

## A Rare Case of Ulcerous Lobular Capillary Haemangioma on the Lateral Side of the Tongue



### Dental Science

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**Dr. DILIP KUMAR R**

mds, professor, department of oral and maxillofacial surgery Dayananda Sagar College of Dental Sciences

**Dr. MADHUKAR NATEKAR**

Post graduate student, department of oral and maxillofacial surgery, Dayananda Sagar College of Dental Sciences

**Dr. PRITHVIRAJ K**

Post graduate student, department of oral and maxillofacial surgery, Dayananda Sagar College of Dental Sciences.

### ABSTRACT

*Lobular capillary haemangiomas are well known benign lesions that occur on both the cutaneous and mucosal surfaces. In the oral environment they show strong predilection for gingiva and mucosa of the lips, cheeks, gingiva, and palate but rarely develop on the ventral side of the tongue. As they mimic physical characteristics similar to epulis, varicocele, telangiectasia and pyogenic granuloma, the clinical diagnosis becomes a challenge. Differentiation through histological features is a feasible method to arrive at a definitive diagnosis of lobular capillary haemangioma. The objective of this article is to report an unusual case of benign lobular capillary haemangioma on the lateral side of the tongue in a 26 year old male patient.*

### INTRODUCTION:

Haemangiomas are a group of benign vascular lesions that share similar histological features. Haemangiomas are a common occurrence in the head and neck region but relatively rare in the oral mucosa [1]. They can be classified on the basis of histological features into capillary, cavernous and sclerosing variety with fibrosis [1]. Capillary haemangiomas are further divided into two sub-types: Lobular capillary haemangioma (LCH) and Non-Lobular capillary haemangioma (non-LCH). LCH earlier known as pyogenic granuloma is composed of proliferating blood vessels aggregated in a lobular configuration although superficially does not show any granulation tissue or oedema. On the contrary, non-LCH is characterised by higher proliferation of vasculature, resembling granulation tissue. LCH can develop on lips, cheeks, tongue, gingiva, palatal mucosa, jawbone and salivary glands [1-4]. These lesions are commonly identified at a young age and have a greater predilection for females than males (3:1 ratio)[5, 6]. Clinically they appear as small, red or pink, sessile lesions with characteristic ulcerated and friable surface covered by yellow, fibrinous membrane [6, 7]. Haemangiomas mimic other lesions like epulis, varicocele, telangiectasia and pyogenic granuloma in terms of clinical, radiological and histopathological features and therefore require accurate diagnosis to rule out other aggressive lesions. Management generally includes excision, ligation, thermocautery or remission through sclerosing agents. Extensive review of literature did not reveal any cases of ulcerated LCH, thus we report a rare case of a 26 years old male patient with ulcerated LCH on the lateral-ventral border of the tongue.

### CASE REPORT:

A 26-year-old male patient with a chief complaint of ulcerous lesion on the right lateral-ventral border of the tongue was evaluated. The patient had undergone surgical excision of a similar lesion earlier 2 months back at the same location. The patient reported that he had noticed the growth one month after surgical excision was done, which gradually increased to the present size. General physical examination, medical, surgical, and family history were all non-contributory.

Intraoral examination revealed a pale, pinkish, solitary, broad based lesion with a central crater like depression located at right lateral surface of the tongue [Fig 1]. It was

measured 3cm x 2 cm in size and had distinct, well-defined borders. The lesion was non-tender and firm in consistency. Lymph nodes were not palpable.

Differential diagnosis of epithelioid haemangioendothelioma and pyogenic granuloma were considered.

Excisional Biopsy was performed under local anaesthesia and the specimen was sent to a NHA Accredited laboratory at St John's Medical College, St John's Laboratory Services, Bangalore for histological examination. The microscopic examination of the H & E stained section revealed an overlying parakeratinised stratified squamous epithelium with an underlying connective tissue stroma [Fig 2]. Focal ulcerations covered with fibrinopurulent membrane were seen. The sub epithelium showed the presence of numerous small to medium endothelium lined blood capillaries. The characteristic plump endothelial cells were seen proliferating around the lumen and separated by connective tissue septa dividing them into lobules [Fig 3]. On the basis of the histological picture and clinical features, we diagnosed the lesion as Lobular Capillary Haemangioma.

The management of the lesion included complete excision using electrocautery under local anaesthesia [Fig 4]. The lesion has healed uneventfully and showed no evidence of recurrence during follow-up examination after 6 months [Fig 5].

### DISCUSSION:

LCH is a polypoid, non-neoplastic form of capillary haemangioma occurring on mucosal surfaces of the oral cavity but rarely on the ventral side of the tongue as seen in our case. Several studies have attributed the aetiology to trauma, infections, hormones, chronic irritants, medications, defective fillings, gingival inflammation and periodontitis [8]. The mechanism of development is not well established, but Kuo and colleagues in their study have demonstrated the role VEGF and bFGF – angiogenesis enhancers, contribute in the growth of haemangioma. They further observed that vascular morphogenic factors were elevated in LCH of the healthy gingiva [9]. Chen et. al, also suggested that the overexpression of transcription factors like p-ATF2, p-STAT3 and p53 may play a role in the tumorigenesis of vascular lesions [10].

