Case Report

Management of a Patient with Hepatic-Thoracic-Pelvic and Omental Hydatid Cysts and Post-operative Bilio-Cutaneous Fistula: A Case Report

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ABBREVIATIONS: Magnetic Resonance Imaging (MR); Endoscopic Retrograde Cholangiopancreatography (ERCP)

SUMMARY

In humans, most hydatid cysts occur in the liver and 75% of these are single. Our patient was a 31 year-old male. His magnetic resonance imaging (MR) showed one cyst (15x20cm) in the right lobe and three cysts (5x6cm, 8x6cm, and 5x5cm) in the left lobe of the liver, two cysts (4x5cm and 5x5cm) on the greater omentum, and two cysts (15x10 and 10x10cm) in the pelvis. The abdomen was entered first by a bilateral subcostal incision and then by a Phennenstiel incision. Partial cystectomy+capitonnage was done on the liver cysts; the cysts on the omentum were excised, and the pelvic cysts were enucleated. The cyst in the right lobe of the liver was in communication with a thoracic cyst. An air leak developed from the thoracic cyst which had underwater drainage and bile drainage from the drain in the cavity of the right lobe cyst. Sphincterotomy was done on the seventh post-operative day by endoscopic retrograde cholangiopancreatography (ERCP). No significant effect on mean bile output from the fistula occurred. Octreotide therapy was initiated, but due to abdominal pain and gas bloating the patient felt and could not tolerate, it was stopped on the fourth day; besides, it had no decreasing effect on bile output during the 4 days. Because air and bile leak continued and he had bile stained sputum, he was operated on on post-operative day 18. By right thoracotomy, the cavity and the leaking branches were closed. By right subcostal incision, cholecystectomy and T-tube drainage of the choledochus were done. On post-operative day 30, he was sent home with the T-tube and the drain in the cavity. After 3 months post-operatively, a second T-tube cholangiography was done, and a narrowing in the distal right hepatic duct and a minimal narrowing in the distal left hepatic duct were exposed. Balloon dilatation was done by way of a T-tube. Bile drainage ceased. There was no collection in the cavity in follow-up CT scanning, so the drain in the cavity, and the drainage catheter in the right hepatic duct were extracted. Evaluation of the biliary ductal system is important in bilio-cutaneous fistulas, and balloon dilatation is very effective in fistulas due to narrowing of the ducts.

INTRODUCTION

In humans, the most commonly effected organ by *Echinococcus granulosus* is the liver. It may cause no symptoms or signs for a long time, and the diagnosis is usually established incidentally by examinations for malaise, dyspepsia or, rarely, for abdominal mass. Disseminated intraabdominal hydatidosis is very rare. The most common complication of hepatic hydatid cysts is intrabiliary rupture. Treatment of biliocutaneous fistula after hepatic hydatid cyst surgery is difficult and time consuming. It requires long periods of time and the collaboration of many branches of medicine.

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CASE REPORT

A 31 year-old male patient presented with malaise, headache, nasal bleeding, and dyspnea in activity. Two months previously, he felt chills and malaise. In his abdominal MR, a hydatid cyst of 14x16x7cm in the posterior and anterior segments of the right lobe of the liver and another cyst of 18x9x21cm in the medial and lateral segments of the left lobe of the liver, which displaced the stomach to the posterior abdominal wall were found **(Figure 1)**. Another 9x15x12cm cyst in the pelvis, which displaced the bladder to the left, and near to this, above the bladder, a 7x10x5cm cyst **(Figure 2)** were visualized. The liver was hypertrophic and reached over the pelvis. In his thoracic computed tomography (CT), 20 days after his abdominal MR, no intra-thoracic lesions were visualized. In the right lobe of the liver a 16x15x10cm cyst which lay in the cranial part of the liver and extended to the whole of the right lobe was present **(Figure 3)**. In the left lobe of the liver two cysts, 14cm and 12cm, were present. In his clinical examination, the liver edge was palpated 10cm below the right costal arch. The PT time was prolonged (50%).

