# Cutaneous Lupoid Leishmaniasis: A Case Report

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GOAL

To gain a better understanding of cutaneous lupoid leishmaniasis (LL) to better manage patients with the condition

OBJECTIVES

Upon completion of this activity, dermatologists and general practitioners should be able to:

1. Recognize the clinical presentation of LL.
2. Discuss the differential diagnosis for LL.
3. Describe the treatment options for patients with LL.

CME Test on page 37.

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*Cutaneous leishmaniasis is a disease transmitted by the sandfly. During the course of the disease, all classical stages of the development of leishmaniasis from small erythematous papules to nodules to ulcerative lesions can be seen. We report a case of lupoid leishmaniasis (LL) treated with daily intramuscular injections of meglumine antimoniate for 20 days with marked improvement of clinical features.*

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Cutaneous leishmaniasis has several types of lesions, all of which tend to occur on exposed parts because the disease is transmitted by the sandfly. The resulting syndrome depends upon a complex interaction between a specific species of *Leishmania* and the genetic and immunologic status of the host. Ultimately, either the patient's immune response is able to eliminate the infection and effect

a spontaneous cure, or the immune response fails and a chronic form of leishmaniasis develops. The lupoid type spreads peripherally on a common erythematous base and occurs most commonly with the urban type of disease, caused by *Leishmania tropica*.[1,2]

## Case Report

A 69-year-old woman was admitted with coalescent, erythematous, papulo-infiltrative, nodular and verrucous plaques on her whole face. The lesion on her nose started one year ago as a slowly growing, indurated, livid, indolent papule that gradually enlarged and spread peripherally to cover a large part of her face in a few months. There was no response to topical antibacterial therapy. Results of a dermatologic examination revealed extensive confluent erythematous papules and infiltrative nodular and verrucous plaques with ulceration that were crusted over. The papules and plaques were localized on the patient's forehead, glabella, eyelids, nose, cheeks, and upper lip (Figure 1). The chin and lower lip were spared. Results of a complete clinical evaluation showed obesity and hypertension. There was no evidence of systemic involvement.

The results of the routine laboratory tests, including complete blood counts and serum biochemistry, were within reference range except for the erythrocyte sedimentation rate, which was 70 mm/h. *Staphylococcus aureus* was isolated from the bacterial culture of the lesion.

Amastigotes of *Leishmania* species were seen both in monocytes and extracellularly in cutaneous scrapings from the center of the ulcer (Figure 2). A punch biopsy (4 mm in diameter) was performed at the edge of the plaque. Results of a histologic examination revealed a heavy infiltrate of histiocytes, lymphocytes, polymorphonuclear leukocytes, and a few plasma cells under the epithelium. Numerous organisms were present (mostly in histiocytes), which were nonencapsulated and contained a nucleus and a paranucleus (Figure 3). Absence of parasites in the smear prepared from the bone marrow biopsy eliminated the diagnosis of visceral leishmaniasis.

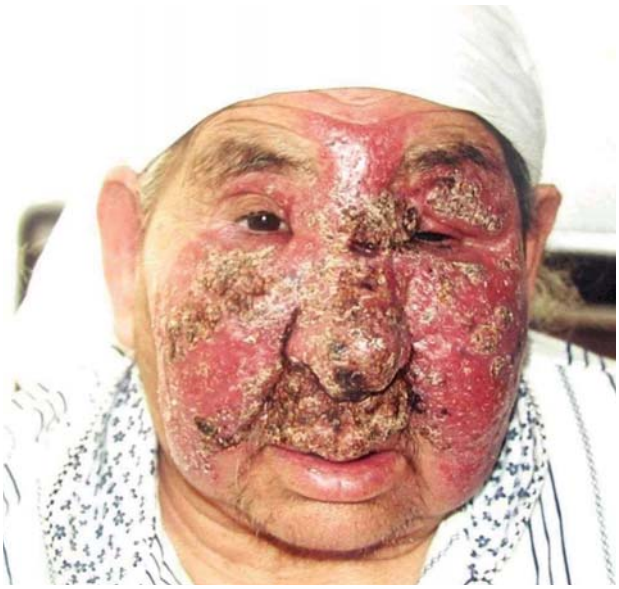

**Figure 1.** Extensive confluent erythematous papules and infiltrative nodular and verrucous plaques with crusted ulceration localized on the face.

