A Giant Tuberculous Lymphangioma Extending From the Mediastinum to the Inguinal Region*

Eytan Z. Blumenthal, M.D.; Nathan P. Gottehrer, M.D.; Marcel Dollberg, M.D.; and Jonathan Halevy, M.D.

A 23-year-old Ethiopian new immigrant presented with a giant lymphangioma extending from the posterior mediastinum through the retroperitoneum, ending as a herniated fluid-filled sac in the inguinal region. Chylous fluid aspirated from within the lymphangioma was cultured positive for *Mycobacterium tuberculosis*. Considerable regression occurred following 6 months of antituberculous treatment. ***(Chest 1994; 105:1279-80)***

Lymphangiomas residing in the abdomen and thorax are rare tumors, usually found incidentally. We report a patient with a lymphangioma containing chylous fluid cultured positive for tuberculosis. To our knowledge, this is the first report of intrathoracic tuberculous lymphangioma.

Case Report

A 23-year-old Ethiopian man, who had immigrated to Israel 1 year prior to his hospital admission, presented with a right inguinal mass. The mass was noted 1 month before admission, and enlarged gradually. The patient complained of fatigue for several months and moderate weight loss, but had no fever, respiratory complaints, or any other systemic symptoms.

Two years before hospital admission, the patient suffered from a right shoulder abscess, later evolving into a chronic, intermittently discharging sinus in the posterior axilla. At the time, no therapeutic measures were taken. No other significant events were recalled in the patient's history. Family history included a father who presumably died of tuberculosis (TB).

Physical examination revealed a 10-cm fluctuating and completely reducible right inguinal mass, a 10-cm nontender fluctuating right abdominal mass, and a scarred sinus in the right posterior axilla. No lymph nodes were palpable.

Results of laboratory investigations included the following: anemia (hemoglobin, 9.5; mean corpuscular volume, 80), a sedimentation rate of 74 mm, and hypergammaglobulinemia (IgG, 2,100; IgA, 367). The white blood cell count was 6.1×10^9/L, with a normal differential cell count. Serologic tests for syphilis and HIV were negative. A PPD tuberculin test (1 U/ml) resulted in 10-mm induration after 48 h. The axillary sinus discharge was cultured negative for TB.

A chest radiograph showed a widened posterior mediastinum due to a paravertebral mass, and anterior scalloping of the dorsal and lumbar vertebral bodies. There was no evidence of acute or past pulmonary TB. A plain abdominal radiograph revealed displacement of bowel loops to the left, and a mass occupying the right side of the abdomen and pelvis. An abdominal ultrasound revealed a large cystic mass.

On computed tomographic scan (CT) scan, a very large elongated cystic mass of cylindrical shape was demonstrated, extending continuously from the superior-posterior mediastinum, down to the retroperitoneum and right inguinal region, displacing the right psoas muscle and bowel loops (Fig 1).

A diagnostic-therapeutic puncture of the inguinal mass yielded

*From the Department of Internal Medicine, Shaare Zedek Medical Center, the Hebrew University, Jerusalem, Israel.

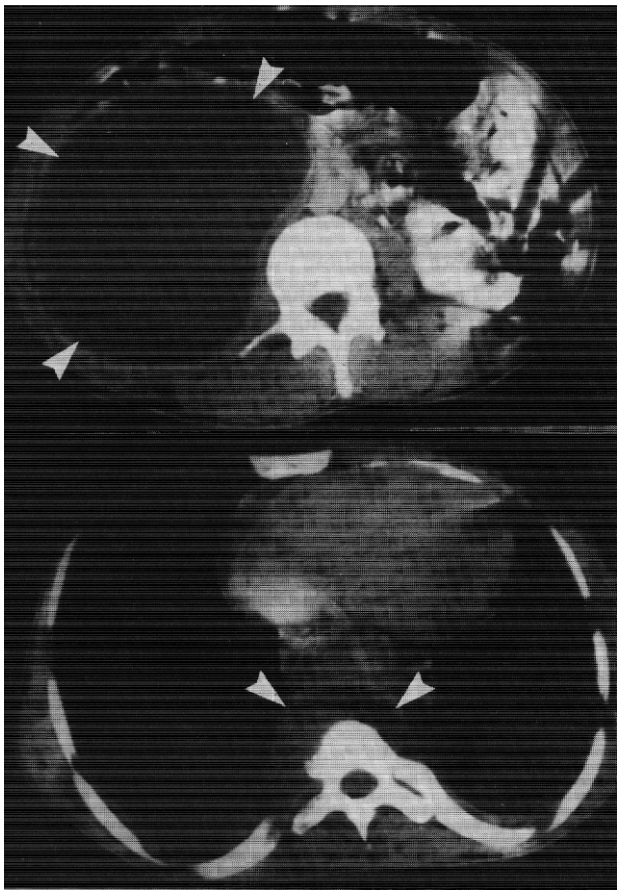

FIGURE 1. Computed tomographic scan, when the patient first presented: A *(top)*. Abdominal section; B *(bottom)*. Mediastinal section. Arrowheads delineate lymphangioma mass.

2,400 ml of a chylous aspirate, stained positive for neutral fat. The aspirate contained epithelioid cell aggregates and few neutrophils. A repeat CT scan showed considerable shrinkage of the mass, implying a continuous, communicating structure. *Mycobacterium tuberculosis* was cultured from the aspirate.

The patient was started on a regimen of the following: isoniazid, 300 mg/d; rifampin, 300 mg×2/d; and pyrazinamide, 1,250 mg/d. Six months following the commencement of treatment, a third CT scan revealed considerable improvement. The abdominal and inguinal segments of the mass disappeared almost entirely, while the mediastinal mass shrank to a third of its pretreatment size.

DISCUSSION

This patient was admitted to the hospital with a mass in the posterior mediastinum extending to the retroperitoneum and right inguinal canal. The mass was found to contain chylous fluid infected with *M tuberculosis*. Although this case lacks tissue biopsy of the lesion, both the fluid contents and the typical radiologic findings confirm the diagnosis of lymphangioma.

The association of lymphangioma and TB is extremely rare, with only two cases reported in the Medline literature since 1966.[1,2] In these cases, biopsy specimens that were taken from small lesions on the arm were diagnosed histologically as lymphangiomas and later cultured positive for TB. Although tuberculous chylous peritonitis[3] and tuberculous chylous ascites[4] have been reported, no case is described, (to our knowledge) in which lymphangioma infected with TB resided in the thorax or abdomen.[5] The fact that considerable regression of the mass occurred during a 6-month period of antituberculous treatment makes this case unique.

REFERENCES

1 Frahm R, Wimmer B, Goth D. Tuberculous bilocular cystic hygromas of the right hand and forearm. Radiologe 1989; 29:454-56

2 Gay B. Tuberculous hygroma of the hand. Beitr Orthop Traumatol 1971; 18:596-601

3 Jhittay PS, Wolverson RL, Wilson AO. Acute chylous peritonitis with associated intestinal tuberculosis. J Pediatr Surg 1986; 21:75-6

4 Howard PH, Knudsen KB, White RR. Chylous ascites due to retroperitoneal tuberculous lymphadenitis. Surg Clin North Am 1972; 52:493-96

5 Roisman I, Manny J, Fields S, Shiloni E. Intra-abdominal lymphangioma. Br J Surg 1989; 76:485-89