ORIGINAL ARTICLE

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

Sami Akbulut, Ridvan Yavuz, Nilgun Sogutcu, Bulent Kaya, Sinan Hatipoglu, Ayhan Senol, Firat Demircan

Sami Akbulut, Ridvan Yavuz, Firat Demircan, Department of Surgery, Diyarbakir Education and Research Hospital, Diyarbakir 21400, Turkey

Nilgun Sogutcu, Department of Pathology, Diyarbakir Education and Research Hospital, Diyarbakir 21400, Turkey

Bulent Kaya, Department of Surgery, Kanuni Sultan Suleyman Education and Research Hospital, Istanbul 34303, Turkey

Sinan Hatipoglu, Department of Surgery, Adiyaman University Faculty of Medicine, Adiyaman 02040, Turkey

Ayhan Senol, Department of Radiology, Ergani State Hospital, Diyarbakir 21950, Turkey

Author contributions: Akbulut S, Kaya B and Hatipolgu S designed the report; Akbulut S and Yavuz R were the attending doctors for the patients; Akbulut S, Yavuz R and Demircan F performed the surgical operation; Akbulut S organized the report and wrote the paper; Senol A provided the radiological information; Sogutcu N provided the histopathological information.

Correspondence to: Sami Akbulut, MD, FICS, FACS, Department of Surgery, Diyarbakir Education and Research Hospital, Uckuyular Mevki, Kayapinar, Diyarbakir 21400, Turkey. akbulutsami@gmail.com

Telephone: +90-412-2580052 Fax: +90-412-2580050
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Abstract

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 coun-try. 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.

RESULTS: A total of 58 patients, including our one new 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

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.

Key words: Echinococcosis; Hydatid cyst; Pancreas; Pancreaticoduodenectomy

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[1,2]. 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[1,2]. 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[1,2]. 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[2-5].

Pancreatic hydatid cyst (PHC) disease is rare, even in regions where hydatidosis is endemic[4-7]. 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[6]. 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[4]. 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 ultasound (EUS), endoscopic retrograde cholangiopancreatography (ERCP), and serological testing[4]. Despite the advanced radiological imaging instruments in use, though, it is not always easy to differentiate hydatid cysts from common cystic neoplasms of the pancreas[4,8]. 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

