

Ayşe ÖKTEM
Başak YALÇIN
İrem GENÇ
Güneş GÜR



CASE REPORT

UNUSUAL CLINICAL VARIANT OF CUTANEOUS LEISHMANIASIS: ERYSIPELOID LEISHMANIASIS

ABSTRACT

Although cutaneous leishmaniasis was an important health problem in the Southeast Anatolia region of Turkey before the 1950s, it was effectively controlled by successful health policies thereafter. Classical clinical presentation of cutaneous leishmaniasis is the painless erythematous papules or nodules developed in the areas of infected sandfly bite which are ulcerated and healed gradually with a scar formation. Cutaneous leishmaniasis can be presented in its atypical forms as well, such as zosteriform, sporotrichoid, psoriasiform, chancriform, paronychia cutaneous leishmaniasis. In this case report, we present a 67 years old woman with erysipeloid cutaneous leishmaniasis, a rare atypical form of cutaneous leishmaniasis, who was successfully treated. Cutaneous leishmaniasis is an increasing health problem in Middle Eastern countries and countries receiving refugees nowadays. Regional and international health organizations should be aware of this problem and support the fight against this disease accordingly.

Key Words: Aged; Cutaneous leishmaniasis; Erysipeloid leishmaniasis.



OLGU SUNUMU

KUTANÖZ LEİSHMANİASİSİN NADİR VARYANTI: ERİZİPELOİD LEİSHMANİASİS

Öz

Türkiye'nin Güneydoğu Anadolu bölgesinde 1950 yılları öncesinde endemik olarak görülen kutanöz leishmaniasis başarılı sağlık politikaları sayesinde bu tarihten sonra etkin olarak kontrol altına alınabilmektedir. Kutanöz leishmaniasis klasik olarak tatarcık sineğinin soktuğu alanda gelişen ağrısız papül ve nodüler lezyonların ülser olup daha sonrasında skar dokusu bırakarak iyileşmesi ile karakterizedir. Kutanöz leishmaniasis zosteriform, sporotrikoid, psöriaziform, şankriform, paronişyal gibi atipik klinik tablolarla da karşımıza çıkabilmektedir. Bu olgu sunumunda atipik kutanöz leishmaniasis nadir görülen alt tiplerinden olan erizipeloid leishmaniasis tanısı olarak başarılı şekilde tedavisi tamamlanan 67 yaşında kadın hasta sunulmaktadır. Kutanöz leishmaniasis orta doğu ülkeleri ve göç alan komşu ülkelerde önemli bir sağlık problemi olarak son yıllarda karşımıza çıkmaktadır. Bölgesel ve uluslararası sağlık örgütlerinin, artan bu sağlık probleminden haberdar olup hastalıkla mücadelede desteklerini artırmaları gerekmektedir.

Anahtar Sözcükler: Yaşlı; Kutanöz leishmaniasis; Erysipeloid leishmaniasis.

Correspondance

Ayşe ÖKTEM
Ankara Numune Training and Research Hospital,
Dermatology Clinic, ANKARA

Phone: 0312 508 56 43
e-mail: ayseoktem@yahoo.com

Received: 08/08/2015

Accepted: 15/10/2015

Ankara Numune Training and Research Hospital,
Dermatology Clinic, ANKARA



INTRODUCTION

Cutaneous leishmaniasis (CL) is a protozoal infection caused by intracellularly located *Leishmania* species that are transmitted through *Phlebotomus* genus vectors (1). Although CL was an important health problem in the Southeast Anatolia region of Turkey before the 1950s, it was effectively controlled by successful health policies thereafter (1). However, due to the recent increase in the Middle Eastern refugee population in Turkey, CL cases have remarkably increased and a great variability of atypical forms of the disease have also begun to frequently appear, which may cause some difficulties in diagnosis and treatment. Here we describe the case of a 67-year-old woman with erysipeloid CL, a rare atypical form of CL, who was successfully treated.

CASE REPORT

A 67-year-old female was admitted to our clinic with enlarging areas of redness on her face. The patient's history revealed the development of a painless papule 0.5–1 cm in diameter on the nose 2 years previously, which began to extend to her cheeks and forehead. On clinical examination, erysipelas-like erythematous, edematous plaques with superficial ulcerations and crusts were observed on the midline of the face, including the dorsum of the nose, cheeks, lower forehead, and the philtrum (Figure 1). She was otherwise healthy. Laboratory examinations, including the complete blood count, blood and urine biochemistry, and chest radiography showed nor-



Figure 1— Before treatment.



Figure 2— After treatment.

mal results. Microscopical examination of Giemsa-stained smears prepared from the lesion revealed intracellular and extracellular amastigote forms of the *Leishmania* protozoa, confirming the diagnosis of cutaneous leishmaniasis (CL). Since it was a clinically atypical erysipeloid CL and the lesion was located on the face, meglumine antimoniate 15 mg/kg/day was administered intramuscularly, and the treatment was continued for 15 days without any side effects. At the end of the treatment, she recovered completely (Figure 2).

