Maxillomandibular Advancement Surgery as Alternative to Continuous Positive Airway Pressure in Morbidly Severe Obstructive Sleep Apnea: A Case Report

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ABSTRACT: Obstructive sleep apnea syndrome (OSAS) is a sleep-related breathing disorder, characterized by disrupted snoring and repetitive upper airway obstructions. Oral appliance therapy is an effective alternative to continuous positive airway pressure (CPAP) and is especially effective in mild and moderate OSAS cases. Successful oral appliance therapy has been suggested as a predictor for successful maxillomandibular advancement (MMA) surgery in OSAS patients. MMA surgery has gained increasing popularity in this field since this procedure is associated with an enlargement of the entire velo-oro-hypopharyngeal airway. The authors present an unusual case of a CPAP-intolerant morbidly obese female (body mass index (BMI)=40) with morbidly severe OSAS (apnea-hypopnea index (AHI)=139, lowest oxyhemoglobin saturation level = 73%), who underwent a successful combination of MMA surgery, a modified genioplasty and cervicomental liposuction after oral appliance therapy. The post-surgical AHI was 6, and the lowest oxyhemoglobin saturation level increased to 86%. The authors suggest that this surgical combination should be seriously considered in obese patients with severe OSAS and CPAP intolerance.

Dr. Michiel H. Doff studied Mechanical Engineering at the Hanzehogeschool, Groningen, the Netherlands. In 2002, he started his dental education at the University of Groningen, where he received his D.D.S. degree in 2007. He began his Ph.D. research project on the long-term efficacy and side effects of CPAP and oral appliance therapy in obstructive sleep apnea patients, and subsequently received his Ph.D. in 2012 from the University of Groningen. He is vice president of the Dutch Society of Dental Sleep Medicine and a recognized specialist in dental sleep medicine. He is also a member of the interdisciplinary sleep apnea research group at the University Medical Center Groningen. He began his residency in oral and maxillofacial surgery in Groningen in 2011, and combines his clinical training with his medical training at the same university.

Obstructive sleep apnea syndrome (OSAS) is a sleep-related breathing disorder, characterized by disrupted snoring and repetitive upper airway obstructions. Patients suffering from OSAS have a predisposition to repetitive upper airway collapse during sleep, which can be the result of anatomical abnormalities, physiologic alterations of the upper airway, or fat depositions surrounding the upper airway of which the latter is most pronounced in overweight patients. OSAS is associated with an increased risk of cardiovascular events and associated morbidity and mortality. The condition also results in excessive daytime sleepiness, fatigue and neurocognitive deficits. Severity of OSAS is usually expressed by the apnea-hypopnea index (AHI), i.e., the mean number of apneas and hypopneas per hour of sleep. By convention, OSAS is classified as mild (AHI 5-15), moderate (AHI 15-30), or severe (AHI>30).

The standard treatment, continuous positive airway pressure (CPAP), relieves upper airway obstructions and improves quality of life. Because of the cumbersome nature of CPAP, patients may abandon or adhere poorly to this treatment. Oral appliance therapy is an effective alternative and is especially effective in mild-to-moderate OSAS cases. Most oral appliances used in a
clinical setting are mandibular advancement devices (MADs), which keep the mandible and its attached musculature in a protruded position. Successful oral appliance therapy has been suggested as a predictor for successful maxillomandibular advancement (MMA) surgery in OSAS patients. MMA surgery has gained increasing popularity in this field since this procedure is associated with an enlargement of the entire velo-oropharyngeal airway. This specific surgical procedure may be indicated for treatment of severe OSAS patients who cannot tolerate or are unwilling to adhere to CPAP therapy. MMA surgery may also be contemplated in patients in whom oral appliances, which are more often appropriate in mild and moderate OSAS patients, have been considered undesirable. The aim of this case report was to describe the value of MMA surgery after (partially) successful oral appliance therapy in a patient with morbidly severe OSAS and CPAP intolerance.

Case Report

A 32-year-old female was referred by the Department of Home Mechanical Ventilation to the Department of Oral and Maxillofacial Surgery of the University Medical Center Groningen because of respiratory problems and CPAP therapy intolerance. She suffered from morbidly severe obstructive sleep apnea syndrome (Apnea-Hypopnoea Index (AHI)=139) and was morbidly obese (body mass index (BMI)=40). Her minimal oxyhemoglobin saturation during sleep was 73%. Daily functioning was severely compromised because of excessive daytime sleepiness. Furthermore, she was diagnosed with left ventricular hypertrophy (LHV) and was treated for essential hypertension. She tried CPAP therapy (pressure 9 cm H2O) for a month, but this resulted in discomfort in the paranasal sinuses and distress during the night. During physical and radiological examination, a mandibular retrognathia and pronounced cervicomedial fat deposition was observed (Figure 1A). Intra-oral examination revealed a bulky tongue, which obscured visualization of the soft palate and uvula (i.e., modified Mallampati class IV). Further intra-oral examination revealed a healthy dentition without active periodontal disease. The lateral cephalogram showed a narrow posterior airway space at the level of the base of the tongue (Figure 2).

