

PRESENTACIÓN DE CASO

Zosteriform cutaneous leishmaniasis due to *Leishmania (Viannia) panamensis* and *Leishmania (Viannia) braziliensis*: Report of three cases

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In Colombia, zosteriform leishmaniasis is a little-known and infrequent clinical variant of cutaneous leishmaniasis. Its clinical features include one or more plaques made up of papules and pseudo-vesicles, which conform to a lineal pattern, as well as satellite lesions that affect one or more dermatomes, without crossing the median line.

We present three zosteriform cutaneous leishmaniasis cases in which *Leishmania panamensis* and *Leishmania braziliensis* were identified as the infective species. In light of the fact that the disease occurs infrequently, diagnosis was reached by taking into account epidemiological and clinical suspicion.

Key words: *Leishmania*, *Leishmania braziliensis*; leishmaniasis, cutaneous; herpes zoster, diagnosis, epidemiology.

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Leishmaniasis cutánea zosteriforme causada por *Leishmania (Viannia) panamensis* y *Leishmania (Viannia) braziliensis*: reporte de tres casos

La leishmaniasis zosteriforme es una variante clínica de la leishmaniasis cutánea, infrecuente y poco conocida en Colombia. Clínicamente se caracteriza por una o varias placas conformadas por pápulas y pseudovesículas que siguen un patrón lineal, y por lesiones satelitales que comprometen uno o varios dermatomas sin sobrepasar la línea media.

Se presentan tres casos de leishmaniasis cutánea zosteriforme en los que se identificaron *Leishmania panamensis* y *Leishmania braziliensis* como especies infectantes. La sospecha epidemiológica derivada de la procedencia de los pacientes, así como la sospecha clínica a partir del reconocimiento de una presentación infrecuente de la enfermedad, permitieron hacer el diagnóstico.

Palabras clave: *Leishmania*, *Leishmania braziliensis*, leishmaniasis cutánea, herpes zóster, diagnóstico, epidemiología.

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Leishmaniasis is an endemic disease in Colombia. A striking number of new cases has been reported in the past decade: up from 4,130 cases in the

year 2001 to 9,684 cases in the year 2011. It accounts for 20% of all leishmaniasis cases registered in the Americas over the period 2001-2011, and places the country second in incidence, surpassed only by Brazil (1). In Colombia, 98% of all cases are classified as cutaneous leishmaniasis (2), generally presenting as a papule or nodule on an exposed area that grows progressively and turns into a painless ulcer with infiltrated edges (3,4). Other clinical manifestations designated

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Camilo Andrés Morales and Juliana Palacio participated in providing the clinical information of these cases and wrote the first draft. Gerzaín Rodríguez performed the histological evaluation. Yenny Carolina Camargo performed the microbiological studies. All authors participated in the design, literature search, writing, review and approval of the final version of this manuscript.

as “atypical” because they are infrequent and little known may also occur. These are given different names according to the lesion’s clinical characteristics (annular, chancriform, cicatricial, keloid, tumoral, acneiform, panniculitis, verrucous), its similarity to other diseases (lupoid, acneiform, eczematous, erysipeloid, sporotrichoid, psoriasiform, rhinophymatous, mycetomatous, zosteriform) and the anatomical localization (palmoplantar, genital, conjunctival, whitlow, paronichial) (4-9).

Among the atypical cutaneous leishmaniasis manifestations, zosteriform is one of the least frequent, accounting for a scant number of reported cases in the Americas (10). Its global prevalence varies between 0.17% and 2.5%, according to some case series from the Middle East (6,11). It is clinically discernible by one or more plaques composed of papules or pseudo-vesicles, located on the head (10,12), back (8,9), abdomen (13) or lower limbs (7), forming a lineal pattern and satellite lesions that involve one or more dermatomes that do not cross the median line (6,8,12). The appearance and distribution of lesions can cause the disease to be confused with herpes zoster, thus complicating initial diagnosis. A biopsy may be ordered which, in turn, may make histological pattern identification difficult since it differs from that observed in typical lesions of cutaneous leishmaniasis.

In the three cases described below cutaneous leishmaniasis diagnosis was confirmed using either microbiological or direct methods. In two cases, the etiological agent was identified using monoclonal antibodies, whereas in the case with biopsy sample (case 2), the species was identified using molecular methods.

Clinical cases

Case 1. A 21-year old male farmer, born in and resident of Falan (Department of Tolima), presented 3 month-old slightly itchy lesions on the abdomen that had spread progressively, and for which he had received several cycles of oral antibiotics with no signs of improvement. Physical examination revealed on the abdomen numerous small infiltrated confluent papules with shiny surface, that formed five erythematous plaques distributed over dermatomes T8, T9 and T10 (figure 1). Direct smear exam and lesion aspirate culture tested positive for leishmaniasis. *Leishmania panamensis* was cultured and identified by means of species specific B4 and B11 monoclonal antibodies. This species is compatible with the reference strain MHOM/PA/71/LS94. Treatment was begun with

meglumine antimoniate (Glucantime®) at a dosage of 20 mg/Kg per weight/day for 20 days, with subsequent lesion involution and healing.

