

SURGICAL AND ENDOSCOPIC MANAGEMENT OF BILIO-BRONCHIAL FISTULA SECONDARY TO RUPTURED HYDATID CYSTS OF THE LIVER: -EXPERIENCE OF A MOROCCAN CENTER-

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ABSTRACT

Introduction: The bilio-bronchial fistula (BBF) is an anomalous communication between the bile ducts and the tracheobronchial tree, which complicates several hepatobiliary pathologies but the most common, in our location, remain the hydatid origin. Through 5 clinical cases, we report our experience of multimodal management of this pathology. **Patients and Methods:** The positive diagnosis was based on clinical, radiological and endoscopic arguments. All patients were treated according to two components: the first is an assessment and a correction of any associated biological disorders with a drainage of any pleural collection. The second therapeutic component consists in performing an endoscopic sphincterotomy to remove any individualized bile duct's obstacle. Surgical treatment was indicated only in case of the failure of the medical and endoscopic management. **Results:** Five patients were included. In 03 patients, follow-up showed a clear clinical, biological and radiological improvement and the surgical cure of bilio-bronchial fistula was not indicated. In the 02 other patients, thoracic surgical approach was decided for a destruction of the right pulmonary lower lobe and because of the persistence of a pleural collection. The surgical follow-up was simple. The two operated patients were discharged 07 days after surgery and anti-helminthic treatment was initiated. **Conclusion:** An adequate preoperative patient's preparation combining both the correction of the biological disorders and the introduction of endoscopic sphincterotomy made possible the exclusive thoracotomy with satisfactory results and fistula healing.

Keywords: Bilio- bronchial fistula (BBF); Cholangiography; Hydatidosis; Sphincterotomy; Thoracotomy.

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INTRODUCTION

The bilio-Bronchial fistula is a condition that is defined by an abnormal communication between the bile ducts and the bronchial tree. It is an uncommon but serious complication due to the multiplicity of lesions and the biological repercussions. In developed countries, the bilio-bronchial fistula may be a complication of several aetiologies such as post-traumatic, iatrogenic, post-hepatic surgery or even congenital origin. In Morocco, were this study has been conducted, the hydatid origin is by far the most frequent cause especially in endemic regions [1- 3].

The mortality rate may reach 50% [4, 5]. The prognosis, associated to biliary damage has been always known to be pejorative until the inclusion of the endoscopic retrograde cholangio-pancreaticography (ERCP) in the therapeutic arsenal and the improvement of patients' preparation prior to the surgical procedure. Through the following 05 case series, we report our experience in the management of BBF complicating rupture of hepatic hydatid cysts.

PATIENTS AND METHODS

The study we conducted is a retrospective examination of five cases of bilio- bronchial fistula of hydatid origin, gathered in the thoracic surgery department at Hassan II^d University hospital in Fes, over a 12 months period, between January 2019 and January 2020. The diagnosis was made on the basis of a number of arguments:

- ✓ History of liver Hydatid Cyst whether operated or not.
- ✓ Suggestive clinical symptoms (hemoptysis, jaundice...).
- ✓ Suggestive radiological signs based on chest radiography, hepatobiliary ultrasound and especially chest-abdominal CT.
- ✓ Bronchoscopy findings.
- ✓ Endoscopic retrograde cholangiography findings.

The therapeutic strategy for all our patients was as following:

- ✓ First component is an assessment and correction of associated biological disorders with a drainage of any pleural collection
- ✓ The second therapeutic component consists in the performing of an endoscopic sphincterotomy whenever an obstacle from the bile duct to retrograde cholangiography or to biliary liver ultrasound got individualized.

The surgical treatment was indicated only in case of medical and endoscopic management failure.

