10.5005/jp-journals-10037-1025

CASE REPORT

Cheilitis Glandularis Treated with Intralesional Steroids: A Rare Case Report and Review

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ABSTRACT

Cheilitis glandularis (CG) is a rare disorder characterized by swelling of the lip with hyperplasia of the labial salivary glands. CG is most commonly seen in the lower lip, in middle-aged to older Caucasian men; however, rarely reported in Asians. The exact cause of CG is unknown, but various agents have been attributed like smoking, chronic irritation, poor oral hygiene, allergy, bacterial infections, syphilis, chronic exposure to sunlight and wind, compromised immune system, and genetic transmission. In this paper, we report a rare case of CG of the superficial suppurative type in a 43-year-old Indian female affecting lower lip which was diagnosed based on clinical and histopathological findings and was treated with intralesional steroid injections. Even though the incidence and occurrence of CG in clinical practice is rare, it presents a diagnostic challenge to dentists because the etiologic factors are uncommon with variations in clinical presentation. Early diagnosis with definitive treatment and frequent monitoring should be carried out to prevent further complications.

Keywords: Cheilitis glandularis, Suppurative, Minor salivary glands, Intralesional steroids.

How to cite this article: Mohammed RB, Rani GS, Sandya MP, Jyotsna S. Cheilitis Glandularis Treated with Intralesional Steroids: A Rare Case Report and Review. Oral Maxillofac Pathol J 2014; 5(2):510-513.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

Cheilitis glandularis (CG) is a rare chronic inflammatory disease affecting the minor labial salivary glands and characterized clinically by edema and focal ulceration.[1,2] The term CG was first used by Volkmann in 1870 to describe a disorder that presented as a chronic, suppurative inflammation of the lower lip, characterized by swelling of the mucous glands and associated with mucopurulent discharge through dilated ductal openings.[3] CG is a rare condition most commonly seen in the lower lip of adult male with little predilection to female and children,[4] in middle aged to older Caucasian men;[5] Japanese and Asian cases are very rare. In the literature, three types of CG have been described, the simple, superficial suppurative and deep suppurative.[6] However, it is now believed that these represent three stages in progression of a single disease entity.[7] Clinically, CG may resemble many other conditions. Differential diagnoses includes multiple mucocele, chronic sialadenitis of the minor salivary glands, sialolithiasis of minor salivary gland, factitious cheilitis, actinic cheilitis, cheilitis granulomatosa, angioedema, and benign and malignant salivary gland tumors.[2,8]

Histologically, CG is a nonspecific chronic inflammatory lesion characterized by localized dense accumulation of inflammatory cells and inflammation of mucous glands associated with glandular distension and ductal dilatation with notable loss of acinar structure.[9] Roda and Yacobi and Brown consider duct ectasia and inflammation of minor glands and adjacent tissue to be the essential microscopic features of CG.[10, 11]

Various treatment modalities have been described for CG based on clinical presentation, diagnostic information obtained from histopathologic analysis and the identification of likely etiologic factors responsible for the disease. It includes administration of antihistamines, antimicrobials, topical or intralesional steroids[12,13] and topical immunosuppressants. In cases with severe dysplasia, surgical stripping, vermilionectomy, cryosurgery, laser surgery, or topical chemotherapy with 5-fluorouracil found to be the effective treatment modalities.[4,12]

Herein we report a rare case of CG of superficial suppurative type in a 43-year-old Indian female, successfully treated with intralesional steroid injections.

CASE REPORT

A 45-year-old Indian female reported with burning sensation and ulceration of lower lip since 1 year. She had history of trauma to lower lip region 2 years back which later healed

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with pigmentation. After a year, she developed a small ulcer in the middle of lower lip and it spread to the corner of mouth on both sides. She also noticed swelling of lip with frequent alteration in size. Even on slight touch, there was bleeding from lip region. During night times, lips stick together and early morning she had severe pain and bleeding with dry, cracked lower lip. She consulted general physician for the same problem but the lesion did not subside on using the medication prescribed. She was not allergic to any drugs. She also revealed that she was a house wife with no frequent sun exposure.

