

Hydatid Cyst of the Pancreas: A Report on Ten Cases

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ABSTRACT

Objective Hydatidosis pancreatic site remains outstanding even in countries with endemic hydatid. The diagnosis can be difficult, confusing with other cystic lesions of the pancreas. The aim of this study was to describe clinical, radiological and therapeutic aspects of hydatid cysts of the pancreas. **Methods** This study was carried out in the Department of Digestive and General Surgery in Sahloul University Hospital, Sousse in Tunisia during the period 1993–2013. Ten patients were treated for hydatid cyst of the pancreas, four men and six women with an average age of 31 years. **Results** The clinical signs were variable according to size and location of the cyst within the pancreas and the degree of biliopancreatic involvement. The main symptoms were pain in left upper quadrant, jaundice, fever and epigastric and right upper quadrant pain. The preoperative diagnosis was established in six patients by ultrasonography and computed tomography. However, the diagnosis was established during laparotomy in two cases. The echinococcal immunological test was positive in only two cases. Surgical treatment included resection of the prominent lump (cystectomy) in five cases, total cystectomy in three cases and distal pancreatectomy with splenic preservation in one case and with splenectomy in one case. The postoperative period was uneventful in six cases. Acute postoperative pancreatitis occurred in one case. The patients were free of symptoms and were followed up for 24 months without any recurrence as revealed by abdominal ultrasonography and computed tomography scan. **Conclusions** Hydatid cysts in the pancreas are rare. Even if rare, the diagnosis should be considered in any pancreatic cystic mass in endemic countries like ours. Diagnosis is difficult when it is a unique and isolated location, may even pose therapeutic problems. The surgical treatment is to be considered whenever possible.

INTRODUCTION

Echinococcosis, an endemic disease in the Tunisia is caused by a tapeworm, the lifecycle of which has man as an accidental intermediate host. Cysts occur in any tissue – the liver (70%), lung (10%), muscles (4.7%), spleen (2.1%) and brain (1.4%) are most commonly affected, with rare involvement of the bone, thyroid, breast and pancreas [1]. Hydatid cyst of the pancreas is rare since it accounts for less than 1% of that of various other sites, even in countries where it is endemic [2, 3]. The diagnosis is rarely made before surgery but is currently facilitated by ultrasonography and computerized tomography (CT) scan. Herein we present a series of ten cases of hydatid cyst of the pancreas.

PATIENTS AND METHODS

Ten patients were diagnosed in the Department of

General Surgery in Sahloul University Hospital in Tunisia as hydatid cyst of the pancreas during the period 1993–2013.

RESULTS

The ten cases consisted of four men and six women with an average age of 31 years ranging from 19 to 42 years. Among the ten patients, six presented with pain in the left upper quadrant, three for jaundice, fever and pain in epigastrium and right upper quadrant (**Table 1**).

Physical examination showed abdominal mass in three cases (epigastric region: one patient, left upper quadrant: two patients). Laboratory tests revealed a high serum bilirubin and alkaline phosphatase level in three cases (cases 5,6 and 8). Hydatid serology performed by ELISA was positive in only two cases. Complete blood count revealed eosinophilia in only two cases.

Abdominal ultrasonography and CT scan performed in all cases revealed cystic tumour of the pancreatic head in three cases (cases 5,6 and 8) the body in three cases (case 3,9 and 10) and the tail in four cases (cases 1, 2, 4 and 7) (**Figure 1**). The ultrasound classification of the cyst according to Gharbi showed a type III cyst in five cases. The hepatic duct was compressed by the cyst resulting in dilatation of the intrahepatic bile ducts in three cases (cases 5,6 and 8). The cysts were associated with hydatid

Received June 22nd, 2015-Accepted July 25th, 2015

Key words Pancreas; surgery

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Table 1. Summary of data of the ten cases of hydatid cyst of pancreas.