He was operated on for multiple liver and pelvic hydatid cysts. The abdominal cavity was entered by bilateral subcostal incision. Exploration revealed three cysts of 5x6cm, 8x6cm and 5x5cm in the medial end lateral segments of the left lobe, a cyst of 15x20cm in the posterior segment of the right lobe, and two cysts of 4x5cm and 5x5cm on the greater omentum. First, the cysts in the left lobe were emptied. Fifty percent diluted 7.5% povidone iodine was used to isolate the abdominal cavity while aspirating and emptying the cysts. Then, partial cystectomy and capitonnage was used to get rid of the cystic cavities. The cyst in the right lobe was partially calcified and fixed to the right diaphragm. The cyst was also emptied in the same manner. In the cystic cavity on the diaphragmatic side, there was a hole about 3cm in diameter through which many daughter vesicles were extracted. The liver was freed as much as possible and we saw that the cyst which appeared to be in the liver in MR was in the thoracic cavity and was connected with the cyst in the right lobe of the liver. The cyst was about 10x10cm and it was emptied through the hole in the diaphragm.

An air leak occurred from this cyst. Drains were placed in the cavity of the thoracic cyst and the right pleural cavity and connected to underwater drainage. Partial cystectomy and capitonnage was done on the anterior surface of the cyst, but because the diaphragmatic part of the cyst was extremely calcified and fixed to the diaphragm nothing could be done on the posterior surface of

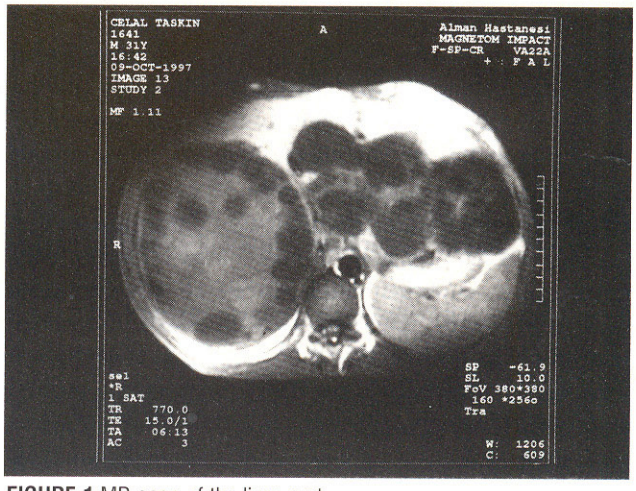


FIGURE 1 MR scan of the liver cyst.

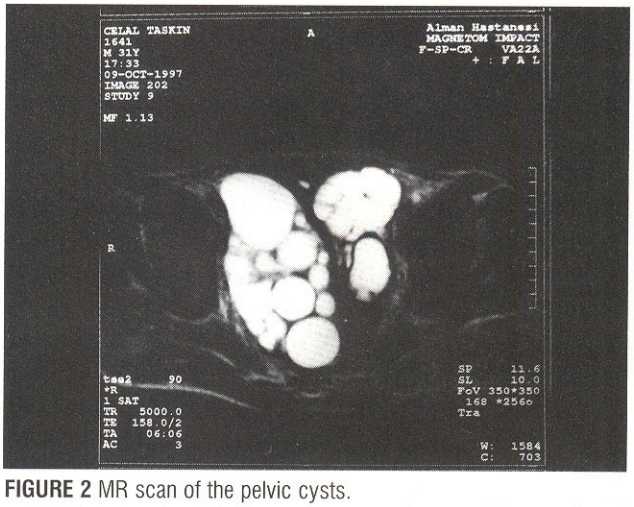


FIGURE 2 MR scan of the pelvic cysts.

