A case of mutilating localized cutaneous leishmaniasis caused by *Leishmania donovani* from Bhutan

Sir,

A 57-year-old man, a farmer from Bhutan, presented to us with a large reddish lesion on the left side of his face of around 6 months duration. He noticed a reddish nodular growth on the left lower eyelid 10 years back, which persisted and in the past 6 months grew to involve the left side of the face. He could not recall any prior trauma or insect bite on the site. He had never traveled outside his country. A topical steroid cream was used briefly, without improvement. He had a history of intermittent fever and weight loss one year prior, for which he was given anti-tuberculosis treatment for suspected underlying tuberculosis.

On examination, there was a large erythematous, indurated and crusted plaque (10 × 9 cm) on the left side of the face from the left eyebrow to the upper lip with complete ankyloblepheron and loss of architecture of the left eyelids [Figure 1]. Atrophy, scarring and telangiectasia were seen on the nose. There was no regional lymphadenopathy. The rest of the cutaneous and systemic examinations were normal. Our differential diagnoses included lupus vulgaris, atypical mycobacterial infection, sporotrichosis, histoplasmosis, cutaneous leishmaniasis and sarcoidosis.

Investigations revealed a hemoglobin of 8.9 g/dL. Enzyme-linked immunosorbent assay for human immunodeficiency virus and Mantoux test were negative. A Giemsa-stained skin smear showed numerous oval amastigotes within histiocytes [Figure 2a]. A biopsy revealed numerous amastigotes with characteristic bar-shaped paranuclear kinetoplasts and granulomatous inflammation, consistent with cutaneous leishmaniasis [Figure 2b]. The polymerase chain reaction of the tissue confirmed *Leishmania* [Figure 3a] and the polymerase chain reaction-reverse fragment length polymorphism analysis corresponded to *Leishmania donovani* [Figure 3b].

The rK-39 immunochromatic dipstick test from the blood sample was positive. Bone marrow aspirate and peripheral blood polymerase chain reaction were negative for *Leishmania*. Imaging of the face, chest and abdomen showed no significant abnormalities. We diagnosed him with localized cutaneous leishmaniasis due to *L. donovani*. He

was given a course of intravenous liposomal amphotericin B (3mg/kg/day) for 15 days. At the end of treatment, he had marked reduction in swelling of the lesion with signs of healing [Figure 4]. He is scheduled for follow-up.

Leishmaniasis is a zoonotic protozoal disease transmitted by sandfly vectors and is endemic in 88 countries in the tropical and subtropical zones.[1] Its presentation ranges from cutaneous leishmaniasis of varied morphology to systemic multi-organ involvement in visceral leishmaniasis.

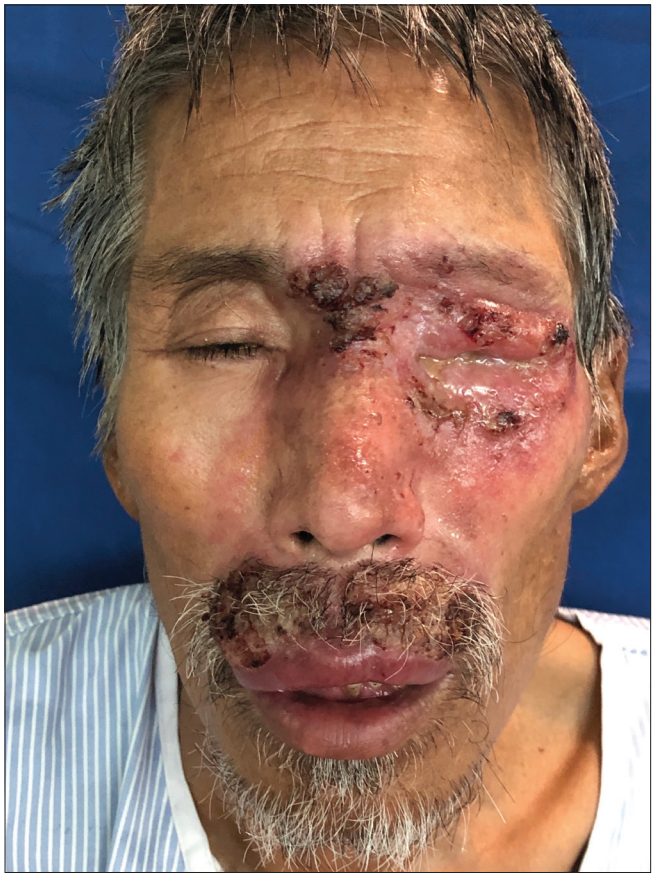

Figure 1: Large erythematous plaque with crusting on the face and destruction of the left eyelids

Localized cutaneous leishmaniasis is caused commonly by *L. tropica* and *L. major* in the Old World - the Mediterranean basin, Northern Africa, Kenya, the Middle East and in Rajasthan, Punjab and Himachal Pradesh in India.[1-3] *L. donavani-infantum* complex primarily causes visceral leishmaniasis in Asia and East Africa but has been recently reported to cause localized cutaneous leishmaniasis in Sri Lanka, the Middle East, Kenya and Himachal Pradesh and Kerala in India.[1,3-6] Localized cutaneous leishmaniasis has not been reported from Bhutan to date, making our case unique.

