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Acquired Immunodeficiency Syndrome-Related Visceral Leishmaniasis Presenting in a Pleural Effusion*

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Visceral leishmaniasis is increasingly reported in immunocompromised patients, including patients with AIDS. We report a case of visceral leishmaniasis in an AIDS patient who presented with pulmonary symptoms and bilateral pleural effusions. Histologic evaluation of pleural fluid and bone marrow revealed histiocytes with intracellular Leishmania amastigotes. Visceral leishmaniasis should be considered in AIDS patients with a significant travel history who present with unexplained pulmonary symptoms.

(Chest 1993; 103:648-49)

Infections with protozoal pathogens are common in AIDS. Recently, there have been increasing reports of reactivation of visceral leishmaniasis (kala-azar) in AIDS patients.1-3 We report an unusual case of visceral leishmaniasis in an AIDS patient manifesting as bilateral pleural effusions with no apparent splenomegaly.

CASE REPORT

A 46-year-old male homosexual with diffuse cutaneous Kaposis sarcoma had a two-month history of progressive weight loss, dyspnea, and cough productive of brownish sputum. Previous infections included hepatitis B, gonorrhea, amebiasis and giardiasis. He had traveled to Manila, Borneo, Saudi Arabia, India, Indonesia, Morocco and Cairo, and had lived in Athens, Greece, for five years prior to his return to Rochester, NY, for hospitalization.

On admission, oral temperature was 35.2°C and diffuse bluish-nodules and plaques were noted on the skin. Diffuse adenopathy was present, as well as a 3 x 4-cm pedunculated nodular mass in his posterior pharynx. There were clear signs of bilateral pleural effusion. Cardiac examination was normal. His liver span was 14 cm, but no splenomegaly or abdominal masses were detected. His stools were guaiac-positive. Pitting edema (2+) was present below the knees. Neurologic examination was, with the exception of diffuse weakness, within normal limits. The white blood cell count was 2.1 thousand/ccm with 80 percent polymorphonuclear cells, 16 percent lymphocytes and 4 percent monocytes. The hematocrit was 23 percent. Clinical chemistry values were normal. Sputum cultures grew 1+ yeast and herpes simplex virus. While breathing room air, he had a Po2 of 77 mm Hg, and a chest x-ray film showed large bilateral pleural effusions. Thoracentesis revealed a serosanguineous fluid with 111,000 red and 467 white blood cells/per cubic millimeter with a predominance of histiocytes. Blood chemistry analysis of pleural fluid included a glucose value of 113 mg/dl, a protein value of 4.8 g/dl and an LDH level of 158 IU/L (serum LDH, 170 IU/L). Histiocytes in the pleural fluid contained abundant Leishmania amastigotes (Fig 1), from which Leishmania promastigotes were cultured. Growth was insufficient to allow speciation. Bone marrow examination revealed macrophages containing similar intracellular amastigotes. The patient refused further evaluation and was treated with sodium stibogluconate (Pentostam) and periodic therapeutic thoracentesis.

Initially, pleural fluid decreased and after 10 days intracellular amastigotes no longer could be detected in the pleural fluid. However, the patient then developed elevated pancreatic enzyme levels and pleural fluid began to reaccumulate. Subsequently, his hematocrit value decreased and he developed hypotension, bilateral alveolar infiltrates and intractable seizures. The patient and his family refused all investigational efforts, and he expired less than one month after admission. Terminal blood cultures grew both Klebsiella pneumoniae and Mycobacterium avium intracellulare. An autopsy was refused.

DISCUSSION

Visceral leishmaniasis, usually caused by L donovani or

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Visceral leishmaniasis often presents with atypical features in the immunocompromised patient. Marked splenomegaly, which is a classic physical finding, was absent in 8 of 47 HIV-related cases in a recent review. Our patient presented prior to the availability of HIV testing, but had classic clinical criteria for AIDS. It is noteworthy that splenomegaly was also absent at the time of physical examination. Antileishmanial antibodies frequently are not detectable in HIV patients, whereas approximately 95 percent of immunocompetent patients will have high antibody titers. Bone marrow aspiration, however, has a very high yield in immunocompromised patients and several studies suggest that bone marrow aspiration with microscopic examination and culture for Leishmania is the best diagnostic procedure for visceral leishmaniasis in HIV-infected patients.

An unusual feature of this patient was the initial presentation of pulmonary symptoms and pleural effusions. The pleural fluid contained large numbers of intracellular amastigotes. Treatment with pentavalent antimonials resulted in disappearance of the organism and a sustained decrease in pleural effusions. Pleural fluid reaccumulated later, presumably secondary to pancreatitis. This location of Leishmania amastigotes is very rare, but has been reported previously in a similar immunocompromised patient. In a recent review, many HIV patients with visceral leishmaniasis died of diffuse respiratory disease of unknown etiology. One patient was found to have amastigotes in macrophages from a bronchoalveolar lavage specimen, and a second patient had amastigotes detected in lung tissue at postmortem examination. These findings suggest that, in the immunocompromised patient, Leishmania species may cause respiratory disease more often than previously recognized.

The course of visceral leishmaniasis in HIV patients often is chronic and subject to relapses. Treatment with pentavalent antimonials, which is the standard therapy in non-immunocompromised patients, often is ineffective in HIV patients. This finding is not surprising, given the usual role of cell-mediated immunity in the host response to leishmaniasis.

In summary, as the incidence of HIV increases in areas in which Leishmania species are endemic, visceral leishmaniasis is being recognized more frequently as an opportunistic infection in patients with AIDS. Therefore, the diagnosis of visceral leishmaniasis should be considered in AIDS and other immunocompromised patients returning from endemic areas with unexplained pulmonary or systemic illness.

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