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

Ref.	Year	Country	Language	Paper type	Case count	Age (yr)	Sex	Complaint/examination findings	Cyst location	Cyst size (mm)	Serology
Trigui *et al*[4]	2013	Tunisia	English	Article	12	21	F	Epigastric mass + epigastric pain	Tail	NS	NS
						13	M	Epigastric pain + RUQ pain	Tail + Body	NS	NS
						15	M	Jaundice + RUQ pain	Head	NS	NS
						26	M	Epigastric pain	Head	NS	NS
						50	F	Epigastric pain	Head	NS	NS
						37	F	Jaundice + RUQ pain	Head	25	Negative
						8	M	Jaundice + RUQ pain	Head	83 × 76	Positive
						26	F	Pancreatitis + epigastric pain	TaİL + body	40	Positive
						61	F	Epigastric pain	Tail	NS	NS
						11	F	Jaundice + RUQ pain	Head	NS	Negative
						16	F	Epigastric pain	Body	NS	Positive
						11	F	Jaundice + epigastric mass + RUQ pain	Head	50	NS
Yarlagadda *et al*[5]	2013	India	English	Case Report	1	43	M	Epigastric mass	Tail	180 × 170	NS
Patil *et al*[6]	2013	India	English	Case Report	1	47	M	Epigastric mass	Tail + Body	100 × 80	Positive
Kaushik *et al*[7]	2013	India	English	Case Report	1	18	F	LUQ pain	Tail	65 × 63	NS
Baghbanian *et al*[8]	2013	Iran	English	Case Report	1	46	M	Epigastric pain + fever	Tail	60 × 50	NS
Gundes *et al*[9]	2013	Turkey	Turkish	Case Report	2	24	F	Back pain	NS	70	NS
						50	F	Abdominal pain	Head	50	NS
Mandelia *et al*[10]	2012	India	English	Case Report	1	6	M	Jaundice + fever + epigastric mass	Head	54 × 41	Not-done
Kushwaha *et al*[11]	2012	India	English	Case Report	1	40	M	Epigastric mass + epigastric pain	Tail + Body	NS	Positive
Makni *et al*[12]	2012	Tunisia	English	Review	1	38	M	Abdominal pain + vomiting + nausea	Tail + Body	100 × 90	Positive
Karaman *et al*[13]	2012	Turkey	English	Case Report	1	32	M	Epigastric pain	Neck	55 × 45	Positive
Rayate *et al*[14]	2012	India	English	Case Report	1	30	F	Abdominal pain	Tail	62 × 57	Positive
Suryawanshi *et al*[15]	2011	India	English	Case Report	1	20	M	Epigastric mass + epigastric pain	Head	80 × 80	NS
Varshney *et al*[16]	2011	India	English	Case Report	1	35	M	Abdominal pain + vomiting + nausea	Tail	NS	Positive
Somani *et al*[17]	2011	India	English	Case Report	1	30	F	Epigastric mass + epigastric pain	Head	NS	NS
Masoodi *et al*[18]	2011	India	English	Case Report	1	45	M	LUQ mass	Tail	70 × 60	Positive
Makni *et al*[19]	2011	Tunisia	English	Case Report	1	30	F	Epigastric mass + discomfort	Body	80	Positive
Bhat *et al*[20]	2011	India	English	Case Report	1	4	F	Jaundice + epigastric mass	Head + Body	150 × 100	Negative
Cankorkmaz *et al*[21]	2011	Turkey	English	Case Report	1	7	F	Epigastric mass + weight loss	Tail + Body	70 × 60	Negative
Dalal *et al*[22]	2011	India	English	Case Report	1	48	M	Epigastric fullness + fever	Tail	80 × 50	Not-done
Agrawal *et al*[23]	2011	India	English	Case Report	1	5	F	Jaundice + abdominal pain	Head	120 × 100	NS
Küçükkartallar *et al*[24]	2011	Turkey	Turkish	Case Report	1	48	F	Abdominal pain	Head	28 × 25	Negative
Tavusbay *et al*[25]	2011	Turkey	English	Case Report	1	50	M	Abdominal mass	NS	NS	Positive
Derbel *et al*[26]	2010	Tunisia	English	Article	7	25	F	LUQ mass + LUQ pain	Tail	60	Negative
						19	F	Epigastric mass + RUQ pain	Tail	70	Negative
						32	F	Epigastric pain	Tail + body (two cyst)	150	Positive
						41	M	LUQ mass + LUQ pain + fever	NS	150	Negative
						38	M	Jaundice + epigastric pain	Head	50	Negative
						29	M	Jaundice + epigastric mass	Head	60	Negative
						25	F	LUQ pain + vomiting	Tail + Body	90	Positive
Bansal *et al*[27]	2010	India	English	Case Report	1	30	F	Jaundice + epigastric mass	Head	80 × 60	Not-done
Boubbou *et al*[28]	2010	Morocco	English	Case Report	1	38	M	Jaundice + epigastric pain	Head	NS	NS
Shah *et al*[29]	2010	India	English	Article	6	46	M	Epigastric pain	Tail	28	Positive
						37	F	Epigastric mass + vomiting	Body	26	Positive
						18	M	Dyspepsia	Body	33	Positive
						22	F	Epigastric pain	Tail	48	Negative
						28	M	Jaundice	Head	50	Positive
						68	M	Jaundice	Head	35	Negative
Karakas *et al*[30]	2010	Turkey	English	Letter	1	18	M	Abdominal pain + fever	Body	70 × 45	Negative
Diop *et al*[31]	2010	France	English	Case Report	1	29	M	Acute pancreatitis	Tail	35 × 25	Positive
Szanto *et al*[32]	2010	Romania	English	Case Report	1	49	F	Epigastric pain + bloating + vomiting	Tail	NS	NS
Cağlayan *et al*[33]	2010	Turkey	English	Article	1	70	M	NS	NS	NS	Positive
Orug *et al*[34]	2010	Turkey	English	Case Report	2	26	M	Abdominal pain + fatique + vomiting	Tail	115 × 95	NS
						57	F	Epigastric pain + weight loss	Tail	45 × 35	NS
Chammakhi-Jemli *et al*[35]	2010	Tunisia	French	Case Report	1	32	F	Acute pancreatitis + epigastric mass	Tail	80	Negative

