**Case Report**

A rare case of gingival plasma cell granuloma of the gingiva masquerading as a gingival epulis

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Plasma cell granuloma is a non-neoplastic lesion rather uncommonly seen in the maxillofacial region. Its etiology, biological behavior, ideal treatment and prognosis are still unclear and rather controversial. A rare case of this lesion affecting the gingiva is presented here, which could easily have been mistaken for a pyogenic granuloma, a peripheral giant sell reparative granuloma or a fibrous gingival epulis, had it not been for a detailed histological and Immunohistochemical study conducted on this lesion. Histopathological examination of the excised specimen revealed a proliferation of inflammatory cells, among which there was a typical predominance of plasma cells with the typical cartwheel shaped nuclei, set in a fibrovascular connective tissue stroma. Immunohistochemistry was employed for confirmatory diagnosis, and characteristically demonstrated the lambda and kappa light chains as seen in the polyclonal plasma cell population.

**Key words:** Plasma cell granuloma, plasma cells, kappa and lambda light chains.

**INTRODUCTION**

Plasma cell granuloma is an uncommon non-neoplastic lesion that was first described in 1973 by Bahadori and Liebow [1]. This lesion’s incidence, etiology, biologic behavior and most appropriate treatments are unclear, and little is known about the prognosis. Histologically, it consists of a proliferation of inflammatory cells, with a predominance of plasma cells, in a fibrovascular background. It has been called by different terms, that is inflammatory myofibroblastic tumour, inflammatory pseudotumour, inflammatory myofibrohistiocytic proliferation and xanthomatosus pseudotumour [1]. It primarily occurs in the lungs [2]. It is also seen to occur in the brain [3], kidney [4], stomach [5] and heart [6]. Rarely seen in the oral cavity the lesions are usually single, seen primarily on the periodontal tissue, mainly the gingiva, followed by tongue, lips, buccal mucosa and palate [7-10].

**CASE PRESENTATION**

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A 56 year old male patient presented with a localized,
Fig 1(a, b, c): A localized extraoral swelling in the region of the upper lip just below the ala of the nose on the right side, which intra-orally appeared as an irregular pinkish red, firm, lobulated, pedunculated, gingival growth in the upper anterior gingiva in 12, 13, 14 and 15 region.

Figure 2. (a, b): Lesion excised leaving a raw bleeding surface. A palliative Coe–pack periodontal dressing given to cover the wound.

non-tender swelling over the right side of his upper lip, measuring 3cm x 2cm, just below the right ala of the nose, causing obliteration of the nasolabial sulcus (Fig 1a). It was related to an intraoral gingival growth in the upper anterior gingiva in 13, 14 and 15 region (Fig 1b, c).

The intraoral mass was oval in shape and measured around 2 cm x 1cm. It was an irregular pinkish red in colour, lobulated, non-tender and pedunculated, attached to the gingiva by a narrow stalk (Figure 1c). The surface was smooth with no evidence of secondary changes such
Figure 4 (a,b,c): Haematoxylin & Eosin stained Sections of the lesion at 10X, 30X and 60X magnifications, showing a hyperplastic parakeratinized stratified squamous epithelium, with underlying richly fibrocellular connective tissue stroma containing a few lymphocytes, and abundant plasma cells.

Figure 5. (a, b, c): Immunohistochemical staining showing strong positivity for the kappa light chain seen in the polyclonal plasma cell population, a weak immunohistochemical expression noted for the lambda light chain; and a kappa to lambda light chain ratio of be 2:1 (the kappa light chains staining a dark intense brown, while the lambda light chains taking up a light tan colored IH stain; confirming the diagnosis of Plasma cell granuloma and its inflammatory origin.

as ulceration or any secondary discharge of pus. It was firm in consistency and bled readily on probing. The patient also complained of inability to maintain oral hygiene as the intraoral enlargement interfered with and bled on brushing. The patient's medical history was not contributory and he was on no long standing medications for any other medical condition. Routine blood and urine examination of the patient was carried out and all the values were within normal limits. A differential diagnosis of pyogenic granuloma, peripheral giant cell reparative granuloma, fibrous epulis, giant cell epulis and fibroma were considered.

