SKIN

SKINmages

An Elusive Case of Cutaneous Leishmaniasis

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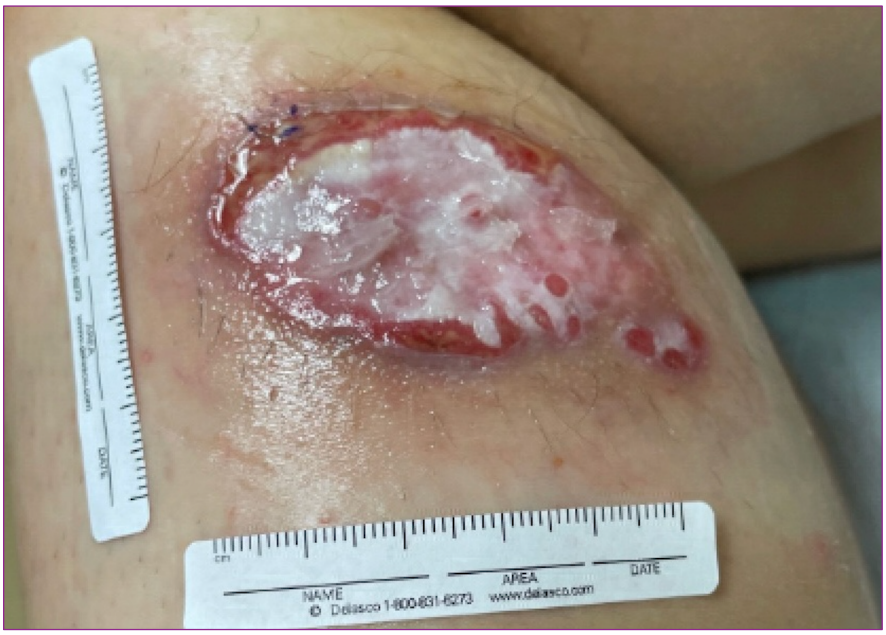

Figure 1. Nonhealing ulcer of the left posterior lateral leg at the time of presentation.

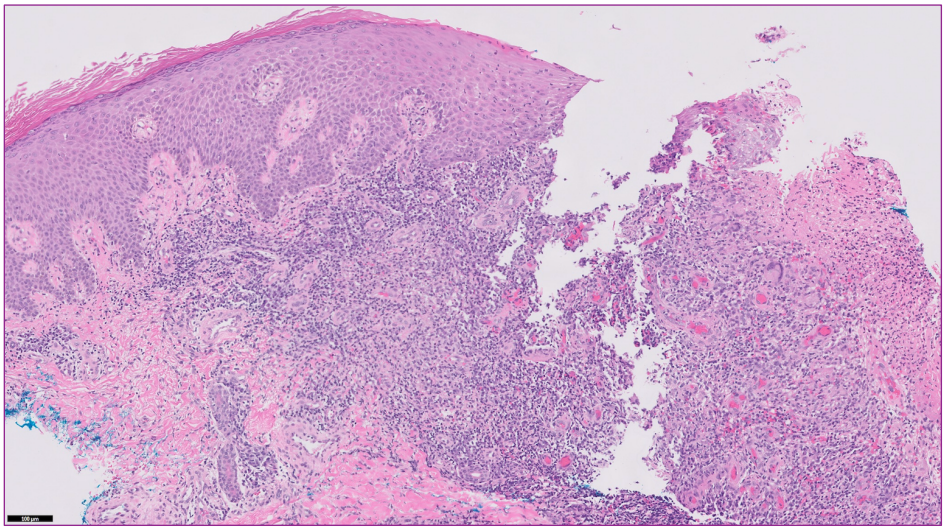

Figure 2. Punch biopsy taken from the ulcer edge on the left posterior lateral leg, 10x magnification. No evidence of amastigotes was noted on tissue culture. Black scale bar indicates 100 µm.

CASE REPORT

A 17-year-old otherwise healthy woman presented with a seven-month history of a nontender, nonhealing ulcer on the left posterior lateral leg that developed two months after vacationing in Eastern Costa Rica.

Six months prior, she presented to an outside institution and started empiric topical bacitracin without improvement. An initial punch biopsy revealed superficial and deep perivascular dermatitis with sparse plasma cell aggregates. After a second adjacent ulcer formed, a repeat punch biopsy showed nodular diffuse infiltration of lymphocytes, histiocytes, and plasma cells. Infectious workup including acid-fast bacilli (AFB), Fite, periodic acid Schiff with diastase (PAS-D), and Brown-Brenn stains were negative. Tissue culture grew *Enterococcus* and *Pseudomonas putida*, favored to be contaminants or colonization. Laboratory workup including complete blood counts, C-reactive protein (CRP), erythrocyte sedimentation rate (ESR), antinuclear antibody (ANA), anti-neutrophil cytoplasmic antibody (ANCA), anti-cardiolipin, cryoglobulin, antithrombin 3 activity, protein C/S, and complement C3 and C4 were unremarkable.

