



## Case Report

## Lymphangiohemangioma of buccal mucosa: Report of a rare case



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## ABSTRACT

Vascular anomalies are divided in two groups of vascular tumors and vascular malformation. Hemangioma and lymphangioma are both examples of vascular tumors. Hemangiomas are known as proliferative vascular lesions. They present at birth and are characterized with increased cellular turnover. On the other hand lymphangiomas are hamartomatous tumors of lymphatic vessels. In contrast to these vascular tumors, vascular malformations could be considered as structural anomalies of blood vessels of different origin such as vein, capillaries, lymphatic vessels or combinations.

In this article, we presented a case of 23 year-old male with a chief complaint of swelling of the buccal mucosa, which caused some asymmetry in the lower lip. Clinical and histopathological findings clear out this lesion as a combination form of vascular malformations.

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## 1. Introduction

Vascular anomalies are best classified by International Society for the Study of Vascular Anomalies (ISSVA) [1]. In this biological classification, Mulliken and Glowacki are describing two major groups of vascular tumors and vascular malformations according to the cellular turnover, histology, clinical findings, and natural history [2–4] pertaining to such tumors and malformations.

Hemangioma is classified as a vascular tumor or a proliferative vascular lesion, which is characterized by increased endothelial turnover. However vascular malformations are stable lesions populated by a mature endothelium that grows with a child, and they can be categorized according to the type of vessel involved including lymphatic, capillary (port-wine stains), venous, arteriovenous and mixed malformations and according to hemodynamic features (low flow or high flow) [3,5].

Hemangioma is the most common benign vascular tumors. It usually appears at birth, and its growth exceeds the child's growth. However, many hemangiomas regress spontaneously before or during puberty [6]. Hemangiomas mostly occur in female. Majority of hemangiomas involve the head and neck. However there are rare in the oral cavity [7]. Hemangiomas are classified by Enzinger and Weiss into capillary, cavernous, and miscellaneous forms [4].

Lymphangioma is a benign hamartomatous tumor of lymphatic vessels. It is widely considered as a developmental [8] and congenital [9] lesion rather than a true neoplasia [8]. Although it is rarely diagnosed in adults [8], about 50% of lymphangiomas diagnosed in infancy and about 90% of them are developed in early childhood as lobular masses or cystic lesions [9]. Superficial lesions are pebbly, vesicle-like and the so-called “frog-egg” or “tapioca-pudding” appearance. Deeper lymphangioma may present as a submucosal mass [10]. Depending on the anatomical location, it may determine damages such as tissue swelling, obstruction of upper airways, pain, tongue's extrusion, sialorrhoea, deformity of jaws, as well as difficulties in mastication, speech, and so on [8].

Secondary hemorrhage into the lymphatic spaces may cause some of these “vesicles” to become purple [5]. Lymphangioma is classified by the diameter of the vessels into Capillary lymphangioma, Cavernous lymphangioma, and Cystic lymphangioma.

Cavernous lymphangiomas are more frequent in the mouth, where the denser surrounding connective tissue and skeletal muscle limit vessel expansion [5].

<sup>\*</sup> Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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Fig. 1. Clinical feature of patient. Asymmetry of lower lip.

Complete surgical excision remains the most accepted treatment option when it is possible [11]. While the common location of lymphangioma is head and neck, it is rarely reported in oral cavity and the preferred oral location is the tongue. The other sites of involvement in the oral cavity may include palate, gingiva, lips, and alveolar ridge of mandible [9]. In the case that we are going to present in this article we encountered the histopathologic structures of both lymphangioma and hemangioma in the oral cavity. These mixed vascular malformations are named as lymphangiohemangioma or hemangiolympangioma according to the dominant tissue structure. Although combined vascular malformation is not common in the oral cavity, some articles reported the occurrence of mixed vascular malformations in the oral cavity [12–15]. Here we present a patient with a lymphangiohemangioma of the buccal mucosa.

## 2. Case report

A 26 year-old male patient from the north of Iran reported to the Department of Oral & Maxillofacial Medicine of Babol Dental

School with the chief complaint of a painless swelling in the left buccal mucosa.

Patient noticed swelling over the left buccal mucosa 2–3 years back. He mentioned repeated episodes of bleeding on mechanical trauma to the lesion such as chewing, brushing, speaking, and so on. In an extraoral examination, there was an asymmetry around the lip area (Fig. 1).

