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Case Report

CASE REPORT

A successful treatment of severe lupoid cutaneous leishmaniasis in an elderly man: a case report

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Abstract

Lupoid cutaneous leishmaniasis (LCL) is a rare, atypical presentation of cutaneous leishmaniasis (CL). In this report, the authors present the case of a severe LCL in an elderly patient who presented to our dermatology department with severe, painful ulcerated lesion on his midface with cosmetic deformity to his nose. He had a history of CL 3 years ago at the same place. Histopathologic examination showed epidermal and dermal changes with chronic inflammatory infiltrate and no leishmaniasis bodies were detected. He was admitted and treated with systemic glucantime (60 mg/kg) for a month followed by hydroxychloroquine (200 mg twice a day) for another month with favorable outcome. Countries with endemic CL should consider LCL in patients with a history of leishmaniasis and a similar clinical presentation, especially that it could be misdiagnosed with other granulomatous cutaneous conditions, thus leading to cosmetic deformities that can be avoided with early adequate treatment.

INTRODUCTION

Cutaneous Leishmaniasis (CL) is an insect-borne disease transmitted by the female sand fly. In this report, we describe the case of an elderly man with lupoid cutaneous leishmaniasis (LCL), which is a rare type of CL.

CASE REPORT

A 52-year-old man was referred to our dermatology department with painful, coalescent, erythematous, papulo-infiltrative, nodular plaques on his midface. He has a history of uncontrolled diabetes mellitus Type II and a history of CL that started 3 years ago as a slowly growing papule on his nose that evolved to a nodule. Direct smear was positive for leishmaniasis bodies. The patient was treated with systemic meglumine antimoniate (glucantime) for 20 days; the lesion was healed with a scar. Two years later, the patient had a recurrence of the lesion at the border of the scar; he sought folk medicine that resulted in severe ulcers and dissemination of the lesion, which led to deformity of the nose. Results of dermatologic examination revealed extensive erythematous papules and nodular plaques on his nose, upper lip and cheeks with telangiectasia on his cheeks and severe ulcers and scarring on the nose and upper lip (Fig. 1). The other parts of his face were spared. There was no evidence of systemic involvement. The results of the routine lab tests, including complete blood count (CBC) and serum biochemistry are listed in (Table 1). Smear was not made since the patient is known to have a history of leishmaniasis. Histological examination showed changes in the dermis with epidermal atrophy. There was a mixed inflammatory infiltrate composed of lymphocytes, histiocytes, plasma cells and multinucleated cells of Langhans (Fig. 2). Neither parasites within dermal macrophages (Fig. 3), nor tuberculoid granulomas with caseation necrosis were detectable. Diagnosis of LCL was established based on the clinical aspect, facial localization,

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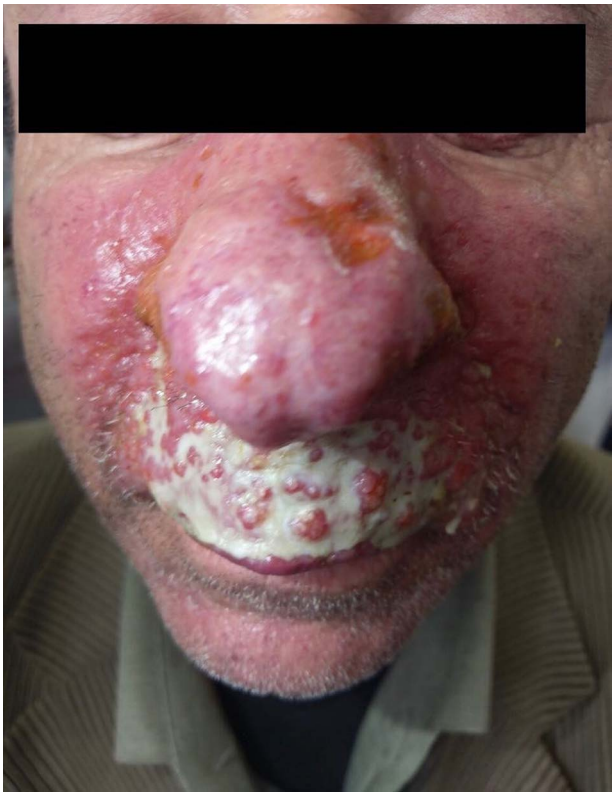

Figure 1: Infiltrative nodular plaques and ulcers on cheeks, upper lip and nose.

