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Chapter

Primary Cystic Echinococcosis of the Pancreas

Azize Saroglu and Alexander Julianov

Abstract

Primary cystic echinococcosis of the pancreas is rare, even in endemic regions. The clinical presentation depends on the affected part of the pancreas and the presence of local complications, but there are no specific symptoms, which can be a clue to diagnosis. Imaging studies usually reveal avascular lesions in the pancreas that are most frequently misdiagnosed as pseudocysts or neoplastic cystic tumors. The treatment options vary from evacuation and drainage to formal resection of the pancreas, with no evidence of the best treatment strategy. This chapter provides a comprehensive review of the current knowledge of the clinical presentation, diagnosis, and treatment of primary cystic echinococcosis of the pancreas. Acute thrombosis of the splenic artery which leads to massive splenic infarction and abscess, a previously unreported initial manifestation of cystic echinococcosis of the pancreas is also presented, as well as the first use of intraoperative pancreaticoscopy to clear the main pancreatic duct from membranes of the parasite.

Keywords: cystic echinococcosis, pancreas, diagnosis, treatment, pancreaticoscopy

1. Introduction

Cystic echinococcosis is a zoonotic disease caused by the larval stage of the *Echinococcus granulosus* parasite, representing an endemic problem in many regions of the world such as the Mediterranean countries, Australia, New Zealand, South America, South East and Far East Asia, and Middle Eastern countries [1]. The parasite was named in 1801 by Rudolphi who wrote: “*Echinococcus*, that’s what I call the granular bladderworms...” (**Figure 1**) [2].

The prevalence of isolated pancreatic cystic echinococcosis (PCE) is very low, ranging from 0.14 to 2% of total systemic echinococcosis [3–5]. Pancreatic cystic echinococcosis (PCE) may develop as a primary isolated disease involving the pancreas only, or as a secondary disease with multiple organ involvement (**Figure 2**), and can masquerade as more common lesions of the pancreas such as pseudocysts or cystic pancreatic neoplasms.

The lack of specific clinical manifestations of PCE is clearly demonstrated by the majority of cases published in the literature, thus explaining why preoperative diagnosis is challenging [5–8]. In symptomatic cases, clinical findings may be similar to those of other pancreatic diseases. However, the imaging features of PCE are almost

Echinococcus, so nenne ich die körnigen Blasenwürmer. Goeze *) unterschied die geselligen Blasenwürmer in solche, wo viele Würmer auf einer gemeinschaftlichen Blase sitzen, ohne eine weitere Außenblase oder Decke zu haben, und in solche, wo viele Würmer in einer gemeinschaftlichen Blase befindlich sind, die noch eine kaltsche Außenblase haben, jene nannte er *Taenia vesicularis*; *cerebrina*; *multiceps*, diese aber *Taenia visceralis socialis granulosa*. Zeder **) macht hietaus

Figure 1.
The original text of Rudolphi introduces the name "Echinococcus".

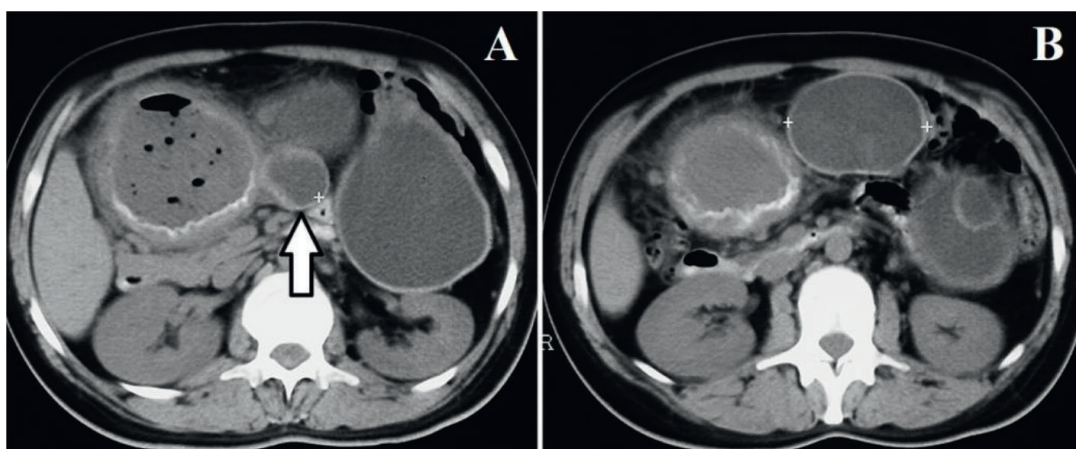


Figure 2.
(A, B) axial computed tomography scans of a patient with abdominal cystic echinococcosis with multiple locations including body of the pancreas (arrow, a).

identical to those of other cystic diseases of the pancreas, and given the rarity of the problem, isolated primary pancreatic cystic echinococcosis is often misdiagnosed even in endemic areas of the world.

