Cysto-cholecystostomy: A new technique for management of hilar biliary disruption due to a large hydatid cyst liver

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Abstract
Introduction
The disruption of a major bile duct due to intra-biliary rupture of a large hydatid cyst at liver hilum is a potentially lethal condition. An early diagnosis and appropriate surgical intervention is life-saving in these cases. An internal biliary drainage remains the only option since defect in the biliary track is usually large and primary repair is not feasible. Roux-en-Y cysto-jejunostomy is the standard recommended treatment in such cases. This report describes one such patient that was successfully treated by cysto-cholecystostomy, a procedure that has not been described earlier in the literature.

Case report
A 35 years old male was admitted with the diagnosis of hydatid cyst liver. CT scan of the abdomen revealed a large hydatid cyst in right lobe of liver near hilum that had ruptured into the biliary system. On exploration there was a 10 x 8 cm cystic mass in right lobe of liver at the hilum containing 400 ml thick bile stained inspissated hydatid material that was evacuated. On cleaning the cavity, bile was seen trickling from divided biliary openings due to destruction of intra-cystic part of right hepatic duct. Through a cholecdochootomy incision, a Kehr’s T-tube was placed into the common bile duct with its proximal limb reaching into the cyst cavity through divided right hepatic duct. The adjoining gallbladder was mobilized from the liver bed, a longitudinal incision was given on fundus of the gallbladder and side to side cysto-cholecystostomy was performed with interrupted 2-0 vicryl sutures. Post-operative period was uneventful. MR cholangiogram was done at six weeks that demonstrated patent cysto-cholecystostomy anastomosis with normal biliary radicles and shrunken cyst cavity. The T-tube was removed at six weeks.

Conclusion
The gallbladder can be used as a conduit for internal biliary drainage in cases of large hydatid cyst of liver lying at hilum with intra-biliary rupture by performing cysto-cholecystostomy. It is a simple, safe, quick, physiological and less technically demanding procedure in comparison to Roux-en-Y cysto-jejunostomy.

Introduction
Hydatid disease is caused by Echinococcus granulosus and it usually affects liver (60-70%) and lungs (20-30%).1 Although it remains asymptomatic in most of the cases, the symptoms may appear due to mechanical effects, toxic reactions or presence of complications. 2 One of the most common and potentially fatal complications is rupture of the cyst into biliary tract that presents as biliary colic, jaundice and cholangitis.3 Intrabiliary rupture is reported to be seen in a range of 1-17% in such cases.4 Cyst diameter of 7.5 cm and above is an independent factor that is associated with a high risk of biliary-cyst communication.5 It should be diagnosed early and managed promptly since it carries high mortality. The principle of treatment is killing and evacuation of parasite, management of residual cavity and restoration of internal biliary drainage. In cases of major biliary disruption, it is usually achieved with Roux-en-Y cystojejunostomy.3,6 We describe a new and simple technique of cysto-cholecystostomy performed successfully in one such case of large intrabiliary rupture of hepatic hydatid cyst in to the right hepatic duct at liver hilum.

Case report
A 35-year-old male presented one year ago with painless progressive lump in the right hypochondrium that was diagnosed as hydatid cyst liver. He was advised surgery that he refused and did not report back. He started developing dull aching pain abdomen, jaundice, hepatomegaly and melena two months ago and presented in emergency in a state of hypovolumic shock with tense and tender abdomen. He was resuscitated with intravenous fluids, antibiotics and blood transfusions. Abdominal sonography revealed a large multiculated cystic mass in the right lobe of liver occupying segments V, VI and VIII along with ipsilateral right intrahepatic bile duct dilatation. Laboratory tests revealed an elevated leukocyte count (18,000/cmm), polymorphonuclear leucocytosis (92%), direct hyperbilirubinemia (3 mg/dl) and elevated serum alkaline phosphatase (343IU/L). Serum antibody screening was positive for antiechinococcal IgG antibodies. Non-contrast CT scan of the abdomen revealed hydatid cyst in the right lobe of liver near hilum that had ruptured into the biliary system. His jaundice subsided spontaneously and his condition improved with conservative treatment over a period of one week. He was discharged on tablet albendazole (10 mg/kg/day in two divided doses) and readmitted after one month for surgery. On MRCP performed at the time of surgery, he was found to have 8 x 8 cm hydatid cyst at

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Figure 1: Preoperative MRCP showing ruptured hydatid cyst into the right hepatic duct. Right hepatic duct and its branches are displaced and dilated whereas left hepatic duct is normal.

