



American Tegumentary Leishmaniasis of The Penis: An Atypical Location

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Abstract

Leishmaniasis comprise a heterogeneous and extensive group of infectious and non-contagious diseases caused by protozoa of the genus *Leishmania* spp. To date, more than 20 species have been identified that can cause visceral or tegumentary impairment in humans. Considered endemic in 92 countries, with around one million new cases of tegumentary forms annually. They have a strict relationship with poverty, environmental changes, population displacements resulting from catastrophes or wars and a lack of public health policies to adequately control them. The World Health Organization considers that more than one billion people are at risk of becoming infected because they live in endemic regions. Classically, the lesions appear in exposed areas of the tegument that have been bitten by the winged vector. In the present report we present a case of a patient with a skin lesion clinically compatible with cutaneous leishmaniasis, in an atypical and rare location on the penis, resulting from the patient's work habit as he performed his physiological needs in the forest area, the place where he was probably bitten by the winged agent. The diagnosis was confirmed by histological examination of the lesion, aspiration from the edge with identification of the agent, and the patient was treated with liposomal amphotericin with complete regression of the lesion.

Keywords: American cutaneous leishmaniasis; Liposomal amphotericin B; Atypical location; Leishmaniasis in the penis; Differential diagnosis; Treatment.

INTRODUCTION

The initial records of American cutaneous leishmaniasis are found in ceramics from the Mochica culture in Peru between 300 and 700 AD, which show several changes found in mucous forms, such as mutilations and deformities in the nose and lips. Tegumentary Leishmaniasis is a disease strongly linked to environmental and ecological factors, with a wild cycle whose vector-host balance remains in balance as a zoonosis of wild animals. Human beings can become infected when they start visiting wooded or forested regions to carry out work activities where there is a need for deforestation (agriculture, livestock) or leisure excursions. The peri-urban or rural cycle occurs in endemic regions where humans begin to inhabit deforested areas, with the disease also affecting women, children and the elderly [1].

In the Americas, eleven species of *Leishmania* spp have already been isolated and identified as agents of this disease. In Brazil, *Leishmania (Viannia) braziliensis* predominates and is present throughout the national territory.

Leishmaniasis are a clinically heterogeneous group of diseases caused by protozoa of the genus *Leishmania*. There is growing evidence that the true incidence of the disease is underestimated, especially in hyperendemic regions. Moreover, climate changes together with the increasing movement of humans and animals raise concerns about the possible introduction of *Leishmania* infection in previously spared areas [2].

It is caused by numerous *Leishmania* protozoa species, which are responsible for its clinical diversity.

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The wide variety of *Leishmania* species responsible for human American cutaneous leishmaniasis combined with the immune mechanisms of the host results in a large spectrum of clinical, histopathological, and immunopathological manifestations. At the middle of this spectrum are the most frequent cases of localized cutaneous leishmaniasis, caused by members of the subgenera *Leishmania* and *Viannia*, which respond well to conventional therapy. At the middle of the clinical spectrum, American cutaneous leishmaniasis with one or multiple ulcerated skin lesions represents the most frequent form of the disease, having as the etiologic agent any member of the neotropical subgenera *Viannia* and *Leishmania*. *L. (V.) braziliensis* [3].

Atypical clinical forms of cutaneous leishmaniasis are uncommon, particularly that classified as "sporotrichoid form", characterized by the presence of lymphangitis, generally accompanied by adenopathy and the presence of ulcers along the path, being described in leishmaniasis caused by *L. guyanensis* and *L. braziliensis* [4,5]. The presence of cutaneous leishmaniasis lesions in unconventional locations is uncommon and requires diagnostic accuracy to distinguish them from other pathologies [6-9].

We report below a case of American cutaneous leishmaniasis with an ulcerated lesion on the penis, an unusual location, resulting from the patient's physiological habits of exposing himself in a forest region during his work activity in the fields of a farm where he carried out deforestation work for agriculture.

CASE PRESENTATION

32-year-old patient, worker in a rural area, where his work activity consists of promoting deforestation of farm areas to plant soybeans. He states that he lives in a house on the farm and that he leaves his home for work at dawn and only returns home at the end of the day. It performs its feeding and physiological needs in the forest area. He reports that approximately 1 month and a half ago, a painless lesion appeared on the penis, with slight serous secretion. When seeking medical attention, he was diagnosed as having syphilis and was treated with intramuscular benzathine penicillin, in a total dose of 2,400 million units, reporting that there was no change in the lesion after this medication. On his own, he started applying antibiotic cream with neomycin, also without results. He presented to our service presenting an ulcerated lesion, approximately 1 cm in diameter, oval-shaped, lymphatic base, slightly erythematous and granular, infiltrated edges, elevated and painless on palpation, with a general "picture frame" appearance (Figure 1),



accompanied by discrete and painless inguinal adenopathy. As a result of the clinical and epidemiological history, the diagnostic hypothesis of cutaneous leishmaniasis was established. Histopathological examination was performed, which revealed an epidermis with hyperparakeratosis, hypogranulosis, and irregular acanthosis. In the dermis, there is a lymphohistiocytic infiltrate with numerous superficial and deep plasma cells with outlines of granuloma, presenting rounded bodies compatible with leishmania (Figure 2). The specific histochemical examination on this material was positive for leishmania, as well as the PCR exam which confirmed the species *braziliensis*. Simultaneously with the biopsy, material was collected from the edge of the ulcer and an imprint of the material was made with Giemsa stain, and material was also collected from the edge of the ulcer by fine needle aspiration, both material collections revealed the presence of amastigote forms in the collected material. Simultaneously, culture was carried out in NNN medium of the material obtained by aspirating material from the edge of the lesion. The IDR Montenegro reaction test showed a positive result of 9 mm. Biochemical tests and serology for syphilis, HIV and viral hepatitis were normal or negative.

