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## Original Article

# Rapid maxillary expansion (RME) for pediatric obstructive sleep apnea: a 12-year follow-up

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## ABSTRACT

**Objective:** The objective of this study was to prospectively evaluate the long-term efficacy of rapid maxillary expansion (RME) in a group of children with obstructive sleep apnea (OSA).

**Material and method:** Thirty-one children diagnosed with OSA were involved in the study. These children had isolated maxillary narrowing and absence of enlarged adenotonsils at baseline. Twenty-three individuals (73% of the initial group) were followed up annually over a mean of 12 years after the completion of orthodontic treatment at a mean age of 8.68 years. Eight children dropped out over time due to either moving out of the area ( $n = 6$ ) or refusal to submit to regular follow-up ( $n = 2$ ). Subjects underwent clinical reevaluation over time and repeat polysomnography (PSG) in the late teenage years or in their early 20s. During the follow-up period, eight children dropped out and 23 individuals (including 10 girls) underwent a final clinical investigation with PSG (mean age of 20.9 years). The final evaluation also included computerized tomographic (CT) imaging that was compared with pre- and post-initial treatment findings.

**Results:** Yearly clinical evaluations, including orthodontic and otolaryngological examinations and questionnaire scores, were consistently normal over time, and PSG findings remained normal at the 12-year follow-up period. The stability and maintenance of the expansion over time was demonstrated by the maxillary base width and the distance of the pterygoid processes measured using CT imaging.

**Conclusion:** A subgroup of OSA children with isolated maxillary narrowing initially and followed up into adulthood present stable, long-term results post RME treatment for pediatric OSA.

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## 1. Introduction

Treatment of prepubertal children with obstructive sleep apnea (OSA) is a difficult task. Since the 1970s, adenotonsillectomy (T&A) has been considered the first-line treatment in non-syndromic children. However, short- and long-term follow-up studies have shown that many children have residual symptoms and elevated apnea–hypopnea indexes (AHIs) after T&A. Several studies have also found that OSA frequently recurs following T&A either alone [1–7] or in combination with rapid maxillary expansion (RME) as initial treatments [8–10]. Nasal continuous positive airway pressure (CPAP) is a successful treatment for pediatric OSA, although compliance can be problematic. This is particularly true for adolescents and young adults. Additional data indicate that the CPAP mask necessary to administer positive pressure can have a detrimental effect on the orofacial growth of a child over time, that is, worsening the anatomic

facial growth abnormalities, leading to increased collapsibility of the upper airway during sleep [11]. We have prospectively followed up children seen during prepubertal years for OSA who were treated with RME. The initial data obtained on these children have been previously reported [12]. Following initial posttreatment evaluation, these children were asked to undergo yearly orthodontic evaluations, at which time information was collected on the development of any new symptoms. This report presents the clinical and polysomnographic (PSG) findings obtained during the prospective orthodontic follow-up. This study was approved by the University of Tor Vergata institutional review board.

## 2. Materials and methods

### 2.1. Subjects

There were initially 31 Caucasian children (19 boys). The children were seen at a mean age of 8.68 years (range: 6–12 years), and all were considered as prepubertal based on clinical evaluation (Tanner stage 1 [13]). They had a specific anatomic presentation: none had enlarged T&A confirmed by a fiber-optic ear nose throat

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(ENT) evaluation, clear maxillary deficiency was present, and narrow hard palates with unilateral or bilateral cross-bites were noted. The studied children were distributed into three skeletal classes based on the skeletal–sagittal relationship (class I,  $n = 9$ ; class II,  $n = 14$ ; and class III,  $n = 8$ ). This distribution was expected, as maxillary constriction is an abnormality of the transverse diameter.

## 2.2. Follow-up evaluations

Following initial posttreatment evaluation, the children were asked to undergo a yearly follow-up with an orthodontic specialist and an ENT specialist. Information was also collected from questionnaires to evaluate for the development of any new pediatric symptoms. Not all children came back for such an evaluation; eight children dropped out of the follow-up group, including six who moved out of the geographic area and two whose parents did not want to return for a regular follow-up. The final group consisted of 23 individuals (73% of initial group).

