

Case report

Primary hydatid cyst of pancreas mimicking mucinous cystic neoplasm

Running title: Hydatid cyst of pancreas

Keywords: Echinococcus, Hydatid cyst, Mucinous cystic neoplasm, Pancreas

Disclosures

Informed consent was obtained from the patient for this case report.

Abstract

Hydatid cyst (HC) is a major health problem in endemic countries. Primary pancreatic hydatid disease (PHC) is exceedingly rare entity which may mimic other, more commonly encountered cystic neoplasms of pancreas. We report the case of a 55-year-old female who presented with abdominal pain. The treatment consisted of a distal pancreatectomy for suspected mucinous cystic neoplasm (MCN). A diagnosis of PHC was established during the surgery. Primary PHC, though exceedingly rare, should be considered in the differential diagnosis of cystic lesions of the pancreas in patients from endemic regions.

Keywords

Echinococcus, Hydatid cyst, Mucinous cystic neoplasm, Pancreas

Introduction

HC is a zoonotic parasitic disease caused by tapeworm *Echinococcus*. The disease is endemic in many countries and human infestation occurs when they accidentally ingest tapeworm eggs. HC may develop in almost any organ of the body; the liver (58%), lung (26%), kidneys (4%), spleen (3%) and brain (3%) are the most affected organs, with infestations of the bone, pancreas, thyroid and breast

rarely encountered.[1] Reported incidence of PHC varies from 0.1–2% even in those countries where the disease is endemic.[2] Pre-operative diagnosis of PHC can be difficult due to rarity of the disease as well as overlapping clinical and radiological features with that of other common cystic neoplasm of pancreas. We report a case of a 55-year-old woman who was suspected as a case of MCN of pancreas on pre-operative radiological imaging.

Case Report

A 55-year-old woman without history of acute or chronic pancreatitis presented with mild abdominal pain of 6 months duration. On physical examination, there was a palpable non-tender mass in left upper quadrant. All routine laboratory studies were within normal limits. Serum CA-19.9 was 14.5 U/ml. Abdominal sonography suggested a multiseptated cystic mass at the tail of pancreas. Contrast enhanced computed tomography (CECT) scan of abdomen revealed a 7.6 x 8.1 cm thick-walled cystic mass with enhancing margins and internal septations in the tail of pancreas [Figure 1]. Endoscopic ultrasound (EUS) revealed complex solid-cystic tumour arising from pancreatic body and tail region, measuring 7.2x6.2 cm with thick irregular wall, multiple cysts of varying sizes and intramural solid component. There was no communication seen between cyst and pancreatic duct on EUS. Fine needle aspiration cytology (FNA) of cyst fluid was not performed due to lack of facility. A pre-operative probable diagnosis of MCN of the pancreas was considered. On abdominal exploration, a mass was found to be arising from body and tail of the pancreas and having dense adhesion to the adjacent small bowel and omentum [Figure 2]. Spleen preserving distal pancreatectomy was performed without rupturing the cyst. Rest of abdominal organs were normal. On-table gross examination of resected specimen revealed multiple daughter cysts confirming the diagnosis of primary PHC [Figure 3]. Patient was discharged on post-operative day five after having uneventful recovery.



Figure 1. Computed tomography scan showing cystic mass with internal septation in body and tail region of pancreas.

