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Lymphangioma Circumscriptum of Lower Lip – A Rare Case Report

Abstract:
The lymphangioma is a benign hamartomatous, uncommon, hyperplasia of lymphatic system. It is common in head and neck but rarely found in oral cavity. In oral cavity, it occurs mostly in tongue. In this article, we will report a case of lymphangioma circumscriptum of lip in a young male which is unusual.

Key-words: Lymphangioma, Mucocele, Lower Lip

Introduction:

Redenbacher first described lymphangioma in 1828. Lymphangiomas are uncommon, benign malformations of the lymphatic system that can occur anywhere on the skin and mucous membranes and are localized to head and neck region in about 50% - 75% of cases. Lymphangiomas is divided as deep or superficial on basis of size & depth of the abnormal lymphatic vessels or as congenital or acquired[1]. Cavernous lymphangiomas and cystic hygromas are deep forms of lymphangioma. These are two distinct congenital types. Lymphangioma circumscriptum and acquired lymphangioma, which is also referred as lymphangiectasia are of superficial forms. Even though both the type shows similar clinical and histologic features, lymphangioma circumscriptum denotes lymphatic channel dilation due to a congenital malformation of the lymphatic system and lymphangiectasia, or acquired lymphangioma, indicate dilated lymphatic channels of earlier normal lymphatics that have become obstructed by an external cause. The most frequent site in oral cavity is dorsum of the tongue, lips, buccal mucosa, soft palate, and floor of the mouth. Lymphangiomas are painless mass of soft tissue and is slow growing[2]. Superficial lesions consist of elevated nodules with pink or yellowish color or seen as transparent grouped vesicles, which can be red or purple due to secondary hemorrhages. Deeper lesions are described as soft, diffuse masses with normal color[3].

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Case Report:

A 19 year old male reported to our department of oral and maxillofacial surgery with chief complaint of painless swelling on the inner aspect of right lower lip since one month. Swelling was initially small and then was increasing gradually to attain the present size. There was no relevant past medical and dental history.

On intraoral examination, an oval, solitary, fluctuant swelling was seen on inner aspect of lower lip at right canine region [**Fig.1**]. Swelling was approx. 10x5mm in size. There was no differentiation of colour in relation to oral mucosa.

Based on the location and clinical features, it was diagnosed as a case of mucocele. Excisional biopsy

[Fig.2] under local anesthesia was done by placing an incision circumferentially and wound was primarily closed

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[Fig.4]. The surgical specimen was sent for histopathological examination

[Fig.3]. The histopathological examination of the lesion revealed acanthosis and hyperkeratosis of stratified squamous cell with dilated lymph vessels. It is lined by a layer of a flattened epithelial cells which is suggestive of lymphangioma. Wound healing is sufficiently good and no complication is seen post operatively
[Fig.5].

