Schistosomiasis—An Unusual Cause of Abdominal Pseudotumor

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Schistosomiasis is a common parasitic disease in the tropical and subtropical regions of Africa, Asia, South America and the Caribbean. It is the second most common parasitic infection of humans after malaria. Acute and chronic clinical presentations of *S. mansoni* are well described. Presentation as a pseudotumor is considered rare. We present a case of a 58-year-old Nigerian who presented with recurrent abdominal pain and abdominal mass of one-year duration. Stool was negative for schisostomal eggs, but histological specimen obtained from surgical resection of part of the caecum showed *S. mansoni*. The case was treated successfully by excisional biopsy and praziquantel therapy. A review of the literature is discussed.

Key words: abdominal pseudotumor ■ Nigeria

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INTRODUCTION

Schistosomiasis—also called bilharziasis after the German tropical disease specialist, Theodore M. Bilharz, 1829–1862—is the second most common parasitic infection of humans after malaria.[1] Approximately 200 million people are infected globally in 76 countries and about 600 million are exposed to infection in tropical and subtropical regions of Africa, Asia, South America and the Caribbean.[1] Of all people suffering from schistosomiasis, 85% live in sub-Saharan Africa where *S. mansoni, S. haematobium and S. intercalatum are endemic. S. mansoni, S. intercalatum and S. japonicum* largely cause hepatobiliary and gastrointestinal symptoms, while *S. haematobium* causes urogenital symptoms. It is a chronic worm infection caused by a species of trematodes, the Schistosomes, which are blood flukes that parasitize the venous channels of definitive human hosts. The infection is transmitted by freshwater snails. Based on their organ localization, we may distinguish a urinary form from *S. haematobium*, and an intestinal or hepatosplenic form from *S. mansoni*, *S. japonicum*, *S. mekongi* and *S. intercalatum*.[2,3] *S. mansoni*, a main agent of the intestinal form, is a trematode that infects human beings more frequently and also infects other primates too.[4] Adult females of *S. mansoni* deposit the eggs in the small veins around the large intestine of infected individuals. Some of the eggs may be trapped in the gut wall or break loose into it and are then eliminated by defecation. The eggs trapped in the gut wall are responsible for inflammatory and immunopathologic responses, leading to erythema, edema, granulomas, ulcerations, hemorrhages and fibrosis. The infection is characterized by nausea, meteorism, abdominal pain, bloody diarrhea, rectal tenesmus, and hepatosplenomegaly.[5,6]

About a century ago, Symmers[7] classically described the characteristic gross picture presentation of hepatic pathology in advanced schistosomiasis, after performing autopsies in Egypt. The lesion resulted from the deposition of numerous schistosome eggs along the periportal tissues, which provoked chronic granulomatous inflammation with consequent fibrous expansion of the portal spaces and intrahepatic portal vein obstruction. The parenchyma usually maintains its normal architecture in a good correlation with the preservation of the normal hepatic function, as exhibited by the patients.[8]

Infection with *S. mansoni* is still a major health issue in Africa, Asia and South America,[9-13] but recent epidemiological studies showed that its prevalence increases in Europe and the United States due to international travelers and immigrants.[4]

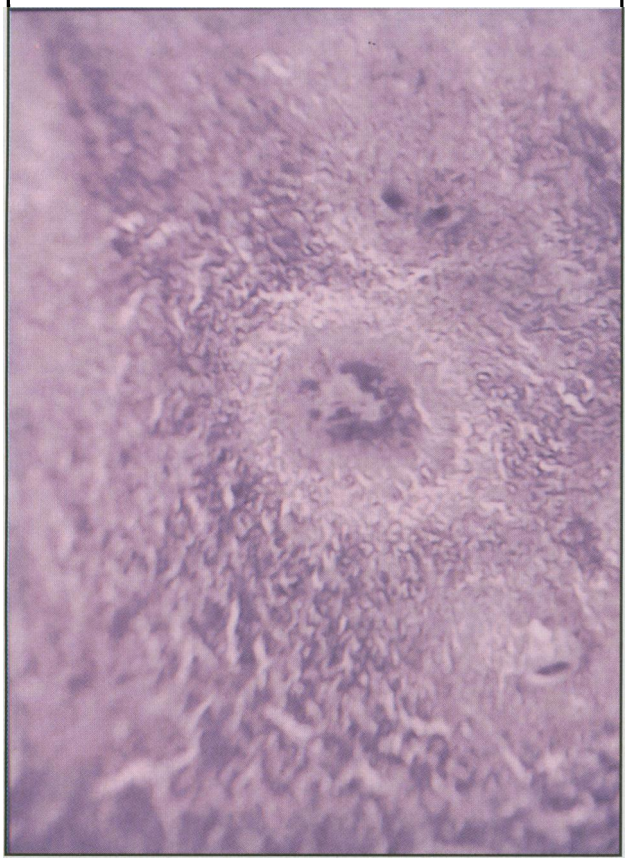
Acute clinical presentations include cercarial dermatitis and katayama syndrome presenting as fever, lethargy, myalgia, cough, rash, anorexia and headache.[14] Chronic presentations include gastrointestinal presentations (hepatosplenomegaly, periportal fibrosis, abdominal pain, diarrhea and polyposis) and renal symptoms (dysuria, frequency and terminal hematuria).[13] Others include cardiopulmonary manifestations (pneumonitis,

