

Diagnosis and surgical treatment of intrabiliary ruptured hydatid disease of the liver

IBRAHIM TAÇYILDIZ, M.D.
MUSTAFA ALDEMİR, M.D.
NEDİM ABAN, M.D.
CELALETTİN KELES, M.D.

Department of General Surgery, Dicle University, Diyarbakir, Turkey

Summary

Background and objectives. The major complication of hydatid disease of the liver is intrabiliary rupture of the cyst. The purpose of this study was to evaluate the outcome of patients with intrabiliary ruptured hydatid disease of the liver.

Method. Using a standardised data collection instrument, case records of patients who were operated on for hydatid disease of the liver diagnosed between January 1990 and December 2001 at Dicle University Hospital (DUH) were searched and 192 patients who had been operated for hydatid disease of the liver were detected. Of these, 20 patients (16 females, 4 males) were retrospectively reviewed for intrabiliary ruptured hydatid disease of the liver.

Results. Intrabiliary ruptured hydatid disease of the liver was determined in 10.4% ($N = 20$) of the patients ($N = 192$) operated for hydatid disease of the liver. The average age of patients was 38.9 ± 14.05 years (range 20 - 72 years). The duration of the symptoms was 3.4 ± 2.13 years (range 1 - 8 years). The most frequent symptoms were right upper quadrant/epigastric pain, dyspepsia, jaundice and pruritus. Diagnosis of hydatid cyst was principally made using ultrasonography. Twelve cysts (60%) were located in the right lobe, 5 (25%) in the left

lobe, and 3 (15%) in the right and left lobes. The size of the cysts was 12.6 ± 5.79 cm (range 6 - 20 cm). The average diameter of the common bile duct (CBD) was 20.45 ± 8.54 mm (range 10 - 40 mm). Dilated CBD in 16 patients (80%) and daughter cysts and debris in the CBD in 10 patients (50%) were found during operation. Partial cystectomy and capitonnage were performed in all patients. In addition, T-tube drainage in 17 patients, omentoplasty plus T-tube drainage in 2 patients and choledochoduodenostomy in 1 patient were carried out during operation. An internal opening of the biliary fistula was found and sutured in 12 patients (60%). Wound infections developed in 6 patients (30%), suppuration of the residual cavity in 4 patients, and wound dehiscence in 2 patients. Two patients (10%) died from sepsis-multiple organ failure and hepatic failure. The average period of hospitalisation was 28.75 ± 19.1 days (range 10 - 103 days).

Conclusions. If bile-stained cystic fluid and a dilated CBD is found in patients with hydatid disease of the liver, choledochal exploration should be performed during operation. T-tube drainage may be preferred in the management of intrabiliary ruptured hydatid disease because of low morbidity, the ability to decompress intrabiliary pressure, easier monitoring of the biliary drainage and smaller alteration of the anatomy.

Hydatid cyst is a manifestation of the infestation of a cestode (*Echinococcus granulosus* (EG)) larva, which exists in its adult form in the canine intestine. Although the natural intermediate carriers are sheep, deer, cattle, and caribou, humans may become an accidental intermediate host, with the disease manifesting as hydatid cysts.¹ The natural history of EG localised in the liver is closely related to the biliary tract. The biliary tract lesions are various and explain the diversity of clinical features and the multiple surgical procedures suggested.^{2,3}

The major complication of hydatid disease of the liver is intrabiliary rupture of the cyst, which is seen in 9 - 17% of cases.^{4,7} Intrabiliary rupture of a cyst occurs as a result of augmented intracystic pressure and necrosis of the growing cyst and less frequently due to trauma. The necrotic cyst wall fragments and daughter cysts may lead to obstructive jaundice and related complications such as acute cholangitis and

recurrent pancreatitis.^{3,5,8} We evaluated results of surgically managed patients with intrabiliary ruptured hydatid disease of the liver in our hospital.

Material and methods

Dicle University Hospital (DUH), a tertiary referral centre and the largest hospital in south-east Turkey, cares for the vast majority of patients with hydatid disease of the liver who are referred from other hospitals. Using a standardised data collection instrument, case records of patients who were operated on for hydatid disease of the liver diagnosed between January 1990 and December 2001 at DUH were searched and 192 patients who had been operated on for hydatid disease of the liver were detected. Of these, 20 patients (16 females, 4 males) were retrospectively reviewed for intrabiliary ruptured hydatid disease of the liver. The

average age of patients was 38.9 ± 14.05 years (range 20 - 72 years). Most of the patients were originally from rural areas, and all had been exposed to sheep or dogs. The duration of the symptoms was 3.4 ± 2.13 years (range 1 - 8 years). The most frequent symptoms were right upper quadrant/epigastric pain, dyspepsia, jaundice and pruritus (Table I).

