

CASE SERIES AND REPORTS

Effects of simultaneous palatal expansion and mandibular advancement in a child suffering from OSA

Effetti di simultanei espansione palatale e avanzamento mandibolare in un paziente pediatrico con apnee ostruttive notturne

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SUMMARY

This clinical report describes a child suffering from obstructive sleep apnoea (OSA) and class II skeletal malocclusion with maxillary contraction and anterior open bite. He presented moderate obstructive sleep apnoea with large impact on quality of life of patient and parents. He was treated using an innovative orthodontic device (Sleep Apnea Twin Expander) to simultaneously carry out palatal expansion and mandibular advancement. After orthodontic therapy, the OSA-18 questionnaire demonstrated an improvement of the main respiratory symptoms, while cardiorespiratory sleep study revealed a reduction in obstructive sleep apnoea events. Post-treatment, clinical assessment and cephalometric analysis showed a reduction of sagittal maxillary discrepancy and an extension of upper airway space. In conclusion, this case report suggests that orthodontic treatment might be a valuable alternative treatment in children with obstructive sleep apnoea related to craniofacial anomalies.

KEY WORDS: Obstructive sleep apnea • Palatal expansion • Mandibular advancement appliances

RIASSUNTO

Questo caso clinico illustra il trattamento di un bambino affetto da apnee ostruttive nel sonno (OSA) che presenta una malocclusione di classe II scheletrica da retrusione mandibolare con contrazione del mascellare superiore e morso aperto anteriore. Il paziente presenta apnee ostruttive del sonno di grado moderato con un alto impatto sulla qualità della vita del paziente e dei genitori. Il paziente è stato trattato utilizzando un dispositivo ortodontico innovativo (Sleep Apnea Twin Expander), al fine di realizzare l'espansione del palato e l'avanzamento mandibolare contemporaneamente. Dopo la terapia ortodontica, il questionario sulla qualità della vita ha evidenziato un miglioramento dei principali sintomi respiratori e lo studio cardiorespiratorio del sonno ha rivelato una riduzione degli eventi di apnee ostruttive. Al termine della terapia, la valutazione clinica e l'analisi cefalometrica hanno evidenziato una riduzione della discrepanza sagittale e verticale tra il mascellare superiore e la mandibola e un ampliamento dello spazio delle vie aeree superiori. In conclusione, questo case report suggerisce che il trattamento ortodontico può essere una valida terapia alternativa nei bambini con apnea ostruttiva del sonno associata ad anomalie cranio-facciali.

PAROLE CHIAVE: Apnee ostruttive notturne • Espansione palatale • Apparecchi di avanzamento mandibolare

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Introduction

Obstructive sleep apnoea (OSA) in children is a "sleep breathing disorder characterised by prolonged partial airway obstruction and/or intermittent complete obstruction that interrupts normal ventilation and normal sleep patterns"¹. OSA in children is associated with a series of daytime and night time symptoms, such as daytime sleepiness, morning headache, snoring, laboured breathing, restless sleep and nocturnal enuresis². Untreated OSA can result in serious cardiovascular, neurocognitive, behavioural and metabolic problems². The "gold standard" for the diagnosis of OSA is

polysomnography even if nocturnal cardiorespiratory sleep study (CRSS) represents a good alternative as an abbreviated diagnostic tool³. The main risk factors of OSA in children include adenotonsillar hypertrophy, obesity, neuromuscular disorders and craniofacial anomalies³. The first line therapy for children with OSA is adenotonsillectomy, but it is not universally effective due to multifactorial aetiology of the disease⁴. Some authors have suggested the presence of a strong association between sleep breathing disorders and mouth breathing, abnormal placement of the tongue and anomalies in orofacial anatomy⁵. The most common craniofacial anomalies reported are retrognathism, maxillary con-

traction usually associated with unilateral or bilateral cross bite and open bite⁶. The potential benefit of an orthodontic approach in patients with OSA and malocclusion was previously described by several authors⁷⁻⁹. We report our experience on treatment of paediatric OSA with an innovative oral device that we called the “Sleep Apnea Twin Expander”.

