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Long-Term Management of a Patient With Obstructive Sleep Apnea Using Multimodal Combination Treatment

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Received: 16 March 2024 | **Revised:** 20 February 2025 | **Accepted:** 3 March 2025

Funding: This work was supported by Japan Society for the Promotion of Science (KAKENHI No. 23K09326).

Keywords: continuous positive airway pressure | mandibular advancement | obstructive sleep apnea | orthognathic surgery | treatment

ABSTRACT

In this case, we combined oral appliance, continuous positive airway pressure, and sleep surgery for the management of obstructive sleep apnea. It will require a combination of therapies according to the patient's chief complaint and symptoms and not just one therapy for the long-term management of obstructive sleep apnea.

1 | Introduction

Continuous positive airway pressure (CPAP) and oral appliance (OA) are widely known treatments for obstructive sleep apnea (OSA). Recently, sleep surgeries, such as maxillomandibular advancement (MMA) and genioglossus advancement (GA), which are fundamental treatments for OSA, have become widespread. Besides these, there are multiple treatments for OSA [1, 2].

Each treatment for OSA has advantages and disadvantages. CPAP and OA are minimally invasive; however, patient adherence must be maintained to achieve therapeutic effects. In contrast, sleep surgery provides a therapeutic effect without a device but is more invasive. While some reports on the long-term persistence of the therapeutic effects are available, the permanence of these effects is unknown [3]. Notably, as management with a single treatment modality is difficult for the long-term management of OSA treatment, patients must be managed with a combination of multiple therapies depending on their symptoms.

Herein, we report a case of long-term management of a patient with OSA using a combination of OA, CPAP, and sleep surgery.

2 | Case History

A 32-year-old man visited a sleep clinic in 2010 with the main complaint of sleepiness and difficulty staying awake. Polysomnography (PSG) showed an apnea-hypopnea index (AHI) of 8.1/h and a supine AHI of 19.1/h. His body weight (BW) was 76.4 kg. The patient was diagnosed with mild OSA with positional dependence, and OA therapy was then initiated. The mandible position of OA was set at 7 mm forward (70% mandibular advancement). In 2011, the patient underwent PSG with OA, and his AHI was 0.8/h (BW was 79.5 kg).

As the OA treatment led to an improvement in sleepiness, it was continued. However, the patient became aware of occlusal changes caused by 8 years of OA use in 2018. Comparing the 2010 and 2018 cephalography images, the overjet changed from 4 to 0 mm, and the overbite changed from 1 to 0 mm. The

inclination of the anterior maxillary teeth (U1-SN; upper incisor angle to SN plane) changed from 101.5° to 91.0°, that of the anterior mandibular teeth (L1-MP; lower incisor angle to mandibular plane) changed from 94.5° to 98.5°, and the occlusal relationship of the first molars changed from Angle Class I to Class III (Figure 1a; Before OA treatment of cephalometric radiograph, Figure 1b; after 8 years OA treatment of cephalometric, and Table 1).

PSG was performed again in 2019 without OA to identify adaptation to treatments other than OA. PSG showed a worsening AHI of 12.2/h and a supine AHI of 36.3/h (BW was 75.4 kg). The patient desired treatment for the occlusal changes and OSA; thus, he visited our Center for Dental Sleep Medicine in 2019.

3 | Methods

When the patient visited our center, he had asked for OSA and occlusal treatment. He had perceptions of the effectiveness of OA treatment for OSA (reduces snoring and improves sound sleep) and was concerned about occlusal changes because another dentist told him that he had occlusal change and needed occlusal treatment. To find a treatment that would improve the patient's chief complaint, we performed endoscopy and cephalography. Endoscopy did not reveal tonsillar hypertrophy or overlengthening of the soft palate; however, cephalography revealed a small mandible. Therefore, we planned orthodontic treatment for occlusal changes, as well as sleep surgery with MMA and GA for OSA and small mandible.

The OA therapy changed the patient's occlusion, and his jaw position was unstable; therefore, we decided to initiate orthodontic treatment in the Department of Orthodontics at our hospital. The patient had been using OA to treat his daytime sleepiness; when the patient slept while using OA, his Epworth Sleepiness Scale (ESS) score was 3, whereas his ESS score was 10 when he did not use OA. During the orthodontic treatment, the patient could not use the OA because he was wearing orthodontic appliances; thus, he needed another treatment for OSA. His OSA was eventually treated using automatic CPAP available in the Department of Internal Medicine at our hospital, delivering a minimum pressure of 4 cm H_2O and a maximum pressure of 8 cm H_2O. CPAP adherence during the orthodontic treatment (2019–2021) was good (AHI 0.9–1.5/h; number of days used per month, 97%; and number of days of CPAP use, over 4 h/month, 63%).