	Case #1	Case #2	Case #3	Case #4	Case #5	Case #6	Case #7	Case#8	Case #9	Case #10
Age	25	19	32	41	38	29	25	42	24	35
Gender	Female	Female	Female	Male	Male	Male	Female	Female	Female	Male
Clinical symptoms	Pain in left upper quadrant	Pain in right upper quadrant	Epigastric pain	Acute abdominal pain in left upper quadrant fever 38.3 C	Epigastric pain jaundice	Epigastric pain jaundice	Pain in left upper quadrant vomiting	Epigastric pain, jaundice	Epigastric pain	Epigastric pain
Physical examination	Left upper quadrant mass 6 cm	Hepatomegaly epigastric mass 7 cm	Normal	Mass in left upper quadrant 10 cm	Jaundice tenderness in right upper quadrant	Hepatomegaly epigastric mass 7 cm	normal	normal	normal	normal
Hydatid serology (ELISA)	Negative	Negative	Positive	Negative	Negative	Negative	Positive	-	Negative	Negative
Ultrasonography CT scan	Cystic mass in tail pancreas 6 cm type III (Gharbi)*	Multiple hydatid cyst liver (13) cystic mass tail pancreas 7 cm type III (Gharbi)	Two cystic masses in body and tail pancreas 15 cm type III (Gharbi)	Heterogeneous cystic mass 15 cm	Multiple hydatid cyst liver cystic mass in head pancreas 5 cm type III (Gharbi) dilatation of common bile duct 10 mm.	Important dilatation of common bile duct and cystic mass in head of pancreas	Cystic mass in body and tail pancreas 9 cm type III (Gharbi)	Cystic mass in head of pancreas 6 cm and dilatation of common bile duct	Cystic mass in body 8 cm type II(Gharbi)	Cystic mass in body 5 cm type III (Gharbi)
MRI	None	None	None	None	None	Dilatation of biliary ducts and cystic mass in head of pancreas 6 cm	None	None	None	None
Preoperative diagnosis	Hydatid cyst pancreas	Hydatid cyst liver and tail pancreas	Hydatid cyst pancreas	Infected cyst of pancreas 15 cm	Hydatid cyst liver and head of pancreas	Cystic dilatation of bile duct	Hydatid cyst pancreas	Hydatid cyst pancreas	Hydatid cyst pancreas	Hydatid cyst pancreas
Surgical treatment	Cystectomy no duct communication	Cystectomy for all liver cyst and pancreas	Cystectomy for both cyst	Cystectomy splenectomy no duct communication	Cystectomy for all hepatic cysts and cholecystectomy compression of cbd by hydatid cyst of pancreas. cystectomy no duct communication	Cystectomy of head cyst of the pancreas no duct communication	Distal pancreatectomy splenectomy	Total cystectomy No duct communication	Total cystectomy No duct communication	Total cystectomy No duct communication
Postoperative complication	None	None	None	None	Acute pancreatitis	None	None	none	None	None
Follow-up	35 months Ultrasonography shows no recurrence	9 months CT scan shows no recurrence	6 months Ultrasonography shows no recurrence	9 months Ultrasonography shows no recurrence	14 months Ultrasonography and CT scan normal	16 months CT scan shows no recurrence	2 months CT scan shows no recurrence	15 months CT scan shows no recurrence	16 months Ultrasonography shows no recurrence	18 months CT scan shows no recurrence

* Gharbi classification: Type I: pure fluid collection – univesicular cyst. Type II: fluid collection with a split wall – detached laminated membrane – ‘water lily’ sign. Type III: fluid collection with septa – daughter cysts. Type IV: heterogeneous appearance – presence of matrix – mimics a solid mass. Type V: reflecting thick walls – calcifications.

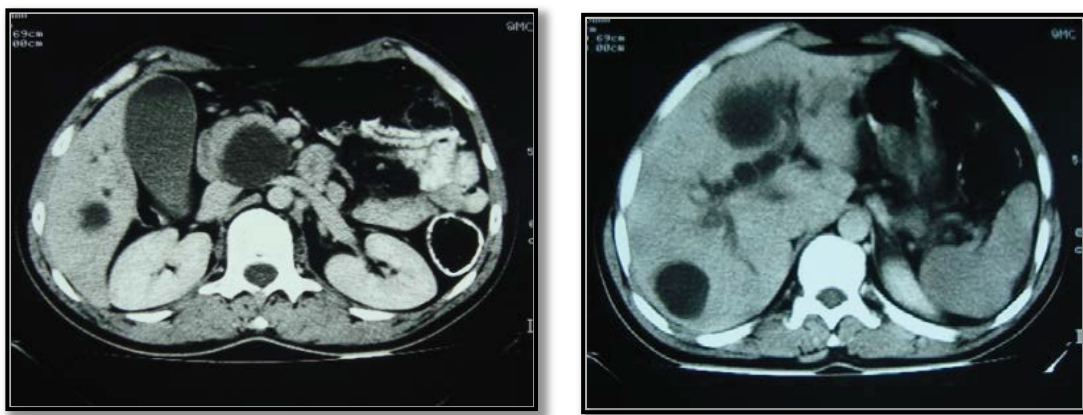


Figure 1. Abdominal CT showing hydatid cyst liver and head of pancreas (case #5).

