Plasma-cell cheilitis: successful treatment with intralesional injections of corticosteroids

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Summary

Plasma-cell cheilitis is a rare inflammatory disorder of the lip characterized histologically by a band-like infiltrate of plasma cells in the upper dermis. It is considered an oral counterpart of plasma-cell balanitis. Clinically, it presents as a circumscribed, flat to slightly raised, eroded area of the lip. The cause of plasma-cell cheilitis is unknown, and the treatment is often disappointing. We describe a 55-year-old woman who had a long-lasting painful, swollen, and eroded area on her lips, which responded poorly to various topical treatments. Biopsy showed a band-like infiltrate composed mainly of mature plasma cells in the dermis. A diagnosis of plasma-cell cheilitis was made after excluding contact dermatitis, lichen planus, bacterial, fungal and spirochaete infections, and an extramedullary plasmacytoma. Dramatic improvements were observed after intralesional injections of corticosteroids. The lesion cleared up after two treatments, and there has been no recurrence in 1 year of follow-up.

First described by Zoon in 1952,[1] plasma-cell balanitis (Zoon's balanitis) is an inflammatory disorder of the glans penis showing a characteristic band-like infiltrate of plasma cells in the upper dermis. Subsequent reports disclosed similar conditions in other mucosal areas.[2] Plasma-cell cheilitis is considered the oral counterpart of Zoon's balanitis.[3] Clinically, the lesion shows a glistening reddened lip. Like Zoon's balanitis, plasma-cell cheilitis represents a stage of an immune response to various stimuli, either benign or malignant.[3] It is often resistant to topical treatments.[4] In this report, we describe an illustrative case of plasma-cell cheilitis, successfully treated with intralesional injections of corticosteroids.

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Report

A 55-year-old Taiwanese woman presented with a recalcitrant, painful, swollen lower lip (Fig. 1a). The lesion had persisted for 7 months; intermittent erosion and bleeding were noted during this period. The remainder of the skin and mucous membranes was normal.

The patient had undergone various treatments in the past without improvement, including topical corticosteroids, an antibiotic (erythromycin ointment), a topical aciclovir cream and a calcineurin inhibitor (0.03% tacrolimus cream). She had also applied beeswax to the lesion. Her condition led to marked food restriction and social impairment.

The patient did not smoke and she had no habit of lip-biting or excessive lip-licking. She denied any use of lipstick or history of drug use. She also denied any history of exacerbation of lesions by food and liquids, excessive sun exposure or any family history of lip disorders. There was no history of dental treatments or trauma.

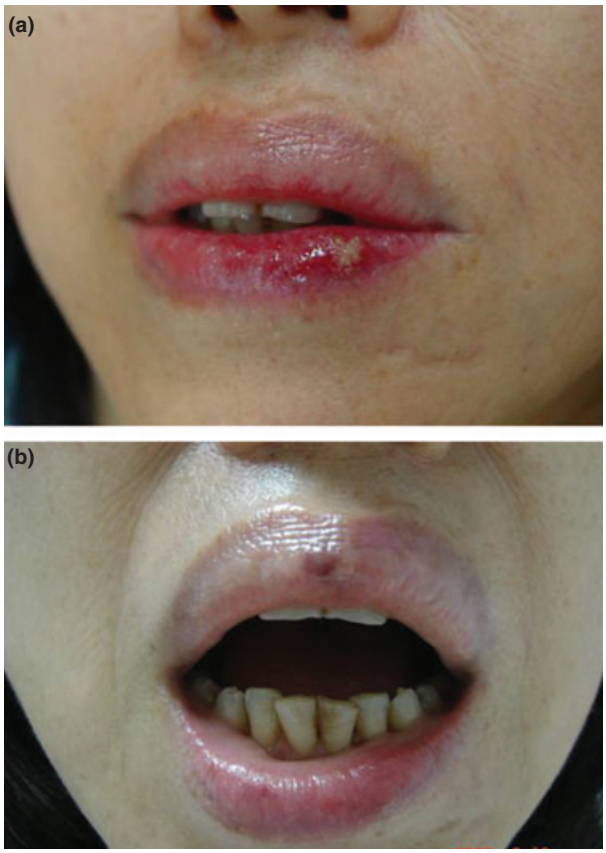

Figure 1 (a) A poorly demarcated, brightly erythematous, crusted patch on the left vermilion surface of the slightly swollen lower lip; (b) dramatic response after one intralesional injection of triamcinolone acetonide 2.5 mg/mL.