After the preoperative orthodontic treatment was completed, the amount of jawbone movement by the MMA was determined using an articulator mounted on the upper and lower jaw models attached to the articulator based on the Frankfurt plane, Sassouni analysis, and the following four points [4]:

(i) the therapeutic effect of OA was shown with a 7 mm anterior movement of the mandible, (ii) the pharyngeal airway was expanded by the maxilla moving horizontally by at least 3 mm and upward by at least 4 mm [5], (iii) the length between the nasal floor and the inferior nasal turbinate was 3 mm, and (iv) the patient did not want facial changes due to excessive jawbone movement. Considering these points, the maxilla was moved 5 mm horizontally and 3 mm upward via Le Fort I. Subsequently, the mandible was moved 8 mm anteriorly on the right side and 6 mm anteriorly on the left side using bilateral sagittal split ramus osteotomy (SSRO), and the mental was moved 5 mm horizontally for postoperative cosmetic reasons.

4 | Conclusions and Results

After completing the orthodontic treatment, the patient underwent MMA and GA (Le Fort I for the maxilla and SSRO and genioplasty for the mandible) under general anesthesia in 2022 (Figure 1c; After sleep surgery of cephalometric radiograph).

A sleep test (type 3) was performed 2 months after surgery; the respiratory event index (REI) was 2.3/h, and the supine REI was 0.8/h (BW was 72.0 kg). As the effect of the surgery was observed, the CPAP treatment was stopped. A comparison of preoperative and 6-month postoperative computed tomography (CT) scan airway morphology showed that both the surface area of the narrowest area and airway volume were expanded (Figure 2a; before surgery, Figure 2b; after surgery) Airway volume and minimum axial area were evaluated by measuring between

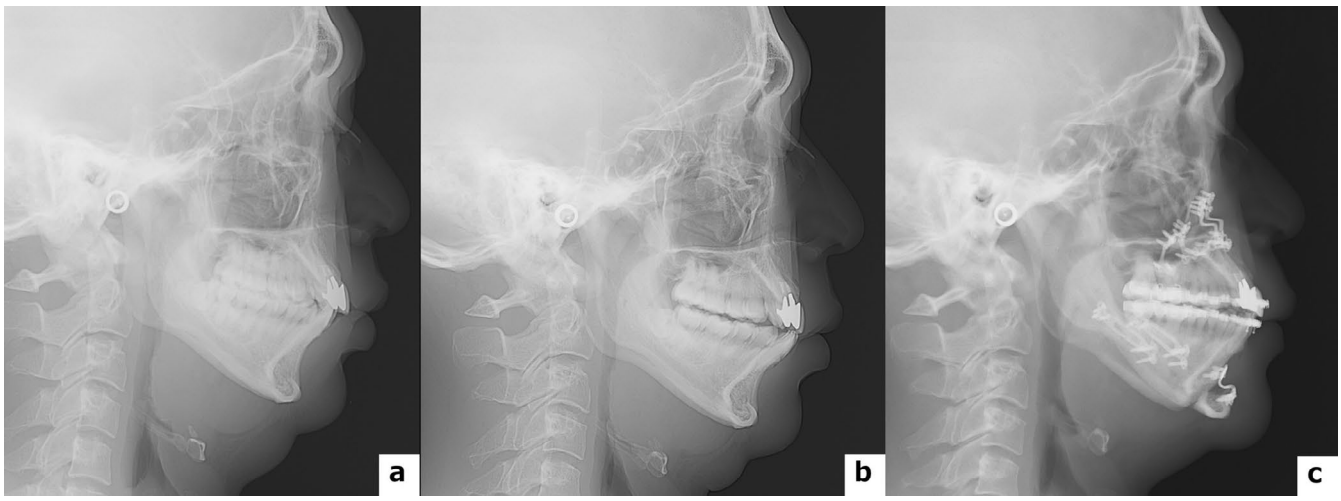

FIGURE 1 | Cephalometric radiographs. (a) Before treatment. (b) After oral appliance treatment. (c) After sleep surgery (6 month).

TABLE 1 | Result of polysomnography and cephalometric measurements.

