

## RELATO DE CASO

## LEISHMANIOSE TEGUMENTAR AMERICANA COM PADRÃO CUTANEOMUCOSO DISSEMINADO: UM RELATO DE CASO

## AMERICAN CUTANEOUS LEISHMANIASIS WITH DISSEMINATED MUCOCUTANEOUS PATTERN: A CASE REPORT

Murilo Bazzo Carvalho<sup>1</sup>, Natália Ferrer Simões de Sousa<sup>1</sup>, Luísa Lopes Dias da Silva<sup>1</sup>, Rafael Nogueira Araújo de Lima<sup>2</sup>, Mônica Silva Dias Franco<sup>3</sup> Beatriz César de Oliveira<sup>4</sup>.



ACESSO LIVRE

**Citação:** Carvalho MB, Sousa NFS, Silva LLD, Lima RNA, Franco MSD, Oliveira BC. (2021) Leishmaniose tegumentar americana com padrão cutaneomucoso disseminado: um relato de caso. Revista de Patologia do Tocantins, 8(4).

**Instituição:** <sup>1</sup>Interno de Medicina, discente da Universidade Federal do Tocantins (UFT), Palmas, Tocantins, Brasil. <sup>2</sup>Médico infectologista pela Universidade Federal do Tocantins (UFT), Palmas, Tocantins, Brasil. <sup>3</sup>Médica residente de infectologia pela Universidade Federal do Tocantins (UFT), Palmas, Tocantins, Brasil. <sup>4</sup>Médica, formada pelo Instituto Tocantinense Presidente Antônio Carlos (ITPAC), Porto Nacional, Tocantins, Brasil.

**Autor correspondente:** Murilo Bazzo Carvalho; [mbcmurilo@gmail.com](mailto:mbcmurilo@gmail.com). Orla 14, Rua 5, Lote 1, Residencial Graciosa, Palmas, Tocantins, Brasil.

**Editor:** Carvalho A. A. B. Medicina, Universidade Federal do Tocantins, Brasil.

**Publicado:** 10 de janeiro de 2022.

**Direitos Autorais:** © 2022 Carvalho et al. Este é um artigo de acesso aberto que permite o uso, a distribuição e a reprodução sem restrições em qualquer meio, desde que o autor original e a fonte sejam creditados

**Conflito de interesses:** os autores declararam que não existem conflitos de interesses.

## RESUMO

**Introdução:** A Leishmaniose Tegumentar Americana (LTA) se configura como uma patologia endêmica ao Estado de Tocantins, sendo responsável por importante morbidade e estigma social. A doença pode se apresentar de diversas maneiras, variando de formas cutâneas localizadas a cutaneomucosas disseminadas, sendo esta última uma apresentação rara e objeto deste trabalho. **Desenvolvimento:** Paciente com lesões dolorosas ulceradas disseminadas em pele e região cutânea, diagnosticado com leishmaniose tegumentar americana, tratado com anfotericina B lipossomal, apresentando boa resposta à terapia. **Considerações finais:** Pela gravidade do caso e pela endemidade das patologias que integram o diagnóstico diferencial, a descrição desta patologia pode auxiliar futuras pesquisas e no reconhecimento precoce de novos casos.

**Palavras-chave:** Leishmaniose Tegumentar Americana; Diagnóstico; Tratamento.

## ABSTRACT

**Introduction:** The American Cutaneous Leishmaniasis (ACL) is an endemic pathology in the Brazilian state of Tocantins. It is a frequent cause of morbidity and social stigma. This disease can present itself in different kinds of ways, from the traditional form, to the disseminated one, which is a rare entity. **Development:** Patient presenting painful ulcerated and disseminated lesions on the skin and mucosa, diagnosed with American cutaneous leishmaniasis, treated with liposomal amphotericin B, showed a good response to therapy. **Final considerations:** Because of the severity of the case and the endemicity of diseases which integrate the differential diagnosis, the description of this pathology may help with future research and with the early identification of new cases.

