American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2021; 22: e930439
DOI: 10.12659/AJCR.930439

Received: 2020.12.12
Accepted: 2021.03.05
Available online: 2021.04.11
Published: 2021.05.11

Colorectal Schistosomiasis Infection After Cytoreductive Surgery and Hyperthermic Intraperitoneal Chemotherapy for Recurrent Metastatic Colon Adenocarcinoma: A Case Report

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Conflict of interest: None declared
Source of support: Deanship of Scientific Research, King Saud University, through the Vice Deanship of Scientific Research Chairs

Patient: Female, 53-year-old
Final Diagnosis: Schistosomiasis colitis
Symptoms: Intolerable anal pain • rectal bleeding
Medication: —
Clinical Procedure: —
Specialty: Oncology • Surgery

Objective: Rare co-existance of disease or pathology

Background: Colorectal cancer is one of the most common cancers in men and women worldwide. There are several studies showing an association between chronic schistosomiasis infection and colorectal cancer.

Case Report: A 53-year-old woman presented with recurrent metastatic colon cancer involving the peritoneum and bilateral adnexa. The patient then underwent exploratory laparotomy that involved abdominal wall deposit resection, omentectomy, redo left hemicolectomy, peritonectomy, diaphragmatic stripping, and total abdominal hysterectomy with bilateral salpingectomy-oophorectomy, as well as hyperthermic intraperitoneal chemotherapy (HIPEC). She also underwent adjuvant chemotherapy, but on her 6th cycle, the patient suffered intolerable anal pain, diarrhea, and rectal bleeding. Her colonoscopy showed extended circumferential inflammation with loses of vascular pattern and a few rectal ulcers going up to the anastomosis site. Biopsy revealed *Schistosoma mansoni* eggs and marked ischemic changes. She was then managed with a single dose of Praziquantel.

Conclusions: Colorectal schistosomiasis infection is a rare cause of such common presentations especially in postoperative settings in a patient with recurrent metastatic colon cancer. The use of multimodality investigations and high clinical suspicion were needed for the diagnosis and to exclude other common etiologies.

Keywords: Colonic Neoplasms • Schistosomiasis • Schistosomiasis mansoni

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/930439

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Background

Schistosomiasis is a tropical parasitic disease affecting more than 240 million people globally, more endemic in tropical and subtropical areas with poor hygiene [1]. Schistosomiasis disease is more endemic in certain geographic areas depending on the species subtypes: the *S. mansoni* species is mainly found in the sub-Saharan African region and North America, the *S. japonicum* species is found in southeastern and eastern Asia including China, and the *S. haematobium* group is seen in Africa, Indian Ocean Islands, the Arabian Peninsula including Saudi Arabia, and Mediterranean regions [2]. There are several old studies suggesting that chronic intestinal schistosomiasis infection has etiological association with colorectal cancer [3]. However, different studies showed a lack of causal relationship between schistosomiasis infection and colorectal cancer [4]. In this case report, we present the first case of colorectal schistosomiasis infection in postoperative cytoreductive surgery (CRS) and hyperthermic intraperitoneal chemotherapy (HIPEC) for a patient with recurrent colorectal adenocarcinoma.

Case Report

A 53-year-old woman presented with a history of sigmoid cancer, which was managed with surgical resection and adjuvant chemotherapy. One year later, she was referred to our hospital with findings of recurrence on computerized tomography (CT) scan.

A full work-up was repeated for her, including tumor markers, colonoscopy, and CT chest, abdomen, and pelvis. Carcinoembryonic antigen (CEA) was high, with a level of 1225 ng/mL. Apart from internal hemorrhoids, colonoscopy reported unremarkable study with normal stapler line. CT abdomen showed bilateral adnexal masses inseparable from the uterus with multiple peritoneal soft tissue nodularities. CT chest reported unremarkable study. The case was discussed in the Tumor Board and the plan was to proceed with cytoreductive surgery (CRS) and hyperthermic intraperitoneal chemotherapy (HIPEC).

