Hydatid cyst of the pancreas: Report of an undiagnosed case of pancreatic hydatid cyst and brief literature review

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RESULTS: A total of 58 patients, including our one case, (age range: 4 to 70 years, mean ± SD: 31.4 ± 15.9 years) were included in the analysis. Twenty-nine of the patients were female, and 29 were male. The information about cyst location was available from studies involving 54 patients and indicated the following distribution of locations: pancreatic head (n = 21), pancreatic tail (n = 18), pancreatic body and tail (n = 8), pancreatic body (n = 5), pancreatic head and body (n = 1), and pancreatic neck (n = 1). Extra-pancreatic locations of hydatid cysts were reported in the studies involving 44 of the patients. Among these, no other focus than pancreas was detected in 32 of the patients (isolated cases) while 12 of the patients had hydatid cysts in extra-pancreatic sites (liver: n = 6, liver + spleen + peritoneum: n = 2, kidney: n = 1, liver + kidney: n = 1, kidney + peritoneum: n = 1 and liver + lung: n = 1). Serological information was available in the studies involving 40 patients, and 21 of those patients were serologically positive and 15 were serologically negative; the remaining 4 patients underwent no serological testing. Information about pancreatic cyst size was available in the studies involving 42 patients; the smallest cyst diameter reported was 26 mm and the largest cyst diameter reported was 180 mm (mean ± SD: 71.3 ± 36.1 mm). Complications were available in the studies of 16 patients and showed the following distribution: cystobiliary fistula (n = 4), cysto-pancreatic fistula (n = 4), pancreatitis (n = 6), and portal hypertension (n = 2). Postoperative follow-up data were available in the studies involving 48 patients and postoperative recurrence data in the studies of 51 patients; no cases of recurrence occurred in any.

AIM: To overview the literature on pancreatic hydatid cyst (PHC) disease, a disease frequently misdiagnosed during preoperative radiologic investigation.

METHODS: PubMed, Medline, Google Scholar, and Google databases were searched to identify articles related to PHC using the following keywords: hydatid cyst, hydatid disease, unusual location of hydatid cyst, hydatid cyst and pancreas, pancreatic hydatid cyst, and pancreatic echinococcosis. The search included letters to the editor, case reports, review articles, original articles, meeting presentations and abstracts that had been published between January 2010 and April 2014 without any restrictions on language, journal, or country. All articles identified and retrieved which contained adequate information on the study population (including patient age and sex) and disease and treatment related data (such as cyst size, cyst location, and clinical management) were included in the study; articles with insufficient demographic and clinical data were excluded.

In addition, we evaluated a case of a 48-year-old female patient with PHC who was treated in our clinic.
patient for an average follow-up duration of 22.5 ± 23.1 (range: 2-120) mo. Only two cases were reported as having died on fourth (our new case) and fifteenth days respectively.

CONCLUSION: PHC is a parasitic infestation that is rare but can cause serious pancreato-biliary complications. Its preoperative diagnosis is challenging, as its radiologic findings are often mistaken for other cystic lesions of the pancreas.

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Key words: Echinococcosis; Hydatid cyst; Pancreas; Pancreatocoduodenectomy

Core tip: Hydatid disease is a zoonotic disease caused by the Echinococcus parasite, which belongs to the Taeniidae family of the Cestode class. Although hydatid cysts can be found in almost any tissue or organ of the human body, the liver, lung, spleen, and kidney are the most commonly affected. Pancreatic hydatid cyst (PHC) disease is rare, even in regions where hydatidosis is endemic. Yet, PHC disease is associated with severe complications, such as jaundice, cholangitis, and pancreatitis. These complications often develop as a result of fistulization of the cyst content into pancreato-biliary ducts or external compression of those ducts by the cyst.

INTRODUCTION

Hydatid disease, also known as echinococcal disease, is a zoonotic disease caused by the Echinococcus parasite belonging to the Taeniidae family of the Cestode class. Four different Echinococcus species have been defined as causative agents of hydatid disease in humans. The most common species encountered in humans are the Echinococcus granulosus (E. granulosus), which causes cystic echinococcosis, and the Echinococcus multilocularis, which causes alveolar echinococcosis. E. granulosus is responsible for 95% of the human hydatid cases reported. In the biological life cycle of hydatid disease, carnivores are the definitive hosts while herbivores are the intermediary hosts. Humans themselves have no role in the biological life cycle and are usually infected after inadvertent ingestion of Echinococcus eggs in canine feces. The disease continues to be a major public health issue in many regions of the world where agriculture and stockbreeding are primary sources of income. Although hydatid cysts can localize to almost any tissue or organ of the human body, the liver (50%-77%), lung (15%-47%), spleen (0.5%-8%), and kidney (2%-4%) are the most commonly involved organs.