The patient's history, together with the clinical and histopathologic findings and the parasites in the

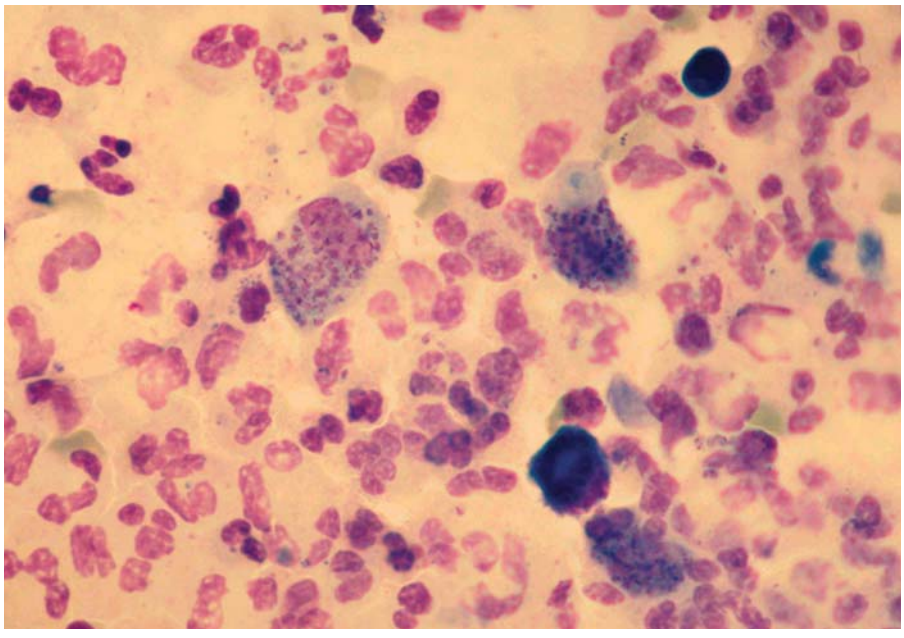

**Figure 2.** Amastigotes of *Leishmania* species were seen both in monocytes and extracellularly in cutaneous scrapings from the center of the ulcer (Giemsa, original magnification ×100).

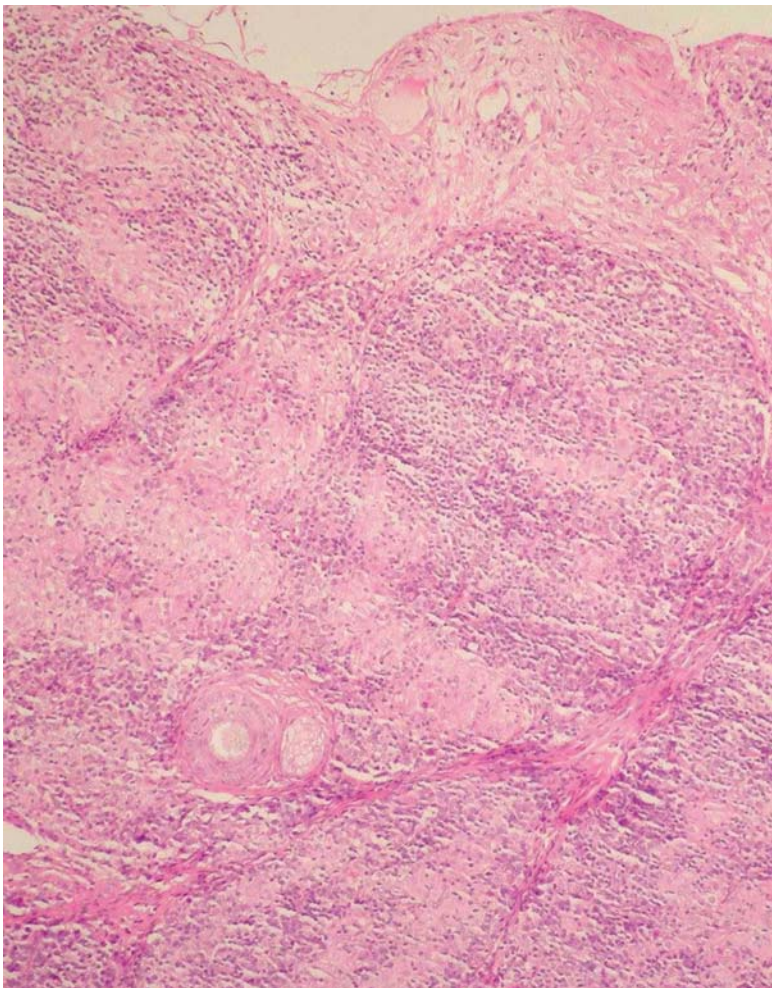

**Figure 3.** A dense inflammatory infiltrate in the dermis composed of mostly epitheloid histiocytes formed as granulomas and lymphocytes (H&E, original magnification ×100).