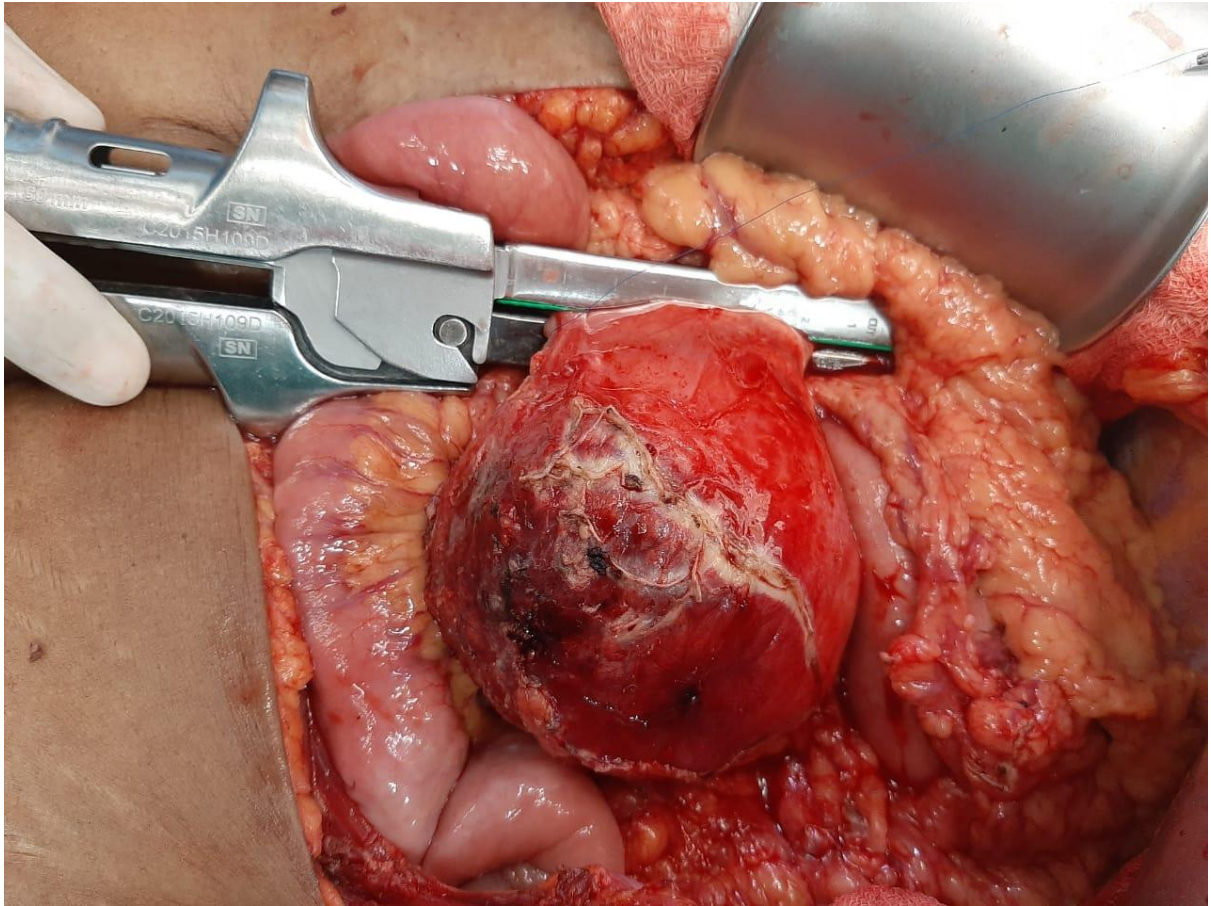


Figure 2. Intraoperative appearance of cyst after mobilizing the pancreatic tail.

UNDER PEF



Figure 3. Gross examination of specimen revealing multiple daughter cysts.

Discussion

Hydatid cyst (HC) is a major health problem in endemic countries. There are four known different species of *Echinococcus*. Among the four known species of *Echinococcus*, three are of medical importance in humans. These are; *Echinococcus multilocularis*, causing alveolar echinococcosis; *Echinococcus granulosus*, causing cystic echinococcosis and *Echinococcus vogeli*. This disease is caused by larval stage of *E granulosus*. Liver and lung are the most common sites to be involved. Pancreatic location of hydatid disease is rare (less than 1%) compared to the other sites of hydatid disease. The mode of infestation is either hematogenous, when there is a failure of trapping oncosphere by the liver and lung filters, or more rarely through lymphatic spread.[3] The cyst can be found in the head (50- 57%), in the body (24-34%) or in the tail region (16-19%) of pancreas.[4] Clinical manifestation of PHC depends upon the anatomical location and potential complications of cyst in pancreas. Patient may present with jaundice when the cyst is present in head due to external compression of bile duct, however, patient may be asymptomatic or present as abdominal lump and

recurrent abdominal pain when the cyst is present in body or tail of pancreas. Rarely, patient may develop acute pancreatitis, infection, biliary or intestinal fistula, segmental portal hypertension, vascular thrombosis, acute or chronic pancreatitis, rupture of cyst in peritoneal cavity or communication with pancreatic duct.[5] Preoperative diagnosis of hydatid cyst of pancreas can be exceedingly difficult unless hydatid disease is suspected and modern serology tests such as indirect fluorescent antibody test, latex agglutination test, and enzyme-linked immuno-absorbent assay test for echinococcal antigens are positive. However, these serological tests are not widely available in countries where hydatid disease is endemic. Most cystic lesions in the pancreas are pseudocysts (80%) followed by cystic neoplasms including MCN (10%).[6] Most MCNs are oligolocular lesions and their similarity to the detached membranes of PHC in such cases can be confusing on imaging. Considering radiological findings, our patient had type CE III-B HC involving the pancreatic body and tail region. In favour of similarity, most MCNs are typically located in the body and the tail of the pancreas (93%) and in almost all cases are seen in women (>90%).[6] Some authors suggested EUS guided FNA biopsy to differentiate a hydatid cyst from other common cystic lesions.[7] However, it is operator dependent and available only in high-volume centres. Surgery remains the treatment of choice for primary PHC. Type of surgery depends on the site of cyst and communication with pancreatic duct. Surgical procedures ranging from deroofting of cyst to some type of pancreatic resection. Role of percutaneous aspiration and chemotherapy in primary PHC is not well established.[8]

Conclusion

Primary pancreatic hydatid cyst, though exceedingly rare, should be considered in the differential diagnosis of cystic lesions of the pancreas in patients from endemic regions.

Disclosures

Author's contribution: Zuber Ansari was part of surgical team and drafted the manuscript. Somak Das, Sukanta Ray and Jayanta Biswas were part of surgical team and contributed to literature search and critical revision of manuscript. All authors read and approved the final manuscript.

Financial disclosure: None to report.

Informed consent was obtained from the patient for this case report.

References

1. Amr SS, Amr ZS, Jitawi S, et al. Hydatidosis in Jordan: an epidemiological study of 306 cases. *Ann Trop Med Parasitol*. 1994 Dec;88(6):623-7.
2. Hamamci EO, Besim H, Korkmaz A. Unusual locations of hydatid disease and surgical approach. *ANZ J Surg*. 2004 May;74(5):356-60.
3. Dziri C: Hydatid disease—continuing serious public health problem: introduction. *World J Surg*. 2001, 25:1-3.
4. Hammad A, Mentouri B: Acute pancreatitis in Algeria. Report of 221cases. *Am J Surg*. 1985, 149:709-711.
5. 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 Jul 8;6(4):354-8.
6. Kalb B, Sarmiento JM, Kooby DA, et al. MR imaging of cystic lesions of the pancreas. *Radiographics*. 2009 Oct;29(6):1749-65.
7. Sharma S, Sarin H, Guleria M, et al. Endoscopic ultrasound-guided FNA: Emerging technique to diagnose hydatid cyst of pancreas. *J Cytol*. 2015;32(3):211-212.
8. Dziri C, Dougaz W, Bouasker I. Surgery of the pancreatic cystic echinococcosis: systematic review. *Transl Gastroenterol Hepatol*. 2017 Dec 8;2:105.