Case Report

First Schistosomal Cholecystitis Complicated by Cholangitis and Liver Abscess: Case Report and Review of Literature

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Schistosomiasis is one of the most prevalent parasitic infections in the developing world. When it affects the gastrointestinal system specifically the liver, it causes periportal fibrosis followed by cirrhosis. Cholecystitis however is a rare presentation, and associated liver abscess has certainly never been reported to date. We report a case of acute cholecystitis complicated by cholangitis and liver abscess in a 46-year-old man. After complex course of treatment, he had laparoscopic cholecystectomy, and the histology report confirmed schistosomiasis. Gallbladder schistosomiasis is an uncommon disease that is associated with dense fibrotic changes that strongly mimics xanthogranulomatous cholecystitis. Liver abscess may occur during the disease evolution especially in patient originating from endemic backgrounds. We present the case and a comprehensive literature review.

1. Introduction

Parasitic infections remain a problem in the developing countries [1]. Schistosomiasis is responsible for more than 200 thousands deaths yearly [1]. Schistosomiasis may present acutely as febrile illness [2] or more commonly in chronic form due to eggs that are trapped in the tissues during the peri-vesical or peri-intestinal migration or after embolization in the liver, spleen, lungs, or cerebrospinal system [3]. Chronic lesions in these tissues are usually characterized by chronic inflammation and fibrosis which gives rise to the clinical manifestation of the disease (i.e., cirrhosis for hepatic involvement, chronic cystitis, and fibrosis for urinary involvement) [1].

In spite the fact of the high frequency of hepatic involvement particularly by Schistosoma mansoni, schistosomiasis of the gallbladder (GB) is remarkably uncommon [4]. About twelve cases have been retrieved from the literature. However, none of them was associated with liver abscess or cholangitis. A literature review in a comprehensive approach was carried out (Table 1). We report this case in line with the updated consensus-based surgical case report (SCARE) guidelines [5].

2. Case Presentation

A 46-year-old Egyptian man presented to the emergency department with one-day history of epigastric and right upper abdominal pain, associated with nausea and vomiting. He denied any other associated gastrointestinal or urologic symptoms. Apart from being type 1 diabetic, he declared no other significant past medical history.

The patient presented in good shape, and there was no hemodynamic instability. He showed epigastric tenderness without any signs of peritonism with the rest of the abdomen unremarkable. White blood cell count (WBC) was 17.4 K/uL (4-10 K/uL). Liver function tests (LFTs) were abnormal: total bilirubin 47 umol/L (0-21 umol/L), direct bilirubin 35 umol/L (0-3 umol/L), ALT 156 U/L (0-40 U/L), and
Table 1: Summary of characteristics of current case and other reported cases of gallbladder schistosomiasis identified from the review of the literature.

