A 73-year-old white woman had a 14-month history of an extensive, disfiguring facial lesion involving the cheeks, nose, and eyelids, resulting in exposure keratopathy. A biopsy of the facial lesion established the diagnosis of cutaneous leishmaniasis, and the lesion responded to treatment with itraconazole.


Leishmaniasis, a parasitic infection caused by a hemoflagellate protozoan of the genus *Leishmania*, is rarely seen in the United States. It is endemic, however, in the Mediterranean littoral, the Middle East, Africa, and Central Asia. With the current regularity of worldwide travel, physicians around the world must have an index of suspicion for this diagnosis. We describe a patient from Italy who had facial cutaneous leishmaniasis and associated bilateral ptosis, ectropion, and exposure keratopathy.

REPORT OF A CASE

A 73-year-old woman who had immigrated 4 months previously from Italy, where she worked on a farm, was first seen with a chief ocular complaint of facial sores, associated with itching, burning, photophobia, and mucoid discharge. Fourteen months previously, she noted a small pimple on her nose, which gradually developed into an extensive facial lesion that did not respond to treatment with a variety of antibiotics. The medical and surgical histories were unremarkable.

External examination revealed an extensive disfiguring, verrucous facial lesion with multiple areas of crusting, lichenification, and vegetation primarily involving the nose and cheeks with extension to the eyelids (Figure 1). There was bilateral ectropion and ptosis with weakened levator palpebral superior function, requiring use of her frontalis muscles to assist in lid elevation. Visual acuity was correctable to 20/25 OD and 20/30 OS. Slitlamp examination revealed bilateral inferior superficial punctate keratopathy and palpebral conjunctival injection and hypertrophy with granulation and diffuse papillae. The results of the remainder of the ocular examination were normal. Systemic examination did not reveal any lymphadenopathy or hepatosplenomegaly.

A biopsy specimen was obtained from the edge of the lesion on the cheek. Hematoxylin-eosin staining of the formalin-fixed specimen showed chronic inflammatory infiltration with focal areas of granulomatous inflammation (Figure 2, top). The inflammatory cells consisted primarily of histiocytes, plasma cells, and lymphocytes without evidence of necrosis. Multiple Leishman-Donovan bodies were present within the cytoplasm of histiocytes as well as in free form in the dermis, confirming the diagnosis of leishmaniasis (Figure 2, bottom). After 2 months of treatment with itraconazole, 100 mg orally daily, lubricating drops, and ointment, a significant improvement was noted in the facial lesions, ptosis, ectropion, and keratopathy.

COMMENT

Cutaneous leishmaniasis, which is caused by *Leishmania major* and *Leishmania tropica* and transmitted by sand flies, typically be-
gins as a small erythematous papule on the face or extremity at the site of inoculation. It progresses slowly to an indurated, verrucous plaquelike nodule, reaching 1 to 2 cm in diameter, and after several weeks or months develops into a shallow ulcer containing a central crust.1 Small satellite nodules are characteristically present at the edge of the lesion. Involvement of the lid and ocular adnexa may simulate dacryocystitis, a chalazion, or a tumor of the lid.2 As in the present case, ocular complications of cutaneous leishmaniasis may include cicatricial ectropion and ptosis. Other infectious conditions that can mimic the clinical picture typical of leishmanias include lepromatous leprosy, sporotrichosis, herpes zoster ophthalmicus, herpes simplex, vaccinia, anthrax, toxoplasmosis, histoplasmosis, blastomycosis, and candidiasis.3

The diagnosis of the infection is established based on clinical suspicion and confirmed by biopsy. On microscopic examination, the cutaneous lesion discloses a moderate dermal infiltrate of lymphocytes and plasma cells intermixed with a granulomatous inflammatory reaction composed of histiocytes, epitheloid cells, and multinucleated giant cells.4 The Leishmania organism within the cytoplasm of the histiocytes appears as a round to oval body, measuring 2 to 3 µm in diameter, containing a nucleus and a small kinetoplast (paranucleus).1

In most immunocompetent individuals, the infection and cutaneous manifestations spontaneously resolve. Otherwise treatment with pentavalent antimonial, sodium stibogluconate (pentostam), is typically curative. However, serious systemic adverse effects, such as fatal cardiac arrhythmias, hepatotoxicity, nephropathy, and gastrointestinal systemic effects, such as nausea and vomiting, limit its use,1 and more tolerable antifungal agents (ketoconazole, itraconazole, and amphotericin B), as well as pentamidine and allopurinol, may be equally effective. Systemic therapy with interferon gamma has also been shown to be effective in the treatment of cutaneous leishmaniasis.5 Residual scarring and deformity may require surgical correction.

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REFERENCES


A look at the past . . .

ISRAELOHN (Smolensk) gave an account of 42 cases of Distichiasis and trichiasis cured by transplantation of a piece of mucous membrane from the lip. Kuhnt proceeded in the same manner, only without the use of sutures, and Matkovec had had success by the same method.