Forcierte Gaumennahterweiterung vor und nach Adenotonsillektomie bei Kindern mit obstruktiver Schlafapnoe

P. Pirelli · M. Saponara · C. Guilleminault
In children adenotonsillectomy (AT) is the recommended treatment in the presence of obstructive sleep apnea (OSA) [1]. It should be performed, if needed, in association with nasal inferior turbinate reduction usually with radiofrequency [2]. But as shown in previous reports, upper airway soft tissues treatment is not always successful in completely controlling abnormal breathing during sleep in children, despite the fact that subjective improvement is often reported [3, 4, 5, 6].

Recently, a randomized study performed in Caucasian prepubertal children with OSA mostly recruited from an otolaryngological practice showed that many children would benefit from both otolaryngological and orthodontic treatment; in that report, one out of all the children treated initially with orthodontics maneuvers did not need further otolaryngological treatment [7]. Our investigation looks further at this interaction between otolaryngological and orthodontic treatment in OSA. We hypothesized that orthodontic treatment alone, specifically rapid maxillary expansion (RME), may resolve OSA without the need for adenotonsillectomy.

Patient and methods

Study participants were recruited among children referred to our ear nose and throat (ENT) otorhinolaryngological and odontological ambulatory clinic during an 8-year period; all children were from the Rome (Italy) urban area. The selected group included 225 children (119 boys and 106 girls) ages 6–13 years (average 7.3 years).

Main inclusion criteria were the following: presence of clinical complaints usually noted in children with sleep-disordered breathing (SDB) and signs of mouth breathing, chronic snoring; adenotonsilar hypertrophy (2+ or 3+); OSA demonstrated at polygraphy with a minimum apnea–hypopnea index (AHI) of 1 and a minimum respiratory disturbance index (RDI) of 5 [8]; and narrow upper jaw at examination, diagnosed clinically and confirmed by cephalometric assessment according to Ricketts parameters [9]. Presence of other significant pathologies that could influence upper airway patency was an exclusion criteria.

The following cases were excluded: 41 children with vasomotor rhinitis, allergy and turbinate hypertrophy, 19 children with mandibular retrognathia, 63 children with associated obesity based on weight and height tables for age, 16 children with macroglossia due to medical causes, and 6 children with associated chronic illnesses. A total of 80 children (43 male and 37 female) with a body mass index (BMI) <24 kg/m² participated in the study. Protocol and informed consent were approved by the Tor-Vergata University ethics committee, Rome (Italy).

Group allocation

All children underwent the same baseline investigation; all had a clinical interview and pediatric evaluation, additional information on their prior health status was obtained from their pediatricians. Based on the current complaints, past medical history, and clinical evaluation, suspicion of the presence of obstructive sleep apnea led to polygraphy that demonstrated an abnormal amount of SDB events. Subjects then had simultaneous EN) and orthodontic evaluations including rhinoscopy and cephalometric X-rays.

Patients were subdivided into two groups based on results of this investigation:

Those with indicators of chronic adenotonsillar inflammatory problems—diagnosed based on clinical evaluation with history of chronic tonsillitis and lateral enlargement of tonsils based on Friedman scale [10]—were placed in the group to be initially treated with adenotonsillectomy (group II), while those not clearly presenting this ENT problem were placed in the initial orthodontic treatment group (group I). The goal was to recruit a similar number of subjects in both groups in order to simplify comparison of results. A total of 40 subjects per group was obtained.

Parents were informed that their child had two different morbidities and that both ENT and orthodontic treatment may be needed. In our community, standard of care for inflammatory tonsils is surgical.
excision, but children with enlarged but noninflammatory tonsils were referred for orthodontic treatment as a first step. For each treatment modality, patients were followed by the specialist responsible for application of the selected treatment. Independent of treatment order, all children underwent an evaluation of the effect of treatment and impact on OSA. This evaluation consisted of clinical interview and examination, polygraphic recording, and cephalometric X-rays analyses. A first assessment was made 4 months after end of the first treatment (T1).

In the AT group, patients with resolution of OSA did not enter the second phase of the study. For the remaining children, parents could decide to proceed with orthodontic treatment either within the research protocol or independent of any study schedule. Similarly in the RME group, children with complete resolution of SDB at T1 did not undergo follow-up AT surgery.

