ISSN: 2643-3907

Mustafa. Res Rep Oral Maxillofac Surg 2021, 5:059
DOI: 10.23937/2643-3907/1710059
Volume 5 | Issue 2
Open Access

Research Reports in Oral and Maxillofacial Surgery

MINI REVIEW

Lymphangioma of the Tongue Revisited

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Abstract

Lymphangioma is a benign, congenital hamartomatous malformation of the lymphatic system, it is more common in the head and neck region. Oral lymphangiomas are rare but if it occurs, the tongue is the most commonly affected site, rarely on other sites such as the palate, gingiva, and alveolar ridge of the mandible. This study aims to throw light on lymphangioma of the tongue regarding pathogenesis, clinical signs and symptoms, and the different treatment modalities. Although lymphangioma is benign and its occurrence in the tongue extremely rare, a health care provider like a dentist must be conscious of the existence of such lesion to promote a precise diagnosis, and therefore, proper treatment can be rendered for this disorder, to avoid the serious complications that might occur when it becomes traumatized tor infected in which case it obstructs the airway and lead to the death of the patient if not promptly rescued.

Introduction

Lymphangioma is benign tumors involving lymphatic channels the majority are confined to the head and neck region in about the majority of cases [1]. Lymphangiomas occur commonly at birth or before the age of two [2]. In the oral cavity, the anterior two-thirds of the tongue is the most commonly involved site [3]. This study aims to throw light on lymphangiomas of the tongue regarding pathogenesis, clinical signs and symptoms, classification, and the different treatment modalities.

Materials and Methods

An electronic search of the databases was conducted, within PubMed, using a combination of keywords and control terms (MeSH) were used wherever possible. The search terms included lymphangioma and oral cavity, lymphangioma and tongue, lymphatic malformations, and tongue.

Literature review

Lymphangioma is a benign, congenital hamartomatous malformation of the lymphatic system; it is more common in the head and neck region [2]. It arises as a result of hyperplasia of sequestered lymphatic vessels that have lost connection with the rest of the lymphatic channels. Virchow was the first to describe lymphangioma in 1854 [2]. Lymphangioma has a high for the head and region with about 75% occurring at this site. 50% of lymphangiomas are present at birth and 90% develop by 2 years of age [2]. The tongue is the most commonly affected site in the oral cavity, rarely on other sites such as the palate, gingiva, and alveolar ridge of the mandible [4]. Lymphangiomas usually present as papillary lesions with the same colour as adjacent mucosa [5]. Occasionally oral lymphangiomas are associated with syndromes like Turner's syndrome, Noonan's syndrome, trisomies, cardiac anomalies, fetal hydrops, fetal alcohol syndrome, and Familial pterygium colli [6]. Tongue lymphangiomas are usually superficial with a pebbly surface resembling a cluster of translucent vesicles [7]. The anterior two-thirds of the tongue is the most commonly affected site causing enlargement of the tongue [7]. Patients with tongue lymphangioma tend to have speech disturbances, poor oral hygiene, and bleeding from the tongue associated with trauma [8].

The complications of lymphangioma affect the patients in many ways including aesthetic, occlusal, functional, and psychological aspects [9]. The most serious complications are those related to infection which can result in Ludwig's angina associated with

Citation: Mustafa AHK (2021) Lymphangioma of the Tongue Revisited. Res Rep Oral Maxillofac Surg 5:059. doi.org/10.23937/2643-3907/1710059

Accepted: October 06, 2021; **Published:** October 08, 2021

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an infected base of the tongue lymphangioma [10]. Postoperative complications include seroma formation, infections, minor bleeding, recurrent cellulitis, and lymph fluid leakage [11].

Lymphangiomas have been classified into:

- Lymphangioma Simplex (Capillary Lymphangioma) consists of small, capillary sized vessels.
- Cavernous Lymphangioma is composed of large, dilated lymphatic vessels.
- Cystic Lymphangioma (Cystic Hygroma) exhibits large macroscopic cystic spaces [12].

Lymphangiomas are characterized histologically by the formation of lymphatic channels lined by endothelial cells, often the three types may be seen at the same lesion. The most common of the three types is the cavernous lymphangioma and is seen commonly in the tongue and floor of the mouth [13].

Another classification of the lymphangioma of head and neck based on the anatomical involvement had been proposed by De SerresLM.

- 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 [14].

According to their clinical presentation lymphangiomas are classified into macrocystic (cavities larger than about 2 cm^3), microcystic (cavities smaller than about 2 cm^3), and mixed (combining these two types). The objectives of the treatment of lymphangiomatous macroglossia.

The differential diagnosis for lymphangioma includes haemangioma, congenital hypothyroidism, Amyloidosis, Neurofibromatosis, Primary muscular hypertrophy [15].

The treatment of lymphangioma depends upon their type, size, the involvement of anatomical structures, and infiltration to the surrounding tissues.

Different treatment modalities have been tried for the management of lymphangiomas such as laser therapy, cryotherapy, embolization, and electrocautery. Various types of sclerosing agents have been used for the treatment of lymphangiomas including that affecting the tongue, bleomycin, and OK432. OK432 is a lyophilized mixture of streptococcus pyogenes and penicillin G potassium is considered as the first line of treatment of lymphangiomas because of the absence of perilesional fibrosis when using OK432 [16]. The sclerosing agent OK-432 is effective for macrocystic lymphatic malformations but showed less promise for microcystic lesions, mixed lesions, and lesions outside the head and neck region [8,17].

The after-effects of sclerosing agents include fever and swelling at the site of injection [16]. Because of the high fluid content in the tissues and poor blood supply, cryosurgery has also been tried for treating lymphangiomas [4]. Laser photocoagulation has been reported useful in controlling the tongue size and removing superficial lymphangioma in some cases [18]. Surgical excision is the ultimate choice of treatment for lymphangiomas. Surgical excision is preferred as spontaneous regression of lymphangiomas is rare [2] and most of the adult lymphangiomas are encapsulated or partially circumscribed [16].

Conclusion

Although lymphangioma is benign and its occurrence in the tongue extremely rare, a health care provider like a dentist must be conscious of the existence of such lesion to promote a precise diagnosis, and therefore, proper treatment can be rendered for this disorder, to avoid the serious complications that might occur when it becomes traumatized tor infected in which case it obstructs the airway and lead to the death of the patient if not promptly rescued.

Conflict of Interest

None declared.

Ethical Approval

Not applicable.

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