TABLE I. CLINICAL MANIFESTATIONS IN PATIENTS WITH INTRABILIARY RUPTURED HYDATID DISEASE OF THE LIVER

	Number of patients	%
Abdominal pain	16	80
Dyspepsia	16	80
Jaundice	14	70
Pruritus	14	70
Anorexia	10	50
Palpable hepatic mass	8	40
Fever	6	30
Allergic reactions	3	15

In all cases ultrasound (US) was performed for diagnosis of the hydatid cysts (Fig. 1). Computed tomography (CT) scan and magnetic resonance (MR) cholangiography were other imaging modalities used to identify hydatid cysts (Fig. 2). Pre-operative bilirubin, alkaline phosphatase (AP), alanine aminotransferase (ALT), aspartate aminotransferase (AST), gamma-glutamine transferase (GGT) and amylase were examined. Criteria for rupture into the biliary tract were the presence of daughter vesicles in the biliary tract, staining of the cystic contents with bile, connection demonstrated by pre-operative US/CT or intra-operative cholangiography. All patients were treated surgically under prophylactic antibiotic coverage. Depending on the location and size of the cyst, three types of operations were performed: (i) partial cystectomy, scolical irrigation, capitonnage and T-tube drainage; (ii) partial cystectomy, scolical irrigation, capitonnage, omentoplasty and T-tube drainage; and (iii) partial cystectomy, scolical irrigation, capitonnage and choledochoduodenostomy. Biliary gaps in the cyst cavity were carefully inspected and ligated using chromic catgut sutures. Germinative membrane, daughter cysts, scolices, and fluid were removed but the pericyst wall was not. Before any manipulation, the peritoneal cavity was isolated using moist towels. About 20 cc of fluid was then aspirated and the cyst cavity filled with 10% povidone-iodine. All patients under-



Fig. 1. US of the abdomen showing a multi-loculated cystic lesion in the right lobe of the liver.

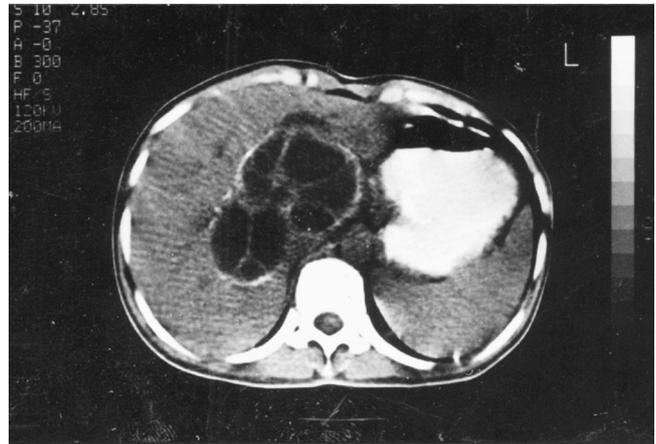


Fig. 2. CT scan of the abdomen showing multi-loculated cystic lesion in the right lobe of the liver.

went cholecystectomy and common bile duct (CBD) exploration. After complete evacuation of the cyst contents from the CBD, it was irrigated with saline. Subsequently, T-tube drainage or choledochoduodenostomy were undertaken in all patients. The residual cavity of the small cysts was treated by capitonnage, and the large or deeply situated cavities were treated with omentoplasty with or without external tube drainage.

Results

Intrabiliary ruptured hydatid disease of the liver was determined in 20 (10.4%) of the 192 patients operated for hydatid disease of the liver. Twelve (60%) of the cysts were located in the right lobe, 5 (25%) in the left lobe and 3 (15%) in both the right and left lobes. US was performed on all patients while CT was performed on 7 patients and MR cholangiography on 1 patient. Highest diagnostic sensitivities were achieved for hydatid disease using US and CT examinations (95% and 100%, respectively). The ALT and AST in 10 patients (50%), bilirubin in 14 patients (70%), AP in 8 patients (40%), GGT in 8 patients and amylase in 1 patient (5%) were found to be elevated. Leukocytosis was determined in 14 patients (70%) and eosinophilia in 10 patients (50%) (Table II). Cholelithiasis in 1 patient, choledocholithi-