The patient underwent exploration laparotomy, abdominal wall deposit resection, omentectomy, redo left hemicolectomy, peritonectomy, diaphragmatic stripping, and total abdominal hysterectomy with bilateral salpingectomy-oophorectomy. After completion of CRS, HIPEC with oxaliplatin-based protocol for 30 minutes was done. The postoperative course was smooth with no complications except for reactivation of herpes labialis in the left upper lip, managed conservatively. She was discharged on day 14 in a stable condition.

The surgical pathology report was positive for recurrence at sigmoid and anastomosis site with moderately differentiated invasive adenocarcinoma. The omentum was positive for multiple nodules showing metastases; the largest was 3 cm. The ovaries and fallopian tubes showed metastatic moderately differentiated adenocarcinoma of colonic origin. A diaphragm nodule was positive for metastatic adenocarcinoma. An abdominal wall lesion showed metastatic adenocarcinoma. A pyloric lesion was positive for metastatic adenocarcinoma with focal necrosis. The lesser curvature and portal hepatic lymph nodes were positive for metastatic adenocarcinoma.

The case was discussed in the Tumor Board with a plan to proceed with 8 cycles of oxaliplatin and capecitabin (XELOX)

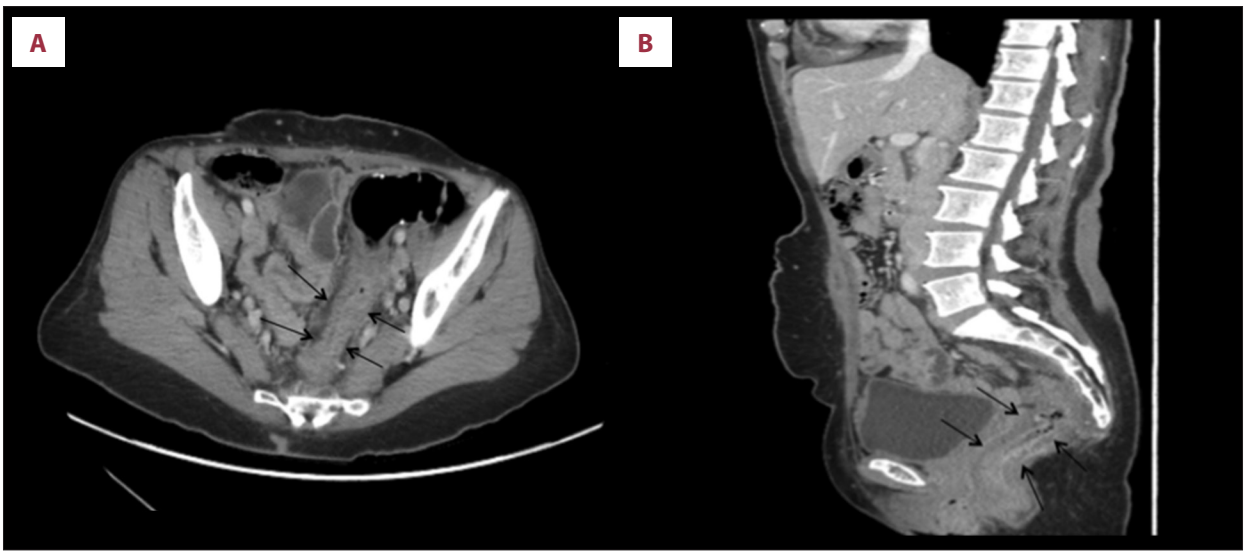

Figure 1. CT scan showed rectum is slightly collapsed with interval decrease of the rectal wall diffuse mural thickening suggestive of infectious/inflammatory changes. (**A**) Axial view and (**B**) sagittal view.

