Plasma Cell Granuloma of Gingiva- A Rare Case Report

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Introduction
Bahadori and Leibow in the year 1973 studied a pulmonary lesion and termed it as “Plasma cell granuloma”. Plasma cell granuloma (PCG) is a rare lesion primarily showing polyclonal plasma cells. Various synonyms of plasma cell granuloma add confusion to the disease terminology, such as atypical gingivostomatitis, idiopathic gingivostomatitis, inflammatory pseudotumor, inflammatory myofibroblastic tumor, histiocytoma, and fibroxanthoma, to mention a few¹²³. It is a rare non-neoplastic lesion and etiopathogenesis of this disease remains unclear. It affects primarily lungs along with some extrapulmonary sites such as the central nervous system, endocrine gland, paranasal sinuses, larynx, lips, tonsils, oral cavity, ears, and orbit. Plasma cell granuloma of gingiva is rarely seen and very few case reports have discussed the same in literature. This case report is a rare case of Plasma cell granuloma of gingiva and highlights the clinical as well as histological findings.²

Case Report
A 13 year old female patient reported with a chief complaint of poor esthetics due to swollen gums, foul smell and bleeding gums involving all her teeth for four months. The enlargement was first noticed four months back and was not associated with pain or bleeding. The size of the gums increased gradually to the present size [Fig 01]. Foul smell and bleeding while brushing was noticed two months back. Patient’s medical history was not relevant and there was no history of drug intake, fever, anorexia, weight loss, seizures, hearing loss or any other systemic illness. She had undergone extraction of lower left back tooth at a government hospital, one year back due to acute pain and the healing was uneventful. Familial and personal history was non-contributory. General physical examination showed no signs or symptoms suggestive of any systemic illness. The intra oral examination revealed that the patient had 28 teeth with two (65 & 74) retained deciduous teeth and mesiodens.
Gingiva was highly inflamed, extending from free gingival margin to the MGJ in both the maxillary and mandibular anterior sextant. The entire tissue was a bright fiery red, moderately thick and edematous with profuse bleeding on gentle manipulation. The gingiva on the posterior region of both arches was relatively less edematous. Some teeth were barely visible in the anterior region as the enlarged gingiva covered till the occlusal third of the teeth. Grade I mobility was observed in upper left first premolar. The gingival examination also revealed deep pseudopockets in all her teeth. Heavy plaque deposition was noticed around exposed teeth in maxillary and mandibular anterior sextants. Nikolsky’s sign was negative with no blister formation. Orthopantomograph molar teeth and minimal horizontal pattern of generalized bone loss. Blood and urine examinations were normal. Blood estradiol and progesterone levels were within normal limits and in the luteal phase. Based upon the history and clinical findings patient was provisionally diagnosed as a case of inflammatory gingival overgrowth. Phase I therapy was carried out following patient education and motivation. The patient was on maintenance recall for three weeks. The probing pocket depth reduced in fourth following phase I therapy. Surgical therapy was planned to establish normal gingival contour. Hence, external bevel gingivectomy was planned to maintain the gingival size, shape, and contour of gingiva for the region from 11 to 17 and 21 to 25. Internal bevel gingivectomy was planned to remove the excess fibrous tissues while maintaining the contour of gingival tissues for the region 33 to 37 and 45 to 33. Internal bevel incisions were made and full thickness periodontal flap was raised. Thorough debridement with root planning was carried out. The flap was repositioned and sutured [Fig 02]. All the excised tissues were sent for histopathological examination. The periodontal pack was placed following gingivectomy. Analgesics and antibiotics were prescribed for five days. Appropriate instructions for wound care were given. The patient was recalled after one week for suture removal. Clinically, there was an excellent resolution of inflammation and uneventful. The patient was advised to maintain strict oral hygiene and was recalled for review after one month, three months and six months.[Fig 03] Currently, the patient is asymptomatic and on regular recalls.

Histopathological examination comprising of Hematoxylin and Eosin stained sections showed stratified squamous lining epithelium exhibiting marked acanthosis with minimal keratinization along with anastomosing rete pegs. The subepithelial tissue was edematous and showed lympho-mononuclear inflammatory infiltrate. Higher magnification of sections revealed predominant plasma cell infiltrate having the characteristic eccentrically placed nucleus. Immunohistochemistry with CD 138 was positive and plasma cells did not show any light chain restriction with immunohistochemical staining for kappa and Lambda chains. Sections did not show any abnormal or malignant cells. Sections were negative for fungi with PAS & Grocott special stains. Based on the histopathological examination and immunohistochemistry study final diagnosis of Plasma cell granuloma of gingiva was given [Fig 04].

