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ENDOSCOPIC MANAGEMENT OF RUPTURED HEPATIC HYDATID CYSTS: CLINICAL REFLECTION THROUGH 03 ILLUSTRATIVE CASES

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ABSTRACT

Hydatosis is an infection which affects mostly the liver. Although rare, Rupture of hydatid cysts in biliary ducts is one of the most serious complications of hepatic hydatidosis. Many authors recommended ERCP as the safest therapeutic approach for management of ruptured hydatid cysts, but there is still a real need for recommendations regarding per-procedure aspects and follow-up. We present the experience of a secondary hospital in the management of inaugural biliary complications of hepatic hydatid disease with clinical reflection for improvement of the quality of care.

Keywords: Hepatic hydatid cysts; Endoscopy; ERCP; Rupture.

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INTRODUCTION

Even rare, the rupture of hepatic hydatid cysts (HHC) in biliary ducts is a serious complication of hepatic hydatidosis. If endoscopic retrograde cholangiopancreatography (ERCP) is recommended in the therapeutic strategy of ruptured HHC, there are many challenging technical aspects which need to be discussed and clarified especially the benefit of systematic stent placement, nasobiliary drainage and/or the role of systematic post-procedure albendazole medication in the prevention of possible recurrence. In this article, through 3 consecutive and illustrative cases, we highlighted some issues related to the endoscopic management of patients with biliary complications of hepatic hydatid disease.

CASE REPORTS

Three patients (2 men and one women aged respectively 70, 40 and 32 years) benefited from ERCP for ruptured HHC. The diagnosis was based on clinical presentation and abdominal ultrasonography (US) findings (**Fig. 1**). All patients presented with cholangitis. US examination founded hepatic hydatid cyst(s) associated to the dilation of both intrahepatic biliary ducts (IHBD) and Common bile duct (CBD) (**Fig. 1**). Mean dilated CBD size was 14 mm [12- 18]. The cavity of the ruptured cyst was located in the right lobe in two patients. Involved hepatic segments were segment II (n=1) and VIII (n=2). The mean size of ruptured HHC was 50 mm [34-78]. Cysto-biliary communication was seen in one case.

ERCP with large sphincterotomy was performed in all patients, hydatid cyst material was extracted (**Fig. 2**) and CBD cleared. No biliary stent was placed. Post-procedure evolution was good with jaundice disappearance and no reported complications. Clearance of CBD and jaundice disappearance were considered as success criteria.

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Figure 1: US findings showing voluminous multilocular hydatid cyst (a), dilated IHBD (b) and ruptured hydatid cyst with dilated CBD.

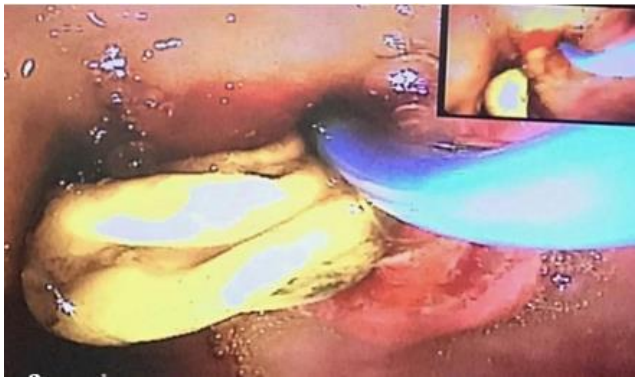

Figure 2: Endoscopic extraction of hydatid cyst membrane using balloon.

Two patients remained asymptomatic during the follow-up period of 6 months. One patient was admitted 4 months later with severe acute abdominal pain and absence of jaundice. US and CT scan showed a hydatid residual cavity with significant reduction of the size of the cyst (>50%) without dilation of biliary ducts. Surgery was decided, the residual cavity explored and a T-tube placed in CBD with good evolution. The **table I** is summarizing all patients' characteristics.

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Table I: Summary of patients' characteristics, procedures details and outcome.

