Rev Soc Bras Med Trop 51(1):108-110, January-February, 2018
doi: 10.1590/0037-8682-0306-2017

Case Report

American cutaneous leishmaniasis triggered by electrocoagulation

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Abstract

Cutaneous leishmaniasis is usually transmitted by infected phlebotomine sand fly bites that initiate local cutaneous lesions. Few reports in the literature describe other modes of transmission. We report a case of a previously healthy 59-year-old woman who underwent electrocoagulation to remove seborrheic keratosis confirmed by dermatoscopy. Three months later, a skin fragment tested positive for *Leishmania* culture; the parasite was identified as *L. (V.) braziliensis*. Trauma may generate inflammatory cascades that favor *Leishmania* growth and lesion formation in previously infected patients. American cutaneous leishmaniasis is a dynamic disease with unclear pathophysiology because of continually changing environments, demographics, and human behaviors.

Keywords: Leishmaniasis. Trauma. Atypical presentation.

INTRODUCTION

Cutaneous leishmaniasis (CL) is an endemic infectious disease, representing a large public health problem worldwide[1]. In Brazil, an average of 30,000 cases are reported annually. Usually, CL is transmitted by infected phlebotomine sand fly bites, and a subclinical infection in the host has been proven[1]. There are a few reports that describe unusual modes of parasite infection, such as through direct contact, rat bites, accidental laboratory inoculation[2,3], localized trauma[4,5], and the use of immunomodulators such as anti-tumor necrosis factor (TNF)-alpha[6,7]. Here, we report a case of American tegumentary leishmaniasis (ATL) following electrocoagulation for a seborrheic keratosis skin lesion.

CASE REPORT

A previously healthy 59-year-old female Brazilian biologist who often travels to ATL endemic areas underwent an electrocoagulation procedure to remove two hyperchromic squamous stable lesions, that appeared after 50 years of age, when the patient was no longer traveling to endemic areas. Clinically and dermoscopically, both lesions were diagnosed as seborrheic keratosis: one on the nose and the other on the thigh. Two months later, with no reported travel to endemic ATL regions, erythematous nodules emerged on the surgical scars. One month later, the nodules were removed and sent for histopathology, fungal, bacterial, and mycobacterial direct examinations and cultures, which were reported to be negative. Only 6 months later and after the third biopsy, the histopathology showed a lymphohistiocytic infiltrate with granulomas and multinucleated cells, and some rare roundish forms compatible with amastigotes, which were confirmed by immunohistochemistry. A skin fragment tested positive for *Leishmania* culture, and the parasite was identified as *L. (V.) braziliensis* using a polymerase chain reaction (PCR) assay and restriction enzyme analysis[8]. Montenegro skin test and indirect immunofluorescence assay results were positive and human immunodeficiency virus (HIV) serology test results were negative. She was treated with 2g of liposomal amphotericin B and had completely healed by the time of this report (**Figure 1 and Figure 2**).

DISCUSSION

Here, we reported the case of an immunocompetent patient with no previous sign of leishmaniasis. After undergoing a very common dermatological procedure for the removal of two well-defined benign lesions on distant parts of the body, after a short period of time, she presented with simultaneous

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Received 18 August 2017
Accepted 1 December 2017

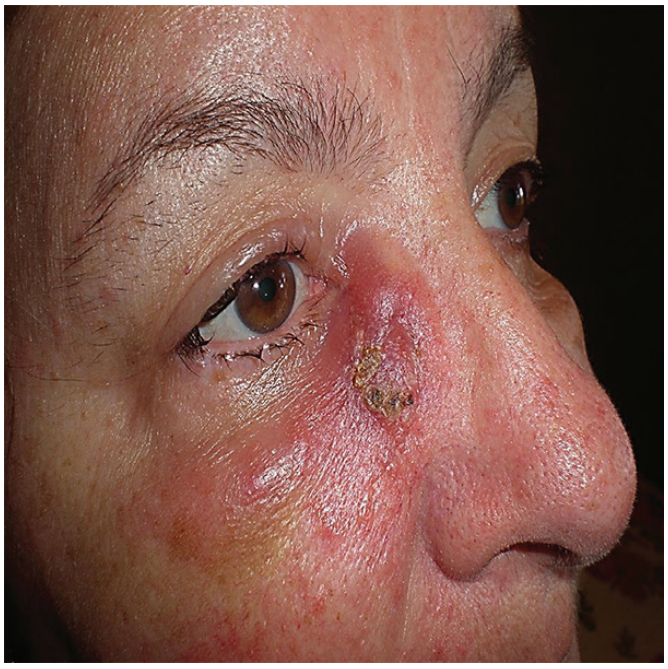

FIGURE 1: Ulcerated erythematous infiltrated plaque on the right malar and nose lateral region 3 months after seborrheic keratosis electrocoagulation.

