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RESEARCH ARTICLE

HYDATID CYST OF THE LIVER RUPTURED INTO THE BILIARY TRACT: SERIES OF 52 CASES

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Abstract

The rupture of the hydatid cyst in the bile ducts is a frequent and serious complication whose evolution of the disease which from parasitic becomes hepatobiliary and involves the vital prognosis. The aim of our work is to review the clinical and therapeutic aspects of this complication. For this, we conducted a retrospective study of operated cases of ruptured hydatid cyst in the bile ducts. Between January 2015 and December 2022, 52 cases of hydatid cyst of the liver ruptured in the bile ducts were collected in our department, these are 30 women and 22 men, with an average age of 38.6 years with extremes between "18 and 70 years old". The clinical picture was marked in the majority of cases by an association of hepatic colic (84%), jaundice (28%), febrile syndrome (13%) and episodes of cholangitis in 8% of cases. Clinical examination found hepatomegaly in 25% of cases. The fistula between the cyst and the bile ducts was incidentally discovered in 2% of cases. The contribution of radiological examinations, in particular ultrasound, is invaluable, supplemented in case of doubt by computed tomography. The treatment of the cyst consisted of conservative treatment in most patients 71.15%, pericystectomy in 23.07%. This conservative treatment was completed by suture of the cystobiliary fistula 67.30%, catheterization 3.84%, Perdomo technique 13.46%, drainage of the main bile duct in 57.69% of cases, a bipolar drainage in 2 cases and finally a case of bilio-digestive anastomosis. The surgical procedure was radical in 3 cases. There was no known recurrence or late complications (jaundice) in the patients followed regularly in 82.69% of cases, while 11.53% patients presented postoperative complications.

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Introduction:-

The hydatid cyst is an anthropozoonosis due to the development in humans of the larva of Echinococcus granulosus [1,2,3].

It is endemic in Morocco and poses both a public health problem and an economic problem. the trunk carries the parasite [4,5].

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The hydatid cyst of the liver often evolves for several years before the onset of clinical manifestations [6].

Pathology benign in itself but worrying by its complications, the most frequent and the most serious of which is the opening in the bile ducts [1] which can sometimes be life-threatening.

The rupture of the cyst in the bile ducts is often asymptomatic except when it sits at the level of the main bile duct, thus responsible for cholangitis.

This complication has aroused special interest, especially on the therapeutic level, so surgical treatment must be as conservative as possible in order to reduce the mortality and morbidity rate of this benign condition..

Methods:-

Our retrospective study based on a series of 52 cases of hydatid cyst of the liver ruptured in the bile ducts collected in the Surgery B department of the Avicenne Hospital in Rabat over a period from January 2015 to December 2022 , aims to identify the clinical and paraclinical aspects of this complication and to better understand the surgical treatment of this condition**.**

Results:-

Between January 2015 and December 2022, among 207 patients operated on for hydatid cyst of the liver in our service, 52 cases were complicated by biliary fistulas (25.12%).

The average age of our patients was 38 years old with extremes ranging from 18 to 70 years old.two most affected age groups are between “30 and 39 years old” and between “20 and 29 years old”

The female sex marked a great predominance with a frequency of 57.70% against 42.30% of the male sex.

Our patients were in 56% of cases of rural origin, while 44% of patients were of urban origin.

The notion of contact with dogs was found in 23 patients in our series, (44.23%.)
Six of our patients already had a history of pulmonary or hepatic hydatid cyst. the appearance of the symptoms that led our patients to consult dates back to a variable duration, ranging from 10 days to 6 years with an average of 9 months. Whatever the type of symptom, the installation was most often insidious and progressive.

Pain in the right hypochondrium was found in 44 of our patients (84.61%), in the form of hepatic colic or heaviness.

Retentional jaundice is found in 15 patients (28.84%)

It's cholestatic jaundice. Some patients have reported spontaneously regressive jaundice not found on clinical examination.

Hepatomegaly was found in 13 patients (25%).

Febrile syndrome was encountered in seven patients (13.46%). It reflects suppuration of the cyst or cholangitis.
Vomiting was present in 20 patients(38.46%) of cases.

Four patients presented with acute cholangitis (8%).

Weight loss was noted in 8% of our patients.

In only one case, the hydatid cyst was asymptomatic, discovered fortuitously during a radiological assessment.

Hepatobiliary ultrasound was performed in 45 patients. It objectified the presence of a single hydatid cyst in 32 patients and multiple hydatid cysts in the rest.
- The location of the cysts was in 32 of our patients in the right liver, i.e. 61.53% of cases, in 11 patients in the left liver, i.e. 21.15% of cases and in 17.30% of cases, it was horse between the left liver and the right liver.