Clinical case

Patient: A 5-year-old boy D.S. referred to “Bambino Gesù” Children’s Research Hospital for sleep apnoeas since the age of 30 months. The patient underwent a baseline home nocturnal cardiorespiratory sleep study (CRSS) which revealed moderate OSA with apnoea/hypopnoea index (AHI) of 6 events per hour of sleep and no alteration in oxygen saturation (mean of 98%, minimal 92%). Clinical history revealed poor appetite, no allergies, diurnal and nocturnal mouth breathing, loud snoring and repeated apnoeas during sleep. On physical and ear nose and throat (ENT) examination the patient showed good general conditions, pectus excavatum and pathological tonsillar hypertrophy grade III) with no signs of inflammation. The initial extraoral and intraoral photographs are shown in Figure 1. The lateral cephalogram (Fig. 1) showed adenoidal hypertrophy (75% of obstruction) confirmed by fiberoptic evaluation. The cephalometric analysis (Table I) showed a class II skeletal malocclusion, increased maxillary-mandibular

Table I. Cephalometric analysis. Maxillary and mandibular skeletal assessment: maxillary position in relation to cranial base (SNA angle), mandibular position in relation to cranial base (SNB angle), maxillomandibular sagittal differential (ANB angle). Vertical skeletal assessment: maxillo-mandibular divergence (ANS-PNS/GO-GN angle). Airway dimensions: rhinopharyngeal airway dimension (AD1-PNS), oropharyngeal airway dimension (PAS-PPAS).

	Pre-treatment	Post-treatment	Normal value
SNA	73.5	73.4	81.4 ± 1.7
SNB	66.1	67.1	76.4 ± 1.4
ANB	7.4	6.3	5.0 ± 1.5
PNS-ANS/GO-GN	39.8	34.4	29 ± 5.0
AD1-PNS	7.3	7.9	18.9 ± 5.2
PAS-PPAS	3.1	4.8	12.0 ± 3.2

ular divergence, reduced rhinopharyngeal and oropharyngeal airway dimension. The quality of life (QOL) of the patient was measured with a questionnaire (OSA-18)¹⁰. The score reported in our patient was 81 which suggested a large impact on QOL for both the patient and parents.

Treatment: after multidisciplinary assessment, the child was prescribed a conservative treatment including orthodontic treatment and medical therapy. In order to correct malocclusion, we developed an innovative custom-made device, that we called “Sleep Apnea Twin Expander”. For development of the device, anatomical impressions and a wax bite registration in maximal intercuspation position



Fig. 1. Pre-treatment extraoral, intraoral photographs, lateral cephalogram.

