Massive lymphoid hyperplasia presenting with obstructive sleep apnea secondary to lingual and palatine tonsil hyperplasia

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Abstract

Lingual tonsil hyperplasia is a rare condition that may cause obstructive sleep apnea (OSA). In the management of OSA, the lingual tonsils should be evaluated during the otorhinolaryngologic examination. We report the case of a 66-year-old man with findings of upper airway obstruction secondary to excessive lingual and palatine tonsil hyperplasia and with MRI findings of bilateral cervical lymphadenopathy. We review the clinical, radiologic, and histopathologic aspects of this case, and we discuss the surgical options for treating massive reactive lymphoid hyperplasia in conjunction with OSA.

Introduction

Lingual tonsil hyperplasia is a rare and serious disease that can be detected during a standard oropharyngeal examination. A hyperplastic lingual tonsil may compromise the upper airway, particularly in the retrolingual area, and it can lead to severe complications such as cardiac arrest, cerebral anoxia, and death, primarily because of the risk of a difficult intubation.[1] Exposure and removal of the lingual tonsils is difficult, especially in patients with obstructive sleep apnea (OSA).[2] Therefore, in patients with known or suspected OSA, it is essential to examine the area from the base of the tongue to the larynx with indirect rigid and/or flexible endoscopy. Polysomnography is another important technique for assessing this disorder.[3]

We describe a case of excessive lymphoid hyperplasia of the lingual and palatine tonsils presenting with OSA, and we discuss the diagnostic workup, histopathology, and treatment options for this disorder. We also review the literature.

Case report

A 66-year-old man presented to the Department of Otorhinolaryngology at the Istanbul University Cerrahpaşa Medical School with a 2-year history of snoring with snorting, choking, and apnea in addition to restless sleep and open-mouth sleep. Four months earlier, he had been evaluated for the same complaints at a public hospital, where cervical computed tomography (CT) with intravenous contrast had demonstrated bilateral mass lesions that extended from the palatine tonsils and tongue base to the aryepiglottic folds and epiglottis.

The mass was stained with contrast diffusely, and it was found to contain microcalcifications. The possibility of a lymphoma was considered, and a punch biopsy was performed on both palatine tonsils. Histopathologic evaluation of the biopsy material revealed diffuse reactive lymphoid hyperplasia.

Two months prior to this patient's presentation at our ENT department, contrast-enhanced magnetic resonance imaging (MRI) of his neck revealed bilateral hyperplasia of both the lingual and palatine tonsils and multiple small, bilateral cervical lymphadenopathies at the superior, middle, and inferior jugular chains (figure 1). The largest lymph node measured 18 × 15 mm.

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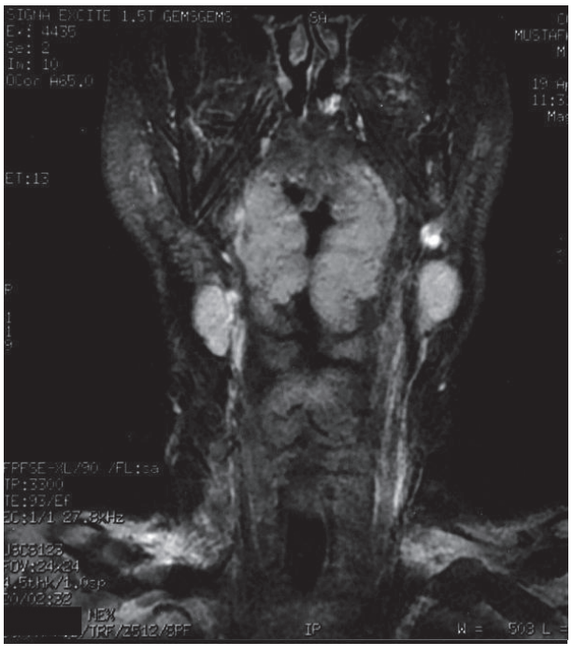

Figure 1. Prior to referral, this contrast-enhanced, T2-weighted, fat-saturation MRI shows the bilateral hyperplasia of the lingual and palatine tonsils and the jugular lymphadenopathies.

Also, abdominal and thoracic CT scans were obtained to screen for any additional lesions, and these findings were negative. Because lymphoma was still suspected, a left palatine tonsillectomy under general anesthesia was performed; because of intubation difficulties, a preoperative tracheotomy was performed.

