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Atypical gingivitis: A rare case report of recurrent and familial history, and its surgical management

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Abstract

Plasma cell gingivitis, an infrequent benign entity, is an inflammatory or reactive condition of the gingiva to certain allergens or to an unknown factor. It is clinically characterized by erythematous, edematous, granular/cobblestone appearance of gingival surfaces with clear demarcation from the mucogingival junction. It easily bleeds on manipulation and may be associated with a burning sensation to spicy food consumption. As the name suggests it is diffuse and massive infiltration of plasma cells into the sub-epithelial gingival tissue. It is a hypersensitivity reaction to some antigen, often flavouring agents or spices found in chewing gums, toothpastes and lozenges. Plasma cell gingivitis is also known as atypical gingivostomatitis, allergic gingivostomatitis, unusual gingivostomatitis and idiopathic gingivostomatitis. Histologically, the lesion shows dense plasma cell infiltrate of the connective tissue. We report a case of a rarely seen mucosal pathology of a 51 yr old male patient with a chief complaint of bleeding swollen mass in the upper and lower front tooth region who presented with this rare condition and its histological diagnosis and its management.

Keywords: Plasma cell gingivitis, periodontitis, enlargement, biopsy, allergic

Introduction

Plasma cell gingivitis is a rare benign condition of the gingiva characterized by sharply demarcated erythematous and edematous gingivitis often extending to the mucogingival junction [1]. Other terminologies previously used for this unusual type of conditioned enlargement were atypical gingivitis, allergic gingivitis, plasmacytosis of the gingiva, plasma cell gingivostomatitis and idiopathic gingivostomatitis [2]. Plasma cell gingivitis is considered a hypersensitive reaction to some antigen, often to chewing gum, toothpaste, and other foreign substances. The allergens identified were mostly cinnamonaldehyde and cinnamon, used as flavouring agents in chewing gums and dentifrices. Clinically, the gingiva appears red, friable and bleeds easily. Histopathologically, a dense and massive infiltration of mature plasma cells in the lamina propria is noted [3]. Early diagnosis is essential as PCG has similar pathologic changes seen clinically as in leukemia, HIV infection, discoid lupus erythematosus, atrophic lichen planus, desquamative gingivitis, or cicatricial pemphigoid that must be differentiated through hematologic and serologic testing [4].

Clinical findings

A 51 year old male patient reported to the department of periodontics and implantology, D.Y.Patil school of dentistry, Navi Mumbai. With a chief complain of Bleeding gums and swollen mass in the upper and lower front tooth region. Patient noticed these swelling 6 months ago in his upper jaw in front and right region, which was slowly increasing in size. Oral examination revealed a fiery red gingival enlargement of the marginal, papillary, and attached gingiva between the right and left maxillary central incisor, with a small 2 × 2 x 2 cm outgrowth from the mesial portion of the marginal gingiva. There was bleeding on probing, with loss of stippling and contour. It had a soft consistency and it was non-tender in nature. Probing depth ranged from 7 mm to 9 mm with an attachment loss of 5-7 mm in the maxillary anterior teeth region.

Grade I mobility around the maxillary central incisors and mandibular central and lateral incisors. Patient had a poor oral hygiene, was systemically healthy and he not report a positive drug history. The recent change of oral hygiene products like toothpaste or consumption of chewing gum was denied by the patient. There was no loss of appetite, fever or lack of sleep. The patient was an occasional smoker since the last 25 years. He has supari chewing habit for the past 25 years after taking his meals.

After taking a detailed history, it was found that the patient

had similar signs and symptoms 12 years back for which the treatment was done at D.Y. Patil School of dentistry. Patient also gave familial history in which he mentioned that his sister had same swollen gums and was treated for the same. Radiographs showed a moderate amount of bone loss in the maxillary anterior region. A blood sample was obtained in order to rule out systemic diseases or any other blood dyscrasias. Provisional diagnosis of the chronic generalized gingivitis with localized gingival enlargement was made.