- By analyzing the frequency of ultrasound types of liver hydatid cysts according to the Gharbi classification; there is a predominance of type III found in 15 patients; i.e. 48.64% of cases then type IV found in 21.62% of our patients. In the other cases, the ultrasound type was not specified.

It is noted that in four cases , there was the association of two ultrasound types.
Dilation of the bile ducts was noted in eight cases(15.38%)

Abdominal computed tomography (CT): was requested for 18 patients (34.61%).
The CT had made it possible to make the diagnosis of rupture of the hydatid cyst in the bile ducts in eight cases of our series when the ultrasound had not shown them.
- It had also made it possible to confirm the diagnosis in five cases already suspected on ultrasound.
- In the five remaining cases, CT only showed hydatid cysts without visualization of bile duct abnormality.

MRI was performed for four patients who presented with cholangitis (7.69%).

Chest x-ray: was performed in 36 cases, it showed in two cases a cystic image (3.84%).

Hyperleukocytosis was found in 13 patients
This sign is synonymous with infection of the cyst or rupture in the bile ducts.
Hypereosinophilia was observed in three patients. It is a synonym of parasitosis but also a witness to the cracking of the hydatid cyst.

Hepatic cholestasis was found in 13 patients;however, the cholestasis assessment was not carried out in five cases, 13 patients also had signs of cytolysis

The association of the two biological abnormalities was found in eight patients in our series.

Hydatid serology was a very effective diagnostic tool. It was performed in 48 patients and was positive in 41 cases (78.84%).

However, the methods used were not specified in all cases, but it was the ELISA technique and immunofluorescence that were most often used.

All our patients have benefited from an exclusively surgical treatment.

The approach used in our series was Kocher subcostal incision
for all patients.

Intraoperative exploration essential operating time aiming to examine the entire abdominal cavity in a meticulous way in order to search for peritoneal grafts and to evaluate the number, topography and content of the cysts after mobilization of the liver. in summary Intraoperative exploration allowed us to take stock of localizations or associated pathologies

4-Number, location and size of the hydatid cyst
The hydatid cyst involved the right lobe of the liver in 42 patients or 80% of cases, the left lobe was affected in six patients or 11.53% and in four patients, the cyst was on both lobes.

After laparotomy, four patients had double or multiple liver hydatid cysts and in two cases an associated extrahepatic localization was noted.

In our series, most of the cysts are large, with an average size of 9 cm.

The contents of the cyst were bilious or even purulent bilious in five patients.

Site of the fistula
It generally coincided with the location of the liver hydatid cyst.

In eight cases, the fistula was located at the bottom of the residual cavity.
It should be noted that in 12 patients, multiple biliary fistulas were found on exploration.

Our attitude towards the cyst and the residual cavity,
Before any maneuver on the cyst, the operative area was excluded from the rest of the abdomen by a drape soaked in parasiticidal product to avoid any parasitic dissemination. The parasiticidal product used in our series was hydrogen peroxide diluted in isotonic saline.

In our series, the treatment of the residual cavity after sterilization and evacuation of the cyst consisted of:
- conservative treatment for 37 patients or 71.15%.
- Partial pericystectomy in 12 patients or 23.07%.
- Total pericystectomy wasperformed in a single patient
- A controlled hepatectomy was performed in two of our patients.
- Drainage of the residual cavity was systematic in all our patients.

Our attitude towards biliary cystic fistula:
The treatment of the biliary-cystic fistula consisted in our series of:
1. A simple suture of the fistula in 35 cases or 67.30%.
2. Catheterization using a ureteral catheter in two patients.
3. Cysto-biliary disconnection or transhepatic-cystic choledocotomy according to Perdomo was performed in seven cases, i.e. 13.46%.
4. Drainage of the common bile duct in 30 cases.
5. Bipolar drainage in two cases.
6. End-to-side choledocojeujenal anastomosis in a single patient

Gesture on the gallbladder and cholecystectomy in 12 patients, or 23.07%.

The duration of hospitalization of our patients varied from 13 to 70 days with an average of 20 days.
Postoperative stay: extended from 3 to 22 days in our patients with an average of 12 days, this more or less long stay was due to postoperative morbidity as well as to the time required for drainage of the residual cavity and bile ducts.

The postoperative stay in patients who received conservative treatment reached 22 days, but did not exceed seven days in the radical techniques.

Mortality: No case of death was reported in our series.

Morbidity: There was no known recurrence or late complications (jaundice) in the patients followed regularly. the follow-up was simple in 43 patients, or 82.69%.