Elmadi *et al*[36]	2010	Morocco	French	Case Report	1	7	M	Jaundice + fever + RUQ pain	Head	NS	Negative

RUQ: Right upper quadrant; LUQ: Left upper quadrant; NS: Not-stated.

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[4-36]. Of these, 15 articles were from India, 8 from Turkey, 5 from Tunisia, 2 from Morocco, and 1 each from Iran, France and Romania. Twenty-two cases were reported from Tunisia, 20 from India, 10 from Turkey, 2 from Morocco, and 1 each 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 hyperintense 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.	Radiology	Surgical approach	Complication	Medical treatment	Recurrence	Follow-up (mo)	Other hydatid cyst focuses
Trigui et al[4]	NS	Distal pancreatectomy	No	NS	No	24	NS
	USG + CT	Partial cystectomy + drainage	Hemorrhage	NS	No	36	Liver + spleen + peritoneum
	USG + CT	Partial cystectomy + drainage	Pancreatic fistula	NS	No	24	Liver
	USG + CT	Partial cystectomy + drainage	Cavity infection	NS	No	NS	Liver + lung
	USG + CT	Partial cystectomy + drainage	Pancreatic fistula	NS	No	5	No
	USG + CT	Pancreaticoduodenectomy	Hemorrhage	NS	No	9	NS
	USG + CT	Partial cystectomy + duodenal fistula treatment	No	NS	No	24	NS
	USG + CT	Distal pancreatectomy + splenectomy	No	NS	No	36	Liver
	USG + CT	Distal pancreatectomy + splenectomy	No	NS	No	36	NS
	USG + CT	Cysto-duodenal anastomosis	No	NS	No	NS	Liver
	USG + CT	Partial cystectomy + drainage	No	NS	No	24	NS
	USG + CT	Cysto-duodenal anastomosis	No	NS	No	NS	NS
Yarlagadda et al[5]	USG + CT	Distal pancreatectomy + splenectomy	No	Albendazole-6 mo	No	6	No
Patil et al[6]	USG + CT	Distal pancreatectomy	No	Albendazole-3 wk	No	6	No
Kaushik et al[7]	USG + CT	Total cystectomy + marsupialisation	No	Albendazole-3 wk	No	6	NS
1Baghbanian et al[8]	USG + CT	Distal pancreatectomy + necrozectomy + partial nephrectomy	Died (POD15)	No	Died	Died	Kidney
Gundes et al[9]	NS	Partial cystectomy + omentoplasty	Wound infection	Albendazole-4 mo	No	NS	NS
	CT	Partial cystectomy + omentoplasty	Wound infection	Albendazole-4 mo	No	NS	No
2Mandelia et al[10]	USG + MRCP	Enucleation + cholangiography	No	Albendazole-3 wk	No	24	No
Kushwaha et al[11]	USG + CT	PAIR + epigastric cyst excision	Pancreatitis	Albendazole-3 mo	No	6	Kidney + peritoneum
Makni et al[12]	CT	Distal pancreatectomy + splenectomy	No	Albendazole-3 mo	No	8	No
3Karaman et al[13]	USG + CT	Percutaneous drainage	No	Albendazole-2 mo	No	18	No
Rayate et al[14]	USG + CT	Distal pancreatectomy	No	Albendazole-2 mo	No	7	No
4Suryawanshi et al[15]	USG + CT	Cyst evacuation + omentoplasty-laparoscopic	No	Albendazole-3 mo	No	3	No
5Varshney et al[16]	CT	Distal pancreatectomy	No	Albendazole-3 wk	No	10	No
6Somani et al[17]	USG + CT + ERCP	Pancreaticoduodenectomy	No	Albendazole-6 mo	NS	6	NS
7Masoodi et al[18]	USG + CT	Distal pancreatectomy + splenectomy	Hyperglisemia?	Albendazole-1 mo	No	6	No
Makni et al[19]	USG	Distal pancreatectomy + splenectomy	Pancreatic fistula	Albendazole-3 mo	No	8	No
Bhat et al[20]	USG + CT	Partial cystectomy + drainage	No	Albendazole-3 mo	No	24	No
8Cankorkmaz et al[21]	USG + CT	Partial cystectomy + drainage	Pancreatic fistula	NS	No	24	No
9Dalal et al[22]	USG + CT	Cyst excision along with tail of the pancreas	No	Albendazole-6 mo	No	8	No
10Agrawal et al[23]	USG + MRCP	Enucleation + cholangiography + cholecystectomy + cystography	No	Albendazole-2 mo	NS	NS	NS
11Küçükkartallar et al[24]	USG + CT + MRCP	Partial cystectomy + omentoplasty	No	Albendazole-4 mo	No	10	No
12Tavusbay et al[25]	USG + CT	Partial cystectomy + omentoplasty + splenectomy + mesenteric cyst excision	Wound infection	Albendazole-6 mo	No	12	Liver + spleen + peritoneum
13Derbel et al[26]	USG + CT	Partial cystectomy	No	No	No	35	No
	USG + CT	Partial cystectomy (for all cysts)	No	Albendazole-6 mo	No	9	Liver (13 cyst)
	USG + CT	Partial cystectomy for both pancreatic cysts	No	No	No	6	No
	USG + CT	Distal pancreatectomy	No	No	No	9	No
	USG + CT + MRI	Partial cystectomy (for all cyst)	Acute pancreatitis	Albendazole-6 mo	No	14	Liver
	USG + CT	Partial cystectomy	No	No	No	16	No
14Bansal et al[27]	USG + CT	Distal pancreatectomy + splenectomy	No	No	No	2	No
Boubbou et al[28]	USG + MRCP + EUS	Pancreaticoduodenectomy	No	Albendazole-6 wk	No	6	No
	USG + CT	PAIR + drainage + cystogastrostomy laparoscop	No	NS	NS	NS	NS
Shah et al[29]	USG + CT	Distal pancreatectomy + splenectomy	No	Albendazole-6 wk	No	120	No
	USG + CT	Central pancreatectomy + reconstruction	No	Albendazole-6 mo	No	96	No