RESULTS

Five consecutive patients were considered for the study; there were 4 men and 1 woman with an average age of 50 year old [37- 65]. Two patients had a history of hydatid liver cyst (only one patient was already operated). Major symptoms were dyspnea in 3 patients, hemoptysis in one patient and jaundice in one case. Physical examination revealed signs of severe under-nutrition in one case, a right pleural fluid effusion syndrome in two patients and a mixed pleural effusion syndrome in one case. Acute angiocholitis was reported in one patient. Chest X-ray showed a right side basi-thoracic opacity with air fluid level in 3 cases, a right side basi-thoracic opacity in one case and an aspect of right side extensive pleural effusion in one case. The abdominal ultrasound showed an aspect of a hydatid cyst of the hepatic dome in all patients. Bile ducts were dilated in only one case. The thoracic

abdominal CT scan objectified in all cases the presence of a hepatic dome hydatid cyst communicating either with a pulmonary abscess in 1 case, with a single pleural collection in 1 case, with both a pulmonary abscess and a pleural collection in 2 cases and with a pulmonary abscess and bile ducts in 1 case. All patients underwent a bronchoscopy which established the presence of purulent sputum in 4 patients and bile-tinged sputum in one case. All patients underwent a retrograde per endoscopic cholangiography that detected an obstacle in the bile duct. Therefore, an endoscopic sphincterotomy was performed allowing obstacle removal and the extraction of hydatid membranes. The whole set of iconographies is represented in figures 1 to 5.

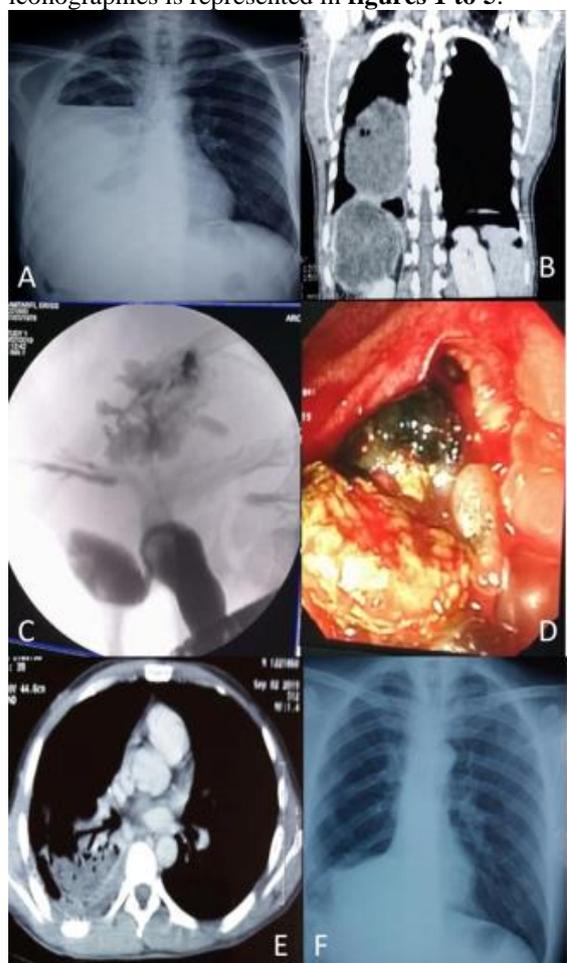


Figure1: Patient #1 imaging and endoscopic iconography:
A. Chest x-ray showing opacity with air fluid level
B. Abdominal chest CT showing hepatic dome hydatid liver cyst communicating with lung abscess
C. ERCP showing common bile duct obstruction.
D. Post endoscopic sphincterotomy image bile and hydatid material
E. Post sphincterotomy chest CT showing disappearance of lung abscess but constitution of lower right lobe condensation
F. Post-operative chest x-ray

