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Case Report

Cutaneous leishmaniasis with atypical clinical manifestations: Case report

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ABSTRACT

This case report alerts to the existence of atypical forms of cutaneous leishmaniasis (CL). A woman with nodular cutaneous lesions over a neck with papules and pustules located deep in the hypodermis that formed plaques with subcutaneous induration and satellite papules was confirmed to have CL. After confirmation, the patient was treated with remission of the lesions, scarring and thickening of the skin.

Case presentation

A 61-year-old female with lesions on the neck, was referred under physician order in July 2011 to the LEPAC of the Universidade Estadual de Maringá for diagnosis of cutaneous leishmaniasis (CL). The patient reported that approximately 7 months previously (December 2010) a single vesicular lesion appeared on her neck, not ulcerated, with prominent fluid accumulation, intense pruritus, and subsequent swelling and redness, without fever. A physical examination revealed pale dry skin, decreased subcutaneous tissue, a swollen and erythematous neck, with nodular lesions containing papules and pustules located deep in the hypodermis, forming plaques with subcutaneous induration and satellite vesicles. According to the patient, these vesicles ruptured and new lesions appeared nearby. Lymphadenitis was also present, as well as three open skin lesions with crusts and exudate, up to 1.5 cm in diameter (Fig. 1A and B). This clinical picture, together with the rupture of the vesicles and the appearance of new lesions adjacent to the original lesion, is not typical of CL. Thirteen months after the initial infection and two months after completing the treatment, in January 2012 the patient returned to the laboratory for new tests in order to manage her treatment. At this time we observed remission of the lesions, with scarring and thickening of the skin (Fig. 1C), and search for anti-*Leishmania* IgG antibodies by indirect immunofluorescence (IIF) test was nonreactive. Until March 2013, the patient has shown no indication of relapse according to the criteria recommended by the Ministry of Health [1].

Discussion

Leishmaniasis is a group of protozoonoses, transmitted by the bite of naturally infected sandflies (*Lutzomyia*), that can take forms with destructive and disabling lesions [2]. Leishmaniasis have a high incidence [3], and the different clinical forms of the disease are characterized in terms of the biological complexity of the parasite, reservoirs, vectors, environment, and immune response of the host [4]. In Brazil, CL cases are mainly due to *Leishmania*

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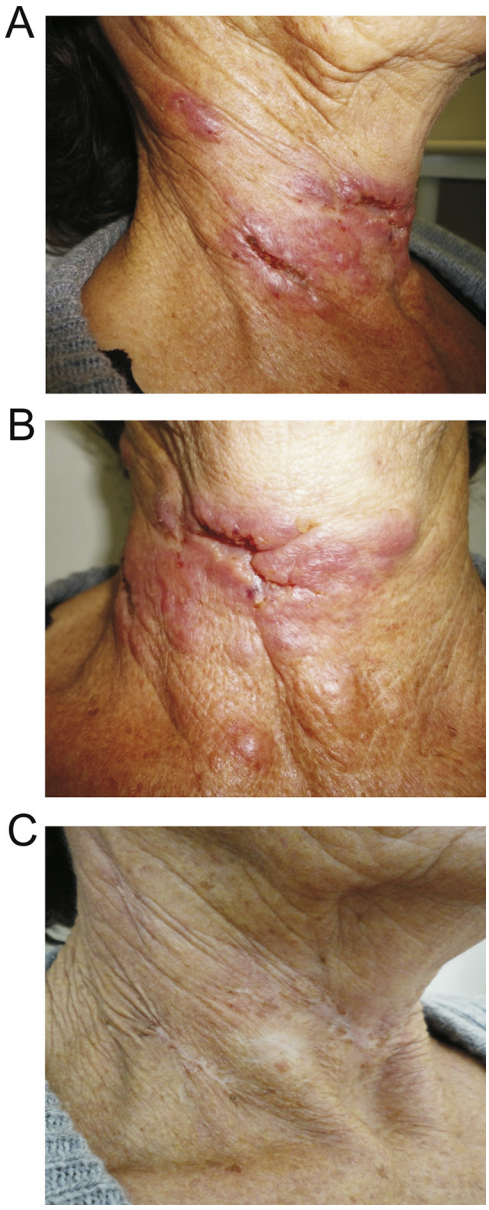

Fig. 1. Patient with cutaneous leishmaniasis. (A and B) Clinical manifestations of cutaneous leishmaniasis, 7 months after the initial infection and before treatment. (C) Healing of cutaneous lesions after two months of treatment.