Figure 2A: End of T tube (arrow) seen projecting into de-roofed cyst cavity through common bile duct. GB-Gallbladder.

Figure 2B: Gallbladder mobilized and opened longitudinally (arrow) against cyst cavity.

porta hepatis with intra-biliary rupture into the right hepatic duct. The lesion was pushing the caudate lobe medially. The biliary ducts on right side were dilated and displaced by the cyst. Left bile duct and common bile duct were normal (Figure 1). The patient was taken up for elective surgery.

On exploration there was a 10 x 8 cm crumpled cystic mass in right lobe of liver at the hilum. After packing the adjoining bowel with hypertonic saline soaked sponges, de-roofing of the cyst was done and 400 ml thick bile stained inspissated hydatid material was evacuated from the cyst cavity. The cavity was thoroughly irrigated with saline. On cleaning the cavity, bile was seen trickling from multiple divided biliary openings draining into the cyst cavity. Through a choledochotomy incision in common bile duct, a 6 F catheter was advanced retrogradely. It entered the cyst cavity through stretched and divided distal end of right hepatic duct. Due to expansion of the cyst wall at liver hilum, 2 cm length of intra-cystic part of right hepatic duct was lost. A Kehr’s T-tube was then placed through choledochotomy into the common bile duct with its proximal limb reaching into the cyst cavity (Figure 2A).

The gallbladder was normal and distended with bile and was lying close to the wide opening of the cyst cavity. The fundus and body of the gallbladder were mobilized from the liver bed and it was further approximated with the cyst cavity. A longitudinal incision was given on fundus of the gallbladder and side to side cysto-cholecystostomy was performed with interrupted 2-0 vicryl sutures (Figure 2B, C, D and Figure 3A, B). The wound was closed after placing a closed drainage tube in the subhepatic space. Initially bile and then bile stained fluid drained through the subhepatic drain that gradually decreased in amount and the drain was removed on eighth postoperative day. T-tube was clamped intermittently on 15th post-operative day without any complications. Patient was discharged on tablet albendazole with clamped T-tube in situ.

MR cholangiogram was done at six weeks that demonstrated patent cysto-cholecystostomy anastomosis with normal biliary radicles and shrunken cyst cavity (Figure 4A). The T-tube was removed at six weeks and tablet albendazole was continued for six cycles of four weeks each at two weeks interval. The patient was kept on regular follow up. At one and half years after surgery, the patient was asymptomatic. There was no icterus and abdominal examination was also normal. Laboratory findings revealed normal liver function tests. Abdominal ultrasonography was normal. MRCP showed almost resolved cyst cavity that was draining well into the gallbladder through cysto-cholecystostomy and biliary radicles were also noldilated (Figure 4B). This work conforms to the values laid down in the Declaration of Helsinki (1964). The protocol of this study has been approved by the relevant ethical committee related to our institution in which it was performed. All subjects gave full informed consent to participate in this study.

Discussion

Treatment of large intrabiliary rupture of hydatid cyst liver has remained controversial. The main aims for
management of such a case include evacuation of cyst contents, management of residual cavity, clearance of cystic material from biliary cavity and restoration of bile flow.7,8,9

Some authors have performed radical procedures such as selective hepatic lobectomy, total cystectomy or partial cystectomy followed by Roux-en-Y pericysto-jejunostomy.7 On the contrary, others have suggested conservative procedures such as suture of cysto-biliary fistula, double side drainage and cysto-biliary disconnection.10 In patients with intra-biliary rupture of hydatid cyst, common bile duct exploration should be done using intraoperative cholangiography and cholangioscopy. After clearance of hydatid debris from the biliary tree, various procedures described for biliary drainage are Kehr’s T-tube drainage, sphincteroplasty and choledochoduodenostomy.7,8,9 The main complication following these procedures is external biliary fistula.11,12 Endoscopic sphincterotomy has proved to be an effective technique for treating persistent extended external biliary fistulae.13

