

Lymphangioma in Tongue- A Case Report with Review

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Case Report

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ABSTRACT

Lymphangioma is a benign hamartomatous lesion caused by congenital malformation of the lymphatic system. This benign tumor is detected most commonly at birth or in early childhood but rarely in adults. On clinical examination, most lymphangiomas contain clear lymph fluid, but some may present as transparent vesicles containing red blood cells due to hemorrhage. In addition, lymphangioma may occur in association with hemangioma. This tumor occurs most commonly in the head and neck area but rarely in the oral cavity. The dorsum of the tongue is the most common location in the mouth, followed by the lips, buccal mucosa, soft palate, and floor of the mouth. There are various treatment approaches for lymphangioma, but surgical excision is the preferred method. We present a case of a 13-year-old girl with lymphangioma on the anterior dorsal part of the tongue, not associated with any dysfunction in mastication or speech disorders.

Keywords: Lymphangioma, Dorsum of Tongue, Hamartomatous Growth.

Introduction

A thirteen-year-old female patient reported to the Department of Oral Medicine and Radiology with a chief complaint of growth on the left side of her tongue for one year. The patient reveals a history of growth noticed one year ago which was smaller in size in the beginning and gradually increased to the present size. The patient gave a history of disturbance in speech and bleeding from growth. The patient also reported difficulty in chewing and swallowing. No history of any pain associated with the growth.

On intraoral inspection, multiple papular appearances were seen on the left dorsal surface of half of the tongue of size approximately 4cm×3cm which was exactly within the midline and not crossing the midline. Extending antero-posteriorly from the tip of the tongue till 0.5cm anterior to circumvallate papillae. The surface appeared to be irregular and granular.

On palpation, growth was soft, non-tender, and pebbly. A Diascopy test was done which was negative and there were no palpable pulsations felt. Without any loss of sensation and motor function, there is a reduction in the movement of the tongue.

Based on the clinical findings, the case was provisionally diagnosed as the lymphangioma of the left dorsal surface of the tongue. hemangioma, granular cell tumor, and neurofibromatosis were considered as a differential diagnosis.

On ultrasound examination, a submucosal hypoechoic lesion involving the left half of the dorsal side of the anterior 2/3rd of the tongue was seen. Features consisted of a clinical diagnosis of lymphangioma.

The Hemogram of the patient was within normal limits. The growth was surgically incised under local anaesthesia and was sent to histopathological evaluation. Microscopic evaluation revealed- at an area close to the tongue epithelium: numerous thin-walled lymphatic capillaries covered by flat endothelium and consist of lymphatic fluid with few lymphocytes were evident.

Based on history, clinical and histological features a final diagnosis of lymphangioma of the left dorsal surface of the tongue was made.



