Rapid maxillary expansion in children with obstructive sleep apnea syndrome: 12-month follow-up

Maria Pia Villa a,*, Caterina Malagola b, Jacopo Pagani a, Marilisa Montesano a, Alessandra Rizzoli a, Christian Guilleminault c, Roberto Ronchetti a

a Pediatric Clinic, Sant’Andrea Hospital, II Faculty of Medicine, University “La Sapienza”, Rome, Italy
b Orthodontic Clinic, Sant’Andrea Hospital, II Faculty of Medicine, University “La Sapienza”, Rome, Italy
c Sleep Disorders Clinic, Stanford University, Stanford, CA, USA

Received 11 April 2006; received in revised form 11 April 2006; accepted 5 June 2006
Available online 18 January 2007

Abstract

Objectives: To assess the outcome of rapid maxillary expansion in the treatment of obstructive sleep apnea syndrome (OSAS) in children, we studied 16 patients (mean age 6.6 ± 2.0; 9 males) with dental malocclusion, a body mass index ≤ 85 percentile, and OSAS confirmed by polysomnography.

Methods: At baseline and after the trial, all patients underwent physical examination, standard polysomnography and orthodontic assessment. The Brouillette questionnaire investigating symptoms of OSA was administered to parents before and during the trial to assess the clinical severity of their sleep-disordered breathing. Two treated patients were lost to follow-up and excluded from the final study.

Results: In the 14 treated subjects who completed the study and follow-up, polysomnography showed a significant decrease in the apnea-hypopnea index (p = 0.005), hypopnea obstructive index (p = 0.002) and arousal index (p = 0.001). Questionnaire responses before and after treatment showed a significant decrease in the severity of symptoms.

Conclusion: A rapid maxillary expander is an effective appliance for treating children with OSAS.

Keywords: Children; Obstructive sleep apnea; Rapid maxillary expander; Treatment

1. Introduction

The most common cause of childhood obstructive sleep apnea syndrome (OSAS) is adenotonsillar hypertrophy [1–3]. As a surgical approach, adenotonsillectomy is limited by surgical risks [4–6] and, in children with OSAS, by recurrence [7]. In a recent study, Guilleminault et al. reported on a cohort of OSAS children who initially responded to adenotonsillectomy but in whom symptoms recurred during adolescence [8]. Childhood OSAS, therefore, emerges as a dynamic process resulting from a combination of structural and neuromotor abnormalities rather than from structural abnormalities alone [8–10]. Correction of craniofacial deformities such as mandibular or maxillomandibular deficiency, both among the known syndromic risk factors, has been shown to improve OSAS [10,11].

A therapeutic option that reportedly improves daytime symptoms and polysomnographic indexes in children with OSAS is repositioning of the jaw. In a six-month trial, a personalized oral jaw-positioning appliance proved clinically useful and was well tolerated in children with malocclusion [11].

Abnormal mandibular development and malocclusion can involve the jaws as well as the skeletal
structures of the respiratory dynamic space [12,13]. Children with habitual snoring and OSAS have a special cranio-facial morphology [8,9]. The craniofacial dysmorphism that leads to OSAS may involve delayed growth of the mandible, producing the mandibular retroposition commonly found in patients with OSAS. Mandibular retroposition is also associated with posterior displacement of the tongue base [14]. This abnormality narrows the upper airway, predisposing it to collapse and contributing to the development of OSAS [15]. Another common abnormality in patients with OSAS is a high-arched (ogival) palate by which posterior tongue displacement may force the lateral palatine processes to expand over the abnormally placed tongue before fusing at the midline [14].

A study investigating the treatment of OSAS with a rapid maxillary expander (RME) reported that in 9 of the 10 patients maxillary expansion reduced symptoms [16]. An RME was shown to be valid treatment for OSAS in children without enlarged tonsils and adenoids [17], and maxillomandibular expansion was successful also in adult OSAS [18].

Orthodontists today play an important role in the management of snoring and OSAS with the increasing use of oral mandibular advancement appliances and rapid maxillary expansion, but available data come mainly from studies in adults [16,18] in whom bone changes are slower than those noted in children and not always attainable without recourse to an invasive surgical approach.

The possibility that early usage of rapid maxillary expansion could not only improve the symptoms associated with snoring, OSAS and abnormal respiratory effort, but could also change the natural history of OSAS is an intriguing idea that deserves further investigation.

No studies are currently available on the long-term efficacy of treatment with an RME in children or in children with adenotonsillar hypertrophy.

In this open trial we assessed at 12-month follow-up the effectiveness of an RME as an early orthodontic treatment for OSAS in young children with dental malocclusion.