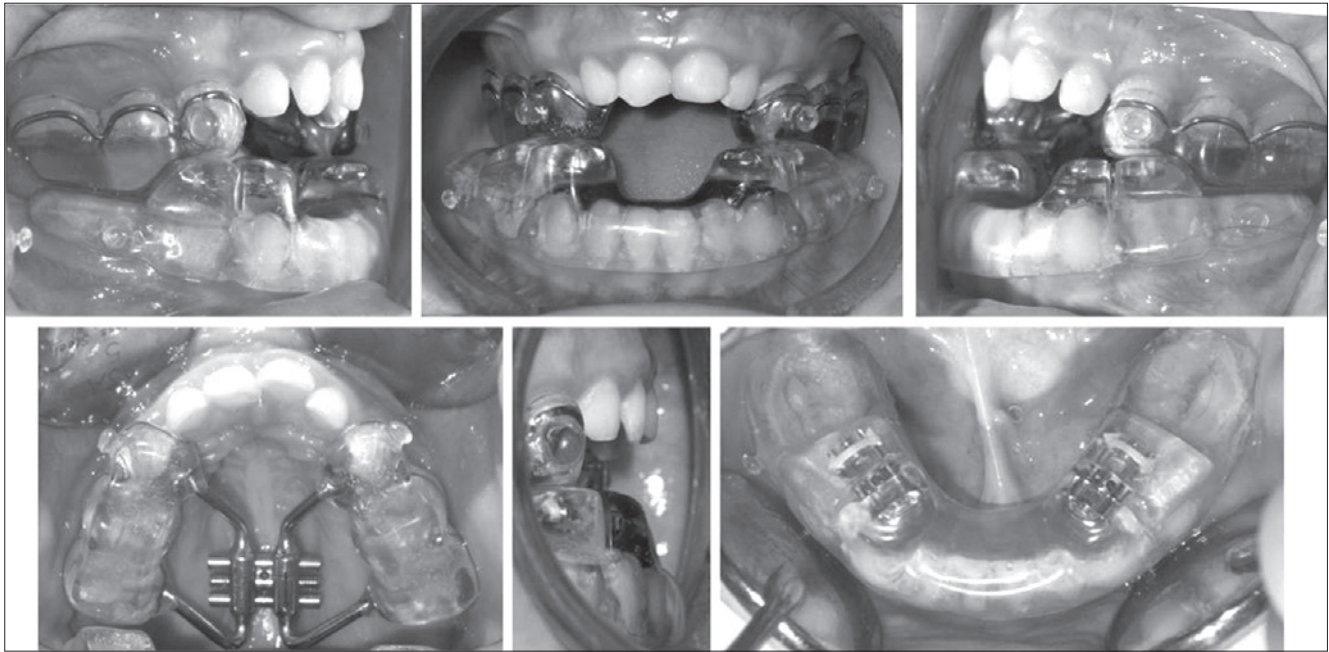


Fig. 2. Sleep Apnea Twin Expander in situ.

and in maximum mandibular protrusion were taken. The device (Fig. 2) is a double plate consisting of two separate elements, an upper fixed and a lower removable. It is a combination between the bite block expander by McNamara¹¹ and twin block appliance by Clark¹². The upper part is a modified bite block palatal expander, consisting of a Hyrax-type screw 10 mm (Philosophy 1, Lancer) secured to the maxillary arch with a 0.045-inch wire framework imbedded in acrylic covering the buccal, lingual and occlusal surfaces of the deciduous canine and first and second deciduous molars. The acrylic was 2 mm thick at the canines and 7 mm thick at the first and second deciduous molars creating steep inclined planes interlocked at about 70° to the occlusal plane. The lower appliance consisted of removable bite plane of acrylic retained with 0.9-mm ball clasps placed in the mandibular interproximal areas of first and second deciduous molars. The mandible was guided to protrude forward by the inclined planes in acrylic. Re-activation of the advancement was carried out with an adjustable screw mechanism on the lower splint to achieve movement of inclined planes for gradual advancement of the mandible. This appliance allows to carry out palatal expansion and mandibular advancement simultaneously in order to obtain an extension of upper airway area and a possible improvement of OSA symptoms in a short period. The upper bite block expander was bonded to the maxillary arch with resin reinforced glass ionomer cement (Fuji Ortho, GC America). The mother of the patient was instructed to advance the screw once a day for 30 days. In addition, after one week the patient was instructed to wear the lower device during the night. The otorhinolaryngologist prescribed intranasal corticosteroids. The advancement screw on the lower appliance was initially activated at 50% of the

maximum mandibular protrusion. Thirty days after the oral device placement, the patient's mother referred a reduction of snoring and disappearance of apnoea events. Intraoral examination revealed an expansion of the transverse diameter of the upper jaw and an upper interincisal diastema. The screw of the lower plate was reactivated to increase mandibular advancement until 60% of maximal protrusion, the patient was instructed to wear it 14 hours per day, to stop activations in upper expander and was instructed about nasal breathing exercises. The patient was seen at 4-week intervals for an average of 10 additional months. After four months of orthodontic therapy the patient performed a second home nocturnal CRSS with device in situ in order to evaluate the effectiveness of the occlusal device in reducing