On clinical examination, lips were dry, incompetent and lower lip presented with well-defined solitary ulcerated lesion above vermillion border of size 5 × 1 cm with bloody crustations. Lower lip was inflamed and swollen. On palpation, lower lip was edematous, soft in consistency with exudates over the lesion and severe bleeding from the cracks even on slight touch with moderate tenderness (Figs 1 to 3). Right submandibular lymph nodes were palpable but asymptomatic. Oral mucosa was not affected.

On the basis of above clinical findings, it was provisionally diagnosed as CG affecting lower lip. Differential diagnosis included was actinic cheilitis, cheilitis granulomatosa and syphilitic ulcer. Routine blood investigations performed were within normal limits except for elevated erythrocyte sedimentation rate. C-reactive protein levels, serum glucose, hepatic and renal parameters were normal. The treponema pallidum hemagglutination test, the HIV-screening test was nonreactive. Incisional biopsy was done to confirm the diagnosis.

Histopathology revealed nonspecific inflammatory infiltration with lymphocytes, plasma cells and neutrophils. The labial glands were hyperplastic with focal infiltration of lymphocytes and plasma cells with ductal dilatation and mucin pooled areas (Fig. 4). Treatment consisted of administration of intralesional steroid injections of dexamethasone

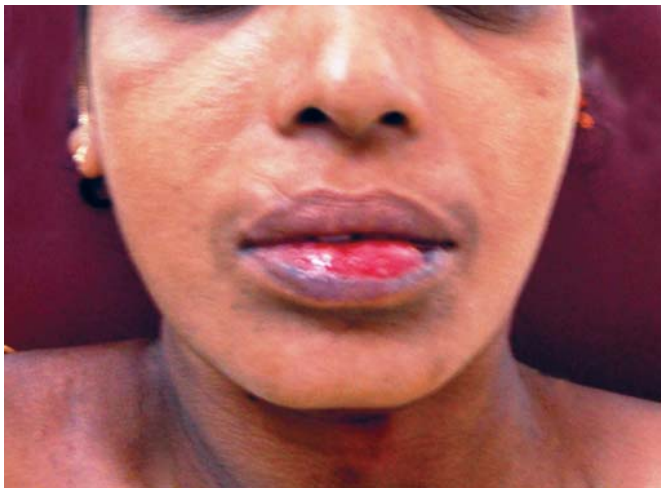

Fig. 1: Extraoral photograph showing ulcerated lower lip

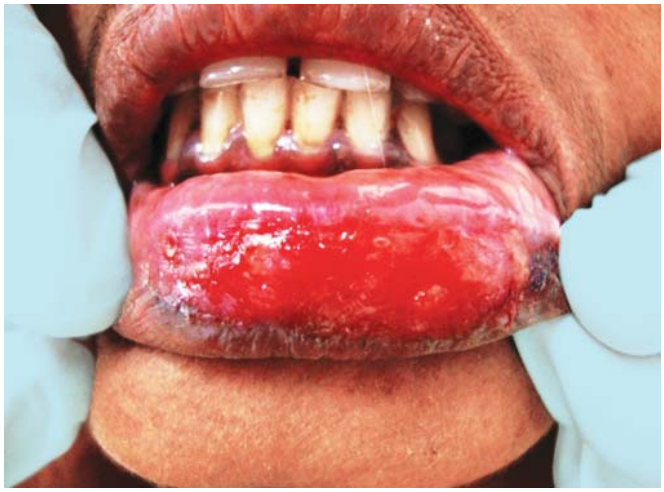

Fig. 2: Inflamed, swollen, edematous lower lip with exudates over the lesion

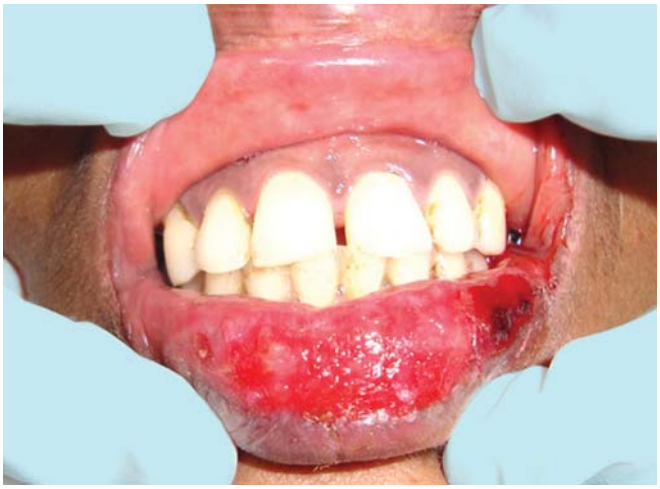

Fig. 3: Severe bleeding from the lower lip even on slight touch

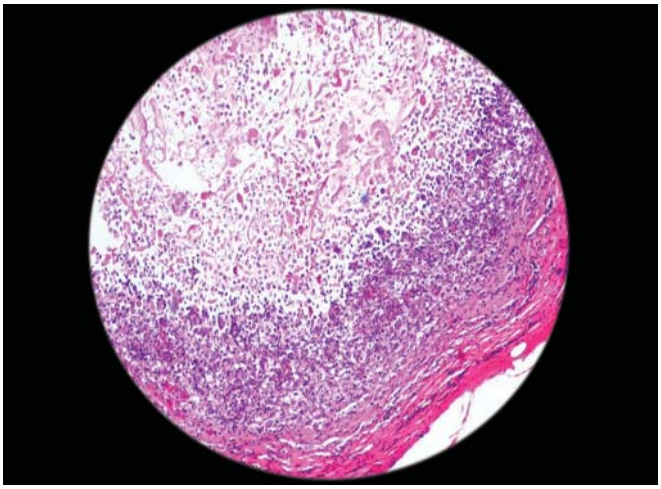

Fig. 4: Photomicrograph showing nonspecific inflammatory infiltration with lymphocytes, plasma cells and neutrophils. The labial glands were hyperplastic with focal infiltration of lymphocytes and plasma cells with ductal dilatation and mucin pooled areas (H & E, ×10)

2 ml weekly twice for 1 month along with analgesics and topical antimicrobial metronidazole gel. The lesion resolved spontaneously within 3 months and recurrence was not noticed during 3 years of regular follow-up (Figs 5 and 6).

DISCUSSION

Cheilitis glandularis is a rare disorder characterized by swelling of the lip with hyperplasia of the labial salivary glands. Sutton stated that the characteristic lip swelling was attributable to a congenital adenomatous enlargement of the labial salivary glands.[14] Swerlick and Cooper stated that there was no evidence to support the assertion that submucosal salivary gland acinar hyperplasia is either responsible for or a consistent feature of established CG.[15]

