ORIGINAL ARTICLE

Intrabiliary Rupture of a Hepatic Hydatid Cyst

Associated Clinical Factors and Proper Management

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Hypothesis: The prediction of an intrabiliary rupture of a hepatic hydatid cyst using associated clinical factors is important for early diagnosis and proper management.

Design: Case series of patients with hepatic hydatid cysts treated between January 1, 1992, and January 1, 2000, in a single institution.

Setting: A tertiary care teaching hospital.

Patients: The clinical findings in 116 patients with a hepatic hydatid cyst were reviewed. Of the 116 patients, 24 (21%) had a cyst-biliary communication: 15 (13%) had an occult rupture, and 9 (8%) had a frank rupture.

Main Outcome Measures: The following variables were analyzed as potential predictors of an intrabiliary rupture: age, sex, type and duration of symptoms, findings on physical examination, leukocyte count, liver function test results, serologic test results, suggestive ultrasonographic findings, ultrasonographic cyst features (type, diameter, number, and localization), and whether the cyst is primary or recurrent.

Results: The independent clinical factors for the presence of an occult rupture were a history of nausea and vomiting ($P=.004$), alkaline phosphatase level greater than 144 U/L ($P=.004$), total bilirubin level greater than 0.8 mg/dL (>13.5 µmol/L) ($P<.001$), and cyst diameter greater than 14.5 cm ($P<.001$) in multivariate analysis. Multivariate analysis also showed that history of jaundice ($P<.001$), jaundice found on physical examination ($P=.05$), cyst diameter greater than 10.5 cm ($P=.009$), a type IV cyst ($P<.001$), and suggestive ultrasonographic findings ($P<.001$) were the independent clinical predictors for the presence of a frank intrabiliary rupture. Patients with cyst-biliary communications had increased morbidity rates (13 [54%] of 24 patients vs 13 [14%] of 92 patients; $P<.001$) and longer mean postoperative hospital stays (13.7 vs 9.4 days; $P=.03$) compared with others.

Conclusion: Clinical predictors should be considered for early diagnosis and proper management of intrabiliary ruptures in patients with hepatic hydatid cysts.

Arch Surg. 2001;136:1249-1255

HYDATID DISEASE (echinococcosis) is a zoonosis caused by the larval stage of *Taenia echinococcus*. Humans are intermediate hosts and become infected directly by contact with canines or indirectly by food, water, and contaminated objects. Echinococcosis is endemic in many Mediterranean countries, the Middle and Far East, South America, Australia, and east Africa.[1] However, the distribution of the disease is worldwide, since the dog, a definitive host, is a common animal all over the world. Furthermore, because of population migration, its prevalence in nonendemic regions has been increasing.

Most patients have a single organ involved and harbor a solitary cyst, localized in approximately two thirds of the patients in the liver. Some cysts may grow (average increase, 1-30 mm/y) and then persist without a noticeable change for many years; others may collapse and can completely disappear.[2] An enlarging cyst may cause compressive atrophy of surrounding hepatocytes and fibrosis. Compression and displacement of biliary ducts are frequent. At the point of contact with a biliary duct, a spontaneous rupture may occur. This communicating intrabiliary rupture has been classified as a frank perforation with overt passage of hydatid material into the biliary tract and as occult leakage with signs of suppuration only.[3]

Early diagnosis and treatment of an intrabiliary rupture of a liver hydatid cyst are mandatory, since it may lead to obstruction of the biliary system, with a 50% mortality rate.[4] It has also been suggested

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PATIENTS AND METHODS

A prospective database containing 116 patients with hepatic hydatid cysts (the cyst was primary in 106 and recurrent in 10) treated between January 1, 1992, and January 1, 2000, was evaluated. A cyst-biliary communication was detected in 24 (21%) patients. These patients were examined in 2 groups: 15 (13%) had an occult perforation and 9 (8%) had a frank intrabiliary rupture. A *frank intrabiliary rupture* was defined as an overt passage of the hydatid material into the biliary system. An *occult perforation* was defined as the presence of bile in the cyst without an overt passage of intracystic content into the bile duct.