| Study* | Sex | Age | pH | LEA | Presentation | D | PE | Labs | US | CT | Others | Surgery | Intra-op | Histo | F Up |
|--------|-----|-----|----|-----|-------------|---|----|------|----|----|---------|----------|----------|-------|------|
| Current study Qatar 2020 | M | 46 y | DM | Yes | RUQ pain | 1 d | Epigastric tenderness | WBC: 17.4, Hb: 14.4, bili: 47, direct bili: 35, ALT: 156 U/L, AST: 182 U/L, lipase: 106 U/L, CA 19-9: 303 U/ml, IgE: 432 K units/L, postop positive S serology | 1st US: GS, Di IHBD, CBD: 7 mm | Newly developed liver abscess | MRCP: acute Chol, Cholang. | Lap Chole | Omental adhesions to the GB which was densely adherent to the liver | Chronic Chol, Gr Inf secondary to S | Prazi 40 mg/kg divided into 3 doses |
| Hedfi 2019 Tunisia [4] | F | 51 y | DysL | No | Hepatic colic | 2 m | N | N | Thin-walled GB, GS 10 mm | NR | NR | Lap Chole | | | Slightly thick-walled GB, fine cystic duct | Calcified S ova in the wall of GB stained positively for periodic acid-Schiff | CT urography: N |
| Majrashi 2018 Saudi [20] | M | 50 y | DM | Yes | Elective surgery for biliary colic | 9 y | RUQ tenderness | Positive S serology postop others: N | Wall thickness (4 mm), GS 8 mm | UR | NR | Lap Chole | | | Thick wall GB, with necrotic spots, firmly attached to the liver bed | Gr Inf around calcified S. haematobium eggs | Referred to ID team |
| Azoulay 2016 France [15] | M | 53 y | NR | Yes | Elective after 2 episodes of Chol, recent 4 kg weight loss | 5 m | N | N | Hyperechogenic thick GB wall, no GS | Thicl GB wall 12 mm, contained calcifications and lesion protruding into GB and the liver, increased density of peri-vesicular fat, enlarged 2 hilar LN’s (7 mm) | Lap to open radical Chole (en bloc omental adhesions and LN resection) | | Tense retraction of the right colon, duodenum, and omentum to the inferior aspect of the liver hampered Lap GB exploration | Acute and chronic Chol with dense fibrosis, S eggs in GB wall | Single dose of 2.4 mg of Prazi 15 d after surgery |
| Study* | Sex | Age | pH | LEA | Presentation | D | PE | Labs | US | CT | Others | Surgery | Intra-op | Histo | F Up |
|--------|-----|-----|----|-----|--------------|---|----|------|----|----|--------|---------|----------|-------|------|
| Manes 2014 Greece [19] | M | 77 y | NR | Yes | Elective 3 months after Chol | 3 m | RUQ tenderness | N | Thick-walled GB (6.8 mm) GS 1.7 cm impacted at GB neck | NR | NR | Lap converted to open Chole | GB inflamed and thick with necrotic spots and wood-like consistency | Gr Inf around calcified S. mansoni eggs | Praziquantel 20 mg/kg every 4 h for 3 doses |
| Sharara 2001 Lebanon [8] | F | 47 y | Smoker | No | RUQ discomfort | 3 d | RUQ tenderness | AEC: 660/mm³ UA: Mic hem | Thick GB wall, 1 cm echogenic structure without acoustic shadow at GB fundus | Markedly thick GB wall, 2 hypodense liver lesions | NR | Lap Chole | Thick nondistended gallbladder firmly adherent to the liver surface and an enlarged cystic LN, no GS | Gr Inf around multiple S. mansoni eggs, with the lateral spine, likely S. mansoni | Praziquantel 20 mg/kg every 4 h for 3 doses |
| Bakhotma 1996 Saudi Arabia [21] | M | 30 y | NR | RUQ pain, HU | NR | NR | UA: S. haematobium | GS | NR | NR | Lap Chole | Thickened wall | Gr Inf around S. mansoni with S. mansoni infection | Praziquantel, received before surgery |
| Al-Saleem 1989 Iraq [7] | M | 27 y | NR | Yes | Biliary colic, hematemesis | 2 m | Enlarged spleen down to the pelvis | NR | Huge spleen, thick GB wall, no GS | NR | NR | Lap Chole | Chronic Chol with S. mansoni infection | Huge spleen, cirrhotic liver, GB grey, irregular in thickness, infiltrating into the liver bed. Thick cystic duct | Extensive S fibrosis | NR |
| Al-Saleem 1989 Iraq [7] | M | 25 y | NR | Yes | Epigastric pain | 2 m | NR | Thick GB wall, large GS | NR | NR | Chole | Thick walled grey GB, the fibrosis so deep into the bed, thickened fibrotic, and calcified cystic duct | Extensive fibrocalcific GB S, due to S. mansoni | Extensive fibrocalcific GB S, due to S. mansoni | NR |
| Al-Saleem 1989 Iraq [7] | M | 62 y | Childhood | Yes | RUQ pain | NR | NR | NR | Thick-walled grey GB, attached tightly to the liver and infiltrating it | Extensive fibrocalcific GB S, due to S. mansoni | Extensive fibrocalcific GB S, due to S. mansoni | NR |
Table 1: Continued.