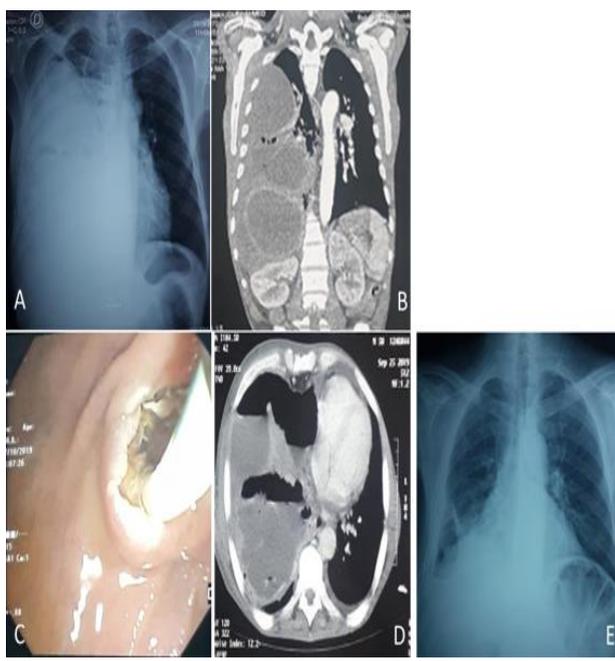


Figure 2: Patient#2 imaging and endoscopic Iconography: **A.** Chest x-ray showing profuse pleural fluid effusion **B.** Abdominal chest CT scan showing hepatic dome hydatid cyst communicating with lung abscess and a pleural collection. **C.** Post endoscopic sphincterotomy image with bile and removed material. **D.** Post sphincterotomy and post-pleural drainage chest CT showing a regression of pleuro pulmonary lesions but persistence of a basal pleural collection. **E.** Post-operative chest x-ray.

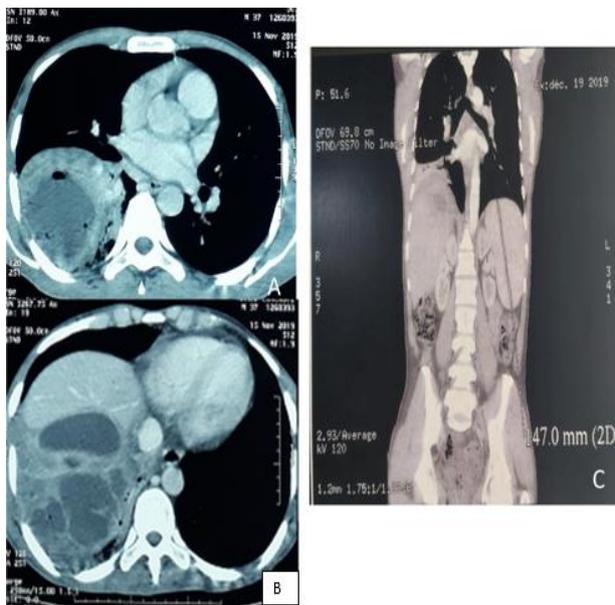


Figure 3: Patient # 3 X-Ray and Endoscopic Iconography: **A & B** Abdominal chest CT showing fistulised liver hydatid cyst in bile duct and communicating with lung abscess

C. Post- sphincterotomy abdominal chest CT showing a disappearance of the lung abscess with a significant decrease in liver lesion size.

