Plasma-Cell Gingivitis a Challenge to the Oral Physician

Abstract
Plasma-cell gingivitis (PCG) is an uncommon inflammatory condition of idiopathic etiology. PCG is a rare condition, appearing as erythema (redness) and edema (swelling) of the attached gingiva. The diagnosis of PCG is based on arrant history taking, clinical examination, and appropriate diagnostic tests. Here, we are presenting a case of PCG in a 12-year-old boy, its management and treatment outcome after 3 years of regular follow-up in tertiary health-care center in North Kerala.

Keywords: Gingivectomy, laser therapy, plasma-cell gingivitis

Introduction
Plasma-cell gingivitis (PCG) was first described in the late 1960. A wave of cases occurred during this period, thought to be caused by allergic reactions to a component in chewing gum spices, foods, candies, or dentifrices.[1] Plasma-cell gingivitis is a rare and unique gingival disorder, characterized histopathologically by a dense chronic inflammatory infiltration of the lamina propria, mainly of plasma cells. The importance of this lesion is that it may cause severe gingival inflammation, discomfort, and bleeding and may mimic more serious conditions.[2] Since the number of cases has decreased, they are still occasionally reported. According to the etiology, PCG is categorized as lesions of unknown cause, lesions owing to some allergen, and lesions due to the neoplastic origin. Bleeding on the slightest provocation is an invariable feature. Periodontal signs such as the loss of attachment are usually absent. However, architectural changes such as the loss of stippling are commonly observed.

The incidental clinical resemblance of the PCG to the gingival changes as seen in leukemia, lichen planus, and cicatricial pemphigoid does require thorough serologic and hematologic testing. PCG being a benign condition remits after detection and elimination of the etiologic agent.[3] Lesions may mimic that of acute leukemia and histologically imitate multiple myeloma and extramedullary plasmacytoma. Hence, the diagnosis requires hematological screening in addition to clinical and histopathological examinations.[4] The case presented here is a PCG associated with unusual gingival enlargement in a 12-year-old patient and treatment outcome with regular follow-up for 3 years.

Case Report
Here, we present a case of PCG in a 12-year-old boy who presented with unusual gingival enlargement and erythema. The treatment outcome with a regular follow-up is also discussed. A 12-year-old boy reported to the Department of Oral Medicine and Radiology, Government Dental College Calicut (Kozhikode), North Kerala, with a chief complaint of painless swelling of gums in the upper and lower regions of teeth for 3 months with bleeding while brushing. The medical, dental, and personal histories of the patient were noncontributory. No history of any familial diseases was reported. Clinically no extra Oral anomaly detected [Figure 1].

Intraoral examination revealed a diffuse severe gingival erythema and enlargement covering up to the cervical third of the clinical crown involving attached and marginal gingival of facial aspect maxillary and mandibular anterior teeth region in relation to the right maxillary central incisor to the right side maxillary first premolar and the left maxillary lateral incisor to the left side maxillary canine and the right mandibular lateral incisor to the right side mandibular second premolar.
region [Figure 2]. There was no loss of attachment and pus discharge from the gingival sulcus. The gingiva appeared edematous with the loss of stippling [Figure 2]. Bleeding on slight manipulation was present. The lesion was irregular in margins and rough surface. Generalized pseudo pockets ranged from 5 mm to 8 mm [Figure 2]. All the teeth except the third molars were erupted, with crossbite of right maxillary central incisor, right maxillary lateral incisor, and right mandibular lateral incisor, right mandibular canine was found.

In PCG, oral manifestation of hematological disorders, such as leukemia and granulomatous lesions, was
considered as the differential diagnosis [Figures 3 and 4]. Radiographic and hematologic investigations were carried out but did not reveal any significant finding [Figure 4].

An incisional biopsy was performed from the gingiva, and the tissue specimen was sent for the histopathologic examination. The histopathologic examination revealed bits of the tissue covered by hyperplastic parakeratinized
The underlying connective tissue shows intense chronic inflammatory cells predominantly the plasma cells intermingled with numerous endothelium-lined vascular spaces filled with red blood corpuscles into moderately collagenous stroma [Figures 5 and 6]. Immunohistochemistry staining for kappa [Figure 7] and [Figure 8] lambda light chain was carried out to check the presence of plasma cells. A strong positivity for kappa [Figure 7] and [Figure 8] lambda light chain was seen in the polyclonal plasma cell, and a final diagnosis of PCG was made.

The patient was prescribed topical steroids, antiseptic mouthwashes and antihistamines, which give only temporary results. [Figures 9 and 10]. An oral prophylaxis was carried out initially, followed by gingivectomy in the maxillary gingiva [Figure 11]. However, the results were not promising with only mild improvement [Figure 12]. Laser biostimulation was done with soft-tissue diode laser after obtaining the informed consent from the patient [Figure 13]. A diode laser of a 980-nm wavelength was opted with a beam diameter of 12 mm to cover the entire affected area in a scanning motion. The power setting of the laser was set at 1.5W and was used in a continuous noncontacting defocused mode in a scanning motion, starting at a distance of 2–5 mm away from the lesion and slowly approaching the lesion within for about 30–40 s for the first pass [Figure11]. Whenever the patient complained of warmth, the distance was suitably increased. The patient required two more passes of 3.2W and 4.0W, for about 30–40 s each. A cooling interval of 15 s was given in between the passes. The patient is reviewed after 7 days of therapy which showed improvement of the symptom [Figure 14]. Subsequent therapy was used at a weekly interval in further follow-up for three visits and has shown satisfactory results till date [Figure 15].

