Hydatid cyst of the pancreas causing both acute pancreatitis and splenic vein thrombosis

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ABSTRACT

Hydatid cyst of the pancreas is a rarely seen entity even in endemic countries. Cyst may causes several symptoms due to external compression or fistulisation to pancreaticobiliary tract or small bowel. A 23-year-old female patient was referred with a complaint of abdominal pain. Preoperative imaging revealed an undefined cyst in the tail of pancreas. She underwent distal pancreatectomy and splenectomy, with a diagnosis of acute pancreatitis due to cystopancreatic duct fistula and also left-sided portal hypertension due to splenic vein thrombosis. Pathological examination reported a final diagnosis of hydatid cyst. To the best of our knowledge, coincidence of cystopancreatic duct fistula and splenic vein thrombosis due to pancreatic hydatid cyst has not previously been reported.

KEYWORDS

Acute pancreatitis – Echinococcosis – Hydatid cyst – Portal hypertension – Splenic vein thrombosis

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Introduction

Hydatid cyst disease is still an endemic public health problem in some regions, especially in South Africa, the Middle East and Southern Europe. Hydatid cysts, caused by the larval stage of *Echinococcus granulosus*, mostly occurs in the liver and lungs but rarely in any organ or tissue. In the literature, the incidence of isolated pancreatic hydatid disease (of which 50% of cases are located in the head of the pancreas) is less than 2%, even in endemic areas.^{1,2} Most of infected patients stay asymptomatic for many years owing to the slow growth pattern of the hydatid cyst. Cyst size, location and relationship with adjacent organs play a role in the occurrence of different symptoms.² We present the diagnosis and treatment of a patient with a pancreatic hydatid cyst causing both splenic vein thrombosis and acute pancreatitis.

Case history

A 23-year-old female patient was admitted to the emergency service with epigastric pain, intermittent for the past 3 months and increased for the past 2 days. She had no history of abdominal operation, chronic disease, alcohol consumption or medication. On physical examination, tenderness was found in the epigastric region. Investigations revealed a leucocyte count of 14550/mm³ and haemoglobin level of 11.5 g/dl, levels of amylase and lipase of 794 iu/l

and 910 iu/l, respectively, above the normal range. Levels of liver enzymes and tumour markers such as carcinoembryonic antigen and cancer antigen 19-9 were normal. Abdominal and chest radiographs showed no significant findings. Abdominal ultrasound was suspicious for a cystic mass on the pancreas and no gallbladder pathology was detected. Contrast-enhanced abdominal computed tomography (CT) detected mild oedematous pancreatitis with a dilated main pancreatic duct and a hypodense cystic mass on the tail of the pancreas with a diameter of 45 mm causing left-sided portal hypertension due to external compression-related splenic vein thrombosis (Fig 1). The patient was admitted and intravenous fluid and anticoagulant therapy were administered. Magnetic resonance imaging (MRI) was employed to evaluate the malignant potential of mass, and revealed a splenic vein thrombosis and a 44 × 36 mm cystic mass in the tail of the pancreas (Fig 2). A decision was made to operate.

During surgical exploration, a mass of approximately 5 × 4 cm in the tail of the pancreas and mild splenomegaly due to splenic vein compression were detected. En bloc resection of distal pancreas and spleen was performed (Fig 3). The main pancreatic duct in the resection margin was sutured and fibrin sealant Tisseel (Baxter Healthcare) was used to reduce the risk of a pancreatic leak. A drain was placed and the abdomen layers were closed. The postoperative period was uneventful and the patient was discharged on postoperative day three.

Figure 1 Axial computed tomography images. Hydatid cyst in the tail of the pancreas and splenic vein compression of the cyst (arrow), (a) cystopancreatic connection (yellow arrow) and dilated main pancreatic duct (white arrow) are seen (b), cystopancreatic connection (arrow) in the beginning of the fistulation region (c)

Figure 2 Magnetic resonance image (coronal view) of the hydatid cyst containing daughter vesicles (arrow)

Figure 3 The hydatid cyst in the tail of the pancreas and the congested spleen after resection

The patient was given pneumococcal, meningococcal and haemophilus influenzae vaccinations at the time of first outpatient visit, which was 2 weeks after the operation.

Pathological examination revealed a congested spleen and a hydatid cyst with a thick fibrous wall, cuticular membranes, scolices and cystopancreatic duct fistula. After the final diagnosis of hydatid cyst, albendazole treatment was administered for 3 months at a dose of 800 mg/day for a cystopancreatic duct fistula. The patient remained well, with no sign of recurrence or complications in the six months' follow-up period. Her levels of blood glucose remained within normal limits during follow-up. Written informed consent was obtained from the patient who participated in this study.

Discussion

Hydatid cyst of the pancreas is a rare entity even in endemic countries. Affected patients may remain asymptomatic or may present with complaints of abdominal pain, vomiting, discomfort or weight loss.³ Fistulisation into the pancreatobiliary tract or external compression of that tract may cause severe conditions including jaundice, cholangitis or acute pancreatitis.⁴ Splenic vein compression of the hydatid cyst, which is located in the distal pancreas, may lead to left-sided portal hypertension due to direct compression or indirectly via splenic vein thrombosis.^{2,5} The patient we present had a diagnosis of acute pancreatitis due to a cystopancreatic duct fistula and left-sided portal hypertension due to external compression-related splenic vein thrombosis. To the best of our knowledge, coincidence of these conditions has not previously been reported.

Preoperative diagnosis is challenging owing to the rarity of disease. The differential diagnoses include a group of

cystic diseases including pseudocyst, serous cystadenoma and intraductal papillary mucinous neoplasm.⁵ Diagnostic imaging tools include ultrasound, CT and MRI. Invasive methods such as endoscopic ultrasound and endoscopic retrograde cholangiopancreatography can be used to biopsy or aspirate cyst contents, for biochemical and cytological sampling or to place a stent in terms of obstruction. The malignant potential of the cyst in our patient was suspicious for intraductal papillary mucinous neoplasm owing to the presence of a cystopancreatic duct fistula.

Surgery is considered to be the best treatment option.^{3–5} Several surgical methods have been reported, depending on location of the hydatid cyst. Spleen-preserving distal pancreatectomy may be a good choice for a hydatid cyst in the tail in the absence of splenic vascular involvement.² Cystogastric or cystoenteral anastomosis are also recommended as alternative techniques in cases with cystopancreatic duct fistula, to prevent postoperative pancreatic leak.² Our patient underwent distal pancreatectomy and splenectomy two days after hospitalisation following the CT findings. We used fibrin sealant after suturing the resection margin of main pancreatic duct to avoid fistula, and no fistula was detected in the follow-up period.

Conclusion

Primary hydatid cyst of pancreas is a rare entity, even in endemic regions. Preoperative differential diagnosis from

malignant or premalignant cystic diseases of pancreas is hard. CT and MRI should be used to evaluate the malignant potential of the cyst and the presence of any connection with main pancreatic duct. In the case of a cystopancreatic connection and splenic vein thrombosis, distal pancreatectomy and splenectomy is a reliable surgical procedure. In addition, fibrin sealant may be useful to avoid postoperative pancreatic fistula.

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