

Disseminated Cutaneous Leishmaniasis with Ocular Involvement in An Immunocompromised Patient: A Case Report

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Abstract

Purpose: To introduce a case of cutaneous and lid leishmaniasis in an immune suppressed woman

Case report: We report an unusual clinical course of old world cutaneous leishmaniasis (OWCL) in an immune suppressed 51 years old women recipient of kidney transplant. She developed disseminated cutaneous leishmaniasis (DCL) with bilateral upper and lower eye lids lesions. Antimonial compounds were not available and she did not tolerate amphotricin B. After replacing mycophenolate mofetil and cyclosporine by sirolimus, her Lesions though partially improved, were still persistent in the last visit after six months.

Conclusion: Ocular leishmaniasis should be considered in the differential diagnosis of nodular or ulcerative of the eyelids specially in the endormic cases.

Keywords: Disseminated Cutaneous Leishmaniasis, Ocular Leishmaniasis, Immunocompromised, Hordeolum, Conjunctiva

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Introduction

Old world cutaneous leishmaniasis (OWCL) occurs in an extensive geographic area extending from Mediterranean basin and North Africa to Middle East and India.¹ Causative species include *L. major*, *L. tropica*, *L. aethiopica*, *L. donovani* and *L. infantum*, which are responsible for almost all cases of cutaneous leishmaniasis (CL) in this area.² Clinical manifestation of the disease are

protean, but in most cases can be summarized into a sequence of nodule, crusting, ulceration and healing with scarring.³ Although the natural history of the lesions due to each of these four species has been described somewhat distinctly, there seems to be much overlap due to variation in host's immunological responses.⁴

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Introduction

The two most prevalent leishmania species in Iran are *L. major* and *L. tropica*.⁵ Lesions due to *L. major* are described as rapidly evolving, inflammatory, "wet" ulcers, whereas *L. tropica*, which is the predominant etiologic organism of CL in urban areas in north east of Iran, province of Khorasan Razavi, Mashhad, often causes slowly evolving, less inflammatory, "dry" type lesions.^{6,7} Versatility of clinical manifestation may go as far as "diffuse" or "disseminated cutaneous leishmaniasis" (DCL), which has previously been specified with *L. aethiopia*,⁸ but has increasingly been associated with other leishmania species, specially in immunocompromised hosts.^{9,10} DCL typically manifests as slowly growing non ulcerative papulonodular lesions that can disseminate to wide areas of skin and less commonly oral and nasal mucosa.¹¹ To our knowledge, eye mucosa has rarely been reported as a disease site in DCL. We present a case of DCL in a kidney recipient patient with ocular involvement.

Case report

A 51-year-old female patient, recipient of kidney transplant and undergoing immunosuppressive treatment attended dermatology clinic with complaint of multiple erythematous-ulcerative papules on extremities and inflamed lid margins for almost a year (Figure 1). She was otherwise healthy and received 2 g/d mycophenolate mofetil, 100 mg/d cyclosporine and 5 mg/d prednisolone as immunosuppressant. Baseline laboratory examinations were normal. She mentioned a prior localized

leishmaniasis infection on her elbow which has been treated with local injection of glucantime and an atrophic scar was visible on the site. Detailed ocular examination revealed nodular lesions in upper and lower eye lids. Lesions were 2×2 mm in size in biomicroscopic examination. Tarsal conjunctiva was hyperemic with papillary reaction in the region of the lesions. Cornea was not involved. Other anterior segment and intraocular pressure were normal. Bilaterally visual acuity (VA) was $20/20$.

All lesions mimicked hordeolum and were treated with antibiotic eye drop and ointment. No improvement was noticed and small ulcerations developed after five days. A tissue smear from conjunctiva was prepared which showed Leishman bodies. Biopsy from both skin and conjunctiva revealed granulomatous infiltration of foamy and epithelioid histiocytes, some of them containing Leishmania parasite (Figure 2) with few giant cells and scanty lymphocytic infiltration.

Finally the patient was diagnosed as DCL with conjunctival involvement.

In order to reduce immunosuppressive burden, her regimen was changed to sirolimus 1 mg/d and prednisolone 5 mg/d. She received a five days course of intravenous amphotericin B injection, which was held because of inflammation of injection site. Subsequently the patient refused to comply with the treatment anymore. After six months she attended the clinic once more, with marked improvement of all lesions specially the ophthalmic ones, but again refused treatment options.



