

CASE REPORT

Atypical manifestations of cutaneous leishmaniasis in a boy from Afghanistan

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Abstract

Cutaneous leishmaniasis is one of the most common forms of leishmaniasis in Afghanistan, which is caused by different *Leishmania* species that are transmitted to humans by sandflies. Its clinical manifestations are very diverse and depend on a variety of parasite and host factors.

KEYWORDS

Afghanistan, atypical presentations, cutaneous leishmaniasis, *Leishmania major*

1 | INTRODUCTION

Leishmaniasis is a tropical zoonotic disease of protozoan origin that infects vertebrate hosts by the bites of *Phlebotomus* and *Lutzomyia* sandflies (depending on the geographical area).¹ Malnutrition, population movement, poor housing, a weak immune system, and a lack of economic deposits are all associated with the disease, which affects some of the poorest individuals.^{1,2} Due to the species of parasite and the site of involvement in the vertebrate host, three main clinical manifestations of this disease include cutaneous leishmaniasis (CL), mucocutaneous leishmaniasis (MCL), and visceral leishmaniasis (VL).² CL is one of the most common clinical forms of leishmaniasis throughout the world,^{1,3} which endemic in

about 90 countries from the Americas, the Mediterranean basin, the Middle East and Central Asia, with an annual global incidence of 600,000 to 1 million. According to the WHO recent report, more than 85% of CL cases were reported from Iran, Afghanistan, Pakistan, Iraq, Syria, Tunisia, Peru, Bolivia, Algeria, Colombia, and Brazil.⁴ Over the last decades, the number of cases of CL has been increasing in Afghanistan, mainly due to war and political conflicts.^{1,5} This country is one of the main global foci of CL, with an annual estimated incidence of about 200,000 cases and a 13 million population at risk.⁵ In general, *Leishmania major* (*L. major*) and *Leishmania tropica* (*L. tropica*) are the causative agents of CL in the country, which are transmitted in a zoonotic and anthroponotic cycle, respectively.^{6,7}

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Early clinical manifestations of CL include a small erythema at the site of a sandfly bite that develops into a papule and nodule over time. These lesions are injured over a period of 2–6 months. Eventually, the uncomplicated wounds will cure spontaneously even if left untreated.^{8,9} Classical CL presentation is characterized by skin ulcers, or nodules, papules, or erythematous plaques less frequently. It may, however, present with atypical or unusual manifestations such as zosteriform, erysiploid, lupoid, sporotrichoid, nodular, hyperkeratotic, and so on. Since atypical forms of CL resemble various dermal diseases; the clinical differentiation of them seems to have been difficult.³ However, leishmaniasis dermal lesions, although not fatal, have a great impact on the social life of infected people, especially women. Because it may cause a social stigma in people with severe skin lesions.^{8,9} Here, we describe an unusual CL case in a child with disseminated dermal lesions on the face, nose, and hands.

2 | CASE PRESENTATION

A 5-year-old boy presented to our clinic with an 18-month history of skin lesions on his nose, face, and hands. The patient was from Herat, western Afghanistan, where CL is hyperendemic.¹ Physical examination was remarkable for painful and boggy disseminated ulcerative and exudative lesions with overlying plentiful crusts and presenting disfiguring skin tissue accompanied by several swollen nodule (approximately 5 cm in diameter) and ulcers on the both hands and many small crusted ulcers on forehead. It's covering approximately two-thirds of face and dorsal of the hands (Figure 1). A dermal scraping specimen of the both hand and face was obtained. Next, direct smear was prepared and stained with Giemsa stain as a gold standard procedure. Then, direct stained smear showed intracellular/extracellular Leishman bodies (amastigotes) under a light microscope (see Figure 2). Moreover, using species-specific polymerase chain reaction (PCR) assay,¹⁰ *L. major* species identification was confirmed. The patient was treated with intramuscular injections of sodium stibogluconate (SSG; Pentostam, UK) in a dose of 20 mg/kg/

day for 28 days and also topical 25% zinc oxide cream and 2% Mupirocin ointment twice a day on the wound surfaces for two months later. The skin lesions cured, with huge scarring after 5 months follow-up. All facial lesions withdrew and re-epithelialization of the lesions established. This research was carried out in accordance with the principles outlined in the Helsinki Declaration. Also, CARE guidelines and methodology have been followed in this study.

