A case report of diffuse lip swelling: Our approach to diagnosis and management

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ABSTRACT

Lip swellings may present as a common finding in clinical practice, and the causative factors could include a variety of local and systemic factors. Here we report a case of recurrent lip swelling in a 48-year-old male patient of Indian origin with no specific etiology or signs. The patient underwent a detailed investigation, and the diagnosis was made as ''cheilitis glandularis-CG'' on histopathological examination. CG is a rare disorder characterized by hyperplasia of the labial mucous glands and heterotopic mixed salivary glands as well as ectasia of glandular ducts. The patient was treated successfully with oral and topical corticosteroid with satisfactory improvement. The present case may provide an insight to the clinicians for a collaborative approach in the diagnosis of lip swelling and its management.

Introduction

Lip swellings may be a sign of systemic diseases with a known fact that more than 180 conditions can manifest as lip swellings and the pertinent diagnostic features do not always appear simultaneously (1). The etiology can be simplified by grouping the findings into broad categories such as trauma, inflammation, infection, metabolic diseases, neoplasm and idiopathic conditions which should all be considered in the differential diagnosis and ruling them out may require detailed investigation to arrive at a definitive diagnosis (2). Lip swelling can be a perplexing presentation, and common etiologies must first be excluded before more invasive and expensive testing is done. Other granulomatous and edematous causes of lip swelling must be investigated prior to diagnosis (3)

In our case the 48 year old male patient presented with a complaint of recurrent lip swelling without a clear etiology and non specific symptoms that responded to medications temporarily which indicated a comprehensive work up of the patient. Thus the present case highlights the prominence of approaching such lesions with broader perspectives to eliminate specific granulomatous diseases that could predispose to lip enlargement .The article also reveals the implication of lip biopsy to confirm the diagnosis of cheilitis glandularis which was effectively treated with a combination of oral and topical corticosteroid.

Presentation of Case

A 48 year old male patient complains of swelling and burning discomfort in the upper and lower lip region since 3 months. The swelling gradually initiated from the left side of the lower lip with no change in size and the onset was without a clear identifiable precipitating event. There was no history of trauma in the region and the family history was not contributory. The patient was an employee at a medical store and would chew betel quid occasionally. He was being treated with prednisolone 10mg, nimesulide 100 mg and serratiopeptidase 10 mg which subsided the swelling markedly. He was also being treated with fexofenadine 180mg for a suspected fungal infection of the toe since 6 months. Within a week the lip swelling reappeared with a sensation of rawness and the patient reported to the department of oral medicine.

On examination there was no facial asymmetry seen and the skin extraorally was intact. Prominent crusted

areas with dry scaly appearance was noted. The left side of the lower lip appeared diffusely swollen measuring about 1.5cms whereas the upper lip swelling was not discernible. Lower lip appeared erythematous along with multiple fissures and there was irregularity in the contour of lip mucosa. The junction between the vermillion border and lip mucosa was obfuscated. (Figure 1,2 &3). On palpation the swelling was non-tender, soft, compressible continuous with overlying mucosa with no pus discharge or bleeding from the region. There was absence of bruit and no rise in temperature. Intraorally mild gingival enlargement with glossy appearance was seen. There was absence of stippling, the dentition was intact and oral hygiene was satisfactory. All other sites appeared normal. There was no cervical lymphadenopathy.

Based on the aspect of the lesions, the case was clinically diagnosed as cheilitis granulomatosa with a suspicion of an underlying granulomatous condition initiating the lip swelling. The differential diagnosis of oral crohns, angioedema, actinic cheilitis and cheilitis glandularis was considered. Local factors orally were excluded primarily on the basis of the patients orthopantamograph (OPG) which revealed no odontogenic factors contributing to the lip swelling (Figure 4).

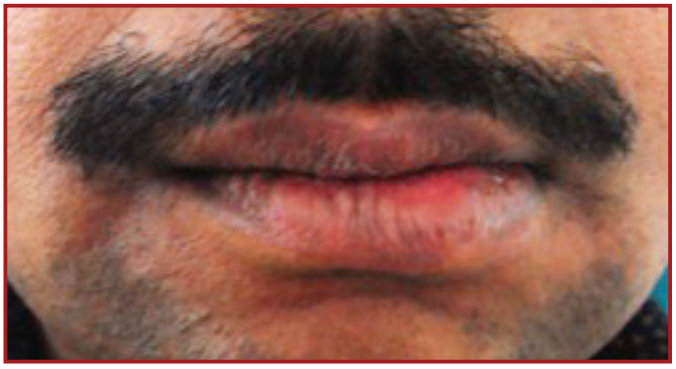

Figure 1. Clinical image shows diffuse swelling of the lower lip with dry scaly areas and multiple fissures

