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Critical role of myofascial reeducation in pediatric sleep-disordered breathing

C. Guilleminault [a,*], Y.S. Huang [b], P.J. Monteyrol [d], R. Sato [a], S. Quo [e], C.H. Lin [c]

[a] Stanford University, Sleep Medicine Division, United States
[b] Pediatric Sleep Medicine, Taiwan
[c] Cranio-Facial Center, Chang-Gung University and Memorial, Taiwan
[d] Clinique Oto-Laryngologique, Bordeaux, France
[e] Orthodontic Department, University of California, San Francisco Dental School, United States

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ABSTRACT

Background: Limited studies suggest that pubertal development may lead to a recurrence of sleep-disordered breathing (SDB) despite previous curative surgery. Our study evaluates the impact of myofunctional reeducation in children with SDB referred for adenotonsillectomy, orthodontia, and myofunctional treatment in three different geographic areas.

Methods: A retrospective investigation of children with polysomnographic analysis following adenotonsillectomy were referred for orthodontic treatment and were considered for myofunctional therapy. Clinical information was obtained during pediatric and orthodontic follow-up. Polysomnography (PSG) at the time of diagnosis, following adenotonsillectomy, and at long-term follow-up, were compared. The PSG obtained at long-term follow-up was scored by a single-blinded investigator.

Results: Complete charts providing the necessary medical information for long-term follow-up were limited. A subgroup of 24 subjects (14 boys) with normal PSG following adenotonsillectomy and orthodontia were referred for myofunctional therapy, with only 11 subjects receiving treatment. Follow-up evaluation was performed between the 22nd and 50th month after termination of myofunctional reeducation or orthodontic treatment if reeducation was not received. Thirteen out of 24 subjects who did not receive myofunctional reeducation developed recurrence of symptoms with a mean apnea–hypopnea index (AHI) = 5.3 ± 1.5 and mean minimum oxygen saturation = 91 ± 1.8%. All 11 subjects who completed myofunctional reeducation for 24 months revealed healthy results.

Conclusion: Despite experimental and orthodontic data supporting the connection between orofacial muscle activity and oropharyngeal development as well as the demonstration of abnormal muscle contraction of upper airway muscles during sleep in patients with SDB, myofunctional therapy rarely is considered in the treatment of pediatric SDB. Absence of myofascial treatment is associated with a recurrence of SDB.

1. Introduction

Obstructive sleep apnea (OSA) has become increasingly recognized as a notable health concern in children given its consequences on behavior, function, and quality of life. The importance of early recognition and treatment in children is paramount to maximizing resolution of symptoms and potential avoidance of OSA syndrome during adulthood. Adenotonsillectomy and palatal expansion have established their roles in the treatment of OSA after demonstrating considerable improvement related to adenoid or tonsillar hypertrophy, maxillary or mandibular deficiency, and orthodontic or craniofacial abnormalities. However, the implementation of other treatment modalities such as myofascial reeducation also may play a role in the optimization of sleep-disordered breathing (SDB).

Functional myofascial reeducation in children has been well-established in the treatment of abnormal orofacial development for more than 40 years [1]. However, few studies have been published supporting the benefits of orofacial reeducation compared to the numerous studies reinforcing the utility of surgical and orthodontic treatments in SDB [2]. Although the role of orofacial education remains largely variable between institutions, the most notable results have been described when myofunctional therapists and orthodontists worked in collaboration to manage orofacial weakness. Although promising, the efficacy of myofunctional therapy in combination with surgical and orthodontic treatment is unclear. The purpose of our study was to evaluate the impact of myofunctional reeducation protocols on orofacial muscle weakness and the treatment of SDB in children following surgical and orthodontic optimization.

* Corresponding author. Tel.: +1 650 723 6601; fax: +1 650 725 8910.
E-mail address: cguil@stanford.edu (C. Guilleminault).

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2. Methods

Our retrospective analysis involving prepubertal children diagnosed with OSA, who were referred for orthodontic treatment after presenting with residual symptoms of abnormal breathing following adenotonsillectomy, could only draw a small number of subjects.

Data collection was performed in three different regions of the world, including the San Francisco Bay area, Taiwan, and France. Our analysis involved three different pediatric sleep centers working with otolaryngologists, orthodontists, and functional therapists. The three sleep centers performed all sleep monitoring and were referral centers for large geographic areas. The participating sleep clinics and the orthodontic practices had a collaborative working relationship spanning from 6 to 14 years.

Retrieval of health information for children was variable but targeted those initially seen between the ages of 3 and 6 years preceding confirmation of SBD by nocturnal polysomnography (PSG). If a child was confirmed to have OSA by PSG, the second step was to determine the presence of adequate follow-up and appropriate documentation including subsequent PSGs and documentation from other specialists. Most charts did not fulfill these criteria and were excluded from our study. Charts that had systematic PSG at different phases of follow-up were those of children seen by orthodontists either postadenotonsillectomy or without otolaryngologic intervention. Children often were referred to both a functional reeducation specialist and to an orthodontist in an effort to perform the investigation where myofunctional therapy was practiced. Children were followed in sleep medicine and orthodontic clinics with variable schedules.

