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CASE REPORT

A CLINICAL CASE OF TONSILLAR LYMPHANGIOMATOUS POLYP

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Abstract. Introduction: *Tonsil lymphangiomas are extremely rare benign tonsil tumors. They could be asymptomatic, especially when they are small, and in these cases, they are most often found by chance on physical examination.* ***Clinical case:*** *The authors present a 67-year-old man with complaints of discomfort, a sore throat, and an unspecific formation on his right tonsil. Upon microscopic examination, we found a polypoid mass covered by squamous epithelium with a stroma composed of lymphoid tissue.* ***Discussion:*** *Benign tonsillar tumors are significantly more common than malignant ones. Lymphangiomatous polyps located in the tonsillar region, on the other hand, have been described as very rare, and their etiology and pathogenesis remain unclear. They tend to occur in areas where lymph vessels are abundant, with more than 90% of all lymphangiomas occurring in the head and neck region.* ***Conclusion:*** *Lymphangiomatous polyps should be considered in the differential diagnosis of all benign tumors, and it is extremely important to differentiate them from malignant tonsil lesions.*

Key words: *tonsillar lymphangiomatous polyps, benign tumors*

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INTRODUCTION

Benign tonsil tumors are very rare compared to malignant tumors. There are various types of benign polypoid neoplasms, such as angiomas, polypoid tonsil lymphangiomas, hamartomatous tonsil polyps, lymphoid polyps, and tonsil lymphangiomatous polyps, that have been described in the literature. According to Kardon et al., there are ten well-documented cases of tonsillar lymphangiomatous tumors [1]. Due to the different descriptions of the same or similar histological forms, it is difficult to determine the actual frequency of the tumor itself [2, 3].

The pathogenesis of tonsillar lymphangiomatous polyps is controversial. Chronic tonsillogenic inflammation and the associated obstruction of the draining lymph canals are discussed as the most likely mechanisms of occurrence. These disorders of lymphedema eventually lead to mucosal prolapse and the appearance of polypoid edema [4].

Tonsil lymphangiomas are most commonly asymptomatic when they are small in size, and in these cases, they are found by chance on physical examination. Rarely, symptoms could be manifested by complaints, e.g., dysphagia or a feeling of a foreign body in the throat, which can cause an unproductive cough. Reaching larger sizes can compress the sur-

rounding structures, creating breathing difficulties, stridor, or nausea. Difficulty swallowing may progress and lead to profuse salivation [5].

Mesopharyngoscopy most often reveals the tumor formation located on the surface of the palatine tonsils. They are almost always unilateral, but in isolated cases could be bilateral [6]. It's not typical, but when they are symptomatic, as in our case, they can hardly be evaluated as benign lesions before pathohistological examination and provoke a wide differential-diagnostic process.

CLINICAL CASE

This case is about a 67-year-old man from Stara Zagora with complaints of discomfort and sore throat, more pronounced on the right side 4 months ago. Two weeks before the visit, he noticed a formation on his right tonsil that began to interfere with food intake.

A physical examination of the oral cavity was conducted and showed a pale pedicular formation originating from the right palatal tonsil. Upon palpation, the formation presented as a painful, dense mass with a smooth surface. No crepitations, surrounding hyperemia, exudates, or ulcerations were observed.

No pathological changes were found in the rest of the oral, pharyngeal, and laryngeal cavities. No cervical lymphadenopathy was detected, and no deviations from the norm were found in the blood count.

The neoplasm was assessed as an inflamed tonsillar papilloma. Intra-oral excision/biopsy was performed under local anesthesia. The macroscopic dimensions were 14x11 mm. The tumor mass was firm and smooth, with a small foot base. No recurrences had been observed upon follow-up until 11 months after the procedure.

Microscopic findings

As shown in the images below, the polypous mass was covered by squamous epithelium. In the stroma, lymphoid tissue is observed, resembling lymphoid follicles. In the connecting tissue stroma, many vessels are found. The vessels are of lymphatic and blood origin (Figure 1 a, b).