DISCUSSION

The classical clinical presentation of CL is characterized by the development of painless erythematous papules or nodules in the areas of infected sandfly bites that have ulcerated and healed gradually with the formation of a scar. However, CL can also present in its atypical forms, such as zosteriform, sporotrichoid, psoriasiform, chancriform, paronychia, and erysipeloid CL (2-4). Erysipeloid CL is a rare type of CL that was first reported in Iran in 1994 (5). The frequency of erysipeloid CL and other atypical forms of CL are not exactly known due to a lack of reports related to this subject. However, the studies that have analyzed the frequencies of atypical CL and erysipeloid CL are shown in Table 1.

The causes for the formation of atypical erysipeloid CL instead of classical CL are not known, although the patient's immune status may be responsible. Erysipeloid and other atypical forms of CL may appear in patients with defective im-

**Table 1**— Frequencies of Atypical CL and Erysipeloid CL

Studies	Total Number of the Patients	Frequency and (Number) of Atypical CL	Frequency and (Number) of Erysipeloid CL
Gurel et al. (6)	2120	1.9%	–
Bari et al. (3)	718	5.7% (41)	–
Momeni et al. (5)	1250	–	1.36% (17)
Bongiorno et al. (4)	50	40.0% (20)	34.0% (17)
Raja et al. (7)	1709	2.1% (37)	0.05% (1)

mune systems, such as a previous patient of ours who has diabetes mellitus (8). Secondly, erysipeloid CL has been reported to commonly appear in middle aged and elderly females like our patient (5). This may indicate a possible role of some hormonal factors in the development of this subgroup (9). Thirdly, as was seen in our patient, mid-face involvement seems to be common in erysipeloid CL (9,10). Increased fragility of the skin in this area may facilitate the spread of the parasite in this location (5). Finally, erysipeloid CL may be associated with a new leishmania species (3).

Based on this case, we want to indicate two important points: first, the diagnosis of atypical CLs including erysipeloid CL can be difficult and this may cause a delay in treatment. To prevent late diagnosis, we recommend giving special attention to patients with atypical lesions, traveling from endemic regions. Additionally, in patients with chronic lesions, lesions ≥ 5 cm, lesions on the face, fingers or toes, or in cases of immunosuppression, we emphasize the necessity of systemic treatment.

Cutaneous leishmaniasis is an increasing health problem in Middle Eastern countries and countries currently receiving refugees. Regional and international health organizations should be aware of this problem and support the fight against this disease accordingly.

REFERENCES

- Uzun S, Uslular C, Yucel A, Acar MA, Ozpoyraz M, Memisoglu HR. Cutaneous leishmaniasis: evaluation of 3,074 cases in the Cukurova region of Turkey. *Br J Dermatol* 1999 Feb;140(2):347-50. (PMID:10233236).
- Iftikhar N, Bari I, Ejaz A. Rare variants of Cutaneous Leishmaniasis: whitlow, paronychia, and sporotrichoid. *International Journal of Dermatology* 2003 Oct;42(10):807-9. (PMID:14521695).
- Bari AU, Rahman SB. Many faces of cutaneous leishmaniasis. *Indian J Dermatol Venereol Leprol* 2008 Jan-Feb;74(1):23-7. (PMID:18187818).
- Bongiorno MR, Pistone G, Arico M. Unusual clinical variants of cutaneous leishmaniasis in Sicily. *International Journal of Dermatology* 2009 Mar;48(3):286-9. (PMID:19261018).
- Momeni AZ, Aminjavaheri M. Clinical picture of cutaneous leishmaniasis in Isfahan, Iran. *International journal of dermatology*. 1994 Apr;33(4):260-5. (PMID:8021082).
- Gurel MS, Ulukanligil M, Ozbilge H. Cutaneous leishmaniasis in Sanliurfa: epidemiologic and clinical features of the last four years (1997-2000). *International Journal of Dermatology* 2002 Jan;41(1):32-7. (PMID:11895511).
- Raja KM, Khan AA, Hameed A, Rahman SB. Unusual clinical variants of cutaneous leishmaniasis in Pakistan. *Br J Dermatol* 1998 Jul;139(1):111-3. (PMID:9764160).
- Akilov OE, Khachemoune A, Hasan T. Clinical manifestations and classification of Old World cutaneous leishmaniasis. *International Journal of Dermatology* 2007 Feb;46(2):132-42. (PMID:17269962).
- Ceyhan AM, Yildirim M, Basak PY, Akkaya VB, Erturan I. A case of erysipeloid cutaneous leishmaniasis: atypical and unusual clinical variant. *Am J Trop Med Hyg* 2008 Mar;78(3):406-8. (PMID:18337335).
- Karincaoglu Y, Esrefoglu M, Ozcan H. Atypical clinical form of cutaneous leishmaniasis: erysipeloid form. *International Journal of Dermatology* 2004 Nov;43(11):827-9. (PMID:15533066).