

Unusual Presentations of Cutaneous Leishmaniasis in an Endemic Area, Kerman Province, Iran

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Abstract

Leishmaniasis is a infectious disease with broad spectrum of clinical features from typical noduloulcerative lesion to atypical lesions. In this article, we presented 4 cases with atypical presentation of cutaneous leishmaniasis lesions (including eczematous, sporotrichoid and lichenoid) in an endemic area in Iran, Kerman. The first case was a 28- year- old man presented with a pruritic purplish -red plaque with lichenoid appearance around proximal nail fold of right third finger since a few months ago. The second case was a young woman with an indurated yellowish-brown plaque with peripheral depigmentation firstly diagnosed as keloid with subsequent intralesional triamcinolone injection. The third case was a middle-aged woman with multiple ulcerated reddish-brown nodules and plaques in a linear array on both legs and one forearm (sporotrichoid pattern). The forth case was a man with a painful indurated reddish-brown plaque with yellowish crust and swelling resembling infected contact dermatitis.

Introduction

Leishmaniasis is a common protozoan infectious disease which is transmitted by sand fly from phlebotomous family (1). Depending on parasite genetic background, number of parasite inoculation, host immune response to parasite the clinical presentation is classified to acute and chronic types (2).

Acute form may present with typical morphologies such as papule, plaque, ulcerative nodule and impetiginous (3). Atypical forms are classified based on their morphology and location. Atypical morphology can present as zosteriform, eczematous, erysipeloid, verrusiform and psoriasiform (4-7).

Atypical locations include lips (macrocheilia), palmoplantar, periungual (paronychial) and genital area (chancriform) (8, 9).

Chronic type of leishmaniasis may present as lupoid form in site of previous scar from cutaneous leishmaniasis, or as a lesion with duration more than 1 year without any improvement (10).

In this article we describe 4 patients with uncommon presentation of cutaneous leishmaniasis.

Case reports

Case 1

A 28-year-old man presented with a history of a pruritic lesion around proximal nail fold of right third finger since a few months ago. Previously, the lesion was treated by general practitioner with impression of eczema with topical corticosteroid and emollient with no improvement. On physical examination we observed an indurated shiny purplish-red plaque with scale and minimal superficial ulcer. (Fig 1). Since, the patient was living in an endemic area of leishmaniasis treated with topical steroid without response, we decided to biopsy the lesion. Histologically, a granulomatous infiltration with numerous leishman bodies was reported. We referred our patient to leishmaniasis treatment center that treated with weekly Intralesional meglumine antimoniate in addition to cryotherapy every other week. The lesion was cleared after treatment course of 8 weeks.



Fig 1. An indurated shiny purplish-red plaque with scale and minimal ulcer on the surface

Case 2

An 18-year-old woman referred to dermatology center with a severely pruritic lesion on volar aspect of left forearm,

since 1 year ago. Initially, she was treated by general practitioner with intralesional triamcinolone as keloid without any improvement. On examination, a 2 cm² indurated yellowish-brown plaque with apple-jelly appearance and some scales on the surface with depigmented margin was observed (Fig2). Due to the chronicity and induration of the lesion we performed skin biopsy, considering chronic or lupoid leishmaniasis, sarcoidosis and lupus vulgaris as differential diagnosis. After confirming the diagnosis of chronic leishmaniasis, the lesion was treated with combination of weekly intralesional meglumine antimoniate and biweekly cryotherapy for 3 months with complete cure.



Fig 2. An indurated yellowish-brown plaque with apple-jelly appearance and some scales on the surface and depigmentation at the margin of it

Case 3

A 53-year-old woman complaining of multiple ulcerated lesions in both legs and one forearm admitted in Afzalipour hospital in Kerman for further evaluation. In her past history she had suffered from diabetes mellitus taking insulin for 10 years. She had no other problem in her medical and drug history. On physical examination, no fever, lymphadenopathy or constitutional symptoms were detected. Multiple painless

ulcerated reddish-brown nodules and plaques with crust formation in a linear array were observed (Figure 3).

Direct smear for bacterial infection was positive for pseudomonas which treated by ciprofloxacin. Skin biopsy from one of the lesions showed leishman bodies. The patient responded to intramuscular injection of meglumine antimoniate with dosage of 20 mg/kg/day for 3 weeks.



Figure 3. Multiple painless ulcerated reddish-brown nodules and plaques with crust formation on the surface in linear array

Case 4

A 47 year old man with history of painful indurated plaque on dorsum of hand from 1 year ago referred to our clinic. Despite of treatment with combination of intralesional glucantime and cryotherapy for 4 weeks, the lesion continued to spread as indurated reddish-brown plaque with yellowish crust and swelling resembling infected contact dermatitis (Fig 4). Bacterial smear and culture from the lesion was positive and patient received intravenous cefazoline 1g, twice a day. After remission of bacterial infection, as induration of the lesion persisted, skin biopsy was performed that confirmed leishmaniasis. So he was treated with parenteral

administration of glucantime for 3 weeks and oral allopurinol for 3 months.



Fig4. An indurated reddish-brown plaque with yellowish crust and multiple ulcers on the surface and swelling of hand resemble to infected contact dermatitis

Discussion

Atypical forms of cutaneous leishmaniasis can be seen with alteration (both increased and decreased) in host immune response. Cutaneous leishmaniasis in patients with cell mediated immunodeficiency secondary to HIV infection, diabetes mellitus, organ transplantation and immunosuppressive drugs can present as diffuse, disseminated, lupoid and sporotrichoid forms (10-12).

Sporotrichoid pattern is one of the uncommon presentations of CL characterized by spreading of the infection along the lymphatic drainage system. Other causes of the sporotrichoid lymphocutaneous infection are sporotrichosis schenchei, mycobacterium marinum, nocardia brasiliensis, bacillus anthracis, staphylococcus aureus and histoplasma capsulatum (13, 14).

Trauma due to intralesional injections, and immune system failure are the most common hypotheses proposed for sporotrichoid pattern of the infection (15).

Currently, first choice treatment of sporotrichoid pattern of cutaneous leishmaniasis is systemic therapy with meglumine antimoniate. In case of contraindication or intolerance, antifungal drugs such as itraconazole can be used (15, 16).

Two out of 4 of our cases (case 2 and 4) have a history of treatment with local (topical and intralesional) corticosteroid. There are some reports of reactivation and/or alteration in clinical features of cutaneous leishmaniasis after topical and systemic corticosteroid that can be explained by local immunosuppression and anti-inflammatory effects (17).

Eczematous type is one of the atypical presentations of CL reported in previous articles. Occasionally, lesions are clinically similar to eczema and only biopsy and parasitological studies may lead to correct diagnosis. Although the exact mechanism of this form of CL isn't completely revealed, exaggerated cell-

mediated immune response secondary to parasite invasion into the skin has been proposed (18, 19).

To date, there are several reports of leishmaniasis involving periungual area (20, 21), but our case (case 4) had a shiny and purplish lichenoid appearance with moderate pruritus. This can be due to repeated contact with irritant and allergic substance. According to our knowledge, this is the first report of lichenoid form of the leishmaniasis mimicking lichen simplex chronicus.

Conclusion

Atypical forms of cutaneous leishmaniasis can be seen in patients with cell mediated immunodeficiency or after application of topical and systemic corticosteroid. So we must consider atypical cutaneous leishmaniasis in differential diagnosis of indurated skin lesions unresponsive to usual treatments in endemic regions.

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