CASE REPORT

Keratoconjunctivitis and corneal ulceration in a patient with erosive lichen planus

Zahra Arooba[1], Anam Batool[2], Faizaan Asghar[3]

Abstract

Lichen planus (LP) is an immune mediated mucocutaneous disease with a broad clinical spectrum, primarily affecting the but skin and oral cavity. Erosive lichen planus (ELP) is a rare variant of LP that manifests as erosions in the oral cavity as well as chronic erosions of the soles accompanied with severe disabling pain. Ocular LP is very uncommon presenting usually as keratoconjunctivitis and if left untreated can lead to vision impairment. We report the case of a woman who presented with characteristic skin lesions of lichen planus, accompanied by severe erosive oral and plantar lesions as well as bilateral keratoconjunctivitis and corneal ulceration.

Key words: Lichen planus, erosive LP, ocular LP, keratoconjunctivitis.

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Introduction

Lichen planus (LP) is a common inflammatory disorder affecting the skin, nails, hair and mucous membranes, either concomitantly or sequentially. The condition most commonly presents in middle aged adults, with T-cell mediated autoimmunity is proposed to play a major role in its pathogenesis.[1,2] The cutaneous lesions typically appear on the wrists, forearms, ankles and sacral area, presenting as classic violaceous polygonal, plain-topped papules with an overlying fine network of white lines known as Wickham's striae. Mucosal involvement is characterized by reticular and erosive white patches, typically affecting the buccal mucosa, lips and genitalia.[3,4] The erosive variant of LP is characterized by slowly evolving, painful erosions mainly localized in the soles, palms, scalp, oral and genital mucosae, accompanied by typical LP lesions on the of skin and nails. Progressive toe nail destruction and dorsal pterygium formation are characteristic as the disease evolves. Malignant transformation to squamous cell carcinoma has been reported in mucosal erosions.[5] Ocular LP is extremely rare mainly affecting the eyelids and conjunctiva and can lead to severe scarring. Involvement of corneal epithelium is even rarer, with very few cases reported worldwide. Corneal involvement severely jeopardises eye's function and can even lead to corneal perforation. Clinically, ocular LP is indistinguishable from other causes of keratoconjuntivitis. Extraocular lesions of LP are present in most, though not all, patients with ocular lichen planus,[4-6] The clinical morphology of well-established lesions aids in making the diagnosis, which is confirmed by histolopatholgy. High-potency topical corticosteroids are the therapy of first choice for all forms of lichen planus. Systemic immunosuppression should be considered for severe and widespread disease.[7,8]

Case Report

On 25th June 2024, a 36-year-old female presented to the Dermatology Department of Mayo Hospital Lahore with generalized pruritic lesions over her entire body along with severely painful eroded plaques on hands and feet for past 8 months. The erosions at the tips of her digits had led to progressive loss of toenails. She also had painful erosions and ulcers in the oral and genital mucosa. Additionally for the past 3 months, she had been experiencing photophobia, ocular pain, and decreased vision in both eyes. Cutaneous examination revealed numerous grouped, violaceous, flat-topped papules and plaques, distributed symmetrically on the front and back of the trunk and distal extremities. There were eroded plaques on soles of both feet, around ankles and on tips of all fingers and toes (Fig1). All toenails were absent while the fingernails were dystrophic with dorsal pterygium formation (Fig2). In the oral cavity, she had confluent erosions on the buccal mucosa with superimposed candidiasis, confirmed by smears. There was no preceding history of any drugs or blood transfusion. Eye examination revealed conjunctival congestion, profuse watery discharge and the patient was unable to open her eyes due to pain and photophobia (Fig3). Vision in both eyes was reduced to light perception. Detailed evaluation from the ophthalmologist established the diagnosis of keratoconjunctivitis and corneal ulceration in both eyes.

[1,2]Department of Dermatology, Mayo Hospital, Lahore, Pakistan. [3]Final Year MBBS Student, King Edward Medical University, Lahore, Pakistan.
Correspondence: Zahra Arooba. **Email:** zahraarooba@gmail.com
ORCID ID: 0009-0008-3582-767X

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Figure-1: Painful eroded plaques bilaterally on soles of feet.

Figure-2: Lichenoid papulosquamous lesions over distal extremities, dystrophic finger nails and absent toenails.

