Clinically granulomatous cheilitis with plasma cells

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ABSTRACT

Plasma cell cheilitis, also known as plasma cell orificial mucositis is a benign inflammatory condition clinically characterized by erythematous plaque on lips that may be ulcerated. Histopathologically it is characterized by dense plasma cell infiltrates in a band-like pattern in dermis, which corresponds to Zoon's plasma cell balanitis. On the other hand, granulomatous cheilitis, as a part of orofacial granulomatosis, manifests as sudden diffuse or nodular swelling involving lip and cheek. Initial swelling is soft to firm, but with recurrent episodes swelling gradually become firm rubbery in consistency. We hereby report a case of cheilitis in a 52-year-old man with diffuse swelling involving lower lip, which clinically resembles granulomatous cheilitis, but histopathological examination showed diffuse infiltrate of plasma cells predominantly in upper and mid-dermis.

Key words: Cheilitis, granulomatous, plasma cell

INTRODUCTION

Plasma cell cheilitis is characterized by sharply demarcated, infiltrated, reddish plaque with glazing surface on lip. It is histologically characterized by diffuse plasma cell infiltration in dermis. On the other hand, granulomatous cheilitis is clinically characterized by diffuse, progressive, and chronic enlargement of lips, without any fissuring, scaling, or ulceration. The differential diagnosis of granulomatous cheilitis includes different causes of macrocheilia, angioedema, glandular cheilitis, contact cheilitis, Ascher's syndrome, and others. Histologically, granulomatous cheilitis is characterized by a chronic inflammatory reaction consisting of lymphocytes, histiocytes, and tuberculoid granuloma having epithelioid cells and Langhans giant cells. Here we present a case, which is clinically resembling granulomatous cheilitis at presentation, but histologically resembling plasma cell cheilitis.

CASE REPORT

A 52-year-old male patient, mason by profession presented with diffuse swelling involving his lower lip since four months. Lip lesion started initially as a localised swelling that disappeared spontaneously and again reappeared as a diffuse swelling of lower lip. The swelling was progressive in nature. There was no associated itching, pain, or burning sensation. The lesion did not exacerbate on sun exposure. The patient was nonsmoker and nonalcoholic. The patient had no history of any bowel problem, abdominal cramp, chest pain, respiratory distress, and visual problems. No family history of similar lesion was noted. He did not take any medication prior to onset of lesion.

Examination revealed diffuse skin-colored swelling of the lower lip, which was firm in consistency and nontender. There was no surface change and the swelling was limited within the vermilion border of the lip [Figure 1]. Examination of the upper lip, and oral mucosa was within normal limits. Regional lymph nodes were not enlarged. Examination of hair, nail, and genital mucosa was unremarkable. Respiratory, cardiovascular, gastrointestinal, neurological,

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and ophthalmological examination were within normal limits. Complete hemogram showed peripheral eosinophilia (10% and reference range 1%–6%).Other routine blood investigations such as blood sugar, liver function test, and renal function tests were normal. Mantoux test showed and in duraton 8 mm × 6 mm. Serology for retrovirus and hepatotropic viruses was negative. His thyroid hormone profile revealed the T4 level to be 2.15 μg/dL (reference range for male, 4.4–10.8 μg/dL) and thyroid stimulating hormone (TSH) to be 75.4 μlu/mL (reference range for adults, 0.30–6.02 μlu/mL). Both T4 and TSH was done by Lilac, Neo Lumax (CLIA). Upper gastrointestinal tract endoscopy, colonoscopy, and chest X-ray revealed nothing. Histopathological examination with hematoxylin and eosin stain showed normal epidermis and normal rete ridges. Dermis showed diffuse infiltration of plasma cells in upper and mid dermis [Figures 2 and 3]. Histopathology of the upper lip was within normal limits. Considering the clinical features, laboratory investigations, and histopathological findings, we diagnosed plasma cell cheilitis.

