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Case report
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Primary retroperitoneal hydatid cyst in pancreatic head region

Pankreas başında primer retroperitoneal hidatik kist

Shilpi Karmakar [1], Sanjay Kala [2], Saurabh Karmakar [3]

[1] Department of Plastic Surgery, King George's Medical University, Lucknow, India
[2] Department of General Surgery, Ganesh Shankar Vidyarthi Memorial Medical College, Kanpur, India
[3] Department of Pulmonary Medicine, All India Institute of Medical Sciences, Phulwarisharif, Patna, India

ORCID ID of the author(s)
SK: 0000-0001-7423-9186
SK: 0000-0001-8669-7512
SK: 0000-0002-8138-4864

Abstract
A hydatid cyst in retroperitoneal space is uncommon and one in the pancreatic head region is rare. We hereby report a case of a 20-year-old male presenting with a retroperitoneal hydatid cyst of the pancreatic head region. Investigations revealed no hydatid cysts in other organs of the body. The diagnosis was confirmed at laparotomy and partial pericystectomy was performed. Hydatid cyst should be considered in the differential diagnosis of cystic lesions of retroperitoneal area.
Keywords: Retroperitoneal cyst, Hydatid disease, Echinococcosis, Abdominal mass, Cystectomy

Öz
Hidatik kistler genel olarak retroperitoneal boşlukta görülmez, pankreas başı bölgesinde ise oldukça nadir saptanır. Bu vaka raporunda, pankreas başı bölgesinde retroperitoneal hidatik kist ile başvuran 20 yaşında bir erkek olguyu sunmaktayız. Araştırmalar vücudun diğer organlarında hidatik kist olmadığını ortaya çıkardı. Tanı laparotomide doğrulandı ve parsiyel perikistektomi yapıldı. Retroperitoneal alandaki kistik lezyonların ayırıcı tanısında hidatik kist düşünülmelidir.
Anahtar kelimeler: Retroperitoneal kist, Hidatik hastalık, Ekinokokkoz, Abdominal kitle, Kistektomi

Introduction

Hydatid disease (HD) is an anthropozoonosis that is present endemically in the grassland or temperate regions of the world, particularly the Mediterranean region, South America, and Australia. However, due to increased migration, hydatid disease is becoming a worldwide health problem [1].

HD is caused by the infection of the larval stage of *Echinococcus granulosus* parasite belonging to family Taeniidae of the Cestode class [3].

In 85-95% of the cases, the liver and/or the lungs are involved and in only 5-15% of patients, the cyst occurs at other sites [2]. Hydatid cyst (HC) developing in retroperitoneal space without accompanied lesion in other organs is rare, and one in the pancreatic head region is extremely rare. To the best of our knowledge, no such case of HC developing in retroperitoneal region in close proximity to the pancreatic head, without its involvement, has been reported. Rarity of this entity prompted us to report the case.

Corresponding author / Sorumlu yazar:
Shilpi Karmakar
Address / Adres: Department of Plastic Surgery, King George's Medical University, Lucknow, India
E-mail: drshilpikarmakar@rediffmail.com

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Case presentation

A healthy looking, 20-year old male visited the outpatient clinic of General Surgery with complaints of early satiety, a feeling of fatigue and fullness in the upper abdomen for three months. He had no history of trauma, nausea, vomiting, anorexia, yellowish discoloration of eyes or alteration of bowel habits. His medical history was not significant. He had no history of prior surgery.

On examination, his vital signs were within normal limits. A cystic lump measuring approximately 15 cm x 12 cm was palpated over the epigastrium, right hypochondrium and part of the left hypochondrium. The lump was spherical, well-defined, had a normal surface temperature, and non-tender. It did not move with respiration and was not contiguous with liver dullness. On examining the patient in the knee-elbow position the lump did not fall forward, signifying retroperitoneal location of the swelling. Hemogram revealed a hemoglobin of 11.2 g/dl, total leukocyte count of 14,900/cu.mm and differential count revealed 10% eosinophilia. His biochemical investigations including liver function test and renal function tests, coagulation profile and serum amylase and lipase levels were within normal limits. Contrast enhanced computed tomogram (CECT) of abdomen revealed thin walled, decreased attenuating lesions with multiple thin walled septations, measuring around 94 x 98 mm in longest dimensions (Figure 1). Focal posterior wall calcification was seen in the pancreatic head region. The mass displaced and compressed the common bile duct posterolaterally and the gall bladder posteriorly, with mild dilatation of intra hepatic biliary tract. It medially abutted the superior mesenteric artery (SMA) and vein and posteriorly abutted the right kidney and right renal vessels (Figure 2). Pancreas was mildly displaced. Investigations revealed no other cyst in liver, lungs or other organs.

