Case Report

# Oral lymphangioma

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**Abstract**

Lymphangioma is a benign hamartomatous neoplasm of the lymphatic vessels. Head-and-neck lymphangiomas are rare but when it affects, tongue is the most common site. These tumors usually manifest within the 1st year of life. When it affects the tongue, there may be either a localized or diffused growth that can enlarge to cause macroglossia. The complications associated with lymphangiomas include occlusal disturbances, disturbances in speech, and poor oral hygiene. Here, we present a case of lymphangioma of the tongue in a 26-year-old male patient who was diagnosed following a routine dental examination.

**Keywords:** Benign, hamartoma, lymphangioma, tongue

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## INTRODUCTION

Lymphangioma is a benign, congenital malformation with unknown etiology, arising from the lymphatic channels. Virchow first described it in 1854. They have a high prepondence to the head-and-neck region, but the oral cavity is a rare site.[1,2] It comprises about 6% of the benign soft-tissue tumors of the head and neck in individuals <20 years of age. It is a congenital anomaly causing a blockage of the lymphatic drainage due to the failure of anastomosis of the clusters of small lymphatic vessels with the main channel.[3] It has also been proposed that lymphangioma can be considered as a true neoplasm of the transformed lymphatic cells rather than being a congenital malformation.[4] Here, we report the case of lymphangioma of the tongue in a 26-year-old patient.

## CASE REPORT

A 26-year-old male patient reported to the department with a complaint of reddish spots on the tongue for 20 years. The patient was aware of the lesion since childhood with associated burning sensation and was left untreated. The lesion was initially present only in a small area which further involved the whole part of the tongue as age advanced. On intraoral examination, erythematous areas with bluish discoloration were observed on the lateral borders, ventral, and dorsal surfaces of the tongue with papillary hyperplasia. The lesion was soft on palpation and nontender [Figures 1 and 2a, b]. A provisional diagnosis of lymphangioma of the tongue was considered with a differential diagnosis of capillary hemangioma.

Excisional biopsy was taken from the lateral borders of the tongue and was sent for histopathological examination which showed the presence of numerous intervening dilated lymph vessels in loose fibrovascular stroma below the epithelium. Juxtaepithelially enlarged tortuous vessels are lined by the thin single layer of endothelial cells, with presence of red blood cells and

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lymphocytes in the surrounding connective tissue. The deeper muscle layer also showed the presence of dilated lymphatic vessels, which lead to a confirmed diagnosis of capillary lymhangioma [Figure 3]. The patient was informed about the lesion but denied further treatment due to the absence of any symptoms. Instructions for the maintenance of oral hygiene and tongue cleaning habits were given to the patient. The patient is kept on long-term follow-up.

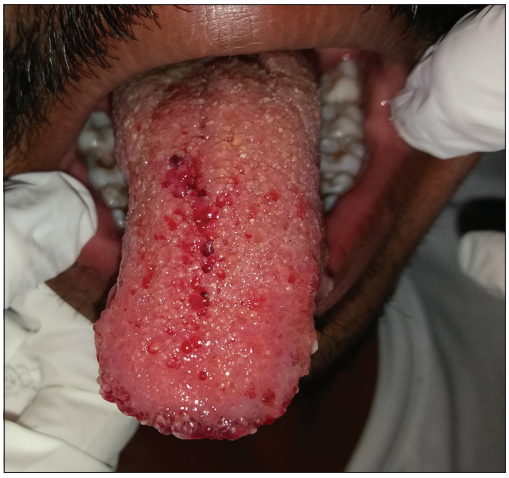

**Figure 1:** Clinical photograph of the patient showing pebbled appearance on the dorsal aspect of the tongue

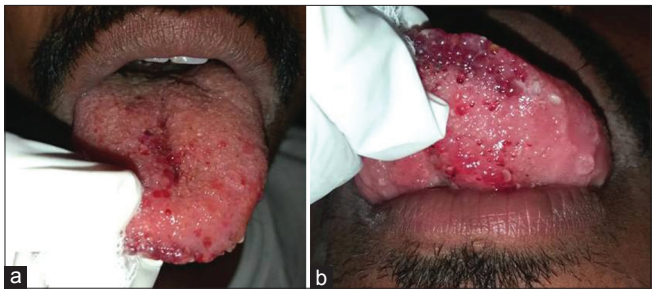


**Figure 2:** (a and b) Clinical photograph of the patient showing multiple red and blue vesicles on the tip, dorsum, and ventral surface of the tongue

