Eyelid leishmaniasis in a patient with neurogenic ptosis

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Abstract
A patient with chronic progressive external ophthalmoplegia contracted cutaneous leishmaniasis of the upper eyelid. Infection of this site is rare because eyelid movements usually prevent the sandfly vector from biting the skin there. It is postulated that the relative immobility of the upper eyelid in this patient was a major predisposing factor for the infection.

Leishmaniasis is a protozoan zoonosis transmitted to man by sandfly vectors. The vertebrate host includes rodents, dogs, cats, and humans. It is endemic to the tropical and warm temperature zones of Central and South America, Africa, the Middle East, the Indian subcontinent, and many countries in the Mediterranean basin.1-3

Leishmaniasis of the eyelid is uncommon. The highest incidence has been reported in Algeria, where it has been described in 2–5% of all cases of facial leishmaniasis.4 As there have been only three previous reports of eyelid leishmaniasis in visitors to endemic areas in the English literature,5-7 it has been suggested that the rarity of eyelid infestation is due to eyelid movements preventing the fly vector from biting the skin in this area.8

We report a case of eyelid leishmaniasis occurring in a patient with reduce lid movements due to a chronic progressive external ophthalmoplegia.

Case report
A 46-year-old Caucasian female developed a papule on her left upper eyelid in December 1988 while visiting rural Yemen. She stayed in a mountainous district near Ta’izz (syn. Taiz). The patient had a documented past history of chronic progressive external ophthalmoplegia and had presented as a teenager with bilateral ptosis. Her unaided visual acuity was recorded as 6/24 right and 6/18 left prior to her trip to the Yemen. Her corrected acuity was only one Snellen line better in each eye. L levator palpebrae superioris function measured less than 2 mm in each eye. There was bilateral inferior corneal exposure and lower facial weakness. She had bilateral total external ophthalmoplegia and pigmentary retinopathy. Electoretinographic responses to blue and white scotopic, photopic, and flicker stimuli were all within normal limits. Goldmann fields were full. An edrophonium (Tensilon) test gave a normal result. Her electrocardiogram did not show any conduction defect.

The eyelid lesion enlarged slowly over the next four weeks and the entire lid became oedematous, with a small ulcer developing in the centre. Her vision was unchanged, and systemically she was in good health. On returning to the United Kingdom she was treated with 2 g of fluclotaxillin daily in divided doses for four weeks. The lesion persisted despite antibiotics, and she was referred to our eye casualty for further treatment. Oriental sore was suspected and she was therefore referred to a dermatologist.

Eight weeks after the onset of symptoms the area of left upper lid induration was recorded as 25 mm in diameter while a central area of ulceration measured 5 mm (Fig 1). There was no mucosal involvement. Systemic examination did not reveal any lymphadenopathy, hepatomegaly, or splenomegaly. Her haemoglobin, white cell count, differential count, and blood film were all normal. A biopsy was taken from the non-ulcerated edge of the lesion, and cutaneous leishmaniasis was diagnosed histologically on haematoxylin and eosin staining of a formalin fixed specimen. This showed an infiltrate of plasma cells and pale histiocytic cells containing multiple Leishman–Donovan bodies in the dermis. No further identification of the species was made.

She was treated with sodium stibogluconate (Pentostam) 10 mg/kg body weight daily for 21 days, with regular monitoring of her hepatic transaminases and ECG. She had no adverse reactions to treatment. The ulcer healed two months after treatment, and the residual oedema settled two months later (Fig 2).

Discussion
Old World leishmania causes four distinct forms of cutaneous disease depending on the degree of the host’s cellular immune response. Oriental
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sore is the commonest form. It causes an ulcer which usually heals within six months. Leishmaniasis recidiva and diffuse cutaneous leishmaniasis are chronic recalcitrant forms of cutaneous leishmaniasis seen in subjects with hyperergic and anergic responses respectively. The development of immunity following infections with visceral leishmaniasis may be accompanied by the occurrence of post-kala-azar dermal leishmaniasis.

Old world cutaneous leishmaniasis (without visceral involvement) is caused by Leishmania tropica, L major, L infantum, and L aethiopica. It is possible that any of these strains caused this infection, as the Leishmania strain was not identified and there is limited knowledge about the strains of leishmaniasis present in Yemen. Epidemiological studies have demonstrated the presence of Leishmania tropica, L donovani, and L infantum in rural, coastal, and mountainous Yemen. Enzyme linked assays used in this recent study showed that L tropica (strain LON 71) caused all cases (24/24) of cutaneous leishmaniasis tested in the study area.

Cutaneous leishmaniasis should be considered in the differential diagnosis of eyelid ulcers, abscesses, or nodules in patients who have been to areas endemic for leishmaniasis. It may mimic chalazion, skin tumours, or epidermoid cyst. It is important for medical staff to be aware of the prevalence of leishmaniasis with increasing international travel and also to know that the incubation period may extend to several months.

Treatment includes control of secondary infection, the intralesional or systemic administration of antimony, and cryotherapy.

Morgan suggested that the rarity of eyelid infestations was due to movement of the upper eyelid preventing the sandfly vector from inoculating this otherwise exposed site. As this patient had virtually absent levator function, her case gives further support for this hypothesis.

We thank Mr G Sutton for his permission to report this patient.

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