Written informed consent was obtained from the patient for the surgery, photography and for publication of photographs. Under general anesthesia, laparoscopic surgical exploration was planned. Cyst was approached via the intraperitoneal route. It was adherent to the medial wall of duodenum, pancreas and SMA. It was decided to convert to open laparotomy. Abdomen was opened in upper midline. The lesser sac was entered by taking the omentum off the transverse colon and the attachments of the posterior gastric wall to the anterior surface of the pancreas. The large cyst was seen nested in the retroperitoneal tissue in duodenal C-curvature. The cyst was isolated from the abdominal cavity and other structures by four 20% hypertonic saline soaked mops. After placing stay sutures, a stab incision was performed in the cyst wall, cyst fluid was aspirated and hypertonic saline was poured in the cavity, which was re-aspirated after fifteen minutes. Due to thick adhesions with the SMA, partial pericystectomy with cauterization of the residual endocyst was performed. There was no communication with the pancreatic duct. A drain was placed and the abdomen was closed in three layers.

The drain showed minimal output. The patient, who recovered uneventfully, was discharged after three days on oral albendazole. The drain was removed on day seven. Sutures healed well and were removed on day 10. Histopathological examination of the aspirate from the cyst and tissue from cyst wall confirmed the diagnosis. The patient is in our follow-up for more than a year now and has no recurrence.

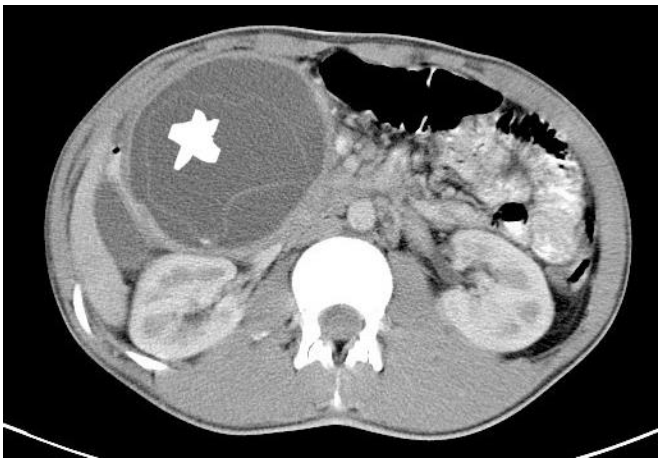

Figure 1: CT of abdomen showing thin walled decreased attenuating lesion (indicated by star) having multiple thin walled septations, measuring approximately 94 x 98 mm in longest dimensions.

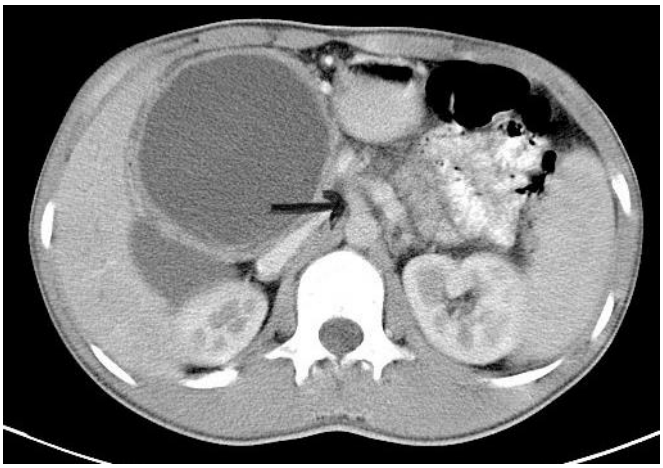

Figure 2: CT of abdomen showing Superior Mesenteric Artery (SMA) arising from the aorta. (indicated by arrow) Superior Mesenteric Vein is to the right of the SMA. SMA and SMV are adjacent to the cyst.

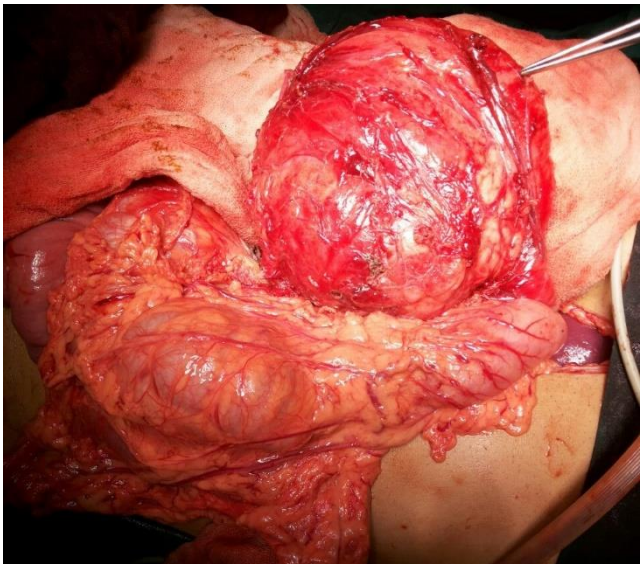

Figure 3: The transverse colon is reflected inferiorly, and mops are placed to cover the duodenum and liver. The forceps holds the pancreatic tissue. The pancreas head is stretched over the large cyst.

