

# Intrabiliary rupture: An algorithm in the treatment of controversial complication of hepatic hydatidosis

Kenan Erzurumlu, Adem Dervisoglu, Cafer Polat, Gokhan Senyurek, Ibrahim Yetim, Murat Hokelek

Kenan Erzurumlu, Adem Dervisoglu, Cafer Polat, Gokhan Senyurek, Ibrahim Yetim, Murat Hokelek, Department of Surgery, Medical School, Ondokuz Mayıs University, Kurupelit, Samsun, Turkey

Correspondence to: Kenan Erzurumlu, MD, Professor, Department of Surgery, Medical School, Ondokuz Mayıs University, 55139 Kurupelit, Samsun, Turkey. kerzurum@omu.edu.tr  
Telephone: +90-362-4576000-2470 Fax: +90-362-4576029  
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## INTRODUCTION

Intrabiliary rupture (IBR) is the most common and serious complication of hepatic hydatid cyst (HHC). It has also been reported as 'cystobiliary fistula' or 'cystobiliary communication' in the literature. The incidence varies from 1% to 25% although an incidence of 64.75% has been reported from a multicentric study in Tunisia<sup>[1-12]</sup>.

Obstructive jaundice, fever, right-upper quadrant pain, nausea and vomiting, flatulence, palpable hepatic mass are the most commonly encountered symptoms in intrabiliary rupture<sup>[2,3,9]</sup>.

Although there is some consensus on the medical and surgical treatment of hydatid cyst diseases, the treatment of intrabiliary rupture still remains controversial. Intracystic suturing of the orifice, T-tube drainage, double side drainage, cystobiliary disconnection, choledochoduodenostomy, choledochojejunostomy, endoscopic sphincterotomy and nasobiliary stent application have been used.

In this report, eight cases of intrabiliary rupture were presented and discussed in the light of literature review.

## MATERIALS AND METHODS

In this study, eight cases of hepatic hydatid cyst with cystobiliary fistulae treated by our team between 1994 and 2003 were presented. They began to undergo intra-operative ultrasonography (US) in 1997.

All patients' records were reviewed and evaluated as to the age, sex, clinical findings, cyst number and stages according to Gharbi's classification, abdominal ultrasonography and CT-scan results, surgical methods, complications, results and coincidental diseases.

## RESULTS

### Patients' data

Between 1994 and 2003, a total of 70 cases of hepatic hydatidosis were treated surgically by our team, of these cases eight (11.42%) had cystobiliary fistulae, diagnosed either pre-, intra- or post-operatively. Female/male ratio was 1/7. Mean age was 52.12±18.26 years (range 24-69 years).

Right upper quadrant pain, flatulence, palpable hepatic mass were the symptoms present in all patients. Cholestatic jaundice was found in four cases.

Four patients had one, two patients had two and the

## Abstract

**AIM:** Intrabiliary rupture (IBR) is a common and serious complication of hepatic hydatid cyst. The incidence varies from 1% to 25%. The treatment of IBR is still controversial. We aimed to design an algorithm for the treatment of hepatic hydatidosis with IBR by reviewing our cases.

**METHODS:** Eight cases of IBR were analyzed retrospectively. Patients were evaluated according to age, sex, clinical findings, cyst number and stage, abdominal ultrasonography and CT-scan, surgical methods, complications, results and coincidental diseases.

**RESULTS:** Female/male ratio was 1/7. Mean age was 52.12±18.26 years (range 24-69 years). Right upper quadrant pain, flatulence, palpable hepatic mass were symptoms common in all patients. Cholestatic jaundice was found in four cases. In all patients, cyst evacuation and omento-plasty were performed, followed by either choledochoduodenostomy, T-tube drainage, intracavitary suturing of the orifice, two cases in each. Whereas in two patients diagnosed post-operatively percutaneous drainage of biliary collection or ERCP and sphincteroplasty were added. Morbidity and hospital stay were higher in these cases.

**CONCLUSION:** When the diagnosis of IBR can be done pre- or intra-operatively, morbidity decreases. If a biliary fistula is seen post-operatively, endoscopic procedures such as ERCP, sphincteroplasty or nasobiliary drainage can be applied.

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**Key words:** Intrabiliary rupture; Hepatic hydatidcyst

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**Table 1** Patients' characteristics (*n* = 8)

Mean age	52.12±18.26
Female/male	1/7
Symptoms	
Right upper quadrant pain, palpable hepatic mass, flatulence	8
Cholestatic jaundice	4
Number of cysts	
1	4
2	2
3 or more	2
Stages of cysts (According to Charbi's classification)	
Stage II	3
Stage III	5
Choledochal image at US and CT-scan	
dilated (= 2 cm)	3
Normal	5

other two patients had three or more cysts. The majority of the cysts were in stage III (62.5 %) and stage II (37.5 %) (Table 1). Seven cases had their cysts in the right hepatic lobe, one in the left lobe. The cyst sizes were in the range of 2.5-15 cm. Table 1 summarizes the patients' data.