**Keywords:** American cutaneous leishmaniasis; Diagnosis; Treatment.

**INTRODUCTION**

The American Cutaneous Leishmaniasis (ACL) is a noncontagious, infectious disease, caused by the protozoa of the *Leishmania* gender, which are transmitted to humans by the bite of a phlebotomine sandfly. The most common specie on the Legal Amazonia, including Tocantins, it is the *Leishmania amazonenses*, but there are described cases by the specie *Leishmania brasiliensis* as well.<sup>1,2,3</sup>

The most common clinical feature is the single or multiple ulcerated lesions, with elevated borders, typically painless.<sup>1</sup> These lesions are common in exposed areas of the body, where the mosquito gains access.<sup>2</sup> Nonetheless, if the lesions are located on the mucosa, frequently will be found on the nose, mouth and throat, appearing in the form of the following signs and symptoms: nasal congestion, epistaxis, running nose, hoarseness and cough.<sup>4</sup>

ACL may manifest solely on the skin, mucous membranes, or both. Furthermore, the lesions are classified by their site of occurrence in localized or disseminated, the latter being the rarest one, observed only in 2% of cases. The dissemination phenomenon occurs, probably, via blood or lymphatics.<sup>2</sup> After searching in the bases Medline, PubMed and Scielo, the authors did not find any article describing this pattern of disseminated ACL in the State of Tocantins.

**CASE REPORT**

A.F.S., male, 50 years old, married, admitted to the hospital with fever, multiple ulcerated and painful lesions (Image 1), located on left infraorbital, infrascapular, inguinal regions, left gluteus, penis, and palate. They appeared with well delimited, elevated, and erythematous borders, grainy floor, without exudate. The patient awaited almost three months to seek the doctor, for which the lesions worsened progressively within that timeline. His BMI was 28,4, therefore he was overweight.

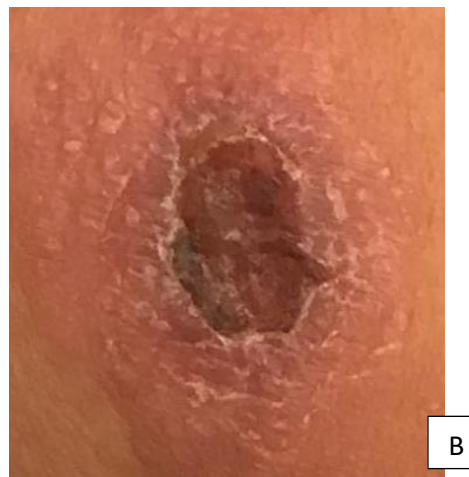
He has also had splenomegaly (which was confirmed by further investigation with an abdominal ultrasound), and altered renal function with creatine of 1,5g/dL and BUN 64g/dL. His hemoglobin was 11g/dL and hematocrit 32%. Before that presentation, the diagnosis of disseminated american cutaneous leishmaniasis was suggested and confirmed by histopathology.

The patient was treated with liposomal amphotericin B 350mg and glucose serum 5 % (200ml) intravenous for 11 days. During the hospitalization, the blood exams to analyze his renal and liver function were made daily, and he had a renal dysfunction during the treatment as the graphics on the Table 1 show.

Nephrotoxicity is a common adverse effect of the amphotericin B and it can be responsible for causing acute renal injury. But whereas the treatment went by his creatinine and BUN got back to normal, therefore it was not necessary to stop treatment. The drug chosen was not pentavalent antimoniate because of his age (> 50 years) and because of his renal

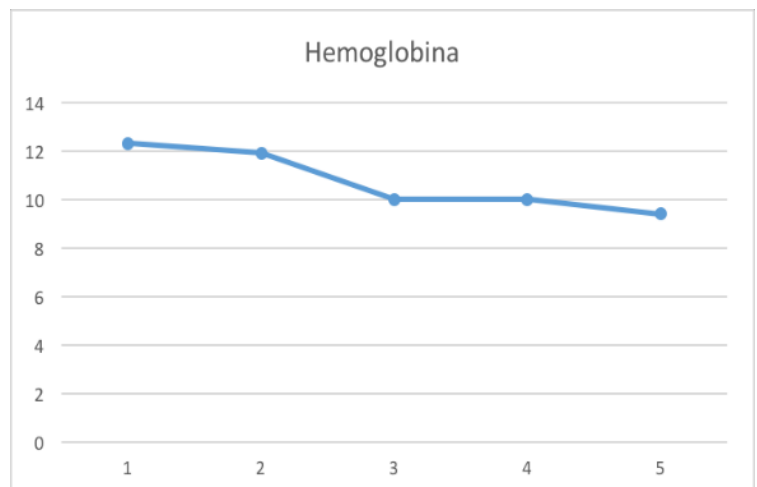
dysfunction. The patient's hospitalization was a short and relatively calm time, and his treatment was a success.

