

RISK FACTORS AND SURGICAL SOLUTIONS OF COMPLICATED LIVER HYDATID CYSTS

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(ABSTRACT): Aim of this study was to evaluate the predisposing factors for peritoneal perforation and intrabiliary rupture and the effects of these complications on surgical outcome in liver hydatid disease.

Material and Methods: A total of 254 patients with liver hydatid cysts who had undergone surgical treatment were evaluated retrospectively. Twelve patients with peritoneal perforation, 43 patients with spontaneous intrabiliary perforation, and 199 patients with noncomplicated hydatid cysts were treated in our clinics. **Results:** When the predisposing factors for complications were evaluated, younger age, superficial position, and larger cyst dimensions ($P < 0.05$) increased peritoneal perforation rates. It was shown that older age increased cyst dimensions, and presence of multiple and bilobar cysts increased intrabiliary rupture rates ($P < 0.05$). Partial pericystectomy and drainage was the most frequent surgical procedure in all groups (82.7%). The incidence of post-operative complications in the peritoneal perforated group, in the intrabiliary ruptured group, and in the noncomplicated group was 27.6%, 18.3% and 4.7%, respectively. When length of hospital stay was compared, there was no significant difference between the groups ($P > 0.05$). The overall recurrence rate was 4.3% (11 patients), but there was not any statistical difference among the patient groups ($P = 0.13$). **Conclusion:** In peritoneally perforated and intrabiliary ruptured cases, the most important steps are irrigation of the peritoneal cavity and clearance of the cystic material from the biliary tree.

KEYWORDS: COMPLICATED LIVER HYDATID CYSTS, PREDISPOSING FACTORS, SURGICAL TREATMENT, SURGICAL OUTCOME.

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INTRODUCTION

Echinococcosis is a near-cosmopolitan zoonosis caused by adult or larval stage of tapeworms (cestodes) belonging to the genus *Echinococcus* (family Taeniidae). Both sheep and humans are intermediate hosts [1].

The two major species of medical and public health importance are *Echinococcus granulosus* and *Echinococcus multilocularis*, which cause cystic echinococcosis and alveolar echinococcosis, respectively. These are both serious life-threatening diseases, the latter especially so, with a high fatality rate and poor prognosis without careful clinical management.

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In humans, 50% to 75% of hydatid cysts occur in the liver, 25% are found in the lungs, and 5% to 10% are distributed along the arterial system [2]. The clinical picture can be severe, and complications may occur, making the already difficult treatment even more so [3-5].

Complications are observed in one third of patients with liver hydatid cysts. Without treatment, cysts grow and eventually may form fistulas into the peritoneal cavity or intrabiliary rupture, requiring emergency surgery [2-5]. Operative treatments vary from complete resection to minimal invasive procedures, but the ideal treatment is still controversial [6,7]. The choice of therapy depends on several factors: number and localization of the cysts, surgeon expertise, and presence of complications.

Surgery is the mainstay of treatment, although there is no consensus on the respective advantages of conservative and radical methods. Besides, in the treatment of peritoneal perforated cases, profuse peritoneal lavage with hypertonic solutions appears to be more necessary; in the setting of intrabiliary rupture, T-tube drainage, sphincteroplasty, and choledochoduodenostomy are appropriate treatment strategies to reduce the pressure in the biliary tree. In this study, we aimed to evaluate the predisposing factors for peritoneal perforation and intrabiliary rupture and the effects of these complications on morbidity, length of hospital stay (LOS), mortality, and recurrence in hydatid disease.