Patients who only had a partial benefit with orthodontics were referred to their pediatric ENT specialist, but some were not submitted to surgery, based on the unilateral decision of the specialist who felt that the polygraphic findings at T1 were too mild to justify adenotonsillectomy [11]. Thus, in this group the second treatment was reserved only to subjects who clearly had persistence of problems at T1.

Procedures
Orthodontic evaluation
All children were examined by an orthodontist with a large experience in pediatric facial growth and in abnormal breathing awake and asleep. The orthodontist evaluated each patient on:

- facial thirds:
  - The face is normally divided in three thirds of equal length. A flattening of the area of the face located between the superior orbital margin (superior limit) and the occlusal plane (inferior limit) can be observed with abnormal growth. This clinical finding can be evaluated in relation to the “aesthetical analysis of the human profile” based on the “Holyday line of harmony” (H-line) which is defined as the distance from the deepest point of sulcus at the upper lip and the most prominent point of the chin (ideally −5±−2 mm).
  - The presence/absence of labial incompetence defined as absence of contact between upper and lower lips when at rest position with presence/absence of hypotonia of upper lip manifested by an open mouth with a tended upper lip, resulting in an increase of the nasal labial angle (angle formed by the labial surface of the upper lip at the midline and the inferior border of the nose (normally 85–90°) with a tender upper lip increasing this nasal-labial angle.
  - The presence/absence of mouthbreather facies.
  - intra-oral evaluation showed presence/absence of anterior cross-bite; presence/absence of an ogival shape of the upper maxillary arch (the result from a high palatal vault and a narrow maxillary arch), related to a contraction of the upper jaw at its base.
  - cephalometric assessment: the narrowness of the upper jaw was diagnosed clinically and confirmed by cephalometric assessment according to Rick-ets parameters on posteroanterior cephalograms [11]. The following measurements were obtained from the posteroanterior tele-radiograms (Fig. 1): to define a narrow maxilla, we assessed the maxillary cross-section studying the distance of JR and JL (bilateral points located at the depth of the concavity of the lateral maxillary contour at the junction of the maxilla and the zygomatic buttress; Fig. 1); and from the frontofacial plane, the values ZR-AG and ZL-AG (normal value at 8 years of age =10±1.5 mm, and until 14 years =8±1 mm) with a larger distance indicating a narrow upper jaw. We also assessed the nasal cavity width NC-CN (bilateral points located at the larger cross section of the nasal cavities with normal value at 9 years of age =25.2±2 mm, and +0.6±1 mm/year), with a shorter distance confirming the diagnosis of a narrow upper jaw (Fig. 2). In addition, the interincisive space A1-1A and the intermolar width A6-6A were measured.

The ENT evaluation, as mentioned above, was a comprehensive visual examination of upper airway including rhinopalatoscop[y, and cephalogram analyses indicating size and location of adenoids and tonsils.

The polygraphic recording was a nocturnal home study performed with a validated against polysomnography cardiorespiratory recorder, the SOMNOscreen™ plus RC (SOMNOmedics GmbH, Randersacker, Germany) [12], monitoring: body position, SpO₂, thoracic effort, abdominal effort, snore, pulse rate, pulse transit time (PTT), and oral–nasal flow. The software...
Abstract
Adenotonsillectomy is not always successful in controlling obstructive sleep apnea (OSA) in children, and orthodontic treatment may be a helpful adjunction and sometime an important alternative. A total of 80 nonoverweight children (37 girls) with a high and narrow hard palate, based on an indepth orthodontic evaluation, were subdivided into two groups based on presence/absence of chronic adenotonsillar inflammatory problems as determined by full otolaryngological examination, with recruitment of 40 children in each group. Patients with evidence of chronic inflammation were treated with adenotonsillectomy first (group II), while all other children had rapid maxillary expansion (RME) first (group I). In all children, clinical inter-
views and clinical evaluations, frontal and lateral cephalometry, and polysomnography were performed at entry. Four months after end of treatment, the initial evaluation was repeated in all children. Children incompletely treated were offered to cross-over to the other treatment venue. At entry, there was no significant difference between the monitored respiratory polygraphic variables between the two groups. At the 4-month follow-up, 15 subjects treated with RME were considered as cured compared to 6 patients after adenotonsillectomy, and absence of improvement was observed in 8 children with RME and 16 with adenotonsillectomy. After cross-over to the other treatment, involving 42 subjects due to 15 drop-outs between treatment 1 and 2, three children were still having residual sleep disordered breathing at the 12-month follow-up, while normal breathing during sleep was observed in all others. With appropriate clinical investigation, prepubertal children with OSA and narrow maxilla may have better treatment outcome when treated with orthodontics than with adenotonsillectomy; and polysomnographic or even better polysomnographic documentation of clinical impression is always needed posttreatment.