Pancreatic hydatid cyst (PHC) disease is rare, even in regions where hydatidosis is endemic. While the reported incidences of PHC have varied in different studies, the rates are consistently below 1%. PHC may develop as a primary (involving the pancreas only) or secondary (with multiple organ involvement) disease. Since hydatid cysts grow slowly, a considerable portion of affected patients may remain asymptomatic for years. In symptomatic patients, however, the symptoms are varied and depend on location, size, and position relative to neighboring organs. The most serious complications in PHC disease are jaundice, cholangitis, and pancreatitis, all of which can develop as a result of fistulization of the cyst content into pancreato-biliary ducts or external compression of those ducts by the cyst. Clinical tools routinely used to diagnose PHC are ultrasonography (USG), computed tomography (CT), magnetic resonance cholangiopancreatography (MRCP), endoscopic ultrasound (EUS), and serological testing. Despite the advanced radiological imaging instruments in use, it is not always easy to differentiate hydatid cysts from common cystic neoplasms of the pancreas. Thus, hydatid cyst disease should be considered in the differential diagnosis of pancreatic cystic lesions, especially in patients living in endemic areas. In this study, we review the cases of PHC in the literature and present a new PHC patient who was treated at our clinic to provide a comprehensive discussion of this disease and its features relevant to diagnosis and management.

