

Maxillomandibular advancement surgery after long-term use of a mandibular advancement device in a post-adolescent patient with obstructive sleep apnea

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Patients with obstructive sleep apnea (OSA) whose phenotype belongs to a craniofacial vulnerability are referred from sleep doctors to orthodontists. In adults, for osseo-pharyngeal reconstruction (OPR) treatment, permanent maxillomandibular advancement (MMA) surgery and use of a temporary mandibular advancement device (MAD) are applied. This case report demonstrates successful treatment of OSA through application of phased MAD and MMA in a 16-year-old male with craniofacial deformity and residual growth potential. This patient showed skeletal and dentoalveolar changes after 7-year MAD use throughout post-adolescence, which affected the design and timing of subsequent MMA surgery, as well as post-surgical orthodontic strategy. This case report suggests that OPR treatment can be useful for treatment of OSA in post-adolescent patients, from an orthodontic point of view, in close collaboration with sleep doctors for interdisciplinary diagnosis and treatment.

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INTRODUCTION

Widely used medical treatment for patients with obstructive sleep apnea (OSA) comprises conservative non-surgical methods, phase I soft tissue surgery, and phase II skeletal surgery in a stepwise manner.¹⁻³ It is generally accepted that positive airway pressure (PAP) is the first-line treatment modality and that a mandibular advancement device (MAD) can be prescribed when the patient becomes intolerant to the PAP.⁴ Maxillomandibular advancement (MMA) has been regarded as an effective treatment modality, as it is a permanent osseo-pharyngeal reconstruction procedure.⁵⁻⁷

MAD or MMA is a good choice for adult patients whose OSA is caused by a primary craniofacial vulnerability, especially those with retruded mandibles.⁸ MAD enlarges the oropharyngeal airway space only during sleep by advancing the mandible. This leads to sagittal enlargement by forward movement of the tongue base and soft palate through stretching of the genioglossus and palatoglossus muscles; moreover, it causes transverse enlargement of the lateral pharyngeal wall by stretching the pharyngeal dilator muscles.^{9,10} In contrast, MMA permanently opens the entire velo-, oro-, and hypopharyngeal airway spaces.² By advancing the maxilla and mandible, anterior and lateral pharyngeal tissues attached to the posterior maxilla, mandible, and hyoid bone can be pulled forward sufficiently to enlarge the whole pharyngeal airway. However, MMA does not always completely cure OSA and its impact varies substantially among patients with different phenotypes of OSA.⁸ Although it remains challenging to establish therapeutic predictors for successful outcomes of MMA, response to MAD therapy or MAD-titration polysomnographic analysis have been suggested as predictors.¹¹

MMA is typically delayed until craniofacial growth is completed because of the unpredictable nature of residual growth,¹² or because of concerns regarding iatrogenic growth inhibition after surgery.¹³ This delay is

included even for patients with OSA who are expected to be good responders to MMA, due to phenotype, severity, and craniofacial factors. For these patients, MAD may be indicated for symptomatic management until MMA surgery can be applied. However, there have been no reports of the effect of MAD on the surgical and orthodontic planning of subsequent MMA treatment in patients with OSA who have residual growth potential.

The aim of this case report was to describe three considerations involved with long-term MAD use as a temporary aid throughout the post-adolescent period, which affected planning of the final MMA treatment in a patient with OSA.

DIAGNOSIS AND ETIOLOGY

A 16-year-old male adolescent was referred from the Department of Otorhinolaryngology of Kyung Hee University Medical Center because he exhibited severe snoring and excessive daytime sleepiness. He was diagnosed with moderate OSA based on polysomnography analysis that showed an apnea-hypopnea index (AHI) of 24.8 and a respiratory disturbance index (RDI) of 34.4. His body mass index (BMI) was 17.1 kg/m², and his subjective symptoms were severe, such that he exhibited a lowest oxygen saturation (LSaO₂) of 81.0% (Table 1). Facial and intraoral examination (Figure 1) revealed that he had a convex profile with protrusive upper lip, retruded chin, and short throat length. Class I molar and canine relationships, proclined maxillary and mandibular incisors with crowding, and shallow overbite were observed (Figure 2). Lateral cephalometric analysis showed skeletal Class II relationship with bimaxillary retrusion, hyperdivergent vertical pattern, low hyoid bone position, and narrow oropharyngeal airway space, particularly at the retroglossal airway; furthermore, the patient exhibited low tongue posture, such that the dorsum of the tongue was separate from the palate (Figure 3A and Table 2). He had no history of adenotonsillectomy or temporo-

