REVIEW ARTICLE

Primary Hydatid Cyst of the Pancreas: a Review

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ABSTRACT

Cystic Echinococcosis caused by adult or larval stages of Echinococcus granulosus. Primary pancreatic involvement (without liver or lung disease) is found in less than 1% of the various sites of hydatid disease. Endoscopic ultrasound provides more detailed morphological information as well as the opportunity for biochemical and cytological sampling which helps in differentiating hydatid cysts from other cystic pancreatic lesions.

Introduction

Cystic Echinococcosis is a near-cosmopolitan zoonosis caused by adult or larval stages of tiny cestode parasites belonging to the genus *Echinococcus* and the family *Taeniidae* [1, 2]. The genus *Echinococcus* consists of six species but four are of public health concern: *Echinococcus granulosus* (which gives rise to cystic hydatid disease), *Echinococcus multilocularis* (which causes alveolar echinococcosis), *Echinococcus oligarthus* (which cause polycystic echinococcosis). Recently two new species have been identified: *Echinococcus shiquicus* on the Tibetan plateau and *Echinococcus felidis* in African lions but to date no human infection have been described [2, 3, 4]. On a global basis, *Echinococcus granulosus* is the most common species and is responsible for 95% of the human cystic echinococcosis cases reported [5].

Infected Organs

The annual incidence of hydatid disease has been reported to be 18 to 20 cases per 100.000 inhabitants [6]. The liver and lungs act as the primary filtering beds with their vast capillary network and hence are the organs most frequently (90% of cases) involved in human echinococcosis. Primary pancreatic involvement (without liver or lung disease) is found in less than 1% of the various sites of hydatid disease, about 0.2 % of abdominal locations and less than 1% in those countries where the disease is endemic [7-12]. Pancreatic infestation is mainly by hematogenous dissemination, local spread via pancreatobiliary ducts, and peripancreatic lymphatic invasion [12-15]. Clinical presentation varies with size of the cyst and anatomic location. The hydatid cyst of the pancreas is mainly located in the head (57%), followed by the body (24-34%) and the tail (16-19%) [14]. The relatively higher frequency in the pancreatic head can be explained by the fact that the head region is the most vascularized [16].

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Clinical Symptoms and Complications

The clinical presentation of hydatid disease of the pancreas is variable depending on the size and anatomical location of the cyst [17]. An abdominal mass, epigastric pain, weight loss, discomfort and vomiting are the main clinical symptoms [18-20]. Cyst in head of pancreas can cause obstructive jaundice [14, 19-23]. Accordingly, two hypotheses are posited: main pancreatic duct compression caused by the cyst itself [24] and main pancreatic duct obstruction by hydatid scolices' migration from the hydatid cyst [25-28]. Also, cholangitis, duodenal stenosis or fistula, acute and chronic pancreatitis, pancreatic abscess and pancreatic fistula are unusual complications of hydatid cysts involving the head of the pancreas [29-33]. Cysts located in the body or tail of the pancreas can be symptomless and may be detected by the presence as an abdominal mass [34,35]. Mesenteric vein thrombosis, the compression of splenic vein causing portal hypertension, rupture into the biliary three or into the peritoneal cavity, gastrointestinal tract and abcess formation are uncommon presentations of cysts in the body and tail [21, 23, 36]. Increased bilirubin, ALP, ALT, AST, amylase and lipase have been reported in these patients [37-49].

Diagnosis- Imaging techniques

The preoperative diagnosis of hydatid cysts is very difficult. Establishing a precise diagnosis may be difficult because the presenting symptoms and the findings of clinical investigations may be similar to some other more commonly encountered cystic lesions of the pancreas [50]. Cystic lesions of the pancreas (CLP) are relatively common and differentiating among these cysts is challenging. CLPs include pseudocysts, retention cysts, duplication

cysts, lymphoepithelial cysts, congenital epithelial cysts, cysts which occur in association with polycystic disease or von Hippel-Lindau disease, serous cystic adenomas (SCA), mucinous cystic neoplasms (MCN), intraductal papillary mucinous tumors (IPMN), solid pseudopapillary tumors (SPN), cystic metastases from cancer of the lung, ovary, or melanoma, vascular tumours (haemangioma or lymphangioma) and primary hydatid disease of the pancreas (Table 1) [51-57].

