

Colorectal Schistosomiasis: Report of Three Cases*

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SCHISTOSOMIASIS is prevalent in several Caribbean Islands; however, the Puerto Rican infestation rate of 14 per cent is of particular concern, as more than one million former inhabitants of Puerto Rico now reside principally in large urban areas of the mainland United States.

Three unusual cases of schistosomiasis seen on the surgical service of our 300-bed municipal hospital emphasized the variations of pathologic anatomy and clinical presentation and stressed the need to maintain clinical vigilance to detect this disorder.

Report of Three Cases

Patient 1: A 35-year-old Puerto Rican woman was first seen because of diffuse abdominal pain, vomiting, and scant diarrheal stools. She had been a resident of the United States for the preceding ten years. Past medical history included two years of peptic ulcer symptoms. An upper gastrointestinal series had been done elsewhere one week prior to admission.

The patient appeared cachectic, and her abdomen was distended. On abdominal examination the bowel sounds were decreased and moderate tenderness was present in all quadrants. No abdominal mass could be palpated. The rectal examination disclosed no abnormality, and the stool was negative for occult blood.

Hematocrit was 44 per cent. Leukocyte count was 3,700 per cu mm, with 80 per cent neutrophils, 19 per cent lymphocytes, and 1 per cent stab cells. Serum sodium was 134 mEq/l, potassium 3.0 mEq/l,

chlorides 93 mEq/l, blood urea nitrogen 15 mg/100 ml, glucose 3.3 mg/100 ml, alkaline phosphatase 43 IU/l, creatine phosphokinase 46 IU/l, lactic dehydrogenase 200 IU/l, SGOT 85 IU/l.

Abdominal roentgenograms showed barium from the prior upper gastrointestinal series filling the distended colon down to the distal sigmoid. The rectal gas shadow appeared narrow and constricted. Roentgenogram of the chest disclosed no abnormality.

Sigmoidoscopy revealed a rectal stricture 10 cm from the anal verge, with the lumen narrowed to 0.5 cm. Examination of a biopsy specimen obtained at the site of narrowing disclosed fresh schistosomal eggs in the submucosa.

The patient was treated with enemas and nasogastric suction. However, failure of nonoperative decompression necessitated performance of a right transverse colostomy 48 hours after admission. A four-week course of stibophen was given to treat the schistosomiasis. A barium-enema study done at this time revealed incomplete mechanical obstruction in the sigmoid colon. Diffuse mucosal edema extending to the lower descending colon was evident proximal to the obstruction (Fig. 1). Spot films of the sigmoid colon showed a sharp narrowing of the distal sigmoid with reduction of the lumen to the size of a pinhole (Fig. 2).

Two months after the transverse colostomy operation, an anterior resection of the rectosigmoid colon was performed to excise the distal sigmoid stricture. Examination of the resected specimen revealed a dilated proximal segment with muscular hypertrophy (Fig. 3). The area of stricture had a 1-2-mm orifice. Microscopic sections showed that the stricture was caused by fibrous replacement and marked puckering of the submucosa and muscle wall, with numerous fresh and old eggs of *Schistosoma mansoni* in the fibrous tissue. The overlying mucosa contained ova in various stages of extrusion, but was not ulcerated.

A barium-enema study one month later, prior to closure of the colostomy, established the presence of a patent anastomosis and normal-appearing mucosa. At follow-up examination one year later, the patient had no gastrointestinal complaint and had gained 30 pounds in weight.

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Patient 2: A 34-year-old black man was admitted because of massive upper gastrointestinal bleeding. At operation for control of a bleeding duodenal ulcer, the sigmoid colon was found to have multiple white, 1-2-mm subserosal nodular excrescences. Frozen-section examination of a biopsy specimen revealed numerous schistosomal granulomas within a thickened, fibrous serosa. Permanent section of an operative liver biopsy specimen revealed numerous schistosomal granulomas in various stages of evolution, as well as hepatic fibrosis. Following full recovery from the operation, the patient was treated with stibophen.

Patient 3: A 52-year-old Puerto Rican man, resident of New York City for the preceding ten years, was seen because of a full-thickness rectal prolapse, 5 cm long. The prolapse had occurred intermittently over the past five years but had always been reducible until two days prior to admission. Prior episodes had been accompanied by diarrhea, but at this time the patient had not had a bowel movement for two days.