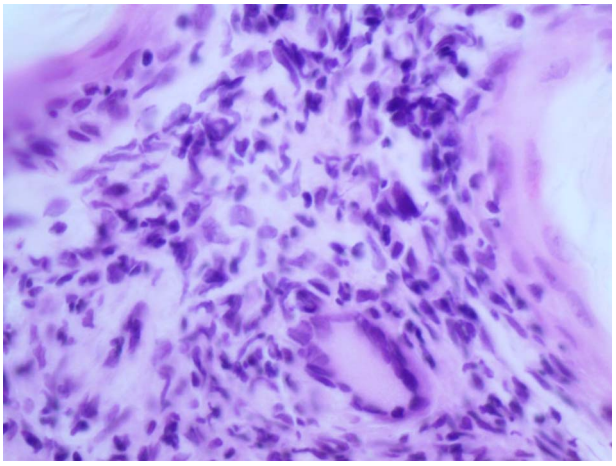

Figure 2: Histopathologic examination shows a granulomatous infiltrate with a multinucleated cell of Langhans in the center (hematoxylin and eosin).

chronic evolution, the history of CL and histopathologic findings even with the absence of intra-amastigote in the biopsy. Patient was admitted in the hospital and was started on systemic meglumine antimoniate (60 mg/kg/day) divided into two doses for 20 days in addition to ceftriaxone (500 mg twice a day) with application of topical cream diprogenta (betamethasone dipropionate and gentamicin sulfate). During the treatment, the patient was monitored daily for any clinical or laboratory signs of chemical pancreatitis, liver function tests, CBC in addition to a daily electrocardiography (ECG) test. On Day 10, the ECG showed a slight prolongation of QTc (QTc = 459 msec) for only 1 day. No other significant side effects were observed. At the end of the Day 20, improvement was noted and the patient was discharged and started on hydroxychloroquine (plaquenil) (200 mg twice a day) for a month. At the end of the month, he came for a follow-up and the lesion was completely healed (Fig. 4).

DISCUSSION

CL is one of different types of leishmaniasis that are caused by morphologically indistinguishable protozoa of the family Trypanosomatidae, called *Leishmania*. They have different clinical features and geographical distribution. According to the

Table 1: Results of Complete Blood Count and Serum Biochemistry.

Parameter	Result	Unit	Normal range
WBC	16.5	10^3/µl	3.5:10.0
GRAN	12.8	10^3/µl	1.2:8.0
RBC	5.90	10^3/µl	3.50:5.50
HGB	16.8	10^3/µl	11.5:16.5
MCV	80.1	fl	75.0:100.0
PLT	263	10^3/µl	120:400
GLU	340	mg/dl	70–105
CREA	0.88	mg/dl	0.90–1.30
UREA	35	mg/dl	5.0–45.0
URIC ACID	2.9	mg/dl	3.5–7.2
AST	25	U/l	0–40
ALT	17	U/l	0–40
AMYLASE	37	U/l	20.0–80.0
CA	7.9	mg/dl	8.6–10.3
CRP	7.53	mg/dl	
INR	1		0.8–1.1
PT	12.7	sec	10–14
CHOL	208	mg/dl	80–200

WBC, white blood cell; GRAN, granulocyte; RBC, red blood cell; HGB, hemoglobin B; MCV, mean corpuscular volume; PLT, platelet; GLU, glucose; CREA, creatinine; AST, aspartate aminotransferase; ALT, alanine aminotransferase; CA, carbohydrate antigen; CRP, C-reactive protein; INR, international normalized ratio; PT, prothrombin time; CHOL, cholesterol.

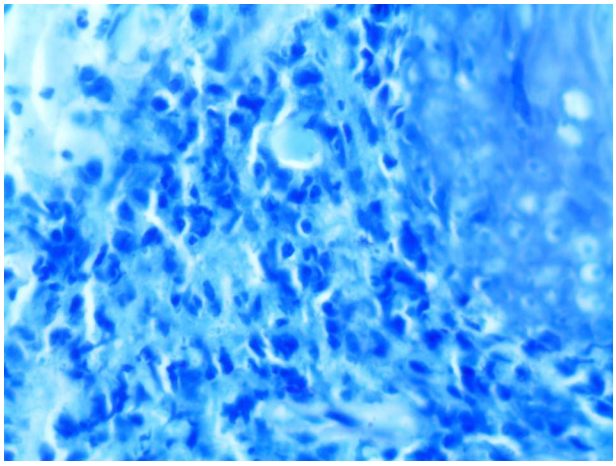

Figure 3: Giemsa staining showed no parasites within dermal macrophages.

