



Management of liver hydatid cyst with cystobiliary communication and acute cholangitis: a 27-year experience

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Abstract

Purpose The rupture of the hydatid cyst into the bile ducts is a common and serious complication. The rupture can be latent or revealed after a complication such as acute cholangitis. The objective of this study was to discuss the clinical features, radiographic findings, and surgical treatment of this rare complication.

Methods A retrospective analysis of patients operated for acute cholangitis caused by hydatid cyst between January 1990 and May 2016 was conducted.

Results Our study included 55 cases of acute cholangitis caused by ruptured hydatid cyst: 35 were men and 20 were women. Five patients had medical history of hydatid cyst. On imaging, all patients had a dilated bile duct. In 51 cases, there was a single hydatid cyst. Hydatid cysts with fistula were located in the right lobe of the liver in 49 cases and in the left lobe in 6 cases. Seven patients underwent an emergency surgery after admission, while others were operated after 3–8 days. A conservative procedure associated with drainage was applied in 49 cases. Endoscopic treatment was performed in four cases which failed in two cases. Twenty-eight cases developed postoperative complications, among which the most common complication was biliary fistula ($n = 17$). There were eight cases of postoperative deaths (14.5%).

Conclusions Liver hydatid cyst with cystobiliary communication and superadded acute cholangitis is a serious clinical problem requiring the early diagnosis and surgery in the absence of endoscopic therapy.

Keywords Liver · Echinococcosis · Biliary fistula · Cholangitis

Introduction

Liver hydatid cyst is a cosmopolitan disease. It is very frequent in endemic countries. It presents a serious public health problem in our country. Although it is a mild disease, some of its complications are serious and lethal. Fistula of the cyst in the bile ducts is very formidable [1, 2]. Acute cholangitis is an uncommon but serious complication of liver hydatid cyst. Most of these cases are managed by the initial endoscopic therapy to achieve biliary clearance

followed by surgery for hydatid cyst [3–5]. However, the success rate of endoscopic treatment varies from 80 to 90% [6, 7]. In addition, little is known about the outcomes of emergency surgical treatment in such patients in the absence or failure of endoscopic therapy. This study was conducted to analyze the short-term outcomes of primary surgical treatment for liver hydatid cyst with cystobiliary communication and acute cholangitis.

Patients and methods

In this retrospective study, we retrieved the data of all the cases of abdominal hydatid cyst with acute cholangitis who underwent surgery in our Department of digestive surgery at Fattouma Bourguiba Hospital between January 1990 and May 2016. We selected the patients who were diagnosed having an acute cholangitis. We excluded patients with acute cholangitis due to other causes, cases of hydatid cyst without

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acute cholangitis and cases with incomplete records. The data collected included age, gender, clinical findings, laboratory data, radiological findings, endoscopic treatment, surgical treatment, mortality, morbidity, and recurrence, and the diagnosis of hydatid cyst was based on intraoperative findings. This study was approved by the Institutional Ethics Committee and has, therefore, been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Hydatid cyst were classified in to subtypes based on Gharbi classification as: Type I (univesicular cyst), Type II (cyst with detached membranes), Type III (multivesicular cyst with daughter cysts), Type IV (cyst with heterogenous matrix), and Type V (reflecting thick walls) [8].

Medical therapy

To prevent recurrences, albendazole therapy was started preoperatively (10 mg/kg/day) and given for mean duration of 9 months.

Endoscopic treatment

In selected cases, we used endoscopic treatment with sphincterotomy, to overcome the critical period especially in those patients with high risk of anesthesia provided that endoscopic therapy is available.

Surgical technique

The peritoneal cavity was liberally protected with scolicalid agents. The cyst contents were aspirated. We used 20% or 30% saline solution as the scolicalid agent for the cyst. The biliary fistula was identified through careful exploration of the remaining cystic cavity, if necessary by the blue methylene test. Intraoperative cholangiography was performed in all patients. Choledochotomy was performed and thoroughly cleared from hydatid contents and/or stones. Complete clearance was confirmed by instrumentation, intraoperative cholangiography, or choledochoscopy.

The cystobiliary communication was dealt according to the size of the fistulae as follows:

A. Radical method

We conducted total pericystectomy associated with clearance of CBD (common bile duct) and drainage with Kehr tube.

B. Conservative method

– For small fistulas (less than 5 mm), we realized a partial pericystectomy and simple suturing of the fistula com-

bined with drainage, omentoplasty of the residual cavity, and drainage of the CBD with Kehr drain.