Table 1. Summary of polysomnographic records at each observation time point

Polysomnography	Initial	MAD-titration	7 yr-post-MAD	Post-treatment	1.5 yr-retention
BMI (kg/m ²)	17.1	18.8	22.5	22.0	20.8
Sleep efficiency (%)	96.2	98.5	98.4	99.8	99.8
AHI	24.8	0.0	29.2	10.5	6.0
RERA	9.6	9.8	2.4	2.8	2.6
RDI	34.4	9.8	31.6	13.3	8.6
Supine AHI	24.9	0.0	35.2	10.5	6.4
LSaO ₂ (%)	81.0	95.0	81.0	89.0	90.0

MAD, Mandibular advancement device; BMI, body mass index; AHI, apnea-hypopnea index; RERA, respiratory effort-related arousal; RDI, respiratory disturbance index; LSaO₂, lowest oxygen saturation.



Figure 1. Pretreatment facial and intraoral photographs.



Figure 2. Pretreatment dental casts.

mandibular disorder, and exhibited a favorable condylar shape and position on cone-beam computed tomography (CBCT) analyses.

TREATMENT OBJECTIVES

The primary objective was to relieve the symptoms

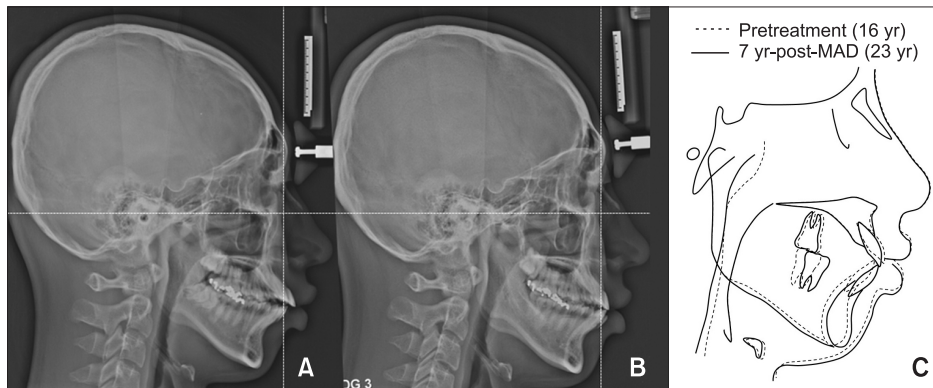


Figure 3. Lateral cephalograms. **A**, Pretreatment. **B**, After 7 years of wearing the mandibular advancement device (MAD). **C**, Superimposition of **A** and **B**.

of OSA by using temporary mandibular advancement to open the airway during sleep until the cessation of growth. The final objectives were to (1) treat the OSA permanently by surgical MMA with surgical flattening of the occlusal plane; (2) obtain an esthetically pleasing appearance by increasing throat length; and (3) establish favorable occlusion while maintaining incisal inclination, in order to avoid interrupting the tongue space.

TREATMENT ALTERNATIVES

The following treatment options were considered: (1) early MMA surgery during post-adolescence; (2) delayed MMA treatment after growth completion, with pre-surgical 4-premolar extraction treatment; (3) delayed MMA treatment with a surgery-first approach (SFA), followed by 4-premolar extraction treatment; (4) delayed MMA treatment with SFA, followed by mandibular incisor extraction, with temporary MAD support until the time of surgery.

MMA was chosen as radical osseo-pharyngeal reconstruction, considering the patient's definite craniofacial deformity, which contributed to his severe OSA symptoms without obesity or para-pharyngeal soft tissue deformities. Regarding the timing of surgery, it was delayed until mandibular growth had ended and the patient could afford the overall medical expenses; until that time, MAD was prescribed as a temporary aid. Pre-surgical orthodontic treatment, combined with regular premolar extraction, was omitted for this patient; such treatment was expected to aggravate symptoms of OSA by pushing the tongue backwards. Thus, SFA followed by atypical extraction treatment (the fourth option) was chosen for this patient with OSA.