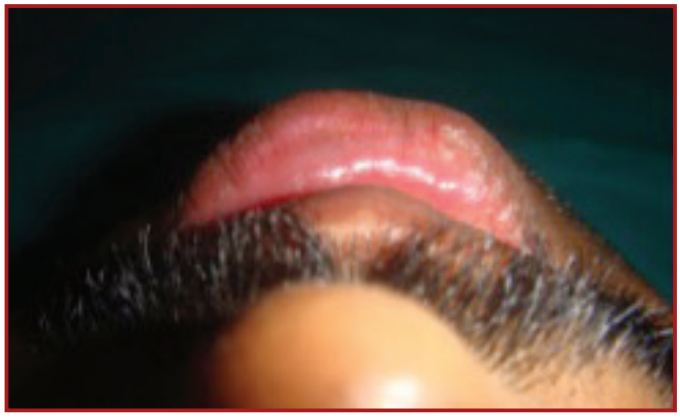

Figure 2. Clinical image shows swelling and irregularity of the lip contour in the axial view

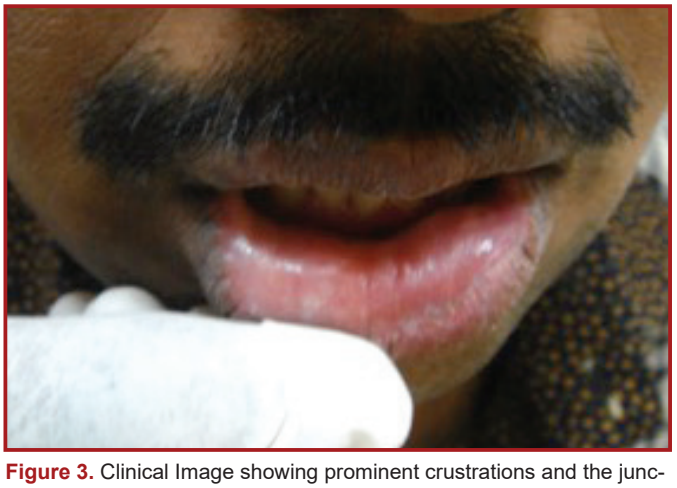

Figure 3. Clinical Image showing prominent crustrations and the junction between the vermillion border and lip mucosa was obfuscated.

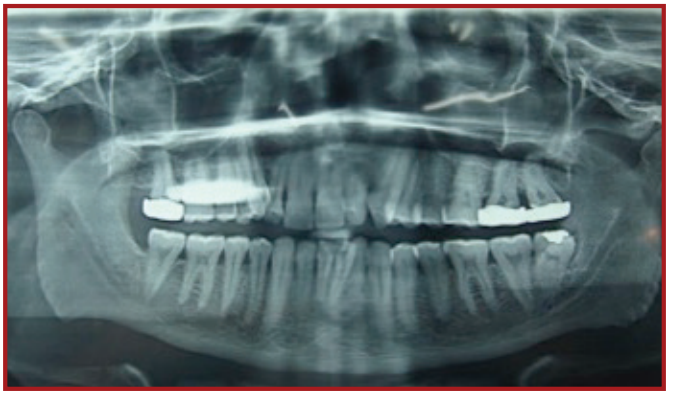

Figure 4. Orthopantamograph showing no signs of odontogenic infections

To rule out systemic granulomatous diseases like Chrons disease, Tuberculosis (TB) and Sarcoidosis, the patient was advised for a chest xray, a barium meal follow through and a complete blood count. Blood parameters were normal as were C- reactive protein, serum glucose, hepatic and renal parameters as well as the HIV screening test and HBsAg. No abnormalities were eminent in the radiographic studies too. Hence the patient was advised for a lip biopsy as the difficulty was in differentiating cheilitis granulomatosa and cheilitis glandularis. Histopathological examination revealed labial glands showing lobular architecture of acini with inflammatory cell infiltration (Fig 5). Replacement of acini by dense inflammatory cells along with atrophy of acini. Dilated ducts and focally fibrous stroma were also seen (Fig 6 & 7). Thus the case was concluded with the diagnosis of Cheilitis Glandularis simplex. Patient was treated initially with oral prednisolone 40 mg (Wyeth, India)

