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Case Report

A rare case of oral lymphangioma of tongue

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ABSTRACT

Lymphangiomas are congenital benign tumors of lymphatic vessels, which are localized in the head and neck area in about 75% of cases. Most cases occur in the head and neck region. Oral cavity lymphangiomas are rare. We here describe one such rare case of lymphangioma involving the tongue and was successfully treated with laser therapy.

Keywords: Lymphangiomas, Congenital benign tumors, Laser

INTRODUCTION

Lymphangiomas are congenital benign tumors of lymphatic vessels, which are localized in the head and neck area in about 75% of cases.[1] Most of the cases are present since birth, and about 80% are developed before 2 years of age and are rarely diagnosed in adults.[2] They indicate localized abnormal development of the lymphatic system. Lymphangiomas are classified as microcystic (capillary lymphangiomas), macrocystic (cavernous lymphangiomas) and cystic hygromas according to the size of the lymphatic cavities incorporated.[3] Other common sites, outside the head and neck, include the axilla, shoulder, chest wall, mediastinum, abdominal wall and thigh.[4]

In the oral cavity lymphangioma represents a very rare entity with more common predilection in the anterior two-third of the tongue resulting in macroglossia. Cases have also been described in palate, gingiva, lips, and mandibular alveolar ridge.[5]

Surgical resection still remains the best treatment for lymphangiomas; other treatment modalities include sclerotherapy with sodium morrhuate, dextrose, tetracycline, doxycycline, bleomycin, ethibloc as sclerotherapeutic agents. Radiation therapy, cryotherapy, electrocautery, steroid administration, embolisation, ligation, and laser surgery have also been proposed to treat lymphangioma.[6,7]

We present a rare case report of lymphangioma in the tongue, which was successfully treated by laser therapy.

CASE REPORT

A 16-year-old female patient presented to the Department of Oral and Maxillofacial Pathology with a painless swelling on the dorsal surface of anterior tongue. The lesion was there since three years and increased in size over the last four months. An intraoral clinical examination revealed the presence of a reddish pale colored lesion on the tongue (Figure 1). On palpation it was pebbly and soft in consistency giving the tongue a granular appearance.

To confirm the diagnosis, an incisional biopsy was made and the sample was referred to the Oral Pathology

department for examination. Histological examination revealed a cystic dialated, thin-walled lymphatic vessels that contain lymph with a few erythrocytes and lymphocytes located at subepithelially occupying the lamina propria (Figure 2). Thus, the diagnosis was confirmed as lymphangioma.

The lesion was treated with laser therapy. The procedure was facilitated due to the superficial location of the lymphatic vessels. After one year of following-up, there was no relapse of the lesion.

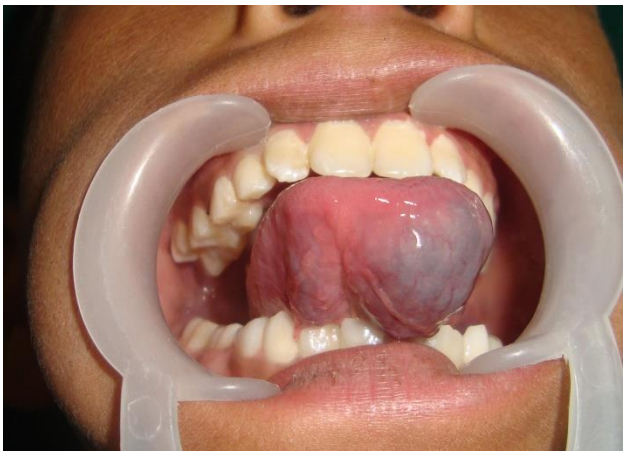

Figure 1: Clinical photograph showing papular lesions on tongue.

