World J. Surg. 26, 704–708, 2002
DOI: 10.1007/s00268-002-6259-y

WORLD
Journal of
SURGERY

Significance of Bile Leaks Complicating Conservative Surgery for Liver Hydatidosis

George Skroubis, M.D.,[1] Constantine Vagianos, M.D.,[1] Andreas Polydorou, M.D.,[2]
Evangelos Tzoracoleftherakis, M.D.,[1] John Androulakis, M.D.[1]

[1]Department of Surgery, University of Patras, Rion University Hospital, 26500 Patras, Greece
[2]Department of Surgery, University of Athens, "Evgenidion" Hospital, Papadiamantopoulou 18-20, 11528 Athens, Greece

Published Online: March 26, 2002

Abstract. Hepatic hydatidosis presents a challenge in liver surgery, and there is still controversy regarding the appropriate surgical technique. A high incidence of postoperative bile leaks is reported as a significant disadvantage of conservative surgical procedures. The purpose of this study was to examine the incidence and clinical importance of bile leakage in patients being treated exclusively by a conservative surgical technique. From January 1985 to November 2000 a total of 187 patients were operated on at our department for hepatic hydatidosis. They were subjected to the standard conservative surgical technique (wide unroofing and cyst drainage). A total of 18 complications were related to bile leakage (10%), 3 of them bile abscesses (1 drained surgically and 2 percutaneously), 1 case of bile peritonitis due to an accessory bile duct in the gallbladder bed (treated surgically), and 14 fistulas (1 bronchobiliary and 13 biliocutaneous). Five of the fistulas, including the bronchobiliary one, were treated successfully by endoscopy; and the remaining nine healed after conservative treatment. Bile leakage, representing a significant complication following conservative operations for hepatic hydatidosis, can be effectively treated conservatively or endoscopically, not justifying more aggressive surgical approaches.

Cystic hydatidosis is a parasitic disease endemic in Mediterranean countries. Caused by the tapeworm *Echinococcus granulosus,* it is characterized by the formation of one or more expanding cysts located mainly in the liver (70–75%) [1] and lungs (20–25%) [1].

Medical therapy has been disappointing for treating primary hepatic hydatidosis; and as suggested by the World Health Organization (WHO), it should be restricted to inoperable cases or to preventing recurrence [2, 3]. Surgery remains the treatment of choice, but controversy still exists regarding the most appropriate surgical method by which total extirpation of the parasite is achieved, avoiding at the same time postoperative complications, which may be particularly troublesome.

Numerous surgical techniques have been proposed. The more aggressive operations, hepatectomy and total pericystectomy, entail removing the whole cyst with the pericyst, the thick surrounding layer that results from compression. There is often an immunologic reaction and infection of the hepatic parenchyma [4]. On the other hand, conservative operations mainly aim at deroofing the cyst and evacuating its contents, taking special precautions to avoid intraperitoneal spillage of the hydatid fluid and the daughter cysts [4]. Despite increased morbidity and unnecessary loss of normal hepatic parenchyma, in experienced hands radical operations seem to be highly effective in reducing bile leakage and disease recurrence [5, 6]. The more conservative operations are technically easier and safer but are related to a high incidence of postoperative bile leakage, reported to approach 28% [4]. This high incidence and a significant local recurrence rate are regarded as the main disadvantages of conservative surgery [5–9].

The purpose of this study was to examine the incidence, clinical significance, and treatment options of bile leakage in patients suffering from hepatic hydatidosis when a conservative surgical technique is applied.

Materials and Methods

We evaluated all patients with hepatic hydatidosis who had been operated on at our institution from January 1985 to November 2000. In the few cases of a peripheral or a "hanging" cyst, total cystectomy or pericystectomy had been performed, and these cases are not included in our study material. In all other patients (n = 187) the same conservative surgical technique was applied, which we have described in the past [10]. The special characteristics of patients are shown in Table 1.

