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CASE REPORT

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Schistosomiasis: Two cases with unusual presentation

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Summary

Background: Schistosoma live in bowel lumen and their eggs migrate through mesenteric and portal veins where they cause granulomatous response, fibrosis and various complications.

Case Report: Two cases of schistosomiasis with hepatic and intestinal manifestations are presented. One of them presented as colonic malignancy and the second masquerading as appendicitis. Plain x-ray, Ultrasound and CT findings are discussed.

Conclusions: Established cases of schistosomiasis may be seen far from endemic areas due to migration of populations across the globe. It is therefore important to recognize the radiological findings and its possible associations.

Key words: schistosomiasis • calcification • adenocarcinoma colon • mosaic liver

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Background

Schistosomiasis is one of the most common and significant tropical parasitic infectious diseases in the world [1]. Schistosomal infestation constitutes a risk factor not only for those living in but also for those travelling to endemic areas [1,2].

Schistosomiasis was known in ancient Egypt since 1200 BC. It became a major health hazard in 19th century, when irrigation systems were extensively developed. Occurrence of schistosomiasis in Americas was likely dated with the beginning of African slave trade.

The three pathogenic species of *Schistosoma* for humans are *S. haematobium, S. mansoni and S. japonicum.*

The species *S. haematobium* was discovered by Theodor Bilharz in 1851 in Cairo, hence also called *bilharziasis*. *S. haematobium* attacks predominantly urinary bladder, ureters and genital tract. This species is distributed mostly in Egypt, Eastern and Central Africa, Iraq, Iran, Syria, Yemen, Southern Saudi Arabia, Lebanon, Israel, Turkey and Cyprus [2]. Smaller pockets of the disease are also seen in Portugal and India.

S. mansoni affects mostly the large bowel. This species is endemic in river Nile delta, Southern Sudan, East and West Africa (Senegal, Cameroon and Zaire) as well as in Arabia [2]. Less commonly it is also seen in South America, Antilles and Puerto Rico.

S. japonicum generally affects biliary tract, small bowel and proximal large bowel. This species is seen in China, Japan, Thailand, Vietnam, Burma and Philippines [2].

In the present day, with people migrations across the globe, full blown cases may present far from the endemic regions. We present two cases of histologically proved schistosomiasis with radiological findings of hepatic and intestinal manifestations, which presented as colonic malignancy and appendicitis. Both cases were diagnosed in Qatar, far away from endemic areas.

Case Report

Case 1

A 35-year-old Philippino female presented with a history of the left upper quadrant pain and constipation of approximately two weeks duration.

Initial plain x-ray of abdomen showed multiple punctuate and large calcifications of colonic wall as well as in the mesocolon (Figure 1). Ultrasound examination showed irreg-

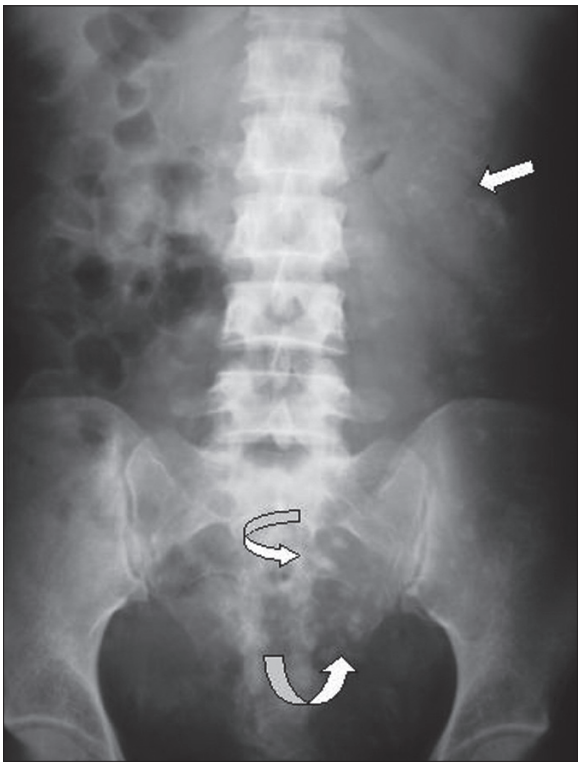

Figure 1. Case 1. Plain x-ray of abdomen: multiple punctuate and large calcifications along colonic wall (straight arrow) and mesocolon (curved arrows).