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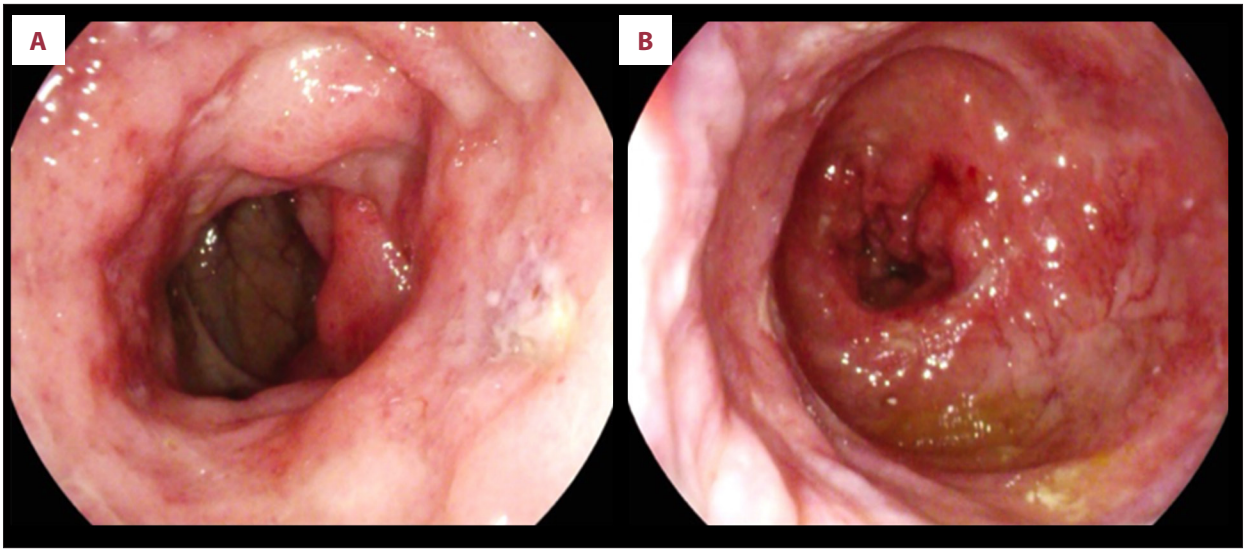

Figure 2. Colonoscopy findings showed (**A**) circumferential inflammation with loses of vascular pattern (**B**) significant narrowing with few rectal ulcers.

adjuvant chemotherapy. The patient was originally from a rural area in a southern province, with exposure to fresh water and soil. She came regularly for her chemotherapy sessions, but after the 6th cycle, she was very keen to stop because she started complaining of intolerable anal pain with defecation, diarrhea, and minimal amounts of bright red PR bleeding. On examination, there was an ulcerated left lateral pile. A full work-up was performed, including CEA, colonoscopy, and CT abdomen, with local conservative management of the anal fissure. CEA was unremarkable. CT abdomen showed interval decrease of the rectal wall diffuse mural thickening suggestive of infectious/inflammatory changes (**Figure 1**). Colonoscopy showed extended circumferential inflammation with loses of vascular pattern as well as a few rectal ulcers up to the anastomosis site. The proximal 10 cm of area of inflammation was significantly narrowed but not obstructed (**Figure 2**). Multiple biopsies were taken, which showed *Schistosoma mansoni* eggs and marked ischemic changes with regenerative atypia (**Figure 3**). These findings reflect schistosomiasis of the large intestine. In addition, the rectal mucosa showed features suggestive of mild chronic ischemic injury in the form of patchy mildly atrophic ("drop out") glands, reduction in goblet cells, mild hyalinization of the lamina propria, minimal chronic inflammation, mild edema, and extravasated red blood cells. There was no dysplasia or invasive carcinoma seen. The patient was managed with a single dose of Praziquantel 600 mg with follow-up 1 week after. All her symptoms were completely resolved. She was arranged for a follow-up colonoscopy, which kept getting postponed until months after. Results revealed normal mucosa with no pathologies in the terminal ileum, anastomosis site, or rectum.

