Research Article

Surgical treatment of hydatid cysts with large biliocystic fistula results from a monocentric surgical experience

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ABSTRACT

Background: Hydatid cyst opened in the biliary tract constitutes an evolutionary turning point in the hydatid disease and risks to be life-threatening. Liver hydatid cysts with large biliocystic fistula is defined by the presence of a biliocystic fistula which is more than 5 mm in diameter and / or the presence of hydatid material in the common bile duct. The diversity of the pathological lesions of this disease and the lack of randomized controlled trials studying the different therapeutic techniques make those attitudes varied and sometimes debated. The purpose of this study was to study the different therapeutic possibilities as well as the difficulties posed by the large biliocystic fistula.

Methods: This is a retrospective study of all consecutive patients (n=51), operated on for liver hydatid cysts with a large biliocystic fistula, between January 2003 and December 2012. The average age in this series was 39 years. There were 21 men and 30 women. An acute cholangitis was present in 47% of cases.

Results: The treatment of hydatid cyst was conservative (cyst unroofing) in 90% of cases and radical in 10% of cases. The large biliocystic fistula was treated by internal transfistular drainage (21%), cystobiliary disconnection by trans-parieto-hepatocystic fistulization (35%), bipolar drainage (25%), direct suture of the fistula (8%) and liver resection (10%). The specific morbidity rate was 27% (n = 14). No deaths have been observed. The average follow-up was 22 months. Six cases of recurrence were observed.

Conclusions: The treatment of intrabiliary rupture of echinococcal cysts is not consensual. The internal transfistular drainage and the trans-parietohepatocystic fistulization represent the techniques of choice in case of conservative surgery. Prevention remains the best treatment.

Keywords: Hydatid cyst, Large biliocystic fistula, Surgery, Morbidity

INTRODUCTION

Hydatid cyst of the liver (HCL) is still a major public health problem in Tunisia, which has serious consequences both economically and sanitarilly. Indeed, despite the efforts developed for decades in the field of primary prevention of this disease, our country still ranks among the areas of high hydatid endemicity. Among the

complications that may arise, the opening in the biliary tract constitutes an evolutionary turning point in its natural history.¹⁻³ Hydatid cyst of the liver largely opened in the bile ducts is defined by the presence of a large biliocystic fistula (LBCF), of which the size is greater than or equal to 5 mm^{1,4} which allows two thirds of the cases the migration of small daughter vesicles or membranes into the bile ducts.¹ The therapeutic choices

deserve to be well structured and codified. Surgical treatment of this complication is varied and object of controversies between a radical treatment that can treat at the same time the cyst and the fistula, and conservative treatment with several techniques. The aim of our work was to study the therapeutic difficulties posed by the LBCF.

METHODS

It is a retrospective study of all the patients who underwent surgery for hydatid cysts of the liver largely opened in the bile ducts, treated at the Department of General and Digestive Surgery of Farhat Hached University Hospital of Sousse (Tunisia). We held the cases where the bilio-cystic fistula was confirmed intraoperatively, either by the visual examination of the residual cavity, or by using intraoperative cholangiography. The LBCF was defined by a diameter upper or equal to 5 mm, or by the presence of hydatid material in the biliary tract. We were interested in the files of all the patients operated on between January 1st 2003 and December 31st, 2012. We didn't include hydatid cysts with intrathoracic and intraperitoneal rupture. All individual data were collected on data collection forms, containing 146 items. We used the Microsoft Excel spreadsheet for statistical analyses. A descriptive analysis was performed for all parameters.