Case 2. A 30-year old baker, born in and resident of Bogotá, presented asymptomatic lesions on the dorso-lumbar region with 2-month evolution, for which he had received various topical and oral acyclovir treatments with no signs of improvement. Physical examination revealed a violet erythematous plaque on the right lumbar region over dermatomes L2, L3 and L4 that was composed of pseudo-vesicles and yellowish erythematous papules. Some of these were shiny and infiltrated, with slight scaling and a small central scab (figure 2). Under the diagnostic impression of sarcoidosis, a skin biopsy was performed. Histologically, the epidermis showed discrete acanthosis with dense histiocytic-lymphocytic infiltrates throughout the dermis, suggesting the presence of a live agent



Figure 1. Shiny, confluent papules forming brown erythematous plaques with lineal distribution on abdomen



Figure 2. Violet erythematous plaque with numerous shiny and pseudovesicle papules on the lumbar region

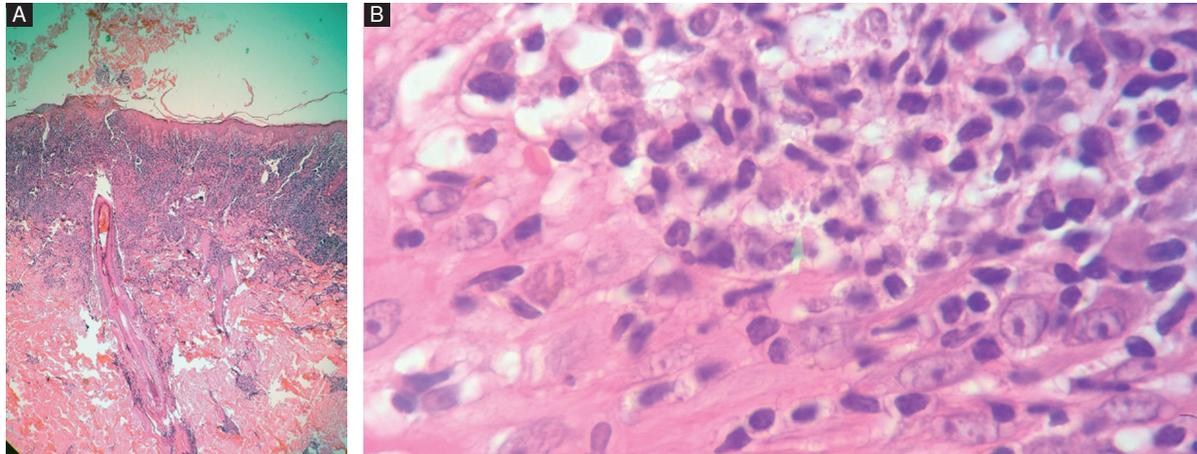


Figure 3. A. Skin biopsy without epidermal hyperplasia and with dense histiocytic-lymphocytic superficial dermal infiltrates. Hematoxiline-eosin (HE), 10X. **B.** Presence of amastigotes. Hematoxiline-eosin (HE), 100X

(figure 3A). The patient was interviewed once again and he mentioned that the lesions had appeared 2 weeks after visiting Otanche (Department of Boyacá). Direct smear exam and lesion aspirate culture were ordered, the results of which resulted positive for leishmaniasis. In a later review of histological slides, numerous amastigotes were observed (figure 3B), and by means of molecular methods *L. braziliensis* was identified as the etiologic agent. Montenegro skin test results were also positive. Treatment began with meglumine antimoniate (Glucantime®) at a dosage of 20 mg/Kg per weight/day for 20 days, with subsequent lesion involution and healing.

Case 3. A 27-year old male miner, born in and resident of Pacho (Department of Cundinamarca) underwent physical examination with a 1-month history of asymptomatic lesions on his back. Examination of the right infra-scapular region revealed a brown erythematous plaque, over dermatomes T5 and T6 made up of papules and pseudo-vesicles, with a lineal distribution on its upper border (figure 4). Direct smear exam and the culture of the lesion aspirate tested positive for leishmaniasis. *Leishmania panamensis* was identified by means of species specific B4 and B11 monoclonal antibodies. This species is compatible with the reference strain MHOM/PA/71/LS94. The patient was treated with meglumine antimoniate (Glucantime®) at a dosage of 20 mg/Kg per weight/day for 20 days, with subsequent lesion involution and healing.

Discussion

The clinical manifestations of cutaneous, mucosal or mucocutaneous and visceral leishmaniasis, depend



Figure 4. Brown erythematous plaque with papules and pseudovesicles with lineal distribution (upper edge) on the right infra-scapular region

upon the host's immunological response and the virulence of the infecting strain. Therefore, the spectrum of the disease varies from asymptomatic cutaneous infection to visceral involvement, which is potentially mortal (14). In the Americas, 95.7% of the cases for which clinical information is available are classified as cutaneous leishmaniasis (1). In Colombia, 98% of leishmaniasis cases are cutaneous (2), and lesions are generally caused by species belonging to the subgenus *Viannia*: *L. panamensis*; *L. braziliensis* and *L. guyanensis* (15,16).