An excisional biopsy and gingivoplasty was performed under local anesthesia (Fig 2a) and the excised specimen (Figure 3b) was sent for histopathological examination. A Coe-Pak dressing (Fig 2b) was given and the gingival healing was quick and uneventful (Figure 3a). Coe-Pak is a palliative surgical dressing and a periodontal pack, that protects raw areas and promotes their cleanliness and healing post operatively. It has no unpleasant taste or disagreeable odor and produces no burning sensation. It is marketed as a two tube system - one tube containing the base and the other containing the catalyst. Depending upon the size of the wound, required
Figure 4a, b, c) revealed a stratified squamous parakeratinized hyperplastic epithelium, with underlying fibrocellular connective tissue stroma, with fibroblasts, few lymphocytes, and abundant plasma cells with typical eccentrically placed hyperchromatic, cartwheel-shaped nucleus. The histopathological appearance, when correlated with the clinical features, was suggestive of plasma cell granuloma of the gingivae. Immunohistochemical staining for kappa and lambda light chains was carried out, to check for the presence of plasma cells and for confirmatory diagnosis (Figure 5a,b,c). A strong positivity for the kappa light chain was seen in the polyclonal plasma cell population (Figure 5a), whereas, a weak expression was noted for the lambda light chain (Figure 5b), with a ratio of 2:1 (Figure 5c), thus confirming the diagnosis of Plasma cell granuloma.

DISCUSSION

Plasma cells are terminally differentiated B lymphocytes which are typically found in the red pulp of the spleen, medulla of the lymph nodes, tonsils, lamina propria of the entire gastrointestinal tract, mucosa of the nose and upper airway, and sites of inflammation. They are characterized by a basophilic cytoplasm with an eccentrically placed nucleus. They range in size from 14 to 20 micrometers. A plasma cell's main function is to produce immunoglobulins or antibodies [1].

The phenomenon of plasma cell infiltrate was first described by Zoon in 1952 when he described balanitis plasma cellularis. Plasma cell infiltrates have also been found on the vulva, buccal mucosa, palate, nasal aperture, gingiva, lips, tongue, epiglottis, larynx and other orificial surfaces [1].

During the late 1960s and early 1970s, cases of plasma cell infiltrates of the lips, gums, and tongue were described primarily in the dental literature under the names of atypical gingivostomatitis, idiopathic gingivostomatitis, and allergic gingivostomatitis [11]. The lesions were thought to be a result of a reaction to chewing gum, dentrifices, and other foreign substances, although extensive allergy testing had been inconclusive [11].

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 granulomas may be benign, but plasmacytomatas may show early stages of multiple myeloma [11].

The pathogenesis of the plasma cell granuloma 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 [11]. The presence of polyclonal plasma cells, lymphocytes, and histiocytes suggests an infectious or autoimmune origin [12]. This lesion has no sex predilection and may occur at any age. Plasma cell granuloma is thought to result from inflammation following minor trauma or surgery or to be associated with malignancy [13, 14, and 15], however, this was not so in the present case and our patient was free of any history of trauma or malignancy. However his oral hygiene was extremely poor with generalized chronic destructive periodontitis, which could have contributed to the genesis of this lesion, which was present on the gingival and was solitary.

The immunohistochemistry determines the clonality of the lesion, wherein in a reactive lesion, the kappa to lambda light chain ratio is 2:1, and in the case of malignancy the ratio may be greater than 10:1 [11]. In the present case, ratio was approximately 2:1, suggestive of an inflammatory etiology.

A Coe-Pak palliative surgical dressing and a periodontal pack, was used to protect the raw area after excision of the lesion, which promoted a very quick, comfortable and uneventful healing of the wound post operatively.

CONCLUSION

Plasma cell granuloma can sometimes be mistaken for a malignant lesion due to its aggressive clinical appearance. In this case, cauliflower-shaped, pedunculated appearance also raised the suspicion of a verrucous carcinoma. Excisional biopsy followed by histopathological examination using Haematoxylin and Eosin stains exhibited the characteristic Plasma cells in a richly fibrocellular connective tissue stroma. Immunohistochemistry greatly aided in making a difficult and challenging diagnosis, by positively identifying the plasma cells and their origin. As a plasma cell granuloma is rarely encountered in the oral and maxillofacial region, the diagnosis of this lesion could have been very easily missed, had it not been for the detection of the large numbers of plasma cells with their typical eccentric cartwheel shaped nuclei scattered in the connective tissue stroma. Further immunohistochemistry was emplo-
yed for clinching the diagnosis. In this way we see that an appropriate identification of any lesion is important to avoid needless extensive and radical surgical procedures.

REFERENCES


