

Endoscopic Treatment of Biliary Complications of Hydatid Cysts of the Liver

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Abstract

Case Report

Objective: To evaluate the contribution of endoscopic retrograde cholangio-pancreatography (ERCP) in the diagnostic and especially therapeutic management of biliary complications of hydatid liver cysts (HHF). **Patients and method:** We included 19 patients hospitalized for endoscopic management of biliary complications of hydatid cyst of the liver over a four-year period, with an average age of 42 years and a male predominance in 62% of cases. **Results:** The prevalence of biliary complications of hydatid cyst of the liver was 2.5% in our series. ERCP revealed a cystobiliary fistula in 31% of cases, and lacunar images in the main bile duct (MBD) in 69% of cases related to hydatid material. Sphincterotomy was performed in all patients, allowing extraction of hydatid material by balloon extraction, and dilatation of the MBD by candle. On average, jaundice disappeared 7 to 12 days after the endoscopic procedure. No complications related to endoscopic treatment were observed. **Conclusion:** Endoscopic treatment of the biliary complications of hepatic hydatidosis is a safe and effective therapy with low morbidity and mortality.

Keywords: Liver hydatid cyst - Biliary complication - Biliary fistula - Endoscopic retrograde cholangiopancreatography - Endoscopic biliary sphincterotomy.

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INTRODUCTION

Hydatid cyst of the liver is a parasitic condition caused by the development in the human body of the larval form of a dog taenia known as “Echinococcus granulosus” [1]. The disease is endemic in Morocco, but despite its frequency, diagnosis is often delayed, and complications arise. The most frequent complication is fistulization of the cyst in the bile ducts, which in most cases occurs in young working people [2, 3]. The development of endoscopic and interventional radiology techniques has improved the management of hydatid cysts of the liver (HCH), and in particular of their main complications [3]. The aim of this study is to evaluate the contribution of endoscopic retrograde cholangio-pancreatography (ERCP) to the diagnostic and, above all, therapeutic management of complicated hydatid cysts in the bile ducts.

OBSERVATION

Given the diversity of diagnostic and therapeutic modalities for bilio-cystic fistulas, a retrospective study was conducted based on a series of 50 cases of KHF rupture in the bile ducts collected in the

visceral surgery department of ARRAZI at the MOHAMMED VI University Hospital Center in Marrakech, over a four-year period from May 2016 to May 2020 ; ERCP and endoscopic biliary sphincterotomy were performed in 19 patients, with an average age of 42 years and a male predominance in 62% of cases. Two hundred ERCPs were performed over this four-year period. In 19 cases, the indication was hydatid cyst of the liver complicated in the bile ducts. 2.5% of indications for ERCP in our series. Clinical symptomatology was marked by persistent external biliary fistula postoperatively in 30% of cases, retentional jaundice in 30% and angiocholitic syndrome in 40%. All patients presented with cytotoxicity and/or biological cholestasis. Abdominal ultrasonography and abdominal tomendositometry (Figures 1, 2, 3, 4) were performed in all patients, and revealed cystic lesions in all, intra- and/or extrahepatic bile duct dilatation in 80%, and the presence of hydatid material in the main bile duct (MBD) in 40% of patients. ERCP was performed in all patients: 8 preoperatively and 11 postoperatively. A cystobiliary fistula was found in 31% of cases, and lacunar images in the MBD (Figure 5) in 69% of cases, associated with hydatid material. Endoscopic biliary

sphincterotomy was performed in all patients, with extraction of hydatid material by balloon extraction. The cyst lumen communicating with the intrahepatic bile ducts could only be reached and drained in half the cases. In the other half of cases, the cyst lumen could not be

reached endoscopically. Progression was marked by the disappearance of jaundice on average 7 to 12 days after the endoscopic procedure, and the external biliary fistula dried up after 10 to 12 days. No complications related to the endoscopic treatment were observed.



Figure 1: Ultrasound section showing dilatation of the MBD containing hydatid echogenic material



Figure 2: CT section of left hydatid cyst of the liver with Intrahepatic bile duct dilatation

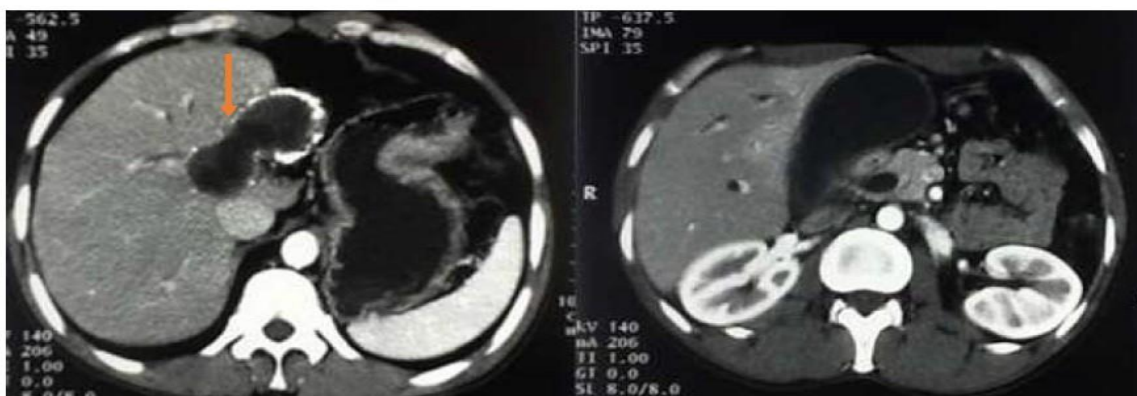


Figure 3: Axial cross-section CT scan of a bilobed, partially calcified hydatid cyst with dilatation of the Intrahepatic bile duct and MBD

