Mouth breathing, “nasal disuse,” and pediatric sleep-disordered breathing

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Abstract
Background Adenotonsillectomy (T&A) may not completely eliminate sleep-disordered breathing (SDB), and residual SDB can result in progressive worsening of abnormal breathing during sleep. Persistence of mouth breathing post-T&As plays a role in progressive worsening through an increase of upper airway resistance during sleep with secondary impact on orofacial growth.

Methods Retrospective study on non-overweight and non-syndromic prepubertal children with SDB treated by T&A with pre- and post-surgery clinical and polysomnographic (PSG) evaluations including systematic monitoring of mouth breathing (initial cohort). All children with mouth breathing were then referred for myofunctional treatment (MFT), with clinical follow-up 6 months later and PSG 1 year post-surgery. Only a limited subgroup followed the recommendations to undergo MFT with subsequent PSG (follow-up subgroup).

Results Sixty-four prepubertal children meeting inclusion criteria for the initial cohort were investigated. There was significant symptomatic improvement in all children post-T&A, but 26 children had residual SDB with an AHI>1.5 events/hour and 35 children (including the previous 26) had evidence of “mouth breathing” during sleep as defined [minimum of 44 % and a maximum of 100 % of total sleep time, mean 69±11 % “mouth breather” subgroup and mean 4±3.9 %, range 0 and 10.3 % “non-mouth breathers”). Eighteen children (follow-up cohort), all in the “mouth breathing” group, were investigated at 1 year follow-up with only nine having undergone 6 months of MFT. The non-MFT subjects were significantly worse than the MFT-treated cohort. MFT led to normalization of clinical and PSG findings.

Conclusion Assessment of mouth breathing during sleep should be systematically performed post-T&A and the persistence of mouth breathing should be treated with MFT.

Keyword Sleep-disordered breathing · Adenotonsillectomy · Mouth breathing · Myofunctional treatment · Apnea-hypopnea index worsening

Introduction
Adenotonsillectomy (T&A) improves but often does not completely eliminate pediatric obstructive sleep apnea (OSA) at systematic post-surgical follow-up [1–6]. A long-term study showed that persistence and recurrence of the syndrome with slow worsening of the apnea-hypopnea index (AHI) may frequently occur within 3 years even in the setting of shorter-term postoperative benefit [7]. Recent work has indicated that a substantial portion of those with pediatric SDB will have persistence of SDB up to 4 years later [8]. Based on short-term follow-up periods, children with atopy (allergies, asthma) are thought to have increased risk of having persistence of sleep-disordered breathing (SDB) with snoring, flow limitation, and/or low amounts of apnea-hypopnea during sleep post-T&A, but this finding was not confirmed in the 3-year follow-up study [7].
Data from Rhesus monkey investigations [9, 10] and from human orthodontic studies demonstrated that mouth breathing leads to abnormal orofacial growth that can be readily observed [11–14]. Similarly, there are data showing that abnormal orofacial growth is associated with sleep-disordered breathing [15, 16]. Finally, it has been previously shown that mouth breathing leads to a significant increase in upper airway resistance [17]. Chronic mouth breathing is detrimental in developing individuals, and it has been shown that nasal breathing is the primary route of airflow responsible for about 92 and 96 % of inhaled ventilation during wakefulness and sleep, respectively [18]. We previously found that mouth breathing was a commonly seen finding in children who were later found to have symptomatic abnormal breathing during sleep [19]. Despite this knowledge, no systematic attention is paid to restoration of nasal breathing when treating sleep-disordered breathing with surgical approaches such as T&A and nasal surgery when assessing response to treatment, including with polysomnography (PSG).

We questioned as a first step how frequent mouth breathing during sleep was before T&A surgery in SDB children and how much improvement of this abnormal behavior was noted at post-surgical evaluation. Also, as a second goal, we search for a possible approach in treating the persistent mouth breathing noted during sleep in the studied children.