Figure-3: Lichenoid patches over face, watery discharge from eyes with inability to open the eyes due to photophobia, erosions along borders of tongue.

Histological examination of skin biopsy revealed saw toothed acanthosis, thickened granular layer, dense band like lymphocytic infiltrate along the dermoepidermal junction, vacuolated basal layer cells, clumps of colloid bodies and pigment incontinence into the dermis. Direct immunofluorescence of the biopsy tissue showed deposition of IgM and fibrin at dermoepidermal junction and in colloid bodies. All these histopathological and immunofluorescence finding were consistent with the diagnosis of lichen planus. A paraclinical assessment in collaboration with the internal medicine department was conducted to exclude associated co-morbidities: biochemical profiles including blood sugar levels were found normal; serologies for HBV, HCV, HIV and antinuclear antibodies were negative.

The patient was started on oral prednisone at a dose of 1.5mg/kg/day. Artificial tears, lubricants and Moxifloxacin eye drops were advised by the ophthalmologists. After resolution of oral candidiasis, intralesional steroids were given in oral erosions. After two weeks of treatment, conjunctival congestion and photophobia had improved but corneal ulceration had not begun to heal. A corneal abscess was seen in the left eye, for which topical antibiotics were continued. Skin lesions were not showing satisfactory response so at two weeks, Azathioprine at the dose of 2mg/kg was started and steroids were gradually tapered. The patient was closely monitored for her cutaneous, mucosal and ocular symptoms. Her skin erosions were almost healed after 6 weeks, but oral erosions showed slower healing. The corneal ulcers showed signs of healing and re-epithelialization, with development of neovascularized opacification. The patient remains under follow-up by both dermatologist and ophthalmologist and keratoplasty is considered as part of her long-term management plan.

Discussion

Lichen planus is a multifaceted, chronic inflammatory disease. Differential diagnosis for papulosquamous cutaneous lesions of lichen planus includes a wide range of conditions such as drug eruption, eczema, psoriasis, pityriasis rosea, secondary syphilis, the lichenoid variant of mycosis fungoides, and graft versus host disease.[1,7,8] In our patient, the chronic nature of the lesions, absence of

a history of blood transfusion or use of any suspect medications, along with the characteristic morphology and distribution of the skin lesions and accompanying mucosal involvement, helped distinguish lichen planus from other conditions. The diagnosis was subsequently confirmed through histopathological analysis of skin biopsy samples.

Ocular involvement in Lichen planus is nonspecific but is usually associated with cutaneous and oral lesions, which aids in the definitive diagnosis. Very few cases of isolated ocular LP have been reported, making diagnosis very difficult.[5,9] Histopathologic examination of tissue samples, either from conjunctiva or other body sites, helps in distinguishing lichen planus from other cicatrising disorders, such as pemphigus vulgaris, mucous membrane pemphigoid, Stevens-Johnson syndrome, graft-versus-host disease and atopic keratoconjunctivitis, as these conditions may appear clinically.[4] Cutaneous biopsies are better and more specific than mucosal biopsies where ulceration masks the specific histopathological features needed for diagnosis. In our patient, all the screening done by ophthalmologist to rule out other known causes of corneal ulceration were negative. The presence of typical cutaneous and oral lesions suggested lichen planus as the aetiological factor.

Due to its chronic and refractory course, multiple treatment options have been suggested for treating erosive lichen planus with varying outcomes. These include systemic, local and intralesional glucocorticoids, azathioprine, cyclosporine, phototherapy, thalidomide, dapsone and biologicals targeting IL 12/23.[5] A systematic review published in 2012 intended to assess the systemic therapies for erosive LP revealed Azathioprine to be the most effective, while second in efficacy were systemic steroids. In our case, patient didn't respond well to corticosteroids alone, however the addition of azathioprine gave good results in terms of significant pain relief and healing of lesions. Long term follow up of all patients is mandatory to tackle with disease recurrence and for early detection of malignant transformation.[10]

Conclusion

Lichen planus is relatively a common disease with a wide range of clinical manifestations. Although ocular involvement is rare, it can lead to significant morbidity if not recognised and managed promptly. Early diagnosis, meticulous management and long term treatment is needed to avoid complications.

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AUTHOR'S CONTRIBUTION:
ZA: Concept, design, writing, final approval and agreement to be accountable for all aspects of the work.
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FA: Drafting, revision and agreement to be accountable for all aspects of the work.