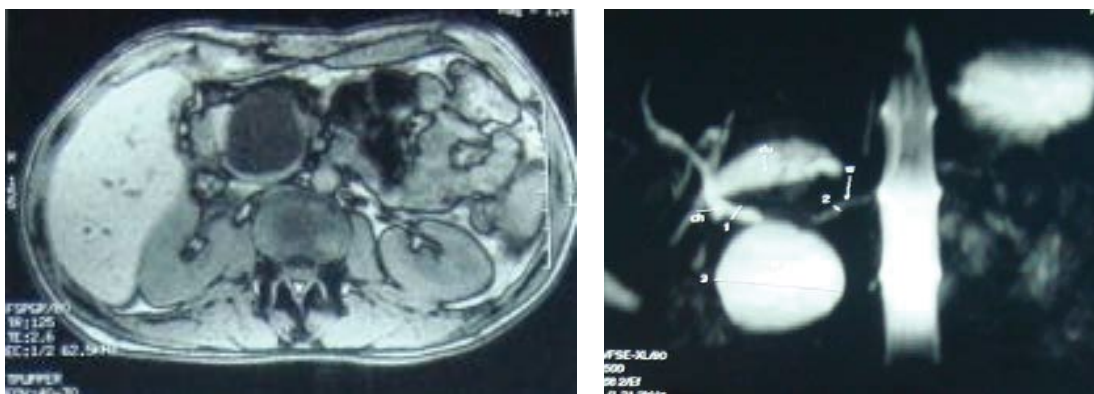


Figure 2. Abdominal MRI showing Dilatation of biliary ducts and cystic mass in head of pancreas 6 cm simulating a cystic dilatation of bile duct (case #6).

cyst of the liver in two cases (cases 2 and 5).

The MRI, performed in one case (case 6), showed a cystic mass of the head of the pancreas. The preoperative diagnosis was a congenital dilatation of the common bile duct (**Figure 2**). The endoscopic ultrasound was not performed due to its unavailability in our centre. The size of the pancreatic masses ranged from 7 to 15 cm in diameter. Laparotomy was performed in all cases. Partial cystectomy was performed in five cases and no communication was found between the cyst and the pancreatic or the biliary ducts (**Figure 3**) Distal pancreatectomy was performed in two cases associated with splenectomy in one case (**Figure 4**), total cystectomy was performed in three cases. The postoperative period was uneventful in nine cases. However, a postoperative pancreatitis occurred in one case (case 5). Patients were free of symptoms and were followed up for 24 months without any recurrence at abdominal ultrasonography and CT scan.

DISCUSSION

Hydatid disease, which may have a prevalence exceeding 5% of the general population in endemic countries, affects mainly the liver and the lungs. Pancreatic localization is exceptional with a prevalence of about 0.2%.

Infestation of the pancreas occurs by arterial route after passage of the hepatic and pulmonary filters. Pancreatic hydatid cyst is isolated in 91% of the cases. The localization may be in the head of pancreas (57%), body (24%) and tail (19%). The relatively higher frequency in the pancreatic

head can be explained by the fact that the head region is the most vascularized [3–9]. In our study, the body and tail were the most frequent localizations of the disease (70%).

The cyst size is very variable, ranging from some millimetres up to three litres. The localization within the pancreas is more often peripheral (two-thirds of the cases) than central.

The clinical presentation of pancreatic cysts depends on their size and anatomic locations. Pancreatic hydatid cysts located in the head may present with obstructive jaundice due to either compression of the common bile duct or fistulization and spillage of scolices into the biliary tree [10–18]. In some cases, continuous undetected passage of scolices into the biliary tree may lead to recurrent attacks of acute pancreatitis. Cysts located in the body and tail of the pancreas have a less specific clinical presentation which includes abdominal pain, discomfort, vomiting, fullness and occasionally early satiety because of compression of the stomach by large cysts in the lesser sac. It is suspected that these cysts have a long preclinical latency period during which clinical manifestations of the disease are sparse until the size of the cyst results in clinical symptoms due to compression of adjacent organs. Many such cysts have been reported to present as an asymptomatic abdominal mass that is detected on physical examination. However, the compression of the splenic vein by the cyst can lead in 14% of the cases to a partial portal hypertension that was the case in our patient (case 4). Some other complications are rare as thrombosis of the superior mesenteric artery and subsequent intestinal infarction. Rarely, cysts can