TABLE II. LABORATORY AND RADIOLOGICAL MANIFESTATIONS IN PATIENTS WITH INTRABILIARY RUPTURED HYDATID DISEASE OF THE LIVER

	Number of patients	%
Elevated ALT and AST	10	50
Elevated bilirubin	14	70
Elevated AP	8	40
Elevated GGT	8	40
Elevated amylase	1	5
Leukocytosis	14	70
Eosinophilia	10	50
US diagnosis	20	100
US + tomographic diagnosis	7	35
US + MR cholangiographic diagnosis	1	5

ALT = alanine aminotransferase; AST = aspartate aminotransferase; AP = alkaline phosphatase; GGT = gamma-glutamine transferase; US = ultrasound; MR = magnetic resonance.

asis in 2 patients, recurrent pancreatitis in 1 patient, and pregnancy (6 months) in 1 patient were determined coincidentally.

All patients underwent operative therapy. Cholecystectomy was undertaken for all patients. Hydatid cyst fluids were seen to be bile-stained in all cases during operation. The size of the cysts was 12.6 ± 5.79 cm (range 6 - 20 cm). The CBD was explored in all cases. The average diameter of CBDs was 20.45 ± 8.54 mm (range 10 - 40 mm). The dilated CBD in 16 patients (80%) and daughter cysts and debris in the CBD of 10 patients (50%) were found during operation. The mean total bilirubin level was 5.02 ± 3.04 mg/dl (range 2.5 - 11.8 mg/dl) in 14 jaundiced patients. Partial cystectomy and capitonnage were performed in all patients. In addition, T-tube drainage in 17 patients, omentoplasty plus T-tube drainage in 2 patients and choledochoduodenostomy in 1 patient were carried out during operation. The internal opening of the biliary fistula was found and sutured in 12 patients (60%) (Table III). Wound infections developed in 6 patients (30%), suppuration of the residual cavity in 4 patients and wound dehiscence in 2 patients. Two patients (10%) died from sepsis-multiple organ failure and hepatic failure. The average period of hospitalisation was 28.75 ± 19.1 days (range 10 - 103 days).

Discussion

Hydatid disease has been a major worldwide health problem since the time of Hippocrates. Although more prevalent in certain sheep-raising areas, recent reports indicate that the disease has also become increasingly widespread in other regions.^{1,9,10} Hydatid cyst disease causes very few symptoms until complications occur. Usually the cyst enlarges gradually, becomes manifest by its size, and then the patient may become symptomatic. The cause of symptoms and signs are local pressure (abdominal pain and dyspepsia), infection (fever and leukocytosis), displacement of the liver (abdominal mass and abnormal liver function test) and rupture into the abdominal cavity (obstructive jaundice, eosinophilia and pruritus).^{8,11} In our study, the most frequent symptoms were right upper quadrant/epigastric pain (80%), dyspepsia (80%), jaundice (70%) and pruritus (70%). Occasionally, symptoms of biliary colic, jaundice, recurrent pancreatitis, or cholangitis occur with rupture of the cyst into the biliary tract. Daughter cysts and debris may obstruct the CBD and cause dilatation and operative intervention then becomes mandatory.^{1,5,12,13}

A definite pre-operative diagnosis of hydatid disease can be achieved by taking a complete history, physical examination, and detailed investigation. However, in hydatid liver disease, the diagnosis of intrabiliary rupture is very difficult and must first be suspected. In areas where the disease is endemic, any patient having fever with rigor, jaundice and pain in the right upper quadrant, with or without enlargement of the liver or a palpable abdominal mass, should be regarded as a case of intrabiliary rupture of hydatid cyst until proved otherwise.¹⁴⁻¹⁶

Although the diagnosis is usually best established by US, occasionally CT examination and MR cholangiography are required for a precise pre-operative diagnosis.^{11,16,17} Our findings indicate that the sensitivity of US and CT for the diagnosis of hydatid disease of the liver are 95% and 100%, respectively. The presence of a dilated CBD, jaundice, or both in addition to a cystic lesion of the liver is strongly suggestive of a hydatid cyst with intrabiliary rupture.^{14,18} In suspected cases, endoscopic retrograde cholangiopancreatography (ERCP) allows the patient to undergo elective surgery in an improved condition once the suppurative cholangitis has settled.^{18,19} However, we have no experience in pre-operative ERCP for the diagnosis of intrabiliary rupture in hydatid liver disease.

