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Large bowel obstruction secondary to schistosomiasis-related colonic stricture

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ABSTRACT

INTRODUCTION: Intestinal involvement of schistosomiasis uncommonly involves the formation of non-obstructive polypoid lesions; however, obstructing fibrotic stenoses and strictures secondary to chronic infection are extremely rare with only nine reported cases in the literature.

PRESENTATION OF CASE: An 85-year-old Southeast Asian female originating from the Philippines presents with a one-day history of obstructive symptoms in the setting of chronic constipation over the past four months. Subsequent CT imaging and colonoscopy biopsy revealed a nodular cecal mural wall thickening with chronic inflammation and a single *Schistosoma* egg. Despite treatment with praziquantel, and medical optimization the patient did not improve. Additionally, a malignancy as the underlying cause of obstruction could not be ruled out as such, she had a right hemicolectomy. Final pathology confirmed the diagnosis of intestinal submucosal schistosomiasis causing fibrotic stenosis.

CONCLUSION: Obstructing lesions including fibrotic stenoses secondary to Schistosomiasis infection can be managed safely with medical co-morbidity optimization when possible, treatment with Praziquantel and surgical resection of the involved area of colon. Given the risk of malignancy and the inability to clinically distinguish between infectious and neoplastic processes, surgical management is recommended.

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1. Introduction

Schistosomiasis is a chronic infection caused by trematodes, which develop in the human host after exposure to fresh water snail larvae [1]. While uncommon in North America, schistosomiasis affects approximately 200 million people globally and is endemic to 70 countries, where it accounts for approximately 300,000–500,000 deaths annually [1,2]. Schistosomiasis is asymptomatic in 30–40% of cases [1,3]. More commonly, it presents with non-specific symptoms related to affected organs, chronic anemia, or malnutrition [1]. Rarely, the deposition of *Schistosoma* ova can cause severe symptoms such as portal venous stricture leading to portal hypertension, or colonic stricture leading to bowel obstruction [1,4]. Treatment of uncomplicated Schistosomiasis is accomplished with a single 40–60 mg/kg dose of Praziquantel, an anthelmintic drug, however, pathology related to egg deposition may require surgical intervention [1].

In this case report we present a rare case of large bowel obstruction (LBO) due to severe intestinal schistosomiasis causing a colonic fibrotic stricture seen in a non-endemic region. The formatting of

this report is congruent with the Consensus Surgical Case Report (SCARE) criteria [5].

2. Case report

An 85-year-old Southeast Asian female who immigrated from Philippines in 1987, with a past medical history of hypertension, chronic kidney disease, depression, 50-pack year smoking history, and a previous squamous cell carcinoma of the lung requiring surgical resection, presented to tertiary-care academic center with abdominal pain.

The onset of acute pain leading to her presentation had started 12 hours prior to arrival to the emergency department. Although mild and generalized diffusely, the most prominent pain was localized to the right lower quadrant. She described obstructive symptoms including five episodes of non-bloody, non-bilious emesis, persistent nausea, and abdominal bloating. She had a small bowel movement and passed flatus the previous day. She denied any systemic symptoms of fevers, chills, or night sweating but had lost approximately 10–15 pounds over a four-month period. Although, the patient had chronic constipation and intermittent mild abdominal pain during the same time period. On physical examination, she had a blood pressure of 123/58 and was slightly tachycardic at 105 beats per minute. Her abdomen was soft, moderately distended and non-tender with no palpable organomegaly