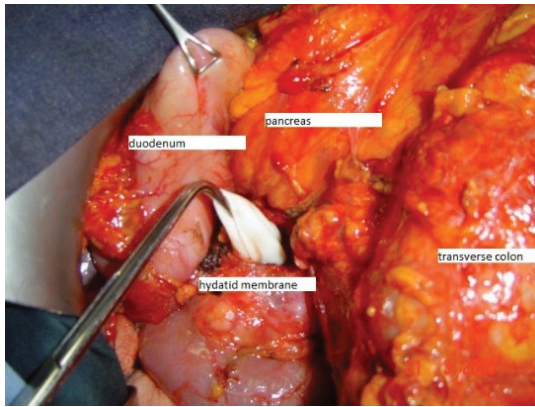


Figure 3. Hydatid cyst of the head of pancreas (case #5).

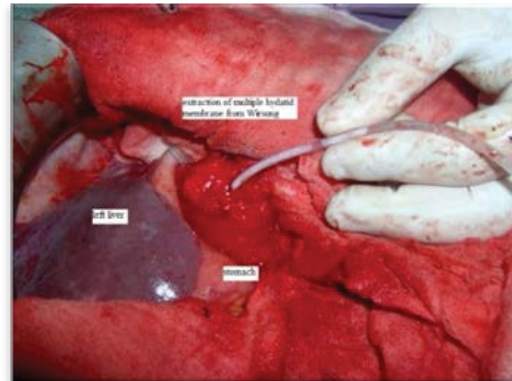
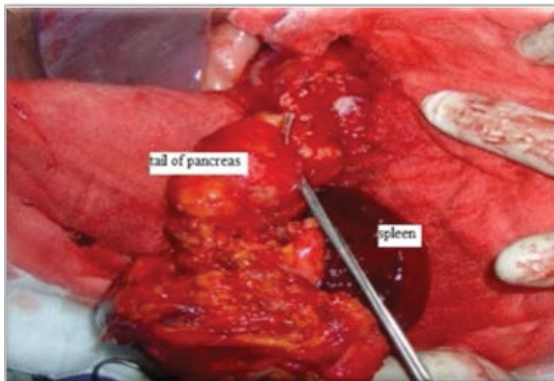
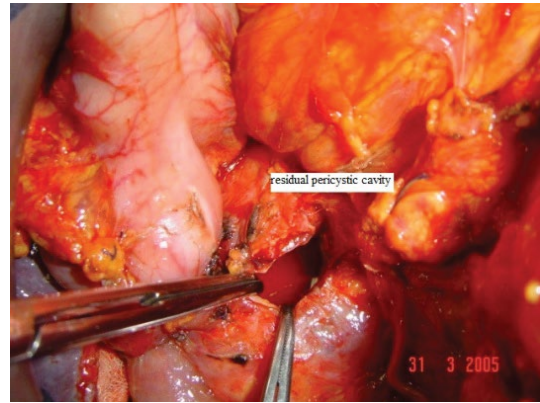


Figure 4. Distal pancreatectomy with splenectomy for hydatid cyst of tail (case #7).

communicate with the Wirsung duct leading to acute pancreatitis or wirsungorrhagia. [3-5, 12, 13].

The rarity of pancreatic hydatid cysts makes the proper preoperative diagnosis difficult. It is important to consider hydatid cyst in the differential diagnosis of pancreatic and parapancreatic cystic lesions. This is particularly true for patients living in regions where the disease is endemic, but it may also be encountered in immigrants from endemic areas to developed countries where the disease may not be prevalent or may not exist. Establishing a precise diagnosis may be difficult because the current symptoms and the result of investigations may be similar to other more commonly encountered cystic lesions of the pancreas.

The peripheral eosinophilia and hydatid serology are not positive in all cases. In our study, hydatid serology was positive in only two cases and eosinophilia was present in also two cases.

