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Case Report

Recurrence of OSAHS in a Patient Previously Treated with MAD and Multilevel Surgery: The Role of Epiglottis

Giulia Anna Marciante*, Federico Leone, Alessandro Bianchi, Chiara Re and Fabrizio Salamanca

Unit of Otorhinolaryngology, Head and Neck Surgery, Humanitas San Pio X, Milan, Italy

***Corresponding Author:** Giulia Anna Marciante, Unit of Otorhinolaryngology, Head and Neck Surgery, Humanitas San Pio X, Milan, Italy.

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Abstract

The introduction of sleep endoscopy in the diagnosis of sleep apnea-hypopnea syndrome (OSAHS) has led to the development of a customization in the surgical approach of patients refusing nocturnal ventilation. Sometimes follow up polysomnography can show a recurrence of OSAHS, even in patients with no changes in weight or head and neck anatomy. We report a case of a patient undergone uvulopalatopharyngoplasty and epiglottoplasty showing a worsening of OSAHS years after surgery.

A 69-year-old man with moderate OSAHS refusing overnight ventilation underwent sleep endoscopy showing collpase of palate, tongue base and epiglottis. Uvulopalatopharyngoplasty and epiglottoplasty were then perfomed in addiction to application of mandibular advancement device (MAD), with normalization of parameters, as shown by the following polysomnographies. Over time, symptomes started to recur, and the patient underwent a new polysomnpography showing increase of apnea-hypopnoea index (AHI). A second sleep endoscopy was therefore performed, pointing out the role of the collapse of the residual epiglottis in developing apneas. Epiglottis stiffening operation was consequently perfomed with resolution of symptoms and reduction of AHI.

Patients treated with surgery and MAD should always be monitored with follow up polysomnography. Worsening of symptoms should be evaluated by means of sleep endoscopy in order to identify and treat the new causes of obstruction.

Keywords: OSAHS; Mandibular Advancement Device; Epiglottoplasty; Sleep Endoscopy

Introduction

Overnight ventilation represents the gold standard treatment for obstructive sleep apnea/hypopnea syndrome (OSAHS). Since the compliance to this therapy varies from 29% to 80% [1], other options can be proposed. The correct choice depends on the results of sleep endoscopy, that allows to directly visualize sites of obstruction along the airways, from the nose to the larynx. Its introduction, in fact, has led to the identification of peculiar patterns of collapse and detection of obstruction also at laryngeal level in patients without evident morphological laryngeal anomalies, proving the importance of an appropriate dynamic evaluation of the airways during sleep [2]. Based on sleep endoscopy, surgery and/or application of oral appliances can be considered. Since several factors may be involved in the pathogenesis of OSAHS [3], polysomnography is usually performed during follow up in patients surgically treated, in order to avoid possible recurrence or worsening of the disease due to variations in weight or muscular tone. Here we report a case of a man with a recurrence of OSAHS ten years after surgery and application of MAD. Written informed consent was obtained from the patient.

Case Description

A 69-year-old man with observed apneas was referred to the Sleep Apnea Research Center of Humanitas San Pio X Clinic of Milan. Polysomnography revealed the presence of a moderate OSAHS (AHI: 19.7), and overnight ventilation was therefore proposed as first line treatment. After a two-month trial the patient refused the treatment. Since positional therapy was not indicated, propofol sleep endoscopy was performed, showing collapse at palatal, tongue base and epiglottic level. Mandibular pull up manoeuvre improved tongue base collapse, without resolution of collapse at palatal and epiglottic level. Uvulopharyngoplasty (UP3) and laser epiglottoplasty were then performed. A three-month follow up polysomnography with appplication of MAD showed the efficacy of the combined treatment (AHI: 2.8), and 6 months after surgery significant increase of the laringeal posterior airways space was seen at fiberoptic endoscopy (Figure 1). Although strongly recommended, the patient decide not to perform periodic polismonographies. Over time, snoring and sleepiness started to recur, so ten years after surgery the patient decided to repeat polysomnography. Surprisingly, the exam showed a pathologic apnea/hypopnoea index (AHI 18.3) including central and mixed apneas. A second sleep endoscopy was therefore performed: collapse of the residual epiglottis during apneas was still appreciated, with no efficacy of both MAD at maximal titration and mandibular pull up manoeuvre after MAD removal (Figure 2).

