2020

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Recommended Citation
DOI: 10.38209/2708-2636.1088
Available at: https://www.tjo.org.tw/tjo/vol32/iss4/2

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CASE REPORT

Interdisciplinary Approach of Pediatric Obstructive Sleep Apnea with Congenitally Missing Second Premolars

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ABSTRACT

This report is a case of 11-year-old young boy who was diagnosed with pediatric obstructive sleep apnea (OSA) by sleep surgeon and was referred for further evaluation of co-therapy with intervention of orthodontic appliances. This patient is a growing case of skeletal class II jaw relation, hypodivergent facial pattern, large overjet and anterior deep overbite with congenitally missing premolars. The treatment contained two folds. In the first stage, functional appliance was designed to help with the skeletal jaw discrepancy. By doing so, the expansion over upper airway was also expected. In the second stage, fixed edgewise appliances were bonded for preparation of prosthetic space and improvement of occlusion. This case report will present and analyze the mechanism of treatment and the pre- and post-treatment changes over the condition of OSA, facial profile as well as the occlusion.

Keywords: Pediatric obstructive sleep apnea; Obstructive sleep apnea (OSA); Functional appliance; Growth modification; Congenital missing

INTRODUCTION

Obstructive Sleep Apnea (OSA) is a common disease which is highly regarded nowadays. It is a breathing disorder during sleep time. The upper airway is partially obstructed or intermittently completely obstructed.1 OSA is not only a grown-up disease but also a pediatric issue. The origin of OSA can be divided into two condition. One is central neurologic (5%); the other is obstructive, main source accounting for 95%. In addition to OSA in adults, familiar sign and symptoms (S/S) can also be evaluated in pediatric OSA during sleep time and night time. During sleep time, snoring, diaphoresis, restless sleep, frequent awakenings, enuresis and even parasomnias can be noted. In the day time, pediatric OSA can lead to daytime sleepiness, poor school performance, attention deficit hyperactivity disorder (ADHD), aggressive behavior, depression, and morning headache.2

Concerning this important issue, prevalence and incidence are useful references for clinical diagnosis and treatment in pediatric OSA. The peak incidence is 2-8 years old, relating to the large adenoid tonsil size and undeveloped airway.2,3 Between males and females, the distribution is almost equal. The global prevalence of pediatric OSA is estimated from 1-5%, varying in different regions. In Asia, the prevalence are 0-4.8% in China and 0.2-1.3% in Thailand. In the U.S., it accounts for 0-1.2%. Also, the recorded results showed 1-1.8% in Italy and Turkey.1,2

The etiology can be classified by different aspects, including anatomic obstruction, neuromechanical dysfunction, obesity, genetic or environmental factors, and inflammation. The anatomic obstruction may be...
resulted from adenoid-tonsillar hypertrophy which is one of the most common pathogenesis of pediatric OSA, hypertrophy of cervical lymphoid tissue, and nasal obstruction, etc. Also, obesity is a serious issue in pediatric OSA. The fatty tissue infiltration oppresses the upper airway and decreases the function of lung capacity. In addition, rhinosinusitis, asthma, nasal polyps consist in the inflammatory reasons. Craniofacial morphology may also be a factor that influence the condition of pediatric OSA. For example, mandibular retragnathism, long face, high mandibular angle, anterior open bite, deep and narrow palate and midface deficiency increase the risks of OSA.

The main treatment of pediatric OSA include surgery, continuous positive airway pressure (CPAP), medication, functional appliance, and myofunctional therapy. No matter which method is used, the main goals are expanding the airway size and/or removing the obstructive structure. The surgical techniques are adenotonsillectomy (AT) and uvulopalatopharyngoplasty (UPPP) which are the major management of pediatric OSA. In the cases that surgery is contraindicated, CPAP is the most common non-surgical treatment. However, certain functional appliance and myofunctional therapy generally practiced by orthodontists were also reported to increase the volume of upper airway effectively. Some studies also indicated improved polysomnography (PSG) outcome with obvious decrease of value of apnea-hypopnea index (AHI). The most often selected appliances are maxillary-expansion devices and mandibular anterior repositioning appliances. Maxillary expansion is conventionally used to correct the transverse deficiency of maxilla. However, we can gain the benefit in treatment of OSA by the secondary effects, reducing nasal airway resistance and increasing volume of nasopharynx and nasal cavity. Mandibular anterior repositioning appliances are used in treatment of Class II skeletal discrepancy. By the stimulation of mandibular growth and anterior positioning, the volume of upper airway may also be increased, especially in oropharynx. These appliances are frequently used by orthodontists. In addition, the effect of improving pediatric OSA can be expected.

