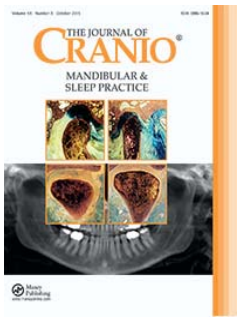

CRANIO®
The Journal of Craniomandibular & Sleep Practice

ISSN: 0886-9634 (Print) 2151-0903 (Online) Journal homepage: http://www.tandfonline.com/loi/ycra20

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

Michiel H.J. Doff, Johan Jansma, Rutger H. Schepers & Aarnoud Hoekema

To cite this article: Michiel H.J. Doff, Johan Jansma, Rutger H. Schepers & Aarnoud Hoekema (2013) Maxillomandibular Advancement Surgery as Alternative to Continuous Positive Airway Pressure in Morbidly Severe Obstructive Sleep Apnea: A Case Report, CRANIO®, 31:4, 246-251, DOI: 10.1179/crn.2013.31.4.001

To link to this article: http://dx.doi.org/10.1179/crn.2013.31.4.001

■ CASE REPORT

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

Michiel H.J. Doff, D.M.D., Ph.D., B. Eng.; Johan Jansma, M.D., D.M.D., Ph.D.; Rutger H. Schepers, M.D., D.M.D.; Aarnoud Hoekema M.D., D.M.D., Ph.D.

0886-9634/3104-246$0.25/pp, THE JOURNAL OF CRANIOMANDIBULAR & SLEEP PRACTICE,

Manuscript received June 6, 2013; revised manuscript received July 23, 2013; accepted July 29, 2013

Address for correspondence:
Dr. Michiel Doff
Dept. of Oral and Maxillofacial Surgery
University Medical Center Groningen
University of Groningen
Hanzeplein 1
P.O. Box 30.001
9700 RB, Groningen
The Netherlands
Email: m.h.j.doff@umcg.nl

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 Gronigen. 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.[1] 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.[2] 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).[3]

The standard treatment, continuous positive airway pressure (CPAP), relieves upper airway obstructions and improves quality of life.[4] 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.[5] 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.[6] MMA surgery has gained increasing popularity in this field since this procedure is associated with an enlargement of the entire velo-oro-hypopharyngeal airway.[7,8] 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.[9] 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 H_2O) for a month, but this resulted in discomfort in the maxillary sinuses and distress during the night. During physical and radiological examination, a mandibular retrognathia and pronounced cervicomental 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 cervicomental 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,[10] with advancement of the suprahyoid 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.[11] 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 buttresses at the Le Fort 1 level.

After preoperative placement of orthodontic brackets, MMA surgery was performed under general anesthesia. A bilateral sagittal split osteotomy (BSSO) of the mandible

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.