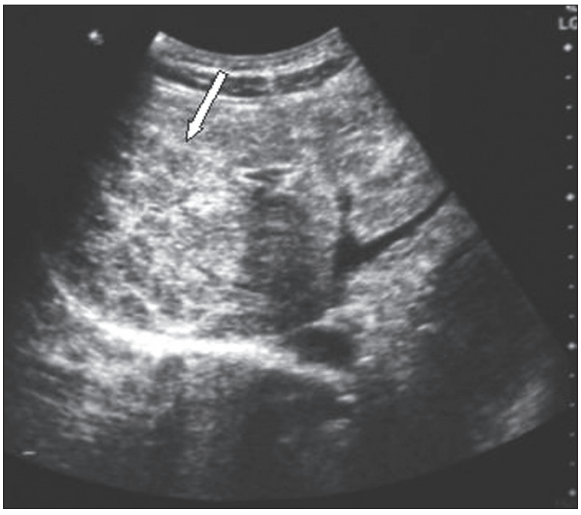

Figure 2. Case 1. Ultrasound of liver: echogenic septa (arrow) outlining normal areas of liver parenchyma.

ular liver borders with mosaic echopattern due to echogenic septa outlining polygonal areas of apparently normal appearing liver parenchyma (Figure 2). A CT scan was recommended with a provisional diagnosis of schistosomiasis.

CT scan showed features considered pathognomonic of schistosomiasis [3]. The scan demonstrated irregular hepatic contour, capsular and septal calcification as well as periportal fibrosis and fat (Figure 3). Calcifications in the colonic wall and in the mesocolon seen on plain x-rays were re-demonstrated on CT. These calcifications were punctu-

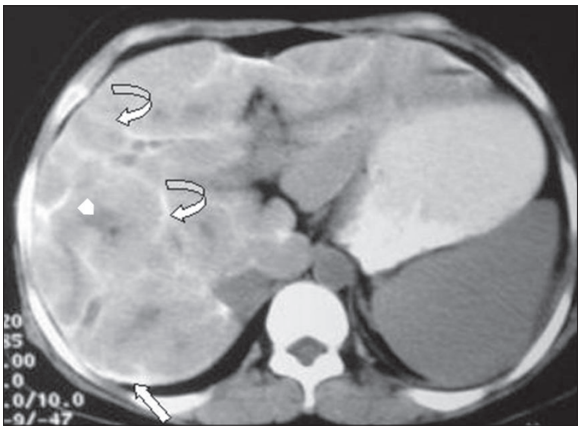

Figure 3. Case 1. Un-enhanced CT of abdomen: capsular (straight arrow) and septal calcifications (curved arrows) giving a pathognomonic appearance. Other features are irregular hepatic contour, periportal fat and fibrosis (arrow head).

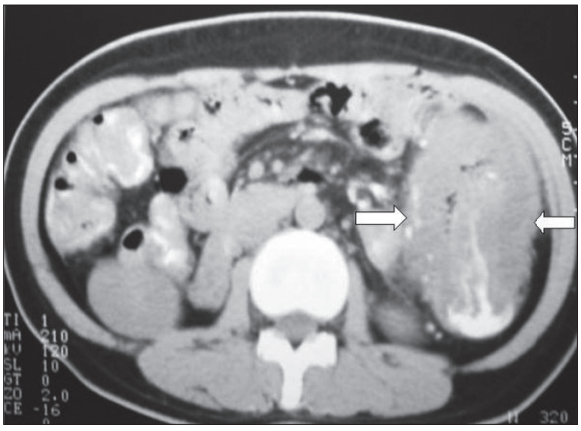

Figure 4. Case 1. CT of abdomen: mass in proximal descending colon (arrows). Colonic wall calcifications seen.