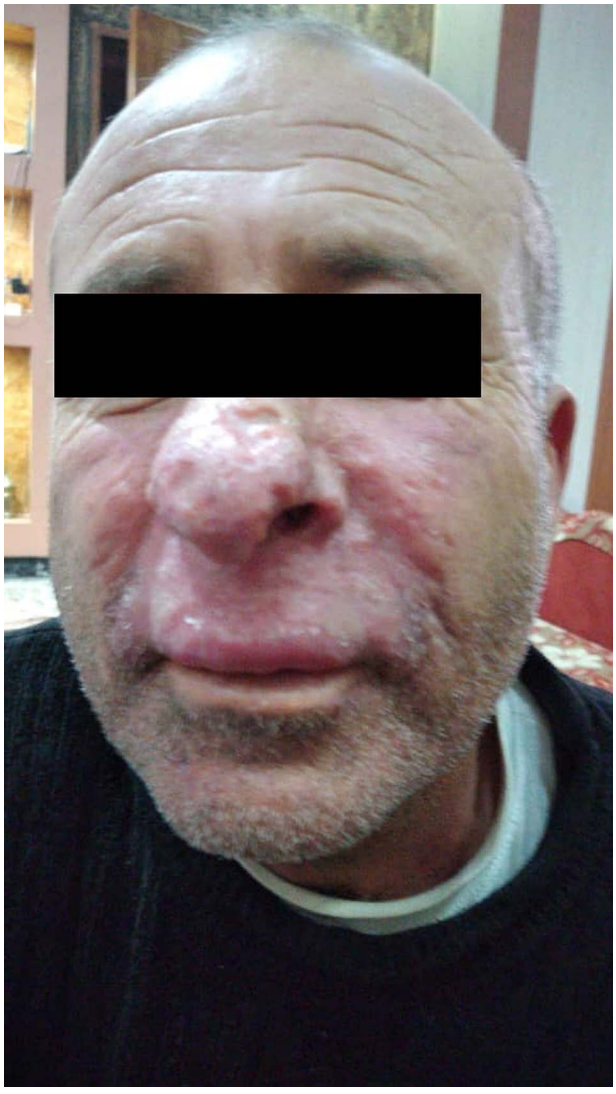

Figure 4: Patient at follow-up.

World Health Organization, old world leishmaniasis is one of the most prevalent insect-borne diseases in Eastern Mediterranean Region [1]. The most common form in Syria is anthroponotic CL due to *Leishmania tropica*, which is transmitted between humans by *Phlebotomus sergenti* sand fly [2]. During the Syrian war (2011–present), the number and distribution of CL have evolved dramatically. According to the Ministry of Health in Syria, the number of CL cases was 42 221 in 2010 and increased to reach up to 82 275 in 2018, with incidence rate 20.08 cases per 10 000 in 2010 that doubled in 2018 to reach up to 44.99 cases per 10 000 in 2018 [2]. These dramatic changes can be attributed to the mass-scale displacement of populations within Syria and to other countries, bad environmental conditions and obstacles affronted by the Ministry of Health and Agriculture to control vector and reservoir in many areas, and the impeded access to health facilities [2]. Usually CL affects unclothed areas of the body. The primary lesion is a slowly growing, painless, red papule that enlarges in a few months to a plaque or nodule that may form an ulcer that is well circumscribed with a violaceous border. The painful lesion in our patient is due to secondary bacterial infection. LCL is a chronic condition that typically follows an acute CL lesion; after the initial lesion is healed, a few red papules may appear at the border of it, covered with whitish scales; these papules spread peripherally on an erythematous base [3]. The lupoid type is also known as 'leishmaniasis recidivans' or 'relapsing leishmaniasis'; however, according to Oliveira-Neto *et al.* [4], LCL is the initial clinical presentation, whereas CL is the recurrent lesion. Spontaneous healing in acute leishmaniasis usually takes place within 12–18 months. While in CL it may last up to 20–40 years without treatment [3]. Usually LCL occurs most commonly with the urban type of disease caused by *L. tropica*, which is the most prevalent type in Syria, and LCL accounts for ~4–10% of *L. tropica* in the old world [5]. There are many different differential diagnoses for LCL including cutaneous tuberculosis, rosacea granulomatosa, lupus pernio, vegetate type and squamous cell carcinoma vegetant. The history of CL and the development of the lesion, with the clinical and histopathologic aspects, rule out other diagnoses. The histopathologic changes are seen in epidermis and dermis. The epidermal changes are variable; meanwhile, the dermis exhibits chronic inflammatory infiltration and non-caseating granulomatous inflammation. In chronic lesions, it is difficult to find any parasite on histopathologic examination [3, 6]. There are no guidelines or systematic reviews for the treatment of atypical CL [7]. However, pentavalent antimonials remain as first-line treatment for all forms of leishmaniases even for atypical forms. In Syria, a slightly different pentavalent antimoniate, meglumine antimoniate, is used. In literature, we could find many therapeutic options for leishmaniasis like fluconazole, amphotericin B, allopurinol and others with variant outcomes [8]. We could also find many randomized controlled trials discussing the effect of hydroxychloroquine on CL with excellent results [9]. To our knowledge, we are the first to recommend the use of hydroxychloroquine after meglumine antimoniate in the treatment of LCL.

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Conflict of interest statement. None declared.

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ETHICAL APPROVAL

No formal ethical approval is needed for this work.

CONSENT

A consent form to submit this case was completed and signed by the patient.

GUARANTOR

H.S.

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