A CASE REPORT ON INTRAORAL PLASMA CELL GRANULOMA IN AN UNUSUAL SITE

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ABSTRACT: Plasma cell granuloma is a rare non-neoplastic lesion that occurs most often in the lungs, but it is not commonly seen in the maxillofacial region. Its etiology, biological behavior, ideal treatment and prognosis are still unclear and rather controversial. Very few cases of intraoral plasma cell granulomas have been reported in the past. Hence, we present an unusual case of plasma cell granuloma in a 60-year-old female which was presented clinically as ill-defined swelling over left upper buccal vestibule region extending to the midpalate. Histological examination revealed fibro-cellular connective tissue stroma with inflammatory cell infiltrate containing plasma cells and lymphocytes. The plasma cells are abundant varying in size and shape, with very few large and binucleated plasma cells. Both clinically and histopathologically, it may be misinterpreted as various pathological entities thus proper evaluation of patient and histopathological examination of the tissue to rule out other lesions is mandatory.

KEYWORDS: Plasma cell granuloma, immunohistochemistry, reactive lesion.

INTRODUCTION

Plasma cell granuloma (PCG) was first described by Bahadori and Liebow in 1973. Intraoral PCGs have been reported on the tongue, lip, and buccal mucosa and in the gingiva. PCG in the oral cavity can be easily mistaken clinically as peripheral giant cell granuloma, pyogenic granuloma, thus necessitating the histopathological examination to confirm the diagnosis. Hence, the clinical, radiological, histopathological findings must be correlated for an accurate diagnosis. This present case report describes a rare case of plasma cell granuloma on the buccal vestibule region extending to the palate which was clinically thought as ameloblastoma.

Case Report

A 60-year-old female patient reported to the department of Oral Medicine and Radiology, SVS institute of Dental Sciences, Mahabubnagar, with a chief complaint of pain and swelling in the upper left back tooth region for the past 6 months. The swelling was initially small in size which gradually increased in size. Extraorally, a 5 X4 cm solitary diffuse swelling was noticed in the left middle third of the face. The swelling was initially small in size which gradually increased in size. Extraorally, a 5 X4 cm solitary diffuse swelling was noticed in the left middle third of the face. The swelling extended anteroposteriorly from the ala of the nose to 5cms in front of the ear. Superoinferiorly, it extended 2cms below the left eyelid to 1cm above the corner of the mouth. The skin over the swelling was normal. On intraoral examination, a solitary growth was extending from 26 to 28 over the left side of upper buccal vestibule region which extended up to the midpalate (Fig.1). The swelling was well-defined, oval in shape measuring about 5X4cms approximately. The colour of the swelling was slightly red as compared to that of the surrounding mucosa. On palpation, the inspectory findings were confirmed. The swelling was fluctuant in the palatal region, soft in consistency in the center and hard at the periphery and it was tender.

On radiographic investigations OPG revealed ill-defined radiopacity in the left maxillary sinus causing downward displacement of the arch and teeth (23 to 26) and loss of lamina dura of involved teeth and root apices (Fig. 2). The occlusal radiograph revealed a well-defined multilocular radiolucency distal to 27. It also showed expansion of the buccal cortex and erosion of palatal bone. CT scan revealed extension of the swelling from maxillary arch posteriorly and involving and obliterating maxillary sinus and superiorly extending till inferior border of mandible. A provisional diagnosis of ameloblastoma was made and the patient was sent to Department of Oral and Maxillofacial Surgery. Routine blood investigation was done. The findings were within normal limits.

An incisional biopsy was done under local anaesthesia and the specimen was sent for
Fig. 1. Solitary growth from the left side of upper buccal vestibule region extending to the midpalate.

Fig. 2. OPG revealed ill-defined radiopacity in the left maxillary sinus.

Fig. 3. High power magnification (40x) revealed a fibro-cellular connective tissue stroma with intense chronic inflammatory cell infiltrate chiefly composed of plasma cells and lymphocytes. The plasma cells with varying in size and shape and few large and binucleated plasma cells.

Fig. 4. Immunohistochemical study reveals the plasma cell infiltrate uniformly positive for CD138 antibody, a marker for plasma cells.
histopathological examination. Histopathological examination revealed a fibro-cellular connective tissue stroma with intense chronic inflammatory cell infiltrate chiefly composed of plasma cells and lymphocytes. The plasma cells were abundant varying in size and shape, with very few large and binucleated plasma cells. The connective tissue also shows numerous endothelial lined budding capillaries surrounded by clusters of plasma cells. Areas of hemorrhage, foci of ossification and extravasated RBCs were also evident. Immunohistochemical study of the biopsy showed the plasma cell infiltrate uniformly positive for CD138, a marker for plasma cells (Fig. 4).

