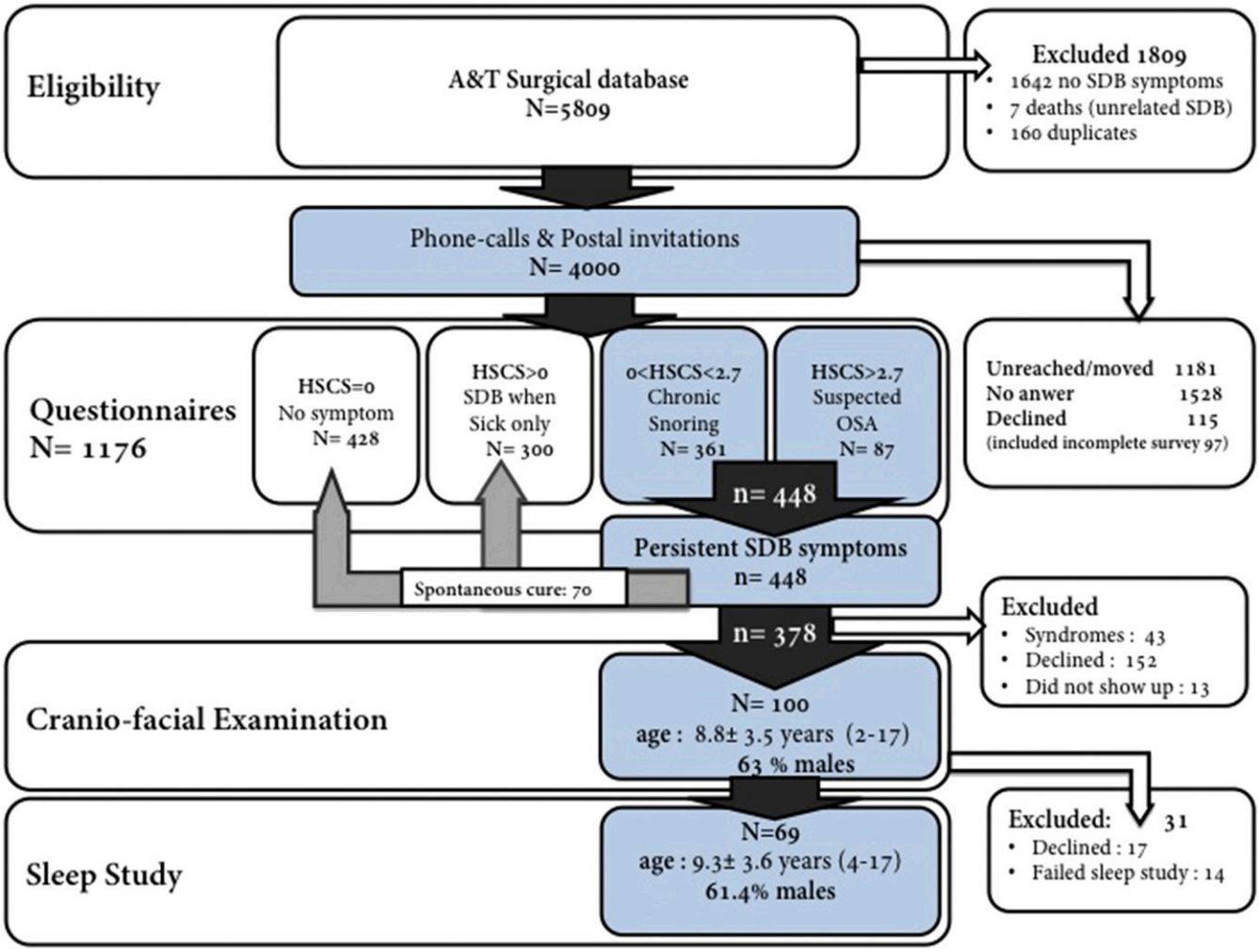
The craniofacial morphology and oronasal functional assessments at V2 were completed by a single trained orthodontist (JCL). Results from the physical examination were used to calculate the Sleep Clinical Record total score. This tool was developed by Villa et al[18] to identify children with OSA and validated for persistent OSA after treatment[19] (see supplemental material) and includes the clinical history and physical examination of each participant.

Physical examination comprised the nose (septal deviation, Glatzel mirror test, sniff test, Gudin's reflex[20], Rosenthal's test), tongue (relative volume, frenulum[21], protraction/elevation tests), oropharynx (Friedman palate and tonsils scales), craniofacial proportions, and occlusion (see supplemental material). Children's height (in cm) and body weight (kg) were measured on an electronic scale (Seca column scale).The BMI was calculated as the body weight divided by the squared height (kg/m^2), BMI z-scores (ie, BMI measured in terms of standard deviations from the mean according to age and sex) were calculated using the WHO Anthroplus software (available at https://www.who.int/growthref/tools/en/; see supplemental material). Obesity was defined by BMI z-score of > +2 standard deviations. Clinical history comprised parental report of Attention Deficit Hyperactivity Disorder or other neurological symptom, daytime somnolence, headache, frequent awakenings or agitated sleep, enuresis, and medication.