Polymerase chain reaction confirmed the presence of *Leishmania* DNA in the biopsy and reverse fragment length polymorphism analysis confirmed the causative species, in this case, *L. donovani*. The rK-39 immunochromatic dipstick test qualitatively detects circulating antibodies to a recombinant *Leishmania* antigen rK-39.[7] The antibody response to rK-39 is largely restricted to *L. donovani-infantum* complex that causes visceral leishmaniasis and there is virtually no response in localized cutaneous leishmaniasis due to *L. tropica* infection.[7] However, positive results in localized cutaneous leishmaniasis patients suggesting infection by *L. donovani-infantum* complex have been reported in India, strongly suggesting cutaneous infection by *L. donovani-infantum* complex.[2] Thus, with the parasitological, serological and polymerase chain reaction results and in the absence of evidence of past or current visceral leishmaniasis, we diagnosed him with localized cutaneous leishmaniasis due to *L. donovani*.

Our patient was a farmer from a rural area in the Wangdue Phodrang district in Bhutan. Nineteen cases of visceral leishmaniasis have been reported since 2006 in Bhutan, where

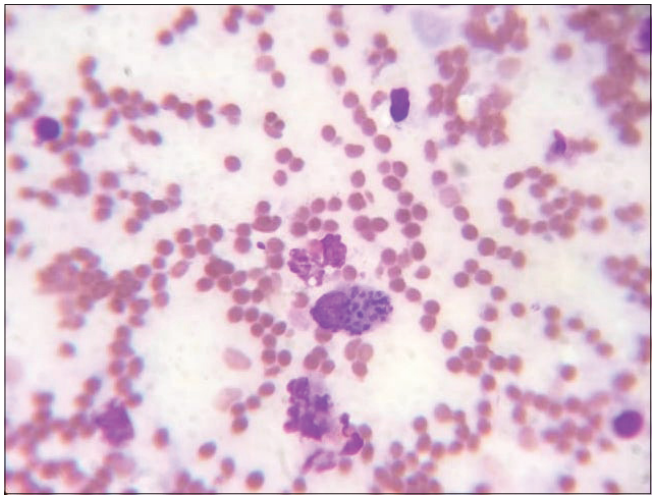

Figure 2a: Skin scraping with numerous amastigotes seen within a macrophage (Giemsa, ×100)

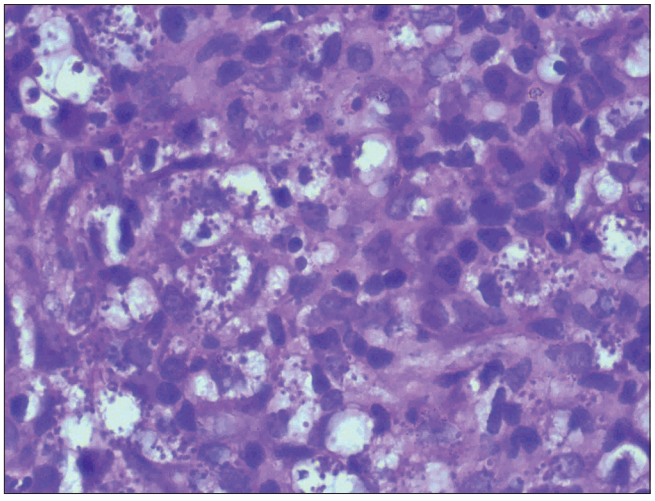

Figure 2b: Numerous oval amastigote forms of *Leishmania* within macrophages in the dermis (hematoxylin and eosin, ×100)

