

CASE REPORT

A Case Report of the Unusual Presence of Hydatid Disease in the Pancreas and Breast

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ABSTRACT

Context Hydatid cyst disease can occur in all viscera and soft tissues although the liver and the lungs are the most commonly involved organs. Pancreatic and breast involvement with the disease are extremely rare and the presence of this disease in these organs without any other organ involvement has not been previously reported.

Case report We present herein a patient with isolated pancreatic cyst hydatid treated successfully with complete cyst excision. However, 18 months after the pancreatic surgery, hydatid disease was found in the breast and again the cyst was completely removed. No other organ involvement has been detected during a 2-year follow-up.

Conclusions Hydatid disease should be considered in the differential diagnosis of all cystic masses in all anatomic locations, especially in regions where the disease is endemic. The ideal treatment is the complete excision of the cyst without any spillage.

INTRODUCTION

Hydatid cyst disease still constitutes a serious public health problem in areas in which it is endemic. It is a tissue infestation caused by

the larval stage of a parasite, *Echinococcus granulosus*. Although the liver and lung are the most commonly involved organs, hydatid disease can occur in all viscera and soft tissues [1]. Primary pancreatic hydatid disease is extremely rare and it accounts for 0.19-2% of all hydatid cases [2]. The breast is another rare location for the disease; it accounts only for 0.27% of all occurrences [3]. To the best of our knowledge, pancreatic and breast involvement without any other organ involvement has never been reported. We present herein such a case.

CASE REPORT

A 38-year old woman was admitted to hospital with a six-month history of epigastric pain associated with nausea and vomiting. She had no history of weight loss, jaundice, alcohol abuse, abdominal trauma, previous pancreatitis or abdominal surgery. Physical examination revealed nothing but fullness in the epigastric area. Routine blood tests and serum amylase levels were all within the normal limits. An upper gastrointestinal endoscopic evaluation was also normal. Abdominal ultrasonography (US) revealed a multiloculated cystic mass with a diameter of 90x60x70 mm located anteriorly in the corpus of the pancreas. Abdominal computerized tomography (CT) was done to delineate the borders and the relationship of the cyst to the



Figure 1. An abdominal CT scan revealed a multiloculated cystic mass with a diameter of 90x60x70 mm which had an internal septa which was originating from the corpus of the pancreas and bulging anteriorly, hence, distorting the lesser curvature of the stomach by its mass effect.

surrounding structures. It showed a cystic mass with internal septa which originated from the corpus of the pancreas and bulged anteriorly hence distorting the lesser curvature of the stomach by its mass effect (Figure 1). The liver and other abdominal structures were all normal.

She underwent surgery for a suspected cystic neoplasm of the pancreas. At laparotomy, we observed that the cystic mass was bulging from the pancreas anteriorly and was surrounded by the stomach, transverse colon, left lobe of the liver and the omentum. The cyst was carefully freed from all adhesions and surrounding structures. Then, the cyst

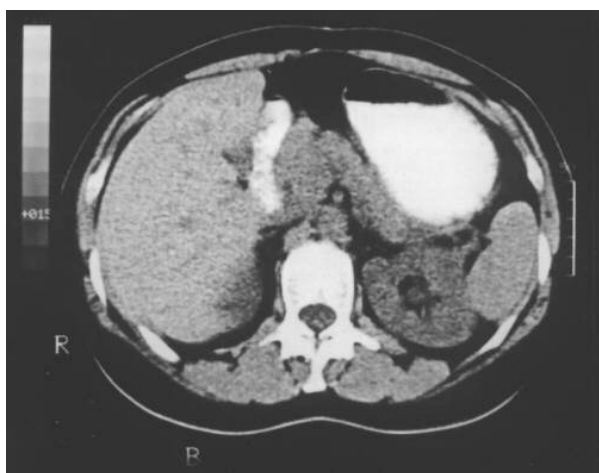


Figure 2. Postoperative abdominal CT showed a completely normal pancreas without any evidence of an intra-abdominal or retroperitoneal hydatid cyst.