– For large fistulas (greater than 5 mm), we used:

- DITFO trans-fistula Oddien drainage
- Biliary kysto-disconnection according to Perdromo technique by drainage of the bile duct and catheterization fistula

Statistical analysis

The quantitative variables were expressed as mean with standard deviation and the qualitative variables were defined by frequency (%).

Results

Patient characteristics

Fifty-five patients were included during the study period, 35 men and 20 women with the mean of 42 years (range 15–74 years), and the standard deviation of 13.41 years. Five patients had past history of hydatid cyst disease. All patients presented with right upper quadrant abdominal pain, fever, and cholestasis, 50% with jaundice, and hydatid serology was positive in 30 of 47 tested patients. Acute renal failure was diagnosed in 7 patients.

Radiological findings

Abdominal ultrasound, computed tomography (CT), magnetic resonance imaging (MRI) were performed in 55, 30, and 2 patients, respectively.

In 48 cases, the cyst was single, and in 7 cases, there were multiple cysts (two cysts in 3 cases, three cysts in 2 cases, and four cysts in 2 cases). In 4 cases, an associated mesenteric hydatid cyst was found. Hydatid cysts were located in the right lobe of the liver in 49 (89%) cases and in the left lobe in 6 (11%) cases. The hydatid cysts of the right lobe were located in the segment 1 ($n=2$), segment 4 ($n=19$), segment 5 ($n=15$), segment 6 ($n=3$), segment 7 ($n=3$), and segment 8 ($n=7$). Among the left lobe cysts, 2 hydatid cysts were located in segment 2 and 4 cysts in segment 3. There were 32 cases of Gharbi type III and 23 cases of Gharbi type IV hydatid cyst. The mean diameter of hydatid cyst was 13.5 cm (range 7–16 cm). Dilatation of the common bile duct (CBD) was present in all cases. Defect in the cyst wall near the dilated duct was seen in 22 cases (Figs. 1, 2). Intraluminal echogenic or hyper-dense content within CBD was visible in 52 cases. Incidental gallstones were detected in 4 patients. None of the patients had pulmonary hydatid cyst.

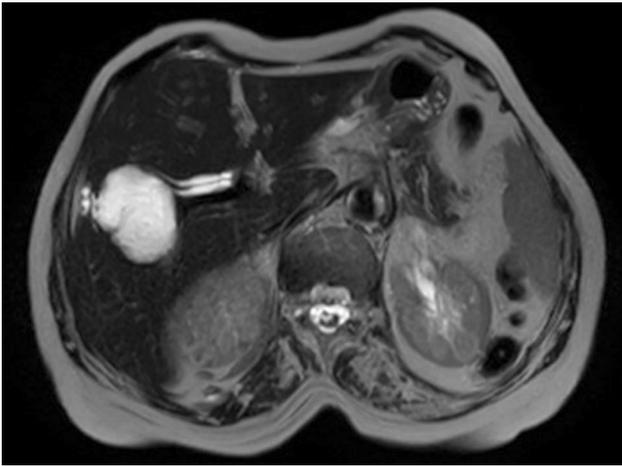


Fig. 1 Hepatic MR (axial T2 sequence) showing hydatid cyst located in segment 6 and communicating with the bile duct

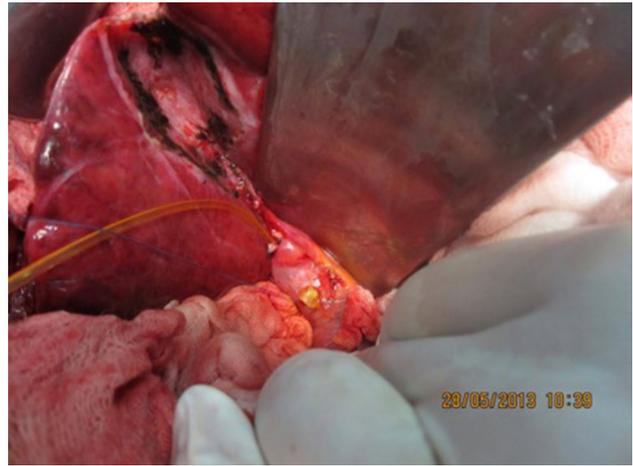


Fig. 3 Intraoperative photograph showing hydatid membrane being removed from the common bile duct through the choledochotomy

