Hepatosplenic Schistosomiasis Presenting as Spontaneous Hemoperitoneum in a Filipino Immigrant

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Abstract: Hepatosplenic schistosomiasis is due to chronic parasitic trematode infections with various *Schistosoma* sp. The *Schistosoma* life cycle requires contamination of surface water by infected human or animal excreta, specific freshwater snail intermediate hosts and human skin contact with water. The disease is prevalent in many developing tropical areas, particularly in sub-Saharan Africa as well as in Southeast Asia. Deposition of *Schistosoma* eggs in the hepatic portal system leads to periportal fibrosis, cirrhosis and portal hypertension but little hepatocellular damage. Portal hypertension of any etiology may cause gastrointestinal varices. Rarely, ectopic varices may rupture into the peritoneal cavity and result in a hemoperitoneum. The authors describe a case of a Filipino immigrant who presented with a hemoperitoneum associated with previously unrecognized hepatosplenic schistosomiasis due to *Schistosoma japonicum*.

Key Indexing Terms: Hepatosplenic schistosomiasis; *Schistosoma japonicum*; Hemoperitoneum; Portal hypertension. **[Am J Med Sci 2013;346(4):334–337.]**

INTRODUCTION

Schistosomiasis, caused by the blood-dwelling flukes of the genus *Schistosoma*, is one of the most common tropical parasitic diseases in the world.[1,2] Both the burden and the duration of infection contribute to clinical disease. Chronic schistosomiasis often involves the liver, causing hepatosplenic schistosomiasis with subsequent cirrhosis, portal hypertension, splenomegaly and gastrointestinal varices. Here, we present what we believe is the first reported case of hemoperitoneum associated with hepatosplenic schistosomiasis.

CASE REPORT

A 26-year-old Filipino woman, who immigrated to the United States 3 years before, presented to a primary care physician to establish care. A routine blood test revealed thrombocytopenia (45×10^9/L) without anemia (hemoglobin of 12.8 g/dL). A hematology consultant diagnosed immune thrombocytopenic purpura and recommended corticosteroid therapy, but the patient did not begin treatment. Over the ensuing month, the patient noted the gradual onset of diffuse abdominal discomfort; 3 days before admission, she had a progressive increase in abdominal pain and was hospitalized. Three years earlier, she was diagnosed with latent tuberculosis for which she completed a 9-month course of isoniazid therapy without complication. She had no history of prior liver or gastrointestinal disease. Her medical history was otherwise unremarkable. She had no family history of liver or hematological diseases. She was born and raised in the Philippines. After moving to the United States, she was working in a candy factory. She rarely consumed alcohol. She denied use of tobacco or illicit drugs.

On physical examination, she was alert, oriented and in mild distress because of abdominal pain. She was afebrile, and her other vital signs were normal. She had diffuse mild abdominal tenderness, hepatomegaly, splenomegaly and shifting dullness but no jaundice or dermatologic stigmata of liver disease. The remainder of her physical examination was unremarkable.

A complete blood count showed a leukocyte count of 4.4×10^9/L, a hemoglobin level of 8.4 g/dL and a platelet count of 35×10^9/L. Liver-associated enzyme levels were normal except for a slightly elevated aspartate aminotransferase of 60 IU/L (normal range, 0–40 IU/L). Prothrombin time (37.0 seconds) and serum albumin (4.1 g/dL) were normal, and viral hepatitis serological tests were unrevealing. HIV antibody was negative. An ultrasound of the liver demonstrated heterogeneous liver echotexture and moderate ascites (Figure 1). Computed tomography (CT) of the abdomen with intravenous contrast was consistent with liver cirrhosis, portal hypertension, moderate volume ascites, splenomegaly and esophageal varices (Figure 2). On abdominal CT, the density of the ascitic fluid measured approximately 10 to 15 HU greater than the bladder fluid, suggestive of hemoperitoneum. The patient underwent paracentesis, which showed bloody fluid with a hematocrit of 27.8% (peripheral blood hematocrit, 27.9%) and an albumin of 1.7 g/L (serum ascites-albumin gradient of 2.4 g/L) consistent with hemoperitoneum and ascites from portal hypertension. Results of Gram stain, bacterial and mycobacterial cultures and cytology studies were all negative. The patient remained hemodynamically stable; however, she required 4 units of red blood cell transfusion and 24 units of platelet transfusion to maintain her hemoglobin level and platelet count.

