A case of hyperkeratotic cutaneous leishmaniasis on the hallux: an unusual presentation of American tegumentary leishmaniasis

Marcus H. de S.B. Xavier[a], Sabha Mushtaq[b], Luiz A.B. Pôrto[a], Ana C.R. Fonseca[a], Moisés S. Pedrosa[a]

[a]Hospital Foundation of the State of Minas Gerais, FHEMIG, Belo Horizonte, MG, Brazil, [b]Department of Dermatology, Jawaharlal Nehru Medical College (JNMC), Aligarh, Uttar Pradesh, India

Correspondence to Sabha Mushtaq, MD, Department of Dermatology, Jawaharlal Nehru Medical College (JNMC), Aligarh, Uttar Pradesh 202001, India. Tel: +91-9796444136; e-mail: smqazi.gmc@live.in

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Cutaneous leishmaniasis (CL) is endemic in various parts of Brazil and is known for its clinical pleiomorphism in humans. Beyond the classical picture, CL may appear at unusual sites or with atypical morphologies which may elude the diagnosis at first instance. Herein, we report the case of a 48-year-old Brazilian male, who presented with a 2-year history of persistent hyperkeratotic plaque on the left hallux, which turned out to be cutaneous leishmaniasis. The wide variety of cutaneous findings and unusual presentations of CL often pose a diagnostic challenge especially for inexperienced physicians. The objective of this case report is to emphasize on the unpredictability of clinical expressions of CL and to apprise the clinicians of its masquerading potential.

Keywords:
cutaneous leishmaniasis, hyperkeratotic, meglumine antimoniate, mucocutaneous leishmaniasis, new world leishmaniasis

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Introduction

Cutaneous leishmaniasis (CL) is considered an important public health issue, causing considerable morbidity in many parts of the world including Brazil [1,2]. According to the WHO's estimates, nearly 1.5 million cases of CL occur every year in 98 countries and territories on five continents [3]. CL is widely distributed, with approximately one-third of cases occurring in each of the three epidemiological regions: the Americas, the Mediterranean basin, and Western Asia [4]. A recent rise in cases of CL in the United States has been noted, and it is attributable to international travel to and from endemic regions, whether by immigrants, refugees, tourists, or soldiers [4,5].

Although the typical cases of CL are easy to diagnose in endemic regions, the unusual forms may dodge even seasoned physicians. In the recent past, there has been an increase in reporting of atypical/unusual clinical presentations of CL, both in Old and the New World [6]. Dermatologists, therefore, should be aware of the clinical manifestations of CL, as its clinicopathological picture is variable, often making diagnosis difficult besides being a condition sometimes forgotten by physicians.

Case report

A 48-year-old male from a rural settlement to the North of Minas Gerais state of Brazil presented with a 2-year history of an asymptomatic plaque over his left big toe. It began as an erythematous papule which gradually developed into a plaque over a period of 6 months. There was no history of obvious trauma at the site. Since 1 month, the patient had noticed thick crust covering the plaque and a small ulceration at one place. The patient had consulted several family physicians and dermatologists without a definitive diagnosis. He had taken multiple courses of antibiotics without any improvement. His medical history was otherwise unremarkable.

Cutaneous examination revealed a well-defined plaque measuring 3 cm×2.5 cm over dorsum of left hallux. The margin of the plaque had a violaceous hue and the surface was hyperkeratotic, covered with thick adherent yellowish-gray crust. A small ulcer was seen at the anterior end of the plaque (Fig. 1). However, there was no active ooze or discharge. There was no regional lymphadenopathy. The differential diagnosis included tuberculosis verrucosa cutis, cutaneous leishmaniasis and fixed cutaneous sporotrichosis.

Biopsy specimens were obtained for histological examination and culture (mycobacterial and fungal). Histopathology revealed pseudoepitheliomatous hyperplasia with the dermis showing nodular infiltrate of histiocytes, lymphocytes, and tuberculoid-type

histiocytic granulomas. Leishman-Donovan bodies in the cytoplasm of histiocytes could be demonstrated on Giemsa stain, thus confirming the diagnosis of CL (Fig. 2a–c). The mycobacterial and fungal culture was negative. The results of chest radiograph, blood chemistry, blood count, and urinalysis were within normal limits.

The patient was given intralesional meglumine antimoniate administered fortnightly for 8 weeks. At a 3-month follow-up, the lesion had completely healed.

Discussion

Leishmaniasis is a diverse vector borne disease caused by different species of protozoa *Leishmania*. It is transmitted to humans by the bite of a tiny (2–3 mm long) infected sand-fly (*Phlebotomus* and *Lutzomyia*) [3]. As per geographic categorization, leishmaniasis is divided into (a) Old World leishmaniasis (transmitted by vector Phlebotomus and caused by *Leishmania* species found in Africa, Asia, the Middle East, the Mediterranean, and India), which produces cutaneous or visceral disease and (b) New world leishmaniasis (transmitted by vector Lutzomyia and caused by *Leishmania* species found in Central and South America), which produces cutaneous, mucocutaneous, and visceral disease [3,6].