MATERIALS AND METHODS

The primary aim of this study was to review cases of PHC published in the literature within the last 4.5 years. To this end, a literature search was made of the PubMed, Medline, Google Scholar, and Google databases using the keywords hydatid cyst, hydatid disease, unusual location of hydatid cyst, hydatid cyst and pancreas, pancreatic hydatid cyst, and pancreatic echinococcosis (alone or in different combinations). All identified abstracts, case reports, letters to the editor, review articles, original articles, and other documents were reviewed. No language filter was set and the review period was set from January 2010 to April 2014. Reference lists of the retrieved articles were also examined to identify citations that complied with our inclusion criteria. Corresponding authors of the articles were contacted by email to obtain more detailed information about the patients. Articles without an accessible full-text version or those providing insufficient information or insufficient data for comparison with other studies were excluded. A table (Table 1) was generated using the following information: publication year, country, and language; patient age, sex, and complaint; cyst...
| Ref.          | Year | Country | Language | Paper type | Case count | Age (yr) | Sex | Complaint/examination findings                                                                 | Cyst location | Cyst size (mm) | Serology |
|--------------|------|---------|----------|------------|------------|----------|-----|-------------------------------------------------------------------------------------------------|--------------|---------------|----------|
| Trigui et al. | 2013 | Tunisia | English  | Article    | 12         | 21 F     | Epigastric mass + epigastric pain + RUQ pain + vomiting + nausea                              | Tail          | NS            | NS       |
|              |      |         |          |            |            |          | M    |                                                                                                   | Tail + Body   | NS            | NS       |
|              |      |         |          |            |            |          | 13 M | Epigastric mass + RUQ pain + vomiting + nausea                                                   | Head          | NS            | NS       |
|              |      |         |          |            |            |          | 15 M | Jaundice + RUQ pain + vomiting + nausea                                                          | Head          | NS            | NS       |
|              |      |         |          |            |            |          | 26 M | Epigastric pain + RUQ pain + vomiting + nausea                                                   | Head          | NS            | NS       |
|              |      |         |          |            |            |          | 30 F | Epigastric pain + RUQ pain + vomiting + nausea                                                   | Head          | NS            | NS       |
|              |      |         |          |            |            |          | 37 F | Jaundice + RUQ pain + vomiting + nausea                                                          | Head          | NS            | NS       |
|              |      |         |          |            |            |          | 8 M  | Jaundice + RUQ pain + vomiting + nausea                                                          | Head          | NS            | Negative |
|              |      |         |          |            |            |          | 26 F | Pancreatitis + epigastric pain + RUQ pain + vomiting + nausea                                   | Tail + body   | 40            | Positive |
|              |      |         |          |            |            |          | 61 F | Epigastric pain + RUQ pain + vomiting + nausea                                                   | Tail          | NS            | Positive |
|              |      |         |          |            |            |          | 11 F | Jaundice + RUQ pain + vomiting + nausea                                                          | Head          | NS            | Negative |
|              |      |         |          |            |            |          | 16 F | Epigastric pain + RUQ pain + vomiting + nausea                                                   | Body          | NS            | Negative |
|              |      |         |          |            |            |          | 11 F | Jaundice + epigastric mass + RUQ pain + vomiting + nausea                                       | Head          | 50            | NS       |
| Yarlagadda et al. | 2013 | India    | English  | Case Report | 1          | 43 M     | Epigastric mass + RUQ pain + vomiting + nausea                                                  | Tail          | 180 × 170 | NS       |
| Patil et al. | 2013 | India    | English  | Case Report | 1          | 47 M     | Epigastric mass + RUQ pain + vomiting + nausea                                                  | Tail + Body   | 100 × 80 | Positive |
| Kaushik et al. | 2013 | India    | English  | Case Report | 1          | 18 F     | RUQ pain + vomiting + nausea                                                                   | Tail          | 65 × 63 | NS       |
| Baghbanian et al. | 2013 | Iran     | English  | Case Report | 1          | 46 M     | Epigastric mass + RUQ pain + vomiting + nausea                                                  | Tail          | 60 × 50 | NS       |
| Gundes et al. | 2013 | Turkey   | Turkish  | Case Report | 2          | 24 F     | Back pain + fever + nausea                                                                    | NS            | 70            | NS       |
| Mandelia et al. | 2012 | India    | English  | Case Report | 1          | 6 M      | Jaundice + fever + epigastric mass + RUQ pain + vomiting + nausea                               | Head          | 54 × 41 | Not-done |
| Kushwaha et al. | 2012 | India    | English  | Case Report | 1          | 40 M     | Epigastric mass + epigastric pain + RUQ pain + vomiting + nausea                                | Tail + Body   | NS            | Positive |
| Makni et al.  | 2012 | Tunisia  | English  | Review      | 1          | 38 M     | Abdominal pain + vomiting + nausea                                                              | Tail          | 100 × 90 | Positive |
| Karaman et al. | 2012 | Turkey   | English  | Case Report | 1          | 32 M     | Epigastric mass + RUQ pain + vomiting + nausea                                                  | Neck          | 55 × 45 | Positive |
| Rayate et al. | 2012 | India    | English  | Case Report | 1          | 30 F     | Abdominal pain + vomiting + nausea                                                              | Tail          | 62 × 57 | Positive |
| Suryawanshi et al. | 2011 | India    | English  | Case Report | 1          | 20 M     | Epigastric mass + epigastric pain + RUQ pain + vomiting + nausea                                | Head          | 80 × 80 | NS       |
| Varshney et al. | 2011 | India    | English  | Case Report | 1          | 35 M     | Abdominal pain + vomiting + nausea                                                              | Tail          | NS            | Positive |
| Somani et al. | 2011 | India    | English  | Case Report | 1          | 30 F     | Epigastric mass + epigastric pain + RUQ pain + vomiting + nausea                                | Head          | NS            | NS       |
| Masoodi et al. | 2011 | India    | English  | Case Report | 1          | 45 M     | RUQ mass + vomiting + nausea                                                                   | Tail          | 70 × 60 | Positive |
| Makni et al.  | 2011 | Tunisia  | English  | Case Report | 1          | 30 F     | Epigastric mass + RUQ pain + vomiting + nausea                                                  | Body          | 80            | Positive |
| Bhat et al.  | 2011 | India    | English  | Case Report | 1          | 4 F      | Jaundice + epigastric mass + RUQ pain + vomiting + nausea                                       | Head + Body   | 150 × 100 | Negative |
| Cankorkmaz et al. | 2011 | Turkey   | English  | Case Report | 1          | 7 F      | Epigastric mass + RUQ pain + vomiting + nausea                                                  | Tail + Body   | 70 × 60 | Negative |
| Dalal et al.  | 2011 | India    | English  | Case Report | 1          | 48 M     | Epigastric mass + RUQ pain + vomiting + nausea                                                  | Tail          | 80 × 50 | Not-done |
| Agrawal et al. | 2011 | India    | English  | Case Report | 1          | 5 F      | Jaundice + abdominal pain + vomiting + nausea                                                  | Head          | 120 × 100 | NS       |
| Küçükkartallar et al. | 2011 | Turkey   | Turkish  | Case Report | 1          | 48 F     | Abdominal pain + vomiting + nausea                                                              | Head          | 25 × 20 | Negative |
| Tavusbay et al. | 2011 | Turkey   | English  | Case Report | 1          | 50 M     | Abdominal mass + vomiting + nausea                                                              | NS            | NS            | Positive |
| Derbel et al. | 2010 | Tunisia  | English  | Article     | 7          | 25 F     | RUQ mass + LUQ pain + vomiting + nausea                                                        | Tail          | 70            | Negative |
| Bansal et al. | 2010 | India    | English  | Case Report | 1          | 30 F     | Abdominal mass + vomiting + nausea                                                              | Tail + body   | 150          | Positive |
| Boubbou et al. | 2010 | Morocco  | English  | Case Report | 1          | 38 M     | Jaundice + epigastric mass + RUQ pain + vomiting + nausea                                       | Tail          | 60            | Negative |
| Shah et al.  | 2010 | India    | English  | Article     | 6          | 46 M     | Epigastric mass + RUQ pain + vomiting + nausea                                                  | Tail          | 25            | Positive |
| Karaman et al. | 2010 | Turkey   | English  | Letter      | 1          | 18 M     | Abdominal mass + RUQ pain + vomiting + nausea                                                  | Body          | 80 × 60 | Not-done |
| Karakas et al. | 2010 | Turkey   | English  | Letter      | 1          | 18 M     | Abdominal mass + RUQ pain + vomiting + nausea                                                  | Body          | 80 × 60 | Not-done |
| Diop et al.  | 2010 | France   | English  | Case Report | 1          | 29 M     | Acute pancreatitis + epigastric mass + RUQ pain + vomiting + nausea                             | Tail          | 35 × 25 | Positive |
| Szanto et al. | 2010 | Romania  | English  | Case Report | 1          | 49 F     | Epigastric mass + bloating + vomiting + nausea                                                  | Tail          | NS            | NS       |
| Caglayan et al. | 2010 | Turkey   | English  | Article     | 1          | 70 M     | NS                                                | NS            | NS            | Positive |
| Orug et al.  | 2010 | Turkey   | English  | Case Report | 2          | 26 M     | Abdominal mass + fatigue + vomiting + nausea                                                   | Tail          | 115 × 95 | NS       |
| Charmakhi-Jendi et al. | 2010 | Tunisia  | French   | Case Report | 1          | 32 F     | Epigastric mass + weight loss + vomiting + nausea                                              | Tail          | 45 × 35 | NS       |

