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REVIEW ARTICLE

Gingival plasma cell granuloma: An enigmatic inflammatory pseudo-tumor with literature review

Gaurav Pandav[1], Harjit Kaur[1], Sanjeev Jain[1], Sakshi Pandav[2]

Department of Periodontology & Oral Implantology, Guru Nanak Dev Dental College & Research Institute, Sunam, Punjab India, [2]Post Graduate Institute of Medical Sciences, Rohtak, Haryana, India

Correspondence
Dr. Gaurav Pandav, Department of Periodontology & Oral Implantology, Guru Nanak Dev Dental College & Research Institute, Sunam, Punjab India. Phone: +91-9041417176, E-mail: gauravpandav18@gmail.com

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Abstract

Plasma cell granulomas (PCGs) also known as inflammatory pseudotumors are tumor like proliferations consisting chiefly of plasma cells, found most commonly in lungs. They can occur in areas such as orbit, head, neck and liver, but rarely they occur in the oral cavity. We here report an exceedingly rare case of gingival PCG in a 52-year-old woman who presented with unusual maxillary gingival overgrowth by excisional biopsy. Histopathological examination of the lesion showed a dense mass consisting of thin keratinized epithelium with proliferating rete ridges. The connective tissue showed hypercellularity. A dense inflammatory infiltrate consisting primarily of plasma cells and lymphocytes was seen in the connective tissue. Focal budding capillaries were seen in the stroma confirming the diagnosis of PCG.

Keywords: Gingival neoplasm, inflammatory pseudotumor, plasma cells, plasma cell granuloma

Introduction

Polyclonal and monoclonal proliferations of the gingiva have been reported under a variety of names. The monoclonal varieties of plasmacytoma and multiple myeloma are well-defined and reorganized as neoplasms. However, the polyclonal plasmacytic proliferations are often signed out descriptively. These polyclonal reactive lesions are the result of various stimuli, which often must be addressed to resolve the plasmacytic proliferation. The term plasma cell granuloma (PCG) has been used primarily to describe a fical mass effect caused by polyclonal plasmacytic infiltrates.[1] In 1968, Bhaskar, Levin and Firch first reported the cases of gingival PCG. Although PCG occurs most commonly in lungs, other organs may be involved. In the head and neck region, areas which are most commonly involved are the orbit and paranasal sinuses, but they have also been reported in larynx, pterygomaxillary space, tonsils, ears, tongue, lip, oral mucosa, periodontal tissues and rarely gingiva.

Literature review shows that gingival PCG's are rare and very few case reports of gingival PCG have been observed. The reported cases were in the wide range of 19 months to 63 years, but most of the cases are observed in fourth and fifth decades of life.[2] These lesions have no sex predilection.[3]

Clinically gingival PCG appears as a nodular, polypoidal mass with a smooth surface. It does not produce significant systemic symptoms. Routine laboratory tests are normal and microbiological culture is usually negative; but a certain subset of inflammatory pseudotumor occurs secondary to infection. E.g. Mycobacterial infection is found to be associated with spindle cell tumor, whereas splenic and nodal pseudotumors are caused by Epstein-Barr virus. Pulmonary pseudotumor are most commonly associated with nocardia and mycoplasma infection, whereas infection with actinomycetes results in hepatic pseudotumors.[4] Other organisms associated with it are *Mycobacterium avium*-intracellular complex, *Corynebacterium equi*, *Escherichia coli*, *Klebsiella*, *Bacillus sphaericus*, *Pseudomonas*, *Helicobacter pylori* and *Coxiella burnetti*.[5]

Radiographically, infiltrative margins are seen in some oral lesions giving them an appearance of malignant tumor. Histopathological examination of these lesions is must to decide the exact nature of these lesions and to rule out other potential

plasma cell dyscrasias and neoplasms, including multiple myeloma.[2,6]

The exact incidence of PCG is unclear. The lesion's etiology, biologic behavior and most appropriate treatment are unclear and very little is known about the prognosis. It may arise due to periodontitis, periradicular inflammation or idiopathic antigenic cue.[7] In some cases, it results from inflammation following minor trauma or surgery or found to be associated with malignancy.[8-10] An autoimmune mechanism has also been implicated. It is also associated with alteration of blood flow imposing congestive vasodilatation.[11] The most common treatment for PCG is complete resection; however in some cases complete resection is not possible.[12]

Case Report

A 52-year-old woman was referred to the Periodontology Department for the evaluation of a painless mass in the left upper back tooth region since 2 years, which had gradually increased in size. There was no history of associated symptoms such as pain, parasthesia/numbness but the patient had a history of occasional bleeding on provocation. There was no history of trauma/surgery in the past.