There was no lymphadenopathy or any paresthesia. On palpation, the lesion was soft, non-tender and no obvious bleeding was identified.

Intraoral examination revealed approximately 3.5 cm × 1.5 cm exophytic lesion. It was a diffused, lobulated swelling that was extended from the anterior border of the left buccal mucosa to the level of premolar teeth. The lesion was red-yellowish in color. Close inspection revealed a few pinhead-sized, clear vesicles throughout the lesion.

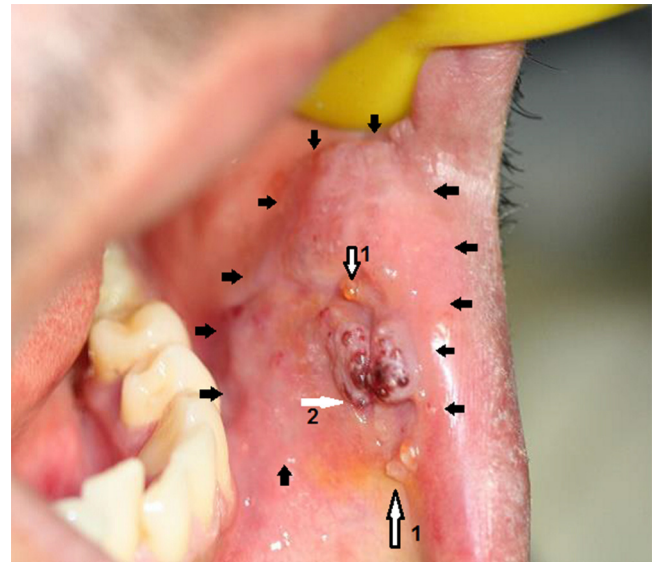


Fig. 2. Intraoral view of lobulated swelling of the anterior buccal mucosa (black arrows). Pinhead-sized, clear vesicles (arrow 1). The reddish exophytic masses (arrow 2). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

There were two separated, sessile, exophytic and less than 1 cm masses on the anterior aspect of the lesion which were granular and reddish in color (Fig. 2). Patient reported that these two masses have occurred recently in 3 months.

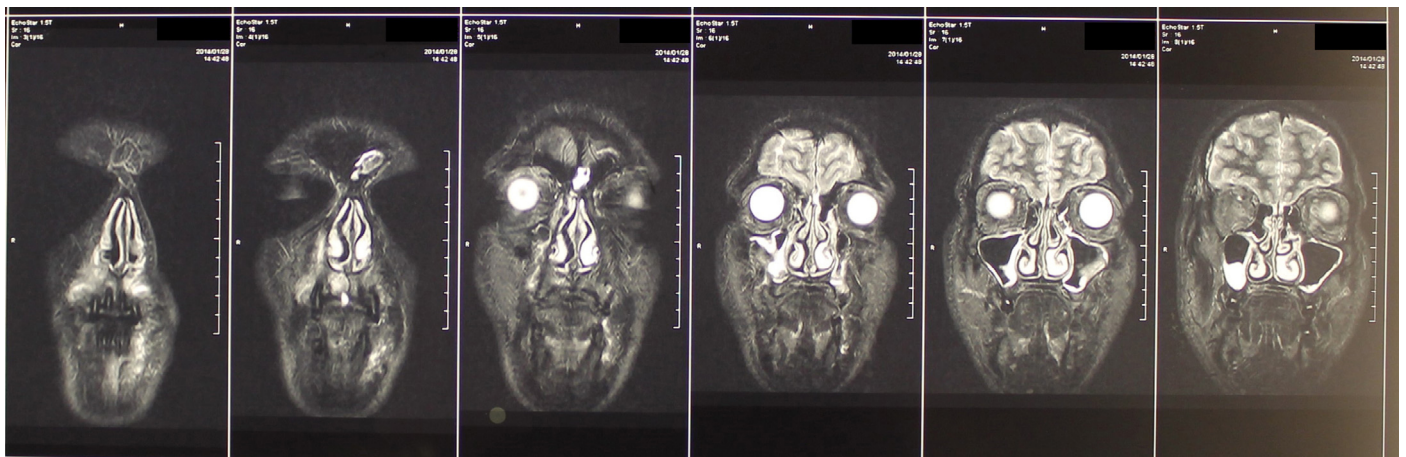


Fig. 3. MRI images of coronal view fat saturated T2 weighted show heterogeneous high signal intensity, ill-defined lesion.

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