Keywords
Adenotonsillectomy · Rapid maxillary expansion · Prepubertal children · Obstructive sleep apnea · Treatment

Forcierte Gaumennahterweiterung vor und nach Adenotonsillektomie bei Kindern mit obstruktiver Schlafapnoe

Zusammenfassung

Schlüsselwörter
Adenotonsillektomie · Forcierte Gaumennahterweiterung · Präpubertäre Kinder · Obstruktive Schlafapnoe · Therapie
Rapid maxillary expansion (RME) was performed, as previously reported [13, 14], applying orthopedic forces on the midpalatal sutures using the first molars and permanent premolars as anchor teeth; while in deciduous dentition, the second primary molars were selected as long as they could provide the required firmness. The device was comprised a central expansion screw with four arms: two front arms and two back arms, anchored to the selected anchor teeth. The bone distraction at the suture level enables an effective enlargement of the maxillary skeletal base. Enlargement was visually appreciable as the bone distraction leads to an interincisive space—a diastema—and with X-rays as the gain appears as a radiotransparency corresponding to the visually seen space. The procedure usually lasts 3 weeks with daily turning of a midline screw that allows enlargement (distraction) of the space at the level of the midline suture: the transpalatal force, which exceeds the orthodontic force, produces an orthopedic force that opens the midpalatal suture leading to maxillary movement without tipping of the teeth. Once the needed extension was obtained (end of the activation phase), the midline screw was blocked and the device was kept in place at least 3–4 months more in order to allow the newly formed bone to strengthen.

Prior to distraction, child and parents had been briefed on daily care (turning the screw), diet, and oral hygiene in order to avoid any complications, which could interfere with the expansion [14, 15]. Based on the screw activation system that we used, the following schedule was followed: day 1 (morning and evening) three consecutive activations at 10 min intervals, and from day 2 onward, one activation every morning and evening; one activation consists of one turn of the screw, i.e., 0.25 mm.

All modifications induced by RME on the upper jaw and adjacent anatomical structures were studied with posteroanterior cephalometric evaluation at T0, T1, and T2 and with CT Dentascan at T0 and T1. The Ricketts parameters were adopted for posteroanterior cephalometric evaluation (Fig. 1, 2).

**Adenotonsillectomy**

Patients were instructed to avoid heavy exercise and to remain at home for approximately 1 week.
Analysis

At each evaluation, each child was seen by both the ENT and orthodontist team. Polygraphic recordings were analyzed following the recommendation of the American Academy of Sleep Medicine (AASM) and apnea and hypopnea were scored as recommended in the atlas [8].

Statistical analyses

The t test for repeated measure was used to compare groups at different stages of treatment. The Kruskal–Wallis analysis of variance (ANOVA) was performed when comparing several groups with post hoc analysis, if the ANOVA was significant. The χ² statistics were used to compare percentages.

Results

During the treatment period (Fig. 3), no complications were observed in either group.

Polygraphic results

Entry (T0)

At T0 there was no difference between the two groups in demographics and polygraphic variables (see Table 1). For both treatment groups, the mean AHI was 12.8 events/h, which is considered to be in the “moderate” range. There was no significant difference in age, BMI, AHI, and lowest oxygen saturation between the two groups at entry (T0).

Initial posttreatment evaluation (T1)

In the RME group (group I), 15 subjects (37.5% of subjects) had normal clinical evaluation and normal polygraphy at the initial posttreatment evaluation (T1) 4 months after completion of treatment; 17 presented a significant improvement (AHI 6.5 ±3.1) and 8 minimal or no improvement (AHI 13±3.5; ANOVA p=0.05).

Progression to the second treatment phase (T2)

From this initial orthodontic treatment group (group I), AT was performed only on the 8 patients who show only minimal or no improvement. The 17 children with incomplete improvement were deemed to have a too mild sleep disordered breathing problem by local standard of practice and were dropped out of the protocol.

These results contrasted with the AT group (group II) where only 6 patients (15% of subjects) presented total remission, 18 presented an improvement of OSA (AHI 6±3.1) and 16 minimal or no improvement (AHI 15±2.9; ANOVA p=0.05). The second treatment with RME was performed on all 34 patients who did not have resolution of their OSA with AT treatment.