	Before treatment	After OA treatment (with OA)	After OA treatment (no OA)	After surgery (2 month)	After surgery (6 month)
Age (year)	24	25	32	35	36
BW (kg)	76.4	79.5	75.4	72	73
BMI (kg/m^2)	24.8	25.8	24.6	23.4	23.7
ESS score	16	4	3	14	5
Result of sleep test					
Type of sleep test	Type 1	Type 1	Type 1	Type 3	Type 1
AHI (/h)	8.1	0.8	12.7	[2.3]	15.4
AI (/h)	0.2	0	6.5	[2.0]	5.3
HI (/h)	8.0	0.8	6.3	[0.3]	10
Supine AHI (/h)	19.1	0.6	36.3	[0.8]	13.9
LowestSpO_2 (%)	88	93	88	90	90
3% ODI (/h)	6.0	0.8	12.2	2.3	11.8
SE (%)	76.0	67.2	72.7		91.1
Sleep stage N1 (%)	10.6	12	4.1		11.2
Sleep stage N2 (%)	53.7	78.2	74.7		53.7
Sleep stage N3 (%)	23.4	0.3	6.7		17.1
Sleep stage REM (%)	12.3	9.5	14.5		17.9
Cephalometric measurements					
SNA (°)	79.5		79.1		84.3
SNB (°)	73.0		71.0		77.6
Fx (°)	78.0		76.4		82.4
U1-SN (°)	101.5		91.0		104.1
L1-MP (°)	94.5		98.5		96.4
PNS-P (°)	32.4		30.7		33.9
MPT (mm)	10.2		10.1		10.7
MP-H (mm)	30.2		22.7		27.1
SPAS (mm)	9.8		11.9		15.9
MAS (mm)	5.8		8.7		10.8
IAS (mm)	7.5		7.9		12.0

Abbreviations: AHI, apnea-hypopnea index; AI, apnea index; BMI, body mass index; BW, body weight; ESS, Epworth Sleepiness Scale; HI, hypopnea index; OA, oral appliance; OCST, out-of-center sleep testing; ODI; oxygen desaturation index; REM, rapid eye movement; SE, sleep efficiency. Cephalometric measurements: Fx, angle between the line from Ba to N and the line from Pt to Gn; IAS, the thickness of the airway along a line extending through the Go-B line plane; L1-MP, lower incisor angle to mandibular plane (MP); MAS, the thickness of the airway along a line parallel to the Go-B point plane through P; MP-H, linear distance between the MP and H; SPAS, the thickness of the airway behind the soft palate along a line parallel to the Go-B point plane; MPT, maximum thickness of the soft palate along a line perpendicular to PNS-P; PNS-P, distance between PNS and P; SNA, angle between the nasion-sella line (SN) and the line from A to N; SNB, angle between SN and the line from B to N; U1-SN, upper incisor angle to SN. Cephalometric landmarks: A, deepest anterior point in the concavity of the maxilla; B, deepest anterior point in the concavity of the mandible; Ba, most inferior point in the sagittal plane on the anterior rim of the foramen magnum; Gn, deepest antero-inferior point of the body chin; Go, mid-plane point at the gonial located by bisecting the posterior borderline of the mandible; H, most antero-superior point of the hyoid bone; Me, most inferior point of the mandible; N, anterior point of the frontonasal suture; P, most inferior tip of the soft palate; PNS, most posterior point of the hard palate; Pt, point at junction between the Ptm and foramen rotundum; S, midpoint of the fossa hypophysialis.

the most posterior point of the hard palate (PNS) and epiglottal base(Eb). Furthermore, PSG showed a significant improvement in supine AHI from 36.3/h to 13.9/h (BW was 73.0 kg), and sleep efficiency improved from 72.7% to 91.1% (Table 1). Currently, the patient's sleepiness and difficulty staying awake are improved without using OA or CPAP, and he is still being followed up.

