

Ileocolonic schistosomiasis resembling Crohn's disease

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GW GARDINER, A ZALEV, R WARREN. Ileocolonic schistosomiasis resembling Crohn's disease. *Can J Gastroenterol* 1995;9(6):345-348. A case of ileocolonic schistosomiasis mimicking Crohn's disease of the ileum is presented. Surgical resection of ileum was performed in a 40-year-old man because of intestinal obstruction with fistulas, which were diagnosed surgically and histologically as Crohn's disease. It was only with tissue review six months later that the diagnosis of ileal schistosomiasis was made. The difficulties in diagnosis and recognition of this disease are discussed.

Key Words: Crohn's disease, Schistosomiasis

Schistosomiase iléocolique ressemblant à la maladie de Crohn

RÉSUMÉ : Un cas de schistosomiase iléocolique imitant la maladie de Crohn au niveau de l'iléon est présenté ici. Une résection chirurgicale de l'iléon a été effectuée chez cet homme de 40 ans à cause d'une obstruction intestinale accompagnée de fistules, suivant un diagnostic chirurgical et histologique de maladie de Crohn. Ce n'est qu'à l'examen des tissus, six mois plus tard, que le diagnostic final de schistosomiase iléale a pu être posé. Les difficultés relatives au diagnostic et à l'identification de cette maladie sont présentés.

ALTHOUGH INTESTINAL SCHISTOSOMIASIS is well recognized in patients with a recent travel history, an insidious onset and long periods of remission may result in clinical omission of this entity from gastrointestinal differential diagnosis (1). Chronic schistosomiasis progresses to fibrostenosis similar to Crohn's disease and may assume similar radio-

graphic features. Furthermore, at the time of operation and gross pathological inspection, the opened bowel may look similar to Crohn's disease frequently seen in North America (2). We report a case of ileal schistosomiasis that was mistaken for Crohn's disease, and that resulted in surgery and significant delay in appropriate medical treatment.

CASE PRESENTATION

A 40-year-old Portuguese man who had lived in Canada for nine years presented with a three-year history of diarrhea, characterized by four loose stools daily, abdominal cramps and periodic vomiting. Abdominal examination disclosed a soft tender mass in the right lower quadrant. Intravenous and retrograde pyelography and abdominal ultrasonography disclosed right-sided hydronephrosis and hydroureter secondary to an irregular 4 cm stricture of the lower right ureter which was treated by stenting. An upper gastrointestinal small bowel series showed two narrowed segments in the distal ileum separated by a short dilated bowel segment. The more distal stenotic segment showed linear ulcers and sacculations (Figure 1).

Air contrast barium enema and colonoscopy to the cecum disclosed normal mucosa except for a symmetric, slightly narrowed segment extending 13 to 18 cm from the anal sphincter. Mucosal biopsies were superficial and interpreted as showing chronic non-specific inflammation. Ileal Crohn's disease complicated by hydronephrosis and hydroureter was diagnosed. Prednisone therapy was initiated and a short course of trimethoprim-sulphamethoxazole and metronidazole was administered.

Three months later, the patient was having one normal bowel movement daily and the abdominal mass was no longer detectable. A small bowel series

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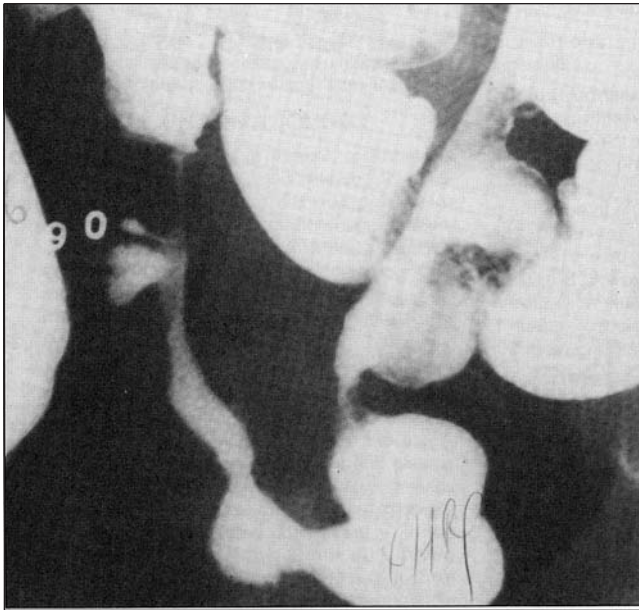


Figure 1) Spot view of distal ileum showing two narrowed segments with irregular contours and sacculations separated by a dilated segment

