Introduction

Leishmaniasis refers to a wide clinical spectrum diseases caused by a heterogeneous group of protozoans of the Leishmania genus, which consists of more than 2 dozen species. Infection is transmitted by the bite of sandfly vectors of the genera Phlebotomus and Lutzomyia. Different species of mammals, including rodents and even humans act as reservoirs of the disease. There are many species of Leishmania and the species present vary according to geographic area.
Leishmaniasis is an endemic disease in Spain, where it exists in cutaneous and visceral forms. The only species that causes leishmaniasis in Spain is *Leishmania infantum* though, due to the gradual increase in the immigrant population, it is possible to observe clinical cases of imported leishmaniasis that are caused by other species and have different clinical characteristics. We present a case of cutaneous leishmaniasis in a Senegalese patient, with clinical characteristics somewhat different from the forms of cutaneous leishmaniasis usually found in Spain. These characteristics may be attributable to the species *L. major*, which is responsible for leishmaniasis in Senegal.

**Case Description**

A 38-year-old Senegalese man who had been in Spain for only 20 days and who had no relevant medical history visited our department due to asymptomatic cutaneous lesions on the right wrist. The lesions had appeared a month and a half previously (when the patient was in Senegal) and had gradually grown and become ulcerated. The patient had tried different treatments with topical and systemic antibiotics, with no response.

A physical examination revealed 2 ulcerated nodules on the back and volar aspect of the right wrist. The nodules measured 5 cm along the major axis, with a necrotic, friable center and raised, infiltrated edges (Figures 1 and 2). The rest of the physical examination was normal and there were no enlarged lymph nodes.

The differential diagnosis included essentially bacterial, fungal, and mycobacterial infections, malignant tumors, and tropical ulcer. A biopsy was taken from the edges of the lesion using a 6-mm punch. The resulting cultures for bacteria, fungi, and mycobacteria were negative.
The biopsy result revealed a dense inflammatory infiltrate in the superficial and deep dermis and there were a large number of oval bodies measuring approximately 6 µm both inside and outside the macrophages (Figures 3 and 4). These bodies corresponded to the amastigotes—the intracellular form taken by Leishmania in the host, as opposed to the promastigote form taken in the mosquito vector.

Cutaneous leishmaniasis due to L. major was diagnosed with microbiological techniques, in line with clinical and epidemiological criteria. Because our patient had large, highly inflammatory, multiple lesions, we opted for systemic treatment with intramuscular meglumine antimonate, at a dosage of 20 mg/kg/d for 28 days. After 1 month of treatment, the lesions showed clear signs of regression in the form of somewhat depressed and unsightly hypopigmented scars.

Discussion

Leishmaniasis represents a considerable problem in public health and is endemic in 88 countries (16 developed countries and 72 developing countries). It affects 12 million people, with 2 million new cases each year. Furthermore, official figures underestimate the real situation because it is a highly underreported disease (reporting is obligatory in only 40 countries).1

There are many species of Leishmania in different geographic regions and they have traditionally been divided into “Old World” forms (Asia, Europe, and Africa) and “New World” forms (Americas). Clinical manifestations are varied but they can be grouped into three main syndromes: visceral leishmaniasis or kala azar, which is fatal and untreatable; mucocutaneous leishmaniasis or “espundia,” which produces erosive lesions of the nasal and oropharyngeal mucosa leading to severe disfigurement; and cutaneous leishmaniasis, which we will now focus on.

The characteristic form of cutaneous leishmaniasis in the Old World is the “oriental button” and it can be caused by the following species, according to geographic area: L. infantum, L. tropica, L. aethiopica, and L. major.2,3

In Spain, as in the other countries of southeastern Europe (such as France, Italy, Cyprus, and Malta), the only Leishmania species identified as causing cutaneous leishmaniasis is L. infantum, for which several zymodemes (strains identified by their different enzyme profiles) can be distinguished by means of enzyme analysis using electrophoresis.4 There are 2 species of Phlebotomus that have been identified as vectors: P. perniciosus and P. ariasi.2

Cutaneous leishmaniasis is endemic in Senegal and is caused by L. major (MON-26 zymodeme)5 (Figure 5). The
reservoir consists of several species of rodent (*Tatera, Aricanthus, Mastomys*, etc) and the main vector is *P. duboscqi.*

The clinical manifestation of cutaneous leishmaniasis is highly polymorphic and the type of lesion depends on 3 factors: parasite (pathogenicity, virulence, etc), vector (number of bites, type of saliva, etc), and host (genetic susceptibility and immune status). Lesions tend to affect uncovered areas of the body such as the face and limbs and are normally asymptomatic unless secondary infection occurs. It is not possible to distinguish the *Leishmania* species from the clinical aspect of the lesion but, in our atypical case, we wondered whether there were clinical signs characteristic of *L. major* that could differentiate it from the lesions produced by *L. infantum.* There are, in fact, clinical characteristics that serve as guides to each species.

In Spain, typical lesions due to *L. infantum* tend to begin with small, single or multiple erythematous papules, similar to a mosquito bite. They are asymptomatic or mildly pruritic with small, single or multiple erythematous papules, similar to a mosquito bite. Under the crust is an ulcer with an irregular base and raised edges. The lesion may remain for months or crust over. Transmission is usually takes place in and around urban areas and the dog is the main reservoir.

*L. major* on the other hand, produces the so-called rural ulcer, which is wet and zoonotic. The 2 most common clinical forms are the ulcerated and crusty forms. Lesions are often multiple, highly inflammatory and exudative and are often multiple, highly inflammatory and exudative and there are reported cases of sporotrichotic infections (multiple lesions with a lymphangitic spread), forms of diffuse leishmaniasis,11 and necrotizing and suppurating lymphadenitis.12 The reservoir consists mainly of rodents (*Tatera gambiana, Mastomys erythroloecus*, etc) and transmission usually takes place in rural areas.

Diagnosis is mainly by microscopic examination for parasites in exudate smears, skin biopsies, and fine-needle aspiration samples. Other techniques such as polymerase chain reaction are not normally used, except for research purposes.

Localized cutaneous leishmaniasis, especially the Old World varieties, do not always require treatment as they naturally tend to cure spontaneously and, when they are treated (because the lesions are large or multiple or because unsightly scarring is expected), intralesional treatment with pentavalent antimonial drugs is the therapy of choice. Meglumine antimonate is normally used at dosages varying from 1 to 2 mL per lesion, twice weekly, fortnightly, or monthly. Other alternatives include topical treatment with paromomycin, imiquimod, and cryosurgery for small lesions. Oral fluconazole at doses of 200 mg/d for 6 weeks has proven to be an effective and safe treatment in cases of cutaneous leishmaniasis due to *L. major* MON-26, as shown in the study carried out by Alrajhi et al.17 Species other than *L. major,* however, may not respond to this treatment. Systemic treatment with antimonial drugs is not usually necessary although some forms do require it, as in the case of our patient.

By reporting this case of imported leishmaniasis with a clinical manifestation that is rare in Spain, we hope that our admittedly limited experience will highlight the need for dermatologists to familiarize themselves with tropical skin diseases as the progressive increase in the immigrant population and in travel abroad means that cases of such diseases will become an important part of our day-to-day practice in the very near future.

Conflicts of Interest

The authors declare no conflicts of interest.

References