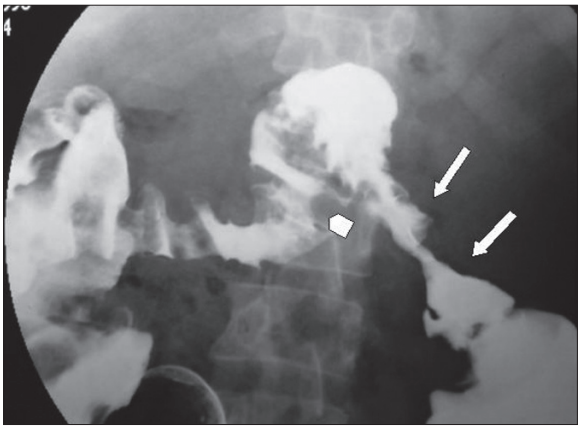

Figure 5. Case 1. Barium enema: stricture at splenic flexure (arrows) with adjacent thumb-printing (arrow head).

ate and tram-like. Furthermore, CT demonstrated a poorly enhancing, circumferential mass, approximately 8×6 cm, in the proximal descending colon (Figure 4).

A complementary barium enema showed a stricture at the splenic flexure with "thumb-printing" appearance of a large segment of adjacent colon (Figure 5).

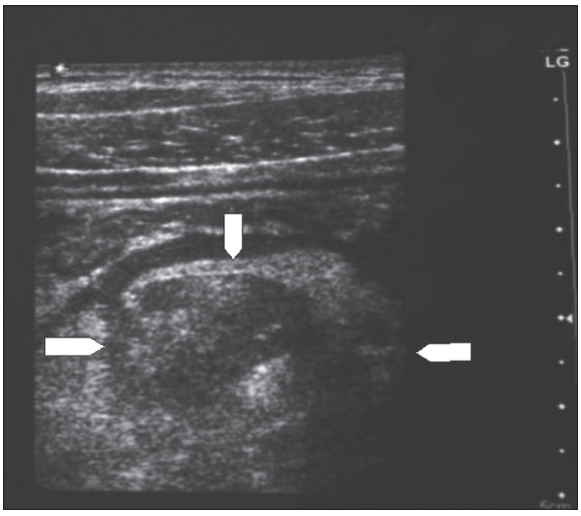

Figure 6. Case 2. Ultrasound of the right iliac fossa: heterogeneous appendicular mass.

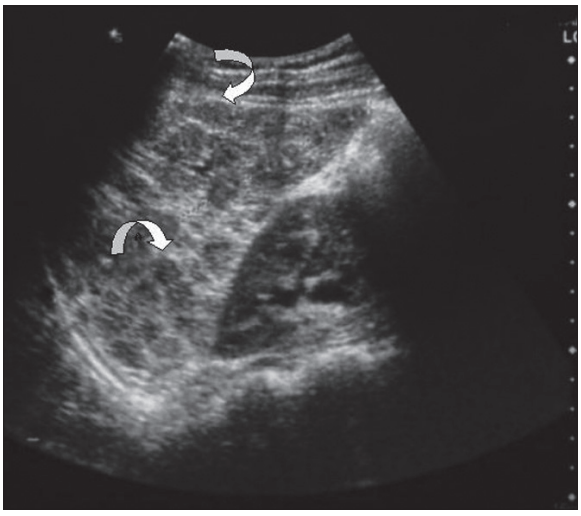

Figure 7. Case 2. Ultrasound of the liver: echogenic septa (curved arrows) outlining normal areas of liver parenchyma.