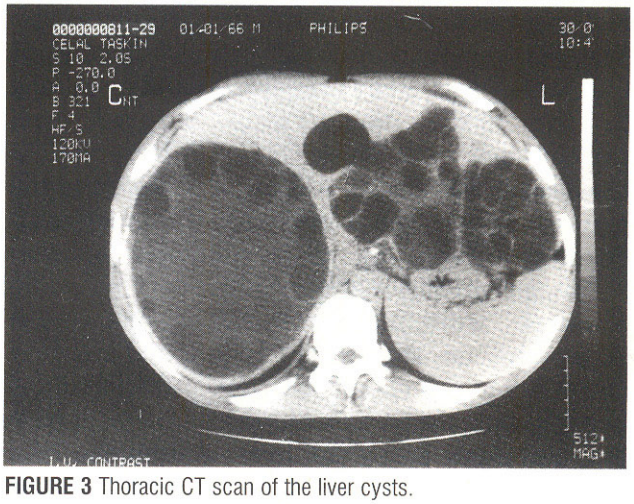


FIGURE 3 Thoracic CT scan of the liver cysts.

the cyst. The omental cysts were excised. The remaining greater omentum was very short and omentoplasty was impossible. Drains were placed in the cavity in the right lobe, below the right diaphragm, below the right and left lobes of the liver, and the incision was closed. Then, a Phannenstiel incision was done. The cysts in the pelvic cavity were freed and completely enucleated. A drain was placed to the Douglas and the incision was closed. The drain below the left lobe was extracted on the fifth post-operative day, the drain below the right diaphragm on the sixth, and the drain in the pelvis on the seventh day. By the drain in the cystic cavity in the right lobe of the liver, bile drainage of about 400-700cc/day occurred. Sphincterotomy by ERCP was done on the seventh post-operative day but no significant effect on mean bile output from the fistula occurred. Bile drainage of about 700cc/day continued. Octreotide therapy was initiated, but due to the abdominal pain and gas bloating the patient felt, it was ended on the fourth day of therapy, and it had no decreasing effect on bile output during these 4 days. The patient had bilioptysis on the tenth post-operative day. Mezlocillin sodium (2x1g) and gentamycin (2x80mg) were started. His temperature was around 37°C. He was operated on on the 18th post-operative day. First right thoracotomy was done. It was seen that the cystic cavity was isolated from the pleural cavity and no bile or air leak was present in the pleural cavity. The cystic cavity and the lower part of the right lung were freed. Four bronchial radicals opening to the cystic cavity were sutured, and capitonnage was done. The defect in the diaphragm was closed. A drain was placed to the pleural cavity and connected to underwater drainage. Then, by right subcostal incision cholecystectomy+cholodochotomy+T-tube drainage were done. A drain below the diaphragm, in the cystic cavity, and another drain below the right lobe were placed and the incision was closed. On the third post-operative day, the drain below the liver, and on the fifth day, the thoracic drain were extracted. T-tube cholangiography on post-operative day 21, revealed two small biliary radicals opening to the cystic cavity **(Figure 4)**. The general condition of the patient was very good, and he had oral feeding. Ten mg/kg/day albendazole was started. He was sent home with the T-tube and the drain. In his weekly follow-ups, the mean bile output from the drain was about 400-500cc/day.

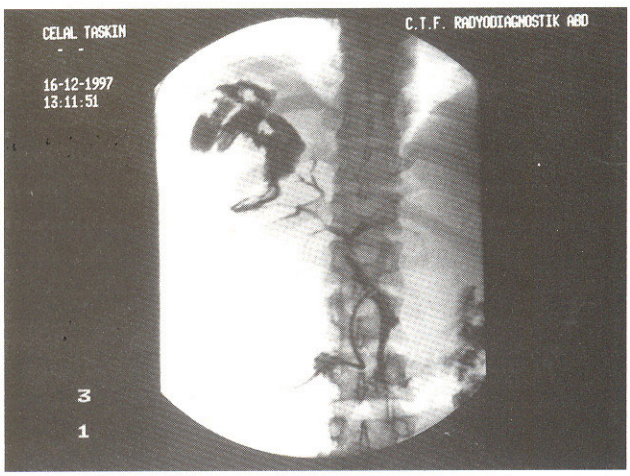


FIGURE 4 T-tube cholangiography revealing the leaking biliary radicals.

