Primary hydatid cyst of pancreas: Case report and review of literature

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A B S T R A C T

INTRODUCTION: Hydatid disease is caused by the larval stage of Echinococcus granulosus. It most commonly affects the liver and lung. Pancreatic hydatid cyst (PHC) is very rare with incidence of 0.14%–2%.

PRESENTATION OF CASE: A 40 year old lady presented with epigastric pain for last 3 months. A 5 × 5 cm abdominal lump occupying the epigastric and left hypochondrial region was noted on physical examination. Ultrasonography (USG) and Contrast enhanced Computed Tomography (CT) revealed a 55 × 57 mm cystic structure in the pancreatic body. Endoscopic ultrasound guided fluid aspiration cytology revealed normal Carcinoembryonic antigen and Amylase levels. Cytological examination was noncontributory. During open surgical exploration, it was found to be a hydatid cyst. After irrigation with stadalid agent and evacuation of cystic contents, Partial cystectomy with external drainage was done. Histopathological biopsy revealed Hydatid cyst. Post-operative ELISA (Enzyme linked immunosorbent assay) for Echinococcal antigen was positive.

DISCUSSION: PHC is a rare entity. Most common mode of spread is hematogenous. Cysts in pancreatic head can present as obstructive jaundice. Cysts in body and tail are usually asymptomatic. USG, CT and Hydatid serology can help in diagnosis and monitoring recurrence. Surgical exploration is treatment of choice. Options include pericystectomy, partial cystectomy + − external drainage/omentectomy, marsupialisation or cysto-enterostomy. Preoperative and Post-operative anti helminthic (Albendazole) is recommended.

CONCLUSION: PHC can masquerade as pseudocyst or cystic neoplasm of pancreas. It should always be considered in the differential diagnosis of cystic pancreatic lesion in patients from endemic regions.

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1. Introduction

Hydatid disease is a zoonosis caused by the larval stage of Echinococcus granulosus which is endemic to regions where stock-breeding and agriculture are a common occupation [1]. These include the Mediterranean region, Africa, South America, Australia, Middle East and India [2]. For Echinococcus granulosus, dogs are the definitive host whereas sheep and goats are the intermediate host. Man is an accidental dead end host who is infected after consuming vegetables contaminated with dog feces containing Echinococcal eggs [3]. Hydatid cysts can be found in almost any organ of the body but the most common sites are liver (50%–77%), lung (15%–47%), spleen (0.5%–8%), and kidney (2%–4%) [1]. We describe a case which presented with epigastric pain and cystic pancreatic mass.

2. Presentation of Case

A 40 year old lady presented to us with chief complaint of epigastric pain for 3 months. The pain was insidious in onset, continuous, non-progressive, 4/10 intensity, radiating to the back and dull aching in character with no aggravating or relieving factors. Patient had no other significant complaints. Physical examination revealed approximately 5 × 5 cm lump occupying the epigastrum and left hypochondrium. It was spherical in shape, had ill-defined margins, smooth surface, not moving with respiration, not contiguous with liver dullness and firm in consistency. On examining the patient in the knee-elbow position the lump did not fall forward implying retroperitoneal location of the swelling.

Base line hematological and biochemical investigations were within normal limits. Serum Amylase (103 U/L) and Lipase (165 U/L) were non diagnostic for Acute pancreatitis. Previous Serum Amy-
Lase and Lipase done at the onset of symptoms 3 months earlier were within normal limits.

Chest X-ray was within normal limits. Ultrasonography of abdomen noted a cystic structure in the body and tail region of pancreas approximately 5.6 × 5.7 cm in size. No mass lesion was noted. (Fig. 1) Contrast enhanced CT abdomen showed well defined round to oval cystic lesion of size approximately 55 × 57 mm noted in the epigastric region near pancreatic body causing compression over adjacent stomach and pancreatic parenchyma with minimal adjacent fat stranding with well-defined peripherally enhancing margins. (White arrows).

Pancreas distal to the lesion was atrophic with prominence of Main pancreatic duct proximal to it. The main radiological differential diagnosis was between a pseudopancreatic cyst and a cystic pancreatic neoplasm.

