Combined Orthodontic and Surgical Treatment in a 8-Years-Old Patient Affected By Severe Obstructive Sleep Apnea: A Case-Report

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An eight-years-old girl showed a restless sleep with snoring and severe apnea episodes, a mandibular retrognathia, mouth breathing, maxillary transverse discrepancy, mandibular transverse discrepancy, moderate crowding and anterior open-bite. The CBCT showed an anterior collapse of the epiglottis. The treatment consisted in a rapid palatal expansion, an epiglottoplasty and a reduction of the tongue base. Polysomnography revealed that apnea-hypopnea index improved from 21,8 episodes/hr at the baseline to 0,6 episodes/hr, average oxygen saturation from 96,5% to 98,1%, oxygen desaturation events from 23,4 episodes/hr to 1/hr.

Key words: Obstructive sleep apnea, children, orthodontics, epiglottoplasty,

INTRODUCTION

Sleep disordered breathing (SDB) incudes different sleep disorders ranging in severity from primary snoring to obstructive sleeping apnea (OSA). OSA is characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction that implicates a complication in physiological ventilation during sleep and alters the normal sleep pattern[1].

In a systematic review published in 2008[2], authors assessed that in paediatric population the prevalence of snoring was 7.45% while the prevalence of OSA was 1-4%.

Several studies demonstrated a wide range of medical implication caused by OSA that can affect children health such as cardiovascular, endocrine and metabolic disorders but also their behavioural development[3]. More precisely, untreated OSA in children can evolve in aggressive behaviour, attention-deficit and hyperactivity disorder (ADHD) and even in an IQ reduction and poor school performances [4,5]. Because of this wide range of complications, a prompt and efficient therapy is absolutely necessary in order to restore the physiological ventilation during sleep.

The gold standard test for the diagnosis of obstructive SDB is an overnight ambulatory polysomnography (PSG). The apnea-hypopnea index (AHI) is the most commonly used PSG parameter for the quantification of SDB severity. It comprises the number of mixed, obstructive and central apneas and hypopneas per hour of total sleep time. An obstructive AHI ≤1 episodes/hr is considered to be normal, 1< AHI ≤5 to be mild OSA, 5< AHI ≤10 to be moderate OSA, and an AHI >10 episodes/hr as severe OSA.

Actually only a few treatments resulted efficient. Although surgical treatment has not been proven to fully resolve OSA in all patients, most children are managed with tonsillectomy and adenoidectomy[6]. Regarding non-surgical treatments, continuous positive airway pressure (CPAP), that is the most common treatment in adult patients, seemed not ideal for children due to a very poor compliance rate[7].

Orthodontic treatment such as rapid maxillary expansion and mandibular advancement devices showed positive outcomes even only a few studies have been conducted until now. In a recent review on orthodontic treatment in OSA patient, Huynh *et al*., 2016 concluded that further studies with larger sample size, a more accurate study design and strong inclusion/exclusion criteria are needed in order to confirm data resulted from preliminary studies[8].

Case Report

An eight-years-old girl came to Padua University Hospital Dental Clinic for an orthodontic evaluation. General medical history questionnaire was complete. Her parents reported that their daughter was in good health but suffered of a restless sleep with loud snoring and intensive apnea episodes. In addition the patient showed a severe fatigue during daytime to the extent that her normal recreational and athletic daily activities were limited.

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Moreover, parents reported that their daughter had already undergone adenotonsillectomy when she was six years old. The extraoral examination showed a hyperextension of the neck, a severe mandibular retrognathia and mouth breathing.

The intraoral exam showed a maxillary transverse discrepancy without cross-bite, mandibular transverse discrepancy, mild crowding on the upper and on the lower arches and anterior open bite and a class II malocclusion (Fig.1).

In order to analyse more extensively her condition an ENT evaluation was required. A Cone Beam Computed Tomography (CBCT) of the craniofacial complex was requested

An ambulatory PSG was prescribed to evaluate the severity of her sleeping disorder and verify the diagnosis of OSA.

Polysomnography

Ambulatory PSG was performed in a private clinic specialized in sleep medicine (Fig.2a). The patient was prepared and slept there monitored by remote video. The complete exam lasted 10 hours and the patient slept in both supine and non-supine positions. During the exam the following parameters were registered: electroencephalogram (EEG), electrooculogram (EOG), electromyogram (EMG), electrocardiogram (ECG), plethysmogram, oxygen saturation, blood pressure.

