CASE REPORT

Two Cases of Severe Obstructive Sleep Apnea Induced by Neck Radiotherapy Treated with an Oral Device

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Abstract Sleep apnea has been shown to be more prevalent in patients who underwent radiotherapy and or surgery for head and neck cancer. Here we describe two cases of severe OSAS induced by surgery and radiotherapy, treated by Mandibular Advancing Devices as an alternative option to continuous positive airway pressure.

Keywords Obstructive sleep apnea · Head and neck cancer · Oral appliance

Introduction

Obstructive sleep apnea syndrome (OSAS) is a chronic sleep-related disorder breathing, characterized by repetitive complete (apnea) or partial (hypopnea) airway collapse. The intermittent hypoxia, consequent to obstruction, and the sleep fragmentation lead to an increased cardiovascular risk and daytime sleepiness. Some authors reported a high prevalence of OSAS in patients treated for head and neck cancer, underlining the importance to screen them to increase their quality of life [1–3].

Structural alterations, as a flaccid epiglottis, and neuromuscular dysfunction can occur after surgery and/or radiation, thus affecting the balance between airway dilating and collapsing forces and leading to sleep apnea [4]. Unfortunately, most of patients included in these studies (67 %) discontinued use of Continuous Positive

Airway Pressure (CPAP), due to pain and xerostomia, secondary to treatment [1]. We describe two cases of severe OSAS induced by surgery and radiotherapy, treated by Mandibular Advancing Devices (MADs).

Case 1

A 47-year-old man was referred to the ENT Department of S. Orsola Malpighi Hospital (University of Bologna, Italy). His main complaints were loud snoring, witness apneas and choking. The patient had a history of tonsil squamous cell advanced carcinoma, treated by surgery and chemoradiotherapy. The chemoradiotherapy finished 6 months before the onset of symptoms. On physical examination he weighed 67 kg, his height was 178 cm and BMI was 21 kg/m².

The Epworth Sleepiness Scale (ESS) administration revealed a score of 14, confirming the daytime sleepiness reported by the patient. At the home sleep test evaluation, performed by a portable monitoring (type III monitor, EMBLETTA GOLD, 6 channels), he was diagnosed to have severe OSAS. He presented an apnea/hypopnea index (AHI) of 49 events per hour; an AHI of 15 eV/h in supine position and of 51 eV/h in non-supine position; an oxygen desaturation index (ODI) of 31 events per hour; a lowest minimum oxygen saturation recorded during sleep (O₂ Sa) of 82 % and the cumulative percentage of time spent at saturations below 90 % (CT < 90 %) was 9.4 %.

The endoscopic examination with Muller maneuver showed a grade 3 retropalatal upper airway collapse and a grade 2 at the retrolingual site.

Because the patient refused C PAP therapy, a drug induced sleep endoscopy (DISE) was performed. DISE showed flaccid supraglottic tissues. The airflow induced

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collapse of the epiglottis into the laryngeal inlet causing upper airway obstruction, strongly improved by mandibular pull-up. The oropharynx appeared normal.

The patient was referred to the orthodontist to evaluate the possibility to prescribe a MAD.

A careful assessment of the patient was carried out to evaluate TMJ, dental, periodontal and oral mucosa health.

The patient showed multiple missing teeth. In the upper arch, only 13 and 23 root were present and were provided with ball anchorages for an upper overdenture. In the lower arch, thirteen elements were present, with several dental restorations and non carious lesions (abfractions). Gingival bleeding on probing was present.

No pain to palpation of the masticatory, cervical or TMJ regions, no TMJ noises or dysfunction were detected. He presented a normal range of motions and the maximal protrusion was 12 mm.

The treatment plan provided the reopening of the tracheostomy, waiting for the construction, the titration and the outcome evaluation of the MAD.

Before any further treatment, periodontal health was achieved by performing a full-mouth scaling and root planning. Due to this treatment and to a good patient compliance, periodontal parameters improved and subsequently stabilized within healthy values. To preserve a good oral health we suggested the patient to have dental prophylaxis every 3 months and a clinical examination by his general dentist every 6 months.