Imaging modalities range from simple to complex and invasive. Ultrasound is readily available and cost effective. The diagnostic sensitivity of abdominal ultrasound in abdominal echinoccosis in general ranges from 93-98% [3, 58, 59], but it may be less accurate with pancreatic lesions, due to the difficulty in visualisation of the pancreas. Abdominal CT scan shows the presence of a cyst in the pancreas with no enhancement on contrast. The classical imaging features of hydatid cysts include a multivesicular cyst containing daughter cysts, 'hydatid sand' (layering of scolices in the cyst), 'water lily sign' (floating membrane within the cyst), Cyst wall calcification and hyper-intense cyst wall on plain CT [60]. Magnetic resonance imaging shows the characteristic low-signal intensity rim of hydatid cyst on T2-weighted images and is superior in demonstrating irregularities of the rim. These irregularities represent incipient detachment of the membranes [61]. Although the presence of cystic lesions of the pancreas are easily identified by ultrasound, CT scan and magnetic resonance imaging (MRI), these methods have limited sensitivity in making a specific diagnosis because of the considerable overlap of imaging features [62]. A specific diagnosis is seldom made preoperatively unless hydatid disease is suspected. Since its early introduction in the early 1990's, EUS has emerged as a safe and accurate technique for the diagnosis, stage, and treat a variety of lesions. A particularly useful aspect of EUS is the enhanced imaging of the pancreas [63].

Endoscopic ultrasound involves placing small, high-frequency ultrasound transducers on the tips of fiberoptic or video-endoscopes [64]. With several technological improvements, such as the introduction of electronic transducers and color-doppler capability, EUS is able to gain accurate images, with high negative predictive value, ranging from 87 to 100% [65, 66]. By placing the transducer within the gut lumen, EUS overcomes the two major technologic problems for pancreatic imaging by transcutaneous ultrasound; obscuring overlying gas filled bowel and the necessity to use low frequency and therefore low resolution ultrasound to penetrate to the depth of the pancreas [64].The strict proximity between the transducer and the lesions allows for a very precise definition of the structural component of the cysts and some components of pancreatic cysts, are better visualized with EUS than with other modalities [67]. Sensitivity of EUS in pancreatic mass detection can reach 97%, with a better yield than transabdominal US and conventional CT scan, mainly with small size lesions (<3 cm) [68]. EUS can accurately show the relationship between the cyst and pancreatic duct [69]. Endoscopic ultrasound provides more detailed morphological information as well as the opportunity for biochemical and cytological sampling which helps in differentiating hydatid cysts from other cystic pancreatic lesions. Aspiration of cyst fluid for (cytologic and/or chemical) analysis or biopsy of the cyst wall has been recommended as methods of distinguishing hydatid cysts of the pancreas from more commonly occurring pseudocysts or cystic tumours [70]. EUS now allows combining the biopsy (EUS-guided fine needle aspiration) and therapeutic capabilities of EUS (eg. tumor injection therapy or celiac neurolysis) with the initial diagnostic procedure [67]. The first EUS-FNA of a pancreatic lesion was reported in 1994 and there have been numerous series since then [71-77]. EUS-FNA techniques has been described extensively elsewhere [78-82] and involve passing an 18 to 25 (usually 22) gauge stainless steel, echogenic aspiration needle through the biopsy port of an echoendoscope under real-time guidance into an endosonographically visualized pancreatic lesion or fluid collection. The needle is moved back and forth through the lesion with varying degrees of suction applied to it and the sample is deposited on a cytology slide(s) for immediate staining and cytopathologic examination [64]. EUS-FNA is the most cost-effective modality as primary approach and it is able to change patient management as previously reported [83, 84]. Moreover, EUS-FNA has a lower risk of needle tract seeding when compared with a percutaneous approach due to a short needle track, which does not pass through peritoneal or pleural surfaces [85]. EUS-FNA can provide material for a cytologic diagnosis in up to 80% of cases of pancreatic cystic lesions [86-90]. Major complications related to EUS-FNA of pancreatic cystic lesions include acute pancreatitis (0.85%) [91,92], abdominal pain (0.56%) [91], hemorrhage within the cyst (<1%) [87, 88, 89], infection and fever (0.85%) [88-92]. The history of acute or chronic pancreatitis was not associated with higher risk [91]. Overall, the accuracy for diagnosing various cystic lesions by EUS-FNA is 54% to 97% [86-90, 93]. Finally, when EUS is combined with its sister endoscopic procedure, therapeutic ERCP, it results in a powerfully efficient combination of diagnostic, staging and therapeutic techniques that are very difficult to match with any other set of procedures [94]. However, EUS, although operator dependent, can be very useful, particularly when CT or RMN are equivocal [67] and due

Table 1.Differential diagnosis of pancreatic cysts.

Nonneoplastic lesions	Neoplastic lesions
Pseudocysts	IPMN
Syndromes causing multiple cysts	MCN
(i) Autosomal dominant polycystic disease	SCN
(ii) Cystic fibrosis	SPN
Infectious cysts	Cystic variants of solid tumors
(i) Hydatid cysts	(i) Cystic teratoma
(ii) Abscess	(ii) Cystic ductal adenocarcinoma
Lymphoepithelial cysts	(iii) Cystic neuroendocrine tumor
Congenital epithelial cysts	(iv) Cystic acinar cell carcinoma
Duplication cysts	(v) Cystic metastases
Retention cysts	

Table 2. Summary of demographic and clinic characteristics patients with hydatid cyst published in the medical literature between 1975 to 2014