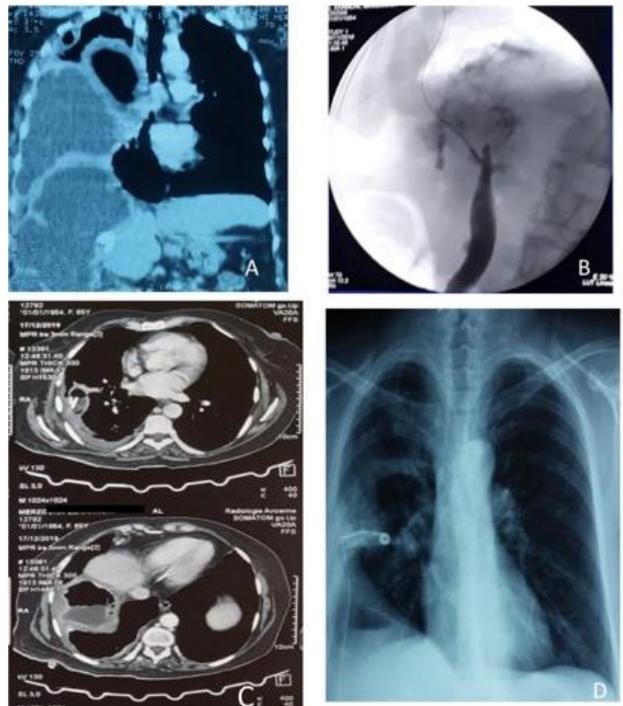


Figure 4: Patient # 4 X-Ray and Endoscopic Iconography: **A.** Abdominal chest CT showing hydatid liver cyst communicating with pleural collection. **B.** ERCP showing common bile duct obstruction. **C.** Post sphincterotomy and post pleural drainage abdominal chest CT showing a pleural collection regression. **D.** Chest x-ray check made one month later and after pleural drainage showing significant radiological improvement.

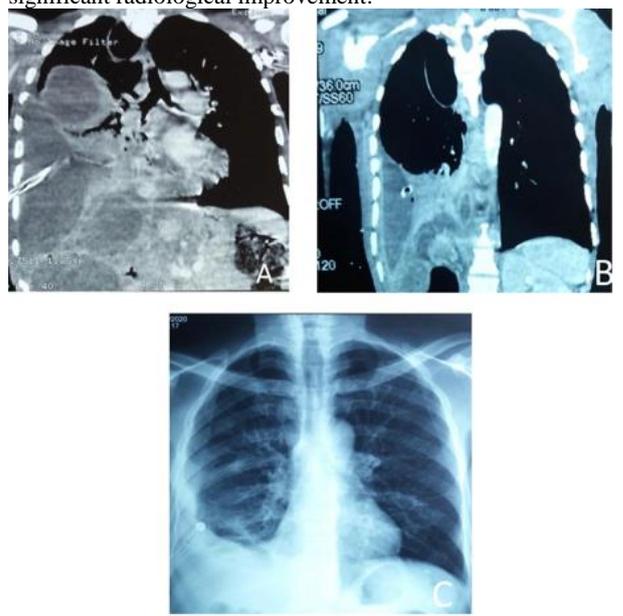


Fig. 5: Patient #5 imaging and endoscopic iconography: **A.** Abdominal chest CT scan showing liver hydatid cyst communicating with lung abscess and pleural collection **B.** Post sphincterotomy and post pleural drainage abdominal chest CT showing regression. **C.** Chest x-ray showing improvement.

chest CT control showing a regression of pleuro pulmonary lesions. C. Chest x-ray one month later showing significant radiological improvement. Biological findings were a hyperleukocytosis associated to anemia with an elevated C reactive protein in all patients. Hypoalbuminemia and hepatic cytolysis were reported in only one case. As a result, the medical management of all patients consisted first in a correction of biological and metabolic disorders associated with broad-spectrum antibiotherapy and a drainage of pleural collection if any. In 3 cases, the drainage of collections allowed the retrieval of purulent liquid with hydatid membranes.

On the five patients, 03 showed a clear clinical, biological and scannographic improvement and thus, surgical abstention for bilio- bronchial fistula cure was decided. However, they were referred to the abdominal surgery unit for management of hepatic hydatid cyst. The other two patients were operated via thoracic approach. The indication was the destruction of the right lower lobe of the lung in the first patient who benefited from a right lower lobectomy and the persistence of a pleural collection in the second patient who benefited from pleuro-pulmonary decortication. In both operated cases, the surgical approach was performed by a posterolateral thoracotomy through the 6th or 7th intercostal space, followed by a phreno-pulmonary disconnection (Fig. 6) meant to avoid the risk of bronchial flooding and bleeding, then the treatment of pleuro-pulmonary lesions seeking a surgery as conservative as possible, alongside the protection of the bronchial stump in case of lobectomy.