Home sleep apnea test

The HSAT was a 1-night polygraph study done with a type 3 ambulatory device, the NOX T-3 Sleep Monitor (Nox Medical, Reykjavik, Iceland). The system comprises a microphone, a wireless oximeter/cardiac pulse meter, thoracic and abdominal belts, a nasal canula, and oronasal thermistor. This system demonstrated very good measurement agreement compared with in-laboratory PSG and a high degree of sensitivity for detecting even mild OSA.[22] Respiratory events were manually scored by an external sleep laboratory (Sleepstrategies.com, Ottawa, Canada) following the guidelines of the American Academy of Sleep Medicine and indexes calculated accordingly:[23] REI (number of respiratory events/monitoring time), obstructive REI (OREI number of obstructive respiratory events/

Figure 1—Flowchart.



A&E = adenotonsillectomy, HSCS = hierarchic severity clinical scale, SDB = sleep-disordered breathing.

monitoring time), and oxygen desaturation index (ODI; number of 3% desaturation/monitoring time events).

Statistical analysis

All results were presented as mean ± standard deviation for continuous variables and as percentage (%) for nominal variables. Statistical analyses were carried out using IBM-SPSS Statistics for Windows (Version 24) for descriptive statistics, parametric (*t* test), and nonparametric tests (Mann-Whitney *U* test, Kruskal-Wallis test). The null hypothesis was rejected at $P < .05$.

RESULTS

Demographics

Overall, persistent SDB symptoms were reported in 448 participants among those who completed V1[21] (**Figure 1**). They were invited to continue onto V2 and V3 between April 2016 and May 2017. However, 70 participants reported spontaneous resolution in the meantime (60 from chronic snoring group, 10 from suspected OSA group). Mild SDB was suspected in 301 children (HSCS > 0 with chronic snoring, 25.59%), whereas 77 (6.54%) had an HSCS > 2.72, suggesting persistent OSA. Among 378 candidates for V2 and V3, 43 were excluded because of a syndromic or compromised medical condition, 152 declined to participate, and 13 missed their scheduled appointments. Thus, 100 consecutive participants with reported persistent SDB symptoms who were representative of the entire cohort underwent the physical examination (V2, see **Table 1**). Finally, 69 participants completed the HSAT (V3), since 17 declined and 14 refused to repeat their HSAT after it failed initially. For 2 participants, sleep study results were taken from an in-hospital PSG study done at the same study center and within 3 months of V1.

General characteristics of the participants at V2 and V3 are described in **Table 1**. These visits were done between 1 and 15 years after surgery. The study sample was predominantly male, between 63 and 64.4%. Male patients had been operated significantly younger than girls (3.63 ± 1.84 vs 4.92 ± 2.67 years, $P = .02$). Patients were from various origins (**Table 2**): 53%

Table 1—Characteristics of the surgical cohort, questionnaire, and examined children samples.

	Male Sex	Age of Surgery (years)	Age at Survey (years)	Years from Surgery
Database: surgical SDB cohort n = 4,000	57.7% n = 2,306	4.23 ± 2.1 (0–18)	10.57 ± 3.7 (2–19)	6.30 ± 3.4 (1–17)
V1 Questionnaire Sample n = 1,176	56.3% n = 663	4.34 ± 2.2 (0–18)	9.54 ± 3.7 (1.9-19.5)	5.19 ± 3.4 (1–16)
V2 Examined Symptomatic Children n = 100	63% n = 63	4.09 ± 2.2 (0–12)	8.78 ± 3.5 (2.5-17.5)	4.64 ± 3.1 (1–15)
V3 Sleep study n = 69	61.4% n = 43	4.11 ± 2.3 (1–12)	9.27 ± 3.6 (4–17)	4.11 ± 2.3 (1–15)

SDB = sleep-disordered breathing.

Table 2—General anthropometric and surgical factors tested in the symptomatic sample.

Studied Factors	Frequency (V2 n = 100)	REI (V3 n = 69)	*P* Value and Test
Sex			
Men	63% (n = 63)	2.12 ± 2.55 (60.9%, n = 42)	**$P = 0.042$ Mann-Whitney *U***
Women	37% (n = 37)	1.19 ± 1.65 (39.1%, n = 27)	
Ethnicity*			**Mann-Whitney *U* tests:** /other non-East African $P = .638$ /other non-North African $P = .064$ /other non-European $P = .229$ **Kruskal-Wallis test:** $P = .187$
East African/Haiti	36% (n = 36)	2.29 ± 2.67 (30.5%, n = 21)	
North African	13% (n = 13)	0.84 ± 2.40 (17.4%, n = 12)	
European	53% (n = 53)	1.80 ± 2.24 (55.1% n = 38)	
Other	5% (n = 5)	-	
Type of surgery			
Sole adenoidectomy	21% (n = 21)	1.89 ± 1.89 (20.3% n = 14)	/ A&T $P = .160$ / sole adenoidectomy $P = .02$/A&T $P = .07$ (Kruskal-Wallis test)
Tonsillectomy (previous adenoidectomy)	13% (n = 13)	1.11 ± 0.70 (10.1% n = 7)	
Adenotonsillectomy	66% (n = 66)	1.91 ± 2.45 (69.6% n = 48)	
Age at surgery			
< 3 years of age	26% (n = 26)	2.93 ± 3.18 (27.6% n = 19)	**$P = .007$ *t* test** (bilat)
vs ≥ 3 years	74% (n = 74)	1.31 ± 1.64 (72.4% n = 50)	
> 7 years of age	8% (n = 8)	1.25 ± 1.82 (11.6% n = 8)	$P = .504$ *t* test (bilat)
vs ≤ 7 years of age	92% (n = 92)	1.83 ± 2.33 (88.4% n = 61)	
BMI z-score (long term after surgery)			
< 2 SD	74% (n = 74)	1.82 ± 2.65 (65.2% n = 45)	$P = .837$ *t* test (bilat)
≥ 2 SD	26% (n = 26)	1.70 ± 1.42 (34.8% n = 24)	
WtHR (follow-up)			
≥ 0.5 (weight dependent risk)	48% (n = 48)	1.79 ± 1.67 (50.7% n = 35)	$P = .898$ *t* test (bilat)
< 0.5 (low risk)	52% (n = 52)	1.72 ± 2.78 (49.3% n = 34)	
HSCS			
≥ 2.72	30% (n = 30)	1.85 ± 1.90 (34.8% n = 24)	$P = .802$ *t* test (bilat)
< 2.72	70% (n = 70)	1.71 ± 2.46 (65.2% n = 45)	
Sleep Clinical Score			
≥ 6.5	56% (n = 56)	2.72 ± 2.67 (55.1% n = 38)	**$P < .001$ *t* test (bilat)**
< 6.5	44% (n = 44)	0.58 ± 0.55 (44.9% n = 31)	

