CASE REPORT

PLASMA CELL GINGIVITIS ASSOCIATED WITH INFLAMMATORY CHELITIS: A REPORT ON A RARE CASE

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ABSTRACT

BACKGROUND: Plasma cell gingivitis (PGC) is a rare disease of gingival tissues which is difficult to treat. It has a higher rate of reoccurrence and needs a detailed and careful analysis of etiology. Further, its association with chelitis is rare, only few cases have been reported and the condition with this presentation poses a diagnostic dilemma.

CASE REPORT: This paper reports a 16 year old male with a complaint of bleeding and swelling of gingiva since 3 years. The gingival enlargement occurred on facial aspect of upper and lower anterior teeth involving attached gingival. He also presented with swelling of both lips which was recurrent and for the same duration of 3 years.

DISCUSSION: Based on clinical features and histopathological findings, a diagnosis of plasma cell gingivitis with inflammatory chelitis was made. A detailed history of possible allergen exposure was taken and patch test was conducted to identify any such allergens. Other conditions were ruled out by blood investigation, detailed medical and drug history.

CONCLUSION: A close collaboration between Periodontist and Dermatologist is essential to manage such a case.

KEYWORDS: Plasma cell gingivitis, inflammatory chelitis

INTRODUCTION

Plasma cell gingivitis (PCG) is a rare inflammatory benign condition of gingival tissue characterized by a marked infiltration of plasma cell into sub epithelial connective tissue (1). It is one of the few diseases known to involve attached gingiva. Other conditions which involve attached gingiva are leukemic enlargement and hereditary idiopathic gingival enlargement. Exact etiology of PCG is still obscure and some believe it is a hypersensitivity reaction to certain allergens such as mint-candy or herbal ingredients of toothpaste, red pepper, cinnamon, clove, khat leaves and food flavouring agents (2, 8).

The occurrence of PCG is associated with a number of allergens and hence it is also known by various names such as Allergic gingivostomatitis, Stomatitis venenata, Irritant contact stomatitis, atypical gingivostomatitis. Granulomatous cheilitis like PCG is a rare disorder of unknown etiology. Possible role of some food additives and metals has been proposed infrequently. We here report a 16 year old boy with plasma cell gingivitis and cheilitis possibly granulomatous secondary to some contact allergen. The coexistence of both conditions together suggests that they have a common etiopathogenesis and the fact that this association is rare needs reporting. However, on conducting a search on medline database for publications in English language on 29 January 2013 with keywords 'plasma cell gingivitis' and 'inflammatory chelitis', we came across only one such case report similar to ours.

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CASE DESCRIPTION

A 16 year old, 45 kg student of Indian nationality was referred from the Department of Dermatology to the department of dental surgery for gingival enlargement and bleeding gums along with swelling of lips. On collecting a detailed medical and drug history, no significant contributing factors was identified and the patient was systemically healthy. The patient had a mixed diet and his blood investigation and radiographic examination were within normal limits. His complaints were bleeding from gums for the past 3 years spontaneously or following minor trauma of brushing. Subsequently, there was enlargement of gingiva along with recurrent swelling of both lips. The gingival enlargement was persistent and progressed slowly to reach its present size and extent involving upper and lower anterior on buccal aspect. Swelling of lips was temporary initially lasting for few days with recurrences after 2-3 weeks involving the upper lip more than the lower one. However, since the past 6 months, it became persistent and was slowly progressing. The patient reported history of not using herbal toothpastes, khat leaves and mint-candy.

On clinical examination, the patient's lips were diffusely enlarged without any surface change, the upper lip more than the lower lip with loss of philtrum. The inner surfaces of both lips were smooth, and on squeezing, did not show any pin-point oozing of saliva. Attached gingiva of facial aspect was involved from mesial aspect of maxillary right second premolar to left second premolar and mesial aspect of mandibular right second premolar to left second premolar. No involvement was reported on lingual and palatal gingiva. Pseudo pockets were present in the upper and lower incisors and in the canine region. Differential diagnosis kept for lip swelling was granulomatous chelitis and plasma cell chelitis, and for gingival enlargement was plasma cell gingivitis. A score of 3.7 of Oral hygiene index (OHI) suggested poor oral hygiene and sub gingival band of calculus was present. Complete blood counts, peripheral blood picture and routine biochemical investigation results were within normal limits. An external bevel gingivectomy was carried out to remove the excess gingival tissue and was sent for histopathology examination. Gingivoplasty was performed to attain scalloped gingival contours. Protective periodontal dressing was applied and the patient was instructed not to brush over it and to return after 7 days. Simultaneously, a punch of biopsy was taken from the mucosal surface of upper lip. The patient was kept on chlorhexidine mouthwash 10 ml twice daily for 1 week. He was also advised to refrain from possible contact with allergens including mint-candy, herbal tooth pastes and spices. After a week, significant reduction in lip swelling was reported and philtrum became visible. Healing of gingival tissue was uneventful.

