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Case Report

# Unusual presentation of cutaneous Leishmaniasis in pregnancy: A case report

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ABSTRACT

Cutaneous Leishmaniasis is a parasitic infection characterized by significant clinical variability. Unusual and atypical clinical aspects of infection have been reported in immunodeficient patients or associated with particular parasite species. We report a pregnant woman with unusual presentation of cutaneous Leishmaniasis.

**Key words:** Leishmaniasis; Skin, Pregnancy

## INTRODUCTION

Worldwide, Leishmaniasis affects 112 million people in 88 countries, with a yearly incidence of 2 million cases [1]. The majority of these cases are cutaneous Leishmaniasis (CL), which is most common in adolescents and young adults from extremely poor rural areas [2,3].

Cutaneous Leishmaniasis (CL) is caused by a parasite from the genus Leishmania infection, and is transmitted to humans by the bite of female sand flies [4].

The clinical features of cutaneous Leishmaniasis (CL) may vary in terms of type and extension, ranging from single, chronic ulcerative lesions to disseminated nodular ones; however, several unusual and atypical clinical features of the disease have been reported in the literature [5-7].

Pregnancy is associated with an altered of human cell-mediated immuneresponse and an increased susceptibility to many infectious agents with atypical cutaneous presentation. Herein, we present a case of a particular clinical aspect of CT on the leg of a pregnant patient. The hypothesis of a possible role of pregnancy in this particular clinical presentation shall be discussed.

## CASE REPORT

A 34-years-old woman,who was five months pregnant, lived in an area endemic for Cutaneous Leishmaniasis in northern Morocco; she was referred to our department because of a painful skin lesion that had been progressing for six months at the front of her right leg.

Dermatological examination showed that Tumoral and cauliflower-like lesion measured 10 cm long axis, adhered to deep planes and painful on palpation in the centerin the right leg (Fig. 1). It was thought to be Sarcoma, amelanotic melanoma, squamous cell carcinoma. The rest of the physical examination was unremarkable.

Histopathological examination of the biopsy obtained specimens demonstrated ulcerative changes and irregular acanthosis in the epidermis, namely, pseudoepitheliomatous hyperplasia. In dermis infiltration of lymphocyte, plasma cell and histiocyte with multinucleated Giant cell were shown. Leishman bodies were seen in (Fig. 2). With regards to the clinical and histopathological findings, the diagnosis of CL was made.

Complete biological tests were normal and HIV serology was negative for eliminating an immunosuppression.

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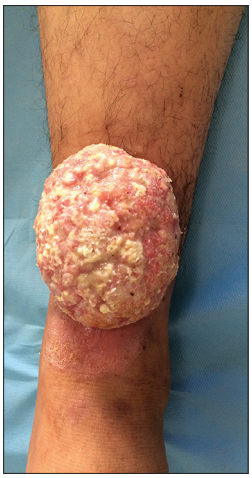

**Figure 1:** Cauliflower-like lesion of the right leg.

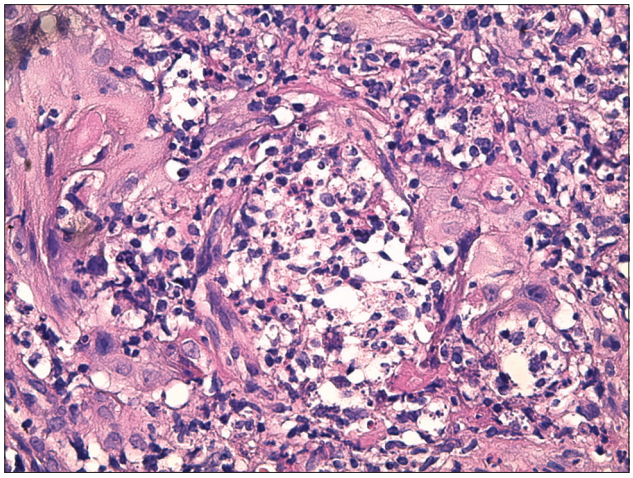

**Figure 2:** Leishman bodies.

The treatment consisted of surgical excision to relieve the patient because the pentavalent antimony is potentially abortogenic.

## DISCUSSION

The term "Leishmaniasis" defines a group of vector-borne diseases caused by species of the genus Leishmania and characterized by a spectrum of clinical manifestations.

Parasite properties (infectivity, pathogenicity, and virulence), host factors, and host responses regulate heterogeneous disease expression [7].

The most common clinical form is the classical ulcer, with an indurate raised outer border and sharply incised central crater that usually self-heals over a period of months. The usual clinical presentations of Leishmaniasis are easily diagnosed by clinicians in endemic regions, but unusual forms may give rise to difficulties in the diagnosis and appropriate treatments. Cutaneous Leishmaniasis can produce a large variety of atypical and rare forms. There has been an increase in the number of papers reporting unusual clinical presentations, both for the Old and the New World [8-14].

In our Moroccan experience atypical and unusual clinical aspects of LC are observed especially with *Leishmaniatropica* [15].

This raises the question of the possible involvement of this species in our patient. For technical reasons, we did not perform the identification of the responsible strain.

Another factor that could be responsible for these particular clinical aspects would be pregnancy as in our case.

In contrast to the typical presentation of a well-demarcated ulcer with raised borders, CL during pregnancy is characterized by larger lesions with a highly atypical, exophytic appearance. As in our case, which raised concerns for other diseases, such as tuberculosis, chromomycosis or neoplasms? [3].

In a C57BL/6 mouse *L. major* model, larger CL lesions occurred during pregnancy, which correlated with decreased Th1 cytokine production [16]. The human cell-mediated immune response is altered during pregnancy [17], with an overcomepensation immediately after delivery.

The study of Conceição-Silva et al showed transient modulation of maternal immune responses during pregnancy as indicated by exacerbated cutaneous lesions, increased parasite burdens, and decreased levels of IFN-γ and NOS2 [18].

All hypotheses about the influence of pregnancy on the expression and evolution of CL converge to a possible decrease in the quality of the T-cell response against antigens of leishmaniasis during pregnancy. This immunodeficiency, possibly associated with other factors inherent in the host and parasite, could contribute to a change in the lesion appearance.

Treatment of pregnant CL patients is a debated issue [19-21], since there is no description of congenital infection, and many antileishmanial drugs, such as pentavalent antimony or miltefosine, are teratogenic [21,22]. Consequently, alternative therapies

should be evaluated in order to warrant safety and efficacy in this group of patients. Guimarães et al [23] showed that 40% of patients with atypical manifestation of CL were pregnant women and suggested that Amphotericin B should be considered as the drug of choice for all patients diagnosed with atypical CL. Intralesional treatment with meglumineantimoniate was successful in over 83% of patients treated who had contraindication to systemic therapy [24], but there is a lack of evidence demonstrating safety in pregnant women. As spontaneous healing has been reported to occur after delivery [25], several groups avoid the use of specific treatments and follow the patients by using local heating and/or antibiotic ointments to control lesion development and secondary infections. In our case surgery gave good results.

## CONCLUSION

CL during pregnancy is characterized by larger lesions with a highly atypical exophytic appearance. No therapy is known to cure the disease during pregnancy, although postpartum cure has been found to be complete. CL during pregnancy has a notably different clinical presentation. It is important for physicians who are caring for patients in regions where the disease is endemic to recognize this presentation.

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