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CASE REPORT **OPEN ACCESS**

LYMPHANGIOMA OF TONGUE IN A PAEDIATRIC PATIENT– A RARE CASE REPORT

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AB S T RACT

Lymphangiomas are uncommon, non-cancerous congenital malformations involving the lymphatic vessels. They are most frequently observed in the head and neck area. Intraoral involvement is relatively rare, with the dorsal surface of the tongue being the most frequently affected site, followed by the lips, buccal mucosa, soft palate, and floor of the mouth. When present in the oral cavity, these lesions can lead to complications such as enlarged tongue (macroglossia), difficulty in speaking, and challenges with feeding. Surgical removal is typically the preferred method of treatment.

Keywords: congenital malformations, macroglossia, lymphangioma..

BACKGROUND

Lymphangiomas are uncommon, non-malignant anomalies that arise due to developmental disturbances in the lymphatic system during embryogenesis. These lesions typically present in infancy or early childhood and are rarely seen in adults. Although lymphangiomas can occur in different regions of the body, approximately 75% are located in the head and neck

region. Within the oral cavity, the anterior dorsum of the tongue is the most frequently affected site [1,2].
Other intraoral locations include the lips, buccal mucosa, soft palate, and floor of the mouth. Lymphangiomas in these areas may lead to a variety of functional and aesthetic problems, including macroglossia, articulation difficulties, problems with mastication and deglutition, and in more severe cases, airway obstruction [3,4].

Histopathologically, lymphangiomas are composed of dilated lymphatic vessels lined by endothelial cells within a loose connective tissue stroma [5]. These lesions are not encapsulated and often infiltrate surrounding tissues, which makes complete surgical excision challenging and contributes to a risk of recurrence [6].

Surgical removal is considered the treatment of choice, particularly in cases where functional compromise is present. Early diagnosis and intervention are essential to minimize complications and optimize treatment outcomes . The current report details the case of a 7-year-old malediagnosed with lymphangioma of the tongue, outlining the clinical presentation, diagnostic approach, histological findings, and surgical management. On Histopathology it was confirmed as Lymphangioma.

CASE REPORT

A 7 year old male child presented with a painless swelling on ventral surface of midline of tongue that had been progressively increasing in size over last 2 years. . The patient had difficulty in swallowing food . there was no associated symptoms such as pain, bleeding, pus discharge , change in voice .

On examination, the lesion was approx. 2 x 2cm ,reddish mass, solitary, flat raised swelling ,firm in consistency, non-tender, adherent on surface ,non-mobile in the junction of ant. 2/3rdand post. 1/3rd of ventral surface of the tongue .The child exhibited mild macroglossia with interference in articulation and mastication .There was no evidence of systemic involvement or lymphadenopathy. The patients vital signs were stable, and there was no signs of infection or other oral pathologies .

The differential diagnosis include conditions like mucocele , hemangioma, and dermoid cyst. To further evaluate, patient underwent a series of diagnostic tests .

The management of lymphangioma is influenced by factors such as the size of the lesion, its anatomical location, and the severity of associated symptoms. In the present case, due to the lesion's progressive enlargement and its position on the tongue, surgical removal was performed after preanaesthetic approval **(IMAGE1,2,3).**

