CASE REPORT

Solitary Fibrous Tumor of the Tongue: An Uncommon Cause of Obstructive Sleep Apnea

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Abstract

Introduction Solitary fibrous tumor is an uncommon mesenchymal neoplasm that may be found in any location. To date, only a few cases of solitary fibrous tumor involving the tongue have been reported.

Case Summary We present the case of a 31-year-old man with a history of progressively worsening snoring and daytime sleepiness. Polysomnography revealed severe obstructive sleep apnea. An attempt to treat sleep apnea by continuous positive airway pressure and oral appliance led to a poor clinical response. CT and MRI scans findings revealed a large mass in the tongue base partially obstructing the airway. After the excision of the mass all symptoms, included daytime somnolence, disappeared and a polysomnographic examination showed the normalization of the somnographic parameters.

Discussion Although OSA is rarely caused by tumors, each patient with sleep disorders breathing should be examined carefully for the potential presence of an upper aero-digestive tract neoplasm that may contribute to obstruction.

Keywords Solitary fibrous tumor · Obstructive sleep apnea · Polysomnography

Introduction

Obstructive sleep apnea syndrome (OSAS) is caused by anatomic abnormalities combined with neuromuscular dysfunctions, leading to nocturnal repetitive episodes of upper airway (UA) obstruction. In rare case, OSAS can be associated with the presence of uncommon slow-growing neoplasms of the upper aero-digestive tract or parapharyngeal spaces [1–3]. We report a rare case of a large atypical solitary fibrous tumor (SFT) of the tongue causing OSAS.

Case Report

In 2012, a 31-year-old male patient, with a BMI of 23, was referred to the Sleep Medicine Center of our Hospital because of suspected sleep-disordered breathing.

Some months before daytime sleepiness and mood changes had emerged interfering with his daily activities (Epworth sleepiness scale score 20). 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 56.9 events per hour; an oxygen desaturation index (ODI) of 38.3 events per hour; a lowest minimum oxygen saturation recorded during sleep (O2Sa) of 75% and the cumulative percentage of time spent at saturations below 90% (CT < 90%) was 4%.

The patient was referred for continuous positive airway pressure (CPAP) therapy by the neurologist; the ventilatory treatment provided a temporary improvement of the symptoms, but poor compliance led to a poor clinical response.

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Six months later, the patient underwent an orthodontic consultation in order to try a custom-made oral appliance therapy. Despite the good compliance, he complained the persistence of OSAS-related symptoms.

A PSG control, performed with the oral device, revealed a residual mild positional OSA (AHI = 9; AHI supine = 30) for which the dentist suggested an evaluation to our ENT department.

Clinical examination of the oral cavity and oropharynx showed a suspected disproportionate macroglossia (Mallampati score 4). The fiberoptic nasopharyngoscopy revealed a huge hypertrophy of the base of the tongue, covered by healthy mucosa, filling the vallecula and pushing back the epiglottis. The patient did not complain any speech or swallowing dysfunction.

A CT scan with intravenous contrast showed a large well-circumscribed 4×5 cm mass with contrast heterogeneous enhancement, deeply located between the tongue base and the floor of the mouth, with a pedicle reaching the left sublingual region.

On magnetic resonance angiography, there was early and homogeneous enhancement without neovascularity or early venous drainage. Based on this pattern of enhancement, initial diagnosis favored vascular malformation such as hemangioma (Fig. 1).

As the initial treatment, the tumor was completely resected by a suprahyoid transcervical approach [4] involving clinically negative surgical margins, after a tracheostomy under general anesthesia (Fig. 2). The surgical specimen was an encapsulated bilobed red-tan lesion of firm consistency measuring 7.5 cm in its greatest dimension (Fig. 3).

Histologic examination showed a moderately cellular spindle cell neoplasm. Immunohistochemical evaluation showed strong expression of CD99 and CD34, but S100 and cytokeratin were negative. These findings were compatible with an atypical SFT. The patient was discharged 7 days after surgery, after removal of the tracheostomy tube.

Fig. 1 Sagittal MRA revealed early and homogeneous enhancement without neovascularity

Fig. 2 Surgical approach: tumor removal by a suprahyoid transcervical approach

Fig. 3 Surgical specimen

After 12 months, an MRI excluded relapse of the lesion. All symptoms, included daytime somnolence, disappeared (ESS = 4) and a PSG examination showed the normalization of the somnographic parameters (AHI = 1.2). Informed consent was obtained from the patient.

Discussion

UA and craniofacial abnormalities are said to play a role in OSAS together with neurological dysfunction. Less common causes of OSAS are tumors of the UA and parapharyngeal spaces. As far as we know, this is the first case report of a SFT leading to OSAS. SFTs are uncommon mesenchymal neoplasms that may be found in any location. To date, only 11 cases of SFT involving the tongue have been reported [5]. SFT is usually painless and slow growing and thus often remains unnoticed until it reaches a certain size. Although generally benign, malignant variants have been identified [6] and surgery is regarded as the treatment of choice [7, 8].

Conclusion

The complex interplay of multiple conditions that can be responsible of OSAS mandates a comprehensive evaluation that often requires the consultative input of various specialists. ENT evaluation, including flexible laryngoscopy, is warranted to identify anatomic factors that may contribute to OSAS. Although CPAP is the gold standard treatment for OSAS, a partial therapeutic result should alert clinicians to consider the possibility of a lesion obstructing the UA; therefore, each patient with OSAS should be examined carefully for the potential presence of an upper aero-digestive tract lesion.

Compliance with Ethical Standards

Conflict of interest The authors 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 Declaration of Helsinki 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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