The cysts were large and mostly located in the right liver lobe; among them, 69 presented with clinical and laboratory findings of infection. The commonest presenting symptom was right upper quadrant pain (80%) followed by fever (30%), jaundice (20%), and allergic reactions (7%). In 36 (19%) patients the disease was completely asymptomatic and was discovered incidentally. The diagnosis in all cases was established by computed tomography, ultrasonography, or both; immunologic tests and detection of anti-echinococcal antibodies were occasionally undertaken but only in doubtful cases.

Our technique consisted of the widest possible unroofing of the cyst, careful evacuation of the content avoiding contamination of the peritoneal cavity, interlocking suturing of the cavity edges to

Correspondence to: C. Vagianos, M.D.

Table 1. Characteristics of patients.

Total no. of patients	187
Men/women (no.)	78/109
Age (years)	16–85 (median 61)
Cyst diameter (cm)	5–25 (median 10)
Cyst location (right/left/bilateral)	138/29/20
Infection	69 (37%)
Reoperation	24 (13%)

avoid bleeding and bile leakage, and drainage of the residual cavity using negative-pressure closed-system drains [10]. The abdominal cavity was carefully isolated using compresses soaked in 15% hypertonic saline. Omentoplasty was not performed routinely; it depended on the intraoperative decision of each surgeon.

Cholecystectomy was added in cases of coexisting gallbladder disease, when cholangiography was needed, or when the gallbladder was located near the cyst. Intraoperative cholangiography through the cystic duct was performed in cases where duct pathology was suspected, and the duct was explored in the presence of intraductal daughter cysts, hydatid debris, or (uncommonly) bile stones.

When the cyst fluid was bile-stained, a cystobiliary communication was suspected. Such communications were identified by retrograde infusion of normal saline through the cystic duct after performing cholecystectomy and obstructing the peripheral common bile duct using an atraumatic vascular clamp. Back-flow of saline revealed these communications, which were then sutured [11].

The surgical procedure followed for the infected hydatid cysts was essentially the same, and antibiotics were administered for a longer postoperative period. Imidazole compounds (albendazole 10 mg/kg body weight per day) were administered 1 week prior to and 3 months after the operation, particularly in cases where intraoperative contamination was suspected.

Results

There was one perioperative death in our series (mortality 0.5%): an 81-year-old man who died of cerebral hemorrhage on the fourth postoperative day. Hospitalization time ranged from 6 to 94 days (mean 11 days).

Cholecystectomy was performed in 74 patients (39.5% of the total). Bile duct pathology was discovered in 24 patients (13%), and the duct was explored and a T-tube inserted. Omentoplasty was performed in 35 patients (19%), with diminishing frequency over the years.

Almost all postoperative complications were related to bile leakage (Table 2). One patient developed biliary peritonitis on the first postoperative day and had to be urgently reoperated. It could not be directly attributed to the type of surgery, however, as the leakage was due to an accessory bile duct at the gallbladder bed not recognized at the time of the surgery, which also included cholecystectomy. The accessory duct was sutured, and the patient recovered well.

Three patients developed postoperative perihepatic abscesses (one subphrenic and two subhepatic). Two of the abscesses were successfully drained percutaneously under the guidance of ultrasonography, and one was treated surgically after an unsuccessful percutaneous attempt. All patients received antibiotics based on bacterial cultures and presented no further morbidity.

Table 2. Bile leak-related complications and their treatment.

Complication	No.	Percent of total	Treatment
Total	18	10.0	
Bile abscess	3	1.5	Operation (n = 1) Percutaneous drainage (n = 2)
Bile peritonitis	1	0.5	Operation
Bronchobiliary fistula	1	0.5	Percutaneous drainage + endoscopy
Biliary fistula	13	7.0	
Low-output (< 300 ml/day)	10	5.0	Conservative (n = 9) Endoscopy (n = 1)
High-output (> 300 ml/day)	3	1.5	Endoscopy (n = 3)

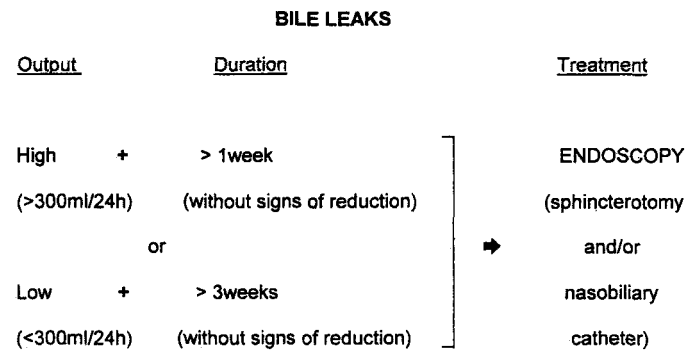

Fig. 1. Suggested treatment of significant bile leaks.

