

## Two Cases of Plasma Cell Cheilitis Treated with Intralesional Injection of Corticosteroids

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Plasma cell cheilitis is a rare inflammatory disorder which shows a dense, band-like infiltrate of plasma cells in the upper dermis. Although the histological picture is shared with other diseases of the lips, plasma cell cheilitis is not associated with any known dermatoses. Some authors have shown the effectiveness of topical or intralesional corticosteroids; however, there have been many reports describing poor therapeutic response to topical steroids. We describe two patients with plasma cell cheilitis whose clinical condition responded rapidly to the intralesional injection of corticosteroids. (*Ann Dermatol* 15(1) 34~38, 2003).

*Key Words* : Plasma cell cheilitis, Corticosteroids, Intralesional injection

Plasma cell cheilitis (PCC) is a rare, idiopathic, benign, inflammatory condition characterized by a dense plasma cell infiltrate in the oral mucosa. It appears as a circumscribed, flat to slightly elevated, eroded plaque, usually on the lower lip of an elderly person<sup>1</sup>. The lesion may be accompanied by tenderness and sensitivity to certain food<sup>2</sup>. PCC is regarded as a benign idiopathic disorder that is not associated with any known dermatoses, although the histological picture is often overlapped with diseases such as actinic keratosis, Bowen's disease, squamous cell carcinoma, and syringocystadenoma<sup>3</sup>. The pathogenetic mechanism for the infiltrate remains obscure<sup>3</sup>. Various topical treatments have been tried, producing conflicting results. Some authors have reported that steroids, both topical<sup>4</sup> and intralesional<sup>5</sup>, have produced remission, whereas many others have reported resistance to these steroids and recommended surgical excision, ra-

diotherapy, or cryotherapy for severe cases<sup>2</sup>. Recently, systemic administration of griseofulvin has been found to be effective for the clinical improvement of the lesion<sup>1,6</sup>. We present two elderly patients with PCC who showed rapid clearing of the lesions after intralesional injection of corticosteroids.

### CASE REPORT

**Case 1.** A 69-year-old, otherwise healthy man visited our clinic because of a painful erosive plaque on the lower lip. The lesion developed three years before his visit and gradually became thickened and fissured. Pain was evoked by drinking and eating. He had been treated with topical corticosteroids without benefit. There was no history suggestive of trauma or contact allergy. Physical examination showed a well-demarcated erythematous to yellowish erosive plaque on the lower lip (Fig. 1). Histopathology of the biopsy specimen revealed acanthosis and a dense, band-like infiltrate of plasma cells in the upper dermis (Fig. 2, Fig. 3). Results of serologic tests for syphilis were negative. Local treatment consisted of topical application of 0.05% clobetasol-17-propionate ointment under plastic occlusion for three weeks. Because topical steroid produced only minimal improvement, intralesional injection of triamcinolone

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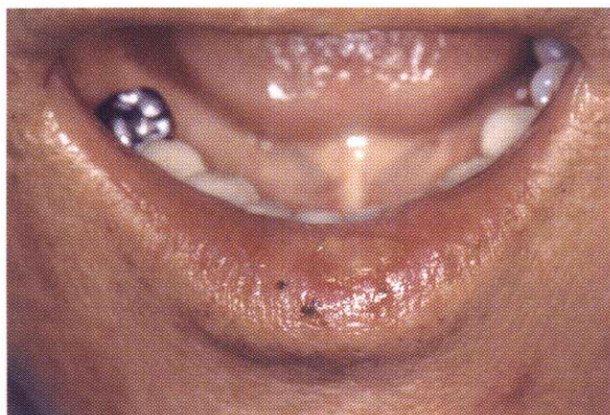


Fig. 1. A well-demarcated, erythematous, erosive plaque on lower lip (Case 1).

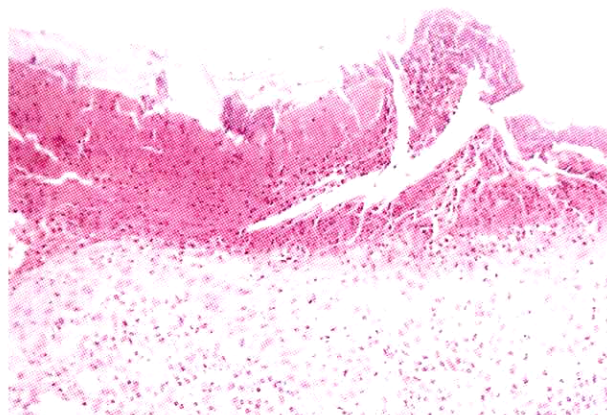


Fig. 2. Dense, band-like infiltrate in the upper dermis (Case 1, H & E,  $\times 40$ ).

