

## **Lymphangioma of the Tongue - Case Report**

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**Citation of this Article:** Pratap Movaniya, Hirakben Patel, Nimisha Desai, Tushar Makwana, Kalpesh Makwana, “Lymphangioma of the Tongue - Case Report”, IJDSIR- May - 2020, Vol. – 3, Issue -3, P. No. 127 – 132.

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**Type of Publication:** Case Report

**Conflicts of Interest:** Nil

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### **Abstract**

Lymphangiomas are benign tumours resulting from a congenital malformation of the lymphatic system. They are relatively uncommon and usually diagnosed in infancy and early childhood. Commonly located in head and neck, they are rarely seen in the oral cavity. Intraoral lymphangiomas are seen more frequently on the dorsum of tongue, followed by palate, buccal mucosa, gingiva, and lips. These lesions usually do not required surgical intervention.

**Keywords:** tongue, lymphangioma of anterior tongue, congenital malformation

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### **Introduction**

Lymphangioma is a benign proliferation of lymphatic vessels and represents hamartomas of malformed lymphatics. They have poor communication with the normal lymph systems and therefore collect lymph.<sup>[1-3]</sup> The most common location in the mouth is the dorsum of tongue, followed by lips, buccal mucosa, soft palate, and floor of the mouth.<sup>[3]</sup> The clinical appearance of lymphangioma depends on the extension of the lesion. Superficial lesions consist of elevated nodules with pink or yellowish colour. Deeper lesions are described as soft, diffuse masses with normal colour.<sup>[3]</sup> These tumours typically present in children younger than 2 years and in a

significant number of cases are present at birth<sup>[1-7]</sup> uncommon in adults<sup>[8]</sup>.

In children, intraoral lymphangioma, especially tongue lymphangioma leading to macroglossia, may cause speech disturbances, poor oral hygiene, mandibular prognathism, open bite, yawning, chewing difficulties, and maxillofacial deformities<sup>[8,9]</sup> It is the common opinion that severe life-threatening functional impairment necessitates early treatment. When there is no significant functional deficit, treatment can be delayed well past infancy and may consist of surgery, sclerotherapy, or observation. Treatment timing relative to the age of the patient is somewhat debatable. Lymphangiomas of small dimensions, without functional impairment or cosmetic disfigurement, do not necessarily require treatment. The possibility of spontaneous regression in low-stage macrocystic Lymphangioma suggests that observational monitoring may also be appropriate.<sup>[1,10]</sup>

This article describes a case of lymphangioma of the tongue and review of literature for its management.

### Case Report

A 34 year male patient reported to our department with chief complain of pain in lower right third molar region due to unerupted third molar since 2 months. Past medical history was negative. Patient had noticed growth on left side of tongue before 27 year which was not related to the present history of pain. Growth did not increase in size according to patient. Before the examination informed consent was taken from patient. On examination patient was moderately built and nourished. On extra oral examination Mouth opening was within normal limit.



Figure 1: Clinical Pictures of The Lesion

On intraoral inspection, dorsal aspect of the tongue appeared enlarged with pebble like multiple red, blue, grey and clear vesicles.(Figure -1)Tip of the tongue was also involved. A prominent mass was seen on left dorsal and ventral surface of tongue approx. 4×2cm in size which was irregular and granular in appearance. There was marked deviation of tongue on right side with macroglossia. (Figure - 2)Tongue movements were normal. There was no complain of slurring of speech and dysphagia. On palpation lesion was hard and non-tender. Occlusion was normal.

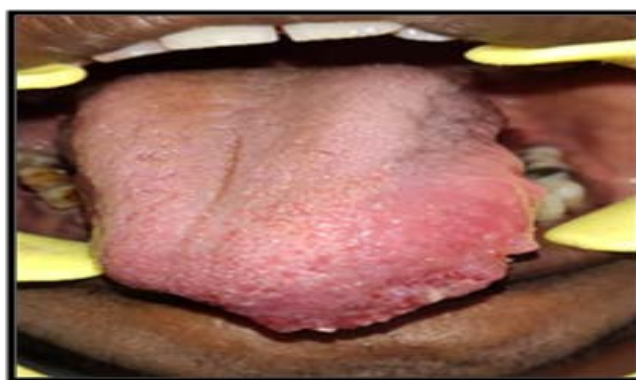


Figure 2: Marked Deviation Of Tongue On Right Side With Macroglossia

Punch biopsy was taken from left side of tongue under local anaesthesia. Histopathological examination revealed that the lesion was lymphangioma of tongue. (Figure 3,4)The patient was advised for surgical excision. However, the patient was not willing for treatment.