By far the commonest type of bile leakage was a postoperative bile fistula. Biliary fistulas were sought even when only small volumes of bile drained daily over a short period of time. When a fistula was identified, the patient was first evaluated carefully to exclude insufficient external drainage of the cavity and thereby intraperitoneal bile accumulation. In doubtful cases computed tomography, ultrasonography, or both were necessary to exclude fluid accumulation. Conservative therapy was the treatment of choice, consisting of careful daily follow-up of the patient, measuring the daily bile output, and replacing the patient's daily fluid and electrolyte losses. Low-output fistulas (< 300 ml bile/day) could be treated on an outpatient basis. High-output fistulas (> 300 ml bile/day) of more than 1 week's duration with no signs of reduction and low-output fistulas of more than 3 weeks' duration without signs of reduction were treated by endoscopy (Fig. 1).

At endoscopy a nasobiliary catheter was inserted, with or without sphincterotomy, connected to low-pressure suction (10–15 cm H_2O). The catheter was left in place until the biliary fistula stopped draining, and then the drain was removed. A cholangiogram through the nasobiliary catheter, obtained a few days later, revealed complete healing of the cystobiliary communication (Fig. 2). The catheter was then removed and the patient discharged.

Fourteen patients developed biliary fistulas, one of them bronchobiliary. The latter appeared in a patient who underwent right thoracotomy to treat a right lung hydatid cyst, while at the same time a coexisting cyst of the right liver lobe was drained through the diaphragm. Postoperatively, an abscess formed in the residual cavity, which resulted in penetration of the diaphragm and com-

Fig. 2. Treatment of biliary fistula by placing a nasobiliary catheter. Before (**a**) and after (**b**) treatment.

Table 3. Classification of low-output biliary fistulas.

Parameter	No.	Percent of total	Treatment
Total number	10	5	
Output			
< 50 ml/day	8	4	Conservative (n = 8)
> 50 ml/day	2	1	Endoscopy (n = 1); conservative (n = 1)

munication with the bronchial tree. The patient presented with bronchospasm and was treated successfully by a combination of percutaneous drainage and endoscopic placement of a nasobiliary catheter under low suction. Occlusion of the bronchobiliary communication was verified by cholangiography 1 week later, and the nasobiliary catheter and drain were removed within 5 days.

Among the remaining 13 biliary fistulas, 10 were low-output and 3 high-output. Among the low-output fistulas, eight were simple bile leaks from the drainage catheters, accounting for 20 to 50 ml daily and in most cases lasting not more than 1 week (Table 3).

Nine of the biliary fistulas healed with conservative treatment, requiring an average of 10 days until the drain was removed. Patients in this group were treated on an outpatient basis. The remaining four (three high-output, one low-output) were subjected to therapeutic endoscopy, as previously described. In one case a sphincterotomy was performed first, but because of insufficient results endoscopy had to be repeated and a nasobiliary catheter inserted. There was no further morbidity related to biliary fistulas.

Discussion

Surgery for hepatic hydatidosis aims at totally extirpating the parasite while avoiding immediate and late complications. Thus treatment of the residual cavity and avoidance of bile leaks and spillage of hydatid fluid resulting in anaphylaxis and peritoneal dissemination are the main concerns.

Although many do not recommend the use of negative-pressure closed drains, probably because of increased postoperative bile leakage, we prefer the morbidity of a bile leak to complications related to improper drainage of the residual cavity or the hazard of contamination using gravity drains [12]. Negative-pressure closed drains may be used to drain adequately for a long time without introducing secondary contamination.