TREATMENT PROGRESS

The MAD was fabricated with one-piece vacuum-formed splints (Forestadent, Pforzheim, Germany) in the calibrated mandibular position, with a setting of 6 mm

of advancement (65% of patient's maximum protrusion); this allowed 3 mm of vertical opening between the maxillary and mandibular incisor tips. MAD was designed not to cover the mandibular incisors, due to the potential for their unwanted proclination. The patient adapted to this initially guided mandibular position without requiring additional adjustment; he wore the appliance every night and reported that his subjective symptoms had nearly disappeared. MAD-titration polysomnographic analysis revealed that his AHI had decreased to zero, which indicated complete relief of airflow obstruction; this ruled out the possibility of non-anatomic vulnerability. RDI decreased from 34.4 to 9.8, and $LSaO_2$ improved from 81.0% to 95.0% (Table 1).

Seven years later, the patient decided to undergo MMA surgery as originally planned; his facial, skeletal, and dentoalveolar changes were noticeable. Unexpected forward mandibular growth occurred even after the period of peak height velocity, decreasing his facial convexity and improving his chin profile (Figure 3B and 3C). Molar and canine relationships were shifted from Class I to Class III, accompanied by mesial movement of the mandibular posterior teeth (Figures 4 and 5). Maxillary incisors were retroclined, reducing overjet and overbite. Mandibular incisor inclination was maintained, while incisor irregularity was increased. The patient's AHI had increased to 29.2, possibly due to his increased BMI, despite forward mandibular growth after long-term MAD use (Table 1).

Regarding the surgical treatment objective, maxillo-mandibular rotational advancement with advancing genioplasty was planned such that respiratory functional enhancement by sufficient airway expansion could be obtained in combination with an esthetically pleasing appearance. Advancing genioplasty was chosen in order to compromise insufficient chin advancement by limited counterclockwise jaw rotation, due to the lack of maxillary incisor exposure. A SFA was applied because patients with OSA may exhibit aggravated symptoms during the pre-surgical orthodontic period, due to the

Table 2. Comparison of the lateral cephalometric measurements of craniofacial, dentoalveolar, and pharyngeal airway parameters among the four observation time points

Cephalometric parameter	Initial	7 yr-post-MAD	Post-treatment	1.5 yr-retention
Craniofacial				
Nper-A (mm)	-2.5	-1.5	2.0	2.0
Nper-B (mm)	-18.0	-16.0	-8.0	-8.0
Nper-Pog (mm)	-17.0	-15.0	-4.0	-3.5
SNA (°)	78.0	78.5	82.0	82.0
SNB (°)	72.0	74.0	78.0	78.0
ANB (°)	6.0	4.5	4.0	4.0
MPA (°)	35.0	35.0	32.0	32.5
PFH/AFH	61.5	61.0	62.5	62.5
OP-FH (°)	12.0	14.0	11.0	11.0
Dentoalveolar				
U1-FH (°)	122.0	113.0	118.0	118.0
IMPA (°)	100.0	101.0	96.0	96.0
IIA (°)	103.0	114.0	115.0	115.0
Overjet (mm)	4.0	1.0	2.5	2.5
Pharyngeal airway				
PNS-ad1 (mm)	27.5	27.0	31.0	31.0
PNS-ad2 (mm)	21.0	24.5	26.0	27.0
SPAS (mm)	12.0	13.0	16.0	17.0
MAS (mm)	8.0	9.0	13.0	14.0
IAS (mm)	7.0	8.0	12.0	12.0
VAL (mm)	74.0	75.0	72.0	72.0
MPH (mm)	23.0	20.0	20.0	21.0