All patients underwent a complete blood cell count and liver function test determinations. A real-time abdominal ultrasonographic examination was performed in all patients. Computed tomography was not used routinely. An indirect hemagglutination test and a radioallergosorbent test were used for serologic confirmation of the diagnosis. Endoscopic retrograde cholangiography (ERC) could be performed only in a few patients because of unavailability, although it was indicated in more. Patients were operated on through either a right subcostal or a midline incision. All patients underwent chemotherapy (albendazole) for at least 3 months, beginning from 10 to 15 days before their operations or any other invasive procedures.

Liver cysts were classified into 5 types according to ultrasonographic appearance[6]: type I, a simple hydatid cyst with pure fluid collection; type II, the cyst contains undulated hyperechogenic membranes that float in the cystic fluid (detached germinative layer of the endocyst); type III, the cyst contains secondary vesicles (daughter and granddaughter cysts); type IV, the cyst has a heterogeneous echopattern and is filled with a matrix or an amorphous mass; and type V, the cyst has a thick and calcified wall. The presence of irregular linear echogenic structures without acoustic shadowing in the bile duct and/or the dilated biliary tract was accepted as suggestive ultrasonographic findings for a cyst-biliary communication.[7]

The following variables were analyzed as potential predictors of a cyst-biliary communication: age, sex, type and duration of symptoms, findings on physical examination, leukocyte count, liver function test results (alanine aminotransferase [ALT], aspartate aminotransferase [AST], alkaline phosphatase [ALP], γ-glutamyl transpeptidase [GGT], and bilirubin levels), serologic test results (indirect hemagglutination test), suggestive ultrasonographic findings, ultrasonographic cyst features (type, diameter, number, and localization), and whether the cyst is primary or recurrent. The upper normal limits for liver function test results in our laboratory were as follows: AST, 37 U/L; ALT, 40 U/L; ALP, 133 U/L; GGT, 50 U/L; total bilirubin level, 1.2 mg/dL (20 µmol/L); and direct bilirubin level, 0.4 mg/dL (6 µmol/L). None of our patients had acute liver disease or chronic hyperbilirubinemia. *Leukocytosis* was defined as a leukocyte count greater than 0.01×10^6/µL.

Postoperative morbidity, mortality, and length of hospital stays of patients with cyst-biliary communications were compared with those of others. Long-term results were not compared because of lack of data. A comparison was also made between patients with a frank rupture and patients with an occult rupture for all the examined clinical features.

Quantitative data were compared using the *t* test, and categorical data were compared using the Pearson χ^2 test or the Fisher exact test (where appropriate) for the presence of a frank intrabiliary rupture and an occult rupture. Receiver operating characteristic curve analysis was used to determine ideal cutoff values for those variables found to be significant on a *t* test. Logistic regression analysis was used for multivariate analysis. Comparisons between subgroups of patients (patients with frank intrabiliary ruptures, patients with occult communications, and others without a cyst-biliary communication) were performed using the *t* test, the χ^2 test, or the Fisher exact test. $P \leq .05$ was considered statistically significant. Some percentages may not total 100 because of rounding.

that biliary cirrhosis could be a late sequel.[5] Imaging techniques are helpful in making the diagnosis. However, some of them are quite expensive and invasive. Moreover, they have limited value in the diagnosis of an occult rupture. Some noninvasive clinical factors associated with intrabiliary rupture of a cyst can be useful in establishing the proper diagnosis and management. These factors have not been well-defined before.

This study investigates the clinical factors associated with an intrabiliary rupture of a hepatic hydatid cyst for early diagnosis and management of cyst-biliary communications in patients with liver echinococcosis and presents our experience with this condition.

RESULTS

Of the 116 patients, 37 (32%) were male and 79 (68%) were female. The age range was from 8 to 75 years (mean, 40 years). The most common presenting symptom was right upper quadrant abdominal pain (93 patients [80%]). Fifty-four patients (47%) presented with nausea and vomiting. Twenty-six patients (22%) had flatulence. A history of jaundice was found in 9 (8%) of the patients. Thirteen patients (11%) were asymptomatic. The duration of symptoms ranged from 1 month to 7 years (median, 3 months).