**Discussion**

PCG is a rare condition characterized by a massive infiltration of plasma cells into the subepithelial gingival tissue. Clinically, the illness presents as a diffuse enlargement with edematous swelling of the gingiva in the maxillary and mandibular anterior segments. Although the exact mechanism behind this condition is not known, the presence of plasma cells suggests an allergic origin. However, most of the cases seen are without any known or identifiable causes.

Gargiulo et al.\(^5\) in 1995 classified PCG into three types – caused by an allergen, neoplastic nature, and of unknown origin. The case report which we depicted here also is of unknown etiology and hence belongs to Type 3. PCG usually occurs in the anterior gingiva, most frequently in the maxilla. Even in our case, the patient showed pronounced enlargement of the facial gingiva in the anterior maxillary and mandibular regions. Plaque-induced gingivitis would normally involve the marginal gingiva alone and not the entire width of attached gingiva. In the present case, there was an inflammation of marginal and attached gingiva, which was not responding to local therapy and hence inconsistent with a plaque-related etiology. Biopsy had
helped to rule out oral granulomatous lesions. Blood picture and bone marrow tests cleared doubts about hematologic malignancies.

S Dhir et al.\textsuperscript{[3]} suggested that \textit{candida albicans} as an etiological factor; however, the absence of fungal hyphae under microscopic examination excluded any such infection in this case. Differential diagnoses for the lip lesion included cheilitis granulomatosa, dermatitis venenata, actinic cheilitis, and plasma cell cheilitis. Plasma cell cheilitis is a very rare and benign inflammatory mucosal condition characterized by a dense plasma cell infiltrate within the mucosa. The classic clinical appearance is a flat-to-slightly raised, eroded area, usually on the lower lip of an elderly patient. Plasma cell cheilitis was ruled out here because of a lack of erosion or crusting and also as there was enlargement of the upper lip alone. Granulomatous cheilitis was ruled out by biopsy. Actinic cheilitis was excluded since this usually involves the external aspect of the lips.

DA Kerr et al.\textsuperscript{[6]} reported the occurrence of PCG, cheilitis, and glossitis in patients and identified the culprit as the cinnamaldehyde in chewing gums. The condition regressed completely on the discontinuity of the use of these gums. S Jr Silverman described plasma-cell gingivostomatitis as a syndrome, consisting of gingivitis, cheilitis, and glossitis.\textsuperscript{[7]} In the above case, when the clinical and histological findings are correlated, the case can be diagnosed with a part of plasma-cell mucositis. Mucous membrane plasmacytosis of the upper aerodigestive tract is a rare benign disorder, in which the mucous membranes are infiltrated by plasma cells.\textsuperscript{[8]} Cases have been reported about plasma-cell infiltrates on the buccal mucosa, palate, nasal aperture, gingiva, lips, tongue, epiglottis, larynx, and other orificial surfaces.

In 1986, JW White et al.\textsuperscript{[9]} grouped all such lesions under the name plasma-cell orificial mucositis because all the cases reported had clinical and histologic findings that were indistinguishable from one another. The management of plasma cell mucositis involves both medical and surgical approaches. Although several treatment modalities have been tried, including corticosteroids (topical, intralesional, and systemic), antibiotics, destruction of the tissue (liquid nitrogen, carbon dioxide laser, and electrocauption), excision of the tissue, and radiation therapy, no treatment clearly stands out as consistently effective.

**Laser interaction with biological tissues**

Broadly, the tissue effects of lasers can be groups as:
- Photochemical interaction
- Photothermal interactions
- Photomechanical interactions
- Photoelectrical interaction.

**Photochemical interactions**

Specific wavelength absorbed by naturally occurring chromophores. Wavelength-specific light-absorption substances that can induce certain biochemical reactions at the cellular level. Photochemical interactions are subdivided into photodynamic therapy biostimulation.

**Photodynamic therapy**

Therapeutic use of lasers to induce biochemical reactions in the tissues for the treatment of pathologic conditions.

**Biostimulation**

The stimulatory effects of the laser light on biochemical and molecular processes that normally occur in the tissues such as heating or repair.

Babu et al. reported case series “versatility of diode lasers in low-level laser therapy (LLLTI) for the management of recurrent aphthous stomatitis” and considered that the LLLT as a prime treatment modality for managing RAS (Recurrent Aphthous Stomatitis) in their case series provides immediate pain relief to the patient and augment healing of the lesion.\textsuperscript{[10]} The possible mechanisms for immediate reduction in pain in their case series might be attributed to the fact that LLLT stimulates the production of β-endorphins, which are considered as our body’s natural painkiller, thus causing pain relief. It has a profound effect on C-fibers, leading to a decreased activity of these fibers and altering the pain threshold.\textsuperscript{[11]}

The patient in this case report had responded to diode bio-laser biostimulation and has shown no recurrence till date.

**Conclusion**

PCG 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. Although 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 nonmucosal surfaces by allergic and irritant stimuli were not successful. Roman hypothesized that plasma-cell gingivitis may be associated with low levels of serum immunoglobulin A (IgA) and secretory IgA, which allows localized, repetitive, subclinical infections that could lead to the plasma-cell infiltrate.

In this case, the PCG suggests a contact allergy to some antigenic agent which could not be identified. The patient should be 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, surgical excisions may be required for the gingival enlargement to facilitate plaque control. PCG presenting
with nonspecific enlargements of the gingiva and histological appearance, which cannot be attributed to any other disease entity, often presents a therapeutic challenge to the specialist.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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