The etiology of CG is still unknown, but it has been suggested to be an autosomal dominant hereditary disease for which smoking, chronic irritation, poor oral hygiene, allergy, bacterial infections, syphilis, chronic exposure to sunlight and wind, compromised immune system, genetic transmission are predisposing factors.[2,5,16] The present case was unusual because of its occurrence in Indian female with no obvious etiology detected.

Cheilitis glandularis most commonly affects lower lip, in middle-aged to older Caucasian men. Schauermann classified CG into three types: simple, superficial suppurative, and deep suppurative. The simple type consists of multiple painless lesions that exhibit openings and dilated ducts. If these lesions are infected, the disease may progress to the superficial or deep suppurative types. Superficial suppurative type is characterized by painless crusting, swelling and induration of the lip with superficial ulceration. The surface mucosa produces clear to cloudy fluid at the sites of ductal openings. All these features were associated with the present case. Deep suppurative CG is a deep seated chronic infection accompanied by abscess formation and fistulous tracts.[6,7] The latter two types have the highest association with dysplasia and carcinoma.

Differential diagnosis may include actinic cheilitis, atopic dermatitis, angioedema, exfoliative cheilitis, chronic sialadenitis of the minor salivary glands, factitious cheilitis, and cheilitis granulomatosa.[17] Reported cases of CG were associated with other clinical conditions. Stoopler et al[8] reported a papillary cystadenoma like ductal growth pattern in one patient with CG. Musa et al[2] reported a case with a more generalized presentation of CG, termed as suppurative stomatitis glandularis. Reichart et al[18] reported a retention cyst in the upper lip of an elderly patient with the 'simplex' form of CG. Leao et al[1] reported a case with CG which was later discovered to have undiagnosed HIV infection. Butt et al[19] reported a case of CG that progressed to squamous cell carcinoma in an HIV-infected patient. Carrington and Horn[20] reported a case of CG related to actinic damage following vermilionectomy for squamous cell carcinoma of the lower lip. Dhanapal et al[21] reported a case in which a 14-year-old girl with double lip developed CG, raising a possibility of a genetic predisposition for occurrence of CG.

