An Unusual Presentation of Cutaneous Leishmaniasis: The Role of Skin Ultrasound

Leishmaniasis cutánea de presentación inusual. Papel de la ecografía cutánea

To the Editor:

Cutaneous leishmaniasis (CL) manifests as a papule or nodule that forms in response to the bite of mosquitoes that carries this parasitosis. These lesions tend to grow to form a well-defined plaque with a raised violaceous border that may ulcerate, leading to subsequent formation of a crust. Atypical presentations of CL are increasingly common, and pose a diagnostic challenge.

Diagnosis is based on histological and microbiological findings (i.e., a positive result in polymerase chain reaction [PCR] analysis of blood or tissue). However, skin ultrasound can be a useful complementary technique for CL diagnosis and a tool to monitor treatment response in CL patients.

We describe 2 cases of CL with an unusual, erysipelasoid presentation, and the corresponding ultrasound findings.

Case 1

A 62-year-old man with a personal history of hepatic porphyria cutanea was seen for a lesion covering a large portion of the external aspect of the shoulder and upper left arm. The lesion had appeared 2 months earlier and was occasionally suppurative. The patient had been previously treated for suspected cellulitis with multiple oral antibiotics, without improvement. Physical examination revealed an indurated, erythematous plaque (9 × 7 cm) with poorly defined borders that was hot to the touch (Fig. 1A). Histology revealed non-necrotizing granulomatous dermatitis and the presence of Leishmania bodies within the cytoplasm of the histiocytes. Skin ultrasound (SonoScape, 15-MHz linear probe) was performed to evaluate the extent of the lesion.

References


M. Mazzeo,* A. Dattola, M.V. Cannizzaro, L. Bianchi

Departamento de Dermatología, Universidad de Roma «Tor Vergata», Roma, Italia

*Corresponding author.
E-mail address: mauromazzeo@hotmail.it (M. Mazzeo).

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and showed diffuse thickening of the dermis in the affected area (Fig. 1B). Because of the lesion’s large size, the patient was treated with intravenous amphotericin B (5 mg/kg/d) for 3 days (total dose, 15 mg/kg). The lesion resolved in response to treatment (Fig. 1C), as confirmed by subsequent ultrasound (Fig. 1D).

**Case 2**

A 66-year-old man with a personal history of diabetes mellitus type 2 and dyslipidemia was referred from the otorhinolaryngology department with an asymptomatic lesion on the right pinna that had appeared 5 months earlier. He reported no previous trauma or insect bites in the affected location. The patient had been diagnosed with erysipelas, for which he was treated with several oral and topical antibiotics, with no improvement. Physical examination revealed diffuse erythema on the right pinna. On the earlobe and antitragus the erythema was more pronounced and was accompanied by marked thickening (Fig. 2A). Histology revealed similar findings to those described for Case 1. The result of a PCR test for *Leishmania* DNA was positive. Ultrasound (SonoScape, 15-MHz linear probe) revealed an unencapsulated, hypoechoic structure in the superficial dermis that was well-delimited near the surface and less so at deeper levels, and exhibited increased low-resistance flow on Doppler imaging (Fig. 2B). The lesion resolved after treatment for 4 weeks with weekly infiltrations of intraleisional meglumine antimoniate (injected subdermally until a wheal formed) (Fig. 2C and D).

Examination of both patients after 6 months revealed no signs of lesion recurrence or reactivation.

Leishmaniasis is a group of diseases caused by infection with any of over 20 protozoan species of the genus *Leishmania*, and is transmitted by the bite of insects of the genera *Phlebotomus* and *Lutzomyia* in the Old World and New World, respectively. The most common reservoirs are domestic mammals such as the dog, cat, rat, hare, and rabbit.  

CL has many different presentations, and recent years have seen an increase in atypical forms. The clinical presentation of leishmaniasis depends, among other factors, on the species involved and the immune response of the host.  