We decided to go for an Endoscopic Ultrasound (EUS) which showed a 6 cm cystic lesion in body and tail of pancreas. Analysis of EUS guided aspirated fluid showed a clear fluid with CEA (1.22 ng/ml) and Amylase (30 U/L) levels within normal limits. The centrifuged deposits were acellular with no inflammatory cells, malignant cells or parasitic elements seen in the cytosmears.

The patient did not have a previous history of trauma or pancreatitis making a pseudocyst less likely. The other differential was a cystic neoplasm of the pancreas. The location of the cyst and analysis of the cystic fluid aspirate (low CEA, low Amylase, mucin negative) were in favor of a serous cystic neoplasm (SCN) but CT did not show the characteristic radiological findings of SCN.

A decision was made to explore the patient through mid-line laparotomy incision. The main reason for operating the patient was the patient’s symptoms. Our primary clinical suspicion was a serous cystic neoplasm of pancreas despite no characteristic findings on the CT scan. Intraoperatively, an approximately 5 × 6 cm cystic structure was found to be originating from the upper border and body of pancreas pushing the stomach forwards. Intraoperative spillage from the cyst was minimal as we took care to aspirate the cyst with a syringe first. Aspiration of the cyst showed clear fluid. The cyst was opened and gerimal membrane was found. (Fig. 3) The cyst was injected with a scolicidal agent, 0.5% Cetrilmide solution and the contents evacuated after packing the operative field with Cetrilmide soaked sponges. Abdomen was irrigated with the scolicidal agent. Partial cystectomy was done along with exter-

Fig. 1. Abdominal Ultrasonography a cystic structure in the body and tail region of pancreas approximately 5.6 × 5.7 cm in size (White arrows). No mass lesion was noted.

Fig. 2. (A) Axial section (B) Coronal section of Contrast enhanced CT abdomen showing well defined round to oval cystic lesion of size approximately 55 × 57 mm noted in the epigastric region near pancreatic body causing compression over adjacent stomach and pancreatic parenchyma with minimal adjacent fat stranding with well-defined peripherally enhancing margins. (White arrows).
nal drainage of the residual cavity. There was no communication with the pancreatic duct. Post-operative histopathological analysis confirmed hydatid cyst. ELISA for Echinococcal antigens done post-operatively was positive. Post-operative drain output was minimal. The drain was removed on Post-operative day 3. Patient was discharged on Oral Albenzole tablets (10 mg/kg/day) for 8 weeks. Patient has been asymptomatic for the last 4 months since operation. Repeat Chest X-ray and USG abdomen are normal.

3. Discussion

Pancreatic hydatid cysts (PHC) are rare entities with incidence ranging from 0.14% to 2% [4]. PHCs are usually solitary (90%–91%) and distributed unevenly throughout the head (50%–58%), body (24%–34%) and tail (16%–19%) [1]. Hematogenous dissemination is hypothesized to be the most common mode of spread to the pancreas. The other possible modes of spread of cystic elements to the pancreas described are passage through the biliary system, lymphatic spread from the intestinal mucosa, direct passage via the pancreatic veins and retroperitoneal dissemination [1,4].

Clinical presentation depends on the location of the cyst within the pancreas. Cysts located in the head can present as obstructive jaundice due to external compression of the common bile duct and masquerade as a choleodochal cyst [5,6]. Cysts located in the body and tail of the pancreas are usually asymptomatic until they become large enough to present as an abdominal lump or cause symptoms due to compression of adjacent structures like epigastric pain, nausea and vomiting [1,4]. Rarely, cysts located in the pancreatic tail can result in splenomegaly and portal hypertension [7]. Complications like cholangitis, rupture into the biliary tree or peritoneal cavity, pancreatic fistula, recurrent pancreatitis and abscess have also been described [8].

Imaging modalities commonly employed to diagnose a pancreatic cyst are Ultrasonography (USG), Computed Tomography (CT) and Magnetic Resonance Imaging (MRI). Abdominal USG is a sensitive tool for diagnosing Hydatid Cyst with characteristic findings like floating membranes, hydatid sand and daughter cysts though the sensitivity is decreased due to retroperitoneal location and bowel gas in case of PHCs [9]. Presence of an undulating membrane and multiple daughter cysts within a mother cyst can suggest the diagnosis on CT and MRI [4,10]. Endoscopic Ultrasound (EUS) guided aspiration of pancreatic cystic fluid and cytological/biochemical evaluation can help in excluding pancreatic cystic neoplasms and pseudocyst of pancreas [2]. 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 [11]. Endoscopic Retrograde Cholangio-Pancreatography (ERCP) is appropriate for palliative stenting in case of cholangitis or pancreaticitis secondary to biliary/pancreatic ductal compression by PHCs [1]. Tests for detecting specific serum antibodies and circulating echinococcal antigens include indirect hemagglutination assay, immunoelectrophoresis, enzyme linked immunosorbent assay, complement fixation test and immunofluorescence assay [1]. They are useful in follow-up monitoring also. Enzyme-linked immunosorbent assay for Echinococcal antigens is positive in more than 85% of cases [12,13].