During that period, we listed 498 patients operated for hydatid cysts of the liver, among whom 51 were operated for liver hydatid cysts largely opened in the biliary ducts (10% of all HCL). The average age was 39 years with extremes of 17 and 79 years. They are distributed in 21 men and 30 women (Sex-ratio of 0.7). No patient underwent a first-line endoscopic retrograde cholangiopancreatography (ERCP). 36 patients (71 %) underwent an emergency operation. The size of the hydatid cysts varied between 3 cm and 30 cm with an average size of 11 cm. The contents of the cyst were bilious in 34 cases (64%), purulent in 10 cases (20%) and gelatinous in 8 cases (16%). A single LBCF was found among 50 patients (96%) and in 1 patient, we have noticed the presence of 2 large fistulas on the same cyst. Its size was between 5 mm and 12 mm. The biliocystic fistula was judged by terminal nature in 25 cases (46%) and lateral nature in 15 cases (28 %). In 12 cases (26%), the nature of the fistula was not specified. There were 20 simple hepatic cysts and 3 intraperitoneal hydatid cysts.

The Table 1 resumes the clinical, biological, radiological and intraoperative characteristics of our series.

RESULTS

The Unroofing procedure (Figure 1a) was performed on 46 HCL (90% of cases). Drainage of the residual cavity was realized to 18 patients in our series (39%). Some technical devices associated to the unroofing procedure were made. Omentoplasty filling of the remaining cavity

(Figure 1b) was done in 18 cases (39%), pericystorrhaphy (Figure 1c) in 3 cases (6%) and intralamellary pericystectomy (Figure 1d) in 10 cases (22%). Subtotal pericystectomy was conducted for 1 HCL (2.2%) in contact with the inferior vena cava. Liver resection was performed on 5 cases. They consisted in a left hepatectomy, a left lobectomy, a right hepatectomy, and 2 pericystoresection. Cholecystectomy was performed in all patients. Intraoperative cholangiography was performed in 42 patients (82%) through a cystic duct drain. It was considered normal in 11 cases (26%), abnormal in 30 cases (71.4%) and inconclusive in one case (2%). Cholangiography allowed viewing the large biliocystic fistula in 24 cases (57%). It was used to study the bile ducts showing lacunar images within the common bile duct (CBD) in 8 cases (19%), dilatation of the CBD in 17 cases (40%) and dilated intrahepatic bile ducts (IHBD) in 7 cases (16.7%). Twenty-seven patients (53%) had had a choledochotomy. In 13 cases (48%), we extracted hydatid material from the CBD. In all cases, the CBD was closed on a T-drain.

Figure 1: Conservative treatment of the liver hydatid cyst and associated devices; Figure 1a: cyst unroofing; Figure 1b: Drainage and omentoplasty in the remaining cavity; Figure 1c: Pericystorrhaphy “capitonnage”; Figure 1d: Intralamellary pericystectomy.

The internal transistulary drainage (Figure 2a) was performed on 11 cases (21%) ,for posterior HCL in 7 cases, for Centro-parenchymal hydatid cysts in 3 cases and for a hydatid cyst of the left lobe measuring 7 cm in 1 case. Bipolar drainage was used in 13 cases (25%). All patients underwent drainage of the large biliocystic fistula with (8 cases) or without (5 cases) omentoplasty. The biliary drainage was performed by a cystic duct drain in 5 cases and a T-drain in 8 cases. This method was

carried out for 9 posterior cysts of the right liver and 4 cysts of the left lobe. Cystobiliary disconnection by directed fistulization (Figure 2b) was carried out in 18 cases (35%) among patients having lateral fistulas with a thick pericyst. In 8 cases, the size of the cyst exceeded 10 cm. In 4 cases of superficial cysts with flexible pericyst and a terminal fistula (8%), a direct suture of the fistula by slowly absorbable wire was performed. It has been associated with omentoplasty in 3 cases and drainage of the residual cavity in one case.

Figure 2: Management of the large bilocystic fistula after conservative treatment. (2a) internal trans fistulary drainage. It consists in a partial pericystectomy reducing the size of the remaining cavity. The pericyst is then sutured and a T-tube is inserted in the common bile duct. (2b) Transparieto hepatocystic fistulization. This procedure uses an adapted tube introduced with rubbing through the fistula and pushed into the biliary ducts 3 to 4 cm. Then, it is exteriorized through sufficient hepatic tissue thickness with a short intracystic course (3 cm). The common bile duct is approached (T-drain) and the remaining cavity is also drained.