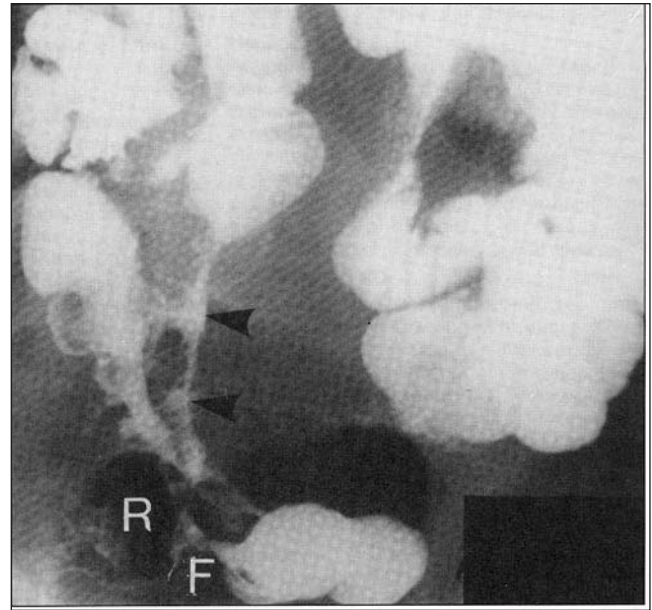


Figure 2) Spot view of distal ileum showing narrowed segment with deep linear ulcers (arrowheads). Ulceration has caused a fistula (F) to the rectum (R)

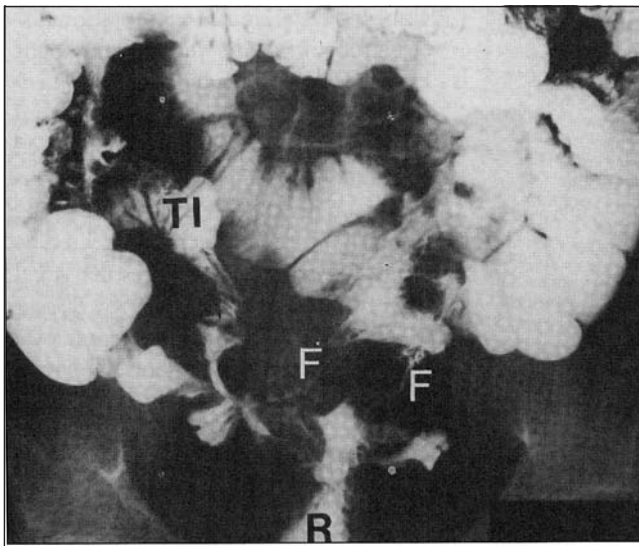


Figure 3) Overhead view of small bowel showing a normal terminal ileum (TI). The rectum (R) fills via a fistula (F). Ileo-ileal fistulas have developed and the loops are separated because of mural thickening and mesenteric inflammation

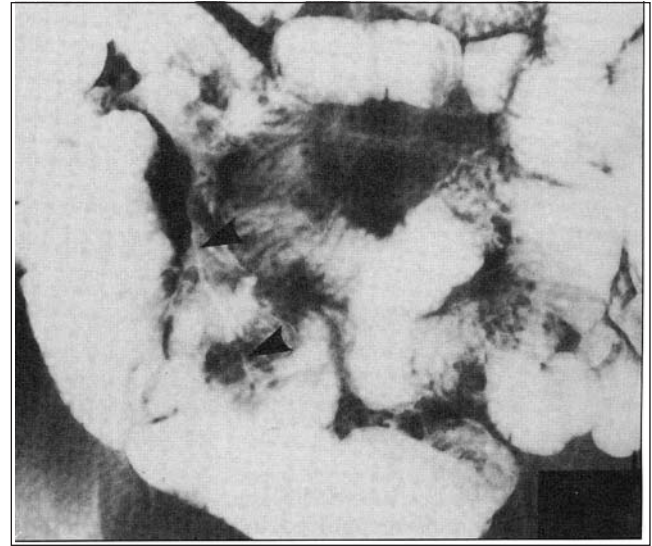


Figure 4) Spot view of ileocecal region showing linear ulcers (arrowheads) in the terminal ileum

showed that the proximal stenotic segment in the distal ileum had narrowed further. Deep linear ulcers and a fistula from this segment to the rectum were noted (Figure 2). Abdominal ultrasound showed complete resolution of the hydronephrosis and hydroureter.

Eight months later, a small bowel series showed that the previously dilated segment of distal ileum between the two stenotic segments had narrowed.

