Plasma cell gingivitis with aggressive periodontitis: A rare presentation of an uncommon condition

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Abstract

Background: Plasma cell gingivitis is a rare benign condition typified by non-neoplastic infiltrate of plasma cell into the gingival connective tissue. In most of the cases the underlying etiology is allergic reaction to a foreign substance such as cinnamon, chewing gum, spices or dentifrices. The diagnostic workup includes comprehensive history taking, examination and laboratory tests to rule out neoplastic involvement.

Case Details: This paper depicts a rare case of type 3 plasma cell gingivitis in a young male patient presenting as generalised gingival enlargement associated with generalised aggressive periodontitis.

Conclusion: This case underscores the importance of comprehensive history taking and diagnostic investigations for varying clinical presentation of gingival conditions. Since plasma cell gingivitis is generally not associated with loss of attachment, periodontal pockets and bone loss, a plausible explanation of their concurrence is given.

Keywords: Plasma cell; Gingival enlargement; Allergy; Periodontitis

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Introduction

Gingival overgrowth or gingival enlargement is a common trait of gingival diseases featured either isolated or generalised with varied appearances. Numerous types of gingival enlargement are classified on the basis of etiology, systemic factors and pathological findings and its diagnosis may require extensive investigations due to varied clinical and histopathological features. One of the causes of conditioned gingival overgrowth is plasma cell gingivitis (PCG). It is an uncommon condition characterised by diffuse infiltration of non-neoplastic plasma cells in the sub-epithelial tissue. Clinically, it manifests as bright red oedematous gingival swelling usually localized and sharply demarcated from the mucogingival junction[(1,2)]. Previously, different terminologies were used for PCG such as atypical gingivitis, plasma cell gingivostomatitis, plasmacytosis of the gingiva and allergic gingivitis[(3)].

PCG is generally regarded as a hypersensitive reaction to an allergen present in chewing gum, flavouring dentifrice, spices or herbal products[(2,4)]. Almost two decades ago, Gargiulo et al. classified PCG into three types: type 1 caused by an allergen, type 2 of neoplastic origin and type 3 as idiopathic[(5)]. It is a benign condition usually not associated with tissue destructive changes. This case description outlines an unusual presentation of type 3 PCG in association with generalised aggressive periodontitis (GAP).

Case Report

A 25 year old male presented with a chief complaint of generalised swelling of gums with associated mobility of teeth since six months. The patient detailed initial appearance of the swelling on posterior maxillary buccal aspect. Over the period of two months, gingival enlargement occurred in other quadrants with progressive increase in size and was accompanied with progressive mobility of teeth which restricted patient's dietary habits. The medical history was non-contributory. Extraoral examination did not reveal any significant finding. Right and left submandibular lymph nodes were enlarged, mobile and tender. Intraoral examination revealed bright, erythematous, generalized gingival enlargement (grade III), with loss of stippling covering almost two-third of the clinical crown with no area of spontaneous bleeding. Thin deposits of plaque with little calculus were evident **(Fig. 1).** Periodontal condition of teeth was compromised. Generalised pseudo and true pockets with grade II-grade III mobility of teeth was present. Increased probing depth (>6mm) was recorded with grade II furcation (Glickman's classification) in #16 and grade III furcation in #26, #36, #37, #46 and #47. Panoramic dental tomography was noticeable for generalised moderate-severe bone loss (posterior>anterior) **(Fig. 2a)**. The clinical presentation and radiographic findings were consistent with GAP.

Routine hemogram and biochemical tests were with in normal limits and immunoassay for HIV was negative. Sputum and chest radiograph was negative for

tuberculosis **(Fig. 2c)**. Microscopic examination of the representative gingival biopsy tissue showed stratified squamous keratinised epithelium with inflammatory oedematous stroma comprising chiefly of plasma cells with few interspersed lymphocytes. No cytological atypia was evident. These features were consistent with PCG. Gingival enlargement associated with rapid alveolar bone loss and predominance of plasma cells on histology made us consider the possibility of plasma cell dyscrasia for which further investigations were carried out.

FNAC of the lymph nodes was suggestive of reactive hyperplasia. Serum immune electrophoresis showed diffuse band in gamma globulin region with kappa/ lambda free light chain ratio of approximately 2:1, suggestive of polyclonal gammopathy. Myeloma band (M-band) was not detected. Roentgenological examination of skull and cervical vertebrae did not show any lytic lesion **(Fig. 2b).** Laboratory tests revealed no evidence of systemic disease or haematological abnormality **(Table 1)**. Thereafter, to clarify allergy as the cause of gingival enlargement, patient was thoroughly questioned regarding habitual use and/ or recent change in oral dentifrices or mouth fresheners. However, no significant information was obtained.

