Multiple ulcerated lesions in a patient from Amazon, Brazil



Nathalia Miranda Souto, MD, Sidharta Quercia Gadelha, MD, Virginia Vilasboas Figueiras, MD, Rosilene Viana de Andrade, MD, and Luciana Mendes dos Santos, PhD *Amazon, Brazil*

Key words: Amazon region; direct smear test; leishmaniasis; multiple painless ulcers; PCR.







A 36-year-old woman from Manaus (Amazon, Brazil) presented with a 1-year history of multiple papules and plaques that evolved to painless ulcers on the lower portion of her left limb. Different types of oral antibiotics and corticosteroids (up to 120 mg/d of prednisone) were prescribed, without any improvement. A physical examination revealed papules; plaques; extensive scarring areas; and multiple painless ulcers with elevated, well-defined edges on the lower portion of her left limb (Figs 1 and 2). A direct mycologic examination and direct smear test for leishmaniasis yielded negative results. A polymerase chain reaction (PCR) for leishmaniasis; tissue culture for bacteria, atypical mycobacteria, and fungi; and skin biopsy were performed (Fig 3).

Question 1: What is the most likely diagnosis?

- **A.** Pyoderma gangrenosum
- **B.** Chromoblastomycosis
- **C.** Cutaneous leishmaniasis
- **D.** Mycetoma
- **E.** Cutaneous tuberculosis

Answers:

A. Pyoderma gangrenosum – Incorrect. Pyoderma gangrenosum is a chronic neutrophilic dermatosis with an unknown etiology characterized by multiple painful ulcers usually associated with inflammatory gastrointestinal diseases, with a good response to corticosteroids, which differs from the response in this case.¹

B. Chromoblastomycosis – Incorrect. Chromoblastomycosis lesions are usually verrucous, with the presence of black dots. Medlar bodies are seen via histopathologic and direct mycologic examinations. In the Amazon region, the culture is usually positive for *Fonsecaea pedrosoi*, which is not present in this case.¹

C. Cutaneous leishmaniasis – Correct. Cutaneous leishmaniasis is an infectious disease caused by the protozoan *Leishmania* that can be classified as follows: cutaneous, mucosal, and mucocutaneous forms.¹ Amazon is the Brazilian state that has the highest incidence of cutaneous leishmaniasis (an average of 50 cases per 100,000 inhabitants),² and *Leishmania (Viannia) guyanensis* is the most prevalent species (about 91% of cases).² Clinically, the appearance and extension of skin lesions depend on the species, host genetic factors, and immune system activation. A positive smear test result; a direct examination; the presence of amastigotes, determined using histology; and/or a positive PCR result are used to

From the Tropical Medicine Foundation, Amazon, Brazil.

JAAD Case Reports 2022;21:192-4.

confirm the diagnosis of leishmaniasis. In our case, the PCR result was positive for leishmaniasis, and few amastigotes were seen using histology.¹⁻⁵

D. Mycetoma – Incorrect. Mycetoma is a chronic infection of cutaneous and subcutaneous tissues that can be caused by filamentous bacteria or fungi. Mycetoma is characterized by the following classical triad: tumor or soft tissue swelling, sinus tracts, and characteristic macroscopic grains.¹

E. Cutaneous tuberculosis – Incorrect. Cutaneous tuberculosis is a rare extrapulmonary form of pulmonary tuberculosis. Its clinical manifestations range from papules to verrucous plaques, nodules, and ulcers. Histologically, bacilli and granuloma with caseous necrosis may be present.¹

Question 2: About cutaneous leishmaniasis, choose the correct answer:

A. It is more common in women in the Amazon region.

B. Histopathologic examination reveals a diffuse infiltrate with xanthomized histiocytes and lymphocytes.

C. The diagnosis is confirmed using the direct smear test, which has high specificity and sensitivity.

D. PCR is the most sensitive diagnostic test for cutaneous leishmaniasis.

E. The Montenegro test is a noninvasive method with high rates of positivity in patients with HIV.

Answers:

A. It is more common in women in the Amazon region – Incorrect. In the Amazon region, cutaneous leishmaniasis is more common in men

Funding sources: None.

IRB approval status: Not applicable.

Correspondence to: Nathalia Miranda Souto, MD, br Alaska street, 1071, Building Porto Seguro apartment 1702, Ponta Negra neighborhood, Amazon 69037-057, Brazil. E-mail: nathaliamiranda22@yahoo.com.