After full examination of the temporomandibular complex, it was decided to make an oral appliance. The oral appliance (Thornton Adjustable Positioner, Airway Management Inc., Dallas, TX, USA) consisted of two separate parts, fixing the patient’s mandible in a protruded and downward position. The mandibular protrusion could be adjusted with a propulsion screw, which was incorporated anteriorly in the oral appliance. When initiating oral appliance therapy, the mandible was set at 60% of the patient’s maximum protrusion. After having adapted to this position during a two-week period, the patient was allowed to further adjust the oral appliance until symptoms abated or adjustments became uncomfortable. The final amount of mandibular advancement was close to 88% of the maximal mandibular protrusion. At the final follow-up review, four weeks later, the patient reported that she slept and felt much better. She also reported a substantial decrease in her daytime sleepiness. To quantify the result of oral appliance therapy, a polysomnographic study with oral appliance in place was conducted six weeks after initiating therapy. The AHI decreased from 139 to 40, which was considered a major reduction. Minimal oxyhemoglobin saturation during sleep improved from 73% to 76%. The patient was very positive about the treatment effects and was treated with an oral appliance for about four months. However, her severe OSAS persisted despite oral appliance therapy. Furthermore, she indicated to prefer a more “permanent” solution for her condition. Since she also complained about a lack of chin projection in her profile, she opted for maxillomandibular advancement (MMA) surgery, combined with a genioplasty and cervicomedial liposuction. Presurgical orthodontics was regarded cumbersome in combination with the oral appliance.

Based on soft- and hard-tissue cephalometric analyses, using the true vertical line (TVL), a surgical plan was made. MMA surgery was planned in such a manner that an optimal airway enlargement was combined with an aesthetically pleasing surgical outcome. For this reason, a counter clockwise rotation of the maxillomandibular complex was planned. As a result, a large advancement of the mandible is accompanied with a posterior downgraft of the maxilla and thereby relatively less advancement of the upper jaw. This results in less paranasal puffiness, which may be expected when a straightforward advancement of the maxillomandibular complex of 10 mm would be planned. A modified genioplasty comparable to Plenier and Delaire, with advancement of the suprhyoid and genioglossal musculature was also planned. This procedure consists of a trapezoid-shaped advancement osteotomy of the chin to establish an additional forward stretching of the tongue and hyoid musculature. With this procedure, the upper part of the labial cortex of the advanced chin can be removed and used as a bonegraft at the zygomatic buttears at the Le Fort I level.

After preoperative placement of orthodontic brackets, MMA surgery was performed under general anesthesia. A bilateral sagittal split osteotomy (BSSO) of the mandible...
was performed with counterclockwise rotation and advancement of 10 mm at B point and fixed with two miniplates on both sides. After this, a standard Le Fort I osteotomy was performed with posterior downgraft of 3 mm at the level of the first molar and advancement of 8.5 mm at the incisal edge of the central incisors leaving the existing occlusion practically unchanged. The maxilla was fixed with four miniplates. The modified genioplasty, as described previously, was performed with an advancement of 6 mm. After this, a cervicofacial liposuction was performed using a small stab incision just behind the submental fold in the facial midline. At the end of the surgery, the zygomatic butresses were grafted with mandibular bone grafts from the chin. The bony defect between the proximal and distal segments of the mandible that resulted from the BSSO was grafted with a mix of bone particles and hydroxyapatite. The wounds were closed primarily and miniscrews were placed in the anterior region of the mandible and maxilla in order to put the patient on postoperative skeletal elastics in combination with dental elastics on the brackets. The patient was extubated at the end of the surgery and stayed overnight in the recovery room to guarantee continuous (oxyhemoglobin) monitoring. There were no surgical or medical complications in the direct post-operative period. Four months after surgery, another polysomnographic study was performed. There was almost a complete resolution of the patient’s OSAS with a postoperative AHI of 6, which consisted only of hypopneas and no apneas. The lowest oxyhemoglobin saturation during sleep was 86%. The patient reported to feel ‘reborn’ after surgery. She experienced her sleep as undisturbed and reported a resolution of her excessive daytime sleepiness. Furthermore, she was very pleased with her new facial appearance.

Figure 1 (A & B)
Thirty-three-year-old female with morbidly severe obstructive sleep apnea syndrome. Photograph of the (A) preoperative profile and (B) postoperative profile.

Figure 2 (A & B)
(A) Cephalogram of the preoperative situation. (B) Cephalogram after maxillomandibular advancement surgery, modified genioplasty, and cervicofacial liposuction.
The tracings of the pre-operative and postoperative cephalograms illustrate the profound skeletal and soft-tissue changes as a result of surgery (Figure 3).