2. Methods

The study subjects were children between 4 and 11 years old who were referred from our Pediatric Sleep Center (Rome, Italy) and met the following three inclusion criteria: clinical signs of malocclusion (all presented with a high, narrow (ogival) palate associated with deep bite, retrusive bite or crossbite); signs and symptoms of OSA, including habitual snoring, apneas, and restless sleep witnessed by parents; and obstructive apnea/hypopnea index >1 proven by laboratory polysomnography and whose parents refused adenotonsillectomy. Children were enrolled between November 2004 and April 2005. Informed consent was obtained from the parents of each child, and assent was obtained from children >6 years old. Children were excluded if they had any of the following conditions: obesity (BMI ≥ 85 percentile) [19], acute or chronic cardiorespiratory or neuromuscular diseases, dysmorphism, major craniofacial abnormalities or associated chromosomal syndromes.

2.1. Study design

After recruitment, all participants underwent a detailed personal and family history and general clinical examination and had an oto-rhino-laryngologic and orthodontic assessment. The parents also completed a questionnaire, and children underwent overnight polysomnography.

2.2. Questionnaire data

Parents of participants completed a modified version of the Brouillette questionnaire on symptoms of OSAS [20]. The questionnaire elicited information on daytime symptoms (including sleepiness, irritability, headache, school problems, tiredness and oral breathing) and nighttime symptoms (including habitual snoring, apneas, restless sleep and nightmares).

A clinical score was assigned for each nocturnal and daytime symptom, with one point per question, yielding a maximum total score of 10.

2.3. Polysomnography

Standard overnight polysomnographic recordings were performed on a Grass Heritage polygraph. Variables recorded included a 16-channel electroencephalogram (EEG), electro-oculogram (EOG), submental electromyogram (EMG) and electrocardiogram (ECG). Chest and abdomen movements were measured by inductive plethysmography. Oronasal airflow was recorded with a thermocouple. Arterial oxygen saturation was monitored with a pulse oximeter.

Polysomnography variables were scored according to the criteria of Rechtschaffen and Kales [21] and the Italian Guideline [22].

The apnea-hypopnea index (AHI) was defined as the average number of apneas and hypopneas with or without desaturation per hour of sleep, lasting >5 s [22]. Other variables calculated were the obstructive hypopnea index (OHI), arousal index, obstructive apnea index (OAI), mean SaO2 value, and mean duration of rapid eye movement (REM) and non-rapid eye movement (NREM) sleep [22].

2.4. Oto-rhino-laryngologic assessment

Before orthodontic assessment children underwent an oto-rhino-laryngologic assessment to grade tonsillar
hypertrophy according to a scale ranging from 1 to 4 [23]: tonsils hidden within the pillars (size 1), tonsils extending to the pillars (size 2), tonsils are beyond the pillars but not to the midline (size 3), and tonsils extend to the midline (size 4).

2.5. Orthodontic assessment and orthopedic therapy

The orthodontic assessment detected the presence of jaw deviation from normal occlusion: deep bite, retrusive bite and cross-bite. Based on the results of this evaluation children had an endo-oral RME device placed. The device was a fixed, two-band RME appliance with an expansion screw fitted to the second deciduous molars of the upper jaw (Leone Sesto Fiorentino-Florence). The screw was turned two turns a day for the first 10 days until the palatal cusp of the upper molar came into contact with the buccal cusp of the lower molar. After this first treatment phase, when the maxillary arch was sufficiently over-expanded, the device was assembled with two round stainless steel wires (arms), soldered to bands placed on the second primary molars. The RME was removed after 12 months. All patients underwent monthly follow-up assessments until the trial ended.

The diameter of the upper dental arch was calculated as the distance between the two deciduous canine cuspid of the upper arch and the distance between the second deciduous molars of the upper arch. Variables were measured from plaster casts.

2.6. Statistical analysis

Data are expressed as means ± SD (standard deviation). The \( \chi^2 \) test was used to analyze the questionnaire responses. An analysis of variance (ANOVA: Friedman test) was used to assess significant differences between measurements obtained at each step against measurements at baseline. The Mann-Whitney test was used to calculate \( p \) values for differences between cross-bite and deep/retrusive bite. The SPSS statistical software program (SPSS 10.0, Chicago, III) was used for calculations. \( P \) values less than 0.05 were considered statistically significant.

3. Results

Out of the 260 outpatients who underwent polysomnography during the study period, 35 were eligible for recruitment, 16 of whom (mean age 6.9 ± 2.2 years, range 4.5–10.5; 9 boys) consented to participate in the study. All participants had normal height and weight for their age and none were obese.