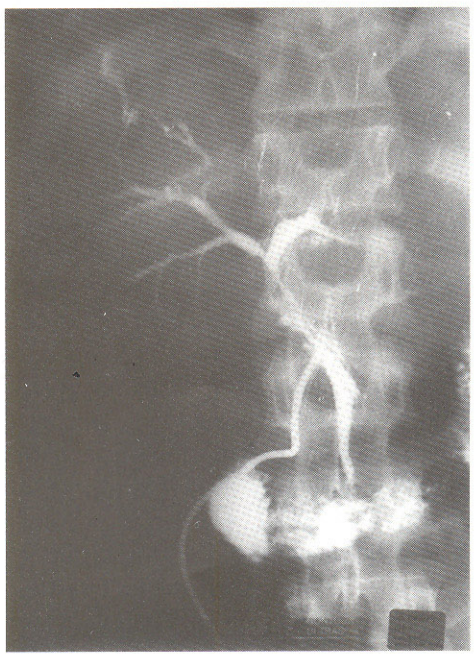


FIGURE 5 T-tube cholangiography 3 months post-operatively, demonstrating the narrowing of the distal right hepatic duct.

Three months post-operatively, T-tube cholangiography was done and a narrowing of the distal right hepatic duct was revealed **(Figure 5)**. It was thought that this narrowing was the cause of continuing bile fistula, in spite of the application of endoscopic sphincterotomy, octreotide therapy and total parenteral nutrition. There was minimal narrowing of the distal left hepatic duct, so bilateral dilatation was planned. In the radiology department, by way of a T-tube and with the help of a 4F Berenstein catheter (Bern Berenstein 4F Torque catheter, Medi-tech, Boston Scientific corporation, USA) **(Figure 6)**, road runner guides (Roadrunner wire guide slip coat hydrophilic coating, William Cook Europa, AIS, Denmark) were placed to the right and left hepatic ducts **(Figure 7)**. Then the Berenstein

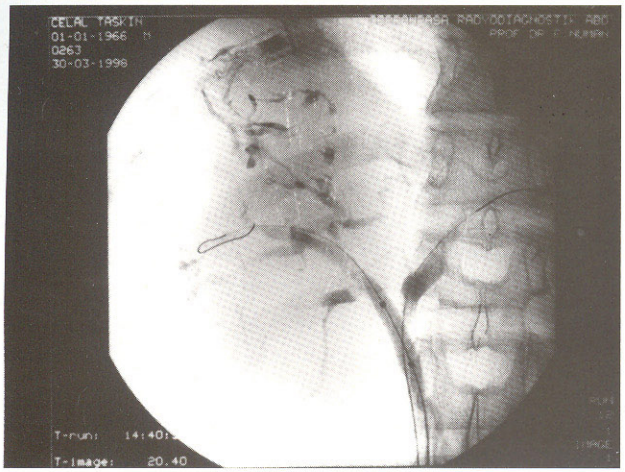


FIGURE 6 The Berenstein catheters by T-tube.

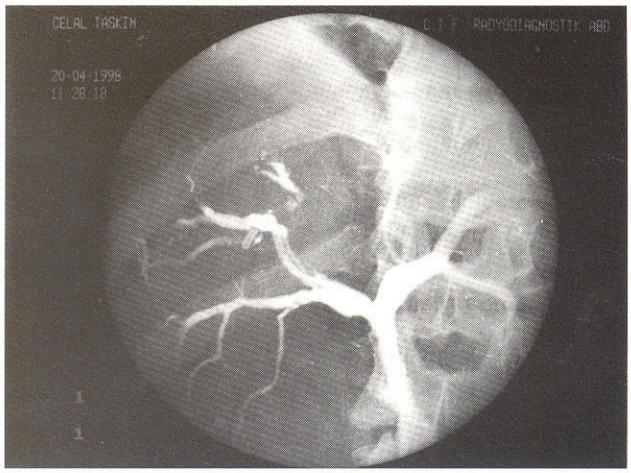


FIGURE 8 The drainage catheters after the dilatation.

