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Angiocholitis Due to Hydatid Cyst of the Liver Fistulized in the Bile Ducts: Comparative Study between Surgical Treatment and Endoscopic Treatment

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Abstract **Original Research Article**

Background: Liver hydatid is a parasitic disease considered benign but can become dangerous because of its complications. The fistula in biliary tract remains one of the most feared complications by its frequency, morbidity and mortality. ***Materials and methods***: We present a descriptive and comparative retrospective study at the university hospital Hassan II of Fez. This study was carried out on 63 cases of angiocholitis on hydatid cyst of the liver fistulized in the bile ducts and extends over a period of 5 years from January 2015 to December 2019. The objective of this study is to evaluate and compare the contribution of different therapeutic modalities in the treatment of cystic biliary fistula (Surgical treatment against endoscopic treatment). ***Results:*** Surgical treatment was used in 22 patients against 41 patients who underwent endoscopic treatment. The success rate of endoscopic treatment was higher than that of surgical treatment (83% against 73 %, p = 0.350). The mortality rate was reduced in the endoscopy group compared with patients in the surgery group (5% against 14 %, p = 0.226). General morbidity and major morbidity were higher significantly in patients treated surgically (General morbidity: surgery 41% against 2% for endoscopy, P0.05). ***Conclusion***: Endoscopic treatment should be considered as the treatment of choice for angiocholitis due to hydatid cyst of the liver fistulized in the bile ducts.

Keywords: Angiocholitis – Bilio cystic fistula – ERCP – Surgery.

BACKGROUND

Hydatid cyst of the liver is a parasitic disease caused by Echinococcus granulosus. It is considered as an endemic disease in most parts of the world, especially in the Mediterranean where sheep husbandry is common, and remain an important medical health problem in these regions. Hydatidosis may remain asymptomatic for a long time, but can also cause serious complications. Common complications of hydatid cyst of the liver include rupture into intrahepatic bile ducts or the peritoneal cavity, invasion of other organs, pressure on the biliary tree and other neighboring structures, and infection. The communication between the cyst and the biliary tree is the most common and serious complication. Most of patients presenting cholangitis due a hydatid cyst fistula are young and usually have severe cholangitis. The management of hydatid cyst ruptured into the biliary tract is not consensual. Although surgery is the classic method of treating bilio cystic fistula, endoscopic retrograde cholangiopancreatography (ERCP) has emerged as an alternative choice for the management of those complications. The goal of our study is to compare the two methods (surgery vs endoscopy) based on the experience in our center and comparing it to the results in the literature.

METHODS

Our series is a retrospective descriptive and comparative study, performed at the department of general surgery, gastroenterology and intensive care unit of the university hospital Hassan II in Fez. This study covers a period of 5 years from January 2015 to December 2019.

In this study we included all patients admitted for angiocholitis due to a Cyst biliary communication confirmed by ultrasound or CT scan and having undergone surgical or endoscopic treatment. We excluded angiocholitis due to a compressing of the bile ducts, uncomplicated hydatid cyst, hydatid cyst with a complication other than bilio-cystic fistula and Incidental discovery of bilio-cystic fistula during hydatid cyst surgery because they were asymptomatic.

We report in this study 63 cases of angiocholitis due to a Cysto-biliary communication which have been divided into two groups:

Group 1: Patients who received surgical treatment (N = 22);

Group 2: Patients who received endoscopic treatment (N = 41)

For the surgical treatment, three techniques were used:

- Direct suture of the fistula, indicated in small and lateral fistula. This method is contra-indicated when the pericyst is thick, or the fistula is deep or near the biliary convergence.
- Bipolar drainage (consist after resection of the protruding dome, in locating and suturing the fistula, the common bile duct is drained by a T tube and the residual cavity by a large drain).
- Cystobiliary disconnection by percutaneous hepatic choledochostomy: after resection of the protruding dome the cystobiliary trajectory is intubated via a choledochotomy and the drain exits through the skin after traversing a healthy portion of liver. The common bile duct is drained by a T tube and the residual cavity by a large drain, this method is used for large cysts (diameter greater than 10 cm) with thickened pericyst.

Endoscopic treatment consists on a sphincterotomy and removal of membranes with the balloon, with Dormia basket or with both.

The two groups are comparable in terms of age, sexe, comorbidity and number of severe angiocholitis (Table 1).