As mentioned above, there are data demonstrating that oral breathing impact on oral-facial growth. Also, orofacial muscle training and reeducation of normal oral-nasal functions alongside orthodontic treatment has been implemented for many years because of the successful results of treating open bites and crossbites when combining both approaches [16, 20–24]. In teenagers with low to moderate AHI, daily orofacial muscle training (termed “myofunctional therapy” [18, 19]) has been reported to help eliminate abnormal breathing during sleep, including detrimental mouth breathing, at follow-up [20, 24–27]. Similarly, in young school-aged children, oropharyngeal exercises performed after T&A improved residual symptoms of OSA. Indeed, similar findings have been seen in adults, with specific orofacial muscle training that significantly reduced AHI [28, 29]. We questioned, as a preliminary investigation, if such approach can be a helpful addition to treatment particularly when mouth breathing was present post-surgery, collaborating with myofunctional therapists that are well aware of sleep-disordered breathing in our region.

One of the goals of myofunctional therapy in an orthodontic setting is to modify the swallowing pattern, mastication, and suction and eliminate mouth breathing that may interfere with or reduce the results of orthodontic treatment [20]. Long-term follow-up of children treated with T&A shows that even with systematic administration of montelukast and nasal allergy treatment, recurrence or worsening of abnormal breathing during sleep is possible. As there exists some component of SDB that can remain after T&A and anti-inflammatory therapy, we have also recommended regular clinical and PSG follow-up to evaluate long-term evolution; this recommendation has not been systematically followed by pediatricians in the community and parents, but some results are however available.

This study reports the results of a retrospective analysis of children with SDB who underwent post-T&A polysomnography (PSG) with quantifiable data on mouth breathing. We investigated whether myofunctional reeducation was effective to alter the mouth breathing pattern in children and whether this had an impact on nighttime respiratory parameters in SDB children. This retrospective investigation performed on data rendered anonymous was approved by the IRB.

Methods

Protocol

Inclusion criteria. To be in the study, children had to be prepubertal at entry. They must have had complete clinical charts indicating the clinical presentation at entry, with demonstration by examination of the absence of nasal allergies and the absence of orthodontic crossbites or significant dental crowding. All subjects had an in-laboratory polysomnogram (PSG) and those children referred to otolaryngology and who had adenotonsillectomy performed, with a post-T&A PSG taken, were included.

Exclusion criteria. Overweight/obese children, children with syndromic craniofacial malformations, and children with other medical problems including asthma and desensitization for upper airway allergies were excluded from the review.

Taking into account inclusion and exclusion criteria, we created a retrospective cohort. All children with complete data and successively seen during the 24-month period ending in December 2012 were included in the review. We then collected follow-up data available for this cohort, evaluating clinical data, the presence or absence of myofunctional therapy recommendations, whether the recommendation was implemented, and PSG if it was performed about 12 months later. We ended with an “initial cohort” and a “follow-up subgroup.” The goal of the follow-up subgroup was to obtain a preliminary investigation on possible means of restoring nasal breathing if this normal function was lacking post-surgery.

Data collection

All children responding to inclusion and exclusion criteria are included in the study. At entry, all children completed the Pediatric Sleep Questionnaire [30] and underwent a systematic sleep-medicine evaluation guided by a standardized form.
Anatomic scales evaluating the upper airway (Mallampati-Friedman scale, Friedman tonsils scale inferior nasal turbinates, dental crowding, presence of overjet, overbite, and facial harmony) were used [31]. Systematic evaluation for presence of nasal allergies and rhinitis and presence of orthodontic problems was also performed with referrals to specialists if needed. An in-laboratory PSG was performed with a test lasting a minimum of 7 nocturnal hours with light-out at regular home sleep time and one parent sleeping on a fold-out bed in the same room as the child.