MATERIALS AND METHODS

We retrospectively evaluated medical records of 254 patients with liver hydatid cysts who were admitted to the Department of General Surgery of “Sf. Ioan” Emergency Hospital and I Surgery Clinic of “Sf. Spiridon”, Medicine and Pharmacy University “Gr. T Popa”, Iasi, Romania, between February 2004 and December 2010. In this period, 12 patients with peritoneal perforation (group I), 43 patients with spontaneous intrabiliary perforation (group II), and 199 patients with noncomplicated hydatid cysts (group III) were treated in our clinics. The ratios of intrabiliary rupture and peritoneal perforation cases to noncomplicated cases were 21% and 6%, respectively. From the point of view of surgical technique adopted, there were 215 patients with laparotomy, 27 by celioscopic technique, and 12 conversions to laparotomy. Patient age and sex, initial complaints, physical findings, cyst characteristics, imaging results, surgical procedures, reasons for peritoneal perforation and intrabiliary rupture, morbidity, LOS, recurrence rates, and mortality were evaluated. The patients with extrahepatic organ involvement were excluded.

The preoperative evaluation included blood tests (complete blood count, blood type, liver function tests), chest radiography, abdominal ultrasonography (US) or computed tomography (CT). Chest and abdominal radiography and abdominal US were performed in all patients at admission. The most important indications for CT were a need for additional anatomical and cystic details, the presence of multiple hydatid cysts, findings of a solid appearance, and findings of cyst infection.

In the cases of liver hydatid cysts operated by classical technique, the area around the cyst was covered with packs soaked in 3% hypertonic saline solution to prevent the further spread of the parasite during evacuation of the cyst. Cyst contents were aspirated and the germinative membrane and daughter cysts were removed with forceps or spoons. With the roof excision of the redundant part of the cyst, an excellent exposure was obtained. Any orifices of bile ducts observed on the inner surface of the cavity were sutured with nonabsorbable sutures.

Next, a surgical procedure, such as partial pericystectomy (PP) and capitonnage, PP and omentoplasty, or PP and drainage or liver resections, was performed. All the complicated cysts was operated initially or converted to “open” technique.

Since 1993, medical treatment for hydatidosis has consisted of albendazole 10 mg/kg per day, beginning after surgery and continuing for 3 months. Over the last 5 years, the drug has been administered continuously with monthly blood liver function tests and complete blood count controls.

During follow-up, US were performed twice a year for 2 years, and then annually. CT scans were performed as needed.

Differences among categorical variables were compared using χ^2 tests, and Kruskal-Wallis tests were used for age, diameter, and number of cysts. Significance was set at $p < 0.05$.

RESULTS

The mean age (mean \pm SD) of peritoneal perforated and intrabiliary ruptured patients was 36.32 ± 11.86 and 50.26 ± 13.88 , respectively. The mean age factor in the peritoneal perforation group was significantly different from others ($p = 0.001$). The sex ratio of the patients was not statistically different among the groups ($p = 0.95$), (Table 1).

Table 1
The repartition of patients according to sex, age and number and position of cysts

Characteristic	Group I (n = 12)	Group II (n = 43)	Group III (n = 199)	P
Sex				
Male	6 (50)	23 (53)	112 (56)	0.950
Female	6 (50)	20 (47)	87 (44)	
Age (yr)				
mean \pm SD	36.32 ± 11.86	50.26 ± 13.88	45.62 ± 16.01	0.001
Median (range)	37.0 (17-76)	53.0 (23-76)	43.0 (18-79)	
Previous hydatid disease surgery				
No	10 (83)	34 (79)	173 (87)	0.204
Yes	2 (17)	9 (21)	26 (13)	
Position				
Superficial	10 (83)	24 (56)	122 (61)	0.017
Deep	2 (17)	19 (44)	77 (39)	
Cyst diameter (cm)	8 (5-21)	9 (6-26)	7 (4-29)	0.001
Localization				
Right lobe	8 (66,6)	25 (58)	141 (71)	0.109
Left lobe	2 (16,6)	11 (25)	38 (19)	
Bilateral	2 (16,6)	7 (17)	20 (10)	
No. of cysts				
1	9 (75)	28 (66)	153 (77)	0.284
2	2 (16,6)	8 (18)	31 (15)	
3	1 (8,3)	4 (9)	12 (6)	
4	-	3 (7)	3 (2)	
Cyst infection				
Negative	11 (91,7)	41 (95)	185 (93)	0.683
Positive	1 (8,3)	2 (5)	14 (7)	

Previous hydatid disease surgery, cyst localization, number of cysts, and presence of infection were not statistically different among groups ($P > 0.5$; range, 0.109-0.683), whereas the differences in cyst diameters and positions (superficial or deep) were significant ($P = 0.001$ and 0.017 , respectively).