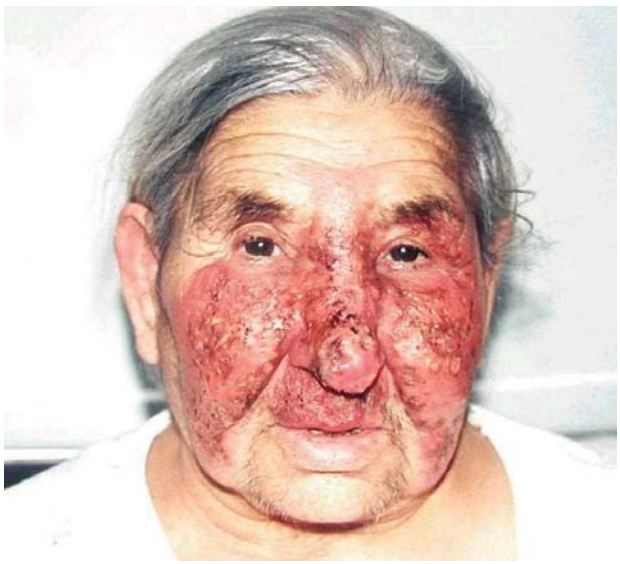

**Figure 4.** Less indurated erythematous plaques after 20 days of therapy.

smear obtained from the lesion, supported the diagnosis of lupoid leishmaniasis (LL). Systemic meglumine antimoniate treatment was started with 20 mg/kg per day intramuscularly and continued for 20 days. In addition to this therapy, an oral systemic antibiotic (ampicillin and sulbactam), 375 mg twice daily for 10 days, a topical antiseptic dressing, and an antibacterial ointment were used for secondary bacterial infection. During the treatment, transient elevations were detected in serum aminotransferase levels. After 20 days of therapy, lesions became less indurated and smooth (Figure 4), and no parasites were found in the smear obtained from the lesion. The patient was followed for one year after completion of treatment, and no relapse was observed.

## Comment

*Leishmania* infection following a bite from an infected sandfly may remain subclinical or may develop after an incubation period of 1 to 12 weeks. During the course of the disease, all classical stages of the development of leishmaniasis from small erythematous papules to nodules to ulcerative lesions can be seen. Secondary bacterial infection is common, which can lead to pain.[2]

Although LL has been used as a synonym for leishmaniasis recidivans, Oliveira-Neto et al[1] stated that there is a clear difference between leishmaniasis recidivans and LL. This difference can be defined as LL being the initial clinical presentation, and leishmaniasis recidivans being a recurrent lesion. Therefore, it is not appropriate to use these 2 names synonymously. LL is a chronic condition that typically follows an acute cutaneous leishmaniasis infection. While the acute lesion heals with scarring, papules and nodules become apparent. The papules have a granulomatous, lupoid appearance and are often associated with ulceration and crusting, as in our case. The papules characteristically are present at the edge of the scarred area. Reported cases are associated predominantly with Old World rather than New World strains of leishmaniasis, with *L tropica* being the causative agent in most cases.[3] Incidences reported for LL, following simple acute cutaneous leishmaniasis, range from 0.5% to 6.2% and are most prevalent in endemic areas of leishmaniasis, particularly in the Middle East.[3,4]

The clinical differential diagnosis of cutaneous leishmaniasis includes lupus vulgaris, verrucous skin tuberculosis, chronic leishmaniasis, discoid lupus erythematosus, and basal cell carcinoma. The histopathologic changes include epidermal atrophy or sometimes hyperkeratosis and acanthosis, follicular plugging, and a diffuse dermal granulomatous infiltrate composed of histiocytes, lymphocytes, giant cells, and plasma cells. The recurrent lesions generally resemble lupus vulgaris, with tuberculoid granulomas surrounded by a rim of lymphocytes and histiocytes and some giant cells. However, caseation necrosis is generally absent.[1,5]

Cutaneous leishmaniasis can become disseminated especially in immunosuppressed persons. There is still some question about the pathogenesis of this form of leishmaniasis, though factors such as the specific species involved, the host's immune response, the hormonal changes encountered with increasing age, and the changes in skin barrier with aging can be considered important points in causing such an unusual presentation.[6,7] Indeed, parasites cause a defect in T-lymphocyte activation that the macrophages cannot kill.[1,8] We think that the lesion became disseminated because our patient was elderly and obese. LL should be considered in all patients from endemic areas (like our country) of leishmaniasis who present with cutaneous large nodulo-plaques.

There is no standardized treatment for this condition and thus multiple treatments have been reported with varying degrees of success.[2,9] The pentavalent antimony derivatives sodium stibogluconate and meglumine antimoniate remain the mainstay of systemic treatment. Their mode of action is not known, though they inhibit glycolysis and fatty acid oxidation in *Leishmania*. Their efficacy is well established provided they are given for an adequate length of time.[3] This case was treated with daily intramuscular injections of meglumine antimoniate for 20 days with marked improvement of clinical features.

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