Postoperative cholangiography was carried out among 49 patients (96%), through a T-drain in 27 cases and a cystic duct drain in 22 cases. In 2 cases, the patients had no bile drainage. The cholangiography was made 7 days after the intervention on average (2 to 10 days). Thirty-five patients had an uneventful postoperative course (69%). Sixteen patients (31%) had complications. They were 4 non-specific complications and 14 specific complications (Table 2). An external biliary fistula was declared among 5 patients (11%). Four fistulas dried up spontaneously after a mean period of 3 weeks. In one patient, the depletion of the fistula was obtained after performing ERCP with endoscopic sphincterotomy. Three patients had a suppuration of the residual cavity. This has followed unroofing with bipolar drainage in the 3 cases. In 2 cases, the abscesses were treated by irrigation aspiration associated with an adapted antibiotherapy. In the other case, ultrasound-guided percutaneous drainage allowed the evacuation of the collection. A postoperative choleperitonitis occurred to 2 patients who underwent reoperation. Finally, a residual obstacle on the CBD was observed in 4 patients. They had ERCP which objectified the presence of hydatid material in the CBD. An

endoscopic sphincterotomy was carried out allowing the extraction of hydatid debris. The average length of postoperative stay was 14 days (5 to 41 days). The total hospital stay was between 6 and 42 days with an average of 15 days.

The median follow up of our patients was 26 months, with extremes of 6 and 60 months. Ten patients received a postoperative medical treatment by Albendazole. The duration of the treatment varied between 3 and 12 months with a mean of 5 months. The hydatid immunology was performed after one year and a half on average in 25 cases. It was positive in 16 cases and negative in 9 cases. We identified 6 cases of hydatid recurrence after a mean period of 20 months (15-34 months). In all these cases, hydatid immunology was positive. Recurrences were hepatic in 5 cases and peritoneal in one case. Of these, 3 patients were reoperated. The others received medical treatment.

DISCUSSION

Conservative treatment is the most used technique in the treatment of hydatid cysts of the liver with LBCF. Internal trans fistulary drainage and in the second place the cystobiliary disconnection by directed fistulization represent the treatment of choice for large bilocystic fistula, with a lower morbidity and a shorter hospital stay.

Despite the contribution of medical therapy, percutaneous and endoscopic treatments, treatment of HCL largely opened in the biliary ducts remains essentially surgical.^{5,6} The surgery of HCL with a LBCF has several targets: the parasite, the pericyst (with the problem of the residual cavity), the bilocystic fistula and the obstruction of the cbd.⁷ Radical treatment allows the management of both the cyst and the fistula, while avoiding having a residual cavity. However, it exposes to a high morbidity. Benignity of this disease and its endemic state in our mediums make conservative methods our first choice.⁸ The radical methods treat both the residual cavity and the LBCF. In case of conservative methods, a specific treatment of the LBCF is required. The attitude towards the LBCF depends on the topography of the fistula and on the location of the hydatid cyst. The LBCF is mostly treated at the same time as the hydatid cyst, or rarely at a second time when it had been passed unnoticed or deliberately neglected by the operator.⁴ Internal trans fistulary drainage is a process reported, for the first time in 1985 by Goinard.² The works of Ennabli allowed to update this process and to specify its modalities and indications.^{9,10} It consists in a partial pericystectomy reducing the size of the residual cavity. The pericyst is then sutured and a T-tube is inserted in the CBD. This technique constitutes the most effective method according to Zaouche.⁴ Cystobiliary disconnection by directed fistulization (transparietohepatocystic fistulization) was described by Perdromo and praderi.¹¹ This procedure uses an adapted tube introduced with rubbing through the fistula and pushed into the biliary

ducts 3 to 4 cm. Then, it is exteriorized through sufficient hepatic tissue thickness with a short intracystic course (3 cm). The common bile duct is approached (T-drain) and the remaining cavity is also drained. The T-tube should be kept between 4 and 6 weeks. It could be withdrawn after a cholangiography. Despite its good results (low mortality and morbidity), this method requires a long hospital stay to achieve the exclusion of the fistula.¹² In the literature, this method was used in 16.6 to 24% of cases.¹³ Bipolar drainage presents several complications,