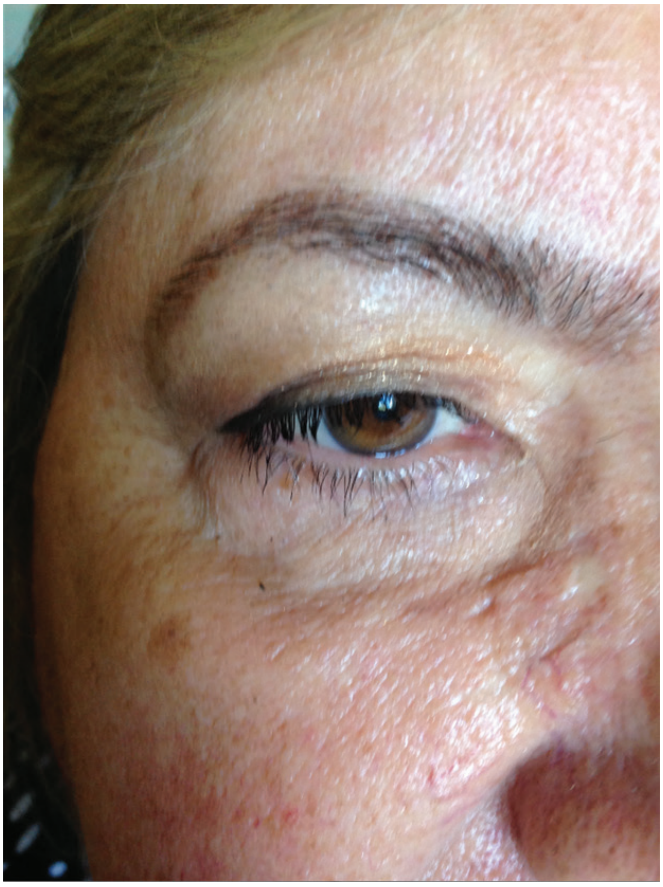

FIGURE 2: Complete healing after liposomal amphotericin B treatment.

CL lesions on both sites. This presentation reinforces the idea that electrocoagulation may have triggered ATL.

In the Americas, the main species that causes ATL is *L. (V.) braziliensis*[8]. Previous studies of *Leishmania* serology, Montenegro skin tests, and the presence of *Leishmania* DNA identified using PCR techniques have shown that in Brazilian endemic areas, 10% of the population has subclinical infections[7]. It is also important to note that there are some reported cases of primary or secondary CL lesions after localized trauma, and these lesions can appear months later[4,5]. In our experience of more than 2,000 cases of ATL, we have observed 3 cases triggered by trauma: one after a bicycle trauma on the ankle cause by *L. (L.) amazonensis*[9]; one after a snake bite; and one after a laser hair removal procedure. In addition, previous studies in murine models infected with *Leishmania* support the possibility of metastatic cutaneous lesions at sites of trauma[5].

Considering the concepts presented above, we may consider that trauma generates an inflammatory cascade that favors *Leishmania* dissemination and lesion formation in previously infected patients. This hypothesis is supported by the local formation of immunosuppressive cytokines and transformation of growth factor-p that exacerbates lesion development. Previous studies in Balb-c mice infected with *L. (V). braziliensis* have shown that these factors could reactivate the subclinical infection[10,11]. Our patient was a biologist that lived for 9 years in Rio de Janeiro, Brazil, and travelled several times to endemic regions. She presented typical lesions of seborrheic keratosis, the diagnosis of which was confirmed by clinical and dermatoscopy examinations, and that appeared when she no longer travelled.

In this case and in others found in the literature, we note that the clinical presentation is usually characterized by nodules, plaques, and papules that are not typical unique ulcers, making the diagnosis even more difficult. It is important to differentiate this case, wherein an immunocompetent patient had a previous subclinical *Leishmania* infection, from the previously referenced cases in which the parasite was inoculated by agents other than phlebotomine sand flies[2,3]. It is believed that disease activation was triggered by the electrocoagulation procedure, characterized as the known *locus minoris resistentiae phenomenon*[12].