P values appear in bold when < .05. *Total exceeds 100 as some children had mixed origins. A&T = adenotonsillectomy, HSCS = hierarchic severity clinical scale, REI = respiratory event index, WtHR = waist-to-height ratio.

of white European origin, 36% from black Africa or Haiti, 13% from North Africa, 5% from other regions. Ethnicity did not influence respiratory events ($P = .187$ Kruskal-Wallis). Still, children of black African origin tended to be operated at younger age compared with non-African children (3.50 ± 1.79 vs 4.45 ± 2.43 years, $P = 0.051$). Mean BMI z-score was

1.23 ± 1.51 (range: −1.71 to 5.94), whereas mean waist-to-height ratio was 0.53 ± 0.22 (range: 0.38 to 1.71).

Sleep

A subgroup of 69 participants with reported SDB symptoms completed a level 3 HSAT. Results showed 32 participants (46.37%) with persistent OSA (REI > 1), with a mean REI of 3.28 ± 2.60 events/h (range 1.1–12.8 events/h), mean OREI of 2.22 ± 2.16 events/h, and mean ODI of 3.87 ± 2.36 events/h. Their mean age was 9.06 ± 3.79 years, and they were evaluated 5.66 ± 3.84 years after surgery. Male sex was significantly associated with higher REI and ODI (P = .04 and P = .03, respectively).

Five patients (7%) had moderate OSA (REI > 5), and only 1 had severe OSA (REI > 10). They were not under any medication and had a variety of phenotypes and ages. These patients with moderate to severe OSA were effectively screened with SCS (all had an SCS score > 6.5) but only 2 of 5 had a positive HSCS score (> 2.72).

In the full sample (N = 100), 66% had had an A&T, 21% a sole adenoidectomy, and 7% had had a tonsillectomy as a second surgery (previous adenoidectomy in early childhood). In the HSAT subgroup, revision surgery had significantly reduced REI when compared to A&T and sole adenoidectomy (Kruskal-Wallis test with Bonferroni correction, P = .07 and .02, respectively), but REI was not significantly different in the long term when comparing adenoidectomy with A&T. Children who had been operated before 3 years of age (n = 19 of 69) had significantly more abnormal respiratory events than patients operated at a later age (REI of 2.93 ± 3.18 vs 1.31 ± 1.64 years); 8 children were operated after 7 years of age and they did not have more severe REI than the rest of the sample.

Obesity affected 24–26% (26 of 100 in V2, 17 of 69 in V3): 15 were obese (BMI z-score comprised between 2 and 3 standard deviations; mean REI = 1.77 ± 1.55), 11 were severely obese (BMI z-score > 3 standard deviations, mean REI = 1.59 ± 1.25 for 9 patients who completed the HSAT).

Obese participants did not have more severe REI than lean patients in this sample when comparing both BMI z-score (1.82 ± 2.65 for < 2 standard deviations vs 1.70 ± 1.42 for ≥ 2 standard deviations) and waist circumference to height ratios (1.72 ± 2.78 for waist-to-height ratio < 0.5 vs 1.79 ± 1.67 for waist-to-height ≥ 0.5).

Craniofacial, oronasal anatomical and functional factors

Children from the full sample showed long/dolichofacial faces in 45%; 39% had a labial incompetence with lip strain (**Table 3**). Almost 3 of 4 children had a convex profile (74%), 56% had an increased facial height, and 53% a malocclusion: posterior crossbite 13%, anterior crossbite 10%, Angle's class II 31%, class III 5%, increased overjet 52%, dental crowding 61%; 8% had had an orthodontic treatment. None of these craniofacial features was associated with differences in REI. A narrow palate (42% of subjects) tended to be associated with higher REI (P = .05; **Table 4**).

With children of 8 years of age or less (n = 34) from the HSAT subgroup, the OREI was significantly higher when presenting with at least 1 dental malocclusion (2.19 ± 2.65 events/h, n = 19) compared with normal occlusion (0.63 ± 1.04 events/h, N = 15) but not for a specific malocclusion type.

