A CARE-compliant article: Lymphangiomatous polyps of the palatine tonsils in a miner

A case report

Weigang Gan, MDa,d, Yu Xiang, MBb, Xinrong He, MMc, Yiyuan Feng, MMd, Hongbin Yang, MDd, Hai Liu, MDd, Shixi Liu, MDa, Juan Meng, MDa∗

Abstract
Rationale: Lymphangiomatous polyps of the palatine tonsils are benign tumors that are rare in both adults and children. Most patients suffering from this disease present with nonspecific symptoms similar to those of chronic tonsillitis.

Patient concern: We report a case of a 21-year-old male miner who presented with a chronic history of a foreign body sensation in the oropharynx and an intermittently sore throat.

Diagnosis: The patient was preoperatively diagnosed with the palatine tonsils neoplasm.

Interventions: The neoplasm with palatine tonsils was completely resected under general anesthesia. The tissue was sent for histological examination, and the diagnosis was lymphangiomatous polyps of the palatine tonsils.

Outcome: The surgical outcome was good, and no surgical site infection was recorded. After 12 months of follow-up, the miner was asymptomatic with no recurrence.

Lessons: Tonsillectomy is a curative method to address lymphangiomatous polyps (LAPs) of the tonsils which resulted in no recurrence during the clinical follow-up period. The etiology of this rare disorder and potential pathogenesis should be studied in the future, which would help prevent its occurrence.

Abbreviations: ENT = Ear, Nose and Throat, LAPs = lymphangiomatous polyps, VEGF-C = vascular endothelial growth factor C.

Keywords: lymphangiomatous, neoplasm, palatine tonsils, polyp

1. Introduction

Polyps occurring the upper respiratory tract are usually found in the nasal or laryngeal cavity but are rarely located in the oropharynx. Bilateral diffuse lymphangiomatous polyps (LAPs) of the palatine tonsils have rarely been reported. They may cause symptoms related to local irritation, such as chronic tonsillitis, and their diagnosis is confirmed by postoperative histopathology. LAPs are a kind of benign lesion of the tonsils and are different from acute and chronic tonsillitis. We report a case of tonsillar neoplasms with a unique appearance, showing diffuse mamillary lesions that were histopathologically diagnosed as LAPs. Of note, the patient had worked underground as a miner for a long period of time, which may suggest that LAPs of the palatine tonsils are related to a dusty or polluted environment. A literature review and discussion are included.

2. Case report

A 21-year-old male patient presented to the ENT (Ear, Nose and Throat) outpatient department with a longer than 1-year history of a foreign body sensation and an intermittent sore throat, which were refractory to medical treatment. The patient had no history of swallowing or breathing difficulty, fever, arthralgia, or palpitation and was prescribed drugs to treat gram-positive bacteria when he visited a doctor for consultation, which resulted in a temporary alleviation of the condition, followed by a prompt recurrence. The patient had never used tobacco or alcohol and did not suffer from allergies or take drugs regularly. He was a miner working underground 8 hours a day for >3 years. Pre- and postoperative physical examinations showed that both tonsils appeared villous, papillomatous, and enlarged (Fig. 1). The results of routine laboratory tests, including a biochemical analysis, blood cell count, coagulation function, and urinalysis, were normal.

The patient underwent tonsillectomy with traditional dissection without complication. Specimens were sent for routine postoperative histological analysis. Both tonsils showed a papillary appearance, with nodules measuring from 0.2 to 0.5 cm. Overall, each tonsil measured 4.0 cm × 2.0 cm × 1.5 cm
Microscopically, the lesion was composed of dilated lymphatic channels packed with lymphatic cells and a hyperplastic fibrous stroma and was covered with a squamous epithelium (Fig. 2).

After 1 year of follow-up, the patient confirmed that he was asymptomatic.

The postoperative pathology confirmed the diagnosis as lymphangiomatous polyps of the palatine tonsils.

(Fig. 1). Microscopically, the lesion was composed of dilated lymphatic channels packed with lymphatic cells and a hyperplastic fibrous stroma and was covered with a squamous epithelium (Fig. 2).

After 1 year of follow-up, the patient confirmed that he was asymptomatic.

The postoperative pathology confirmed the diagnosis as lymphangiomatous polyps of the palatine tonsils.

3. Discussion

Lymphangiomatous polyps of palatine tonsils are rare benign lesions in the oropharynx, and 30 or fewer cases had been reported by the end of the first decade of the 21st century. However, Kardon et al. reported that LAPs represented approximately 1.9% (26/1389) of all tonsillar neoplasms, that is, they were not as rare as previously reported. Barreto et al. reported an extremely small number (14) of LAP cases recorded.
between 1983 and 2008 at the Medical School of the University of Campinas, including 9 men and 5 women. The youngest patient was 9 years old, and the oldest patient was 53 years old. Head and neck LAPs are less frequently found in the tonsils than in subcutaneous tissue, the larynx, parotid gland, mouth, and root of the tongue.\(^{[5]}\) LAPs originate from the lymphoid stroma, which is morphologically different from papillary hyperplasia, and blend with the tonsillar parenchyma.\(^{[6]}\) The earliest recognition of LAPs of tonsils dates to the beginning of the 1960s and was due to the development of histopathological techniques.\(^{[7]}\) Before the development of these techniques, LAPs on tonsils were identified as benign lesions but were referred to by various names including angiomas,\(^{[8]}\) angiofibromas,\(^{[9]}\) and fibroangiomas.\(^{[10]}\) LAPs are characterized by symptoms of a foreign body sensation, sore throat, and tonsillar mass, similar to tonsillitis, and should be distinguished from lymphangiectasia, fibroepithelial polyps, lymphoma, and papilloma.\(^{[11]}\) When tonsillar neoplasms or unhealthy tonsils grow sufficiently large, dysphagia and dyspnea may occur.

