Case Report

Atypical extragingival occurrence of lobular capillary hemangioma: A report of two cases

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Abstract:
Capillary hemangiomas and pyogenic granuloma (PG) are well-recognized benign entities of the oral cavity which are vascular in origin. PG is said to have a predilection for gingival area, whereas capillary hemangioma involves the lips, cheek, and tongue. They may mimic more serious conditions such as malignancies making the clinical diagnosis quite challenging. There have been cases reported where PG was found on extra-gingival sites such as the palate, lips, and tongue. We present the two cases of lobular capillary hemangioma (LCH) on extragingival sites, namely the palate and buccal mucosa, which manifested as a different lesion clinically and histopathological examination was indicative of LCH. The cases reported here show that relatively common pathologies can present on rare or atypical regions inside the oral cavity, and hence, it is imperative for the clinician to consider such lesions while formulating a diagnosis.

Key words:
Buccal mucosa, capillary hemangioma, extragingival, gingiva, palate, pyogenic granuloma

INTRODUCTION

Hemangiomas are usually benign tumors originating from proliferating blood vessels, categorized into different types such as capillary, cavernous, or central according to their histological appearance.[1] They can present as flat or raised reddish-blue, generally solitary, with three-to-five fold greater incidence in young females.[2] In the head-and-neck region, hemangioma is common, but its manifestation in the oral cavity is quite rare and unusual.

Pyogenic granuloma (PG) is a tumor-like lesion comprising of moderately small-sized proliferating blood capillaries. The term “lobular capillary hemangioma (LCH)” has been used by some authors for PG on the basis of histological features.[3] Here, we intend to present the two cases of PG associated with sites other than the gingiva, which had a clinical appearance different from its typical appearance and were then histopathologically proven to be PG.

CASE REPORT

Case 1
A 57-year-old male patient reported with a complaint of a growth on the hard palate for 4–5 months which was gradually increasing in size with no associated pus discharge. Medical and dental history was noncontributory. Extraorally, the face appeared symmetrical with adequate mouth opening and normal movements of the temporomandibular joint. On intraoral examination, an irregularly shaped solitary reddish pink pedunculated mass was noted on the hard palate 3 cm in diameter. The growth was firm in consistency and covered with superficial slough [Figure 1a].

The patient had earlier visited a private physician where a computed tomography (CT) scan of paranasal sinuses was advised. Hence, no other additional radiographs were taken. Coronal section of the scan showed a fairly well-defined soft-tissue density lesion approximately 1.8 cm × 2.1 cm × 0.8 cm (anteroposterior × mediolateral × superoinferior) arising from the anterior midline aspect of the soft tissue of the hard palate showing early intense homogeneous enhancement and focal thinning with osteolysis (white arrow) of the hard palate on the left side [Figure 1b]. Blood investigations were within the normal limits.

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Provisional diagnosis given was minor salivary gland tumor. Differential diagnosis of PG was given. Excisional biopsy of the lesion was done under local anesthesia [Figure 1c]. Histopathological examination with hematoxylin and eosin staining at ×10 magnification showed capillary sized blood vessels lined by single layer of endothelial lining separated by fibro collagenous septae with lymphocytic infiltrate [Figure 1d]. A follow up after 7 days showed uneventful healing [Figure 1e]. The final diagnosis formulated after histopathological examination, was suggestive of PG LCH [Figure 1f].

**Case 2**
A 58-year-old male patient reported with a complaint of growth on the left inner side of cheek for 6 months which was increasing in size with a history of difficulty in chewing and brushing. There was no history of any pain or tendency of bleeding accompanying the growth. The medical and dental history was nonsignificant and noncontributory. The patient did not give any history of trauma, tobacco chewing, or alcohol consumption.

On intraoral examination, a well-defined, solitary, sessile, round to oval, pinkish growth with distinct borders were noted on the left buccal mucosa in relation to mandibular left first and second molar. The growth was approximately 1.5 cm × 1 cm × 1.5 cm. (Anteroposterior × Mediolateral × Superoinferior). On palpation, the growth was non tender, soft in consistency, and with no tendency for bleeding [Figure 2a]. Since the history and clinical appearance of the growth was not characteristic of a specific lesion, a provisional diagnosis of soft-tissue overgrowth was made, and the patient was advised for an excisional biopsy to reach a conclusive diagnosis. The differential diagnosis included fibroma and peripheral giant cell granuloma.

A white, nonscrapable, homogeneous patch was also noted on the left buccal mucosa posterior to the swelling extending to the buccal vestibule and gingiva in relation to the mandibular left first and second molar region. The patient was again questioned in detail for any adverse habit such as tobacco chewing in any form, but the patient denied any habits such as tobacco chewing or smoking. Based on the clinical appearance of the white patch, a provisional diagnosis of homogeneous leukoplakia on the left buccal mucosa was made. Frictional keratosis was considered as a differential diagnosis.

Preoperative hematological assessment revealed all parameters within the normal limits. After obtaining informed consent from the patient, excision of the growth was done under local anesthesia. Bleeding was controlled by applying pressure with gauze to excision site. The patient was recalled after 1 week, where the healing at the site of excision was uneventful and satisfactory [Figure 2b].

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**Figure 1:** (a) Intraoral presentation of the growth on the hard palate; (b) Coronal section of the computed tomography scan showing focal osteolysis on the left side of the hard palate (white arrow); (c) Excised tissue sent for histopathological evaluation; (d) Histopathological section stained with hematoxylin and eosin showing endothelial-lined blood vessels (black arrows) of various sizes at ×10 magnification; (e) Postoperative picture after 2 weeks; (f) Histopathological report suggestive of lobular capillary hemangioma.

