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Asymptomatic occult cysto-biliary communication without bile into cavity of the liver hydatid cyst: A pitfall in conservative surgery

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ABSTRACT

Background: An occult cysto-biliary communication in liver hydatid disease is still a major problem in surgical practice. Radiologic and intraoperative findings may not be helpful to detect cysto-biliary communications in some asymptomatic patients with liver hydatid disease. Biliary leakage is a troubling complication that arises after conservative surgery in patients who have occult "insidious" cysto-biliary communications. We aimed to identify the factors which are associated with the risk of occult insidious cysto-biliary communications in patients preoperatively who developed biliary leakage after surgery.

Patients and methods: We investigated the records of 183 asymptomatic patients treated for liver hydatid cyst and analyzed potential predictors of occult insidious cysto-biliary communication, retrospectively.

Results: There were 115 female and 68 male patients; the mean age was 42.3 years. Occult insidious cysto-biliary communications which presented as postoperative biliary leakage found in 24 (13.1%). Independent clinical predictors were alkaline phosphatase >133 U/L, total bilirubin levels >1.2 mg/dL, white blood cell count >10,000/mm^3 and cyst diameter >10 cm on multivariate analysis. Seventeen of 24 were low output biliary fistula which resolved spontaneously within 9.2 days. The remaining 7 were high output biliary fistula for which endoscopic sphincterotomy was performed in all patients, fistulas resolved within 22.6 days. Average interval between endoscopic sphincterotomy and fistula closure was 10.3 days. Mean hospital stay was longer in patients with biliary leakage than in those without (9.8 vs. 4.2 day $p < 0.001$). There was no hospital mortality.

Conclusion: The predictors demonstrated in this study should allow the likelihood of occult insidious cysto-biliary communication to be determined and, thus, indicate the need for additional procedures during operations to prevent the complications of biliary leakage.

1. Introduction

Liver hydatid disease which is caused by *Echinococcus granulosus*, is a common public health problem all around the World.[1,2] Infestation of humans with larval form incidentally causes hydatid cyst formation in different organs, most commonly in the liver. Clinical features of hepatic hydatid cyst are generally nonspecific. The main objective of surgical treatment of hepatic hydatid cyst is to treat the current disease with minimal complications. Surgical treatment includes a broad range of interventions from radical surgeries such as hepatic resection and cystopericystectomy to conservative methods as removal of cyst ingredients and capsulorrhaphy, or external drainage.[3,4]

Cysto-biliary communications or intrabiliary rupture are the most common complications of hepatic hydatid cyst.[5] The communication between the biliary tree and the hydatid cyst can be frank or occult.[6] In frank cysto-biliary communication, biliary colic, obstructive jaundice and cholangitis due to complete or partial obstruction of the common bile duct by the elements of the hydatic cyst are common.[7,8] Diagnosis of frank communication is generally easy and can be managed as surgical and/or endoscopic procedures.[9,10] However, diagnosis of occult cysto-biliary communication is challenging, because the symptoms and radiological findings in preoperative period are not clear.[11] If the cysto-biliary opening cannot be detected and repaired during conservative surgery, postoperative biliary leakage may arise in such cases.[8,12] In some cases cysto-biliary communication can be detected by observing the bile in cyst cavity or cyst contents in the bile duct (occult honest cysto-biliary communication). In such cases, even if the cysto-biliary communication cannot be found and sutured,

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precautions should be taken during surgery in order to take bile leakage under control and treatment can be supported by further endoscopic methods. However, if bile in cyst or cyst content in bile duct is not detected and the patient is asymptomatic with regard to rupture (occult insidious cysto-biliary communication), prevention of biliary complications that occur during conservative surgery is challenging; therefore, prediction of such patients is of great significance.

The aim of this study was to evaluate possible risk factors for occult insidious cysto-biliary communication in patients with hepatic hydatid cyst.

2. Methods

The records of 252 patients treated for hepatic hydatic cyst between 1998 and 2008 were reviewed retrospectively and 183 patients were included in this study.