Discussion

Colorectal cancer is the third most common cancer in men and the second most common cancer in women worldwide [5]. According to the American Cancer Society, colorectal cancer is the third leading cause of cancer deaths in men and women in the United States [6]. In Saudi Arabia, colorectal cancer is the first most common cancer among men and the third most common cancer among women, with the estimated incidence of 12.2%, which represents 1465 newly diagnosed cases in the 2015 report by the Saudi Cancer Registry [7].

Schistosomiasis is a parasitic disease caused by parasitic flatworms of the genus *Schistosomes* [8]. There are 3 major schistosomes infecting humans: *Schistosoma mansoni* and *S. japonicum,* primarily affecting the intestine and liver, and *S. haematobium,* which affects the bladder and urogenital system [8.] Many studies reported a possible link between colonic schistosomiasis and colorectal cancer, and a study in China showed that out of 179 schistosomiasis infection patients, 32 had colorectal cancer [9]. Moreover, many studies reported a link between *Schistosoma japonicum* in developing colorectal cancer more than for *S. mansoni* [10,11]. A few cases were reported of colorectal cancer with *S. mansoni*, but a clear link was not yet established, and they reported that more studies are needed [3,12]. A study investigating the pathogenesis in developing colorectal cancer in patients with schistosomiasis infection showed that the chronic inflammation, immunomodulation, Schistosoma toxins, and bacterial coinfection may have a major role in the pathogenesis [10].

Acute schistosomiasis typically presents with a sudden onset of fever, malaise, myalgia, headache, eosinophilia, fatigue, and

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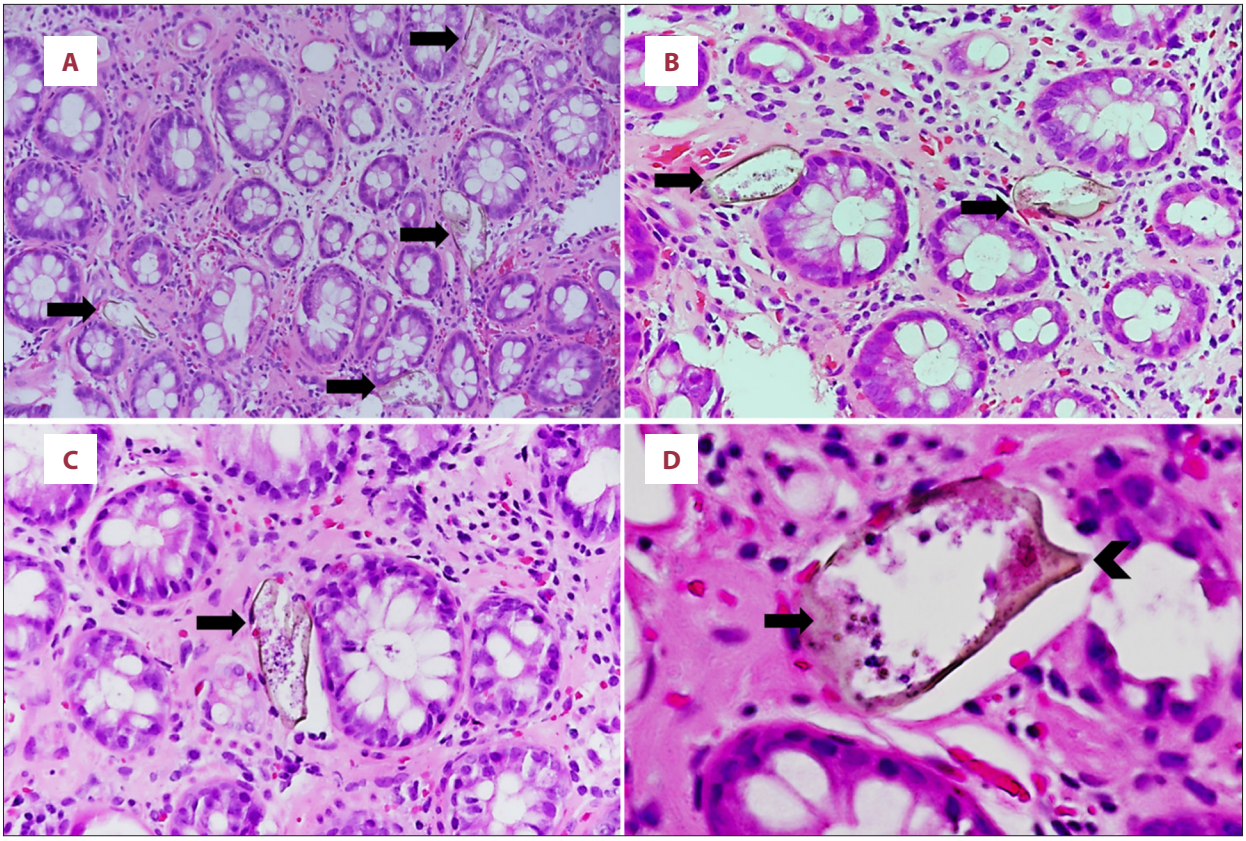