Figure 2: Evidence of the residual epiglottis collapse persistent after pull up manoeuvre during DISE 10 years after surgery and MAD application

Epiglottic collapse was treated by means of Epiglottis Stiffening Operation (ESO) according to Salamanca., *et al*. [4]. Informed consent was obtained from the patient. A three-month follow up polysomnography showed a significant reduction of the apnoic events (AHI: 4.7), even if persistence of central apneas was still recorded. The six-month post operative fiberoptic endoscopic evaluation showed complete adhesion of epiglottis to the tongue base without any complained discomfort or episodes of penetration/aspiration (Figure 3).

Figure 1: Fiberoptic evaluation of the posterior hypopharyngeal-laryngeal space 6 months after UP3, epiglottoplasty and MAD.

Figure 3: Fiberoptic evaluation of the results of the epiglottis stiffening operation 6 months after ESO.

Discussion

Obstructive sleep apnea hypopnea syndrome (OSAHS) is a multifactorial chronic disease with an estimated prevalence of 2-4% in men and 1-2% in women [5]. Overnight ventilation is the first-line and gold standard for OSAHS, but the low compliance rate still represents a problem in treating this condition [6]. Surgery and oral appliances are effective alternatives in selected patients refusing nocturnal ventilation, as it has been widely demostrated in literature [7-10].

Indication to surgery or MAD is essentially based on sleep endoscopy, which helps to identify the sites and the patterns of collapse, suggesting the possible efficacy of MAD or palatal, tongue base or epiglottis surgery [11].

In fact, if pull up monoeuvre is effective during sleep endoscopy, MAD can be proposed alone or associated to surgery, improving not only polysomnographic parameters, but also quality of life. In spite possible short term side effects, which are well known and described (a.e. dry mouth, excessive salivation, teeth discomfort, muscle tenderness, temporomandibular joint pain, myofascial pain, and gum irritation) [12], a little is known about the long term ones. Among the several surgical approaches proposed, uvulopalatopharyngoplasty (UP3) is one of the most spread treatment for palatal collapse, which is a common finding in OSAHS patients. It consists in removal of the tonsils and excision of soft palate and uvula, leading to a significant increase of the posterior airway space and a reduction of the palatal collapse [13]. Collapse of epiglottis, instead, is rarer, and it has been reported up to 12% in patients with OSAHS, even if DISE has shown higher rates of obstruction at this level [14].

In these subjects, epiglottoplasty is effective in reducing the collapse of the floppy epiglottis by means of its resection [15]. However, in case of extreme laxity of the glossoepiglottic ligaments, epiglottoplasty could be not as efficacious as expected. In these selected patients Epiglottis Stiffening Operation proposed by Salamanca., *et al*. [4] has proven to be effective, especially in addition to MAD.

Conclusions

Surgery and MAD can be effective therapeutic options for OSAHS either alone or in association in patients refusing overnight ventilation. Post-operative periodic polysomnography is necessary for identification of possible recurrence of apnoic episodes. Onset or worsening of symptoms should always be evaluated by means of sleep endoscopy in order to identify the new causes and sites of obstruction, which can recur at laryngeal level also in patients previously treated with epiglottoplasty.

Main Points

- OSAHS is a multifactorial disorder in which collapse may be observed at different levels
- Patients treated for obstructive sleep apnea hypopnea syndrome (OSAHS) should always undergo periodically instrumental follow up by means of polysomnography even when surgically treated
- When worsening of polysomnographic parameters is observed, sleep endoscopy should be considered in order to identify the sites of obstruction
- Epiglottis could represent a cause of obstruction even after partial epiglottoplasty.

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