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Case Report

Plasma cell granuloma of lower lip: A rare case entity

Mubeen[1], Manpreet Kaur*[1], Kondajji Ramchandra Vijyalakshmi[1] and Ashwini B.R[2]

[1]*Department of Oral Medicine & Radiology, Government Dental College & Research Institute, Bangalore, Karnataka, India*
[2]*Department of Pathology, Bangalore Medical College & Research Institute, Bangalore, Karnataka, India*

***Correspondence Info:**
Dr. Manpreet Kaur
Post Gradute Student
Department Of Oral Medicine & Radiology
Government Dental College & Research Institute,
Bangalore, Karnataka, India
E-mail: manpreetkaur_behniwal589@yahoo.in

Abstract

Plasma cell granuloma is a benign non-neoplastic lesion which is described under the pseudo-inflammatory tumor category. The etiologies of this lesion are complex and still remain unclear. It occurs primarily in lungs, but also been reported in other extrapulmonary sites. In oral cavity, plasma cell granuloma is exceedingly rare and present as a rapidly growing mass which show aggressive behavior and mimic a malignant tumor, posing a diagnostic dilemma. Histologically, it is composed of polyclonal plasma cells with variable components of fibroblast and myofibroblast, owing to microscopic diversity and thus inconsistent nomenclature. Simple excision plasma cell granuloma is curative with no evidence of recurrence.

This is a case report of plasma cell granuloma of lower lip in a 62 years old female patient which was initially diagnosed as carcinoma of lip because of its clinical ulceroproliferative form. However, detailed radiographic and histological investigation diagnosed it to be plasma cell granuloma of lower lip.

Keywords: inflammatory pseudotumor, labial mucosa, lip, plasma cell granuloma.

1. Introduction

Plasma cell granuloma also known as inflammatory pseudotumor, inflammatory myofibroblastic tumor, and myofibrohistiocytic proliferation, is a rare non-neoplastic, reactive, tumor like proliferation composed chiefly by plasmacytic infiltration.[1] The etiology of this lesion is still unknown. However, various hypothesis have suggested that the pathogenesis has infectious, autoimmune and vascular origin.In some cases, it is thought to result from inflammation following minor trauma or surgery, or to be associated with other malignancy.[2]

The term "plasma cell granuloma" was coined by Bahadori and Liebow in 1973 to describe a pseudotumor lesion of lung.[3] This rare lesion is commonly seen in the lungs and some other anatomic locations such as brain, kidney, stomach, heart. In the head and neck region it has been reported on the oral cavity, temporal bone, tonsil, sub-mandibular region, paranasal sinuses.[4]

The first case of plasma cell granuloma in oral cavity was reported by Bhaskar et al, involving gingiva, subsequently case have been reported involving gingiva and periodontal tissue, tongue and buccal mucosa.[5] Intraoral plasma cell granuloma occurs in the age range of 19th months to 63rd years with slight female predominance.[4]

Clinically, plasma cell granulomas is a great mimicker and simulate malignant neoplastic process due to their infiltrating and destructive nature.[6] The distinction between plasma cell granuloma and malignant tumor is of paramount clinical importance to avoid redundant radical surgery.

With respect to prognosis, plasma cell granuloma is generally benign, non-recurring condition; however, local aggressiveness and recurrences may complicate the outcome of the disease.[7] We hereby report a case of plasma cell granuloma in lower lip clinically mimicking a malignant tumor which posed a diagnostic dilemma.

2. Case Report

A 62 years old female patient, visited department of Oral Medicine & Radiology with growth on lower lip since 3 months (Figure 1). Patient noticed pea nut sized growth in lower lip which gradually increased in size and was associated with dull persistent pain and thick purulent foul smelling exudation from the growth. Patient also experienced discomfort while talking and chewing food. Her medical and family history was non-contributory towards diagnosis. However, patient had habit of quid placement in lower labial vestibule four times a day since 30 years. The aged lady was

apparently healthy and oriented to environment and her vital signs were within the normal limit. Extraorally, an ulceroproliferative growth with broad base measuring approximately 5x6cm in its greatest dimension noted involving lower lip extending to both lip commissure. The surface of the growth appears erythematous intermingled with diffuse whitish areas. On palpation the growth was firm, tender with active pus discharge. Intraoral examination of lesion revealed an irregular ulceroproliferative growth with pebbled surface involving lower labial mucosa extending to lower labial sulcus with broad base measuring approximately 5x5.5cm in its greatest dimension. The surface of the growth appears erythematous intermingled with diffuse whitish areas (Figure 2).

