

Sporotrichoid dissemination of cutaneous leishmaniasis possibly triggered by a diagnostic puncture

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Case report

A 31-year-old Spanish woman attended Hospital Clínic in Barcelona in August 2018, after working in La Paz (Bolivia) for six months. She remembered an insect bite on her right thigh, during a trip to Beni (Brazilian border) with progression to a papule with a central crater within one month. Treatment with oral amoxicillin/clavulanic acid yielded no improvement. Thereupon, a diagnostic puncture of the ulcer was performed, with a positive result of polymerase chain reaction (PCR) for *Leishmania* spp. without bacterial isolates in the culture. She then decided to return to Spain for management. She reported a 3-month history of a painless 3 cm diameter ulcer with friable granulation tissue and raised erythematous edges on the back side of her right thigh (figure 1), with multiple painful erythematous subcutaneous nodules on a linear path from the ulcer to the right groin (figure 2). Bilateral inguinal nodes were found, larger on the right side. Laboratory workup was unremarkable except for eosinophilic count of 0.5 x 10⁹/L. Serology for *Leishmania*, Syphilis and HIV were negative, along with Interferon-gamma release assay. Histopathologic examination of the ulcer revealed granulation tissue and lymphoplasmacytic infiltrate, while the nodule revealed an abscessed lesion with necrotizing granulomas. Giemsa stain and bacterial culture were negative. Molecular techniques for mycobacteria, 18S rRNA and 16S rRNA were negative in both samples. An in house kDNA qPCR for *Leishmania* (1) was inconclusive. Clinical diagnosis of cutaneous leishmaniasis (CL) was made and intralesional meglumine antimoniate 4ml/weekly was prescribed. Ultrasound and a Nuclear Magnetic Resonance suggested nodular lymphangitis and lymphadenitis of the right thigh, with suspicion of a sporotrichoid leishmaniasis. Resolution of the ulcer and reduction in size of all subcutaneous nodules was achieved after two months of treatment. Treatment was interrupted due to an accident that required admission in another centre. Six weeks later, a small erosion appeared on the edge of the previous ulcer (figure 3), with positive qPCR for *Leishmania* spp. from a biopsy. At present, the patient is improving under treatment with amphotericin B.

Discussion

Few diseases can produce a sporotrichoid dissemination of chronic ulcers; namely infections by *Sporothrix schenckii*, atypical mycobacteria, *Nocardia brasiliensis* and *Leishmania* spp., among others (2). Clinical and epidemiological features of this case prompted a diagnosis of CL, which was confirmed by qPCR. PCR assays showed a greatest sensitivity in contrast to classical diagnostic methods (3). There is not a consensus on the procedure to be used and the performance of the PCR assay could be influenced by the sample, the target and the PCR method used as well as by the presence of different *Leishmania* species, as in the American region (4). Bolivia is an endemic country for CL, being *L. braziliensis* the most common in the country, but also *L. amazonensis*, *L. guyanensis* and *L. lainsoni* are present (5). In South America, *L. braziliensis* has been identified causing sporotrichoid forms in 4.7% of CL, mainly in women and affecting upper limbs principally (6). It is usually described after receiving local treatment (7). Some authors suggest that tissue damage caused by local treatment triggers the spread of parasite into the subcutis and lymphatic vessels (7). Our patient had not received any local treatment before the lymphatic manifestation, but two months before our evaluation the ulcer had been punctured in order to obtain a sample. This puncture could have been the trigger for the sporotrichoid dissemination.

Author statements:

Author contribution:

PLR, IF, AA, IL, PG, DC, JM and NRV contributed to the patient care. MG and AFA contributed to the diagnosis techniques. All authors contributed to the writing of the manuscript and reviewed the literature.

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Figure Legends

Figure 1: Chronic ulcer caused by *Leishmania* spp.



Figure 2: Sporotrichoid dissemination of leishmaniasis



Figure 3: Small erosion on the edge of the previous ulcer