The mean follow-up from the first evaluation to the last evaluation of these 23 children was  $12.3 \pm 1.5$  years, and the mean time from the initial posttreatment evaluation to the last evaluation was  $12.0 \pm 0.5$  years.

## 2.3. Testing

Evaluation consisted of clinical interview and evaluation, completion of the Pediatric Daytime Sleepiness Scale (PDSS) [14] or the Epworth Sleepiness Scale (ESS) [15] based on the age of subject, and the Italian translation of the Pediatric Sleep Questionnaire [16]. Each child underwent annual otolaryngologic and orthodontic evaluation until the final follow-up.

## 2.4. PSG evaluation

An ambulatory PSG was performed at entry and at final evaluation. Electroencephalogram (EEG), eye movements, chin, leg electromyogram (EMG), and respirations were monitored with a nasal cannula, a mouth thermistor, an uncalibrated inductive plethysmograph, thoracic and abdominal bands, a snore microphone, a position sensor, and a finger pulse oximeter.

Computerized tomography (CT) imaging was performed at entry and at final follow-up. Data were compared with initial pre- and posttreatment findings.

## 2.5. Analysis

PSGs were scored (or rescored for initial studies) based on the 2007 American Academy of Sleep Medicine (AASM) recommendations [17]. Statistical analyses used paired *t*-test when data were normally distributed and Wilcoxon signed-rank test otherwise; percentages were analyzed using chi-squared statistics, considering the significant level at  $p = 0.05$ . Calculations were performed using Statistical Package for Social Science (SPSS) version 17.

## 3. Results

Due to dropout during the follow-up period, the final group consisted of 23 individuals (73% of initial group). As mentioned, the mean follow-up from the first evaluation to the last evaluation was  $12.3 \pm 1.5$  years, and from the posttreatment evaluation to the last evaluation  $12.0 \pm 0.5$  years. There were 13 boys with a mean age (standard deviation, SD) of  $20.9 \pm 1.2$  years and 10 girls with a mean age of  $21.3 \pm 1.5$  years at the time of final evaluation. None of the subjects was overweight (mean body mass index, BMI =  $22.7 \pm 1.3$  kg/m<sup>2</sup>). All of them were at least Tanner stage 5 [13].

**Table 1**

Polysomnographic results immediately post treatment and at long-term (12 years) follow-up.

	Results after completion of initial RME (n = 31)	Results at long term follow-up (n = 23)	p-value
PSG parameters			
AHI	$0.4 \pm 1.1$	$0.3 \pm 0.9$	NS
range	0–2.1	0–1.8	NS
Nadir SpO <sub>2</sub> (%)	$95.3 \pm 1.7$	$97.2 \pm 1.5$	NS
% sleep time with SpO <sub>2</sub> < 92%	$1.3 \pm 1.1$	$1.1 \pm 1.0$	NS
Sleep efficiency (%)	$89.2 \pm 7.7$	$90.1 \pm 6.5$	NS

Legend: AHI: apnea-hypopnea-index % sleep time with SpO<sub>2</sub> < 92%: percentage of sleep-time spent below 92%.

As initially reported at the end of the expansion at a mean age of 8.57 years in the 23 followed-up children, there was an opening of the inter-incisive space of  $2.95 \pm 0.3$  mm. PSG showed a change in the mean AHI from  $12.20 \pm 2.6$  to  $0.4 \pm 1.6$  and oxygen saturation nadir from  $78.9 \pm 8.6\%$  to  $95.1 \pm 1.9\%$ . Complete resolution of clinical complaints had occurred [12].

Documents obtained at the yearly follow-up were reviewed: Clinical evaluation and questionnaires indicated normal development and absence of complaints or symptoms related to sleep-disordered breathing. Over time, questionnaires always showed very low scores. Regular orthodontics and ENT evaluations did not uncover any abnormalities.

