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CASE REPORT

Lymphangioma of the Tongue: A Case Report

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ABSTRACT

Lymphangioma is a benign growth caused by the developmental malformation of lymphatic vessels that usually occurs within the first two decades of life. The clinical features are painless, nodular, vesicle-like swellings of the soft tissues, most commonly found on the anterior two-thirds of the dorsal surface of the tongue. Histopathological analysis demonstrates multiple dilated lymphatic vessels that contain proteinaceous fluids and occasionally leukocytes. We report a case of a 7-year-old Thai boy who presented with papillary and vesicle-like swelling on the left dorsal aspect of the tongue that had been present for 5 years. The lesion was surgically excised and monitored for 6 months, during which time there were no signs of recurrences.

Key words: dorsal tongue, lymphangioma, histopathology, Thailand

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INTRODUCTION

Lymphangioma is a benign, hamartomatous abnormality of lymphatic vessels caused by the developmental malformation of lymphatic tissues.¹ It usually manifests within the first two decades of life and frequently in the head and neck area. The clinical features are painless, nodular, vesicle-like swellings of the soft tissues, most commonly found on the anterior two-thirds of the dorsal surface of the tongue.² Histopathological features are numerous dilated lymphatic vessels lined by a single layer of endothelial cells. The lumina are filled with lymph fluid and occasionally white blood cells.¹

CASE REPORT

A 7-year-old Thai boy presented to the Dental Unit of Chiangraiprachanukroh Medical Center, Chiang Rai, Thailand with an asymptomatic growth on the dorsal tongue that had been present for 5 years. The patient's

medical history and dental history were documented, and physical examination was performed. The patient did not have any systemic diseases. Intraoral examination revealed a nodular swelling on the anterior two-thirds of the dorsal surface of the tongue measuring 2.5 × 1.5 × 0.5 cm. The nodular surfaces exhibited a reddened, papillary, and vesicle-like swelling, resembling the appearance of frog eggs or tapioca pudding (Figure 1). Histopathological analysis revealed that the surface of the mucosal specimen was acanthotic parakeratinized stratified squamous epithelium. Numerous dilated, lymph-filled vessels were identified, and a focal group of white blood cells in the lumina was noted. The submucosa exhibited striated muscle bundles, adipose tissue, blood vessels, extravasated erythrocytes, and nerve bundles (Figures 2 and 3). The lesion was definitively diagnosed as a lymphangioma, and it was surgically removed in its entirety with the patient under general anesthesia. No complications ensued. The patient was monitored for 6 months and exhibited complete healing of the lesion and no signs of recurrence (Figure 4).



Figure 1: The clinical features of the dorsal aspect of the patient's tongue. Multiple pink nodules are visible on the left side.

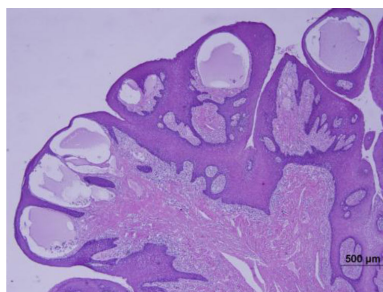


Figure 2: The microscopic features of a nodular specimen whose surface consisted of parakeratinized stratified squamous epithelium. The underlying connective tissue exhibits multiple lymphatic vessels. (Hematoxylin and eosin stain; original magnification, $\times 40$.)

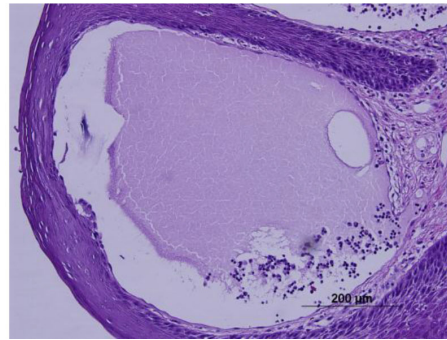


Figure 3: High-power view of the lesion depicted in Figure 2, showing a lymphatic vessel with a single layer of endothelial cells. The lumen is filled with pink lymph fluid and a group of leukocytes. (Hematoxylin and eosin stain; original magnification, $\times 200$.)