2.1. Terminology

A *cysto-biliary communication* was defined as a loss of continuity of the cyst wall adjacent to the bile duct. A *frank intrabiliary rupture or cysto-biliary communication* was defined as large tears involving large bile ducts with an overt passage of the hydatid material into the biliary system. An *occult perforation or cysto-biliary communication* was defined as small tears between the cyst wall and small biliary radicles without an overt passage of intracystic content into the bile duct. Occult intrabiliary rupture presents with biliary leakage, if the cysto-biliary opening cannot be detected and repaired at operation.[12] We divided occult cysto-biliary communication into two subgroups as follows. (1) An o*ccult honest cysto-biliary communication* which was defined as the presence of bile in the cyst or cysto-biliary opening detected, and (2) an o*ccult insidious cysto-biliary communication* is defined as development of biliary leakage following conservative surgery despite the absence of bile in the cyst cavity, preoperatively.

2.2. Exclusion criteria

Patients with frank or occult honest biliary communication, patients who received percutaneous drainage or curative resection, patients who had perforated or recurrent cyst or incomplete clinical records were excluded from the analysis (totally 69 patients) since we aimed to investigate risk factors for "occult insidious cysto-biliary communication". Patients with hyperbilirubinemia due to external compression of the hepatic biliary system were included in the study if a cysto-biliary fistula not identified in ERCP or during operation.

2.3. Preoperative evaluation

All patients underwent a complete blood cell count and liver function test determinations. To diagnose and evaluate liver hydatid cyst, Casoni's skin test and complement fixation test of Ghedini–Weinberg, plain radiography, and abdominal ultrasonography were performed in all patients but computed tomography was not used routinely.

Cysts were classified radiologically as unilocular (Gharbi types I and II), multilocular (Gharbi type III), or degenerate (Gharbi type IV).[13] The presence of irregular linear echogenic structures without acoustic shadowing in the bile duct and/or the dilated biliary tract was accepted as suggestive radiologic findings for a cyst-biliary communication.[14,15]

2.4. Medications

All patients underwent chemotherapy (albendazole) for at least 3 months, beginning from 10 to 15 days before their operations. A single dose of a 2nd generation cephalosporin was used for infection prophylaxis; no other antibiotics were used in patients who did not develop any infective complication during the postoperative period.

2.5. Surgery

Open approach was used in all surgical procedures. Following the incision that allowed the surgeon to visualize the area of interest, gauze compresses embedded in hypertonic saline as the scolicidal agent were used to cover the cysts. A 10–30 ml of cyst fluid was replaced with the same volume of hypertonic saline. Ten to 15 min later, pressure on cyst surface was reduced by aspirating a volume of cyst fluid again. After the cyst was unroofed, all contents were removed and the cavity was then wiped with scolicide-soaked swabs. The cavity was examined carefully for sites of biliary leakage for 5 min. After partial cystectomy, cavities were managed with external drainage, omentopexy, or introflexion, according to the choice of the surgeons. The cavities were drained to control biliary leakage.

2.6. Postoperative period

Drains were removed on the third postoperative day if they provided no biliary drainage was seen. Patients who continued to have biliary drainage in the first 10 days postoperatively were considered to have biliary leakage. Longer biliary drainage was classified as biliary fistula. The fistulas were categorized into low- and high-output types depending on whether the fistula output was less than or greater than 300 mL/day, respectively.

Drains were not removed in patients who developed biliary fistulas and flow rates of these fistulas were recorded daily. Endoscopic sphincterotomy was performed in patients in whom no reduction in biliary flow rate was observed or a spontaneous termination of biliary leakage was unlikely.

2.7. Variables

The data of the patients that were analyzed as potential predictors of occult insidious cysto-biliary communication are age, sex, leukocyte count, liver function test results (alanine aminotransferase [ALT], aspartate aminotransferase [AST], alkaline phosphatase [ALP], γ-glutamyl transpeptidase [GGT], and bilirubin levels), and ultrasonographic cyst features (type, diameter, number, and localization). 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 leukocyte count, $<10{,}000/mm^3$.

