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Oral Lymphangioma: An Original Series of Three Cases with Review

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Abstract: Lymphangiomas are benign hamartomatous tumors of lymphatic origin which results from sequestration of lymphhatic tissues that do not communicate with other lymphatic channels.They may occur with hemangiomas and have high predilections for head and neck region (50-70%). They may occur at birth (60-70%) or till two years of age (90%) and rarely in adults. The most common site of occurrence in the mouth is dorsum of tongue being followed by lips, buccalmucosa, soft palate and floor of mouth. Tongue lymphangioma may present as localized or diffused growth that may result in macroglossia, impaired speech and difficulty in mastication. There are different treatment options; surgical excision being the most effective mode of treatment. Here we have reported three different cases of oral lymphangiomas, two of which occurring in the tongue and one in the buccal mucosa.

Keywords: Hamartomatous, lymphangioma, lymph vessels, tapioca pudding.

Case Series

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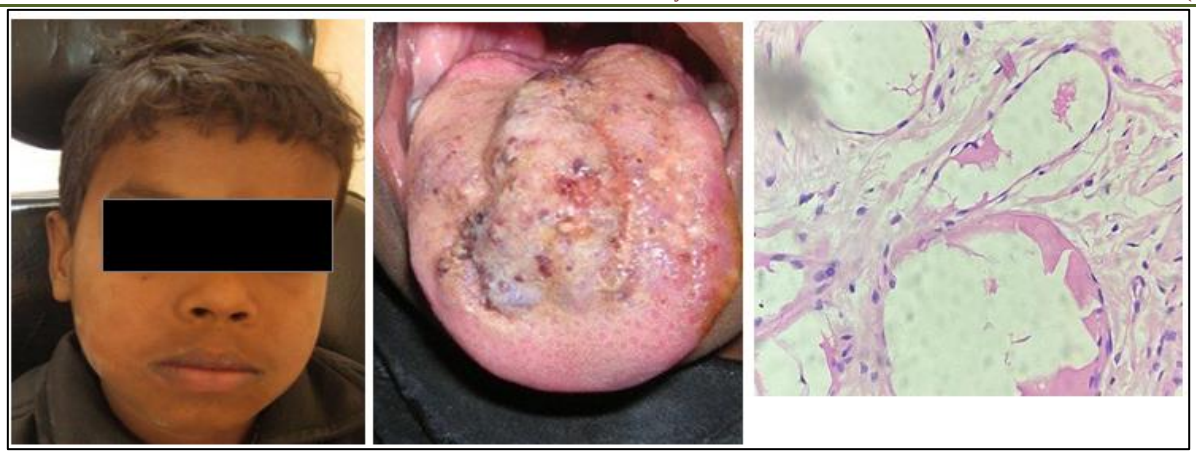
INTRODUCTION

Lymphangiomas are congenital low flow vascular hamartomas - about 75% of which has predilection for head and neck tissue. Their incidence ranges from 1.2-2.8 per 1000 newborns and around 80-90% cases are found before 2 years of age. They arise from sequestration of malformed lymphatics which fails to connect to body's lymphatic or venous system and hence accumulate lymph and becomes dilated and enlarged [1]. The projection of the endothelial sprouts from the lesional wall further destroys surrounding tissue and invades tissue planes.Other theories revolve around either arrest of normal growth of primitive lymph channels or lymphatic tissue development at a wrong site. They can be classified as capillary lymphangioma or lymphangioma simplex, cavernous lymphangioma and cystic hygroma. In oral cavity the mostcommon site is tongue where the lymphangioma is developed in dense tissue and presents mostlyas cavernous type [2]. The present article reports 3 cases of oral lymphangiomas with review of literature.