2. Clinical presentation

The clinical presentation of PCE is a result of: 1) the pressure generated by the cyst on the pancreatic tissue and adjacent structures, which depends on the size and anatomic location of the cyst; 2) the local inflammatory reaction to the parasite involving neighboring anatomical structures, and; 3) the presence of local/systemic complications.

According to data from the literature, 50–58% of pancreatic echinococcosis is found in the pancreatic head, 24–34% in the pancreatic body, and 16–19% in the pancreatic tail [8–10]. It is considered that the embryos of hydatid cysts end up in the

pancreas mainly by hematogenous dissemination [11]. The rich vascular network on the head of the pancreas explains the more frequent involvement of this part of the gland by the parasite. Other possible mechanisms for the involvement of the pancreas include local spread by passage of cystic elements via the bile duct into the pancreatic duct, direct passage of cystic components through the intestinal mucosa into the peripancreatic lymphatic plexus, and retroperitoneal spread [11–13].

Pancreatic echinococcosis is considered to be asymptomatic for a long period, due to its slow growth rate of 0.3–2.0 cm per year [14]. All data for the clinical symptoms of cystic echinococcosis of the pancreas come from a small published series or case reports in the literature. An abdominal mass, epigastric pain, weight loss, discomfort, and vomiting are the main nonspecific clinical symptoms [5, 9, 10, 14–22]. PCE located in the pancreatic head most commonly causes cholangitis, obstructive jaundice, or acute pancreatitis [3, 12, 13, 16, 21–28]. Cysts of the parasite located in the body or tail of the pancreas can be asymptomatic and usually present as an abdominal lump when they enlarge [3, 19, 22, 29–31]. Infrequently, PCE located in the pancreatic tail can result in splenomegaly and segmental portal hypertension owing to splenic vein compression/thrombosis [32]. Other reported uncommon complications include mesenteric/portal vein thrombosis, upper gastrointestinal bleeding with splenic artery pseudoaneurysm, intracystic bleeding, and rupture into the biliary system or peritoneal cavity, pancreatic fistula, recurrent pancreatitis, and pancreatic abscess [6, 7, 32, 33].

As primary PCE can mimic any other pancreatic disease, it is still frequently a clinical surprise rather than a preoperatively established diagnosis. We observed a previously unreported initial manifestation of PCE in a patient who presents with splenic infarction and abscess secondary to acute splenic artery thrombosis due to isolated cystic echinococcosis located in the body of the pancreas (**Figure 3**). The patient was erroneously diagnosed preoperatively as having a complicated malignant pancreatic tumor with splenic artery involvement, and a correct diagnosis was made during laparotomy.