The most important index is the number of apnoea and hypopnoea (AHI) episodes per hour and resulted 21,8 episodes/hr. This is a severe result because it doubled the cut-off for diagnosis of severe OSA. Nevertheless, such a condition is even more serious in child of eight years old.

The other results were an average oxygen saturation 96,5% and oxygen desaturation events (OD) 23,4 episodes/hr. All these findings confirmed the diagnosis of severe paediatric OSA, a condition that need an immediate and efficient therapeutic strategy.

CBCT evaluation

The CBCT was analysed using Dolphin Imaging 3D Software to obtain the analysis of sagittal, coronal and axial sections and a three-dimensional reconstruction of the facial complex and airways volume (Fig.3-4). From the sections a low tongue posture and the palate mucosa hypertrophy emerged. Regarding the upper airways a left nasal septal deflection, an inferior turbinate hypertrophy and a chronic sinusitis of the maxillary sinuses and of the ethmoid cells were found. In physiological conditions paranasal sinuses appear radiolucent in radiographs and tomography; chronic inflammation and inflammatory exudate are confirmed by radiopaque findings in the paranasal sinuses.

Figure.1: Pretreatment photographs.

Figure.2: a) pre-treatment polysomnography; b) polysomnography 24 months after the surgery.

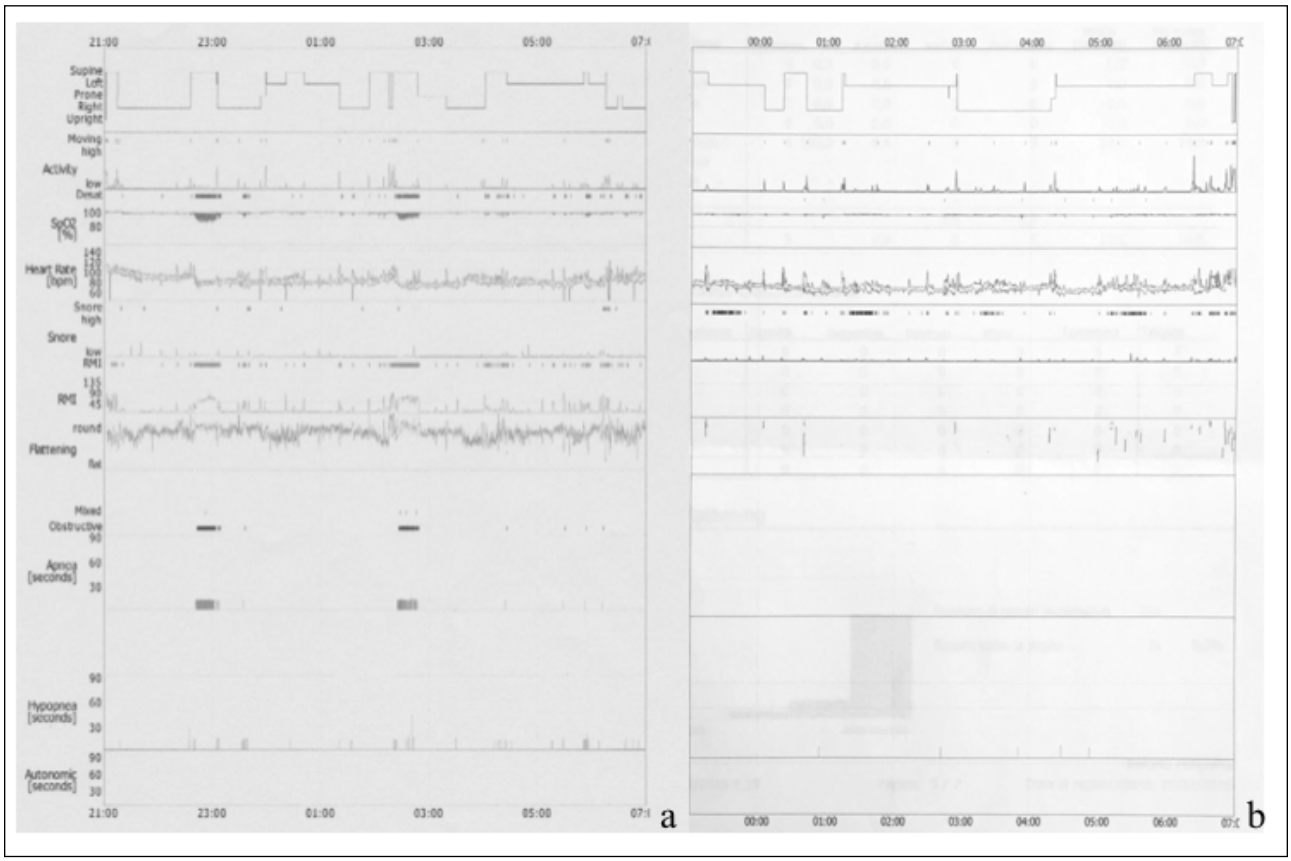


Figure.3: a) sagittal section from the CBCT; b) obstruction at the epiglottis level; c) rapid palatal expansion,