		Case 1	Case 2	Case 3
General Characteristics				
Age		40	32	70
Gender		M	F	M
Clinical Symptoms		Acute jaundice, Severe Pain	Jaundice, fever, moderate pruritus, Severe acute epigastric and right upper quadrant pain	Jaundice 3 months before admission Moderate pruritus.
US Findings		IHBD and CBD dilation		
	Cysts Size (mm)	12	13	18
	Cysts number, respective size, type* & location	n=1; Large cyst (78 mm) Type III* Segment II/III	n=1; 46.5mm Type III* Segment VIII	n=2 46mm / 34mm Type III/IV* Segments VIII/II
Cholestasis Enzymes markers and lipase value		Elevated ALP and GGT. Normal lipase level		
CRP		Elevated	Elevated	Normal
ERCP procedure details				
Large Endoscopic sphincterotomy			Yes	
Need for Needle-knife precut; papillotomy		No	Yes	No
Membranes removal with an extraction balloon and CBD clearance in cholangiogram			Yes	
Need for: -Endoscopic balloon dilatation -Dormia basket -Biliary stenting -Nasobiliary drainage			No	
Patients' outcome and clinical evolution				
Jaundice		Disappearance 5 days after the endoscopic procedures	Disappearance 07 days after the endoscopic procedures	Disappearance 04 days after the endoscopic procedures
Abdominal US		Normal CBD size after 02 days in US control.	Normal CBD size after 02 days in US control.	Normal CBD size 20 days later. US revealed significant reduction of the liver cysts size (>50% of initial diameter).
Post-procedure Complications		No complications related to the ERCP procedure were reported. Discharge on post-procedure day 2. All the patients remained asymptomatic during the follow-up period of 3 months after procedure.		
Albendazole medication (after hepatic enzymes normalization)		Yes	Yes, with no observance	Yes
Outcome (6 months follow-up)		Good evolution. Surgery was indicated later (after 6 months) in an asymptomatic patient in order to treat the hydatid disease.	After 4 months, the patient was admitted with severe acute abdominal pain. US and CT scan show significant reduction of cyst size (>50%) without CBD dilation (Recurrence?). Surgery exploration of the residual cavity was decided and T-tube placed in CBD with good evolution. Follow-up was uneventful	Good evolution. Surgery was not indicated. Decision for albendazole medication only.

DISCUSSION

Management of the hepatic hydatid disease is possible by percutaneous approach, [**1**] surgery [**2**] and/or albendazole medication. [**3**] Intra-biliary rupture is a rare but serious complication of HHC with an estimated incidence of 3-17%. [**4**] ERCP is recommended in ruptured HHC; however, some technical aspects are still not clearly stated especially the benefit of systematic stent placement, nasobiliary

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drainage (NBD) and/or systematic post-procedure albendazole medication with no available guidelines. In this article, we're presenting issues related to the endoscopic management of ruptured HHC through 3 illustrative cases.

Common clinical presentation is severe pain, jaundice and angiocholitis.[**1**] Abdominal US in the hand of experimented radiologist and gastroenterologist can easily assess the diagnosis. It shows hydatid cyst cavities and allows measurement and classification.[**1**] CBD and IHBD dilation can be easily identified in US and in some cases, biliary communication could be shown. [**5**] Computed tomography (CT) increases the diagnosis accuracy when coupled to US, [**6**] but is not easily performed in our institution. If scanographic findings are mandatory before hydatidosis surgery, endoscopic approach doesn't require systematically CT exploration. In our case-series, it was necessary to perform CT scan in one patient in order to eliminate a malignant cause of biliary obstruction (choloangiocarcinoma...) considering both patient's age (70 years old) and the long delay between jaundice appearance and admission to the hospital (3 months). Magnetic resonance cholangio-pancreatography (MRCP) is not available in our institution, and if MRCP exploration provides an added value in the management of CBD stones [7], our conviction is that MRCP is useless in case of ruptured HHC. Some authors recommend MRCP only in difficult cases when US and CT scan were not conclusive. [**6**] In some centers, ERCP is still indicated as a part of the diagnostic approach in case of rupture of hydatid cysts [**8].** This is subject to controversies; as ERCP is an invasive technique with potential risk of complications, we believe that it should be limited only to therapeutic indications.

