Sporotrichoid Cutaneous Leishmaniasis

M López-Escobar, M Drake-Monfort, R Salesa-Gutiérrez de Rozas, and S Hermana-Ramírez

Servicio de Dermatología, Servicio de Microbiología, and Servicio de Anatomía Patológica, Hospital Universitario Marqués de Valdecilla, Santander, Cantabria, Spain

To the Editor:

The leishmaniases are parasitic diseases caused by intracellular protozoa of the Leishmania genus. These microorganisms are transmitted by the bite of the female of the fly belonging to the Phlebotomus genus in the Old World and Lutzomya in the New World. The clinical manifestations of the leishmaniases are extremely varied, and cutaneous, mucocutaneous, and visceral forms of the disease have been described.1,2 We present a case of cutaneous leishmaniasis with sporotrichoid spread, a rare variant of this entity that is unusual in Spain.

A 63-year-old woman of Ecuadorian origin and no history of interest consulted 3 weeks after her arrival in Spain for a lesion in the facial region that had been present for 2 months. The lesion showed progressive growth and was occasionally pruritic. The patient reported no other associated symptoms and had no recollection of insect bites or trauma prior to appearance of the lesion.

The physical examination showed an infiltrated plaque of about 3×3 cm in diameter, located on the left mandibular ramus. The surface was smooth, shiny, and erythematous, and presented a small ulceration covered by a crust. In addition, indurated lymph nodes and small subcutaneous nodules were palpable around the periphery of the lesion, along the path of the proximal lymph node chains (Figure 1). Complete blood count, blood biochemistry, protein analysis, chest x-ray, and bone marrow aspirate were normal. Exudate cultures were negative for bacteria, fungi, and mycobacteria. Giemsa stain of the exudate smear showed amastigotes inside and outside the histiocytes.

Histology revealed a dense inflammatory infiltrate composed of lymphocytes, plasma cells, and some eosinophils, along with macrophages infested with oval basophilic microorganisms with an eccentric kinetoplast and with no capsule (Figure 2).

References

Based on the clinical and cytology findings, a diagnosis of sporotrichoid cutaneous leishmaniasis was established. The patient was treated with meglumine antimoniate (Glucantime) at doses of 20 mg/kg/day of intramuscular pentavalent antimony for 28 days, with good tolerance and complete resolution of symptoms.

Cutaneous leishmaniasis usually manifests as wine-red papules or plaques, generally solitary, infiltrated, frequently ulcerated and covered by a bloody crust. These lesions are also known by the name “oriental sore.” Some rare clinical variants have been described, such as the sporotrichoid form, characterized by subcutaneous nodules and indurated nodes due to local and regional lymphatic spread of the disease. These nodules represent an immunological reaction against the direct lymphatic expansion of the parasite or its antigens. Likewise, there have also been reports of other unusual forms of cutaneous leishmaniasis such as eczematiform, hyperkeratotic, verrucous and papillomatous, zosteriform, erysipeloid, lupoid, phagedenic, and tumoral forms, or clinical manifestations such as macrocheilia or paronychia.

This particular presentation of sporotrichoid cutaneous leishmaniasis will require differential diagnosis with a number of conditions, including cutaneous tuberculosis, tuberculoid leprosy, infection with atypical mycobacteria, sporotrichosis, nocardiosis, actinomycosis, mycetomas, chancriform pyoderma, and tumors, particularly squamous cell carcinomas and lymphomas.

Sporotrichoid spread also has therapeutic implications, as systemic therapy must be started in these cases. As a first choice, pentavalent antimonials such as intramuscular or intravenous meglumine antimoniate (Glucantime) or sodium stibogluconate (Pentostam) are used. Liposomal amphotericin B, itraconazole, ketoconazole, and pentamidine have also been used as alternatives.

In summary, although sporotrichoid cutaneous leishmaniasis is not common in Spain, we feel it should be considered in order to ensure early diagnosis and rapid instigation of specific therapy.

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References