Postoperative microscopic examination of the tonsillectomy specimen was again consistent with diffuse reactive lymphoid hyperplasia. The tracheotomy opening was closed within 24 hours of the tonsillectomy. One month later, one of the cervical lymph nodes was excised under local anesthesia. Histopathology of the specimen revealed diffuse lymphoid hyperplasia and mature plasma cell infiltration, mostly in the trabeculae. The lymphoid cells were primarily CD20 and CD3 cells; CD30 cells were observed in some immunoblasts.

The patient was referred to our clinic the following month. On physical examination, we noted the left tonsillectomy cavity and right palatine tonsil hyperplasia. On endoscopy, the lingual tonsils extended from the base of the tongue to the epiglottis inferiorly and to the retropharyngeal wall posteriorly, and they were hyperplastic on both sides. At the level of the hypopharynx, a marked obstruction of the airway was seen.

Two months after presentation, the patient underwent polysomnography. His apnea-hypopnea index (AHI) was 59.4, and his minimal saturation level was 77%. Because both lingual tonsils and the right palatine tonsil were obstructing the upper airway (figure 2, A), we removed all of them via sharp dissection and electrocautery under general anesthesia (figure 2, B). Again, a tracheotomy was necessary to prevent intubation difficulties. Microscopic evaluation of the resected material revealed reactive follicular hyperplasia (figure 3). No immediate complications were observed, and the patient was decannulated within 24 hours. Postoperatively, all the patient's complaints had resolved, and endoscopy revealed an open airway.

Five months postoperatively, follow-up MRI with contrast at the levels of the oropharynx, hypopharynx, and larynx found no mass in the airway (figure 4). Three months later, follow-up polysomnography revealed that the patient's AHI had fallen to 6 and his minimal saturation level had risen to 85%. More than 1 year later, the patient was well with no clinical obstructions.

Discussion

The lingual tonsils are part of the Waldeyer ring of lymphoid tissue. During routine otolaryngologic examinations, they are often unnoticed. Active inflammation or hypertrophy of a lingual tonsil can cause severe airway obstruction.[4] In addition to causing difficulties in intuba-

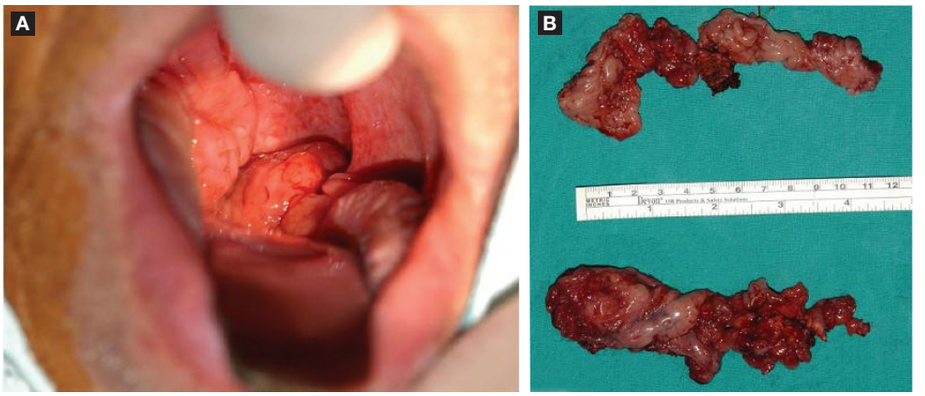

Figure 2. ***A:*** *The preoperative photograph shows the bilaterally hyperplastic lingual tonsils and the right palatine tonsil.* ***B:*** *Postoperatively, the masses resected from each side measure roughly 11 × 2 cm.*

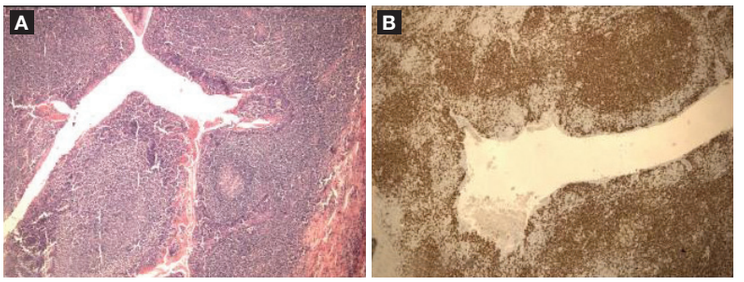

Figure 3. ***A:*** *On microscopic analysis, reactive follicles with large germinal centers are seen beneath the epithelium (H&E, original magnification ×40).* ***B:*** *The reactive follicles are mostly made up of CD20-positive B cells (CD20, original magnification ×40).*

tion, lingual tonsil hyperplasia may aggravate sleep disturbances. Before instituting any treatment for presumed lingual tonsil hyperplasia, it is important to perform a differential diagnosis to rule out cancer and specific granulomatous disorders such as tuberculosis.[5]