A transjugular liver biopsy specimen unexpectedly showed normal hepatic parenchyma with calcified *Schistosoma japonicum* ova, confirmed by Centers for Disease Control and Prevention, Division of Parasitic Diseases and Malaria (Figure 3). Stool examination showed no ova or parasites. The patient was treated with praziquantel and referred to a liver center for the management of portal hypertension. However, the patient was lost to follow-up. We were unable to determine what region of the Philippines she had lived in or what risk factors she had for schistosomiasis.

DISCUSSION

Schistosomiasis affects approximately 207 million people worldwide, mainly those in subtropical and tropical countries.[3] Five species of *Schistosoma* cause human infections (*S mansoni*, *S japonicum*, *S mekongi*, *S intercalatum* and *S haematobium*). *Schistosoma mansoni*, *S japonicum* and *S mekongi* typically cause intestinal or hepatosplenic schistosomiasis, whereas *S haematobium* typically causes genitourinary schistosomiasis. *Schistosoma japonicum* infection occurs in China, Indonesia and the Philippines, and *S mekongi* is found in the Mekong river regions of Cambodia and Laos.[4] In the Philippines, the major foci for transmission are located on the

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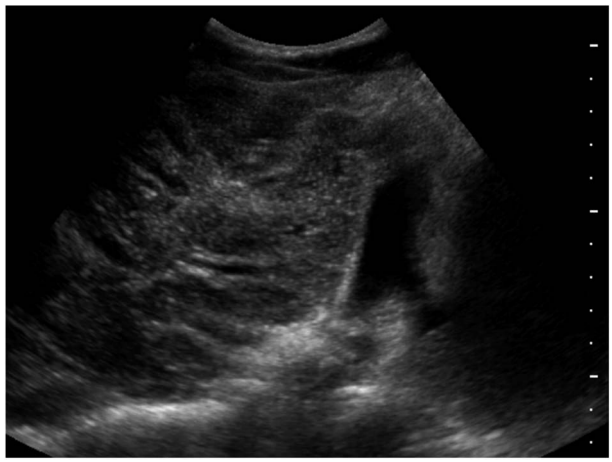

FIGURE 1. Ultrasound of the liver. Transverse image demonstrating left lobe with heterogeneous echotexture and moderate ascites.

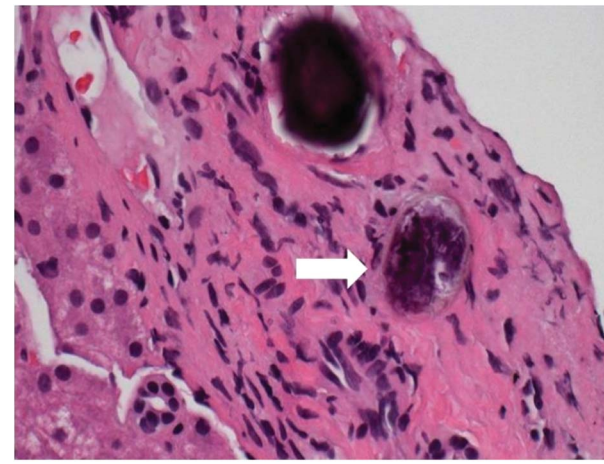

FIGURE 3. Biopsy specimen of the liver. A high-power view showing a calcified ova (arrow) consistent with *Schistosoma japonicum* (hematoxylin and eosin stain).

southern islands, Samar and Mindanao.[5] In the Philippines and China, an estimated 560,000 and 1 million people, respectively, are estimated to be infected.[6] In nonendemic countries, schistosomiasis may occur among travelers, refugees and immigrants from endemic areas.[7,8] However, physicians in nonendemic areas often are unfamiliar with schistosomiasis, and diagnosis is often challenging.