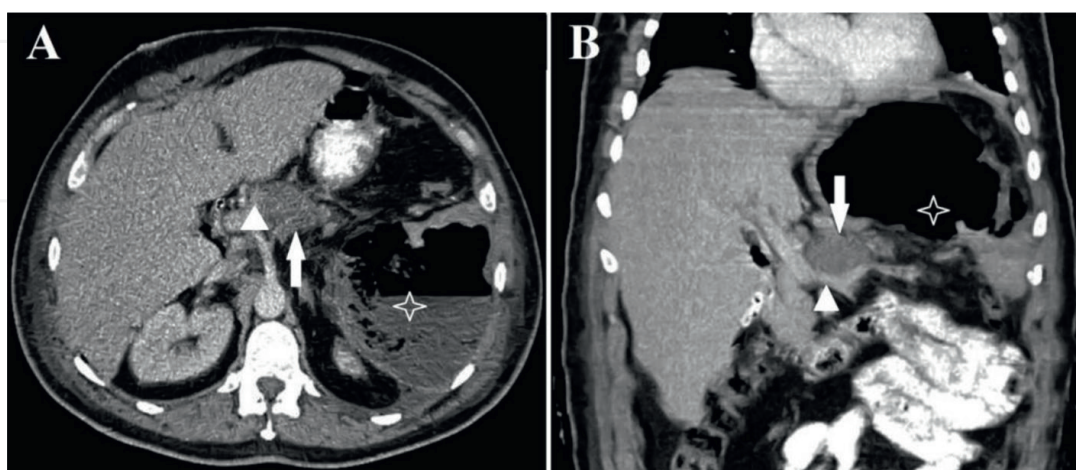


Figure 3. Computed tomography in a patient with primary PCE. (A) Axial image demonstrating pancreatic cystic lesion (arrow), thrombosis of splenic artery at its origin (arrowhead), and splenic abscess (asterisk). (B) Coronal image demonstrating patent splenic vein (arrowhead) along with the pancreatic cyst (arrow) and splenic abscess (asterisk).

3. Diagnosis

The first and probably most important step in the diagnosis of PCE is clinical suspicion, particularly in endemic regions. The diagnosis is much easier when associated with other localization(s) (**Figure 4**).

Characteristic imaging findings of echinococcosis are often missing in isolated PCE, with considerable imaging overlap between other cystic lesions of the pancreas and peripancreatic regions (**Figure 5**), such as pseudocysts, choledochal cysts, serous or mucinous cystadenomas, and cystadenocarcinomas, which complicates the diagnostic process. The higher prevalence of mucinous cystadenomas of the pancreas and, on the other hand, the rare occurrence of pancreatic echinococcosis leads to the fact that it is rarely taken into account in the differential diagnosis [34, 35], and pancreatic echinococcosis is, as a result, often misdiagnosed [20, 36–38]. According to data from the literature, the vast majority of PCE patients are not diagnosed preoperatively [7, 37].

In daily clinical practice, blood tests for detecting specific serum antibodies and circulating echinococcal antigens usually include indirect hemagglutination assay, immunoelectrophoresis, enzyme-linked immunosorbent assay, complement fixation test, and immunofluorescence assay [3, 7, 22]. According to a systematic review by Dziri et al. [3], hydatid serology has a low sensitivity (62%). In another literature review by Akbulut et al. [7], the sensitivity of hydatid serology was even lower (54%).

The most commonly performed imaging modalities for the diagnosis of pancreatic cysts are transabdominal ultrasound (US), contrast-enhanced computed tomography (CT), and magnetic resonance imaging (MRI). Cases that require further workup are examined using invasive diagnostic tools such as endoscopic retrograde cholangiopancreatography (ERCP) and endoscopic ultrasound (EUS); the latter has emerged rapidly as an effective technique to gain diagnostic information and access to retroperitoneal organs such as the pancreas.

Transabdominal ultrasound is a widespread, cost-effective, and sensitive method for detecting internal cyst structures, including membranes, septa, hydatid sand, and daughter cysts. It can show a well-defined anechoic lesion with a hyperechoic thick double-lined wall and internal echogenic material, although the sensitivity is decreased in cases of PCE due to the retroperitoneal location and superposing bowel gas [37, 39].

Localization and size of the pancreatic cyst can be detected accurately on CT, that moreover, may provide information about the relationship between the cyst and bile

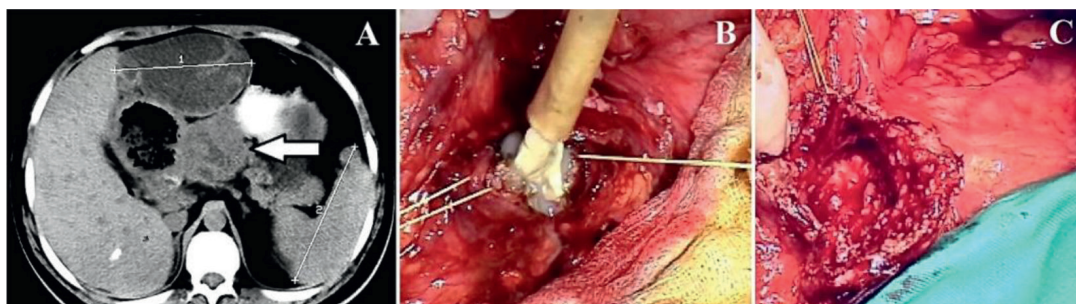


Figure 4. Abdominal cystic echinococcosis. (A) Axial computed tomography demonstrating cystic lesion in the head of the pancreas (arrow) and ventrally located cyst in the left liver with typical appearance of cystic echinococcosis. (B, C) Intraoperative photograph shows removal of the parasite and remaining defect in pancreatic head.

