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CLINICAL CASE REPORT

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Intrahepatic Duct Stones Harboring Ascariasis Ova

A Case Report

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Abstract: *Ascariasis lumbricoides* is one of the most common helminthic infestations in humans. Despite the fact that the prevalence of ascariasis in developed countries has been decreasing, biliary ascariasis can cause serious complications, such as acute cholangitis, pancreatitis, and liver abscess. Here we presented a rare ascariasis-related complication—hepatolithiasis.

A 60-year-old female patient had symptoms of recurrent cholangitis. Abdominal computed tomography scan revealed left intrahepatic duct stones with left liver lobe atrophy. Endoscopic retrograde cholangiopancreatography was performed, but the stones could not be removed due to left main intrahepatic duct stenosis. The patient was treated with left hemi-hepatectomy. Unexpectedly, *Ascaris* ova were found on the histopathological examination. She received antihelminthic therapy orally and was on regular follow-up without any complications.

Our study indicates that clinicians should be aware of biliary ascariasis in patients with hepatolithiasis, though not living in endemic areas.

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Abbreviations: CBD = common bile duct, CT = computed tomography, ERCP = endoscopic retrograde cholangiopancreatography, IHD = intrahepatic duct.

INTRODUCTION

Ascariasis lumbricoides is one of the most common helminthic infestation in Asia, Africa, and South America.[1–3] In Taiwan, the prevalence was high before and around 1960 when up to 80% of school children were infected.[4] However, it has dramatically been decreasing during the past decades due to the successful national control activities and the increase in standard of living. Only 10 cases (0.12%) had been diagnosed with biliary ascariasis among 8160 cases who were admitted for biliary tract diseases in a single center in Taiwan from 1982 until 1987.[5] Although the majority of infected individuals are asymptomatic, the adult worms may migrate into the bile ducts or the pancreatic duct, leading to serious complications.[6–10] Here we presented a case of ascariasis-related hepatolithiasis. The diagnosis was made from the pigment stones harboring *Ascaris* ova, instead of round worms.

CASE PRESENTATION

A 60-year-old female patient presented with right hypochondrial dull pain radiating to the back and upper shoulders bilaterally. She had been living and working at a farm in rural area since the age of 15 without special traveling history. There was a history of increased frequency of intermittent abdominal pain and occasional fever for at least 2 years. The patient looked ill, poorly nourished, but no jaundice; physical examination revealed right upper abdominal tenderness, but no rebounding pain. Vital signs were within normal limits. Blood tests were as follows: hemoglobin 9.2 g/dL, hematocrit level 29.4%, alkaline phosphatase 112 U/L, whereas the white blood cell count, serum amylase level, and the remaining liver function tests were within normal limits. The stool microscopic examination was negative. Abdominal computed tomography (CT) scan revealed left intrahepatic duct (IHD) stones with atrophy of left lobe of liver (Figure 1A), whereas endoscopic retrograde cholangiopancreatography (ERCP) could not remove the stones due to left main IHD stenosis (Figure 1B).

As a result, the patient was taken up for surgery. Grossly, the left liver parenchyma was markedly atrophied with fibrotic change. Left hemi-hepatectomy with cholecystectomy was performed; pigment stones were removed after opening the left IHD, which contained white bile due to obstruction (Figure 2). The common bile duct (CBD) was explored and main biliary trees were examined by a choledochoscope. Biliary stricture and pigment stones were found in the left hepatic duct. Final pathology reports showed that the liver parenchyma had inflammatory and fibrotic change (Figure 3A), whereas it consisted of dilated bile ducts with pigment stones harboring *Ascaris* ova (Figure 3A and B). The histological examination revealed active fibrosis and an inflammatory infiltrate consisting of lymphocytes, eosinophils, and plasma cells around the dilated bile ducts in which pigmented stones resided. Marked proliferation of small bile ductules with atrophy of hepatic parenchyma was noted. The hepatic parenchyma showed pronounced fibrous expansion of portal tracts and frequent bridging fibrosis (Figure 3A and B). The postoperative period was uneventful. The patient received antihelminthic therapy orally and was on regular follow-up for 4 years without any complications.

The Chang Gung Memorial Hospital ethics committee approved this study (CGMH IRB No. 103-2474B), and informed consent was obtained from the patient.

DISCUSSION

Ethnic and environmental factors are also thought to be related to hepatolithiasis since its prevalence is much higher in East Asia than in Western countries.[11] From the view of pathogenesis, metabolic problems, bacterial infection, and bile stasis with bile duct strictures cause this disease.[12] Parasitic

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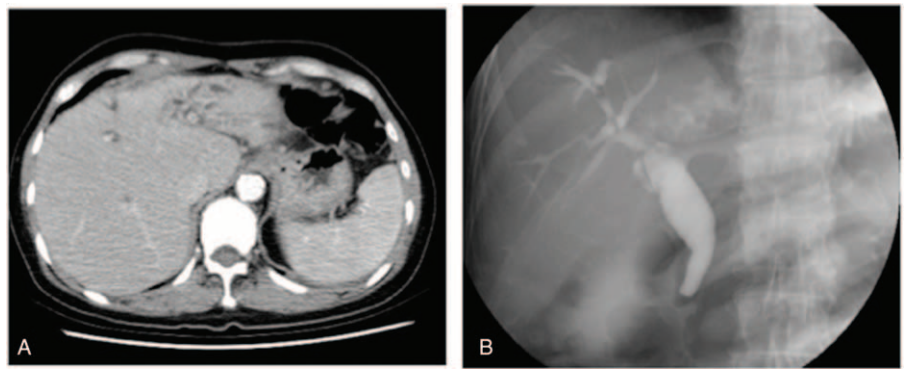