An initial diagnosis of chronic cheilitis was made, and an incisional biopsy was taken. Histologically, there was a dense, band-like infiltrate composed mainly of plasma cells in the upper and mid dermis (Fig. 2a–c). A few eosinophils and histiocytes were admixed. Histochemical staining for fungi and bacteria (periodic-acid–Schiff and Gram stain, respectively) revealed no microorganisms. Immunostaining for *Treponema pallidum* (Biocare Medical, Concorde, CA, USA) was also negative. The plasma cells expressed both kappa and lambda light chains of immunoglobulins (Fig. 3a,b). Serological tests for antinuclear antibodies, C3, C4 and rheumatic factor were all within normal limits.

Under a clinicopathological diagnosis of plasma-cell cheilitis, the patient received an intralesional injection of 0.1 mL triamcinolone acetonide (2.5 mg/mL). The lesion responded rapidly. Marked clinical improvement was observed 2 weeks after the first treatment (Fig. 1b). An additional session of injections performed at 2 weeks resolved the lesion. The patient has shown no recurrence in the ensuing 1 year of follow-up.

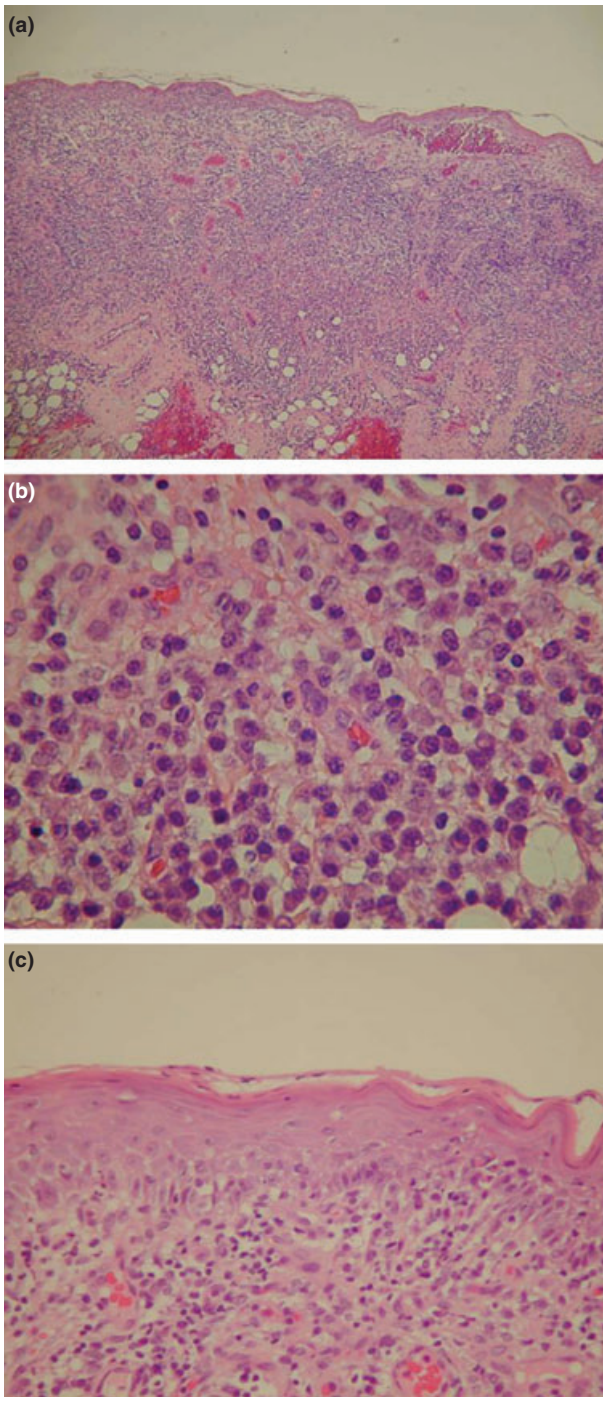

Figure 2 (a) Dense, band-like infiltrate in the upper dermis; (b) predominance of mature plasma cells in the infiltrate; (c) close-up view of the epidermis showing no evidence of viral cytopathic changes or cellular dysplasia. Haematoxylin and eosin; original magnification (a) × 40; (b) × 400; (c) × 200.