Epidemiologically, LAPs are found more often unilaterally than bilaterally and are more frequent in adults than in children.\(^{[11]}\) Histologically, the typical appearance of LAPs is usually represented by dilated lymphatic channels filled with fibrous stroma, adipose stroma, and/or lymphoid tissue, and the composition ratio can result in different phenotypes, some of which include pedunculated polyoid masses, whereas others are papillary. Some of the lesions have a more papillary architecture and minimal stromal fibrosis. In some but not all LAPs, nested epitheliotropism of lymphocytes can be observed on the surface.\(^{[12]}\) Heffner\(^{[12]}\) proposed that LAPs should be classified as hamartomatous proliferations rather than neoplasms because the 3 mentioned elements observed in dilated lymphatic channels are routinely found in the tonsillar fossae and are merely arranged in a distinct type in LAPs. The pathogenesis of LAPs is not very clear and is mainly described by 3 hypotheses, including the overgrowth of lymphatic tissue out of the sac, hyperplasia of endothelial fibrillar membranes regulated by Prox-1 and vascular endothelial growth factor C (VEGF-C), and mucosal congestion leading to a polypoid lesion, caused by chronic tonsillitis.\(^{[13]}\)

Based on the patient reported, whose work place was underground and dusty, we propose that heavy dust, especially in the presence of chemical stimulation, can obstruct the tonsillar crypt, which may cause lymphangiectasia and polypoidal swelling. In our opinion, environmental factors may be involved in the development of LAPs to some extent. Consequently, the etiology of this rare disorder should be studied in the future, which would help prevent its occurrence.

Therapeutically, conventional treatment with surgical excision has been demonstrated to be an effective therapy for LAPs of the palatine tonsils, which comprise a rare type of benign tumor. In this case, which we followed up, no recurrence was confirmed for 1 year after the operation, similar to Kardon observations.\(^{[13]}\)

4. Conclusion

In conclusion, we report a rare case of LAPs of the palatine tonsils in a patient who had worked for a long time in an environment containing mineral dust, which may suggest that obstruction of the tonsillar crypt, followed by subsequent lymphangiectasia and polypoidal swelling, can play an important role in the pathogenesis of LAPs. LAPs are a kind of benign tonsillar lesion, rarely reported in the literature that can be precisely diagnosed by histopathology based on their characteristic manifestations. Tonsillectomy is a curative method to address LAPs of the tonsils that will prevent recurrence during clinical follow-up.

Author contributions

Conceptualization: Juan Meng.
Data curation: Yu Xiang, Yiyuan Feng.
Methodology: Xinrong He.
Resources: Hai Liu, Hongbin Yang.
Supervision: Shixi Liu.
Writing – original draft: Weigang Gan.
Writing – review & editing: Juan Meng.

References

[1] Iliadou E, Papapetropoulos N, Karamatzanis E, et al. Primary lymphangiomatous polyp of the tongue in a 9-year-old boy: a case presentation and literature review. Case Rep Otolaryngol 2016;2016:1503202.
[2] Chen HH, Lovell MA, Chan KH. Bilateral lymphangiomatous polyps of the palatine tonsils. Int J Pediatr Otorhinolaryngol 2010;74:87–8.
[3] Kardon DE, Wenig BM, Heffner DK, et al. Tonsillar lymphangiomatous polyps: a clinicopathologic series of 26 cases. Mod Pathol 2000;13:1128–33.
[4] Barreto I, Costa AF, Martins MT, et al. Immunohistochemical study of stromal and vascular components of tonsillar polyp: high endothelial venules as participants of the polyp’s lymphoid tissue. Virchows Arch 2011;459:65–71.
[5] Stal S, Hamilton S, Spira M. Hemangiomas, lymphangiomas, and vascular malformations of the head and neck. Otolaryng Clin North Am 1986;19:769–96.
[6] Harrison GI, Johnson LA. Lymphangioma of the tonsil. Report of a case with a critical review of the literature. Ann Otol Rhinol Laryngol 1960;69:961–8.
[7] Pallestrini EA, Ameli M. Polyoid lymphangiomia of the palatine tonsil. Arch Ital Otol Rinol Laringol 1966;77:343–8.
[8] Ormerod FC. Angioma of the tonsil. J Laryngol Otol 2007;41:797–800.
[9] Hyams VJ. Differential diagnosis of neoplasia of the tonsil. Clin Otolaryngol Allied Sci 1978;3:117–26.
[10] Hara HJ. Benign tumors of the tonsil - with special reference to fibroma. Archiv Otolaryngol 1933;18:62–9.
[11] Cengiz BP, Acar M, Giritli E. A pedunculated lymphangiomatous polyp of the palatine tonsil: a case report. Braz J Otorhinolaryngol 2013;79:402.
[12] Heffner DK. Pathology of the tonsils and adenoids. Otolaryngol Clin North Am 1987;20:279–86.
[13] Hockstein NG, Carpentieri D, Shah UK. Pathology quiz case 2. Tonsillar lymphangioma. Arch Otolaryngol Head Neck Surg 2002;128:12101212.