Figure 4: Postoperative appearance of the tongue after 6-month follow-up, demonstrating complete healing of the lesion and no signs of recurrence.

DISCUSSION

Lymphangioma is a rare, benign lymphatic malformation. It was proposed to develop from a congenital obstruction of the lymphatic vessels or a proliferation of the remnant of the primitive lymphatic tissue. Lymphangiomas can be classified into three subtypes on the basis of size of the lymphatic vessels: (1) macrocystic, which is composed of cyst-like spaces measuring ≥ 2 cm in diameter; (2) microcystic, with vascular channels < 2 cm in diameter; and (3) mixed.¹ Lymphangioma of the head and neck are common, accounting for approximately 50%–75% of all cases of lymphangiomas. Within the head and neck region, the most common location of these lesions is the cervical area, mainly at the posterior triangle of the neck (the area bordered by the upper middle third of the clavicle, the anterior edge of the trapezius muscle, and the posterior border of the sternocleidomastoid muscle). The clinical manifestation is usually a soft or

fluctuant mass.³ Although lymphangioma is a benign lesion and usually causes no harm except discomfort during chewing, large lesions on the tongue may be life-threatening because they can cause upper airway obstruction.

Oral lymphangioma is found mostly on the anterior two-thirds of the dorsal surface of the tongue, which results in enlargement of the tongue (macroglossia).⁴ Approximately 90% of oral lymphangioma develop by the age of 2 years.⁵ In our patient, the lesion was first observed when the patient was aged 2 years. Clinically, a lymphangioma usually appears as a translucent, vesicle-like lesion, white or yellow in color, resembling frog eggs or tapioca pudding. Secondary hemorrhage by rupture of blood vessels may cause some of these lesions to become red or purple.⁶ In our patient, the clinical appearances were similar to the classic features of lymphangioma of the dorsal surface of the tongue previously described.

Lymphangioma may be treated by surgical excision, cryotherapy, sclerotherapy, embolization, ligation, or radiation therapy.³ Surgeons must evaluate cases individually because the sizes and locations of the lesions vary. In cases that involve the vital structures, surgical excision may not be appropriate, and alternative approaches should be used.⁷

CONCLUSION

Lymphangioma is an idiopathic, rare lymphatic malformation that commonly involves the head and neck region, particularly in children. Pediatric dentists may be the first health care providers who recognize the lesions, and cases are referred to oral and maxillofacial surgeons for proper management.

CONFLICTS OF INTEREST

The authors declare that there were no conflicts of interest related to this case report.

REFERENCES

1. Neville B, Damm DD, Allen C, Chi A. Oral and Maxillofacial Pathology, 4th edition. Canada: Elsevier; 2016.
2. Eren S, Cebi AT, Isler SC, Kasapoglu MB, Aksakalli N, Kasapoglu C. Cavernous lymphangioma of the tongue in an adult: a case report. *J Istanbul Univ Fac Dent*. 2017; 51(2):49-53.
3. Erugula S, Mvs S, Sameera A, Vujhini S, Kandukuri M. A rare case of oral lymphangioma of tongue. *Int J Contemp Pediatrics*. 2016;3(3):1112-4.
4. Iamaroon A, Pongsiriwet S, Srisuwan S, Krisanaprakornkit S. Lymphangioma of the tongue. *Int J Paediatr Dent*. 2003;13(1):62-3.
5. Usha V, Sivasankari T, Jeelani S, Asokan GS, Parthiban J. Lymphangioma of the tongue - a case report and review of literature. *J Clin Diagn Res*. 2014; 8(9):ZD12-4.
6. Vasconcelos MG, Santos BC, Lemos LC, Ribeiro BF, Iglesias DP, Vasconcelos RG, Medeiros AM. Oral lymphangioma: case report. *RSBO*. 2011;8(3):352-6.
7. Kotrashetti V, Pammar C, Nayak R, Hosmani J. Lymphangioma of the buccal mucosa: A case report with a literature review on lymphangioma of buccal mucosa. *J Orofac Sci*. 2015; 7(2):129-31.

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