Case Report: A Case of Erysipeloid Cutaneous Leishmaniasis: Atypical and Unusual Clinical Variant

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Abstract. Cutaneous leishmaniasis has a number of morphologic variants such as acute paronychial, chancriform, annular, palmoplantar, zosteriform, and eczematoid, which depend on the immune condition of the patients, the subspecies of the Leishmania, and the area of the localization. In recent times, the number of reports of new and rare variants of cutaneous leishmaniasis has been increasing. In this report, we describe a very rare variant of Cutaneous leishmaniasis, presented with ulceration on glabellar region and infiltrative erythematous lesions covering the center of the face, resembling erysipelas.

INTRODUCTION

Cutaneous leishmaniasis (CL) is a parasitic infection encountered in daily dermatologic practice. In our country (Turkey) the majority of the cases are of dry type caused by Leishmania minor. Infected patients and dogs are the main reservoirs of disease. Phlebotomus papatasi and Phlebotomus sergenti are the main vectors of transmission.1,2 Leishmaniasis is endemic in 88 countries, with a total of 350 million people at risk. The incidence of leishmaniasis is increasing on a global level due to population and environmental changes.3 Depending on the type of parasite and the immune response of the patient, Leishmania species may cause a wide spectrum of disease including cutaneous (acute, chronic, recidiva, and disseminated anergic forms) and visceral forms with prevalence of over 12 million people worldwide and 1.5 to 2 million new cases each year.3–5

Cutaneous leishmaniasis has many different clinical presentations. Recently, there has been an increase in the number of reports for new and rare variants of CL.5–8

A rare and unusual presentation of CL is the erysipeloid type, which has been reported from Iran, Pakistan, and Turkey.6,9,10 This clinical form is not only unusual in its clinical features, but also in the specific category of patients whom it seems to afflict.11 We described a 73-year-old woman who has erysipeloid type CL presented with ulceration on glabellar region and infiltrative erythematous lesions covering the center of the face, resembling erysipelas in a nonendemic area of Turkey.

CASE REPORT

A 73-year-old woman was admitted to our clinic for ulceration on the forehead and erythematous plaque on the face. As it was concluded from her history, the initial lesion started in the form of an asymptomatic, painless papule, 0.5–1 cm in diameter on the glabellar region, which slowly coalesced and formed a red infiltrative plaque. Two months after the initial papule occurred, an ulceration developed on the lesion. Erythematous lesion was reported to have enlarged and spread on her nose, cheeks, and forehead despite local and oral treatment with numerous antibiotics in the past 3 months.

There was no history of trauma or insect bite. She had no significant past medical history. She denied having fever, chill, night sweating, weight loss, facial trauma, or traveling to an endemic area for CL.

Upon physical examination, a sharply defined erythematous indurated plaque with a central crusted ulcer on glabella, spreading to forehead, upper portion of the nose, and both cheeks was observed (Figure 1). No other systemic abnormalities were detected.

Total blood count and erythrocyte sedimentation rate of the patient were within normal limits. Routine biochemical tests, urine analysis, chest radiography, and intradermal purified protein derivative (PPD) skin test were normal. Culture of the biopsy specimen and Gram smear prepared from the

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FIGURE 1. Erythematous and ulcerated lesions on the face before treatment. This figure appears in color at www.ajtmh.org.
lesion were negative for bacteria, fungi, and mycobacteria, whereas a number of amastigotes belonging to Leishmania were determined in the intracellular and extracellular area (Figure 2) with Giemsa stain. Biopsy of the primary lesion showed a dense mixed inflammatory infiltrate in the upper dermis comprising lymphocytes and neutrophils. However, no organisms were identified.

Considering asymptomatic nature of the lesion and unresponsiveness to previous treatments, CL was clinically suspected. Skin smears and/or skin biopsy also revealed the diagnosis of CL and the patient was treated intramuscularly with 10 mg/kg/day systemic meglumine antimoniate, divided into 2 doses. After receiving a total of 20 days of systemic meglumine antimoniate, ulcerated lesion flattened and improved markedly. Four weeks after the end of treatment, only slightly erythematous skin was observed in the place of the ulcerated lesion (Figure 3).

