World J. Surg. 25, 28–39, 2001
DOI: 10.1007/s002680020005

WORLD
Journal of
SURGERY

Management of Liver Hydatid Cysts with a Large Biliocystic Fistula: Multicenter Retrospective Study

Abdeljélil Zaouche, M.D.,[1] Karim Haouet, M.D.,[1] Mohamed Jouini, M.D.,[2] Adnène El Hachaichi, M.D.,[1] Chadli Dziri, M.D.,[3] Tunisian Surgical Association[4]

[1]Surgical Unit A, Charles Nicolle Hospital, Bd9 Avril, 1006 Tunis, Tunisia
[2]Surgical Unit, La Rabta Hospital, 1077 Jebari, Tunis, Tunisia
[3]Surgical Unit B, Charles Nicolle Hospital, Bd9 Avril, 1006 Tunis, Tunisia
[4]Institut National de la Santé de l'Enfance, Bab Saadoun, 1006 Tunis, Tunisia

Published Online: December 18, 2000

Abstract. The large biliocystic fistula (> 5 mm) encountered with hydatic cyst of the liver produces clinical manifestations only when it allows the hydatic cyst content to pass into the common bile duct. Various therapeutic problems occur. The aim of this study was to evaluate the results of the therapeutic methods used by 14 Tunisian centers to treat this specific aspect of the hydatic cyst of the liver associated with a large biliocystic fistula. This study concerned a period of 5 years between January 1988 and December 1992, and it included 244 cases associated with hydatic content in the common bile duct (158 cases) and with cholelithiasis and choledocholithiasis (2 cases); 127 patients underwent an emergency operation (52%). The surgical procedures performed consisted in radical procedures (24 cases, 9.8%) and conservative procedures (220 cases, 90.2%). The latter included 52 cases of internal transfistulary drainage, 140 unroofing procedures associated in 20 cases with the fistula, in 93 cases with suture of the fistula, and in 27 cases with direct fistulization. In the 28 remaining cases, through the choledoctomy evacuation of the parasite was performed. The common bile duct was approached in 180 cases (73.7%). The postoperative course was uneventful in 57% of the cases and complicated in 38.5% others. The mortality rate was 4.5%. In conclusion, the presence (or not) of hydatic material in the common bile duct did not seem to be a determinant of the surgical procedure choice and did not influence the results. The only difficulty with treatment was the large biliocystic fistula itself. The internal transfistulary drainage on one part, and the unroofing procedure associated with suturing healthy fistula tissue and to omentoplasty or capitonnage of the remaining cavity on the other part, were easily performed and constituted efficient methods. Radical methods constituted operations that had excellent results, but they were feasible in only 10% of the cases.

The natural history of *Echinococcus granulosus* localized in the liver is closely related to the biliary tract. The biliary tract lesions are various and explain the diversity of clinical features and the multiple surgical procedures suggested [1, 2].

Hydatid cysts of the liver (HCLs) can rupture into the biliary tract, as defined by the presence of hydatic material in the common bile duct [3–6]. This situation constitutes an anatomic and clinical entity characterized by the occurrence of life-threatening acute cholangitis [7–10].

Correspondence to: A. Zaouche, M.D., e-mail: abdeljelil.zaouche@rns.tn

The large biliocystic fistula (LBCF), defined as a fistula with a diameter ≥ 5 mm [10–14], manifests clinically only when it allows hydatic cyst content to pass into the common bile duct. The LBCF increases the therapeutic problems [1, 2, 5, 12, 13, 15–21]. Indeed, once the parasite is evacuated and the obstacle in the main biliary tract is removed, the problem of the residual cavity of the cyst and the LBCF remains. Independent of the fact that various surgical procedures are available for treating a biliocystic fistula and the residual cavity, the LBCF may be responsible for the high morbidity rate. The aim of this study was to evaluate the results of various therapeutic methods used by the teams working on this multicenter study to treat HCLs associated with an LBCF.

Patients and Methods

Inclusion Criteria

Fourteen Tunisian surgical centers took part in this retrospective study over 5 years between January 1988 and December 1992. The study included 244 HCLs with an LBCF. They were associated with hydatic content in the common bile duct (CBD) 158 times and with cholelithiasis and choledocholithiasis twice.

Twenty-five other LBCF cases were excluded from this group because of the simultaneous presence of destruction of the upper biliary confluence or a concomitant thoracic opening. These associated complications posed specific problems.

The observation data were obtained from a basic program consisting of 68 variables relative to clinical presentation, complementary investigation, intraoperative findings, surgical procedures, and outcomes. To evaluate the results, we have used judgment criteria regarding specific complications: external biliary fistula, suppuration and retention in the remaining cavity, residual hydatic obstacles in the CBD, postoperative choleperitonitis, mortality rate, and median hospital stay.

Clinical Presentation

The 168 women (68.9%) and 76 men (31.1%) studied here ranged in age from 9 to 84 years with a mean age (± SD) of 42.7 ± 18.1

Table 1. Intraoperative findings concerning hydatid cysts of the liver with a large biliocystic fistula.

Parameter	No.
Size (cm)	
≤ 5	24
6–10	116
11–20	56
> 20	9
Not mentioned	39
Site of the HCL	
Right liver	179 (73.4%)
Dome (VII–VIII)	76
Right paramedian (V–VIII, IV–VIII)	24
Right lateral (VI–VII)	22
Anterior segments (IV–V–VI)	57
Left lobe	46 (18.8%)
Caudate lobe	7 (2.9%)
Complex systematization	12 (4.9%)
Cyst contents	
Plurivesicular and bile-stained	238
Univesicular	6
HCL exteriorization	
Widely exteriorized	126
Partially exteriorized	86
Inaccessible intraparenchymatous	28
Approachable after hepatotomy	4
Cyst wall	
Soft	18
Fibrotic	46
Calcified	44
Not mentioned	136
Site of the LBCF	
Segmentary	131
Sectorial	70
Left hepatic duct	9
Right hepatic duct	7
Not mentioned	27

HCL: hydatid cyst of the liver; LBCF: large biliocystic fistula.

years. At admission 56 patients had an uncomplicated HCL, 39 had an isolated infectious syndrome, 138 had acute cholangitis (among which 25 were severe), and the remaining 11 patients had isolated jaundice. The patients underwent preoperative ultrasonography complemented eight times by computed tomography (CT) and twice by endoscopic retrograde cholangiopancreatography (ERCP).

Intraoperative Findings

A total of 127 patients underwent an emergency operation (52%) and 117 selective surgery (48%). During intervention, exploration of the liver and the extrahepatic biliary tracts was first manual and visual. The HCL was isolated in 177 cases; in 67 other cases there were one or several associated, noncomplicated HCLs.

The lesions observed during the operation concerned only the HCLs that were sites of an LBCF (Table 1). Because of the retrospective aspect of the study, various parameters were not always specified on the operative reports.

The HCL measured more than 10 cm 65 of 205 times. In 179 cases it was located in the right liver (73.4%) among which 57 were directly accessible on anterior segments (IV, V, VI) and 46 were in the left lobe (18.8%). In 28 cases the HCL did not protrude onto the liver surface, and 4 were approachable after a 2- or 3-cm hepatotomy.

Table 2. Intraoperative findings concerning the extrahepatic biliary tracts.

Finding	No.
Gallbladder	
Normal	125
Not attached to the cyst	14
Pathologic	105
Choleliathiasis	42
Acute cholecystitis	63
Hydatic material	28
Hydatic material and cholelithiasis	3
Purulent gallbladder bile with neither hydatic material nor lithiasis	32[a]
Common bile duct	
CBD obstacle	160 (65%)
Hydatic	153
Hydatic and lithiasic	5
Lithiasic	2
Free CBD	84 (35%)

CBD: common bile duct.
[a]Among which 25 had hydatic material in the CBD.

The LCBF was at least 5 mm, sometimes reaching 2 cm. Its exact site was determined in only 217 cases. It was segmental in 131 cases and sectorial in 70 (right paramedian 21; right lateral 20; left lateral 29). It was a left hepatic duct fistula in nine cases, whereas a right hepatic duct fistula far from the upper biliary confluence was observed in seven cases.

The terminal or lateral nature of the biliary fistula on the biliary ducts was determined only 10 times. The LBCF was sometimes associated with other small biliary fistulas. The status of the extrahepatic biliary tract was always determined (Table 2). Among the acute cholecystitis cases, parietal gallbladder inflammation and purulent vesicular bile but with no hydatic material or gallstone occurred in 32 cases. Among the latter, hydatic material in the CBD was in evidence 25 times. No HCL fistulizations in the gallbladder were found in this group of 244 patients.

