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### RESEARCH ARTICLE

#### CENTRAL HYDATID CYST OF THE LIVER FISTULIZED IN THE BILE DUCTS, DIFFICULT SURGICAL ACCESS AND THE CONTRIBUTION OF ENDOSCOPY A STUDY OF THREE CASES

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#### Abstract

Hepatic hydatid cyst is one of the common public health problems in Maghreb, remains a benign pathology, but its seriousness concerns its complications, which are largely dominated by cystic rupture in the bile ducts. The aim of this study is to highlight the difficulty of surgical treatment of hydatid cyst, localized in central part of liver and ruptured in biliary ducts with the place of endoscopy we report the cases of three patients with central hepatic hydatid cyst opening in biliary ducts which are treated differently by surgery and endoscopy.

**Results:** The surgery with the radical and conservative treatment in compared to endoscopy procedure was technically more difficult and efficient, but at the expense of morbidities and a longer hospitalization period.

**Conclusion:** Cysto-biliary fistula is the most frequent and the most frightening complication of liver hydatid cyst, the surgery is the gold standard treatment with the conservative and radical methods. However, some locations such as central part of liver make surgery very difficult and dangerous which open the topic of the usefulness of the endoscopy in this problem.

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#### Introduction:-

The Hydatid cyst (HC) is a non-immunizing cosmopolitan anthroponosis, which occurs endemically in large sheep-breeding countries in the presence of a dog, it is said that "hydatidosis follows the dog like its shadow" (1,2).

Humans are affected accidentally by the ingestion of Echinococcus granulosus eggs transmitted by the dog, the usual definitive host. In the Maghreb, according to figures from the Ministry of Health, Morocco ranks 3rd after Tunisia and Algeria with a surgical incidence of 5.2 per 100,000 in 2008 (1). It is a real public health problem, with a very high economic impact, especially in developing countries. The pathology considered as benign, frequently affects the liver, and its seriousness is linked to its complications.

The fistulation in biliary trunks remains the most frequent and most dreadful complication, which can lead to angiocholitis with severe septicemia leading to death. According to most authors, it varies between 10 and 30 % of cases (3,4,5). In several series, the most frequent predictor in Cysto-biliary fistula (CBF) is the large size of HC (6,7). The cysto-biliary communication may be latent and discovered incidentally during surgery or in forms of hydatid angiocholitis (8). The diagnosis is based on a CT scan and abdominal MRI. In addition, an endoscopy is very

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useful for both diagnosis and treatment (9). The management of a liver hydatid cyst (LHC) with CBF, of which surgery occupies an unavoidable place, has known several advances with the progress of endoscopic methods such as the endoscopic retrograde cholangio-pancreatography (ERCP) with sphincterotomy (ES) (10). Through this study we present the surgical difficulty caused by the central localization of a hydatid cyst ruptured in the bile ducts with the contribution of endoscopy.

### **Clinical Presentation:**

#### **Case N°1:**

A 22-year-old patient, with some notion of contact with dogs, who presents a clinical and biological cholestasis with a positive hydatid serology.

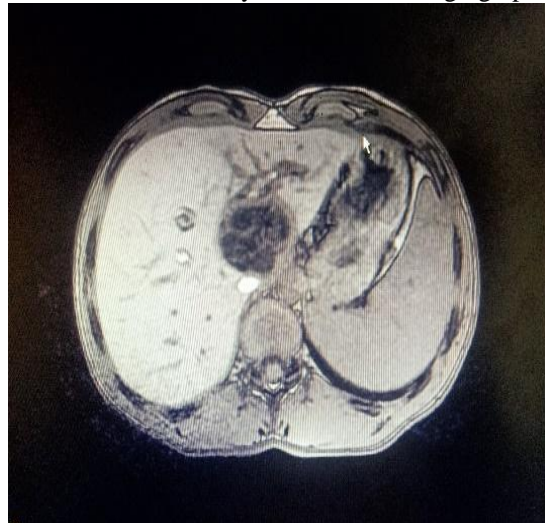
The abdominal CT scan shows: The liver cystic mass of segment I of type III compressing the main hepatic duct and the portal vein with a dilatation of the upstream intrahepatic BD.

The BILI-MRI demonstrates a centro-hiliary multilocular liver hydatid cyst (LHC) at the level of segment I, II, and III compressing the left bile ducts (BD) with probably microcracks. Figure (1)

The patient has been treated by radical surgery with total pericystectomy and cysto-biliary disconnection (large fistula) according to the Perdromo technique. At the end of the procedure, a cholangiography by Kher drain showed a vacuity of the BD.

The postoperative follow-up was simple, with the regression of jaundice and cholestasis after 4 days.

The hospitalization lasted 11 days and the patient was discharged under medical treatment for 6 months. The evolution was good. The Kher drain was removed 45 days after two cholangiographies of control.



**Figure 1:-** Hepatic MRI showing a centro-hiliary LHC type III located at the level of segment I, II and III in intimate contact with the dilated left BD with probably microcracks.

#### **Case N°2:**

A 57-year-old patient with a history of LHC under medical treatment for 6 months. Admitted for hepatic colic with febrile attacks and chills.

Biologically, he presents an inflammatory syndrome and cytolysis with a positive hydatid serology.