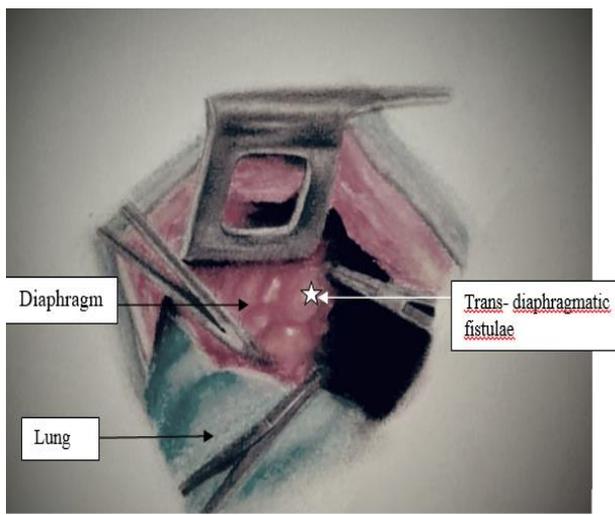


Figure 6: Phreno-pulmonary disconnection leading to evacuation of hydatid membranes interspersed with infected bile through the transdiaphragmatic fistula (Star)

At the level of the diaphragm: This step consists of a hepatic- diaphragmatic disconnection with a resection of the sclerotic edges of the diaphragm (Fig. 7).

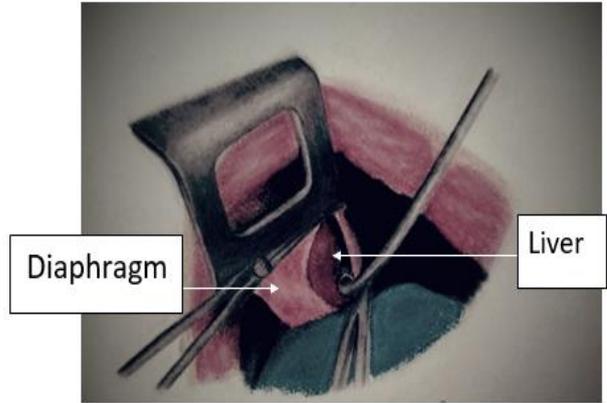


Figure 7: hepato-diaphragmatic disconnection.

At the hepatic level, we usually perform a hepatic pericystectomy, with suture or catheterization of the biliary fistula depending on its diameter (Fig. 8).

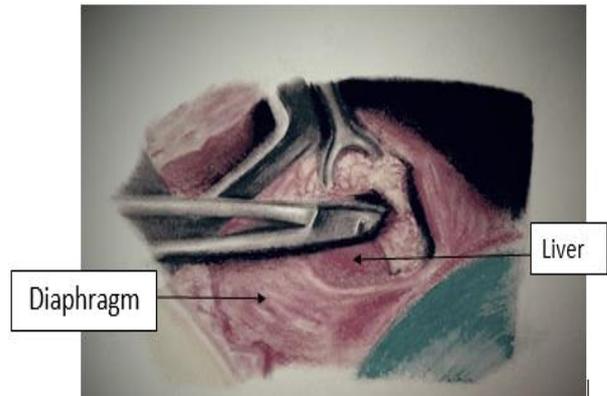


Figure 8: Peri-cystectomy of liver with suture of the biliary fistulae

The procedure was completed with the insertion of a pleural drain and an inter-hepatic-diaphragmatic drain taken out through the abdominal wall, so as to promote a return to normal bile flow. The closure of the diaphragm was accomplished simply by bringing the muscle fibres closer together. The surgical follow-up was simple and both operated patients were discharged within 7 days with the implementation of an anti-helminthic treatment. The whole set of epidemiological, clinical, biological, radiological and therapeutic data is summarized up in Table 1, hereafter:

Table I: Epidemiological, clinical, biological, radiological and therapeutic data of our patients case series.