Of these 16 children selected for RME treatment 14 completed the therapeutic trial; two children (12.5%) were excluded, one because weight increased (BMI from 19.59 to 26.7 kg/m\(^2\)) and the other because severe bronchial asthma developed, necessitating prolonged medical therapy.

According to questionnaire responses, 11/14 subjects (78.5%) who completed the therapeutic trial were chronic, heavy snorers and 12 of the 14 had obstructive sleep apneas (Table 1). The mean clinical score for the severity of OSAS improved significantly in treated subjects (Fig. 1). After the 12-month trial, the clinical score in 13/14 (92.8%) improved (a fall of at least two points in the score) and in 10/14 patients (71.4%) regressed (a fall of 50% in the score). Polysomnographic recordings (Table 2) showed that the AHI measured when treatment ended diminished significantly from baseline (5.8 ± 6.8 vs 1.5 ± 1.6; \( p = 0.005 \)). Similarly, the OHI improved significantly (3.1 ± 3.2 vs 0.9 ± 1.3; \( p = 0.002 \)), as did the arousal index (17.2 ± 3.5 vs 9.2 ± 1.6; \( p = 0.001 \)). None of the other measured polysomnographic variables changed significantly.

<table>
<thead>
<tr>
<th>Table 1</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Questionnaire answers of treated subjects before and after the trial</strong>^a^</td>
</tr>
<tr>
<td><strong>Nighttime symptoms</strong></td>
</tr>
<tr>
<td>Habitual snoring</td>
</tr>
<tr>
<td>Apneas</td>
</tr>
<tr>
<td>Restless sleep</td>
</tr>
<tr>
<td>Nightmares</td>
</tr>
<tr>
<td><strong>Daytime symptoms</strong></td>
</tr>
<tr>
<td>Sleepiness</td>
</tr>
<tr>
<td>Irritability</td>
</tr>
<tr>
<td>Headache</td>
</tr>
<tr>
<td>School problems</td>
</tr>
<tr>
<td>Tiredness</td>
</tr>
<tr>
<td>Oral Breathing</td>
</tr>
</tbody>
</table>

\(^a\) Data are expressed as a percentage of total category subjects treated.

\(^b\) \( \chi^2 \) test significance.
The AHI changed differently according to the type of malocclusion, diminishing more in subjects with deep and retrusive bite than in those with crossbite (6.7 ± 8.4 vs 1.0 ± 1.0, \( p = 0.03 \); and 4.3 ± 2.1 vs 2.5 ± 2.2, \( p = 0.04 \)) (Fig. 2). In two subjects (14.3%), treatment with the RME left the AHI unchanged. The first subject, a boy who had undergone adenoidectomy and had a right-convex deviation of the nasal septum with a monolateral crossbite, had a pre-post respiratory disturbance index (RDI) of 1.4 vs 1.6 events/h; the second, a boy who had recurrent upper airway infections with concomitant semi-obstructing tonsillar hypertrophy, a reduced epipharyngeal airway lumen caused by adenoidal vegetations, and deep bite, had a pre-treatment AHI of 1.5 and a post-treatment AHI of 3.2 events/h.

The oto-rhino-laryngologic examination before treatment detected mild tonsillar hypertrophy (clinical scale +2) in 35.7% (5/14), (clinical scale +3) in 35.7% (5/14), and severe hypertrophy (+4) in 7.1% (1/14) of the cases; only three patients (21.4%) had no tonsillar hypertrophy. Three subjects (21.4%) had adenoidectomy two years before the study. In patients with mild tonsillar hypertrophy (clinical scale 0 to +2), the mean AHI before orthodontic treatment was 5.6 events/h, and after 12 months of treatment decreased to 1.0 events/h \(( p = 0.034 \text{ by ANOVA})\), whereas in patients with severe tonsillar hypertrophy (grades +3 and +4) the mean AHI decreased from 6.2 events/h before treatment to 2.3 events/h thereafter \(( p = \text{ns})\).

All patients, in association with a high narrow palate, had occlusal anomalies: five subjects (35.7%) had crossbite, and nine subjects (64.3%) deep or retrusive bite or both abnormalities. The mean maxillary expansion achieved was 3.7 ± 0.7 mm for the intercanine diameter and 5.0 ± 2.2 mm for the inter-premolar diameter. No significant difference was found in the intercanine and inter-premolar diameters measured before and after therapy with RME in patients with deep or retrusive bite versus those with crossbite (before: 29.5 and 33.0 mm; after: 40.2 and 45.4 mm vs before: 29.5 and 33.1 mm; after: 39.0 and 44.6 mm).