A lithiasic or hydatic obstacle was encountered in the CBD in 21 cases with no suggestive clinical signs, such as fever or jaundice (21/56, 37.5%). In contrast, among the 188 patients who were admitted for acute cholangitis, isolated jaundice, or isolated infectious syndrome, 139 (73.9%) exhibited an obstacle in the CBD.

The liver and biliary tract were explored using intraoperative cholangiography 219 times (89.8%), as follows: 170 times through the cystic duct after cholecystectomy of a pathologic gallbladder ($n = 105$), a gallbladder not stuck to the cyst ($n = 14$), and a healthy gallbladder ($n = 51$); 39 times through a cholecystostomy catheter; and 10 times through a catheter pushed into the biliocystic fistula. Twenty-five patients did not undergo cholangiography, but they underwent choledochotomy right away because of the strong clinical presumption of an obstacle in the CBD and the intraoperative observation of an LBCF together with a distended CBD.

Surgical Procedures

When the HCL is not deeply seated in the liver, treatment starts by parasite evacuation. After having protected the operative site by gauze packs soaked in H_2O_2, the cyst content is aspirated. The HCL is approached either directly or after having been injected with a scolicidal agent in variable quantity, generally H_2O_2 or

Table 3. Surgical procedures performed on hydatid cysts of the liver with a large biliocystic fistula.

Procedure	No.
Radical treatment	24 (9.8%)
Left lobectomy	7
Pericystectomy	17
Conservative treatment	220 (90,2%)
Internal transfistulary drainage	52
Unroofing procedure	140
Respected fistula	20
External drainage	10
External drainage + omentoplasty	8
External drainage + "capitonnage"	2
Sutured fistula	93
External drainage	49
External drainage + omentoplasty	28
External drainage + "capitonnage"	16
Directed fistulization	27
Transcholedochral evacuation	28

Respected: not operated on.

saline solution. Once the protruding dome is removed in part or totally, the proligerous membrane and the hydatic fragments are eliminated. The pericyst is cleaned and smoothed out, and possible daughter cysts developing in the pericyst are removed. The residual cavity is examined for biliary fistulas or pericyst multistratification, which could be softened by intralamellary pericystectomy.

The surgical procedures performed for the HCLs with an LBCF are reported on Table 3. A radical procedure was performed 24 times (9.8%). It consisted in total pericystectomy or hepatic resection. The pericystectomies, including cleavage of the healthy liver pericyst with progressive hemostasis and bilistasis, were total or subtotal pericystectomies, leaving a small portion of pericyst in front of a large vessel, or pericyst resections. The hepatic resections were formal and consisted in a lobectomy. This radical treatment was performed for cysts easily accessible in the left lobe or anterior liver with a thick or calcified pericyst. In 18 of 24 cases (75%) a choledochotomy was performed (Table 4).

A conservative procedure was undertaken in 220 cases (90.2%), and in 52 cases it was internal transfistulary drainage [12]. Once the cyst was approached and evacuated under visual control, the pericyst, softened when necessary by an intralamellary pericystectomy, was sutured with slowly resorbing thread, with no drainage of the residual cavity. The cavity drainage was made "naturally" in the CBD through the fistula. If the CBD was obstructed a choledochotomy was performed to ensure its being emptied, and the CBD was drained by a T-tube. If the CBD was free, a choledochotomy was not done, and no surgery was performed on the extrahepatic biliary tract. No surgical sphincterotomy was undertaken. In 36 of 52 cases (69.9%), a choledochotomy was performed, and the CBD was drained by T-tube. The remaining 16 cases, in which a choledochotomy was not performed, the intraoperative cholangiography was normal.

An extensive unroofing procedure of the cyst was performed in 140 cases. After unroofing, the residual cavities were opened 51 times toward the dome of the liver, 37 times toward the inferior face of the liver, and opened laterally 19 times. Ten unroofing procedures of frontal cysts or external cysts left a shallow residual cavity. The remaining cavity aspect was not described in 23 cases. As for the LCBF, it was treated in variable ways (Tables 3, 4). In 20 cases the fistula was left alone, with the residual cavity aspirated using external drainage; it was filled in with omentoplasty 8 times. In 15 cases there was CBD drainage using either a T-tube (12 cases) or a biliodigestive anastomosis (3 cases); in 3 cases a cystic duct drain was used. Bipolar drainage was thus performed in 18 cases. In the remaining 2 cases, the CBD was free, and neither a choledochotomy nor drainage was performed.

The fistula was sutured in 93 cases associated 69 times with CBD drainage using a T-tube; in 24 cases the CBD was not approached, although in 4 cases there was a cystic duct drain. In two-thirds of cases the suturing mode on sclerotic tissues or on healthy biliary tissue after a perifistulary pericystectomy was not described. Once the biliocystic disconnection was performed, the remaining cavity was drained by Redon drains. In 28 cases omentoplasty and in 16 cases suture obliteration capitonnage of the remaining cavity was performed.

Directed fistulization was performed 27 times. A Nelaton catheter with a caliber adapted to the fistula diameter was introduced with rubbing through the fistula and pushed into the intrahepatic biliary tracts 2 to 3 cm. The catheter end was multiperforated. The catheter was exteriorized through sufficient hepatic tissue thickness with an intracystic course of 3 to 4 cm (as short as possible) without completely crossing the residual cavity. In these cases it was a matter of "transparietohepatocystic" fistulization, and the biliary tract was approached 14 times. Among the remaining 13 patients, 2 had a cystic duct drain. Evacutation of the parasite was done through the choledocotomy 28 times, with cleaning of the remaining cavity, which could not be approached from the liver surface. This cleaning was performed either blindly or with the help of choledochoscopy. In all cases there was a central, deeply sited HCL in the liver with direct opening of the biliary tract. This technique differs from internal transfistulary drainage, as the cystic cavity was not approached from the liver surface or cleaned under visual control, but backward through the choledocotomy.

A total of 214 cholecystectomies were performed: 105 times for a pathologic gallbladder, 14 times to approach the HCL, and 95 normal times after having opened the CBD or having performed transcystic duct cholangiography. Among the 30 gallbladders that were not removed, 24 times it had been totally untouched, three times it had been sutured after having removed a cholecystostomy catheter placed there for cholangiography, and three times a cholecystostomy catheter was maintained.

The common bile duct was approached in 180 cases (73.7%) (Table 4). It was the site of a coincidental gallstone or hydatic obstacle in 160 cases. In 20 cases, it was free when approached, either on principle or because a choledochal obstacle was suspected at cholangiography. The surgical procedure was completed 174 times with installation of a T-tube and six times by a biliodigestive anastomosis performed on patients with a blocked or highly distended CBD. The six biliodigestive anastomoses were performed three times for HCLs evacuated transcholedocally and three times for HCLs treated by an unroofing procedure without approaching the fistula. Among the 64 cases where the CBD was not approached, there were 30 nonremoved gallbladders, 9 cases of cystic duct drainage after cholecystectomy, and 25 cholecystectomies with cystic duct ligation.

Statistical Methods

For comparisons, we used the chi-squared test, the Fisher exact test, the Student *t*-test, and the Mann-Whitney U-test. The significance level was fixed at $p = 0.05$.

Table 4. Surgical procedures on the biliary tracts according to treatment of the remaining cavity and the large biliocystic fistula.

Procedure	No.	Radical treatment ($n = 24$)	Internal transfistulary drainage ($n = 52$)	Unroofing procedure			Transcholedochal evacuation ($n = 28$)
				Fistula respected ($n = 20$)	Fistula sutured ($n = 93$)	Directed fistulization ($n = 27$)	
Approached CBD							
Obstacle	160	16	31	13	60	12	28
No obstacle	20	2	5	2	9	2	0
Nonapproached CBD	55[a]	6	16	2	20	11	0
Nonapproached CBD + cystic duct drain	9	0	0	3	4	2	0

[a]There were 25 cholecystectomies and 30 nonremoved gallbladders.

Table 5. Global results.