Physical examination disclosed no abnormality except a markedly edematous 5-cm rectal prolapse. The rectal mucosa was obscured by a yellow-green

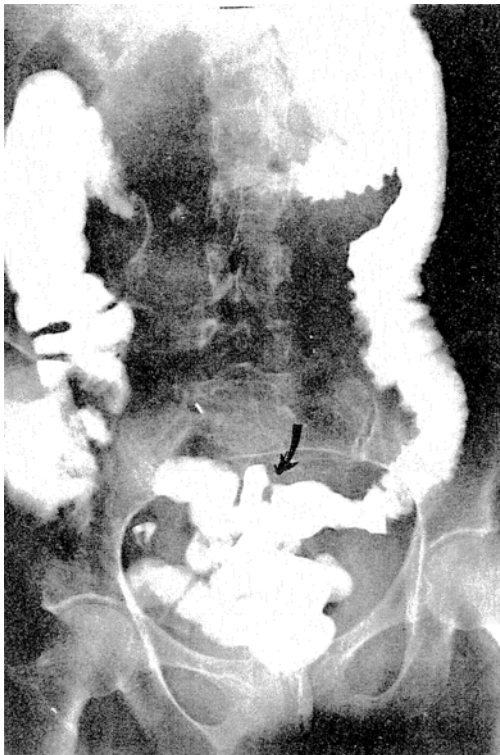


FIG. 1. Barium-enema study (Patient 1), showing incomplete mechanical obstruction of the sigmoid colon (arrow) with diffuse mucosal edema of the descending colon proximal to the stenosis.

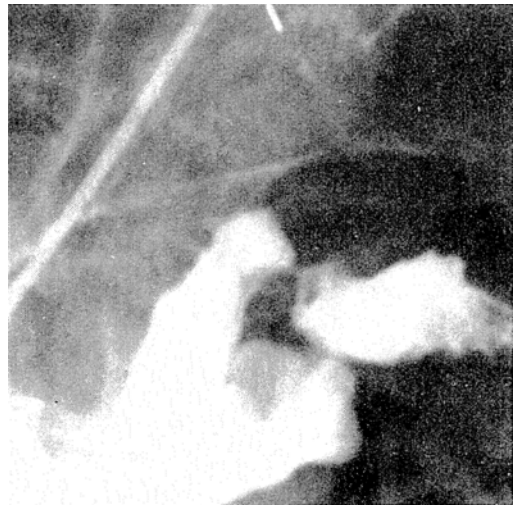


FIG. 2. Spot film of the sigmoid colon (Patient 1), revealing severe stenosis.

pseudomembrane (Fig. 4). Reduction of the rectal prolapse in the operating room was accomplished utilizing general anesthesia. A temporary Thiersch-type circumferential wiring at the rectal sphincter was done to maintain reduction of the rectal prolapse. Sigmoidoscopy at the time of surgery failed to reveal any abnormality above the prolapse. Multiple rectal mucosal biopsies disclosed necrotizing ulceration of the mucosa with schistosomal granulomas in the mucosa and submucosa. The patient was treated with stibophen and on follow-up examination, was found to be doing well, without recurrence of the prolapse.

Comment

The erythematous, papular skin rash associated with penetration of the parasite and the rare involvement of the central nervous system are only two examples of the myriad clinical manifestations due to infestation with schistosomiasis. Virtually every system of the body can be involved by this parasite.

The usual clinical picture of schistosomiasis previously seen at this hospital has included mucous, bloody diarrhea associated with the intestinal phase of parasitic oviposition. Presinusoidal hepatic cirrhosis, sometimes complicated by bleeding esophageal varices, has been the usual chronic manifestation encountered. That



FIG. 3. Resected rectosigmoid colon (Patient 1) with dilated proximal portion (right side), muscular hypertrophy, and two areas of stricture (arrows).

adult worms can live for 26 years or longer in the human host explains the recrudescence of the clinical intestinal disorder.¹ Moreover, a constant supply of eggs provides the etiology for the chronic visceral involvement exemplified by our first case.

Our first case illustrates the end result of many years of intestinal infestation by *Schistosoma*. Repeated cicatrization of acute inflammation of the bowel wall, subsequent fibrosis, and eventual hyalinization explain the rare occurrence of a stenotic, rigid bowel wall. The lesions associated with *S. mansoni* and *S. hematobium* infestation are usually localized in the descending colon or rectum, as in our patient. Stenotic intestinal lesions due to *S. japonicum* infestation can involve the entire large bowel and portions of the small intestine.^{1-3, 5-7}

Patient 1 had almost total colonic obstruction, and eventually required a colostomy. Barium-enema studies of patients who have schistosomiasis may reveal multiple small polypoid masses, especially in the sigmoid colon. There may be diffuse

bowel-wall induration with extensive pericolic infiltration.⁴ The radiologic findings are essentially those of a granulomatous colitis with strictures and polyoid lesions. Other disorders to be considered in the differential diagnosis are carcinoma, foreign-body granulomatous reactions, vascular occlusive disease, diverticulitis, pelvic inflammatory disease, carcinoid tumor, and metastatic disease. The finding of characteristic ova in the rectal biopsy specimen implicates schistosomiasis as the cause of a colonic stricture, but does not exclude carcinoma. A barium-enema study will help to differentiate the two disorders: a long, smooth, concentric stricture favors schistosomiasis, while an irregular, shorter stricture, particularly with overhanging edges, favors carcinoma. The colonoscope now provides the means for direct biopsy of these stenotic lesions in the colon. While it is possible that a carcinoma may be present in the area of stricture, there is no evidence to suggest that schistosomiasis predisposes to intestinal carcinoma. At operation the gross findings may be indis-