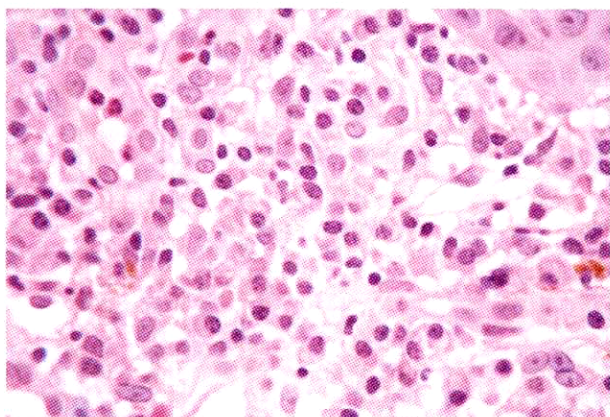


Fig. 3. Dermal infiltrate consisting largely of plasma cells (Case 1, H & E,  $\times 200$ ).

acetodide (5.0 mg/ml, in buffered diluent) was started. The lesion responded rapidly and marked clinical improvement was observed in two weeks. Additional two sessions of injection produced virtual clearing of the lesion. The patient was observed for 10 months without evidence of recurrence.

**Case 2.** A 62-year-old woman who had been suffering from liver cirrhosis was referred to our department because of a painful lesion on the lower lip for four years. The lesion developed spontaneously without history of preceding trauma or contact allergy and became erosive. Treatment with various topical corticosteroids had been completely unsuccessful. Examination disclosed an erythematous thickened plaque with erosion and

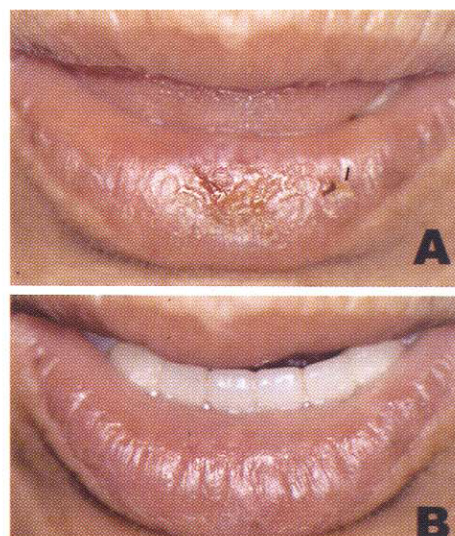


Fig. 4. An encrusted, thickened plaque on lower lip (A), which later shows marked clinical improvement after intralesional injection of triamcinolone acetonide (B) (Case 2).

crusts on the lower lip (Fig. 4A). Laboratory tests revealed hepatic dysfunction (AST, 111 units/l; ALT, 153 units/l;  $\gamma$ -GT 47 units/l), elevated level of alpha-fetoprotein (31.0 ng/ml), and the presence of anti-HCV IgG antibody. Serology for syphilis was negative. Liver ultrasonography showed a diffuse hepatosplenomegaly and multiple hemangiomas. Punch biopsy was performed on the lip lesion and histopathology revealed acanthosis and an upper dermal dense band-like infiltrate which consisted mainly of plasma cells (Fig. 2). Intralesional injection

**Table 1.** Summary of selected reports of plasma cell orificial mucositis

| Reference                    | Age (years)/Sex | Site                          | Treatment   |   |
|------------------------------|-----------------|-------------------------------|---|---|
|                              |                 |                               | Ineffective   | Effective   |
| Tamaki et al. <sup>1</sup>   | 56/male         | lower lip                     | topical corticosteroids, oral prednisolone  | griseofulvin 500mg/day for 2 weeks                              |
|                              | 71/male         | lower lip                     |   |   |
| White et al. <sup>2</sup>    | 47/female       | upper lip, tongue, epiglottis | nystatin oral suspension, 0.25% desoximetasone cream, intralesional injection of triamcinolone acetate (10.0 mg/ml) | griseofulvin 500mg/day for 3 weeks                              |
| Jones et al. <sup>4</sup>    | 52/female       | lower lip                     |   | clobetasol propionate ointment for 3 weeks                      |
| Baughman et al. <sup>5</sup> | 65/female       | lower lip                     | 0.025% fluocinolone acetonide cream   | intralesional injection of triamcinolone acetonide (2.5mg/ml)   |
| Yoon et al. <sup>6</sup>     | 70/male         | lower lip                     |   | griseofulvin 500mg/day for 8 weeks                              |
| Curto et al. <sup>8</sup>    | 13/female       | gingival                      | 0.5% hydrocortisone ointment, 0.1% triamcinolone acetate ointment, 2% fusidic acid ointment                         | Electrocoagulation  |
| Noorily <sup>10</sup>        | 67/male         | lower lip                     |   | lower lip reduction with primary closure                        |
| Mahler et al. <sup>11</sup>  | 53/female       | gingiva                       | topical corticosteroids, local anesthetics, carbon dioxide laser treatment  | 2% fusidic acid ointment for 10 weeks                           |
| present case (1)             | 69/male         | lower lip                     | 0.05% clobetasol-17-propionate ointment   | intralesional injection of triamcinolone acetonide (5.0mg/ml)   |
| Present case (2)             | 62/female       | lower lip                     | topical corticosteroids   | intralesional injection of triamcinolone acetonide (10.0 mg/ml) |

of triamcinolone acetodide suspension (10.0 mg/ml) produced remarkable thinning of the plaque as well as complete loss of subjective symptoms. Two more injections (5.0 mg/ml) were undertaken and the lesion nearly disappeared (Fig. 4B).