2.8. Statistics

Statistical analyses were performed by a statistician who was blinded to the study. All the collected data were analyzed using the SPSS (v.11.0) statistical program (SPSS, Chicago, IL, USA). The results are presented as the mean ± SD and number. 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). A p value less than 0.05 was considered significant. Variables were first subjected to univariate analyses. Those suggesting an association whereby $p < 0.05$ were included in multivariate logistic regression analyses which used to estimate the

factors associated with biliary leakage. Some percentages may not reach total of 100 because of rounding.

3. Results

There were 115 (37.2%) female and 68 (62.8%) male patients; the mean age was 42.3 (range, 15–83) years. Cysts were located in the right hepatic lobe in 148 patients (80.9%), in the left lobe in 30 patients (16.4%), and in both lobes in 5 patients (2.7%). The majority of patients ($n = 131$; 71.6%) had 1 cyst; the remaining 52 patients (28.4%) had multiple cysts. The mean cyst diameter was 8.5 cm (range, 4–16). Cyst diameter was greater than 10 cm in 41 (22.4%) patients. Of 183 dominant cysts, 107 (58.5%) were unilocular. Omentoplasty was performed in 38 (20.8%) following unroofing procedure. Mean hospitalization time for all patients was 5 days (range 3–20). There was no hospital mortality.

3.1. Patients with occult insidious cysto-biliary communications

Postoperative biliary leakage occured in 24 (13.1%) out of 183 patients. Fifteen (62.5%) were female and 9 (37.5%) were male. The mean age was 46 (range, 15–83) years. Nineteen patients (79.2%) had a single cyst. The cysts were localized in the right lobe of the liver in 22 (91.7%) patients and in the left lobe in 2 (8.3%). Of the 24 patients, 8 (33.3%) were unilocular, 13 (54.2%) were multilocular, and 3 (12.5%) were degenerate. The cyst diameter ranged from 7 to 16 cm (mean, 10.9 cm). Omentoplasty was performed in 7 (29.2%). Mean total bilirubin, ALP, GGT, AST and ALT levels were 2.9 mg/dL (range, 0.5–10), 182.2 U/L dL (range, 85–314), 80.1 U/L dL (range, 22–157), 59.4 U/L (range, 10–145) and 50.6 U/L dL (range, 8–100), respectively. The mean white blood cell count was 14,400/mm^3 (range, 6400–22,400). Median fistula output was 263 ± 166.3 mL/day (range, 65–550); the fistula had a low output in 7 patients (29.1%) and high output in 17 patients (70.8%). Biliary leakage in all cases remained as controlled fistula, and none of the cases developed postoperative intra-abdominal biloma, biliary peritonitis or cyst cavity biliary abscess. In all 17 cases, low output fistulas were closed spontaneously in an average of 9.2 days (range, 4–16). In all 7 cases with high output fistulas, the drainage was terminated gradually in an average of 22.6 ± 7.4 days (range, 17–38) following endoscopic sphincterotomy. Average interval between endoscopic sphincterotomy and fistula closure was 10.3 ± 7.3 days (range, 3–26). Average fistula closure time in all 24 cases was 13.1 ± 7.7 days (range, 4–38). Mean hospital stay was found 9.8 ± 4.4 day (range, 5–20).