especially prolonged external biliary fistulas. This technique was performed in 36% of the cases for Daali and 38% of the cases for Zaouche.^{12,4} Its high morbidity has led authors to condemn this technique. The suture of the orifice of the fistula by a slowly absorbable wire should be performed with a normal hepatic parenchyma or a soft pericyst. It should be banned in case of thick pericyst, deep or fistula close to the biliary convergence.¹⁴ It may be associated to an omentoplasty.

Table 1: Clinical and diagnostic characteristics of patients.

	Characteristics	Number / Percentage
History	Medical history	6 (1 diabetes, 2 hypertension, 3 coronary heart disease)
	Surgery for hydatid cysts	11 (6 liver, 4 right lung, 1 spleen)
Clinical symptomatology	Asymptomatic	3 (6 %)
	Cholangitis	24 (47 %)
	Abdominal pain	10 (20%)
	Hepatomegaly	5 (10%)
	Fever	9 (18 %)
Biology	Hyperleukocytosis	23 (45%)
	Positive blood culture	8/15 (53%)
	Cholestasis	25 (49%)
	Hepatic cytolysis	17 (33%)
	Hepatic failure	6 (12%)
	Renal failure	1 (2%)
	Hydatid serology (ELISA)	Positive in all cases (16 patients)
Radiology	Chest radiography (indirect signs)	12 (23%)
	Fistula's diagnosis by ultrasonography	5/49 (10%)
	Fistula's diagnosis by CT	11/21 (52%)
Cyst's location	Right liver	27 (53%)
	Left liver	11 (21%)
	Bilateral	13 (25%)
Fistula's location	Right ducts	37 (72%)
	Left ducts	14 (27%)

Table 2: Postoperative course according to treatment of the hydatid cyst of the liver and to management of the large biliocystic fistula.

Postoperative course	Radical (n=5)	Conservative (n=47)			
		Internal transistifistulary drainage (n=11)	Directed fistulization(n=18)	Bipolar drainage (n=13)	Suture (n=4)
Uneventful	4	10	15	4	0
Specific complications	0	1	2	8	3
EBF	0	0	0	3	2
RC suppuration	0	0	0	3	0
CBD obstacle	0	1	1	2	0
Choleperitonitis	0	0	1	0	1
Non-specific complications	1	0	1	1	1
Wound infection	0	0	1	0	1
Medical complications	1	0	0	1	0
Reoperation	0	0	1	0	1
ERCP	0	1	1	3	0
Total hospital stay (days)*	12	8.7	17.2	23	12

EBF: external biliary fistula; RC: residual cavity; CBD: common bile duct; ERCP: endoscopic retrograde cholangio pancreatography.*: mean.

Table 3: Frequency of specific complications after treatment of liver hydatid cysts with large biliocystic fistula according to the literature.

Complications	Zaouche ⁴	Ennabli ¹⁰	Daali ¹²	Medarh ¹³	Moume ¹⁸	Bouzidi ¹⁹	Our series
EBF	1.3%	15.2%	13.1%	19.3%	24.2%	14%	11%
RC suppuration	12.7%	8%	0	0	16.2%	0	6%
CBD obstacle	2.4%	0	0	0	0	0	8%
Choleperitonitis	0.4%	0	0	0	0	1.2%	4%

EBF: external biliary fistula; RC: residual cavity; CBD: common bile duct.

Table 4: Morbidity and mortality depending on surgical procedures treating liver hydatid cyst with large biliocystic fistula.

Series	Type of treatment	Number of patients	Specific complications	Mortality	Hospital stay (days)
Zaouche ⁴	Conservative	220	32.7%	5%	20*
	Radical	24	2%	0	15*
Daali ¹²	Conservative	54	24.7%	3.1%	15 to 60
	Radical	10	0	0	NS
Medarhri ¹³	Conservative	74	31%	1.3 %	24.3*
	Radical	14	0	0	11.6*
Our series	Conservative	46	25%	0	21*
	Radical	5	0	0	12*

*: mean; NS: not specified.