Ref.	Imaging	Procedure	Complication	Albendazole				Location
	USG + CT + MRI	Pericystectomy	No	Albendazole-6 mo	No	No	52	No
	USG + CT	Distal pancreatectomy + splenectomy	No	Albendazole-6 wk	No	No	50	No
	USG + CT + MRI	Partial cystectomy + evacuation + T-tube drainage	No	Albendazole-6 mo	No	No	30	Liver
	USG + CT + MRI	Partial cystectomy + evacuation + T-tube drainage	No	Albendazole-6 mo	No	No	4	No
[15]Karakas et al[30]	CT + MR + EUS	Distal pancreatectomy + cystectomy	Pancreatic fistula	NS	No	No	4	No
[16]Diop et al[31]	USG + CT + EUS	Distal pancreatectomy + polar nephrectomy + partial cystectomy-Liver	No	Albendazole-18 mo	No	No	48	Liver + kidney
Szanto et al[32]	NS	NS	Hemoperitoneum	Not-used	No	NS	NS	NS
Cağlayan et al[33]	USG + CT + EUS	Distal pancreatectomy + splenectomy	NS	Albendazole	NS	NS	NS	NS
Orug et al[34]	USG + CT	Distal pancreatectomy + splenectomy	No	Albendazole-6 mo	No	No	24	No
	USG + CT	Distal pancreatectomy + splenectomy	No	Albendazole-3 mo	No	No	24	No
Chammakhi-Jemli et al[35]	USG + CT + MRI	Distal pancreatectomy + splenectomy	No	NS	No	NS	NS	NS
Elmadi et al[36]	USG + MRI	Partial cystectomy + evacuation	No	NS	No	NS	24	No

