

Original Article

Surgical approach to giant hepatic hydatid cysts: a single-center experience from Istanbul

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Abstract: Background: The aim of this study was to determine the presence of cystobiliary communication (CBC) and to assess the treatment and follow-up of existing and postoperative fistulae in giant hepatic hydatid cysts managed by either conservative or radical surgery. Methods: A total of 40 patients with hepatic hydatid cysts of 10 cm or greater in diameter were examined. These patients were seen in the General Surgery Clinic between January 2005 and June 2015. Conservative and radical surgeries were performed in 25 (62.5%) and 15 patients (37.5%), respectively. In patients that underwent conservative surgery, perioperative cholangiography was performed following pericystectomy and cholecystectomy by passing a 6F nelaton catheter through the cystic duct. Presence of CBCs and daughter vesicles was assessed. Results: Perioperative fistula was detected in 15 patients (60%) in the conservatively managed group. Successful closure with primary suturing was achieved in 14 patients (93%). One patient (7%) developed a postoperative biliary fistula (PBF). Of the 10 cases that were free of fistulae perioperatively, 4 (16%) developed PBF. True positivity and false negativity rates for detecting perioperative fistulae were 84% and 16%, respectively. Choledochoduodenotomies with T-tube drainage were performed in 2 out of 5 patients in which daughter vesicles were detected in the biliary tree. Duodenotomies with sphincteroplasty were performed in the remaining 3. Conclusion: Cholecystectomy with partial cystectomy and perioperative visualization of the biliary tree is a safe and useful procedure for management of giant hepatic hydatid cysts.

Keywords: Giant hepatic hydatid cyst, biliary fistula, surgical treatment

Introduction

Hydatid disease is a worldwide parasitic disease. Although hydatid cysts can grow in any organ of the human body, they most commonly involve the liver. In fact, 75% of all hydatid cysts are found there [1]. Entry of the offending parasite into the body via the gastrointestinal tract and its passage primarily to the liver through the portal vein explain why the liver is the most commonly involved organ.

Management of hepatic hydatid cysts remains a topic of debate. Three main types of treatment modalities are available, medical therapy, conservative surgery, and radical surgery. Surgical management is the most commonly utilized therapy in parts of the world where the disease is endemic [2]. It includes a wide array of interventions, ranging from radical surgeries, such as hepatic resection and cystopericystec-

tomy, to more conservative methods, including removal of cystic material and capsulorrhaphy or external drainage [3, 4]. Hydatid cyst disease commonly leads to cystobiliary communication (CBC) or intrabiliary rupture [5]. Although not well established, incidence of CBC in clinical practice may range between 6.6 and 26% [6]. Several studies have even reported rates exceeding 80% [7-9]. CBC may have variable clinical presentations and may be predicted by the size of the communication [10-12].

CBC may sometimes be diagnosed by the presence of bile in the cyst cavity or bile duct containing cystic material (occult honest CBC). When CBC cannot be localized and sutured, appropriate measures should be taken intraoperatively to control bile leakage. Even endoscopic methods have been found to be beneficial. However, when the presence of bile in the cyst cavity or cystic material remains undetect-

ed in the bile duct, when impending rupture is silent (occult insidious CBC), it is difficult to control biliary complications during conservative surgery. This makes accurate prediction vitally important [13]. The main reason for the procedure used in this study was to avoid complications of postoperative biliary fistula (PBF). The aim of the present study was to assess the reliability of the procedure used to detect CBC and/or treatment of existing CBCs caused by giant hepatic hydatid cysts managed by conservative and/or radical surgery.

