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Case Report

Oral lichen planus with palmoplantar involvement: a case report

Wei Chen[1,2*], Jianjun Liu[2*], Jianzhong Zhang[3], Jinlian Zhou[3], Junlian Liu[1,2]

[1]The 306 Clinical College of PLA, Anhui Medical University, Beijing, PR China; Departments of [2]Dermatology, [3]Pathology, 306 Hospital of PLA, Beijing, PR China. [*]Equal contributors.

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Abstract: Lichen planus (LP) is a chronic autoimmune inflammatory dermatosis that is characterized by purple, pruritic plain topped papules, involving the skin, mucous membrane, nail and hair follicles. According to the site of involvement there are various forms of LP. Palmoplantar LP is one of the rare clinical variants, which is meanwhile accompanied with the oral involvement being extremely uncommon. Herewith, we report a case of a woman who has oral LP with palmoplantar involvement, treated with oral acitretin and topical corticosteroids. Now the patient is responding well to the treatment.

Keywords: Lichen planus, palmoplantar, oral, acitretin

Introduction

Lichen planus (LP) is a common chronic inflammatory disorder that is characterized by purple, flat-topped, polygonal pruritic papules and plaques with Wickham striae, which affects the skin, mucous membranes, nails, and scales [1]. Although pathophysiology has not yet been fully defined, LP mainly appears to be T cell immune-mediated [2]. Usually LP occurs on the extremities and trunk. According to the site of involvement there are various forms of LP [3]. Palmoplantar LP is one of the rare clinical variants, which is meanwhile accompanied with the oral involvement being extremely uncommon. Little literature describes oral LP accompanied with palmoplantar involvement. In this paper, we present a case of LP simply involved in the palm, plantar and oral mucosa.

Case description

A 67-year-old woman visited us for pruritic, painless papules and plaques eruption of approximately five months' duration on the oral left mucosae. Subsequently, the lesion spread to right buccal mucosa in the case of having no treatment and gradually the initial pompadour papules evolved into grayish-white plaque. Confluent plaques and discrete papules had simultaneously emerged on her palms and soles, with mild itch. Then the patient went to a dental hospital with a complaint of friction sensation in the mouth. Consequently, the manifestations were diagnosed as oral leukoplakia by the dentist. After she had been treated with hydroxychloroquine 100 mg daily for two weeks, the lesion persisted and had gradually increased in size with no positive improvement. She denied any additional skin eruptions and hair or nail lesions. There was no history of any other systemic or skin disease. There was no history of any drug intake prior to the lesions.

Physical examination revealed lesions with papules or plaques involving the oral mucosae, palm and sole. As shown in **Figure 1**, buccal mucosa presented as reticulate, grayish-white, flat-topped plaques, which were symmetrically distributed over both right and left buccal mucosa on an erythematous background. Lesions that touched slightly hard had well-defined edges, without hyperemia and erosion on the surface. As shown in **Figure 2A**, the palm skin lesions presented as purple flat-topped papules and partially fused into plaques, whose surface was dry and whose edge was scattered with slight scaling. **Figure 2B** highlights confluent,

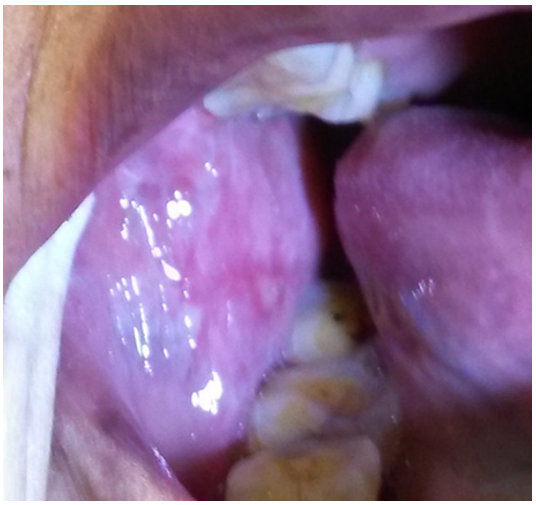

Figure 1. Patient with reticulate, grayish-white, flat-topped plaques on right buccal mucosa.

erythematous papules and plaques with scaling on the lateral proximal aspect and the internal plantar arch of the foot. No other skin lesions were seen on the body. Nails and hair were not involved.

Laboratory examination demonstrated a complete blood count and comprehensive metabolic panel, and lipid panel were all within normal limits. A hepatitis viral, herpes simplex virus panel, syphilis rapid plasma regain trial and treponema pallidum hemagglutination test were all negative.