Recent data [13, 14] seem to demonstrate a clear advantage of omentoplasty for reducing postoperative complications during conservative surgical treatment of hepatic hydatidosis. However, this has not been our experience [7, 9], and we have chosen not to add it routinely, leaving the decision to the surgeon.

Cystobiliary communications are quite common with hepatic hydatidosis, and the incidence of intrabiliary rupture of cysts has been reported to reach 25% [15]. In the presence of a competent, functioning mechanism at the sphincter of Oddi, after cyst drainage and even in the absence of obvious bile duct pathology there is a pressure gradient between the bile duct and the residual cavity, facilitating flow of bile through these communications

toward the cavity rather than the duodenum [16]. This bile leakage represents the main source of immediate postoperative complications. If not properly drained, it may result in abscess formation in the residual cavity or passage to the peritoneum and bile peritonitis. If drained effectively outward, an external biliary fistula develops, representing the commonest complication of such operations [4]. Because of the remaining thick, calcified pericyst, such fistulas may be particularly persistent; the postoperative course may be prolonged, and occasionally hazardous reoperations are required before the patient is cured [8]. In most cases the fistula closes spontaneously, provided the intrabiliary pressure is not high. Reduction of such high pressures by endoscopic sphincterotomy, removal of the intraductal pathology if present, and insertion of an endoprosthesis promotes healing, even in the absence of obvious obstruction [16, 17].

Endoscopic intervention is required for high-output fistulas or fistulas not responding to conservative treatment. Prolonged external bile loss may lead to substantial fluid and electrolyte depletion. In addition, chronic external biliary fistulas may be complicated by steatorrhea, calcium and vitamin D malabsorption, infection, and poor wound healing [18].

Endoscopic biliary drainage by inserting an endoprosthesis is a technique originally applied for decompression in patients with malignant [19] or benign [20] obstruction of the biliary system. Although prophylactic preoperative reduction of bile duct pressure in selected patients has been reported to reduce the incidence of bile leaks successfully [21] it seems that it cannot be used routinely in asymptomatic patients [22].

Like most fistulas of the gastrointestinal tract, biliary fistulas are best managed by lowering the pressure in the bile duct, thereby facilitating bile flow toward the duodenum. Factors contributing to nonhealing include the volume of the bile, the size of the defect (breach of biliary integrity), and the outflow pressure. Endoscopic sphincterotomy has been used to address the latter factor and seems to promote healing by lowering pressures, even in the absence of bile duct pathology. In the case of ongoing leakage, one may proceed to more aggressive endoscopy by inserting a stent [23].

An alternative to stenting is selective nasobiliary tube placement proximal to the fistula with negative-pressure suction [24]. Nasobiliary tube drainage, with or without sphincterotomy, has been reported to treat bile leaks successfully after cholecystectomy [25] and acute cholangitis [26]. Despite the fact that an internal endoprosthesis is better tolerated and avoids bile loss, nasobiliary catheters have the advantage of being easy to flush if occluded; moreover, cholangiography may be performed at any time, and the endoprosthesis can be easily removed, not requiring new endoscopy.

There is hardly any indication for open surgery nowadays in the treatment of external biliary fistulas. Endoscopic retrograde cholangiopancreatography (ERCP) is the method of choice for dealing with bile leakage, being safe, well tolerated, and effective [27]. Utilization of ERCP seems extremely meaningful in hydatid liver surgery [22].

The incidence of postoperative bile leakage in our material remains significant (10%) although lower than that in other reports [5–8]. It should be pointed out, however, that 8 of 14 fistulas (57% of the total) were simple bile leaks accounting for less than 50 ml of bile drained daily, in most cases lasting not more than 1 week (Table 3) and adding no actual morbidity.

Endoscopic intervention with sphincterotomy, biliary stenting, or selective nasobiliary drainage in persistent fistulas has eliminated the need for complex reoperations. We consider these steps the procedures of choice.

Conclusions

Bile leakage represents a significant complication following conservative surgery for hepatic hydatidosis. It can be effectively treated conservatively or by endoscopy [28]. Consequently we do not believe bile leaks justify more aggressive surgery during treatment of hepatic hydatidosis.

