



Acute Recurrent Pancreatitis Secondary to Biliary Cystic Fistula of Liver Hydatid Cyst. An Exceptional Cause of Acute Recurrent Pancreatitis Treated by Endoscopic Sphincterotomy

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Abstract

Human hydatid cyst is an endemic disease and its distribution is worldwide affecting many countries in the Northern Africa as well as other parts of the world. This disease is a major public health problem in our country. The incidence of the disease has been increasing in the absence of an effective prevention policy. In addition to liver and lung localization representing 90% of the disease, other organs and tissues are spared. Acute recurrent pancreatitis caused by the presence of hydatid membranes in the biliary tract is very rare disease. This is considered unusual. We report a case of acute recurrent pancreatitis in a female patient who had presented an acute pancreatitis six months ago.

Keywords: Acute recurrent pancreatitis; Hydatid cyst; Biliary cystic fistula; Surgery; ERCP and Endoscopic Sphincterotomy

Introduction

Hydatidosis is a zoonosis that is generally caused by infection with tapeworms (*Echinococcus granulosus*). When the parasite eggs are ingested by human beings, the infective forms penetrate the intestinal wall and reach the liver via the portal circulation. In some cases, the hydatid cyst subsequently ruptures into the biliary tract [1]. Patients who have pancreatitis caused by hydatid membranes in the biliary tract have been treated, generally, by surgery. There are a few previous case reports in the international literature of acute pancreatitis an exceptional cases as acute recurrent pancreatitis as a complication of hepatic hydatid disease ruptured into the biliary tree. We present a single case of acute recurrent pancreatitis caused by the presence of hydatid membranes in the biliary tract treated by endoscopic sphincterotomy.

Case Presentation

A 46 years old female patient was oriented to our hospital with epigastric pain radiating to the back, fever, nausea and vomiting over a period of 48 h. She is followed up and treated for diabetes mellitus (type 2) for 9 years. She had a history of a first attack of acute pancreatitis six months ago with diagnosis of hydatid cyst of liver (segment VII with 6 cm of diameter). Laboratory investigations showed serum lipasemia =1255UI/L (>3xn). She was managed medically for her acute pancreatitis and operated for the hydatid cyst of the liver with cholecystectomy (October 2018). There was fistula between hydatid's cyst cavity and biliary tree treated by drainage. One month later the patient developed a second attack of acute pancreatitis "acute recurrent pancreatitis". Biliary RMI showed voluminous residual cavity of hydatid cyst in the segments VII & VII of the liver measuring 60 cm × 70 cm accompanying with discrete dilatation of CBD (7 mm) with biliary cystic fistula allied with a material of hydatid cyst in the CBD. Pancreas was globally enlarged associated with castings of necrosis. The Computed Tomography showed enlarged pancreas gland classified as pancreatitis stage E of Balthazar (Figure 1) with CTSI of 2. There was an extended thrombosis of splenic vein with a small intra-abdominal collection of 1.9 cm × 2 cm.

Discussion

Hydatid cystic disease is a illness that has been known since antiquity and was described by Hippocrates with the particular term "liver filled with water", followed by the famous Arabian

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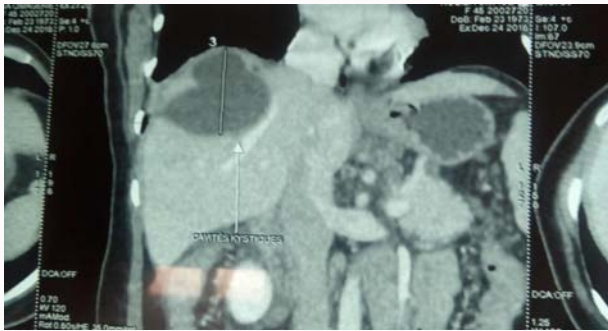


Figure 1: Residual cavity of Hydatid cyst of the liver.