Despite being followed in these clinics, postorthodontic treatment PSG records often were unavailable and complete documentation often was absent, excluding a large number of cases. Once the necessary clinical data and PSG reports were confirmed, identifiers were removed and data were extracted (Fig. 1). Anonymous analyses of clinical and polysomnographic data were performed. Retrospective analyses of unidentified PSG and of clinical information were approved by the internal review boards.

All surveyed subjects were prepubertal children between the ages of 3.6 and 6.6 years at the time of their initial visit. Initial assessment of each child included clinical interview, pediatric and sleep clinical evaluation, completion of the pediatric sleep questionnaire (PSQ), a questionnaire validated in different languages [3,4], and nocturnal PSG. Following clinical and PSG evaluation, all children diagnosed with OSA were referred to otolaryngology for surgical evaluation. All subjects except for one had adenotonsillectomy performed and all were followed up after surgery with repeat clinical evaluation and PSG. Subjects with residual OSA detected on postsurgery PSG were sent for orthodontic evaluation [5]. Once the decision regarding orthodontic treatment was made (i.e., rapid maxillary expansion or bimaxillary expansion), recommended myofunctional reeducation also was performed [1].

Subjects were followed at an orthodontic practice during the application of orthodontic treatment and also were followed at their sleep clinics 6 to10 months following initiation of their orthodontic treatment. Concomitant use of myofunctional reeducation was documented as being implemented or as recommendation not followed. Repeat PSG was performed following orthodontic treatment with or without functional reeducation. Data from myofunctional reeducation clinics were used solely to monitor compliance with follow-up appointments and to monitor duration of treatment. Subjects were most often seen during their scheduled orthodontic follow-up. Less frequently they were seen several years after initiation of orthodontic treatment due to planned follow-up visits or due to recurrence of sleep-related symptoms; in this case, they were referred back to sleep clinics. During long-term follow-up visits, the reassessment always involved clinical interviews, PSQ, clinical pediatric evaluation and sleep evaluation, determination of height and weight based on body mass index, sleep medicine examination, myofunctional orofacial status, and nocturnal PSG.

All long-term follow-up PSGs (i.e., last investigation performed) were transferred to new compact discs with recordings formatted in European Data Format. This transfer allowed analysis of all PSGs performed on various sleep programs to be anonymously rescored by a single scorer. PSG rescoring could not be performed on the initial PSGs in the same fashion. However, all centers used the same atlases and guidelines for scoring sleep and breathing variables.

All subjects were evaluated by full-night PSG performed in a sleep laboratory and included the following electrophysiologic parameters, electroencephalogram (EEG) (three channels), electrooculogram (two channels), electrocardiogram, chin electromyogram (EMG), leg EMG (one channel), nasal pressure cannula, oral thermistor, thoracic and abdominal belts, snoring sensor, pulse oximetry, position sensor, and video recording. Variations to the montage included an additional second leg EMG, a fourth EEG, transcutaneous CO_2 or end-tidal CO_2, and the thoracic and abdominal belts were either piezoelectric or inductive plethysmography. All recordings lasted a minimum of 7.5 hours. Individuals were assigned corresponding identification numbers and their data were compiled using the Microsoft Excel program to perform statistical analyses of the results.

Myofunctional reeducation specialists were trained in various countries and were divided into two categories of either speech therapists or specialists in muscle reeducation. Speech therapists were trained in the United States, whereas muscle reeducation specialists were trained outside of the United States. Myofunctional specialists obtained university degrees in functional reeducation with a subspecialty in myofunctional reeducation and practiced validated therapeutic protocols. Treatment protocols are similar in different countries [1]. In the United States, if not sanctioned by a diploma, courses are administered (particularly in California) by trained individuals often trained in other countries. The myofunctional re-educators involved in the three participating sleep centers had similar myofunctional reeducation training, including several years of experience with treatment modalities and use of the same type of report forms. Similar exercise regimens and daily durations of treatment were recommended to parents. Frequency of visits varied not with the sleep center but with the individual and were based on the needs of each case. Visits were more frequent at the initiation of treatment and less frequent as time passed. Daily exercise performance was recorded by parents in a log and reviewed by re-educators at visits. Reeducation programs were completed after 2 years.

3. Reeducation

Myofunctional reeducation involves strengthening of the tongue and orofacial muscles by teaching individuals how to reposition muscles to the appropriate position. The tongue should be kept in a high position during sleep with its dorsal-terminal end in constant contact with the palatine striae located on the anterior aspect of the palate. Reeducation typically is easier in children ages 6 years and older, but it is largely related to the degree of effort parents make in reinforcing a subject to perform his or her exercises. Exercises are initially repeated several times per day with a quick initial increase in frequency during the earliest phase of treatment. This phase requires the subject and one parent to frequently follow-up with a specialist during the first 6 months and less frequently thereafter. The amount of follow-up depends on the duration of therapy needed, but once the subject has gained the desired tongue position along with appropriate strength the