MAD, Mandibular advancement device; Nper-A, perpendicular distance between A point and nasion perpendicular line; Nper-B, perpendicular distance between B point and nasion perpendicular line; Nper-Pog, perpendicular distance between pogonion and nasion perpendicular line; SNA, angle between lines sella-nasion and nasion-point A; SNB, angle between lines sella-nasion and nasion-point B; ANB, angle between lines point A-nasion and nasion-point B; mandibular plane angle (MPA), angle between lines gonial-menton and Frankfort horizontal line; posterior facial height/anterior facial height (PFH/AFH), ratio between linear distance from sella to gonion and from nasion to menton; OP-FH, angle formed by the lines between occlusal plane and Frankfort horizontal line; U1-FH, angle formed by the lines between long axis of upper incisor and Frankfort horizontal line; incisor mandibular plane angle (IMPA), angle formed by the lines between long axis of lower incisor and mandibular plane angle; inter incisor angle (IIA), angle formed by the lines between long axis of upper incisor and lower incisor; PNS-ad1, linear distance from posterior nasal spine to the point ad1 (ad1, intersection of the line PNS-Basion and the posterior nasopharyngeal wall); PNS-ad2, linear distance from posterior nasal spine to the point ad2 (ad2, the intersection of the posterior nasopharyngeal wall and the midpoint on the line joining sella and basion); superior posterior airway space (SPAS), the thickness of the airway behind the soft palate along a line parallel to gonion-point B; middle airway space (MAS), the thickness of the airway along a line parallel to gonion-point B through the lowest point of soft palate; inferior airway space (IAS), the thickness of the airway along a line extended through gonion-point B; vertical airway length (VAL), the linear distance between posterior nasal spine to epiglottis base; mandibular plane to hyoid (MPH), linear distance along a perpendicular from hyoid bone to the mandibular plane.

reduced oral volume and subsequent backward displacement of the tongue by mandibular incisor retraction.¹⁴ Computer-assisted three-dimensional surgical planning was performed using Simplant Pro software (Materialize Dental, Leuven, Belgium); three-dimensionally printed

surgical wafers and a surgical guide for advancing genioplasty were fabricated (Figure 6).



Figure 4. Facial and intraoral photographs after 7 years of mandibular advancement device use. Facial convexity was reduced, while the Class III molar and canine relationship with shallow overjet and overbite showed progression.



Figure 5. Dental casts after 7 years of mandibular advancement device use.

RESULTS

As a result of surgery, the maxilla was advanced by 4.5 mm at the A-point level; its counterclockwise rotation

moved the maxillary incisor tip upward by 2.0 mm and forward by 5.5 mm. Surgical flattening of the occlusal plane by 3° (angle formed by the lines between occlusal plane and Frankfort horizontal line: 14° to 11°) was ac-