Methods

Patients

This study retrospectively screened medical records of 385 patients diagnosed with and operated for hydatid cysts at Istanbul Haydarpaşa Numune Training and Research Hospital, between January 2005 and June 2015. A total of 40 patients (10.4%) with hydatid cysts, with sizes of 10 cm or larger, were enrolled. All patients provided informed consent prior to enrollment into the study. The hospital is a reference center for hepatobiliary surgery in its region. Patients were divided into two groups. Group I consisted of 25 patients (62.5%) that were managed conservatively. All patients in this group underwent cholecystectomies, on top of additional procedures, as follows. Twenty patients had additional partial cystectomies (PC), 2 had PC and choledocotomies with a T-tube drainage, and 3 had PC and duodenotomies with sphincteroplasty. Choledocotomies and duodenotomies were performed in cases where the cysts opened into bile ducts, but cystic material could not be detected preoperatively. In Group II, radical surgery was conducted in the form of left lateral segmentectomy in 11 out of 15 patients (37.5%), along with pericystectomies in the remaining 4 patients. Patients with recurrent disease and concurrent non-hepatic organ involvement underwent sphincterotomies with preoperative endoscopic retrograde cholangiography (ERCP). Those with alveolar hydatid cysts were excluded. All procedures were carried out by the same surgical team. This study received approval from the İstanbul Haydarpaşa Numune Training and Research Hospital Ethics Committee.

Diagnosis

Patients were assessed based on age, sex, preoperative findings, cyst size, laboratory results,

imaging results, surgical procedures, intrabiliary rupture, PBF, duration of hospital stay, rates of follow-up and recurrences, and mortality and morbidity rates.

Preoperative evaluation

Complete blood counts, liver function test results, hemagglutinin antibody levels, chest radiography findings, abdominal ultrasonography (USG) findings, and whole abdomen computerized tomography (CT) findings were obtained preoperatively. CT was used to assess cyst size, localization, and possible compressive effects. Intraoperative ultrasonography was used as a guiding tool in hepatic resections. All patients received albendazole at 10 mg/kg/day (Andazol, Biofarma, Istanbul, Turkey) for 1 month preoperatively.

Surgical procedure

For conservative surgical management of giant hepatic hydatid cysts, they first covered the pericystic and operative fields with gauze soaked in 20% hypertonic saline to prevent contamination with cystic fluid. Afterward, PC was performed. After evacuating the cyst cavity (Figure S1), the area was cleansed with hypertonic saline-soaked gauze and complete cholecystectomy was performed using the antegrade approach. Cholangiography was performed to visualize the biliary tree, with the help of a 6F nelaton catheter advanced through the cystic canal. This was done to determine the presence of any CBC or daughter vesicles within the biliary tree (Figure S2). In cases where daughter vesicles were detected, a choledocotomy with T-tube drainage or a duodenotomy with sphincteroplasty was performed. In cases where only a fistula but no daughter vesicles were detected, the distal common bile duct was clamped and a large amount of isotonic saline was administered through a nelaton catheter. Oozing of the saline from the cystic bed helped to visualize the fistula orifice. The orifice was then sutured with 3/0 or 4/0 prolene.

For radical surgery, the surrounding tissues were again similarly covered with gauze soaked in 20% hypertonic saline prior to the procedure. Removal of the cyst en bloc together with a 1-cm intact hepatic parenchyma was performed, without opening the cavity. Liver resection was preferentially performed for cysts that were peripherally located or involved an entire

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Table 1. Demographic properties of the study groups