Table 1: Summary of demographic and clinic characteristics patients (n = 57) with pancreatic hydatid cyst published in the medical literature between January 2000 and April 2014.
location and size; results of serologic tests and radiologic examinations; surgical approach, intraoperative complications, postoperative medical management, recurrence, and follow-up (months). In addition, important notes from the studies were summarized in a single sentence.

We also present a case of a 48-year-old woman with PHC who was treated at our clinic and who ultimately died after follow-up. The aim of this case presentation is to emphasize the grave consequences of benign hydatid cyst disease when undiagnosed by preoperative radiological examinations or not considered by a radiologist in differential diagnosis.

RESULTS

Literature review

A literature search using the above review criteria retrieved a total of 33 articles containing 57 cases about PHC disease [36]. Of these, 15 articles were from India, 8 from Turkey, 5 from Tunisia, 2 from Morocco, and 1 from Iran, France and Romania. Twenty-two cases were reported from Tunisia, 20 from India, 10 from Turkey, 2 from Morocco, and 1 from France, Iran and Romania. Twenty-nine articles were written in English, 2 in Turkish, and 2 in French. The current analysis, therefore, included a total of 58 patients (including our one new case), represented by 29 (50%) females and 29 (50%) males, aged 4 to 70 (mean ± SD: 31.4 ± 15.9) years. The age range of the males was 6 to 70 (mean ± SD: 33.4 ± 16.2) years and that of the females was 4 to 61 (mean ± SD: 29.4 ± 15.4) years. Cyst location in pancreas was reported for 54 patients, wherein the cyst was localized to the pancreatic head in 21 (38.8%), the pancreatic tail in 18 (33.3%), the pancreatic body and tail in 8 (14.8%), the pancreatic body in 5 (9.2%), the pancreatic head and body in 1, and the pancreatic neck in 1. Extra-pancreatic location of hydatid cysts was reported for 44 patients. Among these, 32 (72.7%) had no other foci other than pancreas (isolated) while the remaining 12 patients had extra-PHC as follows: liver, n = 6; liver + spleen + peritoneum, n = 2; kidney, n = 1; liver + kidney = 1; kidney + peritoneum = 1; and liver + lung, n = 1. Serological data were available from reports of 40 patients, of which 21 (54%) were serologically positive and 15 (38%) were serologically negative; the remaining 4 patients underwent no serological testing. Information about pancreatic cyst size was available for 42 patients; the smallest cyst diameter was 26 mm and the largest cyst diameter was 180 mm (mean ± SD: 71.3 ± 36.1 mm). Postoperative follow-up information was available for 48 patients and postoperative recurrence information for 51 patients. During the average follow-up duration of 22.5 ± 23.1 (range: 2-120) mo, none of the patients developed recurrence. Only two patients (our new case) died on postoperative day 4 and 15 respectively. Tables 1 and 2 provides detailed information regarding chief demographic data of the 57 patients included in the study.