DISCUSSION

Plasma cell cheilitis is a rare benign idiopathic condition of mucous membrane that is characterized histologically by a dense band such as plasmacytic infiltrate in thedermis.[1] It is more common in males, elderly patients and more common in lower lip.[2] Plasma cell cheilitis is the counterpart of Zoon's plasma cell balanitis. It usually presents as flat or elevated patches of erythema on the lower lip. The cause is unknown, but has been successfully treated with intralesional corticosteroid injection,[3]topical tacrolimus ointment,[4] 308 nm monochromatic excimer light, and subsequent tacrolimus application.[5] Cheilitis has been reported in association with plasma cell gingivitis, and angular cheilitis. Plasma-acanthoma is a verrucous tumor with a plasma cell infiltrate. It is mostly found in oral mucosa mainly at the angle of mouth. It may also occur in perianal areas, periumbilical areas, finger webs, and inguinal areas. Plasma-acanthoma has been reported to occur in a patient with plasma cell cheilitis signifying them to be a spectrum of same disease.[6] Candida albicans is considered to be an etiologic factor in plasma-acanthoma.

Granulomatous cheilitis may affect both sexes equally. Earliest manifestation usually develops in childhood and adolescence. Initially it presents as a sudden diffuse or nodular swelling in the lips. Swelling may involve lower lip, upper lip, cheek, or periorbital area. The attack may be associated with fever and other constitutional symptoms such as headache, malaise, and so on. After the first episode, the swelling disappears. But with recurrent episodes, swelling persists and it gradually becomes firmer in consistency.[7] There may be loss of taste sensation and decreased salivary secretion. Regional lymph nodes may be enlarged. Granulomatous cheilitis has to be differentiated from angioedema, which is sometimes very difficult in the absence of fissured tongue and facial nerve palsy. Persistence of swelling in between the attacks and firm rubbery consistency points toward diagnosis. Granulomatous cheilitis may be associated with sarcoidosis or Crohn's disease. It may present as a part of Melkersson–Rosenthal syndrome. Melkersson–Rosenthal syndrome consists of a triad of fissured tongue, facial palsy, and cheilitis granulomatosa. It may be familial in origin. Peripheral facial nerve paralysis occurs in 20% patients and plicated tongue in 40% of patients.[8] It has been reported with other cranial nerve involvements such as glossopharyngeal nerve and vagus nerve. Histological changes are not very specific. Ascher's syndrome associated with blepharochalasis[9] and lymphoma[10] is the differential diagnosis of this condition. Macrocheilia due to different causes such as traumatic, developmental, familial idiopathic, different infections, lymphangioma, hemangioma, neurofibroma,

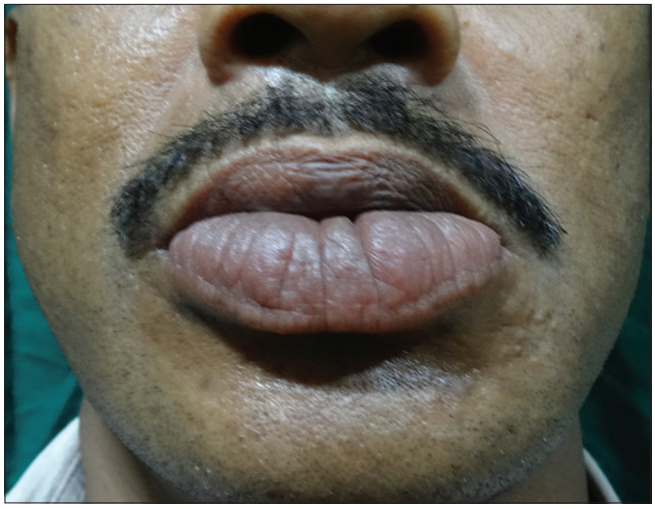

Figure 1: Diffuse swelling of the lower lip at the time of presentation

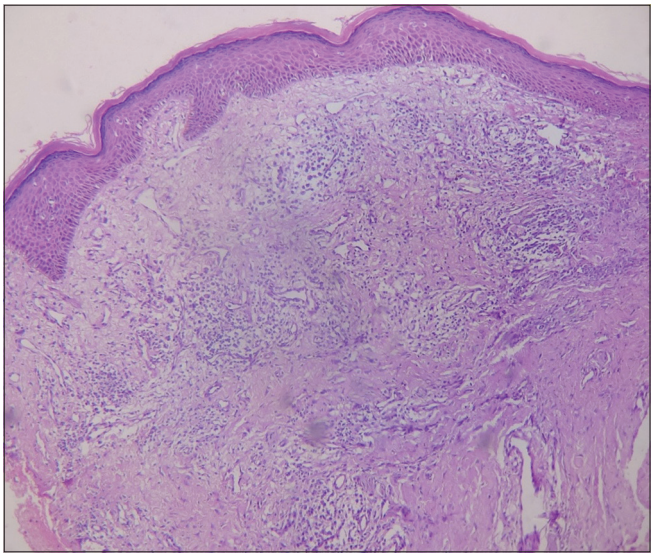

Figure 2: 10X10x view of histopathological section with hematoxylin and eosin stain showing diffuse round cell infiltrate in upper and mid-dermis