The parasitic life cycle of the *Schistosoma* is complex (Figure 4). Infected animal or human hosts excrete eggs into the feces or urine, which then contaminate surface freshwater. The eggs hatch into immature forms, which infect specific freshwater snail intermediate hosts, develop into cercariae, which then emerge from the snail, and contaminate surface freshwater. The infective cercariae swim, penetrate the intact skin of the human host, migrate through several tissues and stages and become adult male and female that dwell in the mesenteric venules. The adults mate and produce eggs that migrate through the gastrointestinal or urinary tract and are excreted into the aquatic environment to complete the life cycle.

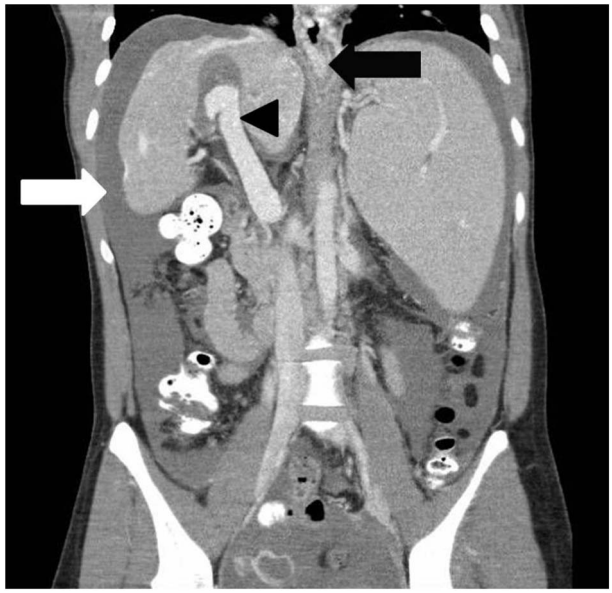

FIGURE 2. Computed tomography of the abdomen and pelvis with intravenous contrast. Axial image showing moderate amount of ascites (white arrow), dilated portal venous system (black arrowhead), splenomegaly and esophageal varices (black arrow).

Hepatosplenic schistosomiasis occurs when *S mansoni* and *S japonicum* eggs embolize via the hepatic portal system to the liver and evoke a host immune-mediated presinusoidal granulomatous inflammation. This leads to periportal fibrosis, progressive occlusion of the portal venous system and portal hypertension.[9] The typical clinical manifestations of chronic hepatosplenic schistosomiasis are splenomegaly, hypersplenism, collateral venous circulation, portacaval shunting and gastrointestinal varices. Imaging studies (ultrasound, CT and magnetic resonance imaging) are useful for diagnosis, particularly in endemic areas. Typical liver ultrasound features of hepatosplenic schistosomiasis include a diffuse coarse reticular pattern, periportal fibrosis and septum-like fibrous bands extending to the liver capsule.[10,11] Hepatosplenic schistosomiasis can mimic other causes of cirrhosis radiologically. However, as demonstrated in our patient, hepatic cellular function is typically preserved in schistosomiasis.[12]

Spontaneous hemoperitoneum is an uncommon condition but can be life threatening. The causes of spontaneous hemoperitoneum are classified into hepatic, splenic, gynecologic, vascular and coagulopathic etiologies. Rupture of visceral organs from trauma, liver masses and gynecologic diseases (ie, rupture of ovarian cyst or gestational sac of ectopic pregnancy) are the most common causes.[13] When hemoperitoneum develops spontaneously in patients with known ascites, it is usually related to the same disease process that caused the formation of ascites.[14] In the absence of a localized hematoma or hepatocellular carcinoma, a rupture of an abdominal cavity varix is the most likely diagnosis.[14] Although much rarer than intraluminal variceal rupture (ie, esophageal, gastric and duodenal), intraperitoneal rupture of ectopic varices has been described as a complication of portal hypertension.[15] Theoretically, any collateral venous circulation can be a culprit. There are reports of rupture of other intra-abdominal vessels, including the mesenteric vein, umbilical vein, omental vein, gallbladder wall vein and retroperitoneal veins.[15–19]

Hemoperitoneum has rarely been reported in schistosomiasis. Picaud et al[20] reported a case of hemoperitoneum from fallopian tube schistosomiasis from *S intercalatum*. However, we found no prior reports of hemoperitoneum associated with hepatosplenic schistosomiasis.