CASE REPORTS

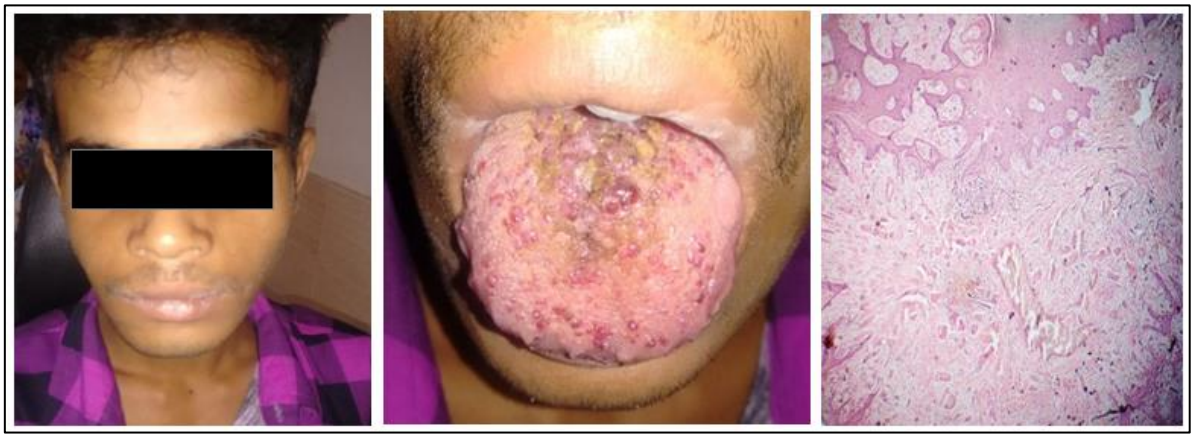
Case No 1

An eight-year-old male patient reported to the department of oral pathology with the complaint of tongue swelling since last 3-4months. On intraoral examination a large dome shaped swelling was present on the tongue dorsum. The surface of the tongue was somewhat pebbled. The swelling was non tender and firm on palpation. Past medical history, family history of the patientwere non-significant. Incisional biopsy was done from the representative site. Histopathology report reveals the presence of lymphatic channels lined by endothelium in the loose connective tissue stroma. The vessels contained lymph which is eosinophilic.



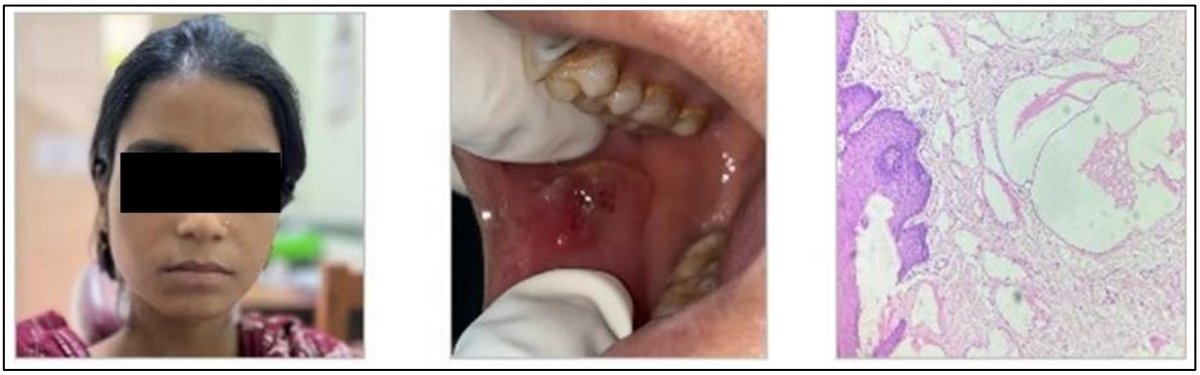
Case No 2

A male patient aged 20 yrs came to our department with complaint of bleeding from the tongue which he noticed few months back. On intraoral examination a firm, non-tendor lesion with papular surface was found on the mid part of the tongue dorsum. Some papules were blood filled and bluish red in appearance. Incisional biopsy was done. Histopathology report showed proliferation of lymphatic vessels lined by endothelium containing eosinophilic coagulum or lymph.