Four cases had obstructive jaundice. Total bilirubin was as high as 7 mg/dL. ALT, AST and ALP levels were thrice the normal levels. Four patients had no elevation at biochemical analysis (Table 2).

US and CT-scan showed choledochal dilatation of 2 cm in three patients. Biochemical tests of these cases were high.

### Treatments of patients

Benzimidazole treatment was started 7-30 d before surgery (mean 14±8.45 d) and continued for 2-5 mo (mean 3.71±1.11 mo) after surgery, in a monthly cyclic protocol.

All cysts were treated by evacuation+omentoplasty+ drainage of the cystic cavity. The cystic cavities were disinfected by 1.7 mg/mL albendazole solution as described previously<sup>13-15</sup>. When choledochotomy was done, all contents in the common bile duct (CBD) were evacuated and biliary tracts were irrigated with 0.9% NaCl solution. Table 3 and Figure 1 show the treatment of intrabiliary rupture.

**Evidence of obstructive jaundice with cystic contents in large common bile duct** There were three such cases. Two of them were treated by choledochoduodenostomy. Cystic content and hemobilia were diagnosed in the CBD of the third case in which choledochus was evacuated and T-tube drainage was performed. The specific cause of hemobilia could not be determined.

**Table 2** Objectives of the cysts and diagnostic investigations

Patient number	ALT, AST, ALP levels	Bilirubin levels (total/conj mg/dL)
1	High	7/5
2	N	N
3	N	N
4	N	N
5	High	5/3
6	High	5/3
7	High	11/8
8	N	N

**Evidence of obstructive jaundice, invisible orifice, normal caliber choledochus** There was only one such patient. Hyperbilirubinemia was 5 mg/dL. There was no bile staining of the cystic liquid. No pathological change was found at the inspection or palpation of the CBD, therefore no surgical procedure was performed for intrabiliary rupture. However, bile leakage in the range of 1 000 mL/d was diagnosed in the early post-operative period. Perihepatic biliary collection was also diagnosed later and drainage under US guidance was performed. Bile leakage subsequently regressed and stopped in 2 wk.

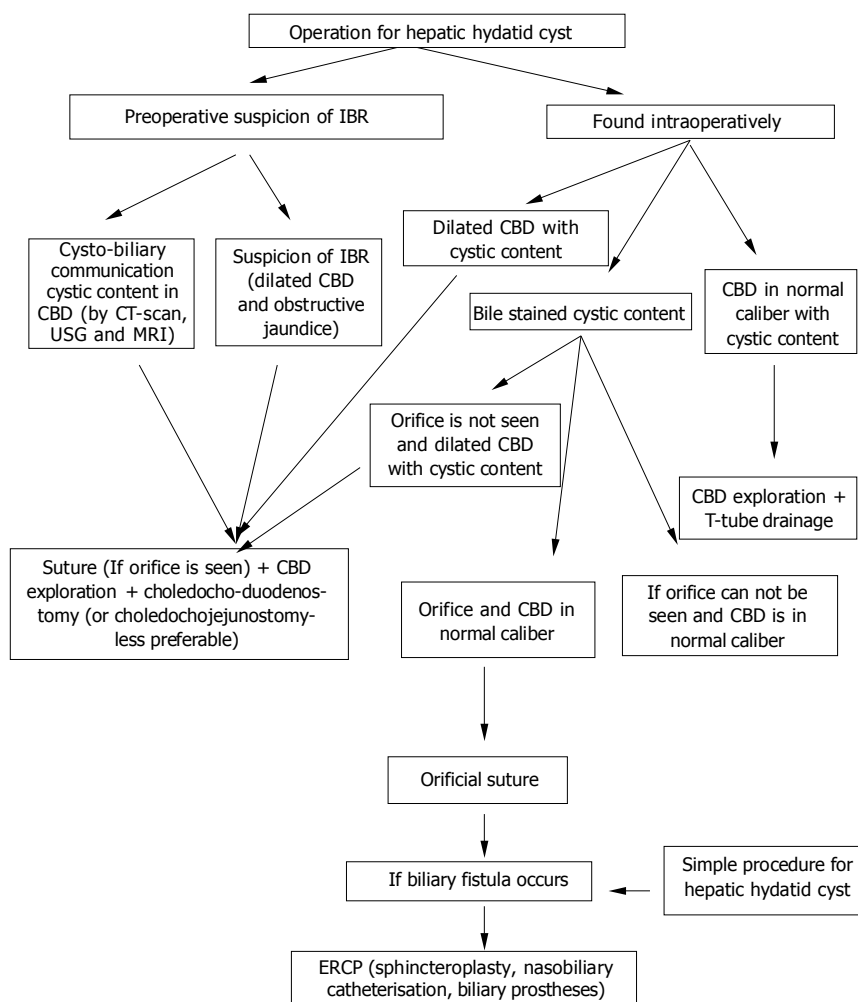
**The cases with bile stained cystic liquid** Two of the three such cases had visible orifices and were treated by suturing with nonabsorbable materials. At last one orifice could not be identified. Choledochotomy was done since it was suspected that there was cystic content in the CBD, but cystic content could not be seen. T-tube drainage was carried out. Biliary fistulae did not occur.