	Age & Gender	Medical history	Clinical presentation	Chest X-ray	US: Bile duct Dilation	CT-scan	Broncho -scopy	ERCP	Biology	Medical Treatment	Evolution	Surgery
1	43 M	0	-Haemoptysis -Pleural fluid effusion syndrome -Fever	Air fluid level opacity	No	Hepatic dome hydatid cyst communicating with a pulmonary abscess	Bile Tainted Purulent Sputum	Bile duct dilation	Hyperleukocytosis Anemia CRP elevated	Antibiotherapy Tranfusion	Persistence of lower right lobe condensation	Yes
2	56 M	Liver hydatid cyst	-Dyspnoea -Pleural fluid effusion syndrome -Fever -Severe under-nutrition	Extensive pleural effusion	No	hepatic dome hydatid cyst communicating with a pulmonary abscess and with a single pleural collection	Purulent Sputum	Bile duct dilation	Hyperleucocytosis Anemia CRP elevated Hypoalbuminemia	Antibiotherapy Tranfusion of albumin Pleural draining	Persistent pleural collection	Yes
3	37 M	0	Acute angiocholitis	Right side basihoracic opacity	Yes	Fistulized biliary hepatic dome hydatid cyst communicating with a pulmonary abscess	Purulent Sputum	Bile duct dilation	Hyperleukocytosis Anemia CRP elevated	Antibiotherapy Tranfusion	Good evolution Fistula healing	No
4	65 F	Surgery for liver hydatid cyst	Dyspnoea crackling noises	Air fluid level Right side basi-thoracic opacity	No	Liver hydatid cyst communicating With a pleural Collection	Purulent Sputum	Bile duct dilation	Hyperleukocytosis Anemia CRP elevated hepatic Cytolysis	Antibiotherapy Tranfusion Pleural draining	Good evolution Fistula healing	No
5	50 M	0	Dyspnea Mixed pleural fluid effusion syndrome	-Air fluid level -Right side basithoracic opacity	No	Liver hydatid cyst communicating with pulmonary abscess and a pleural collection	Purulent Sputum	Bile duct dilation	Hyperleukocytosis Anemia CRP elevated	Antibiotherapy Tranfusion Pleural draining	Good evolution Fistula healing	No

M: male; F: female; CRP: C - reactive protein; US: abdominal ultrasonography.

DISCUSSION:

The Bilio-Bronchial Fistula (BBF) is an abnormal communication between the bile ducts. The most common etiology in our context is hepatic hydatidosis which can progress into the chest depending on several factors [6- 8]:

- ✚ Intimate contact of the hepatic dome with the diaphragmatic cupola
- ✚ The chest-abdominal pressure gradient which tends to suck away the contents of the liver hydatid cyst into the chest cavity;
- ✚ The diaphragm's erosion by ischemia-necrosis caused by a possible cyst infection alongside inflammatory phenomena;
- ✚ The corrosion of all bile contacted tissues [9].

The positive diagnosis is based on several arguments [10]:

- ✚ A past medical history of Liver Hydatid Cyst whether operated or not
- ✚ Suggestive clinical signs like billiptysis, hydatid vomit, acute angiocholitis or pyothorax signs.
- ✚ Suggestive radiological signs based on chest radiography and chiefly chest-abdominal CT that allow:
- ✓ The diagnosis of the hydatid pathology of the liver, whether associated or not with lung

location, and the Bilio-Bronchial Fistula as a complication

- ✓ Carrying out a comprehensive lesion assessment involving the three stages (lung-diaphragm-liver)
- ✓ Enabling a therapeutic approach in accordance with the results of the investigation.

Thus, the approach to be adopted is determined by an assessment of the biliary ducts condition. In this sense, the role of abdominal ultrasound is essentially the search for dilatation of the bile ducts, a crucial element in the therapeutic decision making process [11].