Special staining

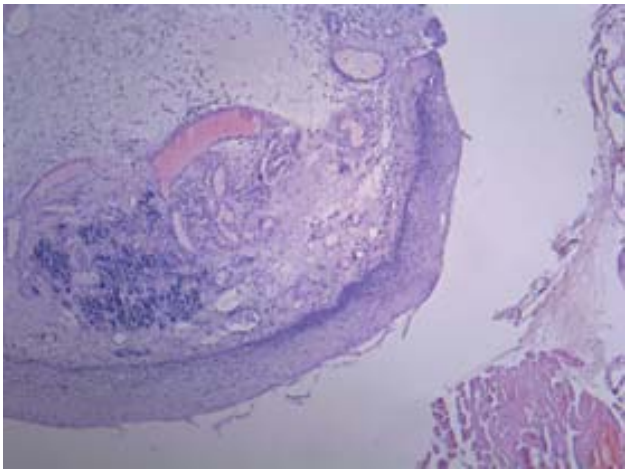
The polyp was stained by markers for blood vessels (CD31 and CD34), lymph vessels (D2-40) (Figures 2a and 2b), lymphocyte cell markers (CD3, CD20, CD45) (Figures 3 a and 3b), and a marker for myoepithelial and myofibroblastic cell markers (α-SMA).

The blood vessels showed reactivity with anti-CD31 and anti-CD34. Lymphatic vessels – with anti-D2-40. Smooth muscle actin is present in the walls of dilated lymphatic blood vessels. The hematologic markers (CD3 [T-cell], CD20 [B-cell], and CD45) are expressed in the lymphoid cells in the lymphoid tissue.

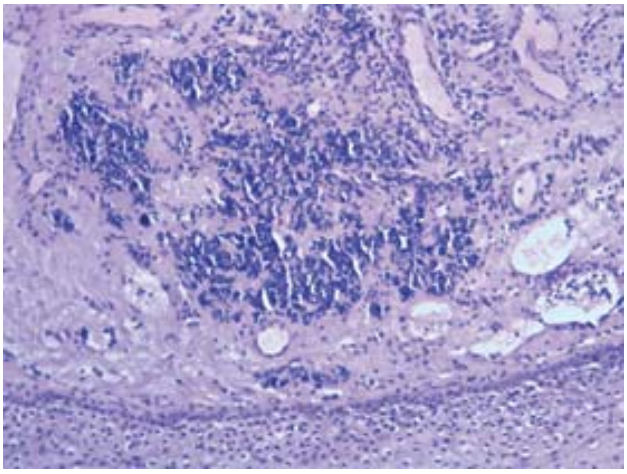
DISCUSSION

Lymphangiomatous polyps located in the tonsillar region have been described as extremely rare, and their etiology and pathogenesis remain unclear. In most cases, they are asymptomatic and are found incidentally.

These tumors tend to occur in areas where lymph vessels are abundant, with more than 90% of all lymphangiomas occurring in the head and neck region. Most commonly, they involve the skin and sub-

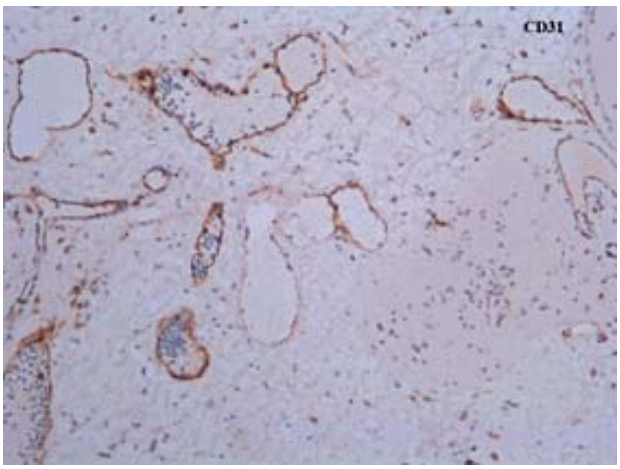


a. Lymphangiomatous polyp HE

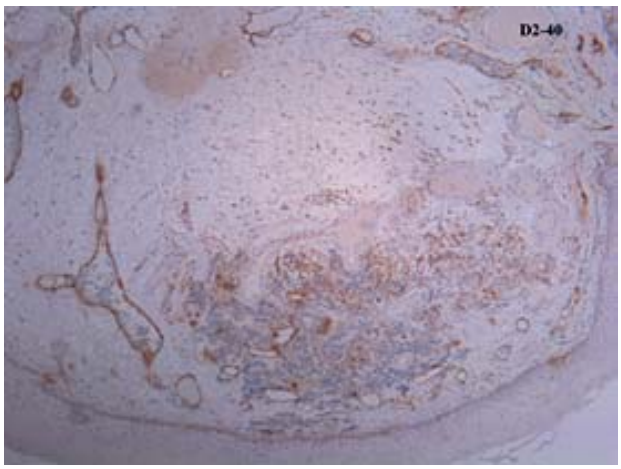


b. Lymphangiomatous polyp HE0001

Fig. 1 a, b. Microscopic findings



a. Lymphangiomatous polyp CD0031-0003



b. Lymphangiomatous polyp D2-40

Fig. 2 (a, b). The polyp was stained by markers for blood vessels (CD31 and CD34) (a) and for lymph vessels (D2-40) (b)