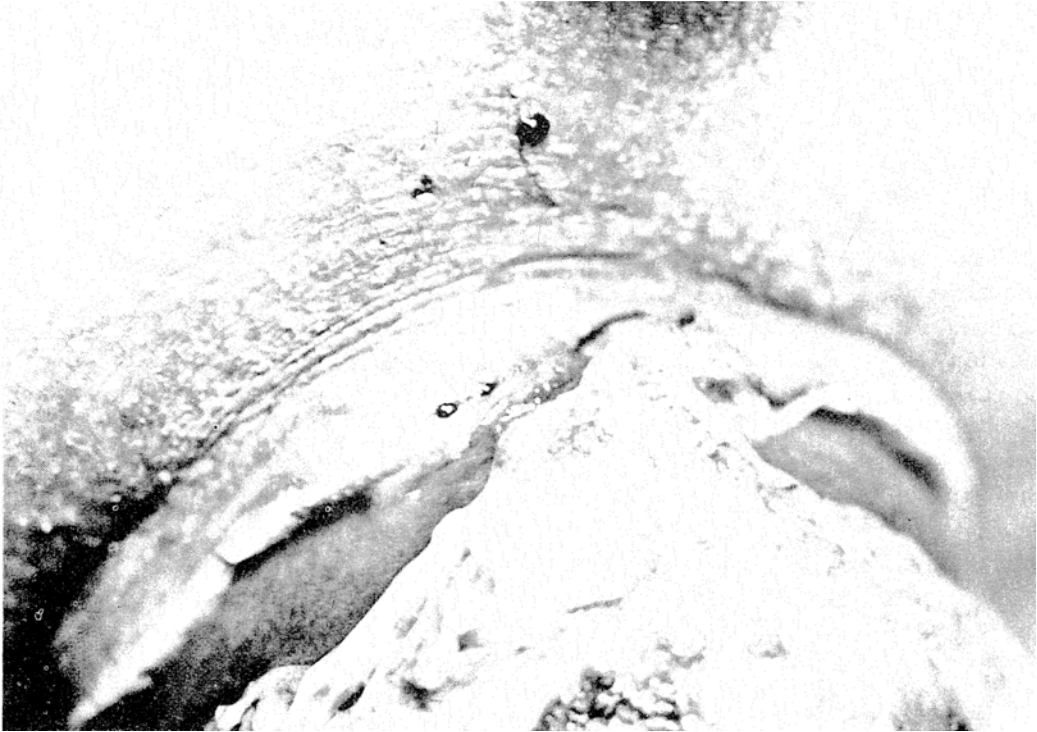


FIG. 4. Mucocutaneous junction of prolapsed rectum. The pseudomembrane was yellowish-green.

tinguishable from those of carcinoma, and only histologic study will verify the diagnosis. Earlier medical treatment could obviate the need for surgical intervention for stenotic lesions of the colon. However, an occasional patient such as ours will require operation.

The case of Patient 2 is presented to encourage consideration of schistosomiasis when unusual serosal inflammatory nodules are encountered during a celiotomy. Biopsy of the nodules is necessary to rule out other considerations such as tuberculosis or carcinomatosis. Although the ova of schistosomiasis usually reach the submucosa in retrograde fashion through venules, they can also lodge in the muscularis and serosa of the colon and incite a granulomatous response there. The latter involvement can produce an exudative focal peritonitis or lead to fibrous adhesions.

Neither of these complications was found in the second case presented.

An early manifestation of the disease is illustrated by our third case. The eggs within the submucosal mesenteric venules incite an inflammatory response, which progresses to rupture of the venules and release of eggs into the colonic submucosal area. The process results in a hyperemic, friable mucosa, frequently with ulceration, which liberates the eggs into the passing fecal stream. The severe mucosal inflammatory response causes intermittent acute or chronic diarrhea. The severe irritative diarrhea in this patient resulted in a massive rectal prolapse. Medical treatment with stibophen controlled our patient's severe diarrhea. Following surgical reduction of the rectal prolapse and proper medical therapy of the schistosomiasis, the prolapse did not recur.

Summary

The facility for mass movement of segments of our world populations creates the need for physicians to recognize and manage diseases not seen in native patients. Such a need has occurred in New York City, where schistosomiasis, with its protean manifestations has been seen with increasing frequency. The cases of three patients who had different clinical manifestations of infestation by *S. mansoni* are presented. Clinical, radiologic, and pathologic features are discussed. To avoid delay in treatment, physicians must consider the possibility of this disorder in cases of patients from areas of endemic schistosomiasis.

References

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Announcement

Symposium on Colorectal Carcinoma

A symposium on colon and rectal carcinoma, sponsored by The American Cancer Society and California Hospital Medical Center, Los Angeles, California, will be presented on Friday, March 26, 1976. This is a one-day session, which will include lunch, and among the outstanding speakers will be Drs. Rupert T. Turnbull, Jr., Maus W. Stearns, Jr., Charles G. Moertel, Kenneth R. Stevens, and others.

B. Richard Jackson, M.D., is Chairman of the symposium. For further information, correspondence should be directed to Kenneth R. Senter, M.D., California Hospital Medical Center, 1414 South Hope Street, Los Angeles, California 90015.