Thus the clinical, roentgenological and laboratory findings were interpreted as type 3 PCG in association with GAP. The treatment plan included oral hygiene maintenance and conventional periodontal therapy followed by surgical management with regular follow-up every month. However, the patient was lost to follow-up after conventional periodontal treatment.

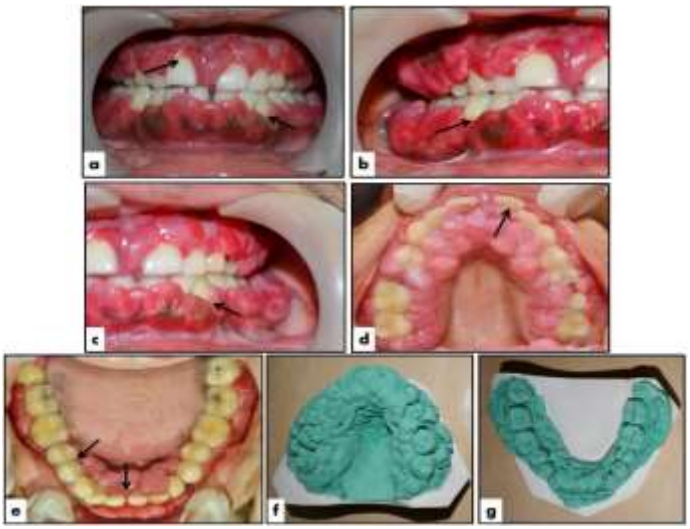

Fig. 1: Intraoral view showing erythematous generalised gingival swelling involving one-third to two-third of the clinical crown with plaque and calculus deposits (arrows) (a-e); dental cast made for pre and post-treatment comparisons (f-g)

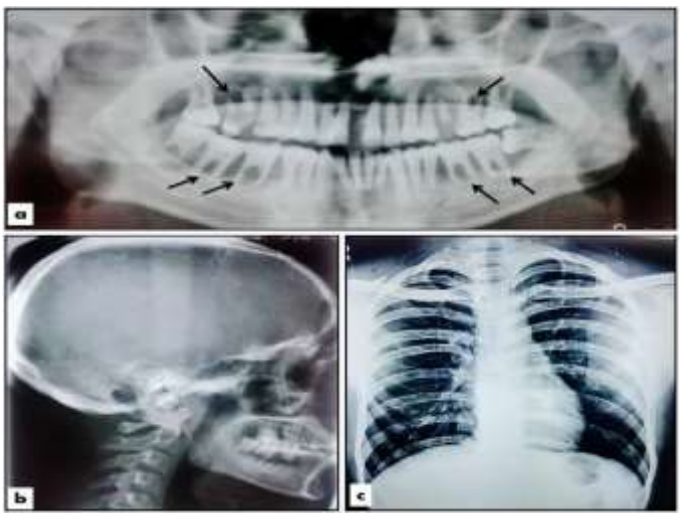

Fig. 2: Panoramic dental tomography showing generalised bone loss extending up to apical third of roots (a); true lateral skull did not show any lytic lesion (b); Chest radiograph findings were negative for tuberculosis (c)

Table 1: Results of laboratory investigations

Test	Value	Normal Range
Hemoglobin	12.7	12-18 gm%
Haematocrit	41	40-55
RBC count	4.8	4-5 million/µl
Total leucocyte count	7800	4000-11000 cells/cumm
Differential leucocyte count		
Neutrophil	76	45-75
Lymphocyte	22	20-45
Eosinophil	02	1-6
Monocyte	00	0-10
Basophil	00	0-2
Platelet	333	150-450 thousand/cumm
ESR (Wintrobe)	45*	0-15 mm/hr
Serum creatinine	0.7	0.7-1.2 mg/dl
Serum Sodium	135	135-145 mmol/l
Serum Potassium	4.6	3.6-5 mmol/l
Serum Chloride	99	98-107 mmol/l
Serum Total T3	1.47	0.95-2.5 mmol/l
Serum Total T4	89.92	60-120 mmol/l
Serum TSH	1.05	0.2-5µIU/ml
Blood urea	23	10-40 mg/dl
Blood urea nitrogen	10.75	7-21 mg/dl
Serum total bilirubin	0.7	0.2-1 g/dl
Alkaline phosphatase	92	30-120 U/L
Serum SGPT	25	5-49 U/L
Serum SGOT	21	6-46 U/L
Serum total protein	7.0	6-8 g/dl
Serum Albumin: Globulin ratio	1.19	1.1-2.0
Serum protein electrophoresis	Polyclonal gammopathy	
Myeloma Band	Not detected	
HIV	Negative	
Sputum	Negative for tuberculosis	
FNAC (lymph node)	Reactive hyperplasia	