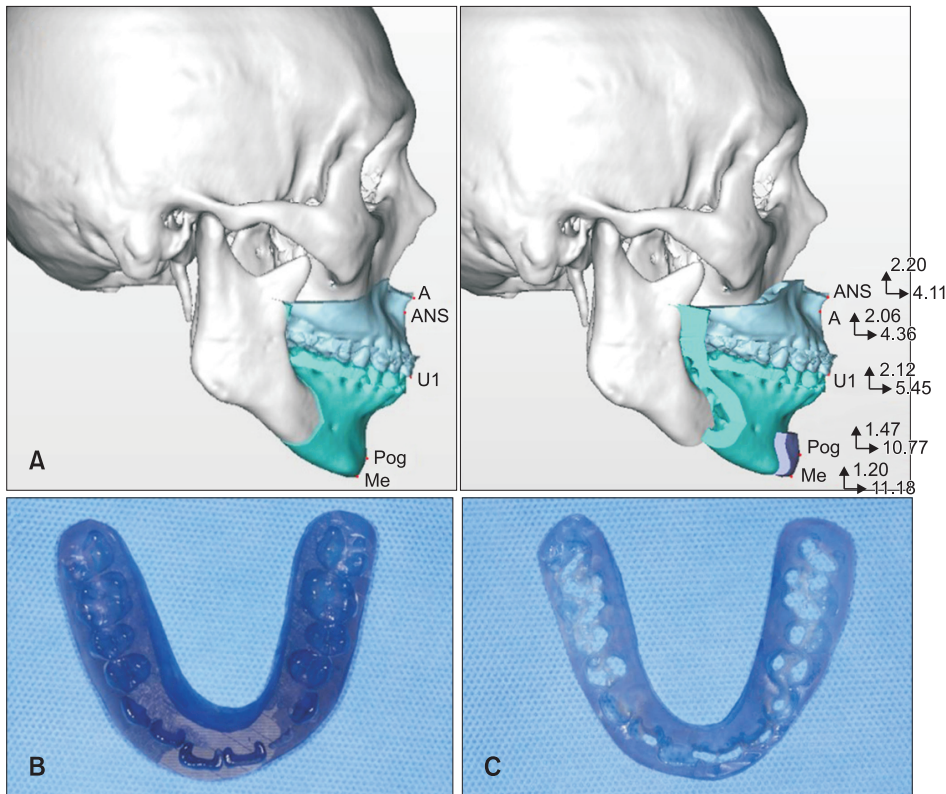


Figure 6. Computer-assisted three-dimensional (3D) planning of maxillomandibular advancement surgery with genioplasty using Simplant Pro software (Materialize Dental, Leuven, Belgium) (A), 3D printed intermediate wafer (B), and final wafer (C). A, Point A; ANS, anterior nasal spine; U1, upper central incisor; Pog, pogonion; Me, menton.

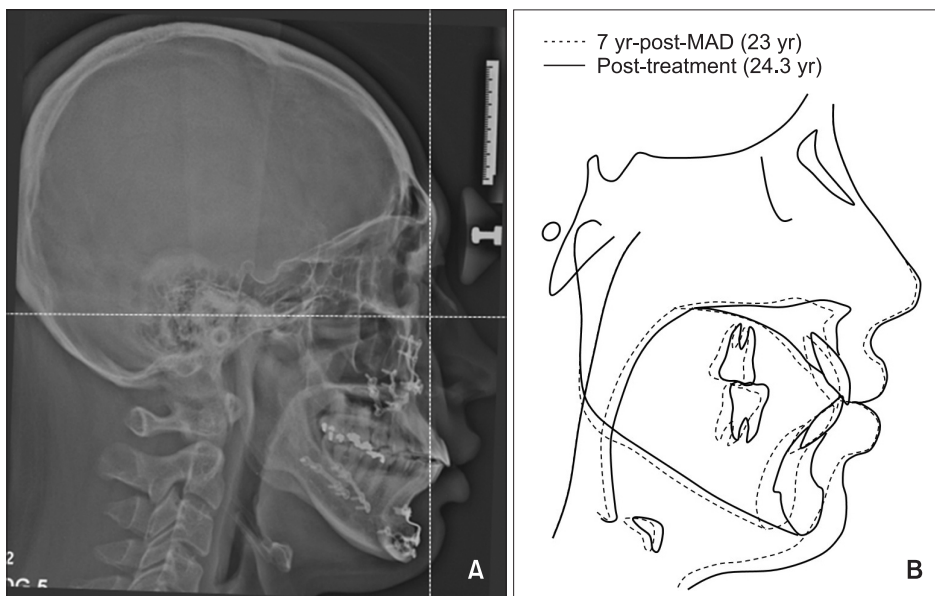


Figure 7. A, Lateral cephalograms taken at post-treatment. B, Superimposition of alignment after 7 years of wearing the mandibular advancement device (MAD) (7 yr-post-MAD) and post-treatment.

accompanied. The chin point was advanced by 11 mm; the hyoid bone was displaced forward and upward, opening the pharyngeal airway (Figure 7). At 6 weeks after surgery, post-surgical orthodontic treatment was initiated, including atypical single mandibular incisor extraction. Because the post-surgical orthodontic goal included relieving the lower anterior crowding with arch form

modification, the anteroposterior position of the incisors was maintained and Class II molar relationship was compensated (Figures 8 and 9). Twelve months of treatment ended with reduction of AHI from 29.2 to 10.5 (64%), reduction of RDI from 31.6 to 13.3, and enhancement of LSAO₂ from 81.0% to 89.0% (Table 1). Post-retention records at 1.5 years after surgery showed stable occlu-



Figure 8. Post-treatment facial and intraoral photographs.