Congenitally missing dentition is also frequently observed clinically. Beside the third molars, missing of second premolars are the most frequently seen position, followed by maxillary lateral incisors. The prevalence ranges from 1.6-9.6% in different countries and regions. The etiology may be multifactorial combining genetic and environment factors. In genetic aspects, MSX-1, PAX-9, EDA and AXIN-2 are highlighted. In environment condition, it is associated with abnormality of morphology and eruption.

CASE REPORT
Clinical examination and diagnosis

An 11-year-3-month-old boy (Height: 142 cm; weight: 42 Kg; BMI:20.8) was first brought to Ear-nose-throat (ENT) and Oral Maxillofacial (OMS) Department in Kaohsiung Medical University Hospital, Taiwan for evaluation of sleep obstructive arena and was referred to Orthodontic department by the sleep surgeon (Figure 1). The patient's parents noted that he had noisy snoring, witnessed apnea and its influenced on school performance. The tonsil size was grading in size 2 by Friedman tonsil grading system. The 60% obstruction of adenoid hypertrophy was noted under fiberscope. The PSG indicated that AHI was 14.5. OSA was diagnosed according to American Academy of Sleep Medicine (AASM) guideline. The guideline contains two criteria; one is clinical symptoms; the other is AHI ≥ 1 or 25% of total sleep time with hypercapnia (PaCO2 > 50 mm Hg) in association with snoring, flattening of the nasal pressure waveform, or paradoxical thoracoabdominal motion. In addition to the PSG, the cone-beam computerized tomography (CBCT) was also taken for evaluation of condition of different part of upper airway and the volume. The 3-dimentional data were further analyzed by Digital software (Dolphin® Version 11, Chatsworth, California, U.S.A.). The patient's volume of velopharynx was 3630 mm³; glossopharynx was 1843 mm³; and hypopharynx was 4554 mm³ (Figure 2A, C, 2D). Though the severity cannot be defined by the data of volume, the progress of treatment can be monitored by the change of volume. Regarding that minimal cross section area plays an important role of evaluating OSA, the lateral surface length (LSL) and the anterior posterior length (APL) of the minimal cross section area were also measured over oropharynx and hypopharynx (Figure 2B). In the minimal cross section area of oropharynx, the LSL was 9.9 mm and the APL was 10.5 mm; they were 26.7 mm and 5.7 mm respectively in hypopharynx.

In cephalometric analysis, the patient had a class II skeletal jaw relation (ANB: 5.8°) with normal-positioned maxilla and retrusive mandible. The cervical development is in cervical vertebral maturation (CVM) stage II ~ III. In vertical aspect, he had a hypodivergent facial pattern (SN-MP: 33.1°) (Figure 1C and Table 1). In his dental condition, the molar relation was Class II over right side and Class I over left side; overjet was 6.5 mm and overbite was 5 mm. The patient was diagnosed as Angle's class II malocclusion, large overjet and anterior deep...
overbite (Figure 1B). Besides, primary teeth 55, 65, 75, and 85 were retained with congenital missing over tooth 15, 34, 35, and 45 and reversely impacted tooth bud of 25 (Figure 1D). For the soft tissue, the patient has a convex facial profile with both lips protruded to E-line (Figure 1A). In evaluation of transversal width of maxilla, the initial inter-canine width was 36.3 mm; inter-molar width was 58 mm;

Figure 1. A, extraoral photographs. B, intraoral photographs. C, lateral cephalometric film. D, panoramic film, initial stage.
basal arch width (BAW) (distance between the centers of resistance) of maxillary canines was 32.1 mm; BAW of maxillary first molars was 48.4 mm.

**Treatment plan**

The treatment in orthodontic department was planned into two stages. In the first stage, the goal was to improve his OSA by expanding the upper airway as well as to stimulate the mandibular growth. In the second stage, the goals were to align the patient's dentition and distribute the space for denture fabrication over missing area in the future. In addition to the orthodontic and orthopedic treatment, AT and UPPP were also planned and
operated by otorhinolaryngologist during the first-stage orthodontic treatment period.