A 48-year-old woman presented to our outpatient clinic with malaise, fatigue, pruritus, yellowish discoloration of the eyes, darkening of urine color, and acholic gaita. She explained that her complaints, except for jaundice, had started 2 mo previous and the jaundice developed 15 d ago. On physical examination, her sclerae were icteric and whole body was jaundiced. Biochemical tests revealed the following results: aspartate aminotransferase (AST): 205 U/L; alanine aminotransferase (ALT): 673 U/L; total bilirubin: 11.6 mg/dL; direct bilirubin: 9.3 mg/dL; hemoglobin: 13 g/dL; platelet count: 244000/μL; white blood cell count: 5000/μL; carbohydrate antigen 19-9: 45 U/mL (normal range: 0-39). Ultrasonography showed that her gall bladder was hydropic and that the common bile duct and intrahepatic bile ducts were dilated. In addition, a 50 mm × 43 mm anechoic lesion consistent with choledococele was detected in the distal common bile duct. A MRCP was performed and showed a common bile duct diameter of 11 mm, dilated intrahepatic bile ducts, and a 4.5 cm mass in the pancreatic head, which appeared hypointense on T2A imaging and caused stenosis in the distal tip of the common bile duct (Figure 1). An ERCP showed no intraluminal mass lesion. The consensus of a gastroenterologist and a radiologist was that this lesion could be a choledococele or duodenal diverticulum. Considering the above findings, a laparotomy was scheduled, in which the abdomen was entered via a midline incision followed by opening of the gastrocolic ligament and application of the Kocher maneuver. A mass lesion of 5 cm × 5 cm was observed in the pancreatic head, and appeared to be malignant. The common bile duct was markedly dilated. Based on preoperative tests and the intraoperative appearance of the pancreatic head, the mass was regarded as a malignant lesion, and a pancreaticoduodenectomy with pyloric preservation was performed without any intraoperative complications. On post-surgery day 1, the patient’s liver function tests were abnormal and her blood pressure dropped. Yet, radiological tests revealed no abnormalities. Since her blood pressure and pulse continued to deteriorate substantially, the patient was taken back to the operating room. During laparotomy, it was observed that all intestinal segments were filled with abundant blood. A regional exploration revealed a pulsatile bleeding focus from a location close to the Wirsung canal in the intestinal lumen. The bleeding was stopped, and the patient was admitted to the intensive care unit. Unfortunately, the profound coagulopathy that developed in the patient could not be reverted and she died on postoperative day 4. A detailed examination of the pathology specimen demonstrated that the mass had characteristics consistent with a hydatid cyst (Figures
Table 2  Detailed information regarding chief demographic data of the 57 patients included in the study

| Ref | Surgical approach | Radiology | Complication | Medical treatment | Recurrence | Follow-up (mo) | Other hydatid cyst focuses |
|-----|-------------------|-----------|--------------|-------------------|------------|----------------|--------------------------|
| 1 | Partial cystectomy + drainage | USG + CT | Hemorrhage | No | 24 | Liver + spleen + peritoneum |
| 2 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | Liver + lung |
| 3 | Partial cystectomy + drainage | USG + CT | Papainic infection | No | 9 | No |
| 4 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 |Liver |
| 5 | Partial cystectomy | USG + CT | Hemorrhage | No | 26 | No |
| 6 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 7 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 8 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 9 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 10 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 11 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 12 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 13 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 14 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 15 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 16 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 17 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 18 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 19 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 20 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 21 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 22 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 23 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 24 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 25 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 26 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 27 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 28 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 29 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 30 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 31 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 32 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 33 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 34 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 35 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 36 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 37 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 38 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 39 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 40 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 41 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 42 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 43 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 44 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 45 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 46 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 47 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 48 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 49 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 50 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 51 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 52 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 53 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 54 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 55 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
| 56 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 24 | No |
| 57 | Partial cystectomy + drainage | USG + CT | Pancreatic fistula | No | 9 | No |
This patient arrived at the hospital with signs of acute pancreatitis. Despite tienam therapy, he developed renal dysfunction and pancreatic necrosis that affected 50% of the organ. Due to deterioration in his overall status, the patient underwent distal pancreatectomy + necrosectomy + partial nephrectomy (for renal hydatid cyst). Unfortunately, the patient was lost on postoperative day 15.

This patient was admitted to the hospital with episodes of obstructive jaundice and elevated liver function tests. Results from MRCP and US were both consistent with a choledocal cyst. An intraoperative cholangiography revealed normal bile flow; however, the cyst perforated into the peritoneal cavity during the procedure, and open surgery was performed and a pancreatic fistula developed. The drain was removed 18 d later.

This patient was initially diagnosed with a pancreatic fistula. MR and EUS localized the fistula between the pancreatic duct and cyst. USG: Ultrasonography; CT: Computed tomography; MRCP: Magnetic resonance cholangiopancreatography; ERCP: Endoscopic retrograde cholangiopancreatography; AST: Aspartate aminotransferase; ALT: Alanine aminotransferase; MRI: Magnetic resonance imaging; NS: Not-stated; FNAC: Fine needle aspiration cytology; US: Ultrasonography; EUS: Endoscopic ultrasonography.

DISCUSSION

Humans have no biological role in the life cycle of hydatids, and they are inadvertently infected upon ingestion of Echinococcus eggs containing live oncospheres in canine feces. The ingested eggs first penetrate the intestinal wall, then pass to the portal systems and ultimately reside in hepatic sinusoids.3,4 Larvae that escape the liver's filtering system (first Lemman's filter) and reach the lungs where they are entrapped by a second capillary filtering system (second Lemman's filter). Larvae that escape the lung then pass to any part of the human body via arterial circulation.5 The organization of the filtering systems explain why hydatid cysts most commonly reoccur in organs that are supplied by arteries rather than veins,6 and why the lung is the most common site of infection.7

Humans

The life cycle of hydatid cysts involves four phases: 

1. Egg ingestion by the intermediate host: 
   - Dogs, wolves, foxes, etc.

2. Larval development: 
   - The oncosphere hatches and migrates to the liver or lungs via the portal vein or lung capillaries.