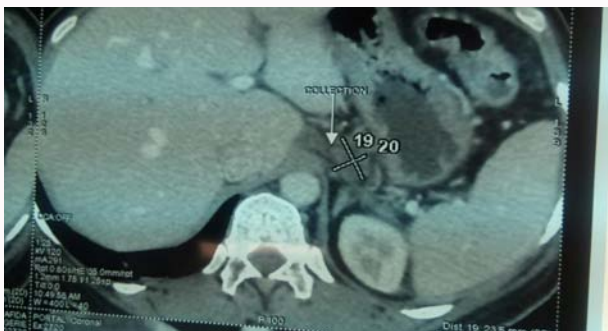


Figure 2: Acute recurrent pancreatitis with enlargement of the pancreatic gland.

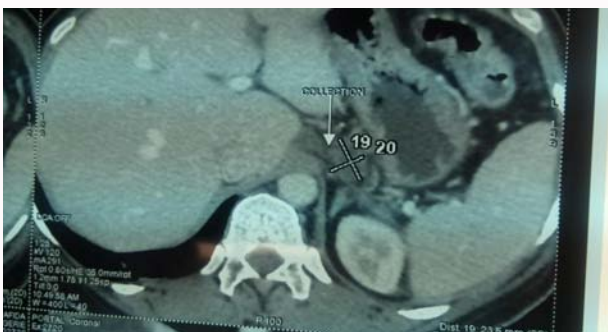


Figure 3: Showing Intra-abdominal collection.

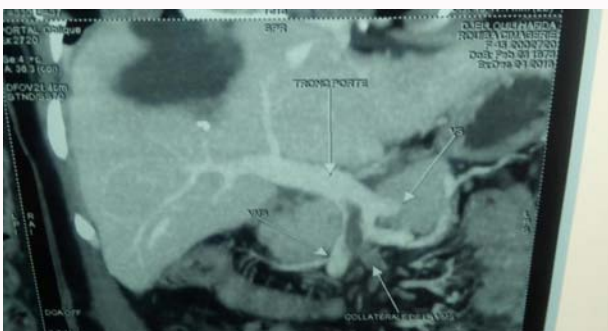


Figure 4: Showing a complication of ARP: venous thrombosis of splenic vein.

physician Al-rahzes who wrote on hydatid cyst of the liver about more than one thousand years ago [2]. The life cycle of the parasite was acknowledged by Dew et al. [3]. It's a parasitic disease caused by the larval form of *Echinococcus granulosus* [4,5]. Primary host of hydatid disease is the dog. Humans become an accidental intermediate host.



Figure 5: MRI shows linear serpiginous shadows in the fluid-filled cyst in the right upper lobe, evoking collapsed membranes. The dilated main left hepatic duct is visible.



Figure 6: ERCP showing intrabiliary hydatid membrane in dilated CBD.

The growth is slow and it may take several years before hydatid cyst become symptomatic. Hydatid cysts may be found in almost any part of the body, but are most often found in the liver (60%) and lungs (30%). Despite the overabundance of publications concerning hydatid cyst, little is known about disease affecting the pancreas. The main causes of acute pancreatitis are gallstones and chronic alcohol abuse, accounting for 75% of all cases [6]. There are rare reports in the international medical literature of acute pancreatitis resulting from intra-biliary rupture of a hydatid cyst [7,8]. There are 16 previous case reports in the international medical literature of acute pancreatitis as a complication of hepatic hydatid disease ruptured into the biliary tree, but only 3 reports of treatment with endoscopic sphincterotomy [9]. We did not find in the literature a similar case (like our) of recurrent acute pancreatitis caused by rupture of a hydatid cyst in the biliary tree. Spontaneous rupture of the hepatic hydatid cyst into the biliary tract occurs in 3.2% to 37% of the cases [1,10], truthful perforation is seen in 3.2% to 17% and small rupture occurs in 10% to 37% [1], while communication between biliary tree and hydatid cyst occurs in 80% to 90% of patients. The physiopathological description of acute pancreatitis due to liver hydatid disease open to the biliary tree is comparable to that of gallstone pancreatitis. Hydrostatic pressure inside the hydatid cyst significantly goes beyond that in bile, and rupture into the bile ducts is frequent [1]. Evident communication can lead to expulsion of hydatid cyst material (membranes, scolices, daughter cysts) into the biliary tree and may cause jaundice and/or recurrent cholangitis. The passage of this material through the papilla of Water can cause transitory obstruction of the pancreatic canal and/or bile reflux into the pancreatic duct and it may produce an acute pancreatitis. A confined allergic reaction to echinococcus antigens inside the ampulla can also be a part in the beginning of pancreatitis [7,11]. The rupture of a hepatic hydatid cyst into the bile