At the National Institute of Dermatology of Colombia, a referral dermatology center receiving patients from almost all regions of the country, most cutaneous leishmaniasis cases (92%), including those with atypical symptoms, are caused by *L.*

panamensis (74.5%) (16). This species had been reported previously as the etiological agent of diffuse cutaneous leishmaniasis in an AIDS patient (16), and in three patients with disseminated, erysipeloid and leishmaniasis recidivans cutis (4). This contrasts with numerous reports on atypical cases from the Mediterranean basin (12) and from other countries, including Pakistan (5-7,11,13,17), India (18), Iran (8,9) and Lebanon (19), where the major infective species is *L. major*. In these publications, the clinical polymorphism of the disease has been related to parasite dependent factors, such as virulence of the species (5,6,8,9,18), the vector (12), and geographical region where infection took place (11,12,18), and to the host, such as lesion localization (5,8) and immune response (5-8,11,12,18).

In the three cases we present, lesions were localized on the trunk, on covered areas (table 1), which could suggest a relation between infection site and atypical clinical presentation due to anatomical and physiological variations related to aspects such as local immune response, skin temperature and microcirculation of the dermis where the parasite develops (20). The influence of anatomical site on the host's clinical and immunological response has been studied in experimental animal models with *L. panamensis*, in which clear differences have been established for time of evolution, quantity of inflammatory infiltrate and tissue necrosis, parasite load, antibody production and treatment response, depending on the site where inoculation took place (20).

It has also been suggested that in other atypical forms, such as erysipeloid leishmaniasis, parasite dissemination in the papillary dermis hinders the

ability of the immune system to control the disease (12,21). Since the three patients in this series had numerous amastigotes, it is possible that the host's deficient immune response, when faced with certain *Leishmania* species (*L. panamensis* and *L. braziliensis* in the New World and *L. major* in the Old World), allows for parasite replication and dissemination with a clinical pattern different from the typical one.

The difficulty of orienting clinical diagnosis in patients with atypical forms of cutaneous leishmaniasis usually calls for a biopsy, which can also be confusing for the pathologist when it means dealing with non-ulcerated lesions due to the lack of epidermal hyperplasia. However, the presence of a diffuse dermal infiltrate rich in plasmocytes and vacuolated macrophages allows for diagnostic orientation and the careful search for the parasite in lesion biopsies of less than 2-to-3-month evolution. In these, it is possible to observe the amastigotes in more than 70% of cases (22). Another scenario where epidermal hyperplasia is seen is in the diffuse form of the disease where the epidermis can be atrophic but the dermal infiltrate has abundant vacuolated macrophages that phagocyte the large quantities of amastigotes present (23).

In addition to herpes zoster, the differential diagnosis in zosteriform leishmaniasis includes other infectious skin diseases such as lupus vulgar and late secondary syphilis; inflammatory ones such as tumid lupus erythematosus, granuloma annulare and cutaneous sarcoidosis; and tumoral ones such as B-cell cutaneous lymphoma, among other diseases. In one of the three cases (figure 2), lesions were confused with another granulomatous disease, and diagnosis was deferred until a history

Table 1. Parasitological characteristics and epidemiological and clinical features of the three patients with zosteriform leishmaniasis

	Case 1	Case 2	Case 3
Gender	Male	Male	Male
Age	21	30	27
Place of origin	Falan, Tolima	Otanche, Boyacá	Pacho, Cundinamarca
Altitude	990 m/ mamsl	1050 m/AMSL	2136 m/AMSL
Average temperature	27° C	23° C	19° C
Lesion localization	Abdomen	Lumbar region	Dorsal region
Lesion type	Plaques and papules	Plaques and papules	Plaques and papules
Evolution	3 months	2 months	1 month
Montenegro skin test	5 x 5 mm (+)	10 x 9 mm (+)	7 x 7 mm (+)
Direct test	Positive (1-5 amastigotes per microscopic field)	Positive (1-5 amastigotes per microscopic field)	Positive (1-5 amastigotes per microscopic field)
Culture test	Positive	Positive	Positive
Infective species ¹	<i>L. panamensis</i>	<i>L. braziliensis</i>	<i>L. panamensis</i>

mamsl: meters above mean sea level

¹ Identification by means of monoclonal antibodies in cases 1 and 3, and by PCR in case 2

of visiting an endemic area of leishmaniasis was known (24). In the other cases, the epidemiological criteria and lesion recognition by an expert dermatologist allowed for disease suspicion, which was later confirmed by microbiological tests that also provided identification of the infective species (table 1).

In conclusion, zosteriform leishmaniasis in Colombia is an infrequent and little known variant of cutaneous leishmaniasis, which, like other atypical manifestations, could be related to the infective species and to the host's immune response, among other factors, and can be confused with different dermatological diseases.

Conflicts of interest

The authors declare no existing conflict of interest.

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