The diagnosis of pancreatic cystic lesion can be performed by ultrasonography, CT scan, MRI and endoscopic ultrasound but the difficulty is to link these lesions to the possibility of hydatid disease as a diagnosis. The radiological imaging features that are useful for distinguishing hydatid cyst from other cystic lesions are the presence of curvilinear calcification in the wall of the cyst (12%), the presence of daughter cysts, or the presence of debris known as hydatid sand, septations, or membrane detachment [3, 10, 12, 19].

Even with these fairly specific characteristics, hydatid cysts with unusual localizations (such as the pancreas)

may present true diagnostic challenges. In fact, it is often difficult to differentiate these from pancreatic pseudocysts or cystic neoplasms.

In our case 6, CT scan and MRI failed to provide a diagnosis. In such difficult situations, hydatid serology (ELISA) could be helpful in establishing the diagnosis of cystic lesion of the pancreas when positive, however, with a negative serology the hydatid disease cannot be excluded.

Aspiration of cyst fluid for analysis or biopsy of the cyst wall has been recommended as methods of distinguishing hydatid cysts of the pancreas from pseudocysts or cystic tumours. This technique can be performed percutaneously or using endoscopic ultrasound. However, if the lesion is malignant, percutaneous CT or ultrasound-guided needle aspiration carries the potential risk of needle tract or peritoneal dissemination of viable parasitic or neoplastic cells as well as spillage of malignant cyst contents [19].

The indication of invasive investigations such as the ERCP and the retrograde wirsungography is limited to complicated cases where these investigations could be used as therapy.

Percutaneous puncture does not have its place as a diagnostic tool as it can lead to complications such as pancreatitis, haemorrhage, peritoneal dissemination, cyst infection and peritoneal rupture causing shock in case of hydatid cyst. Some infectious complications have been reported after fine needle aspiration puncture of a hydatid cyst of the pancreas [5] and the authors think that this technique should be avoided if the diagnosis of hydatid cyst is suspected.

Surgery remains the treatment of choice in hydatid disease. Many surgical procedures are available to remove the cyst. Partial or total cystectomy, marsupialization and external drainage have also been reported in the management of pancreatic hydatid cysts [2, 7, 16, 19]. The choice of the surgical procedure will depend on the localization of the cyst and the existence of complication such as communication with the pancreatic and biliary ducts [4, 5, 15, 17, 19]. The main goals of surgical treatment are total removal of all parasitic elements, avoidance of spillage of the contents of the cyst and management of the residual pericystic cavity with maximum conservation of the host organ. These could be managed either by laparoscopy or by laparotomy. Because of the delicate nature of the laparoscopic procedure and the importance of avoiding spillage of the cyst's contents, open resection has been the standard of care for the treatment of pancreatic echinococcosis.

Because of localization of the cyst near the biliary or pancreatic duct (or vascular structures) and a high risk of pancreatic fistula, a total pericystectomy is not possible in all cases and has to be avoided. Therefore, partial cystectomy is preferred in such cases [4, 15, 17].

If there is a communication with the pancreatic duct, the surgical procedure will depend on the localization of the cyst:

- Distal pancreatectomy is the treatment of choice for the hydatid cyst localized in the tail. This was the case in one of our patients.
- If the cyst is localized in the head of pancreas, a cystojejunal anastomosis can be performed to prevent a postoperative pancreatic fistula [18, 19]. However, this procedure should be avoided with friable pancreatic parenchyma. In this case, some other procedures can be used such as suturing of the fistula using a drain. Pancreatic head resection seems to be not justifiable for this benign parasitic affection.

Even if surgery remains the main therapeutic method of hydatid disease, new interventional techniques are also being used.

Ultrasound guided percutaneous aspiration, injection and reaspiration puncture (PAIR) of the cyst has been used for treatment of hydatid cyst especially when surgery is contraindicated. Multiple trials confirmed that PAIR is appropriate for the treatment of liver, abdominal cavity and renal hydatid cysts. The technique is performed using ultrasound or CT guidance to facilitate aspiration of the cyst fluid. PAIR should not be used in the lung, spine, and brain [20]. Concerning hydatid cyst of the pancreas, this technique is advised against because of the risk of complications [5].

CONCLUSION

We can conclude that pancreatic hydatid disease, even if it is very rare, should be considered in the differential

diagnosis of cystic lesions of the pancreas. Surgical treatment is to be considered whenever it is possible. The conservative treatment (partial or total pericystectomy) is the procedure of choice and resection should be reserved for selective cases.

Conflicts of Interest

The authors declared that there was no conflict of interest.