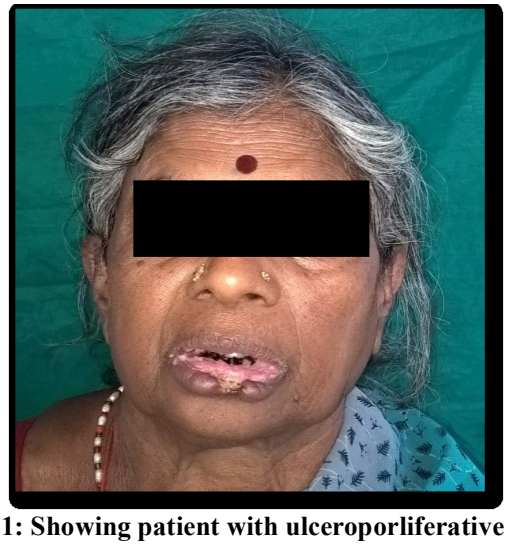

Figure 1: Showing patient with ulceroporliferative growth involving lower lip

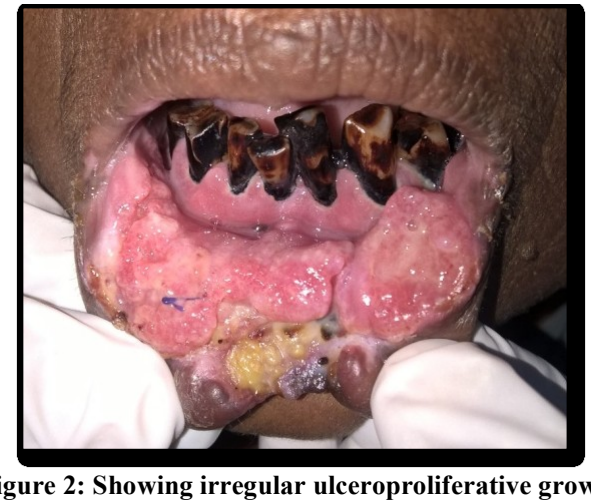

Figure 2: Showing irregular ulceroproliferative growth with pebbled surface involving lower labial mucosa extending to lower labial sulcus

Intraoral palpation of growth elicited the firm consistency of growth at the periphery and soft in the center. The margins of the growth were firm and were non-tender. There was no induration of the base, however thick purulent exudation was evident on application of pressure. Tenderness on percussion noted in lower central and lateral incisor teeth. Based on history and clinical examination, a provisional diagnosis of malignancy of lower lip was considered. An intraoral periapical radiograph of lower anterior revealed severe vertical bone loss in lower central and lateral incisor teeth (Figure 3).

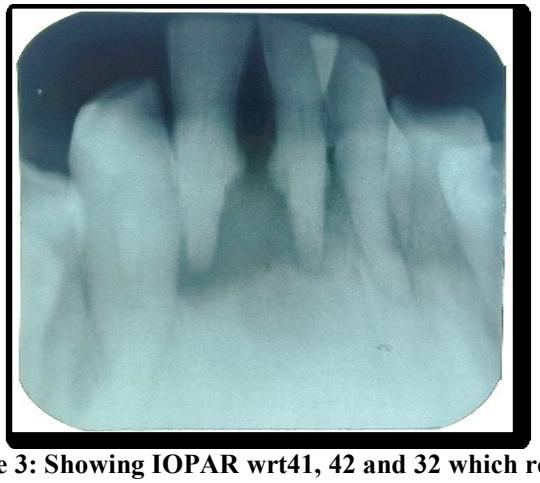

Figure 3: Showing IOPAR wrt41, 42 and 32 which reveals vertical bone loss wrt 32,41 and 42.

Similar findings were observed on orthopantogram which revealed a generalized alveolar bone loss without any significant erosion of bone (Figure4). Further, an incision biopsy was performed and sent for histological examination. Microscopic examination of section showed soft tissue fragment lined by non-keratinizing stratified squamous epithelium with elongated rete ridges (Figure 5a). The subepithelial stroma shows dense sheets of plasma cells, with presence of thin walled elongated blood vessels lined by plump endothelial cells amidst them (Figure 5b). There is also presence of fragments of minor salivary glands composed of lobules of mucinous cell with scattered clusters of lymphocytes in between the lobules (Figure 5c), suggestive of plasma cell granuloma with chronic sialadenitis of the minor salivary gland. Correlating all the investigation with the patient's history and clinical examination a final diagnosis of plasma cell granuloma of lower lip was established. Our case was managed by wide excision of lesion. There was no sign of reoccurrence of lip (Figure 6), however a similar lesion was identified on the right and left buccal mucosa after a follow up of six months.