Reactive follicular hyperplasia is characterized by (1) the presence of enlarged lymph follicles with prominent germinal centers and (2) the appearance of plasma cells, initially in the medullary cords of the lymph node and later throughout the interfollicular pulp of the node. It can be detected in lymph node biopsies. When histologic features do not indicate any specific cause of the reaction, the finding is called *nonspecific reactive hyperplasia.*[6]

In a large autopsy study of 497 cadavers, Breitmeier et al examined the prevalence of lingual tonsil hyperplasia and its possible relationship with tonsillectomy.[1] They found an enlarged lingual tonsil in only 16 cases (3.2%), and they found no association with tonsillectomy.

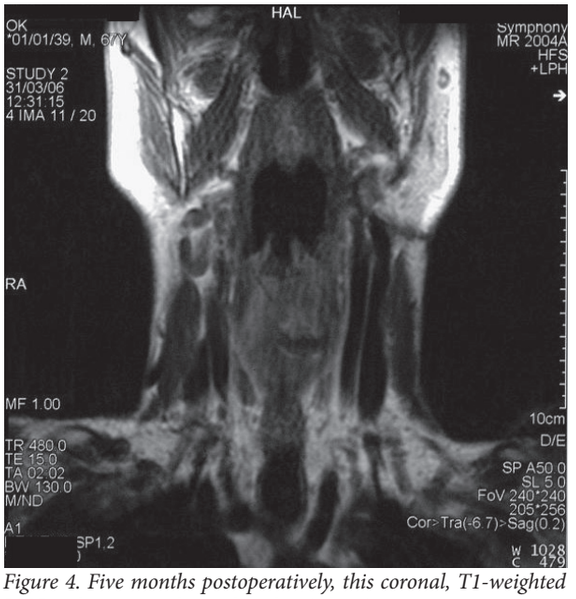

Figure 4. Five months postoperatively, this coronal, T1-weighted MRI with contrast shows that the airway is patent.

Lingual tonsillectomy is difficult to perform because achieving access to the valleculae is difficult. The different techniques for lingual tonsillectomy include sharp dissection, laser ablation, suction diathermy, cryotherapy, ultrasonographic coagulating dissection, and bipolar radiofrequency plasma excision.[7]

In our case, the enlargement of the lingual tonsils, which extended nearly to the supraglottic area, was the main reason for our patient's OSA. The presence of the clinically enlarged lingual and palatine tonsils combined with the finding of multiple small, bilateral cervical lymphadenopathies on MRI raised a suspicion of lymphoma, but a systematic evaluation with multiple biopsies demonstrated the benign nature of the hyperplasia. After the diagnosis of diffuse reactive lymphoid hyperplasia was established, we performed sharp dissection and electrocautery on both lingual tonsils and on the right palatine tonsil to relieve the sleep disorder symptoms. The postoperative polysomnography and MRI demonstrated the effectiveness of this intervention. We believe that sharp dissection and electrocautery are adequate, and that they can be performed without the need for any other sophisticated equipment.

References

1. Breitmeier D, Wilke N, Schulz Y, et al. The lingual tonsillar hyperplasia in relation to unanticipated difficult intubation: Is there any relationship between lingual tonsillar hyperplasia and tonsillectomy? Am J Forensic Med Pathol 2005;26(2):131-5.
2. Robinson S, Ettema SL, Brusky L, Woodson BT. Lingual tonsillectomy using bipolar radiofrequency plasma excision. Otolaryngol Head Neck Surg 2006;134(2):328-30.
3. Johnson JT. Obstructive sleep apnea. In: Gates GA, ed. Current Therapy in Otolaryngology–Head and Neck Surgery. St. Louis: Mosby; 1998:422-4.
4. Caylakli F, Akkuzu B, Avci S. Lingual tonsillar hypertrophy: A case report. Kulak Burun Boğaz Ihtis Derg 2004;13(1-2):28-30.
5. Marcos Ordóñez M, Benito Orejas JI, Blasco Gutiérrez MJ, et al. Oropharyngeal tuberculosis. Report of case in a lingual tonsil [in Spanish]. Acta Otorrinolaringol Esp 1999;50(7):575-8.
6. Stansfeld AG. Inflammatory and reactive disorders. In: Stansfeld AG, D'Ardenne AJ, eds. Lymph Node Biopsy Interpretation. London: Churchill Livingstone; 1992:55-115.
7. Krespi YP, Har-El G, Levine TM, et al. Laser lingual tonsillectomy. Laryngoscope 1989;99(2):131-5.