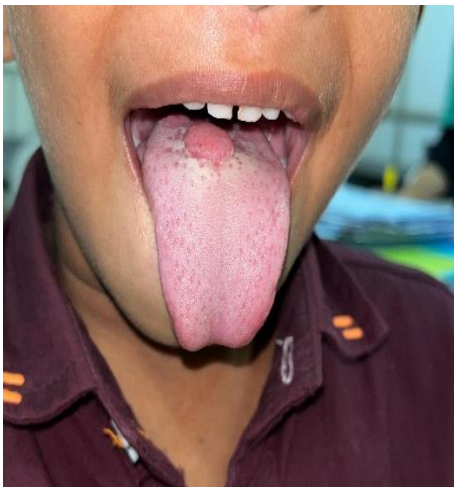

IMAGE 1 – PREOPERATIVE PICTURE

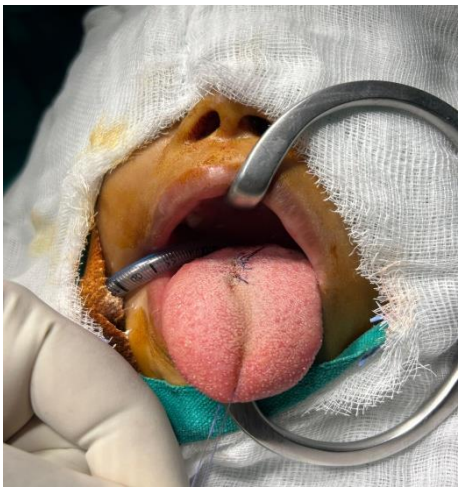

IMAGE 2 – POST OPERATIVE PICTURE

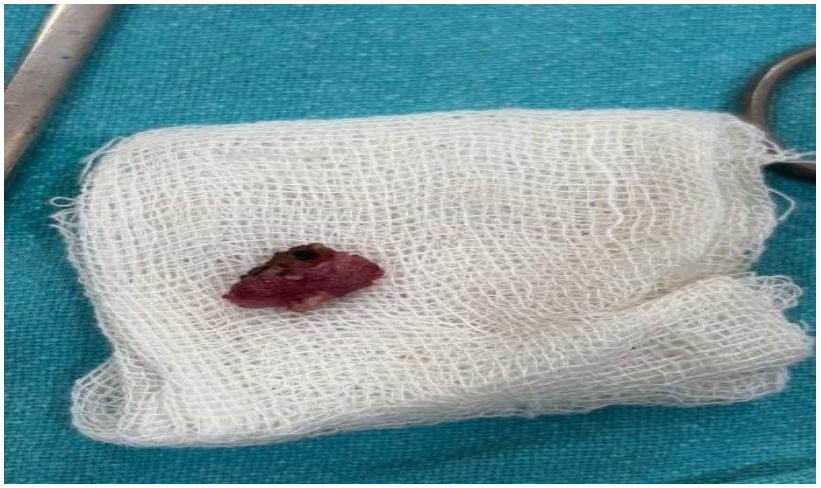

IMAGE 3 – EXCISED SPECIMEN

The excised specimen was subsequently submitted for histopathological evaluation to confirm the diagnosis.

On Histopathology ,**H&E stain** showed stratified squamous epithelium with dilated lymphatic channels confirming it as Lymphangioma**(IMAGE 4)** .

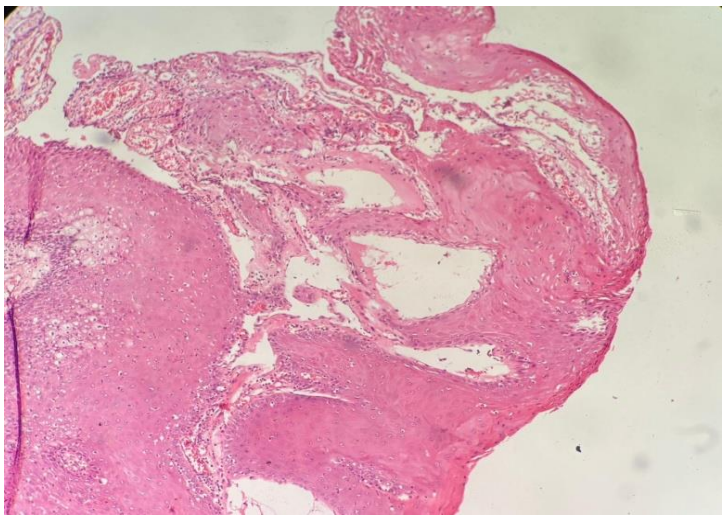

IMAGE 4 – H&E stain showing dilated lymphatic channels.