tree may happen clinically, with symptomatology of cholangitis such as abdominal pain, fever and jaundice [12,13]. Serology can be used to approve diagnosis of hydatid cyst, but a negative serologic result does not exclude the diagnosis. The negative serology can occur in about 50% of the patients [14]. In our patients, the diagnosis of acute pancreatitis was confirmed by clinical presentation, raised serum lipase, by abdominal CT scan and by biliary magnetic resonance imaging particularly as our patient had an episode of acute pancreatitis of biliary origin 9 months ago., the hydatid origin is already known since she was operated for hydatid cyst one month ago before developing acute recurrent pancreatitis.

The habitual treatment for hydatid cysts ruptured into the bile ducts is surgery with exploration of the CBD through a choledochotomy, associated with a T. tube insertion, clearance of cyst fragments and surgical removal of the hydatid cyst or cysts, whichever by Enucleation or by pericystectomy and incomplete hepatectomy. Nevertheless, such procedures are associated with high morbidity, mortality and prolonged hospitalization. In a new study which included 20 patients who had been operated for intrabiliary rupture of hydatid cysts, the mortality was 10% (2 deaths in 20 patients), overall morbidity was 60%, with wound infections in 6 patients, suppuration of the residual cavity in 4 patients, and wound dehiscence in two patients, while mean hospitalization was 28.75 days [15].

ERCP has been used early-on in the preoperative diagnosis of complicated hydatid disease [16,17]. A number of reports have been published showing the efficacy and safety of Endoscopic Retrograde Cholangiogram Pancreatography (ERCP) and Endoscopic Sphincterotomy (ES) in the treatment of intrabiliary ruptured hydatid disease [18,19]. Endoscopic sphincterotomy is an effective, safe and well known method in the treatment of hydatid cysts ruptured into bile ducts [20]. Certain author proposes that endoscopic techniques may altogether replace surgery in patients with hydatid disease open to the biliary tract. Al Karawi et al. [21] have reported on a series of 6 patients with intrabiliary rupture of hydatid cysts, which they treated with ES, hydatid material extraction with balloon and basket, while Akkiz et al. [22] reported treating 5 patients in the same way. Moreover, Rodriguez et al. [23] treated 5 elderly patients unfit for surgery with ES and hydatid material extraction, and Giouleme et al. [24] treated two other patients, who remained asymptomatic thereafter. Albendazole is the most potent anthelmintic drug used in the medical treatment in combination with surgical or percutaneous interventions. On the other hand, it may be used as the primary treatment in patients with severe accompanying diseases, patients with very small or asymptomatic cysts, or patients with multiple cysts, or patients with difficult surgical access. In addition, it can be used as a primary treatment in patients who do not accept surgical treatment [25,26]. Our patient underwent ERCP with Endoscopic Sphincterotomy associated with ablation of hydatid cyst material. She passed a smooth period post ES. All laboratory investigations have become within normal values. Medical treatment Albendazol 400 mg 2 × daily she was discharged in good condition and there was no recurrence of acute pancreatitis. After 6 month the patient had no recurrence neither hydatid cyst nor acute pancreatitis.

Conclusion

Acute pancreatitis is an unusual complication of hydatid disease ruptured into the biliary tree. Hydatid membranes in the biliary tract should be well-thought-out as a probable cause of pancreatitis in

patients with liver hydatid cyst. We presented a single case of acute recurrent pancreatitis related to rupture of hydatid cyst into the biliary tract. In endemic areas, physicians should be attentive of this potential cause of acute pancreatitis. Endoscopic sphincterotomy is safe and real in the treatment of acute pancreatitis induced by hydatid disease. It has become one of the therapeutic options of ruptured hepatic hydatid cyst into the biliary tract adopted by most of surgeons for its safety and effectiveness.

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