3. Cyst formation: 
   - The oncosphere develops into a hydatid cyst, which may grow slowly over many years or rapidly become symptomatic.

4. Cyst rupture: 
   - Hydatid cysts can rupture into the peritoneal cavity, pleural space, or other body cavities, leading to dissemination of the larval form and possible reinfection.
side in the liver, with the second most common residence being the lung.

A number of hypotheses regarding the mode of passage of E. granulosus to pancreas have been postulated, the most accepted is the hematogenous dissemination discussed above. The second route involves passage of cystic elements into the biliary system and then to the pancreatic canal and pancreas. The third route involves passage of cystic elements into lymphatic channels through the intestinal mucosa and then to pancreatic tissue rich in lymphatic network. The fourth route is direct passage of larvae into pancreatic tissue, bypassing the liver, via pancreatic veins. The fifth, and final, hypothesized route is retroperitoneal dissemination. In our literature review, an isolated PHC was detected in 72% and a secondary PHC was detected in 28% of 43 cases where medical data were available. The PHC incidence varies by region, ranging from 0.1% to 2%. Pancreatic cysts are solitary 90%-91% of the time, and their pancreatic distribution is heterogeneous. According to data from the lit-

Figure 1  Magnetic resonance cholangiopancreatography shows a hydropic gall bladder and dilated intrahepatic bile ducts and common bile duct. The distal portion of the common bile duct is narrowed due to external compression. A: A pancreatic cyst compressing the lower tip of the common bile duct is also seen in this section; B: A coronal T2-weighed MR cross-section shows a pancreatic cyst and a dilated common bile duct; C: Intravenous T1-weighed axial cross-section with contrast enhancement shows a pancreatic cyst; D: An axial computerized tomography cross-section with contrast enhancement shows a pancreatic cyst with a thick wall and without central contrast uptake. MR: Magnetic resonance.

Figure 2  Patient’s mass has characteristics consistent with a hydatid cyst. The cyst wall is surrounded by fibrous capsule (also called the pericyst layer). The adjacent parenchyma demonstrates pressure atrophy (hematoxylin-Eosin stain ×100).

Figure 3  Cyst wall of the patient’s mass consists of a laminated faintly stained chitinous membrane (outer layer). Multiple protoscolices are present within the daughter cyst (inner germinal layer, hematoxylin-Eosin stain × 100).
the pancreatic-biliary system. However, the rate of this complication was far below the expected rate. Analysis of patients’ blood tests showed that 8 patients had elevated bilirubin (2.9-11.7 mg/dL), 9 had elevated ALP (280-1843 U/L), 7 had elevated ALT (56-335 U/L), 7 had elevated AST (72-235 U/L), 7 had elevated amylase (610-4965 U/L), and 2 had elevated lipase (103-1390 U/L).

The first and most important step in the diagnosis of PHC is clinical suspicion. Important clues include residence in an endemic region or a previous hydatid cyst surgery. These clues may increase diagnostic yield when assessed in conjunction with results from radiological studies and serological tests. For diagnosis of pancreatic cysts, the most commonly performed radiologic tests are, in descending order, USG, CT, and magnetic resonance imaging (MRI). Complicated cases that require further workup are examined with invasive diagnostic tools, such as EUS and ERCP. USG is a noninvasive, low-cost and sensitive diagnostic instrument. Gharbi defined the typical appearance of hydatid cysts in USG, but application of USG to pancreatic cysts is lower than for liver cysts because of the retroperitoneal location of the pancreas and bowel gas. CT is usually successful in delineating cyst size, location, relation with pancreateo-biliary system, and presence of cysts in other organs. It is also successfully used for treatment monitoring and postoperative recurrence detection. MRI and MRCP are particularly useful to delineate the relationship between cysts and pancreatic and bile ducts. However, results from these techniques may be insufficient when attempting to differentiate between cysts located at the pancreatic head and those located at the common bile duct. In MRI, superposition of the hydatid cyst with the pancreatic duct can be misinterpreted as a fistula. To demonstrate the relationship between cyst and pancreatic duct and to differentiate cysts of unknown nature, ERCP can be used. ERCP is appropriate for palliative stent applications in cases with cholangitis or pancreatitis secondary to biliary or pancreatic duct obstruction. It is also very beneficial in non-operative management of cases that developed biliary or pancreatic fistulae. EUS is not commonly used, but it is capable of delineating pancreato-biliary system anatomy and taking biopsy samples when necessary. It can accurately show the relationship between the cyst and pancreatic duct. Cystography during surgery is especially helpful to demonstrate the relationship of the cyst with the pancreateo-biliary ducts and gastrointestinal tract. In complicated cases, the gall bladder and common bile duct can be entered with a needle and a cholangiogram can be taken, which may show both the anatomy of biliary ducts and their relationship with cyst.