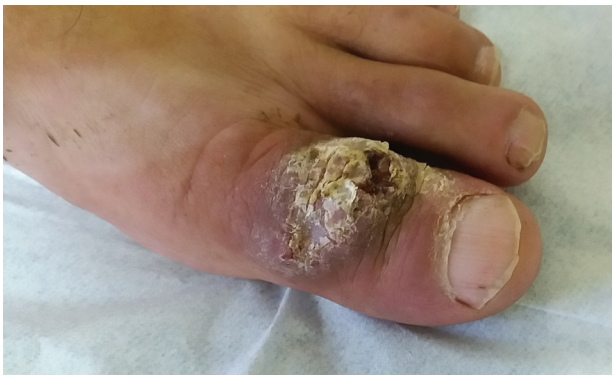
The main *Leishmania* species causing New World CL include *Leishmania mexicana* and *Leishmania (Viannia) braziliensis* species complex. In Brazil, the disease is also known as American tegumentary leishmaniasis (ATL) caused by *L.* (*V.*) *braziliensis*, *Leishmania* (*V.*) *guyanensis*, and *Leishmania* (*Leishmania*) *amazonensis* and is widely distributed in endemic regions of northeast Brazil [2,3,7].

CL is a major public health problem in many countries with 90% of all cases occurring in seven countries including Brazil, where every year ~28 000 cases of CL are reported [7]. The disease is endemic in whole of Brazil, and the major clinical forms include localized or disseminated cutaneous and mucocutaneous disease [5]. Localized CL is most frequently caused by *L. guyanensis* and *L. braziliensis* in Brazil. The disseminated cutaneous and mucocutaneous disease are most frequently caused by *L. braziliens*is [4]. Our patient belonged to Minas Gerais state of Brazil where ATL is endemic, and most cases are of local CL as per the official record [7].

A 'classical' lesion of ATL originates as a macule over the exposed area at the site of inoculation followed by a papule that ulcerates and expands to a typical round-to-oval crateriform lesion or evolves as a nodular lesion [3]. The clinical manifestations of CL are variable and depend not only on the causative *Leishmania* species but also on endemic region, host factors and immunoinflammatory responses [6].

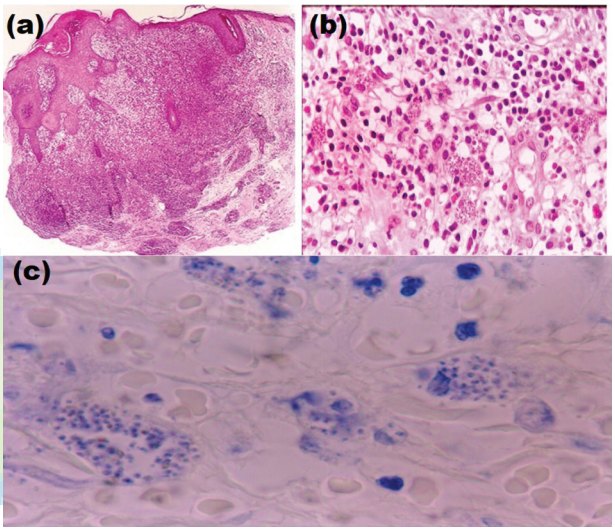
A myriad of atypical presentations of localized CL have been reported over the years: lupoid, eczematous, erysipeloid, verrucous, zosteriform, paronychial, sporotrichoid, chancriform, psoriasiform, and annular [6]. However, there are only two reports of hyperkeratotic form of CL in the literature, and both are from Turkey. Yesilova *et al.* [8] and Gülüm *et al.* [9] reported CL presenting as hyperkeratotic plaque on the nose and penis, respectively. Both these cases had a long standing history similar to the present case.

Figure 1



Hyperkeratotic plaque on the dorsum of left big toe.

Figure 2



(a) Pseudoepitheliomatous hyperplasia with dense lymphohistiocytic infiltrate in the dermis (hematoxylin and eosin, ×40). (b) Higher power showing histiocytes and multiple Leishman-Donovan bodies (hematoxylin and eosin, ×100). (c) Few histiocytes with multiple Leishman-Donovan bodies (Giemsa, ×400).

The hyperkeratotic form of CL is reported in Old World CL though sporadically [8,9], but to the best of our literature search, we could not find any report of the same in the New World.

The most common diagnostic method for CL is microscopic examination, and the causative *Leishmania* species can be identified by PCR [7]. In the present case, species identification could not be performed owing to limitation of resources.

The diagnosis of CL is quite easy in typical cases found in the endemic areas. However, the lesions may appear at unusual sites or with atypical morphologies imitating various infectious, inflammatory, and neoplastic conditions, which may lead to delay in diagnosis and mismanagement [6]. A high index of suspicion in such cases is crucial in arriving at the diagnosis. CL is a self-limiting disease, but treatment may be sought for esthetic reasons, delay in spontaneous healing and disfiguring scar. A battery of treatment options are available, but pentavalent antimonials remain the first line of treatment [3,10].

In our patient, the relatively uncommon site, the atypical clinical presentation and absence of medical suspicion eluded the diagnosis for 2 years. As our patient belonged to an endemic area and owing to the painless nature of the lesion, CL was kept as one of the differential diagnosis, which was confirmed by histopathology.

Conclusion

CL with its protean manifestations may mimic a variety of diseases posing a diagnostic challenge. The delay in diagnosis may not only lead to mismanagement but also financial burden on the patient. The present case report highlights the unusual morphological presentations of CL and emphasizes keeping a high index of suspicion for timely diagnosis and appropriate treatment.

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Conflicts of interest

There are no conflicts of interest.

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