The differential diagnosis of LCH includes lesions such as

bacillary angiomatosis, epulis, telangiectasis, fibroma of the mucosa, pyogenic granuloma, peripheral odontogenic fibroma, Non-Hodgkin's lymphoma, angiosarcoma, squamous cell carcinoma, Kaposi sarcoma and AIDS related complex [5, 11]. Clinically, LCH appear smooth bosselated or lobulated, varying in size – few millimetres to centimetres and usually penetrates into the subcutaneous tissue. Superficial and deep lesions can be differentiated clinically by the colour variance – the former appearing bright red whereas the latter appear purple or blue. Although these lesion are generally painless, yet infection of the ulcerated surface may cause painful complications. These lesions appear to grow from the interdental papilla when they develop in the periodontium and usually spread laterally involving the adjacent teeth [12].

Histologically when observed under lower power magnification, LCH gives a vague lobular appearance. A typical LCH lesion will consist of perpendicularly arranged thin-walled capillaries, lined by a single layer of either flattened or plump endothelial cells with prominent nuclei. Characteristically, they will be surrounded by loose connective tissue filled with reticular fibres, plump fibroblasts and discontinuous layers of pericytes. The capillaries and spaces in between them will be arranged in a lobular configuration. As the haemangiomas mature, the nuclei will decrease in size which results in the endothelial cells assuming a flatter shape and enlargement of the capillary lumina.

Management of LCH is dependent on the age of the patient, size, site and clinical involvement of the lesion. The treatment of choice has been surgical excision for a long time, but other treatment options have been employed by clinicians such oral corticosteroids, sclerosing agents, fibrosing agents, cryosurgery, laser excision, thermocautery, electrocoagulation, injection of interferon  $\alpha$ -2b and embolization [2, 13, 14]. It has been advised that the LCH lesions should be dealt conservatively with non-surgical modalities of treatment before choosing invasive procedures. Incidence of post-operative recurrence, although rare, has been reported in literature [15, 16].

#### CONCLUSION:

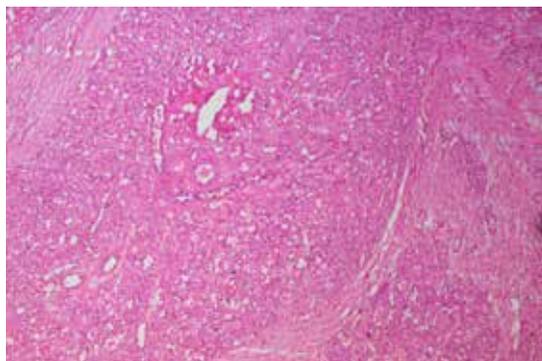
Although, capillary haemangiomas are asymptomatic and non-lethal, yet their appearance in atypical location may require rapid intervention. The case we have presented here is important because of its peculiar and rare location on the lateral - ventral border of the tongue and prominent ulceration. Poor hygiene of the tongue and constant exposure to food particles will increase the susceptibility of the lesion to oral infections. Also, certain neoplastic lesions like Kaposi sarcoma, basal cell carcinoma may appear similar to haemangiomas and an erroneous diagnosis may put life in danger. Thus, early detection and treatment of such lesions are essential to prevent further complications.

#### FIGURES:

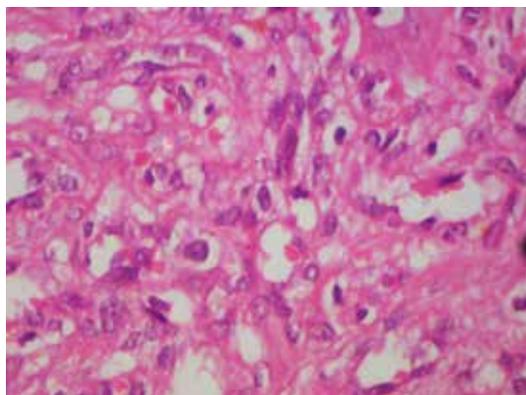
[Fig 1] Gross photograph of the lesion on the right lateral surface of the tongue.



[Fig 2] Microphotograph of the lesion showing parakeratinised stratified squamous epithelium, connective stroma, endothelium lined blood capillaries separated by connective tissue septa dividing them into lobules (H&E x 4)



[Fig 3] Microphotograph of the lesion showing plump endothelial cells were seen proliferating around the lumen. (H&E x 40)



[Fig 4] Gross photograph showing the excised lesion.



[Fig 5] Gross photograph showing the recovery after follow up of 6 months.



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