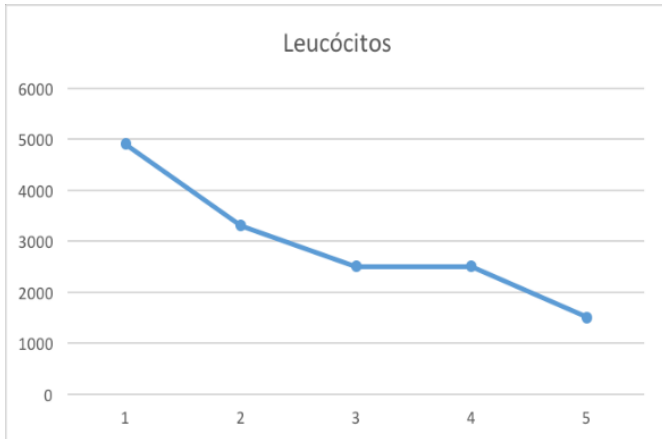
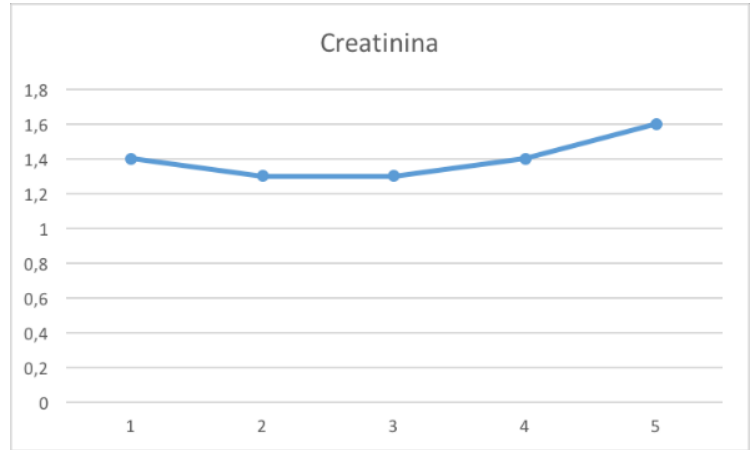
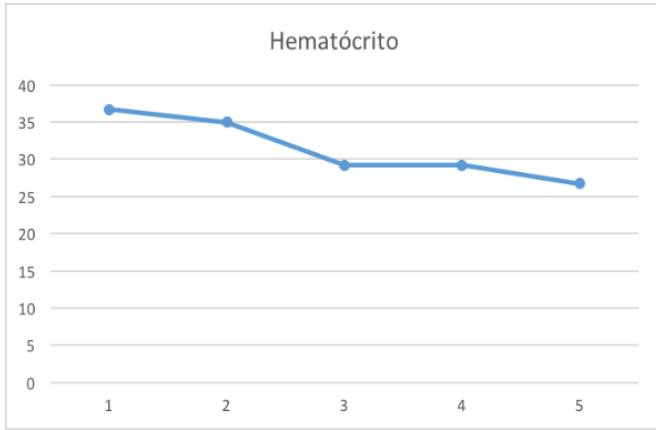
Image 1: Picture of the lesion with Cutaneous Leishmaniasis. A: initial stage; B: healing.



Reference: Own author, 2020.