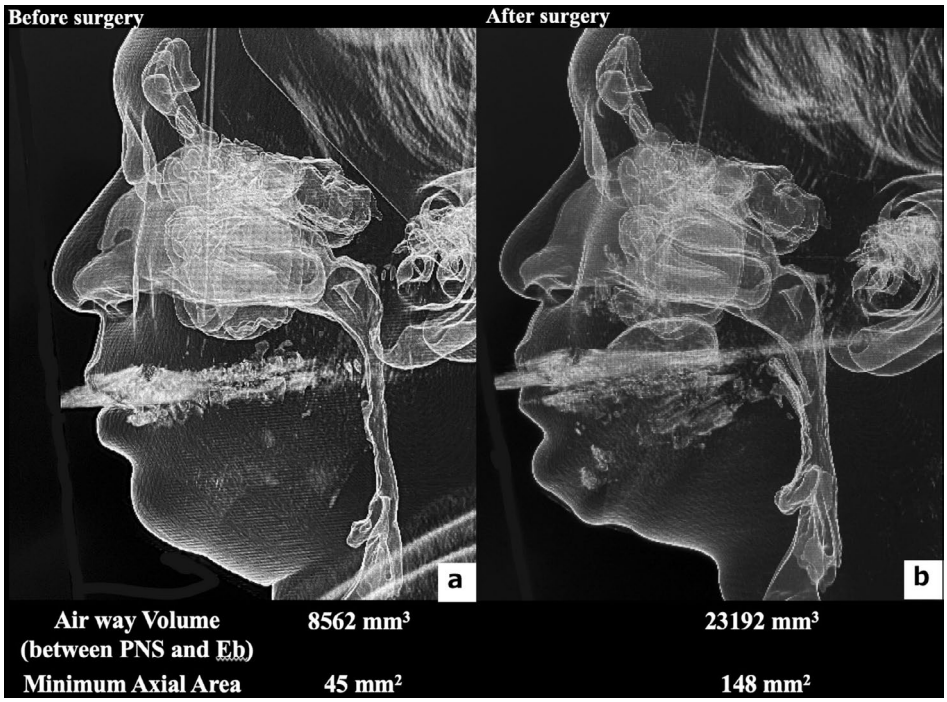

FIGURE 2 | The airway volume and minimum axial area between PNS and Eb were measured. Airway volume changed from 8562 to 23,192 mm^3 by sleep surgery. Minimum axial area changed from 45 to 148 mm^2 by sleep surgery. Data analyses were performed using Dolphin 3D imaging software (Dolphin Imaging & Management Solutions, Chatsworth, CA, USA). (a) Before surgery. (b) After surgery. Eb, epiglottal base; PNS, most posterior point of the hard palate.

5 | Discussion

In this case, we combined OA, CPAP, and sleep surgery for the long-term management of a patient with OSA. Long-term management with OSA therapy requires a combination of therapies according to the patient's chief complaint and symptoms and not just one therapy.

OA is less effective than CPAP in patients with OSA; however, its adherence is superior [6]. OA is used for mild-to-moderate OSA or in cases where CPAP is not indicated. Occlusal changes are one of the side effects of OA. Because the OA is a device that moves the mandible, it forces the teeth during use. When OA is used, the mandible tends to return to its natural position at night, which causes a palatal force to be applied to the maxilla and a labial force to be applied to the mandibular teeth. Therefore, long-term use of OA has been reported to cause changes in the overjet, overbite, and inclination of the anterior maxillary and mandibular teeth [7, 8]. Similar occlusal changes were observed in this case owing to the long-term use of OA.

Because occlusal changes are irreversible, we have to stop the OA treatment if the patient wants occlusal treatment. Dental treatment for malocclusion includes prosthetic and orthodontic treatments; orthodontic treatment is recommended when many natural teeth remain, as in this case. However, restarting OA for OSA even after orthodontic treatment improves occlusion and may cause occlusal changes. Therefore, in this case, we believe that orthodontic treatment alone would make it challenging to manage OSA in the future and consider the curative treatment of OSA by sleep surgery.

This patient had mild OSA that was position-dependent, with a supine AHI of 36.3/h before orthodontic treatment. As the use of OA improved sleepiness and the treatment of OSA was shown to be effective, we considered that the cause of OSA was strongly due to anatomical factors and that curative treatment by surgery was feasible [9]. Because the amount of maxillomandibular movement to effect treatment for OSA is unknown, in this case, the amount of mandibular advancement by OA treatment, the amount of maxillary movement that would expand the pharyngeal airway, and the patient's facial requirements were considered. Surgery did not lead to a radical cure for OSA; however, the supine AHI greatly improved. In addition, the patient did not feel sleepiness postoperatively without devices such as OA or CPAP, and his sleep efficiency improved. There were no complaints of postoperative changes in the facial appearance (Figure 3a; before sleep surgery, Figure 3b; after sleep surgery), and the patient is currently being followed up without using any treatment device.