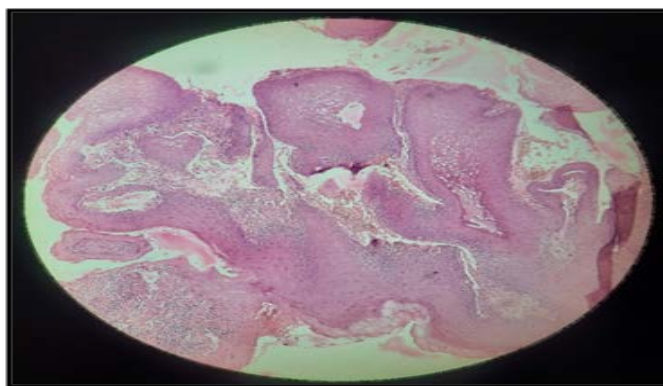


Figure 3: Microscopic appearance of lesion Hematoxylin eosin stain

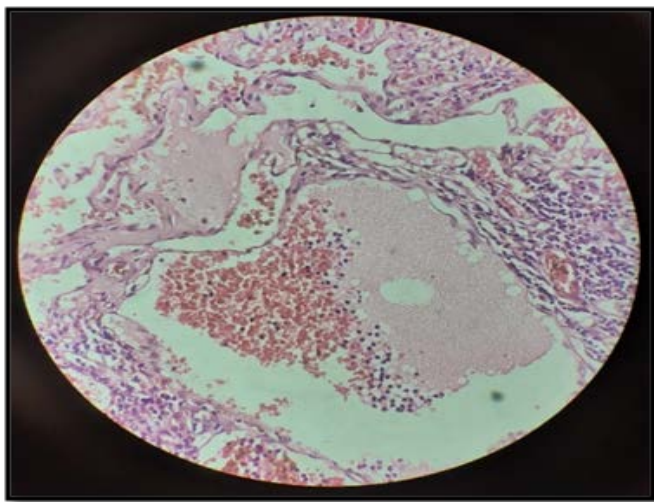


Figure 4: Microscopic appearance of lesion with Magnification

### Discussion

Lymphangioma is a rare, benign, congenital malformation of unknown aetiology that originates from lymph vessels and this entity was first described by Virchow in 1854.<sup>[11,12]</sup> Lymphangiomas are uncommon congenital hamartomas of the lymphatic system.

Lymphangiomas have a **marked predilection** for the head and neck region, which accounts for about 75% of all cases and about 50% of these lesions are noted at birth and around 90% develop by two years of age. However, diagnosis of lymphangioma in adults is a rare occurrence<sup>[13,14]</sup>.

In the oral cavity, this lesion is common in the first decade of life and mostly occurs on the dorsal surface and lateral border of the tongue. It rarely arises on palate, gingiva, buccal mucosa and lips which overlap with the present case.<sup>[15,16]</sup>

The anterior two-thirds on the dorsal surface of the tongue is the most common site for intraoral lymphangiomas leading to macroglossia.<sup>[17]</sup> These patients tend to have speech disturbances, poor oral hygiene, and bleeding from the tongue associated with oral trauma.<sup>[18]</sup>

In this case the anterior two third was involved which was no associated with disturbance in speech.

The clinical appearance of the lesion varies based on its whether it is superficial or deep. Superficial lesion present as papillary lesions with pebbly surface due to the occurrence of several translucent vesicles with same colour as that of adjacent mucosa or occasionally with a mild reddish hue.(figure no 1-3) Interestingly they give a tapioca pudding or frog eggs like appearance. The deeper lesions manifest as diffuse nodules which are soft in consistency and with negligible alterations in colour or texture<sup>[19]</sup>. For lymphangiomas slow progressive enlargement is the rule, but rapid enlargement infrequently occurs secondary to infection or trauma.<sup>[20,21,22]</sup>

The consequences of deep seated lesions may result in swelling of tissues leading to obstruction in upper airway, extrusion of tongue, increased salivation, jaw deformity, pain and poor oral hygiene. Also marked problems with respect to chewing and speaking may result<sup>[23]</sup>.

Marked dilatation of lymphatic vessels is appreciated histopathologically. Small capillary sized vessels, large dilated lymph channels and large macroscopic cystic spaces are respectively appreciated microscopically in capillary, cavernous and cystic lymphangioma<sup>[18]</sup>. Cavernous lymphangioma is the most common intra oral type<sup>[8,24]</sup>. (figure no4-5)

The treatment of lymphangioma depends upon their type, size, involvement of anatomical structures and infiltration to the surrounding tissues. Microcystic lesions do not respect tissue planes, are diffuse and difficult to eradicate, whereas macrocystic lesions are localized and easily excised. The various treatment modalities for lymphangioma are surgical excision<sup>[13,25-27]</sup>, radiation therapy<sup>[23]</sup>, cryotherapy, electrocautery, sclerotherapy, steroid administration, embolization, and ligation, laser

surgery with Nd-YAG<sup>[28]</sup>, CO<sub>2</sub><sup>[29]</sup> and radiofrequency tissue ablation technique.<sup>[23]</sup>

Advancement in intralesional sclerotherapy has shown significant efficacy and reduced the need for other forms of therapy although lymphangiomas do not respond as well as to sclerosing agents as do haemangiomas.<sup>[29]</sup>

Various sclerosing agents such as picinabil, bleomycin, doxycycline, acetic acid, alcohol and hypertonic saline have been used for Lymphangioma. Although the popularity of sclerotherapy is growing, there is no consensus regarding the type to be used.<sup>[30-32]</sup>

Reena et al. used intralesional bleomycin in their patient and concluded sclerotherapy in intra oral lymphangioma can lead to various complications which can be life threatening. Complications can also occur postoperatively or following sclerotherapy. Bleomycin injection (intralesional) is a safe and effective sclerosant for lymphangioma circumscriptum.<sup>[33]</sup>

### Conclusion

Lymphangioma is primarily a childhood disease. It has to be identified and diagnosed early for uneventful treatment, reducing functional, psychological disturbances and cosmetic disfigurement. Oral lesions may be encountered in adults, where they have to be completely excised with clear borders to prevent recurrences. In this case patient did not have any complaints with speech, deglutition, tongue movement and aesthetics. Regular follow up was advised.

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