3.2. Patients without cysto-biliary communications

Of the 159 patients without cysto-biliary communication, 100 (62.9%) were female and 59 (37.1%) were male. The mean age range was 41.7 (range, 15–73) years. A hundred and twelve patients (70.4%) had a single cyst and 47 patients (29.6%) had multiple cysts. The cysts were localized in the right lobe in 126 (79.2%), in the left lobe in 28 (17.6%), and bilobar involvement was seen in 5 (3.1%) of the patients. Of the 159 cysts, 99 (62.3%) were unilocular, 48 (30.2%) were multilocular, and 12 (7.5%) were degenerate. The mean cyst diameter of the 159 patients was 8.1 cm (range, 4–14). Omentoplasty was performed in 31 (19.5%) patients following unroofing procedure. Mean total bilirubin, ALP, GGT, AST and ALT levels were 0.8 mg/dL (range, 0.4–3.6), 102.3 U/L (range, 33–322), 35.8 U/L (range, 8–155), 34.8 U/L (range, 10–98), and 35.8 U/L (range, 8–119), respectively. The mean white blood cell count was 8600/mm^3 (range, 3200–22,000). Mean hospitalization time was 4.2 ± 1 days (range, 3–10). A comparison of various demographic features, clinic factors and laboratory results of patients is presented in Tables 1 and 2.

Table 1
Comparison of characteristic in patients with and without occult insidious cysto-biliary communication.

			Fistula due to occult insidious CBC[a]		
		Patient	Absent (%)	Present (%)	P[b]
Age (year)	Mean	42.3 ± 16.1	41.7 ± 15.3	46 ± 20.8	0.228
	≤60	146 (79.8)	128 (80.5)	18 (75)	0.586
	>60	37 (20.2)	31 (19.5)	6 (25)	
Sex	Female	115 (62.8)	100 (62.9)	15 (62.5)	1.000
	Male	68 (38.2)	59 (37.1)	9 (37.5)	
Number of cyst	Single	131 (71.6)	112 (70.4)	19 (79.2)	0.471
	Multiple	52 (28.4)	47 (29.6)	5 (20.8)	
Size of cyst (cm)	Mean	8.5 ± 2.4	8.1 ± 2.2	10.9 ± 2.5	*0.000*
	<10	142 (77.6)	135 (84.9)	7 (29.2)	*0.000*
	>10	41 (22.4)	24 (15.1)	17 (70.8)	
Localization of cyst	Right Lobe	148 (80.9)	126 (79.2)	22 (91.7)	0.325
	Left Lobe	30 (16.4)	28 (17.6)	2 (8.3)	
	Bilobar	5 (2.7)	5 (3.1)	–	
Type of cyst	Unilocular	107 (58.5)	99 (62.3)	8 (33.3)	*0.027*
	Multilocular	61 (33.3)	48 (30.2)	13 (54.2)	
	Degenerate	15 (8.2)	12 (7.5)	3 (12.5)	
Omentoplasty	Performed	38 (20.8)	31(19.5)	7 (29.2)	0.286
	Not performed	145 (79.2)	128 (80.5)	17 (70.8)	
PHS[c] (day)	Mean	5 ± 2.6	4.2 ± 1	9.8 ± 4.4	*0.000*

[a] Cysto-biliary communications.
[b] Chi-square.
[c] Postoperative Hospital Stay.

3.3. Statistical analysis

In univariate analysis, significant clinical factors associated with the presence of an occult insidious communication were as follows: elevated ALP (>133 U/L), GGT (>50 U/L) and total bilirubin levels (>1.2 mg/dL), high white blood cell count (>10,000/mm^3) and a larger cyst diameter (>10 cm) on ultrasonography. The independent clinical predictors for risk factors of occult insidious cysto-biliary communication in univariate and multivariate analyses are shown in Table 3. Table 4 details the performance of each clinical factor associated with the presence of an occult insidious cysto-biliary communication in univariate analysis.

Table 2
Comparison of biochemical results in patients with and without occult insidious cysto-biliary communication.