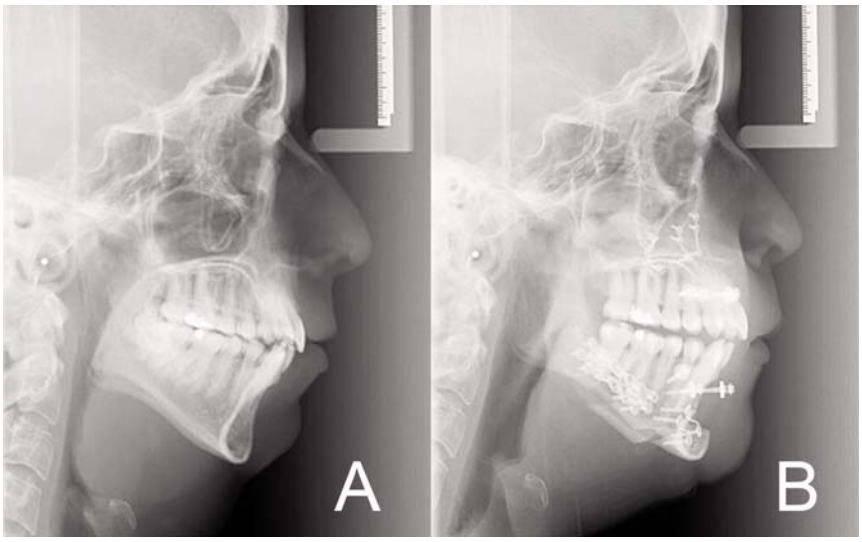

Figure 2 (A & B)
(**A**) Cephalogram of the preoperative situation. (**B**) Cephalogram after maxillomandibular advancement surgery, modified genioplasty, and cervicomental liposuction.
.

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 1 osteotomy was performed with posterior downgraft of 3 mm at the level of the first molars 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 cervicomental 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 buttresses were grafted with mandibular bonegrafts 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 1B**). The tracings of the pre-operative and post-operative cephalograms illustrate the profound skeletal and soft-tissue changes as a result of surgery (**Figure 3**).

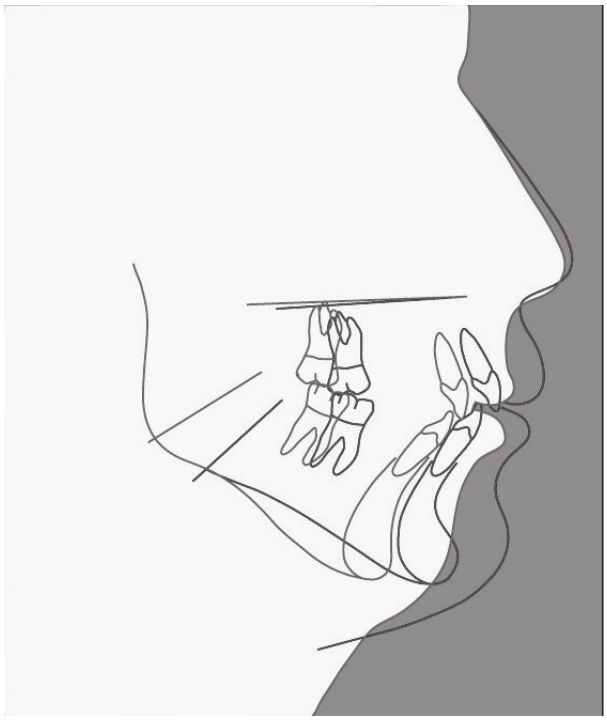

Figure 3
Superimposed (S-N) preoperative (line in gray area) and postoperative tracings (line in white area) of lateral cephalograms.

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 mm^2 to 276 mm^2) and retroglossal (increase minimal cross-sectional area from 109 mm^2 to 188 mm^2) level (**Figures 4** and **5**).

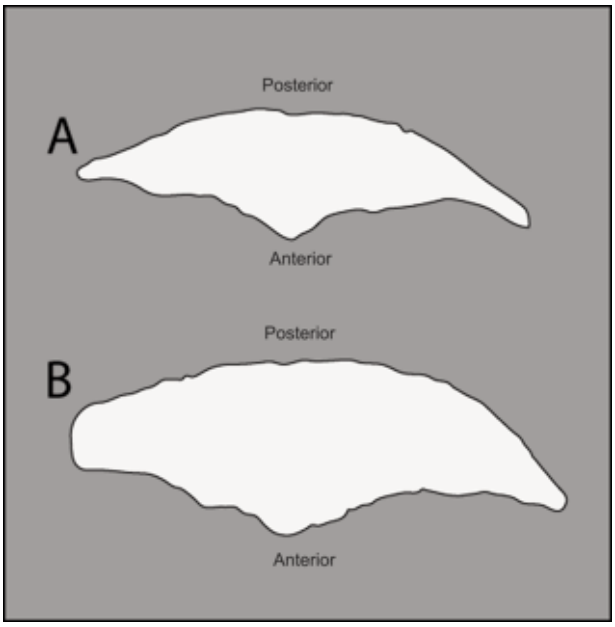

Figure 4
Minimal cross-sectional area of the upper airway at retroglossal level (**A**) preoperative and (**B**) postoperative.

