CASE REPORT

Cholecysto-Hydatid Cyst Fistula

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ABSTRACT
We present a case of recurrent hydatid cyst liver in a 32-year-old female. Previous surgery was performed 8 years ago elsewhere. Pre-operative assessment at presentation revealed a 110 x 105 mm hydatid cyst in the right lobe of the liver. On exploration, a 5 mm fistulous communication was found between the hydatid cyst and the gallbladder. Patient was subjected to endocystectomy (partial cystectomy), cholecystectomy and closure of the fistula followed by obliteration of the cavity with omentum. Postoperative recovery was uneventful.

Key words: Hydatid cyst. Fistula. Gallbladder. Cholecystectomy

INTRODUCTION
Liver is the most commonly affected organ in echinococcosis. The rupture of hydatid cyst liver into the biliary tree is the most common complication.1 The communication is usually minute and occult. It has been described in upto 90% of hepatic hydatid cysts.2 This is due to the incorporation of biliary radicles into the pericyst.3 However, frank rupture into the biliary tree occurs in only 5-15% of cases.2 Although, the communication with the intrahepatic bile ducts is common, its occurrence between a hepatic hydatid cyst and the gallbladder is rare. We describe management of one such case.

CASE REPORT
A 32-year-old female, presented with 3 months history of pain in the right hypochondrium and epigastrium. Pain was intermittent, dull in character and not associated with meals. There was no history of alteration of bowel habits, urinary complaints, jaundice, fever and weight loss. About 8 years back, she underwent a surgical intervention in another setup for hydatid cyst liver. The operative details were not available. On examination, she was a thin-built lady without pallor and jaundice. Abdominal examination revealed a well healed right subcostal scar of the previous surgery. Liver was palpable 5 cm below the right costal margin with deep tenderness in the epigastrium and right hypochondrium. The rest of the clinical examination was unremarkable. Her haemoglobin was 10 g/dl, total leucocyte count of 8.9 x 109/L with 4% eosinophils. Serum bilirubin was in normal (10.2 µmol/L), range and so was serum alanine aminotransferase. Serum alkaline phosphatase was raised (1420 U/L). Ultrasonography (Figure 1) revealed a 110 x 105 mm cystic lesion in the right lobe of the liver with multiple small cysts in the cavity, suggestive of hydatid cyst liver (recurrent). The gallbladder was distended with sludge and the common bile duct/intrahepatic ducts were not dilated. X-ray chest was unremarkable.

Patient was put on oral albendazole for 2 weeks and then surgical intervention was done under general anaesthesia. Abdomen was opened through the previous surgical scar. A lot of adhesions were encountered. Liver was then separated from the duodenum and the diaphragm. Gallbladder found distended and a cyst was noted in the right lobe of liver just below the diaphragm extending to the midline. Twenty percent hypertonic saline was used as the scolicidal agents after partial aspiration. The contents of the cyst could not be emptied probably due to the thick consistency. The cyst was opened along with continuous suction and the contents were emptied. The cyst contained a lot of daughter cysts along with hydatid sand and bile stained debris. After emptying the cyst, the gallbladder was found to be empty as well. On careful evaluation of the cyst, a fistulous communication of about 5 mm was found between the cyst and the gallbladder. Cholecystectomy was planned. Due to
dense adhesions, fundus first technique was used. Gallbladder was slit open in order to localize the fistulous tract (Figure 2). Sub-total cholecystectomy was performed and the fistulous opening in the cyst closed with interrupted sutures. After confirming no other opening of the biliary tract into the cyst, omentum was mobilized and placed to obliterate the cavity of the hydatid cyst in the liver. Postoperative recovery was smooth. Patient was again started on oral albendazole and was followed-up in the OPD regularly. She was symptom-free at 2 months follow-up.

**DISCUSSION**

Hydatid disease is a zoonosis, caused by the larval stage of the genus Echinococcus. Echinococcosis has a worldwide distribution. It is endemic in many Mediterranean countries, the Middle and Far East, South America, Australia, New Zealand and East Africa. Approximately 70% of hydatid cysts are located in the liver and there are multiple cysts in one-quarter to one-third of these cases. The clinical features of hydatid disease depend on the site, size, stage of development, whether the cyst is dead or alive and complications.

The cyst grows in the direction of the least resistance. The rupture is the result of its enlargement. Live hydatid cysts can rupture into physiologic channels, free body cavities or adjacent organs. The other factor responsible for fistulization of hydatid disease is inflammation. Infection and continued expansion of the cyst cause pressure erosion and adhesion to the adjacent structures. In time, with increasing intracystic pressure, the cyst ruptures. Inflammation leads to necrosis and causes fistulization.

The formation of a fistula with the gallbladder is very rare. The cases described in the medical literature are limited. Settef et al. reported 3 cases of gallbladder fistula, Sabat et al., Kumar et al. and Adaleti et al. have documented one case each. In recurrent hydatid disease, the publication by Kumar et al. was the first report of a fistula with gallbladder.

The pre-operative diagnosis of the hydatid cyst showing a fistulous communication with the biliary tract, gallbladder or other sites, can be made directly on different imaging studies. Ultrasonography can identify daughter cysts within the biliary tract, however, in this case the diagnosis was per-operative. Indirect signs of a biliary communication include increased echogenicity on sonography. An air-fluid level within the cyst, previously described as a sign of infection, is considered a sign of either rupture into the biliary tree/hollow viscus or bronchopleural fistula/intrabronchial rupture in a lung hydatid cyst. Computerized Tomography (CT) scan can demonstrate high attenuation material passing through the defect of the cyst wall and filling-up the intrahepatic biliary radicles, gallbladder or common bile duct. Endoscopic Retrograde Cholangiopancreatography (ERCP) and Percutaneous Transhepatic Cholangiography (PTC) can show the communication in more detail. ERCP is the Gold standard in confirming biliary tract involvement and may be of therapeutic benefit as well. Magnetic Resonance Imaging (MRI) and Magnetic Resonance Cholangiopancreatography (MRCP) are used in difficult cases in which CT and sonography may be inconclusive.

Treatment options in such a case consist of drainage and sterilization of the cyst followed by either radical (total cystopericystectomy) or conservative (partial pericystectomy) excision combined with cholecystectomy. Common Bile Duct (CBD) exploration and biliary lavage followed by either T-tube drainage or transduodenal sphincteroplasty can be performed specially in fistulas >5 mm in size. In this case, we did not go for the CBD exploration as there was no biochemical evidence of obstructive jaundice, the isolated rise in serum alkaline phosphatase was probably due to the space occupying lesion in the liver, sonography showed a normal sized CBD with no daughter cysts and lastly due to the previous surgery there were a lot of adhesions around the porta hepatis and further dissection of this area could have been more hazardous. The surgery is usually followed by medical treatment with benzimidazole carbamates (albendazole, mebendazole) to prevent the recurrence, as done in this case.
REFERENCES