Figure 1. Nodulo-ulcerative lesions in the forearm (a); Hordeolum-like lesions of the eyelids (b, c).

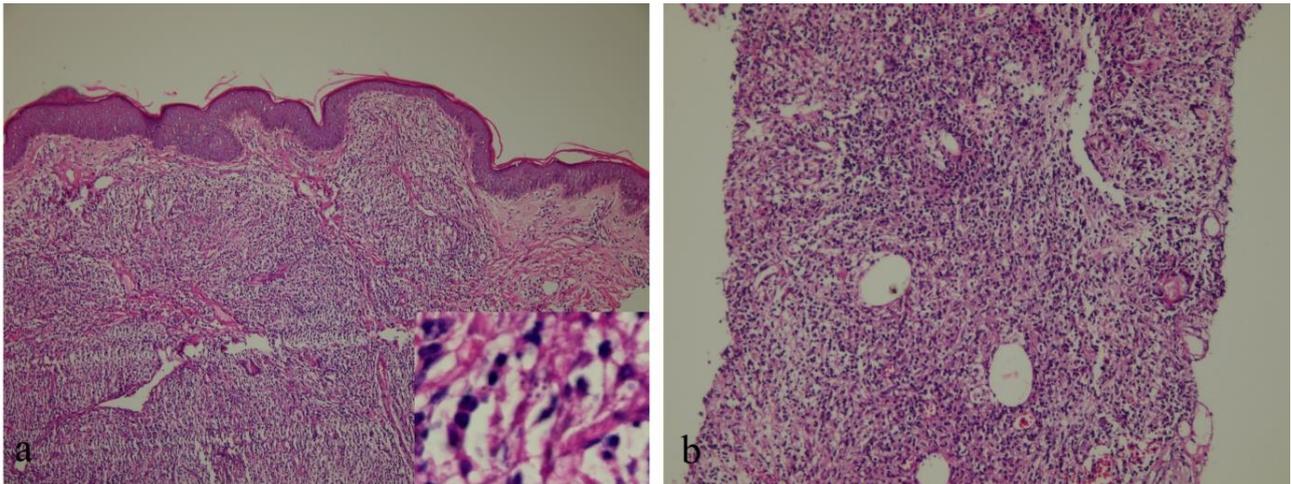


Figure 2. Diffuse nodular infiltration of histiocytes throughout the dermis with few lymphocytes or PMNs are under almost normal epidermis. Most of histiocytes have foamy cytoplasm that is characteristic for disseminated cutaneous leishmaniasis (x100). Parasites are seen in cytoplasm of vacuolated histiocytes with higher magnification (x1000) (a). A 2 mm punch biopsy from conjunctiva shows lymphohistiocytic granulomatous infiltration. Foamy histiocytes are prominent (b).

Discussion

CL is a protozoal disease transmitted by sand fly species. Skin lesions are seen mainly on exposed area, e.g. face and distal extremities. The eyelids are rarely involved, because the movement of the lid prevents the skin of this region being bitten. Ocular lesions may be caused by direct inoculation, contagious or hematogenous spread.¹² Iatrogenic immunosuppression in our patient may explain the disseminated course of the disease and eye involvement. Severe ocular involvement in Leishmaniasis has been described in patients with AIDS.^{13,14}

Ocular leishmaniasis manifests as noduloulcerative lesions on the lids and rarely conjunctiva, cornea and sclera. These lesions may simulate chalazion, epidermoid cyst, tumours, sarcoidosis and dacryocystitis.^{12,15} Because of the acute course initially, conjunctival lesions in our case were misdiagnosed as hordeolum.

Long-term complications of ocular leishmaniasis are lid deformities, dacryocystitis, conjunctival scar, interstitial keratitis and corneal opacity and ulceration.^{12,15}

Diagnosis is made by the detection of Leishman body on direct smear or sometimes biopsied specimens. Granulomatous reaction in histopathology may suggest leishmaniasis, particularly in endemic areas.

Treatment is mainly based upon intralesional or systemic antimony compounds (meglumine antimoniate). Amphotericin has been shown to be effective in the treatment of cutaneous leishmaniasis.^{16,17} We used amphotericin B for the treatment of our case, because antimony compounds were not available in Iran in that time.

Conclusion

In conclusion, ocular leishmaniasis should be considered in the differential diagnosis of nodular or ulcerative lesions of the eyelids and conjunctiva, specially in endemic areas. Early diagnosis and treatment would prevent permanent ocular sequelae.

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