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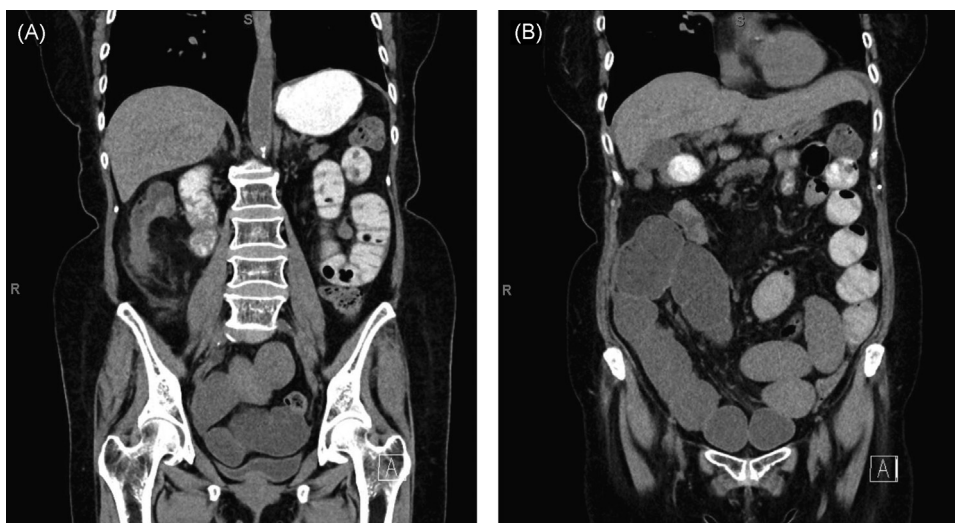


Fig. 1. (A) CT Abdomen and Pelvis illustrating nodular cecal mural wall thickening measuring up to 14 mm and eccentric mural thickening in the terminal ileum measuring up to 7 mm, with marked surrounding fat stranding and numerous prominent surrounding mesenteric lymph nodes measuring up to 8 mm in the short axis. The surrounding peritoneal lining in the right lower quadrant appeared thickened and nodular. (B) The small bowel was diffusely dilated up to 4.9 cm in diameter with air-fluid levels, worst at the terminal ileum proximal to the cecal thickening.

or masses. Digital rectal examination was normal with adequate rectal tone and no masses or blood noted. The patient's relevant laboratory investigations included a white blood cell count of 30.7×10^9 cells/L, hemoglobin on 106 g/L and a creatinine of 231 $\mu\text{mol/L}$; however, her baseline estimated glomerular filtration rate was 14 mL/min. The initial computed tomography (CT) of her abdomen showed nodular cecal mural wall thickening associated with a partial proximal large bowel obstruction with associated small bowel dilatation (Fig. 1).

Given the acute on chronic and partial nature of her LBO, she was placed on conservative management, including bowel rest, nasogastric suction, and intravenous fluid resuscitation. Additionally, as the etiology of her LBO was concerning for a malignancy, we requested a complete colonoscopy, biopsy of the mass, screening chest CT to rule out possible metastatic disease, and a carcinoembryonic antigen (CEA) level. Although, the latter tests were normal, her colonoscopy showed a colonic stricture and lesion proximal to the hepatic flexure with significant associated mucosal edema. The lesion was concerning for a malignancy; therefore, biopsies were taken that demonstrated colonic mucosa with chronic architectural distortion and ulceration, with a single parasitic egg identified that was morphologically compatible with Schistosome species.

The initial infectious workup revealed no ova or parasites in the stool cultures, nonreactive *Schistosoma mansoni*, *Schistosoma haematobium*, and strongyloides antibodies by enzyme immunoassay. However, in consultation with Infectious disease team, given the high suspicion of schistosomiasis as the cause of her colonic stricture, she was treated with 3 doses of praziquantel 900 mg (60 mg/kg). She was subsequently monitored carefully for improvement of her obstructive symptoms over the next week. Furthermore, given her poor nutrition status prior to hospitalization and ongoing bowel rest for five days, we started the patient on total parenteral nutrition (TPN). Despite medical treatment of her schistosomiasis and a week of non-operative management, she did not have substantial improvement of her abdominal symptoms. Therefore, after an interdisciplinary meeting with the patient and her family, a decision was made to schedule her for a right hemicolectomy.

The following day, she was taken to the operating room where she had a laparoscopic converted to open right hemicolectomy. The intraoperative findings mirrored the pre-operative imaging and

colonoscopy, namely a thickened cecum with surrounding inflammatory changes leading to dense adhesions and a bulky mesentery with associated enlarged lymph nodes. Final pathology showed intestinal submucosal schistosomiasis with dense distorting fibrosis, focal mucosal ulceration, architectural distortion of the mucosal crypts and villi, and easily identified calcified eggs (Fig. 2). Notably, the margins were free of disease, and no malignancy or granulomata were identified.