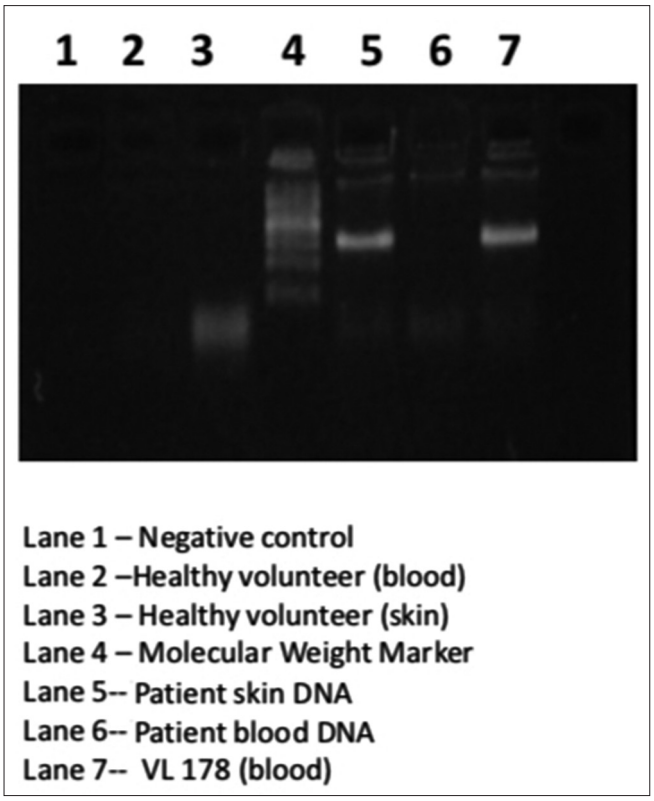

Figure 3a: Polymerase chain reaction tested for parasite DNA in the skin biopsy sample (Lane 5- positive) and in peripheral blood (Lane 6- negative) using *Leishmania*-specific primers (Lane 7 - positive control). Lanes1-3 - negative controls, Lane 4 - molecular weight marker

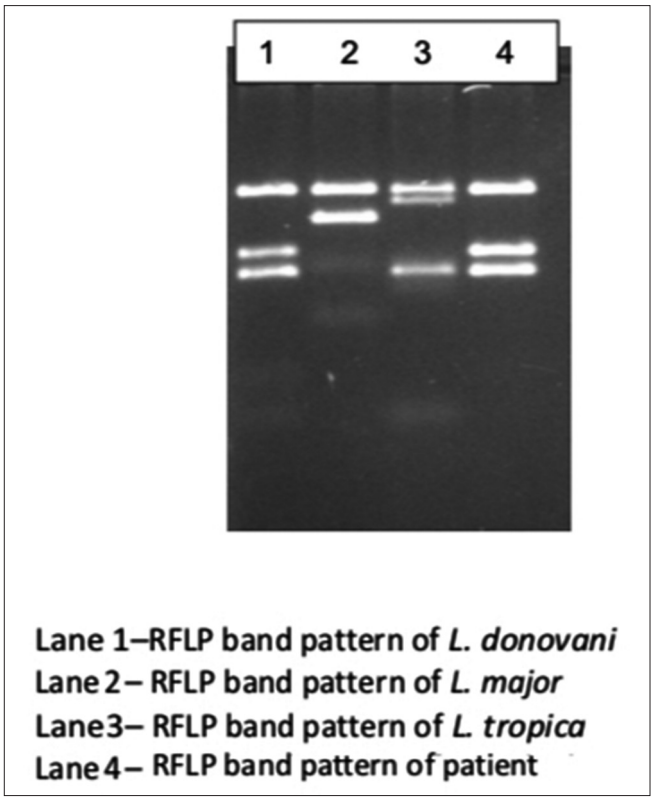

Figure 3b: Reverse fragment length polymorphism of internal transcribed spacer-1 region amplified from *Leishmania* species. The skin biopsy sample (Lane 4) reverse fragment length polymorphism band pattern matched with the reference strain of *Leishmania donovani* (Lane 1).

endemic transmission of visceral leishmaniasis has been suggested to occur.[8] The causative agent for these cases was *L. donovani*, with genotypes similar to Indian *L. donovani* isolates.[8] The presence of *Phlebotomus* and *Sergentomyia* sand flies, known vectors for visceral leishmaniasis and localized cutaneous leishmaniasis in India and China, has been confirmed in several regions here. The increasing prevalence of leishmania skin test positivity with age in the community suggests the presence of parasite for years and becoming indigenous to Bhutan.[8] The occurrence of localized cutaneous leishmaniasis caused by *L. donovani* in such a scenario could be the result of incidental human intrusion into the otherwise well-maintained zoonotic cycle. Other possible factors are the genetic susceptibility of populations and adaptations by parasitic strains.[3,6]

Strong clinical suspicion is the key for early diagnosis of localized cutaneous leishmaniasis especially in non-endemic areas. This will enable early treatment, can minimize patient morbidity and prevent disfigurement. Epidemiological and genetic studies in emerging geographic foci will play an important role in the control of leishmaniasis.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name

Figure 4: Marked reduction in swelling, with signs of healing and surrounding post-inflammatory hyperpigmentation immediately post-treatment

and initials will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There is no conflicts of interest.

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