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# Lymphangioma of the tongue- a rare case report and review of the literature

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## ABSTRACT

Lymphangiomas are considered as hamartomas of the lymphatic system. These malformations are rather infrequent and generally detected either at infancy or at early childhood, rarely in adults. They are commonly seen in head and neck region and rarely in the oral cavity. Here, we report a case of lymphangioma of tongue in a 70 year old male and review its clinical, histopathological features and treatment modalities.

**Key words:** Congenital malformation, Hamartoma, Lymphangioma, Macroglossia, Tongue

## Introduction

Lymphangiomas are rare malformations that are characterized by anomalous proliferation of lymphatic vessels. The commonest sites of these lesions has been the head and neck region (50-70%).[1] Redenbacher (1828) was first to describe a case of lymphangioma and later Virchow (1854) reported lymphangioma of the tongue.[2, 3] Reports have shown that many cases are seen at birth and 90% of them are seen before 2 years of age.[4] Intraoral lymphangiomas are rare.

The commonest intraoral sites has been reported to be dorsum of the tongue, other sites being lips, buccal mucosa, floor of the mouth and soft palate.[5] Lymphangiomas arise as slow growing, painless soft tissue mass. The lesions may be superficial or deep. Superficial lesions show as elevated nodules, pink or yellow color or appear as transparent grouped vesicles, which later due to secondary hemorrhage become reddish or purple.[6] Whereas the deeper lesions show as soft, diffuse masses with normal color.[5, 6]

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Management of lymphangiomas include various procedures like radiation therapy, electrocautery, laser surgery, sclerotherapy, steroid administration etc.[3, 8, 9] Whereas the treatment of choice is surgical excision.[10]

The prognosis in most of the cases is good, while large lesions of neck/tongue might result in airway obstruction and death.[7] Here we report a case of lymphangioma affecting tongue in a 70 year male.

## Case Report

A 70 year old male patient reported to department of oral medicine and radiology with the chief complaint of growth on the tongue.

Patient also complained of slurred speech and bleeding from the growth and difficulty in swallowing and chewing.

Examination revealed that there were multiple papular lesions on the anterior two-thirds of the dorsum and ventral surface of tongue. Inspection showed 1x1 cm size blood filled papules extending from tip of the tongue to anterior two third of the dorsum of the tongue (**Fig 1**).

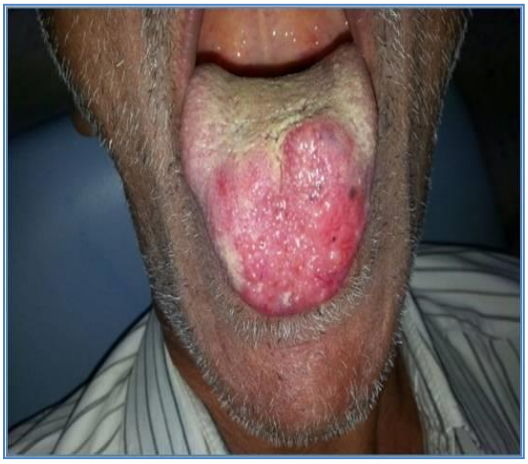

**Fig 1: Intra-oral view showing lesion over dorsal surface of tongue**

These papules were also seen on the ventral side of the tongue extending on either side of the lingual frenum. The lesion was seen having a pebbly surface that resembled as cluster of translucent vesicles. Palpation revealed the growth being soft and non tender. All the inspectory findings were confirmed (**Fig 2**).

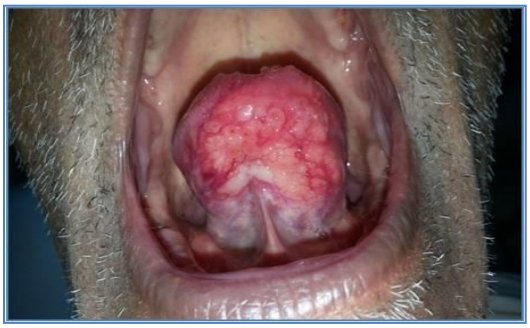

**Fig 2: Intra-oral view showing lesion over ventral surface of tongue**

Based on the basis of history and clinical findings, lymphangioma was made as provisional diagnosis and an excisional biopsy under general anaesthesia was planned. Histopathologic examination revealed many lymphatic vessels with marked dilatations. The spaces contained proteinaceous fluid The lining endothelium was thin. The lymphatic spaces contained lymphatic fluid, lymphocytes and red blood cells (**Fig 3**). The findings confirmed the lesion to be lymphangioma of tongue.

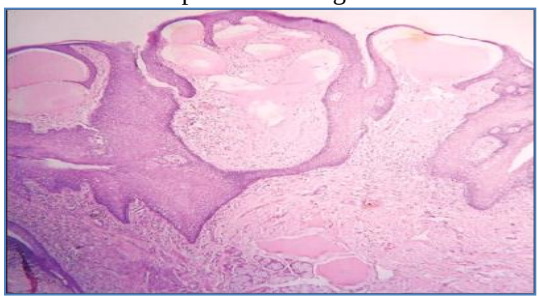

**Fig 3: Photomicrograph**

**Discussion**

Lymphangiomas are rare congenital vascular hamartomas affecting the lymphatic vessels. They constitute about 4% of all the vascular tumors and 25% of all benign vascular tumors in children. Intra-oral

lymphangiomas are rare and most commonest site is anterior two thirds of tongue, where they frequently cause macroglossia. Here these lesions may cause difficulty in eating and speech and sometimes may result in airway obstruction, leading to death. Generally these lesions are superficial and appear as a cluster of translucent vesicles, as was seen in our case.[3-5]Most of the cases are seen at birth. But few cases have been reported in adults. Our case is rare as the patient was elderly male of 70 years age. 2 theories have been proposed regarding the pathogenesis of lymphangioma. The first one explains that the lymphatic system develop from 5 primitive sacs of venous system and endothelial out pouchings spread from jugular sac centrifugally to form the lymphatic system. the second theory suggests that the lymphatic system develops from mesenchymal clefts in the venous plexus reticulum and spread centripetally toward the jugular sac.[8-10]

De Serres et al classified lymphangioma of head and neck based on the anatomical involvement.[10, 11]

1. Stage/Class I: Infrahyoid unilateral lesions
2. Stage/Class II: Suprahyoid unilateral lesions
3. Stage/Class III: Suprahyoid and infrahyoid unilateral lesions
4. Stage/Class IV: Suprahyoid bilateral lesions
5. Stage/Class V: Suprahyoid and infrahyoid bilateral lesions

Our case was class IV type.

The choice of treatment of lymphangioma is surgical excision. Other modalities are cryotherapy, laser surgery, radiation therapy etc. We carried out surgical excision under general anaesthesia. [12, 13] Due to infiltrative nature, a higher rate of recurrence was noted. Orvidas and Kasperbauer noted a recurrence rate of 39%.[14]

**Conclusion**

We report a case of lymphangioma of tongue in a 70 year old male. Even though intraoral lymphangioma is rare, early diagnosis and treatment can prevent complications of these lesions.

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