SDB phenotypes according to SCS scoring (including patients with adenoid faces and obesity) were significantly associated with REI (P < .05). Subjects with convex profiles had been operated on significantly younger (3.63 ± 1.84 vs 4.92 ± 2.67 years, P = .02).

Oronasal abnormalities were overrepresented: cartilage hypotonia was found in 18 children (all white), with spontaneous nasal collapse on forced inspiration in 13 children (unilateral 4, bilateral 9, see **Figure 2A**); septal deviation was found in 5 patients. Nasal obstruction (32%) and oral breathing mode (62%) were both significantly associated with a higher REI. Gudin's and Rosenthal tests were failed in 55% and 44% respectively, a failed Gudin's test being significantly associated with higher OREI (P = .011, Mann-Whitney U test, see **Figure 3A**).

A short lingual frenulum was found in 40% of patients (**Figure 2B**), relative macroglossia in 38%, and tongue motor immaturity in 24% (patients were unable or did not understand how to elevate the tip of the tongue against the palate when asked). This tongue inability/motor imprecision was significantly associated with increased REI, OREI and ODI (Mann-Whitney U test P < .001, P < .01, and P = .08 respectively, **Figure 3B**).

DISCUSSION

The American Academy of Pediatrics suggests that children with SDB "should be reevaluated postoperatively to determine whether further treatment is required…. and objective testing should be performed in patients who are high risk or have persistent symptoms/signs of OSA after therapy."[2] With the limitation of the HSCS questionnaire survey, adenoidectomy and A&T seemed to be effective in the resolution of SDB symptoms in the long term in about 60% of participants of this study. Mild SDB persistence was reported in 25%, and OSA was suspected in about 7% of nonsyndromic children. When objectively analyzed with a HSAT in a subset of patients, mild to moderate OSA was found in half of examined symptomatic patients: 32 of 69 children had an REI > 1 (46.37%), with a mean REI of 3.28 ± 2.60 events/h (1.1–12.8).

A younger age at surgery (under 3 years) was associated with a higher REI in the long term, confirming the findings from Thadikonda et al[24] on adenoidectomy alone and Mitchell et al[25] on A&T. On the other hand, surgery performed after 7 years of age did not affect REI, contrary to the results of Bhattacharjee et al.[10] They had performed a large retrospective study of 578 children who underwent A&T and reported that, when assessed with PSG with a strict criterion of apnea-hypopnea index < 1 event/h, children who were operated on over the age of 7 years were more likely to have persistent OSA.

This different outcome might be related to both timing and sleep study method. We chose an HSAT that is less accurate in the pediatric population than for adolescents and

Table 3—Craniofacial factors tested in the symptomatic sample.

Craniofacial Factors	Frequency (V2 n = 100)	REI (V3 n = 69)	*P* Value and Test
Facial type (Vertical)			
Mesofacial (normal)	38% (n = 38)	2.03 ± 2.08 (31.9%, n = 22)	*P* = .137 Kruskal-Wallis test
Brachyfacial (short face)	17% (n = 17)	1.94 ± 1.60 (20.3%, n = 14)	
Dolichofacial (long face)	45% (n = 45)	1.50 ± 2.63 (47.8%, n = 33)	
Increased facial height	56% (n = 56)	1.93 ± 2.76 (58.0%, n = 40)	*P* = .957 Mann-Whitney *U*
vs Normal/Reduced height	44% (n = 44)	2.40 ± 1.79 (42.0%, n = 29)	
Labial incompetence	39% (n = 39)	1.82 ± 3.02 (37.7%, n = 26)	*P* = .336 Mann-Whitney *U*
vs labial competence	61% (n = 61)	1.71 ± 1.70 (62.3%, n = 43)	
Phenotype			
Positive (adenoid or obese)	62% (n = 62)	2.10 ± 2.48 (68.1%, n = 47)	***P* = .023 Mann-Whitney *U***
Negative	38% (n = 38)	1.03 ± 1.54 (31.9%, n = 22)	
Convexity			
Straight profile	20% (n = 20)	2.27 ± 2.26 (17.4%, n = 12)	*P* = .129 Kruskal-Wallis test
Convex profile	74% (n = 74)	1.74 ± 2.33 (76.8%, n = 53)	
Concave profile	6% (n = 6)	0.42 ± 0.15 (5.8%, n = 4)	
Maxilla position			
Normal	65% (n = 65)	1.73 ± 2.30 (66.7%, n = 46)	*P* = .154 Kruskal-Wallis test
Retruded	14% (n = 14)	0.80 ± 0.59 (17.4%, n = 12)	
Protruded	21% (n = 21)	2.90 ± 2.88 (15.9%, n = 11)	
Narrow palate	42% (n = 42)	2.24 ± 2.67 (40.6%, n = 28)	***P* = .054 Mann-Whitney *U***
vs Normal-shape palate	58% (n = 58)	1.43 ± 1.91 (59.4%, n = 41)	
Mandible position			
Normal	25% (n = 25)	2.96 ± 2.74 (26.1%, n = 18)	*P* = .080 Kruskal-Wallis test
Retruded	67% (n = 67)	1.36 ± 1.99 (69.6%, n = 48)	
Protruded	8% (n = 8)	0.97 ± 0.91 (4.3%, n = 3)	
Dentition			
Primary	18% (n = 18)	2.85 ± 1.53 (15.9%, n = 11)	*P* = .281 Kruskal-Wallis test
Mixed	60% (n = 60)	1.53 ± 2.18 (56.5%, n = 39)	
Permanent	22% (n = 22)	1.60 ± 2.00 (27.5%, n = 19)	
Malocclusion (any)	53% (n = 53)	1.94 ± 2.6 (52.2%, n = 36)	*P* = .750 Mann-Whitney *U*
vs Normal occlusion	47% (n = 47)	1.56 ± 1.9 (47.8%, n = 33)	
Posterior crossbite*	13% (n = 13)	1.07 ± 0.68 (8.7%, n = 6)	*P* = .183 Mann-Whitney *U*
Anterior crossbite*	10% (n = 10)	0.79 ± 0.63 (13.0%, n = 9)	*P* = .909 Mann-Whitney *U*
Open bite*	8% (n = 8)	0.98 ± 1.18 (7.2%, n = 5)	*P* = .258 Mann-Whitney *U*
Deep bite* (> 35%)	45% (n = 45)	1.68 ± 1.88 (46.4%, n = 32)	*P* = .926 Mann-Whitney *U*
Increased overjet*(> 3 mm)	52% (n = 52)	1.82 ± 2.62 (50.7%, n = 35)	*P* = .987 Mann-Whitney *U*
Normal or reduced overjet	48% (n = 48)	1.73 ± 1.85 (47.8%, n = 33)	
Angle's classification*			
Class I (normal)	64% (n = 64)	1.80 ± 2.79 (65.2%, n = 45)	Kruskal-Wallis test: *P* = .894
Class II	31% (n = 31)	1.74 ± 2.08 (29.0%, n = 20)	
Class III	5% (n = 5)	1.32 ± 1.85 (5.8%, n = 4)	
Dental crowding	61% (n = 61)	1.68 ± 2.27 (68.1%, n = 47)	*P* = .624 Mann-Whitney *U*
vs no crowding/spacing	39% (n = 39)	1.93 ± 2.31 (31.9%, n = 22)	