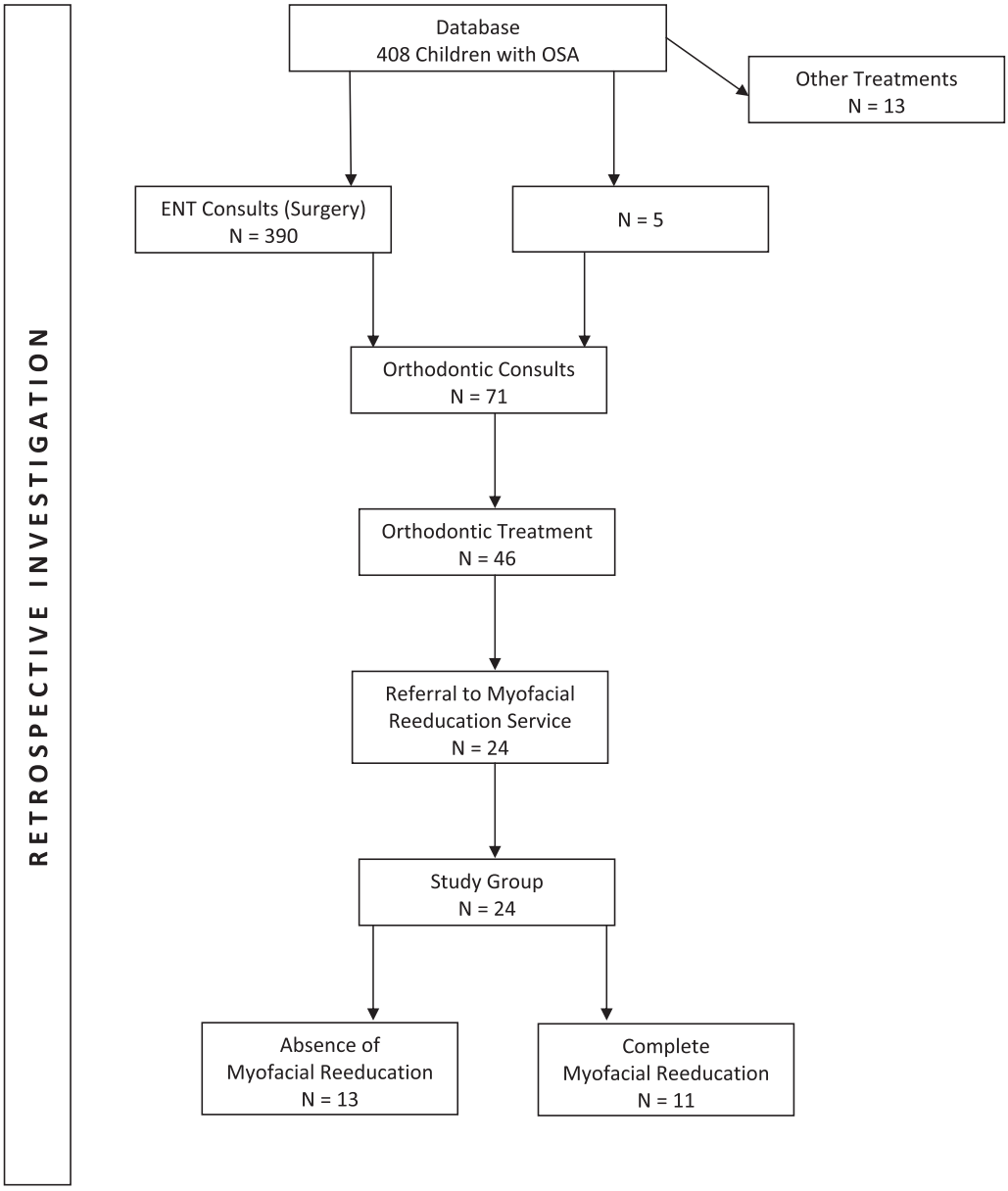

Fig. 1. Graph of initial charts retrieved for study. The graph documents the poor follow-up that occurred for a long time in the past after diagnosis and initial treatment of obstructive sleep apnea (OSA). Many children had no sleep follow-up and absence of sleep recordings postadenotonsillectomy. Seventy one children were seen again posttreatment and had systematic investigation by both a sleep specialist and an orthodontist. Evaluation indicated validity of performing orthodontic treatment in 46 children. Rapid maxillary expansion and bimaxillary treatments were recommended (32 rapid maxillary expansion; 14 bimaxillary treatments). Only 26 children had good documentation of treatment at follow-up. Myofunctional therapy had been recommended in 24 of them in association with orthodontic treatment. However, myofunctional therapy was only performed in 11 children and 13 either did not follow the recommendations or quickly dropped out of the study.

frequency of follow-up can be extended. The subject is then primarily monitored to insure continued appropriate development until the completion of treatment. The investigation took place between the 22nd and 50th month following termination of the myofunctional treatment program, independent of the amount of time spent in the program. If the program was never implemented, subjects were seen for follow-up between the 28th and 34th month following termination of orthodontic treatment.

4. Analysis

The information collected for our study included gender, age at time of each treatment phase and testing, clinical concerns and symptoms, PSQ results, and results of clinical orofacial evaluation. Description of the nasofacial and orofacial examination included Friedman classification tonsil size [6]; modified Mallampati score [7,8]; calculated overjet (mm); evaluation of the hard palate, which was categorized as high and narrow, low lying, or normal; and the presence of enlarged inferior nasal turbinates categorized as occupying less than 50% or 50% or more of the space inside the nostrils. The presence or absence of nasal valve collapse, deviated septum, small mandible, overbite, and awake-mouth breathing also were documented. Absence of anterior short frenulum was affirmed. Head posture was noted in lateral position [9,10] but nuchal Solow angle was not calculated. Clinical information that was recorded during follow-up evaluation but was unavailable at initial presentation included preferential chewing to one side, presence of visualized facial asymmetry, presence of palpable asymmetry of masseteric muscles at maximum clinching, and results of myofunctional evaluation performed by a reeducation specialist. Such information was retrieved from orthodontic and myofunctional reeducation charts. Individual

orthodontists and myofunctional re-educators worked as a team with one orthodontist working with one preferred re-educator.

