



LYMPHANGIOMA OF THE TONGUE – A CASE REPORT

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ABSTRACT Lymphangiomas of the tongue usually manifest within the first few years of life, but may present much later (Litzow, 1961). They are relatively uncommon and usually diagnosed in infancy and early childhood. Lymphangiomas are benign tumours resulting from a congenital malformation of the lymphatic system. Accidental or surgical trauma to a lymphangiomatous tongue may result in rapid enlargement due to bleeding and oedema. Commonly located at head and neck, they rarely occur in the oral cavity. Intraorally it is on the dorsum of tongue, buccal mucosa, lips, gingiva, and palate. We present the case of a 11-year-old male with lymphangioma on the ventral surface of tongue.

KEYWORDS : Macroglossia, lymphangioma, tongue and Congenital malformation

INTRODUCTION

Lymphangiomas are rare, benign tumours resulting from a congenital malformation of the lymphatic system and this entity was first described by Virchow in 1854¹. They have poor communication with the normal lymph systems and therefore collect lymph. Intraoral lymphangiomas occur more frequently on the dorsum of tongue, followed by palate, buccal mucosa, gingiva, and lips^{2,3}. Superficial lesions consist of elevated nodules with pink or yellowish colour and deeper lesions are described as soft, diffuse masses with normal colour. Lymphangioma of the tongue is a common cause of macroglossia in children associated with difficulty in swallowing and mastication, speech disturbances, airway obstruction, mandibular prognathism, openbite and other possible deformities of maxillofacial structures⁴. They have been divided into four categories based on histologic appearance as:

- Lymphangioma simplex (capillary lymphangioma, lymphangioma circumscriptum),
- composed of small, thin-walled, endothelium lined, capillary-sized lymphatic vessels.
- Cavernous lymphangioma, comprised of dilated lymphatic vessels with surrounding adventitia.
- Cystic lymphangioma (cystic hygroma), consisting of huge, macroscopic lymphatic spaces with surrounding fibrovascular tissues and smooth muscle.
- Benign lymphangi endothelioma (acquired progressive lymphangioma), lymphatic channels appear to be dissecting through dense collagenic bundles⁵.

Lymphangiomas are also categorized as macrocystic, microcystic, or mixed based on response to sclerotherapy⁶. 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 appropriate⁸.

Case Report: A 11 years old male patient had reported to the Department of Pedodontics & Preventive Dentistry, KDC, Meerut, with the chief complaint of swelling in the tongue for the past 2 months. Initially it was a small swelling which then gradually increased to the present size. On examination patient was moderately built and well nourished. On extra oral examination Mouth opening was within normal limit. The remainder of the tongue was of normal appearance.

Investigations: Routine hematological examinations including bleeding and clotting time were found to be normal. The differential diagnosis consisted of Mucocele, Lipoma.. fibroma Salivary Gland Neoplasm, oral hemangioma and oral lymphangioma. Based on history and clinical examination a provisional diagnosis of lymphangioma was made.

Treatment: This case of lymphangioma was treated by surgical

excision method using scalpel blade as shown in figures (1-3) below. The patient was explained about the procedure and informed consent was obtained. Local anesthesia was applied around the lesion. Excisional biopsy was done, tissue of size 10 x 6 mm excised. Post-operative instructions were given and analgesics were prescribed. Histopathological examination showed the presence of many cavernous lymphatic spaces within the lamina propria and the underlying muscle and there was no residual deformity or loss of function. Follow-up showed no signs of recurrence.

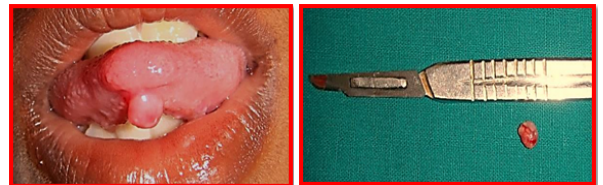


Fig 1 Pre-operative Photograph Fig 2 Excised Lesion



Fig3, Post-operative Photograph

DISCUSSION:

Lymphangiomas are uncommon congenital malformations of lymphatic vessels filled with a clear protein-rich fluid containing few lymph cells. They result from abnormal development of lymphatic system with obstruction to lymph drainage from the affected area. The affected areas in the oral cavity include the tongue, lips, gingiva, palate, and alveolar ridge of the mandible⁹. 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 tongue¹⁰. 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^{11,12}. Intraorally the most common site is anterior two-thirds on the dorsal surface of the tongue leading to macroglossia¹³. 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. 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 texture¹⁴.

A classification of the lymphangioma of head and neck on the base of the **Anatomical** involvement had been proposed by De Serres Lm¹³.

- **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

According to their **clinical presentation** they are classified into;

- **Macrocytic:** cavities larger than about 2cm³
- **Microcytic:** cavities smaller than about 2 cm³
- **Mixed:** combining these two types

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 macrocytic lesions are localized and easily excised. The various treatment modalities for lymphangioma are surgical excision^{11,15,16}, radiation therapy¹⁷, cryotherapy, electrocautery, sclerotherapy, steroid administration, embolization, and ligation, laser surgery with Nd-YAG¹⁸, CO₂ and radiofrequency tissue ablation technique. 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 haemangiomas¹⁹. Various sclerosing agents such as picinabil, bleomycin, doxycycline, acetic acid, alcohol and hypertonic saline have been used for Lymphangioma. Reena et al use intralesional bleomycin 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 (intra-lesional) is a safe and effective sclerosant for lymphangioma circumscriptum²⁰.

CONCLUSION:

Oral lesions may be rarely encountered in oral cavity, their early recognition allows proper initiation of treatment and prevents the occurrence of the complications. These lesions have to be completely excised with clear borders. Regular follow up prevents its recurrence.

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