



# American tegumentary leishmaniasis: an uncommon clinical and histopathological presentation\*

Leishmaniose tegumentar americana: uma apresentação clínica e histopatológica incomum

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**Abstract:** We report a case of an unusual presentation of American Tegumentary Leishmaniasis involving a male patient with a solitary lesion on the ear lobe, persisting with minimal increase for at least six months without ulceration or related symptoms. The histological sections showed epithelial atrophy and a large number of structures consistent with *Leishmania* sp. amastigotes within macrophages. Treatment commenced with meglumine antimoniate resulting in regression of the condition. This report is of importance given the unusual clinical manifestation and histopathological findings in this case and the fact that there was low correlation with the extended duration of the disease.

**Keywords:** Leishmania; Leishmaniasis, cutaneous; Zoonoses

**Resumo:** Relatamos um caso de apresentação incomum de leishmaniose tegumentar americana com lesão solitária no lóbulo da orelha de um paciente do sexo masculino com mínimo aumento em, pelo menos, seis meses, sem ulceração ou sintomas relacionados. Os cortes histológicos mostraram atrofia epitelial e um grande número de estruturas compatíveis com amastigotas de *Leishmania* sp. no interior de macrófagos. O tratamento foi iniciado com antimoniato de meglumina, com regressão do quadro. A importância desse relato de caso é devido à apresentação clínica e histopatológica atípica e sua baixa correlação com a duração prolongada da doença.

**Palavras-chave:** Leishmania; Leishmaniose cutânea; Zoonoses

## INTRODUCTION

American Tegumentary Leishmaniasis (ATL) is an infectious zoonotic disease caused by the protozoan parasite *Leishmania* spp, transmitted to man and other mammals through the bites of *Phlebotomus* sandflies. ATL in the New World is widespread from the south of the United States to the north of Argentina.<sup>1</sup>

In Brazil ATL is found in every state and there has been a high incidence of the disease over the past 20 years, with 26,057 new cases on average per annum.<sup>1</sup> 6% of the cases of ATL in the country are found in the state of Ceará (northeast Brazil), mainly

on the plateaus.<sup>2</sup> We report a case of an uncommon manifestation of ATL.

## CASE REPORT

In September 2010, a 42-year-old male patient from a rural area in Ceará noticed the onset of a nodule on his ear. Over the ensuing months, there was a minimal swelling of the lesion. Treatment with topical steroids and antibiotics produced no improvement.

Physical examination revealed an erythematous nodule of fibrous-elastic tissue on the right ear lobe with no purulent discharge. The patient, with no

Received on 14.01.2012.

Approved by the Advisory Board and accepted for publication on 27.03.2012.

\* Study conducted at the Dermatology Department of the Dona Libânia Dermatology Center (CDERM) - Fortaleza (CE), Brazil.

Conflict of interest: None

Financial funding: None

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history of local trauma, did not experience localized pain, itching or constitutional symptoms (Figure 1).

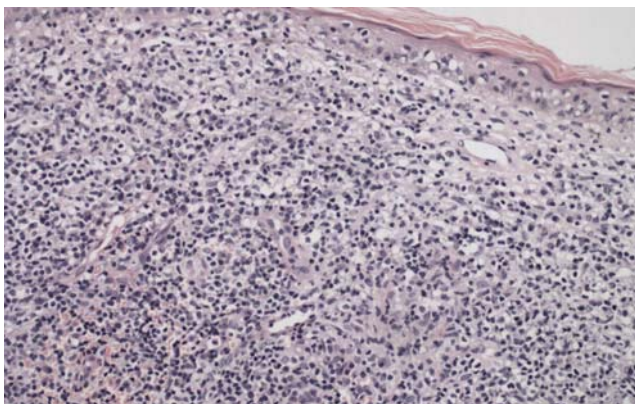
The patient's blood glucose was normal and the VDRL test was nonreactive. Serologic tests for HIV were not undertaken. A biopsy was carried out about six months after the onset of the clinical condition.

The histological sections showed epithelial atrophy, and Grenz's zone in the dermis, with a dense superficial and deep lymphoplasmacytic and histiocytic infiltrate. A large number of structures consistent with *Leishmania* sp. amastigotes was observed in macrophages. The results of the Leishmanin skin test (LST) were positive, with 10 mm of induration (Figure 2 and 3).

