

Case Report

Lid Leishmaniasis: An Atypical Presentation of Cutaneous Leishmaniasis

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ABSTRACT

A typical lesion of the localized form of cutaneous Leishmaniasis (CL) is a painless papule that enlarges over a period of several days to weeks to form a nodule or plaque, usually on exposed parts of the body such as the face, arms or legs. There has been an increase in the number of reports for rare and new forms of cutaneous leishmaniasis. The eyelid is a rare site involved by leishmaniasis and only makes up 2 to 5% of cases with cutaneous leishmaniasis. The diagnosis is based on a high index of suspicion regarding the endemicity of the disease in the region. We describe one unusual clinical form of cutaneous leishmaniasis in a 35-year-old man, who noted a small red coloured lesion on his right eyelid 4 months before. The lesion was nodular and measured 1 cm × 1 cm in diameter. A diagnosis of eyelid cutaneous leishmaniasis was made, and treatment was started with three injections of intralesional sodium stibogluconate (100 mg/ml) given on alternate days. He showed a dramatic response, and the lesion almost completely disappeared.

Key Words: Atypical leishmaniasis, Cutaneous leishmaniasis, Unusual leishmaniasis

INTRODUCTION

Cutaneous leishmaniasis is a vector-borne protozoal infection of the skin caused by several species of *Leishmania*, mainly *Leishmania major*, *Leishmania tropica* and *Leishmania aethiopica* in the Old World and species of *Leishmania braziliensis* and *Leishmania mexicana* in the New World. [1] In addition to classical clinical picture, several unusual variants and atypical features of the disease have been reported in the literature such as paronychia, whitlow, scar, psoriasiform, palmoplantar, chancriform, verrucous, erysiploid, zosteriform, mycetomatous, DLE-like, squamous cell carcinoma-like and eczematous. This clinical diversity is

basically governed by parasite and host factors and immuno-inflammatory response. [2] The usual clinicopathological picture of cutaneous leishmaniasis varies from erythematous papules to noduloulcerative forms, and mostly, the lesions are seen on the exposed parts of the body. [1] Most atypical presentations, like paronychia, whitlow, lid, palmoplantar and chancriform are probably related to the normal host response to the sandfly bite at these atypical sites. [2]

CASE REPORT

A 35 year old man, from Doda area of Jammu division of Jammu and Kashmir State (India) noted a small red coloured

lesion on his right eyelid 4 months before presentation. The lesion enlarged gradually to its present size. The patient did not give any history of itching, burning or pain associated with the lesion. There was no itching, burning, pain, discharge or impairment of vision in the affected eye but the patient had discomfort in opening his eye. There was no history of local trauma or insect bite. There was no history of similar complaints in past but there was history of a similar looking cutaneous lesion one year back over the nose in the patient's father which spontaneously healed with a residual mark within a year without any treatment. The patient had no complaints of anorexia or weight loss during the course of the disease. There were no other systemic complaints. The patient had been treated for bacterial infection by some private practitioner on several occasions with no improvement in the skin lesion. Local examination revealed single well defined, firm, nontender dome shaped nodule over the upper lid of the right eye near the inner canthus, with superficial ulceration in centre and minimal reddish brown crusting. The lesion measured 1cm x 1cm in diameter (Figure 1). The remainder of his comprehensive eye examination was normal. There was no regional lymphadenopathy. Systemic examination was normal.



Figure 1: Single nodular lesion of cutaneous leishmaniasis involving upper eyelid of eye.

CBC (complete blood count), ESR (erythrocyte sedimentation rate), CRP (C – reactive protein) and blood chemistries gave results within normal limits. Montoux test and VDRL test were negative. Sections of a biopsy using Hematoxylin and eosin stain showed a granulomatous reaction in the dermis consisting of lymphocytes, plasma cells, occasional eosinophils, and histiocytes. Many of the latter contained Leishman-donovan bodies, which were also seen extracellularly, thus confirming the diagnosis of Leishmaniasis (Figure 2). Culture could not be done due to non availability of this test in our set up. A diagnosis of uncomplicated eyelid cutaneous leishmaniasis was made, and treatment was started with three injections of intralesional sodium stibogluconate (100 mg/ml) given on alternate days using insulin syringe. He showed a dramatic response, and the lesion almost completely disappeared at the end of one month.