Clinical investigations should be done to rule out neoplastic, immune-suppressive, or inflammatory changes due to local factors. CG histologically characterized by dilated and tortuous minor salivary gland ducts, many of which are lined by oncocytic cells showing foci of hyperplasia and mucous metaplasia. Other features include acinar atrophy with infiltration of the glandular parenchyma by chronic inflammatory cells and the presence of extravasated mucin.[16]

Reiter et al[17] proposed the diagnostic criteria for establishment of a diagnosis of CG. The present case showed both the clinical and histopathological criteria and hence was finally diagnosed as CG of superficial suppurative type involving the lower lip.

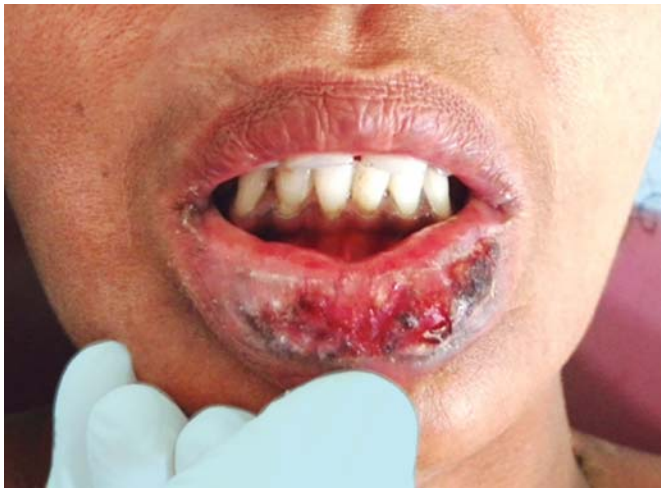

Fig. 5: Follow-up photograph showing scarring of the lesion in 1 month

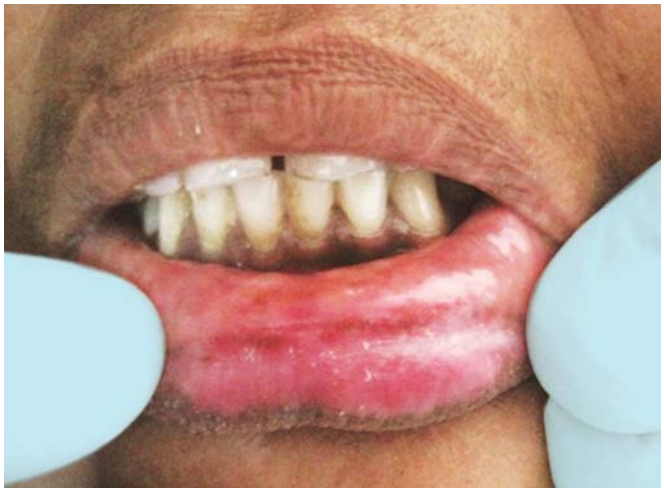

Fig. 6: Follow-up photograph showing complete healing of the lesion after 3 months

Different treatment modalities have been reported for CG and vary accordingly depending upon the predisposing factors, including antibiotics, antihistamines, steroids, immunosuppressants, radiotherapy, surgical stripping, vermilionectomy, cryosurgery, or laser surgery or topical chemotherapy with 5-fluorouracil. Recurrence is rare after surgery. As no obvious predisposing factor was elicited in the present case, intralesional steroid (inj. Dexona 2 ml) was administered at lower lip region with topical antimicrobial application (Metrohex gel) along with systemic analgesics that led to spontaneous resolving of the disease in a month with no recurrence noticed for 3 years. Similar to present case, Schweich and Haldar[22,23] also reported considerable improvement of superficial suppurative CG after treatment by intralesional steroid injections.

CONCLUSION

Cheilitis glandularis represents as an unusual disease, characterized by painless swelling of lips with mucoid draining pores that can be rarely seen in clinical practice. The co-occurrence of lower lip CG lesions and actinic damage should alert for increased susceptibility to develop squamous cell carcinoma. So, early diagnosis and effective treatment with frequent monitoring should be done to prevent further complications of the disease.

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