PSG recording

The following variables were collected: EEG (four leads), eye movement chin and leg EMG, ECG (one lead), and body position. Respiration was monitored using nasal pressure transducer, mouth breathing was monitored using an oral scoop (Braebon Medical, ON, Canada) which was modified to accurately detect oral flow and separate it from any nasal flow alteration [32] (Fig. 1), chest and abdominal movements using inductive plethysmography bands, diaphragmatic-intercostal, and rectus-oblique muscle EMGs, pulse oximetry (Massimo TM) from which both oxygen saturation (SaO2) and finger plethysmography were derived, and continuous video monitoring was done (see Fig. 2). All children, per study design, were referred to ENT, and all had T&A without indication of significant post-surgery complications. In all, six different ENT surgeons performed surgery, and all children were considered healed from surgery when seen again in the sleep clinic for post-surgery evaluation and PSG. Per clinic policy, the pre- and post-surgical PSGs were usually scored by same individuals.

Based on the findings at post-T&A PSG recordings, parents were referred to myofunctional therapy (three different therapists were used) and/or were recommended to have a 6-month follow-up clinical visit and a yearly reevaluation at the sleep clinic with PSG if needed.

Child and parents go to the specialist for training sessions a mean of three times/week initially. Parents and child are instructed how to perform daily exercises, and a log of each daily session and types of exercise is filled on a daily basis; based on the progresses and collaboration of child, the frequency of weekly session with the specialist decrease with time, but daily logging with evaluation of the log at each session by the specialist is carried till end of training. There is a regular interaction between the reeducator-specialist and the sleep-physician, and regular written reports outlying number of sessions, findings from the log, and difficulties with training are sent evaluating compliance with treatment. At regular interval, the reeducator-specialist performs a systematic evaluation of oral-facial muscle activity that is kept in the child file. Only a subset of children came back for the sleep follow-up 6 months and 12-month reevaluation. The evaluation included the same questionnaire, same clinical evaluation, and same PSG protocol as at entry and post-surgery time points. Thus, after the initial evaluation, there were three other appointments for sleep follow-up. The children were seen post-T&A and again at 6 and 12 months post-T&A.

Analysis

As mentioned, anatomical scales were used to analyze oral-facial anatomy following published scoring criteria [28]. Sleep and respiratory scoring of PSGs followed the recommended pediatric scoring, according to the American Academy of Sleep Medicine (AASM) [33]. The presence of nasal flow limitation was determined using criteria published by Palombini et al. [34] and Guilleminault et al. [35]. Mouth breathing during sleep was calculated based on the recording obtained from a modified cannula with oral scoop [31]. Each 30-s epoch of sleep recording was scored for presence/absence of mouth breathing. To be scored as a “mouth breathing epoch,” more than 50% of the epoch must have shown recording of air flow with the oral scoop thermocouple. We defined “mouth breathing during sleep” to occur when a subject spent a minimum of 35% of total sleep time (TST) with mouth breathing. [This cutoff was based on analysis of 10 pediatric PSG children not included in the present study and part of a preliminary investigation: there was absence of clinical complaint when mouth breathing was below 20% and presence of some complaints if mouth breathing was present for more than 40% of sleep monitored with PSG. The decision to select 35% was thus a “preliminary” decision, and all records in the study were scored assessing “mouth breathing”). The compliance to treatment with myofunctional therapy came from the data collected by the reeducator-specialist and were derived from parental report and daily logs.

Fig. 1 Equipment to record mouth breathing (Courtesy of Oscar Carrillo). Legend “Oral breathing is determined by a thermocouple fixed on a scoop that allows collecting flow coming from mouth breathing. The system allows to monitor nasal pressure and end tidal CO2. The system was validated using measurement of end—tidal CO2 collected directly at the mouth and comparing the mouth end tidal CO2 signal to the thermocouple signal [31]”

Statistical evaluation

Data were de-identified and placed in an Excel file for analysis. Chi-squared (percentage) and t tests for repeated measures were used. In cases where datum was not normally distributed, the Wilcoxon signed-rank test was used. To compare data from three successive time points (baseline, post-surgery, and ~12 months post-surgery), a repeated measures analysis using general linear modeling for AHI, flow limitation, and SaO2 was performed. SPSS version 12 was used for statistical analysis.