Table 1: Complementary exams





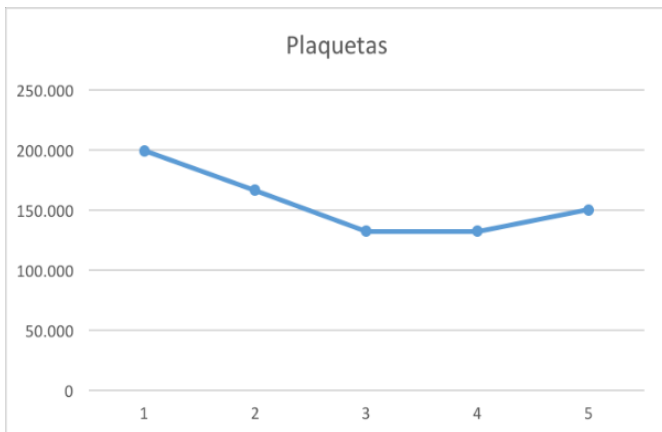
Unities: Hemoglobin (g/dL), Hematocrit (%), Leucocytes (mm<sup>3</sup>), Platelets (mm<sup>3</sup>), Urea (mg/dL), Creatinine (mg/dL)

Reference: Own author, 2020.

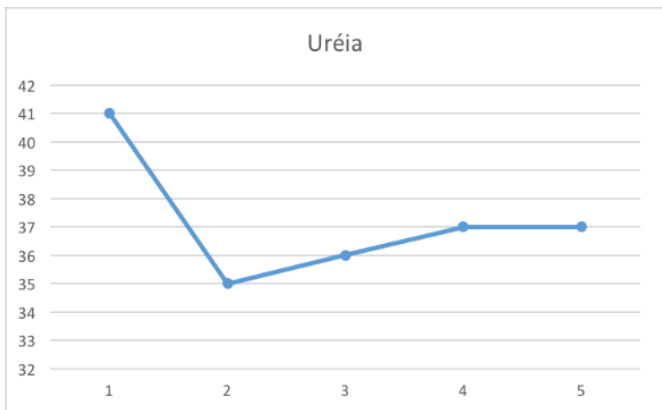
**DISCUSSION**

Leishmaniasis can appear in its cutaneous, mucocutaneous or visceral form, and has a range of varied symptoms, that will depend on the immunocompetence of the affected person and the availability to treatment at his place of residence.<sup>2,3,5</sup> The incidence of this disease has increased in Brazil, and it deserves attention from health authorities, considering the possibility to control the vectors and the access to drugs and treatment, which despite being expensive and with potentially serious side effects, generates good results prognoses.<sup>6,7</sup>

Epidemiological studies are important in places where the environment favors the transmission of this disease. Therefore, factors such as the number of residents of the house, the presence of domestic and wild animals in the vicinity and forest areas close by, facilitate not only the mapping of risk areas, but also the early diagnosis of cases.<sup>2,6,7</sup> It is necessary to search for female phlebotomines and their possible hosts. The presence of animals in the surroundings has been reported in many places where cases have been confirmed, in other Brazilian regions. In 2019, 401 cases were notified in the state of Tocantins, 370 of which were cutaneous and 31 were mucous.<sup>8,9</sup>



The simultaneous involvement of mucous membranes and tegument is a rare clinical entity, which has never been reported in the state of Tocantins. It shows the importance of this case for similar future occasions. The human being usually gets involved in the cycle of the disease in an indirect way, because the infection is essentially zoonotic.<sup>4,6,10</sup> Signs and symptoms can take 10 to 60 days to manifest, considering that the cutaneous tissue is damaged earlier than the mucous membranes.<sup>5,10</sup> Unlike ulcers found on the skin, classic of the disease, mucosal involvement has a destructive, granulomatous aspect, with deep grooves, accompanied by painful symptoms and dysphagia.<sup>10</sup> The most common sites of involvement are lips, hard palate, soft palate and uvula.<sup>2</sup> This incubation period and the manifestation of symptoms were



consistent with the natural course of the disease found in the patient on this case report.