This criterion was not specified in three patients.
Postoperative complications in six patients (11.53%) were as follows:
- An external biliary fistula in three patients, associated with fever in a single case;
- One patient had persistent jaundice and cholestasis on the liver test;
- An isolated fever in two cases.

It is the conservative techniques that were the source of complications, on the other hand, no morbidity factor was noted in the cases treated with the radical technique.

Long-term monitoring and evolution: It is essential in all cases; its main objective is to detect in time a possible recurrence or reinfestation. This follow-up requires an analysis of clinical, radiological and serological data. In our series, the patients were reviewed in consultation, but the follow-up was not transcribed in the medical records**.**

Discussion:-

The main cause of morbidity, the rupture of the hydatid cyst of the liver into the bile ducts is by far the main complication of this benign condition [7, 8, 9].

In our series, there are 52 cases of biliary-cystic fistulas among 207 cases of hydatid cyst of the liver, recruited in the service, during a period of seven years, which represents a rate of 25.12%.

The frequency of this complication varies according to the authors, in fact it represents 20% in the MOUMEN study [10], 31% for BALAFREJ [4], 25% in the DAALI series [5], 18.1% according to BERDILI [11], 25% according to ABI [12] and according to a study carried out by the surgery department of the Avicenne military hospital in Marrakech. Between 1990 and 2008, out of 536 hydatid cysts of the liver operated on in the department, 120 were complicated by rupture in the bile ducts, i.e. a rate of 22.38%. [14]

Pain in the right hypochondrium or epigastrium: is the most frequently found sign. Its intensity, its rhythm and its irradiation are variable. This sign was found in 25% of our patients.

Jaundice: Cholestatic type, testifies to the obstruction of the bile ducts by hydatid material. It varies according to the degree of obstruction, ranging from conjunctival subicterus to frank mucocutaneous jaundice. It can be intermittent, with relapses or continuous without remission [5, 6, 15]. It was detected in 15 patients or 28.84% of cases.
Fever: most often testifies to the suppuration of the cyst but its absence does not eliminate the possibility of a cystic infection. Fever can also be explained by inflammation of the bile ducts [16].

Hepatomegaly: is secondary to deep hydatid cysts, compensatory hypertrophy of the liver parenchyma and venous stasis by compression. It may be the only revelation of a latent KHF [6, 1, 17]. Hepatomegaly was found in 13 patients in our series.

Acute cholangitis: is a serious complication of cystobiliary fistula secondary to the passage of infected hydatid material into the bile ducts. His picture is made up of a triad: pain in the right hypochondrium, cholestatic jaundice and febrile seizures [18] and
Its consequences are often serious

Unlike intraperitoneal rupture, cystobiliary rupture rarely gives rise to allergic manifestations [19]. Allergic reactions such as pruritus and skin rash were encountered in two cases in our series.

The general condition is preserved in the majority of cases until an advanced stage of the disease [20].

Formerly limited to standard X-ray examinations, imaging of hydatid disease has been enriched with effective and non-invasive diagnostic means [21].

Abdominal ultrasound is the key diagnostic test. Due to its complete safety, low cost and reliability, ultrasound has established itself as the key examination for hepatic hydatidosis [21].

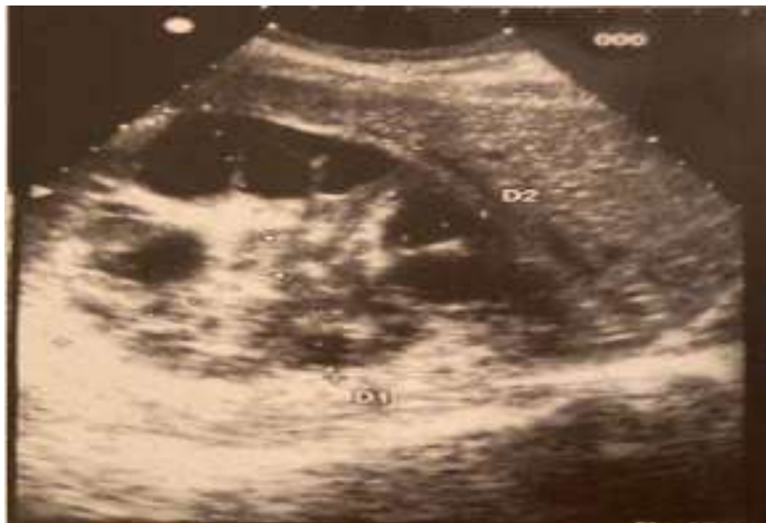

Figure 1:- Ultrasound appearance of a type III hydatid liver cyst, multi-vesicular appearance [22].

