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Case Report

Erythematous Annular Cicatricial Plaques on the Face

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Abstract

Background: Lupoid leishmaniasis (LL) is an unusual chronic form of cutaneous leishmaniasis with clinical and histopathological features resembling lupus vulgaris. It is estimated that lupoid leishmaniasis represents approximately 0.5-6 % of all cutaneous leishmaniasis. In this report, a case of lupoid leishmaniasis that has been misdiagnosed as "lupus vulgaris" and then has been subsequently treated with anti-tuberculoid drugs for a substantial period is described.

Introduction

Cutaneous leishmaniasis (CL), a parasitic skin disease transmitted by sandflies, is characterized by a variety of clinical presentations with acute and chronic forms. Oriental sore is the most frequent acute cutaneous form, while leishmaniasis recidivans, lupoid leishmaniasis, diffuse cutaneous forms are the types of chronic form following simple acute cutaneous leishmaniasis [**1, 2, 3, 4**].

Lupoid leishmaniasis (LL) is the most frequent atypical presentation of CL and represents 0.5-6.2 % of all cutaneous leishmaniasis [**3, 4, 5**]. LL usually follows acute cutaneous leishmaniasis and initiates as a small painless papule or plaque, and then enlarges centrifugally with an active border leaving a scar resembling lupus vulgaris. *Leishmania tropica* is the most common causative agent in the majority of cases [**5**].

Case Report

A 51 year-old man was admitted with a 50-year history of cicatricial plaques over the face. The lesion had initiated as a small, asymptomatic red papule on the preauricular region when he was one-year-old. Slow peripheral growth in size and dissemination had been observed in the following years. The medical history of the patient revealed the repetitive skin biopsies that had shown granulomatous reaction in the histopathology and repeated antituberculoid therapies for four times with the diagnosis of "lupus vulgaris". On dermatologic examination, a number of coalesced erythematous or cicatricial annular plaques extending from the right temporal region to the right cheek and to the left side of the nose with active spreading borders were observed (**Figure 1**). The histopathology revealed non-caseating granulomas composed of epithelioid histiocytes and Langhans type multinucleated cells and some macrophages filled with intracytoplasmic corpuscles (**Figure 2**). *Ziehl–Neelsen* and D-PAS stains were negative for mycobacteria and fungi respectively. The routine laboratory tests revealed eosinophilia (20%) in the

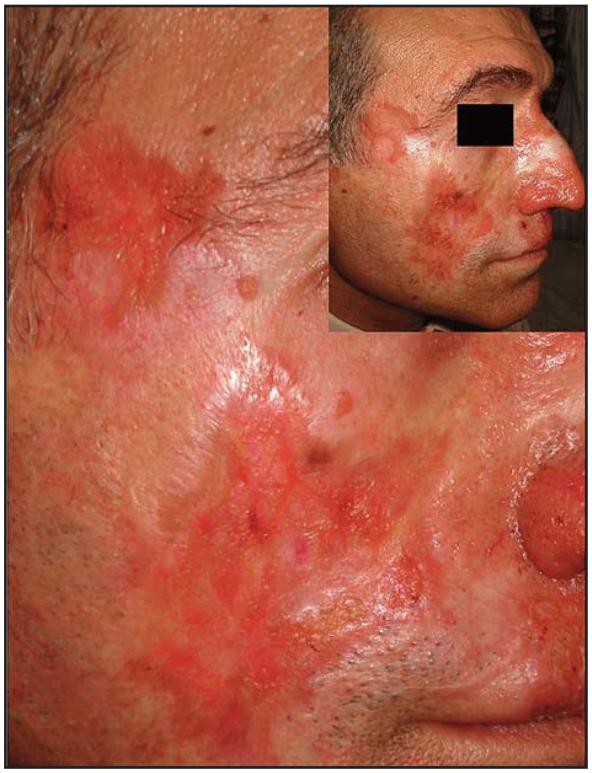

Figure 1. Coalesced erythematous or cicatricial plaques extending from the right temporal region to the right cheek and to the left side of the nose.