Résumé. L'hydatitose hépatique représente un véritable challenge en chirurgie hépatique, alors que la meilleure technique chirurgicale est toujours controversée. L'incidence de fistules biliaires postopératoires est élevée: elles représentent l'inconvénient majeur des procédés chirurgicaux conservateurs. Le but de cette étude a été d'analyser l'incidence et l'importance clinique des fuites biliaires chez les patients traités exclusivement par une technique chirurgicale conservatrice. Entre janvier 1985 et novembre 2000, 187 patients ont été opérés dans notre département pour hydatitose hépatique: le procédé chirurgical, standardisé, était conservateur (résection large du dôme saillant et drainage du kyste). Il y avait au total 18 complications en rapport avec une fuite biliaire (10%), trois abcès biliaires (un drainé chirurgicalement et deux par voie percutanée), une péritonite biliaire en rapport avec un canal biliaire accessoire du lit vésiculaire (traitée chirurgicalement), et 14 fistules (une fistule biliobronchique et 13 biliocutanées). Cinq des fistules, y compris la fistule biliobronchique ont été traitées avec succès par voie endoscopique, alors que les neuf autres ont guéri par un traitement conservateur. La fuite biliaire, une des complications possibles après intervention conservatrice pour hydatose hépatique, peut être traitée efficacement de façon conservatrice et endoscopiquement, et ne justifie pas une approche chirurgicale, plus agressive.

Resumen. La hidatidosis hepática constituye un desafío en el campo de la cirugía del hígado y todavía se mantiene la controversia acerca de cuál es la técnica quirúrgica más apropiada. Se informa una elevada tasa de fuga biliar postoperatoria como una desventaja significativa de los procedimientos quirúrgicos conservadores. El propósito del presente estudio fue determinar la incidencia y la importancia clínica de la fuga biliar en pacientes manejados con técnica quirúrgica conservadora exclusivamente. Entre enero de 1985 y noviembre de 2000 se operaron 187 pacientes con hidatidosis hepática en nuestro Departamento utilizando una técnica quirúrgica conservadora estandarizada (destechamiento amplio y drenaje del quiste). Se registraron 18 complicaciones relacionadas con fuga biliar (10%), tres de ellas fueron absceso (uno drenado quirúrgicamente y dos en forma percutánea), una peritonitis biliar debida a un conducto biliar accesorio en el lecho de la vesícula biliar (tratado quirúrgicamente) y 14 fístulas (una broncobiliar y 13 biliocutáneas). Cinco de las fístulas, incluyendo la bronchobiliar, fueron manejadas exitosamente mediante endoscopia, en tanto que las nueve restantes cicatrizaron con tratamiento conservador. Las fugas biliares, que representan una complicación significante de las operaciones conservadoras en la hidatidosis hepática, pueden ser efectivamente tratadas con manejo conservador o endoscópico, y no se justifican conductas quirúrgicas más agresivas.

References

1. El Mufti M (1989) Surgical Management of Hydatid Disease, London, Butterworths, 31, 73
2. Schantz PM. Effective medical treatment for hydatid disease? J.A.M.A. 1985;253:2095–2097
3. Davis A, Pawlowski ZS, Dixon H. Multicentre clinical trials of benzimidazole carmates in human echinococcosis. Bull. W.H.O. 1986;64: 383–388