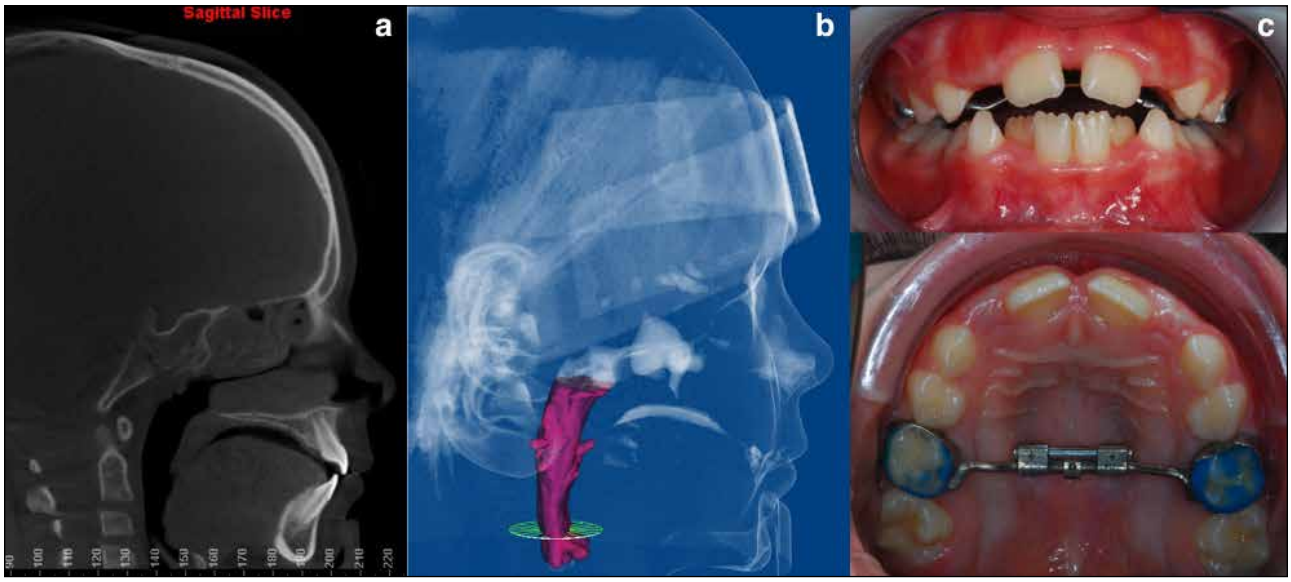
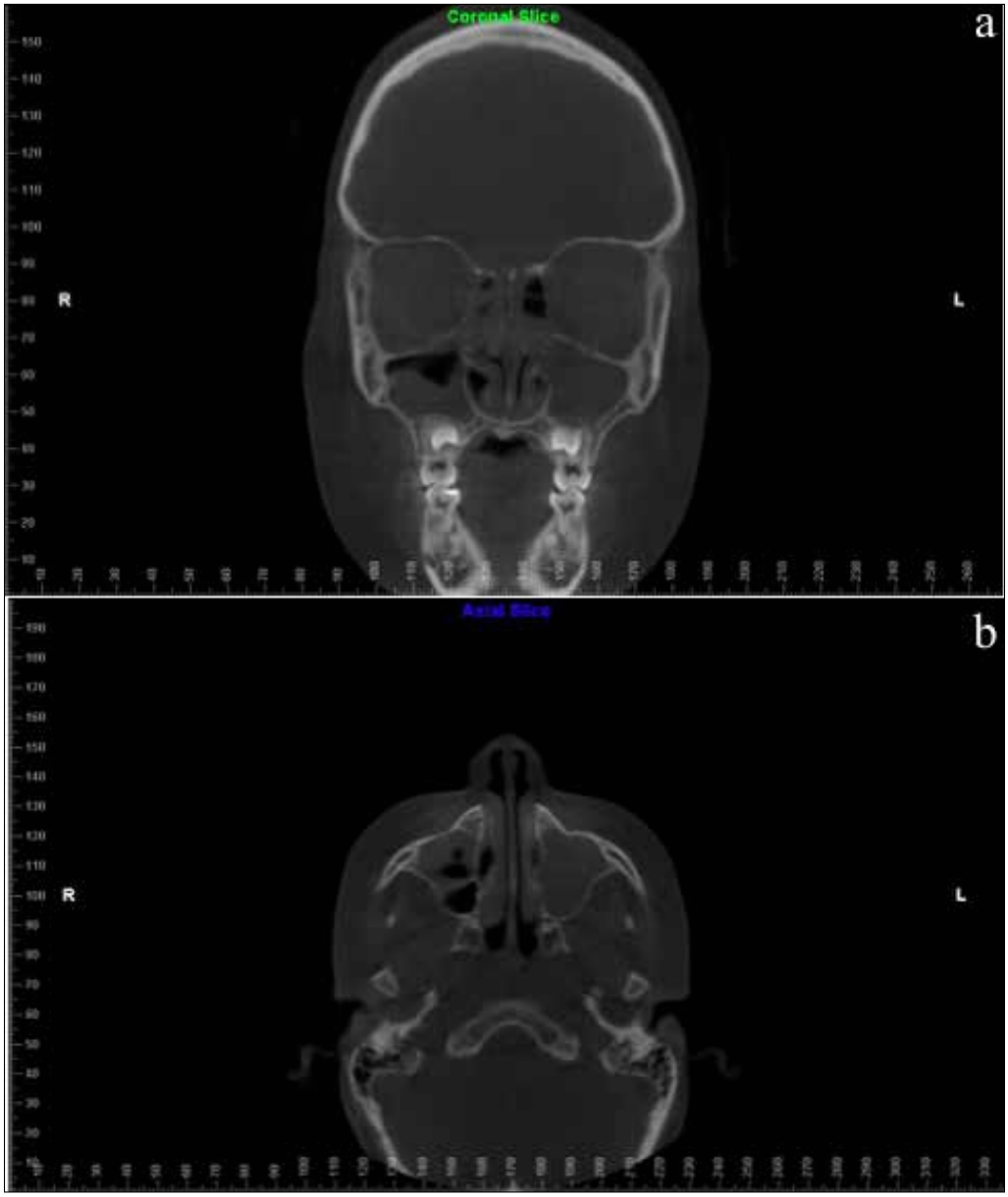