Figure 9. Post-treatment dental casts.

sion with a favorable facial profile (Figure 10 and Table 2), as well as further improvement of AHI (reduction to 6.0; 79.5%) and $LSaO_2$ (enhancement to 90.0%). Based on comparative analysis of CBCT images (version 4.0.70,

InVivoDental; Anatomage, San Jose, CA, USA), total pharyngeal volume and minimal cross-sectional area were increased by 68.9% and 62.5%, respectively. The incremental ratio was greater in the retroglottal area

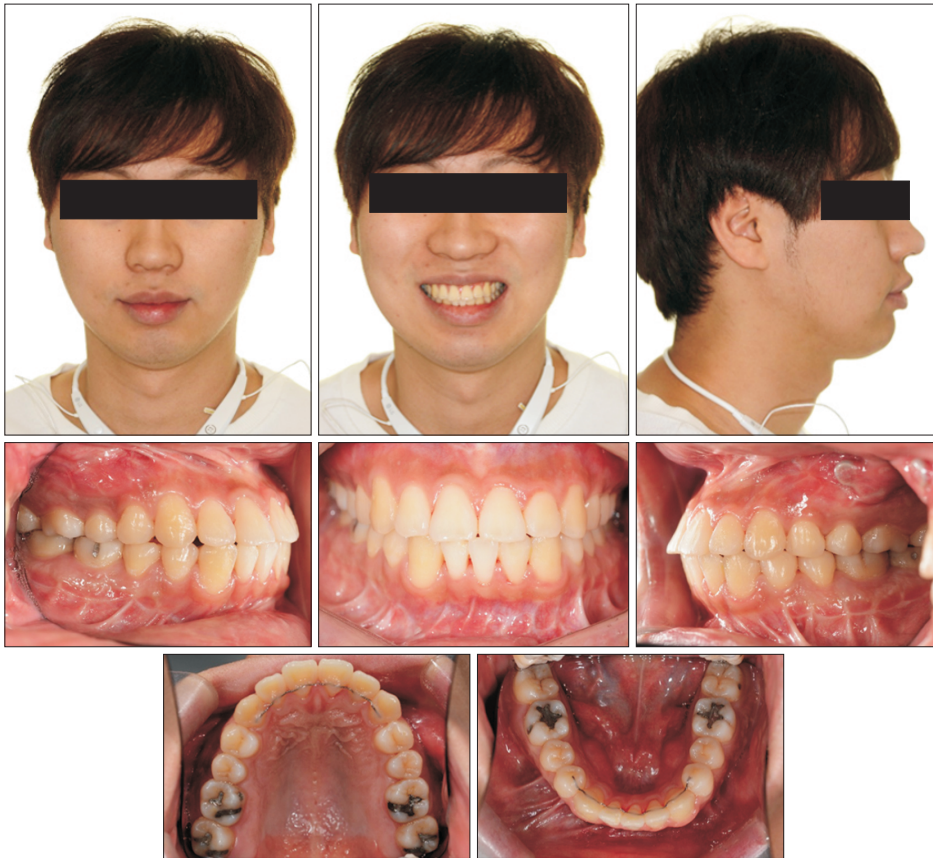


Figure 10. Facial and intra-oral photographs at 1.5 years post-retention.