As clinical features of the patient are very similar to that of oral leukoplakia and lichen drug reactions described in literature, so a biopsy is extremely useful. The histopathology of the oral mucosa is shown in **Figure 3**, hyperkeratosis and mild irregular orthokeratosis overlying an acanthotic epidermis, interface dermatitis with vacuolar degeneration of the basal cell layer and mononuclear cells band-like inflammatory infiltrate in the upper dermis, on which scattered pigmentophage remained. Based on the clinical presentation and histopathological and serologic findings, a diagnosis of oral LP accompanied with palmoplantar LP was made.

The topical therapy with halometasone cream and the systemic administration of acitretin for 20 mg daily were supplied. The patient started on acitretin 20 mg/day for two weeks and then decreased to 10 mg. After one-month treatment the palmoplantar violaceous polygonal flat-topped papules were approximately resolved and oral papules had gradually decreased in size. Two months later, all lesions disappeared with remaining pigment and there was no recurrence of disease as acitretin was tapered and discontinued.

Discussion

LP is an idiopathic inflammatory dermatosis. At present the estimated prevalence of LP is in the range of 0.22 to 5% worldwide [4]. It has different variants based on the morphology of the lesions and the site of involvement. According to the site of involvement, LP may be divided into the following types: palmoplantar LP, mucosal LP, LP of the nails, LP of the scalp, and inverse LP [5], among which palmoplantar LP is a rare topical variant. We reported a rare case of lesion initially appeared on oral mucosa and subsequently on palms and soles. No other skin lesions were seen on the body. According to related literature records, cutaneous lichen planus is seen in approximately 75% of women with oral lichen planus while initial oral involvement that subsequently develops into skin involvement is reported among only 10 to 15% of the patients [6]. The incidence of the above case is very low and there are no documents reporting similar cases.

The condition of the patient with exclusive oral and palmoplantar involvement may cause diagnostic confusion because of the rarity of occurrence and the atypical morphology of lesions at these sites, so early and correct diagnosis with adequate management is very important [7]. Oral LP has several clinical subtypes, including reticular, erosive, atrophic, papular, plaque-like, and bullous subtypes [8]. The case shows that reticulate, grayish-white, flat-topped, plaques are similar to leukoplakia and the clinical presentations belongs to potentially malignant disorder. Palmoplantar LP may be difficultly distinguishable from lichen drug reactions both clinically and histopathologically. Therefore, before making diagnosis of uncommon LP, we should exclude similar presentations disease such as lichen drug reactions, psoriasis, chronic cutaneous lupus erythematosus, and lichen simplex chronicus. Correlation studies suggest that there is lack of clear diagnostic criteria or a uniform methodology for LP. Thus, dermatologists should perform appropriate histopathological examination to avoid misdiagnosis when the diagnosis is ambiguous.