			Fistula due to occult insidious CBC		
		Patient	Absent (%)	Present (%)	P[a]
Bilirubin levels	Mean	112.8 ± 60.5	0.8 ± 0.6	2.9 ± 2.8	*0.000*
	Normal	157 (85.8)	148 (93.1)	9 (37.5)	*0.000*
	Abnormal	26 (14.2)	11 (6.9)	15 (62.5)	
ALP levels	Mean	1.1 ± 1.3	102.3 ± 52.7	182.2 ± 63.7	*0.000*
	Normal	139 (76)	134 (84.3)	5 (20.8)	*0.000*
	Abnormal	44 (24)	25 (15.7)	19 (79.2)	
GGT levels	Mean	41.6 ± 31.8	35.8 ± 27.6	80.1 ± 31.7	*0.000*
	Normal	143 (78.1)	139 (87.4)	4 (16.7)	*0.000*
	Abnormal	40 (21.9)	20 (12.6)	20 (83.3)	
AST levels	Mean	38.1 ± 25.4	34.8 ± 20.3	59.4 ± 41.5	*0.000*
	Normal	122 (66.7)	112 (70.4)	10 (41.7)	*0.009*
	Abnormal	61 (33.3)	47 (29.6)	14 (58.3)	
ALT levels	Mean	37.8 ± 22.2	35.8 ± 21.1	50.6 ± 25.4	*0.000*
	Normal	123 (67.2)	114 (71.7)	9 (37.5)	*0.002*
	Abnormal	60 (32.8)	45 (28.3)	15 (62.5)	
Leukocytosis ($\times 10^3/\mu L$)	Mean	9.3 ± 3.9	8.6 ± 3.4	14.4 ± 3.7	*0.000*
	Absent	122 (66.7)	121 (76.1)	1 (4.2)	*0.000*
	Present	61 (33.3)	38 (23.9)	23 (95.8)	

CBC, cysto-biliary communications; ALT, alanine aminotransferase; AST, aspartate aminotransferase; ALP, alkaline phosphatase; and GGT, γ-glutamyl transpeptidase.
[a] Chi-square.

Table 3
Univariate and multivariate analyses for risk factors of fistula development.

	P^a	OR	%95 CI	P^b
Age	0.577			
Sex	0.828			
Number of cyst (one vs. multiple)	0.896			
Size of cyst (>10 cm)	*0.000*	37.7	4.4–320.3	*0.001*
Localization of cyst	0.929			
Type of cyst	0.071			
Total bilirubin >1.2 mg/dL	*0.000*	28.6	2.4–330.7	*0.007*
ALP > 133 U/L	*0.012*	7.4	1.1–51.4	*0.041*
GGT >50 U/L	*0.016*	3.8	0.4–32.1	0.212
AST Levels >37 U/L	0.899			
ALT Levels >40 U/L	0.572			
Leukocytosis (>10,000/mm^3)	*0.000*	64	4.7–856.4	*0.002*
Omentoplasty	0.383			

[a] Univariate analysis.
[b] Multivariate analysis.

4. Discussion

Some liver hydatid cysts may grow average 1–30 mm per year and cause compressive atrophy of surrounding hepatocytes and fibrosis.[14] A cysto-biliary communication due to spontaneous rupture may occur at the point of contact with a biliary duct.[6,14] The criteria for cysto-biliary communication are visualization of a cysto-biliary opening during operation or demonstration of communication by ERCP in patients with hydatid jaundice or biliary leakage after hydatid liver surgery.[5]

The intrabiliary rupture of a hepatic hydatid cyst may occur in two forms: a frank rupture, or an occult rupture.[11] The incidence of frank and occult cysto-biliary communication ranges 5% to 17% and 10% to 37% of cases, respectively.[5,11,14] In this study the overall incidence of cysto-biliary communication was 25% (64/252) (frank, 7.5%; occult, 17.5%). Diagnosis of frank communication is rarely difficult on ultrasound and CT when typical radiological features are present and can be managed by surgical and/or endoscopic procedures.[9,10,14]

A cysto-biliary communication can be suspected preoperatively by aspiration of bile-stained hydatid fluid. We preferred to define this situation as "occult honest cysto-biliary communication". Occult honest cysto-biliary communication rate was 17.9% (20/252) in all patients who were treated in our clinic. If bile-stained hydatid fluid was determined, the most convenient way of reducing the rate of postoperative bile leakage was to detect and block the cysto-biliary communication. Detection of cysto-biliary communications was done by several ways. First, orifices may visualize in the cavities. Second, biliary drainage from the cavity drains in post-operative period indicates cysto-biliary communications. Last, preoperative ERCP and intraoperative findings have shown the cysto-biliary communications in hydatid jaundice cases.