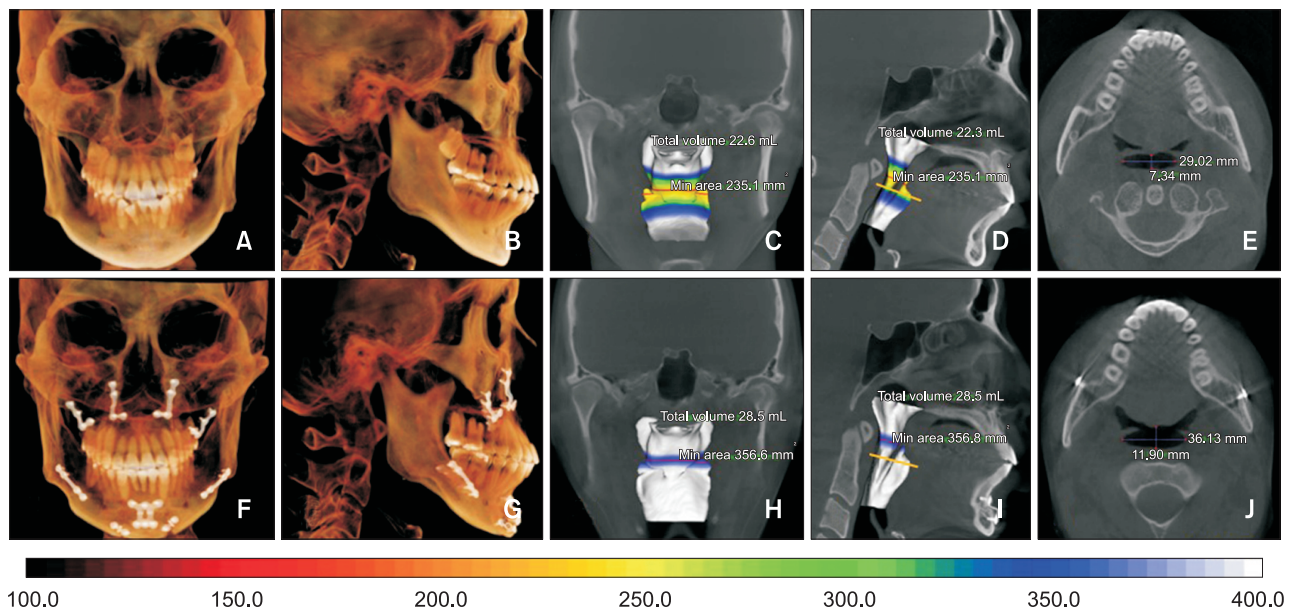


Figure 11. Comparative cone-beam computed tomography images of the craniofacial and pharyngeal airway in three dimensions at pretreatment (A-E) and 1.5 years post-retention (F-J).

Table 3. Post-retention changes of pharyngeal airway measurements on cone-beam computed tomography volume images

Airway	Pretreatment	1.5 yr-post-retention	Increment ratio (%)
Retropalatal volume (cm ³)	12.4	24.2	95.2
Retroglossal volume (cm ³)	10.4	26.3	152.9
Total pharyngeal volume (cm ³)	22.8	38.5	68.9
Minimum cross-sectional area (mm ²)	238.1	386.8	62.5

(152.9%) than in the retropalatal area (95.2%) (Figure 11 and Table 3). The patient reported that his sleep was undisturbed and he experienced complete resolution of excessive daytime sleepiness.

DISCUSSION

It is unclear which treatment option is best for post-adolescent patients with OSA who exhibit craniofacial vulnerability. According to the orthodontic therapeutic protocol for adults with OSA, the MMA approach was indicated for the patient in this case because he exhibited a phenotype involving anatomic vulnerability due to craniofacial deformity, without para-pharyngeal deformity^{1-5,8-11,14}. However, the patient might have been in a growth stage that might have affected the results of early orthognathic surgery, and his symptoms were sufficiently refractory to require early surgery. However, following the established therapeutic protocol for growing patients with OSA,² functional appliances may be indicated for children in anticipation of permanent mandibular growth; this also allows opening of the airway.¹⁵ However, this consideration was not applicable for the post-adolescent patient in this case. Considering that MAD, which is typically selected for elderly patients with OSA, resembles a functional appliance used for pre-adolescent children in its ability to open the airway, MAD appears to be a good temporary option for improving the quality of life of a post-adolescent patient with OSA, regardless of its suspected growth modification effects.

Regarding the significance of MAD use prior to the final MMA surgery, MAD therapy might be a good predictor of a successful outcome in MMA surgery.^{11,16} This noninvasive phased protocol has demonstrated great value in minimizing irrelevant MMA surgery for OSA caused by non-anatomic vulnerability,¹¹ as well as allowing the patient to be accustomed to the mechanism of surgical mandibular advancement to open the airway, and allowing the surgeon to quantitatively estimate the amount of jaw advancement.¹⁶ Thus, MAD was a good predictor for a successful outcome of MMA in this case, because the patient showed an optimal response to MAD in that his AHI decreased to zero. Interestingly, in addition, the patient showed craniofacial and dentoalveolar

changes induced by long-term MAD use; thus, there are three points to consider when MMA surgery is finally performed: surgical design, surgical timing, and post-surgical orthodontic strategy.