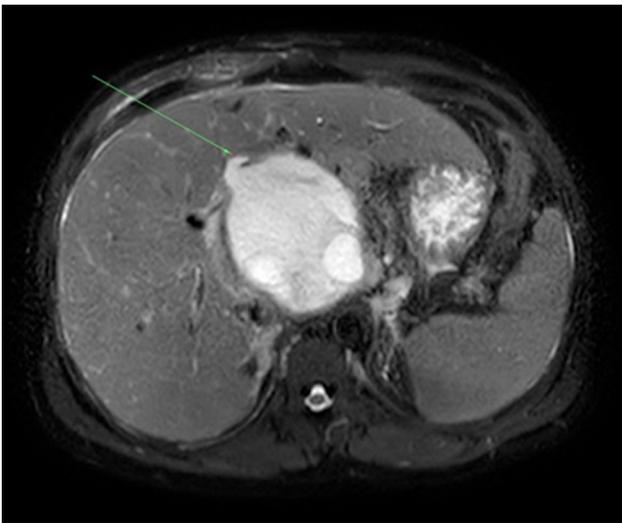


Fig. 2 Hepatic MRI (axial T2 sequence) showing large multivesicular hydatid cyst located in segment 1 and ruptured in the main bile duct

Management

Seven patients underwent emergency surgery after resuscitation because of the presence of septic shock. The others were operated a few days later (3–8 days), because they had a good response to antibiotics and resuscitation. In four cases with high anesthesia risk, we used endoscopic sphincterotomy. Endoscopic procedure failed in two cases and patients were operated in the emergency setting, whereas, in remaining two cases, surgery for the hydatid cyst was performed 1–2 months later.

Total pericystectomy was performed for the mesenteric cysts in four cases.

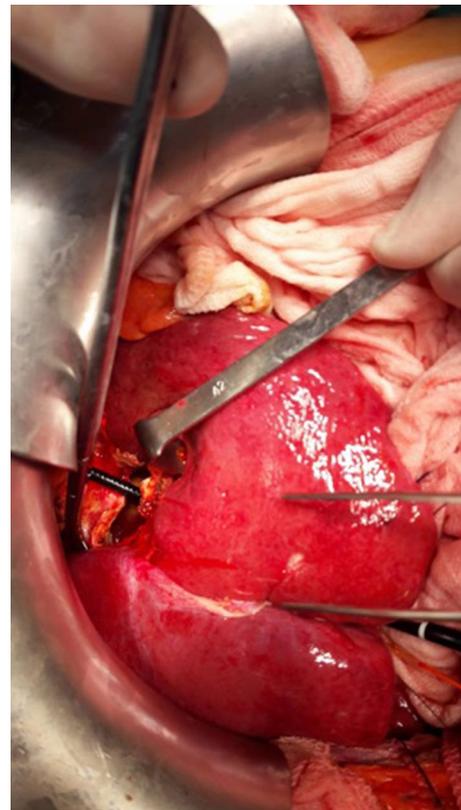


Fig. 4 Intraoperative photograph showing the choledochoscope protruding into the cyst cavity after being introduced in the bile duct through the choledochotomy

All patients underwent CBD exploration with the removal of the hydatid membranes from the bile duct and per operative cholangiography (Figs. 3, 4). We used the radical method with total pericystectomy and CBD

drainage for superficial located liver hydatid cyst in four cases.

We realized the conservative method in 49 patients (47 plus 2 cases of failed endoscopic treatment). We performed partial pericystectomy and suturing of the fistula after perifistular laminectomy that makes the area more pliable for suturing especially when the pericyst is tough, and omentoplasty for the residual cavity in 35 cases of small fistula. For large fistulas (greater than 5 mm), we used DITFO (transfistula Oddien drainage) in 12 cases and biliary kysto-disconnection using Perdromo's technique in 2 cases.

Outcomes

Twenty-eight (50.9%) patients developed postoperative complications (Table 1). The most common complication was biliary fistula which was observed in 17 (60.7%) cases. Nine of these fistulae resolved spontaneously in 3 weeks. Four patients developed pulmonary infection, three patients had pleural effusion, three patients had cavity abscess treated with percutaneous drainage, and one patient developed pulmonary embolism. In this study, the mortality rate was 14.5% (8/55). The causes of death were septic shock ($n=4$), myocardial infarction ($n=3$), and respiratory failure secondary to pulmonary infection ($n=1$). The mean duration of hospitalization was 20 days with the standard deviation of 4.53 days (range 10–60 days).