Eighty-eight patients (76%) had a single cyst and 28 (24%) had multiple cysts. The cysts were localized only in the right lobe of the liver in 77 (66%) of the patients, and bilobar involvement was seen in 13 (11%). A total of 152 hepatic cysts were treated in the 116 patients. Of the 152 cysts, 50 (33%) were type I, 25 (16%) were type II, 39 (26%) were type III, and 38 (25%) were type IV. The cyst diameter ranged from 2 to 30 cm (mean, 10 cm). In 17 (15%) patients, concomitant extrahepatic involvement was present. A splenectomy was performed in 4 (3%) and a partial omentectomy was performed in 7 (6%).

Of the 116 patients, 111 were treated surgically and 4 underwent percutaneous drainage using ultrasonic guidance. A cyst that had been drained by ERC and endoscopic sphincterotomy (ES) healed completely, without need for a surgical intervention.

Table 1. Comparison of Patients With and Without an Intrabiliary Rupture

Clinical Variable*	Patients With a Rupture (n = 24)	Patients Without a Rupture (n = 92)	*P* Value
Categorical†			
Male-female	10:14 (42:58)	27:65 (29:71)	.25
Symptoms			
History of jaundice	6 (25)	3 (3)	<.001
Nausea and vomiting	17 (71)	37 (40)	.007
Abdominal pain	22 (92)	71 (77)	.11
Flatulence	11 (46)	15 (16)	.007
Signs			
Tenderness	5 (21)	16 (17)	.70
Jaundice	5 (21)	3 (3)	.002
Cyst features			
Type			
I	4 (17)	34 (37)	.07
II	4 (17)	15 (16)	.97
III	4 (17)	26 (28)	.25
IV	12 (50)	17 (19)	.001
Single	17 (71)	71 (77)	.52
Multiple	7 (29)	21 (23)	
Primary	22 (92)	84 (91)	.96
Recurrent	2 (8)	8 (9)	
Leukocytosis	2 (8)	4 (4)	.43
Suggestive ultrasonographic findings	8 (33)	0	<.001
Elevated direct bilirubin level	9 (38)	8 (9)	<.001
IHA titer >1/1600	11 (46)	29 (32)	.19
Morbidity			
Total	13 (54)	13 (14)	<.001
Wound infection	5 (21)	5 (5)	.02
External biliary fistula	6 (25)	0	<.001
Pulmonary	2 (8)	8 (9)	1.00
Mortality	1 (4)	1 (1)	.30
Quantitative‡			
Age, y	42.2 ± 18.0	40.0 ± 16.7	.57
Duration of symptoms, mo	8.3 ± 10.1	7.8 ± 14.7	.92
Cyst diameter, cm	13.9 ± 5.9	9.9 ± 3.9	<.001
ALT level, U/L	66.8 ± 89.3	28.4 ± 32.9	.001
AST level, U/L	45.0 ± 52.4	24.6 ± 19.6	.003
GGT level, U/L	117.1 ± 137.7	46.7 ± 66.4	.005
ALP level, U/L	328.3 ± 271.1	130.1 ± 120.5	<.001
Total bilirubin level, mg/dL§	2.6 ± 5.0	0.8 ± 0.6	.001
Postoperative hospital stay, d	13.7 ± 9.2	9.4 ± 8.4	.03

*IHA indicates indirect hemagglutination; ALT, alanine aminotransferase; AST, aspartate aminotransferase; GGT, γ-glutamyl transpeptidase; and ALP, alkaline phosphatase.
†Data are given as number (percentage) of patients.
‡Data are given as mean ± SD.
§To convert total bilirubin from milligrams per deciliter to Système International units of micromoles per liter, multiply milligrams per deciliter by 17.1.

PATIENTS WITH CYST-BILIARY COMMUNICATIONS

Communications with the biliary system were encountered in 24 (21%) patients. Ten (42%) were male and 14 (58%) were female. The age range was between 16 and 70 years (mean, 42 years). Most patients had right upper quadrant abdominal pain (22 [92%]). Seventeen patients (71%) presented with nausea and vomiting. Eleven patients (46%) complained of flatulence. A history of jaundice was found in 6 (25%) patients. Only 1 patient (4%) was asymptomatic. The duration of symptoms ranged from 1 month to 3 years (median, 4 months).

