



Case Report

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Bilateral lobular capillary- The aberrant twins

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Abstract

Lobular capillary haemangioma is a common tumor-like growth of the oral cavity or skin that is considered to be non-neoplastic in nature. Clinically it may appear alarming as a rapidly growing exophytic lesion. Within the oral cavity it shows a striking predilection for the gingiva where it may present unilaterally or bilaterally. Here we present a novel exposition of bilateral lobular capillary haemangioma in a male patient during early adulthood.

Keywords: Lobular Capillary Haemangioma, Bilateral, Gingiva.

INTRODUCTION

Lobular capillary haemangioma (LCH) or Pyogenic Granuloma (PG) as it is commonly called, was first reported in the English literature, by Hullihen in 1844^[1]. Way back in 1897, this lesion was initially termed as "Botryomycosis Hominis", by Poncet and Dor who were the first to describe the lesion in humans. Later in 1904, Hartzell introduced the term "pyogenic granuloma" or "granuloma pyogenicum". The term pyogenic granuloma, is actually said to be a double misnomer as these lesions neither produce pus nor present as a granulomatous lesion. In reality, pyogenic granuloma is a high strung proliferation of a vascular type of connective tissue which emerges as a result of some minor trauma to the tissues^[2].

Pyogenic granuloma is claimed to be a reactive hyperplastic vascular lesion occurring on the skin and mucosal surfaces of the proximal aero-digestive tracts. In the oral cavity it arises most frequently on the gingiva, followed by the lips, tongue and the buccal mucosa^[3]. They show a wide range of occurrence ranging from 4.5–93 years and a slight female predilection^[4]. Clinically, pyogenic granulomas appear as smooth or lobulated, exophytic lesions, which are reddish purple in color, often presenting with a pedunculated base and are usually hemorrhagic.

Histologically PGs are of two types, namely lobular capillary haemangioma (LCH) type and non-LCH type^[5]. In this article, we present an uncommon case of lobular capillary haemangioma (LCH) type of pyogenic granuloma, which manifests itself on both sides of the mandibular lingual gingiva, during the phase of early adulthood in a male patient.

CASE REPORT

A twenty seven year old male patient reported to the private dental clinic with a chief complaint of swelling on the right and left lower gums. The swelling was first noticed by the patient two years back, and the swelling over the right side had progressively increased in size. The patient also experienced difficulty in munching food and pronouncing certain words, due to the interference caused by the swelling. On inspection, the swelling was localized to the lingual gingival margin of 34, 35 on the left side and 44, 45 on the right side (Figure 1). The masses were bluish red in color. On palpation, the masses were soft, fluctuant and had a smooth texture. Lymph nodes were non palpable. There was no associated pain, tenderness

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or induration. Intraoral periapical radiographs taken in relation to 34,35 and 44,45 revealed no foci of calcification. Neither bone or tooth resorption nor any other significant changes were evident in the orthopantomogram. Clinical differential diagnosis of pyogenic granuloma, peripheral giant cell granuloma and localized gingival hyperplasia was given. Surgical excision of the lesion was performed, beginning with local anesthesia to the lingual mucosa, followed by a crevicular incision at the lingual gingival margin. The mucoperiosteal flap was then reflected and the lesion was excised completely. The excised lesion was sent for histopathological evaluation.



Figure 1: Clinical presentation of pyogenic granuloma on lower right and left gingiva

Histopathological examination of the hematoxylin and eosin stained sections from the submitted specimen showed presence of epithelium and connective tissue. The epithelium was stratified squamous parakeratinized in type and was of varying thickness. The underlying connective tissue comprised of loosely arranged bands of collagen fibers, numerous blood vessels with endothelial proliferation, along with chronic inflammatory cell infiltrate. Just adjacent to the collagen bands, numerous extravasated RBCs along with endothelial cells were present in the core of the connective tissue (Figure 2a, 2b). This confirmed the diagnosis of lobular capillary haemangioma.

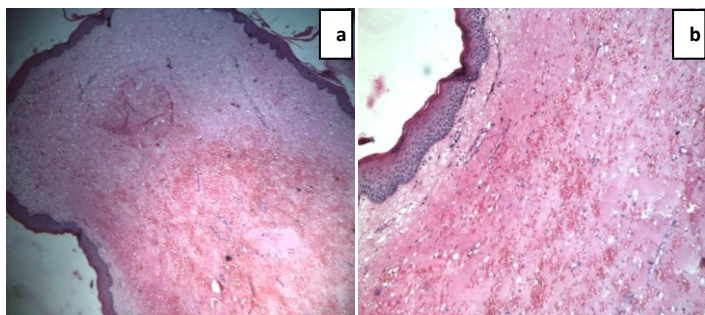


Figure 2: a,b- Photomicrograph showing central core of extravasated RBCs and numerous endothelial cells lined by stratified squamous epithelium (H&E stain, 100x magnifications & 40x magnifications)

DISCUSSION

Pyogenic granuloma is a fast-growing reactive lesion that shows proliferation of endothelial cells that can arise in response to various stimuli such as a chronic low grade local irritation, traumatic injury or hormonal factors. This tissue response harps on the well-known biological principle that any irritant administered to living tissue, may act either as a stimulant or as a pernicious agent, or as both. It is said that all cells release a stimulating agent of which sulfhydryl groups is one of the most important. In the formation of pyogenic granuloma, the destruction of fixed tissue cells is slight, whereas the vascular endothelial stimulation lingers and exerts its impact over a long run^[2].