Discussion

Hydatid cyst is a parasitic disease mainly caused by *Echinococcus granulosus* and less frequently by *Echinococcus multilocularis* [1, 2]. It is common in the Mediterranean, African, and Asian countries. Tunisia is an endemic for liver hydatid cyst. The most common complication of hepatic hydatid cyst is intrabiliary rupture [9]. The incidence of rupture in biliary tree varies from 2 to 42% [9]. The cystobiliary communication has been postulated to occur due to compression of the bile duct by the enlarging hydatid cyst

causing necrosis and rupture. Second hypothesis states that the trapping of small biliary radicals in the pericystic wall leads to high intracystic pressure causing atrophy and the rupture of biliary ducts [3, 10].

Large communication (greater than 5 mm) also called as frank intrabiliary rupture which occurs in 3–17% cases [11]. In this variety, the content of the cyst (hydatid membrane and daughter cyst) passes into the bile duct in 65% of patients. It can be present clinically as obstruction of the biliary system (57–100%) causing jaundice, cholangitis, cholecystitis, or acute pancreatitis [9, 12]. In case of intrabiliary rupture smaller than 5 mm, it is exceptional to find hydatid material in the bile duct. This complication is asymptomatic in 10–37% of cases [11, 13]. Ultrasound and CT are the useful imaging methods for the diagnosis of communication between the cyst and the biliary tree [9, 13]. Ultrasound findings suggestive of intrabiliary rupture are the presence of liver cyst associated with adjacent dilated bile duct and echogenic material in extrahepatic duct without posterior acoustic shadowing. Other radiological signs suggestive of frank intrabiliary rupture are defect in the wall of the cyst near the dilated duct and dilated CBD with low attenuation intraluminal material [9, 11, 14]. MRI may be useful for diagnosing intrabiliary rupture, when CT scan and ultrasound results are inconclusive [4]. In our study, cyst rupture into the biliary tract was detected in 52 cases.

Endoscopic retrograde cholangiography (ERC) is useful for confirming the diagnosis of cystobiliary communication. However, ERC is most commonly used in cases of obstructive jaundice or acute cholangitis to relieve the bile duct obstruction. On ERC, daughter cysts may be seen in the duodenum or the ampulla of Vater or obstructing any part of the biliary tree [4, 5, 9, 11]. The overall success rate is 70–86% with rate of fistula closure of 81% in 1–2 days. At our center, facility for ERC is not available. The four patients who had high anesthesia risk were sent to another hospital for ERC. Among these two patients had a significant improvement in the clinical condition and liver function tests after ERC.

Surgery is the treatment of choice for liver hydatid cyst [9]. The goal of surgical treatment is evacuation of the cystic cavity, cleaning of the bile ducts, and restoration of the bile flow [9]. Several techniques are used for detecting cystobiliary communication. Most commonly used procedures are intraoperative cholangiography and use of methylene blue [14, 15].

After identification of the biliary fistula different methods can be used to close it. The indication depends on the characteristics of the fistula (size, location), proximity to the vessels, involvement of the hilar confluence, and surgeon experience. If the fistula is less than 5 mm, it can be treated by suturing it on the healthy tissue after removal of the capsule of hydatid cyst along with drainage. However, the main disadvantage of this technique is the risk of prolonged

Table 1 Outcomes of the various surgical therapies for cystobiliary communication after partial pericystectomy

Treatment	Number of patients	Morbidity	Mortality
Simple suturing of the biliary opening	33	18	1
Bipolar drainage of CBD	12	2	1
Disconnection of cysto-biliary communication according (Perdromo)	2	0	0

biliary leakage and subphrenic abscess formation. Other techniques in conservative approach include trans-fistular internal drainage. This option offers a lower rate of postoperative biliary fistula, especially for cyst less than 5 cm and cystobiliary communication more than 5 mm as seen in this study [16].

Total cystectomy with resection is a difficult procedure, especially when there is cholangitis, so it is recommended only in peripheral cysts. Liver resection is rarely used in the literature. The advantage of radical techniques, despite their complexity and applicability, is that these definitely treat both cyst and communication. Radical techniques have shown to decrease postoperative morbidity if closing the cysto-biliary communication in healthy tissue [9, 17]. Postoperative mortality in frank intrabiliary rupture patients ranges from 1.25 to 7%, and is mainly caused by sepsis or liver failure. Postoperative morbidity is between 16.7 and 55%, higher than observed in patients operated for liver hydatid cyst without frank intrabiliary rupture. The complications described are biliary fistula (5–27%), wound infection, pulmonary complications, bilioma, bleeding, and abscess formation [9].

