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An Intriguing Encounter: A Rare Case of Tongue Lymphangioma and Its Clinical Implications

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ABSTRACT

Lymphangioma of the tongue, a benign tumor arising from a malformation in the lymphatic system, is a rare occurrence. It typically manifests as a soft, painless swelling on the anterior 2/3 of the tongue. Lymphangiomas are predominantly observed at birth (60-70%) or within the first two years of life (90%), with minimal prevalence in adulthood[1]. This report documents an exceptionally rare case of tongue lymphangioma in a 9-year-old girl, emphasizing its clinical manifestations, diagnostic assessments, and therapeutic alternatives.

Keywords: *developmental malformation, lymphangioma, tongue, vascular*

INTRODUCTION

Lymphangiomas are a type of vascular malformations characterised by abnormal growth of lymphatic vessels. They are rare in the oral cavity, with most cases occurring in the head and neck region[2]. Typically, they are found in the anterior two-thirds of the tongue, but there have been reports of cases in other oral areas such as the lips, palate, gingiva, cheek mucosa, and mandibular alveolar ridge[3]. Clinically, lymphangiomas appear as transparent, grouped vesicles that can be red or purple due to secondary hemorrhages. Deeper lesions present as nodular masses with varying colors and textures on the surface. Larger lesions can lead to difficulty in speaking, swallowing, or feeding, although many cases are asymptomatic. Various treatments were proposed and surgical excision is the most common and preferred treatment for lymphangiomas. The subsequent case report pertains to a patient presenting with lymphangioma of the tongue and its corresponding treatment modalityCASE REPORT

A 9-year-old girl with previously no known medical illness presented with 3 years history of tongue lesion. The mother noticed the lesion was slowly growing and increasing in size. It was otherwise painless, and there were no episodes of bleeding. It did not cause chewing or swallowing difficulties or upper airway obstructive

symptoms. There was no relevant family history.

On intraoral examination, there was an exophytic lesion over the posterior part of the anterior two-thirds of the tongue. It measured 1cm x 2cm, had an irregular surface and margin, was non-tender on palpation, firm in consistency, had a broad base, and was not pedunculated with dilated vessels seen near the lesion. Clinically, such a lesion was diagnosed as lymphangioma with a differential diagnosis of hemangioma and papilloma. The child was generally in good condition with no signs of upper airway obstruction.

Magnetic resonance imaging (MRI) was done as part of the investigation. It revealed a 1.0cm x 0.7cm x 0.3cm small exophytic lobulated superficial lesion seen in the superior aspect of the right posterior tongue. It also showed an intermediate signal on T1WI and a hyperintense signal on T2WI to the muscle with mild enhancement in the post-contrast study with no restricted diffusion and no abnormal signal intensity or enhancement of the intrinsic muscles of the tongue to suggest involvement. The tongue lesion was surgically excised in total with the patient under general anaesthesia. No complications ensued. The excised specimen was sent for histopathological examination and diagnosed as lymphangioma.

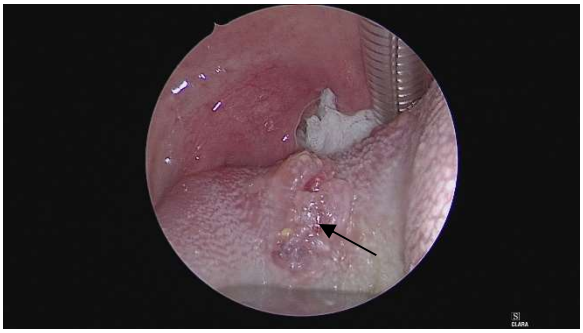

Figure 1: Intraoperatively, multiple pink nodules (black arrow) are visible on the dorsal aspect of the tongue.