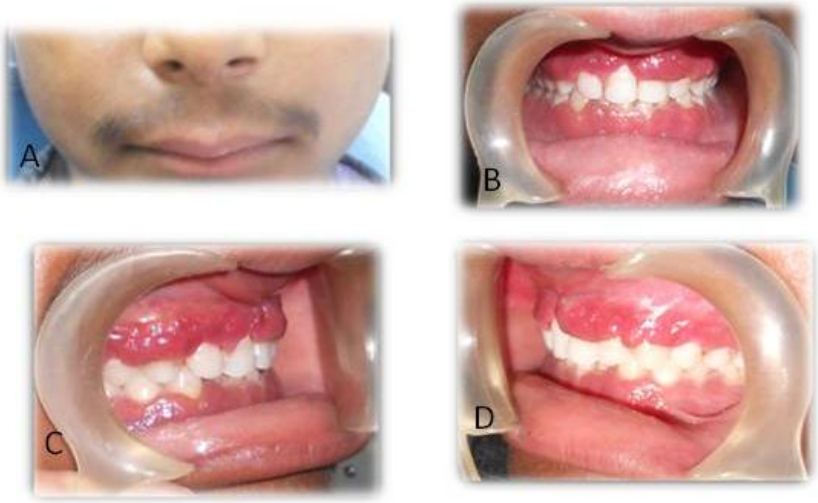

Figure 1: Pre-Op view showing A) chelitis with loss of philtrum and B),C),D gingival enlargement

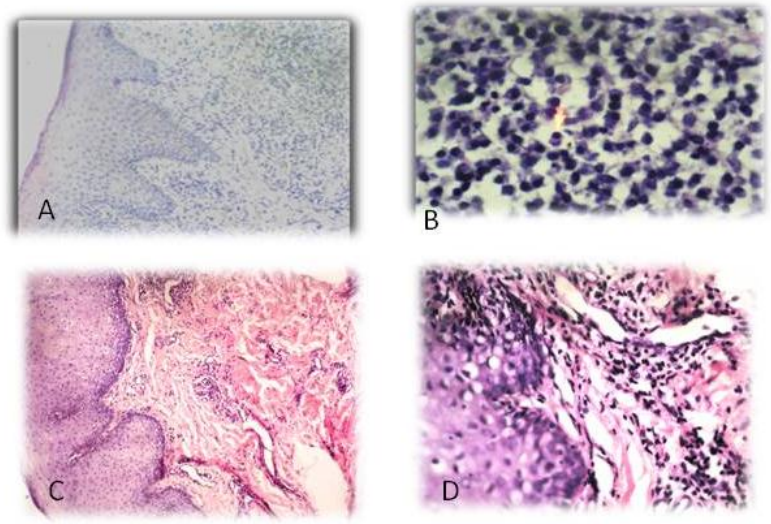

Figure 2: Histopathology: A) gingival biopsy 10 x Hematoxylin and Eosin stain sheets of plasma cell infiltration B) 40 x 10 X plasma cell cart wheel appearance C) lip biopsy 10 x Hematoxylin and eosin stain showing spinous acanthosis; D) 10x40 X showing chronic lymphocytic infiltrates

DISCUSSION

The present case report is of rare disorder of gingival origin associated with granulomatous chelitis. This is a very rare case and to the best of our knowledge is the second case reported in the literature. An interspeciality approach of a periodontist and dermatologist is essential to manage this type of condition.

Plasma cell gingivitis is commonly found in the anterior maxillary region. Leukemic enlargement was excluded as the haematological profile was normal. Hereditary gingival hyperplasia was excluded as no contributing family history was present. Also there was no generalized involvement of attached gingiva. No relevant drug history was present and this excluded the possibility of drug induced gingival enlargement. Granulomatous gingivitis was ruled out by absence of multinucleated giant cells on histopathologic examination, and no association with systemic condition such as crohn disease or wegener granulamatosis was observed. Tuberculosis was ruled out by negative medical history and sputum examination.

Clinical appearance of PCG is striking; it is presented as fiery red erythematous lesion involving the attached gingival extending to mucogingival junction (9, 10). Lesion resembles leukemia infiltration, lichen planus discoid lupus, cicatricial pemphigoid and myeloma. Hence, early diagnosis is essential for effective patient management. Diagnosis is made by selective exclusion through haematological screening, allergen test and histopathological examination.