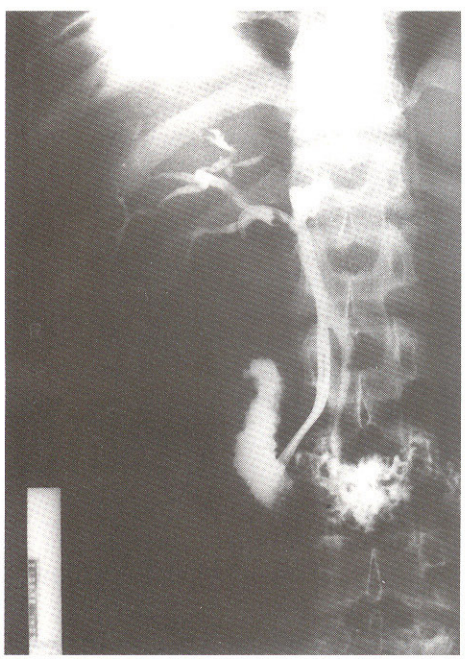


FIGURE 7 The Road runner guides were placed in the right and left hepatic ducts.

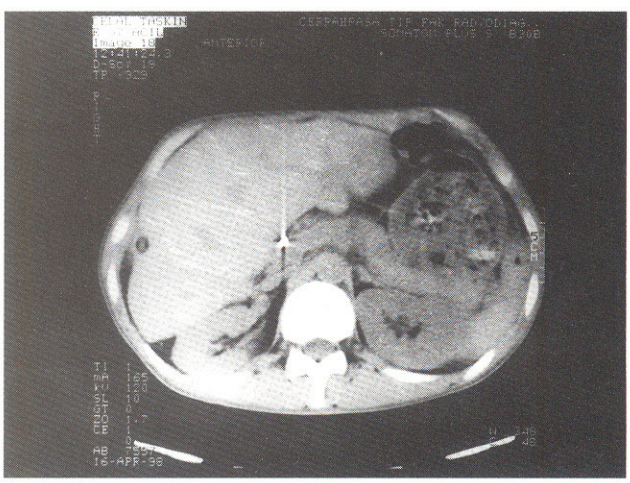


FIGURE 9 Follow-up CT scan of the patient.

catheters were taken out, and dilatation by 8mmx2cm dilatation catheters (8mmx2cm PTA angioplasty ultra-high pressure catheter, Nu Med Canada Inc., Canada) was applied. After the dilatation, the catheters were taken out, and 7F pigtail (Cordis Europa NV, The Netherlands) drainage catheters were placed **(Figure 8)**.

On follow-up 3 days later, the bile output from the drain in the cavity was about 50cc, and in the control cholangiography minimal narrowing of the distal right hepatic duct was still present. A second dilatation to the narrowing of the right hepatic duct was applied 15 days after the first dilatation. On follow-up 1 week later, there was no drainage and the catheter was clamped. On follow-up CT, no collection of fluid was present in the right subphrenic space; the cavity had collapsed **(Figure 9)**. The next day, the drainage catheter in the right hepatic duct and the drain in the cavity were extracted.

On follow-up on day 15 and 30 he had no complaints and had gained 3kg in weight.

DISCUSSION

More than two-thirds of hydatid cysts occur in the liver, and more than 75% of these are single cysts. Although rare, other intraabdominal organs where hydatid cysts are described are: the spleen, the pancreas, the ovaries and the pelvic cavities. Intraabdominal hydatidosis is caused either by extrusion from the cyst in the liver or rupture of a liver cyst. The commonest complication of liver hydatid cyst is cyst-biliary communication and intrabiliary rupture (1,2). As the cyst enlarges, it compresses and stretches bile ducts, and erosion occurs in these bile ducts producing fistulae between the hydatid cyst and

the biliary ductal system. If the fistulae can be seen or visualized pre-operatively, they can be sutured, and this is the best solution. If the bile leak to the cyst is developed, rupture occurs and some of the germinal layer or the scolices can enter the ductal system and may cause obstructive jaundice. Rarely, extrinsic compression of the main bile ducts in the liver, in the porta hepatis or in the pancreas is the cause of the jaundice.

