



Primary Pancreatic Echinococcosis: A Rare Cause of Pancreatic Cyst

Rahul Khullar¹, Hirdaya Nag^{1*} and Sundeep Saluja¹

¹*Department of GI Surgery, GB Pant Institute of Postgraduate Medical Education and Research (GIPMER), New Delhi, India.*

Authors' contributions

This work was carried out in collaboration among all authors. Author RK designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors HN and SS managed the analyses of the study. Author RK managed the literature searches. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Introduction: Primary Hydatid cyst of pancreas is a rare entity even in endemic countries. These cases usually present with non specific symptoms such as pain abdomen, vomiting or feeling of vague lump in abdomen.

Presentation of Case: A 55 year old lady presented to GI surgery department, GB Pant institute of Postgraduate Medical Education and Research (GIPMER), India with complaints of lump in abdomen and pain abdomen which was evaluated with imaging studies (Computed tomography scan Abdomen and Magnetic resonance imaging Abdomen). Initially considered as cystic lesion pancreas, Endoscopic ultrasound (EUS) confirmed diagnosis of Hydatid cyst and patient underwent open deroofting and drainage of cyst with uneventful post operative period.

Keywords: *Abdominal lump; pancreatic echinococcosis; magnetic resonance; hydatid cyst; endoscopic ultrasound.*

*Corresponding author: Email: hirdayanag@gmail.com;

1. INTRODUCTION

Human echinococcosis is a zoonotic infection caused by *Echinococcus granulosus* belonging to taeniidae family. Most common site of Hydatid cyst disease in humans is Liver (50–70%), lungs (20–30%), and less frequently the spleen, kidneys, heart, bones, central nervous system, and other organs [1]. It is endemic in Mediterranean countries, the Middle East, South America, and the Indian subcontinent [2]. Primary pancreatic hydatid cysts are most often confused with cystic lesions of the pancreas and pancreatic pseudocyst. Primary hydatid cyst of pancreas is rare. During the last 30 years, less than 40 cases have been reported in journals on Medline. The reported incidence of hydatid cyst of pancreas is 0.25%. The incidence varies from 0.14% to 2.0% in endemic countries [3]. The prevalence of Pancreatic cystic echinococcosis is low – 0.2 – 0.6% [4]. The Tail of pancreas is most frequent location 47% followed by the head 31% than the tail 22% [5]. Anatomic location of the cyst contributes to clinical symptoms in patients. The main symptoms are pain epigastrium (69%) or in left upper quadrant (31%), vomiting 16%, Jaundice 26%, and Fever in 8%. Association with acute pancreatitis was seen in 15% patients [4]. Enzyme-linked immunoadsorbent assay test for echinococcal antigens is positive in over 85% of infected patients [6]. Magnetic resonance imaging helps in characterizing hydatidosis. Also it can detect complications, such as infection, ductal communication and can reveal the exact extent of the disease [7]. Other useful imaging modalities are Endoscopic ultrasound or Computed tomography scan. After confirmation of diagnosis, Active Hydatid cyst should be treated to prevent its rupture and other associated complications. Various surgical approaches such as cyst excision, pericysectomy and omentoplasty are well established. Cysts located near tail of pancreas are also suited for Distal pancreatectomy [8].

2. CASE PRESENTATION

A 55 year old lady presented to out patient department with swelling in left upper abdomen for past 15 years which was insidious in onset, gradually progressive in size, not associated with pain in abdomen. However for past 6 months, she started complaining of pain in left upper and central abdomen which is dull aching type of pain, radiating to back, no aggravating factors and relieved on oral analgesics. There is no

history of Jaundice, Weight loss, Anorexia, bowel disturbances or severe attack of pain suggestive of Acute Pancreatitis. She had no medical comorbidities or history of previous surgeries or trauma. History of exposure to dogs and live stock present. On examination, Her BP was 124/82 mm Hg, pulse 72 beats/min, no icterus, Eastern Cooperative Oncology Group (ECOG) status 0. Per Abdomen examination revealed 10 x 8 cm lump occupying left upper quadrant of abdomen, smooth surface, margins not well defined, horizontal mobility present but no movement on respiration.

3. INVESTIGATIONS

Laboratory tests including Complete blood count, Liver function test and Kidney Function tests were within normal range. Serum Amylase and Lipase levels were 70 and 28 IU/L respectively. Abdominal Ultrasonography revealed Large cystic well defined Space occupying lesion (SOL) measuring 92.3 x 67.4 mm in epigastrium. Liver, Pancreas and spleen were reported as normal. Contrast Enhanced Computed Tomography (CECT) Abdomen revealed well circumscribed multiseptated, non enhancing intraparenchymal cystic SOL 88 x 66 mm in size involving pancreatic tail. No soft tissue component. Provisional diagnosis of Mucinous cystic neoplasm or pancreatic pseudocyst was given.

