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Lingual Cystic Lymphangioma: About a Case and Literature Review

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Abstract

Lymphangioma is a rare benign tumor of the lymphatic vessels of hamartomatous nature. We report a case of lingual lymphangioma in a 2-year-old child, revealed by macroglossia. The radiology suspected the lesion. Anatomopathological examination confirmed the diagnosis of cystic lymphangioma, and determined its characteristics.

Keywords

Lymphangioma, Tongue, Antananarivo

1. Introduction

Lingual masses in children are a rare entity and are mostly cystic nature. Cystic lymphangioma is a rare benign vascular tumor that can occur anywhere except the brain. It occurs in 90% of cases during the first two years of life. In the oral cavity, the tongue is the most common site of lymphangioma, however, this lesion is exceptionally reported in the floor of the mouth [1]. We report a Malagasy case of cystic lymphangioma of the tongue in a 2-year-old child.

2. Observation

This was a 2-year-old boy seen in stomatology consultation for macroglossia. There was no difficulty in swallowing, or breathing. Moreover, there was no family history of the similar presentation. Physical examination showed an ill-defined limit macrocystic lesion of the tongue. The radiology suspected the lesion. The patient was treated by partial surgical excision of the tongue and he showed significant improvements during the following weeks. The macroscopic

examination of the specimen shows four non-reconstitutable, brown, spongy fragments ranging from 4 to 7 cm in long diameter. The section slices were polymicrocystic. Histological examination showed the mucosa and muscularis of the tongue, the site of vascular proliferation, composed by vessels of various sizes, sometimes enlarged, lined by non-atypical endothelial cells. These vessels were devoid of red blood cells, but occasionally contained lymphocytes. The stroma was edematous, infiltrated with lymphocytes, with formation of lymphoid follicles. The diagnosis retained was that of a lingual cystic lymphangioma (**Figure 1**).

3. Discussion

Lymphangioma is one of the rarest congenital malformations of the neck, representing 6% of benign tumors in children and between 6% and 8% of congenital neck anomalies [2]. It is a benign congenital lesion of the lymphatic system most likely related to aberrant sequestration of lymph tissue and/or vessels occurring during the embryonic development phase, resulting in blocked lymphatic pathways; these progressive expansions under hydrostatic pressure of the lymphatic fluid until balance with the surrounding tissues is reached [3]. Lymphangiomas can occur in any part of the body. In the oral cavity, the tongue is the main site. The neoplasm is present from birth, 80% - 90% are diagnosed before the age of three [4], which concord with our study. The occurrence in adults is very rare [5]. Lingual lymphangioma can be asymptomatic. However, depending on the size of the tumor, complications may be occurred related to the compression and displacement of adjacent structures: breathing difficulties, dysphagia, sometimes infection and hemorrhage. In our case, this neoplasm was revealed by macroglossia limited to the tongue. Extensions to the entire oral cavity are possible, and even to the cervical level [6], and event to the mediastinal level [7].

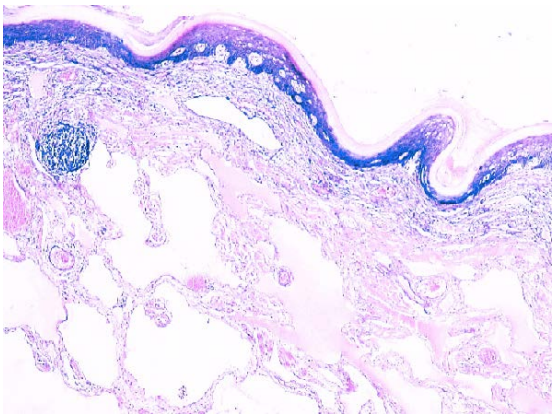

Figure 1. Tongue: cystic lymphangioma: tumor proliferation of lymphatic vessels of various sizes. Staining: Haemathoxylineosin. Magnification: ×200. Source: Unit of anatomy pathology of Hospital University Joseph Ravoahangy Andrianavalona.

Ultrasound, performed in the first intention, can show the cystic nature of the tumor. Computed tomography is helpful in the pre-therapeutic assessment. It allows to appreciate the aspect to the tumor, its limits and to analyze the extensions towards the parotid, parapharyngeal and mediastinal regions, which could modify the therapeutic protocol. Imaging also allows the differential diagnosis of extensive lymphangiomas with other cervico-facial lesions of cystic nature [8].

Histologically, lymphangiomas are classified into: capillary lymphangiomas, composed of small, thin-walled lymphatic vessels; cavernous lymphangiomas, composed of dilated lymphatic vessels surrounded by an adventitia; and hygroma, consisting of large lymphatic cysts. Cystic lymphangiomas called microcystic when the cysts measure less than two centimeters, and macrocytic when cavities are larger than two centimeters [3].

Complete surgical excision is the best management approach but sclerotherapy is also effective according to the literature [9]. In our case, as in the literature, the surgery presented good results. The other treatment modalities that have been employed with variable results include simple drainage, steroids, aspirations, radiation, laser excision, radio-frequency ablation and cauterization. Radiotherapy is currently abandoned [3]. Based on some studies, sirolimus, acting by inhibiting the lymphatic vessel regeneration, invasion, and vascular endothelial growth factor secretion is used when other treatment is failed [10].

4. Conclusion

Cystic lymphangioma of the tongue is a benign tumor known for its occurrence at an early age. It is a rare benign lymphatic malformation, but potentially serious because of its evolution characteristics and its tendency to dissect. The differential diagnosis is mainly the other vascular tumors such as hemangiomas, but the anatomopathological examination makes the positive diagnosis. Surgery is the treatment of choice. Recurrences are frequent, therefore long-term control is needed.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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