Outcome	No.	Hospital stay (days), mean and range
Uneventful postoperative course	139 (57.0%)	15 (4–26)
Complicated postoperative course	94 (38.5%)	31.5 (8–110)
Specific complications	68	
External biliary fistula	32	
Suppuration/residual cavity	23	
Purulent retention/residual cavity	8	
Residual obstacles in the CBD	4	
Postoperative choleperitonitis	1	
Nonspecific complications	26	
Wound infection	14	
Medical complications	12	
Deaths	11 (4.5%)	
Related to the surgical procedure		
Purulent retention/residual cavity	6	
Not related to the surgical procedure		
Severe cholangitis	3	
Pyloroplasty leakage	1	
Cardiac failure	1	

Results

The postoperative course was uneventful in 139 cases (57%) (Table 5). The 11 deaths (4.5%) occurred during the first postoperative month or the same hospitalization. In three cases the deaths were due to an immediate postoperative visceral deficiency. These patients had been admitted with (3) severe acute cholangitis; in on case, a postoperative peritonitis, reoperated on the eighth day, secondary to a leakage of a pyloroplasty performed for an associated complicated duodenal ulcer; and a cardiac failure the third day in an 80-year-old patient. There was a septic retention in the remaining cavity in six cases. Death occurred between days 8 and 49 and was directly related to the surgical procedure performed. One patient needed a percutaneous puncture and the other five a reoperation. The purulent retention evacuation procedures were performed late, when sepsis was obvious. These six deaths occurred after the unroofing procedure, associated twice with external drainage of the residual cavity without treating the fistula, three times with fistula suturing (one of which was associated with omentoplasty) and once with directed fistulization of the fistula.

The 68 specific complications included 32 persistent external biliary fistulas (defined as persisting after the tenth day and accompanied by drainage of dozens of milliliters of clear bile resulting in drainage prolongation); 23 remaining cavity suppurations (defined as being accompanied by fever and a biliopurulent discharge from the drainage, which lengthened the duration of drainage or required irrigation until apyrexia and drying-up); 8 cases of purulent retention in the remaining cavity marked by an obvious infectious syndrome associated with ineffective drainage or a removed drain, necessitating evacuation by percutaneous puncture performed under CT scanning or ultrasonography (five cases) or by reoperation (three cases); 4 residual obstacles to the CBD treated three times by endoscopic sphincterotomy and once by reoperation; and 1 case of postoperative choleperitonitis caused by an improperly intubated LBCF, necessitating reoperation.

On the whole, there were 74 specific complications (SCs) (30.3%), six of which were lethal; eleven early reoperations (4.5%) were required, eight for purulent retention, one for postoperative choleperitonitis, one for residual hydatic obstruction of the CBD, and one for leakage of a pyloroplasty done for an associated complicated duodenal ulcer. The 26 nonspecific complications consisted in 14 cases of wound sepsis and 12 medical complications (postoperative pneumonia 6, phlebitis 2, urinary infection 2, sepsis of the venous catheter 2).

The results of this analysis according to the anatomic lesions is reported on Table 6. Compared to the left lobe and anterior HCLs, the dome and caudate lobe HCLs and those with a complex localization overlapping many segments had a more significant SC rate ($p = 0.7 \times 10^{-4}$) and a longer hospital stay (HS) ($p = 0.01$). The HCL size and the biliary canal (segmentary, sectorial, or principal) that was the site of the fistula did not influence the results. Similarly, the presence of hydatic material in the CBD did not significantly influence the SC, mortality rate, or HS. HCL exteriorization and pericyst thickness was not studied.

As for biliary drainage (Table 7), successful internal or external biliary drainage by biliodigestive anastomosis did not influence the results. In regard to the surgical procedure performed on the HCL (Tables 8, 9), the radical methods that did not leave any remaining cavity, with suturing of the fistula in healthy tissue, were accompanied by the lowest SC rates (8.3% vs. 32.7%; $p < 0.05$) and the shortest median HS (15 vs. 20 days; $p < 0.05$). However, the two groups are not comparable, as the radical procedures were performed on HCLs accessible in patients without cholangitis.

Among the conservative procedures (Table 10), unroofing was most often used (140/244). Internal transfistulary drainage showed better results than the unroofing procedure in terms of the SC rate (1.9% vs. 50%; $p < 10^{-6}$) and median HS (15.0 vs. 23.5 days; $p < 0.05$). The only complication with internal trans-

Table 6. Results according to the anatomic lesions.

Parameter	No.	Specific complications (%) (n = 74)	Mortality (%) (n = 11)	Hospital stay (days), median and range
Hydatid cyst of liver				
HCL site				
Dome	76	34 (44.7%)*	1 (1.3%)	22 (6–90)**
Left lobe and anterior side	103	15 (14.5%)*	5 (4.8%)	17 (2–110)**
Segment I	7	3 (42.8%)*	1 (1.4%)	29 (9–50)**
Others	58	22 (37.9%)*	4 (6.9%)	19 (0–150)**
HCL size				
< 10 cm	140	36 (25.7%)	6 (4.3%)	17 (4–150)
≥ 10 cm	65	22 (32.3%)	3 (4.6%)	19 (0–110)
Large biliocystic fistula***				
Segmentary	131	41 (31.3%)	6 (4.6%)	20 (6–96)
Sectorial or principal	86	33 (38.3%)	5 (5.8%)	20 (0–150)
Hydatic material in CBD***				
Yes	158	50 (31.6%)	6 (3.8%)	21 (2–98)
No	84	24 (28.6%)	5 (5.9%)	17 (0–150)

*$p = 0.7 \times 10^{-4}$.
**$p = 0.01$.
***Difference is not statistically significant.

Table 7. Results according to drainage of the common bile duct.

Parameter	No.	No biliary drainage (n = 55)	T-Tube drainage or biliodigestive anastomosis (n = 180)
Specific complications*	74	16 (29.1%)	55 (30.6%)
Deaths*	11	5 (9.1%)	6 (3.3%)
Hospital stay* (median and range)		18 days (0–150)	21days (2–98)

*Difference not statistically significant.

fistulary drainage was a biliary fistula by the T-tube cutaneous orifice. The death in the group of patients who underwent internal transfistulary drainage was not referable to the method itself but was the consequence of heart failure during the early postoperative course. Yet the unroofing procedure itself was accompanied by various surgical procedures on the LBCF (Tables 8, 11).

When an unroofing procedure was performed, the biliocystic disconnection by suturing or by directed fistulization produced better results than to keep the fistula untouched (SC 43.3% vs. 90%, $p < 0.05$; median HS 22 vs. 33 days, $p < 0.05$).

As for the SCs, fistula disconnection by suturing or fistulization produced the same results. The HS was longer when a directed fistulization was performed, but the difference was not significant.

Once the biliocystic fistula was disconnected by suturing, the residual cavity was externally drained, filled in by omentoplasty, or leveled by capitonnage. There were fewer SCs when omental packing or residual cavity capitonnage was performed than when the residual cavity was simply drained externally ($p < 0.05$), and there was a shorter HS ($p < 0.05$). When filling by omentoplasty and capitonnage were compared, no significant differences were evident. Finally, when internal transfistulary drainage and the unroofing procedure with fistula suturing and omentoplasty were compared (Table 12), we noted that internal drainage produced fewer SCs (1.9% vs. 17.8%; $p = 0.01$).

Discussion

This multicenter series of 244 HCLs with LBCF has reported the largest number of patients. In fact, the 14 surgical centers reported 2013 operated cases [1], although only 244 cases were studied here.

This study revealed the following facts. From the *anatomic point of view,* the HCL was located in the hepatic dome 76 of 244 times (31.1%). The LBCF was segmentary in 131 of 217 cases (60.4%). The CBD was obstacle-free in 35% of cases, despite the existence of an LBCF.

From the *therapeutic point of view,* the most frequently used methods were the unroofing procedure (140/244) and internal transfistulary drainage (52/244). The presence (or not) of hydatic material in the CBD did not seem to be a determinant for the choice of surgical procedure. The only difficulty with treatment was the LBCF itself. It was found in 269 (244 + 25 excluded from this study) of 2013 of the HCLs (i.e., in 13.36% of the cases), and it was associated with hydatic material in the CBD in two-thirds of the cases. The presence (or not) of hydatic material in the CBD did not influence the SC and HS rates. Systematic drainage of a free CBD was not necessary.

The *immediate results* depended on the HCL site and the surgical procedure used. As for the HCL, the dome cysts, caudate lobe cysts, and those that overlapped many segments had a more significant SC rate and a longer HS. As for the therapeutic methods, internal transfistulary drainage, and unroofing procedure associated with suturing the healthy fistula tissue and to an omentoplasty, or suture obliteration capitonnage of the remaining cavity were two easily feasible, effective methods. The radical methods, mainly pericystectomy, were operations that had excellent results but were feasible in only 10% of cases. Directed fistulization was associated with more morbidity and a longer HS, but it was necessary whenever the other methods were not feasible. When the fistula remained untouched the drainage of the remaining cavity with or without CBD drainage was accompanied by a high mortality rate (SCs 16/20; lethal complications 2/20).