cough and sneezing) and neurological manifestations, including seizures, headache and myeloradiculopathy.[14]

Diagnosis of schistosomiasis is based on history and clinical suspicion followed by laboratory studies. These may include the enzyme-linked immunosorbent assay (ELISA), serum antigens and demonstration of the ova.[15] Many cases are diagnosed by endoscopy and biopsy. Schistosome ova may be seen in terminal urine, formol/ether-concentrated stools, and biopsy specimens. Microscopy has a specificity of 100% in expert hands but variable sensitivity. There are a variety of serological methods available for the diagnosis of schistosomiasis. The Hospital for Tropical Diseases in London (HTD) uses an ELISA with crude *S. mansoni* soluble egg antigen (SEA), which has a published specificity of 97% and sensitivities of 96% for *S. mansoni.*[16] The Centers for Disease Control screen sera by falcon assay screening test (FAST)-ELISA using *S. mansoni* adult worm microsomal antigen and, subsequently, confirm the results with a species specific immunoblot.[17] They reported excellent sensitivity and specificity for both the ELISA and the immunoblot. Recently, western blot analysis has been found to be a useful technique for the immunological diagnosis of schistosomiasis.[18]

We report a case of a 58-year-old Nigerian, living in a riverine area, who presented with recurrent abdominal pain and abdominal mass, stool negative for schisostomal eggs, but histological specimen obtained from surgical resection of part of the cecum showed *S. mansoni*. Presentation of *S. mansoni* as a pseudotumor is considered rare; hence, the case report.

Figure 1. Photomicrograph of the section obtained at surgery (H&E x10)



CASE HISTORY

A 58-year-old Nigerian fisherwoman presented with intermittent abdominal pain of one-year duration with recent exacerbation. She is a known peptic ulcer patient, diagnosed seven years ago, and maintained on cimetidine, antacids and anti-*Helicobacter* regimen. The recent exacerbation of the pain occurred three weeks ago, located in the right iliac fossa, colicky in nature but not referred to any organ and not relieved by posture or antacids. There were no known exacerbating or relieving factors. There was no epigastric tenderness. There was associated abdominal swelling located in the right ilica fossa region, associated with weight loss (despite good appetite), not associated with fever, vomiting or change in bowel pattern. There was no tenesmus, no hematochezia or melena stool. There was no tenderness at McBurney's point.

Physical examination revealed a chronically ill-looking woman, wasted (BMI=17.5 kg/m^2), mildly pale, not dehydrated, afebrile and anicteric. There were no significantly palpably enlarged lymph nodes or features suggestive of malnutrition. There was no pedal edema. Abdominal examination showed a right iliac fossa mass, firm and slightly tender. No peristaltic waves were seen. There were no palpably enlarged abdominal organs. Ascites were absent. Rectal examination was normal. Examination of other systems was essentially normal. An assessment of intraabdominal mass, possible appendical mass, was made to exclude an abdominal malignancy.

Laboratory investigations showed a mild anemia Hb =8.7 g%, normal total white cell count (FBC=8,200 cells/m^3) with eosinophilia (14%). Erythrocyte sedimentation rate was normal. Blood film examination showed microcytic hypochromic cells with poikilocytes. No target cells. Stool examination was negative for eggs of *S. mansoni* or any other parasite (ova or cysts). Stool culture was also negative. Electrolytes, urea and creatinine were essentially normal. Serum uric acid was 4 mg%. Liver function tests were normal. Mantoux test, chest x-ray and retroviral screening (ELISA) were normal. Abdominal ultrasound showed a right iliac mass extending to the periumbilical area.