In case of biliary disruption at liver hilum involving major bile duct, chances of developing external biliary fistula are very high. The standard method of treatment in such cases is creating internal biliary drainage with Roux-en-Y cysto-jejunostomy.14 The aim of creating internal biliary drainage is to allow disrupted bile duct to heal and to avoid occurrence of biliary fistula.

The present case had intra-biliary rupture of right hepatic duct and its primary suture repair was not possible due to the large size of fistula (2cm). However we performed cysto-cholecystostomy instead of Roux-en-Y cysto-jejunostomy for internal biliary drainage. We observed that in comparison to Roux-en-Y cysto-jejunostomy, cysto-cholecystostomy was a simple, technically less demanding and short procedure requiring single anastomosis. It can be easily performed in a ruptured cyst near hilum due to close proximity of the gallbladder to the cyst. The gallbladder is a stretchable organ due to its muscular wall and can be easily mobilized from the liver bed by fundus first method. Following mobilization, it hangs like a berry on its pedicle and can be easily rotated anywhere on liver hilum. Then tension free side to side anastomosis can be safely and quickly performed between de-roofed cyst and gallbladder as was done in the present case. Majority of the cases with intra-biliary rupture are reported to have fibrotic cyst wall.7 Hence the wall will effectively hold the sutures leading to secure anastomosis for internal biliary drainage. Moreover, it is more physiological since function

Figure 2C: Gallbladder sutured with the cyst wall using 2-0 interrupted vicryl suture (arrow).

Figure 3: A-Line diagram showing ruptured hydatid cyst into the right hepatic duct.
B-Line diagram showing mobilized gallbladder with cysto-cholecystostomy and T-tube in situ.

of sphincter of Oddi is preserved thus reducing the risk of reflux cholangitis. Following cysto-cholecystostomy, it is unlikely that stone formation and cholecystitis will develop in the gallbladder since it is no longer functioning as a conduit and bile is flowing freely across its lumen whereas cholelithiasis occurs in a stagnant pool of bile. Apparently there is no disadvantage of performing cysto-cholecystostomy, if possible, in such a situation. Following internal biliary drainage, aim of keeping T-tube in the CBD was to ensure free flow of bile and to drain any residual debris so that cysto-cholecystic anastomosis remained decompressed. MR cholangiogram performed at six weeks confirmed sound anastomotic healing and thereafter, the T-tube was removed. However, there remained a concern regarding efficacy of the anastomosis and bile drainage through cystic duct in long term period since the procedure was novice. But the patient remained asymptomatic during follow up and MRCP performed at 1.5 years demonstrated well-functioning cysto-cholecystostomy anastomosis with resolved cyst cavity (Figure 4B).

In case of Roux-en Y cysto-jejunostomy done for ruptured hydatid cyst, cholecystectomy is usually performed to create space for making tension free anastomosis. However, nobody in the past has ever attempted to use gallbladder for creating anastomosis with the de-roofed cyst wall. Nonetheless it seems logical to use the gallbladder as a conduit for internal biliary drainage since it is a natural reservoir for bile. However, the contraindications for this operative procedure could be: a) concomitant cholecystitis with cholelithiasis, b) non-patent cystic duct, c) ruptured cyst cavity located too far away from the hilum where anastomosis with gallbladder is not possible. Hence it is proposed that an attempt should be made to utilize the gallbladder for such anastomosis and in case it fails, cholecystectomy followed by roux-en-Y cysto-jejunostomy always remains a possible option.

**Conclusion**

Cysto-cholecystostomy is a simple, safe, quick, more physiological and less technically demanding procedure in comparison to Roux-en-Y cysto-jejunostomy for internal biliary drainage in large biliary disruption.

**References**