The definition of the large fistula was established for the first

Table 8. Results according to the surgical procedures.

Postoperative course	Radical treatment (n = 24)	Internal transfistulary drainage (n = 52)	Unroofing procedure			Transcholedochal evacuation (n = 28)	Total
			Fistula respected (n = 20)	Fistula sutured (n = 93)	Directed fistulization (n = 27)		
Simple postoperative course	20	45	0	42	9	23	139
Specific complications	2	1	16	36	12	1	68
External biliary fistula	2	1	6	18	5	0	32
RC/suppuration	0	0	6	14	3	0	23
RC/retention	0	0	1	4	3	0	8
CBD obstacle	0	0	3	0	0	1	4
Choleperitonitis	0	0	0	0	1	0	1
Nonspecific complications	2	5	2	10	5	2	26
Deaths	0	1	2	5	1	2	11
Related to the procedure	0	0	2	3	1	0	6
Not related to the procedure	0	1	0	2	0	2	5

RC: remaining cavity.

Table 9. Radical versus conservative procedure.

Parameter	No.	Radical treatment (n = 24)	Conservative treatment (n = 220)
Age (years), mean ± SD		42.8 ± 0.4	42.7 ± 17.8
Gender (F/M)		18/6	150/70
Acute cholangitis*	138	8 (33.3%)	130 (59.1%)
HCL site**			
Dome	76	0	76 (34.5%)
Left lobe	46	16 (66.7%)	30 (13.6%)
Anterior side	57	5 (20.8%)	52 (23.7%)
Caudate lobe	7	0	7 (3.2%)
Other localizations	58	3 (12.5%)	55 (25.0%)
Common bile duct drainage	180	18 (75.0%)	162 (73.7%)
Specific complication*	74	2 (8.3%)	72 (32.7%)
Deaths	11	0	11 (5.0%)
Hospital stay (days), median and range*		15 (6–33)	20 (0–150)

*$p < 0.05$.
**$p < 10^{-6}$.

Table 10. Internal transfistulary drainage versus the unroofing procedure.

Parameter	Internal transfistulary drainage (n = 52)	Unroofing procedure (n = 140)
Age (years), mean ± SD	43.3 ± 16.9	42.8 ± 17.9
Gender (F/M)	33/19	81/59
Acute cholangitis	32 (61.5%)	73 (52.1%)
HCL site*		
Dome	19 (36.5%)	48 (34.3%)
Left lobe	13 (25.0%)	14 (10.0%)
Anterior side	11 (21.2%)	41 (29.3%)
Complex systematization	9 (40.4%)	34 (24.3%)
Caudate lobe	0	3 (2.1%)
HCL size**	9.7 ± 3.3	20.1 ± 27.7
Common bile duct drainage	36 (69.2%)	98 (70.0%)
Specific complication**	1 (1.9%)	70 (50.0%)
Deaths	1 (1.9%)	8 (5.7%)
Hospital stay (days), median and range*	15 (0–96)	23.5 (2–150)

*$p < 10^{-6}$.
**$p < 0.05$.

time by Bourgeon [11] who empirically chose a diameter of 5 mm, which corresponds to the diameter of a no. 16 Nelaton catheter. This size constitutes the frontier between fistulas with significant flow. Beyond this diameter, in 64.8% (158/244) there was hydatic material migration in the CBD; on the other hand, below this diameter hydatic material migration is exceptional [1]. Many authors have retained use of the 5 mm diameter to define large fistulas [10, 12–14].

At the beginning, we intended to individualize, among the biliary complications, the 158 HCLs that ruptured into the biliary tract with the presence of hydatic material in the CBD. This type of HCL is well defined and has constituted the subject of many reports [3–6, 15]. Clinically, it raises the problem of acute cholangitis, observed in 70% of cases. This complication may involve a vital prognosis and frequently necessitates an emergency biliary operation to remove the obstruction [3, 4, 7, 9, 10]. Analysis of the data revealed that in addition to these 158 cases, which constituted an indisputable anatomic and clinical entity, there were 86 other HCLs with an LBCF in the pericyst. They caused the same therapeutic problems and had the same prognosis as the 158 HCLs opening into the biliary tract. Accordingly, these 86 cysts did not constitute a distinct anatomic and clinical entity, as the clinical presentation was relatively poor with much less significant jaundice and a lower incidence of cholangitis.

The decision to pool into one entity the 244 HCLs with a large fistula was reinforced by the following facts. Contrary to the small biliary fistulas, which can be found in one of four HCLs [1] but which were always distinct intraoperatively despite the pericyst unfolding and methylene blue injection into the biliary tract, the large fistulas were recognized intraoperatively. Furthermore, they posed specific problems and required special surgery to avoid postoperative biliocystic flow, which is a source of external biliary fistula and retention in the remaining cavity, resulting in significant morbidity. These particular problems of large fistulas com-

Table 11. Comparison of the procedures associated with unroofing.

Parameter	Biliocystic disconnection		Biliocystic fistula		Biliocystic fistula suture		
	Yes (n = 120)	No (n = 20)	Suture (n = 93)	Fistulization (n = 27)	With omentoplasty (n = 28)	With capitonnage (n = 16)	Without omentoplasty or capitonnage (n = 49)
SC	52 (43.3%)*	18 (90%)*	39 (41.9%)	13 (48.1%)	3 (6.8%)*	5 (3.3%)*	31 (63.3%)
LR	6 (14.6%)	2 (10.5%)	5 (5.3%)	1 (3.7%)	2 (7.1%)	1 (6.2%)	2 (4.0%)
HS (days)	22 (2–110)*	33 (10–150)*	20 (2–98)	26 (10–110)	14 (2–98)*	17 (2–110)*	25.5 (4–90)*

SC: specific complication; LR: lethal rate; HS: hospital stay (days), median and range.
*$p < 0.05$.

Table 12. Internal transfistulary drainage versus unroofing, suture of the fistula, and omentoplasty.

Parameter	Internal transfistulary drainage (n = 52)	Unroofing + fistula suture + omentoplasty (n = 28)
Specific complication*	1 (1.9%)	5 (17.8%)
Deaths*	1[a] (1.9%)	2 (7.1%)
Hospital stay (days), median and range	15 (0–96)	18 (2–98)

[a]Death not related to the surgical procedure.
*$p = 0.01$.

pared to small fistulas have been noted by many authors [2, 5, 16, 20–25]. It was demonstrated that they were the determinant and significant element in HS prolongation [2, 24] and the occurrence of postoperative complications, such as purulent retention or external biliary fistula [24]. In a Tunisian study [1], the SCs, mortality rate, and the HS duration were significantly higher in the presence of an LBCF than with small biliary fistulas.

Moreover, the presence (or not) of hydatic material in the CBD did not influence the results in our series, but the fistula size did influence these results. In the case of a large fistula, the SC, mortality, and HS were similar with or without the presence of hydatic material in the CBD.

Moreover, an HCL with an LBCF but without hydatic material in the CBD may necessitate a choledochotomy for associated choledochal calculus extraction in about one-fourth of cases (22/86 in our series) because of an incorrect lacunar image at intraoperative cholangiography or a decrease in biliocystic biliary flow. Among the 244 HCL cases with an LBCF, the CBD was approached to extract hydatic material in 158 patients, and it was approached without the existence of hydatic material in the CBD in 22 cases (12.2%). Finally, all treatment methods of the residual cavity and large fistulas were undertaken regardless of the existence of hydatic material in the CBD.

In Tunisia the HCL is treated only by surgery. Some puncture–aspiration–injection–respiration (PAIR) procedures and a few laparoscopic treatment attempts have been performed; but in agreement with what has been published [26–30], they were performed only on uncomplicated cysts and in highly selected cases.

Endoscopic sphincterotomy with nasal-biliary drainage allows us to go beyond the acute cholangitis stage, but the large fistula HCL is a subject for classic surgery. The surgical treatment is based on several methods, the indications of which come from the surgical team's choice based mainly on the anatomic lesion observed.

In our series, we have seen a large number of nonspecific factors concerning cyst exteriorization, pericyst thickness, incline of the remaining cavity after the unroofing procedure, and the suturing modalities used on the biliocystic fistula. These imprecisions are due to the fact that these studies are retrospective. They can be found in numerous published studies and are reason to accept the results of the comparisons between therapeutic methods and the value of retrospective studies.

Little et al. [31] and Settaf et al. [32] have rightly insisted on the imprecisions related to the exact anatomy of the HCL treated by one method versus another. Settaf et al. [32] suggested anatomic lesion codification to facilitate interpretation of the results. Even though the elements of this classification are questionable, the fact that it points out the need to standardize the terms used and the need for an anatomic lesion collection for better interpretation of therapeutic methods results may be helpful.