**Discussion**

Patients of PCG may come with a complaint of unesthetic appearance, burning sensation, bleeding from the mouth and diffuse/ single isolated gingival enlargement with erythema with or without desquamation. Differential diagnosis of this condition is very important [4].

Bhaskar, Levin, and Firch first reported this pathologic entity on the gingival tissue and
only very few case reports have been documented since then. The pathogenesis of this lesion remains uncertain. Complete dietary history with records should be taken (Foods, dentrifices, mouthwash, tobacco, alcohol, chewing gum, candy, medication) to rule out possible allergen in an attempt to determine the cause\textsuperscript{[3,5]}.

It has been postulated that the presence of a large number of plasma cells may represent an altered antigen-antibody reaction of the host or an alteration of blood flow imposing congestive vasodilation. A parasitic etiology has also been suggested for this type of lesion\textsuperscript{[6]}. Plasma cell granulomas of the oral cavity are seen primarily in the periodontal tissues. These lesions are often single. Maxillary and mandibular gingiva is equally involved. Bone loss may also occur. These lesions have no sex predilection and may occur at any age\textsuperscript{[7]}.

Pathological changes in this condition are clinically similar to those of pemphigus, pemphigoid, desquamative gingivitis, lichenoid or allergic reactions, and leukemic infiltrate, the latter needing to be differentiated through hematologic and serologic testing. A negative Nikolsky’s sign, a thorough medical history, and age of the patient helps excluding these clinical entities\textsuperscript{[8]}.

Plasma cell granulomas are benign inflammatory lesions for which biopsy and histopathologic/immunologic studies must be performed to rule out potential plasma cell dyscrasias and neoplasms, including multiple myeloma\textsuperscript{[9]}.

Tumors that are mainly composed of plasma cells may be multiple myeloma, solitary myeloma, soft tissue myeloma (plasmacytoma), or plasma cell granuloma. Multiple myeloma and solitary myeloma are tumors of the bone, whereas, plasmacytoma and plasma cell granuloma are soft tissue tumors. Differentiating the type of soft tissue tumor is mandatory, as plasma cell granuloma may be benign, but plasmacytoma may show early stages of multiple myeloma\textsuperscript{[10]}.

Histologically, the lesions consist of a proliferation of mature plasma cells and reticulendothelial cells supported by a stroma of granulation tissue, with varying degrees of myxoid change or collagenization. Diffuse and massive infiltration of plasma cells into subepithelial gingival tissues is also seen in such cases. Angioinvasion within the lesion is observed in 50% of cases\textsuperscript{[11]}. Electron microscopy confirms the benign nature of the plasma cells with fibroblast and myofibroblastic proliferation admixed with that of other inflammatory cells. Serological examination needs to be carried out to rule out lesions such as lupus erythematosus/ lichen planus and benign mucous membrane pemphigoid. Hematological examination needs to be done to rule out leukemia\textsuperscript{[12]}.

Fungal infection can mimic mucous membrane plasmacytosis but is usually distinguished by the absence of fungal hyphae upon microscopic examination, a negative result on maceration with potassium hydroxide, no growth on culture on Saborauds Dextrose Agar and a lack of response to treatment with nystatin\textsuperscript{[11]}. Biological behavior or treatment plan for such a case is unclear. Little is known about the prognosis. The most common treatment is complete resection. But, at times total surgical excision is not possible. In those cases, symptomatic treatment may be offered. Mouthrinses such as Betamethasone, Fluocinonide gel (0.05%), Topical Triamcinolone (0.1%), Topical Fusidic acid (2%) have been tried to provide symptomatic treatment\textsuperscript{[12]}.

**Conclusion**

The clinical picture of Plasma cell granuloma of gingiva mostly points towards a
nonspecific inflammatory component with unknown etiology. These lesions are locally aggressive but rarely recur. Plasma cell granuloma of gingiva is diagnosed based on histopathological findings showing submucosal plasma cell infiltrate as the clinical picture. This lesion also emphasizes the need for submission of all excised tissue for histopathological examination.

References