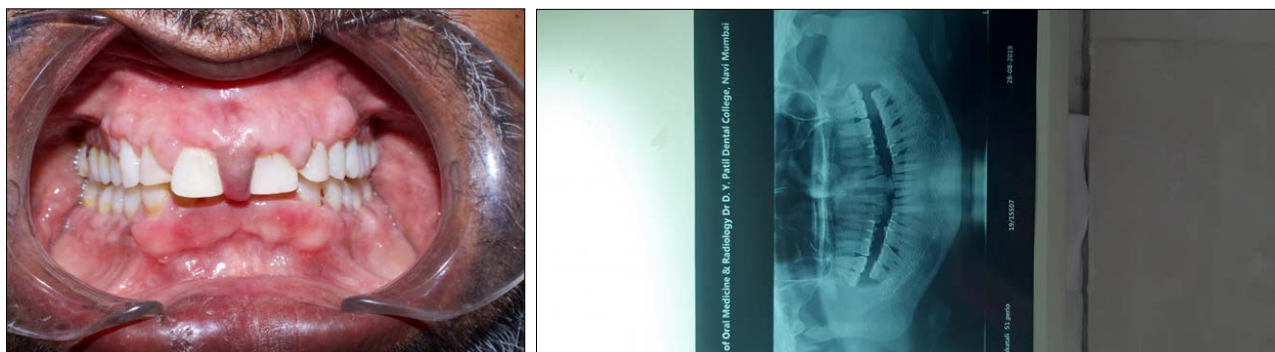


Fig 1: Pre Operative picture.

Table 1: Describes the differential diagnosis of the present case.

Lesion	Clinical features	Histopathological features
Giant cell epulis	A peripheral giant cell granuloma that arises exclusively from the PDL enclosing the root of a tooth.	The connective tissue stroma was highly cellular, consisting of proliferating plump fibroblasts. Numerous giant cells of various shapes and sizes, containing 8–15 nuclei, were seen with proliferating and dilated endothelial lined blood capillaries with extravagated red blood cells (RBCs).
Pyogenic granuloma	They are small, round, and usually bloody red in colour. They tend to bleed because they contain a large number of blood vessels. They're also known as lobular capillary hemangioma.	Pyogenic granuloma shows exuberant granulation tissue which is covered by hyperplastic epithelium that may be ulcerated at times and reveals fibrinous exudates
Multiple myeloma	It is characterized by the neoplastic proliferation of plasma cells producing a monoclonal immunoglobulin.	They have eccentric nuclei that are smooth in contour with clumped chromatin and have a perinuclear halo or pale zone. The cytoplasm is basophilic.

Non surgical therapy included thorough oral prophylaxis, oral hygiene instructions and an antihistamine mouthwash. The patient was instructed to avoid possible allergens like chewing gums, cosmetics and food additives. A strict elimination diet was advised and a change of the regular toothpaste was tried. Herbal toothpastes were not prescribed since reports have shown them as an etiologic factor in the initiation of these types of lesions.

Though all these modalities were followed stringently, there was no true regression of the disease. Hence, a surgical approach was planned for the gingival lesion.

Surgical Therapy Included Modified Widman flap surgery was done in all six sextants and the biopsy were sent for histopathological examination. Non eugenol pack (Coe-pack) was placed and post-operative antibiotics and analgesics were prescribed.

