

Cutaneous Lymphoid Hyperplasia Associated with *Leishmania panamensis* Infection

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Cutaneous leishmaniasis (CL) is an infection caused by protozoa from the genus *Leishmania*. The disease is transmitted by sandflies. Reservoirs are dogs, mice, wild rodents and, more rarely, humans. CL is clinically characterized by a single, polymorphous lesion, usually localized on the face or limbs. Nowadays, CL is more frequently seen among travellers returning from tropical and subtropical countries (1).

Cutaneous lymphoid hyperplasia (CLH), also known as pseudolymphoma, is a reactive proliferation, probably secondary to persistent antigenic stimulation.

We present here a case of CLH associated with *L. panamensis* infection.

CASE REPORT

In May 2005 a 66-year-old Ecuadorian woman presented with a 15-month history of 3 brownish, crusted nodules on her right arm (Fig. 1a), left leg and forehead (Fig. 1b). Clinical history and general physical examination were unremarkable. Histopathological examinations of specimens from both the forehead and the arm revealed a prominent dermal infiltrate of small-to-medium-size lymphocytes, plasmacytes, blasts and histiocytes, with a nodular pattern and reactive germinal centres (Fig. 2a and b). The infiltrate was predominantly of B lymphocytes CD20+, CD79a+ and several plasmacytes CD138+. Light chain restriction was not detected. Bcl-2 revealed a strong lymphocyte staining pattern, while Bcl-6 was expressed only in reactive germinal centres. A considerable number of atypical mitotic features were observed in the infiltrate. PCR failed to demonstrate clonal rearrangement of either T-cell receptors and immunoglobulin genes. Amastigotes were not detected by haematoxylin and eosin or Giemsa stains; only PCR analysis, performed on paraffin-embedded tissue, proved the aetiology of the infection. Sequencing of the amplified fragment revealed a 100% match with *L. panamensis*. A diagnosis of CLH associated with *L. panamensis* infection was therefore confirmed.

The patient was treated with intravenous pentamidine (6 infusions of 200 mg) and oral fluconazole (200 mg/day for 2 months) and remission was observed within 6 months. A careful follow-up was performed in order to detect signs of evolution into cutaneous lymphoma. At the time of writing (February 2010), the patient is alive without clinical evidence of leishmaniasis infection and with a marked regression of the CLH.



Fig. 1. (a) Clinical presentation of cutaneous lymphoid hyperplasia at onset: erythematous papular lesions on the right arm. (b) Brownish crusted nodule on the forehead.

DISCUSSION

L. panamensis belongs to the New World cutaneous leishmaniasis group, specifically to the *L. braziliensis* complex. Histopathological findings of a recent-onset lesion typically show histiocytes with numerous grey-blue dots in the cytoplasm throughout the reticular dermis. Very few organisms in the upper dermis are diagnostic of chronic lesions (2). Sometimes, clinical and histopathological presentations lack the typical features, making the diagnosis very difficult. In fact, in our patient a first misdiagnosis of cutaneous marginal zone B-cell lymphoma was considered. In these controversial cases, a diagnosis is not possible based on morphological findings alone, and PCR can be helpful (3). Infections, drugs and contact allergens are involved in the pathogenesis of CLH.

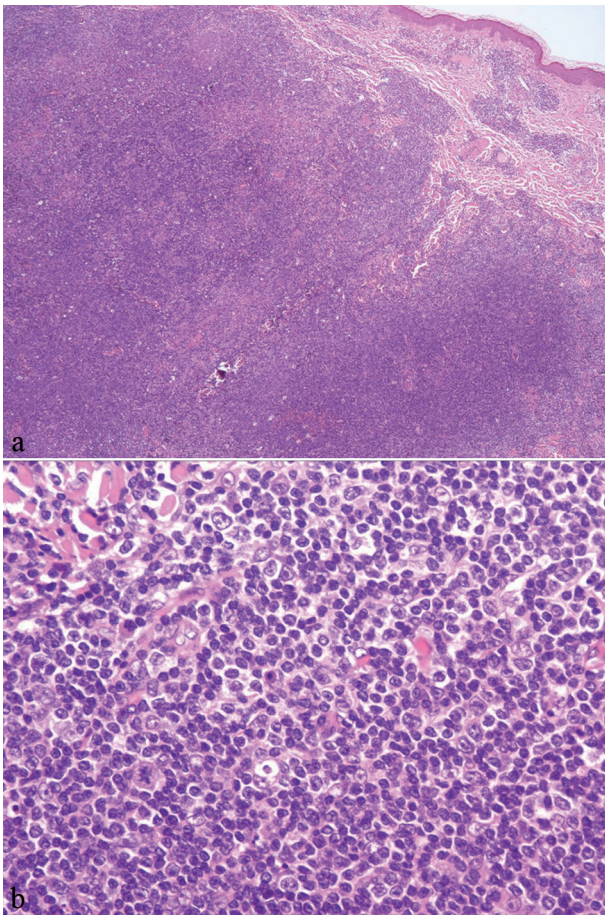


Fig. 2. (a) Histopathology of the dermis showing a nodular pattern infiltrate and reactive germinal centres (haematoxylin and eosin (H&E); $\times 25$); (b) infiltrate of small-to-medium-size lymphocytes, plasmacytes, blasts and histiocytes, in the absence of amastigotes (H&E; $\times 400$).

Only two cases of CLH associated with leishmaniasis infection have been reported in the literature (4, 5), one caused by *L. donovani* (5). To our knowledge this is the first reported case of CLH associated with *L. panamensis* infection. This case may represent an additional clinical variant of *L. panamensis* presentation, and adds a new aspect to the pathogenesis of pseudolymphoma.

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The authors declare no conflict of interest.

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