		Groups			P
		Total (n=40)	Conservative Surgery (n=25)	Radical Surgery (n=15)	
Age	Mean ± SD	45.30±14.74	43.16±17.01	48.86±13.13	0.331 ^a
	Min-Max (Median)	20-74 (45)	20-74 (44)	34-72 (46)	
Sex	Male	11 (27.5)	6 (24.0)	5 (33.5)	0.716 ^b
	Female	29 (72.5)	19 (76.0)	10 (66.7)	
Cyst Size	Mean ± SD	112.80±24.20	115.20±23.42	108.80±25.76	0.238 ^a
	Min-Max (Median)	100-200 (100)	100-180 (100)	100-200 (100)	
Localization	Right	24 (60.0)	20 (80.0)	4 (26.7)	0.001 ^{c,**}
	Left	16 (40.0)	5 (20.0)	11 (73.3)	
Echinococcal serology	Negative	7 (17.5)	7 (28.0)	0	0.033 ^{b,*}
	Positive	33 (82.5)	18 (72.0)	15 (100.0)	
Laboratory (AST-ALT-ALP-GGT)	Normal	27 (67.5)	15 (60.0)	12 (80.0)	0.298 ^b
	High	13 (32.5)	10 (40.0)	3 (20.0)	
Duration of hospital stay	Mean ± SD	11.82±8.44	12.56±9.60	10.60±6.16	0.978 ^a
	Min-Max (Median)	5-45 (8)	5-45 (8)	5-28 (8)	
Duration of follow-up	Mean ± SD	5.55±2.20	5.28±2.11	6.0±2.36	0.504 ^a
	Min-Max (Median)	1-11 (6)	1-9 (6)	2-11 (6)	

^aMann-Whitney U-Test, ^bFisher's Exact Test, ^cPearson's Chi-Squared Test, **P<0.01, *P<0.05.

lobe. Moreover, 1-2 soft drains were placed within the operative field.

Postoperative follow-up and medical treatment

At one-month post-procedure, physical examinations, repeat CT (to check for recurrence), liver function tests, complete blood counts, and indirect hemagglutination (IHA) tests were obtained for each patient. This was followed by abdominal USG and CT examinations with liver function tests, complete blood counts, and IHA test monitoring every six months in the first 2 years after the procedure. Two years later, abdominal CT and USG were performed, as well as AST, ALT, and bilirubin, during follow-ups. Albendazole at 10 mg/kg/day was initiated one month prior to the procedure, continuing for 6 months postoperatively.

Results

The present study involved 40 patients with hepatic hydatid cysts larger than 10 cm. Of these, 11 patients (27.5%) were male and 29 patients (72.5%) were female. The age range was 20-74 years, with a mean age of 45.30±15.74 years. Twenty-five patients (62.5%) underwent conservative surgery (Group I), while 15 patients (37.5%) underwent radical surgery (Group II). Of those in Group I (all underwent

cholecystectomy), 20 patients (80%) underwent additional PC, while 2 and 3 patients (a combined total of 20%) underwent additional PC and choledocotomies with T-tube drainage and PC and duodenotomies with sphincteroplasty, respectively (**Table 1**). No significant differences were detected between Group I and Group II with respect to age, sex, cyst size, laboratory results, duration of hospital stay, and follow-up (P>0.05). There was, however, a significant difference in cyst localization between both groups (P=0.001). This demonstrated that cyst localization significantly predicted the surgical procedure to be performed. Conservative surgery was more commonly selected for cysts localized to the right lobe, in contrast to radical surgery (left lobe). A total of 9 patients (22.5%) suffered complications, while the other 31 patients (77.5%) remained well.

Of the patients developing complications in Group I, one patient (16.7%) suffered hemorrhaging from the hydatid cyst bed, while 5 patients (83.3%) developed PBF. Hepatic artery injuries or wound infections were not observed in this group.

Of the patients developing complications in Group II, one patient (33.3%) suffered hepatic artery injury, while 3 patients (66.7%) developed wound infections. No biloma, hemorrhage,

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Table 2. Distribution of postoperative complications among study groups

		Total (n=40)	Conservative Surgery (n=6)	Radical Surgery (n=3)
Complication	No	31 (77.5)	19 (76.0)	12 (80.0)
	Yes	9 (22.5)	6 (24.0)	3 (20.0)
Hemorrhage		1 (11.1)	1 (16.7)	0
Hepatic Artery injury		1 (11.1)	0	1 (33.3)
Postoperative Biliary Fistula		5 (55.5)	5 (83.3)	0
Wound infection		2 (22.2)	0	2 (66.7)

ge, or biliary fistula was detected in this group (Table 2).

