

Case series

Acute pancreatitis as a rare complication of rupture of hydatid liver cyst: review of 16 cases



Nada Lahmidani^{1,&}, Fatime Zahra Hamdoun¹, Hakima Abid¹, Mounia El Yousfi¹, Dafr Allah Benajah¹, Mohammed El Abkari¹, Sidi Adil Ibrahimi¹

¹Department of Gastroenterology, University Hospital Hassan II, Faculty of Medicine and Pharmacy of Fez, Fez, Morocco

[&]Corresponding author: Nada Lahmidani, Department of Gastroenterology, University Hospital Hassan II, Faculty of Medicine and Pharmacy of Fez, Fez, Morocco

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Abstract

Hydatid disease HD or echinococcosis is a parasitic disease caused by *Echinococcus granulosus*. The prevalence of this parasitic disease in Morocco approximates 2 to 3% in highly endemic regions such Atlas Mountains. Liver is the main affected organ, and cholangitis is the most frequent complication of Hydatid liver cyst, however acute pancreatitis remain a very rare complication of the disease as showed by the very few cases reported in the literature. Its treatment combines the management of acute pancreatitis, endoscopic retrograde cholangiopancreatography (ERCP) and surgical treatment of the cyst. We report a series of 16 patients diagnosed for hydatid cyst of liver with pancreatitis at the University Hospital Hassan II to show the interest and outcomes of the management of such complication . We retrospectively review the clinical presentation and outcomes of management of 16 patients admitted for hydatid cyst of liver with acute pancreatitis at the department of hepato gastroenterology at the university Hospital Hassan II between January 2013 and July 2018. The average age of our patients was 44.33 years [20;66] with a female predominance. Only 12.5% (n = 2) of the patients had past history of surgical cure of liver hydatid cyst. All patients had acute pancreatitis associated with cholangitis. Abdominal CT scan showed oedematous pancreatitis in 87.5% (n = 14) and the necrotizing pancreatitis in 12.5% (n = 2). The common bile duct (CBD) was dilated in all patients and the kystobiliary fistula was identified in 68.75% (n=11). ERCP was performed for all our patients, cholangiogram showed filling defect in distal CBD and a communication between intra-hepatic biliary duct with the hydatid cyst. Sphincterotomy was performed with evacuation of membranes in 87,5%. Acute pancreatitis is an exceptional complication of hydatid liver cyst. It is often associated with cholangitis. The ERCP combined to surgery remain the key treatment.

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Introduction

Hydatid disease HD or echinococcosis is a parasitic disease caused by *Echinococcus granulosus*. This disease is endemic in Mediterranean countries, including Morocco [1]. Liver is the main affected organ, and cholangitis is the most frequent complication of hydatid liver cyst, however pancreatitis remain an exceptional presentation of the disease [2]. Its treatment combines the management of acute pancreatitis and endoscopic retrograde cholangiopancreatography (ERCP) to free the biliary duct and taring the fistula then resection of the cyst under general anesthesia. We report a series of 16 patients diagnosed for hydatid cyst of liver with pancreatitis at the University Hospital Hassan II to show the interest and outcomes of such complication.

Methods

It is a retrospective series of patients hospitalized for hydatid cyst of liver with acute pancreatitis at the Department of Hepato Gastroenterology at the University Hospital Hassan II between January 2013 and July 2018. The study focused on the epidemiological, clinical, CT abdomen and ERCP characteristics as well as their evolution after sphincterotomy. The diagnosis of acute pancreatitis was based on clinical presentation of abdominal pain associated with lipase or amylase movement superior to 2 fold the normal laboratory value. The diagnosis of hydatid origin of the pancreatitis was based on the absence of biliary duct stones in CT and cholangiography with identification of communication between the cyst and the biliary duct or presence of hydatid membranes at the ERCP.

Results

On a series of 16 patients hospitalized for hydatid cyst of liver with acute pancreatitis, the average age was 44.33 years (20;

66) with a female predominance. The sex ratio F/H was 1.3. Only 12.5% (n = 2) of the patients had past history of surgical cure of liver hydatid cyst. All patients had acute pancreatitis associated with cholangitis. More than half of our patients (56.25%) had systemic inflammatory response syndrome at the admission that required hospitalization in intensive care unit. Serum lipase +/-amylase was high in all our patients with a median of 2.6 folds the normal value. The hydatid liver cyst was classified stage II according to Gharbi classification in 68.7% of patients, stage III in 25% ant stage IV in 6.25%. The size of the cyst varied between 4 and 9cm (Table 1). CT abdomen showed edematous pancreatitis in 87.5% and necrotizing pancreatitis in 12.5%. The common bile duct (CBD) was dilated and the kystobiliary fistula was identified in 68.75% (Figure 1). ERCP was performed for all our patients, cholangiogram showed filling defect in distal CBD and a communication between intra-hepatic biliary duct and the hydatid cyst in 10 cases. Sphincterotomy was performed with evacuation of membranes in 87.5%. In 12.5% the CBD was free (Table 2). All patients had surgical cure of their hydatid liver cyst under general anesthesia. We count one anaphylactic shock following postoperative rupture of the cyst, and one infection of the residual cavity treated by percutaneous drainage afterwards.