Patient was planned for Endoscopic Ultrasound which helped in quenching diagnosis. It showed large 7.6 cm x 6.9 sized cystic lesion in tail of pancreas with internal membranes suggestive of Hydatid cyst. Main pancreatic duct normal. ELISA for Hydatid serology was done which was positive. Provisional Diagnosis of Primary pancreatic hydatid cyst was made. Magnetic Resonance Imaging (MRI) Abdomen was done to rule out communication with pancreatic ductal system and surgical planning. Liver and biliary channels were normal, Well defined predominantly cystic lesion seen in relation to pancreatic tail and extending to splenic hilum. No pancreatic duct communication or compression seen. The lesion showed Floating membranes and Daughter cyst.

Patient was started on Anti helmenthic drug Albendazole pre operatively. Patient was taken up for surgery. Midline incision was given. 12 x 10 cm cystic structure was found originating from tail of pancreas pushing the stomach forward. Spleen was free from cystic lump. Left gastroepiploic vein was divided to free attachments surrounding lump. Abdominal cavity

was packed with Hypertonic saline soaked Mops to prevent free spillage into peritoneal cavity. Complete aspiration of cyst was done which showed clear fluid. There was no spillage from cyst. Cyst was opened, large single daughter cyst was present which was again aspirated and opened. Germinal membrane was found which

was extracted carefully and sent for Histopathological examination. Cyst was injected with scolicidal agent 3% Hypertonic saline and kept for 10 minutes. Partial pericystectomy was done. Marsupialization of edges was done. Finally cyst cavity was occluded by omentoplasty. There was no communication with pancreatic duct.

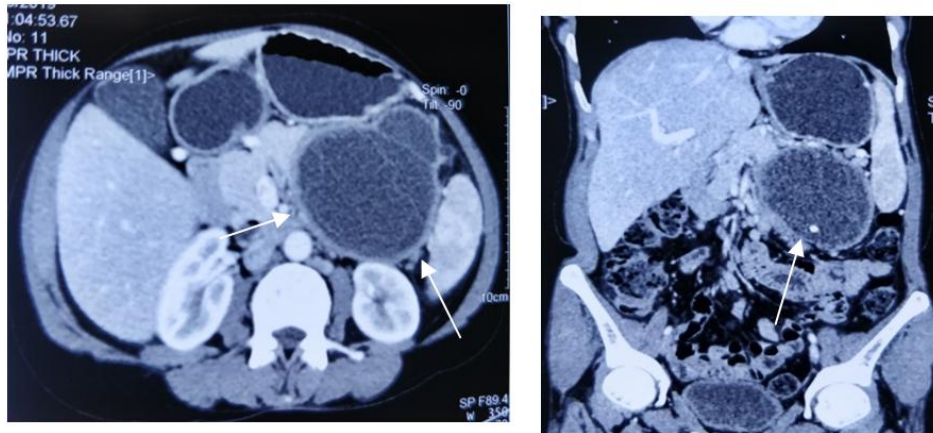


Fig. 1. CT images showing cystic lesion in tail of pancreas

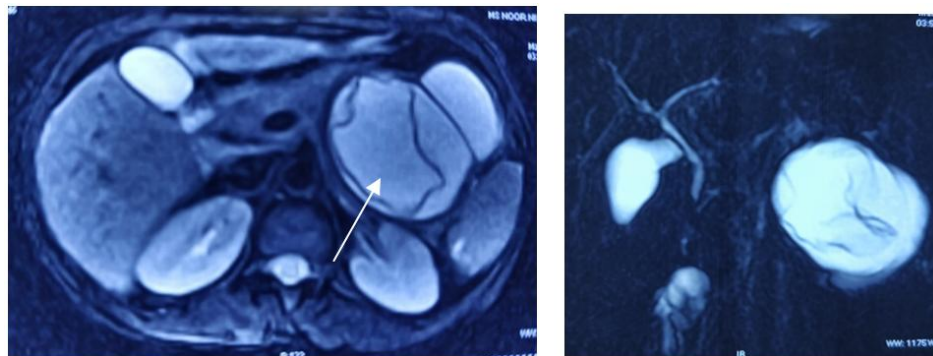


Fig. 2. MR images showing cystic lesion in tail of pancreas alongwith germinal membrane and daughter cyst inside cystic lesion. Arrow indicating germinal membrane and daughter cyst