Treatment of the HCL with an LBCF is the same as that usually applied to HCLs in general but with particularities relative to the existence of a large fistula and to the possible presence of hydatic material in the CBD. Extraction of hydatic material from the cystic cavity and the biliary tract is unequivocal. The surgical procedures for the residual cavity and biliary tract drainage are more doubtful.

The radical treatment applied in our series, as in most consulted series [20, 33–35], showed better results with less mortality and morbidity and a shorter HS. The fistula is sutured in healthy tissue, and the HCL cavity is respected to avoid any possibility of retention. This radical treatment must often consist in total or subtotal pericystectomy, leaving a small portion of the pericyst next to the junction of the vena cava and upper hepatic vein or the lower vena cava. Pericyst resection or hepatic resection can also be performed.

In practice, these operations are not always applicable. A formal hepatic resection is inordinate for benign pathology and is not always easy to perform except when the cyst is located on the left lobe. Pericystectomy may be hazardous or even impossible to perform, especially for some localizations such as the dome or the paramedian sector when the HCL is in the liver or located near a large biliary tract. This surgical treatment requires blood reserve and experience in the field of hepatic surgery; moreover, it is not possible in two of three patients operated on emergently with severe cholangitis. In our series, as for most authors [32–42], the pericystectomies are for accessible, exteriorized, small, peripheral cysts of the anterior liver or the left lobe; formal hepatic resections are often due to left lobectomy. In highly endemic hydatidosis countries, these radical methods are performed, as in our series, on only 10% of the operated cysts [10, 11, 41].

The other argument in favor of radical treatment is to reduce the risk of hydatic recurrence [35]. In fact, the most important problem with large fistulas is to make the early postoperative course easier by decreasing mortality and morbidity. With conservative treatment, the recurrence risk is less than 4% [31], provided the residual cavity is unfolded so we can recognize an exogenous vesiculation and that precautions are taken to avoid operating field contamination by spillage of scolices contained in the hydatic fluid and in the cyst membrane.

When radical treatment cannot be performed, several conservative methods may be used [2, 6, 25]. Concerning the Tunisian teams' choice, it was seen that no center resorted to the marsupialization used by some authors [43], which was once useful as a life-saving procedure but is now abandoned because of its poor results [37, 44, 45]. Similarly, cystic–digestive anastomosis, which is still used by some authors [4, 17, 46], is not free of risks, reported principally when the cyst is the site of a large fistula [20, 47]. This conservative treatment, which is based on nonextirpation of the residual cavity, consists in various procedures related to the large fistula and drainage of the residual cavity in addition to drainage of the CBD [12, 16, 18, 48, 49].

The unroofing procedure without approaching the large fistula, associated with external drainage of the residual cavity, is an inappropriate technique even if the CBD is approached and drained with a T-tube or a biliodigestive anastomosis and even if technical devices of suture obliteration (capitonnage) or of the residual cavity filling-in omentoplasty are performed. This method, known as bipolar drainage because of residual cavity and CBD drainage on both sides of the fistula, which is respected, was used 20 times in our series. No simple postoperative course has been noted. The strategy consisting of preserving the fistula is condemned by many authors [10, 12, 13]. Even when the CBD has been drained (18/20 times in our series) and an omentoplasty performed, the results were poor, as the omentum cannot absorb the large bile flow coming from the biliary tract. This omentoplasty limit has been observed in our series and has been mentioned by others [2, 13, 20, 46, 47]. The T-tube to avoid biliocystic flow and postoperative external biliary fistula were mentioned by Grosdidier et al. [21]. In our series as in others [13], bipolar drainage is a source of important cholerrhagia, septic retention in the residual cavity, and in the best cases a long HS (several weeks). This method should no longer be used [10, 12–14, 20].

Internal transfistulary drainage is based on a protruding dome limited resection that is sutured after the cavity is cleaned out with respect to the fistula. This concept is different from the reduction without drainage used by some authors [43] for uncomplicated HCLs. Thus internal transfistulary drainage is perform in the HCL with large fistulas as it allows residual cavity evacuation without the risk of retention.

Internal transfistulary drainage was initially described by Goinard et al. [50], who pointed out that once the HCL was evacuated under visual control the pericyst could be sutured without drainage. As a result, the residual cavity drains normally into the CBD through the biliocystic fistula. A surgical sphincterotomy is then routinely performed.

Compared with the original technique, surgical sphincterotomy is considered useless and dangerous [12], and the Tunisian teams who adopted transfistulary drainage did not perform it. Transfistulary drainage is an easy procedure that does not generate pericyst suture leakage risks and has good results, provided its modalities and its contraindications are respected [12].

Residual cavity evacuation and cleaning under visual control are effective for leaving no residual hydatic material in the cavity. Intralamellary pericystectomy in the case of a multistratified or calcified pericyst is essential. The protruding dome can be partly resected, but this resection must always allow suturing the pericyst without tension. Neither cholangitis nor infected contents in the cyst are contraindications. The same holds true for multiple fistulas in one cyst or multiple cysts opening into the biliary tract in one patient, and for the incline of a fistula in relation to the residual cavity [12]. The most important condition for use of the method is the fistula width, which must be > 5 mm and at best reach 1 cm or more [12, 50]. If the fistula diameter is smaller, this method is contraindicated and is replaced by another. This method should not be used when the cyst is voluminous, above 1 liter [12], or when the multistratified or calcified pericyst cannot be made supple, because in this case pericyst fragments can detach later and obstruct the fistula [12]. Moreover, some posterior hepatic cysts that cannot be normally explored under visual control and pericysts that cannot be correctly studied constitute contraindications to the method [12]. This method can be applied in only about 30% of HCLs with LBCFs [12].

The postoperative course after this technique is simple: The only SC observed in 52 cases was an external biliary fistula through the T-tube cutaneous opening. The postoperative controls by cholangiography through the T-tube and especially by repeated ultrasonography scan have shown a progressive, rapid reduction in the residual cavity, which collapsed and totally disappeared, leaving a linear scar that was echogenic at ultrasonography [12]. The T-tube was removed between the eighth and twelfth days. This same technique can be used when the HCL is the site of a large fistula without hydatic material in the CBD at intraoperative cholangiography. In these cases, the CBD was not approached as it has been the case in 15 of the 52 patients in whom it was performed in this series. Internal transfistulary drainage gives better results than the unroofing procedure with external drainage of the residual cavity when these unroofing procedures are analyzed globally. As a matter of fact, among the unroofing procedures, the results were markedly improved by biliocystic disconnection.

Other therapeutic methods based on biliocystic disconnection can be used with the unroofing procedure. This disconnection can be performed by suturing or by intubating the fistula, consisting in this last case in directed fistulization of the fistula.

Suturing the fistula has been supported by several authors [6, 13, 17, 21, 45, 47, 51]. It was performed 93 times in our series (38.1%) and was associated with external drainage of the residual cavity and, if necessary, residual cavity suture obliteration capitonnage or an omentoplasty. The postoperative course was uneventful in only half of the patients. In fact, the results of this method depend on the suturing modalities, which were not always specified in the retrospective operative reports. Suturing the fistula on sclerotic tissues is uncertain, whereas suturing the fistula in healthy tissues after perifistulary localized pericystectomy is probably more effective. The latter suturing mode is possible when it is a terminal fistula. When the cyst is central and the fistula is proximal or lateral, suturing is more difficult and must be performed carefully to avoid worsening the bile duct lesion. Moreover, the larger the fistula, and the more difficult is the suturing.

In our series the exact suturing modalities were not always specified.

Intubation or directed fistulization of the fistula (originally called transparietohepatic choledochostomy) was initially described by Perdomo et al. [52]. It derives from the Praderi technique [53], which is used by some authors in its initial conception [10, 16, 19].

Technical modifications have been brought about by Hamdouch et al. [19] and some of the Tunisian teams taking part in the study. Once the parasite is evacuated and the remaining cavity cleaned, a Nelaton catheter, the diameter of which is adapted to that of the fistula, is introduced through the fistula with rubbing. It is then pushed 3 to 4 cm into the biliary tract while respecting its nontraumatic rounded end and adding to it a small lateral opening so as not to obstruct the upstream bile flow in the case of a lateral fistula. Thus the CBD cannot be approached if it is free. In other cases it would be cleared and drained by a T-tube. This modification, introduced by the teams who used it, is easier and respects the original method imperatives (i.e., the fistula directed fistulization in transparietohepatic choledochostomy according to the Praderi technique) and the shortest intracystic distance of the intubation catheter. It has the advantage of avoiding the routine CBD opening. Moreover, there are fewer risks of twisting the drain when using a Nelaton catheter than with a T-tube.