The long-term follow-up PSG recording was scored blindly by a single investigator, whereas all other PSG results were reported without access to the actual recording. The scoring was based on the manual for sleep scoring by Rechtschaffen and Kale [11], the American Sleep Disorders Association recommendation for EEG arousal scoring [12], and the American Academy of Sleep Medicine criteria for scoring hypopneas and apneas [13]. Hypopneas were defined by a 50% reduction in nasal cannula curve amplitude and an associated drop of 3% or more in oxygen saturation. The usual subdivision of obstructive, mixed, and central events was followed. Events defined as postarousal central apneas were eliminated from the apnea–hypopnea index (AHI) score. There was additional scoring of flow limitation based on the definition of Hosselet et al. [14]. The nasal cannula curve was compared to published patterns involving flattening or truncation of the curve during inspiration [15]. The percentage of flow limitation was determined by the number of 30-second epochs containing the presence of flow limitation [16]. An epoch was scored with flow limitation if it was present for more than 15 seconds (i.e., more than 50% of the scored PSG epoch). The percentage of flow limitation was calculated by dividing the total flow limited sleep time (i.e., number of 30-s epochs scored with flow limitation multiplied by two) by the total sleep time [16]. The score of flow limitation was not available for the initial recordings in many subjects and was only systematically obtained in the reports from the postorthodontic treatment and the rescored recordings.

Presence or absence of mouth breathing was noted in the results of each PSG based on the mouth thermistor tracing but was not quantified. As previously mentioned the long-term follow-up PSGs were scored by a single-blinded scorer. Comparisons of PSG results between the subjects with and without myofunctional treatment were performed using Wilcoxon signed rank tests and χ^2 tests.

5. Results

5.1. Subjects involved in retrospective survey

An initial database of 408 pediatric cases diagnosed with OSA by PSG was established and was evaluated by an otolaryngologist who performed surgery and who subsequently had a postsurgical PSG. As previously mentioned many charts were incomplete when looking for further follow-up and were excluded. From this database, 71 subjects with documented visits to an orthodontist postadenotonsillectomy were retrieved. Children seen by orthodontists for evaluation had better documentation than those seen in other places, reflecting the higher representation of this subgroup in the follow-up survey. Children lacking the syndromic presentation but who had close orthodontic follow-up led to closer evaluation and anatomic findings observed in subjects recognized with SDB. Documented charts revealed that 46 of these subjects were considered for orthodontic treatment and simultaneous myofascial reeducation due to persistent OSA at PSG, even if improvement was noted postadenotonsillectomy. Of these 46 subjects, 24 had retrievable follow-up documentation including myofunctional treatment information (Fig. 1 [graph]). These 24 nonoverweight subjects (14 boys) (17% of initial database) formed the study group of those who satisfied the inclusion parameters being evaluated.

5.2. Evaluation at entry

The results at entry are presented in Tables 1 and 2. All subjects presented with clinical concerns, symptoms, and anatomic findings consistent with OSA, with the PSG confirming the diagnosis. Anatomic investigation at entry showed that out of the 24 cases, 23 had a tonsil-size scale of three or four. Twenty-three subjects had a modified Mallampati scale score of three or four, and one subject had a modified Mallampati scale score of two. Inferior nasal turbinates were scored with occupying nasal space >50% in 13 cases. Nasal septum deviation was found in 14 cases, and all of them had a narrow palatal vault. Fourteen subjects had been referred and treated for nasal allergies with treatment consisting of nasal steroids, allergic desensitization, or both. Of the 401 initial nonoverweight subjects being evaluated, 90% had a tonsil score of three or four, 73% had a modified Mallampati scale score of three or four, and 48% were mentioned to have a high and narrow palatal vault. Statistics revealed from the χ^2 test indicated that the 24 studied subjects were significantly different in Mallampati scale score three and four ($P = .01$) and presence of high and narrow palatal vault ($P = .001$), but the initial anatomic description was similar in 46% of the cases.

5.3. Initial treatment

Twenty-three subjects had adenotonsillectomy (T&A). Additionally, five subjects had radiofrequency ablation of the inferior nasal turbinates performed at the time of T&A. One girl was felt to have small tonsils that would not benefit from surgery but was directly referred for orthodontia, given her high and narrow arched hard palate. None of the subjects had an abnormally placed anterior frenulum.

The data retrieved postadenotonsillectomy and postorthodontia treatments are presented in Tables 1 and 2, including the one girl subject who was sent directly for orthodontia treatment. Although symptoms were reportedly improved in all 23 cases following surgery, clinical concerns and symptoms were not completely eliminated, and the persistence of abnormal breathing was confirmed with PSG analysis. The presence of mouth breathing was noted in all postsurgical cases but was not quantified. All 24 subjects were sent to orthodontists and in all cases were expected to benefit from orthodontic treatment.