CT is not a routine examination in the diagnostic assessment of the hydatid cyst, it is an examination that is not suitable for mass screening in endemic areas [20]. It has a sensitivity of 77% in the detection of cystobiliary fistulas [23]. The scanner has the additional advantage of having better reliability in terms of:
- Accuracy of the number, size and topography of the cysts.
- Identification of small cysts and extra hepatic cysts.
- Demonstration of calcifications, whether in mass or parietal
- Diagnosis of infectious or biliary complications (communicating cysts), and ruptures in the peritoneal cavity [21].

Combined with or in addition to ultrasound, CT can facilitate the
diagnosis of hydatid cyst of the liver ruptured in the bile ducts by visualizing a dilation of the bile ducts adjacent to the hydatid cyst but especially by objectifying the cystobiliary communication on the reconstruction images or after injection of the contrast product [5,12], in addition, hydatid debris may be visible in the main bile duct It also provides reference images of the initial assessment, allowing an objective, non-operator dependent subsequent comparison

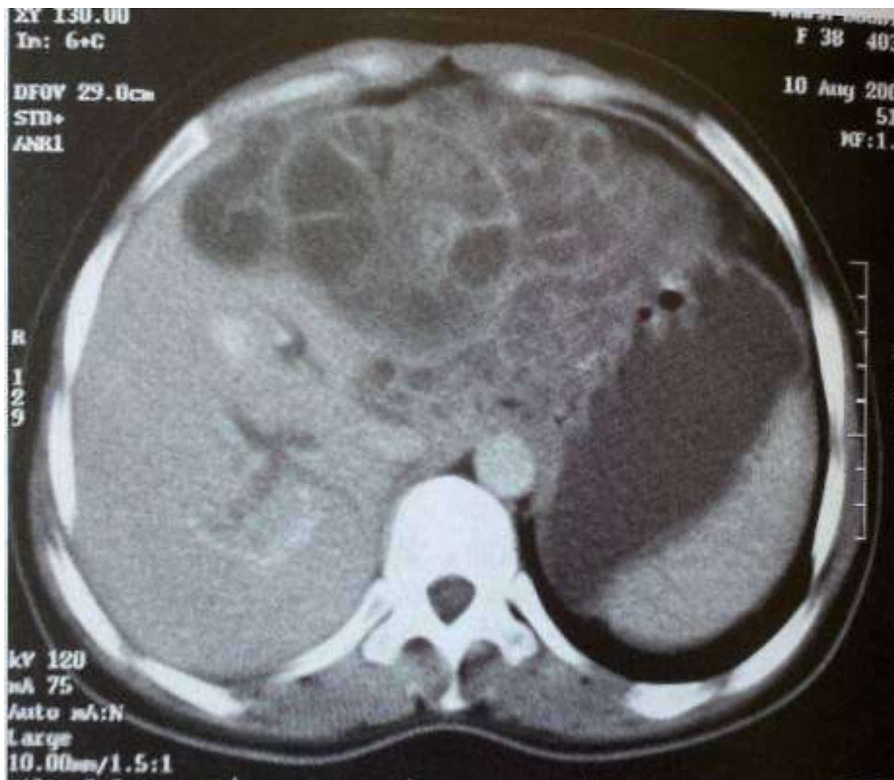

Figure 2:- CT appearance in axial section: hydatid cyst of the left liver complicated by a cystobiliary fistula and pericystic location. [22].

magnetic resonance cholangiopancreatography (MRCP)is totally harmless, and it has an excellent spatial resolution and its great diagnostic performance of the lesional content, and from the study of the vascular and biliary relationships, the bili MRI manages to identify the communication between the cyst and the bile ducts by showing deformation of the cyst, loss of continuity of its wall, dilation of the bile ducts and the presence of hydatid material (daughter vesicles and floating membranes) in the biliary tree [24, 25, 26] which is a specific sign of breaking.
Its sensitivity is around 91.7% and its specificity is 82.8% in the identification of biliary cyst rupture [21].