[1]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; [2]This patient was admitted to physician with episodes of obstructive jaundice and elevated liver function tests. Results from MRCP and US were both consistent with a choledochal cyst. An intraoperative cholangiography revealed normal bile flow; [3]This patient underwent percutaneous drainage after 12 d treatment with albendazole. A cystogram was done, and there was no relationship between cyst and pancreatic duct. The patient received albendazole for 2 mo after the operation. The serologic test was negative during follow-up; [4]This patient was tested for abdominal pain, leukocytosis, and hyperamylasemia and diagnosed with a pancreatic head cyst that compressed the duct externally. Subsequently, the patient developed acute pancreatitis secondary to ductus compression; [5]No differential diagnosis was made on CT. Therefore, a FNAC was performed and cytologic examination revealed hooklets of parasite. The patient was administered a 21 d course of albendazole at pre-operative and post-operative periods; [6]This patient developed jaundice 1 year ago and had elevated levels of AST and ALT. Findings from US, CT, and ERCP were consistent with a choledocal cyst. Thus, a stent was placed in the common bile duct, and a laparotomy operation was performed. After intraoperative exploration, the lesion was considered a cystadenoma. Whipple operation was performed since dissection of the cystic lesion was difficult; [7]The patient was administered albendazole for 4 d preoperatively and 1 mo postoperatively. Postoperative hyperglycemia developed as a complication and was treated with insulin; [8]A pancreatic pseudocyst was considered to exist, and a US-guided drainage was attempted. 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; [9]Clinical presentation and CT findings of this patient were consistent with an abscess. A US-guided aspiration was performed, and the culture result was sterile. Although the patient was sent home on medical therapy, he was admitted to the hospital again with signs of intestinal obstruction. A hydatid cyst was initially diagnosed, and a US-guided cytology confirmed hydatidosis; [10]This patient was initially diagnosed with a choledocal cyst and underwent diagnostic laparoscopy. The cystic lesion originated from the pancreatic head and the aspirated fluid sample from the cyst clear fluid rather than bile. This finding indicated hydatid cyst, and an open operation was performed. Despite findings consistent with a pancreatic fistula on MRCP, no such relationship was detected in cytography. A cholangiography was carried out, and the fluid easily passed to duodenum; [11]MRCP images of this patient were consistent with a Type-II choledocal cyst; [12]This patient had two previous hepatic hydatid cyst surgeries. He had cysts in liver, spleen, pancreas, and the area below the incision. Albendazole treatment was begun 3 wk prior to the operation. The operation team performed splenectomy + total peritoneal cyst excision + partial cystectomy + omentoplasty for two cysts in liver; [13]The authors reported there was no relationship between hydatid cyst and common bile duct or pancreatic duct in any patient. Only 2 patients had bile duct dilatation secondary to compression of the common bile duct by hydatid cyst; [14]USG revealed dilatation in bile ducts. MRCP was consistent with a type-III choledocal cyst. EUS showed a mass lesion originating from the pancreatic head. Since its intraoperative resembled a cystic neoplasm, a Whipple procedure was carried out. Examination of the specimen revealed the relationship between cyst and pancreatic duct; [15]This patient had elevated liver function tests and increased blood amylase levels (acute pancreatitis) in preoperative testing. Radiologically, dilatation of the pancreatic duct and enlargement of the pancreatic body were apparent. The patient received preoperative albendazole treatment. A fluid collection developed at the surgical area at the postoperative period, and a US-guided drainage catheter was placed. In addition, the pancreatic duct was stented, and external drainage dramatically improved after the stenting procedure; [16]A pancreatic cyst was diagnosed in an examination performed for pancreatitis. 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: Ultrasound.