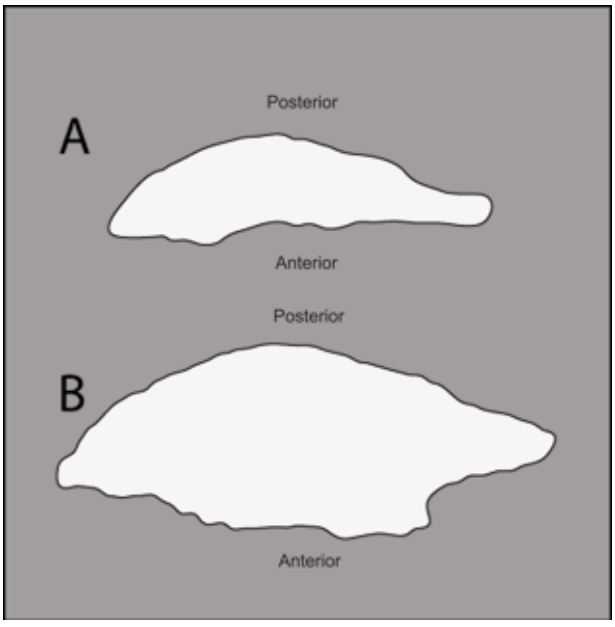

Figure 5
Minimal cross-sectional area of the upper airway at retropalatal level (**A**) preoperative and (**B**) postoperative.

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,[6] 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.[12] 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.[12] 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[13] 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.[6] 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.[14] 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.[15,16] Choi and co-workers[15] 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.[17] 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.

Acknowledgements

The authors would like to thank B. Stegenga and J. Nieuwenhuis for their critical appraisal of the manuscript.

References

1. Malhotra A, White DP: Obstructive sleep apnea. *Lancet* 2002; 360:237-245.
2. Young T, Peppard PE, Gottlieb DJ: Epidemiology of obstructive sleep apnea: a population health perspective. *Am J Respir Crit Care Med* 2002; 1:165:1217-1239.
3. AASM: Sleep-related breathing disorders in adults: recommendations for syndrome definition and measurement techniques in clinical research. The Report of an American Academy of Sleep Medicine Task Force. *Sleep* 1999; 22:667-689.
4. Giles TL, Lasserson TJ, Smith BH, White J, Wright J, Cates CJ: Continuous positive airways pressure for obstructive sleep apnea in adults. *Cochrane Database of Syst Rev* (Online) 2006; 3:CD001106.
5. Hoekema A, Stegenga B, Wijkstra PJ, van der Hoeven JH, Meinesz AF, de

