An atypical presentation of plasma cell gingivitis with generalized skin lesions

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Abstract
This article aims to present a report of an atypical clinical presentation of a plasma cell gingivitis (PCG) case with unusual skin lesions. PCG is a rare benign inflammatory condition which can be classified into Type 4 hypersensitivity reaction. It occurs due to reaction to unknown antigen, often flavoring agents or spices found in chewing gums, toothpastes and lozenges. Histologically, the lesion shows dense plasma cells infiltrate in the connective tissue. Early diagnosis of PCG is essential to differentiate from variety of conditions, namely, leukemia, HIV infection, discoid lupus erythematosus, atrophic lichen planus, desquamative gingivitis, or cicatricial pemphigoid which must be differentiated through hematologic and serologic testing. In this article, we will discuss a case of PCG with unusual skin lesions.

Keywords: Hypersensitivity, plasma cell gingivitis, subepithelial separation

INTRODUCTION
Plasma cell gingivitis (PCG) is a rare benign inflammatory condition of the gingiva. The etiology of the lesion is still unknown but thought to be due to hypersensitivity reaction. The aim of this article is to present a report on the clinical presentation of PCG with unusual skin lesions; no cases with similar findings are published previously with skin lesions and subepithelial separation. Synonyms for PCG are atypical gingivostomatitis and plasma cell gingivostomatitis. These conditions have been reported on the lips, tongue, vulva, conjunctiva, nasal aperture, larynx and epiglottis. It is clinically characterized by erythematous, edematous and cobblestone appearance of gingival surfaces with clear demarcation from the mucogingival junction.

PCG is a Type 4 hypersensitivity reaction. The reaction is often to flavoring agents or spices found in chewing gums, toothpastes and lozenges, identifying the causative agent may not always be possible.[1-3] Histologically, the lesion shows dense plasma cells infiltrate the connective tissue. Early diagnosis is essential of PCG to differentiate from variety of conditions, namely, leukemia, HIV infection, discoid lupus erythematosus, atrophic lichen planus, desquamative gingivitis, or cicatricial pemphigoid which must be differentiated through hematologic and serologic testing.[4] This condition has been classified into following three categories based on their etiology: (1) PCG due to allergens, (2) PCG of neoplastic origin and (3) PCG due to unknown cause.

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CASE REPORT

A 24-year-old female walked in the Department of Periodontics of Saveetha Dental College and Hospital, Chennai, with the chief complaint of painless reddish swollen mass in lower right anterior region with gums which bleed profusely on mild manipulation for 4 months which increased in size with time. On further questions, the patient revealed that she was born out of a consanguineous marriage and she also revealed that she had started using herbal toothpaste approximately 4–6 months back. No history of loss of sleep or fever was given by the patient. Medical history of the patient did not give any alarming history of debilitating systemic diseases except unusual asymptomatic patchy skin lesions since birth [Figure 1], additional peripheral sensory, vision and ocular examination carried out to eliminate Cockayne syndrome. No changes in the condition were observed postantibiotic course prescribed by the physician. Intraoral examination revealed generalized reddish pink gingiva which easily bled on mild provocation. Enlargement of 1 × 1 × 1.5 in respect to lower right canine and first premolar the lesion showed desquamative surface changes. Periodontal examination showed generalized probing depth of 2–4 mm except in 15, 23 and 36 region where probing depth was 5–7 mm and 4–6 mm pseudopocket in relation to 43, 44 region suggesting chronic generalized gingivitis with localized periodontitis in relation to 152,336 region. No mobility in any teeth was recorded. Substantial local deposits were noticed in the oral cavity. The patient was systemically healthy and did not report a positive drug history. Radiographs showed a localized horizontal bone loss of 2–3 mm in relation to 15.23 and angular bone loss in respect to 36 [Figures 1-4].

Blood investigations did not show any abnormality. The patient presents with wrinkled skin and patches of hyperpigmentation all over the skin without any systemic symptoms. With these findings, a provisional diagnosis of chronic gingivitis with inflammatory enlargement was made. Moreover, differential diagnosis of PCG and Cockayne syndrome was made.

Phase 1 periodontal therapy was carried out which included scaling and root planing along with 0.2% chlorhexidine mouthwash twice daily, and the patient was recalled after 10 days for a review appointment; however, patient's failure to respond appropriately to initial periodontal therapy necessitated a biopsy of the involved tissue. Excisional biopsy of the lesion present in lower right anterior gingival mass was taken after acquiring patient consent and was sent for histopathological examination.

Figure 1: (a) Extraoral frontal image. (b) Image showing patchy wrinkled appearance of the skin on hands

Figure 2: Preoperative intraoral frontal photograph showing enlargement in relation to 43, 44 and local deposits

Figure 3: One week postoperative phase 1 therapy image

Figure 4: OPG
Histopathological examination revealed following findings:

1. Fibrocellular connective tissue stroma with sheets of plasma cells admixed with neutrophils in the superficial layers along with areas of endothelial cell proliferation and numerous capillary-sized blood vessels [Figure 5].

2. There was evidence of parakeratinized stratified squamous epithelium of variable thickness with subepithelial plane of separation consisting of red blood cells along with other areas showing ulceration replaced by fibrinopurulent membrane [Figure 6].

Histopathological findings confirmed the diagnosis of PCG. Further examination of K 13 and K 5 along with laminin was suggested to rule out possible mutations of epidermolysis bullosa and Cockayne syndrome, etc.

DISCUSSION

PCG is a rare benign condition of the gingiva. It is marked by a dense infiltrate of normal plasma cells separated into aggregates by strands of collagen. The first case was reported in 1981 by Kerr et al. where they came across gingival enlargement in gum chewers which was resolved with the discontinuation of the habit.[6-9] PCG has been classified in three following group by Garguillo[4,10] and Timmers et al., as an immunological reaction due to neoplasm, hypersensitivity, or unknown cause.[1,2,4,9,11,12] The importance of this lesion is that it may cause severe gingival inflammation, discomfort and bleeding of the gums and may mimic more serious conditions. There is no particular sex predilection or age distribution.[2,13] The diagnosis needs clinical and histopathological examination with hematological screening to rule out other clinically similar pathologic conditions such as leukemia, plasmacytoma and multiple myeloma so that early diagnosis and better treatment can be provided.[11,14] PCG is purely benign, and the detection and elimination of the exposure to the antigenic agent will bring about the remission of the condition. However, the allergen in most cases is unknown, despite extensive allergy testing. Several pharmacological treatment modalities have been tried in the treatment of PCG such as topical or systemic antihistamines, corticosteroids, or antimicrobials. Treatment may also include excision by laser, scalpel, electrosurgery, and so on.[1,2,4,9,11,14]

CONCLUSION

This study emphasizes on detailed history taking, examination and running appropriate diagnostic aids to arrive at an early and prompt diagnosis for better treatment of the condition. An early diagnosis would enable the clinician toward an appropriate treatment plan, especially in cases where common lesions and symptoms mask underlying serious conditions such as multiple myeloma and leukemia.

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 initial s 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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