A left hemicolectomy was performed. The excised mass was sent for histolopathological examination with suspicion of malignancy. Histology showed clusters of malignant epithelial cells at the site of the mass with transmural extension to the pericolic tissue. There were also numerous *Schistosoma japonicum* eggs clustered in groups in lamina propria of the colon. The diagnosis was mucinous carcinoma of colon with background of massive colonic schistosomiasis.

Case 2

A 28-year-old Philippino female presented to our emergency department with right iliac fossa pain. An ultrasound examination was requested with a provisional diagnosis of acute appendicitis. The study revealed a mixed-echoic appendicular mass, approximately 5×4 cm (Figure 6). In addition, the liver showed an irregular outline with echogenic septa outlining polygonal areas of relatively normal liver parenchyma. These findings were considered typical of schistosomiasis (Figure 7). The patient underwent appendectomy. The histological examination of the appendix showed, apart from the inflammatory changes, calcified ova of *Schistosoma japonicum*. The patient left for Philippines without undergoing further investigations.

Discussion

The term schistosomiasis encompasses a group of acute and chronic clinical and pathological disorders caused by infection with one or more of five species of digenetic worms of the genus *Schistosoma* [1]. The species of major global importance are *Schistosoma mansoni, S. haematobium and S. japonicum*. There is a large spectrum of manifestations of schistosomiasis which includes involvement of urinary and gastrointestinal tracts, skin, lung, central nervous system, muscle, adrenal glands and eyes [1,2].

The life cycle involves freshwater snails and man in whom the mature worm pairs reside in the mesenteric or vesical veins. It is described in details elsewhere [2]. The parasite ova are deposited in the submucosa of small and large intestines. The eggs may then migrate into the bowel lumen, remain *in situ* or embolize to the liver, lungs and urinary system [1].

Schistosomiasis has variable clinical and radiological presentation related to the site and severity of infestation [2,3]. *Schistosoma haematobium* is seen in pelvic veins, whereas *Schistosoma japonicum* and *Schistosoma mansoni* are found in portal venous system [4]. The effects of *Schistosoma haematobium* infestation are therefore seen predominantly in bladder and ureters, whereas the consequences of *Schistosoma mansoni* and *Schistosoma japonicum* invasion are seen in the liver (periportal fibrosis, portal hypertension and cirrhosis). In the intestine, the eggs produce granulomatous reaction resulting in mucosal thickening, granulomatous polyps and fibrosis [5,6]. Also it is the calcification of these eggs which are seen on radiographs or CT [5]. Calcification in submucosa and subserosa give curvilinear or tram-like appearance in colon on CT images [3,7]. Calcified eggs in the lymph nodes and the inflammatory tissues of mesocolon appear as punctuate calcifications. In the liver, secondary to *Schistosoma* infestation, there is periportal fibrosis and fatty infiltration with fibrous thickening of the capsule. Fibrous retractions produce liver surface depressions or its irregular outline [5].

The association between *schistosoma* infestation and urinary bladder and hepatic malignancy is well established. Urinary bladder squamous cell carcinoma can occur secondary to *Schistosoma haematobium* infestation [8]. Hepatocellular carcinoma has been described in association with schistosomiasis [4]. However, Xu et al. from Jiangsu province of China, an endemic area of schistosomiasis, in their study of 252 patients of colonic malignancy concluded that risk of colonic malignancy was not significantly increased in presence of schistosomiasis [9].

In reference to the Case 2, the granulomatous reaction, mucosal thickening and polyp formation observed in the course of schistosomiasis were also seen in the appendix [9]. This can potentially lead to appendicolith formation and appendicular inflammation.

Schistosomiasis as the causative factor of either urinary bladder, liver or colonic cancer is not established but is speculative [4,8,9]. However, there is definite increase in incidence of these conditions in endemic areas of schistosomiasis. The potential association of these entities mandates a thorough search and exclusion of malignancy, because schistosomiasis itself is a treatable disease [1]. Therefore, a thorough knowledge of the spectrum of pathological findings in the course of schistosomiasis on various imaging modalities is important as well as the understanding of associations of the disease.

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