

Cutaneous Leishmaniasis of the Lid: A Report of Nine Cases

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Leishmaniasis is a parasitic disease caused by *Leishmania* species and is classified into three forms; cutaneous, mucocutaneous, and visceral. The eyelid is a rare site involved by leishmaniasis and only makes up 2.5% of cases with cutaneous leishmaniasis (CL). Although CL can affect both upper and lower lids on either their outer or inner aspects, the lateral canthus is most often affected. The most common aspect of lid leishmaniasis is chalazion-like lesions but ulcerous, phagedenic, cancer-like forms, and unilateral chronic granulomatous blepharitis may be observed. When the lid is involved, the disease is usually self-limiting; healing usually takes up to one year, hence early diagnosis and treatment are important. The diagnosis is based on a high index of suspicion regarding the endemicity of the disease in the region. Response to treatment in lid CL cases is quite satisfactory. In this article, we report nine cases of lid leishmaniasis with satisfactory responses to intralesional meglumine antimoniate.

Key Words: Iran, Leishmaniasis, Unusual atypical lid

Leishmaniasis is a parasitic disease caused by *Leishmania*, transmitted by the bite of some sandfly species and affects various age groups [1]. Depending on the infecting *Leishmania* species and host immunocompetence, there are cutaneous, mucocutaneous, and visceral forms of the disease [1]. It is estimated that 350 million people are at risk of leishmaniasis [2, 3]. The disease, in its various forms, affects at least 12 million people worldwide [3-5], with 1.5 to 2 million new cases per year [2, 3].

Cutaneous leishmaniasis (CL) is endemic in Iran [4]. It is usually caused by *L. major* and *L. tropica* [4], and typically presents with a skin ulcer over exposed regions of the body after a sandfly bite. It generally heals spontaneously within 3-6 months [1].

In addition to the classical picture, several unusual and atypical clinical features of the disease have been reported in the literature [1, 6]. These features may appear at unusual

sites or may present with atypical morphologies [1]. The eyelid is one of the rare sites for the sandfly to bite [6] and is the cause for 2.5% of CL cases [3, 7].

Abboud et al. [7] describes some cases of lid CL reported by authors from different parts of the world. Herein, we described nine cases of lid CL.

Case Report

Nine cases of lid cutaneous leishmaniasis were referred to us between 2002 and 2008 (Fig. 1). All Leishmaniasis cases were confirmed by demonstration of typical amastigotes in stained smear with Wright-Eosin-Methylene blue prepared from the margin of the lesions (Fig. 2). Characteristics of our cases were summarized in Table 1.

In most of our patients, chalazion-like lesions of leishmaniasis were present. In some cases, lesions were treated as eczema without any improvement. In one case, the diagnosis of chronic blepharitis had been suggested. No complication such as involvement of conjunctiva, cornea, or sclera was seen.