Discussion

PCG is an infrequently observed condition typified clinically as gingival enlargement usually in the maxillary anterior region, erythema with or without gingival bleeding and burning sensation[3]. Based on the clinical presentation differential diagnosis included plaque induced gingival enlargement, gingival hyperplasia associated with systemic conditions or diseases and neoplastic causes. Generalized gingival enlargement along with extensive bone loss necessitates extensive diagnostic work-up. In the case depicted here, inflammation of marginal and attached gingiva was refractory to local treatment, hence inconsistent with plaque related etiology. Negative drug history ruled out drug induced gingival enlargement. On occasions, dense infiltrate of plasma cells in the connective tissue can make the case challenging in differentiating benign conditions like PCG from malignant ones such as plasmacytosis of the gingiva and extramedullary plasmacytoma[1]. Hedin et al. inferred PCG as the result of allergic reaction to bacterial plaque, even after its removal by conventional periodontal treatment[6]. Nitta et al. reported a case of marked gingival enlargement with plasma cells infiltrate along with rapidly progressive periodontitis in which the role of periodontopathic bacteria, porphyromonas gingivalis was speculated[7]. Morphologically non-neoplastic proliferative infiltrate of plasma cells characterises histopathological picture of PCG which serves to distinguish it from plasma cell dyscrasia with gingival manifestations. The present case belongs to type 3 as no etiological factor could be established.

GAP is characterised by "generalized interproximal attachment loss affecting at least 3 permanent teeth other than first molars and incisors"[8]. As these features were identifiable in the present case, a diagnosis of GAP associated with PCG was confirmed. Since rapid bone loss is usually not observed in many cases of gingival enlargement, an intriguing question that arises is: between GAP and PCG which occurred first? The answer is usually difficult to trace when the patient reports to the clinician many months after the condition started. As plasma cell is the predominant inflammatory cell population, a hypothetical explanation is put forth in the following section, linking PCG with alveolar bone loss.

The individual variations in gingivitis and periodontitis in response to plaque accumulation are well established[9]. Not every individual with gingivitis will show progression to periodontitis and not all cases of

periodontitis will advance to tooth loss. Patient's susceptibility is the crucial determinant of the clinical outcome. Numerous studies have demonstrated that gingivitis is predominantly a T cell response while periodontitis primarily involve B cells and plasma cells[10]. Aggressive periodontitis (AP) which usually manifests in young individuals (<30years), is histologically characterized by preponderance of plasma cell infiltrate in the connective tissue[11]. The rapidly occurring bone loss in AP is determined in particular by genetic and environmental factors. As advanced periodontitis is marked with increased plasma cell population, their infiltration into the gingival connective tissue can be expected. This would result in oedematous swelling of the gingiva. Therefore, in the present case there is a possibility that GAP could have started earlier and its progression led to plasma cell infiltration into the sub-gingival stroma causing generalised gingival enlargement. However, this is purely a speculation which fails to justify lesions of PCG not associated with alveolar bone loss.

On the other hand, gingival swelling in PCG may result in the accumulation of plaque and calculus providing conducive environment for bacterial proliferation, and breaching the epithelial barrier in established gingivitis. This may result in activated immune response by secretion of various cytokines such as interleukin (IL)-12, IL-18 by dendritic cells, tumor necrosis factor (TNF)-α, IL-6, IL-8, macrophage inflammatory protein (MIP)-1 alpha by gingival fibroblasts and matrix metalloproteinase (MMP), laminins by periodontal fibroblasts. These cells contribute to bone resorption via cytokine production and receptor activator of nuclear factor-κB ligand (RANKL). Further bacterial antigenic challenge activates adaptive immunity with T cells and B cells playing the central role in progression of the lesion[12,13]. Adding to this, the proliferative plasma cell infiltrate in PCG can further aggravate the condition. The activated antigen specific B cells in PCG differentiate into plasma cells which secrete antibodies against the foreign antigen. It has been observed that activated B cells can induce bone resorption in RANKL dependent manner[14] by production and release of different cytokines[15]. Therefore, the antigenic challenge in PCG can aggravate periodontal tissue breakdown in two ways. Firstly, the individual's innate susceptibility along with inability to maintain proper oral hygiene in area of gingival swelling (PCG) and secondly due to the release of inflammatory mediators, cytokines and proteases, activating bone resorption pathways.

Whether PCG appeared first or GAP can only be known with patient's awareness of the disease process since its initial presentation, as this will help in depicting the exact history, signs and symptoms. To conclude, this case underscores the importance of comprehensive history taking and diagnostic investigations for varying clinical presentation of gingival conditions. PCG is the oedematous swelling of the gingiva generally due to an allergic reaction. The presentation of PCG may be localised or generalised depending on the genetic and immune susceptibility of the individual in association with environmental factors. Further, assessment is needed to underpin the mechanism explaining concurrence of PCG with aggressive periodontitis.

Conflict of interest: None declared

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