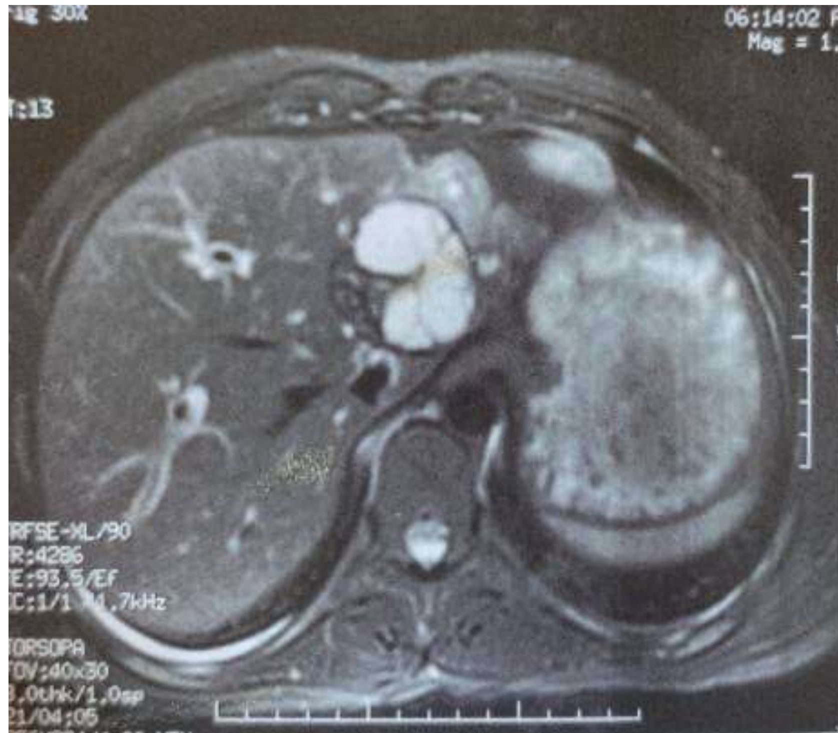

Figure 3:- Hydatid cyst of the left liver fissured in the left intrahepatic bile ducts [22].

Endoscopic retrograde cholangiopancreatographyremains the method of choice in the diagnosis as well as the treatment of biliary complications of the hydatid cyst of the liver. It can detect the rupture of hydatid cyst in the bile ducts in 86.6 to 100% of cases [27,28].

It is an examination systematically requested to search for the pulmonary localization of a hydatid cyst, or a reaction pleurisy to a hydatid cyst of the liver [29,30,31].
g-Abdomen without preparation:

This examination makes it possible to evoke the diagnosis of hepatic hydatidosis without prejudice to the biliary lesions by showing [32, 33]:
- An elevation of the right diaphragmatic dome;
- Calcifications suggestive of cholelithiasis;
- Calcifications

Hyperleukocytosis with polymorphonuclear neutrophils can be encountered in the event of superinfection of the contents of the cyst, which is more frequent in its pulmonary location. [34]

Liver function tests are usually normal due to the absence of hepatic cell damage. Changes such as cholestasis or cytolysis should raise fears of a complication (rupture in the bile ducts or compression) [34].

Surgical treatment is still the treatment of choice and the best alternative in complicated hydatid cysts.
As soon as the diagnosis of simple or complicated hydatid cyst is established, treatment must be undertaken as soon as possible to avoid complications of hydatid cyst of liver ruptures in the bile ducts [35, 36], and regardless of the surgical technique adopted, exploration of conservative treatment and release of hydatid material from the bile ducts should be the surgeon's priority [37].

Surgical treatment must achieve three objectives
- The eradication of the parasite, the first stage common to all radical or conservative techniques;
-Removal of the residual cavity, total and complete with radical methods, incomplete and partial with conservative methods;

- The search for cystobiliary fistulas and their treatment and the control of the vacuity of the main bile duct. [37, 38]

The upper midlineincision :Allows good exploration of the abdominal cavity, but gives a poor view of the right liver.This approach does not allow easy exploration and optimal treatment of posterior seat cysts (posterior sector, dorsal lobe) and hepatic dome (Segment VIII).

Kocher subcostal incision: The approach of choice for liver surgery, the incision can be more or less widened on the left with possibly a vertical partition towards the xiphoid.
It was used in all patients in our series.

The abdominal cavity is meticulously inspected and manually explored in search of other hydatid localizations or a possible peritoneal graft. The liver must be fully mobilized by section of the ligaments of the liver and visceral, peritoneal and diaphragmatic adhesions due to the development of the cyst. The location and number of cysts must be specified. This initial assessment is completed by an intraoperative ultrasound.

Intraoperative ultrasound: Examination that has become essential in liver surgery. It makes it possible to specify the relationship of the cyst with the vasculobiliary pedicles, to orient the therapeutic strategy and to choose the most appropriate technique for each case.

Intraoperative cholangiography: allows to explore the biliary tree and to specify the existence of cystobiliary fistulas, their location and their importance, as well as the existence of associated lesions on the intra and or extra-hepatic biliary tree, in particular daughter vesicles causing bile duct obstruction or bile duct lithiasis.