Table 1: Demographic and clinical variables of patients in each group

		Surgery group	Endoscopy group	P
Sexe	**M**	15 (62%)	20 (51%)	0,186
	F	7 (38%)	21 (49%)	
Mean age		41	43	0,277
comorbidity		5 (23%)	9 (22%)	0,590
Severe angiocholitis (grade 3 of the Tokyo Guidelines)		5 (23%)	7 (14%)	0,184

The parameters evaluated were: mortality and morbidity (assessed according to the Clavien-Dindo classification) within 90 days, length of the hospital stay and treatment success defined by the non-necessity of a complementary endoscopic or surgical gesture after the initial treatment.

RESULTS

We report in this study 63 cases of angiocholitis due to hydatid cyst fistula among 264 cases of hydatid cyst of the liver admitted in our institute within a period of 5 years (24%).

The mean population age in the study is 42 years (range 17–88 years). Our patients were divided into 35 men (56%) and 28 women (44%). The majority of our patients came from rural areas (51 patients, 81%), compared to only 12 from urban areas (19%), and 59 of our patients had a low socio-economic level (95%). 9 of our patients already had a history of hepatic or pulmonary hydatid cyst (14%), between those patients, three had already been operated for a hepatic hydatid cyst and one for a pulmonary cyst.

Tokyo criteria were used for assessment of the severity of the angiocholitis. In the surgery group, 5 patients (23%) had a severe angiocholitis vs 7 patients (14%) in the endoscopy group.

Diabetes and high blood pressure were the most common comorbidities (16% and 12%, respectively) (Table 2).

Table 2: Comorbidities of the population in the study

Comorbidities	Number of cases (percentage)
Blood pressure	8(12%)
Diabetes	10(16%)
Dyslipidemia	2(3%)
Asthma	1(2%)
Dysthyroidia	1(2%)

Hepatobiliary ultrasonography was performed in all our patients. Bilio-cystic fistula was detected in 51 cases (80%). CT scan was performed on 34 patients, (53%); it showed a cyst-biliary fistula in all patients (Table 3).

Table 3: Morphological characteristics on ultrasound

Morphological characteristics	Number of cases (percentage)
Hydatid cyst size	
< 10 cm	31(48%)
≥ 10 cm	32(52%)
Number of hydatid cyst	
Unique	52(83%)
multiple	11(17%)
Location	
Right lobe	35(56%)
Left lobe	18(28%)
both	10(16%)
Radiological stage (GHARBI classification)	
I	3(5%)
II	7(11%)
III	34(55%)
IV	17(26%)
V	2(3%)
Suspicion of cysto-biliary fistula	51(80%)

Biologically, all of our patients had a high level of bilirubin with a mean value at 56,7 mg/l, the level of transaminases was high in 44 of our patients (69%).

Preoperatively, all of our patients have benefits from a correction of hydro-electrolytic disorder, intra veinous antibiotics (Metronidazole and ceftriaxone), Symptomatic treatments to relieve pain, itching and vomiting.

All of our patients were treated surgically or by endoscopy within the 48h of there admission.

The blood transfusions were used for sever cholangitis with hemoglobin inferior to 10 g/dl.

In group 1, the surgical approach was made by a right costal Laparotomy in 95% of the cases (21/22) and a median sus-ombilical Laparotomy in a single patient. The treatment of bilio-cystic fistula was done by direct sutures in 18% of the cases (4/22), Bipolar drainage in 41% (9/22) and by a cystobiliary disconnection by percutaneous hepatic choledochostomy in 41% of the cases (9/22). The mean time of surgery was 3 hours.

In group 2, All patients underwent sphincterotomy and removal of membranes with the balloon in 81% of the cases (34/42), with Dormia basket in 10% of the cases (4/42), extraction with Dormia basket and balloon in 7% (3/42), with a single case of ERCP failure due to a difficulty in the catheterization of the main bile duct.

The mean postoperative stay for patients who received endoscopic treatment was lower compared to that of the surgical group (7 days vs 11 days) with a statistically significant difference (P<0.05).

The mortality of patients in the surgery group was 14% (3 patients), a higher rate than that observed in patients treated with ERCP, which is 5% (2 patients) but without a statistically significant difference (p = 0.226). In the surgery group, all the three patients died by a septic shock, while one patient died due to a septic shock and the other one due to hemorrhage in the endoscopic group.

