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Case Report

Sporotrichoid Cutaneous Leishmaniasis in Central Iran

J. Ayatollahi

Abstract
More than 90% of patients infected with *cutaneous leishmaniasis* live in Afghanistan, the Middle East, Algeria, Brazil, and Peru. *Cutaneous leishmaniasis* in Iran is caused by *Leishmania tropica* and *Leishmania major*. Most skin lesions evolve from papular to nodular, to ulcerative form with a central depression surrounded by a raised indurate border. Some lesions persist as nodules or plaques. Multiple primary lesions, satellite lesion, regional adenopathy and sporotrichoid subcutaneous nodules are variably present. This report presents an 18-yr-old man with sporotrichoid cutaneous leishmaniasis that is not common in the central Iran.
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Keywords ● Sporotrichoid ● cutaneous ● leishmaniasis ● Iran

Introduction

Cutaneous leishmaniasis is classified as New World (American) or Old World disease. Local names for Old World disease include oriental sore, bouton d'Orient, Aleppo evil, and Baghdad boil. Old World CL is caused by *Leishmania* (*L*) *tropica*, *L major*, *L aethiopica*, and *L infantum* and *L donovani*.[1,.2] *cutaneous leishmaniasis* is common and is a major health problem in many areas of Iran but its sporotrichoid form is rare.

Case presentation

A previously well, 18-year-old man from Yazd province, center of Iran, noticed an indurate area near his left elbow. The area gradually grew in size, became erythematous and ulcerated after two weeks. Three weeks later he noticed the development of adjacent bumps extending proximally from the ulcer in a linear pattern, followed by the development of diffuse subcutaneous nodules in the triceps region. The ulcer was painless, the patient had no systemic symptoms and there was no past medical, or drug history of note.

He was received cephalexin without success and was then referred to a dermatologist. Biopsy of the ulcer rim showed necrotizing granulomatous inflammation with angiocentric lymphoid infiltrate. Special stains were negative for fungi, acid-fast bacteria, and common bacteria. The lesion continued to enlarge over two subsequent weeks, so the patient was referred to Infectious and Tropical Diseases Research Center, for further evaluation. His medical and social history was unremarkable. The patient denied risk factors associated with sexually transmitted diseases and reported no fever or constitutional symptoms. Further review of the systems was.

Infectious and Tropical Diseases Research Center,
Shahid Sadoughi Hospital.
Yazd University of Medical Sciences,
Yazd, Iran.

Correspondence:
Jamshid Ayatollahi MD,
Infectious and Tropical Diseases Research Center,
Shahid Sadoughi Hospital,
Yazd University of Medical Sciences,
Yazd, Iran.
Tel/Fax: +98 351 8224228
E-mail: jamshidayatollahi@yahoo.com

unremarkable. Physical examination revealed an apprehensive but healthy-appearing young man. The temperature was 37ºC. There was a round cutaneous ulcer (1.5 cm in diameter) with a granulating base on the lateral aspect of the right elbow. The margins were raised, and the base was relatively clean. A chain of nodules extended proximally from the ulcer. There was no axillary or epitrochlear adenopathy.

Results of CBC and ESR were normal. VDRL and HIV tested negative. Repeated biopsies of the ulcer margin revealed granulomatous inflammation with necrosis. Touch preparations stained with Wright-Giemsa preparation were positive for leishmania.

The patient was treated with meglumine antimonite (Glucantime), pentavalent antimony, at a dosage of 20 mg/kg per day, intramuscularly for 21 days. After completing the course of treatment, the primary ulcer decreased in diameter to 0.5 cm, partially healed, and chain of nodules resolved. Although the patient reported mild myalgia, he tolerated the medication well.

Discussion

Cutaneous leishmaniasis in Yazd is caused by Leishmania major. The vectors are sandflies of the genus phlebotomies, most probably phlebotomus papatasi because approximately 77% of indoor sandflies in this region are of this species.[1]

The incubation period ranges from weeks to months. The lesions appear typically on exposed areas of the body where inoculation occurs.[2] They appear as small nodules, which progressively increase in size and eventually ulcerate. The morphologic characteristics depend on the complex interactions between the virulent characteristics of the infecting leishmania sp. and T-cell mediated immune responses of its human host.[3]The classic lesion is round with raised margins and a granulating center with yellowish exudates.[2]

The sporotrichoid pattern has been reported in cases of American cutaneous leishmaniasis from Panama and Brazil.[2,4-6] Melby et al. reviewed 59 cases of cutaneous leishmaniasis treated at the National Institutes of Health from 1973 to 1991 and found that 90% of cases with a sporotrichoid pattern were due to L (Viannia) braziliensis.[7] In infections caused by these species, regional lymphadenopathy is common and may be accompanied by fever and constitutional symptoms even before skin lesions become apparent.[2,8] Old World cutaneous leishmaniasis is less commonly associated with sporotrichoid presentation, several cases have been reported from the Middle East, though.[9]

Cutaneous leishmaniasis should be considered in differential diagnosis of any chronic localized skin lesion in a person who had been exposed in an endemic area.[10,11] Other etiologies such as infection with atypical mycobacteria or infections with fungi such as Histoplasma capsulatum, Blastomyces, Dermatitidis, or Sporothrix schenckii, which was incorrectly diagnosed in our presented case, should be considered.

The chronic course of cutaneous leishmaniasis makes it more easily distinguished from acute causes of localized skin infections, such as Staphylococcus aureus, Streptococcus pyogenes, Francisella tularensis, and Bacillus anthracis.

The diagnosis of cutaneous leishmaniasis may be difficult to confirm. The lesion should be carefully and repeatedly cleaned before aspiration and biopsy. An aspirate should be taken from the margin after injection of non-bacteriostatic saline. Either a punch or excisional biopsy specimen should also be taken from the margin of the lesion. Touch preparations should be made from the tissue, stain with a Wright-Giemsa preparation, and examine for intracellular amastigotes.[12] A proportion of patients should be sent for histopathologic analysis. Aspirate and biopsy material should be cultured for leishmania, fungi, and mycobacteria.

Sporotrichoid cutaneous leishmaniasis must be treated. The pentavalent antimony meglumine antimonite (20 mg/kg per day for 20 days) remains the drug of choice.[13]

Pentavalent antimony compounds have a number of potential untoward effects particularly when used at higher than recommended doses. These effects include cardiotoxicity, nephrotoxicity, liver enzyme elevations, arthralgia, myalgia, and chemical and clinical pancreatitis.[14-16]

Except for myalgia, our patient tolerated the treatment well. A number of new approaches to chemotherapy are under investigation, but none has yet proven to be superior to pentavalent antimony for the treatment of cutaneous lesions caused by L.major.[16]

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