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Letter to Editor

Glandular Cheilitis Showing Excellent Response with Therapeutic Combination of Intralesional Steroid with Topical Tacrolimus

Parul Bansal[1], Akash Agarwal[1], Maitreyee Panda[1]

[1]Department of Dermatology, Institute of Medical Sciences and Sum Hospital, Bhubaneswar, Orissa, India.

***Corresponding author:**
Akash Agarwal,
Department of Dermatology,
Institute of Medical
Sciences and Sum Hospital,
Bhubaneswar, Orissa, India.

akash.22.1995@gmail.com

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Dear Editor,

A 16-year-old male presented to the dermatology outpatient department with persistent lip swelling associated with intermittent pain and difficulty in lip movements for 2 months. Complaints of adherent crust formation along with lips sticking together on waking up in the morning hours were also present. There was no history of altered bowel movements or fever with chronic cough associated with night sweats. The patient denied a history of repeated lip licking, or manipulation of lips. Previous treatment with oral and topical antibiotics was not satisfactory. There were two similar episodes in the past which resolved on itself within 1 month. On examination, multiple thick yellowish crusting overlying a background of erythematous oedematous lips was seen, predominantly over the lower lip. On palpation, lips were rubbery in consistency. There was no evidence of scarring or lip deformity. On pressing the lips, a few areas depicted extrusion of mucoid discharge [Figure 1a]. On soaking the lips with saline, crusts were removed with difficulty which revealed a glistening erythematous oedematous surface [Figure 1b]. Oral cavity examination revealed poor oro-dental hygiene associated with a foul smell. No other mucosae were involved.

Differential diagnoses of glandular cheilitis, granulomatous cheilitis, contact cheilitis, exfoliative and actinic cheilitis were considered. Granulomatous cheilitis was considered due to persistent oedematous lip swellings; however, it was ruled out due to background erythema, pain and excessive crusting. The diffuse involvement of both lips and a lack of sandpaper-like feel or ulceration over lips were against the diagnosis of actinic cheilitis. A bacterial culture from the discharge showed no growth. Mantoux test was non-reactive and chest X-ray revealed no abnormality. Routine blood investigations including C-reactive protein and erythrocyte sedimentation rate were within normal limits. A punch biopsy from the lower lip revealed focal micro abscess formation with lymphohistiocytic inflammatory infiltrate; however, no minor salivary gland or granuloma could be identified [Figure 2a and b]. There were no features of solar elastosis, or epidermal dysplasia ruling out components of coexisting actinic cheilitis. The patient was apprehensive regarding a repeat biopsy. A patch test was performed which was negative. The neuropsychiatric evaluation did not reveal any underlying psychological cause. Based on the above findings, a clinical diagnosis of a simple type of glandular cheilitis was made. He was started with intralesional triamcinolone acetonide (ILTAC) 40 mg, topical tacrolimus and regular petroleum jelly application. The patient reported significant improvement within 1 week of the above medication [Figure 3a]. He is currently under remission with once-a-month ILTAC and

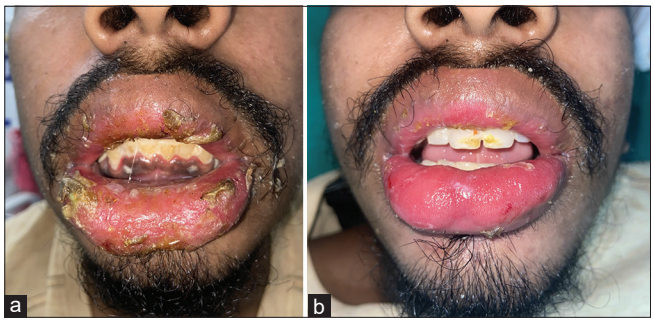

Figure 1: (a) Erythematous oedematous swelling of lips with adherent yellowish crusts showing mucoid discharge from a few areas on squeezing. (b) Underlying erythematous oedematous glistening surface of both lips on removal of crusts with saline-soaked gauze.