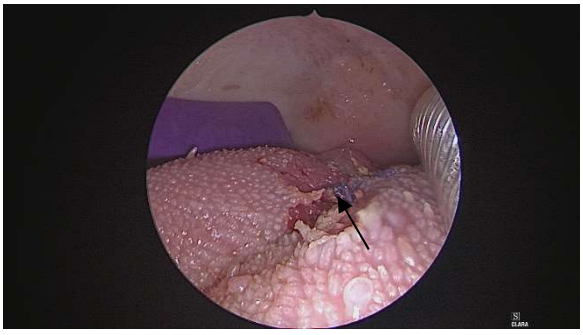

Figure 2: Post-excision of the dorsal aspect of tongue lymphangioma. Arrow pointed at the base of lesion after excision.

DISCUSSION

Lymphangioma is a benign, infrequent, nonmalignant lymphatic malformation. While highly unusual, lymphangiomas represent 25% of benign vascular tumors and 4% of all vascular tumors in children. Extensive research indicates an even distribution between genders and shows no indication of racial prevalence[1]. Notably, lymphangiomas demonstrate a marked affinity for the head and neck region, constituting approximately 75% of cases. Around 50% of these anomalies are identifiable at birth, with 90% appearing by the age of two[3]. Conversely, diagnosing lymphangiomas in adults is an exceedingly rare occurrence[3].

There are two predominant theories concerning the etiology of the condition. It is postulated that the condition may stem from the proliferation of residual primitive lymphatic tissue or from congenital obstruction of the lymphatic vessels. Classifications of lymphangiomas are based on the size of the lymphatic cavities, encompassing macrocystic, microcystic (capillary lymphangiomas), and cystic hygromas[5]. In the specific case under consideration, a lymphangioma situated on the tongue presents as a cavernous lymphangioma.

From a clinical standpoint, a lymphangioma typically presents as a translucent, vesicle-shaped lesion with a white or yellow hue. Some lesions may develop a reddish or purplish discoloration due to secondary hemorrhage resulting from blood vessel rupture. The characteristics of the lesion on this patient's tongue closely resembled the previously documented features of a lymphangioma on the dorsal surface of the tongue.

The diagnosis of lymphatic malformation or oral lymphangioma is clinical. Imaging modalities might be helpful in determining the extension of the lesion or its relation to the surrounding structures. In this case, Magnetic resonance imaging (MRI) and was performed and revealed the tongue lesion was only superficially involved. Treatment of lymphangioma is complex and depends on the size and type of lesion, association with anatomic structures, and infiltration to the surrounding tissues.

The optimal approach for the complete eradication of lymphatic malformations is through surgical intervention. Surgical excision, if not meticulously performed, is often linked to recurrence, with recurrence rates ranging from 10% to 53% [7]. Although alternative options such as radiation therapy, cryotherapy, electrocautery, sclerotherapy, steroid administration, embolization, ligation, Nd-YAG/CO2 laser surgeries, and radiofrequency tissue ablation have been explored, in the case illustrated in this report, surgical removal was the most viable option due to the superficial location of the lesion.

Sclerotherapy is commonly used in lymphatic malformation treatment. The therapy of macrocystic lymphatic malformation has been shown to benefit by the use of sclerotherapy agent which is bleomycin, an antineoplastic antibiotic that damages endothelial cells by causing a nonspecific inflammatory response and vessels occlusion. The major drawbacks of bleomycin reported are pulmonary toxicity, mucocutaneous effects such as skin erythema, pigmentation, edema and alopecia, and fever[10].

Despite the consensus that surgical excision is the best course of treatment for this disease, the infiltrating nature of lymphatic channels frequently raises the risk of surgical complications and lesion recurrences[11]

However in this case, the tongue lesion is superficial which favors surgery as the preferable treatment option as a diagnostic and therapeutic intent. The decision to excise was guided by the imaging study. The girl was follow-up in the clinic with no complication and there was evidence of recurrence at 3 months follow up.

CONCLUSION

Despite being rare in the oral cavity, the timely detection of lymphangioma is of paramount importance. This facilitates the prompt administration of appropriate treatment and helps avert potential complications, emphasizing the significance of attentiveness in clinical practice.

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