Histopathology is the gold standard examination for the diagnosis of seborrheic keratosis, but those lesions are a part of dermatologists' daily practice; seborrheic keratosis lesions exhibit unique clinical characteristics, with no malignant potential, and are cured by electrocoagulation procedures. In addition, dermatoscopy examinations are also commonly performed.

ATL is a dynamic disease, the pathophysiology of which is not well understood because of the continually changing environmental, demographic, and human behavioral factors. It is important for all physicians to be aware of the possibility of CL lesion onset at the sites of dermatological and aesthetical procedures in endemic and nonendemic areas, as the world is globalized and leishmaniasis is considered a re-emerging disease.

Conflict of interest

The authors declare that there are no conflicts of interest.

Acknowledgements

We thank the laboratorial staff from the Dermatomycology laboratory of the Universidade de Brasília, Brasília – Brazil and clinical staff from the dermatology department of the Hospital Universitário de Brasília, Brasília – Brazil.

Financial support

Fundação de Apoio à Pesquisa do Distrito Federal. Process n. 0193.001447/2016.

REFERENCES

1. Mendes DG, Lauria-Pires L, Nitz N, Lozzi SP, Nascimento RJ, Monteiro OS, et al. Exposure to mixed asymptomatic infections with *Trypanosoma cruzi*, *Leishmania braziliensis* and *Leishmania chagasi* in the human population of the greater Amazon. Trop Med Int Health. 2007;12(5):629-36.
2. Dillon NL,Ometto Stolf H, alvarenga Yoshida EL, Alencar Marques ME. Leishmaniose cutânea acidental. Rev Inst Med Trop Sao Paulo. 1993;35(4):385-7.
3. Mardsen PD, Almeida EA, Llanos-Cuentas EA, Costa JL, Megalhães AV, Peterson NE, et al. *Leishmania braziliensis braziliensis* infection of the nipple. Br Med J (Clin Res Ed). 1985;290(6466):433-4.
4. Mulvaney P, Aram G, Maggiore PR, Kutzner H, Carlson JA. Delay in diagnosis: trauma-and coinfection-related cutaneous leishmaniasis because of *Leishmania guyanensis* infection. J Cutan Pathol. 2009;36(1):53-60.
5. Wortmann GW, Aronson NE, Miller RS, Blazes D, Oster CN. Cutaneous leishmaniasis following local trauma: a clinical pearl. Clin Infect Dis. 2000;31(1):199-201.
6. Nicodemo AC, Duailibi DF, Feriani D, Duarte MIS, Amato VS. Mucosal leishmaniasis mimicking T-cell lymphoma in a patient receiving monoclonal antibody against TNFα. PLoS Negl Trop Dis. 2017;11(9):e0005807.
7. Aquino TA, Martins SS, Gomes CM, Carneiro da Motta JO, Graziani D, Rodrigues AMS, et al. First case report of cutaneous leishmaniasis caused *by Leishmania (Leishmania) infantum* in a Brazilian patient treated with adalimumab. J Clin Exp Dermatol Res. 2014;5(6):245.
8. Gomes CM, de Paula NA, Cesetti MV, Roselino AM, Sampaio RN. Mucocutaneous leishmaniasis: accuracy and molecular validation of noninvasive procedures in a *L. (V.) braziliensis*-endemic area. Diagn Microbiol Infect Dis. 2014;79(4):413-8.
9. Sampaio RNR, Marsden PD, Llanos Cuentas EA, Cuba Cuba CA, Grimaldi Jr G. *Leishmania mexicana amazonensis* isolated from a patient with fatal mucosal leishmaniasis. Rev Soc Bras Med Trop. 1985;18(4):273-4.
10. Barral A, Barral-Netto M, Yong EC, Brownell CE, Twardizik DR, Reed SG. Transforming growth factor beta as a virulence mechanism for *Leishmania braziliensis*. Proc Nati Acad Sci USA. 1993;90(8):3442-6.
11. Travi BL, Osorio Y, Saraiva NG. The Inflammatory Response Promotes Cutaneous Metastasis in Hamsters Infected with *Leishmania (Viannia) panamensis*. J Parasitol. 1996;82(3):454-7.
12. Blume-Peytavi U, Tan J, Tennstedt D, Boralevi F, Fabbrocini G, Torrelo A, et al. Fragility of epidermis in newborns, children and adolescents. J Eur Acad Dermatol Venereol 2016;30(suppl 4):3-56.