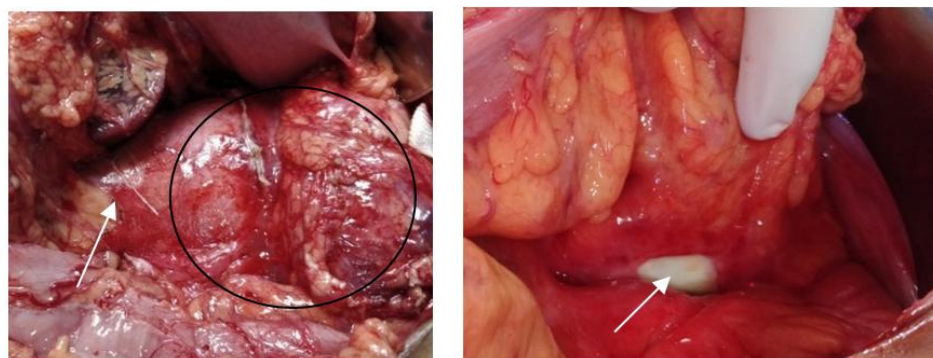


Fig. 3. Clinical picture of cystic lesion in tail of pancreas. a) Arrow showing normal pancreatic parenchyma. Oval figure lump. B) Impression of cyst through transverse mesocolon



Fig. 4. a) Single large daughter cyst visible after excision of wall of cyst. b) membrane removed from cystic lesion

Drain was not inserted. Post operative histopathological analysis confirmed Hydatid cyst. Postoperative day (POD) 1 patient was orally allowed and discharged on POD4 with no complications in post operative and follow up period. Patient was discharged on oral Albendazole tablets (10 mg/kg/day) for 8 weeks. After 2 months of follow up, patient had no symptoms by residual cavity is seen in tail region of pancreas. Patient is planned for repeat USG abdomen after 6 months and CECT abdomen (if cavity persists) after 1 year of surgery as radiological resolution of cavity takes 3 – 9 months.



Fig. 5. Lump after pericystectomy, marsupialization and omentoplasty

4. DISCUSSION

Primary hydatid disease of the pancreas is rare—the largest single study of hydatid disease in 357 cases occurring over 20 years had only 1 case [9]. Infestation of the pancreas occurs mainly by hematogenous or peri-pancreatic lymphatic invasion [10]. Clinical manifestation depends on the anatomical location of cyst and its complications such as infection, rupture, hemorrhage, biliary or pancreatic fistula, vascular thrombosis and acute or chronic pancreatitis.

Most of the patient remain asymptomatic for long period of time or experience non specific complaints such as Abdominal pain, distention or Nausea especially if cyst is arising from body or tail of pancreas. In our case, Patient had awareness of lump for past 15 years but she sought medical opinion only when suffered from progressively increasing abdominal pain. Pancreas when involved by this disease process, it is usually isolated in nature, thus pancreatic Hydatidosis should be considered in differential diagnosis of cystic lesions of pancreas [6].

A very high index of suspicion is required for pre-operative diagnosis of pancreatic hydatidosis. Ultrasonography is the most sensitive for detection of cyst membranes, septa and hydatid sand [9]. Presence of an undulating membrane and multiple daughter cysts within a mother cyst can suggest the diagnosis on CT and MRI. Endoscopic Ultrasound (EUS) evaluation and guided aspiration of pancreatic cystic fluid and cytological/biochemical evaluation can help in excluding pancreatic cystic neoplasms and pseudocyst of pancreas. We would like emphasize the role of EUS as this was the investigation after which Pancreatic hydatidosis was considered for first time in provisional diagnosis.

Magnetic Resonance Cholangio-Pancreatography (MRCP) is helpful in delineating the biliary tree and pancreatic duct when the pancreatic cyst is located in the head of pancreas and/or causing ductal compression. MRI is superior in demonstrating irregularities of the cyst rim, representing incipient membrane detachment. In our case, MRI showed characteristic features of hydatid cyst such as floating membranes and daughter cysts, also there was no biliary or pancreatic communication with the cyst. Enzyme-linked immunosorbent

Table 1. A summary of the literature showing different clinical features of Hydatid cysts in the pancreas