For long time, surgery was the preferred approach for the management of biliary complications of HHC.[**2**] Actually, ERCP with large sphincterotomy is accepted as a safe modality for hydatid membranes removal and for the management of ruptured HHC. [**9, 10]** However, lot of technical aspects of the endoscopic approach are still unclear. In real-life practice, and in the absence of guidelines, the placement of post- procedure endoscopic biliary stenting -or nasobiliary drainage (NBD)- is subject to the personal consideration of the operator or the experience of the centers where ERCP is performed. Some authors advanced that the biliary stenting in association to endoscopic sphincterotomy (ES) may be considered during the initial procedure in a subpopulation of patients (incomplete evacuation of hydatid material, biliary stricture) [**11**]. In some cases where repeat ERCP and stenting was indicated, Akcakaya et al. showed that the procedure failure was secondary to incomplete evacuation of the CBD or probable obstruction by residual hydatid material or unnoticed stones. It was also demonstrated that stenting during the initial procedure reduced the delay for fistula closure and improved ERCP's efficiency rate [**11**]; stenting associated to sphincterotomy reduces significantly the pressure in the biliary tract. In experimental models, stent placement or ES with stent placement significantly reduced CBD pressure as compared to ES alone. [**12**] Biliary stenting provides sufficient drainage as it bypasses sphincter mechanism and reduces delay to fistula closure [**11**]. It's important to mention that placing a biliary stent is certainly costly with a need to repeat ERCP for prothesis removal.

The benefit of the prescription of systematic anti-helminthis medication after the procedure in order to treat the residual cavity and maybe prevent a possible recurrence is not stated. No guidelines or expert recommendations are available.

Our case-series are illustrating 3 different conditions. In the first case, ERCP was successful and cholangitis treated; surgery was indicated 6 months later for the treatment of the residual cavity since it was large (> 5cm). In some specialized centers, percutaneous drainage could be suggested as a non-invasive alternative to surgery. In the second patient, ERCP was also successful with good clinical evolution; however, the patient presented 4 months later with what seems to be a recurrence even after large sphincterotomy and despite of the reduced size of the residual cavity (<50%). Was systematic biliary stenting or NBD an option to be considered in this patient? In the 3rd case, endoscopic approach was sufficient; we started medication by albendazole after normalization of liver enzymes as a treatment for the hydatid disease and surgery was not needed.

If ERCP is indicated for the treatment of the acute cholangitis following the rupture of HHC, surgery (or adjuvant percutaneous drainage in some centers) may be considered later as a complement for the treatment of the hydatid disease. Delayed surgery indications are large ruptured hydatid cyst (**case 1**), a significant persistence of the residual cavity (> 50% of initial cyst's size and/or a diameter >5cm) and the presence of multiple hepatic hydatid cysts. There are no recommendations concerning the delay for surgery in case of persistent residual cyst. In our opinion, eight weeks after ERCP seems to be a good delay for decision. Also, there is no evidence concerning the benefit of systematic biliary stent placement or post-ERCP systematic albendazole medication in order to prevent early recurrence (case 2). Many authors opted for the use of biliary stenting