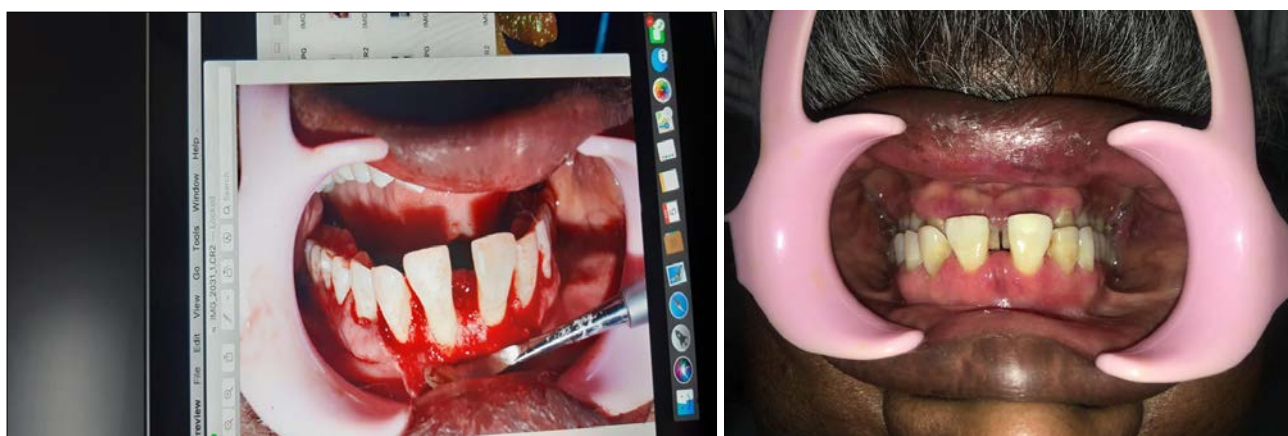


Fig 2: Post-operative picture with 8 months follow up

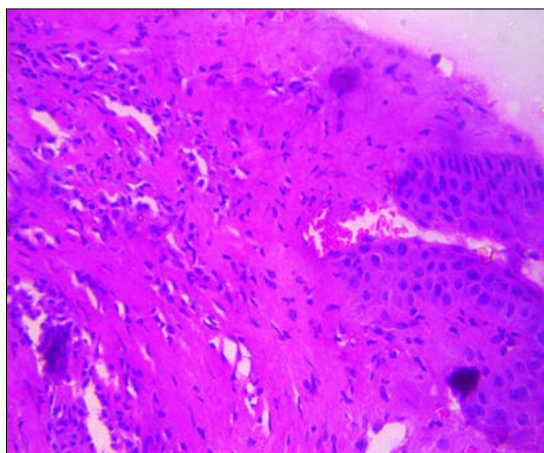
Macroscopic pathology

4 bits of soft tissue measuring 2x2x2cm and smallest measuring 0.5x0.5x0.5 mm irregular in shape, whitish in colour, firm in consistency and the margins are irregular.

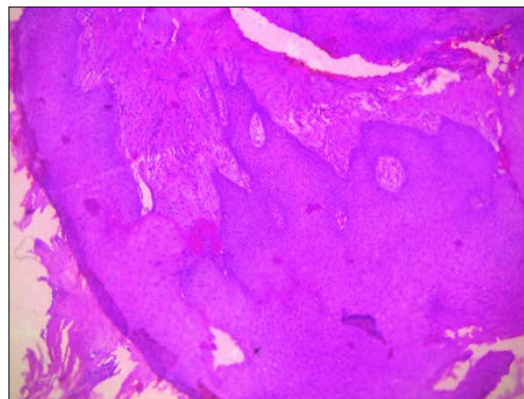


Histological findings

- Microscopic examination revealed a marked squamous hyperplasia with focal ulceration and diffuse dense subepithelial plasmacytic infiltrate consistent with PCG [7]. At higher magnification, plasma cells were seen without cellular atypia. The individual plasma cells had eccentric round nuclei with cartwheel chromatin pattern and an abundant cytoplasm the differential diagnosis of the condition is very important [7]. Most cutaneous disorders were eliminated from consideration by the lack of skin lesions and a negative Nikolsky sign [8]. However, the patient's failure to respond appropriately to initial periodontal therapy necessitated a biopsy of the involved tissue. The histopathological picture revealed replacement of underlying connective tissue by a population of cells predominantly made up of plasma cells thus indicating the diagnosis. Plasma cells are large lymphocytes with abundant cytoplasm and a characteristic appearance on light microscopy. They have basophilic cytoplasm and an eccentric nucleus. The histopathological examination revealed sheets of round, plasma cells, suggestive of plasma cell gingivitis. Plasma cells are normally cells that produce immunoglobulins, the antibodies that help fight infections and support our immune systems.



- H and E stained section shows parakeratinized stratified squamous epithelium which is highly proliferative exhibiting spongiosis and several areas of connective tissue entrapment. Underlying connective tissue stroma is highly fibrocellular showing dense infiltration with plasma cells. Few blood capillaries are seen.



Discussion

Plasma cell gingivitis is considered to be a rare condition of gingiva and represents a hypersensitive reaction with predominant plasma cell infiltration of the connective tissue component of the epithelium with unclear Etiology [6]. However, it has been hypothesized that the immunologic reaction to some allergic antigen might be the possible causative agent. Mint in the toothpaste, chewing gum, cinnamonaldehyde, strong spices, chilies, chewing of khat, and certain constituents of herbal toothpastes have been documented as the reported allergens in the literature. The most common flavourings that are frequently responsible for toothpaste allergies are cinnamal, spearmint, peppermint, carvone, and anethole [3]. Because most toothpaste is flavoured with either a variation of mint or cinnamon, it can be challenging to find toothpaste free of these flavours for those who have an allergy. The second most commonly present allergen in toothpaste was cocamidopropyl betaine (CAPB). Even with this being the second most common allergen, only 16 of the 80 toothpastes actually contained this surfactant. The third most common allergen was propylene glycol, a water-soluble vehicle [11].