Clinical features	Imam et al. [11]	Ozsay et al. [12]	Alsaid et al. [13]	Bakkaly et al. [10]	Sethi et al. [14]	Tezcaner et al. [15]	Ahmed et al. [16]	Hiremath et al. [9]
Gender	Female	Female	Male	Female	Female	Female	Female	Female
Age	18	23	34	5	48	55	40	48
Presenting symptoms	CT - Incidental detection of 3.5-cm cyst in the tail of the pancreas	Epigastric pain for 2 days	Diffuse abdominal pain, episode of acute pancreatitis 2 months ago.	Epigastric pain - two weeks associated with dietary vomiting and diarrhea	Vague abdominal pain and malaise	Epigastric pain	Epigastric pain	Epigastric lump for 5 months
Physical examination	Normal	tenderness in the epigastric region	NA	Normal	Normal	Normal	5 × 5 cm lump occupying the epigastrium and left hypochondrium	8×6 cm non-tender, smooth cystic epigastric mass with horizontal mobility
Serum amylase	NA	794 iu/l	765	NA	NA	NA	105	NA
Elisa serology	NA	NA	NA	Positive	NA	NA	NA	NA
Diagnosed by	CT scan	(CT)- mild oedematous pancreatitis and hypodense cystic mass on tail of the pancreas with a diameter of 45 mm	CT scan - pancreatic cyst measuring 13.5 × 7 cm	MRI - cystic fluid mass in contact with the second duodenum, with a thickened wall septum making 57mm/31mm pushing	MRI - hyperintense lesion in the body and the tail of the pancreas with a hypointense rim and internal serpiginous	MRI - solitary cystic lesion with septations at the pancreatic tail level measuring 24 mm × 18 mm	laparotomy	CT - 8×10×8.2 cm well circumscribed, cystic lesion with a volume of 330 mL, having a cyst wall thickness of 2 mm, arising from the neck of

Clinical features	Imam et al. [11]	Ozsay et al. [12]	Alsaid et al. [13]	Bakkaly et al. [10]	Sethi et al. [14]	Tezcaner et al. [15]	Ahmed et al. [16]	Hiremath et al. [9]
		causing left-sided portal hypertension due splenic vein thrombosis		back the gallbladder and the head of the pancreas of 10cm diameter, with multiple daughter cysts	hypointense structures suggestive of detached membranes			the pancreas
Cyst location	Tail	Tail	Tail	Head	Body and tail	Tail	Body	Neck
Cyst size (in cm) diameter	4 cm	4.4 x 3.6 cm	35 × 20 × 15 cm	5.7 x 3.1	11 x 14 cm	2.4 x 1.8	5 x 6 cm	8 x 10
Surgical treatment	laparoscopic spleen-sparing distal pancreatectomy	En bloc resection of distal pancreas and spleen	Cyst fenestration, multiple daughter cysts and endocyst membrane removed	External drainage of mass with removal of membranes	NA	Laparoscopic spleen-preserving distal pancreatectomy and cholecystectomy	Partial cystectomy was done along with exterparenchymal drainage of the residual cavity	Excision
Postoperative complications	Nil	Nil	Acute edematous pancreatitis after a month of surgery, local retroperitoneal abscess	Nil	NA	NA	NA	NA

assay for Echinococcal antigens is positive in more than 85% of cases [8]. It was positive in this case also. Management of the cystic echinococcosis includes surgery, percutaneous treatment, antiparasitic drug therapy, and observation. Evacuation of the cyst and defacement of the residual cavity are the aims of the surgical therapy. Medical therapy is useful when it is combined with/to surgery and percutaneous therapy. Drug treatment reduces the risk of disseminated disease on intraoperative spillage and recurrence. Albendazole is the most effective among Antihelmenthic medications used [2].

Though Distal pancreatectomy has been done in some cases reported in literature for pancreatic body and tail hydatid cyst. This procedure is not without complications most notably pancreatic fistula formation. Also ductal damage, hemorrhage or less commonly pancreatic insufficiency may develop. If similar results are achievable with less invasive approach as drainage and deroofting of cyst, it can be considered. This was the basis for our surgical plan in this case. Due to paucity of such cases, individualized managements are often undertaken. However more studies with larger number of patients should be conducted to decide whether radical or conservative approach should be used for management of these patients.

Table 1 showing cases of primary pancreatic hydatid cyst and there individualized management.

5. CONCLUSION

Primary Pancreatic Hydatid cysts can mimic pseudocyst or cystic neoplasms of pancreas, thus should be kept in differential diagnosis in pancreatic cystic lesions especially in endemic regions. Imaging studies such as Ultrasonography or CT scan are not always diagnostic that's why high index of suspicion is required to diagnose them preoperatively. Role of Endoscopic ultrasound in pancreatic cystic lesions cannot be over emphasized. Imaging studies to rule out biliary or pancreatic communication should be done preoperatively so that less radical approach can be undertaken while operating on these patients. Major pancreatic resection in benign entities should be avoided. However, further studies with large number of patients and longer follow up can

guide surgical management of these patients in future.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

As per international standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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