Given initial concern for lupus panniculitis, prednisone (1 mg/kg/day for two weeks followed by a six-week taper) was initiated with slight improvement. She then started a second course of prednisone (0.75 mg/kg/day) with topical clobetasol. When the ulcer failed to resolve, prednisone was increased to 1 mg/kg/day with a three-month taper, and the patient was referred to our clinic for further evaluation.

Examination revealed a 5 x 3 cm oval ulcer with yellow-white fibrinous material and violaceous borders on the left posterior lateral leg (**Figure 1**). Upon further discussion, the patient endorsed an extensive recent travel history to Poland, France, Mexico, Canada, Hawaii, the southwestern United States, and Israel, raising suspicion for an atypical infectious process. Punch biopsies from the ulcer edge revealed nonspecific diffuse dermal inflammation with rare giant cells (**Figure 2**), and tissue culture was negative for bacteria, fungi, atypical mycobacteria, and amastigotes. Despite amastigote-negative biopsies and tissue culture, fungal DNA sequencing of tissue culture revealed *Leishmania spp*. Tissue sent to the Center for Disease Control and Prevention for polymerase chain reaction (PCR) confirmed *Leishmania panamensis.*

The patient received a 28-day course of oral miltefosine 50 mg three times daily with complete reepithelialization and resolution of the ulcer three months later (**Figure 3**).

DISCUSSION

A neglected tropical disease, CL remains highly overlooked despite its global prevalence of 3.8-3.9 million cases/year.[1–3] Unfortunately, there is no gold standard diagnostic test for CL. Histopathology is rapid and common, though not sensitive. Parasite culture is highly specific and allows for species identification, but can be limited by poor parasite growth *in vitro* .[1,2] PCR is the most sensitive assay and allows for species identification and treatment selection,[1,2,4] as different *Leishmania* species vary in pathogenicity and drug susceptibility.

Diagnosis of CL can be difficult and obtaining a thorough travel history when assessing

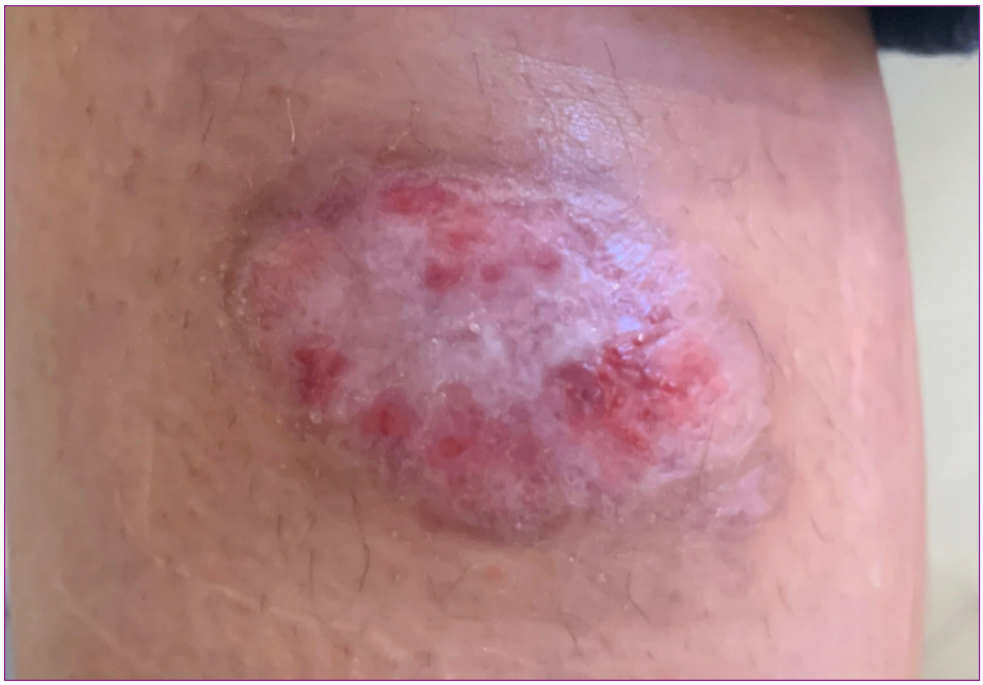

Figure 3. Healed ulcer three months after completing oral miltefosine 50 mg three times daily for 28 days.

patients with nonhealing ulcers are essential, as well as consideration of molecular diagnostics when traditional diagnostic techniques are unrevealing. Ultimately, PCR confirmed the diagnosis of CL in this challenging case and, as next-generation diagnostics are increasingly used in dermatology, providers should consider utilizing molecular techniques including PCR when evaluating nonhealing ulcers.

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