Table 4
Performance of risk factors associated with an occult İnsidious cysto-biliary communication on univariate analysis.

	Sensitivity, %	Specificity, %	PPV, %	NPV, %	LR
Cyst > 10 cm	70	84	41	95	4.6
Total bilirubin > 1.2 mg/dL	62	93	57	76	9.0
ALP > 133 U/L	79	84	43	96	5.0
GGT > 50 U/L	83	87	50	97	6.6
Leukocytosis (>10,000/mm^3)	25	78	17	84	1.1
If these 5 factors positive in a case	33	100	100	90	∞

PPV, positive predictive value; NPV, negative predictive value; LR, likelihood ratio, ALP, alkaline phosphatase; and GGT, γ-glutamyl transpeptidase.

During the operation, the cyst cavity may be filled with saline and air injected from the cystic duct to locate opening of the biliary channel into the cyst cavity.[7] Another method is to pack the cyst with gauze soaked in hypertonic saline and wait for bile stains to appear. We generally prefer this method in our patients. Alternatively, injection of methylene blue into biliary tree or intraoperative cholangiography may also be helpful in detecting cysto-biliary communications. Özmen and Coskun describe an easy technique, using a telescope, for finding the communication via direct visualization during conservative surgery for hydatid liver cysts in situations in which visualization of the cavity is difficult.[16] If biliary opening is not found or safety of ligation is doubtful, decompression of the biliary system through the cystic duct is advocated to prevent bile leakage from occult communications and, therefore, to decrease the risk of infectious complications.[3,7,17,18]

Following the evacuation, the cyst cavity should be drained with closed-suction drains preferably if a bile leak is present.[19,20] However, bile may not be present in the cavity despite cysto-biliary communication. Because biliary system pressure is 15–20 cm H_2O but intracystic pressure is 30–80 cm H_2O and, flow is normally toward the biliary system in these cases.[21,22] If cysto-biliary communication remains undetected or unrepaired, the pressure gradient is reversed. Bile flows into the residual cavity rather than through the biliary tree. Thus, biliary leakage appears following conservative surgery.[4,23] We termed this kind of communication as "occult insidious cysto-biliary communication". This situation is not rare. The rate of occult insidious cysto-biliary communication was 9.5% (24/252) in all patients with liver hydatid disease who were treated in our clinic and 13.1% (24/183) in asymptomatic patients who constituted the study group. In order to prevent postoperative cavity-related complications, it is important to predict whether the liver cysts communicate with the biliary tree in asymptomatic patients or not. Size of cyst,[24] ALP,[7,14,21,24] GGT[14,21,24] and bilirubin[7,14,21] have been reported as risk factors for cysto-biliary communication in literature. An elevated serum ALP level greater than 133 U/L, an elevated total bilirubin level greater than 1.2 mg/dL, white blood cell count more than 10,000/mm^3 and a cyst size larger than 10 cm were the independent risk factors in multivariate analysis, that is, predictors for the presence of an occult insidious cysto-biliary communication in the current study. We found that, positive and negative predictive values were 41% and 95% for cyst size, 57% and 76% for total bilirubin, 43% and 96% for ALP, and 50% and 97% for GGT, respectively. Leukocytosis was poor predictor, with positive predictive value of 17% and negative predictive value of 84%. However, if a combination of these 5 factors is present, 8 cases in our study, the positive and negative predictive values increased to 100% and 90%, respectively. Yalin et al. showed that intracystic pressure increases along with the diameter of a hydatid cyst and leads to a spontaneous intrabiliary rupture.[24] A high intracystic pressure causing intermittent passage of cyst fluid and minor fragments into the biliary system. However, an apparent biliary obstruction does not occur. ALP, GGT and bilirubin levels increase but patients stay asymptomatic.[14,21] Reabsorption of the bile from the cyst cavity may be another mechanism for the increased bilirubin and ALP levels.[14]

