Lupoid cutaneous Leishmaniasis: A Report of Three Cases from Nonendemic Area

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Abstract
The typical clinical presentations of cutaneous leishmaniasis are nodules, ulcer, nodulo-ulcerative lesions and crusted plaques. Besides classical clinical picture, several unusual and atypical clinical presentations of the disease have also been reported. Herein, we report three cases of lupoid cutaneous leishmaniasis to highlight the extended clinical spectrum of CL. Tissue smears were positive for Leishman-Donovan (LD) bodies. All patients were treated by azole antifungals with favorable response.

Key Words: Azoles, cutaneous leishmaniasis, lupus pernio, nonendemic region

Introduction
Cutaneous leishmaniasis (CL) is a vector-borne protozoal infection of the skin caused by several species of leishmania and transmitted by sandfly Phlebotomus. Phlebotomus papatasii is the most common vector for CL in India. The disease is endemic in the Western Thar Desert of Rajasthan.

Clinical presentation of CL varies depending on the leishmania species and host’s immune response. The typical clinical picture is a single or a few skin ulcers on the exposed parts of the body that generally heal spontaneously within 3–6 months. Besides classical clinical picture, several unusual and atypical clinical presentations of the disease have also been reported. Lupoid CL is one such relatively uncommon form of CL. We report three patients with lupoid CL to highlight the extended clinical spectrum of CL.

Case Reports
Three patients (two females and one male) aged 56, 26, and 24 years presented with 3-week to 4-month history of asymptomatic, solitary, sharply demarcated, erythematous to violaceous plaque affecting the tip of the nose [Figure 1a-c]. The clinical picture was almost similar in all the three cases [Table 1]. None of their family contacts had similar lesions. There was no definite history of sandfly bite or travel to endemic zone. The cases did not show clustering to a particular locality. General physical examination and systemic examination were normal. Laboratory investigations including hemogram, erythrocyte sedimentation rate, serum biochemistry, serum angiotensin-converting enzyme, serum calcium, chest X-ray, and X-ray hands were normal. Slit smear examination with Giemsa staining revealed Leishman–Donovan bodies [Figure 2a-c].

Discussion
CL is mainly confined to some pockets of Thar Desert of Rajasthan and Punjab, Himachal Pradesh, and Kerala. In Rajasthan, cases of CL are endemic in North and Northwest districts. All three patients in the index study belonged to South Rajasthan, a nonendemic area. None of the patients had a definite history of travel to endemic areas.

CL is diverse in its presentation and outcome. The clinical diversity mainly relates to parasite and host factors. Different species of Leishmania are associated with different clinical presentations. The disease is treated with azole antifungals with favorable response.
Various morphological forms of CL have been described, of which noduloulcerative form, classically known as oriental sore, is the most common. In addition to classical clinical picture, several unusual and atypical clinical variants including lupoid, sporotrichoid, paronychia, erysipeloid, psoriasiform, mycetomatous, chancriform, zosteriform, palmar/plantar, discoid lupus erythematosus-like, squamous cell carcinoma-like, eczematous, and verrucous have also been reported.

To the best of our knowledge, lupoid CL has not been reported from nonendemic area of Rajasthan. Lupoid CL is an uncommon form of CL with characteristic spread of the initial lesion resulting in solitary erythematous infiltrated plaque commonly on the face. It is generally considered to be a chronic form of leishmaniasis (duration >6 months). However, in a study, on 16 cases of lupoid CL, it was observed that 14/16 (87.5%) patients had a duration of their lesions <6 months. This was seen in all our patients too. Clinical differential diagnoses are lupus vulgaris, lupus erythematosus, lupus pernio (a form of cutaneous sarcoidosis), and facial erysipelas. Another very closely mimicking condition both clinically and histopathologically is leishmaniasis recidivans (LR) which often develops as a recrudescence on a previously healed lesion of CL. The scarring and relative resistant to treatment in LR is also a distinguishing feature.

In lupoid CL, the amastigote forms are rare or absent on a parasitological smear, but all three of our cases had positive parasitological smears. This could be explained on account of the relatively shorter duration of the lesions.

Lupoid CL is usually resistant to conventional therapies, and it spreads slowly for many years. However, all our three patients were treated successfully with 400 mg ketoconazole for 2 months. One patient (Case 3) was regular in follow-up and was in remission when seen 12 months after stopping therapy. The other two patients were lost to follow-up 6 and 8 weeks later. A telephonic enquiry revealed that both of these patients had been staying in remission 8 and 11 months after stopping treatment. Successful treatment response to antimonials and fluconazole in lupoid CL has been reported in other studies also.

Although lupoid CL is an easily diagnosable condition in the endemic areas, the diagnosis may be missed in nonendemic regions on account of lack of familiarity with the condition, and therefore, a high index of clinical suspicion should be kept in mind in such areas. CL both in typical and atypical clinical forms is increasingly reported nowadays from nonendemic regions. This could possibly be due to a new strain of parasite, a different local vector, and increased migratory
activity of population. To substantiate this, further studies should be focused on species characterization in patients presenting with unusual morphology.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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What is new?
- The occurrence of cutaneous leishmaniasis is rare in non-endemic area of South Rajasthan.
- We reported a rare clinical variant; lupoid leishmaniasis from the non-endemic region of South Rajasthan.