In our series, this method produced results almost equivalent to those seen with fistula suturing. It is mainly used in the case of limited-diameter fistulas (4 mm) that cannot be treated with internal transfistulary drainage. Some teams rejected it because it necessitates drainage according to the Praderi technique and runs the risk of catheter twisting or displacement. When performed following a rigorous technique, it constitutes a safe biliocystic disconnection method. Although, some authors [10, 19] support this method's efficiency, it is obvious it cannot be considered the basic treatment for large fistulas. It constitutes only an alternative method in the case of voluminous multilocular cysts or cysts with calcified pericysts or when the fistula is either central and cannot be sutured, or so small it cannot be used for internal drainage. In our series, 10% of the large fistulas were treated using this method.

Once the LBCF was disconnected by suturing or intubation, the remaining cavity problem arises. The residual cavity may be the site of associated small biliary fistulas; and the problem of simple external drainage or filling-in by omental packing or, even more, capitonnage or hepatic introflexion remains to be resolved. In our series, simple external drainage has given significantly less satisfying results, mainly with regard to SCs and the HS. Some authors have used capitonnage [16] and others omentoplasty [13, 16, 17, 21, 36, 39, 45]. A prospective randomized trial reported omentoplasty efficiency when it is rigorously performed and pediculate [54]. A second prospective study comparing omentoplasty with hepatic introflexion [51] concluded that omentoplasty was more appropriate. In our series, there was no significant difference between capitonnage and omentoplasty. It has been confirmed that use of a filling-in technique device omentoplasty or capitonnage significantly improved the postoperative course and shortened the HS (Table 11).

A comparison of the two treatment groups—internal transfistulary drainage, supported by some Tunisians teams [12, 14], and the unroofing procedure with fistula suturing associated with omentoplasty supported by several authors [6, 11, 13, 21, 47, 48]—showed a significantly lower SC rate when internal transfistulary drainage was performed (1.9% vs. 17.8%; $p = 0.01$). This result should be confirmed by a prospective study in which the fistula suturing modalities are well defined.

Evacuation of the cystic content through the CBD was necessary for central HCLs deeply embedded in the liver and opening directly into the biliary tract. It differs from transfistulary drainage, as the cystic cavity was not approached at the liver surface and was cleaned without visual control. It can be compared to transfistulary drainage by its final result (i.e., transfistulary and transcholedochal residual cavity drainage). In this kind of HCL, cystic content evacuation may be incomplete. Similarly, the biliary lesion on cholangiography is difficult to investigate because of superimposition of the biliary pool of contrast fluid in the cyst, making the fistula unreachable by choledochoscopy. The operation may be ended by biliodigestive anastomosis in the absence of complete cystic content evacuation confirmation (2/28 in our series), but it may also be ended by T-tube drainage of the CBD, as a possible residual obstacle can be evacuated by endoscopic sphincterotomy. This consists in spontaneous internal transfistulary drainage; and once CBD evacuation is performed, the remaining cavity progressively retracts [12]. This method was used out of necessity in about 10% of the cases in our series.

Concerning the nonoperability of the central cyst embedded in the liver, several authors reported either spontaneous recovery after complete evacuation of the principal biliary cyst content [55] or endoscopic sphincterotomy [56]. This may not be a generalized situation because of the risk of biliary cirrhosis after repeated cholangitis. In fact, ERCP and sphincterotomy were performed postoperatively for most HCLs ruptured in the biliary tract to evacuate residual material in our series and in other authors' experience [57–59]. During operation, ultrasonography can be impressive enough to recognize the hydatic nature of pseudotumor formation. It can even show the biliocystic fistula and the intraparenchymatous nature of the cyst, as was the case three times in our series. ERCP sometimes shows the biliocystic fistula [39, 59]. In fact, nonoperative perendoscopic treatment appears insufficient in a cholangitis and emergency context because the cystic cavity containing biliopurulent material is situated above the biliary convergence and cannot always be reached through perendoscopic nasobiliary drainage.

Some other problems concerning the biliary tract remain imperfectly investigated. There were no problems during CBD evacuation of hydatic debris. There were problems, however, with the gallbladder (i.e., cholecystectomy or not) and the CBD (i.e. drainage or not) with drainage by a T-tube, a cystic duct drain, or a biliodigestive anastomosis.

The pathologic gallbladder or the gallbladder not attached to the cyst did not raise any problems. Should the externally healthy gallbladder be systematically removed or left alone? In our series, the gallbladder was left in 30 cases, either after having been used for cholangiography or after having performed a cholangiography through a catheter introduced into the biliocystic fistula without consequence. In all these cases, the CBD was free of obstacles.

Should the CBD by systematically drained to diminish the biliary flow going to the residual cavity? This step is supported by some authors who recommended a cystic duct drain [48], a T-tube drain [3, 8, 60], or a biliodigestive anastomosis [4, 5]. It has not been fully demonstrated that CBD routine drainage diminishes

the external biliary fistula risk even if it diminishes its duration and gravity [2, 21]. Nor it is proven that CBD routine drainage by T-tube is associated with morbidity [2]. As for the cystic duct drain, it might only be used for postoperative cholangiography, knowing that no study has demonstrated associated morbidity. Interest in biliodigestive anastomosis with endoscopic sphincterotomy has been lost in most cases; and there are no indications for surgical sphincterotomy.

Whether the CBD had been drained did not make any difference in our series. We can expect improvement more from the quality of the biliocystic disconnection of the HCL with an LBCF than from biliary tract drainage. The T-tube does not avoid an external biliary fistula [2, 20, 21, 23, 49] even if it reduces its gravity [2].

With any method, the mortality rate varies between 3% and 7% [20, 21, 45] and the morbidity rate between 30% and 50% [10, 13, 20, 21, 39, 40, 45]. There is also a long HS, bordering on 30 days [2].

The difficulties encountered during assessment of surgery for HCLs with an LCBF were multifactorial. Most studies have been interested in HCLs in general and comprised few with an LBCD [2, 6, 16, 18, 20–22, 33, 37, 39, 44, 45, 47, 49], when in fact they are different entities based on the variability of the HCL's anatomic and clinical form. There were no published prospective studies that reported enough details regarding the methods used and taking into consideration precise anatomic data. There is no consensus on the anatomic classification used and on the precise description of the different modalities performed. Several authors [10, 18–23, 34, 42, 47] have supported an attitude based on a reduced number of techniques. They defend it with great conviction without taking into consideration the arsenal of therapeutic methods we have at our disposal. This arsenal should be well known, and several methods are sometimes of help.

Conclusions

The LBCF is a frequent anatomic and surgical accompaniment of hepatic hydatidosis. Its treatment is not yet established. A consensus should be established concerning the intraoperative anatomic lesions and precise descriptions of the various therapeutic methods.

Among these methods, none can be used in all cases. Despite the fact that hepatic resection and pericystectomy are efficient techniques, they may be indicated only for cysts on the left lobe and anterior side of the liver. Internal transfistulary drainage is an effective method when the following criteria are respected [12]: fistula diameter ≥ 5 mm and the remaining cavity diameter < 10 cm with a soft pericyst wall. The unroofing procedure with suturing of the biliocystic fistula and filling-in omentoplasty is also an effective method if the suturing is done in healthy tissue. Each of these methods has its own indications. Nevertheless, some cysts can be treated by either method, and a prospective study would allow better definition of their respective place. Directed fistulization necessitates longer hospitalization and is used only when other methods are contraindicated. Hitherto, no study has allowed us to state precisely the exact indications for CBD drainage or its principles and modalities.