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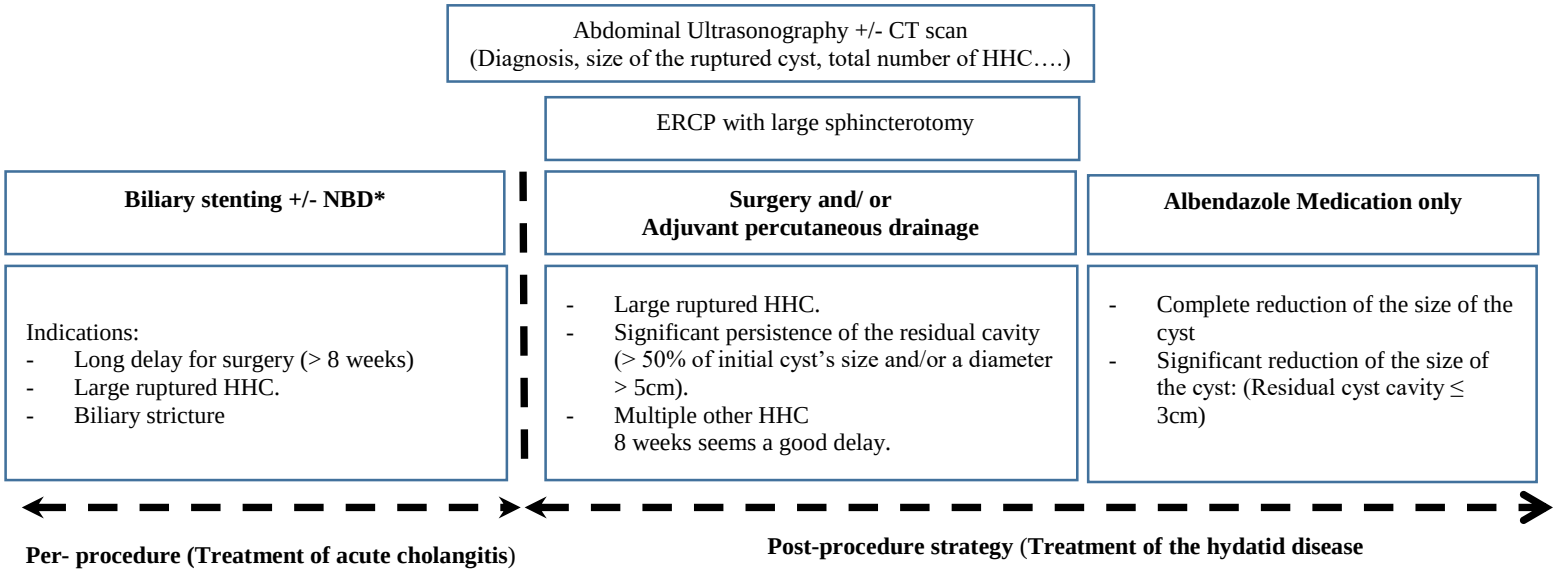
or NBD. [13] Endoscopically placed NBD catheter with saline infusion in the cystic cavity was suggested by some centers as a curative approach avoiding the need for surgery. [13] In patients with large ruptured cyst or multiple HHC still requiring surgical intervention, we believe that biliary stent placement seems to be a good option if the delay for surgery is long (> 8 weeks) in order to prevent early recurrence. Coordination between gastroenterolgist and general surgeon is mandatory! In case of significant or complete reduction of the size of the ruptured cyst (**case 3**), albendazole medication is indicated as an adjuvant therapy after normalization of hepatic enzymes and may be sufficient after endoscopy for the treatment of the hydatid disease. Dehkordi et al stated that albendazole may prevent recurrence of hydatidosis and is associated with the death of the cyst and a reduction of its size. [**14]**
In our patients, no case of pancreatitis was reported. However, the risk of pancreatitis is there and in some cases, necrotizing pancreatitis was inaugural as a consequence of hydatid membranes impaction in the Water ampulla. [**15]** Regarding the intensity of acute abdominal pain, lipase test was performed in all patients to eliminate an inaugural pancreatitis. The following is a suggestion of a decisional algorithm (**Table II**).

Table II: Decisional algorithm for per and post ERCP management of ruptured HHC



(*) With post-ERCP daily saline infusion; HHC: Hepatic hydatid cyst; NBD: Nasobiliary drainage.

CONCLUSION

Intra-biliary rupture of HHC is a rare but serious condition. Clinical observation, biology and abdominal US are sufficient for both diagnosis and follow-up. ERCP is the best therapeutic approach. However, many issues are associated to endoscopic management and need to be discussed, especially the benefit of systematic stent placement and/or the value of systematic post-procedure albendazole medication. Experts meeting should be encouraged in order to make available clear guidelines for the management of ruptured HHC during and following endoscopy.

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CONFLICT OF INTERESTS:
There are no competing interests to declare.

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