Statistical analysis

Descriptive data (mean, standard deviation, median, frequency, and percentage) was used to describe the study data. Mann Whitney-U test was used for comparison of variables showing normal distribution. Qualitative data was compared across groups using the Chi-Squared test and Fisher Freeman Halton test. Results of statistical analyses are presented in a confidence interval of 95%. A *P* value of <0.05 indicates statistical significance. The Number Cruncher Statistical System 2007 software package (NCSS, LLC Kaysville, Utah, USA) was used for all statistical analyses.

Discussion

Some parts of the world still harbor endemic hepatic hydatid disease, with various studies reporting an incidence of 2/1,000,000 to 1/2,000 in Turkey [14-16].

Surgery has been regarded as the gold standard. Patients with cysts larger than 5 cm are considered good surgical candidates, regardless of their symptoms [17]. However, the best surgical technique remains a topic of debate [18-20]. The present study included 40 patients that underwent surgery for hepatic hydatid cyst disease larger than 10 cm.

Surgical treatment aims to eliminate infectious components, prevent contamination (spillage), kill all viable parasitic elements (endocysts), and close any residual cystic cavity. Some experts advocate conservative surgery, while others recommend the pursuit of more radical techniques [21-27]. The latter comprise hepatic

resection total pericystectomies, which consist of cyst wall excision, either totally or partially.

Radical surgery should be performed by surgeons experienced in hepatobiliary surgery. It is more feasible in hydatid cysts with left lobe lateral segment or anterior superior localization, as well as those that are exophytic

and 5 cm or larger [28]. Radical surgery was applied in 15 patients. Cysts were found in the left lobe and left lateral segment in 11 out of 15 patients (73.3%). One patient had hepatic artery invasion, while another had portal vein compression.

Major vessel compression or a close anatomical relationship with cystic walls makes partial pericystectomy a reasonable approach. Conservative surgery has been shown to be more appropriate for deeply located cysts ≥ 10 cm, multiple cysts, bilobar cysts, and cysts with posterior segment localization in the liver [28]. Since these techniques are safer and simpler, they are preferable to radical procedures [21, 29]. Conservative surgery was performed in 25 patients. Of these, 20 patients (80%) had cysts with right lobar localization, close to the main bile ducts, hepatic artery, inferior vena cava, and portal veins. Four patients had intrahepatic bile duct dilatation due to main bile duct compression, while 5 patients had signs of portal vein and inferior vena cava compression.

Several studies have demonstrated that radical surgery increases mortality and morbidity, while others have reported the same for conservative surgery [30, 31]. On the other hand, Daali et al. found no significant differences between the two techniques regarding mortality and morbidity rates [32]. The present study found statistically similar morbidity rates of 20% and 24% for radical and conservative surgery, respectively. Both groups had a mortality rate of zero. No significant differences were found between the two groups in terms of duration of hospital stay.

The most common complication of hepatic hydatid disease is rupture into the bile ducts. In some clinical series, the rate is as high as 42%