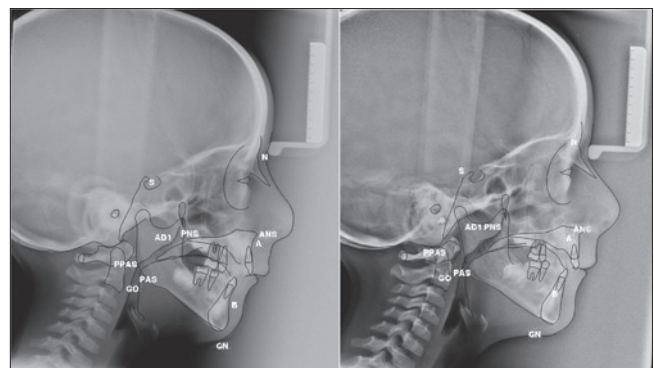


Fig. 3a-b. Lateral cephalogram pre- and post-treatment. Cephalometric points used in the analysis of the child. Points: A, A-point; ANS: anterior nasal spine; B: B-point; Gn: gnathion; Go: gonion; N: nasion; PNS: posterior nasal spine; AD1: adenoid inferior; S: sella; PPAS: posterior pharyngeal wall; PAS: anterior pharyngeal wall around gonial angle

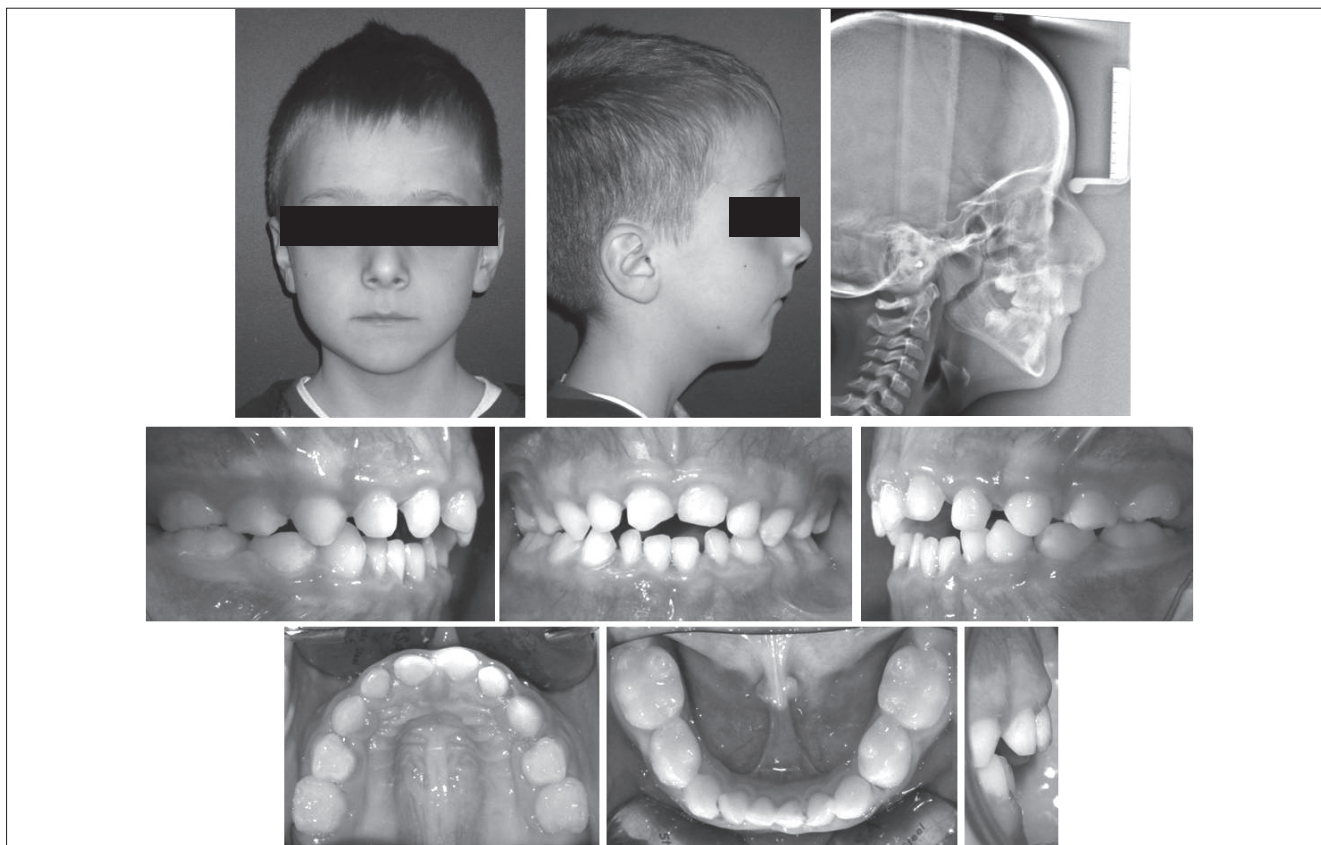


Fig. 4. Post-treatment extraoral, intraoral photographs, lateral cephalogram.

apnoea events at night. The CRSS showed the absence of significant apnoeas with oxygen saturation within normal limits (AHI = 0.6; oxygen saturation mean of 98.2%, minimal 94%). After eight months of retention, the upper appliance was removed, and lateral cephalogram was performed. The patient was prescribed to use the device as removable during the night. The comparison between cephalometric analysis pre- and post-orthodontic treatment (Fig. 3, Table I) showed an increase in the rhinopharyngeal and oropharyngeal space and a reduction in maxillo-mandibular sagittal and vertical discrepancy. The examination of the patient after orthodontic treatment showed an improvement in face appearance, lip competence and dry lips, and the intraoral view showed a class I malocclusion and reduction of the overjet and open bite (Fig. 4). The OSA-18 QOL questionnaire administered to the parents at the end of orthodontic treatment showed that the score was reduced to 20, suggesting a large improvement. ENT assessment at the end of the treatment showed reduction in hypertrophy of palatal tonsils and adenoids to grade II, and with no indication for surgical treatment.

Discussion

In this case report we describe the successful treatment of moderate OSA with a new oral device in a 5-year-old boy.

Although adenotonsillectomy represents the first choice for treatment of OSA in children, we decided to treat him with an orthodontic approach based on the moderate degree of OSA with a normal oxygen saturation index and the possibility of a conservative therapeutic strategy. The Sleep Apnea Twin Expander device was suitable for the patient described because it combined maxillary expansion and mandibular advancement. Orthodontic treatment resulted in an improvement of OSA symptoms through modification of the facial skeletal components. The positive effects of maxillary expansion and mandibular anterior repositioning have been previously described in children⁷⁻⁹. The rapid maxillary expansion can produce significant increment of the total nasal volume¹³ and mandibular advancement enlarges the retrolingual space and at the same time promotes lingual advancement during the night⁷. These aspects, however, have not been confirmed by literature review because of the limited number of studies and further studies are necessary to examine the effect of orthodontic device in a large sample of paediatric patients^{14 15}.