| Study* | Sex | Age | pH | LEA | Presentation | D  | PE | Labs | US | CT | Others | Surgery | Intra-op | Histo | F Up |
|--------|-----|-----|----|-----|--------------|----|----|------|----|----|--------|---------|----------|-------|------|
| Al-Saleem 1989 Iraq [7] | M | 33 y | Childhood | HU | Yes | Dull epigastric pain | 3 m | NR | NR | Large GS | NR | NR | L, Chole | Thick-walled grey GB, with extensive fibrosis | Fibrocalciﬁc GB S, due to S. haemotobium | NR | |
| Al-Saleem 1989 Iraq [7] | F | 40 y | Obese | Yes | Dull RUQ pain | 13 m | No tenderness | NR | Thick GB wall, large GS | NR | NR | Chole | Thick-walled grey GB, GS | Fibrocalciﬁc GB S, due to S haemotobium | NR | |
| Al-Saleem 1989 Iraq [7] | M | 55 y | NR | Yes | RUQ discomfort radiated to Rt shoulder, N&V | 14 m | RUQ tenderness | NR | Thick GB wall, large GS | NR | NR | NR | Pancreatic tumour with multiple hepatic secondaries, thick-walled GB with stones | Biopsy showed extensive fibrosis, ova of S. haemotobium | NR | |
| Rappaport 1975 US [6] | M | 51 | NR | NR | RUQ pain, N&V, diarrhea | Few d | RUQ tenderness | N | NR | NR | IVP: N | Chole | Fibrotic liver, focally mildly thickened GB | Gr Inf, S. mansoni | NR | |

*For space considerations, only the first author is cited. AEC: absolute eosinophil count; Bili: bilirubin umol/L; CBD: common bile duct; Chol: cholecystitis; Cholang: cholangitis; Chole: cholecystectomy; D: duration of symptoms; d: days; DM: diabetes mellitus; Dys.: dyslipidaemia; F: female; F Up: follow-up treatment; GB: gallbladder; Gr: granulomatous; GS: gall stone/s; Hb: hemoglobin g/dl; HU: hematuria; ID: infectious diseases; IHBD: intrahepatic bile ducts; Inf: inﬂammation; Intra-op: intraoperative findings; IVP: intravenous pyelogram; L: laparotomy; Lap: laparoscopic; LEA: lived in an endemic area; LN’s lymph nodes; M: male; m: month/s; Mic: microscopic; N: normal; NR: not reported; N&V: nausea and vomiting; OGD: oesophagogastroduodenoscopy; PE: physical examination; post-op: postoperative; Praz: praziquantel; Rt: right; RUQ: right upper quadrant; S: schistosoma/l; UA: urine analysis; UR: unremarkable; WBC: white blood cells K/ul; y: year/s.
Despite the di
the GB. The GB was also densely adherent to the liver. Patient
lapyaroscopically. Patient’s postoperative course was unremarkable, and he was discharged next day after surgery. Histopathology of the gallbladder showed chronic cholecys-
titis, with granulomatous inflammation secondary to schis-
tosomiasis (Figure 2).

Cultures grew Klebsiella oxytoca and Escherichia coli. The 
towards continuity between the liver abscess and the GB.

He later developed signs of sepsis (tachycardia and fever)
for which blood cultures were taken. He was started on intra-
venous ceftriaxone and metronidazole. Endoscopic retro-
passage cholangiopancreatography (ERCP) showed purulent 
 bile immediately following cannulation and failed to show 
any filling defects. Sphincterotomy and CBD stenting were 
done. ERCP procedure was not extraordinary in difficulty 
to suspect ampullary fibrosis or deformation. Magnetic 
Resonance Cholangiopancreatography (MRCP) later showed 
distended GB containing sludge and tiny stones, hyperen-
hancement of both GB and CBD walls, and mildly thickened 
GB wall in addition to pericholecystic edema and fat strand-
ning, consistent with acute cholecystitis and cholangitis. There 
were no CBD stones nor thickening of the CBD wall. After 
stenting, the patient was kept on piperacillin/tazobactam. 
He improved clinically, and both his LFTs and inflammatory 
markers were trending down till the 4th day post-ERCP 
when he started to spike fever again. Septic work up was 
repeated, and endoscopic ultrasound (EUS) was done. This 
exam showed the stent in place. Both US and computed 
tomography (CT) showed a new lesion 5 × 4 × 5 cm in the 
segment IVb of the liver in continuation with the GB fundus 
(Figure 1). The lesion was compatible with a newly developed 
liver abscess. A percutaneous aspiration was carried out 
under US guidance, during which 100 ml of pus was aspir-
ated and sent for microbiology/culture. During the aspira-
tion, the GB was noticed to be de
ating which pointed 
towards continuity between the liver abscess and the GB. 
Cultures grew Klebsiella oxytoca and Escherichia coli. The 
patient responded well after aspiration and antibiotic 
therapy and showed improved inflammatory markers. He was 
 discharged the next day on oral antibiotics with close clinic 
follow-up to arrange for interval cholecystectomy after ERCP 
and stent removal.