Despite that, it was analyzed other diagnostic possibilities such as sporotrichosis (which may cause local or disseminated pustules, ulcers and nodules, depending on the patient immunologic state).<sup>2,5</sup> Paracoccidioidomycosis (caused by a dimorphic fungus which causes local lesions and pneumonia), lepromatous leprosy (an endemic disease in the State of Tocantins which presented an incidence of 80,57 per 100,000 inhabitants in 2017, whereas the national scenario was 73,8)<sup>11</sup> and tertiary syphilis (characterized by gummas or other clinical features like general paresis or aortitis).<sup>6</sup>

## CONCLUSION

The conclusion was that the patient had disseminated leishmaniasis, caused by *Leishmania amazonenses*, a common etiologic agent in this area. It was confirmed based on histopathology exam. The symptoms included multiple ulcerated lesions, fever, and splenomegaly, which suggested the disease, and guided all management to exclude other potential diagnosis, such as those cited above. This type of presentation of leishmaniasis is rare and was not found any described case in the literature, therefore the authors stated that it was the first case documented in Tocantins. Once the patient started the therapy with liposomal amphotericin B he started to get better, suggesting that he had an efficient treatment.

## REFERÊNCIAS BIBLIOGRÁFICAS

1. Brasil. Ministério da Saúde. Secretaria de Vigilância em Saúde. Sistema Nacional de Vigilância em Saúde. Relatório de Situação. Tocantins. 2ª Ed. Brasília/DF, 2006, 25p.
2. Gontijo CMF, Melo MN. Leishmaniose Visceral no Brasil: quadro atual, desafios e perspectivas. Rev Bras de Epidem, 7(3):338-47, 2004. <http://dx.doi.org/10.1590/S1415-790X2004000300011>
3. Filho JDAF, Valente MB, Andrade WA, Brazil RP, Falcao AL. Flebotomíneos do Estado de Tocantins, Brasil (Diptera: Psychodidae). Rev da Soc Bras de Med Trop, v.34, n.4, p.323-9, 2001. <https://doi.org/10.1590/S0037-86822001000400003>
4. Name RQ, Borges KT, Nogueira LSC, Sampaio JHD, Tauil PL, Sampaio RNR. Estudo clínico, epidemiológico e terapêutico de 402 pacientes com leishmaniose tegumentar americana atendidos no Hospital Universitário de Brasília, DF, Brasil. Anais Bras de Dermatol, v.80, n.3, p.249-54, 2005. <https://doi.org/10.1590/S0365-05962005000300004>
5. Freitas VC, Parreiras KP, Duarte APM, Secundino NFC, Pimenta PFP. Development of Leishmania (Leishmania) infantum chagasi in Its Natural Sandfly Vector Lutzomyia longipalpis. American J of Trop Med and Hygiene, 86(4):606-12, 2012. doi: [10.4269/ajtmh.2012.11-0386](https://doi.org/10.4269/ajtmh.2012.11-0386) PMID: [22492144](https://pubmed.ncbi.nlm.nih.gov/22492144/)
6. Aguiar V, Goncalves GMS, Farias FD. Distribuição dos Casos de Leishmaniose Visceral Humana (calazar) em Pernambuco

- no Ano 2002. Rev da Soc Bras de Med Trop, 36(2), 2003. <https://doi.org/10.1590/S0037-86822003000700012>
7. Ribeiro RSP. Incidência e Epidemiologia da Leishmaniose Visceral no Norte do Tocantins. [Dissertação]. [São Paulo (SP)]: Instituto de Pesquisas Energéticas e Nucleares, Autarquia associada à Universidade de São Paulo. 2010.
8. Brasil. Ministério da Saúde. Portal da Saúde. DATASUS. Acesso em: <http://www2.datasus.gov.br/DATASUS/index.php?area=0201&id=6903>.
9. \_\_\_\_\_. Ministério da Saúde. Portal SINAN. Acesso em: <http://portalsinan.saude.gov.br/dados-epidemiologicos-sinan>.
10. \_\_\_\_\_. Ministério da Saúde. Secretaria de Vigilância em Saúde, Depart de Vigilância Epidemiol. Doenças infecciosas e parasitárias: guia de bolso. 6. ed. rev. Brasília (DF): MS; 2005.
11. Tocantins. Secretaria da Saúde. Anais do 15º Congresso Brasileiro de Hansenologia. 2018. Acesso em: <http://saude.to.gov.br/vigilancia-em-saude/doencas-transmissiveis-e-nao-transmissiveis/hanseniose/15-congresso-brasileiro-de-hansenologia/>