At the last evaluation, the 23 individuals with a continuous follow-up had no indication of OSA recurrence: There was no clinical complaint and no abnormal findings based on scales (with an ESS mean score of  $3 \pm 1$ ). All individuals had normal schooling. The results of the PSG are outlined in Table 1, and they are in a normal range (see the insert in Table 1).

Statistical analyses comparing initial and final results showed that there were no significant changes in PSG results between the results obtained at the completion of initial treatment and at the end of long-term follow-up (mean posttreatment follow-up =  $12 \pm 0.5$  years).

CT imaging was obtained to analyze and compare the maxillary base width and the distance of pterygoid processes at the initial pretreatment and immediate posttreatment and at the end of long-term follow-up. Analyses of these markers confirmed the stability and maintenance of the anatomical changes induced by the orthodontic treatment.

## 4. Discussion

This is the longest follow-up investigation of children treated with orthodontics presented to date, despite the fact that shorter prospective follow-up and retrospective investigations have previously been reported [9,10,18]. As expected, a small percentage of subjects dropped out. However, 73% of the initial group remained until the final follow-up, a percentage sufficient to bring valid information. Our subjects had specific features that are not necessarily present in all children with OSA who are treated with orthodontics: There was no T&A enlargement at entry, and the orofacial abnormalities were restricted to the maxilla. Finally, all children were Caucasians from a very specific geographic area. These are not the most common demographics when considering children with OSA. However, the results indicate that a subgroup of children with the anatomic characteristics described earlier have a strong possibility of achieving a sustained resolution of OSA following RME at a prepubertal age.

One ENT study reported in the literature is related to ours. This study investigated long-term outcomes post T&A in children with sleep-disordered breathing [2]. In contrast to our study, these chil-

dren were only treated by ENT approaches: Tasker et al. [2] evaluated children who were initially scheduled to have T&A. The subjects were surveyed, and they underwent home monitoring. Sixty-one children were studied at baseline and assessed using questionnaires and home ambulatory monitoring. EEG was not recorded, but pulse transit time (PTT) and oximetry in association with recording of snoring were the major variables. Pre- and post-treatment monitoring was compared with 30 age-matched control children without clinical indication of enlarged adenotonsils. There was no yearly follow-up, but 20 of the initial 61 subjects and 20 of the controls (studied around four years) were reevaluated at around 16 years of age. Fifteen of the ENT-treated children had T&A, three had adenoidectomy alone, and two had tonsillectomy alone. The authors reported that with their recording technique, the children who had undergone T&A presented 12 years later with significantly more snoring, and they showed significantly more respiratory efforts measured by PTT compared with controls. The authors attribute the persistence of the differences between the two groups in part to weight (obesity) and to nasal allergies. They also mentioned that the prior T&A group retained “a narrower airway during sleep.”

Since 2002, investigations have shown the importance of determining the impact of enlarged T&A on the growth of orofacial structures in prepubertal children, as the orofacial structures may not have developed in a normal fashion due to abnormal nasal breathing [19,20]. Studies have also shown the importance of orthodontic treatment for upper airway narrowing in the presence of sleep-disordered breathing, including post-T&A [8–10,18,21].

When comparing the literature data on the impact of both treatments (orthodontics and T&A), one may question if orthodontic treatment should not be the first approach when clinical evaluation and testing indicate the presence of both problems. We already know that in many cases, both treatments and retraining for nasal breathing during sleep [8–10,21] will be needed to have stable, long-term gains. Our study, however, shows that certain children with a specific anatomic presentation and small tonsils and adenoids [21] may not need T&A, and they may experience a stable long-term gain when reaching adulthood.

A careful analysis of orofacial abnormalities, including enlarged tonsils and/or adenoids, and maxillary–mandibular involvement in the presence of sleep-disordered breathing is mandatory to consider, as these elements may be associated with incomplete results at the end of the treatment and/or with the recurrence of OSA at a later age [8–10,21]. A subgroup of children with a specific anatomic presentation at the time of initial diagnosis may respond best to palatal distraction, despite the fact that in other cases orthodontic treatment may only be a useful adjuvant to obtain long-term normal breathing during sleep.

### 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.2015.04.012>.

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