Bont LG: Obstructive sleep apnea therapy. *J Dent Res* 2008; 87:882-887.
6. Hoekema A, de Lange J, Stegenga B, de Bont LG: Oral appliances and maxillomandibular advancement surgery: an alternative treatment protocol for the obstructive sleep apnea-hypopnea syndrome. *J Oral Maxillofac Surg* 2006; 64:886-891.
7. Boyd SB, Walters AS, Song Y, Wang L: Comparative effectiveness of maxillomandibular advancement and uvulopalatopharyngoplasty for the treatment of moderate to severe obstructive sleep apnea. *J Oral Maxillofac Surg* 2013; 71:743-751.
8. Prinsell JR: Primary and secondary telegnathic maxillomandibular advancement, with or without adjunctive procedures, for obstructive sleep apnea in adults: a literature review and treatment recommendations. *J Oral Maxillofac Surg* 2012; 70:1659-1677.
9. Aurora RN, Casey KR, Kristo D, Auerbach S, Bista SR, Chowdhuri S, et al.: Practice parameters for the surgical modifications of the upper airway for obstructive sleep apnea in adults. *Sleep* 2010; 33:1408-1413.
10. Plenier V, Delaire J: "Functional" genioplasty. *Rev Stomatol Chir Maxillofac* 1983; 84:54-61.
11. Bruno Carlo B, Mauro P, Silvia B, Enrico S: Modified genioplasty and bimaxillary advancement for treating obstructive sleep apnea syndrome. *J Oral Maxillofac Surg* 2008; 66:1971-1974.
12. Isono S, Tanaka A, Tagaito Y, Sho Y, Nishino T: Pharyngeal patency in response to advancement of the mandible in obese anesthetized persons. *Anesthesiology* 1997; 87:1055-1062.
13. Fairburn SC, Waite PD, Vilos G, Harding SM, Bernreuter W, Cure J, et al.: Three-dimensional changes in upper airways of patients with obstructive sleep apnea following maxillomandibular advancement. *J Oral Maxillofac Surg* 2007; 65:6-12.
14. Li KK, Guilleminault C, Riley RW, Powell NB: Obstructive sleep apnea and maxillomandibular advancement: an assessment of airway changes using radiographic and nasopharyngoscopic examinations. *J Oral Maxillofac Surg* 2002; 60:526-530; discussion 531.
15. Choi JK, Hur YK, Lee JM, Clark GT: Effects of mandibular advancement on upper airway dimension and collapsibility in patients with obstructive sleep apnea using dynamic upper airway imaging during sleep. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2010; 109:712-719.
16. Sutherland K, Deane SA, Chan AS, Schwab RJ, Ng AT, Darendeliler MA, et al.: Comparative effects of two oral appliances on upper airway structure in obstructive sleep apnea. *Sleep* 2011; 34:469-477.
17. Vicini C, Dallan I, Campanini A, De Vito A, Barbanti F, Giorgiomarrano G, et al.: Surgery vs. ventilation in adult severe obstructive sleep apnea syndrome. *Am J Otolaryngol* 2010; 31:14-20.

Dr. Johan Jansma *received both his D.D.S. degree in 1986 and his M.D. degree in 1995 at the University of Groningen, the Netherlands. From 1987 to 1991, he trained as an oral and maxillofacial surgeon at the Department of Maxillofacial Surgery at the same university. In November 1991, he received his Ph.D. for a thesis entitled "Oral sequelae resulting from head and neck radiotherapy." Since 1991, he has been an associate professor at the Department of Maxillofacial Surgery at the University Medical Center Groningen. In 2000, he was board certified as a head and neck oncologist. In the same year, he became Fellow of the European Board of Oro-Maxillofacial Surgeons. Since 2007, he has also worked part-time in private practice in the Martini Hospital Groningen. His main fields of interest are orthognathic surgery (and distraction osteogenesis), cleft palate surgery, and cosmetic facial surgery. Dr. Jansma has been published in national and international journals and has lectured on many different subjects.*

Dr. Rutger H. Schepers *received both his dental and medical degrees from the University of Nijmegen, the Netherlands. He is certified as an oral and maxillofacial surgeon. Currently, he is combining his clinical work with a Ph.D. study, which encompasses 3-D digital planning in surgical head and neck oncology.*

Dr. Aarnoud Hoekema *received both his dental and medical degrees at the University of Groningen, the Netherlands. In 2007, he successfully defended his Ph.D. thesis entitled "Oral-Appliance Therapy in Obstructive Sleep Apnea-Hypopnea Syndrome: a Clinical Study on Therapeutic Outcomes." Dr. Hoekema works as an oral and maxillofacial surgeon at the Tjongerschans Hospital, located in Heerenveen. He is also involved in postdoctoral research at the University Medical Center Groningen, is president of the Dutch Society of Dental Sleep Medicine, and is a recognized specialist in dental sleep medicine. He recently received the prestigious Pierre Robin award as recognition of his scientific work regarding obstructive sleep apnea.*