Non-neoplastic lesions	Neoplastic lesions
<ul style="list-style-type: none"> • Pseudocyst • Syndromes causing multiple cysts • Infectious/parasitic cysts • Lympho-epithelial cysts • Duplication cysts • Retention cysts 	<ul style="list-style-type: none"> • IPMN • MCN • SCN • SPN • Cystic variants of solid tumors: <ul style="list-style-type: none"> <i>teratoma</i> <i>ductal adenocarcinoma</i> <i>neuroendocrine tumor</i> <i>acinar cell carcinoma</i> <i>metastasis</i>

Figure 5.
 Differential diagnosis of pancreatic cystic lesions.

ducts and wall calcification, and CT angiography can show significant displacement of vascular compression in the arterial and venous phases [7, 8, 19–22, 32–34, 37].

In the PCE case series of Li et al. [22], MRI provided a better depiction of the fluid content of cystic lesions and communication with the pancreatic duct due to its higher soft-tissue contrast and capability of multiplanar imaging. Pancreatic cystic echinococcosis showed a hypointense signal on T1-weighted MRI images, a hyperintense signal on T2-weighted images, and a hyperintense signal.

EUS is another diagnostic tool that is commonly used in the evaluation of cystic pancreatic lesions. Nowadays some authors recommended a fine-needle aspiration biopsy to differentiate a hydatid cyst from other common cystic lesions [40, 41].

According to data from the literature, accurate diagnosis cannot be established based on radiological findings alone, and this is especially true for pancreatic cystic lesions demonstrating particular imaging features such as multilocular cysts, presence of internal septations, calcifications, and wall enhancement as they are encountered in both benign and malignant pancreatic cystic lesions [22, 36]. Blood tests may be helpful for diagnosis in some cases, but according to the published experience with PCE, the sensitivity of the routinely used tests is still low [3, 7].

4. Surgical treatment

Surgery remains the main treatment option for patients with PCE as most cases are correctly diagnosed intraoperatively. However, due to a lack of evidence, it is not clear which is the best treatment strategy for patients with isolated PCE. However, Dziri et al. in their review reported that surgery is the main treatment of PCE and the open approach is performed in 95% of the cases [3]. Furthermore, depending on the cyst's

location, several procedures have been suggested, including cyst fenestration, internal derivation, central or distal pancreatectomy with or without splenectomy [3, 6, 7], and presently, available treatment options include formal resection, internal capsule stripping and external capsule removal (subadventitial total exocystectomy) [42].

Regarding the available surgical options to treat PCE, it is clear that formal pancreatic resection is not necessary to treat such benign diseases and should be avoided when possible to spare the patient from complications associated with resection of the pancreas. Superficially located cysts that do not communicate with the pancreatic ductal system can be excised, opened, or drained without substantial complications. On the other hand, the main question to the surgeon after parasite removal (**Figure 6**) is whether the remaining cavity in the pancreas communicates with the ductal system. In cases with ductal communication, the surgical option is to perform a drainage procedure on a Roux-en-Y jejunal limb or to proceed with formal resection. However, the latter is justified only in cases where resection of the body/tail may be sufficient, and formal pancreaticoduodenectomy does not seem justified to treat PCE.

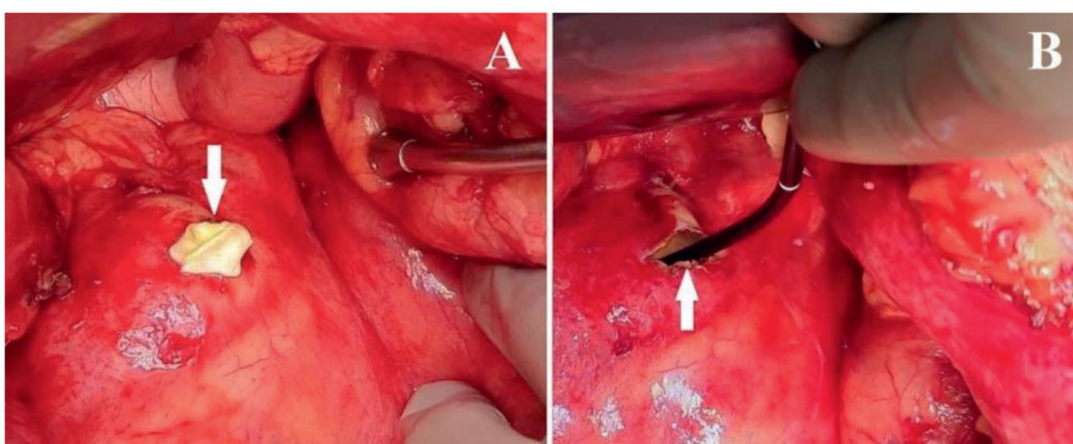


Figure 6. Intraoperative photographs of a patient corresponding to **Figure 3**. (A) Protrusion of parasitic membrane (arrow) through the cystotomy. (B) Pancreaticoscope (arrow) is inserted in the main pancreatic duct toward the head of the pancreas.