The most common complaint was abdominal pain in all groups (63%). Other common complaints were nausea, vomiting, abdominal distension, and allergenic reactions in peritoneally perforated cases, jaundice (26%) in intrabiliary ruptured cases, and abdominal swelling (37%) in noncomplicated hydatid cysts.

The most common physical examination findings were abdominal sensitivity and guarding (100%), acute abdomen findings (42%), distension (27%) in peritoneally perforated cases, hepatomegaly (34%) and jaundice (27%) in intrabiliary ruptured cysts, and finally, hepatomegaly (23%) in noncomplicated patients. In 53 (20%) patients, diagnosis was made incidentally by abdominal US or CT during a medical checkup or after a trauma.

When the predisposing factors for complications were evaluated, younger age, superficial position, and larger cyst dimensions increased peritoneal perforation rates. It has been seen that older age, increased cyst dimensions, and presence of multiple and bilobar cysts increases intrabiliary rupture rates.

Abdominal US were done for all patients and demonstrated hepatic cysts with 96.3% sensitivity CT was used as the second most frequent ($n = 148$, 46%) diagnostic modality, with 96.5% sensitivity.

MRI cholangiography was performed after 1996 to assess the spread of disease to the biliary tree but was not used routinely and was done in 37 patients (9.9%). The indications for MRI cholangiography are the presence of jaundice seen during the physical examination, elevation of serum bilirubin and alkaline phosphatase levels, and other obstructive jaundice symptoms and demonstrated 98% sensitivity.

Partial pericystectomy and drainage was the most frequent surgical procedure in all groups. In all intrabiliary rupture cases, cholecystectomy and common bile duct exploration were performed. Cyst remnants and daughter vesicles in the bile ducts were evacuated with Dormia forceps or a Fogarty catheter in 5 (27.6%) patients. A T-tube drainage was performed in 31 (72%) patients, choledochoduodenostomy was performed in 7 (16.1%) patients, and sphincteroplasty was performed in 3 (6.4%) patients.

The most frequent postoperative complications were wound infection (7.3%) and pulmonary complications (3.6%). In the peritoneal perforation group, 10 surgical complications occurred in 4 (25%) patients; in the intrabiliary rupture group, 18 complications occurred in 7 (16.2%) patients; and in the noncomplicated group, 16 complications occurred in 14 (5.5%) patients (Table 2).

Table 2
Morbidity, recurrence, and mortality rates of the three study groups n (%)

	Group I (n = 12)	Group II (n = 43)	Group III (n = 199)	P
		Medical complications		
Cardiac	1 (8.3)	1 (2.3)	3 (1.5)	0.163
Respiratory	2 (16.6)	3 (6.9)	6 (3)	
Other	1 (8.3)	2 (4.6)	4 (2)	
		Surgical complications		
Wound infection	2 (16.6)	3 (6.9)	5 (3.5)	0.001
Biliary fistula	1 (8.3)	5 (11.6)	1 (0.5)	0.001
Intra-abdominal abscess	-	1 (2.3)	2 (1)	0.855
Incisional hernia	2 (16.6)	2 (4.6)	3 (1.2)	0.087
Recurrence	3 (25)	5 (11.6)	8 (4)	0.132
Mortality	-	1 (2.3)	3 (1.2)	0.844

DISCUSSION

Peritoneal perforation into the abdominal cavity and the spontaneous intrabiliary rupture of liver hydatid disease are not rare complications and cause serious problems. In our study, the ratios of peritoneal perforation and intrabiliary rupture cases to noncomplicated cases were 6% and 21.6%, respectively. The most common complication is rupture of the cyst, either internally or externally, followed by anaphylactic reaction and jaundice [2,4, 5-10]. Systemic anaphylactic reactions have been reported in 1% to 18.3% of patients with intraperitoneal perforation, and these reactions may be life threatening [5,11,12]. Jaundice is the most important sign of intrabiliary rupture within a range of 8% to 34% [13,14]. The presence of complications must be considered in the surgical treatment.