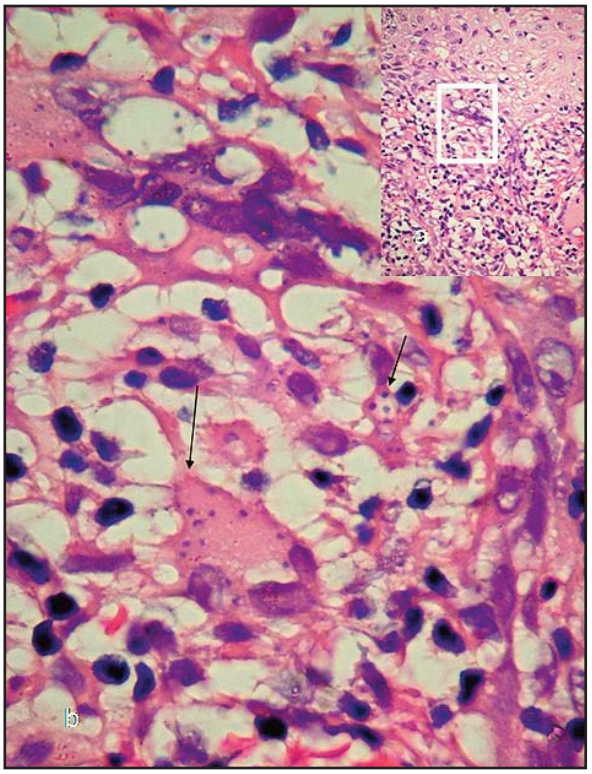

Figure 2. Non-caseating granulomas composed of epithelioid histiocytes (inset: H&E, original magnification x 20); *Langhans* type multinucleated cells and macrophages filled with intracytoplasmic corpuscles (H&E, original magnification x 100).

complete blood count. Direct microscopy of Giemsa stained smears, NNN culture and polymerase chain reaction (with 13A and 13B primers) from samples obtained by needle aspiration were negative. Serological examination of the antibodies against to the Leishmania by indirect fluorescein antibody test (IFAT) (1:1024 titers) and rapid test (K39 antigen) were positive. There was no visceral involvement. Based on the clinic, laboratory, and histopathology the patient was diagnosed as "lupoid leishmaniasis". Treatment with systemic meglumine antimonate (MA) (Glucantime®) 20 mg/ kg/ day intramuscularly for 40 days and intralesional injection of MA twice a week for two months were given complete improvement of the lesions. The treatment was tolerated well without any side effects and no recurrence was observed during a two-year follow-up.

Discussion

Diagnostic methods of cutaneous leishmaniasis include Giemsa-stained smears, culture, histopathology, serology and PCR. However, they may fail to demonstrate the organisms in chronic forms as the number of organisms drops sharply [6]. Serological tests may help to establish the diagnosis especially in the suspected cases as in this case.

Histological features of lupoid leishmaniasis also resemble lupus vulgaris including tuberculoid granulomatous inflammation. Amastigotes of leishmaniasis are frequently absent on microscopy [6]. Thus, the differential diagnosis of lupoid leishmaniasis from lupus vulgaris may be difficult resulting in a delay of diagnosis.

The reason for the delay of diagnosis in this case may be the lupoid presentation over the face and probably overlooked examination of the parasite in previous histopathologic evaluations. In this case, the positive serology supported the diagnosis and a careful histopathologic examination demonstrated leishmania amastigotes.

Treatment options include cryotherapy, topical antimonial compounds and intralesional pentavalent antimony. The pentavalent antimony derivatives and meglumine antimoniate are the first line drugs with a well-established efficacy in the treatment of leishmaniasis [6].

In this case, combined intramuscular and intralesional injections of meglumine antimoniate resulted in complete healing. In conclusion, CL should be considered in differential diagnosis of prolonged granulomatous facial lesions.

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