3 | DISCUSSION

CL, as a major public health issue, is one of the most common types of leishmaniasis in Afghanistan.¹ CL outbreaks have been reported frequently in Afghanistan. This condition is most common in poor and underserved communities with insufficient access to healthcare. Many reasons contribute to the increased incidence, including political instability, war, lack of effective public health system, and cultural and socioeconomically obstacles.⁹

CL is usually diagnosed by looking for amastigote forms in tissue biopsies or smears under a light microscope. However, in atypical and or unusual clinical forms, due to the similarities of CL to other cutaneous diseases such as lupus vulgaris, sporotrichosis, psoriasis, atypical mycobacterial infections, and even skin cancer, this approach usually has a low sensitivity. Consequently, accurate diagnostic procedures such as PCR are required.^{2,3,10}

Although *L. tropica* is responsible for most cases of CL in Afghanistan, *L. major* is more responsible for acute, disfigured, and atypical skin lesions than *L. tropica*. In addition, the course of treatment in *L. major* infected patients is shorter than in *L. tropica* infected patients.² Thus, early and accurate identification of the causative agents of CL is important for the clinical outcome and drug susceptibility.¹¹

The first-line drug treatment with satisfactory clinical outcome for all clinical forms of CL is the use of pentavalent antimony compounds such as sodium stibogluconate (SSG; Pentostam, UK) or meglumine antimoniate (Glucantime, France). Other treatments for CL include



FIGURE 1 (A) Disseminated ulcerative and exudative lesions with overlying plentiful crusts on the face, nose, and forehead; (B) and dorsal of the hands of a boy with cutaneous leishmaniasis caused by *Leishmania major*, Herat, western Afghanistan

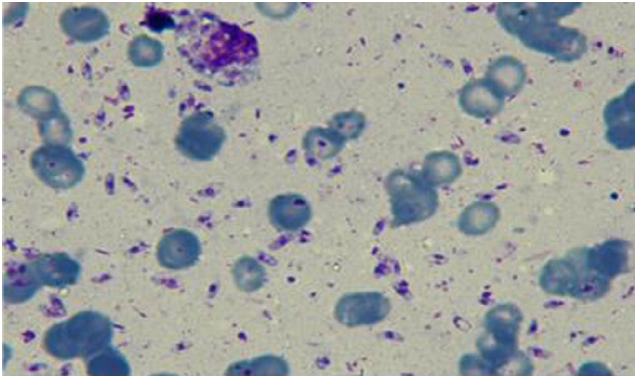


FIGURE 2 Direct Giemsa stained smear obtained from skin scraping specimen showing multiple Leishman bodies (amastigotes). (×1000 magnification)

cryotherapy and heat therapy. Liposomal Amphotericin B (Ambisome) Due to the existence of civil and foreign wars in Afghanistan, the treatment of patients with CL by the Ministry of Health is the only way to control this disease.¹²

Moreover, studies have shown that genetic diversity between *L. major* isolates is associated with different clinical manifestations.^{3,13} Therefore, the diagnosis of unusual clinical forms can be a sign of involvement with the very aggressive isolation of *L. major*.¹⁴ Thus, the clinical manifestations and development of the *Leishmania* parasite can indicate complex parasite interactions and host immune response. However, the immune system plays an important role in the clinical manifestations of leishmaniasis. However, the levels of severity of the disease manifestations depend on parasite virulence, host factors, and the immune response of the host.^{2,3} Untreated and or unusual CL lesions can mimic many other skin disorders non-specifically. In endemic areas, many of these unusual lesions caused by *Leishmania* spp., are not considered by physicians.^{2,3}

Ultimately, the outcome of the infection depends on the ability of the host macrophages to kill the *Leishmania* parasite. Louzir et al., found that high levels of IL-10, IL-12, and IFN- γ mRNA were associated with the negative clinical consequences of CL.¹⁵ As a whole, in some cases, wounds caused by CL due to the complex relationship between the parasite's genetics and the host's immune response can be very unusual and atypical, making it difficult to accurately diagnose the disease.^{2,3}

4 | CONCLUSION

CL should be considered in the differential diagnosis of patients who have lived in endemic areas or have travelled to an endemic region 1 to 6 months before the lesion appears. Although CL affects people of all ages, it is more

common in children and young people, especially in endemic areas. Thus, physicians should also consider the differential diagnosis between cases of CL with atypical lesions and some common and uncommon infectious skin diseases in order to diagnose and treat cases in a timely manner as well as to avoid complications and or disseminated wounds.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

MF and SAR involved in interpretation and collection of data. ESB and MS involved in writing and editing of the manuscript. MF and MS involved in editing and preparing the final version of manuscript. All authors reviewed the paper and approved the final version of the manuscript.

ETHICAL APPROVAL

The study was approved by our local ethics committee.

CONSENT

Written informed consent was obtained from the patient's parents for publication of this report.

DATA AVAILABILITY STATEMENT

The data are available with the correspondence author and can be achieved on request.

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