Résumé

Les fistules biliokystiques larges (>5 mm) survenant dans les kystes hydatiques du foie ne sont accompagnées de manifestations cliniques que quand le contenu du kyste passe dans la voie biliaire principale. Elles posent plusieurs problèmes thérapeutiques. Le but de cette étude a été d'évaluer les résultats des différentes méthodes thérapeutiques utilisées par quatorze centres tunisiens dans le traitement de la fistule biliokystique large secondaire au kyste hydatique du foie. Cette étude concerne 244 cas observés pendant une période de 5 ans entre janvier 1988 et décembre 1992, parmi lesquels 158 avaient des éléments hydatiques dans la voie biliaire principale et deux avaient une lithiase biliaire (vésicule et voie biliaire principale). 127 patients ont eu une intervention en urgence (52%). Les procédés chirurgicaux ont consisté en un procédé radical (24 cas: 9.8%) ou en un procédé conservateur (220 cases: 90.2%). Parmi ces derniers on a relevé 52 cas de drainage transfistuleux interne. II y a eu 140 résections du dôme saillant, sans toucher à la fistule dans 20 cas, avec suture de la fistule dans 93 cas, et enfin, dans 27 cas, en employant un procédé de fistulisation dirigée. Dans les 28 cas restants, le parasite a été évacué par voie transcholédocienne. La voie biliaire principale a été abordée dans 180 cas (73.7%). L'évolution postopératoire a été satisfaisante dans 57% des cas et compliquée dans 38.5%. La mortalité a été de 4.5%. En conclusion: a) la présence ou pas de matériel hydatique dans la voie biliaire principale ne dicte pas le procédé chirurgical et n'influence pas les résultats. La seule difficulté dans le traitement est dans celui des fistules biliokystiques elles-mêmes. b) le drainage interne, transfistuleux, avec une résection du dôme saillant, associée soit à une omentoplastie soit à un capitonnage de la cavité résiduelle sont faciles et efficaces. c) les résultats des méthodes radicales sont excellents mais elles ne sont réalisables que dans 10% des cas.

Resumen

Las fístulas bilio-quísticas de gran calibre (>5 mm) que complican la evolución de los quistes hidatídicos hepáticos, sólo se manifiestan clínicamente cuando el contenido quístico drena al colédoco. En el trabajo se exponen diversos aspectos terapéuticos de esta complicación, evaluando los resultados de las diferentes terapias utilizadas en 14 centros hospitalarios de Túnez, para el tratamiento específico de los quistes hidatídicos hepáticos complicados con fístulas bilio-quísticas de gran calibre. Se estudian 244 casos recopilados en 5 años (enero 1988-diciembre 1992). En 158 casos el contenido quístico se había drenado al colédoco asociándose, en dos a colelitiasis o coledocolitiasis. Se intervinieron de urgencia 127 (52%) pacientes. El tratamiento quirúrgico fue radical en 24 (9.8%) casos y conservador en 220 (90.2%). Dentro de estos últimos casos en 52 se procedió al drenaje transfistular interno; en 140 pacientes se realizaron "capitonages" respetándose la fístula en 20 casos; en 93 se suturó la fístula y en 27 se realizó una fistulización directa. En los restantes 28 casos se extirpó por vía transcoledociana el parásito. Se abordó el colédoco en 180 casos (73.7%). No hubo complicaciones postoperatorias en el 57% de los casos, registrándose éstas en el 38.5% restantes. La mortalidad fue del 4%. Conclusiones: a) La presencia o ausencia de material hidatídico en el colédoco no desempeña papel alguno por lo que

a la elección de la técnica quirúrgica se refiere ni modifica los resultados. La única dificultad verdadera la constituye el tratamiento de la fístula. b) El drenaje transfistuloso interno acompañado de la oclusión, mediante sutura de la fístula por tejido sano, junto a la omentoplastia o "capitonage" de la cavidad residual son dos técnicas fáciles de realizar y muy eficaces. c) Los métodos quirúrgicos radicales, que proporcionan excelentes resultados, sólo pueden realizarse en el 10% de los casos.

Acknowledgments

We thank Professor Jaber Danguir, Ph.D. for helpful review of the English version of this manuscript. The surgeons who participated in this trial are as follows: Tarak Kilani (Ariana); Khiareddine Ben Salah (Kairouan); Hédi Balti, Sami Bouchoucha, Béchir Larabi (La Marsa); Mohamed Fodha, Khaled Sridi (Mahdia); Rachid Amamou (Menzel Bourguiba); Mohamed Hédi Tabka, Khédija Zouari (Monastir); Sadok Sassi, Hassen Touinsi (Nabeul); Tahar Abderrahim, Ridha Belhaj Hamida, Fethi Derbel, Mustapha Kehila (Sousse); Issam Beyrouti, Abderraouf Khiari (Sfax); Mourad Adala, Salem Belaid, Chadli Dziri, Adnène El Hachaichi, Karim Haouet, Mohamed Jouini, Tahar Khalfallah, Abdelmajid Letaief, Nabil Najah, Fethi Najjar, Farouk Sebai, Abdeljélil Zaouche (Tunis).

References

1. Tunisian Association for Surgery , A. Zaouche : La Chirurgie du K.H.F.: A Propos de 2013 Cas. Tunis, Edit. Association Tunisienne de Chirurgie, 1994, p. 351
2. Maillet, B., Le Treut, Y.P., Sabiani, P., Pol, B., Bricot, R.: Kyste hydatique du foie: analyse de la morbidité post-opératoire: a propos d'une série récente de cinquante-deux cas. Ann. Chir. *40*:389, 1986
3. Ulualp, K.M., Aydemir, I., Senturk, H., Eyuboglu, E., Cebeci, H., Unal, G., Unal, H.: Management of intrabiliary rupture of hydatid cyst of the liver. World J. Surg. *19*:720, 1995
4. Lygidakis, N.J.: Diagnosis and treatment of intrabiliary rupture of hydatid cyst of the liver. Arch. Surg. *118*:1186, 1983
5. Alper, A., Ariogul, O., Emre, A., Uras, A., Okten, A.: Choledochoduodenostomy for intrabiliary rupture of hydatid cysts of liver. Br. J. Surg. *74*:243, 1987
6. Safioleas, M., Misiakos, E., Manti, C., Katsikas, D., Skalkeas, G.: Diagnostic evaluation and surgical management of hydatid disease of the liver. World J. Surg. *18*:859, 1994
7. Sebai, F., Laarif, R., Sassi, S., Daoud, A., Mestiri, S.: Les angiocholites graves d'origine hydatique: aspects anatomo-cliniques et résultats thérapeutiques d'une expérience tunisienne. Med. Chir. Dig. *14*:133, 1985
8. Ovnat, A., Peiser, J., Avinoah, E., Barki, Y., Charuzi, I.: Acute cholangitis caused by rupture hydatid cyst. Surgery *95*:497, 1984
9. Kehila, M., Said, R., Alouini, T., Ben, H., Hamida, R., Khalfallah, A., Jerbi, A., Gharbi, S.: Eléments de gravité du kyste hydatique du foie ouvert dans les voies biliaries: a propos de 23 cas. 1977–1984. Tunis Med. *2*:121, 1987
10. El Alami El Farichaa, E.H., El Mahdi, T., Louchi, A., Echarrab, M., Ismail, P., Mohsine, R., Abassi, A., Chkoff, M.R., Errougani, A., Amraoui, M., Benchekroun, A., Balafrej, S.: La rupture du kyste hydatique du foie dans les voies biliaries: résultats du traitement chirurgical et place de la cholédocostomie trans-hépatico-kystique. Lyon Chir. *91*:408, 1995
11. Bourgeon, R.: L'ouverture des kystes hydatiques aux voies biliaires intra-hépatiques. Lyon Chir. *81*:161, 1985
12. Ennabli, E., Zaouche, A., Hachaichi, A.: Les kystes hydatiques du foie ouverts dans les voies biliaires: attitudes thérapeutiques. Ann. Chir. *39*:243, 1985
13. Bouzidi, A., Chehab, F.: Traitement chirurgical des fistules biliokystiques d'origine hydatique. J. Chir. *134*:114, 1997
14. Belkhodja, C., Mestiri, H., Ben Ammar, M.S., Larabi, B.: La chirurgie conservatrice du kyste hydatique du foie ouvert dans les voies biliaries. Tunis Chir. *71*:7, 1993
15. Testas, P., François, M.: La rupture des kystes hydatiques du foie dans les voies biliaires extra-hépatiques. Chirurgie *109*:122, 1983
16. Maaouni, A., El Alaoui, M., Hamiani, O., Benmansour, A., Belkouchi, A., Ahyoud, F., Jalil, A., Faik, M.: Chirurgie des kystes hydatiques du foie: 581 patients; 952 kystes. Chir. *115*:67, 1989
17. Paunescu, V., Ragalie, S., Mateescu, L.: Surgical treatment of pericyst in liver hydatid cysts. Acta. Chir. Belg. *95*:44, 1995
18. Berrada, S., Essadki, B., Zerouali, N.O.: Kyste hydatique du foie: traitement par résection du dôme saillant; notre expérience à propos d'une série de 495 cas. Ann. Chir. *47*:510, 1993
19. Hamdouch, Z., Halhal, A., M'Jahed, A., Oudanane, M., Chekoff, M.R., Foucou, B., Hosni, K., Tounsi, A.: La déconnection kysto-biliaire dans le traitement du kyste hydatique fistulisé dans les voies biliaires. Maroc Med. *8*:309, 1986
20. Chigot, J.P., Langlois, P., Teboul, F., Clot, J.P., Gentilini, M., Mercadier, M.: Le traitement des kystes hydatiques du foie: a propos de soixante dix-sept observations. Ann. Chir. *40*:177, 1986
21. Grosdidier, J., Boissel, P., Bresler, L., Nesseler, J.P., Guirlet, M.N.: Etude rétrospective d'une série de 62 cas de kystes hydatiques du foie. J. Chir. *112*:163, 1985
22. Moumen, M., El Alaoui, M., El Mokhtari, M., El Fares, F.: Les kystes hydatiques du foie: a propos de 670 cas dont 552 compliqués. Sem. Hop. Paris *69*:722, 1993
23. Mokhtari, M., Kadiri, B., Khalidi, M., Gatra, A., A., Zaoui, A., Mansouri, A., : Résultats de la chirurgie "conservatrice" de résection du dôme saillant dans le kyste hydatique du foie: a propos de 183 cas. Lyon Chir. *82*:87, 1986
24. Chipponi, I., Huguier, M.: Les kystes hydatiques du foie opérés en France: epidémiologie, attitudes diagnostique et thérapeutique; etude de 306 cas. Gastroenterol. Clin. Biol. *55*:419, 1984
25. Huguet, C., Nordlinger, B., Zaouche, A., Quilichini, M.A., Hakami, F.: Traitement chirurgical des kystes hydatiques du foie. Med. Chir. Dig. *15*:43, 1986
26. Akhan, O., Ozmen, M.N., Dincer, A., Sayek, I., Gocmen, A.: Liver hydatid disease: long-term results of percutaneous treatment. Radiology *198*:259, 1996
27. Filice, C., Brunetti, E.: Use of PAIR in human cystic echinococcosis. Acta Trop. *64*:95, 1997
28. Khuroo, M.S., Wani, N.A., Javid, G., Khan, B.A., Yattoo, G.N., Shah, A.H., Jeelani, S.G.G.: Percutaneous drainage compared with surgery for hepatic hydatid cysts. N. Engl. J. Med. *337*:881, 1997
29. Alper, A., Emire, A., Hazar, H., Ozden, I., Bilge, O., Acarli, K., Ariogul, O.: Laparoscopic surgery of hepatic disease: initial results and early follow-up of 16 patients. World J. Surg. *19*:725, 1995
30. Massoud, W.Z.: Laparoscopic excision of a single hepatic hydatic cyst. Int. Surg. *81*:9, 1996
31. Little, J.M., Holland, M.J., Ekberg, H.: Recurrence of hydatid disease. World J. Surg. *12*:700, 1988
32. Settaf, A., Mansori, F., Sefrioui, A., Slaoui, A.: Kystes hydatiques du foie: classification à visée thérapeutique et pronostique: 378 observations. Presse Med. *23*:362, 1994
33. Rohner, A.: Traitement du kyste hydatique du foie. Ann. Chir. *42*:635, 1988
34. Belli, L., Del Favero, E., Marni, A., Romani, F.: Resection versus pericystectomy in the treatment of hydatidosis of the liver. Am. J. Surg. *145*:239, 1983
35. Behrns, K.E., Van Heerden, J.A.: Surgical management of hydatid disease. Mayo Clin. Proc. *66*:1193, 1991
36. Tompkins, R.K.: Management of echinococcal cysts of the liver. Mayo Clin. Proc. *66*:1281, 1991
37. Dugalic, D., Djukic, V., Milicevic, M., Stevovic, D., Knezevic, J., Pantic, J.: Operative procedures in the management of liver hydatidosis. World J. Surg. *6*:115, 1982
38. Mentouri, B., Roche, P., Hammami, A., Hammad, A., Meradji, B.: La place de la périkystectomie dans le traitement du kyste hydatique du foie. Chirurgie *101*:882, 1975
39. Pitt, H.A., Korzelius, J., Tompkins, R.K.: Management of hepatic echinococcosis in southern California. Am. J. Surg. *152*:110, 1986
40. Langer, J.C., Rose, D.B., Keystone, J.S., Taylor, B.R., Langer, B.:

Diagnosis and management of hydatid disease of the liver: a 15-year North American experience. Ann. Surg. *199*:412, 1984
41. Langer, B.: Surgical treatment of hydatid disease of the liver. Br. J. Surg. *74*:237, 1987
42. Alfieri, S., Doglietto, G.B., Pacelli, F., Costamagna, G., Carriero, C., Mutignani, M., Liberatori, M., Crucitti, F.: Radical surgery for liver hydatid disease: a study of 89 consecutive patients. Hepatogastroenterology *44*:496, 1997
43. Kayabali, I., Kayabali, M., Kaynac, A., Bac, B., Keles, C., Salih, M., Acar, H.: Sur le diagnostic et le traitement des kystes hydatiques du foie. Lyon Chir. *80*:186, 1984
44. Demirci, S., Eraslan, S., Anadol, E., Bozatli, L.: Comparison of the results of different surgical techniques in the management of hydatid cysts of the liver. World J. Surg. *13*:88, 1989
45. Sayek, I., Yalin, R., Sanac, Y.: Surgical treatment of hydatid disease of the liver. Arch. Surg. *115*:847, 1980
46. Dawson, J.L., Stamatakis, J.D., Stringer, M.D., Williams, R.: Surgical treatment of hepatic hydatid disease. Br. J. Surg. *75*:946, 1988
47. Aktan, A.O., Yalin, R., Yegen, C., Okboy, N.: Surgical treatment of hydatid cysts. Acta Chir. Belg. *93*:151, 1993
48. Salem, A., Ayadi, S., Bereksi, D., Bensafar, S.: La périkystectomie partielle dans le traitement du kyste hydatique du foie à périkyste souple: a propos de 142 cas. Chirurgie *114*:502, 1988
49. Ergüney, S., Tortum, O., Taspinar, A.H., Ertem, M., Gazioglu, E.: Les kystes hydatiques compliqués du foie. Ann. Chir. *45*:584, 1991
50. Goinard, P., Pegullo, J., Pelissier, G.: Sur la sténose oddienne consécutive à l'ouverture des kystes hydatiques du foie aux voies biliaires: déductions opératoires. Mem. Acad. Chir. *84*:619, 1958
51. Mentes, A., Yuser, Y., Ozbal, O., Coker, A., Ilter, T., Musoglu, A.: Omentoplasty versus introflexion for hydatid liver cysts. J. R. Coll. Surg. Edinb. *38*:82, 1993
52. Perdomo, R., Morelli, R., Carriquiry, L., Chifflet, J., Bergalli, L.: Cholédocostomie trans-hépatico-kystique en cas de kyste hydatique ouvert dans les voies biliaires. Nouv. Presse Med. *6*:747, 1977
53. Praderi, R.: Twelve years experience with transhepatic intubation. Ann. Surg. *179*:937, 1974
54. Dziri, C., Paquet, J.C., Hay, J.M., Fingerhut, A., Msika, S., Zeitoun, G., Sastre, B., Khalfallah, T.: Omentoplasty in the prevention of deep abdominal complications after surgery for hydatid disease of the liver: a multicenter, prospective, randomized trial. J. Am. Coll. Surg. *188*:281, 1999
55. Becker, K., Frieling, T., Saleh, A., Haussinger, D.: Resolution of hydatic liver cyst by spontaneous rupture into the biliary tract. J. Hepatol. *26*:1408, 1997
56. Al Karawi, M.A., Mohamed, A.R.E., Yasawi, I., Haleem, A.: Non-surgical endoscopic trans-papillary treatment of ruptured echinococcus liver cyst obstructing the biliary tree. Endoscopy *19*:81, 1987
57. Vignote, M.L., Mino, G., de La Mata, M., de Dios, J.F., Gomez, F.: Endoscopic sphincterotomy in hepatic hydatic disease open to the biliary tree. Br. J. Surg. *77*:30, 1990
58. Shemesh, E., Klein, E., Abramowich, D., Pines, A.: Common bile duct obstruction caused by hydatid daughter cysts: management by endoscopic retrograde sphincterotomy. Gastroenterology *81*:280, 1986
59. Ponchon, T., Bory, R., Chavaillon, A.: Endoscopic retrograde cholangiography and sphincterotomy for complicated hepatic hydatid cyst. Endoscopy *19*:174, 1987
60. Kornaros, S.E., Aboul-Nour, T.A.: Frank intrabiliary rupture of hepatic cyst: diagnosis and treatment. J. Am. Coll. Surg. *183*:466, 1996