According to our results, a clear hydatid fluid without bile in the cyst cavity does not mean an intact cyst wall. A surgeon should suspect an occult insidious cysto-biliary communication of a hydatid cyst if a patient has these risk factors. In this situation, percutaneous drainage and use of scolicidal agents should be avoided because septic complications and risk of sclerosing cholangitis.[24,25] Early management should be planned in these patients, a broad-spectrum antibiotic should be chosen for prophylaxis and suspected patients should be treated surgically as early as possible. If the site of the cysto-biliary communication cannot be identified, methylene blue injection into the common bile duct or a cholangiogram or intraoperative ultrasonography may be helpful to see

the point of communication in patients who have risk for an occult insidious cysto-biliary communication. Because the biliary leakage rate is high, management by external drainage, preferably with suction drainage, is necessary after evacuation of the cyst, even though no active bile leakage is determined.

Although most external biliary fistula close spontaneously, they persist in 4–27.5% of cases.[26] Postoperative biliary fistula should first be treated by conservative methods. If it persists despite all preventive measures, then, endoscopic sphincterotomy is indicated in biliary fistula of more than 3 weeks' duration or with output exceeding 300 mL/d.[7,27]

5. Conclusion

Cysto-biliary communications may be responsible for high recurrence rate of hydatid cysts. If the cyst content is not stained with bile or routine imaging studies do not determinate a cysto-biliary communication in asymptomatic patients, it is difficult to suspect from the connection between the cyst and the bile duct, preoperatively. These occult insidious communications appear with postoperative biliary leakage and its sequelae like bilio-cutaneous fistula, bilomas and bile peritonitis in the postoperative period following conservative surgery. This study demonstrates that occult insidious cysto-biliary communication may be predicted preoperatively by using the predictors that are ALP, total bilirubin, white blood cell count and cyst size.

Conflict of interest
The authors have no conflict of interest.