When a healthy condition was restored a Flex Somnomed® MAS Edentulous (Fig. 1) was applied. A wax superior occlusal rim was performed on a thermoplastic base, maintaining the vertical dimension of his prosthesis. The occlusal rim was necessary to perform the construction bite with a 5 mm George Gauge bite fork (Fig. 2). It allowed the sleep dentist to set the vertical distraction of the mandible and the initial therapeutic advancement. At the start the construction bite determined a vertically opening of 5 mm and a sagittal advancement of 7.5 mm (75 % of maximum protrusion). Final impressions were taken over with individual trays. After the delivery of the device, the patient was informed about appliance use and after 2 weeks, titration protocol was started. The advancement was gradually increased to 0.5 mm a week. After 2 weeks an advancement of 8.5 mm was obtained. During follow-up only a mild muscular pain was reported.

Three months after the delivery of the appliance, a polygraphy (PG) with the same parameters of baseline, blocked tracheostomy and the appliance in situ was carried out. An excellent response was confirmed by the achievement of an AHI of 3/h, an ODI of 2, a lowest desaturation of 89 % and a CT < 90 % of 2 %. Consequently tracheostomy was closed. After 1 month from tracheostomy closure the patient reported no snoring and

Fig. 1 Somnodent edentulous, frontal view

Fig. 2 Superior occlusal wax rim adapted to George gauge bite fork

no daytime sleepiness, using the MAD. The ESS administration revealed a score of 6. Another PSG evaluation was performed after 13 months. The exam revealed a complete resolution of the OSAS, with an AHI of 4 eV/h, an ODI of 5, a lowest desaturation of 89 %, a CT < 90 % of 2 %.

Case 2

A 52-year-old man presented to our department with the chief complaint of loud snoring at night and daytime hypersomnia. The patient had a history of tonsil squamous cell carcinoma, treated by concurrent chemoradiotherapy with a radiation dose of 70 Gy. The chemoradiotherapy treatment ended 2 months before the onset of symptoms. On physical examination he weighed 73 kg, his height was 181 cm and BMI was 22 kg/m².

The Epworth Sleepiness Scale (ESS) administration revealed a score of 13. A PG study (Tipe III monitor, EMBLETTA GOLD, 6 channels) demonstrated an AHI of 55/h (AHI supine 55/h, AHI non supine 55/h), an ODI of 38 events per hour, a LSAO₂ and CT < 90 % of 84 and 4.1 % respectively.

The endoscopic examination with Muller maneuver showed a multilevel upper airway collapse (grade 3 obstruction both retropalatal and retrolingual).

The patient was referred to CPAP therapy but he complained worsening of the daytime symptoms.

DISE showed a flaccid and lax epiglottis prolapsing posteriorly during inspiration and causing complete airway collapse, solved by mandibular pull-up.

The patient was referred to the orthodontist to evaluate the possibility to prescribe a Mandibular Advancing Device.

A careful assessment of the patient was carried out to evaluate TMJ, dental, periodontal and oral mucosa health.

The patient showed multiple non carious lesions (abfractions), several dental restorations and severe dental crowding.

Gingival bleeding on probing was present.

No pain to palpation of the masticatory, cervical or TMJ regions, no TMJ noises or dysfunction were detected. He presented a normal range of motions and the maximal protrusion was 11 mm.

The treatment plan provided the construction of a Mandibular Advancing Device.

Before beginning any further treatment, periodontal health was achieved by performing a full-mouth scaling and root planning. Due to this treatment and to a good patient compliance, periodontal parameters improved and they stabilized within healthy values. To preserve a good oral health we suggested the patient to have dental prophylaxis every 3 months and a clinical examination by his general dentist every 6 months.