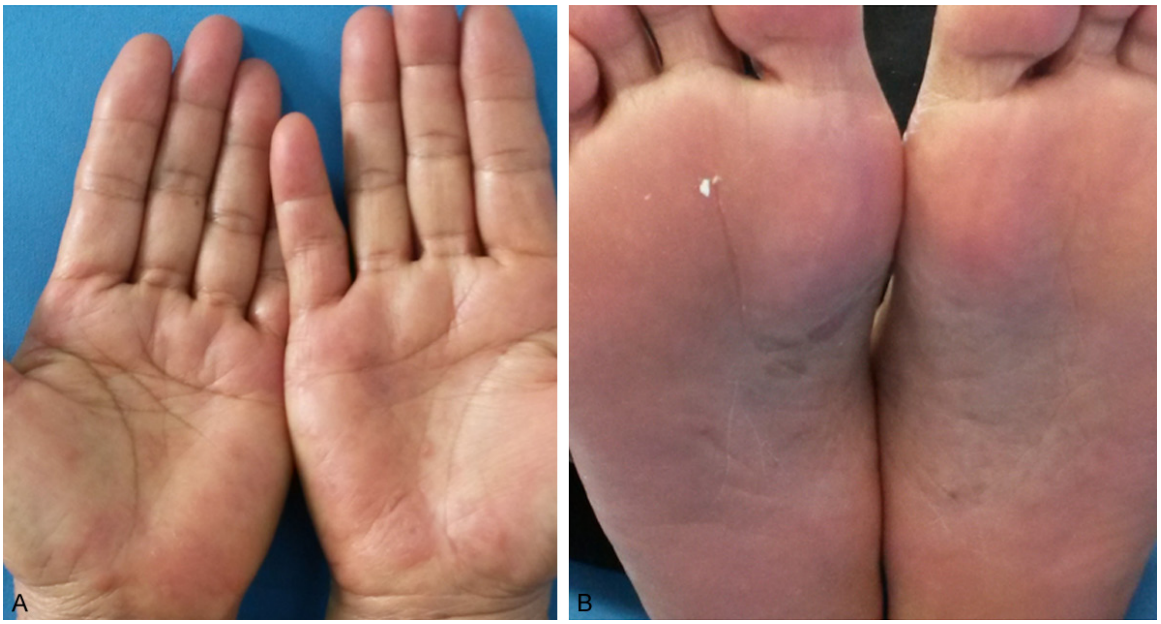

Figure 2. Patient with lichen planus: A. The palm skin lesions with red papules and confluent plaques, at the edge were scatteredly covered with slightly scaling. B. Confluent, erythematous papules and plaques with scaling were on the lateral proximal aspect and the internal plantar arch of the foot.

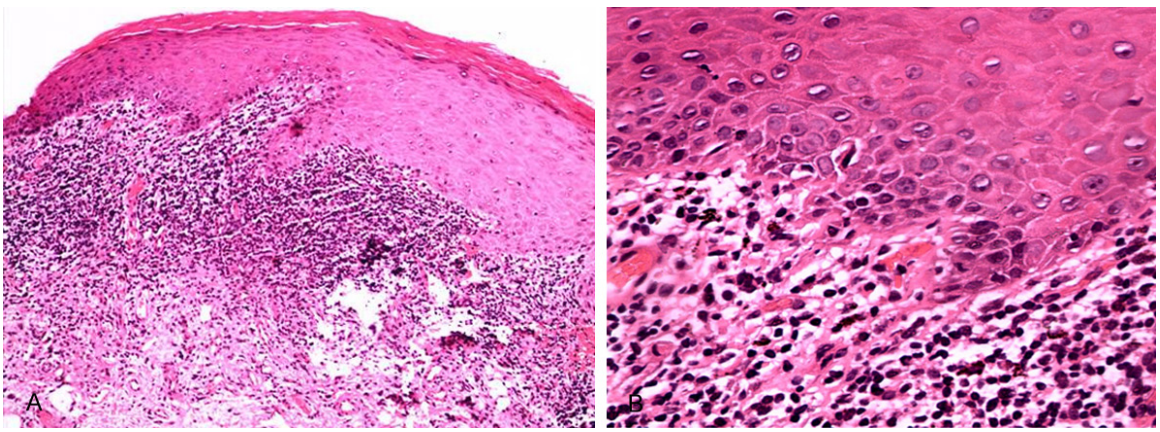

Figure 3. Histological changes of the lesion: A. Hyperkeratosis and mild irregular orthokeratosis overlying an acanthotic epidermis. HE, ×100. B. Interface dermatitis with vacuolar degeneration of the basal cell layer and mononuclear cells band-like inflammatory infiltrate in the upper dermis, on which scattered pigmentophage remained. HE, ×400.

At present, the etiology of LP appears to be multifactorial and complicated. Earlier studies have implicated immune changes, drug use, emotional factors, virus infections, and neurological disorders as the causes for LP, of which T cell immune-mediated responses play an important role in pathogenesis. Langerhans cells recognized external antigens such as, HBV, HCV virus, dental restorative materials and drugs, which produce increasing amounts of interferon-alpha (IFN-α) to activate cytotoxic cell in mediating apoptosis via the keratinocyte caspase cascade [9].

Corticosteroids and the immunosuppressants of hydroxychloroquine are the conventional drugs of therapy for LP. But corticosteroid and hydroxychloroquine usually induce many side effects that lead to poor adherence to the treatment for patients insistently. The patient in this case had received treatment previously of hydroxychloroquine without any relief. We pro-

ceeded with acitretin and topical corticosteroid therapy for the case. Alitretinoin is an endogenous vitamin A derivative, 9-cisretinoic acid. It can regulate keratinocytes proliferation, differentiation and apoptosis. The acitretin is known to possess anti-inflammatory properties, which may alter the cell surface antigens of the keratinocytes, causing an interaction with activated T-cells to mediate destruction of corneocytes [10]. At present, no clinical trials were done to systematically investigate the effect of alitretinoin treatment for LP, with only case reports. The current case we report suggests that acitretin has a good efficacy for treatment of LP of plaque and hyperkeratosis.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Junlian Liu, Department of Dermatology, 306 Hospital of PLA, 9 North Anxiang Road, Chaoyang District, Beijing 100101, PR China. Tel: +86-10-66356729; Fax: +86-10-64871261; E-mail: liujunlian306@sina.com

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