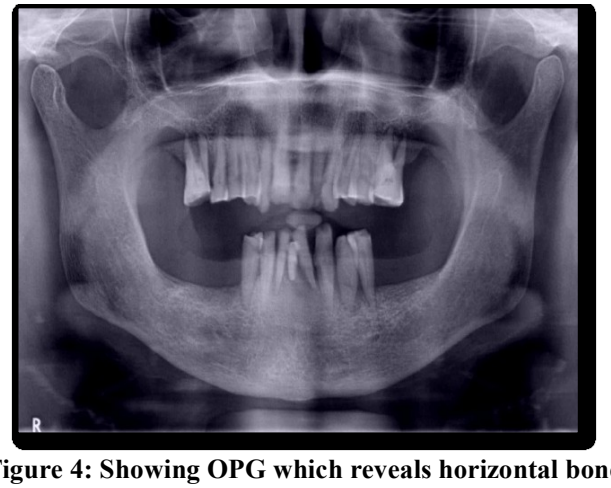

Figure 4: Showing OPG which reveals horizontal bone loss wrt 32, 33,34,35,41, 42, 43 and 44.

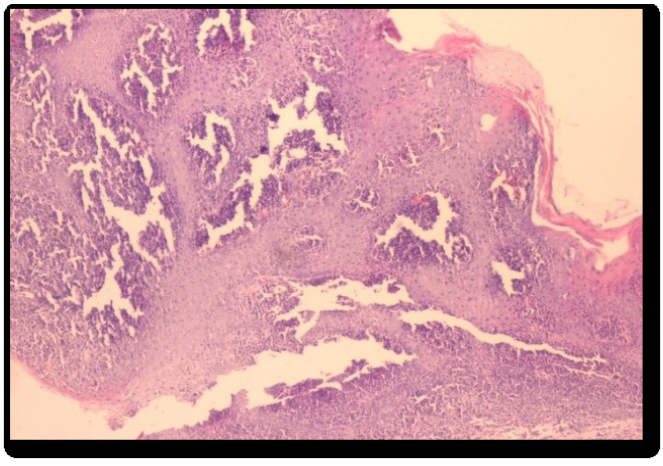

Figure 5a: showing soft tissue fragment lined by non-keratinizing stratified squamous epithelium with elongated rete ridges and the subepithelial stroma shows dense sheets of plasma cells (H & E 100x)

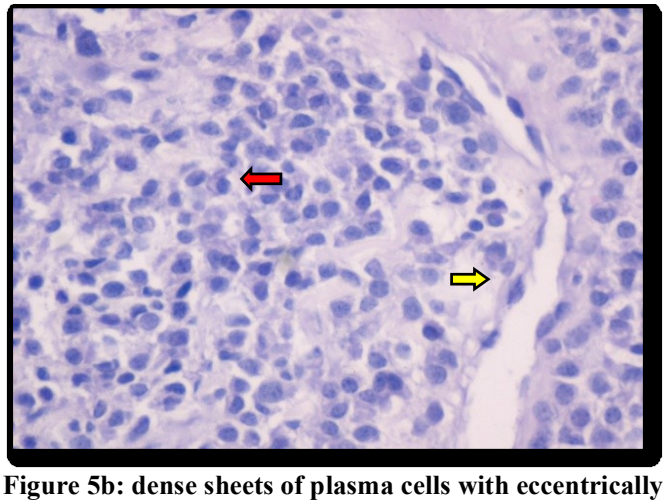

Figure 5b: dense sheets of plasma cells with eccentrically placed nucleus (red arrow)with presence of thin walled elongated blood vessels lined by plump endothelial cells (yellow arrow) amidst them(H & E 400x)

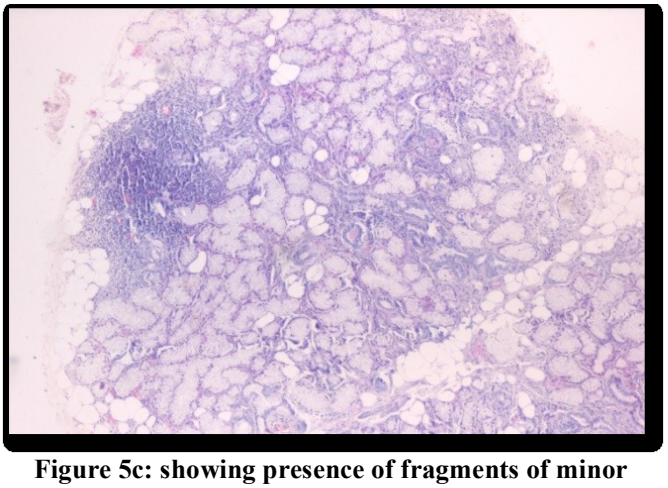

Figure 5c: showing presence of fragments of minor salivary glands composed of lobules of mucinous cell with scattered clusters of lymphocytes in between the lobules (H & E 400x)