Treatment progress

Before the first stage was started, the patient was referred for extraction of tooth 25, which was impacted reversely. In the first stage the total treatment time was 6 months. Functional appliance was designed with a combination of twin block and maxilla expansion device. After intra-oral impression, the bite was registered to an edge-to-edge position, with the mandible positioned forward. It was expected that the appliance not only helps the

<table>
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<th>Post-1st stage Treatment</th>
<th>Post- Treatment</th>
<th>Normal Range</th>
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<td>3</td>
<td>2</td>
<td>1.2–4.4</td>
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growth of mandible but also helps to expand the volume of upper airway and widen the isthmus part (velopharynx). Although no obvious transverse discrepancy was noted, expansion over maxilla was still planned in order to expand the volume of upper airway. After delivery of the appliance, the patient was required to wear at least 14 hours per day including sleep time. The patient was also requested to activate the expander 2 turns (1/2 mm) per week. The total expanding period was 10 weeks. Though the appliance was operated by the patient and his parents, the appointment was still arranged routinely for monitoring (Figure 3).

After the first-stage treatment, there was great improvement of clinical condition (snoring and witnessed apnea, etc.) that was noticed by the patient and his parents. The examination was also arranged after first-stage treatment. The skeletal condition was improved to Class I jaw relation (ANB: 4.8°) with a harmonious facial profile (Figure 4A and B, 4C and Table 1). The situation of the patient’s OSA was also improved obviously according to the PSG data and image of CBCT after first stage treatment. The AHI decreased to 2.6. The volume of upper airway analyzed with CBCT image showed a great expansion. The volume of velopharynx was 4167 mm³; glosso-pharynx was 5679 mm³; and hypopharynx was 5267 mm³. In the minimal cross section area of oropharynx, the LSL increased to 18.6 mm and the APL was 8.7 mm; they were 27.9 mm and 7.5 mm respectively in hypopharynx. All regions showed a dramatical change especial in glosso-pharynx (Figure 2A, 2C, 2D). In transversal change of maxilla, the inter-canine width was increased to 40.8 mm; inter-molar width was 61.2 mm; BAW of maxillary canines was 33.6 mm; BAW of maxillary first molars was 51.5 mm.

In the second stage the fixed edgewise appliance was bonded for alignment (Figure 5). The spaces of missing teeth of 15, 25, 34, 35 and 45 were also be redistributed for fabrication of prosthesis in the future. Over the right side, inter-proximal reduction was operated over tooth 55 and 85 to fit the crown size of definitive prosthesis as well as the to achieve an ideal alignment and interdigitation. In the 9th month of second-stage treatment, gum boil was noted over buccal gingival of tooth 65 and was referred for extraction to prevent further lesion in alveolar bone. Over lower left area, tooth 75 was left in the position for maintaining bone level thought there is a large space to be reconstructed by two-premolar prosthesis. In the 11th month of second-stage treatment, Angle's Class I occlusion and well alignment were also achieved. The midline was corrected to be coincident. Overjet was 1.5 mm; overbite was 1.5 mm (Figure 6A–D). After the two-stage treatment, there were obvious improvement of the patient's profile and dental condition (Figs. 6 and 7). In the retention phase, fixed lingual retainer over lower anterior dentition and removable clear retainers over both arches were delivered for maintaining the treatment outcome as well as the redistributed spaces which were planned for reconstruction after the end of craniofacial growth. The patient is scheduled for routine follow-up for retention in Kaohsiung Medical University Hospital Orthodontic Department.

**DISCUSSION**

In this case, the treatment goal is to achieve ideal airway patency and to create a stable occlusion with appropriate space redistribution for definitive prosthesis after growth. The patient was still in preadolescence with undeveloped upper airway and
oversized lymphoid tissue that may cause airway restriction. Therefore, the treatment strategies were removing the obstructive soft tissues and stimulating skeletal growth to gain greater volume and to improve the clinical symptoms by surgery and removable functional appliance at first. Then, the fixed orthodontic appliances were applied for the precise dental alignment.
In treatment of pediatric OSA, surgery and CPAP are considered as first choices. However, functional appliances that orthodontists apply commonly not only correct skeletal discrepancy by the orthopedic effect but also increase the volume of upper airway in certain aspect. In this case, twin block was selected due to the patient’s skeletal class II jaw relation and hypodivergent facial pattern. Besides the orthopedic effect, the anterior positioned effect over mandible help expand the upper airway space, especially in glossopharynx that contributed to the treatment of this patient’s OSA. In several cases studies, functional appliances that advanced mandible brought obvious widening or expansion of upper airway by analysis with CBCT and lateral cephalometric radiograph. However, effects of the different functional appliances should be considered in different parts of the upper airway. Wang et al indicated that twin block help increase of airway volume and the minimal cross-sectional area of the oropharynx. Shete CS et al revealed significant improvement in oxygen saturation level, airway volume, smallest cross section, anteroposterior width and even transversal width. Regarding the effect of maxillary expansion, the main effect is the increase of nasal cavity three-dimensionally. To evaluate the change under treatment of nasal cavity, acoustic rhinometry is considered more accurate than image analysis by CBCT. Because acoustic rhinometry was not carried in this case, the effect of maxillary expansion could not be defined specifically.