Post-operatively, the patient did remarkably well as she was quickly weaned off her TPN and was tolerating a normal diet. As per the infectious disease team, given the intraoperative findings and final pathology results of severe disease, the patient received an additional 3 doses of 900 mg of praziquantel prior to discharge. At her one-month follow up, the patient reported a return of her normal functioning with no further obstructive symptoms.

3. Discussion

We report a case of acute on chronic partial large bowel obstruction secondary to schistosomiasis-related inflammatory stricture and fibrotic stenosis. Despite treatment with praziquantel, and medical optimization with nasogastric tube suction, TPN, and bowel rest, the patient's abdominal symptoms and obstruction did not improve. Additionally, although the colonoscopy biopsies did not reveal malignant changes or cells, a neoplastic process as the etiology for her obstruction could not be ruled out; hence, the team elected to employ operative management with a right hemicolectomy.

Intestinal involvement of schistosomiasis is rare but most commonly involves the formation of non-obstructive polypoid lesions through the deposition of schistosomal eggs in the superficial layers of submucosa that progresses to the build-up of reactive cellular debris and inflammatory granulation tissue. Eventually, the adjacent layers of the muscularis mucosa hypertrophy and form a nodular polyp [6]. These schistosomiasis-related hypertrophic polyps are usually amenable to medical management with therapies including praziquantel. However, schistosomiasis inducing obstruction with stenosis is exceedingly rare even in endemic regions, therefore, our case is a unique addition to the current literature.

