

Atypical presentations of cutaneous leishmaniasis: a case series

Shahbaz Aman, Shehla Shaukat*, Ahmad Gardezi*

Department of Dermatology, Services Institute of Medical Sciences/ Services Hospital, Lahore

* Department of Dermatology, King Edward Medical University/ Mayo Hospital, Lahore

Abstract

Cutaneous leishmaniasis (CL) is endemic in many areas of Pakistan. The typical clinical presentations of cutaneous leishmaniasis are papulonodular, plaque and ulcer but certain atypical types have also been reported worldwide. Here, we describe eleven patients with uncommon lesions that were difficult to recognize clinically as CL and the diagnosis of CL was made on histopathology. Out of 11 patients, there were 9 males and 2 females. The commonly affected sites were face (55%), upper limbs (36%) and lower limbs (9%). The morphological patterns noted were lupoid (5), sporotrichoid (2), eczematous (2), furuncle-like (1) and tinea faciei-like (1).

Key words

Cutaneous leishmaniasis, atypical types, lupoid, sporotrichoid.

Introduction

Cutaneous leishmaniasis (CL) is a protozoal disease caused by an organism belonging to the genus *Leishmania* transmitted by sand flies (*Phlebotomus*, *Lutzomyia* and *Psychodopygus*).^{1,2} In Pakistan, the disease is mainly found in Dera Ismail Khan, Bajaur Agency, North Waziristan, Dir, Bannu, Kohat, Swat, Chitral, Kashmir, Multan, Khushab, Mangla, Sibbi, Quetta, Loralai, Kohlu, Khuzdar, Turbat and Azad Kashmir.¹⁻⁴ The increased occurrence of leishmaniasis is attributed to more tourism, human migration and alteration of the vector habitat.¹ Lesions usually occur on uncovered parts of the body starting from nodule or plaque and passes through the stages of crust, ulcer and scar formation.^{2,5}

It is a disease of variable morphology, at times closely mimicking various other cutaneous disorders.^{2,6} The present case series was seen

as atypical types which were not diagnosed clinically but confirmed on histopathology.

Case Report

Eleven patients with skin lesions examined in the Outpatient Department of Dermatology, Mayo Hospital, Lahore coming from different areas of Pakistan over a period of 1 year (2014) were included. These cases were difficult to diagnose clinically for CL. Atypical lesions were photographed. All the sufferers had non-healing lesions for the last 3 to 7 months. Lesions were found on exposed parts and started as papules, plaques, nodules and ulcers which gradually increased in size up to a coin. There was no history of fever, weight loss or loss of appetite, pain or itching at lesions. General physical examination and systemic review was normal in these patients. Skip lesions were noted in two patients. Three patients belonged to an endemic area of CL. Past and family histories were insignificant. The patients were non-addict and had normal bowel habits. The socio-economic status of all these patients was poor. All these patients had already been treated with topical and oral

Address for correspondence

Prof. Dr. Shahbaz Aman
Dermatology Department,
SIMS/Services Hospital,
Lahore, Pakistan
E-mail: drshahbazaman@yahoo.com

steroids, antibiotics and anti fungal drugs without any improvement.

Out of 11 patients, there were 9 males and 2 females (**Table 1**). Seven patients had only one lesion, 2 patients had two lesions whereas 2 patients had more than 2 lesions. Commonly affected sites were face (55%), upper limbs (36%) and lower limbs (9%). The morphological patterns noted were lupoid (5), sporotrichoid (2), eczematous (2), furuncle-like (1) and tinea faciei-like (1), **Figure 1-5**.

Laboratory investigations revealed no abnormalities. There was no growth seen on Gram and ZN staining. Abdominal ultrasonography, X-rays chest and of affected bones were normal. Histopathology showed hyperkeratosis and basal layer degeneration at few places. Diffuse granulomatous infiltrate, numerous plasma cells, few eosinophils and macrophages were seen containing LD bodies but there were no giant cells or caseation (**Figure 6a** and **6b**). The diagnosis of CL was made and all these patients were treated with Meglumine antimoniate 20mg/kg for three weeks showing good response.