References

1. Moosavi SR, Kermany HK. Epigastric mass due to a hydatid cyst of the pancreas. A case report and review of the literature. *JOP* 2007; 8:232-4. [PMID: 17356249]
2. Wani NA, Shah OJ, Zargar JI, et al. Hydatid cyst of the pancreas. *Dig Surg* 2000; 17:188-90. [PMID: 10781989]
3. Polat P, Kantarci M, Alper F, et al. Hydatid disease from head to toe. *Radiographics* 2003; 23:475-94. quiz 536-7. [PMID: 12640161]
4. Fadil A, Bolbarod AA, el Fares F. Kyste hydatique du pancréas. A propos d'une observation. [Hydatid cyst of the pancreas. Report of a case]. *Ann Chir* 2000; 125:173-5. [PMID: 10998804]
5. Faucompret S, Farthouat P, Sainton T, et al. Kyste hydatique du pancréas compliqué après cytoponction. [Complicated hydatid cyst of the pancreas after needle biopsy]. *Ann Chir* 2001; 126:491-2. [PMID: 11447807]
6. Errougani A, Chkoff R, Koutani AA, et al. Le kyste hydatique du pancréas. A propos d'une observation. *Méd Maghreb* 1991; 30:27-30.
7. Rey P, Debonne JM, Klotz F. Parasitic pancreas [Pancréas parasitaire]. *EMC Hepato-Gastroenterol* 2005; 2:49-58.
8. Wani RA, Malik AA, Chowdri NA, et al. Primary extrahepatic abdominal hydatidosis. *Int J Surg* 2005; 3:125-7. [PMID: 17462273]
9. Erdener A, Sahin AH, Ozcan C. Primary pancreatic hydatid disease in a child: case report and review of the literature. *J Pediatr Surg* 1999; 34:491-2. [PMID: 10211664]
10. Dahniya MH, Hanna RM, Ashebu S, et al. The imaging appearances of hydatid disease at some unusual sites. *Br J Radiol* 2001; 74:283-9. [PMID: 11338110]
11. Gayral F, Bouree P, Jourdanne P, et al. Kyste hydatique du pancréas. Une observation. [Hydatid cyst of the pancreas. One case (author's transl)]. *Nouv Presse Med* 1981; 10:3787-8. [PMID: 7322913]
12. Khiari A, Mzali R, Ouali M, et al. Kyste hydatique du pancréas. A propos de sept observations. [Hydatid cyst of the pancreas. Apropos of 7 cases]. *Ann Gastroenterol Hepatol (Paris)* 1994; 30:87-91. [PMID: 8067682]
13. Ozmen MM, Moran M, Karakahya M, et al. Recurrent acute pancreatitis due to a hydatid cyst of the pancreatic head: a case report and review of the literature. *JOP* 2005; 6:354-8. [PMID: 16006687]
14. Miguel EE, Amondarain Arratibel JA. Hydatid disease of the pancreas. *JOP* 2004; 5:51-2. [PMID: 14730124]
15. Oruc MT, Kulacoglu IH, Hatipoglu S, et al. Primary hydatid cyst of the pancreas related to main pancreatic duct. A case report. *Hepatogastroenterology* 2002; 49:383-4. [PMID: 11995456]
16. Khiari A, Mseddi MA, Hadiji S, et al. Kyste hydatique de la tête du pancréas fistulisé dans le duodénum. A propos d'un cas. [A hydatid cyst of the head of the pancreas causing a fistula into the duodenum. apropos of a case]. *Ann Gastroenterol Hepatol (Paris)* 1994; 30:57-9. [PMID: 8210204]
17. Ousadden A, Elboughaddouti H, Ibnmajdoub KH, et al. Primary hydatid cyst of the pancreas with a hepatic pedicle compression. *Cases J* 2009; 2:201. [PMID: 20062706]
18. Boubbou M, Boujraf S, Sqalli NH, et al. Large pancreatic hydatid cyst presenting with obstructive jaundice. *Arab J Gastroenterol* 2010; 11:47-9.

19. Krige JE, Mirza K, Bornman PC, et al. Primary hydatid cysts of the pancreas. *S Afr J Surg* 2005; 43:37-40. [PMID: 16035381]

20. Yattoo GN, Khuroo MS, Zargar SA, et al. Case report: percutaneous drainage of the pancreatic head hydatid cyst with obstructive jaundice. *J Gastroenterol Hepatol* 1999; 14:931-4. [PMID: 10535478]