2 and 3). In addition, areas of severe fibrosis were noted in regions neighboring the cyst.

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 system, and ultimately reside in hepatic sinusoids[3,7]. Larvae with a diameter less than 0.3 mm can 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 may then pass to any part of the human body *via* arterial circulation[1-3]. The organization of the filtering systems explain why hydatid cysts most commonly re-
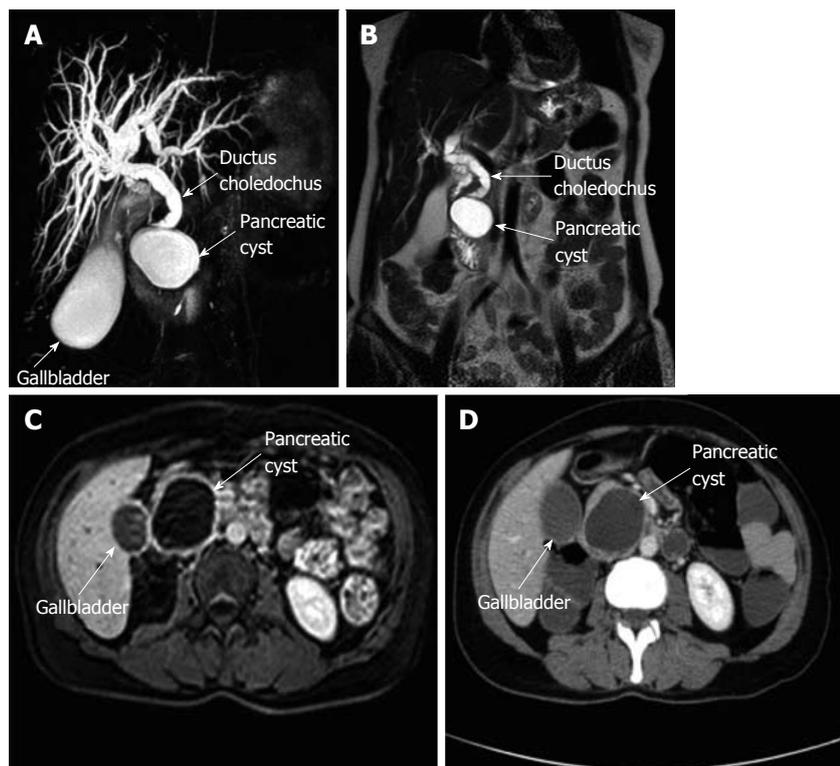
Figure 1 Magnetic resonance cholangiopancreatography shows a hydropic gall bladder and dilated intrahepatic bile ducts and common bile duct. The distal portion of the common bile duct is narrowed due to external compression. A: A pancreatic cyst compressing the lower tip of the common bile duct is also 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.
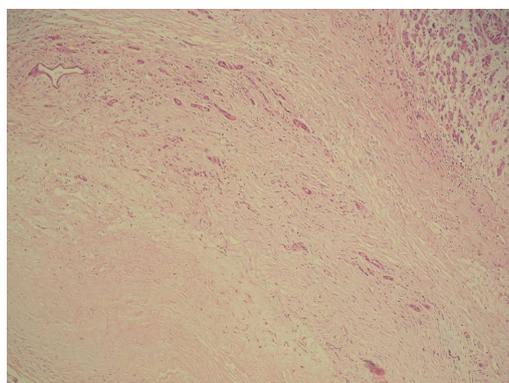
Figure 2 Patient's mass has characteristics consistent with a hydatid cyst. The cyst wall is surrounded by fibrous capsule (also called the pericyst layer). The adjacent parenchyma demonstrates pressure atrophy (hematoxylin-Eosin stain ×100).
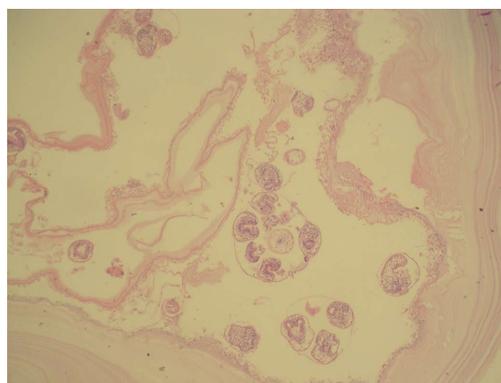
Figure 3 Cyst wall of the patient's mass consists of a laminated faintly stained chitinous membrane (outer layer). Multiple protoscolices are present within the daughter cyst (inner germinal layer, hematoxylin-Eosin stain × 100).