In addition to the nasal cavity, we can discuss more about the upper airway in detail to understand how the treatment contents affected the volume of the upper airway. In the velopharynx, which containing the minimal cross-sectional area of oropharynx, AT and UPPP were considered contributing mainly to the enlargement of this part because that the removed obstructive soft tissues were mostly located in this area. However, some studies have mentioned that mandibular anterior repositioning appliances help with the enlargement of velopharynx or minimal cross-sectional area of oropharynx. In front of the glossopharynx and hypopharynx, the anteriorly positioned mandible due to the simulation by the appliance and natural growth brought obvious effects in increasing the volume of this part. Regarding the decreased value of AHI and reduced symptoms, the contribution of surgery and functional appliance could not be defined individually. Therefore, more cases and further analysis should be recorded in the future.

Mild OSA could still be noticed from the post-treatment data of PSG (AHI = 2.6) though there was a great amount of expansion of upper airway. The possible reasons may be due to residual proliferation of lymphoid tissue or nasopharyngeal origin such as naso-septal anomalies or nasal polyps existed. Further examination by acoustic rhinometry or endoscopy may be needed. However, conservative monitoring and follow-up are considered due to the obvious decrease of value of AHI and the improvement of clinical symptoms.

Though the airway condition could be viewed directly by the image of CBCT, the real situation of upper airway could not be displayed. During the image was taken, the patent is awake and in a standing posture. However, the OSA happened during patients’ sleep time and lie-down posture. Therefore, appropriate sleep and airway examination, like PSG or nasal pharyngoscopy are necessary to help with diagnosis and evaluation.
Figure 6. A, Extraoral photographs. B, intraoral photographs. C, lateral cephalometric film. D, panoramic film, after the 2nd stage Tx was finished.
In treatment of pediatric OSA, surgery, such as AT and UPPP are considered as the first choices. In the situation that surgery is contraindicated due to underlying condition or refused by the patients and family, CPAP is also commonly used to maintain the flow over upper airway. However, the discomfort of wearing CPAP and noise cannot be adapted by every patient. It also requires patient's compliance.

In the second stage of treatment, there were more consideration to manage the spacing of missing teeth. Considering the patient's facial profile and the further growth potential, it was not appropriate to make the both lips more retrusive. Therefore, the space was planned to be maintained. Because the vertical growth of young male was continuous until 21 years old, the implant or tooth-borne prosthesis are not suitable to be applied now. During this period, the space arrangement and maintenance of bone level are the important issues. Once the space is planned to fit a prosthesis of permanent premolar, that the size of retained deciduous second molar need to be decreased. In average, the mesio-distal (MD) width is 11-12 mm in deciduous second molars and 7-8 mm in second premolars. Therefore, 4 mm of reduction is needed for most of cases. However, 4 mm reduction may cause exposure of dentin,
which is more vulnerable to caries than enamel, so a thin layer of resin coverage is necessary to prevent caries. In the aspect of maintaining bone level, retained deciduous tooth can be a natural maintainer. However, when the ankylosis of deciduous tooth is observed, extraction is indicated. Because ankylosed deciduous teeth stop erupting, that may cause an angular-bony-defect like appearance around the ankylosed tooth which does not contribute to periodontal bone level and extraction is needed if there is still a long period before final reconstruction. Diagnosis of ankylosis of deciduous teeth can be confirm from the periapical radiograph. The slope of periodontal bone level from the adjacent permanent teeth to the ankylosed deciduous tooth depress apically due to the infra-occlusal position of the ankylosed deciduous tooth, and that may help prove the occurrence of ankylosis.21 Besides, high-pitched tone of percussion sounds, tipping of adjacent teeth, supra-eruption of opposing teeth are also the clinical appearances that can be considered during making diagnosis.23 In this case, infra-occlusion of tooth 75 could be noticed after the treatment. Therefore, careful following is necessary to monitor the possibility of ankylosis.

CONCLUSION

This report demonstrated a case treated with functional appliance, fixed edgewise appliance, AT and UPPP in treatment of pediatric OSA, mandible retrusion and malocclusion. There was a significant improvement in the patient's sleep quality, facial profile and occlusal condition. Early diagnosis of OSA during childhood and preadolescent period with an effective intervention can further avoid unwanted health issue related with OSA.

Conflicts of interest statement

The authors declare that there is no conflict of interest.

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