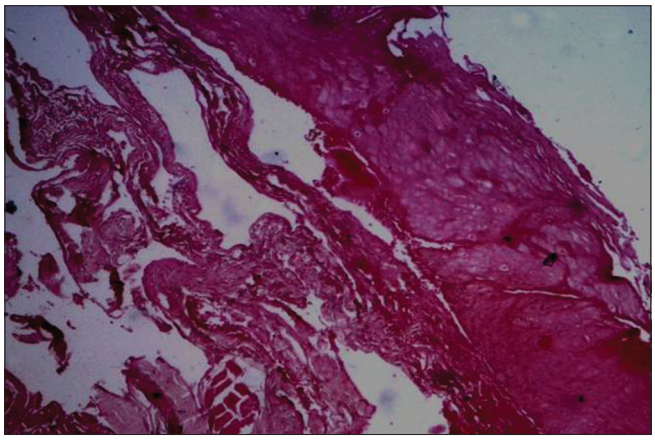

**Figure 3:** Histopathological photomicrograph showing hematoxylin and eosin-stained showed numerous interwining dilated lymph vessels in loose fibrovascular stroma below the epithelium

## DISCUSSION

Lymphangiomas are congenital anomalies of the lymphatic system which are hamartomatous, first described by Redenbacher in 1828.[2] The lesions are manifested either at the time of birth or within 2 years of life and have female prepondence. The common sites are head and neck, proximal extremities, buttocks, and trunk. It rarely affects the oral cavity, but if present, the tongue is the most common site followed by palate, buccal mucosa, gingiva, and lip. Tongue lymphangioma presents with multiple blisters-like nodules resembling translucent vesicles.[4] The similar clinical appearance was seen in the present case also. Two theories were suggested with regard to the development of lymphangiomas. The first theory states that the endothelial outpouching from the jugular sac from five primitive sacs of the venous system, when spreads centrifugally forms the lymphatic system of the head and neck. The second theory states that mesenchymal clefts in the venous plexus that spreads centripetally form the lymphatic system.[4,5] Congenital obstruction of the primitive lymphatic enlargement gives rise to lymphangioma. When affecting the oral cavity, it is mostly seen congenitally or within the first decade of life, involving the dorsal and lateral borders of the tongue in 60% of the cases.[6] When it involves the anterior two-thirds of the tongue, it can lead to macroglossia, resulting in poor oral hygiene, speech disturbances, and bleeding. In the present case, anterior two-thirds of the tongue was involved causing disturbances in speech. The patient had fair oral hygiene but was associated with burning sensation of the tongue. The clinical presentation depends on whether the lesion is superficial or deep. If the lesion has pebbly surface due to the translucency of the vesicles with a mild reddish hue giving a frog-egg appearance, then it is considered as superficially placed.[7] In deep-seated lesions, there are diffuse nodules that are soft in consistency with mild variation in color. These types of lesions can cause complications such as upper airway tract obstructions, poor oral hygiene, and pain.[5] According to the above-said features, the case presented is a superficially seated lymphangioma. A classification for lymphangiomas affecting the head and neck was proposed by De Serres LM based on the anatomical involvement as:[6,8]

- Stage/Class I– Infrahyoid unilateral lesions
- Stage/Class II– Suprahyoid bilateral lesions
- Stage/Class III– Suprahyoid or infrahyoid unilateral lesions
- Stage/Class IV– Suprahyoid bilateral lesions

- Stage/Class V– Suprahyoid or infrahyoid bilateral lesions
- Stage/Class IV– Infrahyoid bilateral lesions

Histopathologically, they can be classified as capillary, cavernous, and cystic lymphangioma.[8,9] The present case was classified as capillary lymphangioma in accordance with the histopathological features. The treatment modalities mainly include surgical excision, cryosurgery, electocautery, sclerotherapy, embolization, laser surgery, and radiofrequency tissue ablation.[8,10] However, in the present case, as the patient remained asymptomatic throughout, he was not willing for any further treatment and is kept on a regular follow-up. The mode of treatment is mainly aimed at minimizing the complications such as macroglossia, difficulty in swallowing, speech and mastication, mandibular prognathism, and airway obstruction.[9]

## CONCLUSION

Lymphangioma is an unusual entity in the orofacial region but their early diagnosis and intervention prevents the occurrence of further complications including orthognathic deformities and also helps patients to maintain good oral hygiene.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

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