Figure 3. (**A-D**) Photomicrographs from a colonoscopic rectal biopsy shows several Schistosoma eggs (arrows) embedded in the lamina propria of the rectal mucosa. The Schistosoma eggs are yellowish brown, elongated to ovoid with a refractile shell. The large eccentric lateral spine projecting near one end (arrowhead) is a characteristic feature of a *Schistosoma mansoni* eggs. In addition, the rectal mucosa shows mild hyalinization of the lamina propria with mild chronic inflammation, edema, and extravasated red blood cells. There is no dysplasia or invasive carcinoma seen. This biopsy was diagnosed as schistosomiasis of the large intestine in which the location and histomorphology of the eggs are consistent with *Schistosoma mansoni* infection (hematoxylin and eosin stain, original magnification ×100, ×200, ×400, and ×600, respectively).

abdominal pain lasting 2-10 weeks [8]. Over time the granulomatous response to eggs is downregulated through several mechanisms, leading to the chronic intestinal manifestation, which includes intermittent abdominal pain, diarrhea, and rectal bleeding [8]. The frequency of these symptoms depends on the intensity of infection. These gastrointestinal features are often present with focal isolated mucosal hyperplasia, pseudopolyposis, and polyposis [8]. More severe forms of intestinal schistosomiasis present extensive fibrosis and subsequent hepatosplenic disease with periportal fibrosis [8]. Clinical features include upper-abdominal discomfort with palpable nodular and hepatosplenomegaly [8]. As a consequence, severe cases might present with the complication of portal hypertension, which can include ascites and hematemesis from esophageal varices, which can rapidly lead to death [8].

Schistosomiasis infection can be difficult to diagnose, as previous studies showed that many cases were misdiagnosed as ulcerative colitis, Crohn's disease, ischemic colitis, and irritable bowel syndrome [13,14]. Many studies concluded that clinical manifestations and endoscopy are insufficient for the diagnosis for schistosomiasis infection, as it shows non-specific features, and multiple biopsies are needed to confirm the diagnosis by the pathological features. Furthermore, they emphasized the importance of follow-up by colonoscopy, as they are at high risk of developing colorectal cancer [9,13,14]. The endoscopic features of schistosomes that were reported in the literature were divided into acute colitis, chronic colitis, and mixed type. In the acute colitis type the mucosa is congested and edematous, while in chronic colitis type the mucosa is hypertrophied with scarring [9,13,14]. Regarding the pathological features, all cases had schistosome ovum deposited in the lamina propria; the ova nodules of acute colitis consisted of one or multiple central ova surrounded by eosinocytes, in contrast to chronic colitis, where the ova are ruptured, calcified, and surrounded by lymphocytes [13,14].

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The challenge in our case was the wide differential diagnosis, including postoperative complications, recurrence of primary disease, and adverse effects of chemotherapy, which are more common than schistosomiasis infection. Therefore, colonoscopy and biopsy were crucial to reach the diagnosis. Schistosomiasis infection is mainly treated medically by Praziquantel, which is the drug of choice for all *Schistosoma* species [8]. However, some severe cases need to be treated surgically, including extreme degrees of portal hypertension, which needs shunting, esophageal varices bleeding, and urinary tract or intestinal obstruction [15].