Surgical intervention is directed at the treatment of the cyst cavity and affected biliary tract. Meticulous evacuation of the cyst contents is required after isolation and local use of scolicedals.¹¹ Although numerous techniques related to the treatment of the cyst cavity have been recommended, their indications remain controversial.^{18,20-22} In the patients in this study, cystectomy, capitonnage and absorbable suture ligation of the open biliary ends to the remaining cavity was the principal method. Capitonnage, omentoplasty and drainage were performed in the large cavities, while capitonnage was performed in small cavities. Intra-operative diagnosis is very important when the pre-operative investigations do not reveal the communication. The presence of a dilated CBD with a liver cyst is an indication for duct exploration. Peri-operative cholangiography or injection of methylene blue should be performed in every case.¹⁸ In our study, the dilated CBD in 16 patients (80%) and daughter cysts and debris in the CBD in 10 patients (50%) were found during operation.

The presence of intrabiliary rupture requires exploration and drainage of the biliary tract. Options for operative management of intrabiliary ruptured hydatid disease of the liver include cyst cavity management and decompression of the biliary tract to assist in healing of communication between the cyst and biliary system. It seems likely that when bile-stained fluid is found within the cyst cavity in a patient with hydatid disease of the liver there is communication between the cyst and biliary tree. Therefore, communication between the cyst and bile duct must be carefully examined and ligated, in order to avoid the possibility of complications during the postoperative period. However, ligation of the internal opening is not a reliable procedure for closing this communication. As long as intrabiliary pressure stays high, closure of the internal opening will be difficult. Decompressing pressure in the biliary system can be achieved by different biliary drainage procedures.^{5,6,8,18} Various techniques including choledochoduodenostomy,^{8,18,23} T-tube drainage^{9,11,14,24} and transduodenal sphincteroplasty^{8,25} have been recommended to decompress intrabiliary pressure. Placement of a T-tube enables easier monitoring of the biliary drainage during the postoperative period and prevents unnecessary alteration of the anatomy and its undesirable long-term consequences. Also, it is a much easier and faster technique in the high-risk patient and free from the peri-operative morbidity of a biliiodigestive anastomosis or sphincteroplasty.¹¹ In this study, we performed choledochotomy and choledochal exploration in all patients. In addition we undertook choledochoduodenostomy in 1 patient in whom CBD size was larger than 20 mm in diameter. The preferred method (95%) was choledochotomy and choledochal exploration with T-tube drainage in our series. Patients were followed up for 15 months - 12 years and no late complications such as choledochal stricture were observed in these patients. If surgeons observe an enlarged CBD and hydatid cyst fluid stained with bile during operations, they should perform choledochotomy and choledochal exploration. Paksoy *et al.*²⁶ reported that they had performed left hepatic resection (atypical, segmentary or lobar) in 12% of their patients with intrabiliary ruptured hydatid disease of

TABLE III. SURGICAL MANAGEMENT OF INTRABILIARY RUPTURED HYDATID DISEASE OF THE LIVER

Surgical treatment	Number of patients	%
Cystectomy + capitonnage + T-tube drainage	17	85
Cystectomy + capitonnage + omentoplasty + T-tube drainage	2	10
Cystectomy+ capitonnage + choledochoduodenostomy	1	5

the liver. Because of localisation and the number of cysts, presence of obstructive jaundice and haemorrhagic diathesis, we did not decide on liver resection in our cases.

In conclusion, if bile-stained cystic fluid and a dilated CBD is found in patients with hydatid disease of the liver, choledochal exploration should be performed during operation. T-tube drainage may be preferred in the management of intrabiliary ruptured hydatid disease because of low morbidity, the ability to decompress intrabiliary pressure, easier monitoring of the biliary drainage and smaller alteration of the anatomy.