Eradication of the parasite and intraoperative protection against the risk of parasitic dissemination. Use of scolicide; We recommend mechanical protection by dry drapes or soaked in scolicide solution (2% hydrogen peroxide or 20% hypertonic saline solution) completely isolating the cyst from the peritoneal cavity and leaving only the cystic dome visible. [38]

After evacuation of the parasite and cleaning of the endocyst, the attitude vis-à-vis the residual cavity depends on several factors: The patient's condition, the location of the cyst, the evolutionary stage of the pericyst, the existence of cystobiliary fistula and the experience of the surgeon.

Conservative techniques (Lagrot technique) leave more or less of the pericyst in place and do not allow the residual cavity to collapse, particularly when the pericyst is thick [39].

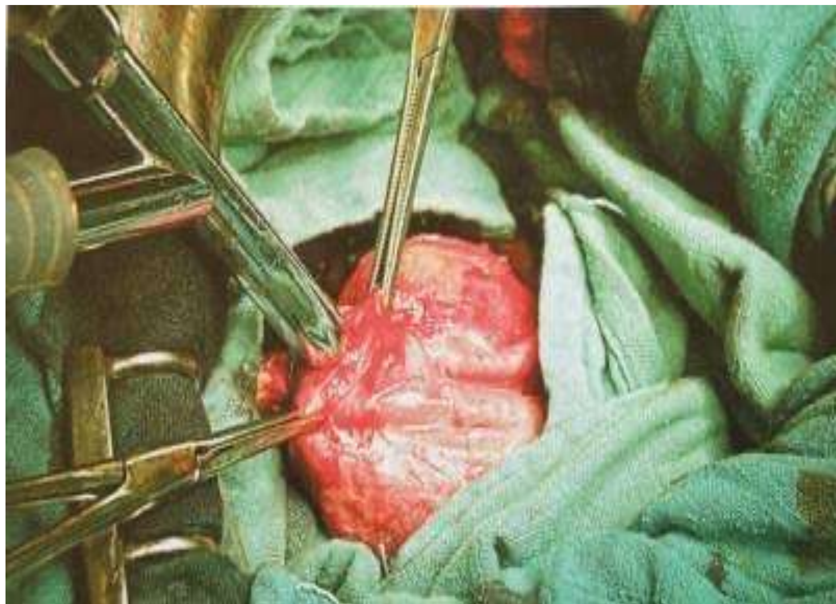

Figure 4:- Emptying the cyst [39].

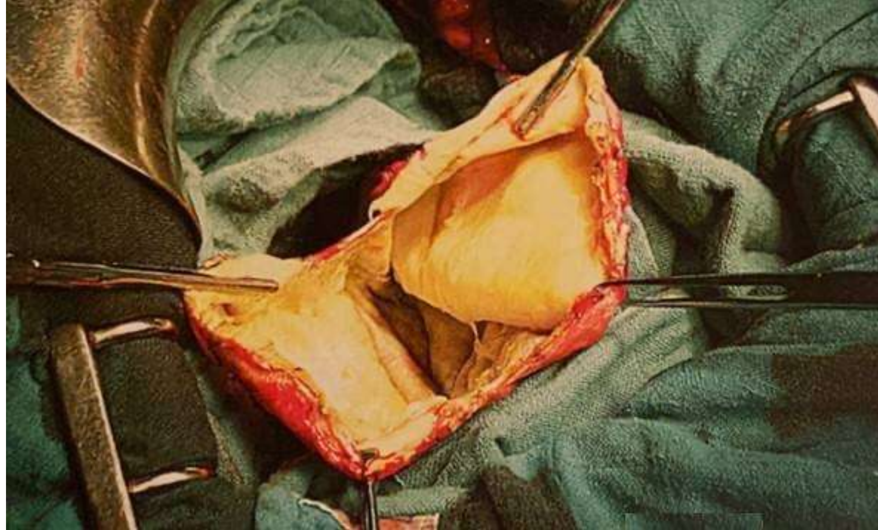

Figure 5:- Thorough cleaning and exploration of the endocyst [39].

Radical techniques resect the entire pericyst and leave no residual cavity (total pericystectomy, pericystoresection, hepatic resection) [39].