A diagnostic work up was done to rule out multiple myeloma and extra medullary plasmacytoma. Chest x-ray and ECG revealed no significant features. The urine analysis for Bence-Jones proteins revealed negative thus ruling out multiple myeloma. Based on the clinical, histopathological findings the final diagnosis of Plasma cell granuloma was given.

Discussion

Plasma cells are terminally differentiated B lymphocytes that provide protective immunity through continuous secretion of antibodies. They are commonly seen in chronic inflammatory infiltrates and were first described by Zoon in balantias plasmas cellularism in the year 1952. Since then, plasma cell infiltrates have been documented in the vulva, buccal mucosa, nasal aperture, gingiva, lips, tongue, epiglottis, larynx and other orificial surfaces. The plasma cell granulomas were considered to be highly uncommon, non- neoplastic, reactive lesion, which were first brought to the attention of health care practitioners during the late 1960s and early 1970. Bhaskar, Levin and Firch first reported this pathological entity on the gingival tissue and only very few case reports have been documented since then. Avecdco and Buchler, Mark and Steven, Karthikeyan and Pradeep and Baltaciaglu et al. have reported this lesion on the gingiva. However, the present case was seen in the posterior palatal region which was well-defined, oval in shape present on the left upper buccal vestibule region and extended up to the midpalate.

The pathogenesis of the PCG remains unclear. The large number of plasma cells may represent an autoimmune reaction or an alteration of blood flow imposing congestive vasodilatation. Lesions occurring due to parasitic infiltration can also not be ruled out. The presence of polyclonal plasma cells, lymphocytes and histocytes suggests an infectious and autoimmune origin.

PCG is thought to result from inflammation following minor trauma or surgery or to be associated with malignancy. However, this was not so in the present case and our patient was free of any history of trauma or malignancy. However, her oral hygiene was extremely poor with generalized chronic destructive periodontitis, which could have contributed to the genesis of this lesion.

Clinically, gingival PCG presents as a mass, which is nodular, polyopoidal lesion with a smooth surface and well circumscribed. It does not produce significant systemic symptoms. Similar findings were seen in the present case.

PCG is microscopically characterized by vascular stroma with reactive inflammatory cells, including plasma cells predominantly and usually surrounded connective tissue septa. No cytological abnormalities are usually present, Russell bodies, which are intra-cytoplasmic eosinophilic hyaline droplets, may also be seen. In our case, microscopic examination revealed fibro-cellular connective tissue stroma with intense chronic inflammatory cell infiltrate chiefly composed of plasma cells and lymphocytes. The plasma cells were abundant varying in size and shape, with very few large and binucleated plasma cells. The connective tissue also shows numerous endothelial lined budding capillaries surrounded by clusters of plasma cells. Areas of hemorrhage, foci of ossification and extravasated RBCs are also evident.

PCG is a diagnosis of exclusion, distinguished primarily on the histological finding of a marked submucosal plasma cell infiltrate. Histologically, it is important to differentiate various plasma cell lesions such PCG, plasma cell gingivitis, and extramedullary plasmacytoma (a possible precursors of multiple myeloma).

Extramedullary plasmacytoma presents as a soft tissue mass outside of bone. The main differentiation between reactive PCG and extramedullary plasmacytoma is based on whether lesion is polyclonal or monoclonal. Histologically, extramedullary plasmacytoma consists of mixture of typical and atypical plasma cells while plasma cell granuloma consists of normal plasma cells and small lymphocytes that are surrounded by connective tissue septa. The immunohistochemical analysis have shown that in the case of malignancy ratio of the kappa to lambda light chain may be greater than 10:1 or 1:10, whereas in a reactive lesion the ratio is 2:1.

Some consider PCG to be a subtype of plasma cell gingivitis, plasma cell gingivitis is a polyclonal lesion of gingival tissue that is usually not a localized nodular lesion, as seen in PCG but presents as generalized edematous and erythematous elevations. In the present case a single, localized nodular mass was present. Plasma cell gingivitis is considered as an allergic hypersensitivity reaction to various flavoring agents used in chewing gums and dentifrices. But in our case, there was no identifiable inciting agent.
PCG is usually treated by simple excision and removal of the underlying inciting agent. With respect to prognosis, PCG seems to be a generally benign, nonrecurring condition: nevertheless, local aggressive, and recurrences may complicate the outcome of the disease.

CONCLUSION

Plasma cell granuloma is a rare benign lesion but its exact etiology, behavior and prognosis is still controversial. Attributing to the fact that it can masquerade as various pathological entity, it is difficult to establish the exact diagnosis alone on the basis of clinical or histopathological examination. Therefore, it is necessary to perform complete lab investigations differentially to diagnose it from other lesions that have a poor prognosis and to avoid unnecessarily extensive and potentially destructive surgery.

References


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