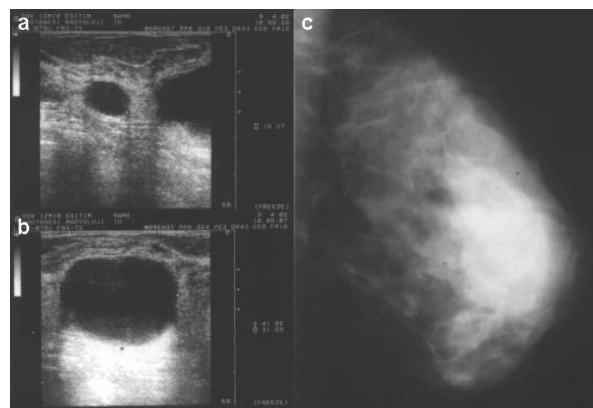


Figure 3. Ultrasonography (a, b) of the breast revealed one large and one small pure cystic lesion adjacent to each other but mammography (c) showed only one pure cystic lesion in the left breast measuring 50x50 mm.

was punctured with a syringe and surprisingly transparent fluid was aspirated. On the possibility of being faced with hydatid disease, a scolicalid agent was introduced first and then the cyst was opened from the anterior wall. We saw the germinative membrane and daughter vesicles and removed them. No communication between the pancreatic duct and the cyst cavity was demonstrated. Partial cystectomy (only the base of adventitial cyst wall was left) was performed and the cystic cavity was obliterated with a patch of the omentum. The postoperative period was uneventful. A CT image of the thorax was performed for screening but was normal. The patient was discharged seven days after the operation.

She was put on a treatment regimen with albendazole at a dose of 10 mg/kg/day for two months. She was well one year after the surgery and a CT of the abdomen revealed completely normal pancreas (Figure 2). However, she was admitted to our clinic with a painless mass lesion in the left breast 18 months after the surgery. Physical examination revealed a soft and mobile mass of 5 cm in size in the upper quadrant of the left breast. Ultrasonography of the breast revealed one large and one small pure cystic lesion adjacent to each other but mammography showed only one pure cystic lesion in the left breast measuring 50x50 mm (Figure 3).



Figure 4. Intraoperative findings of the breast hydatid disease: ruptured cyst and part of the germinative membrane.

Excision of the cyst was planned but the cyst was ruptured during the procedure and the germinative membrane was observed (Figure 4). The area and remaining cavity were immediately irrigated with a scolicidal agent. A CT of the abdomen and the thorax was carried out after the operation and found to be normal. After an uneventful recovery, she was again treated with albendazole for six months. She has been followed without any recurrence or any complaint for two years.

DISCUSSION

Hydatid disease is often manifested by a slow growing cystic mass. The liver and lungs are the organs most frequently involved. The cysts may be single or multiple; uni- or multi-loculated, and thin or thick walled. More specific signs include visualization or calcification of the cyst wall, the presence of daughter cysts and membrane detachment; however, hydatid cysts with unusual localizations may cause serious problems in the differential diagnosis [1, 4].

Echinococcal involvement of the pancreas is extremely rare and it is often associated with hepatic involvement. Possible sources of infestation include hematogenous dissemination, local spread via the pancreatobiliary ducts and peripancreatic invasion [5]. The location of the hydatid cyst in the pancreas has a distinct distribution: 50% at the head, 35% within the corpus and

15% at the tail. The clinical presentation varies and usually results from the compression of the adjacent structures by the cyst; hence, it depends heavily on the size and anatomic localization of the cyst. If it is situated in the head, compression of the common bile duct may result in jaundice. The parasites may lead to acute pancreatitis or even to recurrent attacks and chronic pancreatitis if they occlude the main pancreatic duct [6]. However, cysts located in the body or the tail of the pancreas may be symptomless and may be detected only as an abdominal mass [7]. A cyst located in the tail of the pancreas may also cause portal hypertension [8]. Complications include spontaneous rupture into the peritoneal cavity or gastrointestinal tract [9], and abscess formation [10].