During follow-up visits, the patient was asymptomatic. 
ERCP was done 5 weeks after discharge and showed no fill-
ing defects in the CBD. The stent was removed. The patient 
travelled and was lost to follow-up for 5 months. When he 
came back, an MRCP showed complete resolution of the 
liver abscess. Multiple gall stones in GB were still demon-
strated. He was booked for elective laparoscopic cholecys-
tomy. Intraoperative findings showed omental adhesions to 
the GB. The GB was also densely adherent to the liver. 
Despite the difficult dissection, the procedure was managed 
laparoscopically. Patient’s postoperative course was unremarkable, and he was discharged next day after surgery. Histopathology of the gallbladder showed chronic cholecys-
titis, with granulomatous inflammation secondary to schis-
tosomiasis (Figure 2).

### 3. Discussion

The first case of gallbladder schistosomiasis (GBS) was 
reported in 1975, and since then, speculations were made 
regarding possible pathogenesis [6]. Fourteen cases of GBS 
have been reported; however, none of them presented with 
associated complications. Few theories evolved on how schis-
tosomiasis can cause cholecystitis. Some speculated that the 
fibrosis of the cystic duct, like what is seen in the ureters of 
patients with urinary schistosomiasis, causing a stenosis 
which can contribute to bile stasis and formation of stones 
in the gallbladder [7]. Others suggested that granulomatous 
inflammation in the gallbladder’s wall makes it prone for 
stone formation [8].

The risk factor for contracting schistosomal infection is 
the contact of its larval form with the skin through contami-
nated water in endemic areas [4]. Most of the reported cases 
(Table 1) have been living at one stage in their life in an 
endemic area. Our reported case used to live in Egypt that 
is a well-known endemic area before moving abroad.

Clinical presentation is variable according to the involved 
orган. Infestation of urinary tract may lead to hematuria, 
fibrosis, and obstructive uropathy that may lead to parenchym-
al renal damage [1]. When it involves the liver, early 
inflammatory hepatic schistosomiasis happens in reaction 
to schistosomal eggs trapped in the presinusoidal periportal 
spaces of the liver. It then lead to typical features of sharp-
edged enlargement of the liver nodular splenomegaly [1]. 
Intestinal involvement leads to diarrhea mostly due to muco-
asal granulomatous inflammation, pseudopolyposis, and 
microulcerations [1]. Reported symptoms of GBS are usually 
similar to other gallbladder diseases, including right upper 
quadrate pain that is sometimes associated with nausea and 
vomiting [7]. Abdominal examination shows right upper 
abdominal tenderness especially if the patient is having active 
cholecystitis (Table 1). The reported case first presented to 
the emergency with right upper quadrant abdominal pain. 
His disease progression was completely unique after GBS as 
he developed septic features due to cholangitis and associated 
liver abscess. This is, to our knowledge, the first reported case 
of cholecystitis with a liver abscess in a patient with
biliary stones that usually slip from the gallbladder [9]. In benign and malignant), the most common of which are due to obstruction [9]. Causes of obstruction are variable secondary to a typical gallstone cholecystitis.

The specimen pathology usually reveals a lymphocytic infiltrate; schistosomal eggs can be found in any layer of the gallbladder wall causing fibrocalcific reaction; most of the cases showed granulomatous inflammation surrounding the schistosomal eggs (Table 1).

4. Conclusions

GBS might be considered preoperatively in patients who lived in an endemic area and developed symptoms suggestive of gallbladder disease. This is the first case that report a liver abscess in a patient with cholecystitis with a gallbladder infested by Schistosoma. However, a majority of cholecystitis in patients with schistosomiasis involve the presence of gallstones. This condition carries the same possible complications and should be managed in the same way as usual cholecystitis. Surgeons must however expect a more difficult dissection during operation.

Ethical Approval

The protocol for this research project has been approved by a suitably constituted Ethics Committee of the institution, and it conforms to the provisions of the Declaration of Helsinki. Medical research center committee of Hamad Medical Corporation, approval No. (MRC-04-20-912).

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available on request.

Conflicts of Interest

The authors declare no conflict of interests for this article.
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