Post-operative complications were more pronounced in the group of patients who had surgical treatment, with a statistically significant difference (P<0.05). Major complications: persistence of the biliary fistula, observed in 6 cases or 27%, treated by an endoscopic sphincterotomy; Minor complications: surgical wound infection, observed in 3 cases or 14 %. In patients treated with ERCP, we had one major complication that was mortal, hemorrhage (2% of cases). (Table 4)

Table 4: Post-therapeutic complications within 90 days

	Surgery N=22	**Endoscopy N=41**	**P**
General morbidity	41% N=9	2% N=1	P<0,05
Major morbidity (Grade ≥3 of Clavien-Dindo classification)	27% N=6	2% N=1	P<0,05

The success rate for the endoscopic treatment is greater than that of the surgical treatment but without a statistically significant difference (p = 0.350) (Table 5).

Table 5: Summary table

	surgery N=22	endoscopy N=41	P
Success	73%	83%	0,350
Mortality within 90 days	14%	5%	0,226
Morbidity within 90 days	41%	2%	<0,05
Post-operative stay	11 Days	7 Days	<0,05

DISCUSSION

Although surgery is the classic method of treating bilio-cystic fistulas, ERCP has emerged as an alternative method that has already shown its effectiveness and safety for this type of complication. ERCP can reduce the postoperative stay and avoid a re-intervention, often difficult and hemorrhagic.

Most of the series published in the literature do not differentiate between complicated and asymptomatic bilio-cystic fistula. In addition, to our knowledge, there have been no studies in the literature comparing both methods (endoscopic vs surgery) which makes the comparison of our results with the literature difficult.

In the literature, the success rate of endoscopic treatment was between 86% and 100% [5] [6] [7] [8], which is consistent with our results (83% success rate). The success rate of surgical treatment was 73%, which joins the results of the literature (success rate between 72% and 97%) [1, 2, 3, 9].

For mortality, in the literature the rate is 0% for endoscopic treatment [10, 4, 5, 6, 11, 8] and varies between 1% and 4.5% for surgical treatment [1, 12, 2, 13]. In our series, mortality was higher in both groups compared with literature. It could be explained by the cohort of patients studied (angiocholitis).

Surgical treatment has a significant rate of complications (41% of cases), they were more marked by cysto-biliary fistula, present in 27% of cases, which is consistent with the study of MEDARHRI [1] (20% of cases). Other complications have been reported in the literature, including subphrenic abscess with a rate that varies between 1% and 8% [1, 13, 3], and the suppuration of the residual cavity reported by BARAKET [12] in 9% of cases. For minor complications, they were more marked by infection of the wound (14%), which is consistent with the ALPER study [9] and the DAALI study [2] (respectively 6.5% and 6%). Complications of endoscopic treatment remain well below those of surgical treatment. In our series we had only one post-endoscopy complication but that was fatal (patient died by hemorrhagic shock). In the literature, post-endoscopy complications were more marked by acute pancreatitis as reported by the MURAT series [8].

The duration of hospitalization varies between 6 and 12 days for endoscopy [14, 10, 4, 5] and 13 and 27 days for surgery [6, 1, 3]. In our series, this rate was 7 days for the endoscopy group, and 11 days for the surgery group; which can, of course, be explained by the low rate of complications after endoscopy.

CONCLUSION

In the light of our study comparing the two therapeutic techniques (surgery against endoscopy), we can conclude on the effectiveness, the low morbi-mortality, with a post-intervention stay reduced rate of the endoscopic procedure in the treatment of Angiocholitis due to Hydatid Cyst of the liver fistulized in the bile ducts. The endoscopic treatment should be considered as the treatment of choice for angiocholitis due to hydatid cyst of the liver fistulized in the bile ducts.

List of abbreviations

ERCP: endoscopic retrograde cholangiopancreatography

Limitation of the study:

The nature of the study: retrospective study;
The small number of patients included in the study;
The absence of a long-term surveillance.

DECLARATIONS

Ethics approval and consent to participate

The local institutional review board of surgery at the university hospital Hassan II of Fez has approved this retrospective study (Angiocholitis due to Hydatid Cyst of the liver fistulized in the bile ducts: Comparative study between surgical treatment and endoscopic treatment) and also gave its approval for using medical records without patient informed consent (waiver) with the emphasis on patient's confidentiality.

Availability of data and materials: All data generated or analyzed during this study are included in this published article.

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Authors' contributions

BEB, BDA, ISA, OA, MK and ATK have conceived and coordinated the study
BEB has corrected the manuscript.
MO, HA, AT and AM have done the data collection.
MO has done the redaction of the manuscript.
All authors read and approved the final manuscript.

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Statement

All methods used in this study were carried out in accordance with relevant guidelines and regulations and approved by the head of the division of abdominal surgery and gastro enterology unit in the university hospital Hassan II of Fes, Morocco.

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