The patient missed follow-up but returned three months later with no changes on the lesion. Since no abnormalities were found in the general laboratory tests, treatment was started with meglumine antimoniate (15mg/kg) over 20 days. Two months after the end of therapy the lesions showed clear signs of regression (Figure 4).



**FIGURE 1:** Aspect of the lesion pre-treatment: a nodule of fibrous-elastic tissue on the right ear lobe

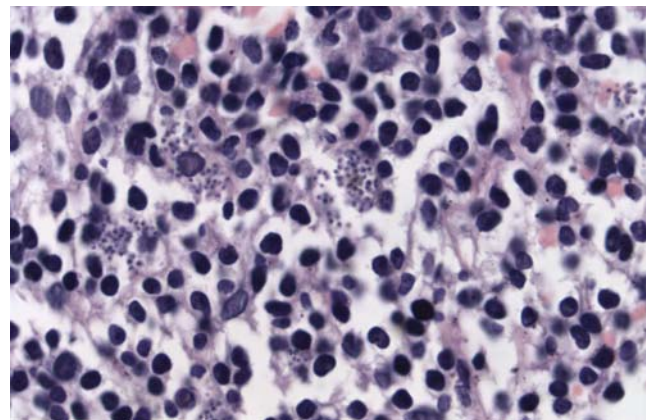


**FIGURE 2:** Epidermal atrophy and infiltrate in the dermis composed of lymphocytes, plasmocytes and histiocytes. There are no granulomatous formations in the sample (H&E 400x)

## DISCUSSION

ATL is characterized by a spectrum of cutaneous manifestations. The classic initial clinical sign of the disease is the appearance of an erythematous nodule, which may be single or multiple, usually located in an exposed region of the tegument where, after a few months, it develops into ulcers with indurated raised outer borders, regular contours and a cross-grained background with or without a seropurulent exudate. The most common presentation of ATL is an ulcer (63%-91% of cases). However, some lesions do not ulcerate as in diffuse cutaneous leishmaniasis. The patient under study had a nodular lesion for at least nine months with no signs of ulceration.<sup>3,4,5V</sup>

When attempting diagnosis of ATL it is important to investigate the history, the clinical findings, the epidemiological information, and to carry out additional tests. When a typical clinical scenario appears in endemic regions, the diagnosis does not usually present any difficulty. However, atypical cases can result in a misdiagnosis, even in endemic regions.



**FIGURE 3:** There is a large number of leishmania amastigotes within macrophages. (H&E 1000x)



**FIGURE 4:** Regression of the lesion four months after the end of the therapy

Some authors argue that ATL is a “great imitator” of other conditions. Tests should be carried out for a differential diagnosis.<sup>3,6</sup>

The ear has not been reported as a common site of ATL involvement. In an observational study by Murback (2011) over 10 years, the ear was the site of inoculation in only one out of 47 patients.<sup>4</sup> In another study in Iraq involving 107 patients with ATL, only one patient had an infection on the ear.<sup>5</sup> According to Martinelli (2005), the ear is an extremely rare site for ATL.<sup>7</sup>

Our patient showed no impairment of cell-mediated immunity, which could be verified by the positivity of the LST, which was consistent with the duration of the skin lesion. This skin test indicates delayed hypersensitivity reaction to *Leishmania*, which plays an important role in disease resolution and healing. The test characteristically becomes positive 4-6 weeks after initiation of the skin lesion.<sup>1</sup>

