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Case Report

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Cheilitis Granulomatosa – A Rare Case Report



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ABSTRACT

Cheilitis granulomatosa (CG) is a disorder affecting the orofacial region with a clinical picture of diffuse, firm, non-tender swelling of the one or both the lips that may or may not occur in conjunction with gingival enlargement. As the etiology remains unknown, the treatment of CG remains challenging. Delay in diagnosis or treating CG leads to permanent induration of the lip that compromises esthetic appearance and impairs function. This paper presents a case of a successfully treated patient with an isolated lesion of CG affecting the lip and the gingiva of a 22-year-old female patient.



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INTRODUCTION

Cheilitis granulomatosa (CG) is a rare disorder with multiple differential diagnosis, underlying systemic diseases and infrequent oral manifestations.

CG is a condition that leads to a painless, symmetric edema of the orofacial tissues. It was first described by Miescher in 1945. It presents in two clinical forms: an isolated lesion or as an incomplete variant of Melkersson-Rosenthal syndrome. The differential diagnosis include allergy, as a manifestation of Crohn's disease, infections such as tuberculosis and sarcoidosis.^{1, 2}

CG is clinically characterized by diffuse increase in the size of the lips, rarely associated with swelling of the gingiva and periodontal disease. The present case has a rare clinical appearance of gingival enlargement with periodontitis along with maxillary lip swelling.^{3, 4, 5}

Case Report

A 22-year-old female presented with diffuse gingival enlargement and associated upper lip swelling with multiple fissuring .She also presented with gingival bleeding, difficulty in pronunciation and mastication and refrained from brushing for 1 year. The dental history indicated that the swelling of the lip occurred prior to the gingiva with no contributory medical, drug, allergic and family history. Also, no traumatic injury to the affected site or masochistic habits was noted.

Extraoral examination revealed diffuse, non-tender, soft, symmetrically swollen upper lip with multiple fissures and prominent median fissuring on its vermillion border (Fig.1). Intraoral examination revealed diffuse, soft, edematous enlargement involving the interproximal, marginal and attached gingiva of the maxillary and mandibular anterior teeth along with the presence of local factors. The degree of enlargement was scored varyingly between Bokenkamp II and III. Generalized exudation with combined pockets ranging between 5-7mm was present (Fig 2). The orthopantogram verified horizontal bone loss of the affected area. Complete hemogram revealed a normal blood picture with increased erythrocyte sedimentation rate of 43 mm/hr. A provisional diagnosis of cheilitis granulomatosa with chronic periodontitis was given considering the history, clinical and radiographic evaluation.

An incisional biopsy of the lip was recommended to establish an accurate diagnosis which the patient refused.

The differential diagnosis included tubercular gingival enlargement, sarcoidosis, Crohn's disease, angioedema, Melkersson-Rosenthal syndrome.

Topical application of clobetasol propionate 20mg once daily for the lip swelling for a period of 2 weeks was prescribed. Thorough scaling and root planning were performed. After a month of phase I therapy, the gingival tissues were firm owing to the reduction of the edematous component of the enlargement (Fig.3). Thereafter, internal bevel gingivectomy was performed to eliminate the combined pockets in the mandibular anterior region (33-43) under local anesthesia (Fig 4). After a week, during suture removal, patient presented with delayed healing. She was referred to the physician who prescribed victofol 100mg once daily for 3 months. During the second surgical intervention of the maxillary anteriors, better postoperative healing was noted (Fig 5). However, complete healing was achieved at one month. The patient was placed on regular follow up for 6 months. During this period, there was complete resolution of the lip and gingival enlargement (Fig 6,7).

DISCUSSION

The etiology of CG has not been clearly identified. Hence, other granulomatous conditions like tuberculosis, sarcoidosis, Crohn's disease, angioedema and Melkersson-Rosenthal syndrome were ruled out before making a diagnosis of cheilitis granulomatosa. Tuberculosis was ruled out on the basis of history, chest radiograph and mantoux test. Sarcoidosis was excluded on the basis of chest radiograph and serum angiotensin converting enzyme levels. Crohn's disease was eliminated on the basis of blood investigations and absence of signs and symptoms of gastrointestinal disorders. Absence of allergic history and blood investigations was the criteria for the exclusion of angioedema. Melkersson-Rosenthal syndrome was ruled out due to the absence of fissured tongue and facial paralysis.⁵

The treatment of CG aimed at reducing lip swelling, improving the esthetic appearance, elimination of periodontal diseases and restoring its health.

Various medicaments like systemic antibiotics, systemic, intralesional topical corticosteroids are used to treat lip swelling. Vano -Galvan *et al* administered corticosteroids (triamcinolone acetonide 10mg/mL) and achieved plausible improvement after three months.⁶ Mirjana *et al* treated CG using chymoral forte and achieved complete remission in the first five days of the

treatment. In the present case, the patient was treated with topical application of clobetasol propionate twice daily for four weeks during which she showed a progressive decrease in the lip swelling.³

The patient was supplemented with Victofol 100mg by the physician because she presented with delayed healing after first internal bevel gingivectomy procedure. However, healing was improved with second surgical intervention with the support of victofol supplementation. This may be because folic acid supplementation increases keratinization and maturation of epithelial cells by increasing DNA synthesis and aid in healing.⁷

CONCLUSION

Merely treating a presenting symptom is not a clinical challenge. Establishing a diagnosis, eliminating systemic involvement and providing an appropriate treatment is a daunting task. Early diagnosis of CG is challenging due to its resemblance with other chronic granulomatous disorders. Thus, dentists may be the preliminary person to diagnose the lesion if systemic involvement is present and play a pivotal role in the multidisciplinary treatment of granulomatous disorders.³

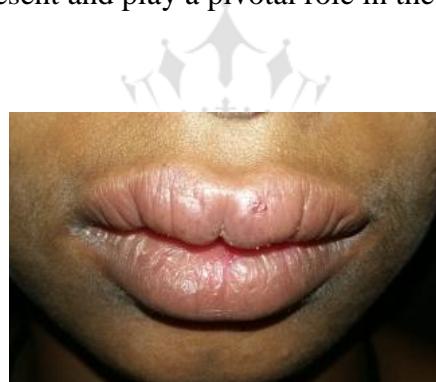


Fig. 1 Extraoral lip swelling



Fig. 2 Gingival enlargement (Bokencamp Classification)



Fig. 3 After phase I therapy



Fig. 4 Postoperative follow up – eventful healing with mandibular anteriors



Fig. 5 Postoperative follow up of maxillary anterior



Fig. 6 Resolution in gingival enlargement



Fig. 7 Resolution of lip swelling

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