P values appear in bold when < .05. *Total exceeds 100% as some children have combined malocclusions. REI = respiratory events index.

Table 4—Nasal, oral, and oropharyngeal factors tested in the symptomatic sample (underlined items are from SCS scoring)

Nasal & Oropharyngeal factors (Sleep Clinical Record)	Frequency (n = 100)	REI (n = 69)	*P* Value, Test
Oral breathing	62% (n = 62)	2.16 ± 2.6 (76.8%, n = 53)	**P = .017 Mann-Whitney *U***
Nasal breathing	38% (n = 38)	1.17 ± 1.50 (23.2% n = 16)	
Nasal cartilage hypotonia	18% (n = 18)	1.35 ± 1.25 (20.3%, n = 14)	*P* = .939 Mann-Whitney *U*
vs no collapsus on inspiration	82% (n = 82)	1.87 ± 2.48 (79.7%, n = 55)	
Deviated nasal septum	5% (n = 5)	0.60 ± 0.20 (4.3%, n = 3)	*P* = .387 Mann-Whitney *U*
Nasal obstruction	32% (n = 32)	2.51 ± 2.42 (30.4%, n = 21)	**P = .017 Mann-Whitney *U***
No obstruction	68% (n = 68)	1.43 ± 2.14 (69.6%, n = 48)	
Palatal tonsils			
Score III or IV	9% (n = 9)	1.13 ± 0.63 (10.1%, n = 7)	*P* = .424 Mann-Whitney *U*
Score 0 (removed), I, II	91% (n = 91)	1.83 ± 2.38 (89.1%, n = 62)	
Palate position			
Score III or IV	58% (n = 58)	1.51 ± 2.4 (55.1%, n = 38)	*P* = .059 Mann-Whitney *U*
Score I or II	42% (n = 42)	2.06 ± 2.1 (44.9%, n = 31)	
Tongue			
Short frenulum	41% (n = 41)	2.25 ± 2.78 (40.6%, n = 28)	*P* = .281 Mann-Whitney *U*
vs normal tongue motility	59% (n = 59)	1.42 ± 1.80 (59.4%, n = 41)	
Relative macroglossia	38% (n = 38)	2.14 ± 2.66 (29.0%, n = 24)	*P* = .126 Mann-Whitney *U*
vs normal tongue volume	62% (n = 62)	1.56 ± 2.03 (65.2%, n = 45)	
Motor immaturity (tasks)	26% (n = 26)	3.63 ± 3.63 (23.2%, n = 16)	**P < .001 Mann-Whitney *U***
vs ability to elevate tongue	74% (n = 74)	1.19 ± 1.19 (76.8%, n = 53)	
Atypical swallowing	54% (n = 54)	2.08 ± 2.60 (52.2% n = 36)	*P* = .327 Mann-Whitney *U*
vs mature swallowing	46% (n = 46)	1.52 ± 1.97 (47.8% n = 33)	
Other factors			
Parafunctionals habits (> 1)	51% (n = 51)	1.58 ± 1.95(50.7%, n = 35)	*P* = .656 Mann-Whitney *U*
vs none	49% (n = 49)	1.94 ± 2.57 (49.3%, n = 34)	
Reported sleep bruxism	27% (n = 27)	1.94 ± 1.95 (29.0%, n = 20)	*P* = .643 Mann-Whitney *U*
No reported sleep bruxism	73% (n = 73)	1.69 ± 1.95 (71.0%, n = 49)	
Nasal corticoid treatment	9% (n = 9)	1.32 ± 1.36 (13.0%, n = 9)	*P* = .986 Mann-Whitney *U*
No medical treatment	91% (n = 91)	1.82 ± 2.38 (87.0%, n = 60)	

P values appear in bold when < .05. REI = respiratory event index.

adults.[26] We also performed the recording a long time after surgery and not in the 6 months to 1 year following surgery; as the maximum peak of lymphoid tissue growth is about age 8 years, residual disease, originating from residual lymphoid tissue, is then more likely to be seen when the study is performed at age 7 or 8 years.