In a case of *unsuspected intrabiliary rupture*, a cyst, 10 cm in diameter was present in the right lobe. The cystic liquid was not stained by bile. Extrahepatic biliary system was found normal intra-operatively. No orifice could be identified because of difficult localization. A high output (1 000 mL/d) biliary fistula occurred on the first post-operative day and continued for a week. ERCP and sphincteroplasty were performed and the biliary fistula stopped at the 4<sup>th</sup> post-operative week.

T-tubes were removed on the 10<sup>th</sup> d in patients with a normal caliber choledoch, while the removal was delayed on the 20<sup>th</sup> d in patients with an enlarged choledoch with daughter vesicles and hemobilia.

**Table 3** Diagnosis and treatment of cystobiliary fistulae

Patient number	Clinical and diagnostic characteristics of the patients	Treatment of common bile duct
1	Evidence of obstructive jaundice with cystic contents in large CBD	Choledochoduodenostomy
2	Bile stained cystic liquid and visible orifice	Suturing orifice
3	Bile stained cystic liquid and visible orifice	Suturing orifice
4	Bile in cystic liquid	T-tube drainage
5	Evidence of obstructive jaundice, invisible orifice, normal caliber choledochus (postop biliary leakage)	Percutaneous drainage under US guidance post-operatively
6	Evidence of obstructive jaundice with cystic contents in a large CBD	Choledochoduodenostomy
7	Evidence of obstructive jaundice, hemobilia, enlargement of the CBD with cystic content	T-tube drainage
8	No suspicion of intrabiliary rupture	ERCP and sphincteroplasty



**Figure 1** Algorithm in the management of intrabiliary rupture (IBR: intrabiliary rupture, CBD: common bile duct).

Four patients had coincidental diseases. Two of them had cholelithiasis (cholecystectomy was done). One of them had coronary disease. Another patient had coronary disease, hypertension and diabetes mellitus. Perihepatic biliary collection noted above was only a complication.

The mean length of hospital stay was  $17 \pm 11.63$  d (range 10-45 d). In one patient with prolonged biliary fistulae the hospital stay was 45 d. When this patient was excluded, the mean hospital stay was  $13 \pm 2.94$  d.

The mean follow-up period was  $57.87 \pm 40.47$  mo (range 3-120 mo). No morbidity or mortality was seen during this period.

## DISCUSSION

Intrabiliary rupture is a common and serious complication of hepatic hydatidosis. It occurs especially in centrally localized and high stage cyst. High intracystic pressure up to 80 cm H<sub>2</sub>O is a predisposing factor. The cyst rupture can occur in three clinical forms. Contained rupture occurs when the cyst contents are confined within the pericyst. Communicating rupture defines tearing of the pericyst and evacuation of cyst contents into the biliary tract or bronchioles. Direct rupture describes complete tear of the cyst wall and spillage of the cyst contents into the peritoneal or pleural

cavity. Small cysto-biliary communications develop in 80-90% of all HHCs<sup>[16]</sup>.

Furthermore, there are two different clinical settings associated with intrabiliary rupture: frank intrabiliary rupture and simple communication. In the former, the cyst content drains to biliary tract and causes cholestatic jaundice. In the latter simple communications are frequently overlooked and could cause biliary fistulae post-operatively<sup>[17]</sup>. If the cystobiliary opening was less than 5 mm, spontaneous drainage of the cystic content was uncommon and could be treated by suturing under the direct vision<sup>[18]</sup>. If the CBD diameter was larger than 5 mm, cystic content migration into the biliary tract would occur in 65% of the cases<sup>[19]</sup>. Vesicles, debris and purulent materials may be found in the biliary collection.

In all patients the most frequent symptoms were right upper quadrant pain and flatulence. Obstructive jaundice and fever have been recorded in 90% and 20% of the cases respectively. Nausea and vomiting were rare<sup>[2,3,9,20]</sup>.