Discussion

Adult E. Granulosus lives in the intestinal tract of infected dogs. The parasite is transmitted to humans by the accidental consumption of soil, water, or food contaminated by the fecal matter of an infected dog. Echinococcus eggs deposited

in soil can remain viable for up to a year. In the stomach, the outer capsule of the egg gets digested and the freed larva (oncosphere) penetrate the duodenal/jejunal mucosa, enter the mesenteric venules and portal vein and reach the liver [3]. Larvae with diameters less than 0.3 mm pass through the sinus capillaries of liver into the vena cava and right side of heart and reach the lungs, where they are entrapped by pulmonary capillaries. Larvae that escape the lung may then pass to any organ of the body via arterial circulation.

Thus, the liver and lung account for the highest percentage of involvement. Isolated HC of the retroperitoneum is extremely rare [4]. A primary retroperitoneal HC without other organ involvement was first reported by Lockhart and Sapinza in 1958 and till 1973, only 9 cases had been reported in the literature [5,6]. Various modes of spread have been suggested to explain the escape of liver and lung involvement via lymphatics from the intestinal vessels to the thoracic duct or via veno-venous shunts within the liver and in the space of Retzius [7-9]

The larvae form cysts within the infected organs. The HC is a fluid-filled, spherical, unilocular cyst that consists of an inner germinal layer of cells (endocyst) and acellular laminated membrane. The outer layer, pericyst, forms due to the host response to the parasite and consists of modified host cells, fibroblasts, giant cells, and eosinophils [10].

Symptoms are usually due to compression as cysts increase in size [11]. When there are complications such as rupture and secondary infection, acute symptoms arise [5,7]. Rupture of the HC is the most common complication (about 15% of all cases). It may occur in the biliary tree causing acute cholangitis, or into the free peritoneal cavity with seeding daughter cysts in the peritoneum [1,2].

The differential diagnosis of a cystic retroperitoneal mass includes cystic lymphangioma, abscess, chronic hematoma, necrotic malignant soft tissue tumor, pancreatic cyst and hydronephrosis [12,13]. The establishment of diagnosis is based on the combination of clinical findings, imaging modalities, laboratory data and cytological information.

Abdominal ultrasonography is a sensitive tool for diagnosing HC with characteristic findings like floating membranes, hydatid sand and daughter cysts [14]. Presence of an undulating membrane and multiple daughter cysts within a mother cyst can suggest the diagnosis on CT and Magnetic Resonance Imaging [4,15]. The characteristic radiological findings described for hydatid cysts are often not present, as in our case.

Chemical tests include Casoni's test which is accurate in 90% of cases and Ghedini-Weinberg test which is accurate in 80% cases [16]. However, neither test is specific for this disease. Recently, many serological methods have replaced Casoni skin test to diagnose human hydatidosis. These methods include latex agglutination, indirect hemagglutination, counter immune-electrophoresis, enzyme-linked immunosorbent assay (ELISA) with whole hydatid fluid and with antigen 5 [17]. Indirect hemagglutination and ELISA are the most sensitive tests. Eosinophilia is detected in 20-50% of patients. [16,18,19]. Thus, serological tests are of limited value. A definite pre-operative diagnosis without histological examination is often difficult [20]. Delayed diagnosis and misdiagnosis increase the risk of impairment, recurrence, and sepsis.

Surgery is the cornerstone for treatment of HD [16,21]. Total cystectomy without contamination of the field is the procedure of choice. If total cystectomy is not possible, because of dense adhesions to important anatomical structures, partial cystectomy should be done [22]. A careful search for other abdominal and liver hydatid cysts should be made because secondary retroperitoneal hydatids far outnumber primary retroperitoneal HC.

Chemotherapy is effective when used in conjunction with surgery. A course of albendazole administered prior to surgery facilitates surgical manipulation of the cysts by inactivating protoscoleces, altering the integrity of the cystic membranes, and reducing the turgidity of the cysts [23].

Conclusion

Due to varied and vague symptomology, suspicion helps in the diagnosis of the HD. HC should be ruled out in cystic retroperitoneal lesions. HC can masquerade as more common lesions of the retroperitoneum. Surgical exploration with histopathological examination is the gold standard treatment. Total cyst excision must be aimed. Precautions should be taken to prevent catastrophic anaphylaxis.

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