Bronchoscopy is recommended in the assessment of the bilio bronchial fistula. It makes possible to assess the severity of the bronchial tree lesions by specifying the origin of biliptysis and the extent of the bronchial mucosa inflammation [12]. The magnetic resonance cholangiopancreatography (MRCP) is a potent non-invasive technique that allows a good exploration of the biliary and pancreatic pathways, and an identification of vascular and biliary complications when other examinations fail to be conclusive [13]. In our experience, whenever there are sufficient suggestive clinical, biologic and radiological signs of bilio-bronchial fistula, gastroenterologists perform an ERCP even if the abdominal ultrasound is normal

because we believe that the obstruction of the bile ducts is too recent to be detected. When the obstruction is confirmed by the bile duct fluoroscopy, sphincterotomy is performed and hydatid membranes are removed. In cases with sufficient presumption of bilio bronchial fistula, MRCP is not needed since it'll show the obstruction leading to the same final indication of ERCP and endoscopic sphincterotomy. On the other hand, the ERCP is easily available in our institution and the management of ruptured hydatid cysts with bilio-bronchial fistula has been the subject of previous consensus with the gastroenterology team [14]. In fact, in our series, ERCP objectified a bile duct obstacle in all cases and the BBF diagnosis was made thanks to the thoracic abdominal CT scan, hepatobiliary ultrasound and endoscopic retrograde cholangiopancreatography.

Regarding the therapeutic strategy, it should be noted that patients with bilio-bronchial fistula are often dehydrated (hydro-electrolytical loss related to bile leakage), undernourished (hypo protidemia; lipid digestion disorder) and suffering from anemia and infectious syndrome. Accordingly, the first stage of therapeutic management consists in customized antibiotherapy associated with good respiratory physiotherapy, blood transfusion in case of anemia, a correction of hydro-electrolytic disorders, hyper caloric protein feeding to promote healing and drainage of pleural collection if any. The second therapeutic component consists in carrying out an endoscopic sphincterotomy whenever a biliary obstacle is individualized. This therapeutic strategy enables:

- Patient preparation for the surgical approach by ensuring a reduction of biliptysis thanks to sphincterotomy, thereby decreasing the rate of parenchymal resections for better surgical outcomes.
- An occasional exclusive bilio-bronchial fistula treatment in high surgical risk, and which sometimes even allows healing, as in the case of patients 4 and 5 and especially of number 3 in whom even the liver cystic cavity regressed clearly.

The other two patients required surgical treatment for their bilio-bronchial fistula with exclusive thoracotomy. However in both of them, the pre-operative preparation made possible to operate under better hemodynamic, respiratory and nutritional conditions, with favorable surgical outcomes.

Concerning the technical aspects of surgery, we prefer surgical approach by thoracotomy as it enables a treatment of the lesions at the 3 levels (pulmonary, diaphragmatic, and the hepatic dome) which cannot be achieved by laparotomy [15-18].

According to our experience, we suggest the following algorithm for the management of bilio-bronchial fistula complicating rupture of hydatid cysts (algorithm #1).

CONCLUSION

The Bilio-Bronchial Fistula is a serious complication of hepatic hydatid cysts. For a long period, the surgical management was considered to be difficult with a very pejorative prognosis due to the metabolic impact and the simultaneous impairment of the 3 levels (thoracic, abdominal and diaphragmatic) of the lesions making difficult the decision of a thoracic or an abdominal approach or for a combination of both. However, due to the introduction of retrograde endoscopic cholangiography with sphincterotomy and improvement of preoperative preparation measures with the correction of biological disorders prior to surgery, exclusive thoracotomy is now possible with satisfactory results allowing complete fistula healing in some cases.