Discussion

Acute pancreatitis is a rare complication of hydatid disease ruptured into the biliary tree [3]. Cystic hydatid disease usually affects the liver (50-70%) and less frequently the lung, the spleen, the kidney, the bones, and the brain [4]. The diagnosis is most easily set by serology test that is sensitive for liver cysts in 80-100% and ultrasound or other imaging techniques such as CT abdomen or MRI [5]. The classic treatment for hydatid cysts ruptured into the bile ducts is surgery with exploration of the CBD through a choledochotomy, placement of a T tube, clearance of cyst remnants and surgical excision of the hydatid

cyst or cysts, either by enucleation or by peri cystectomy and partial hepatectomy [3]. However, such operations are associated with morbidity, mortality and prolonged hospitalization hence the use of ERCP with sphincterotomy. Al Karawi *et al.* have reported on a series of 6 patients with intrabiliary rupture of hydatid cysts, which they treated with endoscopic sphincterotomy, hydatid material extraction with balloon and basket catheters and hypertonic saline lavage through a nasobiliary drain [6]. In the specific setting of acute pancreatitis due to hydatid disease ruptured into the biliary tree, there are many cases reports of treatment with endoscopic sphincterotomy [7]. In our series, acute pancreatitis secondary to hydatid disease was documented by clinical presentation, raised serum amylase and aspect of CT abdomen which revealed dilatation of extra and intrahepatic biliary tree and identified kystobiliary fistula in 68.75%. ERCP and sphincterotomy were performed in all our patients with extraction of hydatid material from the common bile duct in 87.5%, however the common bile duct was free in 12.5%. Our patients avoided painful operations and lengthy hospital stay waiting for resection of the cyst under general anesthesia.

Conclusion

Acute pancreatitis is an exceptional complication of hydatid disease. Imaging technique allows the diagnosis and visualization of the kystobiliary fistula in 2/3 of cases. The couple ERCP and surgery remains the key to treatment.

What is known about the topic

- Hydatid echinococcosis is an endemic parasitic disease in Morocco;
- Pancreatitis secondary to hydatid liver cyst is a rare situation , only few cases have been reported in the literature;
- Surgery of the cyst was the predominant treatment.

What this study adds

- The largest published series in the field;
- Particular Clinical presentation of the cases;
- Highlight of the importance of ERCP to treat the intrabiliary rupture of the liver cyst and to alleviate the severity of the pancreatitis.

Competing interests

The authors declare no competing interests.

Authors' contributions

Nada Lahmidani: writing and drafting the article; Fatime Zahra Hamdoun: co writing of the manuscript; Hakima Abid: registration of cases and performing data collection; Mounia El Yousfi: wrote the paper with input from all authors; Dafr Allah Benajah: planning and supervision of the work; Mohammed El Abkari: revising the manuscript; Sidi Adil Ibrahimi: revising and supervising the manuscript. All the authors have read and agreed to the final manuscript.

Tables and figure

Table 1: radiological characteristics of hydatid cyst

Table 2: results of management of acute pancreatitis with hydatid cyst

Figure 1: hydatid cyst responsible for dilatation of extra-and intra-hepatic biliary tree, with filling defects of varying size and shape due to the presence of hydatid material into the bile ducts

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Radiological characteristics	Results
Number of hydatid liver cyst	Multiple 30%
Hepatic segment	IV and VIII 30%
Hydatid liver cyst size	4 to 9 cm
Common bile duct dilated	All patients 100%
GHARBI classification:	
Stage II	68.7%
stage III	25%
stage IV	6.25%
Balthazar classification the pancreatitis:	
A	40%
B	27%
C	20%
E	12.5%
Identification of the kystobiliary fistula	68.75%

Table 2: results of management of acute pancreatitis with hydatid cyst	
Treatment	results
Endoscopic retrograde cholangiopancreatography (ERCP)	
Sphincterotomy	100%
Evacuation of membranes	81.25%
Free common bile duct (CBD)	12.5%
Surgery resection of the cyst	100%
Postoperative complications	
Anaphylactic shock	6.25%
Infection of the residual cavity	6.25%

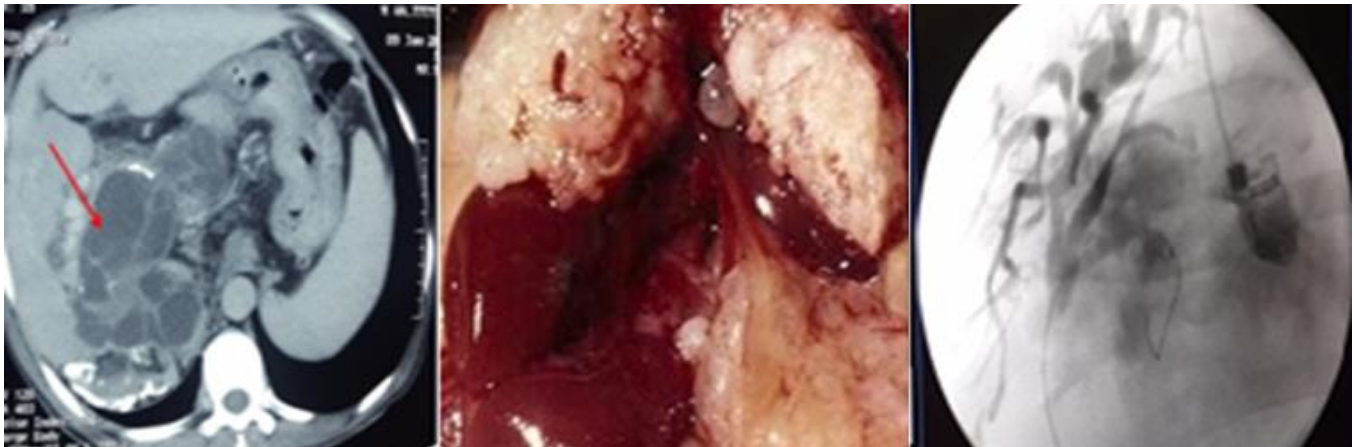


Figure 1: hydatid cyst responsible for dilatation of extra- and intra-hepatic biliary tree, with filling defects of varying size and shape due to the presence of hydatid material into the bile ducts