FIGURE 1. A, Computed tomography (CT) scan showed left liver atrophy with IHD stones and biliary dilatation. The configuration of contralateral lobe was normal without obvious lesions. B, ERCP showed left main IHD stenosis over hilar area with stones in left hepatic lobe. Common bile duct was dilated, whereas right intrahepatic ducts remained intact. ERCP = endoscopic retrograde cholangiopancreatography, IHD = intrahepatic duct.

infestations (eg, *Ascaris lumbricoides* and *Clonorchis sinensis*) have been cited as a cause of biliary complication.[13–15] However, worms and ova are found rarely in cases with hepatolithiasis. Moreover, the endemic areas of infestation are not correlated with the areas with a high prevalence of hepatolithiasis.[16]

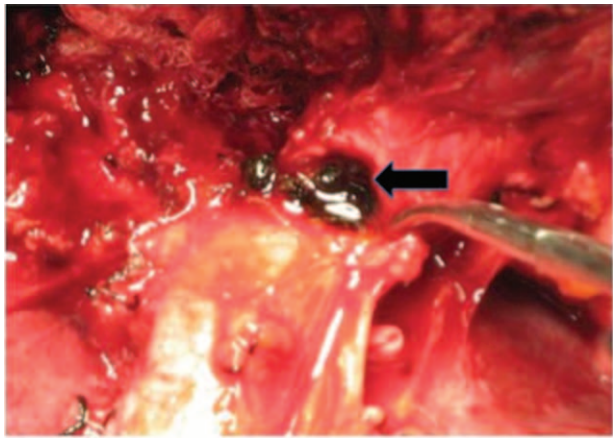

FIGURE 2. Pigment stones (black arrow) in left IHD were shown intraoperatively. IHD = intrahepatic duct.

Ascariasis is a common infestation in tropical and subtropical regions, especially in areas where sanitation is poor.[3] The common symptoms of biliary ascariasis include biliary colic, acute cholangitis, cholecystitis, and acute pancreatitis; however, rare presentations like stricture of bile ducts, liver cirrhosis, hepatic abscess, pancreatic abscess, and granulommataous hepatitis (pseudotumor) can occur.[6,8,17] Ascaris ova have an albumioid membrane that is highly cohesive and thus facilitates the precipitation of calcium carbonate crystals on its surface.[18] Moreover, *Ascaris* worms have high glucoronidase activity that deconjugates bilirubin and form pigment stones.[19] Dead worms or ova serve as a nidus for pigment stone formation; consequently, recurrent suppurative cholangitis can occur even after active infestation has been resolved.[20]

Ascariasis-related complications of the biliary and pancreatic systems are most commonly caused by the adult worms migrating across the ampulla of Vater, leading to obstruction.[21] Ultrasonography is usually the first imaging study for biliary ascariasis[2,22,23]; however, ERCP is the choice of investigation.[7,24] Biliary and pancreatic problems are usually resolved by ERCP and other conservative therapies. Surgical interventions for ascariasis-related hepatobiliary problems are rarely indicated. Major liver resection for ascariasis-related hepatolithiasis with bile duct stricture and liver atrophy is much rarer. The long-standing infestation, stone formation, and recurrent attacks of cholangitis are enough factors to show such a late and complicated condition.

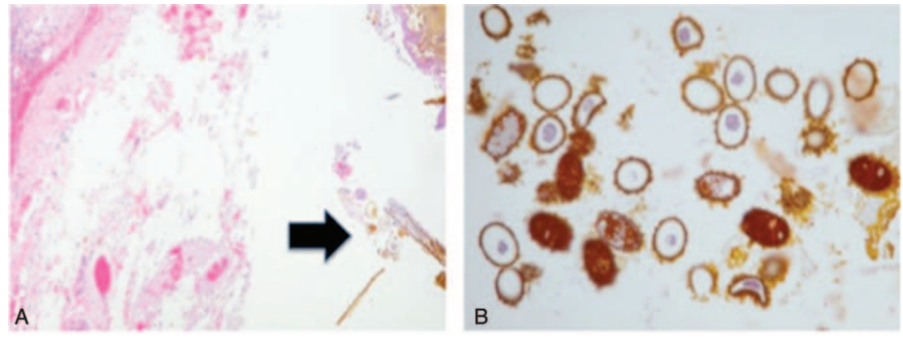

FIGURE 3. A, Histopathology examination of the liver specimen showed acute and chronic inflammation of liver parenchyma with lithiasis harboring *Ascaris* ova (black arrow, 10× objective). B, *Ascaris* ova under higher magnification (40× objective).

The treatment of biliary ascariasis by endoscopic extraction of calculi and worms from the bile duct with or without sphincterotomy provides immediate relief. However, in presence of chronic complications, as in our case, surgical management was the only resort. We were not able to identify traces of round worms in the preoperative studies of the patient. In an interesting comparative study, it was found that choledocholithiasis, hepatolithiasis, liver abscess, and cirrhosis were associated with dead worms, not living worms.[8] Nevertheless, antihelminthic drugs are mandatory to ensure eradication of primary cause, because unbroken ova shells with inner integrity were found on the histopathological examination.

CONCLUSIONS

Our study indicates that clinicians should be aware of biliary ascariasis in patients with hepatolithiasis, though not living in endemic areas. Maintaining a high level of suspicion to diagnose biliary ascariasis is paramount for proper management and avoiding long-term complications.

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