In 3 of the 16 children (18.75%), the device broke but was repositioned within a week. No child experienced adverse effects when RME was applied.

Table 2
Polysomnographic (PSG) indexes at baseline and after 6 and 12 months of orthodontic treatment\(^a\)

<table>
<thead>
<tr>
<th></th>
<th>PSG baseline</th>
<th>PSG at 6 months</th>
<th>PSG 12 months</th>
<th>( p ) Value(^b)</th>
</tr>
</thead>
<tbody>
<tr>
<td>AHI (no/h)</td>
<td>5.8 ± 6.8</td>
<td>2.7 ± 3.5</td>
<td>1.5 ± 1.6</td>
<td>0.005</td>
</tr>
<tr>
<td>OAI (no/h)</td>
<td>2.5 ± 3.8</td>
<td>1.1 ± 2.5</td>
<td>0.6 ± 0.6</td>
<td>NS</td>
</tr>
<tr>
<td>OHI (no/h)</td>
<td>3.1 ± 3.2</td>
<td>1.5 ± 1.3</td>
<td>0.9 ± 1.3</td>
<td>0.002</td>
</tr>
<tr>
<td>Mean SaO(_2) (no/h)</td>
<td>96.0 ± 1.8</td>
<td>96.5 ± 2.3</td>
<td>96.7 ± 2.4</td>
<td>NS</td>
</tr>
<tr>
<td>ARI (no/h)</td>
<td>17.2 ± 3.5</td>
<td>12.1 ± 2.6</td>
<td>9.2 ± 1.6</td>
<td>0.001</td>
</tr>
<tr>
<td>REM (%)</td>
<td>17.2 ± 6.0</td>
<td>19.7 ± 7.1</td>
<td>22.8 ± 5.0</td>
<td>NS</td>
</tr>
<tr>
<td>NREM (%)</td>
<td>82.8 ± 5.9</td>
<td>80.3 ± 7.1</td>
<td>77.1 ± 5.0</td>
<td>NS</td>
</tr>
</tbody>
</table>

Definition of abbreviations: AHI, apnea-hypopnea index; OAI, obstructive apnea index; OHI, obstructive hypopnea index; SaO\(_2\), oxygen saturation; ARI, arousal index; REM, rapid eye movement sleep; NS, not significant.

\(^a\) Data are expressed as means ± standard deviation.

\(^b\) ANOVA Friedman test.
4. Discussion

Early orthodontic treatment with an RME in most children reduced the symptoms of OSAS, improved polysomnographic variables and attained excellent tolerability. In 10 of the 14 patients who completed treatment (71.4%) the symptoms of OSAS regressed and in 11/14 (78.5%) of treated patients the AHI significantly decreased. These promising findings suggest that RME devices should have a useful role in the therapy of OSAS in children.

Our findings in this trial confirm the long-term usefulness of an RME appliance in children with dental malocclusion and OSAS. In an earlier trial in patients with dental malocclusion [11] we found that after six months an oral jaw-positioning appliance reduced the nighttime and daytime respiratory symptoms and the AHI in 64.2% of the patients treated. In this second 12-month trial, we used an endo-oral orthodontic orthopedic device that acts on the maxillary bone structure. The technique consists of applying orthopedic force to the midpalatal suture. This area is mainly composed of compact bone laterally and fibrous tissue with collagen fibers, fibroblasts and blood vessels centrally [24,25]. RME widens the maxillary bone by distraction osteogenesis. Bone distraction at suture level widens the maxilla, increasing the cross-section as well as the volumetric space of the nasal cavity. RME and associated orthodontic repositioning can also indirectly improve the oropharyngeal space by modifying the resting posture of the tongue [26–28].

With one exception, all the children we studied were oral breathers. In nearly all of them, oral breathing disappeared after therapy with an RME. Although we did not measure nasal resistance, we noted that after 12 months of treatment in 13/14 children (92.8%) who were initially oral breathers, in only 2 (14.3%) did mouth breathing persist. The return of nasal breathing in these 11 successfully treated children suggests that the nasal cavity re-opened. In line with these findings, Hershey et al. noted that RME reduces nasal resistance by 45% and effectively widens the nasal passage [29]. Similarly, Bicakci et al. [30] showed that soon after RME in pubertal and prepubertal children, the nasal minimum cross-sectional area of the nasal cavity significantly increased. In another study, Kurol et al. [31] also reported improvement in nasal resistance in 10 prospectively studied children, 8 to 13 years old, treated with RME.