In this case, we combined OA, CPAP, and sleep surgery for the management of a patient with OSA, and the patient was doing well after treatment. However, as the patient ages and gains weight, his OSA may worsen again in the future. Recently, identifying the reasons for OSA (pharyngeal anatomy/collapsibility, upper-airway muscle responsiveness, loop gain, and arousal threshold) in individual patients and providing treatment for OSA that targets one or more of these reasons has been proven important [10]. The treatment we performed in this case (OA, CPAP, and sleep surgery) has affected the pharyngeal anatomy/collapsibility and eventually improved the patient's OSA. However, pharyngeal anatomy/collapsibility is not the only factor affecting OA, and if the patient's OSA becomes worse

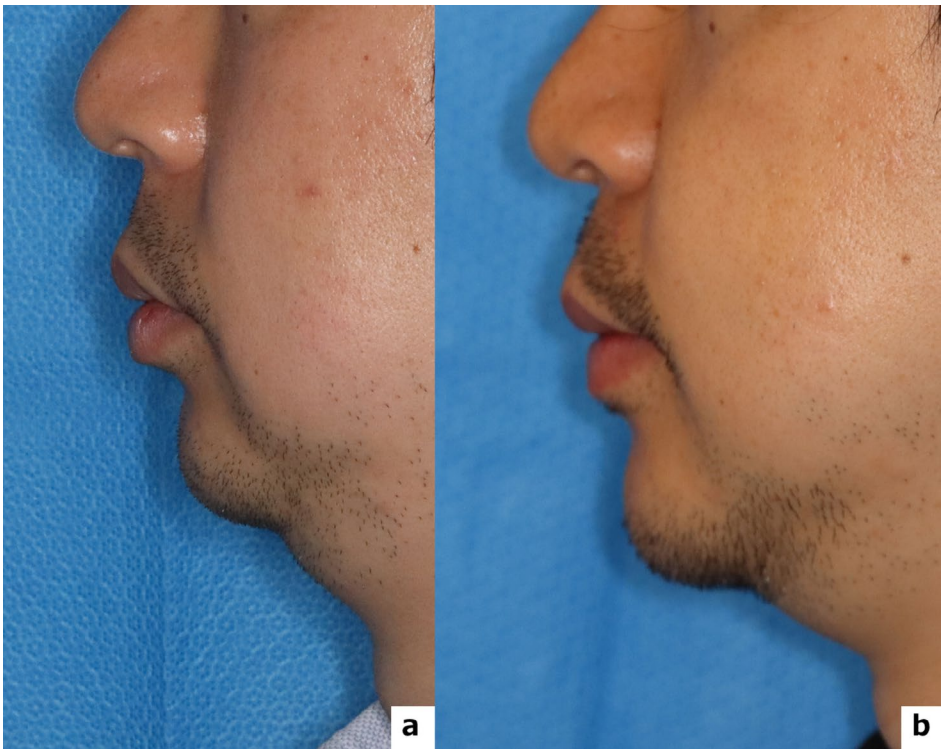

FIGURE 3 | Photograph of the facial appearance. (a) Before sleep surgery. (b) After sleep surgery.

after treatment of pharyngeal anatomy/collapsibility, we have to treat other reasons for OSA. Oropharyngeal exercises and upper airway stimulation are OSA treatments that are currently gaining attention [11, 12]. Both treatments have been effective in upper-airway muscle responsiveness for OSA. Oropharyngeal exercises are effective in moderate OSA, whereas upper airway stimulation is a surgical option for select patients who cannot use CPAP. We believe both treatments may be effective for the residual OSA and exacerbations of OSA after surgery in this case. In summary, OSA requires long-term management, which requires a combination of therapies depending on the patient's chief complaint and symptoms and not just one therapy.

Author Contributions

Akio Himejima: conceptualization, formal analysis, funding acquisition, investigation, project administration, writing – original draft. **Kentaro Okuno:** conceptualization, funding acquisition, investigation, project administration, resources, supervision, writing – review and editing. **Hayato Ikeda:** investigation. **Maho Kono:** formal analysis, investigation. **Hiroaki Yoshida:** investigation. **Yuji Nakayama:** investigation. **Hideo Shimizu:** investigation, resources. **Morio Tonogi:** supervision.

Acknowledgments

We thank Editage (www.editage.com) for English language editing.

Consent

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

The datasets generated and analyzed during the current study are available from the corresponding author upon reasonable request.

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