The patient was hypertensive since 6 years and was taking occasional medication for the same. Extra-oral examination revealed no abnormality. Intra-oral examination revealed a well-defined oval shaped gingival growth bright red in color ranging in 1.2 cm × 0.8 cm in size in relation to 26, 27 [Figure 1]. The color of the overlying mucosa was reddish with white areas interspersed in between. On palpation, growth was soft in consistency, mildly tender, non- compressible, non-fluctuant, non-reducible, pedunculated and appeared to originate from the interdental papillary 26,27. The oral hygiene status of the patient was very poor with no associated habits. Intraoral periapicalradiograph of the region showed horizontal bone loss [Figure 2]. Based on the above clinical findings a differential diagnosis of irritational fibroma, pyogenic granuloma, PCG was considered.

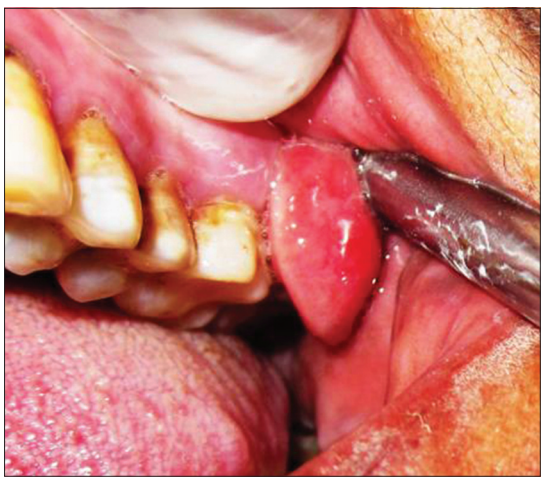

Figure 1: Growth in the interdental papillary region between 26, 27

After a routine blood examination, a thorough oral prophylaxis was done and under local anesthesia excisional biopsy was done using both scalpel and electrocautery device under antibiotic coverage

Extraction of 27 was done along with the excision of the growth [Figure 3]. Thorough curettage of the adjacent periodontal ligament and periosteum was carried out. After controlling bleeding, periodontal dressing was applied, the excised tissue was sent for histopathological examination. The recovery was uneventful [Figure 4].

Histopathological examination

The histopathological examination revealed a mass composed of thin parakeratinized epithelium with proliferating rete ridges. The epithelium showed no dysplastic features. The connective tissue was hypercellular and showed dense inflammatory infiltrate primarily consisting of plasma cells and lymphocytes. Plasma cells were found in the form of sheets and showed no atypia. Focal budding capillaries were also seen in the stroma. The overall features were suggestive of PCG [Figure 5].

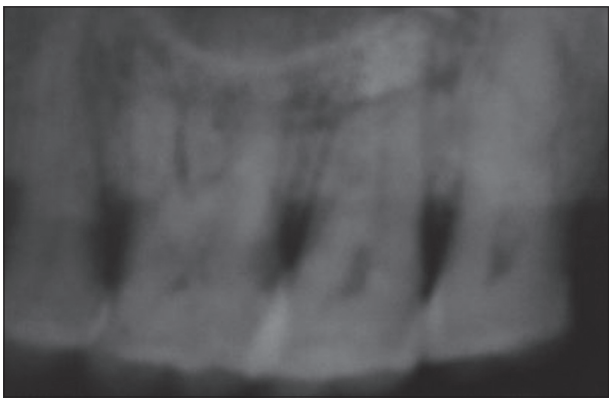

Figure 2: Pre-operative intraoral periapicalradiograph showing horizotal bone loss