Following orthodontic evaluation rapid maxillary or bimaxillary expansion was performed, and orthodontic equipment was kept in place for 8 to12 months. Follow-up evaluation in the sleep clinic with follow-up PSG was performed near the time of orthodontic equipment removal (Tables 1 and 2). Clinical concerns and symptoms related to SDB were absent with the exception of one subject with persistence of attention deficit and hyperactivity disorder that may not have been related to SDB. Despite noted improvement in symptoms following treatment, persistence of intermittent agitated sleep with teeth clenching was reported, for which the subject was referred back to the orthodontist. PSG showed a normal AHI and oxygen saturation. However, in seven cases presence of mouth breathing without indication of frequency was identified in the PSG. Parents also had been referred to myofunctional re-educators. Review of charts indicated that parents regularly followed up for orthodontic treatments. Of the 24 subjects, 10 did not go to myofunctional reeducation and three children missed routine appointments and training sessions, did not adhere to the requested exercise regimen, or did not participate in long-term follow-up with re-educators. Conversely 11 subjects were adherent to myofunctional treatment and were compliant with routine follow-up with their orthodontists. None of the subjects had begun puberty throughout the follow-up period and all remained Tanner stage 1. Children at end of treatment were told to have a yearly orthodontic follow-up to assure persistence of healthy oral development. As part of this follow-up, as subjects were growing orthodontists recommended a follow-up evaluation at the sleep clinic; the timing of this postorthodontic treatment reevaluation ranged between 38 and 50 months postorthodontic treatment.

Table 1
Clinical concerns reported by parents.

	Entry	Post-AT	Postorthodontics	Follow-up study
No. of children (*n*)	24	23	24	24
Age (y)	5.5 ± 1.2	5.10 ± 1.3	7.3 ± 1.5	11.6 ± 1.2
Snoring	24	2	0	5
Agitated sleep	22	11	1	5
EDS	10	0	0	0
Fatigue	15	23	0	11
Insomnia	5	2	0	5
Hyperactivity and inattention	7	2	1	11
Poor school performance	0	0	0	11
Parasomnia	10	0	0	0
Bruxism	3	3	1	3
Morning headache			0	2

Abbreviations: AT, adenotonsillectomy; *n*, number of children; y, years; EDS, excessive daytime sleepiness. One child never had adenotonsillectomy (see text).
The number in each column represents the number of children of which the clinical concern was mentioned by parents.
Parents did not report of school performance concerns in younger children, but they did report concerns of attention and hyperactivity, children often were considered to have possible attention-deficit/hyperactivity disorder.
As previously reported, parental concerns associated with obstructive sleep apnea vary with age [36].

Table 2
Sleep-disordered breathing documented with polysomnography.

	Entry	Post-AT	Postorthodontics	Follow-up study	
				No reeducation	Reeducation
No. of children (n)	24	23	24	13	11
Age (y)	5.5 ± 1.2	5.10 ± 1.3	7.3 ± 1.5	11.8 ± 1.4	11.5 ± 1.2
AHI (event/h)	10.5 ± 2.6	4.3 ± 1.6	0.4 ± 0.3	5.3 ± 1.5	0.5 ± 0.4*
Lowest SaO_2 (%)	90 ± 1.5	92 ± 1	95 ± 1	91 ± 1.8	96 ± 1**
Flow limitation (% TST)	–	–	10 ± 10	72 ± 14	5 ± 8***

Abbreviations: AT, adenotonsillectomy; *n*, number of individuals affected; y, years; AHI, apnea–hypopnea-index; TST, total sleep time.
Percent of flow limitation was determined based on number of 30-second epochs of sleep with abnormal nasal cannula contour not responding to definition of hypopnea with flattening of curve.
If abnormal pattern was present for more than 50% of sleep epoch, epoch was scored as flow limited. The percentage was calculated by number of flow-limited epochs × 2 (i.e., number of min) divided by TST in minute. The percentage was extracted from this calculation. Flow limitation was unavailable in most of the initial reports and initial posttreatment recordings. Flow limitation was introduced in polysomnography scoring later on, and the scoring criteria and definition used were those of the Stanford center, which had trained scorers [33].
Myofunctional treatment: significant differences between treated and untreated children Wilcoxon signed rank test.
* Significant difference ($p = .001$).
** χ^2 test ($p = .01$).
*** χ^2 test ($p = .0001$).