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[3,8,10-12,14,27]. The second route involves passage of cystic elements into the biliary system and then to the pancreatic canal and pancreas[3,10,14,27]. The third route involves passage of cystic elements into lymphatic channels through the intestinal mucosa and then to pancreatic tissue rich in lymphatic network[3,8,10-12,14,20,27]. The fourth route is direct passage of larvae into pancreatic tissue, bypassing the liver, *via* pancreatic veins[20]. The fifth, and final, hypothesized route is retroperitoneal dissemination[27,34]. 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%[5,8,9,11,13,15,16,29]. Pancreatic cysts are solitary 90%-91% of the time, and their pancreatic distribution is heterogeneous[4,19,26]. According to data from the lit-

erature, 50%-58% of PHCs are found in the pancreatic head, 24%-34% in the pancreatic body, and 16%-19% in the pancreatic tail[4,5,12,19]. The rich vascular network found in the pancreatic head suggests that larvae reach this region *via* systemic circulation[4,26]. We did not find any significant difference in location prevalence between pancreatic head (38%) and pancreatic tail (34%). Prevalence of tail localization, however, may be even higher when cysts in the pancreatic body and tail are also taken into consideration.

Pancreatic cysts grow slowly (0.3-2 cm per year)[2], and some patients remain asymptomatic for years prior to obtaining a definitive diagnosis. Such patients are often incidentally diagnosed during tests performed for other indications. In symptomatic PHC patients, clinical presentation and complications depend on the location of the cyst within the pancreas[3-5,8,12,16,19]. Almost all cases we reviewed presented with epigastric pain, 20 had a palpable abdominal mass, and 15 had intermittent/permanent jaundice. Only a small proportion of patients suffered from non-specific symptoms such as fever, nausea-vomiting, weight loss, and abdominal fullness. PHC manifested itself as an intercostal herniation in only 1 patient[6]. However, we were unable to obtain precise information regarding duration of these symptoms.

Hydatid cysts located in the pancreatic head may cause obstructive jaundice or acute pancreatitis by either exerting external compression on or fistulizing into the common bile duct[4,10,12,15,20,23,26-30,31,35,36]. Less commonly, they may lead to cholangitis, duodenal stenosis, or duodenal fistula[4,20]. On occasion, hydatid cysts in the pancreatic head may remain silent but be palpable as an epigastric mass lesion. Hydatid cysts in the pancreatic body and tail usually remain asymptomatic until they grow large enough to compress adjacent organs or anatomical structures[4,8,16,20]. Gastric compression manifests as nausea, vomiting, abdominal pain, and early satiety[4,20]. On rare occasions, splenic vein compression may lead to splenic vein thrombosis with severe complications such as left-sided portal hypertension[4,12,32,34]. A hydatid cyst may also become infected, causing an abscess, or an acute abdomen due to spontaneous intraperitoneal rupture[4,20]. Hydatid cyst may also at times erode walls of gastrointestinal luminal organs, causing a rupture into the lumen[4,20].