Maxillofacial noncontrast conebeam CT (CBCT) scans were taken pre- and postoperative (i-CAT, Imaging Sciences International, Hatfield, USA). At each acquisition, the patient was seated in an upright position and asked to hold her breath at end expiration, with the dentition in maximal occlusion and the lips in relaxed contact position. The CBCT scans were imported in Simplant Crystal software (Materialize Dental, Leuven, Belgium). In the axial slides, the superior boundary of the 3D airway figure was set at the posterior nasal spine and the inferior boundary was set at the base of the epiglottis. The minimum retropalatal and retroglossal cross-sectional area (CSA) of the upper airway was calculated pre- and postoperatively. After surgery, a substantial increase was found in anterior-posterior and lateral airway dimensions of the upper airway at the retropalatal (increase minimal cross-sectional area from 114 mm² to 276 mm²) and retroglossal (increase minimal cross-sectional area from 109 mm² to 188 mm²) level (Figures 4 and 5).

Discussion

This case report shows the effectiveness of MMA surgery after positive results of oral appliance therapy in a patient with severe OSAS and CPAP intolerance. This
finding confirms the results of the study performed by Hoekema and co-workers, in which patients with a substantial reduction in the baseline AHI with oral appliance therapy appeared good candidates for MMA surgery. However, that study was retrospective in design and included only four patients undergoing MMA surgery. It would therefore be of great value to perform a prospective study to investigate the predictive value of oral appliance therapy outcome for MMA surgery in a larger group of patients.

The patient described in this case report had a baseline AHI of 139, which may be considered morbidly severe OSAS. Oral appliance therapy resulted in a major decrease of the AHI to 40 but the remaining OSAS was still severe. It is known that oral appliance efficacy declines with increasing OSAS severity. This is explained by the fact that patients with severe OSAS are generally more obese with oral appliance therapy generally being less effective in these patients. Furthermore, oral appliance therapy only protrudes the mandible while the maxillary position remains unchanged. In MMA surgery, the mandible, as well as the maxilla, are positioned anteriorly, which could explain the superior efficacy of MMA surgery over oral appliance therapy in our patient. The additional maxillary advancement pulls the velum and velopharyngeal muscles forward, most possibly creating more space at the velopharyngeal level of the upper airway.

Fairburn and co-workers reported varying results regarding efficacy of MMA surgery in severe OSAS patients. In a few of their patients, respiratory parameters deteriorated after MMA surgery. One of their explanations for this finding is the negative effect of scarring as a result of previous upper airway surgery (e.g., uvulopalatopharyngoplasty). Our patient did not undergo any type of upper airway surgery before the present procedure, which could have had a favourable effect in this specific case. Younger age, lower preoperative BMI and AHI are considered predictors of successful MMA surgery in OSAS patients. However, our patient was morbidly obese and had an extremely high AHI but was still treated very effectively following this procedure. This finding may be explained by the fact that our patient was additionally treated with a genioplasty and cervicomental liposuction. These procedures may have added to the positive effect of MMA surgery on upper airway patency.

As a result of the enlargement of the lateral and anterior-posterior dimensions of the upper airway following MMA surgery, an increase in the cross sectional area of the upper airway was found at retropalatal and retroglossal levels. This finding is consistent with results published in a previous study. Studies evaluating upper airway changes as a result of wearing an oral appliance also show an increases of the cross sectional area of the upper airway at different levels. Choi and co-workers report a decrease in the supine position of the cross-sectional area (range 36.5%-75.5%) of the upper airway during midazolam-induced sleep. In most studies, including this case report, patients were evaluated during upper airway assessment in an upright position while being awake. However, in this case report, the pre- and postoperative CBCT studies were performed using exactly the same protocol. We therefore believe that the changes found in the upper airway cross-sectional areas after MMA surgery are of clinical importance.

In a recent conducted controlled study, it was found that there was no significant difference in success rate between MMA surgery and CPAP in severe OSAS patients. These findings may indicate and confirm that in the future, MMA surgery can take a prominent place in treating severe OSAS patients who do not tolerate CPAP or in patients who are unwilling to use CPAP.

In conclusion, this case report shows that MMA surgery combined with a modified genioplasty and cervicomental liposuction can be very effective in treating morbidly severe OSAS in a morbidly obese patient. MMA surgery was in fact the only effective treatment for this patient’s life-threatening disorder. Furthermore, this case substantiates the observation that effective treatment with an oral appliance (mandibular advancement device) can be considered a good predictor for successful MMA surgery in OSAS management. Lateral and anterior-posterior upper airway dimensions increase at the retropalatal and retroglossal level as a result of MMA surgery combined with a genioplasty and submental liposuction. To clarify which patients would benefit from MMA therapy, a multidisciplinary evaluation is recommended.

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