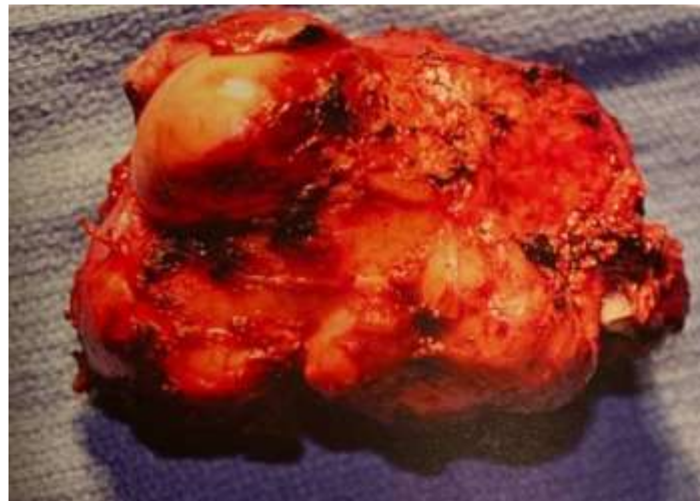

Figure 6:- Closed total cyst pericystectomy is the best guarantee against recurrence [22].

Treatment of cystobiliary fistulas: [40]
a- Conservative treatment:
The treatment is still too often conservative, especially in hydatid endemic countries. Several techniques have been recommended with varying results:

a-1) The simple suture
The simple suturing of the fistula with drainage of the common bile duct by Kehr drain is often complicated by the loosening of the suture when it is done on a thick and sclerotic pericyst. This technique should be abandoned. [40] Achieved in 67.30% of cases

a-2) Bipolar drainage
It is a technique that consists of locating and suturing the fistulous orifice after conservative treatment, with drainage of the main bile duct by a Kehr drain and the residual cavity by a large-caliber drain such as a gastric tube or multiperforated drain.
It has several drawbacks: [41]

Reversal of bile flow in the bile duct-residual cavity direction, maintained by sub-phrenic depression during each inspiration occurring even outside of any obstruction of the main bile duct;

The suturing of the biliary fistula is uncertain in case of thick or infected pericyst; The passage of hydatid material in the bile ducts is responsible for their irritation and infection.
Bipolar drainage was performed in 4% of cases. [40]

a-3) Transfistulo-oddiandrainage :

After evacuation and cleaning of the cyst, and possible resection of part of the pericyst, the cystobiliary fistula is located and widened, the residual cavity cleansed, is then closed tightly with a slow-resorbing fine thread suture. The pericyst can be reduced beforehand and softened by careful resection of the lamellae by endocystic approach.

a-4) Perdomo's transhepatic-cystic choledocostomy [42, 43,44] :

After treatment of the residual cavity by resection of the protruding dome, a choledotomy makes it possible to evacuate and clean the main bile duct.

A metal explorer is ascended through the bile duct to search for the cystobiliary fistula. The Kehr drain is chosen according to the diameter of the kvstobiliary communication.

The short branch of a Kehr drain is then attached to the explorer and draws him towards the choledotomy.

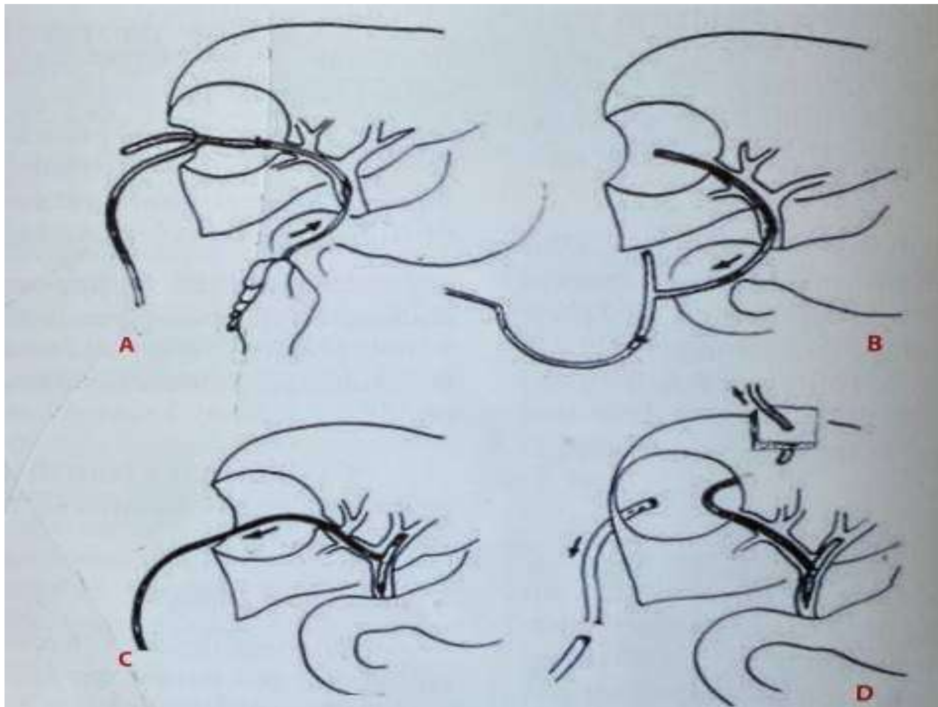

Figure 7:- Transhepaticocysticcholedocostomy technique according to Perdomo [40].

a-5) Cystobiliary Disconnection [45,46]:
This technique, a simplified variant of the Perdomo technique, consists of an anatomical disconnection between the residual cavity and the biliary fistula.