In a series published by Bari and Rahman, up to 5.7% of the 718 cases described were considered unusual forms of CL. These included lupoid, sporotrichoid, paronychial, erysipeloid, palpebral, psoriasiform, mycetoma-like, chancriform, scar, zosteriform, palmar/plantar, verrucous, and eczematous CL. *Lutzomyia mexicana* causes a form of CL that is uncommon in the Old World but is transmitted by *Lutzomyia olmeca* in the New World, where it typically affects rubber workers and in over 50% of cases results in the formation of an ulcerated lesion on the pinna known as a *chiclero ulcer*. Occasionally these atypical presentations can simulate other infectious or inflammatory skin conditions. The erysipeloid form, of which few cases are described in the literature, is characterized by poorly defined erythematous plaques that resemble those seen in erysipelas or cellulitis. In our patients, who had erythematous and indurated plaques on the shoulder and pinna, respectively, the initial diagnostic suspicions were cellulitis and erysipelas, respectively. Both patients responded poorly to subsequent antibiotic treatment.

Although suspected CL is often diagnosed clinically, diagnosis should be confirmed using microbiological (*Leishmania* culture in special media, direct examination, PCR of tissue and/or blood samples) and/or histological techniques.
Skin ultrasound is a fast, safe, and effective technique, and is increasingly used in dermatology. We have found no published descriptions of ultrasound findings in CL patients. In our patients, ultrasound revealed diffuse thickening of the dermis in the first case, accompanied in the second case by a poorly defined and vascularized structure in the superficial dermis and an increase in the thickness of the dermis. These alterations resolved after treatment. Although the ultrasound findings were nonspecific, ultrasound served as a useful additional diagnostic tool to identify dermal alterations and increased Doppler flow. More importantly, in both cases it allowed us to monitor the treatment response, and revealed resolution of the structural alterations in the subepidermal tissue. This would not have been possible by visual examination.

We describe 2 cases of erysipeloid CL. CL should be included in the differential diagnosis of lesions suggestive of erysipelas or cellulitis that do not respond to conventional antibiotics.

We wish to highlight the role of ultrasound in these cases not only as an additional diagnostic technique, but also as a means of monitoring treatment response.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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Circumscribed Palmar Hypokeratosis: Treatment with Cryotherapy

Hipoqueratosis circunscrita palmar: tratamiento mediante crioterapia

To the Editor:

Circumscribed palmar hypokeratosis (CPH) is a skin disorder characterized by a well-circumscribed, depressed, reddish area with a scaly border. Lesions tend to be solitary and are typically located on the thenar and hypothenar eminences of the palms. There are no established treatments. We present 2 cases of CPH treated with cryotherapy on the thenar eminence of the palms of 2 patients.

The first patient was a 60-year-old woman with no relevant medical history who presented with a well-circumscribed, erythematous, depressed, lesion with a stair-like border on the thenar eminence of the right hand (Fig. 1A). The lesion was not hard to the touch and had appeared a year earlier. It had been treated with topical corticosteroids but shown no improvement. The second patient was a 66-year-old woman, also with no relevant past history, who presented with a similar plaque of 7 years’ duration on the thenar eminence of the right palm (Fig. 2A). The lesion had grown progressively over the years. Both patients denied triggers.

Dermoscopy in both cases revealed an erythematous-pink, round, central area with white spots and a stair-like border with desquamation. In both cases, histologic examination showed depression of the epidermis with a sharp stair-like border between the normal and affected skin (Fig. 3). The depressed epidermis showed hypokeratosis and hypogranulosis compared with the surrounding skin. A cornoid lamella was not observed in serial slices. There were also no signs of atypia. The histologic findings were consistent with a diagnosis of CPH. CPH was first described in 2002. It clinically presents as a round, erythematous, circumscribed, asymptomatic, generally solitary, lesion on the thenar or hypothenar eminences of the palms, although plantar lesions have been reported. The condition usually affects women aged between 51 and 70 years, although there has been a report of a congenital case. Numerous hypotheses have been proposed to explain the etiology and pathogenesis of CPH, including human papillomavirus...


Figure 1 Case 1. Before (A) and after (B) treatment.