Discussion

Symptomatic CL is diverse in its presentation and out-

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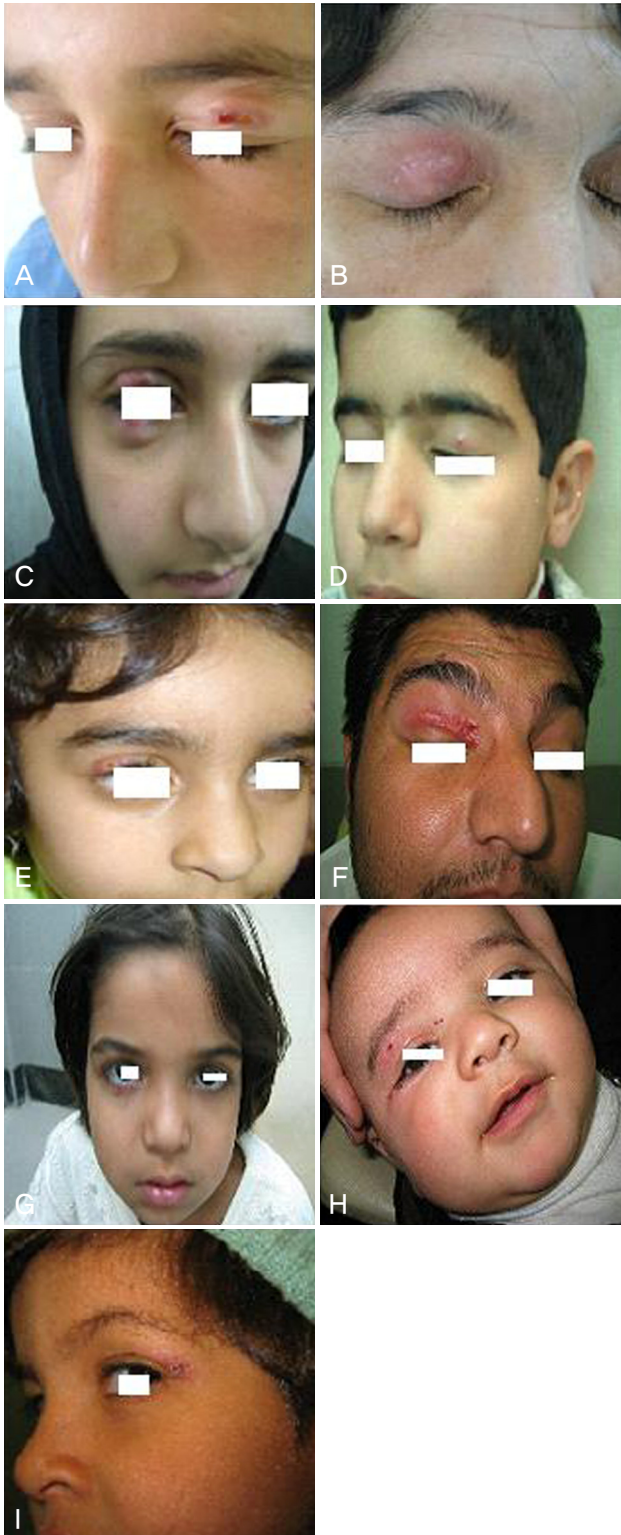


Fig. 1. Cutaneous lid leishmaniasis. (A) A 13-yr-old boy, as ulcerative lesion. (B) A 39-yr-old woman manifested as an erythematous plaque. (C) A 15-yr-old girl, chalazion-like. (D) A 12-yr-old boy manifested as chalazion. (E) A 7-yr-old girl manifested as chalazion. (F) A 30-yr-old man manifested as chronic granulomatous blepharitis. (G) A 7-yr-old girl, chalazion-like. (H) A 6-mon-old baby, eczema-like. (I) A 6-yr-old boy manifested as classic leishmaniasis.

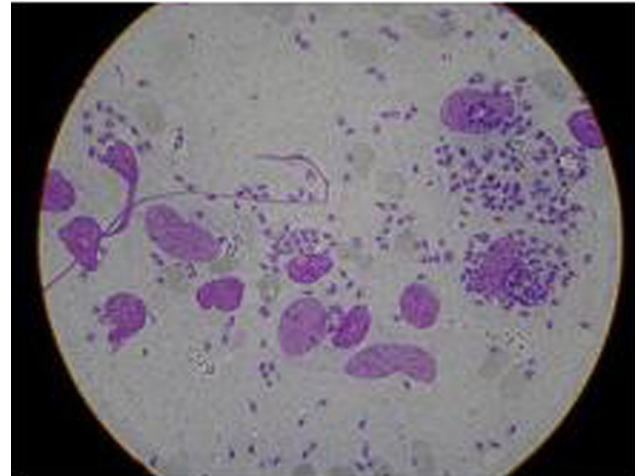


Fig. 2. Typical amastigotes in a stained smear with Wright-Eosin-Methylene blue prepared from the margin of the lesions.

come [1]. This clinical diversity is basically governed by the parasite, host factors, and immune-inflammatory responses [1]. The incubation period of CL varies between several weeks to about a year [5].

The eyelid is rarely involved in the cutaneous form of leishmaniasis, possibly due to movement of the lids preventing the fly vector from biting the skin in this region [2-5, 7]. Lid lesions may be caused by the bite, inoculation of the lid by the patient's fingers, lymphatic dissemination, the Koebner phenomenon (emergence of a new lesion after a mechanical trauma), or contiguous spread from a neighboring site [4]. Lid CL may also occur in mucocutaneous leishmaniasis, due to *L. braziliensis*, when infected material reaches the lid from nasal mucosal lesions via the nasolacrimal duct [2, 5].

The typical CL lesion is a solitary ulcerating granuloma which starts as a small itching papule and then becomes scaled, crusted, and finally ulcerated [5]. When the skin of the lids is affected, both upper and lower lids may be involved on either their outer or inner aspects [5], most often on the lateral canthus [4, 8]. The most common aspect of lid leishmaniasis is a chalazion-like lump but ulcerous, phagedenic, cancer-like forms [8], and unilateral chronic granulomatous blepharitis may be observed [9].