Either US or CT can easily be used to diagnose a hydrated cyst of the liver and lung. A high index of suspicion, peripheral eosinophilia, positive echinococcal antigen immunofluorescence or hemagglutination tests and a history of exposure in an endemic area are also some clues to the diagnosis. Serologic tests, i.e., the enzyme linked immunosorbent assay, indirect hemagglutination, and immunoblot techniques confirm the hydatid origin of a cyst which is diagnosed by imaging techniques. A serologic survey is necessary in the follow-up of operated patients.

Hydatid disease located in rare abdominal sites may lead to diagnostic difficulties, which is especially true in the case of isolated lesions in which there is no liver or lung involvement. Moreover the variable ultrasonographic and tomographic images of isolated hydatid cysts of the pancreas make it difficult to distinguish these cysts from other solid and cystic lesions of the pancreas.

A hydatid cyst of the breast is usually not included in the differential diagnosis of breast lumps due to its rarity even in endemic regions. Systemic dissemination is the source of breast involvement in hydatid disease. Mammography and US are not very helpful for diagnostic purposes; ring-shaped structures inside a breast lesion at

mammography may suggest hydatid disease but this finding is non-specific [11]. A hydatid cyst of the breast may have an appearance indistinguishable from a benign cyst at US. However, the presence of a thicker and more laminated wall than a simple cyst and a thin layer of calcification within the lesion may suggest a hydatid cyst [12]. Magnetic resonance imaging (MRI) may be more helpful; a well-circumscribed cystic lesion with capsular enhancement suggests hydatid cyst but this appearance resembles a breast abscess on MRI [13].

Surgery is still the most effective therapy for hydatid disease which exists in any location [1, 3, 7]. The principal objectives of the surgical treatment are total removal of all parasitic elements, avoidance of spillage of the contents of the cyst, and removal of the cyst with maximum conservation of the stricken organ. The management of a residual pericyst cavity is another important concept. One of the aims of surgery in pancreatic disease is maintaining pancreatic exocrine and endocrine functions. Partial or total cystectomy, cytoenteric anastomosis, marsupialization and external drainage have been reported in the management of pancreatic hydatid cysts [2]. We totally excised the cysts and no communication was present between the cyst and the pancreatic duct. The cavity was filled with omentum and the completely healed cavity was observed one year after the surgery.

In hydatid disease of the breast, total cyst excision is the choice of treatment. But it may be confused with a simple breast cyst and may be aspirated for diagnosis and treatment. The diagnosis of breast hydatid cyst by fine needle aspiration biopsy has even been reported [14] but contamination risk should always be kept in mind.

One of the main problems in patients with a hydatid cyst is the recurrence of the disease. Spillage of protoscoleces from the cyst during surgery can lead to implantation and dissemination of *Echinococcus*. Recurrent disease has been reported in 10% of patients undergoing hydatid cyst surgery [1]. Perioperative chemotherapy using

albendazole has been shown to decrease the incidence of recurrent disease [15]. However, albendazole may not prevent disease recurrence in a distant site, as in our case. After pancreatic surgery, we administered albendazole to the patient for two months and we thought that this short course of therapy might have contributed to the recurrence. We recommended albendazole treatment for six months after the breast surgery. No recurrence was detected after two years. But, we are still wondering what the primary source of the disease was and have followed the patient with annual CT images of the thorax, abdomen and even the brain. But she seems to be free of the disease. Follow-up studies should be complemented with serological tests. It increases the efficiency of the medical treatment. However, it should be kept in mind that the onset of recurrent disease is frequently asymptomatic, and clinical evaluation, even supplemented by serologic tests, may not be diagnostic [16]. Blood titers may decrease slowly over months to years even with complete removal of disease [17]. As a result, a positive serologic test during follow-up is therefore not necessarily diagnostic of recurrence but a rising titer is. The preferred methods for identifying recurrent disease, in addition to clinical and serologic evaluation, are US and CT scanning [16, 17].

CONCLUSION

Hydatid disease should be considered in the differential diagnosis of all cystic masses in all anatomic locations, especially in regions where the disease is endemic. The ideal treatment is the complete excision of the cyst without any spillage.

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