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Case report

Lobular capillary hemangioma: A clinical variant

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ABSTRACT

Lobular capillary hemangiomas (LCH) are a group of vascular lesions characterized by capillary-sized vessels arranged in lobules caused due to external stimuli. Clinically, it is a localized granulation tissue overgrowth in reaction to mild irritation. This report presents a unique case of a male child with LCH on a rare site in the oral cavity and its surgical management.

Keywords: Lobular capillary hemangioma; Pyogenic granuloma; Extragingival.

INTRODUCTION

Pyogenic granuloma (PG) is a benign, angiomatous, proliferative lesion of the skin and mucous membrane caused due to a low - grade chronic external stimuli. Many observers equating it with simple granulation tissue have concluded that PG is a non specific capillary reaction that occurs in a setting of repeated trauma or irritation[1]. The most common site intraorally is the gingiva, followed by lips, tongue, palate and rarely the buccal mucosa[2].

Lobular capillary hemangioma (LCH), on the other hand, is considered as a variant of PG with benign polypoid form of capillary hemangioma, primarily occurring on skin and mucous membranes[1]. Hullihen's description[3] in 1844 was most likely the first PG reported in English literature, but the term "pyogenic granuloma" or "granuloma pyogenicum" was introduced by Hartzell in 1904. Although it is a common disease of the skin, it is extremely rare in the gastrointestinal tract, except oral cavity. In the oral cavity, it is often found on the keratinized tissue. There are two kinds of PG, namely lobular capillary hemangioma (LCH type) and non-LCH type, which differ in their histological features[5]. The age range for LCH is 8-82 years with the mean age of 37±12. LCH is most commonly noticed in females ,having a gender predilection of 2:1 when compared to their counterparts[1].This case report presents a male child with LCH of the buccal mucosa which was surgically excised.

Case Report

A healthy 12years-old boy reported to the pediatric dentistry OPD, with complaint of a painless swelling on the left side of the mouth since two to three months. The patient mentioned that the growth was noticed as a small mass on the left side of the buccal mucosa which gradually increased in size to attain the present dimension. Patient also complained about spontaneous bleeding during mastication and brushing.

Intraoral examination revealed that the lesion was located on the left buccal mucosa coinciding with the occlusal plane in relation to the first molars (Figure 1). The lesion was irregular in shape, measuring approximately 1×1 cm and was white to pink in colour, (Figure 2), which could be related to the chronicity of the lesion. The lesion was

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pedunculated with well-defined borders. On palpation, it had a rough, irregular surface and was soft in consistency. It was also noticed that the mesiobuccal and distobuccal cusp tips of the maxillary left molar were sharp.

Based on the clinical features, the lesion was diagnosed as PG and which was surgically excised by completely excising the pedunculated lesion from its base along with 1 mm of the normal surrounding tissue. Excess bleeding during surgical procedure was controlled by electrocautery and later sutures were placed. The excised specimen was subjected to histopathological investigation (Figure 3). The sharp cusp tips of maxillary left molar were rounded followed by topical fluoride application. After a month the excised area showed good healing (Figure 4).

Fig. 1. Intraoral view of the lesion on the left buccal mucosa

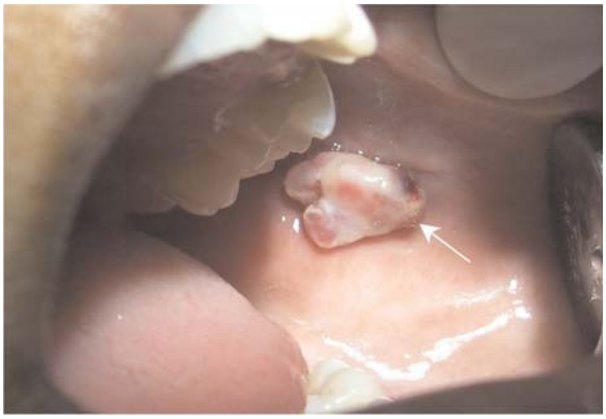


Fig. 2. The excised mass

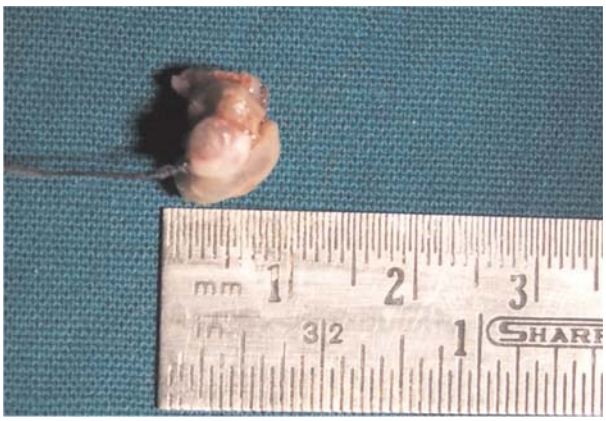


Fig. 3. Histopathological picture (10^X)

