

## Case report

# Intrabiliary rupture of hepatic hydatid cyst presenting as acute pancreatitis and treatment with endoscopic sphincterotomy: report of two cases

A. Beltsis, G. Chatzimavroudis, A. Iliadis, I. Tsavdaridis, I. Vasiliadis, B. Papaziogas, T. Katsinelos

## SUMMARY

We describe the cases of two young patients, one 16 year – old male and one 21 year – old female, who presented acute pancreatitis as a complication of intrabiliary rupture of hepatic hydatid cysts. Both patients underwent diagnostic and therapeutic endoscopic retrograde cholangiopancreatography with endoscopic sphincterotomy and extraction of hydatid material from the common bile duct by balloon and basket catheters, which resulted in prompt resolution of pancreatitis. We were able to discharge both patients five days after the procedure. They were prescribed three cycles of albendazole and have since been free of symptoms for two and one years, respectively. 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. This case study suggests that physicians should be aware of this entity in endemic areas, and it indicates that endoscopic sphincterotomy is safe and effective in the treatment of acute pancreatitis induced by intrabiliary rupture of hepatic hydatid disease. However, surgical resection remains the generally accepted method of radical treatment for hepatic hydatid disease.

**Key words:** Hepatic hydatid disease, endoscopic sphincterotomy, acute pancreatitis.

## INTRODUCTION

Hydatid disease is caused by the larval stage of infection by the tapeworm *Echinococcus Granulosus*, which lives in dogs. Humans, as well as sheep and cattle, are intermediate hosts.<sup>1</sup> The disease is endemic in sheep raising areas, including South Australia, New Zealand, Africa, South America, Southern Europe, Middle East and Far East.<sup>1</sup> The liver is the most frequent site of involvement and is infested in 50 – 70% of patients. Rupture of a hydatid cyst into the biliary tree is the most common complication, occurring in 5 – 25% of patients.<sup>2-4</sup> Pancreatitis, however, has only been associated with the presence of hydatid material inside the biliary tree in a few case reports.<sup>5-20</sup>

We describe the cases of two young patients from Northern Greece, who presented with acute pancreatitis caused by intrabiliary rupture of a hydatid cyst, and who were successfully managed with endoscopic sphincterotomy.

### Case 1

A 16-year-old male was referred to our department two years ago with epigastric pain radiating to the back, nausea and vomiting for a duration of 8 hours. There was no history of alcohol or drug abuse and no recent abdominal trauma. Family history was unremarkable. Physical examination revealed mild jaundice and epigastric tenderness.

Laboratory biochemical data showed: alanine aminotransferase, 165 U/L (normal < 40 U/L); aspartate aminotransferase, 293 U/L (normal < 40 U/L); alkaline phosphatase, 423 U/L (normal 60-120U/L);  $\gamma$ -glutamyl-transpeptidase, 320 U/L (normal < 20U/L); total bilirubin, 6.75 mg/dl (direct: 5.93 mg/dl); and amylase, 865 U/L

Department of Endoscopy and Motility Unit, Central Hospital, Thessaloniki, Greece.

#### Author for correspondence:

Panagiotis Katsinelos, MD, PhD, Head,  
Department of Endoscopy, and Motility Unit, Central Hospital,  
Ethnikis Aminis 41, TK 54635, Thessaloniki, Greece,  
Fax: +302310210401, e-mail: akis\_katsinelos@yahoo.gr

(normal 60 – 100 U/L). Complete blood count revealed leucocytosis (white blood cells, 13300 /mm<sup>3</sup>; neutrophils, 75%; lymphocytes, 20%; monocytes, 3%; eosinophils, 2%) with normal platelet count and hematocrit.

Emergency magnetic resonance cholangiography (MRCP) revealed an edematous pancreas and a dilated common bile duct (CBD), with multiple filling defects within, one of which was embedded into the ampulla of Vater. MRCP also showed a hydatid cyst measuring 6 cm in diameter in segment VI of the liver. Serology was positive for *E. Granulosus* antibodies.

After obtaining a signed informed consent, we performed endoscopic retrograde cholangiopancreatography (ERCP), which confirmed the findings of the MRCP, and further revealed a communication between the biliary tract and the hydatid cyst, while hydatid membranes were seen protruding through the papillary orifice. Endoscopic sphincterotomy (ES) was performed, and hydatid membranes were extracted from the CBD with the use of a balloon catheter and a Dormia basket.

The post-procedure course was uneventful, laboratory values returned to normal within 3 days of sphincterotomy, and the patient was discharged from the hospital in 5 days. Three 28-day cycles of albendazole were prescribed, 400 mg twice daily with 14-day drug – free intervals. Two years later, the patient is still without symptoms.

## Case 2

A 21-year-old female patient was admitted to our department one year ago for moderate pain in the epigastrium and right upper abdominal quadrant radiating to the back, jaundice, chills and fever (38.5° C), for a duration of 12 hours. She also presented nausea and vomiting. She did not have a history of alcohol or drug abuse, was not taking any medication at the time, and there was no history of abdominal trauma. Family history was unremarkable.

Laboratory biochemical data showed: alanine aminotransferase, 174 U/L (normal < 40 U/L); aspartate aminotransferase, 256 U/L (normal < 40 U/L); alkaline phosphatase, 534 U/L (normal 60 – 120 U/L);  $\gamma$ -glutamyl-transpeptidase, 331 U/L (normal <20 U/L); total bilirubin 7.45 mg/dl (direct: 6.21 mg/dl); and amylase, 1071 U/L (normal 60 – 100 U/L). Complete blood count revealed leucocytosis (white blood cells 14200 / mm<sup>3</sup>; neutrophils, 77%, lymphocytes, 15%; monocytes, 4%; eosinophils, 4%), platelet count was 435000 / mm<sup>3</sup> and the hematocrit was normal.