Figure1 Shows Lesion Seen on Dorsal Aspect of Tongue with Pebbled Appearance



Figure2. A Prominent Mass which is Clearly Pebbled Surface on the Tongue

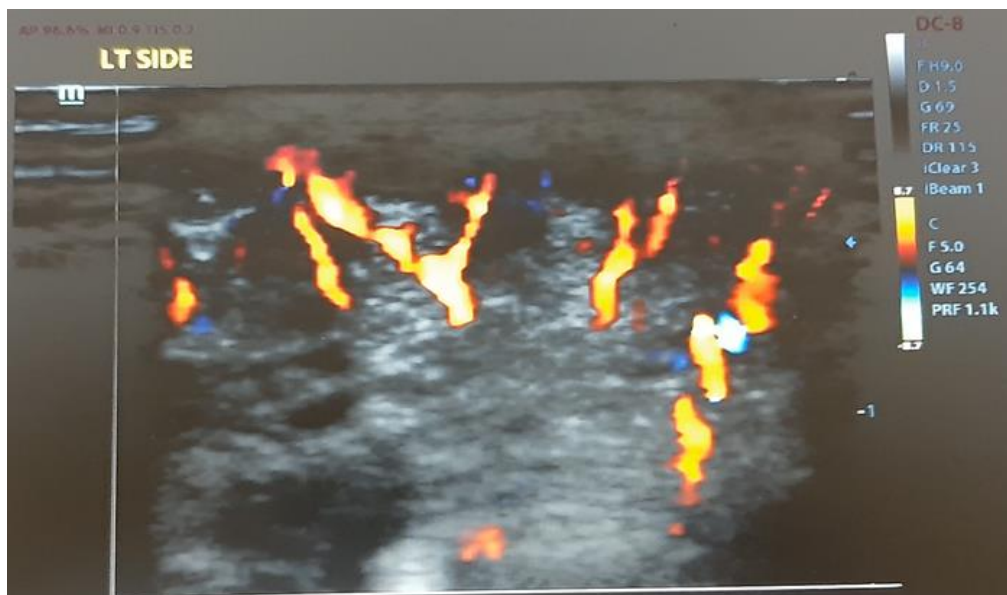


Figure 3 and Figure 4 Represents Hypoechoic Lesion Involving Mucosa and Submucosa of Tongue with Increased Vascularity from Ipsilateral Lingual Artery.

Discussion

Lymphangiomas are benign tumours of lymphatic vessels showing a marked predilection for the head and neck region.¹ They are extremely rare in the oral cavity.^{2,3} The common site of occurrence for lymphangioma in the oral cavity is the anterior dorsum and lateral border of the tongue. It can also be seen in other parts of the oral cavity such as the palate, cheeks, floor of the mouth, gingiva, and lips. Lymphangiomas are rare, they

account for 4% of all the vascular tumors and 25% of all benign vascular tumours in children.^{4,5,6} There is no sign of racial predominance and also equal sex incidence reported in most of the studies.

Histopathologically, lymphangioma are classified by

A. Watson and Mc Carthy as

1. Lymphangioma simplex (composed of small thin-walled lymphatics).
2. Cavernous lymphangioma (comprised of dilated lymphatics vessels with surrounding adventitia).
3. Cystic lymphangioma (consisting of huge, macroscopic lymphatic spaces surrounded by fibrovascular tissue, and smooth muscle).
4. Benign lymphangioendothelioma (lymphatic channels appears to be dissecting through dense collagenic bundles).

B. Serres et al gave a staging system based on the location and extent of the lesions:

1. Stage I is unilateral infrahyoid,
2. Stage II is unilateral suprahyoid,
3. Stage III is unilateral infrahyoid and suprahyoid,
4. Stage IV is bilateral infrahyoid,
5. Stage V is bilateral infrahyoid and suprahyoid.

The clinical diagnosis of the lesion is mainly based on the occurrence of superficial or deep lesions. Interestingly lesion can be easily identified by the appearance of tapioca pudding or frog eggs like appearance.⁷ Deeper lesions occur as diffuse nodules, soft in consistency and there will be changes seen in colour and the texture. Whenever lymphangioma shows rapid growth it is always associated with respiratory tract infection.⁸ The differential diagnosis for lymphangioma includes haemangioma, neurofibromatosis, primary muscular hypertrophy, congenital hypothyroidism.

The treatment of lymphangioma depends upon the size, type, involvement of anatomical structures, and infiltration to the surrounding tissues.⁹ The various treatment modalities available for treating lymphangioma are surgical excision, radiation therapy, cryotherapy, electrocautery, sclerotherapy, steroid administration, embolization and ligation, laser surgery with ND-YAG CO₂, radiofrequency ablation technique.^{10,11,12,13,14} Sclerosing agent application has been successively used in most of the lymphangioma cases. In some of the cases regarding controlling the size of the tongue and removing superficial lymphangioma laser photocoagulation technique is used.^{15,16}

Conclusion

Lymphangioma is rarely encountered in the oral cavity. Although, early diagnosis and proper treatment prevent the reoccurrence; follow-up must be done.

