Role of Therapeutic Endoscopy in Hepatic Hydatid Disease after Surgical Intervention: Case Report
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Citation

Abstract
A 79-year-old female diagnosed as hepatic hydatid disease after hepatectomy was referred with jaundice and right hypochondralgia. An endoscopic retrograde cholangiography (ERC) showed a defect in the bile duct. We performed bile duct stenting, but stent occlusion occurred repeatedly. Bile duct stenting was performed 7 times and endoscopic naso-biliary drainage once. Although bile duct stenting was performed repeatedly, ERC provided an excellent diagnostic and therapeutic modality in the present case; thus, it should be considered as a definitive treatment in similar cases, particularly after hepatectomy or if the surgical risk is anticipated to be high.

INTRODUCTION
Hydatidosis is a zoonosis that is generally caused by infection with Echinococcus granulosus. The disease is endemic in many countries, such as those around the Mediterranean Sea, as well as in Central and Eastern Asia and Latin America (1). The liver is the most common site of hydatid cysts (1, 2). Rupture of a hydatid cyst into the biliary tract is common and is manifested as obstructive jaundice or cholangitis (1, 2). In case of intrabiliary rupture, endoscopic retrograde cholangiography (ERC) with endoscopic sphincterotomy or bile duct stenting achieves decompression of the biliary tract from intracystic debris and prevents the recurrence of obstructive jaundice. We herein describe a case of hepatic hydatid obstructive jaundice after hepatectomy.

CASE REPORT
A 79-year-old woman with jaundice and right hypochondralgia was referred to our hospital in January 2008. Right hepatectomy was performed in April 2003 because of hepatic hydatid disease, and the margin was found to be positive for hydatid cysts. Her height and weight were 151 cm and 42 kg, respectively; she had been treated with albendazole.

Physical examination revealed right hypochondralgia and jaundice; her body temperature was 37.6 ℃. Her white blood cell count was 12,900 /μL, and the levels of C-reactive protein, alkaline phosphatase, and total bilirubin were 12.4 mg/dL, 1,135 U/L, and 18.5 mg/dL, respectively. Abdominal ultrasonography showed a dilated intrahepatic bile duct, and abdominal computed tomography (CT) showed lesions with calcifications and a dilated left intrahepatic bile duct (Fig. 1). Magnetic resonance cholangiopancreatography revealed the obstruction of a 2 cm long bile duct (Fig. 2). A
diagnosis of acute cholangitis with obstructive jaundice was made, and intrabiliary rupture of a relapsing hydatid cyst was suspected to be the cause of this condition.

ERC (Fig. 3A, B) and bile duct stenting (Cotton-Huibregts® Biliary Stent Sets; Wilson-Cook Medical, Inc., NC, USA) were performed on January 22, 2008 (Fig. 4A-D), and the intracystic fluid and debris drained to the bile duct stent. Two months after bile duct stenting, the level of total bilirubin was 1.5 mg/dL, and no dilated intrahepatic ducts were observed on abdominal CT scans (Fig. 5). The patient was discharged in March 2008; however, she was referred to our hospital again in July 2008 with jaundice and right hypochondralgia probably because of bile duct stent occlusion.

The patient was admitted to our hospital 5 times during a period of 3 years, and bile duct stenting was performed 7 times, while endoscopic nasobiliary drainage was performed once. The last bile duct stenting (plastic stent; 8.5Fr, 12 cm) was performed in September 2010, and she survived for a period of 7 years and 8 months after right hepatectomy.
Figure 3
Figure 3: Endoscopic retrograde cholangiography (ERC) showed bile duct obstruction (A) and bile duct stenting (plastic stent; 8.5 Fr, 12 cm; B) ( ).

Figure 4
Figure 4: Bile duct stenting. ERC was performed for cannulation into an orifice of Vater’s papilla (A), exchange of a guidewire (B), and insertion of a bile duct stent (C, D).

Figure 5
Figure 5: Abdominal CT showed the bile duct stent ( ) and disappearance of the dilated intrahepatic ducts.