**Figure 2:** (a) Intraoral presentation of the growth on the left buccal mucosa; (b) Postoperative picture after 1 week; (c) Histopathological section stained with hematoxylin and eosin showing fibrous connective tissue with numerous endothelial lined blood vessels (black arrows) of various sizes at ×10 magnification; (d) Histopathological report suggestive of lobular capillary hemangioma.
Histopathological examination with hematoxylin and eosin staining at \( \times10 \) revealed parakeratinized stratified squamous epithelium of varying thickness and areas of ulceration. The fibrous connective tissue showed numerous endothelial lined blood vessels of varied sizes; suggestive of LCH [Figure 2c]. The final diagnosis after histopathological examination by the oral pathologist was suggestive of PG LCH [Figure 2d]. For the treatment of the white lesion, Retinol A cream was given for the topical application two times a day. The patient was kept under regular follow-up and showed good response to treatment.

**DISCUSSION**

Hemangiomas are benign vascular abnormalities which manifest aggregation and proliferation of blood vessels. Congenital hemangiomas usually present as fully developed solitary lesions at birth in any gender. The pattern of growth is usually proportional, symmetrically round, ranging from 5 to 10 cm in diameter mostly involving the extremities. On the other hand, LCH is described as a reactive lesion which develops mostly in the head, face, and neck region, with 6 years as the mean age of presentation. They grow as rapid bright red papules, few millimeters to 2 cm in size. The frequency of intraoral capillary hemangiomas is exceptionally unusual, and they make for only 0.5%–1.0% of all the intraoral neoplasms and are believed to occur in the second and third decades of life, with a male-to-female ratio of 1:3. This was in contrast, as cases presented here were both males of the older age group. Capillary hemangiomas clinically appear as soft, sessile or pedunculated, smooth or lobulated mass, and are asymptomatic and painless unless traumatized. They blanch on pressure and can vary from deep red to purple in appearance.

PGs are considered to be tumor-like entities encountered in the oral cavity with numerous clinical and histological presentations, the gingival areas being the usually affected. On the basis of histological appearance, two different types of PG have been identified. They have been termed as the LCH type and non-LCH type. The LCH type is identified by the evidence of collections of multiplying blood vessels arranged in a lobular architecture. Epivantanos et al. distinguished two types of PG, with LCH PG being a more frequent sessile lesion and non-LCH PG appearing as a pedunculated soft-tissue entity. Histopathology of the LCH is distinct from that of congenital hemangiomas. LCH type of the lesion displays capillary lobules distinctly separated by the thick bands of fibrous tissue, while the congenital hemangiomas show lobular collections of small, thin-walled vessels, and a large, stellate-shaped vessel at the central portion.

The accurate etiopathogenesis of PG yet remains under debate and unclear. Numerous causative agents, namely trauma or injury to a deciduous tooth, chronic irritation, hormonal influences, drugs, gingival inflammation, preexisting vascular lesions, chronic irritation caused by shedding of primary teeth, eruption of permanent teeth, defective restorations in the area of the tumor, food lodgment, periodontal inflammation, trauma inflicted due to a toothbrush, etc., have been termed as etiological factors where patients presented with these findings.

In one of the case presented here, the lesion was present on the central aspect of the hard palate with some underlying bony changes seen in the CT scan which are not a frequent finding. A similar case was reported by Varma et al. in 2013, where LCH on the palate was associated with bone loss of the related teeth in the maxillary arch. In the other case, there was a broad based, painless soft-tissue growth with an irregular surface. The lesion was present on the buccal mucosa with pinkish color and no tendency to bleed. With such a clinical appearance, diagnosis of a soft-tissue growth was given provisionally; however, histopathological examination confirmed a diagnosis of LCH. Furthermore, the patient showed a nonscrappable white patch which could be considered as nontobacco-induced homogeneous leukoplakia. With no history of tobacco consumption as an etiology, the occurrence of such a lesion in nonsmokers may hint toward a genetic predilection.

LCH has a tendency to have variations in its appearance and can mimic other lesions. This may lead to confusion with other clinical categories, namely chronic inflammatory gingival hyperplasia, growth in a socket post extraction (epulis granulomatosa), telangiectasias, or serious conditions such as squamous cell carcinoma or angiosarcoma. As the types of hemangioma are based on histology, therefore, histopathological assessment becomes the most important means for the diagnosis. Treatment of such lesions is dependent on various factors such as the patient’s age, dimensions and extent of the lesion, and its clinical presentation and characteristics. Small lesions can be treated with excision meticulously and successfully as in the presented cases. The literature has reported small capillary hemangiomas being successfully treated by excision or curettage. Regional complications continue to arise, such as bleeding and postoperative ulcerations. In the presented case, bleeding was controlled during the excision of the pathology by applying local pressure, and postoperative healing of the wound was uneventful.

The occurrence of hemangioma within the oral cavity is relatively rare, and it may resemble clinically with PG and other clinical lesions such as malignancy. Hemangiomas are often presented as asymptomatic pathologies, but their location and size may require instantaneous vigilant intervention and careful management. There have been instances where LCH has presented on extra-gingival sites such as the palate, lips, and tongue. Hence, careful step wise evaluation of every case becomes very crucial. Biopsy of such lesions is necessary for establishing definitive diagnosis and proper management and prevention of various complications.

**CONCLUSION**

The cases reported here show that relatively common pathologies can present on unusual sites in the oral cavity, and hence, it is important to consider such lesions while formulating a diagnosis. It also reasserts the significance of histopathology and also affirms that biopsy is the gold standard and an indispensable tool for the diagnosis of any lesion.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms.
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Conflicts of interest
There are no conflicts of interest.

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