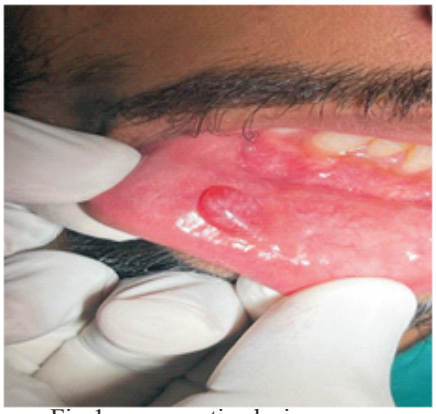

Fig.1 preoperative lesion

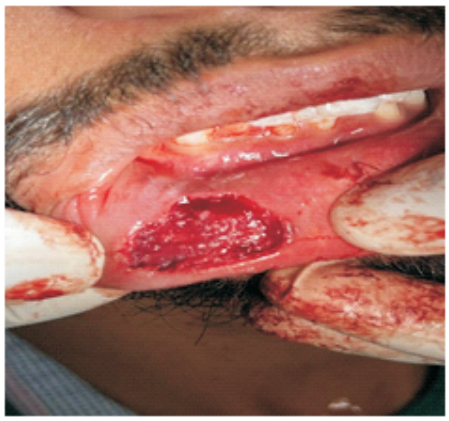

Fig.2 surgical site after excision

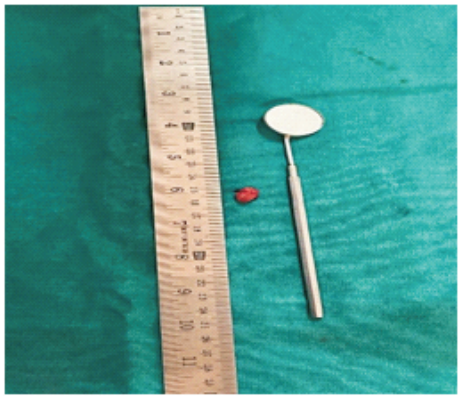

Fig.3 surgical sample after excision

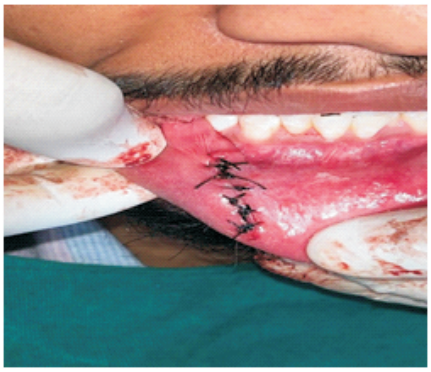

Fig. 4 suturing of the excised area

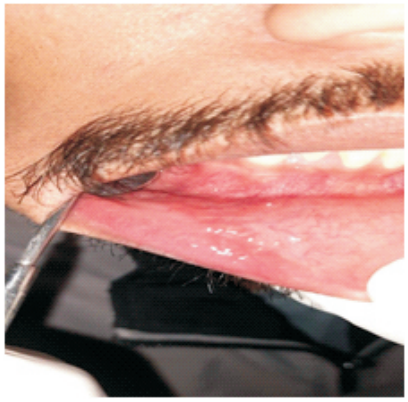

Fig. 5 post operative healing

Discussion:

Lymphatic malformations are benign, hamartomatous tumor like growths of lymphatic vessels. They most likely represent developmental anomalies that arise from sequestrations of lymphatic tissue that do not communicate normally with the rest of the lymphatic system.

Various lesions that take off the clinical appearance of mucocele but had definite histological characteristics of other lesions, like sialolithiasis, canalicular adenoma, collagenoma, and schwannoma[4]. The lesions of lymphangioma may be situated superficially or within deeper tissues.

Depending on size and depth of abnormal lymphatic vessels, lymphangioma is classified in two groups. The superficial is circumscribed angyoma and deeper is cystic hygroma and cavernous lymphangioma. Lymphangioma circumscriptum consist of deep cavernous cisterns within subcutaneous tissue. There can be dilated lymphatics in secondary formation.

Etiopathogenesis of lymphangioma is based on two major theory. First theory explains that the lymphatic system advances from five primitive sacs of venous system. Considering the head and neck, endothelial outpouching spread centrifugally from jugular sac to make the lymphatic

system. Another theory explains that lymphatic system advances from mesenchymal cleft in the venous plexus reticulum and spread toward the jugular sac centripetally[5].

Even though lymphangiomas commonly occur in the head and neck region, lymphangiomas in oral cavity are rare. Intraorally, the dorsum of the tongue is commonly involved site. Clinical examination of the lymphangiomas usually reports a painless, slow growing, soft tissue mass.

There are numerous methods for the treatment of lymphangiomas. When vital structures does not count in, surgical excision is recommended . Procedures such as electrocautery, radiation therapy, cryotherapy, embolization, steroid administration, ligation, laser surgery and sclerotherapy including 25% dextrose, hypertonic saline, ethanol, sodium morrhuate, and doxycycline have been considered to treat lymphangiomas[6]. There is no recurrence of lymphangioma circumscriptum after surgical excision.

Differential diagnosis for lymphangioma includes neurofibromatosis, pyogenic granuloma, hemangioma, amyloidosis, congenital hypothyroidism, granular cell tumor, malignant melanoma, and herpes infection[7,8]. Biopsy and histopathological examination should be made for definitive diagnosis irrespective of clinical features.

Conclusion:

As lymphangioma is rarely seen in oral cavity, it represents a condition that must be recognized. It is recommended to send all lesions for proper examination even if it shows typical clinical characteristic to confirm accurate diagnosis and to avoid future complication by providing adequate treatment.

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