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CASE REPORT

Atypical multifocal cutaneous leishmaniasis in an immunocompetent patient treated by liposomal amphotericin B

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Abstract Multifocal cutaneous leishmaniasis (MCL) is an extremely rare disease in South Europe, and it mainly affects immunosuppressed patients.

We report a case of MCL in an immunocompetent patient affected by type II diabetes mellitus that clinically presented with three large ulcers on the legs with a non-linear distribution and several months later with an erythematous-crusty lesion on the left cheek. Diagnosis of leishmaniasis due to *Leishmania infantum* was formulated by PCR analysis. Given the diffuse and wide lesions, the unresponsiveness to previous local and systemic treatments, a parenteral i.v. therapy with liposomal amphotericin B at a dosage of 3 mg/kg/day for 5 days was started and then repeated on the 14th and 21st days, leading to a clear improvement in the clinical picture.

The different clinical expression and the evolution of leishmaniasis depend on both the parasite subtype and the host's immunity status. *L. infantum* manifests with an atypical clinical feature more frequently than other species. The differential diagnosis for multiple ulcers must include several skin diseases, such as cutaneous TBC, bacterial ulcers, traumatic ulcers, deep mycoses, and sarcoidosis. However, an MCL should always be considered in subjects coming from endemic areas. In our case, the multifocality, the size of the lesions and the unresponsiveness to other treatment indicate a short course treatment with liposomal B amphotericin that proved to be a suitable alternative to traditional drugs used in MCL.

Introduction

Cutaneous leishmaniases (CL) of the old world are generally caused by leishmanias of the major and tropica complexes, but also by *Leishmania infantum*, which is responsible for all cases of CL in the northern Mediterranean littoral.[1]

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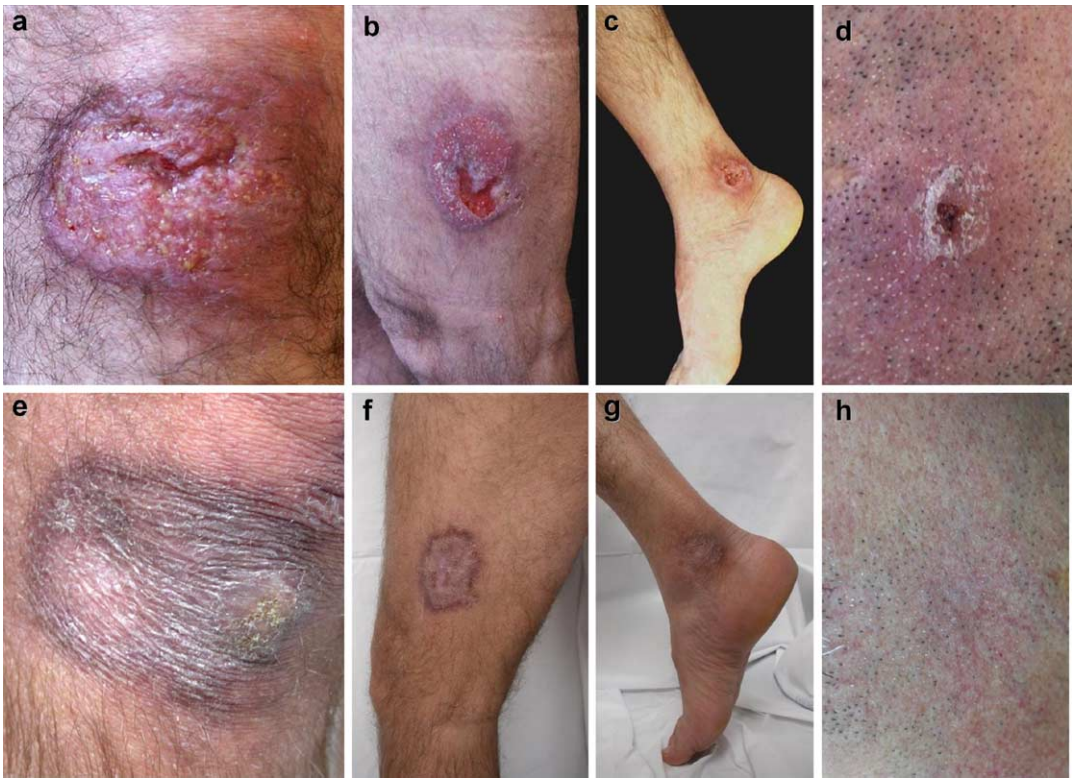

Figure 1 Top: cutaneous leishmaniasis ulcers with multifocal distribution: (a) right knee; (b) lateral left thigh; (c) right medial malleolus; (d) erythematous-scaly and crusty lesion on left cheek. Bottom: complete resolution after treatment with liposomal amphotericin B at 9 months (e)–(h).

Based on the relative tropism of *Leishmania* and the immune status of the host, Leishmaniases are classified into cutaneous, mucocutaneous and visceral. The cutaneous forms are generally divided into Old World CL, related to *Leishmania* of the major and donovani complexes, and New World CL, caused by the *L. mexicana* and *L. braziliensis* complexes.

In 90% of cases Old World CL cause a single cutaneous lesion, usually on uncovered areas, especially the face, whereas New World forms mainly cause multiple, widespread lesions that rarely heal spontaneously. Multifocal CL (MCL) due to old world *L. infantum* is rarely seen in southern Europe.[2]

We report a case of autochthonous CL with multifocal distribution probably due to incongruous steroid applications and successfully treated with liposomal amphotericin B.

Case report

A 56-year-old man from central Italy with a 3-year history of type II diabetes mellitus came to our attention for three large ulcers on his legs. He had noticed a painless papule on his left thigh about 8 months earlier; another appeared on the right knee after 3 months, and a third had developed on the right internal malleolus the previous month. The patient recalled an insect bite on the site of the first lesion. The lesions were treated with topical steroids and antibiotics for several weeks. When he came to our attention he had a wide (about $7\times 6\ cm^2$), painless ulcerative lesion on the right knee with pustulization and exudation growing on an erythematous plaque with raised and hardened borders (Fig. 1(a)); a similar lesion measuring $10\times 6\ cm^2$ on the lateral surface of the left thigh (Fig. 1(b)), and a smaller ulcer ($4\times 3\ cm^2$) with the same features on the internal surface of the right malleolus (Fig. 1(c)). The patient did not have associated lymphadenopathies. An erythematous scaly lesion clinically compatible with CL had developed on the left cheek 2 weeks previously (Fig. 1(d)).

Routine laboratory tests were normal except for glycaemia (147 mg/dl). HIV serology and the Mantoux test were negative. Lymphocyte count and typing were normal. Swabs for common germs and mycetes were negative. A smear followed by a May–Grünwald–Giemsa stain and a blood sample analysed for anti-*Leishmania* antibodies were both