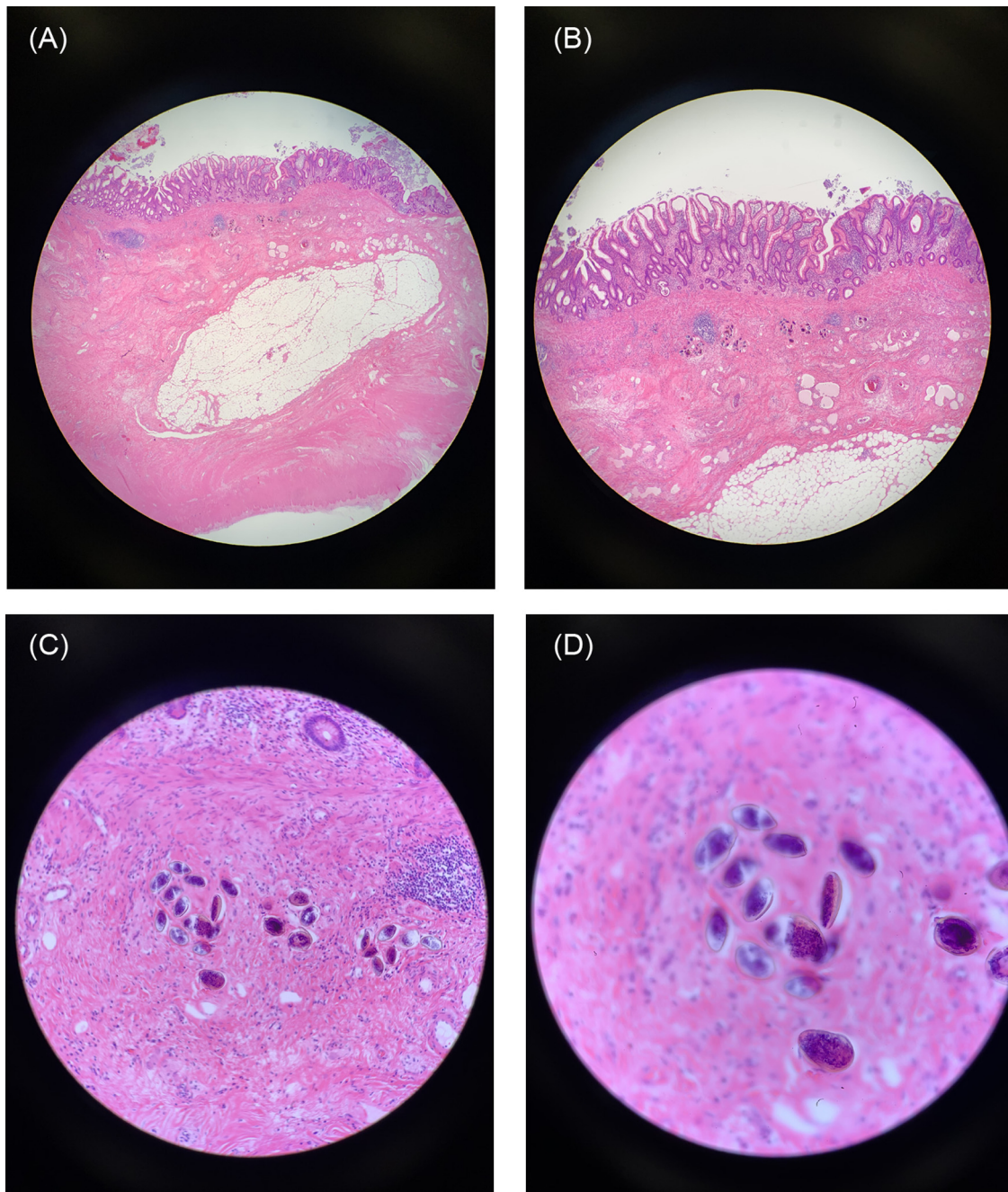


Fig. 2. (A) Cross-section through colonic stricture showing fibrosis and thickening of the muscularis mucosa, submucosa, and muscularis propria. (B) Calcified Schistosomiasis eggs are identified within the submucosa, (C) associated with a mild, predominately lymphocytic, inflammatory infiltrate. (D) Higher magnification of the Schistosomiasis eggs does not identify any prominent terminal spines.

Recognition of intestinal manifestations of schistosomiasis likely differ regionally based on the incidence of infectious complications of this disease process, and availability of diagnostic modalities. In endemic regions, the mainstay of diagnosis and identification of species relies on evidence of parasitic eggs in the stool. Schistosomiasis can also be diagnosed through findings parasitic eggs in tissue biopsies of concerning lesions attained through endoscopy, but often is limited by variance in egg shedding. Anti-schistosomal antibodies can be detected using serologic testing but cannot differentiate between active or previous infection. Additionally, immunoassays are expensive and potentially are limited to specific *Schistosoma* species. Radiologically, the use of abdominal x-rays and barium enemas can be effective in focally identifying polyps or strictures. Despite being easily accessible and low-cost,

these modalities suffer from poor sensitivity and specificity. The sensitivity does improve by using higher fidelity imaging tools such as CT scans.

In non-endemic regions with individuals immigrating from affected countries, some guidelines suggested screening for schistosomiasis as a way of improving identification of infected patients, increase treatment rates, and ultimately reduce any complications of disease progression [7,8]. Previous studies have found high rates of active schistosomiasis in immigrants from *Schistosoma* endemic regions, therefore screening this population with serology and ELISA testing may be effective [7]. However, future studies investigating the cost benefit analysis of establishing screening may prevent complications and long-term sequelae of schistosomal disease.

Table 1
Previously reported cases of large bowel obstruction due to Schistosomiasis.

