Endoscopic and surgical management of intrabiliary rupture of hydatid liver cyst

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S LEONG, Y-I KIM, R GRAY, P KORTAN, G HABER. Endoscopic and surgical management of intrabiliary rupture of hydatid liver cyst. *Can J Gastroenterol* 1992;6(3):135-139. A man with hydatid disease complicated by intra-abdominal cyst rupture 15 years earlier, presented with cholestatic jaundice. There was intrabiliary rupture of a hydatid liver cyst on endoscopic retrograde cholangiography. Sphincterotomy was performed to allow clearance of hydatid material obstructing the bile ducts and insertion of a nasobiliary catheter for irrigation and drainage. Definitive surgery was performed. While endoscopic management is gaining recognition for relieving biliary obstruction in hydatid cystobiliary rupture, surgery is still required for patients who continue to pass hydatid debris obstructing the biliary tree and increasing the risk of cholangitis.

Key Words: Bile duct, Endoscopic retrograde cholangiography, Hydatid cyst, Sphincterotomy

Traitement endoscopique et chirurgical d'un kyste hydatique du foie rompu dans les voies biliaires

RÉSUMÉ: Un homme atteint d'une maladie hydatique compliquée d'un kyste intra-abdominal rompu 15 ans auparavant se présente avec une jaunisse cholestatique. À la cholangiographie endoscopique rétrograde, on a pu observer la présence d'un kyste hydatique du foie rompu dans les voies biliaires. Une sphinctérectomie a été pratiquée afin de permettre l'élimination des tissus hydatiques qui obstruaient les voies biliaires et l'insertion d'un cathéter nasobiliaire pour l'irrigation et le drainage. Le tout a été suivi d'une chirurgie définitive. Bien que l'endoscopie soit de plus en plus employée dans le traitement de l'obstruction biliaire causée par la présence d'un kyste hydatique du foie rompu, la chirurgie est toujours nécessaire chez les patients qui continuent à éliminer des débris hydatiques qui obstruent l'arbre biliaire et qui présentent un risque accru à l'égard de la cholangite.

HYDATID DISEASE IS OFTEN ASYMPTOMATIC until complications occur and this uncommon entity may be overlooked in an emergency situation when a previously healthy patient presents for the first time. Hydatid cyst of the liver is the most common site and it produces symptoms when the cyst becomes infected, compresses or ruptures into adjacent structures or grows to an enormous size (1). A case illustrating the two main complications of hydatid disease (1,2) – rupture of a hydatid liver cyst into the peritoneal cavity and, many years later, another rupture into the biliary tree – is presented. Both conditions may be life threatening and prompt management is vital for a successful outcome.

CASE REPORT

A 37-year-old man of Italian descent presented with jaundice associated with abdominal discomfort for two days. He had emigrated to Canada at 12 years of age. During his childhood he lived with his parents who were farmers, tending cattle, sheep, pigs and dogs.

At 22 years, he experienced severe abdominal pain after being hit by a ball in the abdomen. Emergency laparotomy revealed a ruptured hydatid cyst in the liver. Partial left hepatic lobectomy was performed and he made an uneventful recovery.

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Figure 1) Computed tomography scan of abdomen showing two hydatid cysts in liver

Figure 2) Cholangiogram showing filling defect due to hydatid membranes in the left hepatic duct and common bile duct

At 32 years, an ultrasound and computed tomography scan of the abdomen showed two small liver cysts (Figure 1) at a pre-employment check-up. No

treatment was given as they were thought to be dead cysts.

At 36 years, the patient presented with lower abdominal pain. Imaging

with ultrasound revealed a new pelvic cyst. He underwent a second laparotomy. The cyst was adherent to the bladder wall anteriorly, the rectum posteriorly and laterally to the pelvic wall. It was dissected intact and removed. It measured 5 cm in diameter and on sectioning, multiple daughter cysts were seen. With the exception of the previously noted liver cysts, no other lesions were found.

At 37 years, the patient presented with jaundice associated with upper abdominal discomfort. On abdominal examination, upper and lower midline surgical scars and a tender, enlarged liver were noted. The liver function tests were consistent with cholestatic jaundice and coagulation profile was normal. Ultrasound of the abdomen revealed a distended gall bladder and a cystic lesion in the remnant left lobe, measuring 3.9 by 4.5 cm. The common bile duct was dilated to 1.6 cm. The intrahepatic ducts were also dilated. Endoscopic retrograde cholangiography showed a dilated biliary tree with a cystic dilation in a branch of the left hepatic duct containing hydatid material (Figure 2). Endoscopic sphincterotomy was performed and yellowish gelatinous membranes were seen extruding from the ampullary orifice. A Dormia basket was passed to evacuate hydatid debris from the bile ducts. There was dramatic recovery with relief of biliary obstruction.

Two weeks later the jaundice recurred, associated with fever and chills, suggesting cholangitis due to biliary obstruction from continuing discharge of daughter cysts. At urgent endoscopic retrograde cholangiography, yellowish jelly-like hydatid material and pus were seen protruding out of the ampulla of Vater (Figure 3). There was hydatid debris in the dilated common bile duct and left hepatic radical, which was communicating with the cystic cavity (Figure 4). After evacuation of the debris with a basket, a nasobiliary catheter was inserted into the cyst cavity to improve drainage and allow irrigation with saline. The biliary fluid and sediments did not contain any hooklets or protoscolices on microscopy. Hence there was no evidence for

Figure 3) Hydatid membrane protruding out of opening of ampulla of Vater

Figure 4) Cholangiogram showing metal basket in cyst cavity communicating with left hepatic duct

viability of the parasites and medical therapy with mebendazole was not given. A repeat ultrasound of the abdomen showed a cystic cavity communicating with the left main hepatic duct which was dilated to 8 mm.