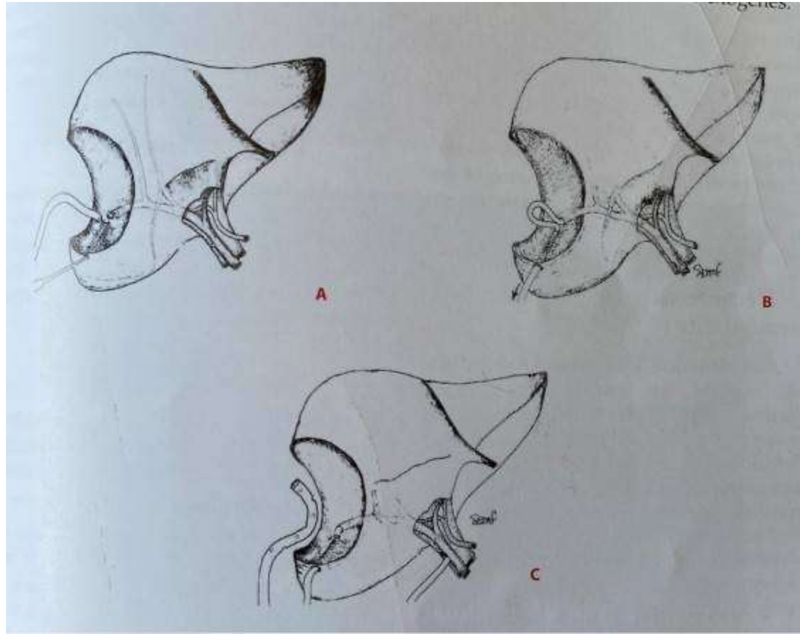

Figure 8:- Cystobiliary disconnection technique. [40].

A: Hydatid cyst of segment VI with large fistula at the expense of the dilated segmental canal of VI. After partial pericystectomy, a long pass creates a subpericystic path until it comes into contact with the fistula. The polyethylene drain is attached to the grommet
B: The grommet is removed causing the drain. This is introduced into the fistula for about 5 cm.
C: The tunneled transfistulous drain provides disconnection
cystobiliary. The common bile duct is drained by a Kehr drain

Medical treatment:
The treatment of the hydatid cyst was for a long time strictly surgical. At the beginning of the 90s, other therapeutic possibilities appeared and modified the management of hepatic hydatidosis. Mebendazole, was used in the first place, but albendazole has become the drug of choice due to its better absorption, better bioavailability, and better clinical efficacy [40]

The usual dose of albendazole is 800mg/day in one or two doses for a month's treatment, for three months, separated (or not) by intervals of a week or 14 days, over a period of six months or more.;

The usual dose of mebendazole is 40/50mg/day for at least three months.

Medical treatment is focused on the elimination of protoscole and the destruction of the germinal layer. Medical treatment can be envisaged with variable objectives: such as The definitive cure of univesicular cysts (type I and type II), after a treatment of three to six months is variable according to the authors.
However, the success rate would be 82% but with a relapse rate of 25%. The majority of relapses occur within two years of stopping treatment. Long-term follow-up is therefore necessary.

Conclusion:-
The rupture of hydatid cysts in the bile ducts is a frequent and serious complication. The treatment is based on radical methods of recognized effectiveness but which seem disproportionate for this generally benign condition and conservative methods which are most often recommended in developing countries. path of development, but the treatment of intracystic biliary fistulas is in return a source of often significant morbidity at the origin of an often long hospital stay.

Indeed, the postoperative morbidity is not negligible represented especially by the external biliary fistulas, the abscesses under phrenic and of the residual cavity. These complications are responsible for a somewhat prolonged postoperative stay.

Finally, prophylaxis, as well as screening and treatment of hepatic hydatidosis at an early stage, would certainly contribute to reducing the incidence of this complication.

Data Availability Statement
The data that support the findings of this article are available from the corresponding author upon reasonable request.

Competing interests:
The authors have no conflicts of interest and source of funding. The subject of the article had no commercial interest, no financial or material support.

Ethics statement
Drs AbdellahMoufid, , SettafAbdellatif ,Benamar Said , khalidlahlou ,Mdaghri Jalil , Mssrouri Rahal declare no conflict of interest.

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