## DISCUSSION

In 1952, Zoon<sup>7</sup> described eight patients with benign circumscribed chronic balanitis characterized by an extensive infiltration of plasma cells with no evidence of dysplasia of the overlying epidermis. Since then, many authors have described equivalent conditions involving buccal mucosa, palate, gingiva,

tongue, epiglottis, larynx, and lips, which later summarized by White et al<sup>2</sup>. under the name of 'plasma cell orificial mucositis', the most widely accepted term for this condition. A tumor-like variant, plasmoacanthoma, was described, implying a broad clinical spectrum of this pathology<sup>8</sup>. Plasma cell orificial mucositis of the lip, or PCC, appears as a circumscribed plaque usually on the lower lip of an aged person<sup>3</sup>. The lip may be thick and fissured, or even ulcerated, and may be accompanied by pain and tenderness<sup>3</sup>.

The pathogenic mechanism for the infiltrate remains unknown. Attempts to identify specific benign or malignant processes have been tried in association with this rare disorder without providing any specific

disease processes<sup>4</sup>. Now it is suggested that PCC, like the balanitis of Zoon or other infiltrates of non-neoplastic plasma cells in various mucosal areas, is not a response specific for any stimulus or any individual disease, but rather represents a stage in the immune response to any one of a variety of stimuli, benign or malignant<sup>5</sup>. However, the role of T cells and macrophages in B cell growth and differentiation has been shown<sup>3</sup>. Furthermore, Aiba and Tagami<sup>9</sup> have shown that the plasma cell infiltrate composed of IgG- and IgA-producing cells is in accordance with the pattern observed in certain epidermal neoplasms frequently accompanied by the plasmacytic infiltrate, such as actinic keratosis, Bowen's disease, squamous cell carcinoma, and syringocystadenoma papilliferum. It is interesting that our second patient was suffering from chronic liver disease which may alter systemic immune function; however, relationship between PCC and systemic diseases including liver cirrhosis has not been described to date.

Allergic or irritant contact dermatitis must be included in the differential diagnosis of PCC. Candidiasis also can mimic this condition but is usually ruled out by culture and lack of response to nystatin. Cheilitis granulomatosa may also share features with plasma cell orificial mucositis, but with cheilitis granulomatosa there is histopathologic evidence of a granulomatous infiltrate. Squamous cell carcinoma must also be considered, but this can be easily ruled out by the biopsy. PCC has to be differentiated from actinic cheilitis because sometimes the latter also demonstrates plasma cell-rich infiltrate. The presence of keratinocyte atypia is a clue for the diagnosis of actinic cheilitis. Personal history of long exposure to sunlight and certain occupations, *e.g.* agricultural workers, help distinguishing PCC from actinic cheilitis. The lack of personal or occupational history suggesting longstanding sun exposure in our patients favored the diagnosis of PCC rather than actinic cheilitis. However, the possibility of actinic cheilitis still remains in our patients.

The treatment of PCC is often disappointing. Some authors<sup>4,5</sup> have shown that topical and intralesionally administered steroids were effective for the clearing of the lesions, whereas others<sup>2,8,10</sup> have found that most of the topical therapies were ineffective in their patients and have recommended that the alternatives such as surgical excision, radi-

ation therapy, electrocauterization, and cryotherapy might be helpful in the severe refractory cases. From a therapeutic point of view, degree of acanthosis seems to be important because a difference in the response of topical steroids have been reported between atrophic, mild acanthotic, and marked acanthotic variants<sup>2,4</sup>. Potency of the topically applied steroids and the manner of application also are likely to be associated with the efficacy of treatment. One well-known modality which increases the effects of topical corticosteroid therapy and bypasses the barrier zone is the establishment of a subepidermal depot by sublesional or intralesional injection. Our patients didn't show the clinical improvement with the topical application of highly potent corticosteroids, but responded rapidly after the intralesional injection of steroids. Topical application of highly potent topical steroids, *e.g.* clobetasol propionate ointment, if applied under plastic occlusion, may be effective in the treatment of PCC, regarding the previously reported cases<sup>4</sup>. One case was reported describing a patient with plasma cell gingivitis treated with topical application of fusidic acid<sup>11</sup>. Some authors<sup>1,6</sup> recommended systemic administration of griseofulvin for the treatment of PCC whereas other authors<sup>12</sup> found that griseofulvin did not help in balanitis of Zoon.

In conclusion, we suggest that intralesional injection of corticosteroids may be valuable treatment of PCC and, possibly, other types of plasma cell orificial mucositis resistant to the conventional topical corticosteroid therapy.

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