Although the fever and jaundice subsided with a repeat clearance of the bile ducts, surgical treatment was elected to eradicate the disease. Resection of the liver containing the cyst was performed together with cholecystectomy and insertion of a T-tube to allow intra- and postoperative cholangiography. Recovery was uneventful and nine months later the patient was asymptomatic and had returned to work without evidence of residual disease on abdominal ultrasound examination.

DISCUSSION

Hydatid disease is a parasitic infection most commonly caused by the cestode *Echinococcus granulosus*. Very rarely it is due to *Echinococcus multi-*

locularis (3). Humans, sheep and cattle are intermediate hosts and the dog is the common definitive host (1). Hydatid disease is endemic in sheep-rearing countries and it is likely that the present patient acquired the infection during his childhood in Italy from handling dogs or eating vegetables contaminated with the ova of the parasite.

With increasing immigration, human echinococcosis may be on the rise in North America since the infection is acquired in the native country during childhood. In Canada it is more common in people of Greek and Italian descent (4).

Rupture of an echinococcal cyst was the first indication of hydatid disease in this patient. Lewall et al (5) described three types of rupture: contained, communicating and direct, the last being most serious because spilling of cyst contents into the peritoneal cavity may cause allergic and infective sequelae. It is thought that patients with direct rupture should receive anthelmintic

drugs to prevent peritoneal hydatidosis (6). The rupture is communicating when cyst contents escape via biliary radicles leading to obstruction of the biliary tree and cholangitis as in the presented patient. There has been no evidence that transbiliary dispersion of scolices leads to dissemination of echinococcal disease.

Cyst rupture may be due to external trauma (direct rupture) as in the initial presentation of this case, or degeneration of the parasitic membranes (communicating rupture) resulting in the release of hydatid daughter cysts and debris into the common bile duct. Hydatid cyst rupture into the biliary tree is a serious complication occurring in 5 to 15% of patients with hepatic involvement (7,8). Diagnosis was made only at laparotomy until sonography became available (7-9). Sonographic visualization of ruptured cyst material into the biliary tree and communication between a cyst and the biliary tract indicate intrabiliary rupture of a

hepatic hydatid cyst (10-12). Together with computerized axial tomography, which can demonstrate the cyst-bile duct communication sometimes difficult to visualize with ultrasound, the diagnostic accuracy can reach 96% for intrabiliary hydatid cyst rupture (13,14).

Cottone *et al* (15) first reported the use of endoscopic retrograde cholangiography in the diagnosis of hepatic hydatid cyst rupture into the biliary tree. The endoscopic picture of yellowish gelatinous membranes protruding out of the ampulla suggest hydatid cystobiliary rupture and this is confirmed by pathological examination of the infected bile which will contain fragmented membranes and daughter cysts. The presence of pus suggests secondary infection and requires administration of appropriate antibiotics. The cholangiogram shows a dilated biliary tree with filling defects representing daughter cysts and hydatid membranes. The leaf-like radiological appearance of these membranes sometimes changes in shape on serial cholangiogram and it differentiates hydatid material from choledocholithiasis (16,17). Entry of contrast into a cystic space points to a communicating type of rupture. Other radiological fea-

tures include extrinsic compression of the biliary tract by a hepatic cyst, complete obstruction of extrahepatic bile ducts and biliary fistula (18). Percutaneous transhepatic cholangiography has been used to demonstrate the rupture of a cyst into the biliary tree (19,20). However, there is the risk of peritoneal seeding or anaphylaxis although diagnostic and therapeutic percutaneous aspiration of an hepatic hydatid cyst has also been described (21).

Before the advent of therapeutic endoscopy, surgery was the only effective mode of treatment for hydatid disease (15). When the hydatid cyst can be completely removed, surgical excision, partial hepatectomy or hepatic lobectomy is performed (4). In addition, cysts that have ruptured into the biliary tract must be treated as infected and the common bile duct must be explored and drained with a large calibre T-tube which would also allow a cholangiogram to be done to ensure common bile duct clearance before its removal (22).

Decompression of the biliary tree by endoscopic sphincterotomy was first reported in 1986 by Shemesh *et al* (23). Evacuation of hydatid debris from the obstructed biliary tree was safe and effective in preventing sepsis (23-25).

Patients with good clearance of the bile ducts avoided surgery (26,27). In post-operative patients, the cause of jaundice was quickly found and endoscopic sphincterotomy with insertion of a nasobiliary catheter for irrigation replaced surgery in some patients (26). Scolicidal agents such as hypertonic saline can be used to irrigate and kill any viable parasites via the nasobiliary drain (27). This was not used in the present patient as the pathologist found no viable parasites. However, frequent irrigation via a nasobiliary catheter helps to clear the bile ducts of debris and this might have prevented a recurrence of cholangitis after the first endoscopic retrograde cholangiography and endoscopic sphincterotomy. More likely, it is the continued discharge of daughter cysts that prevents bile drainage. Other possible causes include incomplete extraction of common bile duct debris and inadequate sphincterotomy.

While endoscopic treatment of intrabiliary rupture of hydatid cyst may temporarily relieve biliary obstruction, surgery should be considered if there is persistent discharge of hydatid debris obstructing the bile ducts. This will prevent cholangitis and eradicate hydatid disease.

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