5.4. Evaluation at long-term follow-up

Results are presented in Tables 1 and 2. There was a clear difference between the subjects who had valid myofunctional reeducation and those who did not. Clinically, none of the 11 subjects with reeducation reported clinical concerns related to sleep disorders, and their PSG showed no evidence of breathing abnormalities during sleep. All subjects had continuous nasal breathing noted on PSG. Wilcoxon rank sum test showed that the AHI was significantly different between the two groups ($P = .001$) and χ^2 test statistics showed that the percentage of lowest oxygen saturation and the percentage of flow limitation also were significantly different ($P = .01$ and $P = .0001$, respectively) At the long-term follow-up, the 13 other subjects reported persistent daytime concerns; parents indicated presence of school difficulties including inattention in school and in some degree attributed these to fatigue in the subject. Interviews and questionnaires also indicated that specific sleep concerns persisted in some subjects, including the presence of snoring, agitated sleep, symptoms of sleep-phase delay, and morning headaches (Table 1). Children with the highest amount of flow limitation (Fig. 2) and the highest AHI scores reported more frequent concerns. All 13 subjects in this subgroup displayed mouth breathing during sleep, as demonstrated by analysis of the mouth thermistor curve on the PSG (Fig. 3). Clinical evaluation reported abnormal head posture in four of the subjects during the daytime (Fig. 4), and the previous improvements noted at the time of removal of the orthodontic equipment were lost after development of a counter-clockwise rotation of the maxilla and high and narrow palatal vault. Such findings were confirmed on evaluation by an orthodontist. These anatomic presentations were not found in the 11 subjects with normal breathing during sleep.

Myofunctional evaluation of the orofacial region showed that subjects had an abnormally low tongue position in the mouth while awake. Among the subjects, 12 were unable to perform appropriate clicking sounds with the tongue, 10 were unable to protrude their tongue upward when asked to try to touch their nose with the tip of their tongue, four had difficulties holding a button between their lips, and one had difficulties swallowing while drinking quickly. All subjects acknowledged having a preferential side for mastication, and nine subjects presented with slight asymmetry of masseteric muscles when evaluated during active contraction. At the end of the evaluation by re-educators, all subjects were scored with abnormal orofacial muscle tone while awake. All subjects without clinical concerns had been scored as normal at myofunctional testing.

6. Discussion

Our retrospective study has typical limitations associated with retrospective studies, particularly when evaluating subjects diagnosed with OSA years ago. First despite the many subjects with

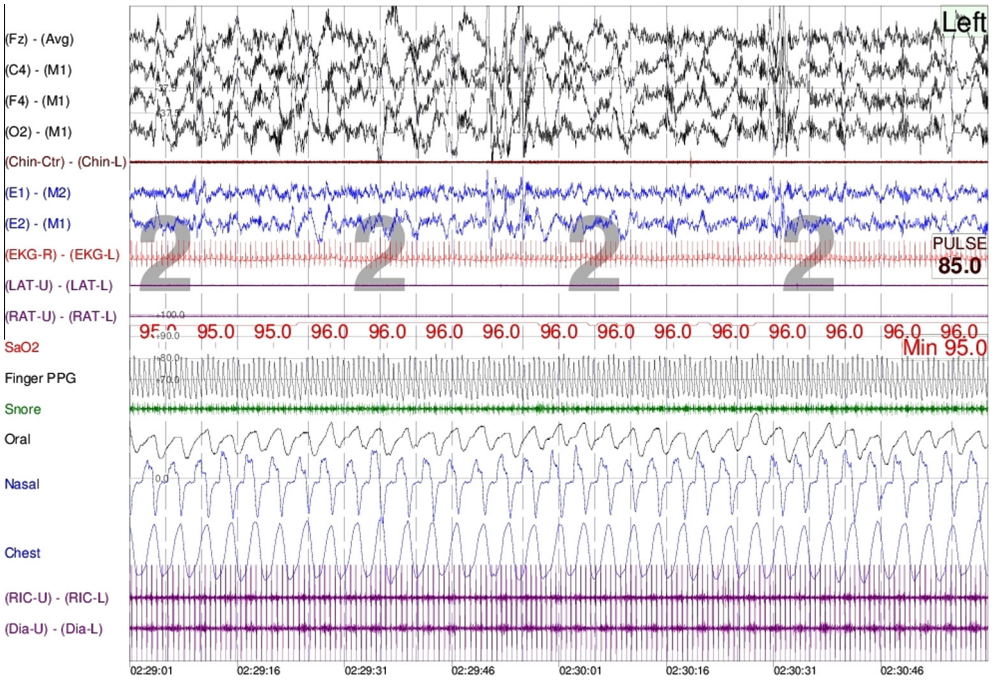

Fig. 2. Example of polysomnography (PSG) segment of a subject with recurrent symptoms postadenotonsillectomy and with orthodontic treatment but without myofunctional therapy. Note the continuous flow limitation expressed as an abnormal curve of the nasal cannula recording (#14 from top).

