Surgical Treatment of Tongue Lymphangioma in a Pediatric Patient: A Case Report

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ABSTRACT

Lymphangiomas are benign tumors that occur predominantly in the head and neck regions. They are very common in children and are usually diagnosed at birth or during the first years of life. Understanding their clinical features is crucial to an early diagnosis. The purpose of this paper was to report the case of a 3-year-old patient with a nodular lesion that evolved for 2 years on the tongue's dorsal surface, highlight the surgical treatment and characteristics of this kind of pathology, and explore the differential diagnosis.

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ymphangiomas are hamartomatous benign tumors of lymph vessels that show predilections for head and neck regions and are frequently diagnosed at birth or during childhood.¹⁻³ Not considered true neoplasms, lymphangiomas probably originate from a sequestration of lymphatic tissue that does not communicate with the rest of the lymphatic system; hence, they constitute a developmental abnormality.^{1,3,4} They are characterized as lobed masses or cystic lesions. In the oral cavity, the lesions may occur on the floor of the mouth, cheek mucous membranes, lips, and commonly

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the tongue. In 36% of cases, they occur on the tongue's dorsal surface. $^{3.5\text{-}7}$

Clinically, the oral lesions are presented as tumoral masses resembling a cluster of translucent blisters that may become reddish blue due to secondary hemorrhage. Some authors describe them as "frog spawn" or "tapioca pudding."⁸ Depending on the lesion's extent, it can involve great portions of the tongue, causing macroglossia and dysfunctional problems in speech, swallowing, and breathing, which makes them a challenge to treat.^{5,7,9} Surgical removal of the tumor is the treatment of choice for this kind of pathology. Conservative treatment or partial resections are normally associated with high rates of recurrence.^{6,10}

The purpose of this article was to report the case of a patient presenting with tongue lymphangioma, pointing out the surgical treatment and most important features for differential diagnosis.

CASE REPORT

A 3-year-old male patient was referred to the maxillofacial surgery and traumatology department of Hospital de Clínicas de Porto Alegre, state of Rio Grande do Sul, southern Brazil, for evaluation of a tongue lesion that

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Figure 1. Nodular lesion on the tongue's dorsal surface.



Figure 2. Tomographic isodense nodular lesion on the tongue's dorsal surface.



Figure 3. Specimen sent for histological examination.



Figure 4. Postoperative, after suture of muscular planes.

had evolved for 2 years. The patient's mother said the lesion bled during breast-feeding and produced some painful episodes.

Upon physical evaluation, a nodular, blister-like, dark, and clear-colored lesion was observed. The clinical features included: sessile insertion; exophytic growth; hardened body; rough surface; size 15 mm x 15 mm; painless when touched; and absence of spontaneous bleeding (Figure 1). The patient underwent a computed tomography angiography, which revealed the vascular nature of the tumor (Figure 2).

Under general anesthesia, the surgical procedure consisted of total removal of the tumoral mass (Figure 3). The histological examination during surgery confirmed healthy tissue on the removed lesion's border. The region was sutured by muscular planes, which allowed maintenance of adequate tongue functioning and taste (Figure 4).

Histopathologic results showed an abundance of dilated lymphatic vessels underneath the tongue epithelium, which confirmed the diagnosis of lymphangioma (Figure 5).

In postoperative consultations, healing was as expected while normal tongue functioning was maintained. Eight months after surgery, no functional abnormalities or recurrences were found (Figures 6 and 7).

DISCUSSION

Lymphangiomas typically occur in the head and neck regions in approximately 75% of cases. Most cases are diagnosed during childhood—50% at birth and 50%

thereafter, with nearly 90% developing in children younger than 2-years-old.^{4,6,11} There is no relation to gender or familiar tendency.^{10,11} The incidence of lymphangiomas among all benign tumors is 6% in chil-



Figure 5. Histopathologic results have shown dilated lymphatic vessels underneath the tongue epithelium.



Figure 6. Postoperative control of 7 days.



Figure 7. Postoperative control of 8 months.

dren.¹² Initially, lymphangiomas follow the patients' development, but after some time they may present a slow regression.¹⁰ Repeated episodes of upper airway infection or occasional traumas in tongue lymphangiomas may cause an enlargement of the tumor size, macro-glossia, and functional and aesthetic complications.^{9,10}

In the oral cavity, lymphangiomas show a predilection for the tongue in approximately 51% of cases, of which 36% are located on the tongue's dorsal surface.⁶ The tumor presented here consisted of a nodular lesion, 15 mm in diameter, with a multiple and transparent blister surface—a very common feature found in superficial lymphangiomas.⁸ There are other tumors, such as pyogenic granuloma, fibroma, and papilloma that occur on the tongue and must be considered in differential diagnosis.¹³⁻¹⁵

Some singularities must be observed in surgical treatment of tongue lesions, such as preservation of taste, when possible. Structural integrity of the tongue is very important for the speech development of pediatric patients, making early diagnosis highly relevant.⁷

The most accepted treatment in the literature consists of total surgical excision of the tumor. Nevertheless, there are many other conservative methods described, including radiation, laser therapy, sclerotherapy, ^{9-11,15} and, most recently, radio frequency ablation.^{9,10,16} Due to the tumor's infiltrative nature, recurrence is present in all kinds of treatments, although more frequently in nonsurgical ones. In total resections, the rate of recurrence is 17%, showing the necessity for delayed postoperatory follow-up.¹⁰

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