REFERENCES

- Dothy JE, Pitt HA. Cystic lesions of the liver and biliary tree. In: Moody FG, ed. *Surgical Treatment of Digestive Disease*, Year Book Medical Publishers, Chicago, 1986: 344-351.
- Maillet B, Le Treut YP, Sabiani P, Pol B, Bricot R. Hydatid cyst of the liver: analysis of postoperative morbidity. Apropos of a recent series of 52 cases. *Ann Chir* 1986; **40**: 389-395.
- Zaouche A, Haouet K, Jouini M, El Hachaichi A, Dziri C. Management of liver hydatid cysts with a large biliocystic fistula: multicenter retrospective study. Tunisian Surgical Association. *World J Surg* 2001; **25**: 28-39.
- Aktan AO, Yalin R, Yegen C, Okboy N. Surgical treatment of hepatic hydatid cysts. *Acta Chir Belg* 1993; **93**: 151-153.
- Erguney S, Tortum O, Taspinar AH, Ertem M, Gazioglu E. Complicated hydatid cysts of the liver. *Ann Chir* 1991; **45**: 584-589.
- Kornaros SE, Aboul-Nour TA. Frank intrabiliary rupture of hydatid hepatic cyst: diagnosis and treatment. *J Am Coll Surg* 1996; **183**: 466-470.
- Paksoy M, Karahasanoglu T, Carkman S, et al. Rupture of the hydatid disease of the liver into the biliary tracts. *Dig Surg* 1998; **15**: 25-29.
- Koksal N, Muftuoglu T, Gunerhan Y, Uzun MA, Kurt R. Management of intrabiliary ruptured hydatid disease of the liver. *Hepatogastroenterology* 2001; **48**: 1094-1096.
- Lygidakis NJ. Diagnosis and treatment of intrabiliary rupture of hydatid cyst of the liver. *Arch Surg* 1983; **118**: 1186-1189.
- Langer B. Surgical treatment of hydatid disease of the liver. *Br J Surg* 1987; **74**: 237-238.
- Ulualp KM, Aydemir I, Senturk H, et al. Management of intrabiliary rupture of hydatid cyst of the liver. *World J Surg* 1995; **19**: 720-724.
- Wong LS, Braghioroli-Neto O, Xu M, Buckels JA, Mirza DF. Hydatid liver disease as a cause of recurrent pancreatitis. *J R Coll Surg Edinb* 1999; **44**: 407-409.
- Becker K, Frieling T, Saleh A, Haussinger D. Resolution of hydatid liver cyst by spontaneous rupture into the biliary tract. *J Hepatol* 1997; **26**: 1408-1412.
- Kattan YB. Intrabiliary rupture of hydatid cyst of the liver. *Br J Surg* 1975; **62**: 885-890.
- Dadoukis J, Gamvros O, Aletras H. Intrabiliary rupture of the hydatid cyst of the liver. *World J Surg* 1984; **8**: 786-790.
- Barki Y, Charuzi I. Intrabiliary rupture of hydatid cyst of the liver: sonographic diagnosis. *J Clin Ultrasound* 1985; **13**: 40-41.
- Perek A, Numan F, Perek S, Durgun V, Kapan M, Aykuter G. Management of a patient with hepatic-thoracic-pelvic and omental hydatid cysts and post-operative bilio-cutaneous fistula: a case report. *Hepatogastroenterology* 1999; **46**: 2202-2207.
- Alper A, Ariogul O, Emre A, Uras A, Okten A. Choledochoduodenostomy for intrabiliary rupture of hydatid cysts of liver. *Br J Surg* 1987; **74**: 243-245.
- Cottone M, Amuso M, Cotton PB. Endoscopic retrograde cholangiography in hepatic hydatid disease. *Br J Surg* 1978; **65**: 107-108.
- Dawson JL, Stamatakis JD, Stringer MD, Williams R. Surgical treatment of hepatic hydatid disease. *Br J Surg* 1988; **75**: 946-950.
- Akinoglu A, Bilgin I, Erkokac EU. Surgical management of hydatid disease of the liver. *Can J Surg* 1985; **28**: 171-172.
- Demirci S, Eraslan S, Anadol E, Bozali L. Comparison of the results of different surgical techniques in the management of hydatid cysts of the liver. *World J Surg* 1989; **13**: 88-90.
- Ovnat A, Peiser J, Avinoah E, Barki Y, Charuzi I. Acute cholangitis caused by ruptured hydatid cyst. *Surgery* 1984; **95**: 497-500.
- Sayek I, Yalin R, Sanac Y. Surgical treatment of hydatid disease of the liver. *Arch Surg* 1980; **115**: 847-850.
- Moreno VF, Lopez EV. Acute cholangitis caused by ruptured hydatid cyst. *Surgery* 1985; **97**: 249.
- Paksoy M, Karahasanoglu T, Carkman S, et al. Rupture of the hydatid disease of the liver into the biliary tracts. *Dig Surg* 1998; **15**: 25-29.