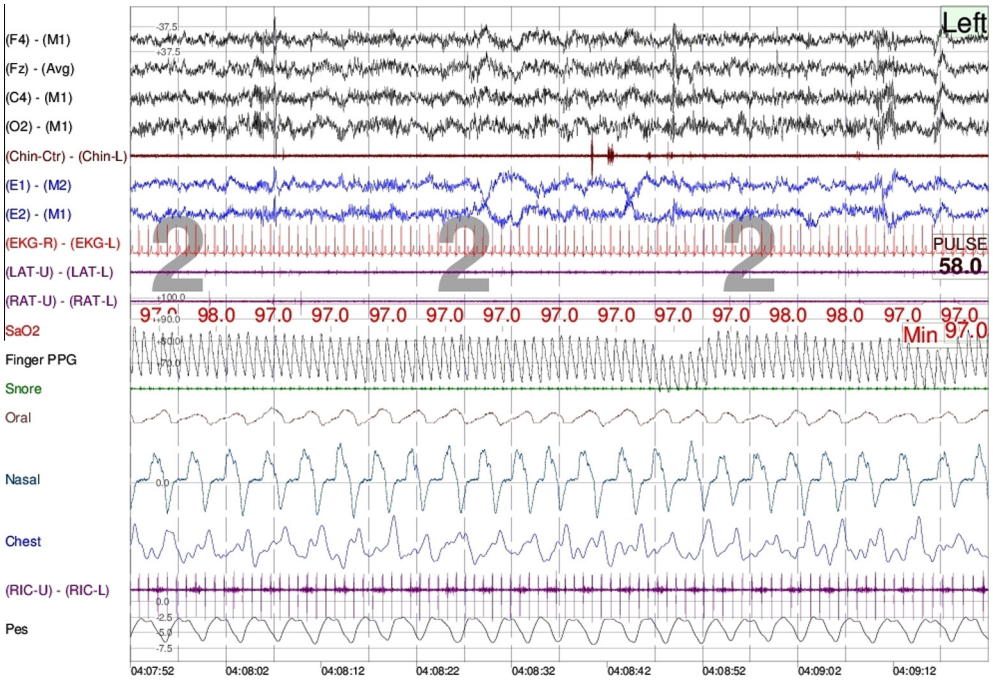

Fig. 3. PSG segment of mouth breathing and flow limitation. In a subject with recurrent symptoms, PSG showed abnormal nasal curve recording contour (tracing from nasal cannula) with flattening of the curve (#14 from top), presence of continuous nasal breathing as indicated by mouth thermistor recording (#13 from top), and presence of continuous increase effort indicated by esophageal manometry (Pes) (#17 from top) with a peak end–inspiratory pressure oscillating approximately 8-cm H_2O compared to baseline supine with normal breathing of 3-cm H_2O.

OSA treated with adenotonsillectomy alone or with adenotonsillectomy and orthodontics, this is a retrospective study with a relatively small number of subjects. This small sample was largely due to the few subjects having the documented data necessary for analysis and the absence of long-term follow-up in OSA patients. Obtaining information from three different locations with differing referral patterns also was challenging. Our goal was to assess the role of myofunctional reeducation, and medical records were not always easily retrievable. Although the different locations worked together in data collection and analyses for other investigations involving pediatric subjects with OSA, the different locations may have created variability in data collection and results. Rescoring of all follow-up PSG records obtained at the last sleep clinic visit was performed by a single-blinded scorer in an effort to avoid interlaboratory and interscorer variability. However, the initial diagnostic PSGs from all the subjects was unavailable for

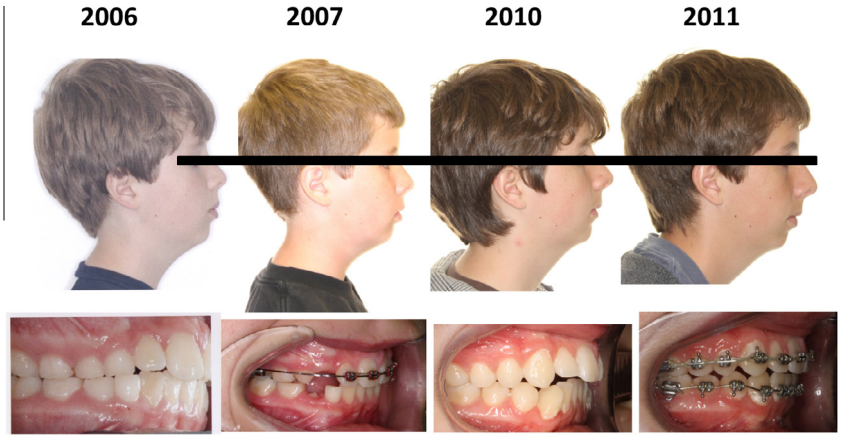

Fig. 4. Abnormal head position during wakefulness with abnormal breathing during sleep. Note the progressive change in head position over time associated with the development of an abnormal nuchal angle. The angle was normal in 2007 with progressive development of abnormal head position associated with abnormal breathing and OSA despite absence of snoring but the presence of mouth breathing during sleep. The last photo clearly shows the abnormal head-neck posture related to the sleep-disordered breathing.

review and results solely relied on the PSG reports kept on file. Finally the different laboratories were affiliated with different healthcare systems that may have influenced subjects' ability to accept or adhere to treatment recommendations. The cost of myofunctional reeducation could have been free, covered by medical insurance, or paid entirely out of pocket. Numbers may have been higher if this type of study had been performed in Brazil where such treatment is routinely included in the management of patients with OSA [2]. Additionally most of the data obtained were from children seen at orthodontic clinics, likely adding another bias. We do not claim that all children with OSA should have myofunctional reeducation, and our study does not show the role of myofunctional treatment performed without orthodontic treatment postadenotonsillectomy; however, clearly more studies are needed. Despite these biases, our study is the first retrospective study investigating myofunctional reeducation and underlining its benefits in the treatment of SDB in the pediatric population. Approximately 46% of nonoverweight children initially diagnosed with OSA had similar anatomic risk factors as in our 24 children. It is possible that adenotonsillectomy itself led back to nasal breathing during sleep, but such changes should be objectively documented several months' postsurgery. The intricate relationship between nasal breathing and orofacial growth has been studied for several years [17–28], and myofunctional reeducation programs were established with the understanding and intention of optimizing orofacial development and breathing in children. Mouth breathing is associated with malposition of the tongue, which further reinforces impaired development and growth of the maxilla and mandible. The intricate relationship between breathing and orofacial growth was studied for many years, supported by experimental animal models that were extensively studied in the 1970s. Harvold et al. [17], Miller et al. [18], and Vargervik et al. [19] showed that abnormal nasal breathing leads to abnormal EMG discharges in tongue and orofacial muscles with secondary impact on the facial skeleton and dentition. Impairment of nasal breathing also has been investigated in children and has demonstrated an impact on facial growth, head posture, and general medical consequences [20–29]. Swedish researchers also have suggested that early mouth breathing without appropriate humidification of air through the nose leads to repetitive tonsillar trauma [29]. Such trauma may lead to an inflammatory response of the tonsils, previously histologically demonstrated and also may lead to progressive enlargement of an already narrow airway.