Results

There were 92 children for potential inclusion between the ages of 3 and 9 years identified during the selected time period. Of these, 64 individuals met the inclusion criteria for the initial cohort. They represented the study group. The clinical symptoms and results of PSG before T&A surgery are presented in Table 1.

Overall, this group of normal weight children without allergy or orthodontic problems had positive surgical results, and parents reported symptomatic improvement in all cases. As shown in Table 1, following surgery, there was significant improvement of an increase of SaO2 nadir and a decrease in mean AHI (8.58 before, 1.71 after, p<0.001). However, there was still residual SDB in 26 children (40.6 %), as they had an AHI equal or higher than 1.5 events/hour, and 35 children had the presence of mouth breathing for at least 35 % of total sleep time (see Fig. 2).

Table 1 shows that the 26 children with residual post-op OSA had some symptoms (particularly reports of “fatigue”, n=25) despite overall improvement. Comparison of children with and without mouth breathing is presented in Table 2. Overall, the mouth breathers had a significantly higher residual AHI compared to the nasal breathing children. Interestingly, nine children with no symptoms, as reported by parents, had an AHI below 1.5 event/hour and mouth breathing for more than 35 % of total sleep time on post-T&A PSG.

Prior to surgery, 63 of the 64 children in the initial cohort showed “mouth breathing during sleep” per our definition, and post-surgery there was 35/64 children with “mouth breathing during sleep.” In this “mouth breathing” subgroup, mouth breathing was present during sleep for a minimum of 44 % and a maximum of 100 % of total sleep time, with a mean percentage PSG with mouth breathing during sleep of

Fig. 2 PSG recording indicating mouth breathing in a 5-year-old child. Legend top segment: 30-s recording during NREM sleep with from top to bottom. EEG (four leads 1—4), chin EMG (one lead—5), electrooculogram (two leads—6, 7), electrocardiogram (one lead—8), pulse-oxymetry (9) with write-up of oxygen saturation (10), finger plethysmography (11), nasal cannula-pressure transducer (12), mouth breathing recording (13), chest and abdomen inductive plethysmography recording (14, 15), leg EMG (16), transcutaneous CO2 (17), and intercostal diaphragmatic EMGs (18, 19). Bottom segment: All-night recording of pulse oximetry (20) and of mouth breathing (21)—bottom signal. As can be seen looking at top segment: there is continuous mouth breathing during the 30-s segment; it is associated with a “flattening” of the inspiratory wave contour of the nasal cannula-pressure transducer and a lengthening of inspiration (per convention, “inspiration” is “up” in the recording). The bottom segment shows that there is very limited change in oxygen saturation during the entire night, but (bottom recording) mouth breathing is observed during a large amount of total sleep time. The segments without mouth breathing do not correlate with specific sleep states or specific body position. One hypothesis that could not be verified was related to the question of the role of the cyclical alternating physiological turbinate turgescence during sleep.
The nasal breathing subgroup had a mean total mouth breathing sleep time of 4±3.9 %, ranging from between 0 and 10.3 % of total sleep time.

Myofunctional therapy

All subjects with persistence of mouth breathing >35 % of total sleep time on postoperative PSG were educated at the follow-up visit on the negative impact of mouth breathing on orofacial growth. Parents were provided with an introduction to myofunctional exercises through web pages (www.myofunctionaltherapy.blogspot.com and www.sleep-apneaguide.com/oropharyngeal-exercises.html) and demonstration of types of possible exercises to perform for at least 6 months. Parents were also systematically given referrals to myofunctional therapists in contact with the sleep clinic. Yearly follow-up recall at the sleep clinic was recommended to assess status. Myofunctional therapy was administered by three different specialists.