Source	Year	Country	Sex	Age (yr)	Location	Size (mm)	Radiology
96	1975	Iraq	F	27	body and tail	7.6 x 10.2 cm	x-ray examination
			M	45	head	...	x-ray examination
97	1997	India	F	22	head	4.3x4.6 cm	USG+CT+ERCP
98	1997	Argentina	F	50	head	6 cm	CT+ERCP
99	2000	Italian	F	28	body and tail	10 x 10 cm	USG, angio-CT and angiography
100	2000	UK	M	20	head	6.37 cm	USG+CT+ERCP
101	2004	French	F	35	body and tail	6xx7 cm	USG+CT
102	2004	Spain	F	34	tail	...	CT
103	2004	Turkey	F	38	body	90x60x70 mm	USG+CT
22	2005	South Africa	F	60	head	10 cm	ERCP
			F	17	head	3 cm	ERCP+CT
			F	52	head	4 cm	CT
			M	17	head	9 cm	CT
104	2005	Turkey	F	18	head	43x35 mm	USG
105	2005	India	M	42	tail	...	USG+CT
			F	28	body and tail	...	USG+CT
106	2005	South Africa	F	11	head	4×2 cm	USG+CECT
107	2006	Turkey	M	21	head	6 x 5 x 5 cm	USG+CT+ERCP
20	2007	Iran	M	30	head	6x8 cm	USG+CT
108	2007	Morocco	M	26	head		USG+CT+MRI
109	2008	Iraq	F	35	body	4x3 cm	USG+CT
110	2009	India	M	21		35x20.8x10.5 cms	USG+CT
111	2009	Jordan	M	33	head	7.3 x 7.2 cm	USG+CT
112	2009	Pakistan	M	32	head	approximately 15 cm	USG
113	2010	Kingdom of Saudi Arabia	M	45	tail	5x6 cm	USG+CT
114	2010	India	F	30	head	8 x 6 cm	USG+MRCP+EUS
115	2010	India	M	46	tail	28 mm	USG+CT
			F	37	body	26 mm	USG+CT
			M	18	body	33 mm	USG+CT
			F	22	tail	48 mm	USG+CT
			M	28	head	50 mm	USG+CT
			M	68	head	35 mm	USG+CT
116	2011	India	M	20	throughout the pancreas	8 cm	USG + CT
117	2011	Turkey	F	7	body and tail	70x55x60 mm	USG+CT
118	2011	India	F	4	head and body	10×15×7 cm	x ray+USG+CT
119	2011	India	M	48	tail	8x5 cm	USG+ CECT
120	2011	India	F	5	head	12 cm × 10 cm	USG +MRCP
19	2012	Tunisia	M	38	Body	100 mm	CT
121	2012	India	F	30	tail	6.2 × 5.7 × 4.5 cm	USG+CECT
122	2012	Turkey	M	33	neck	55 x 44 x 45 mm	USG+CT
123	2012	India	M	6	head	...	USG + MRCP Enucleation + cholangiography
124	2013	Iran	M	46	tail +right kidney	6 cm	USG+CT
17	2013	Tunisia	F	21	tail	...	USG and CT
			M	13	tail and body	...	USG and CT
			M	15	head	...	USG and CT
			M	26	head	...	USG and CT
			F	50	head	...	USG and CT
			F	37	head	...	USG and CT
			M	8	head	...	USG and CT
			F	26	tail and body	...	USG and CT
			F	61	tail	...	USG and CT
			F	11	tail	...	USG and CT
			F	16	body	...	USG and CT
			F	11	head	...	USG and CT
125	2013	Turkey	F	9	head	4 x 5 cm	USG+ MRCP

126	2013	India	M	47	body and tail	10 x 8 x 5 cm	USG+CT
127	2013	India	M	26	body and tail	8.9x6.6 cms	USG+CT
128	2013	India	F	18	tail	65 x 63 mm	USG+CT
129	2013	India	M	43	tail	18 cm × 17 × 15 cm	USG+CT
130	2014	India	F	59	tail	12cm×11cm	CT+EUS
131	2014	India	F	35	body and tail	5.5 × 7.6 × 6.5 cm	USG+CECT
132	2014	Iran	F	33	tail	11 x 7 x 3 cm	USG+CT
5	2014	Turkey	F	48	head	5 cm × 5 cm	USG+MRCP+ERCP

to these advantages, EUS has evolved into an important technique to assess cystic lesions of the pancreas [95].

Conclusion

Pancreatic hydatid cyst is a parasitic infestation that is rare with a reported frequency of 0.1-2% of all cases of hydatid disease. Anatomy of the pancreas may cause difficulties in making the diagnosis. Preoperative diagnosis of pancreatic hydatid cysts is challenging, as its radiologic findings are often mistaken for other cystic lesions of the pancreas. Endoscopic ultrasound is useful in the diagnosis of pancreatic hydatid disease. EUS with its capabilities for fluid sampling for biochemistry and cytology is a very useful discriminates.

Conflicting Interest

The authors had no conflicts of interest

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