Figure.4: a) coronal section from the CBCT; b) axial section from the CBCT.



These findings are typical in patients with obstructive pathologies where the free flowing of the air is not permitted. An interesting finding was the epiglottis anterior collapse on the pharynx. This situation could have been responsible of the sleeping apnoea episodes.

From the three-dimensional reconstruction the airways volume resulted reduced. The software provide a function able to identify the area of maximum restriction of the airways that resulted 137,7 mm2. It corresponded to the epiglottis level confirming the radiograph finding in the sagittal section. A Sleep Nose Endoscopy (SNE) was requested to confirm clinically the obstruction at the epiglottis level.

Sleep Nose Endoscopy

Sleep nose endoscopy (SNE) was prescribed in order to evaluate clinically the airways function during the sleep. The exam was made in day-hospital in an operating room. The sleep was induced by professional pharmacological sedation administered by an anesthetist. After the patient fell asleep, a flexible optical fibre was passed through one side of the nose in order to evaluate the throat and observe the potential blockage of breathing in the palate and tongue regions.

The exam revealed a septal deflection on the left side, inferior turbinate hypertrophy with mucous secretion, no obstruction at rinopharynx level, hypertrophy of the tongue basis, good limits of the larynx. Moreover, the exam revealed III degree antero-posterior collapse of the oral pharynx, hypo-pharynx collapse at the epiglottis level. The minimum oxygen saturation was 79%.

A pharmacological treatment was prescribed using salbutamol and beclomethasone (0,10 mg/kg for ten days) with nebuliser to reduce the inflammation of upper airways.

Treatment alternatives

After all these evaluations, different therapeutic alternatives were examined. In some mild OSA patients, it can be decided to prescribe a pharmacological treatment and revaluate the patient. This is an alternative in moderate obstruction caused by tonsils or adenoids hypertrophy because this condition physiologically tend to decrease during growth.

In this little patient adenoidectomy had previously conducted but it did not resolve her sleeping disorder. This failure probably was due to an inaccurate evaluation of the obstruction at the epiglottis level other than tonsils and adenoids hypertrophy. However, in such a severe condition waiting was not considered a possible solution.