PGs can present anywhere in the body and are common on the fingers and the toes around the nail beds. They account for 1.85 to 7% of all the oral biopsy findings. In the oral cavity they frequently occur on the gingiva, where they are included as part of the differential diagnosis of an epulis^[6].

Although the etiopathogenesis of the pyogenic granuloma is not clear, trauma is considered to be the most common etiological factor. But in our case there was no history of trauma. Hormonal influences have been frequently reported and one such entity is the pregnancy tumor that appears to arise more readily in the hormonally primed gingiva and usually regresses post-parturition. The hormonal imbalance, when coexists with bacterial plaque and gingival inflammation, peaks the tissue response to infection. Increased levels of estrogen during pregnancy contributes to increase in Vascular Endothelial Growth Factor (VEGF) levels. It has been proposed that Angiopoietin-2 (Ang-2), in the absence of VEGF, causes blood vessels to regress. Three cases of oral pyogenic granuloma in bone marrow transplant cases who were taking cyclosporins were reported by Lee *et al.* elucidating the role of cyclosporins in the etiology of pyogenic granuloma. Other etiological factors include origin of these lesions from sites of extraction, usually in response to an irritant that has entrapped into the socket such as calculus, food debris, tooth fragments or bone spicules^[7].

Pyogenic granuloma due to its high vascularity, bleeds spontaneously following minor trauma whereas the older lesions appear to be more collagenized and pink. In our case, the mass appeared collagenized indicating its chronic presence.

A pyogenic granuloma is a lesion which is generally thought to be recognized clinically. However, PGs are clinically inseparable from peripheral giant cell proliferations, haemangiomas and Kaposi's sarcomas. Peripheral giant cell granuloma can be histologically differentiated owing to the presence of multinucleated giant cells and absence of an infectious source. A peripheral ossifying fibroma also resembles a pyogenic granuloma, but the latter is usually more firm and not as red. Histologically, proliferation of blood vessels is seen in both PG and haemangioma, but the later shows endothelial cell proliferation without acute inflammatory cell infiltrate. Presence of proliferating dysplastic spindle cells, vascular clefts and intracellular hyaline bodies are characteristic of Kaposi's sarcoma, none of which are evident in pyogenic granuloma. The clinical resemblance of PG to a primary or metastatic malignancy is of greatest importance. This is because, malignancies such as squamous cell carcinoma, fibrosarcoma, leukemia, non-Hodgkin lymphoma, and metastatic foci from the breast, lung, kidney, prostate, etc, have been known to seed bone and proliferate a soft tissue mass, that mimics a pyogenic granuloma^[8].

A periapical or panoramic radiograph is advised to rule out the possibility of malignancy which is evident from a bony destruction^[6]. Generally there is no radiographic evidence of bone involvement in PG, but in some cases superficial erosion of bone can be seen. Still there are a few reported cases of extensive bone loss associated with this lesion. In a study done by Bhrugeth, Singh and Shenoy, cases of oral pyogenic granuloma with alveolar bone loss have been reported. Martins-Filho, reported a case of aggressive pyogenic granuloma with extensive alveolar bone loss that resembled a Malignant Tumor^[9]. This emphasizes the need for histopathological evaluation, for any clinically or radiographically suspected lesion of pyogenic granuloma, to rule out the possibility of malignancy.

Excisional biopsy is the treatment of choice for oral pyogenic granuloma. The excision should extend down to the periosteum, often to eliminate any source of irritation^[7]. Nd: YAG laser for excision, cryosurgery, injection of absolute ethanol and intra lesion corticosteroid therapy are some of the other treatment protocols reported. Due to the high recurrence rate and the fact that the lesions may resolve spontaneously post parturition, pregnancy tumors are not

excised unless they impose significant functional or esthetic discomforts^[10]. During surgical treatment of pregnancy tumors, if the bleeding is uncontrollable, management should range from supportive therapy such as desiccation of bleeders; local, firm compression to blood transfusion, and also medications to accelerate fetal lung maturity or even termination of pregnancy to save the patient's life^[7].

Recurrence rate ranges upto 16% and in such cases re-excision is necessary. Recurrence is usually believed to result from failure to remove etiological factors or re-injury of the area. An incomplete excision may also lead to recurrence. Of all the oral mucosal sites, gingival cases show a high recurrence. High recurrence rates were noticed in the second third and fourth decades of life in descending order and also in lesions excised during pregnancy.

CONCLUSION

The aberrant bilateral occurrence of LCH, on the less frequently prone lingual portion of the gingiva, in a male patient where hormonal imbalance plays a less vital role, presented in this article, brings to light the versatility of PGs and the significance of its proper diagnosis. This article also highlights the importance of histopathological evaluation to distinguish pyogenic granuloma from the other possible malignancies it mimics. Treatment should be aimed at both, complete removal of the soft tissue lesion and most importantly eliminating the source of the inflammation based on the biological behavior of the lesion. Maintenance of oral hygiene with regular follow up sessions will prevent relapse and increase the quality of life of the patient.

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