Follow-up at +6 and +12 months post-surgery

After the post-T&A evaluation and PSG, subjects, identified with persistent mouth breathing with our definition, were advised to have follow-up clinical appointment 6 months after the post-surgery sleep clinic visit and repeat PSG investigation 1 year after the initial post-surgery sleep clinic visit. Twenty-nine of the 35 children with mouth breathing (91 %) came for a 6-month clinical follow-up, but only 7 of these 29 reported participating in a myofunctional therapy program. At this visit, repeat recommendations for myofunctional therapy and referral for therapy were again made. At 12 months post-T&A, 18 children in the persistent mouth breathing post-T&A group (i.e., 51.4 % of the initial subgroup) were seen again and underwent PSG. In this subgroup, nine children reported having received myofunctional therapy (see Tables 3 and 4). As a total group (n=18), AHI, O2 saturation nadir, and nasal flow limitation were not significantly different 12 months after T&A, compared to immediately after T&A. However, there were significant differences between those who reported undergoing myofunctional therapy compared to those who did not, with all three measures of AHI, O2 saturation nadir, and nasal flow limitation showing improvement in the myofunctional therapy group (see Table 4). Two of the children without symptoms but with mouth breathing at post-T&A study are in the group of the “nine untreated children” at the +12 months post-T&A PSG. These children present worse PSG findings than just post-T&A and have now abnormal findings.

Discussion

In this study, mouth breathing was noted before any treatment, for a minimum of 1/3 of TST on PSG in 63 out of 64 children who met the inclusion criteria. Post-TKA, there were still 35 children (55.5 %) with persistent mouth breathing (as defined)
during sleep. These children tended to have persistence of OSA and the presence of flow limitation despite overall significant improvement of clinical and PSG variables. As specified for inclusion, these children had no evidence of nasal allergies, and there was no indication for orthodontic treatment, which are factors that may play a role in persistent mouth breathing. Findings suggest the presence of “nasal disuse” during sleep in these children who previously had enlarged adenoids and/or tonsils for some time before the decision to perform treatment. The large percentage of residual mouth breathing children post-T&A supports clearly the notion that removal of obstructive upper airway tissues does not systematically mean return to normal nasal breathing during sleep. This is the first study that documents this finding of residual mouth breathing after T&A, even in absence of snoring. Our finding that the normal “nasal breathing children” had on average about 4 % of total sleep time [range 0 to 10 %] is in agreement with an earlier study that showed that normal subjects spend an average of 96 % of their sleep time with nasal breathing [18]. We had selected a cutoff point of 35 % of mouth breathing during sleep based on a small preliminary study. This largest study shows that this cutoff point is most probably incorrect, with our maximum range of 10 % mouth breathing asleep in our asymptomatic children with normal PSGs. To date, we would change our cutoff point to 15 % of total sleep time.

Of interest is the finding that nine children without residual symptoms and an AHI considered as within the normal range presented an abnormal percentage of time with mouth breathing during sleep. Only two of these nine had 1-year follow-up recordings, but both presented mild symptoms and abnormal AHI at this follow-up indicating the need for further investigation.

Myofunctional therapy, prescribed with an aim to eliminate mouth breathing and reestablish nasal breathing, was associated with clinical and PSG improvement in all children who followed the recommendations. No other therapeutic approach was recommended between a second and a third time point except for myofunctional therapy, though it is possible that stochastic factors such as time of year, intercurrent illness, etc., influenced these results.

Our follow-up data are limited. Out of the initial 64, or out of the 35 recognized with mouth breathing during sleep post-T&A, we only had 18 children that came back at 12 months for in-laboratory PSG. Those 18 may represent a bias group; in the children group who had undergone myofunctional therapy, parents were clearly encouraged by reeducators to check results of intervention, and in the 9 children who came back without having had intervention, we cannot eliminate the fact that parents may have observed presence of low-grade symptoms that may have led to obtain a new PSG. This is not a double-blind randomized study. But overall, our investigation even with limited numbers supports our hypothesis: myofunctional therapy may help in eliminating mouth breathing during sleep. There are strong data indicating that chronic mouth breathing leads to change in orofacial growth with impairment of maxillomandibular development relatively early in life.