References

1. Uguru C, Edafioghor F, Uguru N. Lymphangioma of the tongue with macroglossia: a case report. Niger J Med. 2011; 20(1):166–68. <https://pubmed.ncbi.nlm.nih.gov/21970281/>
2. Tasca RA, Myatt HM, Beckenham EJ. Lymphangioma of the tongue presenting as Ludwig's angina. Int J Pediatr Otorhinolaryngol. 1999; 51(3):201–05. [https://doi.org/10.1016/s0165-5876\(99\)00262-1](https://doi.org/10.1016/s0165-5876(99)00262-1)
3. Hancock BJ, St-Vil D, Luks FI, Di Lorenzo M, Blanchard H. Complications of lymphangiomas in children. J Pediatr Surg. 1992; 27(2):220–4. discussion 224–26. [https://doi.org/10.1016/0022-3468\(92\)90316-y](https://doi.org/10.1016/0022-3468(92)90316-y)

4. De Queiroz AM, Silva RA, Margato LC, Nelson FP. Dental care management of a young patient with extensive lymphangioma of the tongue: A case report. *Spec Care Dentist*. 2006; 26(1):20-4. <https://doi.org/10.1111/j.1754-4505.2006.tb01505.x>
5. Guelmann M, Katz J. Macroglossia combined with lymphangioma case report. *J CliPediater Den*. 2003; 27:167-9. <https://doi.org/10.17796/jcpd.27.2.a466020r10440121>
6. Mosca RC, Pereira GA, Mantesso A. Cystic hygroma: characterization by computerized tomography. *Oral Surg Oral Med Oral Pathol Oral RadiolEndod*. 2008; 105(5):65-9. <https://doi.org/10.1016/j.tripleo.2008.01.015>
7. Anurag T, Munish M, Kamakshi, Shuchita G, et al. Anesthetic consideration in macroglossia due to Lymphangioma of Tongue: A Case Report. *Indian J Anaesth*. 2009; 53(1):79-83. <http://www.ijaweb.org/text.asp?2009/53/1/79/60263>
8. Martin SG, Michael G, Jonathan AS. *Burket's Oral Medicine* (11th ed) Hamilton: BC Decker Inc; 2008. pp. 139-41.
9. Brennan TD, Miller AS, Chen S. Lymphangiomas of the oral cavity: a clinicopathologic, immunohistochemical, and electron-microscopic study. *J Oral Maxillofac Surg*. 1997; 55(9):932-35. [https://doi.org/10.1016/s0278-2391\(97\)90062-8](https://doi.org/10.1016/s0278-2391(97)90062-8)
10. Mandel L. Parotid area lymphangioma in an adult: case report. *J Oral Maxillofac Surg*. 2004; 62(10):1320-23. <https://doi.org/10.1016/j.joms.2003.12.040>
11. Yaita T, Onodera K, Xu H, Ooya K. Histomorphometrical study in cavernous lymphangioma of the tongue. *Oral Dis*. 2007; 13(1):99-104. <https://doi.org/10.1111/j.1601-0825.2006.01256.x>
12. Kheur Supriya M, Samapika R, Yashwant I, Desai RS. Lymphangioma of Tongue: A Rare Entity. *Indian Journal of Dental Advancements*. 2011; 3(3):635-37. <http://rep.nacd.in/ijda/pdf/3.3.635.pdf>
13. Sunil S, Gopakumar Devi, Sreenivasan BS. Oral lymphangioma - Case reports and review of literature. *Contemporary Clinical Dentistry*. 2012; 3(1):116-18. <https://dx.doi.org/10.4103%2F0976-237X.94561>
14. Mousumi G, Sanjay S, Gokkulakrishnan, Singh Amit. Lymphangioma of the tongue. *Natl J Maxillofac Surg*. 2011; 2(1):86-8. <https://dx.doi.org/10.4103%2F0975-5950.85862>
15. Ligiastnescu, Georgescu, Cristiana S, Iuliana G. Lymphangioma of the oral cavity. *Romanian Journal of Morphology and Embryology*. 2006; 47(4):373-77. <https://pubmed.ncbi.nlm.nih.gov/17392986/>
16. Vyas T. Biopsy of Oral Lesion -A Review Article. *J Adv Med Dent Scie Res* 2018; 6(1):27-35. <http://jamdsr.com/htmlissue.php?id=1874>

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