In this review, we found that 14 cases had bile duct dilatation due to external compression[4,10,20,23,26-30,36], while 4 had cysto-biliary fistula[4], 6 had pancreatitis due to external compression and fistulization (2 were necrotizing, and 4 were edematous)[8,12,15,30,31,35], 3 had pancreatic ductal dilatation[10,15,36], 4 had cysto-pancreatic fistula[12,27,30,31], 2 had left-sided portal hypertension[4,32], 1 had cysto-duodenal fistula[4], and 1 had splenic vein obstruction[34]. Some of these complications were detected by preoperative radiological examinations, and others were only detected at the time of surgery.

Radiological and clinical properties of the cases in this review suggest that a significant portion were characterized by cyst-induced compression of or fistulization into the pancreato-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)[5,8,10,12,15,17,20,21,23,26,27,29,30,36].

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 diasnostic 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[26]. USG is a noninvasive, low-cost and sensitive diagnostic instrument. Gharbi defined the typical appearance of hydatid cysts in USG[12], 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 pancreato-biliary system, and presence of cysts in other organs. It is also successfully used for treatment monitorization and postoperative recurrence detection[12]. MRI and MRCP are particularly useful to delineate the relationship between cysts and pancreatic and bile ducts[12,31]. 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[10,27]. In MRI, superposition of the hydatid cyst with the pancreatic duct can be misinterpreted as a fistula[23]. 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[17,26]. It is also very beneficial in non-operative management of cases that developed biliary or pancreatic fistulae[12,30]. EUS is not commonly used[31,32], 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[12,31]. Cystography during surgery is especially helpful to demonstrate the relationship of the cyst with the pancreato-biliary ducts and gastrointestinal tract[23]. In complicated cases, the gall bladder and common bile duct can be entered with a needle and a cholangiogram can be taken[10,20,29], which may show both the anatomy of bile ducts and their relationship with cyst[12].

For diagnosis, screening, and recurrence monitoring, the following serological tests are used: enzyme-linked immunosorbent assay, indirect hemagglutination, serum immunelectrophoresis, complement fixation test, and immunofluorescence assay[17,21,28]. 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, gastroenteropancreatic neuroendocrine tumors, vasculary tumors, metastatic cystic lesions) or non-neoplastic (congenital pancreatic cysts, pseudocysts) cystic lesions[16,19,27]. 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[4,18,26]. Using percutaneous fine needle aspiration cytology (FNAC) for the differential diagnosis of cystic pancreatic lesion, Varshney *et al*[16] showed hooklets of hydatid cyst cytologically. In contrast, Dalal *et al*[22] 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[4].

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-reaspiration (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[4,29,31].

Pancreatic head cysts with no communication with biliary or pancreatic ducts can be managed with partial cystectomy + external drainage, partial cystectomy + omentopexy and pericystectomy, marsupialisation, and pancreaticoduodenectomy procedures[4,29,34]. 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 anas-

tomosis techniques[4,26]. In cysts located in the pancreatic body or tail, the most appropriate approach is a spleen-preserving distal pancreatectomy[4,26]. In cases where the spleen cannot be preserved, pneumococcal and meningo-coccal vaccinations should be done immediately to avert postsplenectomy complications[5]. 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*[18] 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[20]. Pancreaticoduodenectomy was performed in only 3 of 19 pancreatic head cysts[4,17,27]. 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, MRCP, 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 hydatic cysts[10,13]. 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[13,18,28]. 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[1]. 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[26]. In cases where minimally invasive surgical therapy has been contemplated, antihelminthic therapy should be administered before ($\geq$ 4 d) and after ($\geq$ 3-4 wk) the procedure in order to reduce intracystic pressure and prevent anaphylaxis[1].

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[15]. 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[32]. 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[19,21]. 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 infestation 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 choledocal 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 panceatic hydatid cyst remains challenging, and it is frequently misdiagnosed preoperatively as other cystic diseases of the pancreas and distal choledocal 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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