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References

1. Sciume C, Geraci G, Pisello F, Li Volsi F, Facella T, Modica G. Treatment of complications of hepatic hydatid disease by ERCP: our experience. *Ann Ital Chir* 2004;**5**:531–5.
2. Saritas U, Parlak E, Akoglu M, Sahin B. Effectiveness of endoscopic treatment modalities in complicated hepatic hydatid disease after surgical intervention. *Endoscopy* 2001;**10**:858–63.
3. Agarwal S, Sikora SS, Kumar A, Saxena R, Kapoor VK. Bile leaks following surgery for hepatic hydatid disease. *Indian J Gastroenterol* 2005;**2**:55–8.
4. Sakhri J, Ben Ali A. Hydatid cyst of the liver. *J Chir (Paris)* 2004;**6**:381–9.
5. Kayaalp C, Bostanci B, Yol S, Akoglu M. Distribution of hydatid cysts into the liver with reference to cystobiliary communications and cavity-related complications. *Am J Surg* 2003;**185**:175–9.
6. Hankins JR. Management of complicated hydatid cysts. *Ann Surg* 1963;**158**:1020–34.
7. Demircan O, Baymus M, Seydaoglu G, Akinoglu A, Sakman G. Occult cystobiliary communication presenting as postoperative biliary leakage after hydatid liver surgery: are there significant preoperative clinical predictors? *Can J Surg* 2006;**49**:177–84.
8. Alper A, Ariogul O, Emre A, Uras A, Otken A. Choledochoduodenostomy for intrabiliary rupture of hydatid cysts of liver. *Br J Surg* 1987;**74**:243–5.
9. Bedirli A, Sakrak O, Sozuer EM, Kerek M, Ince O. Surgical management of spontaneous intrabiliary rupture of hydatid liver cysts. *Surg Today* 2002;**32**:594–7.
10. Giouleme O, Nikolaidis N, Zezos P, Budas K, Katsinelos P, Vasiliadis T, et al. Treatment of complications of hepatic hydatid disease by ERCP. *Gastrointest Endosc* 2001;**54**:508–10.
11. Becker K, Frieling T, Saleh A, Häussinger D. Resolution of hydatid liver cyst by spontaneous rupture into the biliary tract. *J Hepatol* 1997;**26**:1408–12.
12. Magistrelli P, Masetti R, Coppola R, Messia A, Nuzzo G, Picciocchi A. Surgical treatment of hydatid disease of the liver. A 20-year experience. *Arch Surg* 1991;**126**:518–23.
13. Gharbi HA, Hassine W, Brauner MW, Dupuch K. Ultrasound examination of the hydatid liver. *Radiology* 1981;**139**:459–63.
14. Atli M, Aydin NK, Yuksek YN, Doganay M, Gozalan U, Kologlu M, et al. Intrabiliary rupture of a hepatic hydatid cyst associated clinical factors and proper management. *Arch Surg* 2001;**136**:1249–55.
15. Zargar SA, Khuroo MS, Khan BA, Dar MY, Alai MS, Koul P. Intrabiliary rupture of hepatic hydatid cyst: sonographic and cholangiographic appearances. *Gastrointest Radiol* 1992;**17**:41–5.
16. Özmen M, Coskun F. New technique for finding the ruptured bile duct into the liver cyst: scope in the cave technique. *Surg Laparosc Endosc Percutan Tech* 2002;**12**:187–9.
17. Erzurumlu K, Dervisoglu A, Polat C, Senyurek G, Yetim İ, Hokelek M. Intrabiliary rupture: an algorithm in the treatment of controversial complication of hepatic hydatidosis. *World J Gastroenterol* 2005;**11**:2472–6.
18. Vagioanos C, Androulakis JA. Capsectomy and drainage in hepatic hydatidosis. *Dig Surg* 1997;**14**:241–4.
19. Erdem E, Nessar M, Sungurtekin U, Ozden A, Tetik C. The management of hepatic hydatid cyst: review of 94 cases. *J Hepatobiliary Pancreat Surg* 1998;**5**:179–83.
20. Kama NA, Sahin M, Gocmen E, Bayrak M, Kulacoglu H, Akat AZ. The results of surgical techniques in hepatic hydatidosis: treatment with drainage versus treatment without drainage – a 6-year experience. *J R Coll Surg Edinb* 1998;**43**:254–6.
21. Ozaslan E, Bayraktar Y. Endoscopic therapy in the management of hepatobiliary hydatid disease. *J Clin Gastroenterol* 2002;**35**:160–74.
22. Yalin R, Aktan AO, Yeğen C, Döşlüoğlu HH. Significance of intracystic pressure in abdominal hydatid disease. *Br J Surg* 1992;**79**:1182–3.
23. Vignote ML, Mino G, de la Mata M, de Dios JF, Gomez F. Endoscopic sphincterotomy in hepatic hydatid disease open to the biliary tree. *Br J Surg* 1990;**77**:30–1.
24. Kayaalp C, Bzeizi K, Demirbag AE, Akoglu M. Biliary complications after hydatid liver surgery. Incidence and risk factors. *J Gastrointest Surg* 2002;**6**:706–12.
25. Bret PM, Fond A, Bretagnolle M, Valette PJ, Thiesse P, Lambert R, et al. Percutaneous aspiration and drainage of hydatid cysts in the liver. *Radiology* 1988;**168**:617–20.
26. Dolay K, Akçakaya A, Soybir G, Cabioğlu N, Müslümanoğlu M, Iğci A, et al. Endoscopic sphincterotomy in the management of postoperative biliary fistula. A complication of hepatic hydatid disease. *Surg Endosc* 2002;**16**:985–8.
27. Skroubis G, Vagianos C, Polydorou A, Tzoracoleftherakis E, Androulakis J. Significance of bile leaks complicating conservative surgery for liver hydatidosis. *World J Surg* 2002;**26**:704–8.