First, additional forward and downward growth of the mandible may occur, which reduces the amount of surgical jaw advancement. Although this skeletal change cannot be anticipated in every patient—and it remains controversial whether this mandibular growth is fully stimulated by the appliance¹⁷—possible residual growth with the use of MAD must be considered. OSA might be resolved if sufficient mandibular growth to enlarge the entire pharynx can be attained by use of the appliance.¹⁸ If this is not achieved, MMA surgery is still needed for the sufficient advancement of both the maxilla and the mandible. This patient exhibited increased AHI when re-evaluated after use of MAD, due to his increased BMI, despite mandibular forward growth. Accordingly, a modified MMA approach should be considered to achieve airway enlargement without compromising the facial profile. Segmental osteotomy or modified genioplasty can be combined with counterclockwise jaw rotation, in order to achieve maximum advancement of the chin and posterior section of the maxilla, thereby increasing airway patency.¹⁹

Second, the progression of dental compensation by MAD, towards Class III molar relationship with overjet reduction, requires longer orthodontic decompensation before MMA; this is troublesome to patients with OSA. The timing of surgery is especially important in such patients to achieve early resolution of respiratory problems. Therefore, SFA is widely used to enhance sleep quality first without enduring aggravated OSA symptoms during the pre-surgical decompensation period.¹⁴ SFA is also valuable for patients with OSA because the total treatment time tends to be reduced by omitting the pre-surgical orthodontic stage, by immediate surgical correction of most problems due to substitution of durational orthodontic correction, and by rapid post-surgical tooth movement through regional acceleration.²⁰ However, the compensated dental Class I or III relationship disturbs sufficient mandibular advancement, relative to the maxilla with SFA. For successful SFA to sufficiently open the airway, a modified surgical design is required to establish

stable post-surgical occlusion; if this modification is not made, post-surgical orthodontic goals will be compromised. Computer-aided three-dimensional surgical planning and post-surgical prediction are highly efficient and accurate approaches in SFA for OSA patients.

Third, as a post-surgical orthodontic strategy, the potential for extraction is prudent. Without the use of anterior segmental osteotomy, which allows maximum protraction of the posterior maxilla and the chin, non-extraction is preferred in patients with OSA, in order to retain expanded pharyngeal volume after surgery^{21,22} and to complete treatment as rapidly as possible. The results of long-term MAD use included reduced maxillary incisor inclination, enhanced mandibular incisor inclination with reduced overjet and overbite, and Class III molar relationship. In this patient, single mandibular incisor extraction was applied based on the mild maxillary arch length discrepancy; this changed the inclination of both incisors and reduced overjet; moreover, it increased mandibular incisor irregularity, stabilized Class III posterior occlusion, and achieved a favorable Bolton's ratio (anterior Bolton's ratio: 74.0%) and post-surgical facial profile. Despite the presence of proclined incisors with shallow overjet and overbite, the compromised final occlusion was acceptable for this patient, and it has remained stable at 1.5 years post-retention with good periodontal support.

This patient was successfully treated by MMA-assisted orthodontics, based on the surgical success criteria (> 50% reduction of baseline AHI, with post-treatment AHI < 20).²³ Polysomnography analysis confirmed the relief of subjective symptoms, consistent with CBCT morphometric findings showing three-dimensional pharyngeal enlargement. In contrast, MMA may fail in some patients with craniofacial vulnerability, including those who exhibited non-anatomical vulnerability, such as neuromuscular dysfunction. Therefore, in addition to MAD-titration based prediction, craniofacial morphometric analysis and target-based intervention by orthodontists are indispensable for interdisciplinary treatment of OSA. Orthopedic and orthognathic approaches constitute treatment methods for patients with OSA who exhibit definite craniofacial vulnerability, based on the differential diagnosis of the patient's phenotype, primary causative factors, and primary therapeutic targets within craniofacial compartments. Orthodontists should choose intervention or referral to sleep doctors with close collaboration.

CONCLUSION

In conclusion, this case report demonstrated successful treatment with phased MAD and MMA application in a post-adolescent patient with OSA who exhibited

craniofacial vulnerability. This patient showed skeletal changes and progression of dental compensation induced by long-term MAD use throughout post-adolescence, yielding three clinical considerations for planning of subsequent MMA treatment regarding the design and timing of surgery, as well as the post-surgical orthodontic strategy. This suggests a therapeutic protocol for use in patients with OSA in terms of collaborations between orthodontists and sleep doctors.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

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