Case No 3

A 23 years old female patient came to the department with complaint of a swelling inside the mouth. On examination a compressible but non reducible swelling with toad skin appearancewas found on the right buccal mucosa. Other relevant history of the patient was non significant. Incisional biopsy was performed. Histopathology report revealed the presence of connective tissue covered with stratified squamous epithelium showing large dilated vessels filled with eosinophilic coagulum and lined by endothelial cells both in subepithelial and deep connectivetissue. The overall features were suggestive of lymphangioma.



DISCUSSION

Lymphangioma was first described by Redenbacher in 1828 whereas Virchow first described lymphangiomas of the tongue in 1854.Lymphangioma is a hamartomatous lesion which accountsfor 4% of all the vascular tumors and 25% of all benign vascular tumors in children [3]. It occurs mostly in head and neck region but axilla, inguinal or genital region can also be involved. However, intraoral lymphangiomas are rare [4]. In intraoral sites anterior two-thirds of tongue is most common followed by buccal mucosa [5]. In our case series two cases were found on tongue dorsum and one on buccal mucosa.

Lymphangiomas are rare in adults. The onset of lymphangiomas are either at birth (60-70% cases) or upto 2 years of age (90%) [3]. However, lymphangioma can arise in adulthood due to trauma, inflammation, lymphatic obstruction etc [6]. In this case series we have two adult patients and one child patient.

Clinically oral lymphangioma appears as a slow growing painless mass located either superficially or within deep tissue. Superficial lesion demonstrates a cluster of translucent vesicles giving rise to a pebbly surface often referred as frog eggs or tapioca pudding appearance. Sometimes secondary haemorrhage can be found giving the lesion a reddish or purplish tinge. On the other hand deep lesions are described as soft ill-defined elevated mass covered with normal looking mucosa [4]. Lymphangiomas on tongue can cause macroglossia which in turn may lead to speech disturbances, poor oral hygiene, mandibular prognathism, open bite, chewing difficulties, and maxillofacial deformities etc [6].

Histologically there are three types of lymphangiomas: simplex (capillary), cavernous and cystic. Generaly, lymphangiomas when present in fairly dense tissue such as tongue or floor of the mouth, they present as cavernous type whereas when they are found in loose fascia like tissuewhere expansion is permissible such as neck, they become cystic. In our case series two of them them were cavernous type and the rest was capillary type histologically. Slides stained with Heamatoxylin and Eosin stains show the presence of distended lymph vessels lined by flat endothelial cells containing eosinophilic lymph along with lymphocytes and occasional erythrocytes and secondary haemorrhage, if any [4].

Diagnosis is achieved mainly by clinical presentation and histopathological examination along with imaging like USG if needed to detect the cystic nature of the tumor. CT/MRI detects the extension of the tumor and help in planning surgery whereas angiography may be needed to rule out hemangioma [5]. Also, immunohistochemical markers like podoplanin, vascular endothelial growth factor receptor 3, lymphatic vessel endothelial HA receptor1, D240 and Prox 1 can be used to differentiate lymphatics and blood vessels [1] in this case series all the diagnosis were based upon clinical and histological feature since they directed clearly towards lymphamgioma and there was no confusion.

Treatment modalities depend upon size, location, extension into the deeper tissue and proximity to vital structures. Although numerous methods can be applied surgical excision remains the best of them. However in case of head and neck region lesions, to avoid anatomical complexity nonsurgical therapies are usually tried before surgery. Sclerotherapy using Sodium Morrhuate, hypertonic saline, Dextrose, Tetracycline, Doxycyline, Acetic acid, Ethanol, boiling water, and OK-432 etc are often used. Other than these cryotherapy, diathermy, radiation therapy have also been tried [1, 4]. All three cases of ours were sent to the department of oral and maxillofacial surgery for sclerotherapy followed by surgery, if needed.

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