With the understanding of these implications, treatment programs were established with the goal of optimizing orofacial development to improve breathing in children. The benefits of the combination of orthodontic and myofunctional reeducation on breathing, speech, swallowing, orofacial growth, and the elimination of abnormal head-neck posture, with a focus on eliminating tongue and orofacial muscle hypotonia, have been published particularly in the orthodontic literature [2]. This movement also has led to the development of myofunctional reeducation specialists whose expertise is sanctioned in many countries. There is no systematic prospective study involving myofunctional therapy in children with OSA, but there has been an abundance of literature on the benefits of myofunctional treatment on growth and orthodontic development for more than 20 years [1]. This literature emphasizes the importance of nasal breathing and obtaining good orofacial muscle tone to maintain orthodontic gains in children. It also stresses maintenance of obtained gains during pubertal years. However, none of these studies involved systematic PSG evaluation, and the reports emphasized orthodontic development rather than nocturnal breathing. SDB invariably involves abnormal nasal breathing and impaired facial growth associated with mouth breathing. Unfortunately, this concern often has been ignored. Our report indicates that a combined treatment approach including adenotonsillectomy and orthodontia with myofunctional reeducation can be crucial in the elimination of OSA. This finding is especially critical, as the failure to eliminate oral breathing will lead to the reappearance of the OSA syndrome in children. A recent prospective follow-up study [30] lasting 36 months that included clinical and PSG data had followed 67% of an initial OSA children cohort and showed that 68% of the children still involved in the study had either worsening abnormal breathing if adenotonsillectomy had not fully resolved the concern (with complete OSA resolution defined as AHI <1) or had reappearance of OSA, even if complete resolution had been obtained postadenotonsillectomy. Mean AHI of the cohort was approximately six events per hour and none of the children had been monitored for mouth breathing or received myofunctional therapy. Treatment achieving a normal upper airway in children does not guarantee normal tongue position or normal tongue and orofacial muscle strength during sleep. This in turn affects the development of the airway as demonstrated in monkey models [17–19]. Persistence of oral breathing during sleep directly affects tongue position and strength as well as that of the orofacial muscles, leading to abnormal airway development unless myofunctional reeducation is performed to avoid this evolution.

Despite its deficiencies our study highlights the importance of influencing normal facial growth by using the available tools and resources to optimize orofacial development in children with abnormal breathing during sleep. As previously mentioned, although integrated care between myofunctional re-educators and orthodontists may be routine in some countries, such as France, Belgium, Brazil, or Taiwan, this is not the case in all parts of the world. The lack of understanding of these interactions leads to the misconception that pediatric OSA is an upper airway syndrome and not a facial growth dysfunction with secondary impact on the upper airway.

Finally, our study shows that scoring only apneas and hypopneas is not sufficient to recognize abnormal breathing during sleep. Flow limitation [14] is a much more adequate indicator of abnormal breathing in our patients. Previous studies have shown the involvement of flow limitation in parasomnias and in abnormally high amounts of cyclic alternating pattern phases A2 and A3 [31]. Chervin et al. [32] showed that abnormal breaths that do not meet criteria for defined apneas and hypopneas still may have a disrupting effect on the sleep EEG. Recently it was shown that young women with an abnormal amount of flow limitation and a low or normal AHI had the same clinical presentation as women with pathologic AHIs [33]. It also is imperative that we recognize abnormal breathing in children during sleep and have the knowledge to select the appropriate indices to detect SDB [33]. This knowledge requires increased attentiveness for abnormal breathing, as many of our children had no snoring likely due to prior treatment, yet clearly displayed flow limitation disrupts their sleep along with persistent symptoms. We must treat children to optimize and insure normal development of the airway, orofacial muscle strength, and positioning, and in turn normal breathing during sleep. Myofunctional reeducation may be considered to treat adult patients with OSA; however, as demonstrated by Guimaraes et al. [2] even if it is effective, it has a limited impact in adulthood. This finding further reinforces the importance of early identification and intervention during childhood development to optimize normal growth of the airway and to insure a lasting impact in the treatment of SDB. With the help of orthodontists and myofunctional therapists and appropriate testing of nasal resistance in children, we may be able to recognize and treat children at risk for SDB early in life [1,34].

Conflict of interest

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: http://dx.doi.org/10.1016/j.sleep.2013.01.013.

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