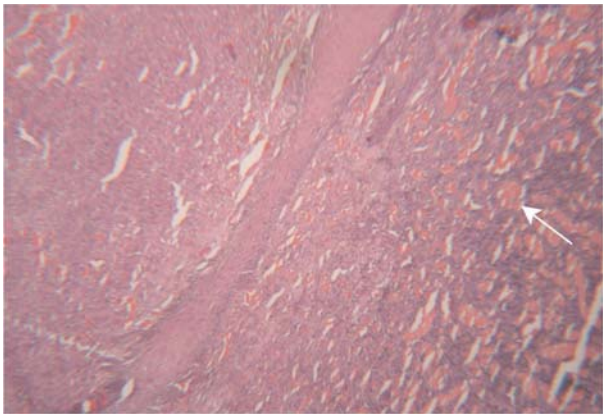
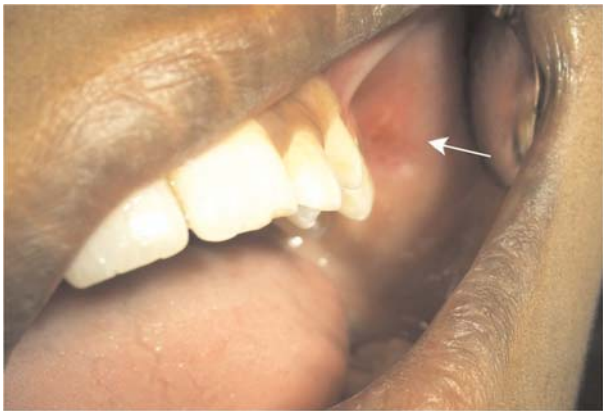


Fig. 4. Intraoral view showing complete healing



DISCUSSION

The term LCH was first suggested by Mills SE *et al* in 1980 which he differentiated it from the non-LCH. LCH can occur both in skin and mucous membrane. LCH may also occur as deeper intradermal, subcutaneous or intravenous masses. The most common cutaneous sites are the trunk, upper extremities and head. Mucosal LCH involve most often the oral mucosa. Oral LCH affects females more than males in a ratio of 1.2:1 to 2.6:1, most commonly in the second and third decades of life[1-6-7-8].Most of the lesions are pedunculated and ulceration is usually present.[7] Mean sizes of the lesions have been reported to be 10–12 mm at the greatest

diamete[6-7]. Some lesions develop rapidly and may be confused clinically with malignancy[1].

The term LCH emphasizes the essential component of the lesion, namely, a circumscribed aggregate of capillaries arranged in one or more lobules. Mills SE et al believed that LCH is a distinct entity due to constancy of capillary lobules in the lesion which allows for its recognition, regardless of site[1]. Gross descriptions of the pathologic specimens are mentioned as a sessile, nodular or polyploidy mass that is gray-brown or, less commonly, pink. Often the central nodule or polyp is ulcerated and covered with a white to yellow exudate. LCH, histologically, is characterized by submucosal vascular proliferation arranged in lobules or clusters composed of central capillaries and smaller ramifying tributaries. Superficial lesions may become ulcerated and secondarily covered with granulation tissue. The microvascular system consists of arterioles, metaarterioles (precapillary sphincter areas), capillaries, pericytic venules and muscular venules. The arterioles have one to three layers of circularly arranged smooth muscle cells. The metaarterioles (precapillary sphincter areas) have a few scattered smooth muscle cells. The pericytic venules have an incomplete and a complete layer of pericytes and no smooth muscle cells. The muscular venules have one or two layers of flattened smooth muscle cells. Thus, regarding the proportion of cell types of the vascular walls, most of the vascular elements in the lobular area resemble more pericapillary (pre- and/or postcapillary) microvascular segments than do capillaries[9-10].

Mills SE et al (1980) verified that LCH does develop in the oral and nasal mucous membranes, although failure to recognize the key feature of this lesion has led to confusion with granulation tissue and other forms of hemangioma in the past. They believed that the superimposed changes in these lesions have resulted in the confusion in literature concerning the basic nature of the lesion[1].

In our case, the histopathologic report revealed discontinued parakeratinised, stratified squamous surface epithelium with fibrocellular connective tissue stroma. There were discontinued areas covered with fibrinopurulent material. Numerous dilated ,well-formed capillaries with extravasated RBCs are seen in the deep connective tissue with numerous endothelial cell proliferation. Minimal chronic inflammatory cell infiltrate with predominant lymphocytes were present. Deep to the muscle fibers, mucous minor salivary glands were evident. This confirmed the diagnosis to be LCH.

Many large LCH, with both elevated and deep components, contain a complete range of lobular appearance; however, between the lobules appearing to connect with them are vascular channels resembling small arteries and veins[1].

As there are numerous capillaries within the LCH, bleeding tendencies from such lesions during surgical excision increases. LCH is a vascular lesion characterized by capillary-sized vessels arranged in lobules which may need certain precautionary measures to avoid excessive bleeding. Excisional surgery is the treatment of choice; some other treatment protocols such as the use of Nd:YAG laser, flash lamp pulsed dye laser, cryosurgery, intralesional injection of ethanol or corticosteroid and sodium tetradecyl sulfate sclerotherapy have been proposed[11].

CONCLUSION

As seen with our case, the treatment carried out for the lesion was surgical excision as it was clinically diagnosed as PG, However, to our surprise, histopathologic report confirmed the diagnosis to be LCH. The histologic characteristic of LCH adds to the increased likelihood for bleeding which can be of concern to the clinician. It becomes mandatory for practitioners to correctly manage any such lesions, as the surgical intervention involves invasive procedures and only the histopathologic report can give a confirmatory diagnosis. Thus, this case report adds a dilemma to these lesions, where a previous

preliminary investigation becomes necessary before undergoing any invasive procedure.

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