For diagnosis, screening, and recurrence monitoring, the following serological tests are used: enzyme-linked immunosorbent assay, indirect hemagglutination, serum immunoelectrophoresis, complement fixation test, and immunofluorescence assay. The seropositivity rate is
higher in hepatic hydatid cysts than cysts in other organs. We calculated a rate of 54% for PHC cases. It should be noted, however, that seronegativity does not guarantee absence of hydatid disease[26].

The differential diagnosis of PHCs include neoplastic (cystadenoma, cystadenocarcinoma, gastroenteroendocrine tumors, vascular tumors, metastatic cystic lesions) or non-neoplastic (congenital pancreatic cysts, pseudocysts) cystic lesions[16,20,21]. Diagnosis of cysts that cannot be made using noninvasive techniques can be made by taking either a biopsy from the lesion or an aspiration cytology sample from cyst fluid via percutaneous or endo-ultrasonographic techniques[18,24]. Using percutaneous fine needle aspiration cytology (FNAC) for the differential diagnosis of cystic pancreatic lesion, Varshney et al[18] showed hooklets of hydatid cyst cytologically. In contrast, Dalal et al[21] had to perform FNAC twice in order to diagnose hydatid cyst. Anaphylaxis and pouring of cyst content into the abdominal cavity are potential complications of the FNAC procedure. Hence, prophylactic antihelminthic agents should be started when FNAC is contemplated in a patient with suspected cysts; otherwise, the procedure should be avoided[46].

All patients presented in this review underwent at least one preoperative radiological or serological test. After these tests, 20 patients were diagnosed with PHC, 14 with benign/neoplastic cystic lesion of pancreas, 8 with choledocal cyst, 4 with PHC/cystic neoplasm of pancreas, 2 with hepatic hydatid cyst and one with splenic hydatid cyst. Minimally invasive surgery was contemplated. No presumptive diagnoses were made for the remaining patients. As seen, only 40%-49% of patients were diagnosed with PHC at the preoperative period. This is true even for the most recent studies performed within the last 4.5 years. Diagnostically, the situation was even worse several decades ago, when the rate of preoperative PHC was far below 30%.

PHC can be treated with one or a combination of several therapies, including open or laparoscopic surgical approach, minimally invasive approach [puncture-aspiration-injection-aspiration (PAIR) or direct percutaneous catheterization], and medical therapy[9]. As is the case for other organ hydatid cysts, open surgery is the gold standard for the treatment of PHC disease. Selection of the appropriate management approach is affected by many factors, such as surgeon’s experience, patient age, presence of comorbid conditions, pancreatic localization of cyst(s), cyst size, and relation of cyst to adjacent structures or the pancreatic and common bile ducts[13,18].

Pancreatic head cysts with no communication with biliary or pancreatic ducts can be managed with partial cystectomy + external drainage, partial cystectomy + omentectomy and pericystectomy, marsupialisation, and pancreaticoduodenectomy procedures[18,26]. Each method has its own advantages and disadvantages. In order to avoid postoperative pancreatic fistula formation, cysts with communication with the pancreatic duct can be treated with cysto-jejunal, cysto-duodenal, or cysto-gastric anastomosis techniques[18,26]. In cysts located in the pancreatic body or tail, the most appropriate approach is a spleen-preserving distal pancreatectomy[18,26]. In cases where the spleen cannot be preserved, pneumocoecal and meningocoecal vaccinations should be done immediately to avert postpancreatectomy complications[19]. Central pancreatectomy may be preferable when cysts are localized to the pancreatic body or neck[29]. The main advantage of this method is the preservation of pancreatic tissue and the minimization of complications, such as diabetes or exocrine pancreatic insufficiency[29]. Masoodi et al[31] reported in a patient that underwent a distal pancreatectomy hyperglycemia high enough to require insulin injection. For management of hydatid cysts of the pancreatic head, the role of pancreaticoduodenectomy is very limited[29]. Pancreaticoduodenectomy was performed in only 3 of 19 pancreatic head cysts[17,21]. A whipple procedure was applied in all three of these cases since the results of preoperative radiological examination and/or intraoperative findings were consistent with a cystic lesion of the pancreatic head. In our case, we experienced similar difficulties. While the preoperative tests, including CT, MRC, and ERCP, were consistent with a choledocal cyst, the intraoperative appearance was totally compatible with a mass in the pancreatic head. Unfortunately, our patient was lost to a misfortunate complication. In retrospect, we realize that patient outcome may have been improved if the diagnosis was made preoperatively and simple partial cystectomy and drainage was performed intraoperatively. Hence, our main objective for writing this manuscript was to heighten awareness about this topic.