Under physiological conditions, the nose accounts for 50% of respiratory resistance; nasal obstruction related to anatomical-structural and functional causes is an important risk factor for OSAS [32]. Reducing nasal resistance is, therefore, one of the main objectives of therapy with an RME. By widening the palatine arch, an RME increases the volume of the nasal and buccal cavities thus helping dynamically to reduce the pharyngeal obstruction by repositioning the tongue so that it can regain its proper collocation within the buccal space, allowing the proper swallowing movements to return. In several studies [33–35], with rhinometry Timms documented well the subjective and objective improvement in nasal resistance in 10- to 20-year-old subjects after RME.

Most of the children we studied (11/14, 78.5%) had enlarged tonsils and were chronic snorers. After wearing the RME, their daytime and nighttime respiratory symptoms nevertheless diminished and AHI significantly improved. Widening the buccal cavity and distending the maxillary bone could reasonably enlarge the space available for the adenoids and tonsils so that they occlude the cavity less [36]. In our earlier study we found that jaw-repositioning reduced the degree of subjective tonsillar hypertrophy [11]. These observations suggest that increasing the oropharyngeal space induces a “relative” reduction in tonsillar hypertrophy.

The increased upper palatal arch diameters after treatment and the fact that all the children we studied withstood the maxillary expansion without experiencing adverse reactions show that the distraction device we used induced well-tolerated facial orthogenesis. In a study conducted on a small group of adult patients, Guilleminault et al., suggested that surgical maxillomandibular expansion improves sleep-disordered breathing in patients with maxillary and mandibular constriction [18]. One of the limitations of distraction osteogenesis is that the distraction devices have to be left in place for two to three months, until the newly generated bone is sufficiently matured. In addition, the need to wear an orthodontic device deters most adult patients from undergoing treatment [18].

In this study, we obtained therapeutic success without recourse to invasive procedures or risks of adverse effects, by beginning treatment early when the bone is still extremely plastic and growth is maximum. Dynamically widening the maxillary bone could induce a change in the natural history of OSAS, especially if treatment is started early. Even though children with deep/retrusive bite and crossbite had a similar increase in interpemolar diameters (in millimeters) in the upper jaw, children with deep/retrusive bite had a greater improvement in symptoms and in polysomnographic variables. This difference might imply that in children with deep or retrusive bite, orthodontic treatment exerts its beneficial effects also by repositioning the tongue and thus improving swallowing. Proper swallowing favors a return of normal lingual tone which reduces the hypotonia that causes the tongue to fall back during the hypotonic stage of sleep [37].

These long-term findings suggest that orthodontic treatment in children with dental malocclusion and OSA should be started as early as possible during
infancy. In as many as 14.5% of patients who undergo adenotonsillectomy, the symptoms of OSA are still seen three months after surgery [8]. Given that adenotonsillectomy will have only a limited and immediate effect on craniofacial morphology [36], orthodontic treatment could be a valid option in patients with recurrent OSAS after surgical treatment. Future studies may clarify whether orthodontic therapy might also reduce the symptoms of OSAS in patients with structural and dental anomalies, who have undergone adenotonsillectomy and whose symptoms of OSAS persist. The two therapies combined might synergistically offer a new fruitful area for research.

In our patients we found no correlation between tonsillar hypertrophy and the severity of sleep-disordered breathing. Children with high clinical grades of tonsillar hypertrophy and those with low grades had similar AHI values. This finding underlines the concept that tonsillar hypertrophy is not the only risk factor for OSAS, especially considering that therapy with an RME improved respiratory symptoms also in children with severe tonsillar hypertrophy. In this study, subjective tonsillar hypertrophy diminished in 50% of the children who completed the RME trial. Enlarging the retro-pharyngeal space and widening the maxillary bone could enlarge the space sufficiently at the stomatognathic level so that the tonsils appear smaller.

An unavoidable limitation of our study is that we had no control group. It is ethically difficult to keep children with OSAS without treatment for 12 months. Open questions needing further study pertain to which skeletal malformations combined might synergistically offer a new fruitful model for the obstructive sleep apnea syndrome. Ann Intern Med 1997;127(8 Pt 1):581–7.


Our findings in this 12-month open clinical trial suggest that young children with OSA may benefit from early orthodontic therapy with an RME. Further studies are warranted to investigate whether children with tonsillar hypertrophy and dental malocclusion might also be suitable candidates.

Our open trial indicates that orthodontists should systematically seek odontic anomalies in children, and then question parents on the presence of chronic snoring and other symptoms of OSA. In addition, the trial indicates that orthodontic treatment with an RME can bring clear benefits to children with OSA.

Our promising findings warrant controlled studies to confirm the value of RME in the therapy of OSAS and to investigate whether RME alone is as effective as RME plus adenotonsillectomy in improving long-term outcome.

References