When a healthy condition was restored, the orthodontist decided to construct for the patient a Thornton Adjustable Positioner (TAP 3[®]) (Fig. 3). A construction bite with a 5 mm George Gauge bite fork was performed to set the vertical distraction of the mandible and the initial therapeutic advancement. At the start the construction bite determined a vertical opening of 5 mm and a sagittal advancement of 7 mm. The advancement was gradually

Fig. 3 Thornton Adjustable Positioner, frontal view

increased to 0.5 mm a week, obtaining a 8.5 mm advancement after 3 weeks. During follow-up only mild muscular pain was reported. Three months after the delivery of the appliance the patient reporting a great improvement on snoring and on daytime fatigue and a PSG control with appliance in situ was carried out. An excellent response was confirmed by the following findings: AHI 4 eV/h, ODI 1 eV/h, lowest desaturation 89 %, CT < 90 % 0.2 %. Another PSG was performed at 15 months follow up. The exam confirmed a good response (AHI 5 eV/h, ODI 3 eV/h, lowest desaturation 91 %, CT < 90 % 0.2 %).

Discussion

Successful treatment of OSAS is based on the accurate identification of the pattern of airway obstruction and targeted effective treatment. The palate is considered the main generator of snoring sounds and at least an important contributor to pharyngeal obstruction in many cases. Therefore the soft palate is the major target of surgery in subjects with snoring or OSAS and uvulopalatopharyngoplasty is one of the main therapy. Other treatment modalities include lateral pharyngoplasty, relocation pharyngoplasty and expansion sphincter pharyngoplasty. In case of severe OSAS, maxillo-mandibular advancement is the most effective treatment.

The management of head and neck cancer, either surgical or radiation treatment, should predispose to sleep apnea. Intuitively there are several factors that may contribute to upper airway collapse and resistance.

These include alteration in suprahyoid and pharyngeal musculature, mandibulotomy, bulky flap reconstruction, soft-tissue fibrosis, and lymphedema [5]. Moreover, these therapies may also create neurosensory dysfunction affecting feedback from the upper airway to the pharyngeal dilators thus clinical suspicion of OSAS may be higher in patients treated for head and neck cancer [6].

The identification and treatment of OSAS might be an important factor in alleviating the fatigue and overall quality of life of these patients.

CPAP is an effective therapy for OSAS, improving the health-related quality of life of patients but acceptance and long-term compliance of CPAP are typically poor. Particularly in head and neck cancer patients, xerostomia occurring after radiation therapy may adversely impact CPAP compliance, because common side effects of CPAP therapy such as dry mouth may be amplified in this population. Moreover when upper airway obstruction is due to epiglottis collapse (lax epiglottis is very common in irradiated patients), positive pressure may exacerbate upper

airway obstruction by further promoting the downward displacement of the epiglottis into the laryngeal inlet.

Our patients were affected by severe OSAS due to epiglottis collapse. In case of OSAS due to epiglottis collapse the management can be surgical and consists of laser assisted or robotic epiglottectomy but patients with head and neck cancer are not likely to be candidates for upper airway surgery because of the risk of poor wound healing and swallowing disfunction [1].

Obviously alternative treatment could be tracheostomy but it can be perceived as a major disability with emotional problems, such as depression or a change in self-image in patients yet at greatest risk for psychosocial dysfunction.

MADs are indicated in patients with a mild to moderate OSA or in severe patients when CPAP cannot be tolerated [7]. Understanding the reason for CPAP therapy dropout in patients submitted to head and neck radiotherapy, could lead to an indication to MAD treatment.

In addition, several authors underline the effectiveness of the functional and anatomic evaluation of obstruction sites to better choose a tailored therapy for OSA patients [8, 9].

Because the site of upper airway obstruction following treatment of head and neck cancer is typically at the level of the larynx, DISE should be considered for these patients. In fact one of the strength points of DISE is the ability to identify laryngeal obstruction.

In the two cases reported above, DISE allowed to understand the dynamics of collapse, due to structural modifications induced by radiotherapy. A flaccid epiglottis was detected in both the patients, which was stabilized by the mandibular protrusion performed during the exam. The MAD treatment was planned on clinical findings and the outcome was a complete response.

Since recognition and treatment of OSAS may play an important role in improving quality of life in head and neck cancer patients, we suggest to screen all patients treated for upper airway carcinoma for the presence of sleep-related breathing problems.

Compliance with Ethical Standards

Conflict of interest Ottavio Piccin, Giovanni Sorrenti and Francesca Milano declare that they have no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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