The characteristic radiological findings described for hydatid cysts are often not present, as in our case [4]. This makes differentiating PHCs difficult from more common cystic lesions of the pancreas like pseudocysts and benign or malignant cystic neoplasms of the pancreas [1,4]. Never the less, PHCs should always be considered in the differential diagnosis of pancreatic cystic lesions in patients from endemic areas. Percutaneous or Endoscopic ultrasonography guided Fine needle aspiration cytology can help in diagnosing equivocal cases but prophylactic antihelminthics should be started to prevent anaphylactic reaction and peritoneal seeding in case of spillage or perforation [14]. It is often not possible to make a definitive diagnosis without surgical exploration [1,4].

Conservative techniques like Puncture-Aspiration-Injection-Reaspiration (PAIR) or direct percutaneous catheterization with medical therapy have been described in patients not fit for surgical intervention [1,4,15].

Open surgery is the treatment of choice [1,4]. The exact procedure depends on the location of the cyst. Care should be taken during the procedure to pack the operative area with sponges soaked in scolicidal agents like 0.5% Cetrizide or 20% Hypertonic saline and avoid spillage of the cyst contents [16]. The cyst should be irrigated with scolicidal agents. Preoperatively diagnosed cases should receive prophylactic antihelminthic agents (Albenzole 10 mg/kg/day) for 2–4 weeks which should be continued post-operatively for at least 4 weeks. This decreases the risk of anaphylactic reaction in case of spillage or perforation and decreases the risk of post-operative recurrence [1].

Cysts present in the head without any communication can be treated with pericystectomy, partial pericystectomy with external drainage or omentopexy, marsupialization or pancreateoduodenec-
tomy procedures [1]. In cysts with communication to the pancreatic duct, cysto-enteric anastomosis is the favored procedure [1]. Cysts located in the tail and body/neck of pancreas are appropriate for distal pancreatectomy and central pancreatectomy respectively [1,4]. Laparoscopic evacuation of the cyst with omentoplasty has also been described using a 10 mm trocar [17].

Primary hydatid cyst of the pancreas is an extremely rare entity. Our intention in presenting the case is to highlight the fact that hydatid cyst can masquerade as more common cystic lesions of the pancreas. Physicians who have not encountered a pancreatic hydatid cyst may not consider it in the differential diagnosis. In patients from endemic areas, a pancreatic hydatid cyst should always be considered in the differential diagnosis.

4. Conclusion

PHCs can masquerade as more common lesions of the pancreas like pseudocyst or cystic pancreatic neoplasms. PHCs should always be kept in the differential diagnosis in case of cystic pancreatic mass in patients form endemic areas. Hydatid serology, Imaging modalities and Cytological examination of cystic aspirate are important but not always diagnostic. Surgical exploration with histopathological examination is the gold standard.

5. Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

6. Conflict of interest

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8. Ethical approval

Ethical approval not required.

9. Author contribution

ZA helped in conception and design of study, analysis and interpretation of data, drafting the article and final approval of the version to be submitted. SC helped in analysis and interpretation of data, revising the article critically for important intellectual content and final approval of the version to be submitted. AM helped in acquisition of data, revising the article critically for important intellectual content and final approval of the version to be submitted. VV helped in analysis and interpretation of data, revising the article critically for important intellectual content and final approval of the version to be submitted. RY helped in conception and design of the study, revising the article critically for important intellectual content, and final approval of the version to be submitted. RB helped in analysis and interpretation of data, revising the article critically for important intellectual content and final approval of the version to be submitted. JK helped in acquisition of data, revising the article critically for important intellectual content and final approval of the version to be submitted. RKJ helped in acquisition of data, revising the article critically for important intellectual content and final approval of the version to be submitted.

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