This study is one of the rare studies in Tunisia reporting an experience of almost 3 decades. In addition, there are some limitations. First, the study was retrospective in nature. Second, the surgeries in this study were not performed by single surgeon. Hence, surgeons' experiences are likely to alter the outcomes. Third, there was limited use of endoscopic treatment due to lack of availability. However, if available, ERCP should be the first choice to deal with acute cholangitis in these situations.

In conclusion, liver hydatid cyst with cystobiliary communication and superadded acute cholangitis is a serious clinical problem requiring the early diagnosis and surgery in the absence of endoscopic therapy.

Compliance with ethical standards

Conflict of interest Omar Toumi, Housseem Ammar, Rahul Gupta, Sadok Ben Jabra, Badii Hamida, Faouzi Noomen, Khadija Zouari, and Mondher Golli have no conflict of interest to declare.

References

- Subramanyam BR, Balthazar EJ, Naidich DP. Ruptured hydatid cyst with biliary obstruction: diagnosis by sonography and computed tomography. *Gastrointest Radiol.* 1983;8:341–3.
- Sparchez Z, Osian G, Onica A, Barbanta C, Tantau M, Pascu O. Ruptured hydatid cyst of the liver with biliary obstruction: presentation of a case and review of the literature. *Roman J Gastroenterol.* 2004;13:245–50.
- Manouras A, Genetzakis M, Antonakis PT, et al. Endoscopic management of a relapsing hepatic hydatid cyst with intrabiliary rupture: a case report and review of the literature. *Can J Gastroenterol.* 2007;21:249–53.
- Özaslan E, Bayraktar Y. Endoscopic therapy in the management of hepatobiliary hydatid disease. *J Clin Gastroenterol.* 2002;35:160–74.
- Bektas M, Dokmeci A, Cinar K, et al. Endoscopic management of biliary parasitic diseases. *Dig Dis Sci.* 2010;55:1472–8.
- Rodriguez AN, Sanchez del Rio AL, Alguacil LV, et al. Effectiveness of endoscopic sphincterotomy in complicated hepatic hydatid disease. *Gastrointest Endosc.* 1998;48:593–7.
- Hilmioglu F, Karıncaoglu M, Yilmaz S, et al. Complete treatment of ruptured hepatic cyst into biliary tree by ERCP. *Dig Dis Sci.* 2001;46:463–7.
- Gharbi HA, Hassine W, Brauner MW, Dupuch K. Ultrasound examination of the hydatid liver. *Radiology.* 1981;139:459–63.
- Ramia JM, Figueras J, De la Plaza R, García-Parreño J. Cystobiliary communication in liver hydatidosis. *Langenbeck's Arch Surg.* 2012;397:881–7.
- El Malki HO, El Mejdoubi Y, Souadka A, et al. Predictive model of biliocystic communication in liver hydatid cysts using classification and regression tree analysis. *BMC Surg.* 2010;10:1.
- Michalopoulos N, Laskou S, Papavramidis TS, et al. Rupture of right hepatic duct into hydatid cyst. *J Korean Med Sci.* 2012;27:953–6.
- Wani NA, Kosar T, Gojwari T, et al. Intrabiliary rupture of hepatic hydatid cyst: multidetector-row CT demonstration. *Abdom Imaging.* 2011;36:433–7.
- Avcu S, Ünal Ö, Arslan H. Intrabiliary rupture of liver hydatid cyst: a case report and review of the literature. *Cases J.* 2009;2:1.
- Pedrosa I, Saíz A, Arrazola J, Ferreirós J, Pedrosa CS. Hydatid disease: radiologic and pathologic features and complications. *Radiographics.* 2000;20:795–817.
- Kilic M, Yoldas O, Koc M, et al. Can biliary-cyst communication be predicted before surgery for hepatic hydatid disease: does size matter? *Am J Surg.* 2008;196:732–5.
- Sakhri J, Ben Ali A. Hydatid cyst of the liver. *J de Chirurgie.* 2004;141:381–9.
- Zaouche AHK, Jouini M, El Hachaichi A, et al. Management of liver hydatid cysts with a large biliocystic fistula: multicenter retrospective study. *World J Surg.* 2001;25:28–39.