



CASE REPORT

The Treatment of Lymphangioma in the Buccal Mucosa by Surgical Excision: A Rare Case Report

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Abstract

Lymphangiomas are benign hamartomatous tumors which are characterized by abnormal proliferation of lymphatic vessels. They are lymphatic malformations which are localized to head and neck region. They are rare located in the oral cavity. Tongue, lips, buccal mucosa, soft palate and floor of the mouth are affected in the oral cavity. However buccal mucosa is infrequent in literature. In this report report, a rare case of lymphangioma of the left buccal mucosa of retromolar area in a 30-year-old female patient has been treated by surgical excision is presented.

Keywords

Lymphangioma, Buccal mucosa, Lymphatic vessels



Figure 1: The view of lesion that is located in buccal mucosa left retromolar region.

Introduction

Lymphangioma is a benign hamartomatous tumor caused by congenital malformation of lymphatic system. Lymphangiomas are usually present at birth, and approximately 80% cases are present by age of 2 years. They are commonly found head and neck region. The most common area of head and neck region is the sub-mandibular region and also parotid gland [1]. The oral cavity can be affected by these lesions. They usually seen at the anterior two thirds of tongue followed by palate, gingiva, lips, alveolar ridge of mandible and rarely buccal mucosa [2]. Various treatment modalities have been used for the cure of lymphangioma. Surgical excision, radiation, laser therapy, and sclerotherapy can be used for the treatment of lymphangioma [3]. In this report, a rare case of lymphangioma of the left buccal

mucosa of retromolar area in a 30-year-old female patient has been treated by surgical excision is presented.

Case Report

A 30-year-old woman was referred to the Department of Oral Maxillofacial Surgery with a chief complaint of growth and blisters on buccal mucosa of mandibular left retromolar region of and halitosis. Patient also complains discomfort during feeding. There is no systemic disease in the medical history of patient.

On intraoral examination, discolorization was seen in left buccal mucosa of retromolar region. The lesion was exophytic with color ranging from reddish purple to yellow was seen (Figure 1). On palpation, the lesion soft and nontender. Diascopy test was carried out which was



Figure 2: The view of buccal mucosa after excision of the lesion.



Figure 3: Closure of the wound.

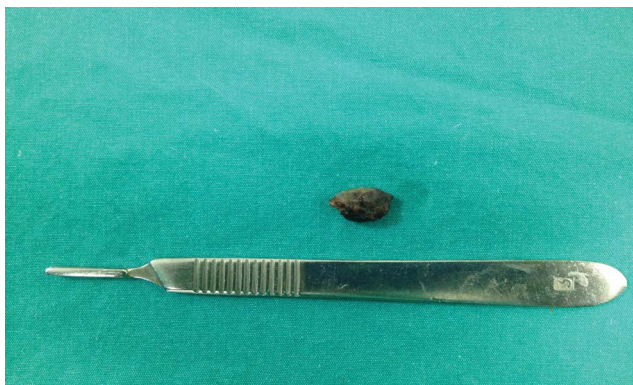


Figure 4: Excised surgical specimen.

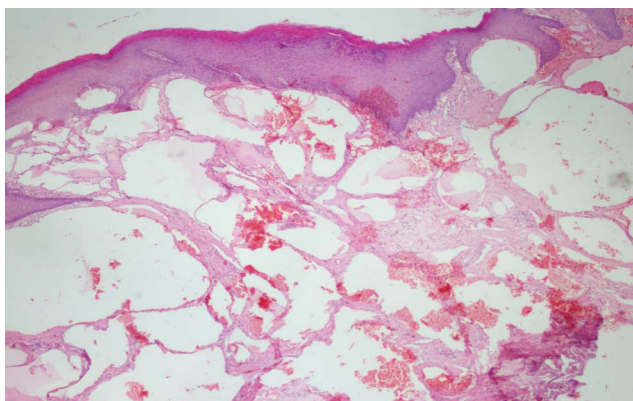


Figure 5: Numerous dilated lymphatics.