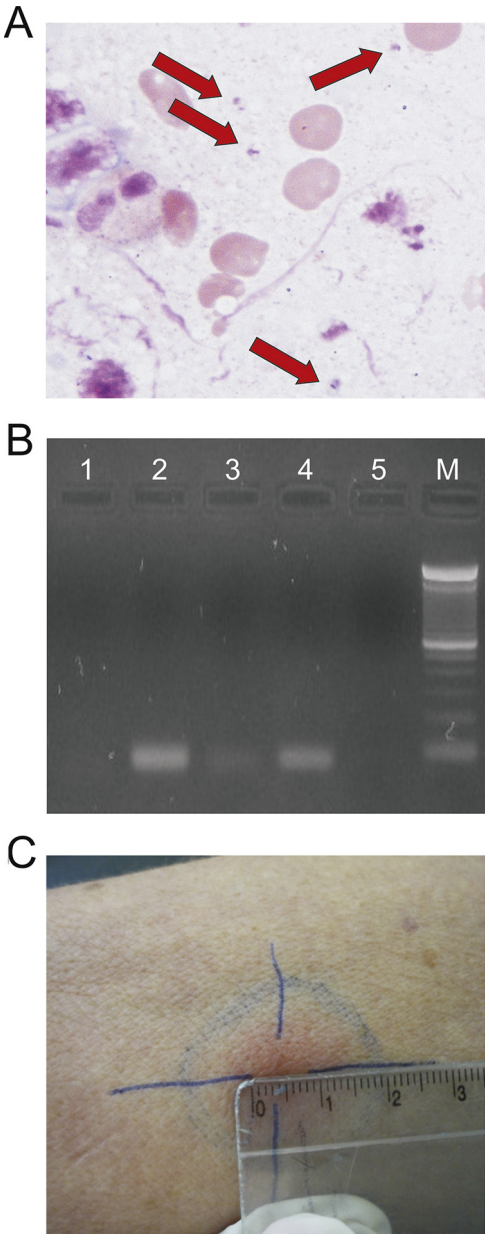

Fig. 2. Laboratory tests to diagnose CL. (A) Direct parasite search in a scraping from the lesion, showing features of amastigotes of *Leishmania* spp.; stained with Giemsa, analyzed by optical microscopy (1000×). (B) Agarose gel showing the 70-bp fragment from the kDNA of *Leishmania* (*Viannia*). Lane 1, negative control (reaction mixture plus water), lane 2, positive control [reaction mixture plus *L.* (*V.*) *braziliensis* DNA]; lane 3, sample of peripheral blood leukocytes; lane 4, sample from lesion scraping; lane 5, negative control extraction (blood of patients without CL); M, 100-bp molecular marker (Invitrogen Life Technologies, São Paulo, Brazil). (C) Result of the Montenegro skin test (0.1 ml of antigen) showing a papule 8.0 mm in diameter that developed following injection of the antigen.

(*Viannia*) *braziliensis*, which causes lesions that if left untreated may result in the mucosal form, which is characterized by disfiguring lesions [1].

The patient, a resident in the municipality of Rio Bom (23°45′ S and 51°24′ W), southern Brazil, reported that the infection likely occurred in a rural locality with woods, a stream, and wild and domestic animals, which are appropriate conditions for sandflies. Sandflies were formerly reported in deforested areas, but more recently have been found in human-impacted rural and urban areas [5,6]. The municipality of Rio Bom belongs to the Paraná-Paranapanema Circuit of CL [7].

Clinical examination is based on epidemiological data and the characteristics of the lesion. Following a sandfly bite, a localized skin lesion appears that develops from an inflammatory wheal and usually leads to an ulcer. In general these skin lesions have a wide variety of forms: round or oval; erythematous base, infiltrated and firm in consistency; well-defined, high edges; reddish background and coarse granules [1]. Vegetating lesions with a papillomatous aspect and a soft moist consistency, and verrucous lesions with a dry rough surface and the presence of small scabs and peeling are less common [1].

The patient sought medical care and was treated with a topical antibacterial ointment and intramuscular benzathine penicillin. Although bacterial superinfection may influence the clinical manifestations, after antibacterial treatment there was no change in the clinical aspect of the lesion.

Laboratory tests of CL include parasite detection and immunological techniques, allowing the identification of *Leishmania* species and providing important information for the prognosis

and the choice of an appropriate therapy [8,9]. Several methods may be used, but the diagnosis can be time-consuming and difficult, especially in atypical infections, because many other diseases show similar clinical pictures including syphilis, leprosy, tuberculosis, fungal infections, and tumors, among others [1].

Six laboratory tests were performed to diagnose CL. A direct parasite search (DS) was conducted in material obtained by scraping the edge of the lesion [2,10], which revealed the presence of characteristic amastigotes of *Leishmania* spp. (Fig. 2A) even after 7 months of infection.

Culture in blood base agar (BBA) supplemented with antibiotics, with negative results. After 7 days at 25 °C, the growth of contaminating bacteria prevented the growth of protozoa. Culture isolation methods are often limited and their performance depends on the species of *Leishmania*; a further complicating factor is the possibility that the culture medium may become heavily contaminated with bacteria.

An IIF test for anti-*Leishmania* IgG antibodies [11] was positive, reaching a titer of 160.

The polymerase chain reaction (PCR) with primers that amplify a 70-bp fragment from kDNA minicircles of the subgenus *Leishmania* (*Viannia*) [9,10], in using peripheral blood leukocytes and scrapings from the lesion, with positive results (Fig. 2B). The positive PCR suggests that the agent was *L.* (*V.*) *braziliensis*, the prevalent species in the region [1,10].

A Montenegro skin test (MST), which gave a positive result, with a papule 8.0 mm in diameter (Fig. 2C).

Searches for fungi in material from nodule puncture by direct microscopy (with KOH and Evans Blue) and culture (in Micosel and Sabouraud culture media) were performed, but no fungi were observed.

In Brazil, systemic therapy is indicated for infections caused by species of the *L.* (*V.*) *braziliensis* complex, to prevent the development of the mucosal form [1]. In this case, as the appearance of lesions did not improve after treatment with the antibacterial, and after confirmation of the CL, the patient started treatment with *N*-methylglutamine antimoniate (Glucantime™) 20 mg/kg by intramuscular injection for 20 days. The treatment ended in November 2011. The patient did not require hospitalization, but upon receiving the last dose of Glucantime™, reacted by fainting and required hospital treatment due to an adverse drug reaction. Several side effects are described for *N*-methylglutamine antimoniate and the need for caution in its use in patients older than 50 years cannot be understated. Although recent studies have recommended local therapy, systemic therapy is indicated in patients with multiple injuries, neck injuries, and lymphatic involvement [12].

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