

# Dual Parasitization of the Biliary Tree: First Reported Case

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## ABSTRACT

Cases of biliary ascariasis and hydatid cysts in liver and elsewhere are common in endemic areas and are routinely encountered in surgical outpatient departments. We describe the diagnosis and management of a unique case, who presented with manifestations of biliary ascariasis, but on further investigation was found to harbor ascarids as well as ruptured hydatids in his biliary passages. To our knowledge, this is the first reported case of simultaneous parasitization of common bile duct by ascarid and hydatid forms, resulting in obstruction of the biliary system. This report highlights the diversity of presentation and challenges in the management of such cases, when encountered in practice.

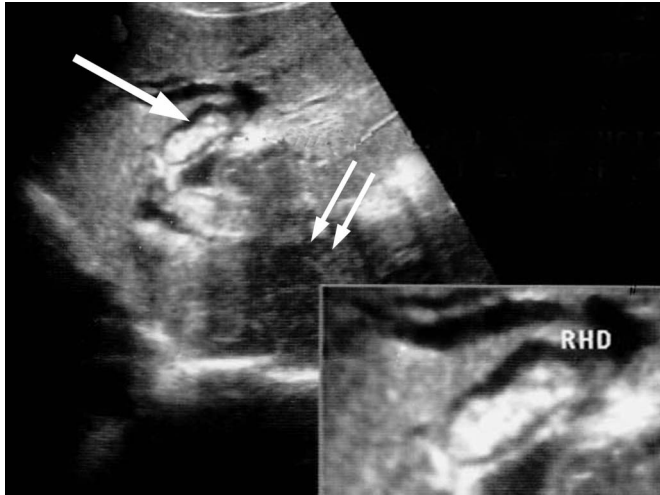
## INTRODUCTION

Ascariasis is a very common parasitic disease in humans, affecting hundreds of millions of people in countries where the standards of public health and personal hygiene are low.<sup>1,2</sup> One of the most common ectopic sites for ascariasis is the hepatobiliary tree, where ascarid invasion could cause biliary colic, recurrent pyogenic cholangitis, cholecystitis, and pancreatitis.<sup>3</sup> Echinococcosis is another endemic disease in the Asian subcontinent, which involves the liver in 50%–70% of the patients.<sup>4</sup> Intrahepatic rupture of hydatid cysts, which has an incidence of 5%–17%, is not an uncommon complication and could present as cholangitis.<sup>5</sup> Although the presence and management of both the parasites in the biliary ductal system have been well documented separately in literature, their concurrent occurrence and management have never been reported.<sup>6</sup> We present a patient simultaneously harboring ascarids and ruptured hydatid cysts in the biliary ductal system, in a state of mutual coexistence.

## CASE REPORT

A 55-year-old man was admitted to the emergency department of our institute with complaints of pain in the right upper abdomen, fever with rigors, and yellowish discoloration of eyes. He had a pulse rate of 110/minute, blood pressure of 112/70 mm Hg, and a temperature of 101°F. There was guarding and tenderness in the right upper abdomen. Laboratory investigations revealed leucocytosis with 85% polymorphonuclear leucocytes, serum bilirubin level of 12.0 mg/dL, serum glutamic oxaloacetic transaminase of 53 U/L, serum glutamic pyruvic transaminase of 48 U/L, and an alkaline phosphatase of 946 U/L. Ultrasound examination of the abdomen revealed a distended gallbladder with dilated extra- and intrahepatic ductal systems. The dilated biliary radicals contained hyperechoic, nonshadowing linear structures consistent with biliary ascariasis. In addition, ultrasound examination revealed an area of mixed echotexture measuring about 3 cm in diameter in the subdiaphragmatic part of the right lobe of liver (Figure 1). A presumptive diagnosis of biliary ascariasis with a liver abscess in the right lobe was made (since it is endemic in nature in this part of the world).

Patient was started on albendazole 15 mg/kg body weight BD and ceftriaxone 1 g twice daily, besides administration of intravenous fluids. Endoscopic retrograde cholangiography (ERCP) revealed a dilated common bile duct (CBD) containing a linear filling defect, representing an ascaris worm along with multiple irregular filling defects (Figure 2). Sphincterotomy was performed and whitish membranes were seen projecting from the CBD into the lumen of duodenum. Basketing of these membranes was performed, which also facilitated the



**Figure 1.** Ultrasound showing dilated intrahepatic biliary channel right hepatic duct containing hyperechoic, nonshadowing, linear structures representing ascarid worms (long arrow). A well-circumscribed area of mixed echogenicity is also seen in the sub-diaphragmatic part of the right lobe of liver (short arrows).

removal of the worm, sludge, and other debris. However, complete clearance of the ductal system could not be achieved. An endoscopic nasobiliary drainage tube was put in place. A preoperative diagnosis of biliary ascariasis with intrabiliary rupture of hydatid cysts was made. Bile culture was also taken and showed growth of *Escherichia coli* sensitive to multiple drugs.

