

SHORT COMMUNICATION

Cutaneous Leishmaniasis Mimicking A Pyogenic Granuloma

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The *Leishmania* genus is formed by parasitic protozoa which are transmitted by the bite of infected female sand flies. Cases of sexual, vertical or transfusional transmission or via infected needles have also been described. In humans, 4 forms of this disease have been described: localised cutaneous (LC), diffuse cutaneous, mucocutaneous and visceral (1). LC counts for 50–75% of all cases (2), it is the mildest form of the disease and can be caused by any species of *Leishmania*. In Spain, the most frequent form is the oriental sore caused by *L. infantum* (2). Most cases resolve spontaneously within one year. In United States and Europe, the incidence is increasing due to tourism and co-infection with HIV. The morphological spectrum of LC is very wide; the most characteristic presentation is the nodular ulcerative lesion, characterised by painless crater-like ulcers with a necrotic base and covered by an adhesive crust. The main complication of LC is its progression in some strains towards the other 3 forms of the disease (3). In patients with AIDS and other diseases associated with immunosuppression the risk of dissemination is much higher than in the immunocompetent. We present a case of LC with clinical and histopathological features similar to a pyogenic granuloma.

CASE REPORT

A 43-year-old man, former drug-user by injection, co-infected with hepatitis C virus and HIV (stage C3), who had begun antiretroviral therapy treatment 2 weeks before visit to our clinic. The patient was consulted due to an exophytic lesion with a 3-month progression, located on the internal lateral side of the thumb of the right hand, after infection probably from a sand fly in Seville, Spain. On clinical examination, it presented a soft red wine-coloured nodular element, 2 cm in diameter and a central haemorrhagic scab (no photos taken). Clinical diagnosis was pyogenic granuloma, thus proceeding to excision by shaving and electrocoagulation. Histopathological study showed vascular proliferation covered by hyperplastic epidermis with peripheral collarettes similar to those seen in pyogenic granuloma. Dermal stroma was oedematous with areas of lymphoplasmacytic inflammatory infiltrate and numerous histiocytes containing intracytoplasmatic microorganisms identified as *L. amastigotes* (Fig. 1). The absence of symptoms and signs suspicious of systemic disease led to the diagnosis of LC with an angiomatoid pattern. At present, 6 months after complete excision, the lesion has not recurred.

DISCUSSION

Morphologically, different types of LC have been described: zosteriform, erysipeloid, lupoid, sporotrichoid,

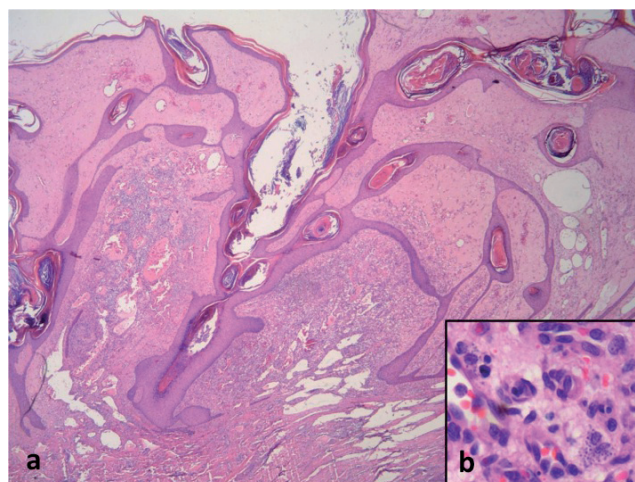


Fig. 1. Pyogenic granuloma-like reactive vascular lesion, formed by a vascular proliferation covered by hyperplastic epidermis with collarettes (a). Lymphohistiocytic inflammatory infiltrates with plasma cells (b).

eczematoid, psoriasiform, chancreform, annular, keloid-like or warty (4, 5), suggesting an inflammatory or tumoural aetiology (6). This morphological variability has been related to the humoral and/or cellular immune response of the host and to the type of *Leishmania* involved (7). Histopathology of the initial lesions of LC usually shows a dermal inflammatory infiltrate, dense and diffuse, formed by histiocytes, lymphocytes and plasma cells. The most effective diagnostic test is identification of amastigotes in the histiocyte cytoplasm (also known as Leishman-Donovan bodies) or in the extracellular space after vacuolar rupture. PCR is a decisive and accurate diagnostic technique despite the fact that it is difficult to perform and its success depends on the stage of the disease (7). As the lesion progresses, the number of amastigotes per microscopic field decreases and histological features are more similar to chronic LC, predominating a granulomatous tuberculoid pattern without nodular/diffuse necrosis (8). In 1980, Ridley (9) proposed 5 different groups by histological response: (i) histologically normal skin with areas of collagen degeneration, (ii) dermal necrotizing process, (iii) diffuse dermal inflammatory infiltrate, (iv) scarce Langerhans' giant cells and epithelioid histiocytes and (v) properly formed epithelioid granulomas. However, there are many publications of cases which do not fit neatly with any of these groups. Atypical forms of presentation usually show a marked epidermal hyperplasia simulating diffe-

rent diseases such as squamous cell carcinoma (10), deep fungal infection or secondary syphilis; granulomatous (sarcoidal and elastolytic) simulating lupoid rosacea or granuloma annulare (11); there are even cases showing overall necrosis. In a series of 57 cases of LC, confirmed by PCR, published in 2012 by Saab et al. (7), other forms such as panniculitis (12), tuberculosis-like lesions, mycosis, anaplastic T-cell lymphoma, sarcoidosis, pityriasis lichenoides, indeterminate leprosy, subacute spongiotic dermatitis or lichen planus are described. However, we have not found any case of LC with an angiomatoid pattern in the reviewed publications.

The most frequently described clinical presentation of leishmaniasis in patients coinfecting with HIV is visceral. These patients present with 2 main types of cutaneous lesions: those specifically produced by *Leishmania*, usually papulo-nodular and simulating multiple dermatofibromas (13) or Kaposi sarcomas (14), and those in which the presence of the parasite is incidental such as Kaposi sarcomas, bacillary angiomatosis, herpes zoster and simplex. In this sense, cutaneous detection of *Leishmania* is frequent in HIV-infected individuals with visceral leishmaniasis. Sometimes *Leishmania* is associated with changes attributable to other dermatological processes, and its presence does not imply a causative role (15). It is also possible to isolate parasites from healthy skin of co-infected subjects. In the case described above, there was no evidence of visceral involvement due to the absence of systemic signs or symptoms. Although the histological background of LC was not characteristic of Kaposi's sarcoma, immunohistochemistry was negative for herpes virus type 8. In patients with combined HIV and LC, neither specific nor characteristic lesions were described, although there is a higher risk of atypical presentation, with mucocutaneous and visceral dissemination as well as numerous recurrences after treatment.

Arrais-Silva et al. (16) proved by immunohistochemistry the presence of type Ia hypoxia-induced factor in tissue infected by *L. amazonensis*. This is a protein with angiogenic properties, which is synthesised by tissue macrophages in hypoxic conditions. Although we have not been able to prove its presence in this study, it would be a causal hypothesis of the observed angiogenic pattern. In the Arrais-Silva study, the presence of type Ia hypoxia-induced factor was in tissue infected by *L. Amazonensis* and due to the most common species in Spain is *L. Infantum*, we cannot extrapolate these findings.

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¹Note added in proof: During the final processing of this paper two related papers have recently been published (17, 18).