Intraabdominal rupture may result in secondary implantations. A cyst below the diaphragm may erode through the diaphragm and rupture into the pleural cavity and a new cyst, or cysts, may develop. If this new cyst erodes and communicates with radicals of the bronchi, broncho-biliary fistulae develop.

Hydatid cysts of the liver are asymptomatic initially and may remain so until they reach a large size. Therefore, patients usually present with painful or painless hepatomegaly and abdominal mass.

Intraperitoneal rupture is a rare complication. Symptoms and signs simulate acute abdomen, but allergic or anaphylactic symptoms accompany or follow the acute abdominal symptoms and signs.

Sometimes the patients present with biliary colic, cholangitis and intermittent jaundice. Because the commonest sign is abdominal mass, the reliable imaging tests are ultrasound, CT and MR scans. For the differential diagnosis of single cysts with no daughter cyst, immunological tests are needed. The most commonly used and reliable immunological tests are: immuno-electrophoresis, indirect hemagglutination test, complement fixation test, and ELISA. The ELISA test is the most reliable, but the complement fixation test is used in post-operative follow-up.

Radiological investigation is useful in the determination of the precise location of the cyst(s) and in planning the incision of the operation. For example, for the cyst on the posterior upper surface of the right lobe of the liver, thoracoabdominal incision through the 9th or 10th intercostal space, or through the bed of the 11th rib is very useful. CT and MR are sensitive in the localization of the anatomical locations of intraabdominal cysts other than liver.

Systemic antihelmintic agents are not effective against human *Echinococcus*. Systemic drug therapy is still an adjuvant to surgery (3).

Surgery remains the main type of therapy for hydatid disease and indicated in all patients with symptomatic disease. The important steps of hydatid disease surgery are: protection of the abdominal cavity from spillage; removal of all living contents of the cyst; instillation of scolocidal agents to sterilize the pericyst lined cavity; closure of the open biliary radical(s) and obliteration of the cystic cavity. Many scolocidal agents are used for sterilization of the cyst. But it is not practiced in adult patients, in whom the cyst is often filled with daughter cysts and has a high pressure, before the cystic contents are removed.

For the aspiration and removal of the cystic contents and instillation of scolocidal agents, a disposable 10mm laparoscopy trocar can be used (4).

For the obliteration of the residual cavity, techniques such as partial pericystectomy with capitonnage, external drainage, omentoplasty, intraflexion or primary closure with saline instillation can be performed. Omentoplasty is especially useful in large cysts which cannot be obliterated with other techniques, or if the cyst walls are calcified or the surgeon cannot be sure about suturing all of the presenting bile ducts. If the patient has jaundice in his history, or presents with cholangitis and jaundice, biliary rupture of the cyst must be considered. In such patients, pre-operative cholangiography helps in identifying the leaking bile ducts in the cavity. After the operation, if the bile leakage continues to be a problem, sphincterotomy by ERCP should be done. If this is not available or is unsuccessful, T-tube drainage should be the method of choice to decompress the biliary tree. If bile leakage by the drain in the cavity continues to be a problem, fistulojejunostomy can be done. Although octreotide therapy is considered to be effective in decreasing bile output, it is not confirmed by everyone (5). For closure of the bile leaks, many non-operative techniques such as endoscopic sphincterotomy, percutaneous microcoil embolization, stenting, nasobiliary drainage and percutaneous drainage, are reported to be successful (6-10). Collaboration with the radiology department in these methods is important. Intrathoracic cysts are approached by thoracotomy and after removal of the cystic contents bronchial radicals are suture ligated and the cavity is obliterated. Although single cyst management is simple and effective,

disseminated hydatidosis possesses many problems in choosing the operative incision, post-operative patient management, hospitalization time and cost.

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