Atypical presentation of cutaneous leishmaniasis: a report of three cases

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Introduction

Over the past three decades increasing numbers of patients with cutaneous leishmaniasis (CL) have been encountered in Sri Lanka. First patient of locally acquired CL was reported in 1992[2]. CL in Sri Lanka is caused by *Leishmania donovani* zymodeme MON - 37[1].

Characteristically patients with cutaneous leishmaniasis present with single erythematous indurated plaque, nodule or ulcer on exposed areas. However atypical presentations occur infrequently.

Herein we report three patients with atypical presentations of cutaneous leishmaniasis.

Case 1

A 40-year old male musician, presented with with large, non-itchy, erythematous plaque of ten years duration. It had started on right lower abdomen and extended to right inguinal region.

He had travelled in India, Middle East and several European countries during the previous few years.

On examination, a 25×35cm, erythematous irregular, indurated plaque with central necrotic ulcers was noted on right lower abdomen (Figure 1). There was a hypopigmented halo around the lesion. Lymphadenopathy was not noted. General examination and systemic examinations were normal.

Histology revealed, mildly hyperplastic epidermis with florid inflammatory infiltrate in dermis which consists of plasma cells histiocytes, lymphocytes and giant cells.

Slit skin smear (SSS) was positive for leishmaniasis. Parasites were scanty. Polymerase Chain Reaction (PCR) for Leishmania antigen was negative. X-ray and Ultra Sound Scan (USS) examination of the lesion showed soft tissue thickening, without calcification, muscle or bony abnormalities.

Haematological and biochemical investigations were normal. HIV screening was negative. USS examination of abdomen were normal.

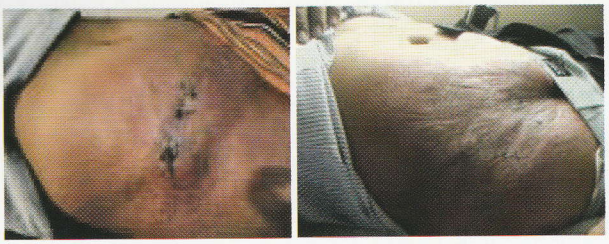

Figure 1. *Before and after treatment.*

Case 2

A 60-year old housewife, who had been employed in Saudi Arabia for several years, presented with multiple itchy erythematous skin lesions on both upper limbs and lower limbs for eight months duration.

On examination, there were multiple, pigmented erythematous, indurated plaques on extremities. Some lesions had sporotrichoid distribution and lichenoid appearance. Large lesions had irregular margins and satellite lesions. Some lesions were ulcerated (Figure 2).

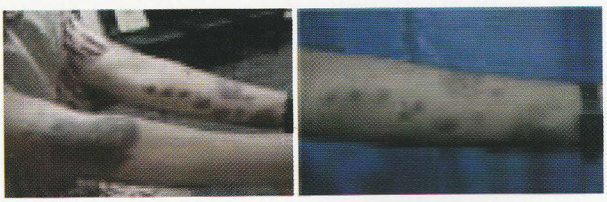

Figure 2. *Before and after treatment.*

Systemic examination was normal. Histology revealed, epidermal hyperkeratosis, acanthosis and dense inflammatory infiltrate in dermis rich in plasma cells. Other cells comprised of histiocytes, lymphocytes, and occasional giant cells. Giemsa stain showed parasitized histiocytes.

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SSS showed fragmented amastigote like structures. PCR-was negative. Other investigations including complete blood count, HIV screenings, chest x-ray, Mantoux test and USS examination of abdomen were within normal limits. She was found to be diabetic.

Case 3

A 54-year old, male, presented to department of dermatology at NHSL with an erythematous, itchy, plaque on abdomen for six months duration. He has been diabetic for six years.

On examination there was an 8x8cm, erythematous irregular indurated plaque on right abdomen, just lateral to the umbilicus (Figure 3). There was no satellite lesions or hypopigmented halo around the lesion. Systemic examinations were clinically normal.

Skin biopsy revealed parakeratosis in epidermis and dense inflammatory infiltrate composed of lymphocytes, plasma cells and macrophages in the dermis. Small clusters of intracellular organisms were noted.