4. Demirci S, Eraslan S, Anadol E, et al. Comparison of the results of different surgical techniques in the management of hydatid cyst of the liver. World J. Surg. 1989;13:88–90
5. Magistrelli P, Masetti R, Coppola R, et al. Surgical treatment of hydatid disease of the liver: a 20-year experience. Arch. Surg. 1991; 126:518–522
6. Moreno Gonzalez E, Rico Selas P, Martinez B, et al. Results of surgical treatment of hepatic hydatidosis: current therapeutic modifications. World J. Surg. 1991;15:254–263
7. Karavias D, Vagianos C, Bouboulis N, et al. Improved techniques in the surgical treatment of hepatic hydatidosis. Surg. Gynecol. Obstet. 1992;174:176–180
8. Dawson JL, Stamatakis JD, Stringer MD, et al. Surgical treatment of hepatic hydatid disease. Br. J. Surg. 1988;75:946–950
9. Vagianos CE, Karavias D, Kakkos SK, et al. Conservative surgery in the treatment of hepatic hydatidosis. Eur. J. Surg. 1995;161:415–420
10. Vagianos CE, Androulakis JA. Capsectomy and drainage in hepatic hydatidosis. Dig. Surg. 1997;14:241–244
11. Tzoracoleftherakis E (1998) Identification of cystobiliary communications in hepatic hydatidosis. In Bismuth M, Galmiche JP, Huguier M, editors. 8th World Congress of the International Gastrosurgical Club, Bologna, Monduzzi Editore (International Proceedings Division), 495–498
12. Balik AA, Basoglu M, Celebi F, et al. Surgical treatment of hydatid disease of the liver: review of 304 cases. Arch. Surg. 1999;134:166–169
13. Dziri C, Paquet JC, Hay JM, et al. Omentoplasty in the prevention of deep abdominal complications after surgery for hydatid disease of the liver: a multicenter, prospective, randomized trial; French Associations for Surgical Research. J. Am. Coll. Surg. 1999;188:281–289
14. Ozacmak ID, Ekiz F, Ozmen V, et al. Management of residual cavity after partial cystectomy for hepatic hydatidosis: comparison of omentoplasty with external drainage. Eur. J. Surg. 2000;166:696–699
15. Alper A, Arioglou O, Emre A, et al. Choledochoduodenostomy for intrabiliary rupture of hydatid cysts of the liver. Br. J. Surg. 1987;74: 243–245
16. Tekant Y, Bilge O, Acarli K, et al. Endoscopic sphincterotomy in the treatment of postoperative biliary fistulas of hepatic hydatid disease. Surg. Endosc. 1996;10:909–911
17. Goldin E, Katz E, Wengrover D, et al. Treatment of fistulas of biliary tract by endoscopic insertion of endoprostheses. Surg. Gynecol. Obstet. 1990;170:418–423
18. Hoffmann BJ, Cunningham JT, Marsh WH. Endoscopic management of biliary fistulas with small calibre stents. Am. J. Gastroenterol. 1990;85:705–707
19. Huibregtse K, Tytgat GN. Palliative treatment of obstructive jaundice by transpapillary introduction of large bore bile duct endoprosthesis. Gut 1982;23:371–375
20. Siegal JH, Harding GT, Chateau F. Endoscopic decompression and drainage of benign and malignant biliary obstruction. Gastrointest. Endosc. 1982;28:79–82
21. Vagianos C, Polydorou A, Vagenas C, et al. Combined endoscopic and surgical treatment of complicated hepatic hydatidosis. Endoscopy 1994;26:367
22. Dumas R, Le Gall P, Hastier P, et al. The role of endoscopic retrograde cholangiopancreatography in the management of hepatic hydatid disease. Endoscopy 1999;31:242–247
23. Marks JM, Ponsky JL, Shillingstad RB, et al. Biliary stenting is more effective than sphincterotomy in the resolution of biliary leaks. Surg. Endosc. 1998;12:327–330
24. Vagianos C, Polydorou A, Vagenas C, et al. Successful treatment of postoperative external biliary fistula by selective nasobiliary drainage. HPB Surg. 1992;6:115–120
25. Chow S, Bosco JJ, Heiss FW, et al. Successful treatment of postcholecystectomy bile leaks using nasobiliary tube drainage and sphincterotomy. Am. J. Gastroenterol. 1997;92:1839–1843
26. Sugiyama M, Atomi Y. The benefits of endoscopic nasobiliary drainage without sphincterotomy for acute cholangitis. Am. J. Gastroenterol. 1998;93:2065–2068
27. Mortensen J, Kruse A. Endoscopic management of postoperative bile leaks. Br. J. Surg. 1992;79:1339–1341
28. Rodriguez AN, Sanchez del Rio AL, Alguacil LV, et al. Effectiveness of endoscopic sphincterotomy in complicated hepatic hydatid disease. Gastrointest. Endosc. 1998;48:593–597