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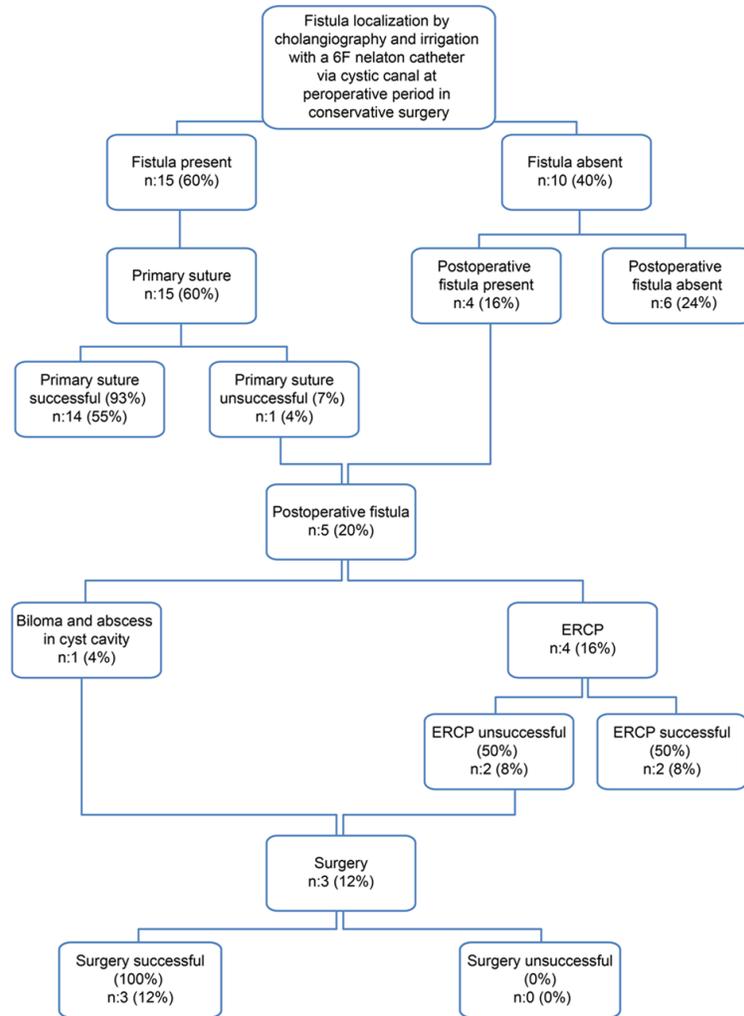


Figure 1. Fistula management flow diagram in the conservative surgery group.

[33-38]. Kilic et al. reported that intraoperative bile leakage and PBF occur more commonly when a cyst diameter is greater than 7.5 cm [39]. In a study by Topcu et al., 22.2% of patients had cysts with a diameter of greater than 10 cm that showed biliary system involvement [40]. Akbulut et al. reported that 67% of cysts (6 out of 9) were associated with CBCs larger than 15 cm and located in the right lobe [28]. The present study demonstrated that 20 out of 25 patients (80%) that underwent conservative surgery had CBCs. Of these fistulae, 15 (75%) were detected at preoperatively and 5 (25%) postoperatively (**Figure 1**). Furthermore, 80% of the cysts were found in the right lobe. Of patients that underwent radical surgery, no biliary fistula developed postoperatively in association with the surgical technique

used (a safe surgical procedure was conducted by constantly performing binding during liver resection). In this group, 73.3% of all cysts were found in the left lobe. The CBC rate of the whole surgical population in this study was 48%.

Several surgical techniques have been introduced for intraoperative control of CBCs, although 10-32% of cases eventually develop a PBF [41]. Zeybek et al. detected a PBF rate of 28% despite having localized CBC orifices and placement of primary sutures perioperatively [42]. A fistula localization study was conducted using cholangiography via cystic canalization in the 25 patients that underwent conservative surgery. In the 15 patients with a CBC, the fistula orifice was localized by administering a large amount of isotonic saline through a 6F nelaton catheter, primarily suturing it with 3/0 and 4/0 prolene sutures. Only one of these patients (7%) developed a PBF. However, PBFs occurred in 4 out of 10 patients in which no fistulas were detected perioperatively. True positivity and false negativity rates were 84% and 16%, respectively.

The biliary system should be assessed with perioperative cholangiography or choledoscopy in every case of CBC [42]. The most effective surgical procedure after elimination of the hydatid material from the biliary tree remains controversial. While some authors advocate the use of the T-tube [43-45], others have reported reduced morbidity and mortality with wide choledochoduodenostomy [46]. Akçan et al. reported complication rates of 21% and 27% in patients with bilioenteric anastomosis and T-tubes, respectively [34]. In contrast to other studies, Akbulut et al. [28] advocated that larger hydatid cysts did not rupture into the biliary system. The present study found daughter

cysts within the bile ducts by cholangiography in 5 patients. Choledocotomies with T-tube drainage were performed in 2 patients, while duodenotomies with sphincteroplasty were performed in the remaining 3 patients. In this study, 12.5% of hydatid cysts that were equal to or larger than 10 cm ruptured into the biliary canal. However, no complications occurred with either of the interventional techniques.