The disease was primary in 22 (92%) patients, and recurrent in 2 (8%). Seventeen patients (71%) had a single cyst, and 7 (29%) had multiple cysts. The cysts were localized in the right lobe of the liver in 14 (58%) patients and in the left lobe in 8 (33%). Two (8%) of the patients had bilobar involvement. Of the 24 communicating cysts, 12 (50%) were type IV, 4 (17%) were type III, 4 (17%) were type II, and 4 (17%) were type I. The cyst diameter ranged from 5 to 30 cm (median, 15 cm). A comparison of various clinical factors of patients with and without cyst-biliary communications is presented in **Table 1**.

MANAGEMENT OF FRANK COMMUNICATIONS

Of the 24 patients, 9 (38%) had a frank intrabiliary rupture with overt passage of hydatid material into the biliary tract and 15 (62%) had an occult communication. Endoscopic retrograde cholangiography with ES was performed in 2 of the 9 patients with a frank communica-

Intraoperative cholangiography performed through the ruptured bile canaliculi using a plastic cannula (arrow) in a patient with an occult cyst-biliary communication.

tion: the cyst was completely drained in one, while the other patient, after passing hydatid material into the common bile duct (CBD), underwent an operation and bile leakage was sutured without CBD exploration. Cholecystectomy, choledochotomy, evacuation and lavage of the CBD, and partial cystectomy were performed in the remaining patients. Closure of the CBD with T-tube drainage was carried out in 2 patients and the other 5 underwent choledochoduodenostomies. Indications for a choledochotomy were as follows: suggestive ultrasonographic findings for a frank intrabiliary rupture in 5 patients and a dilated CBD (>2 cm) in 2. External drainage with or without omentoplasty was performed for the management of the cyst cavities. In the postoperative period, surgical site infections were observed in 2 patients. An external biliary fistula developed in 1 patient after removal of the T tube. It was closed 9 days after the patient underwent ERC and ES.

MANAGEMENT OF OCCULT COMMUNICATIONS

Partial cystectomy, evacuation of the cyst content, and drainage of the cyst cavity were performed in all patients with occult biliary communications. Cholecystectomy was performed in 2 patients for the presence of cholelithiasis. An intraoperative cholangiogram was obtained by one of the following means: through the opening of the bile duct into the cystic cavity using a thin plastic cannula (**Figure**) as described before,[1] through the cystic duct in patients who underwent cholecystectomies, or through the CBD using a needle catheter (a wing needle infusion set). Since the cholangiographic results were normal, a choledochotomy was not performed in any of these patients. The site of the bile leakage was identified and sutured with absorbable sutures in 11 of the 15 patients. Any obvious bile leakage could not be detected during the operations in the remaining 4 patients. These 4 patients and 1 among others with an occult rupture (total, 5 patients [33%]) had postoperative external biliary fistulas. The fistulas healed spontaneously in 2 to 20 days in 4 patients. It was necessary to perform ERC and ES for the closure of a high-output fistula in 1 patient. Other postoperative complications were surgical site infections in 3 (20%) patients and an intra-abdominal abscess in 1 (7%). One of the patients with an occult rupture died of a massive pulmonary thromboembolism on the second postoperative day.

A significantly higher morbidity rate and a longer postoperative hospital stay were noted in patients with an intrabiliary rupture compared with others without a cyst-biliary communication (Table 1).

COMPARISON OF PATIENTS WITH AN OCCULT RUPTURE AND THOSE WITH A FRANK RUPTURE

The suggestive ultrasonographic findings were encountered significantly more in patients with a frank rupture than in patients with an occult rupture (67% vs 13%; $P=.01$). Significantly higher percentages of patients with frank communications have type IV cysts compared with patients with occult ruptures (78% vs 33%; $P=.04$). The mean±SD postoperative hospital stay was longer in patients with frank ruptures than in patients with occult communications (19±9 vs 10±8 days; $P=.02$). All other clinical factors were similar in both groups.

SIGNIFICANT CLINICAL PREDICTORS FOR THE PRESENCE OF OCCULT AND FRANK RUPTURES

In univariate analysis, significant clinical factors associated with the presence of an occult communication were as follows: a history of nausea and vomiting, a history of flatulence, elevated liver function test results (ALT, ALP, and total bilirubin levels), and a larger cyst diameter on ultrasonography. Receiver operating characteristic curves determined the following ideal cutoff values for quantitative clinical factors in patients with occult ruptures: ALT level, 23.5 U/L; ALP level, 144 U/L; total bilirubin level, 0.8 mg/dL (13.5 µmol/L); and cyst diameter, greater than 14.5 cm. **Table 2** details the performance of each clinical factor associated with the presence of an occult rupture in univariate analysis.

