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Original Article

Rapid maxillary expansion outcomes in treatment of obstructive sleep apnea in children

Maria Pia Villa [a,*], Alessandra Rizzoli [a], Jole Rabasco [a], Ottavio Vitelli [a], Nicoletta Pietropaoli [a], Manuela Cecili [a], Alessandra Marino [b], Caterina Malagola [b]

[a] Neuroscience, Mental Health and Sense Organs Department, Paediatric Sleep Disorder Centre, Sant'Andrea Hospital, Faculty of Medicine and Psychology, "Sapienza" University, Rome, Italy
[b] Orthodontic Clinic Sant'Andrea Hospital, Faculty of Medicine and Psychology, "Sapienza" University, Rome, Italy

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ABSTRACT

Objectives: The objectives of this study were to confirm the efficacy of rapid maxillary expansion in children with moderate adenotonsillar hypertrophy in a larger sample and to evaluate retrospectively its long-term benefits in a group of children who underwent orthodontic treatment 10 years ago.
Methods: After general clinical examination and overnight polysomnography, all eligible children underwent cephalometric evaluation and started 12 months of therapy with rapid maxillary expansion. A new polysomnography was performed at the end of treatment (T1). Fourteen children underwent clinical evaluation and Brouilette questionnaire, 10 years after the end of treatment (T2).
Results: Forty patients were eligible for recruitment. At T1, 34/40 (85%) patients showed a decrease of apnea–hypopnea index (AHI) greater than 20% (ΔAHI 67.45% ± 25.73%) and were defined responders. Only 6/40 (15%) showed a decrease <20% of AHI at T1 and were defined as non-responders (ΔAHI –53.47% ± 61.57%). Moreover, 57.5% of patients presented residual OSA (AHI > 1 ev/h) after treatment. Disease duration was significantly lower (2.5 ± 1.4 years vs 4.8 ± 1.9 years, $p < 0.005$) and age at disease onset was higher in responder patients compared to non-responders (3.8 ± 1.5 years vs 2.3 ± 1.9 years, $p < 0.05$). Cephalometric variables showed an increase of cranial base angle in non-responder patients ($p < 0.05$).

Fourteen children (mean age 17.0 ± 1.9 years) who ended orthodontic treatment 10 years previously showed improvement of Brouilette score.
Conclusion: Starting an orthodontic treatment as early as symptoms appear is important in order to increase the efficacy of treatment. An integrated therapy is needed.

1. Introduction

Obstructive sleep apnea (OSA) is a sleep disordered breathing characterized by prolonged partial and/or intermittent collapse of the airway during sleep, that interrupts normal ventilation and normal sleep patterns, with a prevalence of 1% to 5.7% in children [1–3] .

OSA is a multifactorial disease where different risk factors such as craniofacial anomalies, adenotonsillar hypertrophy, obesity, alterations in upper airway neuromotor tone and airway inflammation can co-exist. These lead to a decrease in nasopharyngeal airway dimensions that promotes a wide spectrum of symptoms ranging from primary snoring, to upper airway resistance syndrome, to frank OSA [4,5].

Among all the causative factors, adenotonsillar hypertrophy is the most common cause of childhood OSA [6,7]. Since it is a surgical therapy, adenotonsillectomy (AT) is limited by surgical risks and, although it leads to significant improvements in respiratory indices, residual disease is present in a large proportion of children, especially if aged >7 years, where obesity, severe OSA before surgery and asthma are present [8]. Moreover, Guilleminault et al. reported in a recent study the recurrence of sleep respiratory symptoms on a cohort of OSA patients during adolescence, not depending on the standard therapies [9].

In addition to large tonsils and adenoids, children with OSA may present narrow upper airways deriving from narrow and long faces, maxillary constriction and/or high arched palates and/or some degree of mandibular retrusion [10–12]. However these orthodontic and craniofacial abnormalities in children with OSA have been widely ignored even if, in the last decades, correction of mandibular or maxillomandibular anomalies has been shown to improve OSA [13–18].

Rapid maxillary expansion (RME) is a dentofacial orthopedic treatment procedure commonly adopted in young patients for the

* Corresponding author. Sleep Disorder Centre, Sant'Andrea Hospital, Via di Grottarossa 1035-1039, 00189 Rome, Italy. Tel.: +39 0633775855; fax +39 0633775941.
E-mail address: mariapia.villa@uniroma1.it (M.P. Villa).

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treatment of constricted maxillary arches. Several studies have shown the short-term efficacy of orthodontic treatment with rapid maxillary expander with evidence of a significant improvement of OSA even in children with adenotonsillar hypertrophy [14,15,18].

Pirelli et al. [16] demonstrated that all 31 children studied, with upper jaw contraction, oral breathing, nocturnal snoring, and OSA, achieved a normal anterior rhinometry and an apnea–hypopnea index (AHI) < 1 event per hour after four months of treatment with RME. Our group has previously demonstrated in 14 children with dental malocclusion, a body mass index (BMI) <85 percentile, and OSA confirmed by polysomnography (PSG) a significant decrease in the AHI, hypopnea obstructive index and arousal index after 12 months of RME therapy [17]. Moreover questionnaires on daytime and night-time, fulfilled before and after treatment, showed significant decreases in the severity of symptoms.

Only few studies have investigated the long-term effects of orthodontic treatment in OSA by considering the growing and the skeletal changes occurring through the years [18]. Ten of the 14 children who completed our 12-month therapeutic trial using RME (see above) performed 24 months follow-up after the end of the RME orthodontic treatment. No significant changes in the AHI or in other variables were observed.