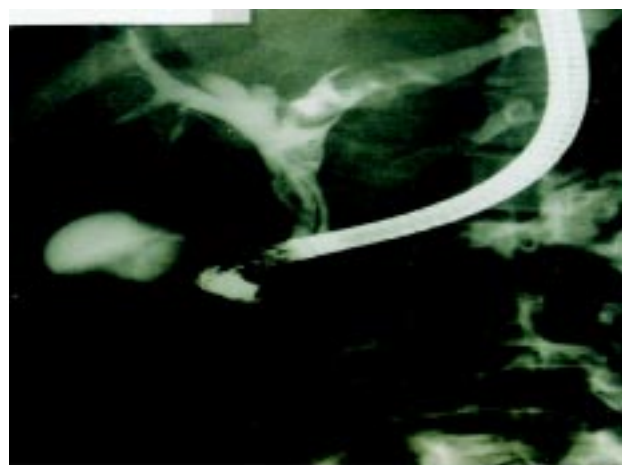
Abdominal CT scan showed a cystic lesion in the left hepatic lobe, with a thickened wall and multiple thickened septae within, and, in addition, a dilation of the intrahepatic bile ducts. After obtaining a signed informed consent, we performed ERCP, which revealed a dilated intra- and extra- hepatic biliary tree with filling defects of varying size and shape (Fig. 1). After ES was performed, hydatid membranes were extracted from the CBD with the use of a balloon catheter and a Dormia basket.

The post-procedure course was uneventful, laboratory values returned to normal within 4 days of sphincterotomy, and the patient was discharged from the hospital in 5 days. Three 28-day cycles of albendazole were prescribed, 400 mg twice daily with 14 -day drug-free intervals. One year later, the patient is still without symptoms.

## DISCUSSION

The main causes of acute pancreatitis are gallstones and chronic alcohol abuse, accounting for 75% of all cases.<sup>1</sup> There are rare reports in the international medical literature of acute pancreatitis resulting from intrabiliary rupture of a hydatid cyst.<sup>5-20</sup> However, some investigators advocate that acute pancreatitis due to hydatid disease, in endemic areas, is not as rare as reported.<sup>8</sup> In a series of 221 Algerian patients operated upon for acute pancreatitis, 4 had hydatid disease as the initiating factor for their pancreatitis.<sup>21</sup>

The pathogenesis of acute pancreatitis due to hydatid



**Figure 1.** A cholangiogram revealing a dilated 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.

disease of the liver open to the biliary tree is similar to that of gallstone pancreatitis. Hydrostatic pressure inside the hydatid cyst greatly exceeds that in bile, and rupture into the bile ducts is frequent.<sup>1</sup> Overt communication can lead to expulsion of hydatid cyst material (membranes, scolices, daughter cysts) into the biliary tree and may cause cholestatic jaundice and recurrent cholangitis. The passage of this material through the papilla of Vater can cause transient occlusion of the pancreatic duct and/or bile reflux into the pancreatic duct and it may initiate acute pancreatitis. A local allergic reaction to echinococcus antigens inside the ampulla may also play a part in the initiation of pancreatitis.<sup>5,15</sup>

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 pericystectomy and partial hepatectomy. However, such operations are associated with morbidity, mortality and prolonged hospitalization. In a recent report that included 20 patients who had been operated upon for intrabiliary rupture of hydatid cysts, mortality was 10% (2 deaths in 20 patients), overall morbidity was 60%, with wound infections developing in 6 patients, suppuration of the residual cavity in 4 patients, and wound dehiscence in 2 patients, while mean hospitalization was 28.75 days.<sup>22</sup>

ERCP has been employed early-on in the preoperative diagnosis of complicated hydatid disease.<sup>23,24</sup> A number of reports have been published showing the efficacy and safety of ERCP and endoscopic sphincterotomy (ES) in the treatment of intrabiliary ruptured hydatid disease.<sup>25-30</sup> Some authors suggest that endoscopic techniques may altogether replace surgery in patients with hydatid disease open to the biliary tract. Al Karawi et al<sup>28</sup> have reported on a series of 6 patients with intrabiliary rupture of hydatid cysts, whom they treated with ES, hydatid material extraction with balloon and basket catheters and hypertonic saline lavage through a nasobiliary drain, while Akkiz et al<sup>27</sup> reported treating 5 patients in the same way. Furthermore, Rodriguez et al<sup>26</sup> treated 5 elderly patients unfit for surgery with ES and hydatid material extraction, and Giouleme et al<sup>24</sup> treated two other patients, who remained asymptomatic thereafter.

In the specific setting of acute pancreatitis due to hydatid disease ruptured into the biliary tree, there are only 3 previous case reports of treatment with ES.<sup>6,8,11</sup> In our patients, acute pancreatitis was documented by clinical presentation, raised serum amylase and, in one of the patients, by magnetic resonance imaging of the pan-

creas. Hydatid disease was strongly suspected by the typical imaging studies, and was confirmed by serology and, eventually, by the extraction of hydatid material from the common bile duct at ERCP. ES with extraction of hydatid material was able to decongest the CBD, relieve jaundice, and induce remission of acute pancreatitis. It should be pointed out that our patients avoided painful operations and lengthy hospital stay.

In conclusion, acute pancreatitis is a rare complication of hydatid disease ruptured into the biliary tree. In endemic areas, physicians should be aware of this possible cause of acute pancreatitis. Endoscopic sphincterotomy is safe and effective in the management of hydatid disease induced acute pancreatitis. However, surgical resection remains the generally accepted method of radical treatment for hepatic hydatid disease.

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