A history of jaundice, jaundice found on physical examination, elevated liver function test results (ALT, AST, ALP, GGT, total bilirubin, and direct bilirubin levels), a larger cyst diameter, the presence of suggestive ultrasonographic findings, and a type IV cyst on ultrason-

Table 2. Performance of Clinical Factors Associated With an Occult Intrabiliary Rupture of a Hydatid Cyst on Univariate Analysis*

Clinical Factor	Sensitivity, %	Specificity, %	PPV, %	NPV, %	LR	*P* Value
A history of nausea and vomiting	80.0	59.8	24.5	94.8	8.6	.005
A history of flatulence	53.3	83.7	34.8	91.7	8.8	.001
ALT level >23.5 U/L	66.7	66.3	25.0	92.2	5.7	.02
Total bilirubin level >0.8 mg/dL (>13.5 µmol/L)	80.0	71.1	33.3	95.2	14.0	<.001
ALP level >144 U/L	73.3	76.5	35.5	94.2	13.7	<.001
Cyst diameter >14.5 cm	66.7	88.0	47.6	94.2	19.5	<.001

*PPV indicates positive predictive value; NPV, negative predictive value; LR, likelihood ratio; ALT, alanine aminotransferase; and ALP, alkaline phosphatase.

Table 3. Performance of Clinical Factors Associated With a Frank Rupture of a Hydatid Cyst on Univariate Analysis*

Clinical Factor	Sensitivity, %	Specificity, %	PPV, %	NPV, %	LR	*P* Value
A history of jaundice	44.4	96.7	57.1	94.7	12.0	.001
Jaundice found on physical examination	33.3	96.7	50.0	93.7	7.6	.009
ALT level >30.5 U/L	88.9	78.7	29.6	98.6	16.8	<.001
AST level >28.5 U/L	88.9	82.0	33.3	98.6	19.0	<.001
Total bilirubin level >1.0 mg/dL (>16.5 µmol/L)	77.8	80.7	30.4	97.1	12.6	.001
Direct bilirubin level >0.4 mg/dL (>6.5 µmol/L)	66.7	90.5	42.9	96.2	14.5	<.001
ALP level >275.5 U/L	77.8	90.6	46.7	97.5	19.9	<.001
GGT level >73.5 U/L	80.0	81.5	25.0	98.1	8.1	.008
Cyst diameter >10.5 cm	77.8	60.9	16.3	96.6	5.1	.04
A type IV cyst	77.8	81.5	29.2	97.4	13.2	<.001
Suggestive ultrasonographic findings	66.7	100.0	100.0	96.8	31.9	<.001

*PPV indicates positive predictive value; NPV, negative predictive value; LR, likelihood ratio; ALT, alanine aminotransferase; AST, aspartate aminotransferase; ALP, alkaline phosphatase; and GGT, γ-glutamyl transpeptidase.

ography were the significant clinical factors associated with the presence of a frank intrabiliary rupture on univariate analysis. Using receiver operating characteristic curve analysis, optimal cutoff values for quantitative clinical data were determined to be 30.5 U/L for ALT, 28.5 U/L for AST, 73.5 U/L for GGT, 275.5 U/L for ALP, 1.0 mg/dL (16.5 µmol/L) for total bilirubin, and 10.5 cm for cyst diameter. The direct bilirubin level was evaluated as a categorical variable, since it was recorded as so. **Table 3** details the performance of each clinical factor associated with the presence of a frank intrabiliary rupture on univariate analysis.

The independent clinical predictors for the presence of occult leakage were a history of nausea and vomiting, ALP level greater than 144 U/L, total bilirubin level greater than 0.8 mg/dL, and cyst diameter greater than 14.5 cm on multivariate analysis. On the other hand, a history of jaundice, jaundice found on physical examination, a cyst diameter greater than 10.5 cm, the presence of suggestive ultrasonographic findings, and a type IV cyst on ultrasonography were the independent clinical factors associated with the presence of a frank intrabiliary rupture (**Table 4**).