The ileorectal fistula was still present and an ileo-ileal fistula had developed. The terminal ileum was spared (Figure 3). Resection of the diseased segment of distal ileum and appendix was performed and the ileorectal fistula closed. The last 10 cm of terminal ileum and ileocecal valve were preserved because they grossly appeared normal. Pathological diagnosis based on fissuring ul-

ceration and transmural aggregated lymphoid hyperplasia histologically was Crohn's disease.

Six months later, because of the recurrence of diarrhea, the patient was referred for a second opinion. Laboratory findings were hemoglobin 132 g/L, white blood cell count 6000/mL with normal differential, serum albumin 3.9 g/L and serum globulin 3.2 g/L, with normal serum vitamin B12, folate lev-

els, aspartate aminotransferase, serum alkaline phosphatase and protein electrophoresis. A small bowel series showed new linear ulceration in the terminal ileum (Figure 4).

Histological review of the previously resected ileum showed deep fissuring ulceration with aggregated transmural lymphoid hyperplasia in a pattern typical of Crohn's disease; however, close scrutiny revealed the presence of schistosoma ova in the granulation tissue of fissures and embedded in the lamina propria (Figure 5). In spite of egg degeneration, the presence of a lateral spine was most consistent with *Schistosoma mansoni* infection. The appendix did not contain schistosoma ova. Flexible sigmoidoscopy showed granular rectal mucosa and mucosal biopsies of the rectosigmoid revealed the presence of schistosoma ova.

In view of these new findings, retrospective review of the patient's history before immigration was carried out. He was born and resided in the Portuguese Azores until 23 years of age when he moved to Lisbon for four months. He then served two years of military duty in Angola, living much of it in primitive conditions. Bathing and recreational swimming frequently occurred in inland waterways.

Praziquantel 20 mg/kg tid for one day was administered and cholestyramine 9 g bid prescribed. Six months later the patient was passing two formed stools daily, which would change to four loose stools if he discontinued cholestyramine. He gained 3.6 kg and abdominal ultrasound showed no evidence of hepatosplenomegaly.

DISCUSSION

Schistosomiasis is a major health problem affecting about 5% of the world population (1,3). Broad geographic areas including Africa, the Middle East, South East Asia and the Caribbean constitute endemic areas; with immigration and travel the incidence of schistosomiasis will increase in North America (1,3). Clinicians, radiologists and pathologists must recognize the lesions of this parasitic infestation outside endemic areas.

