

Short Communication

A Clinical Case of Disseminated Cutaneous Leishmaniasis in a Young Infant: Early Diagnosis

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Keywords

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- Infant

Abstract

Disseminated cutaneous leishmaniasis (DL) is a rare clinical form of American tegumentary leishmaniasis (ATL) caused by *Leishmania* protozoan. The incubation period is usually 2-12 weeks. It is characterized by the presence of numerous pleomorphic lesions distributed on the trunk and face. Here we report a DL clinical case in an infant with the youngest age yet described in literature for this condition. The female patient was 4-months old and had 47 lesions throughout her body. She was successfully treated with liposomal B amphotericin. The results indicate the importance of raising the suspected diagnosis of DL in infants living in urban areas.

ABBREVIATIONS

DL: Disseminated Cutaneous Leishmaniasis; ATL: American Tegumentary Leishmaniasis; PCR: Polymerase Chain Reaction; IgG: Immunoglobulin G; IFN γ : Interferon Gamma; TNF: Tumor Necrosis Factor; Th1: T helper 1 cells

INTRODUCTION

American tegumentary leishmaniasis (ATL) is caused by protozoan *Leishmania* sp. and presents distinct clinical manifestations [1-3] including disseminated cutaneous leishmaniasis (DL), which is a rare clinical form of ATL. Epidemiological and clinical studies of DL have been conducted for more than three decades in Corte de Pedra, Bahia, Brazil, where leishmaniasis is endemic. There was an increase in the prevalence of DL from 0.2% in 1986 [4] when it was first described to 2.4% of the cutaneous leishmaniasis cases diagnosed between 1988 and 2008 [2]. DL is caused by *Leishmania* species of the subgenus *Viannia* mainly *L. (V.) braziliensis*, *L. (V.) panamensis* and *L. (V.) guyanensis*. The main etiological agent in Brazil is *L. (V.) braziliensis* [3], which causes distinct clinical and immunological features. The DL pathogenesis remains unclear, but patients present a decrease in T helper 1 cell (Th1) cytokines such as interferon gamma (IFN γ) and tumor necrosis factor (TNF α), which favor the dissemination of parasites and lesions [4,5]. DL is characterized by the appearance of multiple pleomorphic

lesions (e.g., acneiform, papular, nodular and ulcerated) in the face, trunk and limbs. For a clinical case definition, the number of lesions must be higher than 10 and distributed in different parts of the body [1,6]. The patients typically have a single initial lesion usually on the extremities followed by dissemination that may involve the entire body and is sometimes associated with fever and chills. This supports the hypothesis that these manifestations are produced by the spread of *Leishmania* sp. via the bloodstream [1].

A positive Montenegro skin test and the identification of the parasites in *in vitro* culture of lesion fragments or a histopathological exam can confirm this diagnosis [1,7,8]. DL is one of the most severe clinical forms of ATL due to the number of lesions and the nasal/oral mucosal involvement that is present in ~44% of DL cases [9]. It also is associated with a high frequency of therapeutic failure [4,6,10,11].

There is an increased risk of DL according to gender (male), age (>19) and agricultural occupations [1,5]. This report describes a clinical case of DL in an infant with the youngest age yet reported in the literature [1,5,12,13] presenting a remarkable clinical manifestation of DL after a short incubation period.

CASE PRESENTATION

The subject's mother gave informed consent, and the procedures were approved by a local ethics committee (Ethics



Figure 1 (A)-Lesion features during the treatment with liposomal B amphotericin. (B)-After two months of treatment all lesions healed. (C)-After 9 months of follow up without relapses.

Committee of the Hospital de Doenças Tropicais Anuar Auad, Goiânia, Goiás, Brazil, protocol number 1/16). A 4-month old female infant had an erythematous papule in the upper right eyelid with subsequent formation of central ulceration after seven weeks. The papular lesions disseminated on the face, trunk, buttock and limbs for a total of 47 lesions. The patient was treated for impetigo without any improvement. The cutaneous involvement was initially accompanied by fever. The infant had nasal obstruction and rhinoscopy confirmed the mucosal lesions. A persistent arterial channel was diagnosed at birth. She was born in the West Central Region of Brazil and lived in an urban area close to forest. The laboratory assays revealed a positive indirect immunofluorescence (IgG antibodies at a titer of 160). The humoral immune response was present, and the Montenegro skin test was negative indicating that a cellular immune response had not developed after seven weeks. The initial investigation for primary immunodeficiency was negative. Cutaneous histopathology showed an intense chronic granulomatous inflammation with lymphocytes, plasma cells, epithelioid macrophages and giant cells. Amastigote forms were identified in macrophages. From fragments of the lesion, parasite DNA was extracted and a polymerase chain reaction (PCR) identified the parasite as *Leishmania (Viannia) spp.* All laboratory tests were performed as previously described [14]. Because the patient had previous heart disease, the treatment was liposomal B amphotericin (AmBisome; Giléad-United) at 20 mg/kg for 7 days. The therapeutic response was a success, and all lesions healed after two months of treatment with no relapse after ten months of follow-up. Figure 1 depicts the lesions and the therapeutic response.

DISCUSSION

We describe a clinical case of DL in a young infant with a short incubation period. The diagnosis was difficult because DL typically affects male adults with agricultural occupation, and the incubation period varies from 10 to 90 days [1,5,15]. Mucosal involvement and fever were noted as systemic manifestations, and these have been previously associated with DL [1,4,8]. Although the Montenegro skin test was negative, there were antibodies against the parasites. The cutaneous test was negative at the time of diagnosis and became positive only after four

months of the disease. The extraction of parasite DNA from one lesion fragment identified *Leishmania Viannia* subgenus the same result obtained by Vernal et al., in 17 cases of DL in Southeastern Brazil [13].

Treatment of DL is difficult and relapses are common after standard antimony therapy [1]. The patient received liposomal B amphotericin due to previous heart disease. The therapeutic response agreed with studies showing that this drug has higher efficacy than the pentavalent antimonials as well as shorter treatment time and fewer side effects [9-11].

It is important to notice that DL is now considered an emerging disease with an increased incidence during the last years and an expansion into urban areas [1]. This clinical case highlights the importance of diagnostic suspicion of DL even in infants.

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