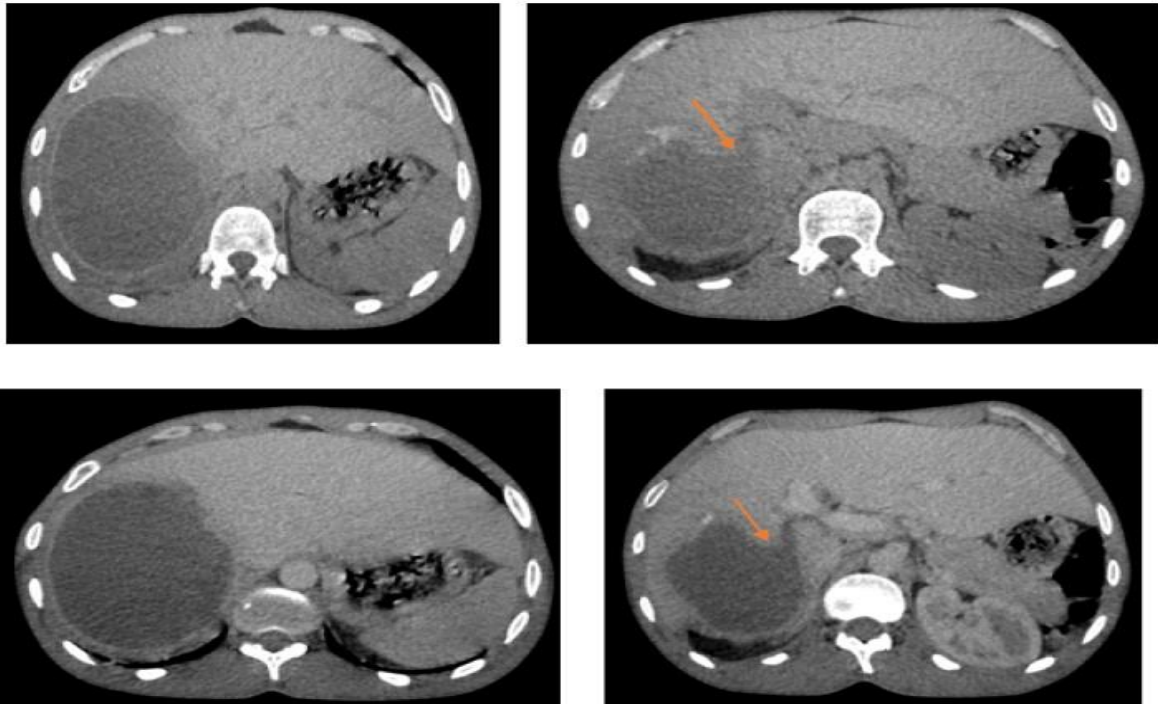


Figure 4: CT sections before and after contrast injection of a right hydatid cyst of the liver communicating with a segmental duct via a kysto-biliary fistula



Figure 5: Endoscopic retrograde per-cholangiography image showing hydatid material within a dilated main bile duct

DISCUSSION

Hydatidosis or echinococcosis is a cosmopolitan anthrozoosis, caused by the development in the human organism of the larval form of a dog taenia known as “Echinococcus granulosus” [1-3]. It is endemic in Mediterranean countries such as Morocco [2,3], and constitutes a real public health problem in terms of frequency, morbidity and potential mortality [1, 3, 4]. A predominant disease in developing livestock-raising countries, hydatidosis can affect up to 5% of the population in highly endemic areas [4]. In the Maghreb, Morocco ranks third in terms of incidence of hydatid disease after Tunisia and Algeria, with an incidence of 4.55 operated cases per 100,000 inhabitants,

according to statistics from the Directorate of Epidemiology and Disease Control in 2006 (DELM, 2006). This parasitosis can affect all organs, but the hepatic localization is the most frequent [4], secondary to the obligatory passage of the parasite through the portal trunk [3]. The hepatic localization of hydatid cysts is classically the most common in adults, accounting for around 2/3 of all hydatid localizations [3]. Liver hydatid cysts (HHC) often evolve for several years before clinical manifestations appear [1]. A benign pathology in itself, but a worrying one due to its complications, the most frequent and most serious of which is an opening in the bile ducts [2], which can sometimes be life-threatening. Indeed, the rupture of the KHF into the bile