DISCUSSION

In the Southeast region of Turkey, CL has been an endemic and major health problem for a long time and the incidence has been rising since the 1980s.12

In the course of typical CL, lesions start as small erythematous papules, which gradually enlarge to 1–2 cm in diameter in about 6 months and then ulcerate. These ulcers are painless with necrotic base and indurated margin and are frequently covered by a firmly adherent crust. Approximately 85% of skin lesions locate on the exposed body sites such as head, neck, arms, wrists, and hands.1,12

Cutaneous leishmaniasis shows a variety of morphologic patterns depending on the immune status of the host and subspecies of Leishmania. Atypical forms of CL such as psoriasisform, paronychial, sporotrichoid, impetiginized, palmoplantar, chancriform, zosteriform, annular, eczematoid, and verrucous types, may be seen very rarely.6–8

The other rare and unusual presentation of CL is the erysipeloid type. The reason for this unusual clinical type is unknown, although an altered host immune response, specific subtypes of parasite, hormonal factors and skin barrier changes, which emerge by skin fragility of the face, especially over the cheeks and nose due to senility can be speculated as the important points. No direct correlation has been reported with age, sex, and clinical features of CL except for the erysipeloid type, which seems to predominantly affect middle-aged and elderly females. It can be considered that a hormonal factor might have played some role in the causation of this rare clinical presentation.9,11,13 Development of erysipelas like lesions in CL may be explained by cell-mediated hypersensitivity as observed in other cutaneous infections or infestations.

Erysipeloid type CL was first reported from Iran in 1994 on the face of female patients predominantly.9 In 1998, Raja reported an erysipeloid form, presented as an erythematous, indurated, ill-defined plaque over the right side of the upper lip and adjacent cheek during a 6-month period.6 Salmanpour reported 5 Iranian patients, predominantly females, between 50 and 70 years of age, presented with infiltrative erythematous lesions covering the center of the face and resembling erysipelas.11 Karincaoglu described erysipeloid-type CL from Turkey in 2004 in a 60-year-old woman, presented with butterfly-shaped infiltrated erythematous plaque on the face,10 a case very similar to ours.

Clinically, localized cutaneous lesions can resemble bacterial skin infections, blastomycosis, sporotrichosis, cutaneous anthrax, eczema, fungal skin infections, leprosy, Mycobacterium marinum infections, sarcoidosis, basal and squamous cell carcinomas, tuberculosis, infected insect bites, and cutaneous metastasis of internal malignancies.3,4,7 When the plaque lesion of CL locates on face, systemic lupus erythematosus, discoid lupus erythematosus, sarcoidosis, lupus vulgaris, and...
erysipelas must be taken into consideration in the differential diagnosis.\textsuperscript{10,14}

The diagnosis is confirmed by the demonstration of amastigotes in a Giemsa-stained smear. When the parasite cannot be detected on smear, Nicolle-Novy-McNeal medium culture, histopathologic examination, and/or leishmanin skin tests can be used. The Montenegro skin test uses leishmanial antigen to induce a cell-mediated response. Polymerase chain reaction—currently a research tool—appears to be the most sensitive single diagnostic test for CL.\textsuperscript{3,4,14}

Although most CL lesions are self-limiting and may heal spontaneously within 1 to 5 years, treatment of CL should be performed in early and multiple, mucosal, or disseminated lesions, also for lesions involving cosmetically sensitive sites and those of immunosuppressed patients.\textsuperscript{15}

There are various treatment options for CL, such as pentavalent antimony compound, cryotherapy, topical paromomycin, local heat, curettage and surgical excision, electrodissection, CO\textsubscript{2} laser, and the antifungal imidazole compounds such as ketoconazole, clotrimazole, miconazole, fluconazole, and itraconazole depending on the clinical type, localization, and diameter of the lesion. The pentavalent antimony compounds still remain the mainstay of the treatment in the majority of cases.\textsuperscript{3,14} Systemic meglumine antimoniate, 10 mg/kg/day intramuscularly for 20 days was administered in the present case. Ulcerated and erythematous lesions were healed almost completely by this regimen.

Considering the different forms of the disease nature, any unusual skin lesion located on the face, resembling erysipelas in a nonendemic area should always be investigated for CL and thus, atypical leishmaniasis should be kept in mind.

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