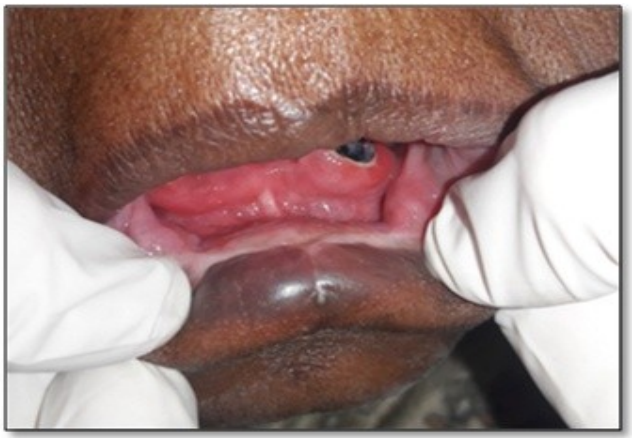

Figure 6: Showing no sign of recurrence on lower lip after follow up of 6 month

3. Discussion

Plasma cell granuloma is a rare benign tumor of inflammatory origin which was recently classified by WHO under soft tissue tumor.[8] Plasma cell granuloma are in head and neck region with an incidence of 15%.[6] Based on reviewed literature in the oral cavity plasma cell granulomas tend to locate primarily on the periodontal tissue and rarely on lip and labial mucosa with only one case reported so far.[1] Intraoral plasma cell granuloma occurs in the age range of 19thmonths to 63rd years with slight female predominance.[4] In present case also, 62year old female patient presented with plasma cell granuloma of lower lip.

Etiopathogenesis of plasma cell granuloma remain uncertain. However, it has been suggested that it represents the inflammation following minor trauma or surgery, or to be associated with other malignancy.[2] In present case, history of placement of quid in labial vestibule can be sorted as chronic trauma leading to inflammation, though the absolute link cannot be established. Not only this, an autoimmune mechanism has also been implicated in the pathogenesis of plasma cell granuloma. Also, association of plasma cell granuloma with infectious agents like mycobacteria, Epstein–Barr virus, actinomycetes, *Mycobacterium avium*, *Corynebacteriumequi, Escherichia coli, Klebsiella, Bacillus sphaericus, Pseudomonas, Helicobacter pylori* and *Coxiella burnetti* has also been anticipated.[2]

Clinically, PCG takes at least two morphological types in the oral mucosa: exophytic/tumor or unilateral ulcerative.[5] In this case it adopted both morphological variant and was simulating a malignant diseases. Diagnosis of such clinical variant of plasma cell granuloma is extremely challenging and imperative as it is benign inflammatory lesion and does not require extensive and potentially destructive surgery.

Coffin *et al,* classified plasma cell granuloma based on the histologic appearance in three patterns. The first pattern consists of myxoid, vascular and inflammatory areas, the second is characterized by compact spindle cells with intermingled inflammatory cells, and the third pattern consists of dense collagen and resembles a scar.[9] Our case presented with first pattern histologically. Present case reflected first pattern histologically with dense sheets of plasma cells, and presence of thin walled elongated blood vessels lined by plump endothelial cells amidst them.

Treatment depends on proper diagnosis; hence it is essential to differentiate plasma cell granuloma from other lesion like carcinoma, extramedullary plasmacytoma and multiple myeloma. The most common treatment for plasma cell granuloma is a complete resection; however, in some cases, total surgical excision is not possible. Radiotherapy and/or steroid therapy have sometimes been successfully used to treat patients with non-resectable lesions.[10,11] Our case was managed by surgical excision of the lesion and antiinflammmatory agents.

Recurrence of extrapulmonary plasma cell granuloma is reported to be upto 25-40%.[12] Our patient showed another lesion in the right and left buccal mucosa after 6 month which can be considered as second primary lesion rather than recurrence.

4. Conclusion

Plasma cell granuloma of lip is rare case entity which can be easily misdiagnosed as malignant lesion. Diagnosis of plasma cell granuloma is of utmost importance to avoid mutilating surgical procedure and for better prognosis.

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