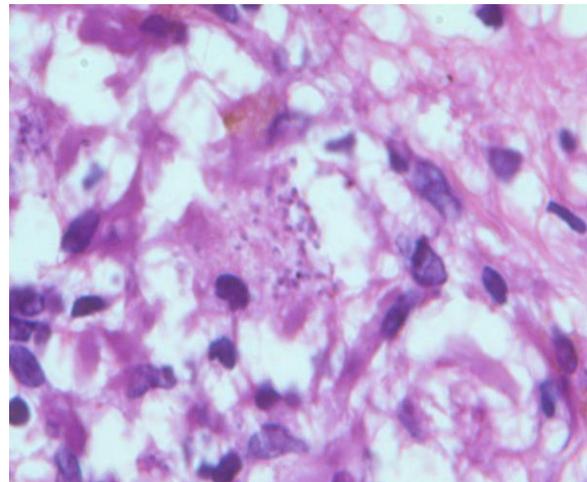


Figure 2: Skin biopsy showing multiple LD bodies both intracellularly within macrophages as well as in the extracellular space (H and E, x1000).

DISCUSSION

Cutaneous leishmaniasis is caused by different species of the Protozoan *Leishmania* with different clinical features. [3] Many of the clinical presentations are typical and present no diagnostic difficulties. [4] Localized cutaneous leishmaniasis typically presents as nodules, or noduloulcerative plaques located mostly on face and extremities.

Various unusual clinical presentations have also been reported in literature. Eyelid is one of the rare sites for the sand fly to bite and produce cutaneous leishmaniasis because the movements of the lids prevent the sandfly from biting the skin in this area. [5-8] The lid is involved in about 2-5 percent of cases of cutaneous leishmaniasis. [5,8,9] In a study on 718 patients on CL, Lid leishmaniasis, as an unusual presentation of CL, was seen in 4.9% of unusual cases in the study. [2] Lid lesions may be caused by the bite, inoculation of the lid by patient's fingers, lymphatic dissemination, or contiguous spread from a neighbouring site. [5] Lid CL may also occur in mucocutaneous leishmaniasis, due to *Leishmania braziliensis*, when infected material reaches the lid from nasal mucosa from the nasolacrimal duct. [6,8]

When the skin of the lids is affected, both upper and lower lids may be involved on either their outer or inner aspects, most often on the lateral canthus. The pathologic pattern is comparative to what can be observed on the rest of the skin, but eyelid fragility results in a special risk for local spread. [5,9] Conjunctiva, sclera and cornea are rarely affected, while lacrimal ducts are sometimes affected with stenosis leading to a chronic discharging fistula and secondary conjunctivitis and uveitis. [5,8,9] When the outer aspect of the lid is involved, the disease is usually self-limiting and healing takes place by scarring. When the inner aspect of lid is involved, contiguous spread will extend to involve the conjunctiva, sclera and cornea with development of interstitial keratitis. [5,6,7] The most common aspect of lid leishmaniasis is a chalazion-like lump but ulcerous, phagedenic, cancer-like forms and unilateral chronic granulomatous blepharitis may be observed. [10,11]

The diagnosis of Lid leishmaniasis is based on a high index of suspicion regarding the endemicity of the disease in the region. Lid leishmaniasis particularly in its early nodular form may simulate

secondarily infected insect bite, eczema, malignancy including basal cell carcinoma, cutaneous tuberculosis (lupus vulgaris), syphilis, sarcoidosis, and other cutaneous disorders. [5-7,10,12] False diagnosis can be best avoided by taking a detailed and accurate history and identifying the pathogens as early as possible on smear, histopathology or culture or by using modern methods like Polymerase chain reaction, electron microscopy and serology. [13]

Pentavalent antimonials with their consistent efficacy remain the drug of choice for treating localized cutaneous leishmaniasis. WHO recommends intralesional sodium stibogluconate therapy for early and localized cutaneous leishmaniasis. This regimen has some side effects such as pain and swelling at the injection site as compared to serious side effects such as cardiovascular toxicity, neurotoxicity, renal toxicity and bone marrow hyperplasia seen with systemic therapy. [14]

Lid leishmaniasis is considered a potentially blinding disorder; therefore dermatologist should consider cutaneous leishmaniasis in the differential diagnosis of patients suffering from chronic granulomatous lid lesions especially in areas where this parasite is endemic, so that early diagnosis and rigorous treatment may prevent blinding complication. In our knowledge, eyelid cutaneous leishmaniasis is unusual, having only been described in few cases in literature and we are probably the first to report this rare presentation of cutaneous leishmaniasis from India.

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