One third of 254 patients who underwent an operation for liver hydatid disease presented with complications. When compared, but the mean age in the peritoneal perforated group was significantly different ($p = 0.001$, Table 1).

Trauma is the most frequent etiological cause of perforation [7,14]. In superficial and bigger cysts, lack of normal liver tissue around the cyst to protect against trauma may be a cause of frequent rupture.

Besides age, cyst diameter and cyst position (superficial or deep) were also significantly different factors ($p = 0.001$ and 0.017 , respectively). Similarly, an increase in cyst diameter may increase perforation risk either by becoming more superficial during growth or by increasing intracystic pressure.

Cyst dimension and age (younger age for peritoneal perforation and older age for intrabiliary rupture) have been determined as significant predisposing factors for the two complications.

Recently, some authors have favoured the use of pericystectomy and liver resections because complete surgical resection is the ideal treatment for hydatid disease [11]. In our study, liver resections were performed only if multiple cysts were localized in one lobe, located peripherally, and were pedunculated, and pericystectomy was performed if the cysts were away from the major vascular and biliary structures of the liver. The need for sufficient technical infrastructure and surgical experience in the field of hepatic surgery limit these treatments in small centers, especially in developing countries. Relatively small-sized subcapsular cysts can be managed by nonanatomic resection of the cysts with a rim of healthy hepatic tissue but routine application of pericystectomy and liver resection may increase the operative complications, such as bleeding and postoperative morbidity and mortality.

In the literature, reported complication rates are between 6% and 47%; recurrence rates are 8% to 15% [15]. In this study, the overall postoperative complication and recurrence rates were 18% and 3.8%, respectively. Most often, surgical complications were wound infection ($n = 8$) and biliary fistulas ($n = 7$). External biliary fistulas developed in 15 (5.9%) patients. Endoscopic procedures have been used not only for diagnostic purposes, but for treatment as well. Several authors have advocated pre-operative or post-operative ERCP [16-18]. If performed in conjunction with a sphincterotomy, an ERCP may be used to clear the biliary tree as a planned procedure, thereby avoiding the need for an intraoperative cholangiography and bile duct exploration.

Recurrence occurred in 14 nonperforation cases (3.8%). Recurrence rates were not significantly higher in peritoneal perforation and intrabiliary rupture cases when compared with noncomplicated cases (Table 2).

Recurrences of hydatid disease are usually due to inadequate cystic content removal, spillage of cystic liquid intraoperatively, undetected cysts, and satellite lesions and reinfestations. Albendazole treatment is indicated in patients who are at high risk for surgery, and in the presence of bilateral multiple small cysts. The importance of medical treatment which is mainly effective on small cysts may indicate the usefulness of the effect on recurrence.

Pre-operative therapy may reduce the risk of intraperitoneal seeding of infection that develops *via* cyst rupture and spillage occurring spontaneously or during surgery.

Mortality rates of the study groups were not statistically different ($p = 0.814$, Table 2). When compared, mortality rates were significantly different among groups ($p < 0.001$). This result requires discussion of the controversial radical methods in hydatid disease.

CONCLUSIONS

According to this study, older age, multiple and larger cysts and bilobar localization are established as predisposing factors for intrabiliary rupture. On the other hand, peritoneal perforation risk increases with younger age, superficial localization and larger dimensions of cysts

Morbidity rates significantly increased in peritoneally perforated and intrabiliary ruptured cases, but there was no difference in recurrence rates and hospital stay times.

The most used surgical technique for liver hydatid cysts is partial pericystectomy and drainage because the rate of postoperative complications, recurrence, and the duration of hospital stay are satisfactory.

We believe that radical methods should not be performed routinely for such a benign pathology. However, randomized controlled studies are needed to establish the best surgical management for peritoneal perforated and intrabiliary ruptured liver hydatid cysts.

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