Study	Age/ Sex/ Country of Origin	Clinical Findings	Imaging/ Surgical Findings /Diagnosis	Treatment
Hariton et al. (1966) [9]	27; M Venezuela	2 yr. history of abdo. pain and, alternating constipation and diarrhea with occasional blood in stool.	Abdo. XR: 4 strictures between transverse and descending colon.	Left hemicolectomy.
Atik et al. (1998) [4]	25; M Brazil	Initial hemodynamic instability, palpable hard LLQ mass, and peritonitis. 2 wk. history of cramping, distension, diarrhea with mucus, and anorexia.	Histopathology: <i>Schistosoma</i> . Histopathology: chronic granulomatous colitis with <i>Schistosoma mansoni</i> ova. Thickened rectosigmoid wall. Perforation of mid sigmoid colon.	Hartman's procedure. Also received single dose of Oxamniquine (15 mg/kg)
Nicodemus et al. (2001) [3]	30; M Zambia	Distention and tender LLQ. 2wk. history of abdo. pain, and 2d of vomiting.	Intra-operative: Stricture and adhesions in sigmoid colon. Histopathology: ulceration with <i>Schistosoma</i> ova.	Sigmoid stricture resection. Also received single dose of Praziquantel (40 mg/kg).
Bessa et al. (1979) [10]	3 cases Egypt ^a	3 patients were referred for General Surgery consultation for intestinal obstruction due to colonic schistosomiasis.	Obstruction secondary to stricture from schistosomiasis in all 3 cases. Two strictures were in the sigmoid colon, and one in the rectum.	Diverting transverse colostomy was deemed the safest treatment by consensus.
Herman et al. (2017) [8]	52; M Tanzania	4d history of abdo. pain, distension, and constipation.	Abdo. XR: LBO. Intra-operative: Proximal sigmoid fibrotic stricture Histopathology: Adenocarcinoma + <i>S. mansoni</i> ova.	Sigmoidectomy
Cadwallader et al. (2011) [2]	44; M South Africa	Short history of symptoms consistent with large bowel obstruction. [ref] HIV positive at time of presentation.	Abdo. XR: LBO. CT: left-sided colonic stricture. Intra-operative: inflammatory stricture, vascular adhesions, and small perforation in descending colon. Histopathology: granulomatous colitis + <i>S. haematobium</i> ova.	Transverse colostomy followed by extended left hemicolectomy when patient improved. Also treated with Praziquantel.
Iyer et al. (1985) [12]	47; M Puerto Rico	48 h. history of generalized abdo. pain, nausea, bilious emesis, and early satiety. 6-months of vague diffuse abdo pain., 35 lb weight loss, and diarrhea with mucus.	Intra-operative: Small white lesions throughout abdo. 6 cm long obstruction from ileum to cecum. Histopathology: cellular and/or fibrosed granulomas. A small bowel sample contained <i>Schistosoma</i> ova.	Ileo-colic resection with side-side anastomosis. Also received single dose of Oxamniquine (14 mg/kg).

Acronyms – Abdo: abdomen, LLQ: left lower quadrant, LBO: large bowel obstruction, CT: computer tomography, XR: X-ray.

^a Study completed in Egypt, individual patient countries of origin not documented.

Given the rare presentation of partial colonic obstruction due to fibrotic inflammation, there are no clear guidelines regarding treatment recommendations. Usually, management of intestinal schistosomiasis involves a single oral dose of 40–60 mg/kg of praziquantel, a safe and effective drug that achieves cure rates between 70–90%. However, identification of *Schistosoma* in colonic polyps or fibrotic strictures, does not rule out the possibility of underlying neoplasm [9]. Therefore, management has been based on expert opinion and published case reports (Table 1) where resection is used to definitively treat the intestinal manifestation of schistosomiasis and confirm the diagnosis with pathological analysis. Furthermore, utilizing a surgical approach is supported by literature suggesting that once a Schistosomal polyp has fully developed, they are not amenable to reduction through medical management [10]. Praziquantel is used in most cases to treat the underlying infection and avoid future sequelae of the disease process [1–3]. Given the inability to distinguish between *Schistosoma* and malignancy, and the inherent underlying risk of future malignancy, surgical resection remains the treatment of choice, as employed in our patient [11,12].

4. Conclusion

In summary, this case report described a patient who presented with a partial large bowel obstruction secondary to *Schistosoma*.

Schistosomiasis should be kept on the differential for immigrant populations from endemic regions. Praziquantel remains the treatment of choice for uncomplicated schistosomiasis. Although rare, if a *Schistosoma* forms obstructing polyps or fibrotic strictures, it must be surgically removed because of the inability to clinically distinguish *Schistosoma* from malignancy.

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Ethical approval

Ethics Approval was not required for this manuscript.

Consent

The patient gave her verbal and written informed consent to be included in this report as well as for publication of anonymized data and images. This can be provided to the editors, if required.

Author contribution

Case Supervision and Study Conception: D'Souza and Garraway; Literature Search: D'Souza and Birnie; Data Collection: D'Souza and Birnie; Data Analysis: D'Souza, Birnie and Garraway; Writing:

D'Souza and Birnie; *Critical revision*: D'Souza, Birnie and Garraway; *Approval of Final Manuscript*: D'Souza, Birnie and Garraway.

Registration of research studies

None. As the submitted work is a case report, there was no study involved.

Guarantor

Dr. Karan D'Souza, Mr. Blake Birnie, and Dr. Naisan Garraway take responsibility for the integrity of the work and the accuracy of the manuscript.

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Declaration of Competing Interest

None.

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