Figure 6

Table 1. Characteristics of the patients with hepatic hydatid obstructive jaundice who underwent endoscopic bile duct stentings from 1983 to 2010 based on the Japanese medical database

<table>
<thead>
<tr>
<th>Patient</th>
<th>Year</th>
<th>Age</th>
<th>Sex</th>
<th>Liver segment</th>
<th>Neoplasm</th>
<th>Neoplasm type</th>
<th>Neoplasm length</th>
<th>Treatment</th>
<th>Post-HDPI segment</th>
<th>Mortality</th>
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<tr>
<td>Takeda 1990</td>
<td>21</td>
<td>F</td>
<td>Rt liver</td>
<td>Curved duct</td>
<td>6.5 cm</td>
<td>PEET</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Takahara 2000</td>
<td>50</td>
<td>M</td>
<td>Lt liver</td>
<td>Expandable metallic stent</td>
<td>6 cm</td>
<td>PEET</td>
<td>Hepatic duct junction</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tanaka 2001</td>
<td>80</td>
<td>M</td>
<td>Lt liver</td>
<td>Expandable metallic stent</td>
<td>8 cm</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Our case</td>
<td>2008</td>
<td>70</td>
<td>F</td>
<td>Rt extrahepatic bile duct</td>
<td>Plastic stent</td>
<td>12 cm</td>
<td></td>
<td></td>
<td></td>
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</tr>
</tbody>
</table>

DISCUSSION
Intrabiliary rupture of a hepatic hydatid cyst is a common complication and may occur in 2 forms: an occult rupture, in which only the cystic fluid drains to the biliary tree and is observed in 10-37% of the patients; and frank rupture, which has an overt passage of intracystic material to the biliary tract and is observed in 3-17% of the patients (3, 4). Intrabiliary rupture mainly occurs in centrally localized cysts, and an intracystic water pressure up to 80 cm is also a predisposing factor for the rupture (5).
Intrabiliary rupture occurs in the right hepatic duct (55-60% cases), left hepatic duct (25-30% cases), hepatic duct junction, common bile duct (CBD), or cystic duct (8-11%); perforation into the gallbladder may be observed in 5-6% of cases (6-8). It was thought that the present case was an occult rupture, because the intracystic fluid and debris drained to the bile duct stent.

Intrabiliary ruptures of hydatid cysts have been diagnosed by imaging and laboratory tests. Although these tests were proven to be ineffective in detecting occult ruptures, certain findings from studies conducted in the USA provide essential clues for the diagnosis of frank ruptures with obstruction. Echogenic material, without posterior acoustic shadowing in extrabiliary ducts, implied the presence of intracystic material (9). An abdominal CT scan may reveal a dilated CBD with low attenuation intraluminal material, suggesting the presence of hydatid sand and cysts (10, 11).

Some cases of definitive endoscopic treatment of obstructive jaundice after intrabiliary rupture have been reported; ERC has become the “gold standard” for the assessment of intrabiliary rupture by achieving a detection rate of 86.6% to 100% (3, 12-15). ERC, which is a minimally invasive procedure, may obviate reoperation.

Endoscopy is a modality serving both diagnostic and therapeutic aims. Postoperative ERC has several advantages: I-clarification of the causes of ongoing or recurrent symptoms and laboratory abnormalities; II-resolution of obstructions or cholangitis due to residual material in biliary ducts; III-management of postoperative external biliary fistulae; and IV-provision of a realistic solution for secondary biliary strictures (12, 13, 16, 17).

ERC with bile duct stenting is performed for the treatment of intrabiliary ruptures associated with obstructive jaundice, but this is rare in Japan. Ichushi Web, a Japanese medical database, showed that only 4 Japanese cases, including the present case, were reported from 1983 to 2010 (Table 1). In all cases, bile duct stenting was repeatedly performed because of stent occlusion, but major complications were not encountered after endoscopic procedures.

In conclusion, therapeutic endoscopy is a safe and valuable procedure for the postoperative management of patients with hepatic hydatid disease.
References

16. Özslan E: Therapeutic endoscopic retrograde cholangiopancreatography and related modalities have many roles in hepatobiliary hydatid disease. World J Gastroenterol; 2006; 12: 4930-1.
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