ducts was first described by Drewen 1928 [5], who considered that only large ruptures in excess of 5mm accompanied by significant migration of daughter vesicles and hydatid debris into the biliary tree were considered to be biliocystic communications. While Bourgeon [6] has demonstrated that even small ruptures in the fine bile ducts should be classified as such complications. The frequency of these cystobiliary fistulas varies from one surgical series to another, with estimates ranging from 9 to 25% [7], and from 17 to 44% of operated hydatid cysts [8]. Hydatid cysts of the liver open into the bile ducts in 2.5 to 6% of cases [8]. In our series, this figure is 2.5%. In its typical form, the opening in the bile ducts manifests itself as acute angiocholitis in 2/3 of cases [4] in our study 40% of cases. Advances in medical imaging have greatly facilitated the diagnosis of KHF and its complications. Indeed, the diagnosis of kystobiliary fistulas can be suggested by indirect images on hepatobiliary ultrasound and abdominal CT scans, while MRI cholangio remains an essential examination for the study of biliary tract anomalies. Indication for endoscopic surgery is based essentially on MRI data, but its cost and lack of availability in most centers limit its use. The contribution of cholangio-MRI is twofold: in the diagnostic and etiological assessment, it provides information on the state of the bile and pancreatic ducts, whether or not they are dilated; while in the therapeutic approach, the information provided helps to guide the therapeutic approach. MRI is currently establishing itself as the second-line morphological examination in the assessment of hepatic hydatidosis. These different explorations help to clarify the anatomical features of the hydatid cyst of the liver, to detect latent complications and to search for other hydatid localizations in order to guide the therapeutic strategy [9]. In our series, all patients underwent abdominal ultrasonography and abdominal computed tomography. ERCP remains the method of choice for the diagnosis and treatment of biliary complications of hydatid cyst of the liver. It detects hydatid cyst of the liver rupture in the bile ducts in 86.6 to 100% of cases [10,11]. It is an invasive technique involving catheterization of the papilla, performed during icterus, preferably the day before surgery. It enables the relationship between ruptured cysts and the bile ducts to be better defined, not only by visualizing intracholecystic hydatid debris, but also the location and size of cyst-biliary communications. In addition to the diagnostic value of ERCP, the second interest is therapeutic. Given the significant complications and morbidity associated with surgical treatment of biliary complications of hydatid cyst of the liver [1,10,11], surgical sphincterotomy has been advocated by Goinard since the 1960s, but still carries a high risk of external biliary fistula [11]. In our series, ERCP was performed in 19 patients, and revealed a cystobiliary fistula in 31% of cases, and lacunar images in the VBP in 69%, related to the presence of hydatid material in the MBD. Surgical treatment is associated with a non-negligible rate of complications (38% of cases), such as biliary leakage, infection of the residual

cavity, recurrence or dissemination of the disease, often necessitating reoperation. Mortality was 4.5% in the series by Zaouche *et al.*, [12]. Endoscopic sphincterotomy ensures better internal bile drainage, with preferential flow of bile to the duodenum, thus promoting healing of the biliary fistula [1,10]. It has been proposed as an alternative to surgery since its value in the management of biliary complications of hydatid cyst of the liver was first reported by Alkarawi *et al.*, in 1985 [13]. It allows easy access to the MBD, reduces pressure in the oddian sphincter, ensures better internal drainage with preferential flow of bile to the duodenum, thus promoting healing of the kystobiliary fistula and reducing the risk of postoperative fistula [12,13]. The MBD can be evacuated using a balloon catheter or a Dormia-type basket catheter [13] in the event of hydatid obstruction. In our series, all patients underwent endoscopic sphincterotomy with balloon extraction of hydatid material. Nasobiliary drainage is indicated whenever bile duct vacuity is uncertain or there is a risk of bile superinfection. It enables bile duct decompression to be monitored, and cholangiographic tests to be repeated if necessary. In the case of MBD stenosis, biliary catheterization is used to ensure that the stenosis is benign by brushing, and to perform balloon or bougie dilatation. In some cases, a plastic biliary prosthesis can be fitted to calibrate the stenosis [12,13]. Despite a success rate of around 90%, biliary catheterization has an overall morbidity rate of between 4 and 10%, and a mortality rate of 0.5% [14]. Indeed, the main complications of biliary catheterization with endoscopic sphincterotomy are, in order of frequency, acute pancreatitis (5.4%), hemorrhage (2%), biliary infection (1.5%) and perforation (0.3%) [14]. All in all, retrograde bile duct catheterization can be proposed as an alternative treatment for biliary complications of hydatid cyst of the liver, either preoperatively, as in the case of angiocholitis, in the presence of indirect signs of kystobiliary fistula, or finally, some authors propose preventive endoscopic sphincterotomy. ERCP is also proposed postoperatively, in cases of persistent external biliary fistula or in the event of a hydatid or iatrogenic obstruction of the MBD [15].

CONCLUSION

Endoscopic sphincterotomy represents a therapeutic advance in biliary complications of hepatic echinococcosis. It is an innovative treatment that has already demonstrated its efficacy and safety. It shortens the post-operative stay and avoids the need for a repeat operation, which is often difficult and hemorrhagic.

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