Previous papers regarding orthodontic treatment, associated or not with AT, studied small-size samples. For this reason, the primary aim of this prospective study was to confirm our previous findings [17] on the efficacy of RME in children with moderate adenotonsillar hypertrophy, with a larger sample. The second aim was to retrospectively evaluate any long-term benefit after onset of puberty in a group of children who underwent orthodontic treatment with RME 10 years ago.

2. Methods

Children between 4 and 10 years of age who had been referred to our Paediatric Sleep Center (Sant'Andrea Hospital, Rome, Italy) and satisfied the following inclusion criteria were included: clinical signs of malocclusion (high, narrow palate associated with deep bite, retrusive bite or crossbite); tonsillar grading I–III [19], signs and symptoms of OSA (including habitual snoring, apnea and restless sleep as witnessed by parents), AHI > 1 as defined by a laboratory PSG recording. All the participants' parents provided written informed consent to the study. The study procedures were approved by the hospital ethics committee.

We excluded patients with a history of previous treatment for OSA (including tonsillectomy, adenoidectomy and AT), severe tonsillar hypertrophy (grade IV), obesity (BMI value ≥95th centile [20]), genetic disorders, cerebral palsy, neuromuscular diseases, cardiac disease, renal disease any systemic diseases or chronic cardiorespiratory or neuromuscular diseases, dysmorphism, major craniofacial abnormalities or associated chromosomic syndrome.

2.1. Study design

The study design is shown in Fig. 1. After recruitment, all participants underwent a detailed personal and family history and general clinical examination and had an ear, nose and throat (ENT) and orthodontic assessment before overnight PSG (T0). Parents were asked when their child started to present daytime and night-time symptoms.

All children who met inclusion criteria underwent cephalometric evaluation and started 12 months of therapy with RME and performed a new polysomnographic assessment (T1). Disease duration was defined as the time between onset of symptoms and the beginning of the treatment. Parents fulfilled a questionnaire at T0 and T1.

Presence of daytime and night-time symptoms in children who had completed the 12-month therapeutic trial with RME [17] was investigated through a questionnaire and clinical evaluation, 10 years after the end of treatment (T2).

2.2. Questionnaire data

The participants' parents completed the previously validated Brouilette questionnaire [21], at T0, T1 and T2. The questionnaire elicited information on daytime symptoms of OSA (including sleepiness, irritability, headache, school problems, tiredness and oral breathing) and night-time symptoms (including habitual snoring, apneas, restless sleep, and nightmares).

2.3. Polysomnography

Standard overnight PSG recordings were obtained at baseline, before starting orthodontic treatment (T0) and after 12 months of treatment (T1) using a Grass heritage polygraph. The variables recorded included an electroencephalogram (EEG) with at least six channels (bilateral frontal, central temporal, and occipital monopolar

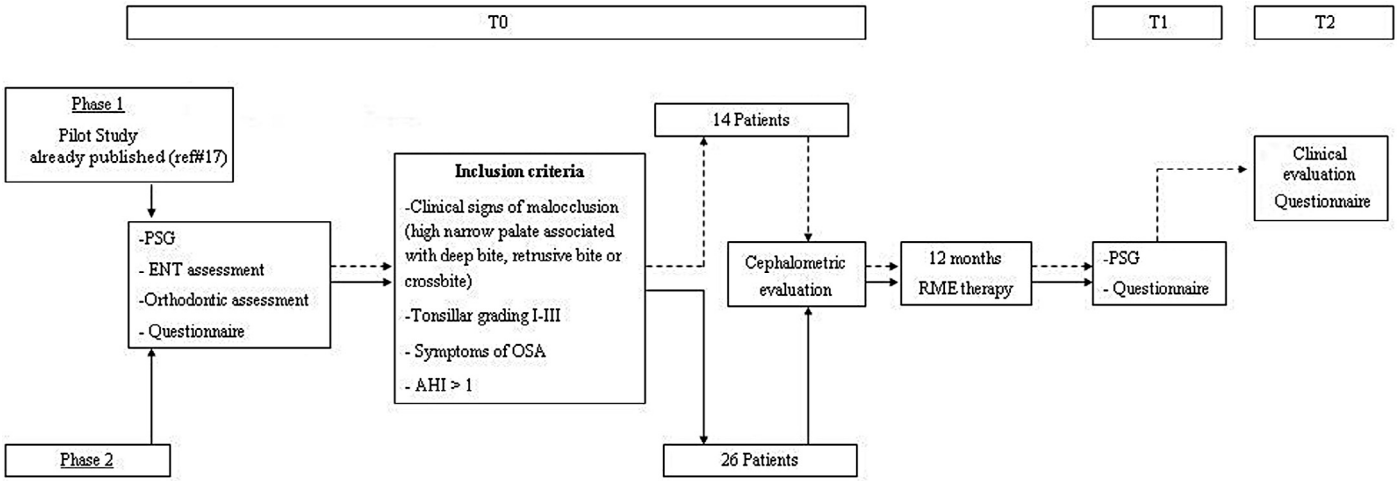

Fig. 1. Study design graph. ----→, pilot study ($N = 14$); →, new sample enrolment ($N = 26$). AHI, apnea–hypopnea index; ENT, ear, nose and throat; OSA, obstructive sleep apnea syndrome; PSG, polisomnography; RME, rapid maxillary expansion.

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