Because there were no facilities for endoscopy or a CAT scan/MRI in the hospital, a surgical opinion was sought. The surgeons opined that an exploratory laparatomy would be beneficial at this stage. At surgery, a cecal mass with adhesions to the omentum was resected. Gross view showed an intestinal tissue matted together with a thickened wall in the cecal region. How-

ever, no peritoneal lavage for cytology was done. Histology showed a transmural acute on chronic inflammatory reaction with focal abscesses (Figure 1). An area of organizing inflammatory reaction was seen in the mucosa extending into the submucosa around calcified ova of *S. mansoni*. The serosal and surrounding tissue were deeply congested and edematous. The histopathological findings in the specimen were negative for chronic inflammatory bowel disease. Thus, a diagnosis of intestinal schistosomiasis was made, confirmed by the detection of *S. mansoni* eggs.

She had praziquantel (40 mg/kg, single dose), and was discharged 15 days after surgery. She remains healthy six weeks after discharge.

DISCUSSION

Schistosomiasis is a major health issue in the tropics and subtropics. Since the transmission of schistosomiasis is linked to the intermediate snail hosts, the disease is more prevalent in subjects whose skin is more susceptible to contacts with water in rivers, lakes, swamps or artificial irrigation systems. This may occur frequently in water-related activities of boatmen, fishermen and rice workers. The infection is also promoted by unsanitary disposal of urine and feces and can occur in animals such as dogs and cows.[19-21] Thus, the reported patient was at a high risk of occupational exposure, having been a fisherwoman.

The classical presentations (acute and chronic) of *S. mansoni* have been well described.[2,5,6,9,10] The symptoms of our patient, except for abdominal pain, are not consistent with those reported,[22] including nausea, meteorism, bloody diarrhea, rectal tenesmus and hepatosplenomegaly. Presentation as a pseudotumor is considered a rare presentation. Adult females of *S. mansoni* deposit the eggs in the small veins around the large intestine. Some of the eggs may be trapped in the gut wall or break loose into it and are then eliminated by feces. The eggs trapped in the gut wall are responsible for the inflammatory and immunopathologic responses, leading to erythema, edema, granulomas, ulcerations, hemorrhages and fibrosis[23,24]—and possibly in this patient, presentation as a fibrotic abdominal mass.

Abdominal tuberculosis, appendical mass, abdominal lymphoma and chronic inflammatory bowel diseases are all differentials diagnoses of this case. Chronic inflammatory bowel disease is very uncommon in Nigeria. Investigations done did not support a diagnosis of abdominal tuberculosis (mantoux test, ESR and chest x-ray) or a diagnosis of lymphoma (uric acid, etc.). Histological appearances in all these conditions differ from what was seen in the specimen of this patient. Finally, at surgery, no appendical mass was seen.

Our patient did not have fiber-optic endoscopic examinations because there are no such facilities presently in our hospital. Previous studies have, however, shown that colonoscopic findings are suggestive of schistosomiasis in about 45.3% of patients, but *S. mansoni* eggs in feces are detectable in only 11.1% of the patients with colonic biopsies positive for *S. mansoni*.[25,26] Endoscopic findings may be atypical, and it is often hard to distinguish Schistosoma infection from chronic inflammatory bowel disease, since these two disorders may show similar patterns. Chronic inflammatory bowel disease is uncommon in Nigeria as noted earlier. Endoscopic examination would have shown the inflammatory changes and would have provided opportunity for biopsy studies in the patient presented.

When a correct diagnosis of *S. mansoni* infestation is made, the complete recovery of the patient can be achieved using a specific antiparasitic therapy with praziquantel.[27,28] The intimate relationship between human beings and infected water leads to schistosomiasis-elevated prevalence. Elimination of the intermediate hosts and avoiding the contact with infected water sources are the measures to prevent schistosomiasis.[29] For the treatment, current drugs include praziquantel, metriphonate[30] and oxamniquine.[31]

We reported in this paper a case of an unsuspected schistosomiasis presenting as an abdominal pseudotumor. We strongly suggest that the detection of the Schistosoma eggs in feces should always anticipate colonoscopy examination; surgical options should always be considered as a last option when indicated. We conclude that Schistosomiasis should be considered in the differential diagnosis of patients with abdominal masses in the tropics.

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