Table 5: Comparison of different techniques managing the large biliocystic fistula after conservative treatment of the liver hydatid cyst.

Series	Technique (number)	Specific morbidity	Mortality	Hospital stay (days)*
Zaouche ⁴	Internal trans fistulary drainage (52)	1.9%	1.9%	15
	Suture (93)	41.3%	5.3%	20
Daali ¹²	Directed fistulization (15)	0	0	27
	Bipolar drainage (24)	4.7%	1.5%	20
	Suture (23)	20.3%	1.5%	15
Medarhri ¹³	Directed fistulization (52)	15.4%	0	25
	Bipolar drainage (17)	63%	0	30
Baraket ²⁰	Internal trans fistulary drainage (8)	0	0	9.5
	Directed fistulisation (6)	0	0	20
	Bipolar drainage (6)	13.6%	0	25
	Suture (2)	4.5%	4.5%	10.5
Moujahid ²¹	Directed fistulization (49)	0	0	28
	Bipolar drainage (25)	5.8%	1.7%	30
	Suture (36)	16.7%	0.8%	40
Our series	Internal trans fistulary drainage (11)	2%	0	8.7
	Directed fistulization (18)	4%	0	17.2
	Bipolar drainage (13)	16%	0	23
	Suture (4)	6%	0	12

*: mean.

The mortality rate varies according to the series from 0 to 6%.^{4, 12,13,15-17} Medical complications are between 1.6% and 7%, and are dominated by respiratory complications

and urinary tract infections.⁴ Non-specific surgical complications are dominated by wound infection encountered in 10-18% of cases. Evisceration of a mechanical or septic origin, non-biliary postoperative

peritonitis, an infection of a badly drained sero-hematic collection can also occur as in any abdominal surgery.⁴ Specific complications are represented by external biliary fistula, suppuration of the residual cavity, residual obstacle in the CBD and postoperative choleperitonitis.^{4,10,12,13,18,19} Table 3 summarizes these complications. The comparison between the results of conservative and radical gestures (Table 4) shows that radical interventions undoubtedly had lower specific morbidity and lower mortality.^{4,12,13} Considering the conservative treatment, which is the most used in endemic areas, the internal transistulary drainage and, in second place, the Perdromo's technique are the procedures that have been proven in terms of morbidity, mortality and the length of hospital stay.^{4,12,13,20,21} On the other hand, the bipolar drainage gave poor results (Table 5).

CONCLUSION

The diversity of the pathological lesions of HCL largely opened in the biliary ducts and the lack of randomized controlled trials studying the different therapeutic techniques make that attitudes varied and sometimes debated. This poses, therefore, a problem of operational tactics and surgical repair. Therapeutic strategy depends on several factors such as the characteristics of the cyst, type of fistula, the technical platform and the surgeon's experience. This treatment is still controversial among the followers of radical methods and those who advocate conservative methods.

It is necessary to avoid the simple bipolar drainage which has a bad reputation leading long and difficult courses. The single suture within a thick pericyst or inflammatory tissue is no longer effective. The internal transistulary drainage remains, by respecting the indications, the technique of choice in the countries where the hydatid cyst is endemic. Transparietohepatocystic fistulization keeps indications inspite of its disadvantages, in particular an increase in the duration of the hospital stay. Radical techniques are the treatment of choice among non-infected patients with a good general condition and whose cyst had destroyed an entire lobe or a hemi- liver. They allow settling the problem of the cyst and fistula, but remain reserved for experienced hepatobiliary surgery centers.

Also let us note that the endoscopic treatment has an important role, either prior to the surgery in severe cholangitis, or as definitive gesture in the central cysts where it can compete with the internal transistulary drainage.

Finally, the best treatment remains preventive, addressed to all the chain links of transmission of this anthroponozoonosis.

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