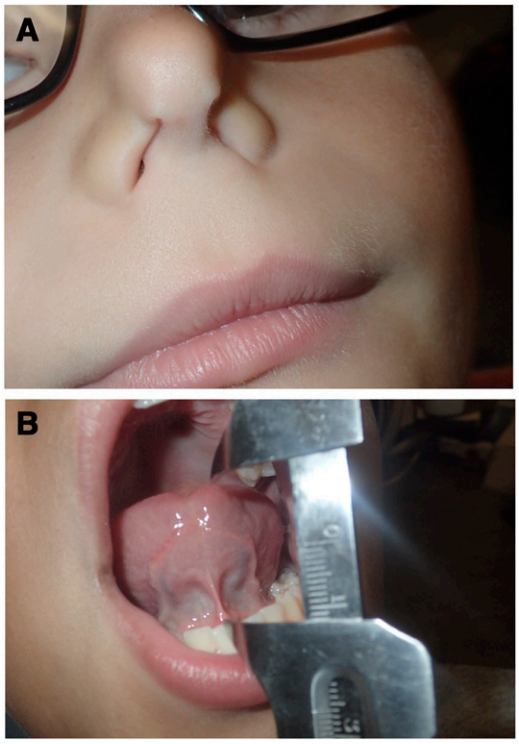
Various craniofacial abnormalities were described in preschool children as predisposing factors for SDB, including decreased mandibular and maxillary lengths, skeletal retrusion, and increased lower facial height.[27]

In this study, we could not establish any direct relationship between abnormal respiratory events during sleep and specific malocclusions, such as crossbite, deep bite, or class II occlusion, whose prevalence in this sample was not significantly different from that in the general population.

This finding was a surprise, as a strong association is depicted in the literature. In the PANIC study,[28] conducted on 491 Finnish children 6–8 years of age, children with crossbite had a 3.3 times higher risk of having SDB (based on a questionnaire) than those without crossbite, and children with a convex facial profile had a 2.6 times higher risk of having SDB than those with a normal facial profile. A recent Italian study,[29] comparing a group of 139 children with OSA to a control group of 137 children (range 2–10 years, all diagnosed with PSG), described several orthodontic factors independently associated with OSA: posterior crossbite (odds ratio = 3.38), reduced overbite (odds ratio = 2.43), increased overbite (odds ratio = 2.19), and increased overjet (odds ratio = 4.25).

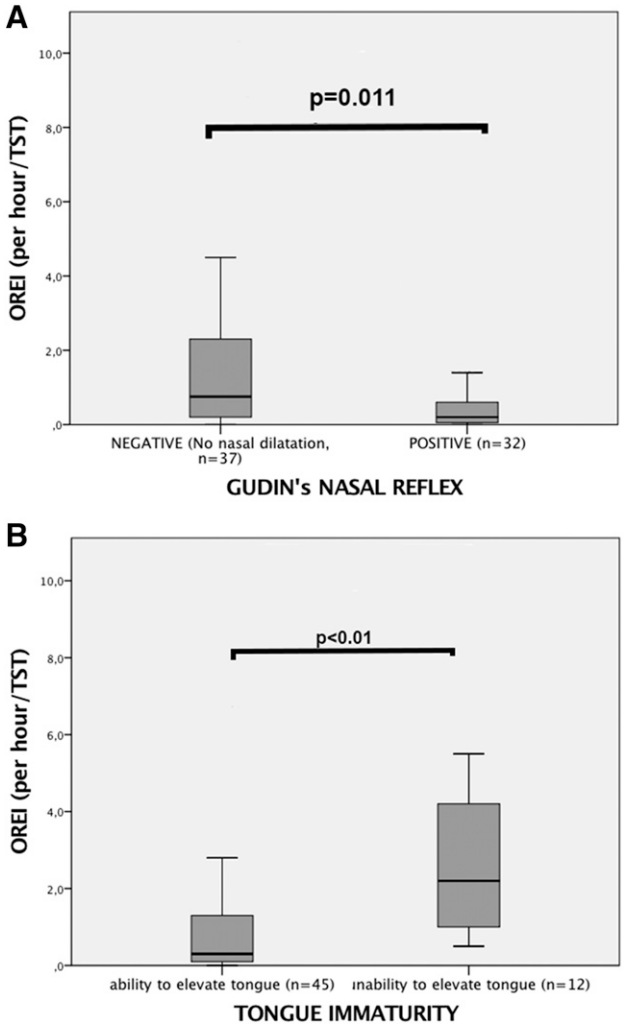
The difference in our trial might be that we studied a different age group. Only when performing a subgroup analysis of children under the age of 8 years were sleep parameters influenced by malocclusion (Mann-Whitney *U* test *P* = .01 for OREI and ODI, respectively).