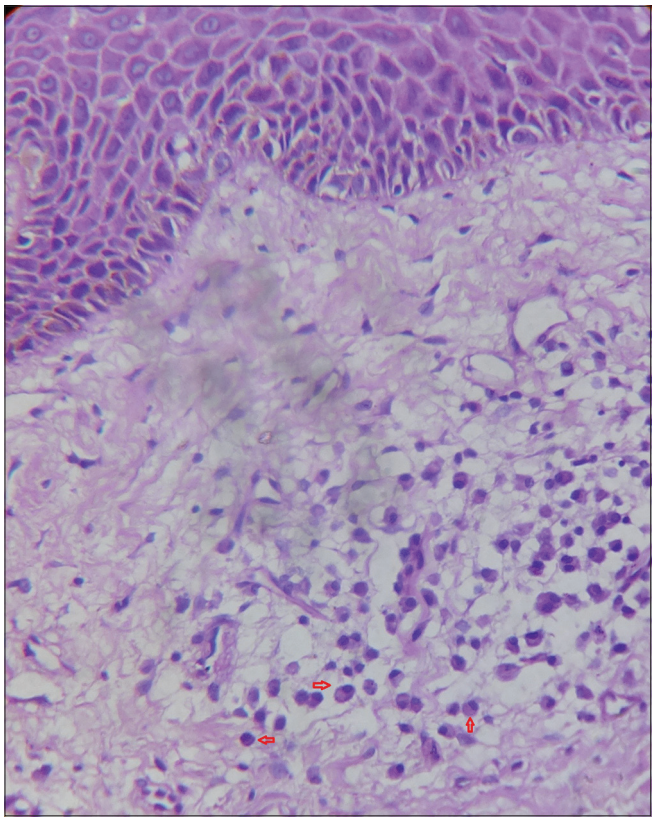

Figure 3: 10X40x view of histopathological section with hematoxylin and eosin stain showing diffuse plasma cell infiltrate in upper and mid-dermis

mucopolysaccharidosis, and neoplasia, also mimic this condition. Intralesional steroid, oral clofazimine, metronidazole are the treatment options for granulomatous cheilitis. Systemic steroids are rarely required.

Our patient is an elderly male. Age and sex of our patient favours plasma cell cheilitis. But contrary to the typical presentation of plasma cell cheilitis as a localized erythematous plaque, it presented as a diffuse firm swelling. Histopathology however clinched the diagnosis in our case.

CONCLUSION

We conclude as besides having typical presentation of erythematous plaque, plasma cell cheilitis might have different morphology.

Our patient was treated with intralesional triamcinolone injection (40 mg/mL), 1 mL every third week along with tacrolimus ointment (0.1%) twice daily. The patient showed slow response to treatment [Figure 4].

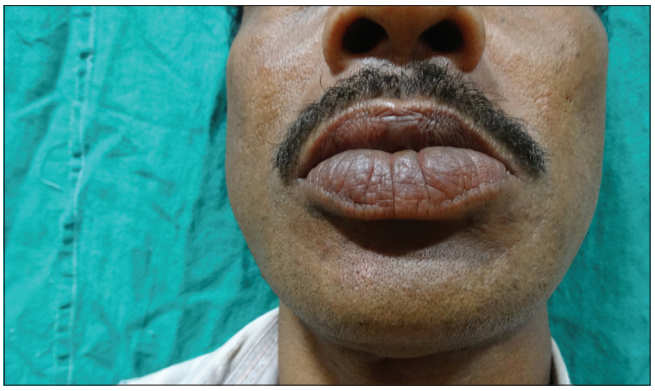

Figure 4: Appearance of lip after third intralesional triamcinolone injection along with topical tacrolimus

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Conflicts of interest

There are no conflicts of interest.

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