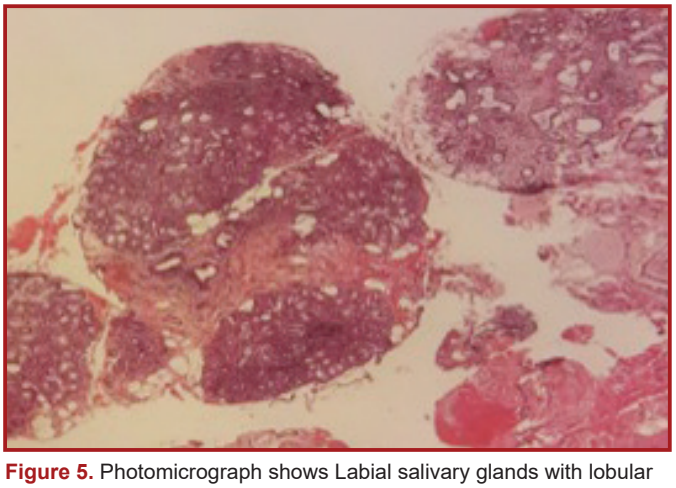

Figure 5. Photomicrograph shows Labial salivary glands with lobular architecture of acini with lymphocytes and plasma cells infiltration in the interlobular region. H & E, 4x

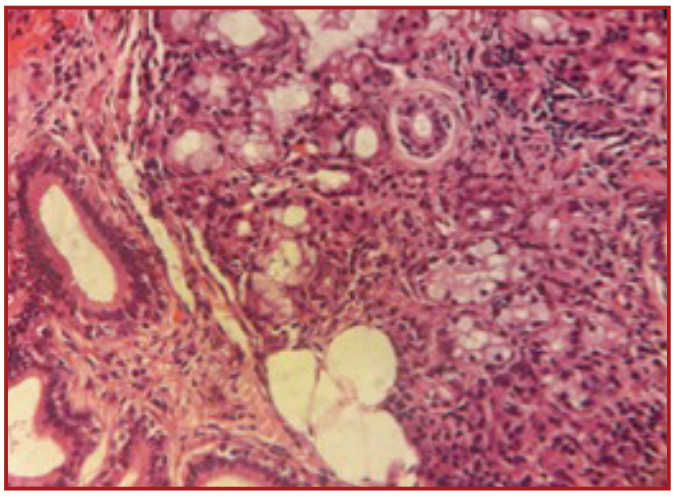

Figure 6. Photomicrograph shows acini with inflammatory cell infiltration i.e Lymphocytes and plasma cells at the interlobular region. H & E, 40x

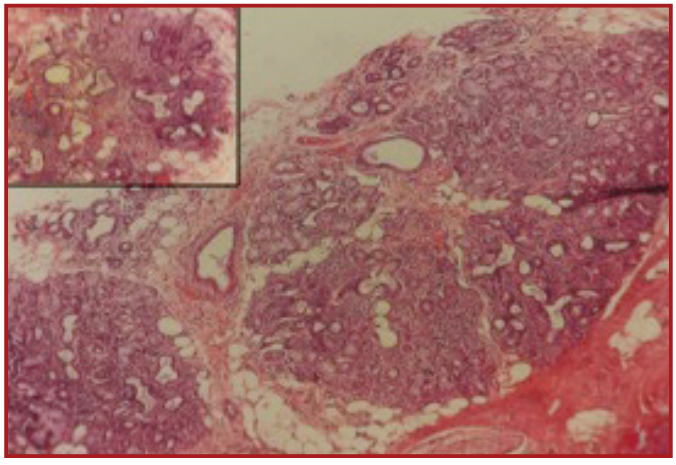

Figure 7. Photomicrograph shows acini are replaced by inflammatory cells with atrophy of acini, dilated ducts and fibrous stroma between acini H & E, 10x, Inset 40x