Diagnosis of IBR is difficult and can be established pre-, intra- and post-operatively. When obstructive jaundice is present, US, CT-scan, magnetic resonance imaging (MRI) and scintigraphic investigation can show the cyst and cystobiliary communications, but in patients with no jaundice, a correct diagnosis can be made in only 25%. Radiodiagnostic

evaluation can also demonstrate cystic content in the gall bladder<sup>[7]</sup> and the CBD<sup>[1,4,19,21]</sup>. On the other hand, cholelithiasis and choledocholithiasis are common (81-61.53%) coincidental diseases<sup>[6,7]</sup>. Laboratory and serological tests can also be helpful for diagnosis.

If obstructive jaundice was not present and cystobiliary connection could not be seen pre-operatively, three findings should raise suspicion of cystobiliary fistulas, namely bile-stained cystic fluid, visualized and sutured bile leak orifice intra-operatively; intra-operative observation of CBD enlargement or the presence of cyst content in the common bile duct; unexpected post-operative bile drainage from the cavity drains<sup>[8,22,23]</sup>.

The orifice of bile leakage could be seen in 11.7-17.07% of the cases during the operation<sup>[3,23]</sup> while this was difficult in posteriorly localized cysts. In these cases, cholangiography could be done by a catheter pushed into the ductus cysticus or the cystobiliary fistula<sup>[3,4,8,22,23]</sup>. As an extreme procedure, puncture of choledochus and injection of radioopaque solution or methylene blue are helpful to diagnose intrabiliary rupture or to see the orifice. Eleftheriadis<sup>[23]</sup> emphasized that choledochoscopy could also be helpful in the diagnosis of IBR.

When an intrabiliary rupture is diagnosed pre- or intra-operatively, there are alternative treatment modalities in addition to conventional surgery of hepatic hydatid cyst. Cystic evacuation, removal of germinative layer, disinfection of cystic cavity are necessary. If cystobiliary orifice is seen and no cystic content is observed in a normal caliber choledochus, suturing the orifice is sufficient to prevent the complications. Videolaparoscopic suturing of the orifice has been reported in literature<sup>[8]</sup>.

When cystic content is observed in a normal caliber choledochus, choledochotomy+evacuation of cystic content and debris from biliary tree±irrigation with 0.9% NaCl solution and T-tube drainage are enough. If CBD enlargement is diagnosed with cystic content in it or in the gallbladder, choledochoduodenostomy is preferable. Some authors have reported the wide application of T-tube drainage in cases with high morbidity rates<sup>[3,4,6,7,9,22,24,25]</sup>. On the other hand, Roux and Y hepaticojejunostomy have been reported for the treatment of bile duct stricture associated intrabiliary rupture<sup>[20,26]</sup>. Open sphincteroplasty was also used in the last decades<sup>[25]</sup>.

The presence of cholangitis has been reported as a risk factor. Even T-tube drainage itself could cause cholestatic icterus<sup>[3,17,27]</sup> and is also a source of infection<sup>[28]</sup>. Some authors have used cystojejunostomy albeit the risk that cystic content drainage into the biliary tract could result in obstructive jaundice and cholangitis<sup>[21,29]</sup>.

When intrabiliary rupture was overlooked during the surgical treatment, biliary fistulae (up to 1 000 mL/d) were unavoidable and if this occurred ERCP would be necessary. Post-operative biliary fistula rate was about 20% in all cases<sup>[22,23,30,31]</sup>.

In the cases of overlooked cystobiliary fistulae, transsphincteric evacuation of the CBD and sphincteroplasty could be applied. Nasobiliary drainage could also be done. Usually, the majority of biliary fistulae could be closed in a few weeks<sup>[2,8,27,30,32]</sup>.

The usage of endoprotheses in biliary fistula was not

common. This can be considered in either high output bile leakage or for intractable fistulae<sup>[31]</sup>.

The morbidity and mortality rates of all patients were 19.44-43.03% and 1.8-4.5% respectively in literature. The most common causes of deaths were sepsis and hepatic failure<sup>[3,4,9,22,23]</sup>.

Hospital stay was the longest in the T-tube group. The patients undergoing choledochoduodenostomy had longer hospital stay than those undergoing simple orifice suturing<sup>[3]</sup>.

In conclusion, IBR has an algorithm in the diagnosis and treatment. If it is not detected pre- or intra-operatively, a biliary fistula is common, its morbidity and mortality rates are high. Detecting and suturing orifices in cystic wall are the best methods of treatment. When cystic content is found in choledochus or when biliary fistula occurs, more complex procedures are necessary.

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