DISCUSSION:

Lymphangiomas are benign, hamartomatous proliferations originating from lymphatic vessels, typically due to disruptions in embryonic lymphatic development. These malformations arise when isolated portions of lymphatic tissue fail to properly connect with the central lymphatic system during fetal development [1]. They are commonly classified into three subtypes based on histological appearance:

- Capillary lymphangioma (also called lymphangioma simplex)
- Cavernous lymphangioma
- Cystic lymphangioma, also known as cystic hygroma [2]

There are two main theories regarding their embryological origin. One suggests that the lymphatic system arises from five primitive sacs derived from the venous system during the sixth week of gestation. The second posits that lymphatic tissues develop ectopically and fail to establish communication with the main lymphatic system, leading to sequestration and subsequent lesion formation [3].

These lesions most commonly appear in the cervicofacial region, particularly in the posterior triangle of the neck. Frequently involved landmarks include the posterior border of the sternocleidomastoid muscle, the midsection of the clavicle, and the anterior margin of the trapezius muscle [4].

Oral lymphangiomas are rare but when present, are most often found on the anterior dorsal surface of the tongue. These lesions can result in macroglossia, potentially causing speech problems, feeding difficulties, and airway obstruction in severe cases . Superficial lesions tend to have a vesicular or pebbly appearance, often described as resembling “frog eggs” or “tapioca pudding.” Deeper lesions are more diffuse and ill-defined, presenting as soft tissue masses without surface change [5].

Papillary projections may be seen in superficial cases, while deeper ones manifest as submucosal swellings. Tongue involvement typically produces uneven nodularity and gray-pink surface projections, with macroglossia being a hallmark feature [6].
Small lymphangiomas (less than 1 cm) are occasionally found on the alveolar ridge and tend to affect males more frequently, with a 2:1 male-to-female ratio . These lesions can impact several domains of the patient’s life, including function, aesthetics, occlusion, and psychosocial wellbeing. Rare complications, such as infection of tongue-based lymphangiomas, can lead to Ludwig’s angina, a life-threatening condition . Postoperative complications may include seroma, infection, minor hemorrhage, recurrence, and lymph leakage [7].

De Serres et al.proposed a classification system for head and neck lymphangiomas based on anatomical distribution:

Class I: Unilateral, infrahyoid
Class II: Bilateral, suprahyoid
Class III: Unilateral, suprahyoid or infrahyoid
Class IV: Bilateral, suprahyoid or infrahyoid
Class V: Bilateral involvement of both suprahyoid and infrahyoid regions

Histologically, lymphangiomas consist of markedly dilated lymphatic channels lined by a thin endothelium. These vessels may contain lymph, inflammatory cells (including lymphocytes, neutrophils, and macrophages), and sometimes red blood cells. The surrounding stroma often shows loose connective tissue with inflammatory infiltrates [8]. In intraoral cases, lymphatic spaces are frequently located just beneath the epithelial surface, sometimes replacing connective tissue papillae.

The differential diagnosis for lymphangioma includes several conditions such as:
Hemangioma, Amyloidosis, Congenital hypothyroidism, Neurofibromatosis, Down syndrome, Muscular hypertrophy

Surgical excision remains the primary treatment modality, especially for functionally compromising lesions like macroglossia. Goals of treatment include restoration of normal tongue size and function, preservation of taste, correction of dentofacial deformities, and cosmetic improvement [9].

However, complete resection is often challenging due to the lesion’s poorly defined, infiltrative margins, contributing to a high risk of recurrence. Lymphangiomas generally do not respond to sclerotherapy as well as hemangiomas do [10].

Additional or adjunctive treatment options include: Laser surgery (e.g., Nd:YAG, CO_2 laser) , Cryotherapy , Electrocautery , Sclerosing agents (limited efficacy) , Embolization , Systemic or intralesional steroids , Radiofrequency ablation , Radiation therapy (rarely used in children due to long-term risks) . Each case requires an individualized treatment plan, depending on lesion size, location, growth rate, and functional impact.

CONCLUSION

Although lymphangiomas are uncommon in the oral cavity, timely diagnosis is crucial for initiating appropriate treatment and reducing the risk of complications. Regular follow-up is equally important to monitor for potential recurrence.

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