There were many cases reported in the literature of schistosomiasis infection found in patients with colorectal cancer at the time of diagnosis [9,13,14]. However, in our case schistosomiasis infection was found postoperatively after CRS and HIPEC, which was more challenging to diagnose. Moreover, being immune-compromised, especially after chemotherapy, as in our case, might have a role in making her more susceptible to schistosomiasis infection. The patient was managed medically with a single dose of Praziquantel 600 mg.

Conclusions

Colorectal schistosomiasis infection is a rare cause of such a common presentation. Although there are many cases published with patients having the parasitic infection at the time of diagnosis, schistosomiasis infection can occur postoperatively, as in our case. The high clinical suspicion of parasitic infection and the use of multimodality investigations is needed for the diagnosis in the presence of other common differential etiologies for postoperative patients.

References:

1. Schistosomiasis (Bilharzia) [Internet]. Who.int. 2020 [cited 4 December 2020]. *https://www.who.int/health-topics/schistosomiasis#tab=tab_3*
2. Jamieson BGM. Schistosoma: Biology, pathology and control, CRC Press, Boca Raton, FL, 2017.
3. H Salim OE, Hamid HK, Mekki SO, et al. Colorectal carcinoma associated with schistosomiasis: A possible causal relationship. World J Surg Oncol. 2010;8:68
4. Darre T, Djiwa T, Dare S, et al. Difficult causality relationship between colorectal cancer and schistosomiasis. Pathol Oncol Res. 2020;26(1):597-98
5. Colorectal cancer statistics [Internet]. World Cancer Research Fund. 2018 [cited 4 December 2020]. *https://www.wcrf.org/dietandcancer/cancer-trends/colorectal-cancer-statistics*
6. Colorectal Cancer Statistics | How Common Is Colorectal Cancer? [Internet]. Cancer.org. 2020 [cited 4 December 2020]. *https://www.cancer.org/cancer/colon-rectal-cancer/about/key-statistics.html*
7. Cancer Incidence Report Saudi Arabia 2015 [Internet]. Nhic.gov.sa. 2018 [cited 4 December 2020]. *https://nhic.gov.sa/eServices/Documents/E%20SCR%20final%206%20NOV.pdf*
8. Colley DG, Bustinduy AL, Secor WE, King CH. Human schistosomiasis. Lancet. 2014;383(9936):2253-64
9. Liu W, Zeng HZ, Wang QM, et al. Schistosomiasis combined with colorectal carcinoma diagnosed based on endoscopic findings and clinicopathological characteristics: A report on 32 cases. Asian Pac J Cancer Prev. 2013;14(8):4839-42
10. Hamid HKS. *Schistosoma japonicum* – associated colorectal cancer: A review. Am J Trop Med Hyg. 2019;100(3):501-5
11. Matsuda K, Masaki T, Ishii S, et al. Possible associations of rectal carcinoma with *Schistosoma japonicum* infection and membranous nephropathy: A case report with a review. Jpn J Clin Oncol. 1999;29(11):576-81
12. Herman AM, Kishe A, Babu H, et al. Colorectal cancer in a patient with intestinal schistosomiasis: A case report from Kilimanjaro Christian Medical Center Northern Zone Tanzania. World J Surg Oncol. 2017;15(1):146
13. Cao J, Liu WJ, Xu XY, Zou XP. Endoscopic findings and clinicopathologic characteristics of colonic schistosomiasis: A report of 46 cases. World J Gastroenterol. 2010;16(6):723-27
14. Ye C, Tan S, Jiang L, et al. Endoscopic characteristics and causes of misdiagnosis of intestinal schistosomiasis. Mol Med Rep. 2013;8(4):1089-93
15. Barsoum RS, Esmat G, El-Baz T. Human schistosomiasis: clinical perspective: Review. J Adv Res. 2013;4(5):433-44

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