Based on etiology, plasma cell gingivitis is categorized as three types: (1) lesions of unknown cause, (2) lesions owing to some allergen, and (3) lesions due to neoplastic origin. This entity is thought to be a B-cell mediated disorder with T-cells augmenting the response. In the present case, because the patient did not give any history of allergy or any recent change in her diet and oral habits, no cause could be elucidated [5].

This benign condition was first described in the year 1952 when Zoon referred to the term "plasma-cell infiltrate" condition involving the glans penis. These conditions have also been reported on the lips, tongue, vulva, conjunctiva, nasal aperture, larynx, and epiglottis. Kerr and his associates in the year 1971 reported a case of plasmacytosis of gingiva and revealed the most causative agent responsible for the allergic response to be chewing gum. He observed that if the patients discontinued the use of the chewing gum, the tissues returned to normal [8]. Kerr *et al.* reported occurrence of plasma cell gingivitis, cheilitis and glossitis in patients, and identified the culprit as the cinnamonaldehyde in chewing gums. The condition regressed completely on discontinuity of the use of these gums [4]. Silverman and Lozada described plasma-cell gingivostomatitis as a syndrome, consisting of gingivitis, cheilitis, and glossitis [6]. In the above case, when the clinical and histological findings are correlated, the case can be diagnosed as a part of plasma cell mucositis. Cases have been reported about plasma-cell infiltrates on the buccal mucosa, palate, nasal aperture, gingiva, lips, tongue, epiglottis, larynx, and other orofacial surfaces. In 1986,

White *et al.* grouped all such lesions under the name plasma-cell orofacial mucositis because all the cases reported had clinical and histologic findings that were indistinguishable from one another.

Management of plasma cell mucositis involves both medical and surgical approaches. Although several treatment modalities have been tried, including corticosteroids (topical, intra lesional, and systemic), antibiotics, destruction of the tissue (liquid nitrogen, Carbon dioxide laser and electrocoagulation), excision of the tissue, and radiation therapy, no treatment clearly stands out as consistently effective.

The patient in this case report had not responded to topical or systemic antibiotics and hence was managed surgically. The enlargement which was very evident previously has regressed spontaneously after gingivectomy, and has shown no recurrence till date.

Plasma cell gingivitis is a diagnosis of exclusion, distinguished primarily by the histologic finding of a marked submucosal plasma-cell infiltrate, after conditions such as infection and plasmacytoma have been eliminated. Careful history taking, biopsy and hematological examinations are mandatory to exclude leukemia and other local manifestations of systemic diseases.

Though recurrences are common, no studies till date report a progression of this condition to a malignancy of any type. The condition is believed to be a nonspecific inflammatory response, in the form of a plasma-cell infiltrate, to an unknown exogenous agent. However, attempts to induce plasma-cell infiltrations on mucosal and non-mucosal surfaces by allergic and irritant stimuli were not successful. Roman hypothesized that plasma-cell gingivitis may be associated with low levels of serum IgA and secretory IgA, which allows localized, repetitive, subclinical infections that could lead to the plasma-cell infiltrate.

In this case, the gingivitis and cheilitis suggest a contact allergy to some antigenic agent which is yet to be found out. The patient is regularly followed up to assess oral hygiene maintenance as well as identification of a possible allergen to avoid recurrences.

During the quest for a true allergen, repeated surgical excisions may be required for the gingival enlargement to facilitate plaque control. Plasma cell gingivitis should be included in the differential diagnosis of nonspecific enlargements of the gingiva with characteristic clinical and histological appearance, which cannot be attributed to any other disease entity. This condition, without any doubt, presents as a diagnostic dilemma and therapeutic challenge to the specialist.

Conclusions

Plasma cell gingivitis is a diagnosis of exclusion, distinguished primarily by the histologic finding of a marked submucosal plasma-cell infiltrate, after conditions such as infection and plasmacytoma have been eliminated. Careful history taking, biopsy and hematological examinations are mandatory to exclude leukemia and other local manifestations of systemic diseases. The condition is believed to be a nonspecific inflammatory response, in the form of a plasma-cell infiltrate, to an unknown exogenous agent. However attempts to induce plasma-cell infiltrations on mucosal and non-mucosal surfaces by allergic and irritant stimuli were not successful.

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Footnotes

Source of Support: Nil

Conflict of Interest: None declared

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