Although rarely reported in the literature, there are some studies describing percutaneous drainage of pancreatic hydatid cysts[16,26]. Percutaneous drainage can be accomplished by puncture, aspiration, injection of hypertonic saline solution, and re-aspiration of cyst content (PAIR) or direct catheterization of the cyst[13,18,26]. These procedures should be specifically carried out in Type I and II PHCs, cysts with a diameter less than 50 mm, patients who refuse surgery, and cases with a higher anesthesia risk[3]. The main advantage of PAIR is the ability to show scoleces in the aspirated cyst fluid cytopathologically within a short period of time. Another advantage is the ability to delineate the relative location of the cyst with the pancreatic duct by contrast material administration during the procedure. However, an unconscious percutaneous drainage procedure or one that is performed without estimating the possible presence of PHC may lead to cyst perforation and surgical complications[21]. The risk associated with release of cyst contents into abdominal cavity is markedly lower with a PAIR procedure that is carried out by passing through parenchyma of solid organs like liver and spleen than it would be with pancreatic and other intraabdominal cysts[29]. In cases where minimally invasive surgical therapy has been contemplated, antihelminthic therapy should be administered before (≥ 4 d) and after (≥ 3-4 wk) the procedure in order to reduce intracystic pressure and prevent anaphylaxis[41].
Although there are numerous articles about laparoscopic excision of hydatid cysts in other organs, there are only a few case reports on the use of the laparoscopic approach for PHCs. In one report, content of a cyst located in the pancreatic head was emptied by directly inserting a 10 mm trochar into the cyst followed by omentoplasty. In our opinion, in order to apply this technique to PHCs, the preoperative diagnosis should be accurately made, the cyst must have an adequate neck, and the surgeon must be experienced in laparoscopy.

Antihelminthic prophylactic therapy (albendazole, mebendazole, or praziquantel) must be administered for 2-4 wk prior to surgery (open, laparoscopic, or PAIR) in order to decrease intracystic pressure and reduce anaphylaxis and postoperative recurrence risks. With radical resections that do not open the cyst cavity, there is no need for medical therapy afterwards. One of the cyclic or continuous medical therapy protocols, however, should be applied during the postoperative period to patients who underwent conservative surgery. During follow-up, these asymptomatic cysts can be followed with medical therapy alone or their size assessed at yearly intervals.

Complications of PHC surgery can be divided into short- and long-term complications. Short-term complications or early postoperative complications include pancreatic fistula, biliary fistula, biloma, intraabdominal abscess, and wound infection. The most suitable approach for treating biloma and intraabdominal abscesses is percutaneous drainage. For biliary and pancreatic fistulae, daily output guides management decisions. ERCP shows well the location of the fistula and the presence of any obstruction due to cystic elements in pancreatobiliary ducts (distal to fistula). Simultaneously, therapeutic procedures like sphincterotomy and/or stent implantation can also be performed with ERCP. Use of somatostatin analogues may hasten closure and reduce output of pancreatic fistulae.

Surgical intervention is rarely needed, and intraoperative cholangiography or cystography may be performed to avoid such complications. In addition, planning surgery in line with cyst location may avert complications. The major long-term complication of cyst surgery is hydatid cyst recurrence. Recurrence is rather common after conservative surgical operations but almost never seen after radical surgery. Recurrence rates can be minimized by applying intraoperative protective measures, which are commonly applied in hepatic hydatid cyst surgery, or by administering preoperative or postoperative medical therapy.

In conclusion, PHC is a rare parasitic infection that can cause serious pancreato-biliary complications. Despite advances in radiological instrumentation, preoperative diagnosis of PHC remains a challenge, and it is often misdiagnosed as other cystic diseases of the pancreas and distal choledochal cysts. Conservative surgical techniques, which are preferred over radical surgical interventions, should be applied, especially in cysts located in the pancreatic head. After confirmation of the diagnosis, cystography is a suitable method to demonstrate the relationship between the cyst and pancreatic duct. While postoperative antihelminthic therapy is not necessary in surgical operations that do not open the cyst cavity, a medical therapy lasting for 3-4 wk is appropriate after more conservative surgical procedures such as partial cystectomy.

**COMMENTS**

**Background**

Pancreatic hydatid cyst disease is rare but can lead to serious pancreato-biliary complications if left untreated. Despite advances in radiological techniques, preoperative diagnosis of pancreatic hydatid cyst remains challenging, and it is frequently misdiagnosed preoperatively as other cystic diseases of the pancreas and distal choledochal cysts.

**Research frontiers**

The authors analyzed previously published articles regarding pancreatic hydatid cyst. For this purpose, a literature search was performed in PubMed, Medline, Google Scholar, and Google databases using different keywords related to pancreatic hydatid cyst. Second, the authors presented a case of a 48-year-old female patient who underwent surgical treatment for pancreatic head hydatid cyst.

**Innovations and breakthroughs**

A review of the literature and personal experience suggest that pancreatic hydatid cyst disease should be considered in the differential diagnosis of pancreatic cystic lesions, especially in patients living in endemic areas.

**Peer review**

Echinococcosis is listed as one of World Health Organizations Neglected Zoonotic Diseases bringing a significant socioeconomic burden, mainly in impoverished and rural areas. The topic of this review is relevant although if performed in a systematic way it would have delivered a stronger evidence-based article for the medical community. Without bringing any new findings, this review stands out over previous attempts as it properly describes the methodology behind the searching and selection process of retrieving articles and represents a comprehensive source of information of reported cases in the last 4.5 years.

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