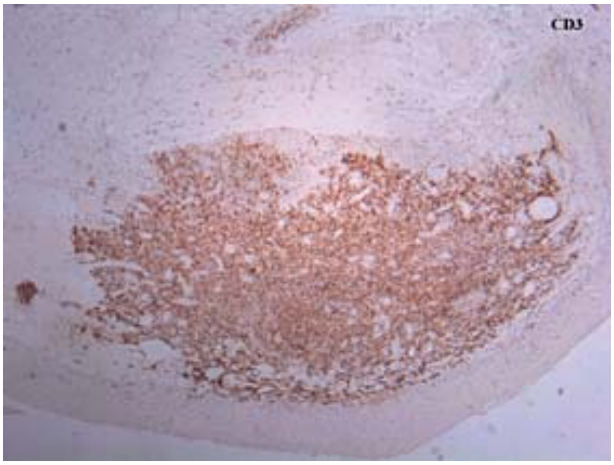


Fig. 3a. Lymphangiomatous polyp CD3

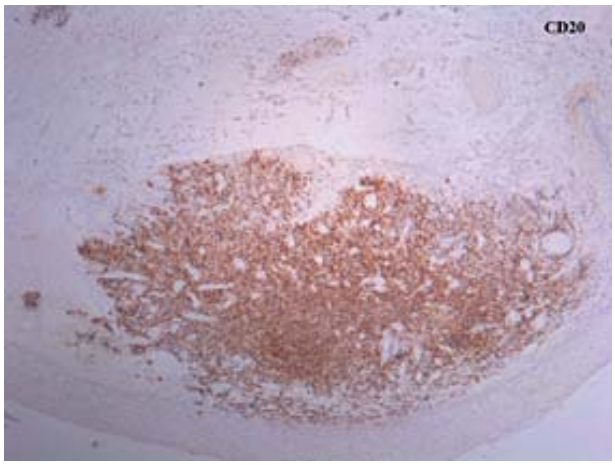


Fig. 3b. Lymphangiomatous polyp CD20

cutaneous tissues, but some articles describe localizations like the nasal cavity, larynx, parotid gland, mouth, and tongue [7].

Histologically, formations are covered with squamous epithelium. The stroma of the polyp consists of various components, mainly collagen tissue with varying degrees of density of collagen fibers, adipose tissue, dilated lymph vessels, and lymphoid tissue [8].

Immunohistochemically, CD34 is less frequent than CD31 for lymphatic vessels [9]. In our case, CD31 and CD34 were detected both in blood and lymph vessels. The hematologic markers, including CD3, CD20, and CD45, showed polymorphous infiltration of the vascular channels and stroma.

There is no consensus on the pathogenesis of these benign tumors, but the most widely discussed hypothesis is that their occurrence is due to obstruction and dilation of lymph vessels provoked by chronically persistent inflammatory processes in the tonsils. The prognosis for tonsillar lymphangiomas is good. In the available literature, we did not find a described case of malignant degeneration. Treatment is either partial excision or, most often, extensive surgical removal, with tonsillectomy being the preferred procedure to ensure complete removal. This is because of the lack of encapsulation, and recurrences of lymphangiomas are generally common if not completely removed [10].

Attempts have been made to treat them with ionizing radiation, but most of the authors conclude that these lesions are not radiation-sensitive, so radiotherapy is ineffective and only unduly increases the risk of developing various complications, including malignancies of the head and neck [11].

CONCLUSION

Lymphangiomatous polyps of the palatine tonsils are very rare, benign lesions that manifest as tonsil-

lar growths. The anamnesis and the clinical examination are integral parts of the diagnostic process, but for their definitive establishment, histological examination and immunohistochemical analysis are required. Lymphangiomatous polyps should be considered in the differential diagnosis of all benign tumors, and it is extremely important to differentiate them from malignant tonsil lesions with detailed histological analysis.

Treatment is only surgical removal with a partial or total tonsillectomy. If partial, the patient should be followed for recurrence.

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Animal and human rights statement: *All procedures performed in this study 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.*

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