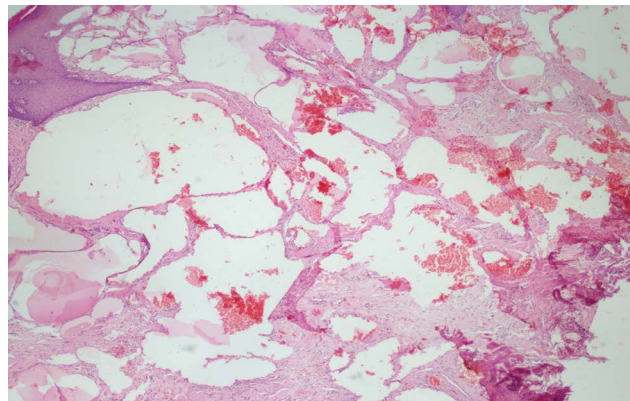


Figure 6: Some of the section revealed the presence of channels filled with red blood cells.



Figure 7: A 6-month follow-up.

negative. There was no evidence of adjacent bone and tooth involvement on orthopantomographic examination. On the basis of history and clinical features a provisional diagnosis of lymphangioma was made and an excisional biopsy under local anaesthesia was planned. The lesion was excised by surgical (Figure 2) and wound was sutured primary (Figure 3). The excised lesion (Figure 4) was sent for histopathological examination and diagnosed as lymphangioma. Microscopic examination showed numerous dilated lymphatics lined by endothelium (Figure 5). Some of the section revealed the presence of channels filled with red blood cells (Figure 6). There were also many of accumulation of lymphocytes. The patient was discharged after 6 months (Figure 7) and 1 year with normal looking buccal mucosa. There was no evidence of recurrence of the lesion on follow up of 1 year.

Discussion

Lymphangioma is congenital vascular abnormality of the lymphatic system which is rare benign lesion. Lymphangioma was first defined by Virchow in 1854 [3]. Several theories have been proposed for pathogenesis of lymphangioma. The first theory is that lymphatic system develops from five primitive sacs arising from the venous system. Concerning the head and neck, endothelial outpouching from the jugular sac spread centrifugally to form the lymphatic system. Another theory proposes that the lymphatic system develops from mesenchymal clefts in the venous plexus reticulum and spread centripetally towards the jugular sac. Lastly, lymphangioma

develops from congenital obstruction or sequestration of primitive lymphatic enlargement [4]. These lesions are frequently reported at birth (60%) and 90% grow by 2 years of age. Lymphangioma is rare present in adults [5]. In this case, a 30-year-old female reported.

De Serres, et al. offered a classification of the lymphangioma of head and neck based on of the spread the anatomical involvement [6].

- Stage/Class I: Infrahyoid unilateral lesions
- Stage/Class II: Suprahyoid unilateral lesions
- Stage/Class III: Suprahyoid and infrahyoid unilateral lesions
- Stage/Class IV: Suprahyoid bilateral lesions
- Stage/Class V: Suprahyoid and infrahyoid bilateral lesions

Lymphangiomas, approximately 75% cases, are located in head and neck region. The oral cavity is rarely affected site including tongue, lips, buccal mucosa, hard and soft palate, gingiva [4]. Tongue is commonly affected by the lymphangioma in the oral cavity. Brennan, et al. reported 49 cases of oral lymphangioma and 17 cases were located in tongue while only one case was placed at retromolar region and one at soft palate [7]. Buccal mucosal lesions of lymphangioma are exactly uncommon in the literature [8].

Clinically, lymphangiomas are divided into three types which are microcystic, macrocystic and mix. Macrocystic lesions are larger than 2 cm, whereas microcystic lesions are less than 2 cm.

Mix type is combining of microcystic and macrocystic types [9,10]. Morphologically, oral lymphangiomas are fairly typical. The superficial types manifest a pebbly surface and vesicle-like feature with so called 'frog egg' appearance and gradually enlarge. The deep lesions usually present as submucosal mass.

Histopathologically lymphangiomas are classified as: [11,12].

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

Treatment of lymphangioma alter their type, size, involvement anatomical structures and infiltration to surrounding tissues. Microcystic lesions are diffuse and difficult to remove in contrast macrocystic lesions are

localized and easily excised. Various methods can be tried the treatment of lymphangiomas. Such as laser therapy, cryotherapy, surgical excision, sclerotherapy, electrocautery [3,13,14].

Conclusion

Oral lymphangiomas are common lesions occurring at the tongue and rarely at the buccal mucosa of retromolar region. Localized lesions can be treated by conservative surgical excision with less recurrence rate. Therefore, the correct diagnosis and therapeutic indication of this type of lesions is crucial.

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