Laparotomy was performed after an interval of 72 hours. On exploration, gallbladder was seen distended; it had



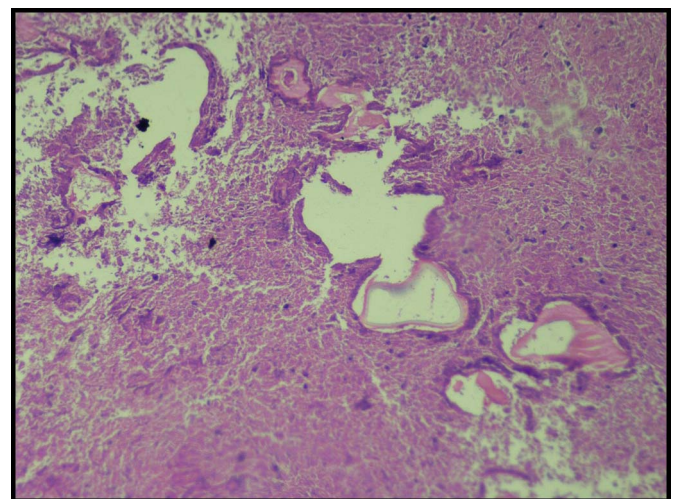
**Figure 2.** Endoscopic retrograde cholangiography showing a dilated common bile duct containing a linear filling defect representing an ascaris worm (long arrow) along with multiple irregular filling defects produced by hydatid membranes (short arrows).

developed multiple adhesions with the duodenum. CBD was also distended (18 mm in diameter) and edematous; it was covered all over with dilated vessels. A cystic swelling of 4 cm in diameter was localized in the superior part of the right lobe of liver. Cholecystectomy and CBD exploration were performed. Fragmented parts of hydatid cyst membranes and fragments of dead worms were removed from the CBD. The right intrahepatic duct contained sludge and one dead worm. All parasitic matter was removed, and the ducts were flushed with normal saline. The cystic lesion in the right lobe of liver was also explored and was found to be an infected hydatid cyst. There was no visible communication between the cyst wall and biliary tree. A tube drain was left in the cavity, and a T-tube was inserted for drainage of the CBD. Postoperative histopathological analysis confirmed the diagnosis of hydatid and ascariasis infestation of liver and biliary tract (Figure 3).

The patient exhibited a remarkable postoperative improvement. Cavitogram and T-tube cholangiogram obtained on the seventh postoperative day showed normal results. Tube drain was removed on the 12th day when bile drainage had ceased, and T-tube was removed on the 14th day. The patient was discharged on the 16th postoperative day after receiving anthelmintic treatment in the form of albendazole 15 mg/kg bodyweight for 1 week. He was also advised for regular deworming at 6-month interval.

## DISCUSSION

While ascariasis is an endemic disease of the humans, echinococcosis is essentially a disease of animals (dogs and sheep) in which man represents a dead-end host who gets infected only incidentally. Ascariasis endemicity is maintained by faeco-oral transmission cycle, while that of echinococcosis is maintained by dog-sheep transmission cycle. Both the



**Figure 3.** Histological image showing the hydatid membranes interspersed with the degenerated hepatocytes.

parasitic infections can occur in unsanitary conditions, when raw night soil pollutes the land and stray dogs have free access to grass fields and vegetable farms. Tropical and subtropical areas provide the ideal environment, and obviously, transmission of both the parasites is not an uncommon event.<sup>7-9</sup> However, simultaneous occurrence of both the parasites within the biliary ductal system is indeed rare, which, to the best of our knowledge, has not been reported before.

Biliary ascariasis accounts for 10%–19% of ascaris-related hospital admissions in an endemic setting.<sup>10</sup> Under exceptional circumstances caused by excessive worm load in the jejunum, or hypermobility of the intestines associated with episodes of acute enteritis, the ascarids enter various internal hollow structures such as CBD, main pancreatic duct, and gallbladder for migration.<sup>11</sup> The invasion of bile ducts can lead to cholangitis, pancreatitis, acalculous cholecystitis, and even to liver abscess.<sup>3,12,13</sup>

Hydatid cysts in the liver exert pressure on the surrounding parenchyma and in some cases, the cysts eventually leak into small bile ducts or perforate into larger ones; some sort of communication can be found in 40%–90% of the cases.<sup>14,15</sup> This could result in biliary obstruction, presenting as obstructive jaundice and cholangitis in 5%–17% cases.<sup>16</sup>

ERC is a confirmatory test both for the presence of hydatid cysts as well as ascariids in the biliary passages. It provides more comprehensive information and elegantly displays daughter cysts obstructing any part of the biliary tree or impacted in the ampulla of Vater.<sup>17</sup> ERC can also demonstrate communication, if any, between the cystic cavity and the biliary tree.<sup>15</sup> In the recent past, the role of magnetic resonance cholangiography in the diagnosis of biliary ascariasis has been described.<sup>18,19</sup> However, sonography continues to be an ideal modality for detecting ascariids in the bile ducts and monitoring their movements.<sup>20</sup> It also helps in identifying hydatid cysts in the liver and elsewhere, and in assessing their impact on the surrounding tissues.<sup>21</sup>