Furthermore, the diagnosis is supported by isolating *Leishmania* sp. in culture or by detecting amastigotes in histological sections, although parasitological demonstration is not always attained. In a study in Rio de Janeiro (2008) a histopathological analysis of 20 cases showed that amastigotes were found in only two samples.<sup>3</sup> Venkataraman's previous study (2001) evaluated the clinical and histopathological correlation of ATL in 40 patients, where four histological patterns were identified: 1) diffuse macrophage infiltration without necrosis, involving over 100 amastigotes in a standard section; 2) macrophage infiltration with necrosis, with 10-100 amastigotes in a standard section; 3) early reactive

granuloma; and 4) established epithelioid granuloma, without amastigotes. The number of amastigotes decreases as the disease evolves. In lesions of 3-12 months duration pattern 2 above was the most common. Venkataraman *et al* also found that the most common epithelial change was pseudo-epithelial hyperplasia. Only 10% of the patients had atrophy of the epidermis, but all of them experienced the chronic phase (over 12 months).<sup>8</sup> In addition, when the histopathological examination shows macrophages filled with amastigotes, we must consider the species concerned. In Ceará, cases of *L. braziliensis* and *L. amazonensis* have been detected. In the infection by *L. braziliensis*, the infected cells are usually evidenced in the first weeks of the disease. After this period the parasites become rarer, with a dense infiltrate of non-infected plasma cells, lymphocytes and macrophages. When dealing with infection by *L. amazonensis*, parasites persist in macrophages for several months (6 or more). While the case under study was apparently an infection by *L. amazonensis*, a molecular biology investigation for confirmation of the species was not however possible.<sup>9</sup>

The treatment of ATL depends on its clinical manifestations as well as on the location and diameter of the lesion. Treatment includes systemic or intraleisional pentavalent antimonials, sodium stibogluconate and meglumine antimonials (Glucantime®) and cryotherapy. We chose the systemic meglumine antimoniate (15 mg/kg/day) for 20 days, with good control.<sup>10</sup> □

## REFERENCES

1. Brasil. Ministério da Saúde. Secretaria de Vigilância em Saúde. Manual de Vigilância da Leishmaniose Tegumentar Americana. 2. ed. atual. Brasília: Ministério da Saúde; 2007. p.13.
2. Brasil. Ministério da Saúde. Sistema Nacional de Vigilância em Saúde. Sistema nacional de vigilância em saúde: relatório de situação: Ceara. 2. ed. Brasília: Ministério da Saúde; 2006.
3. Vieira-Gonçalves R, Pírmex C, Jorge ME, Souza WJ, Oliveira MP, Rutowitsch MS, et al. Clinical features of cutaneous and disseminated cutaneous leishmaniasis caused by *Leishmania (Viannia) braziliensis* in Paraty, Rio de Janeiro. *Int J Dermatol*. 2008;47:926-32.
4. Murback NDN, Hans Filho G, Nascimento RAS, Nakazatol KRO, Dorval MEMC. American cutaneous leishmaniasis: clinical, epidemiological and laboratory studies conducted at a university teaching hospital in Campo Grande, Mato Grosso do Sul, Brazil. *An Bras Dermatol*. 2011;86:55-63.
5. AlSamarai AM, AlObaidi HS. Cutaneous leishmaniasis in Iraq. *J Infect Dev Ctries*. 2009;3:123-9.
6. Sabri A, Khatib L, Kanj-Sharara S, Husseini ST, Nuwayri-Salti N, Semaan R, et al. Leishmaniasis of the auricle mimicking carcinoma. *Am J Otolaryngol*. 2009;30:285-7.
7. Martinelli C, Giorgini S, Minu MB, Orsi A, Leoncini F. Cutaneous leishmaniasis: An atypical case. *Int J Dermatol*. 2005; 44:38-40.
8. Venkataram M, Moosa M, Devi L. Histopathological spectrum in cutaneous leishmaniasis: a study in Oman. *Indian J Dermatol Venereol Leprol*. 2001;67:294-8.
9. Moraes MAP; Silveira FT. Histopatologia da forma localizada de leishmaniose cutânea por *Leishmania (Leishmania) amazonensis*. *Rev Inst Med Trop São Paulo*. 1994;36:459-63.
10. Almeida OLS, Santos JB. Advances in the treatment of cutaneous leishmaniasis in the new world in the last ten years: a systematic literature review. *An Bras Dermatol*. 2011;86:497-506.

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How to cite this article: Adriano AL, Leal PAB, Breckenfeld MP, Costa IS, Almeida C, Sousa ARD. American tegumentary leishmaniasis: an uncommon clinical and histopathological presentation. *An Bras Dermatol*. 2013;88(2):260-2.