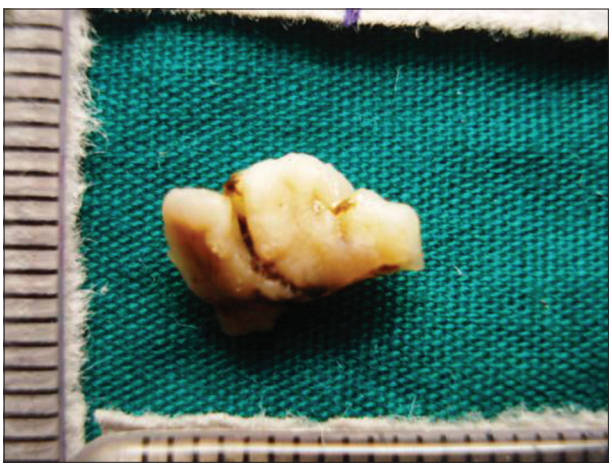

Figure 3: Excised growth and extracted 27

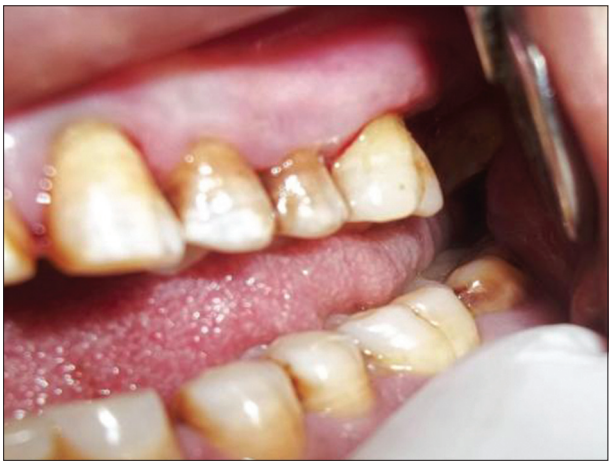

Figure 4: Healing at 2 weeks

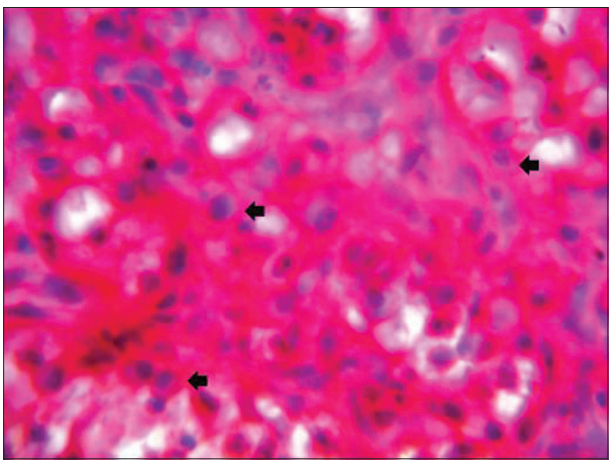

Figure 5: Tissue section stained with Hematoxylin and Eosin, viewed at 40× magnification. The connective tissue is hypercellular and shows dense inflammatory infiltrate primarily composed of plasma cells and lymphocytes. Plasma cells are found in the form of sheets showing no atypia. Focal budding capillaries are also seen in the stroma

Discussion

PCGs are uncommon non-neoplastic lesions, usually pulmonary in location, which are found even more infrequently intraorally.[13] It consists of the proliferation of inflammatory cells, with a predominance of plasma cells, in a fibrovascular background. Its exact incidence is unclear because of numerous different terms by which it is called (i.e. inflammatory myofibroblastic tumor, inflammatory psuedotumor, inflammatory myofibroblastic tumor, inflammatory myofibrohistocytic proliferation and xanthomatous pseudo tumor).[14] It has also been described the names of atypical gingivostomatitis, idiopathic gingivostomatitis and allergic gingivostomatitis.[7]

The phenomenon of plasma cell infiltrate was first described by Zoon in 1952 when he described balaritis plasma cellularis. Since then plasma cell infiltrates been found on the vulva, buccal mucosa, palate, nasal aperture, gingiva, lips, tongue, epiglottis, larynx and other orofacial surfaces.[14]

The origin of development of PCG is unknown, although some speculate either an altered Ag-Ab reaction or an alteration of blood flow imposing congestive vasodilatation (angioplasmocellular hyperplasia).[11,15] Some authors consider PCG to be a purely inflammatory lesion related to infection or an auto-immune disorder, although cases of bacterial, viral, fungal infection have been reported.[16-19]

Histologically, it is characterized by fascicles of spindle mesenchymal cells admixed with chronic inflammatory cells, predominantly plasma cells. It has various components like fibroblast, myofibroblasts, inflammatory cells (plasma cells, lymphocytes, histocytes, mast cells and eosinophills). The stroma is collagenous and/or myxoid. All these components are arranged in varying proportions thus creating a marked histological diversity. The histological diversity had led to conflicting opinion regarding the inflammatory or neoplastic nature of this lesion. The finding of human herpes-8 virus DNA sequences and over expression of human interlukin-6 (IL-6) and cyclin D1 has been recently reported. Kim *et al.* (2002) suggested that IL-6 and phospholipase c-γ1 may induce heavy plasma cell infiltration in cyclosporine induced gingival growth.[20]

PCG of the oral cavity are seen primarily on the periodontal tissue. These lesions are often single. Mesial and distal gingiva are equally involved. Bone loss, which was seen in our case, may occur. These lesions have no sex prediliction and may occur at any age.[21]

The treatment of choice for PCG is a complete resection and removal of underlying inciting agent (e.g. Periodontitis, non-vital tooth, foreign material etc.) radiotherapy and/or steroid therapy have sometimes been successfully used to treat patients with non-resectable lesion but discordant results have been reported. In this case also the complete excision of the lesion was done, and the recovery was uneventful. Even after 2 years post-operative no recurrence of the lesion is reported.

With respect to prognosis, PCG seem to be generally, benign non-recurring condition. Nevertheless, local aggressiveness and reoccurrences may complicate the outcome of the disease.[14]

Conclusion

PCG is a lesion that is diagnosed primarily based on histological findings of marked submucosal plasma-cell infiltrate, after condition such as infection and plasmacytoma have been eliminated. This case lays emphasis on the need to submit any and all excised tissue for microscopic examination regardless of clinical impression and/or perceived surgical success. Only by such practice can rare lesions of this type be documented and studied.

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