Only 42 children were submitted to the second phase of treatment. This was related not solely to the complete absence of clinical and polygraphic findings noted in 21 children, but to the decision made by the ENT to not pursue treatment in 17 cases, despite initial scoring of tonsils as 2+ or 3+. Widening of the palatal vault secondary to orthodontic treatment changes scoring of lateral position of the tonsils, but the AHI score in the dropout group was still abnormal (Fig. 3).

Evaluation after the second treatment phase (T2)

The evaluation at T2 demonstrated the efficacy of the combined treatment procedures: there was complete remission of symptoms in most patients and normalization of AHI and nocturnal SpO₂ with a minimum SpO₂>96% for the total group. However, three cases (2 patients in the initial RME group and 1 patient in the initial AT group) presented incomplete positive results at the end of both treatments with scores of 2.9, 3.2, and 3.8 events/h, respectively. However, parents reported overall clear improvement and great abatement of clinical symptoms.

Orthopedic/orthodontic results

All modifications induced by RME on the upper jaw and adjacent anatomical structures were studied with posteroanterior cephalometric evaluation at T0, T1, and T2 and with CT Dentascan at T0 and T1. The Ricketts parameters were adopted for posteroanterior cephalometric evaluation (Fig. 1).
In all our treated cases, an opening of the midpalatal suture was obtained with the results being confirmed by intraoral occlusal radiographs (Fig. 4) and CT Dentascan (Fig. 5). The objective assessment of maxillary width shows an increment, confirming that the RME maneuver directly influences the skeleton. Through the expansion of the midpalatal suture, RME produces an expansion that occurs not only in the maxillary arch but also in the nasal cavities (Fig. 6). When considering the whole RME-treated group (n=74), the expansion of the midpalatal suture is responsible for the expansion of both maxilllas, with an average cross-sectional increase (IL-JR) of 5.91±0.7 mm. The analysis of the upper intermolar distance (A6-6A) shows an average increase of 8.18±0.3 mm. The interincisive space (A1-1A), typical of midpalatal suture opening, was noted in all treated cases, with an average opening of 4.72±0.2 mm.

The increase in maxillary cross section is also shown by the investigation of the nasal cavities width (NC-CN) that is widened by the RME treatment with an average increase of the pyriform opening of 3.85±0.3 mm. The presence of an important and isolated maxilla narrowing with an important cross-bite was, in our series, the best predictor for important clinical and polygraphic gains post-RME.

**Discussion**

Adenoidian facies was already well described in the 19th century, and between 1960 and 1980, Swedish and UK orthodontists emphasized the role of mouth breathing and enlarged tonsils and adenoid in cranial facial changes [16, 17, 18, 19, 20, 21, 22, 23, 24, 25, 26]. An animal model of the infant monkey demonstrated the important role of abnormal nasal resistance during the developmental period and its consequences leading to the narrowing of dental arches, decrease in maxillary arch length, anterior cross bite, maxillary overjet and increase in anterior face height [27]. The interplay between nasal and oral breathing is well described in the literature, but rarely investigated in clinical practice. However, this interaction has important consequences on the development of the upper airway in children [18, 19, 24] and it can promote the development of obstructive sleep apnea syndrome.

Orthodontic treatment has been shown to improve abnormal breathing related to these developmental changes. Some of these previous reports present typical cases of obstructive sleep apnea in children but without having polygraphic demonstration of SDB [20, 21, 22, 25, 26]. Investigation of the role of orthodontics in nonsyndromic OSA children has been more systematic in recent times [6, 7, 14, 15, 28]. But the first line of treatment for

![Fig. 4](image-url) Intraoral occlusal radiographs before and after opening of the midpalatal suture

![Fig. 5](image-url) CT Dentascan before and after opening of the midpalatal suture
many children with OSA is still AT. Several studies, however, have shown that many children subjected to AT surgery present residual SDB [3, 4, 5, 7, 29, 30, 31, 32]. The addition of orthodontic treatment in these children may help to further reduce the residual AHI, but many questions remain unresolved, i.e., role of orthodontics in the treatment of OSA and its placement compared to adenotonsillectomy.

