

CASE REPORT

Leishmania Braziliensis: Report of a Pediatric Imported Case With Response to Liposomal Amphotericin B

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Abstract. *Leishmania braziliensis* is the main etiologic agent of leishmaniasis in South America. A 9-year-old boy consulted for the presence of round, ulcerative lesions with raised borders that were painful and have appeared after a travel to Bolivia and Brazil. The culture for parasites showed leishmanias and the PCR was positive for *L. braziliensis*. The patient underwent treatment with itraconazol but due to the persistence of lesions he received liposomal amphotericin B with complete resolution of the lesions.

In all lesions by *L. braziliensis* the treatment must be systemic due to the risk of mucosal dissemination. Liposomal amphotericin B is a convenient alternative to pentavalent antimonials given its efficacy and good tolerance.

Key words: cutaneous leishmaniasis, *Leishmania braziliensis*, liposomal amphotericin B.

LEISHMANIA BRAZILIENSIS: DESCRIPCIÓN DE UN CASO PEDIÁTRICO IMPORTADO CON RESPUESTA A ANFOTERICINA B LIPOSOMAL

Resumen. *Leishmania braziliensis* es el principal agente etiológico de leishmaniasis cutánea en América del Sur. Un varón de 9 años consultó por lesiones redondeadas ulceradas de bordes sobreelevados, que eran dolorosas y habían aparecido después de un viaje por Bolivia y Brasil. En el cultivo de parásitos se objetivaron Leishmanias y la reacción en cadena de la polimerasa (PCR) para *L. braziliensis* fue positiva. El paciente realizó tratamiento con itraconazol, y por persistencia de las lesiones inició anfotericina B liposomal con resolución completa de las mismas.

En toda lesión por *L. braziliensis* el tratamiento debe ser sistémico por el riesgo de disseminación mucosa. La anfotericina B liposomal es una buena alternativa a los antimoniales pentavalentes por su eficacia y buena tolerancia.

Palabras clave: leishmaniasis cutánea, *Leishmania braziliensis*, anfotericina B liposomal.

Introduction

Leishmaniasis refers to a group of diseases caused by protozoan parasites of the genus *Leishmania* and is transmitted to humans by the bite of sandflies of the genera *Phlebotomi* and *Lutzomya*. Local cutaneous leishmaniasis is one of the most frequent skin diseases among tourists in tropical areas, and *Leishmania braziliensis* is its main etiologic agent in South America.^{1,2}

Case Description

We report the case of a 9-year-old boy with painful lesions of 6 weeks evolution with no pertinent medical history. One month before the appearance of the lesions, the patient had been on holiday to Bolivia and Peru, visiting the Amazon jungle and Machu Picchu. Physical exploration showed 2 round erythematous lesions, 2-cm diameter, highly infiltrated, with raised edges and an ulcerated exudative center. One was on the back of the second finger of the right hand (Figure 1), and the other on the submandibular area, where there was also a freely moveable swollen lymph node, 2-cm diameter.

Complete blood count, biochemical profile, and coagulation tests were normal. Mycobacterial culture was negative. Leishmaniasis was confirmed by a smear taken from the lesion and a range of *Leishmania* species were present in the parasite culture. The culture medium was

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Manuscript accepted for publication June 26, 2006.

NNN prepared with rabbit blood. Polymerase chain reaction analysis was performed at the Instituto Carlos III (Majadahonda, Madrid, Spain) and proved positive for *L. braziliensis*.

The patient was treated with oral itraconazole for 1 month, with partial initial improvement. Due to persistence of the lesions, other treatments were proposed: systemic antimonials were rejected because of their frequent severe side effects and treatment was begun with intravenous liposomal amphotericin B as it is tolerated better. Treatment continued for 9 days, with complete resolution of the lesions after 1 month (Figure 2).

Discussion

In the Old World, cutaneous leishmaniasis is mainly caused by *L. major*, *L. tropica* or *L. infantum*, whereas in the New World it is caused by *L. braziliensis* or *L. mexicana*. It is important to identify which species is causing the lesion, since *L. braziliensis* can cause mucocutaneous leishmaniasis in up to 3% of the patients who do not receive treatment, whereas *L. mexicana* does not adversely affect the mucous membrane.³ Thus, systemic treatment is recommended for infections by *L. braziliensis*.

In the pediatric population, localized cutaneous leishmaniasis is the most common clinical form (99.13%), followed by mucosal (0.34%), chronic cutaneous (0.27%), and diffuse (0.25%) forms.⁴ The localized cutaneous form becomes manifest as an erythematous papule that develops into an ulcerated node, located in areas exposed to bites. Lesions in patients infected in the New World tend to be isolated (compared to multiple lesions in those infected in the Old World), and in children are often accompanied by swollen lymph nodes, as in the case presented. When the etiologic agent is *L. braziliensis*, lymph nodes are frequently involved (up to 42% in a series of 11 patients), and this involvement can at times precede the cutaneous lesions.

Differential diagnosis should include pyodermitis, mycobacterial infections, epithelioma, sporotrichosis, chromomycosis, etc.

Diagnosis is traditionally performed via smear, culture, or biopsy and is confirmed once the parasite is located. These tests have a sensitivity of around 87%. Polymer chain reaction analysis is also currently available and has a sensitivity of 97%.^{5,6}

All lesions due to *L. braziliensis* should be treated systemically because of the risk of mucosal invasion. Intravenous or intramuscular pentavalent antimonial agents have traditionally been used and have achieved cures in 90% of cases,^{7,8} but side effects are frequent and severe (electrocardiographic disorders, serious arrhythmias, pancreatitis, myelosuppression, myalgia, and arthralgia). In resistant cases, pentamidine or amphotericin is used as



Figure 1. Ulcer with raised edges on the dorsal aspect of the finger.



Figure 2. Healed lesion after treatment with liposomal amphotericin B.

second-line treatment. Liposomal amphotericin B is a macrolide active against fungi and *Leishmania* species that is useful in the treatment of mucocutaneous forms resistant to pentavalent antimonials and in visceral leishmaniasis. Although expensive, treatment is shorter due to its effectiveness and hospital stays are shorter than with pentavalent antimonial agents. It is tolerated better, with fewer serious side effects, and is thus beginning to be used in the treatment of the localized cutaneous forms caused by *L. braziliensis*.^{9,10}

Oral treatment has recently been implemented to avoid hospitalizing the patient. Ketoconazole has an 89% cure rate against *L. mexicana* and 30% against *L. braziliensis*. Another option is fluconazole at 200 mg/day for 6 weeks, with a 79% cure rate against *L. major*.¹¹ Miltefosine, a phospholipid analogue of alkylphosphocholine, seems more promising with a 94% cure rate against leishmaniasis in the New World.^{12,13}

In conclusion, we have presented a case of localized cutaneous leishmaniasis caused by *L. braziliensis* that had excellent response to treatment with liposomal amphotericin B and was well tolerated. Liposomal amphotericin B is a

good alternative to pentavalent antimonial agents because it is effective and well-tolerated, although it is more expensive than traditional treatments.

Conflicts of Interest

The authors declare no conflicts of interest.

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