Case Report: Isolation of *Leishmania tropica* from a Patient with Visceral Leishmaniasis and Disseminated Cutaneous Leishmaniasis, Southern Iran

Abdolvahab Alborzi,* Gholam R. Pouladfar, Mahdi Fakhar, Mohammad H. Motazedian, Gholam R. Hatam, and Mohammad R. Kadivar

*Professor Alborzi Clinical Microbiology Research Center, and Department of Medical Parasitology and Mycology, Shiraz University of Medical Sciences, Shiraz, Iran; Department of Medical Parasitology and Mycology, Mazandaran University of Medical Sciences, Sari, Iran*

**Abstract.** We report a case visceral leishmaniasis with disseminated cutaneous leishmaniasis caused by *Leishmania tropica* in southern Iran. Typing of this parasite was performed by a species-specific polymerase chain reaction and isoenzyme electrophoresis.

**INTRODUCTION**

Disseminated cutaneous leishmaniasis (DCL) is characterized by 10–800 lesions on the head, limbs, and trunk. Most lesions are small, papular, and appear simultaneously with or secondarily to one or several ulcerated lesions of localized cutaneous leishmaniasis.1 This disease has been reported in a small subset of immunocompetent patients infected with *Leishmania braziliensis* and *L. amazonensis* in Brazil.2 A case of DCL caused by *L. guyanensis* was reported from French Guiana.3 We report a patient with DCL accompanied by visceral leishmaniasis (VL) caused by *L. tropica*.

**CASE REPORT**

In September 2006, a 15-year-old woman was admitted to Nemazee Hospital in Shiraz, Iran. In October 2005, a papular lesion appeared on her left forearm that progressed within one month to a painless ulcerative lesion. *Leishmania* amastigotes were seen in a skin biopsy specimen. She was treated with meglumine antimoniate and showed some improvement. Nine months later, numerous, small, papulonodular, painless, non-pruritic, pink lesions appeared on her extremities, back, and face, a few of which became ulcerative (Figure 1), and the primary lesion worsened (Figure 2). A raised superficial lesion was detected on the nasal aspect of a left sclera biopsy specimen (Figure 3). The liver and spleen were palpated 6 cm and 12 cm below costal margins, respectively. Abundant *Leishmania* amastigotes were found in lesions of her skin and left sclera. Because of treatment failure, she was referred to us and hospitalized.

The patient was treated at 1.5 years of age for a disseminated infection with bacille Calmette-Guérin for two years and showed a good response. Results of immunologic analysis for serum immunoglobulins, a nitroblue-tetrazolium test, a complement CH50 test, and flow cytometry of leukocytes were inconclusive. No other pertinent findings were detected in her past history and family history.

The result of an indirect immunofluorescent antibody test for VL was positive with a titer of 1:1,024. A leishmanin skin test (LST) result was positive. Kinetoplast DNA of *L. tropica* was detected by a specific polymerase chain reaction on whole blood, bone marrow, and skin biopsy specimens. Results were compared with those of reference strains *L. infantum* (MCAN/IR/96/LON-49) and *L. tropica* (MHOM/ IR/89/ARD-2). Parasites isolated from cultures of skin biopsy specimens and characterized by isoenzyme analysis with five enzymatic systems (glucose-6-phosphate isomerase, phospho-
glucomutase, nucleoside hydrolase, malate dehydrogenase, and glucose-6-phosphate dehydrogenase) and polyacrylamide gel electrophoresis were identified as *L. tropica*.3

The patient had a hemoglobin level of 8.8 g/dL, a serum total protein level of 10.7 g/dL, and a globulin level of 8 g/dL. Despite positive PCR results, *Leishmania* amastigotes were not seen in bone marrow aspirates and biopsy specimens. Abdominal ultrasonography showed hepatosplenomegaly and multiple intra-abdominal lymph nodes. The patient was negative for human immunodeficiency virus.

The patient did not respond to treatment with amphotericin B (1 mg/kg/day for 45 days) and a four-month course of meglumine antimoniate and interferon-α. Therefore, a 28-day course of miltefosine was given. At the end of therapy, the skin lesions were flattened and disappeared after a four-month follow-up. The size of liver and spleen decreased significantly.

**DISCUSSION**

On the basis of multiple lesions, ulceration at the onset of disease, and positive LST results, the patient was diagnosed as a case of DCL. As in our patient, many reported cases of DCL had *Leishmania* amastigotes in skin lesions.2,4 In differential diagnosis, we considered two other forms of leishmaniasis in which a large number of skin lesions occur: diffuse cutaneous leishmaniasis and post–kala-azar dermal leishmaniasis. In diffuse cutaneous leishmaniasis, parasite-laden nodules do not ulcerate and the T cell response to *Leishmania* antigen is poor.5 Post–kala-azar dermal leishmaniasis, which is common in Sudan and India and has been reported from Iran, is characterized by macular, maculopapular, and nodular skin lesions and usually starts periorally in patients who have recovered from VL.6,7 Therefore, the skin lesions in our patient were characteristic of DCL accompanied by VL. Five cases of VL with DCL have been previously reported in southern Iran for which the cause has not been identified.8

The fact the patient had a positive LST result is unusual for a case of VL. The positive skin test result in our patient after recurrence may be caused by memory T cells generated during the first episode of the infection when the single skin lesion was detected. These T cells may participate in the cellular response in the skin test after the second episode of infection.

*Leishmania tropica*, which is typically more dermatotropic, is one of the most common causes of localized cutaneous leishmaniasis along the Mediterranean basin and in Iran. However, this parasite has been confirmed as the cause of VL in Iran and viscerotropic leishmaniasis among U.S. servicemen in Persian Gulf Conflict.9–11

The diagnosis of leishmaniasis associated with immunodeficiency cannot be rejected because we could not evaluate interferon-γ defects. There was no history of recurrent infection in our patient and immunologic abnormalities in her family but results of immunologic studies were inconclusive. To our knowledge, this is the first case of DCL accompanied by VL caused by *L. tropica* to be reported from southern Iran.

**FIGURE 3.** Raised superficial lesion on the nasal aspect of left sclera of a biopsy specimen of the patient. This figure appears in color at www.ajtmh.org.

Received January 17, 2008. Accepted for publication April 1, 2008.

Acknowledgment: We thank H. Khajehei for editing the manuscript.

Authors’ addresses: Abdolvahab Alborzi, Gholam R. Pouladfar, and Mohammad R. Kadivar, Professor Alborzi Clinical Microbiology Research Center, Shiraz University of Medical Sciences, Shiraz, Iran, Tel: 98-711-6470205, Fax: 98-711-6287071, E-mail: alborziiraj2004@yahoo.com. Mahdi Fakhar, Department of Medical Parasitology and Mycology, Mazandaran University of Medical Sciences, Sari, Iran. Mohammad H. Motazedian and Gholam R. Hatam, Department of Medical Parasitology and Mycology, Shiraz University of Medical Sciences, Shiraz, Iran.
REFERENCES