COMMENT

The intrabiliary rupture of a hepatic hydatid cyst is not a rare complication and may occur in 2 forms: an occult rupture, seen in 10% to 37% of the patients, or a frank rupture, seen in 3% to 17% of the patients.[8] In this series, 13% of the patients had an occult intrabiliary rupture and 8% of the patients had a frank rupture, in accordance with the figures cited. Early diagnosis and treatment are mandatory in these patients, since otherwise serious clinical situations with an increased mortality may ensue. A frank intrabiliary rupture of a cyst may lead to obstruction of the biliary system, cholangitis, and septicemia, with a 50% mortality rate.[4] Moreover, acute pancreatitis and acute cholecystitis caused by hydatid material were described in the literature.[9,10] It has also been suggested that biliary cirrhosis could be a late sequel of an intrabiliary cyst rupture.[5] Although an occult rupture is usually silent, it may give rise to a frank perforation at any time.

Ultrasonography or computed tomography may suggest the diagnosis of a frank intrabiliary rupture in most of the cases.[7,11,12] Similarly, in this series, ultrasonography permitted the diagnosis of a frank rupture in 6 of the 9 patients, with a sensitivity of 66.7% and a specificity of 100%. A cyst-biliary communication can also be detected through ERC, which is superior to ultrasonography or computed tomography in that cleansing the CBD of hydatid material and ES can be performed at the same time. However, it should be performed selectively, since it is an expensive and invasive procedure with well-known short- and long-term morbidities. Moreover, all the imaging techniques fall short as far as detection of occult communications is concerned. In the near future, magnetic resonance cholangiography may become a reliable diagnostic tool, but the experience with its use for the detection of ruptured hepatic hydatid cysts is limited at the moment.[13] In this study, we tried to determine the clinical predictors of an intrabiliary rupture of a hepatic hydatid cyst to establish an early diagnosis and proper management.

Table 4. Results of Multivariate Analysis*

Clinical Factor	Positive Predictive Value, %	*P* Value
For an Occult Rupture		
History of nausea and vomiting	24.5	.004
ALP level >144 U/L	35.5	.004
Total bilirubin >0.8 mg/dL (>13.5 µmol/L)	33.3	<.001
Cyst diameter >14.5 cm	47.6	<.001
For a Frank Rupture		
History of jaundice	57.1	<.001
Jaundice found on physical examination	66.7	.05
Suggestive ultrasonographic findings	100.0	<.001
A type IV cyst	29.2	<.001
Cyst diameter >10.5 cm	16.3	.009

*ALP indicates alkaline phosphatase.

A history of nausea and vomiting, a serum ALP level greater than 144 U/L, a total bilirubin level greater than 0.8 mg/dL, and a cyst diameter larger than 14.5 cm were the independent risk factors, that is, predictors for the presence of an occult rupture in our study. It has been shown that intracystic pressure increases along with the diameter of a hydatid cyst.[14] This increased pressure apparently leads to a spontaneous intrabiliary rupture. The elevations of total bilirubin and ALP levels in patients with occult ruptures are possibly due to an occult passage of hydatid material into the biliary system intermittently in small amounts. Thus, an apparent biliary obstruction does not occur. Reabsorption of the bile from the cyst cavity may be another mechanism for the increased bilirubin and ALP levels. Although significantly associated with an occult rupture, a history of nausea and vomiting, which occurs perhaps due to toxic effects of hydatid material, is somewhat less objective and a poorer indicator of an occult rupture, with a low positive predictive value (24.5%) compared with others. Accordingly, we believe that a surgeon should suspect an occult intrabiliary rupture of a hydatid cyst if a patient has a large cyst and slightly increased ALP and bilirubin levels. In patients who are suspected to have an occult communication, early management should be planned, percutaneous drainage should be avoided, ultrasonography should be performed to rule out a frank perforation, a broad-spectrum antibiotic should be chosen for prophylaxis (because septic complications are expected to be increased in these patients with biliary communications), and use of scolicidal agents should be avoided because of the risk of sclerosing cholangitis.