In our patient, breathing pattern and skeletal improvements were documented: the post-treatment lateral cephalogram showed an increase of rhinopharyngeal and oropharyngeal space, a reduction of maxillo-mandibular sagittal and vertical discrepancy, AHI passed from 6 to 0.6 events/night and the OSA-18 score decreased from 81

to 20. Moreover, a reduction of the adenoidal and tonsillar hypertrophy degree was demonstrated, similar to data reported in a previous study⁷. We hypothesize that this effect may be related to the enlargement of the upper airway and retropharyngeal spaces that produce a relative reduction of the hypertrophy. The successful therapy of this case confirms the importance of a multidisciplinary approach in diagnosis and therapy of paediatric OSA. The early orthodontic approach, aimed to obtain simultaneous palatal expansion of the upper maxillary and the mandibular advancement at the first phase of the treatment, associated with medical and breathing exercises, may modify the skeletal maxillo-mandibular anatomy by preventing obstruction of the upper airway. Considering the results of this case, further studies are warranted to examine the effects of orthodontic treatment with the Sleep Apnea Twin Expander in a large number of paediatric OSA patients with similar craniofacial anomalies.

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References

- 1 American Thoracic Society. *Standards and indications for cardiopulmonary sleep studies in children*. Am J Respir Crit Care Med 1996;153:866-78.
- 2 American Academy of Pediatrics. *Clinical practice guideline: diagnosis and management of childhood obstructive sleep apnea syndrome*. Pediatrics 2002;109:704-12.
- 3 Marcus CL, Brooks LJ, Draper KA, et al. *Diagnosis and management of childhood obstructive sleep apnea syndrome*. Pediatrics 2012;130:e714-55.
- 4 Friedman M, Wilson M, Lin HC, et al. *Updated systematic review of tonsillectomy and adenoidectomy for treatment of pediatric obstructive sleep apnea/hypopnea syndrome*. Otolaryngol Head Neck Surg 2009;140:800-8.
- 5 Huang YS, Guilleminault C. *Pediatric obstructive sleep apnea and the critical role of oral-facial growth: evidences*. Front Neurol 2013;3:184.
- 6 Zucconi M, Caprioglio A, Calori G, et al. *Craniofacial modifications in children with habitual snoring and obstructive sleep apnoea: a case-control study*. Eur Respir J 1999;13:411-7.
- 7 Villa MP, Bernkopf E, Pagani J, et al. *Randomized controlled study of an oral jaw-positioning appliance for the treatment of obstructive sleep apnea in children with malocclusion*. Am J Respir Crit Care Med 2002;165:123-27.
- 8 Pirelli P, Saponara M, Guilleminault C. *Rapid maxillary expansion in children with obstructive sleep apnea syndrome*. Sleep 2004;27:761-6.
- 9 Marino A, Ranieri R, Chiarotti F, et al. *Rapid maxillary expansion in children with Obstructive Sleep Apnoea Syndrome (OSAS)*. Eur J Paediatr Dent 2012;13:57-63.
- 10 Franco RA Jr, Rosenfeld RM, Rao M. *Quality of life for children with obstructive sleep apnea*. Otolaryngol Head Neck Surg 2000;123:9-16.
- 11 McNamara JA Jr, Brudon WL, Kokich VG. *Orthodontics and Dentofacial Orthopedics*. Needham Press; 2001.
- 12 Clark WJ. *Twin-block functional therapy*. London: Mosby-Wolfe; 1995.
- 13 Cordasco G, Nucera R, Fastuca R, et al. *Effects of orthopedic maxillary expansion on nasal cavity size in growing subjects: a low dose computer tomography clinical trial*. Int J Pediatr Otorhinolaryngol 2012;76:1547-51.
- 14 Kiliç N, Oktay H. *Effects of rapid maxillary expansion on nasal breathing and some naso-respiratory and breathing problems in growing children: a literature review*. Int J Pediatr Otorhinolaryngol 2008;72:1595-601.
- 15 Carvalho FR, Lentini-Oliveira D, Machado MA, et al. *Oral appliances and functional orthopaedic appliances for obstructive sleep apnoea in children*. Cochrane Database Syst Rev 2007;18:CD005520.

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