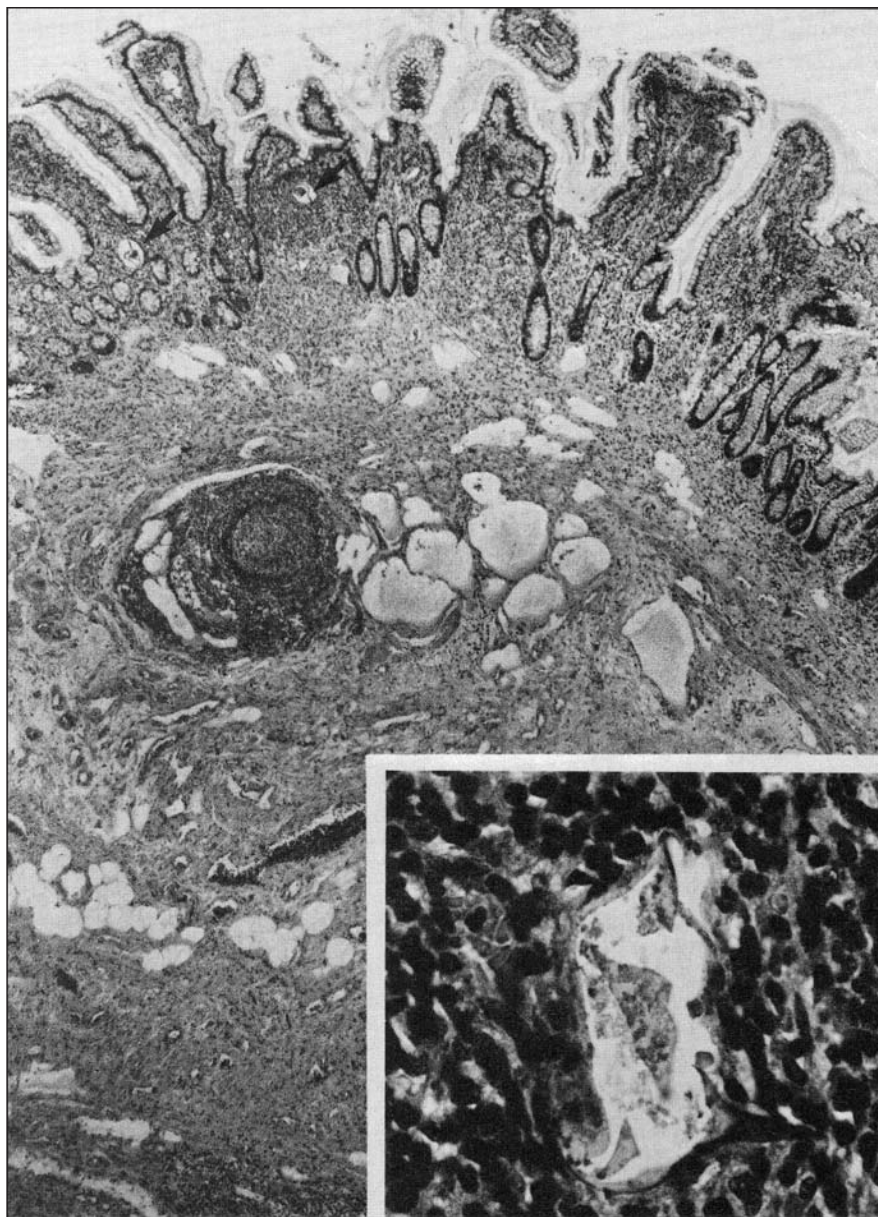


Figure 5) Photomicrograph of ileal mucosa showing an expanded submucosal plane with lymphoid hyperplasia and ova in lamina propria. Inset: *Schistosoma* ova with lateral spine

The blood flukes of *S mansoni* most commonly cause gastrointestinal disease and reside chiefly in the mesenteric veins of the large bowel. They cause disease by deposition of eggs (ova) in the distal venous plexus of the colon. Ova deposition in the intestinal wall may acutely cause ulceration with bleeding, diarrhea and protein loss. The severity of the disease relates to the intensity and duration of the egg burden which progresses from an active to an inactive stage. Furthermore, egg deposition is uneven and may randomly affect any part of the gastrointes-

tinal tract. At endoscopy, heavy egg burden may be associated with 'sandy patches' of the mucosa. Histologically, ova deposition incites a paraovarial granulomatous inflammation with lymphocytes, macrophages, giant cells and usually noticeable eosinophils. The schistosome granuloma is a T cell-dependent host reaction for which the detailed pathophysiology has been well elucidated in experimental animals (3). With down-regulation of the initial infection, only a small percentage of patients develop chronic complications such as fibrostenosis,

fistulization and polyposis (1). Pathologists must be astute to recognize ova as a cause of the granuloma or a misdiagnosis of Crohn's disease can occur, as is evident in this report. Speciation should be attempted by position of the spine of the ova (lateral spine in *S mansoni*), and Z-N staining may accentuate the egg capsule. Negative stools for ova and parasites do not exclude the disease.

Ulcerative colitis occurring in a patient with chronic colonic schistosomiasis has been reported (4). However, schistosomiasis superimposed on Crohn's disease or vice versa has not, to our knowledge, been reported. The presence of a heavy egg deposition in the area of diseased bowel in our case excluded the diagnosis of Crohn's disease. Only a single case report of intestinal schistosomiasis resembling Crohn's disease could be found in the

literature, done by Mynors in 1957 (2). His report was very similar to the current case except that in Mynors's case the disease process destroyed the ileocecal valve and involved the cecum. More recently, ileocolonic schistosomiasis mimicking clinical lymphoma has been reported, with resolution of a right lower quadrant mass after a negative fine needle aspirate and treatment for schistosomiasis detected by rectal biopsy (5). Iyer et al (6) have reported a case of intestinal obstruction in the ileocecal area that was due to schistosomiasis mistaken clinically for colonic carcinoma.

Intestinal schistosomiasis may have many radiological features in common with Crohn's disease (7,8). Extensive transmural disease characterized by deep ulcers, fistulas and stricturing limits the differential diagnosis to Crohn's disease, radiation injury and three

granulomatous infections: schistosomiasis, tuberculosis and actinomycosis. The latter two infections usually show extensive cecal involvement, which was not a feature in this case. Amebiasis is usually limited to the colon. Because of the patient's exposure in an endemic area, schistosomiasis must be included in the radiographic differential diagnosis. Unlike urinary bladder schistosomiasis (*Schistosoma haematobium*), where calcification of dead ova is common, calcification of intestinal ova of *S mansoni* is rare and was not seen in our case. Mural calcification of the large bowel in schistosomiasis has recently been recognized by computed tomography scanning (9).

We emphasize that schistosomiasis can be an imposter of Crohn's disease and can easily be overlooked or misdiagnosed.

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