Approximately 2-5% of facial lesions are localized to the eyelids [4, 5, 8]. In a study on 718 patients with CL, unusual variants were seen in 5.7% of the patients. Lid leishmaniasis, as an unusual presentation of CL, was seen in 4.9% of unusual cases in the study [1]. Most atypical presentations, like paronychia, whitlow, lid, palmoplantar, and chancreiform are probably related to the normal host response to the sandfly bite at these atypical sites [1].

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 [4, 8].

Most atypical morphologies are correctly diagnosed,

Table 1. Summarized characteristics of patients with lid cutaneous leishmaniasis

Case No.	Age (yr)	Sex	Duration of lesions (mon)	Clinical feature of lesions	Sites of involvement on the lid
1	13	M	2	Ulcerative	The left upper lid
2	39	F	6	Erythematous plaque (eczema-like)	The right upper lid
3	15	F	2	Chalazion-like	Two lesions on upper and one lesion on lower lid of the right eye
4	12	M	3	Chalazion-like	The left upper lid
5	7	F	3	Chalazion-like	Margin of the right upper lid
6	30	M	2	Chronic blepharitis	The right upper lid
7	7	F	1.5	Chalazion-like	The right lower lid
8	6	M	2	Classic Leishmaniasis lesion	The left upper lid in lateral part
9	6 mon	M	1	Erythematous plaque (eczema-like)	The right upper and lower lid

keeping a high index of suspicion regarding the endemicity of the disease in the region; but in a few cases, there is substantial delay. It is after therapeutic unresponsiveness and/or further investigations that an ultimate diagnosis of atypical CL can be made [1]. Scraping from lesions, especially early in the infection, show leishman-Donovan bodies which are found mainly within histiocytes, but also can be found extracellularly. The presence of these bodies from a typical skin lesion was first described by Cunningham in 1885 [5].

Clinical diagnosis of ocular leishmaniasis is very difficult [4, 10], especially when appearing in non-endemic areas [2]. Conjunctiva [4, 7], cornea [7], and sclera are rarely [2] affected, while lachrymal ducts are sometimes [8] affected [4]. The disease, particularly its early papulonodular lesion [2], may simulate other more common lesions such as chalazion, tumors (including basal cell carcinoma, especially its ulcerative form) [2-4, 10], dactiocystitis [2, 4], tuberculosis (lupus vulgaris), syphilis [5, 10], sarcoidosis [10], keratoacanthoma (nodule, tumor-like lesions, when ulcerated, in older individuals), histoplasmosis, and rhinoscleroma [5]. Eczema was also added to the differential diagnosis list of lid leishmaniasis.

False diagnoses can be best avoided by taking a detailed and accurate history and identifying the pathogens as early as possible in smear cultures or by electron microscopy [11].

When the outer aspect of the lid is involved, the disease is usually self-limiting and healing takes place by scarring for up to one year. A much more serious chain of events is to be expected when the inner aspect is involved [5]. The contiguous spread from the skin of the lid will extend to involve the conjunctiva, sclera, and even cornea, with development of interstitial keratitis [2-4]. Secondary infection with destruction of underlying soft and bony tissue is common [5]. Long-term complications of ocular leishmaniasis include lid deformity with all its consequences [3, 4]. Stenosis of the lacrimal duct leads to a chronic discharging fistula and secondary conjunctivitis and uveitis [5]. Therefore, ocular leishmaniasis is considered a po-

tentially blinding disorder; early diagnosis and rigorous treatment may prevent blinding complications [2-4]. Residual scarring and deformity may require surgical correction [2]. The lid lesions in all our cases improved completely without any residual significant complication, because of early diagnosis and appropriate therapeutic approach.

Response to treatment in atypical cases is quite satisfactory [1]. Treatment with pentavalent antimonial or sodium stibogluconate (pentostam) is typically curative and more effective. The dosage recommended by the World Health Organization is 20 mg/kg per day for 3 weeks [2]. We underwent intralesional injection of meglumine antimoniate (glucantime) in all our patients. It appeared that this therapeutic regimen resulted in a good and relatively rapid response. Because of the loose tissue in the eyelid, injection is done easily, with the least feeling of pain and in large amount. We believe this is the cause of the satisfactory response.

Treatment failure may be due to both delay in diagnosis and incomplete therapy, as well as other factors such as resistant *Leishmania* species or deficient host immune responses [3].

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