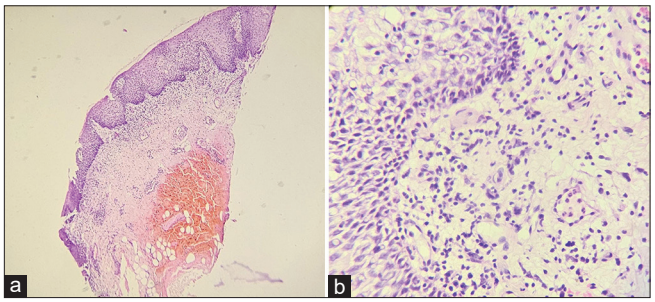

Figure 2: (a) Photomicrograph reveals stratified squamous epithelium with irregular acanthosis, oedematous subepithelial stroma with areas of haemorrhage in deeper aspects of the stroma [Haematoxylin and eosin (H&E) stain, ×10]. (b) Irregular acanthosis with mild transcytosis of lymphocytes and polymorphs with perivascular and interstitial inflammation comprised of lymphocytes and plasma cells. No granuloma or minor salivary glands could be found (H&E stain, ×40).

topical tacrolimus therapy [Figure 3b]. He has been advised for strict photoprotection and to avoid lip licking and smoking.

Glandular cheilitis is an uncommon idiopathic inflammatory condition involving the minor salivary glands of the lips. Classically, three subtypes have been described which include – simplex, superficial suppurative and deep suppurative.[1] The disease is commoner in men with a 1.8:1 ratio. It is regarded as a pre-malignant condition especially the deep suppurative variant with reports of its association with squamous cell carcinoma. Chronic ductal hyperplasia and ectasia drive the disease pathogenesis with inflammation of the glandular parenchyma, leading to a flow of mucin-rich saliva from the ductal openings that leave crusted lesions on drying.[2]

According to the criteria proposed by Reiter *et al*., only two clinical and one histopathological criterion (presence of chronic inflammation) was found in our case. No visible

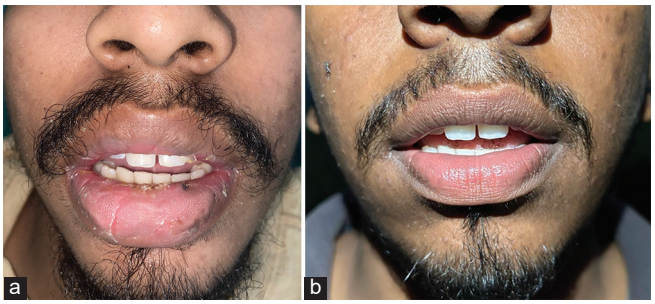

Figure 3: (a) Significant improvement in erythema and oedema after 1 week post-intralesional triamcinolone injection and topical tacrolimus application. (b) Complete remission observed at 1-month follow-up.

minor salivary glands or ducts could be identified. In many cases of glandular cheilitis, the ducts can be normal in size, depth and histology; therefore, a biopsy might not always be diagnostic.[3]

Managing glandular cheilitis poses challenges due to its chronic relapsing nature. Various treatments have been attempted; however, definite guidelines are lacking. Topical options encompass corticosteroids, calcineurin inhibitors and antibiotics such as fusidic acid and mupirocin. Oral agents, including doxycycline, clofazimine and oral steroids, aim to reduce inflammation. Few reports of successful therapy with intralesional steroids have also been documented. Sugaya and Migliari reported significant improvement in a single case following two injections of 10 mg triamcinolone 1 month apart followed by topical tacrolimus ointment as maintenance therapy.[4] Lederman reported treating a case of recurrent glandular cheilitis with intralesional dexamethasone weekly which resulted in a reduction of the severity of the swelling over 4 weeks.[5] Surgical intervention involving labial vermilionectomy and dissection of minor salivary glands is often the therapy of choice. Here, in our case, we used a combination of ILTAC injection and topical tacrolimus with rapid improvement and sustained remission.

Glandular cheilitis represents a rare clinical entity that needs to be appropriately diagnosed and promptly treated given the functional and psychological impairment. Our case depicts the successful use of a less described cost-effective therapeutic modality for the management of this rare condition.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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