²³⁵²⁻⁵¹²⁶

^{© 2022} by the American Academy of Dermatology, Inc. Published by Elsevier, Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-ncnd/4.0/).

https://doi.org/10.1016/j.jdcr.2021.10.041

(approximately 80% of all reported cases) between 20 and 40 years old with a mixed ethnicity (Black and White) and low educational level.^{1,2}

B. Histopathologic examination reveals a diffuse infiltrate with xanthomized histiocytes and lymphocytes – Incorrect. The histopathologic classical findings of cutaneous leishmaniasis are heavy infiltrate of histiocytes, lymphocytes, and plasma cells. Additionally, pseudoepitheliomatous hyperplasia and the presence of amastigotes might occur.^{1,2,3,5}

C. The diagnosis is confirmed using the direct smear test, which has high specificity and sensitivity – Incorrect. The direct smear test is extremely useful for the diagnosis of cutaneous leishmaniasis because it is faster, cheaper, and easier to perform. However, the probability of finding the parasite is inversely proportional to the time of the evolution of cutaneous lesions, which is rare after 1 year.^{2,3,5}

D. PCR is the most sensitive diagnostic test for cutaneous leishmaniasis – Correct. PCR is a method based on the amplification of a parasite's DNA in different types of samples. PCR has the highest sensitivity in typical (100%) and atypical (94%) presentations. Moreover, it has a specificity of 100%, followed by immunohistochemistry, which has a sensitivity of 97% and specificity of 100%.^{2,3,5}

E. The Montenegro test is a noninvasive method with high rates of positivity in patients with HIV – Incorrect. The Montenegro skin test is an invasive technique that shows a false-negative result in patients with HIV because of the lack of a cellular immune response against parasite antigens. Furthermore, the Montenegro skin test may yield a positive result for latent infections and does not distinguish a former infection from a current infection.¹⁻³

Question 3: What is the treatment of choice in this case report?

- A. Intravenous glucantime
- B. Intramuscular pentamidine
- **C.** Intravenous liposomal amphotericin B
- **D.** Intralesional glucantime injections
- E. Pentoxifylline

Answers:

A. Intravenous glucantime – Incorrect. Antimony n-methyl glutamine (glucantime) is the first choice of treatment in patients with a single cutaneous lesion or multiple cutaneous lesions infected by *Leishmania braziliensis* and other species, but with

a poor response to *Leishmania guyanensis*, which is more common in the Amazon region in Brazil.^{2,4}

B. Intramuscular pentamidine – Correct. Intramuscular pentamidine is the first choice of treatment for *Leishmania guyanensis* because this infection is resistant to systemic antimony.^{2,4} In our case, the patient had a great result after 4 mg/kg of intramuscular pentamidine once a week for 3 weeks.

C. Intravenous liposomal amphotericin B – Incorrect. Liposomal amphotericin B in areas of the predominance of *Leishmania guyanensis* is the first choice of treatment in patients with renal, cardiac, and hepatic failure as well as in pregnant women and people older than 50 years old, which differs from our case report.²

D. Intralesional glucantime injections – Incorrect. Intralesional glucantime injections are the first choice of treatment for cases of *Leishmania guyanensis* in which a single cutaneous lesion measures up to 3 cm at its largest diameter, which differs from our case.²

E. Pentoxifylline – Incorrect. Pentoxifylline is a peripheral vasodilator and is not the first choice of treatment for cutaneous leishmaniasis. Some research has shown some benefit of pentoxifylline as an adjuvant therapy by decreasing the time of healing and the toxicity associated with antimoniate.²

Abbreviation used:

PCR: polymerase chain reaction

Conflicts of interest

None disclosed.

REFERENCES

- 1. Teles GD, Fonseca FR, Gonçalves MJ. American tegumentary leishmaniasis in the Brazilian Amazon from 2010 to 2014. Accessed October 7, 2021. https://www.scielo.br/j/rimtsp/a/ hKXGnJm6nxnLFWs65QfRssd/?lang=en
- BRASIL, Ministério da Saúde. Manual de Vigilância da Leishmaniose Tegumentar Americana. Secretaria de Vigilância em Saúde. Accessed February 14, 2022. http://bvsms.saude.gov.br/bvs/ publicacoes/manual_vigilancia_leishmaniose_tegumentar.pdf
- Gurel MS, Tekin B, Uzun S. Cutaneous leishmaniasis: a great imitator. *Clin Dermatol.* 2020;38(2):140-151. https://doi.org/10. 1016/j.clindermatol.2019
- Guerra JA, Maciel MG, Guerra MV, et al. Tegumentary Leishmaniasis in the State of Amazonas: what have we learned and what do we need? *Rev Soc Bras Med Trop.* 2015;48(suppl 1):12-19. https://doi.org/10.1590/0037-8682-0268-2013
- Aronson NE, Joya CA. Cutaneous leishmaniasis: updates in diagnosis and management. *Infect Dis Clin North Am.* 2019;33(1):101-117. https://doi.org/10.1016/j.idc.2018.10.004