Discussion

Cutaneous leishmaniasis (CL) is a major world health problem.³ Pakistan is an endemic country where we also see many typical and atypical morphological types of CL.^{2,4} Typical lesions do not make any diagnostic problem

but atypical presentations need investigations like smear for LD bodies and histopathology of skin biopsy for confirmation of this disorder. Atypical variants of CL have been reported in various previous studies.^{2,4,6-8} Among which mostly noted types are eczematous, erysipeloid, zosteriform, lupoid, paronychia, sporotrichoid, verrucous, chancriform, psoriasiform, mycetoma-like, DLE-like and squamous cell carcinoma-like.^{2,3,4,6-8} Atypical clinical presentations are increasingly seen in Pakistan.^{2,4} The increase in atypical presentation of CL in Pakistan could possibly be due to new parasite strains and altered or over-expressed immune host response.^{2,4,8,9}

Males presenting with atypical CL were more in number than females in our study. This could be due to the fact that men in our set up are usually working and sleeping outdoors which increases their chances of acquiring the disease in endemic areas. The most common type found in our cases was lupoid variety while the commonest site involved was face similar to other study.⁶ Tinea faciei-like type was a newly seen case which has never been reported before. Sporotrichoid pattern is thought to show an immune reaction from direct lymphatic spread of leishmania organisms.^{8,10} Multiple scattered lesions over different parts of body can be correlated with multiple bites by sandfly.

Table 1 Diagnostic work-up of adult LCH.

Sr. No.	Sex	Size	Site	Presentation
1	Male	1 x 2 cm	Face	Lupoid
2	Male	1 x 1.5 cm	Face	Lupoid
3	Female	2.5 x 1cm	Face	Lupoid
4	Male	2 x 1 cm	Face	Furuncle-like
5	Male	2 x 1 cm (2)	Forearm	Ezematous
6	Male	1 x 1 cm (2), 1 x 1.2 cm (2)	Forearm	Sporotrichoid
7	Male	1 x 1.5 cm	Foot	Eczematous
8	Female	1 x 1.2 cm	Face	Lupoid
9	Male	5 x 5 cm	Face	Tinea faciei-like
10	Male	1.5 x 1 cm (2), 1 x 1 cm(2)	Forearm	Sporotrichoid
11	Male	1 x 1.5 cm (2)	Forearm	Lupoid



Figure 1 Lupoid cutaneous leishmaniasis.



Figure 2 Tinea faciei-like cutaneous leishmaniasis.



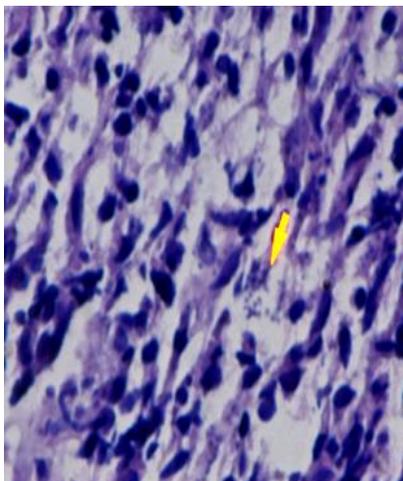
Figure 3 Furuncle-like cutaneous leishmaniasis.



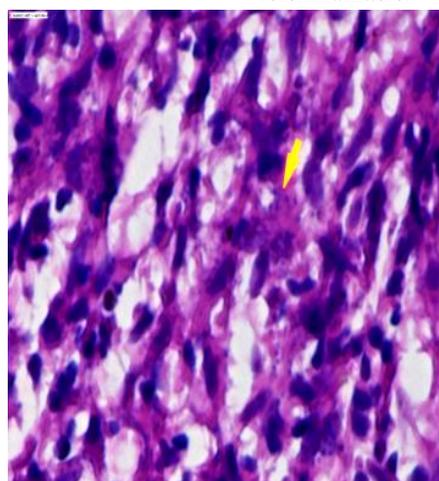
Figure 4 Eczematous cutaneous leishmaniasis.



Figure 5 Sporotrichoid cutaneous leishmaniasis.



a)



(b)

Figure 6 (a and b). Entire dermis is full of granulomas with predominantly histiocytic infiltrate. Leishman-Donovan bodies in cytoplasm and outside the cells. H&E staining 1000 x magnification

CL can present in many atypical forms and it should be kept in mind in differential diagnosis of common skin diseases, especially when patient belongs to an endemic area. The histopathological evaluation is an important tool for diagnostic confirmation in many

disorders for invisible dermatoses. Future studies are also required with isolation of species and better management of this disease.

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