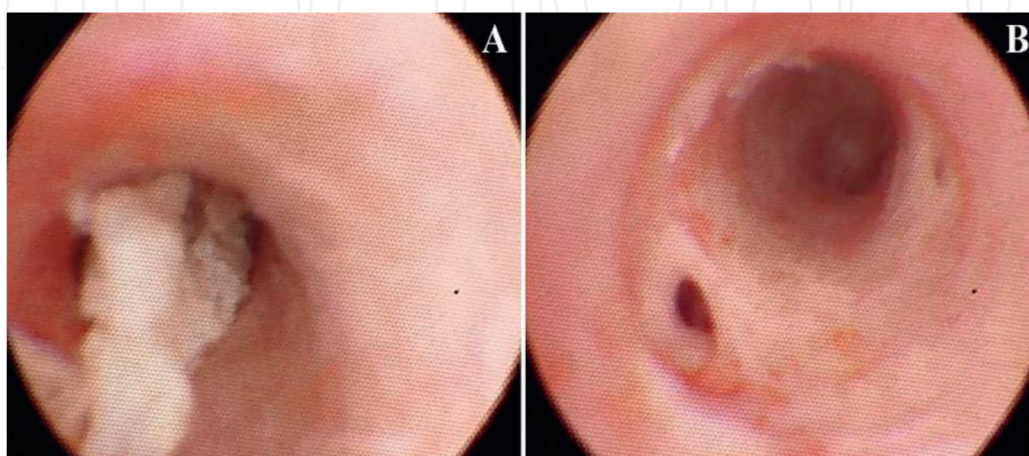


Figure 7. Intraoperative pancreaticoscopy in a patient corresponding to **Figure 5**. (A) Hydatid membrane impacted the main pancreatic duct. (B) the pancreatic duct is cleared from the parts of the parasite.

Intraoperative management of the main pancreatic duct has not yet been resolved. We consider this a topic of concern as parts of the parasite may migrate to the main pancreatic duct in the case of ductal communication of the PCE, causing ductal obstruction, further symptoms, and even disease recurrence. To resolve these problems, the use of intraoperative pancreaticoscopy seems to be a good solution, similar to its use in surgery for chronic pancreatitis [43].

Direct endoscopic inspection of the main pancreatic duct through communication with the residual cavity is simple, securely detects intraductal pathology, and can be easily used to clear the duct (**Figure 7**). Any available flexible endoscope that fits the pancreatic duct can be used for this purpose. Other options to evaluate the involvement of the pancreatic ductal system include intraoperative ultrasonography of the pancreas and intraoperative pancreatography. However, both of them cannot manage the duct in cases of involvement, and the diagnostic accuracy of pancreaticoscopy is unmatched.

5. Conclusion

Primary cystic echinococcosis of the pancreas is rare even in endemic areas. Despite radiological imaging, including transabdominal ultrasound, CT, MRI, EUS, ERCP, laboratory tests, and hydatid serology, the preoperative diagnosis of pancreatic cystic echinococcosis remains difficult, and the correct diagnosis is most often intraoperative.

Clinicians may still encounter undescribed clinical presentations of pancreatic echinococcosis, as presented in this chapter. PCE should be considered in the differential diagnosis of pancreatic cystic lesions, particularly in endemic geographic regions. Surgical treatment combined with albendazole can reduce the recurrence rate and morbidity of pancreatic cystic echinococcosis. As the intraoperative management of the main pancreatic duct in the case of communication with the parasite is not resolved, the use of intraoperative pancreaticoscopy may play an important role as a valuable adjunct to the operative strategy and contribute to more precise surgery.

Primary cystic echinococcosis of the pancreas remains a diagnostic challenge, and it seems that only high clinical suspicion and awareness of the disease can improve the preoperative diagnosis rate.

Conflict of interest

The authors declare no conflict of interest.

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