Remission of lesion on removal of suspected allergen confirms the allergic gingivosomatits when allergen is not identified. It is termed idiopathic gingivostomatitis (11). On conducting a literature search on pubmed data base, we came across only one case report similar to ours. Janam et al reported a 15 year old female with a complaint of mild reddish discoloration involving her upper lip around 5 years back, which progressively increased in size without any apparent discomfort. Gingival changes started at around the same time, the gingiva appearing fiery red and friable with an edematous consistency and granular surface texture (12). Their finding is similar to our case, in which we report a 16 year old male with a similar complaint.

Unlike the case reported by Janam et al, in the present case a lip biopsy was taken to confirm the lip lesion. The histopathological examination gave us a definitive and valid diagnosis for lip lesion. Also, an Indian standard series allergen patch test was done to rule out any standard allergen as a cause for present case. Furthermore, our case report provided a follow-up time of more than a year to show stable result of surgery.

Gingivitis, chelitis and glossitis have been described as a triad for Plasma cell

gingivostomatitis (1). In the present case, removal of gingival lesion by gingivectomy resulted in reduction of lip swelling. This suggests that the combined lesion of lip and gingiva could be due to contact dermatitis as inner aspect of lip was in close contact of gingiva.

Granulomatous chelitis, actinic chelitis and plasma cell chelitis was considered as differential diagnosis for lip lesions (14). Actinic chelitis was excluded as the outer surface of lips was not involved. Plasma cell chelitis and granulomatous chelitis were excluded by histopathological examination.

Cinnamonaldehyde in chewing gums was reported by Kerr *et al*. as an etiological factor for plasma cell gingivitis, chelitis and glossitis (3). The condition regressed completely on discontinuity of the use of these gums. Our case report showed similar results to that of Janam et al for lip and gingival lesions (12). Results regarding gingival lesion were similar to that of Owings, Silverman, and Farrier (11, 13, 14).

Histopathology of gingival tissue revealed dense infiltrate of plasma cell in subepithelial connective tissue with peculiar cart wheel appearance and that of lip showed chronic inflammatory infiltrate of lymphocytes without specific granulomas. Thus, the final diagnosis was plasma cell gingivitis with inflammatory cheilitis. However, absence of granulomas in lip biopsy cannot be taken as strong evidence against granulomatous cheilitis as they are not always present and history of initial remissions and relapses was in favour of granulomatous cheilitis. Patch test was done using Indian standard series and proved negative. No reoccurrence since last 1 year and reduction in lip swelling was reported. Patient is currently not under any medication and is kept on close follow up of 3 months.

The contour and texture of gingival tissues obtained after surgery were stable and no reoccurrence was seen even after 1 year. Lip swelling gradually came down and the lip lesion has also regressed completely and stably for past 1 year. The patient is kept on close follow up of 3 month. Diagnosis by clinical exclusion, haematological and histopathological examination helps to arrive at diagnosis of PCG and inflammatory chelitis. Interdepartmental management of case is crucial to attain treatment success and close follow-up is required to maintain the results achieved.

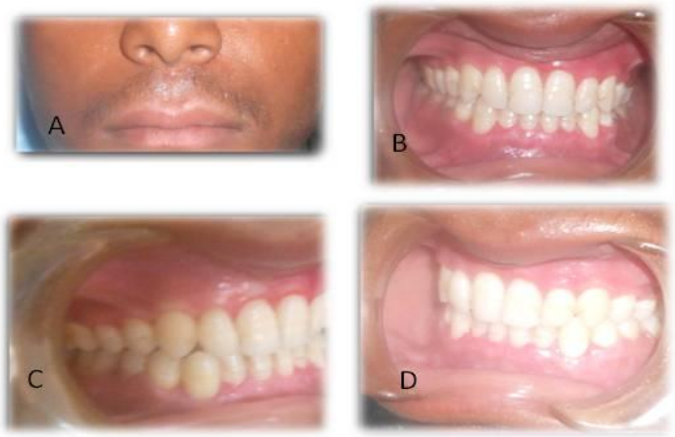

Figure 3: Post op view 12 months A) reduction in chelitis with visible philtrum B),C),D) stable gingival contours and health

Clinical Significance: Plasma cell gingivitis is a difficult entity to manage. It has a high recurrence and poses a severe esthetic and functional problem on the patient, and further association with chelitis make it more problematic for the patient. Finding the allergen and its discontinuation is a prime step in the successful management of the case.

Association of PCG and Chelitis is very rare and proper involvement of Periodontist and Dermatologist is necessary to treat the condition.

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