The abdominal CT scan shows a HC type IV straddling segment IV and V with bile ducts of normal caliber. Figure (2).

The patient was surgically treated by the resection of the prominent dome (RPD) and partial pericystectomy surrounding the fistula with trans-cystic external bile drainage completed by cholecystectomy.

The postoperative follow-up was simple, the hospitalization lasted 9 days.

The kher drain was removed three weeks after two cholangiographics of control.



**Figure 2:-** Abdominal CT scan showing a cystic formation with heterogeneous content evoking a LHC OF TYPE IV straddling segment V and VI without dilation of BD.

### Case N° 3:

A 35-year-old patient aged, operated for a liver hydatid cyst 2 years ago, admitted to the emergency room for angiocholitis.

Biologically, he presented an infectious syndrome, cytolysis and cholestasis.

The CT scan showed a stage III LHC of segment I with a dilation of the main bile duct (MBD).

The BILI-IRM showed a HC of segment I, the dilatation and the presence of hydatid material in the main bile duct (VBP).

The patient was hospitalized in the Intensive care unit, and put on intravenous antibiotics.

The treatment was purely endoscopic by ERCP with sphincterotomy: After opacification, the hydatid material was removed with the help of an extraction balloon with a saline lavage and drainage of the bile ducts, completed by the placement of the naso-biliary drain. Figures (A, B, C, D, E).

The follow-up after surgery was simple with the disappearance of the jaundice in 2 days and normalization of the hepatic balance. The patient left the hospital under medical treatment.

After 4 months of recurrent free disease, the patient presented an abdominal pain, and the abdominal ultrasound and CT scan showed a recurrent hydatid cyst in IV segment of liver without biliary fistula. The patient lost to follow up. Figure 3





- A) scopic view of the central biliary tract pigging,  
 B) Endoscopic view of the extraction balloon.  
 C) Endoscopic sphincterotomy.  
 D, E) Exit of hydatid membranes.  
 F) Vacuity of the main bile duct at the end of the procedure.



**Figure 3:-** Abdominal ultrasound showing a cystic formation evoking a cyst hydatid IV segment of liver.

### Discussion:-

The Hydatid echinococcosis is cosmopolitan and occurs endemically in countries where dogs herd livestock, such as North Africa. The liver is its most frequent location (70%). It is a real public health problem (1,2).

The Hepatic hydatid cysts deemed benign disease, but is worrying because of its complications which can sometimes be life-threatening. The most frequent is the opening in biliary ducts, which is the main morbidity factor of this disease (11). The incidence is variable from one series to another depending on the frequency in the population, the mode of recruitment, and the means of intraoperative screening. They frequently complicate the operated HC estimated at 17-44% (12). In our study, only one case was operated.

This complicated form can be silent, discovered fortuitously intraoperatively or by clinical presentation of acuteangiocholitis and/or deep suppuration (8). An Ultrasound and CT scan allow the characterization of the hepatic KH as well as the visualization of the hydatid material in the VBP. The bilio-MRI remains the examination of choice to explore the bile ducts. The ERCP also allows diagnosis and offers an alternative treatment in emergency (9,13).

Surgery is a gold standard of the treatment; however, the technique choice remains a controversial issue. On one hand, the radical methods, pericystectomy or regulated hepatectomy, recognized by their effectiveness, remain however, disproportionate in regards to abating and endemic disease.

On the other hand, the resection of the protruding dome (RPD) is a simple and rapid method, but still source of significant morbidity. (14, 12, 15)

The treatment of the biliary fistula is potentially morbid, if the fistula is minimal, a simple suture with or without drainage is sufficient, but the risk of prolonged bile leakage is major. Cysto-biliary disconnection is considered the method of choice according to the peridromotom technique (14,16). In our case, it was performed on only one patient.

Surgical disobstruction of the bile ducts is done by transcystic or trans-choleptic approach with the extraction of the hydatid material and external drainage by Kher drain.

Biliary fistulas cause even more therapeutic problems as the site of the HC is in contact with large vessels, especially in the case of centrohepatic HC with a high risk of hemorrhagic and biliary accidents. (15,16)

Surgical drainage may be the only credible treatment for centrohepatic HC that are difficult to access, revealed by severe angiocholitis and largely ruptured in the bile ducts.

Furthermore, studies consider endoscopic drainage as an efficient method in the management of ruptured LHC in the Bile ducts, avoiding surgical re-interventions and allowing a shorter hospital stay. It is indicated for a small HC with a large rupture in the bile ducts and as a standby treatment in severe angiocholitis (13, 17).

Endoscopic sphincterotomy (ES) is intended to ensure better internal biliary drainage with preferential flow of bile to the duodenum, thus promoting the healing of the biliary fistula (10,13,17).

Can endoscopic drainage be considered as an alternative or standby treatment before surgery in the management of ruptured centrohepatic HC in the bile ducts, especially in an emergency situation (angiocholitis) and/or for patients already operated?

### Conclusions:-

The cystobiliary fistulas are a potentially serious complication and the surgical management is the general rule, yet the central liver location with difficult surgical access, can cause technical problems. Endoscopic drainage can be an attractive alternative in the therapeutic arsenal of this condition but its efficiency is still controversial.

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