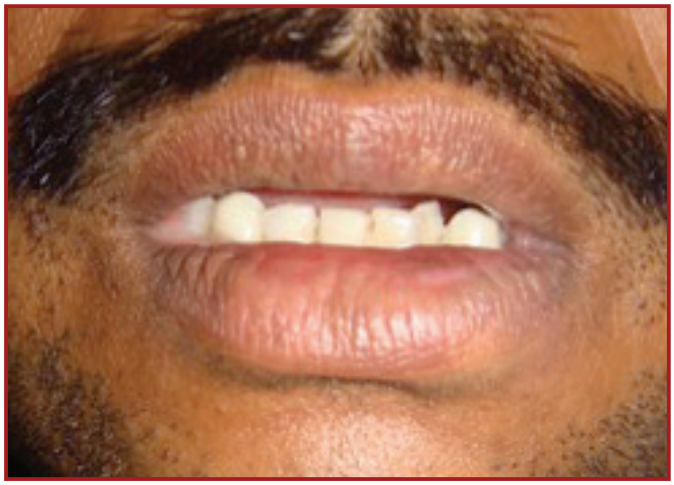

Figure 8. Clinical image shows satisfactory improvement on follow up after 3 months.

once and topical application of triamcinolone acetonide 0.1% (Sarabhai Piramal Pharmaceuticals Ltd., Gujrat, India) thrice daily for a week. Prednisolone dosage was gradually reduced to 5mg per day and the follow up after three months revealed satisfactory improvement in the patient with no fresh complains. (Figure 8)

Discussion

Cheilitis Glandularis (CG) is a clinical diagnosis that refers to an uncommon and poorly understood inflammatory disorder of the lip. Characterised by progressive enlargement and eversion of the lower labial mucosa that results in the obliteration of the mucosal-vermilion interface. Historically three forms of Cheilitis Glandularis have been described. Simple, Superficial suppurative, Deep suppurative. CG simplex being the most common (4).

Cheilitis glandularis represents a clinical reaction pattern to chronic irritation of the lip from a spectrum of highly diverse external causes like use of tobacco products, poor oral hygiene, Chronic exposure to environmental elements (sunlight and wind), compromised immune system. CG appears to favor adult males however cases have been rarely reported in women (5). The present case did not postulate any triggering factors that aggravated the swelling, though the patient was a betel quid chewer ocassionally. It was irrational to suspect betel quid as an etiological factor instigating the oral symptoms since the duration of the swelling was about 3 months and the habit history was present since a decade.

Cheilitis granulomatosa shares some similarities to CG concerning lip swelling and eversion but lacks inflammation in the labial salivary glands, which leads to an increase of the salivary secretion and subsequent crust formation, a feature common. Noberto et al. highlighted that a lip biopsy is helpful in the event of the difficulty in making a differential between cheilitis glandularis and cheilitis granulomatosa as the latter may show non caseating granulomas in 40-50% of cases. In either disease, the exact etiopathogenesis remains unknown (6). The case was successfully treated with a combination of intralesional steroid and topical immunomodulator, whereas in our case the patient was treated effectively with a combination of oral and topical corticosteroid.

Similar case of CG was presented by MAE et.al which did not reveal any clinical or histopathological evidence of chronic inflammation or irritation but there was a positive family history which is not consistent with the present case (7). In another case presented by Hillen U et al the patient presented with discharge of pus from the lower lip on slight pressure and was treated surgically (8). Unlike the present case, the lesion did not have any discharge on palpation hence the diagnosis was considered as Cheilitis glandularis simplex after histopathological examination and was treated with corticosteroids.

CG is a rare disease of the minor salivary glands with a remarkable disposition for local recurrence after local excision. The possibility of a systemic disease with local manifestations should always be considered in causal research. (9) Hence in our case the patient was subjected to detailed laboratory and radiological investigations to exclude the probability of primary systemic conditions that could have predisposed to the lip swelling, which could also intimate diagnostic evidences for the likelihood of recurrence and systematic management of the patient.

Conclusions

This case of cheilitis glandularis emphasizes the nonspecific etiopathogenesis, clinical presentation and the successful management using a combination of oral and topical corticosteroid. The treatment is challenging as many cases of lip swellings that have been reported in literature provide evidences to

the correlation of granulomatous conditions predisposing to lip enlargements. However, the present case did not reveal any significant local or systemic contributory factors, However, the present case did not reveal any significant local or systemic contributory factors. Through this case report, the authors have attempted to provide an insight to the clinicians for a collaborative approach to deal with lip swellings, their recurrences and systematic supervision. It is also mandatory to confirm such cases with histopathological diagnosis for appropriate management of the patients with long term follow up.

Acknowledgement

The patient was evaluated by PP and AS. Data were collected and analyzed by PP, AS and KB. Manuscript was prepared by PP. Histopathology confirmation and photomicrographs were provided by KH. All the authors have participated in the drafting of the manuscript.

Conflicts of Interests

The authors declared they do not have anything to disclose regarding conflict of interest with respect to this manuscript.

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