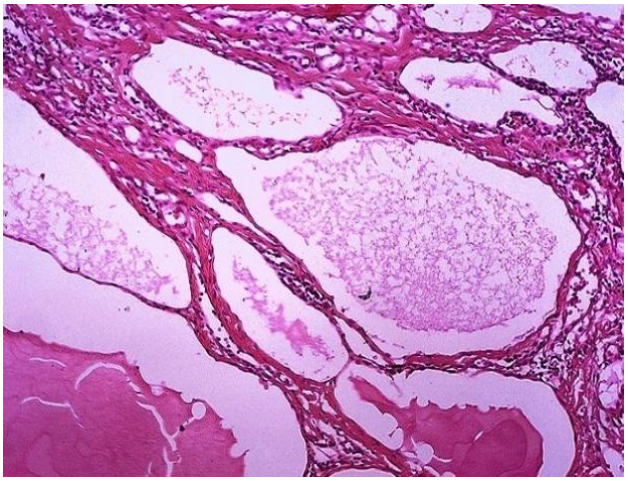

Figure 2: Photomicrograph showing thin walled lymphatic channels filled with lymph and few lymphocytes [H & E stain].

DISCUSSION

Lymphangiomas are rare congenital malformations of the lymphatic system that can occur throughout the body with greater predilection for head and neck. Three theories have been proposed to explain the origin of lymphangiomas. The first suggests that a blockage or arrest of normal growth of the primitive lymph channels occurs during embryogenesis, the second that the primitive lymphatic sac does not reach the venous system, while the third advances the hypothesis that, during embryogenesis, lymphatic tissue lays in the wrong area as a result these cells do not anastomose efficiently with bigger lymphatic vessels, they then provoke areas of lymphatic blockage.[8]

Common in the neck region, the anterior triangle of the neck has been indicated as the most common site, mainly clavicle, trapezius muscle and sternocleidomastoid muscle.[9] The submandibular and parotid regions are the more commonly associated areas to lymphangioma development.[10] Oral cavity rarely represents lymphangiomas mostly restricted to its anterior third however soft palate and mandibular ridge and buccal mucosa are also involved.[5] In our case, the lesion involved the tongue.

Lymphangiomas are presents since birth and therefore rarely diagnosed in adults. In our case, the patient was 16 years of age and the lesion is present since three years only. The incidence of lymphangiomas has been reported to range from 1.2 to 2.8 per 1000 newborns.[11] The most prominent sign or symptom of all lymphangiomas is the presence of a mass. In adult patients, neoplasm can switch to squamous cell carcinoma.[12] The surface is granular due to clear vesicles and color is red or blue due to rupture of underlying blood vessels. The deeper lesion may cause upper respiratory tract disorder or incidental trauma at the site and difficulty in mastication, speech and deformity of the jaws.[13] In this case, the clinical feature was consistent with those of classically described oral lymphangiomas.

Histologically, these lesions are composed of dilated lymphatic channels with one or two endothelial layers, with or without an adventitial layer. These dilated lymphatics can vary in size, depending upon the location and surrounding tissues and is the basis for classification according to Yaita et al.[14] Depending upon cystic space size, they are classified as: macrocystic, microcystic and mixed.[15]

Ultra-sonography, CT and MRI scans can be used to define the relationship of the lesion with the neighboring structures and to help plan surgery.[15] The clinical course of the pathology varies from a spontaneously regressing cyst to an aggressively invasive lesion. Spontaneous or traumatic hemorrhage of the cysts is the common complication.[16]

While treatment of lymphangiomas includes surgical excision, cryotherapy, electro cauterization, sclerotherapy, steroids administration, embolization, and laser therapy.[14] Surgical excision is the best alternative for lesions presenting localized growth.[17] Because the lesion was not a mass rather a superficial lesion we opted for laser therapy. After one year of following-up, there was no relapse of the lesion.

CONCLUSION

Oral lymphangiomas are uncommon lesions occurring at the dorsal region of the tongue. Superficial and localized lesions can be treated by conservative approaches like cryotherapy, laser therapy and surgical excision with low relapse rates. Therefore, knowledge for correct diagnosis is of fundamental importance and for proper therapeutic implications.

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