Case Report

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A RARE CASE OF ORAL LYMPHANGIOMA WITH UNUSUAL PRESENTATION

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ABSTRACT:

Lymphangioma is a change of lymphatic vessels that frequently affects the head and neck region. Its occurrence at oral cavity is rare and it is most commonly identified at the anterior two-thirds of the tongue. At this location, it is clinically characterized as transparent and generally grouped vesicles, which can be red or purple. It can be classified according to the size of vessels into three types: capillary, cavernous, and cystic lymphangioma. To describe a case of oral lymphangioma diagnosed in a 16-year-old female patient with a painless swelling on the dorsal surface of anterior tongue. The lesion is there since 3 years. The lesion has increased in size over four months. An intraoral clinical examination revealed the presence of a reddish pale coloured lesion on the tongue. 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.

Key words: Head and neck, Lymphangioma, Children, Diagnosis, Treatment

INTRODUCTION:

Lymphangiomas are congenital benign tumorus 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 commom predilection in the anterior two-

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third of the tongue resulting in macroglossia. Cases in palate, gingiva, lips, and mandibular alveolar ridge have also been described [5].

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

The following case report is of a patient with a lymphangioma in the tongue, which was successfully treated by laser therapy.

CASE DETAIL:

A 16-year-old female patient presented to the Department of Oral and Maxillofacial Pathology of Sir Ahmed Dental College,Kolkatta with a painless swelling on the dorsal surface of anterior tongue. The lesion is there since 3 years. The lesion has increased in size over four months. An intraoral clinical examination revealed the presence of a reddish pale coloured lesion on the tongue **(Figure 1).**When the lesion was palpated it was pebbly and soft inconsistency 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. Histopathological 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, **(figures2).** Therefore, the diagnosis was confirmed as lymphangioma.

Patient was submitted for laser therapy for lesion .The procedure was facilitated due to the superficial location of the lymphatic vessels. After 1 year of following-up, we did not observe the lesion relapse.

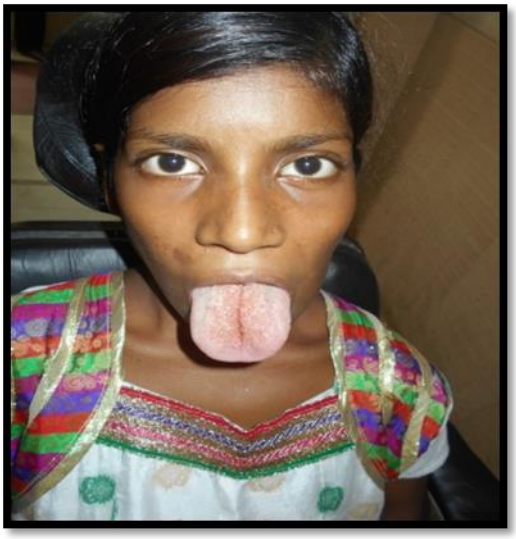

Figure 1: Clinical photograph showing Popular lesions on tongue

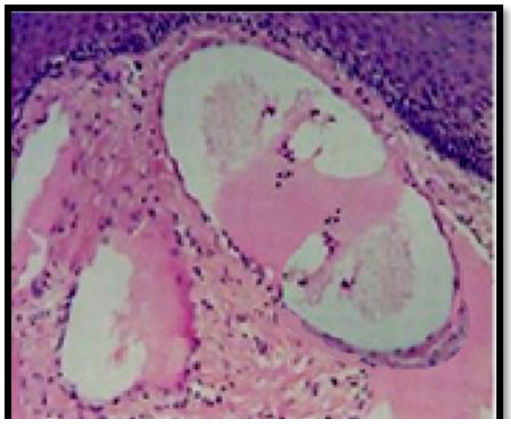

Figure 2:Photomicrograph showing thin walled lymphatic channels filled with lymph and few lymphocytes

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 lymphangioma. 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 [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].

Lymphangioma are presents since birth and therefore rarely diagnosed in adults. In this case reported here the patient is 16 years of age and the lesion is present since 3 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].There 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 there clinical feature was consistent with the form classically described for 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] and microscopic examination in this case exhibited the same as described. Depending upon cystic space size, they are classified as: macrocystic, microcystic and mixed [15].

US, CT and MRI can be used to define the relationship of the lesion with the neighbouring 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 haemorrhage of the cysts is the common complication [16].

While treatment of lymphangiomas includes surgical excision, cryotherapy, electrocauterization, schlerotherapy, 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. No sign of lesion relapse was identified after one year follow up.

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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