In the Rhesus monkey model [9, 10], impairment of nasal breathing leads quickly to oral-facial changes through changes in muscle recruitment. From an orthodontic perspective, the negative role of mouth breathing on orofacial anatomy in children has been documented by many authors [11–16], and maxillomandibular compromised associated with nasal

<table>
<thead>
<tr>
<th>Table 3</th>
<th>Repeated measures analysis of general linear modeling for AHI, flow limitation and SaO2 before T&amp;A, after T&amp;A, and at 12 months after T&amp;A (n=18) in children with persistent mouth breathing post-T&amp;A</th>
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<tbody>
<tr>
<td></td>
<td>Before T&amp;A</td>
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<td></td>
<td>Mean (SD)</td>
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<tr>
<td>AHI SD</td>
<td>9.17 (2.72)</td>
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<tr>
<td>SaO2 SD</td>
<td>89.21 (2.39)</td>
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<tr>
<td>FL SD</td>
<td>79.43 (7.30)</td>
</tr>
</tbody>
</table>

Legend: There is a significant change in the respiratory variables obtained with PSG between pre- and post-T&A, but there is no significant difference for the 18 subjects group between post-T&A PSD results and +12 months post-T&A results with a trend toward improvement overtime.

\[ \text{FL SD} | 79.43 (7.30) | 11.4 (8.19) | 7.50 (12.97) \]

Statistics was performed by Mann-Whitney test.

Legend: two of the asymptomatic and with normal PSG after T&A children are in the nine “untreated children” subgroup and now present abnormal findings at +12-month PSG with persistence of mouth breathing during sleep.

\[ \text{AHI} | 1.1 (1.19) | 2.94 (1.37) | 0.015 \]

\[ \text{Flow limitation} | 0.56 (1.67) | 19.44 (14.24) | 0.003 \]

\[ \text{SaO2} | 96.11 (1.05) | 94.56 (1.67) | 0.037 \]
breathing alteration has also been reported despite T&A [16]. With mouth breathing, there is a “disuse” of nasal breathing with changes in proprioception, posture, and loss of usage of the nose [26]. Such observations were made long ago by orthodontists treating narrow palates, crossbites, and other maxillomandibular problems, as absence of normal nasal breathing and persistence of mouth breathing was found to be a handicap for positive long-term results of some orthodontic treatment approaches [20]. Recently, Souki et al. [36] have looked at the impact of mouth breathing versus nose breathing on cephalometric measurements in children in variable stages of dental development (mean age 4 years and 8 months versus 7 years and 9 months). These authors concluded that a significant difference is noted in the dentofacial patterns of mouth breathing children with some differences dependent on age [36]. But none of these studies has considered sleep and SDB.

Myofunctional treatments were developed to encourage and establish normal orofacial muscle tone associated with normal nasal breathing through daily exercise involving orofacial muscles and stimulation of sensory pathways. In children, these exercises are done under parental supervision and with regular sessions with trained therapists. The training given by the therapist typically involves frequent interaction in the early stages of muscle reeducation, with longer intervals between sessions as therapy continues. Affirming compliance with treatment is always difficult, usage of log books kept by child and parents and regular interaction with the trained therapist are two tools that have been used to maintain compliance.

In conclusion, children are thought of as “obligatory nose breathers” at birth, and nasal breathing is of critical developmental importance for normal oropharyngeal development. The case against mouth breathing is growing, and given its negative consequences, we feel that restoration of the nasal breathing route as early as possible is critical. In fact, restoration of nasal breathing during wake and sleep may be the only valid “complete” correction of pediatric sleep-disordered breathing, although the importance of establishing daytime nasal respiration in affecting night time upper airway properties is not known. What’s more, it appears that we cannot assume that T&A alone can be relied upon to sufficiently restore normal breathing during sleep. Nasal breathing during wake and sleep is the demonstration of normal respiratory functioning in a child, and persistence of mouth breathing is an indicator for the need for further treatment of sleep-disordered breathing. Finally, our findings emphasize the importance of post-T&A PSG investigation with monitoring all important variables including mouth breathing.

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Ethical considerations This retrospective study on data rendered anonymous was approved by the IRB.

Conflict of interest None of the authors has conflict of interest.

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