Previous studies have reported that 6-47% of cases are complicated [47]. The most common complications are wound infections and biliary fistulae [48-51]. Daali M et al. reported that the rates of wound infection and biliary fistulae are 26% and 14.49%, respectively [32]. In the present study, the most common complication was PBF (55.5%), followed by wound infections (22.2%). While there were no significant differences between the groups in terms of complications, the conservative surgery group developed more PBFs (67.7%). Additionally, wound infections only occurred in the radical surgery group (67.7%). Zeybek et al. reported that fistulae spontaneously closed in 12 patients (26%) [42]. In the current study, the fistulae were of high output and showed no signs of closure or reduced output, even after 1 week. Endoscopic procedures are used in PBF treatment [34] and are recommended when no signs of reduction are evident after one week in high-output fistulae or after 3 weeks in low-output fistulae [52]. Some studies have reported successful closure of fistulae by endoscopic sphincterotomy within 5-7 days [53, 54]. Out of 5 patients that developed PBF in the current study, one patient was re-operated due to abscess formation in the residual cyst bed, in addition to fistula. Sphincterotomy by ERCP was attempted in the other 4 patients. This was achieved in 2 patients, in whom fistulae closed within 5-7 days. In one patient, the fistula remained open despite the sphincterotomy. In the other patient, they were unable to cannulate the papilla. A need for re-operation was required in 3 patients that developed postoperative fistula (**Figure 1**).

Hepatic hydatid disease recurrence rates of 8-15% have been reported in the literature [47]. In the current study, the average follow-up time was 5.5 years, with no recurrences encountered. Akbulut et al. reported a lower recurrence rate, which they linked to routine alben-

dazole administration for 3 months, although they could not correlate the use of albendazole to lower recurrence rates [34]. In this study, albendazole was administered for 1 month pre-operatively and 6 months at postoperatively.

Conclusion

In conclusion, the best surgical approach for complicated giant hepatic hydatid cysts remains open to discussion. Various types of surgical interventions are currently performed in different centers. Many different approaches exist with regards to the timing and premedication of surgical intervention, prior need for endoscopic intervention, type of intervention, as well as management of complications. Present study results suggest that cholecystectomy with PC and perioperative visualization of the biliary tree by cholangiography are safe and useful procedures for giant hepatic hydatid cysts. However, this study may have been limited by its retrospective nature. Future randomized and prospective studies are necessary to validate present findings.

Disclosure of conflict of interest

None.

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Rare clinical entity



Figure S1. Pericystic and operative fields with gauze soaked in 20% hypertonic saline to prevent contamination with the cystic fluid. Partial cystectomy and evacuating.

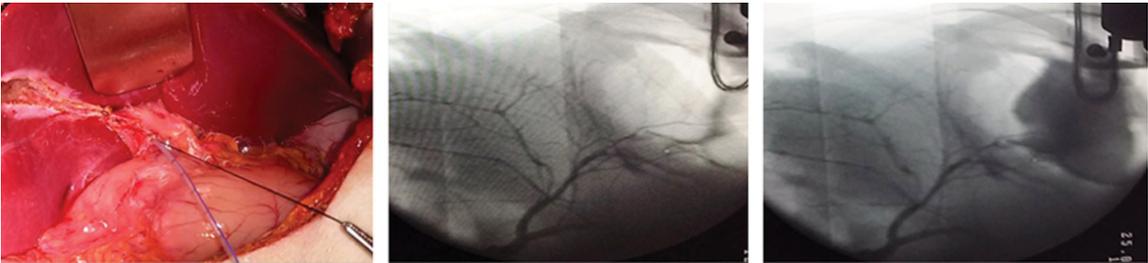


Figure S2. A cholangiography was performed to visualize the biliary tree, with the help of a 6F nelaton catheter advanced through the cystic canal.