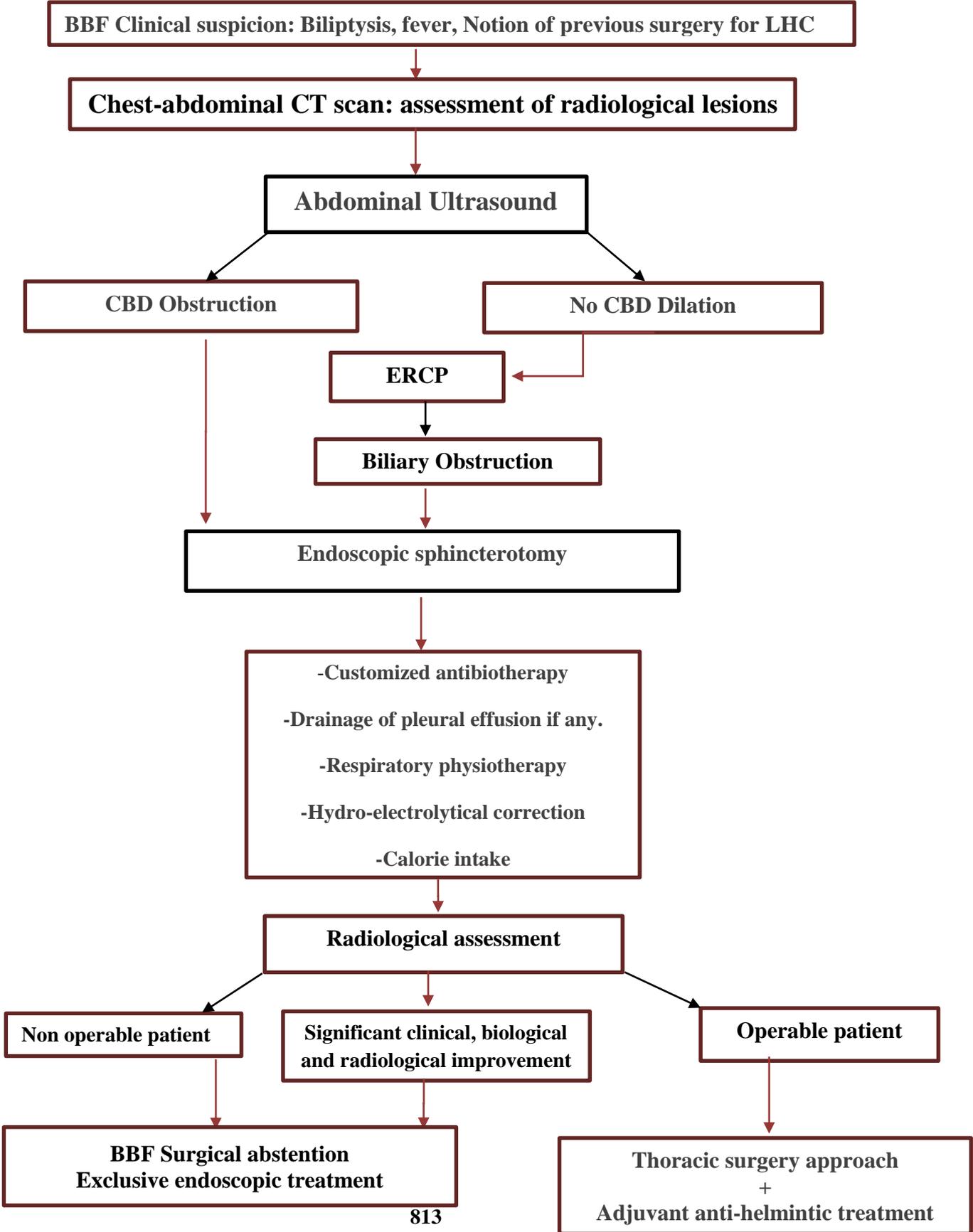
CONFLICT OF INTEREST:

Authors declare no conflict of interest.

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Bilio-bronchial fistula management algorithm



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