In this study, we found greater effectiveness of RME as an initial treatment parameter: a significantly larger number of patients improved with RME alone, and there were fewer patients in the RME group who crossed-over to the second arm of treatment. We acknowledge that this was not a randomized study and that patients with greater chance at clinical inspection to respond to one type of treatment than the other were placed in group I or II. However, selection of patients to have adenotonsillectomy first was not demonstrative of success. The results of our study let us affirm that, in many cases, the functional improvement observed is clearly related to the skeletal expansion caused by the maneuver of RME: the widening of the nasal fossa (Fig. 4) and septal release can restore normal airflow. This anatomical change leads to an increased patency of the upper airways and, as a consequence, to a clear improvement of SDB.

When the AHI distribution at entry was examined, independent of the group allocation, subjects with a higher AHI were more commonly in the "non-cure" group at T1. If we consider all the subjects who were denied the second phase of treatment as "failure" of both treatments, it would mean that 20 out of 80 children failed both treatment, i.e., 25% of the studied children. On the other hand, if we consider only the 3 children who failed after undergoing both treatments and do not include the 17 children who were denied adenotonsillectomy surgery, the failure rate would be 4.8%.

Our investigation also represents a study of beliefs and clinical practices. The decision not to perform AT as a second step indicates a dilemma faced by sleep physicians: many surgeons will easily perform surgery on infectious tonsils, a clear-cut clinical presentation. But, as in the cases above, the surgical indication in the 17 children was related to a different ideology: tonsils were not "infectious" but were occupying space and had an impact on breathing during sleep with only abnormal results at a nocturnal test. Despite initial agreement to have children failing RME to undergo AT; 17 children were denied this procedure. Such a decision by treating community of ENTs was based on the absence of knowledge that presence of adenotonsils may be associated with persistence of mouth breathing and within a few years, disappearance of the beneficial effect of RME with abnormal maxillary growth and re-occurrence of SDB [33].

Several other points must be emphasized:
- Our study was performed with a validated and commonly used home polygraphic recorder. Such home-recording devices are more and more commonly used considering cost-effectiveness. Ambulatory monitoring does not mean necessarily low diagnostic capabilities as some ambulatory devices have even more capabilities than the common in-laboratory sleep systems. But there will always be the absence of the continuous surveillance that the in-laboratory studies provides. It is a limitation of our study. But home studies allow more frequent follow-up of treatment procedures, larger numbers of subjects investigated, more acceptable procedure for parents due to a lower perceived discomfort, and less expensive investigation. We acknowledge, however, that in-laboratory polysomnography may have provided slightly different results at any of the three different studies, but the fact that same methodology was used at each testing period allows valid between-test comparisons.
- Imaging is helpful to investigate children with SDB. At least frontal and lateral cephalometric X-rays should be obtained. Depending on the number of cephalograms obtained, the amount of irradiation related to these tests, which is always a concern in children, must be considered, and newest techniques usually give better results with lower radiation exposure. Usage of a more advance technique such as the CT Dentascan may be considered: this is the most reliable test verifying the extension of the maxillary cross section. Indeed, several studies investigating the improvement of upper dental arch width on cat models showed that final data can be influenced by dental tipping, thus demonstrating a false increase which does not correspond to real skeletal changes and such a tipping may occur in children.
- Depending on the setting in which a child is evaluated, one may expect that the primary complaint may be different, and searching for symptoms confirming OSA may require extra effort.
In addition, the clinical focus may vary based on the dominant anatomical problem being evaluated. For example, large tonsils may mask the involvement of the skeletal structures, or skeletal structures may mask the role of soft tissues. Our study shows that both treatments have beneficial effects, and that often both will be needed. The challenge will be to recognize children where only one treatment may be needed. Systematic evaluation by both orthodontists and ENTs may assist in deciding the first step to take. Imaging with at least frontal and lateral cephalometric X-rays should be obtained.

Finally, despite its limitations, this study demonstrates the importance of a multidisciplinary approach involving orthodontists at the same level as ENT specialists in the diagnosis and treatment of SDB in children. Finally, patients failing treatment with AT and RME should be recognized and closely followed, as they could benefit from an orthognathic surgery performed in the teenage years in order to avoid relapse of symptoms and permanent health problems in adulthood [28, 30].

**References**

9. Langlade M (1979) Cefalometria Ortodontica. Scienze, Firenze, Italy

**Corresponding address**

P. Pirelli
Department of Odontostomatological Sciences, University “Tor Vergata”
Rome
Italy
pirelli@gmail.com

**Conflict of interest.** On behalf of all authors, the corresponding author states that there are no conflicts of interest.