Patients suspected of having an occult rupture should be treated surgically. A laparoscopic approach has also been described.[15] Bile in the cyst cavity does not mean a frank rupture in most of the patients. By obtaining a cholangiogram through the ruptured bile canaliculi (which we prefer), the cystic duct, or the CBD, an unnecessary CBD exploration can be avoided. Intraoperative ultrasonography may also be performed instead of intraoperative cholangiography. If the CBD appears to be normal and a partial cystectomy is to be performed, all that is needed is simple suturing of the rupture. When the communication is side to side, the sutures should be placed carefully so as not to cause an obstruction. In patients with a major communication, internal drainage by a Roux-en-Y pericystojejunostomy has also been shown to be effective in eliminating biliary leakage and fistulas, with a low morbidity rate.[16] Total cystectomy and liver resection are other alternatives. Sometimes, the site of the communication cannot be identified, as in the 4 patients in this series. It was recommended that methylene blue be injected into the CBD in these patients to see the site of occult leakage.[1] We did not have experience with this technique and, therefore, just drained the cyst cavities in these patients. In agreement with previous reports,[17,18] we believe that the cyst cavity should only be drained if a bile leak is present (preferably with closed-suction drains), since it was shown that liberal use of drains accounts for a high morbidity rate and a long hospital stay.[18] We also suggest drainage in patients with clinical predictors of an occult communication, even though no active bile leakage is detected, because the external biliary fistula rate is high in these patients.

When we compared the patients with an occult rupture with those with a frank perforation, all the clinical features were similar except that the patients with a frank rupture had suggestive ultrasonographic findings (irregular linear echogenic structures without acoustic shadowing in the bile duct and/or the dilated biliary system) and significantly more type IV cysts. A type IV cyst, suggestive ultrasonographic findings, a cyst diameter larger than 10.5 cm, jaundice found on physical examination, and a history of jaundice were independent clinical factors for the presence of a frank rupture. Interestingly, the cutoff value for cyst diameter was smaller in patients with a frank rupture than in patients with an occult rupture (10.5 vs 14.5 cm). Similarly, it was reported that when a frank communicating rupture occurs, the hydatid cyst becomes smaller.[4] In addition, a type IV cyst was significantly more frequent in patients with a frank rupture vs those with an occult rupture (78% vs 33%). By using these data, we can propose a scenario. When a cyst becomes larger, an occult cyst-biliary communication may occur due to increased intracystic pressure, whatever the cyst type is; this communication may become frank after a certain time, and the cyst becomes smaller because of the passage of the hydatid material into the biliary system. The cyst also becomes a type IV cyst, possibly due to effects of the bile and the secondary bacterial infection. Preoperative prediction of a frank rupture is important, because by performing preoperative ERC and ES an unnecessary CBD exploration can be avoided. Moreover, a laparoscopic approach may be preferred since it becomes easier. Furthermore, sometimes the cyst drains completely after ERC and ES without the need for surgery,[19] as in one of our patients. This study demonstrates that the presence of suggestive ultrasonographic findings, jaundice found on physical examination, and a history of jaundice are strong predictors for a frank rupture, with positive predictive values of 100.0%, 57.1%, and 66.7%, respectively. It is better to perform preoperative ERC and ES in the patients with 1 or more of these previously mentioned strong predictors. The presence of a type IV cyst and a cyst diameter larger than 10.5 cm

were poor predictors, with positive predictive values of 29.2% and 16.3%, respectively. Patients with any of these weaker predictors may best be examined with intraoperative cholangiography. On the other hand, if a combination of these factors is present, the positive predictive value increases to 63%. Consequently, a type IV cyst with a diameter larger than 10.5 cm is a good indication for preoperative ERC. In most of the patients with a frank communication, surgical treatment is indicated. During the operation, choledochotomy for the evacuation of the debris is performed, unless successful ERC and ES have been performed. Choledochoscopy can be useful for revealing retained hydatid material.[11] The cysts can be managed similar to those in patients with an occult rupture, as discussed before. Choledochotomies can be closed over T tubes[1,11,12] or choledochoduodenostomies can be performed with satisfactory results.[16,20] We preferred a choledochoduodenostomy in patients with a dilated CBD and obtained good results without any related morbidity.

Since a cyst-biliary communication is a life-threatening condition, early diagnosis and treatment are mandatory. Moreover, patients with such communications have significantly higher operative morbidity rates and longer hospital stays compared with others without such a communication. We conclude that better results can be obtained by using the clinical predictors that are demonstrated in this study for the early diagnosis and proper management of an intrabiliary rupture of a hydatid cyst of the liver.

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