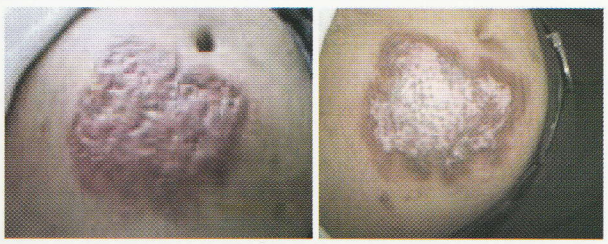

Figure 3. *Before and after treatment.*

Treatment

With intramuscular sodium stibogluconate 20 mg/kg/day for 21 days, all patients recovered uneventfully. Only notable side effects were arthralgia and myelgia.

Discussion

Cutaneous leishmaniasis is almost becoming endemic in certain parts of Sri Lanka. Typical presentations of CL are erythematous slow growing papules, nodules or ulcers, on exposed areas of extremities, head and neck region. However diverse clinical presentations do occur. Clinical diversity is basically determined by parasitic factors and immune-inflammatory response of host which is largely mediated through cellular immunity. Parasitic factors include, number of parasites inoculated and sub species of parasites. Nutritional status of the host[9], HIV infection[10], use of oral steroids[11], old age[12], wound contamination with inorganic materials[13] and site of inoculation, are important host factors.

Several unusual clinical presentations of the disease have been reported in the literature though infrequently[3,4,5,6,7,8].

Lesion of our first patient resembled deep mycosis, malignant deposit, cutaneous lymphoma or large area of panniculitis. Hypopigmented halo which have been described in atypical CL due to *L. chagasi*[23], was present around the erythematous lesion of our patient.

Atypical presentations of this patient may be due to an alteration of immune response or an unusual strain of parasite. He didn't have clinical features of immune suppression. But there is a possibility that his lesion was caused by an unusual strain, since he had been in India and Middle East previously. However we couldn't carry out strain identification due to lack of facilities.

Differential diagnosis for lesions of our second patient included sporotrichosis, atypical mycobacterial infections, lichenoid eruption and leishmaniasis. Several cases of Sporotrichoid pattern of CL were reported in Saudi Arabia[16,19] and from other Middle Eastern countries. In most of these cases the causative organism was *Leishmania major*. Our patient had acquired the disease in Saudi Arabia. This raises the suspicion whether any specific strain of leishmania could have accounted for the different pattern of distribution in her.

Our third patient also presented with a large lesion at an unusual site. Clinical differential diagnosis for his lesion included lupus vulgaris, cutaneous lymphoma and leishmaniasis. He also didn't have clinical features of immune suppression and he had never been out of the country. But he was diabetic.

There are several case reports on widespread CL on patients with immunosuppression[8,14,15]. Unusual multifocal lesions in a diabetic patient[22] has also been described in literature. Diabetes mellitus is indeed recognized by the World Health Organization as a cause of secondary immunodeficiency[21]. The defective host immune factors caused by severe diabetes mellitus in our second patient and third patient could have accounted for their unusual lesions.

We also observed, both diabetic patients (2nd and 3rd) had pruritus on lesions. Usually lesions of CL are asymptomatic. Whether diabetes mellitus of these patients contributed to pruritus is a question.

Other important observation is that lesions of first and third patients were on abdomen. However lesions on trunk as well as lesions on other uncommon sites like eye lid[17], palms and soles[18] were also reported infrequently among patients with CL. Clothing habit of an individual may have determined the occurrence of lesions at their sites.

Since CL is increasing we are bound to come across many atypical presentations. It is essential to report and investigate all atypical presentations of CL to understand contributory factors for these presentations.

In conclusion, atypical manifestations of CL mimic various skin diseases. CL should be included in the differential diagnosis of infective granulomatous diseases, notably leprosy, lupus vulgaris, atypical mycobacterial infection, deep fungal infections, sporotrichosis, as well as panniculitis, malignant deposits and cutaneous lymphoma.

The diagnosis needs a high index of suspicion. Routine investigations are of limited value. Repeated investigations are often necessary to arrive at the diagnosis.

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