The first option was more conservative and consisted in an orthodontic treatment with a RPE and a mandibular advanced device (MAD). The maxillary expansion would have solved the transverse discrepancy and at the same time would have increased the volume of nasal airways. On the other hand the MAD could have solved the epiglottis collapse on the hypo-pharynx level. Treatment with MAD is mainly used in adult patients with mild to moderate OSA and a low compliance with traditional continuous positive airway pressure (CPAP) treatment. In paediatric patient only a few studies have been conducted until now and its efficiency has not already proven especially in severe cases.

The second option was more invasive and consisted in an orthodontic treatment with RPE in order to solve the transverse discrepancy and increase the volume of nasal airways. Moreover a plastic surgery treatment of the base of the tongue and epiglottis would have solved or reduced the collapse of the hypo-pharynx. The MAD would have been used only after a control PSG in case of residual mild to moderate apnoea.

These two solutions were fully illustrated to the parents and we jointly opted for the surgical one. The surgical treatment, even more invasive, guaranteed a more favourable and immediate predictive outcome that was the most important factor in such a severe condition.

Report of case

The orthodontic treatment started in order to solve the maxillary transverse discrepancy using a RPE.

The treatment consisted in the following stages:

1. dental records were taken in order to built the device in a dental technician laboratory
2. after one week the RPE was delivered and applied to the patient (Fig.3c). Daily activations were required (1/die for 20 days). Every activation consists in a 0,2 mm of maxillary transverse expansion, so at the end of the therapy 4 mm of expansion were reached.
3. one month after the application of the RPE the patient came back for a control examination. The maxillary transverse discrepancy was solved so no other activation were required.
4. The device was kept in place for six months in order to waiting for the stabilization of the expansion and at the end of this period the RPE was finally removed (Fig.5).

At the same time the patient was operated of epiglottoplasty and of reduction of the tongue base.

RESULTS

During the following medical examination the parents referred a more comfortable sleep without apnea episodes during the night for the patient. After 24 months from the surgery another PSG was performed. The exam revealed that AHI improved from 21,8 episodes/hr at the baseline to 0,6 episodes/hr, average oxygen saturation from 96,5% to 98,1%, oxygen desaturation events from 23,4 episodes/hr to 1 episodes/hr. The exam duration was of 469,6 minutes and the patient slept both in supine and not-supine position. These records confirmed the resolution of the severe OSA.

Figure.5: Intraoral photographs 24 months after the treatment

DISCUSSION

Paediatric OSA patients need a rapid and effective treatment approach to reduce the complications associated to this syndrome. Altered sleep anatomy can affect cardiovascular and endocrine systems but also children neurological development. Actually only a few treatments have been demonstrated valid. Although surgical treatment has not been proven to fully resolve OSA in all patients, most children are managed with tonsillectomy and adenoidectomy[6]. Regarding non-surgical treatments, continuous positive airway pressure (CPAP), that is the most common treatment in adults, seemed not ideal for children due to a very poor compliance rate[1]. In a recent review on orthodontic treatment in OSA patient, [Huynh et al., 2016][8] concluded that further studies with bigger sample size, a more accurate study design and strong inclusion/exclusion criteria are needed in order to confirm data resulted from preliminary studies. In this case, rapid maxillary expansion associated with epiglottoplasty and tongue base reduction were an efficient therapy and implicated a complete resolution of the severe OSA. Three different treatment options were evaluated to solve the patient's condition: pharmacological, MAD and surgical treatment. The first one, the pharmacological treatment was rejected because it is usually used in moderate or mild obstruction (the patient showed a severe one) caused by adenoids or tonsils hypertrophy. The other two treatment options were discussed with patient's parents. There are no many studies in literature that describe a treatment with MAD in paediatric patients and its efficiency has not already proven in severe cases, so we were not sure about the success of the treatment. This condition influenced patient's quality of life: restless sleep, intensive apnea episodes, severe fatigue during the day. For all these reasons parents decided for a treatment that would have surely improved her quality of life. The CBCT evaluation showed an epiglottis anterior collapse and a hypertrophy of the tongue basis was evaluated in the sleep nose endoscopy (SNE), so it was decided to perform surgery at both these levels in order to completely solved the obstruction and restore an appropriate volume for the air flow. After the surgical treatment the parents referred a more comfortable sleep and an improvement in her quality of life.

CONCLUSION

Orthodontists can play an essential role in the in the interception of obstructive pathologies in the paediatric age. The resolution of the condition of severe OSA can restore a physiological development of these young patients.

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