Surgical approach remains the main modality of management in patients with hepatobiliary echinococcosis, although endoscopic sphincterotomy and extraction of daughter cysts from the bile ducts have been tried with some success.<sup>22</sup> The management of biliary ascariasis ranges from endoscopic extraction to removal by the percutaneous transhepatic approach.<sup>23,24</sup>

## DISCLOSURES

**Author contributions:** All authors had equal contribution in the preparation of manuscript and editing. M. Ilyas is the article guarantor.

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Informed consent was obtained for this case report.

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## REFERENCES

- Gabaldon A, Mofidi C, Morishita K, Moskovskij S, Sankale M, Standen DD. Control of ascariasis (report of a WHO expert committee). *World Health Organ Tech Rep Ser.* 1967;379:6–7.
- Brandborg LL. Parasitic diseases. In: Sleisenger MH, Fordtran JS (eds). *Gastrointestinal diseases.* W.B. Saunders: Philadelphia, 1978, pp 1154–81.
- Wani NA, Shah OJ, Wani MA. Surgical complications of abdominal ascariasis. *Postgrad Doctor Afr.* 2002;24:38–40.
- Beggs I. The radiology of hydatid disease. *AJR Am J Roentgenol.* 1985;145:639–48.
- Al Hashmi HM. Intrabiliary rupture of hydatid cyst of the liver. *Br J Surg.* 1971;38:228–32.
- Uflacker R, Wholey MH, Amaral NM, Lima S. Parasitic and mycotic causes of biliary obstruction. *Gastrointest Radiol.* 1982;7:173–9.
- Bhargava DK. Endoscopy and biliary parasites. *Gastrointest Endosc Clin N Am.* 1996;6:139–52.
- Hamaloglu E. Biliary ascariasis in fifteen patients. *Int Surg.* 1992;77:77–9.
- el Sheikh Mohamed AR, al Karawi MA, Yasawy MI. Modern techniques in the diagnosis and treatment of gastrointestinal and biliary tree parasites. *Hepatogastroenterology.* 1991;38:180–8.
- Ochoa B. Surgical complications of ascariasis. *World J Surg.* 1991;15:222–7.
- Khuroo MS. Ascariasis. *Gastroenterol Clin North Am.* 1996;25:553–7.
- Yang SCH, Laube PL. Biliary ascariasis: report of 19 cases. *Ann Surg.* 1946;123:299–303.
- Khuroo MS, Zargar SA, Mahajan R. Hepatobiliary and pancreatic ascariasis in India. *Lancet.* 1990;335:1503–6.
- Harris JD. Rupture of hydatid cysts of the liver into the biliary tracts. *Br J Surg.* 1965;52:210–4.
- Vicente VF, Garcio EM, Marco AS. Endoscopic retrograde cholangiography (ERCP) and complicated hepatic hydatid cyst in the biliary tract. *Endoscopy.* 1984;16:124–6.
- Alper A, Ariogul O, Emre A, Uras A, Okten A. Choledochoduodenostomy for intrabiliary rupture of hydatid cysts of liver. *Br J Surg.* 1987;74:243–5.
- al Karawi MA, el-Shiekh Mohamed AR, Yasawy MI. Advances in diagnosis and management of hydatid disease. *Hepatogastroenterology.* 1990;37:327–31.
- Hwang MC, Kim TK, Ha HK, Kim PN, Lee MG. Biliary Ascariasis: MR Cholangiography findings in two cases. *Korean J Radiol.* 2001;2:175–8.
- Alper F, Kantarci M, Bozkurt M, Ozturk G, Onbas O, Balik AA. Acute biliary obstruction caused by biliary ascariasis in pregnancy: MR cholangiography findings. *Clin Radiol.* 2003;58:896–8.
- Schulman A, Loxton AJ, Heydenrych JJ, Abdurahman KE. Sonographic diagnosis of biliary ascariasis. *AJR Am J Roentgenol.* 1982;139:485–9.
- Suwan Z. Sonographic findings in hydatid disease of the liver: comparison with other imaging methods. *Ann Trop Med Parasitol.* 1995;89:261–9.
- al Karawi MA, Yasawy MI, El-Sheikh MAR. Endoscopic management of biliary hydatid disease: report on six cases. *Endoscopy.* 1991;23:278–81.
- Zargar SA, Javid G, Khan BA, et al. Endoscopic sphincterotomy in the management of bile duct stones in children. *Am J Gastroenterol.* 2003;98:586–9.
- Ozcan N, Erdogan N, Kucuk C, Ok E. Biliary ascariasis: percutaneous transhepatic management. *J Vasc Interv Radiol.* 2003;14:391–3.

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