Figure 2—Photographs depicting cartilage hypotonia and short lingual frenulum.



(A) Cartilage hypotonia (spontaneous unilateral or bilateral collapse during forced inspiration) was overrepresented in this symptomatic population, but was not related to postoperative sleep parameters nor hierarchic severity clinical scale. **(B)** Short lingual frenulum (limited tongue elevation in relation to maximal aperture of 60% or less).

Figure 3—Obstructive respiratory events index, Gudin's nasal reflex, and tongue immaturity.



(A) Obstructive respiratory events index (OREI) per total sleep time (TST) according Gudin's test (median). **(B)** OREI index per TST according tongue elevation test (median).

Interestingly, we also noted that patients with convex profiles were operated at a significant younger age than those with straight and concave profiles. These anatomical factors might have influenced the surgical decision, as described in the study by Sato et al[30] in a group of Japanese preschool children (mean age 5 years). Surgeons might be more prone to operate on patients with adenoid facies or retrognathia. Whether this facial convexity might have affected the severity of SDB before surgery in our sample remains hypothetical, but the relative importance of skeletal abnormalities in SDB pathogenesis could be reduced with continued growth.

The growth rate of the jaws describes a plateau in childhood before an acceleration at puberty, the mandible having a more extended and pronounced period than the maxilla,[31] so that profile convexity tends to reduce with normal growth. Furthermore, A&T has been shown to normalize maxillomandibular growth compared with baseline,[32] in a group of 17 children (mean age 5.6 years). The authors found no statistically significant differences in the maxillomandibular values at a 5-year follow-up between the patients with OSA and control patients. These results were reproduced in several studies,[33,34,35] where improvements were found in the face and dental arches, 1 year after surgery.[36]

Our results are in accordance with findings of a recent meta-analysis[37] that could not support a direct causal relationship between craniofacial structure and pediatric SDB.

Compilation of worldwide data and large age groups might hide phenotype-specific patterns, with potentially different natural history. These may be elucidated only by complex cluster analysis[38] or longitudinal samples of relatively homogeneous populations.

The airway patency is linked to its anatomical structure and to the muscular tone/activity of its dilator muscles, which counteract the inspiratory depression forces. Oral breathing, "nasal disuse,"[39] and abnormal oral reflexes have been implicated both in the pathogenesis of SDB and malocclusions: oronasal function grading strategies have been widely described in the field of orthodontics and sleep medicine.[40]

Several nasal abnormalities, such as cartilage hypotonia (18%), deviated nasal septa (5%), and nasal obstruction

(32%) were found in this study but do not explain why the majority of these patients with SDB were mouth breathers (62%). Other oral abnormalities, such as a short lingual frenulum (40%) or narrow palate (42%), could favor a low tongue posture and be a factor in the maintenance of an oral breathing mode. This is in accordance with the study of Guilleminault et al,[41] who described a phenotype of patients with narrow high arch palate with a short mandibular lingual frenulum between the ages of 2 and 6 years old, who all had SDB when it was left untreated.

It must be recognized that surgical, orthodontic, and medical treatments of SDB generate structural modification of the airways, but do not necessarily lead to any neuromuscular modification or switching of the child's ventilatory mode.

Myofunctional exercises have been shown to reduce residual OREI in young children after A&T compared with a control group.[42] The critical role of this re-education has already been described,[43] following patients in adolescence after A&T and palatal expansion. When oral rehabilitation was not performed despite an indication, recurrence of SDB symptoms were observed at 1 year, whereas in the same interval of time, rehabilitated children had normalized sleep parameters.[43]

In our sample, residual pathological OREI was significantly associated with a failed Gudin's test. The alar muscles, which control the nostril caliber, have active contraction only during deep inspiration and sniffing; their reflex contraction, during inspiration, makes it possible to oppose inspiratory collapse, and an absence of active contraction could testify of uncommon use of the nasal passage. Then, when asked to elevate the tip of the tongue against the palate or outside of the mouth toward the nose, 26% of patients were unable to spontaneously do so. This motor immaturity was also associated to higher REI, OREI, and ODI.

These simple clinical tests could help identify children requiring re-education in the postoperative follow-up period.

Other risk factors have been involved in pediatric SDB, including male sex and race/ethnicity,[44] but also systemic inflammation, allergy/atopy, asthma, visceral obesity, and prematurity.

Male predominance and male younger age at surgery in pediatric SDB could be explained by a less mature craniofacial skeleton, together with a degree of sexual dimorphism or android pattern of fat distribution of the older adolescent. OSA becomes more prominent in males after puberty.[45]

Anatomical variations with ethnicity have been suggested for black African children, who could present soft tissue differences. These variations could also be present in Asian populations, where some authors could not find any cephalometric characteristic associated with OSA severity in children,[46] except for a lowered hyoid position,[47] which could be related to a low tongue position.

Some of the weaknesses of our study relate to surgical data, which were retrospectively collected and which would not provide precise assessment of SDB severity or obesity status prior to surgery.