Treatment was started with liposomal Amphotericin B, at a dose of 3mg/kg/day intravenously, having received a total dose of 2250 mg. The patient progressed without clinical or laboratory complications, having adequately tolerated the infusion, progressing with significant improvement in the lesion, with a significant reduction in erythema and infiltration, and disappearance of the inguinal lymph node infarction. He returns after 60 days for outpatient follow-up, presenting fully healed as ulcerated lesion. Remains under monthly outpatient follow-up.



Figure 1: Ulcerated lesion of infiltrated "frame" edges on the penile

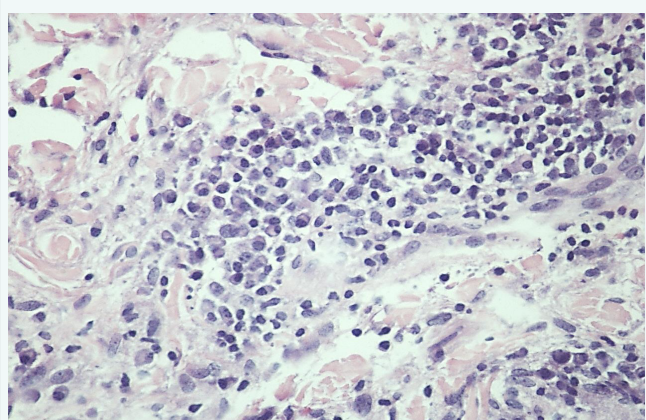


Figure 2: In the dermis, there is a lymphohistiocytic infiltrate with numerous superficial and deep plasma cells with outlines of granuloma, presenting rounded bodies compatible with leishmania.

DISCUSSION

The leishmaniasis afflict the world's poorest populations. Among the two million new cases each year in the 88 countries where the disease is endemic. Human infections with *Leishmania* protozoan parasites, transmitted via the bite of a sandfly, cause visceral, cutaneous, or mucocutaneous leishmaniasis.

Depending on the infecting species, an infection with *Leishmania* parasites can give rise to three clinical manifestations. The first is localized cutaneous leishmaniasis, with single to multiple skin ulcers, satellite lesions, or nodular lymphangitis. The second is leishmaniasis with mucosal involvement and the third is systemic visceral leishmaniasis with involvement of internal organs, such as the liver, spleen, and bone marrow, which is lethal if not appropriately treated [10].

The diagnosis of cutaneous leishmaniasis is based on clinical features (supported by epidemiologic data) and laboratory testing. Numerous diagnostic methods have been described with a huge variation in diagnostic accuracy, including direct parasitologic examination (microscopy, histopathology, and parasite culture) and/or indirect testing with serology and molecular diagnostics [11].

The selection of the diagnostic test employed often depends on the available infrastructure and resources of the diagnostic facility and not on diagnostic accuracy [12]. Parasitologic diagnosis is still considered the gold standard in leishmaniasis diagnosis because of its high specificity. This is typically undertaken by histopathologic examination of fixed tissue or parasite in vitro culture from material from suspected lesions. Microscopical diagnosis of cutaneous leishmaniasis is performed by the direct identification of amastigotes in Giemsa-stained lesion smears of biopsies, scrapings, or impression smears. Amastigotes appear as round or oval bodies, about 2–4 μm in diameter, with characteristic nuclei and kinetoplasts. The material from the ulcer margin usually has the highest yield. A comparative study between widely used scraping smears and fine needle aspiration cytology found a significant difference between the two methods in favor of fine needle aspiration in the detection of amastigotes and microgranuloma, slide background, and patient comfort [13].

Parasite culture in tubes containing Novy-MacNeal-Nicolle medium from suspected lesions is difficult, requires significant technical expertise, is prone to contamination, and is time consuming [14].

In the present work we report the occurrence of an unusual clinical location leishmaniasis on the penis. The therapeutic regimen was carried out with liposomal amphotericin at a dose of 3mg/kg/day intravenously, having received a total dose of 2250 mg [15]. The uncommon location of this lesion makes it necessary to carry out a differential diagnosis with other pathologies such as primary syphilis, chancroid, squamous cell carcinoma, paracoccidioidomycosis and traumatic lesions. Faced with a clinical picture that inspires different differential diagnoses, the doctor must always keep in mind the different diagnostic possibilities, avoiding the use of medications incorrectly, as was done in this patient [16].

CONCLUSION

Cutaneous leishmaniasis is classified as one of the most important neglected diseases. Delay in diagnosis can lead to complications and sequelae in these patients when mucosal involvement eventually occurs. Atypical forms or unusual locations must be remembered, at the risk of not making an early diagnosis, or possibly subjecting the patient to harmless and ineffective therapeutic measures resulting from an erroneous diagnosis.



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