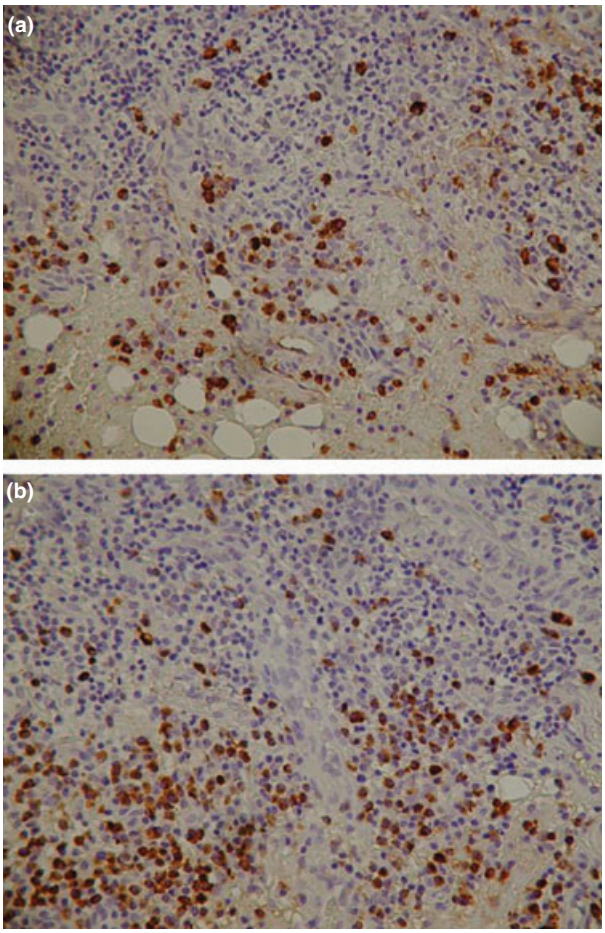

Figure 3 Plasma cells showing positivity for both (a) kappa and (b) lambda light chains (original magnification × 200).

Plasma-cell cheilitis is an idiopathic, benign, inflammatory condition, characterized by a dense plasma-cell infiltrate in the lip mucosa. Besides the lips, lesions can also occur in other organs, such as the penis, vulva, buccal mucosa, palate, gingiva, tongue, epiglottis and larynx.[2,5] White *et al.*[2] proposed 'plasma-cell orificial mucositis' to encompass the various names given to the shared condition at different body sites.

Plasma-cell cheilitis must be differentiated from allergic or irritant contact cheilitis, candidiasis, syphilis, actinic cheilitis, cheilitis granulomatosa, plasmacytoma and mucosal lichen planus.[6] A detailed contact history and the lack of prominent spongiosis or eosinophils reduced the possibility of allergic or irritant contact cheilitis. Fungal and bacteria infections were excluded by the negative microbiological stains. Syphilis was also excluded by the negative result for antibodies against *T. pallidum*.[7] The biopsy specimen showed no keratinocyte atypia, solar elastosis or granulomatous infiltrate, thus a diagnosis of actinic cheilitis or cheilitis granulomatosa was not considered. An extramedullary plasmacytoma was also excluded, based on the polyclonal nature of the infiltrate.[8] Lichen planus shows lichenoid interface dermatitis consisting mainly of lymphocytes rather than plasma cells in the upper dermis. It is also characterized by saw-tooth rete ridges and the presence of colloidal bodies, which were not seen in this case.

The treatment of plasma-cell cheilitis is often disappointing. Most reported cases showed poor or limited responses to topical corticosteroids.[2,6,8] Various other treatments have been recommended, including surgical excision, radiation treatment, electrocauterization, cryotreatment, topical fusidic acid and systemic administration of griseofulvin.[5,6] The degree of acanthosis seems to be an important factor influencing treatment outcomes in those who respond to topical corticosteroids. Different results with topical corticosteroids have been reported for atrophic, mildly acanthotic and markedly acanthotic variants.[2,4] Intralesional injections can increase the effects of topical corticosteroid treatment by bypassing the epidermal barrier zone.[6] Surprisingly, successful treatment with intralesional corticosteroids was not reported until 2001, when Kaur *et al.*[9] treated a case with a combination of topical and intralesional triamcinolone acetonide 10 mg/mL at an interval of 2 weeks for three sessions. Subsequently, Yang *et al.*[6] reported three patients who were successfully treated with intralesional triamcinolone acetonide 4–5 mg/mL at an interval of 2 weeks for three sessions.

There was no acanthosis of the epidermis in our patient; however, the depth and heaviness of the infiltrate, which occupied almost the entire dermis, may have accounted for the poor response to previous topical treatments. Although the dose of intralesional corticosteroids was lower than those reported by Kaur *et al.*[9] and Yang *et al.*,[6] our patient had a good outcome with no recurrence in 1 year of follow-up. Longer follow-up is still needed to confirm true eradication of the lesion.

In conclusion, this case reinforces the importance of a skin biopsy to characterize the nature of 'chronic cheilitis', which is commonly encountered by dermatologists, and accordingly to choose the most effective treatment. Based on this case report, we further support intralesional injections of corticosteroids as a valuable treatment for plasma-cell cheilitis. Compared with topical application, intralesional injection of corticosteroids can deliver the drug more promptly and efficaciously to the lesion. Further studies are warranted to determine the optimum dosage and number of treatments for plasma-cell cheilitis.

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