The American Academy of Sleep Medicine[48] and the American Academy of Otolaryngology Head and Neck Surgery Foundation (AAO-HNSF)[49] clearly stated that an attended full night PSG is the gold standard for diagnosis of pediatric SDB. Unfortunately, PSG is a complex test, expensive, and not readily available. Given the rarity of qualified sleep laboratories to offer pediatric studies, priority is given to severe or syndromic cases, patients with obesity, or patients under 2 years of age,[50] and there can be an extended waiting period between referral and when sleep studies are done. In the examined charts, baseline SDB diagnosis prior to surgery was mostly made through clinical picture (witnessed apnea reported by parents) or oximetry. No PSG was available at baseline to confirm the severity of SDB and to assess whether persistent symptoms were the continuation of a previously severe disease, a recurrence of symptoms, or even a new form of SDB that could be related to weight gain or allometric changes in craniofacial growth.

Katz et al[51] found that the BMI score increased more in children who underwent A&T compared with those in the watchful waiting group, but we could not trace the weight gain of each of the subjects studied. Almost 1 of 4 children in our symptomatic sample and about 40% of the moderate suspected OSA cases (HSAT sample) were overweight or obese, even if no direct relationship could be drawn between BMI z-score and sleep parameters. In obese children, fatty infiltration of the pharyngeal walls and muscles, particularly tongue base and lateral pharyngeal fat pads, gradually reduces the respiratory tract, which mechanically explains the obstruction.

Going further, BMI z-score measure surplus weight relative to height and not body fat, whereas the distribution of adipose tissue seems to be more important than its total mass for the risk of developing obesity-associated diseases. We added a qualitative assessment of body fat distribution, the waist to height ratio, as a marker of central obesity, but neither did we show a significative difference in the REI. Several other methods could have been used, including skinfolds, bioelectrical impedance analysis, air displacement plethysmography, or hydrodensitometry and dual energy x-ray absorptiometry, but these tools were not available.

Still, obesity is not systematically associated with OSA; Yuan et al[52] showed that, despite a narrowed upper airway from adipose tissue, certain adolescents with obesity were protected from developing OSA by upper airway neuromotor activation. More studies are required to assess tongue motor immaturity in different obesity levels at a range of age groups. However, as the association between BMI and the severity of the obstructive apnea-hypopnea index is low before puberty, but becomes significant at the time of adolescence, dietary management and follow-up of these children is recommended.[53]

In the light of our results, it also appears that the HSCS score was not a reliable tool to separate chronic snoring from OSA, but this outcome was not anticipated. We chose this simple and validated pediatric questionnaire, which was developed in both English and French as a first-line tool for pediatric sleep apnea, because to our knowledge, no validated questionnaires existed to screen specifically for persistent SDB following surgery. We can explain this unexpected result by our different

population (a postsurgical sample) and its wide age group, in particular. HSCS was designed on children aged 5 to 9 years and our sample was 4 to 17 years. On the other hand, the sleep clinical record, combining various clinical parameters, seemed to be accurate in screening persistent OSA from snoring, with a cut-off score ≥ 6.5. This is in accordance with the original publications of Villa et al in older age groups and postsurgical populations.[18,19] We had the limitation of a parental report of Attention Deficit Hyperactivity Disorder instead of a clinician diagnosis for most subjects, which could have influenced our results.

Among the other limitations of this study, we could not perform a PSG and the HSAT could not be done in all examined patients (V2). Probably because of these relatively mild symptoms and lack of clinical complaint, children's and parents' motivation was quite low. Many of them refused the sleep study or refused to perform another test after failing a first test. There was a reduced sample size from V2 to V3, with an increase in the populations who were overweight and oral breathers performing the HSAT.

Last, examinations were performed by an orthodontist and not an ear, nose, throat surgeon; no nasofibroscopy was performed to assess directly the location of the obstacle and search for adenoid regrowth, lingual tonsil hypertrophy, turbinates, or nasal mucosa swelling and no allergy diagnosis or tests could be performed. More deviated septa located in a more posterior aspect of the nasal cavities could have been found. Similarly, as enlargement of the lingual tonsils has been described as relatively common in children with persistent OSA after A&T,[54] it could have played some part in the observed macroglossias, which could have been overestimated.

Other factors, such as prematurity, could play a role, as this sample was extracted from a tertiary hospital with a significant number of premature births. A longitudinal study demonstrated that a large number of infants born prematurely had generalized hypotonia,[55] a tendency to exhibit mouth breathing during sleep and a narrow palatal vault.

CONCLUSIONS

Residual SDB was present in a small proportion of children after A&T in the long term: patients in our persistent SDB sample had mild to moderate OSA and presented various phenotypes with a high prevalence of morphological abnormalities and dysfunctions and 24% were obese. The SCS (> 6.5) seemed to individualize patients adequately with persistent OSA from chronic snoring, but should not be substituted for PSG, the gold standard method for pediatric OSA.

Oral breathing, impaired nasal dilator reflex (failed Gudin's test), and tongue motor immaturity were associated with increased OREI in this population, underlying the neuromuscular component of persistent SDB.

Craniofacial risk factors might have had a significant impact in the severity of SDB at a younger age, influencing the age of surgery, but were not associated with sleep parameters in the long term after surgery.

ABBREVIATIONS

A&T, adenotonsillectomy
BMI, body mass index
HSAT, home sleep apnea test
HSCS, hierarchic severity clinical scale
ODI, oxygen desaturation index
OREI, obstructive respiratory event index
OSA, obstructive sleep apnea
PSG, polysomnography
REI, respiratory event index
SDB, sleep-disordered breathing
SCS, sleep clinical score

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DISCLOSURE STATEMENT

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