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A rare case liver hydatid cyst containing multiple calculi

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A rare case liver hydatid cyst containing multiple calculi

The hydatic cyst disease, caused by Echinococcus granulosus, is a potentially lethal, which can be found anywhere in the world, but especially in endemic areas such as the Mediterranean Basin, North Africa, Eastern Europe, the Balkans and Middle East. This parasite is mainly found in the liver (3/4 of the cases), being asymptomatic in most cases and discovered accidentally on a routine abdominal ultrasound or an ultrasound done for diagnosing other pathologies. Liver hydatid cyst threatment is multimodal, which includes medical, surgical, and interventional radiologic techniques.

KEY WORDS: Complication, Echinococcus Granulosus, Lithiasis, Liver Hydatid Cyst

Introduction

Hydatid cyst disease is a parasitic disease caused by a cestode, Echinococcus granulosus, and it most commonly involves the liver. It may also present with involvement of the lung and all other organs. It is frequently seen in developing and underdeveloped countries, in areas where agriculture and animal husbandry is intense. It is an endemic disease in Turkey [1,2]. It is known that mortality increases if the liver hydatid cyst becomes complicated. Infection of the cyst, rupture into the biliary tract causing obstructive jaundice problems caused by compression on the surrounding organs and tissues, free rupture into the abdominal cavity and allergic reactions are all among the most common complications of the hydatid cyst. In this report we present a case with multiple gallstones in liver hydatid cyst.

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Case Report

A 48-year-old male patient was admitted to the emergency department with complaints of right upper quadrant abdominal pain, epigastric fullness, vomiting and daily intermittent fever. On physical examination, tenderness was found in the right upper quadrant. There was no rebound or defense and no icterus was observed. The patient's urine and stool color were normal. Complete blood count and biochemical laboratory values ™were within normal ranges. Abdominal USG, abdominal computed tomography (CT) and magnetic resonance coloangiopancreatographic (MRCP) examination were performed. In imaging studies, a stone with a diameter of 12 mm in the gallbladder and a huge mass 105x95x90 mm in size in the right lobe of the liver were detected. The huge mass was suggested to be a hydatid cyst of Gharbi type 3-4 containing some kind of calcifications inside the cyst. The patient was operated for a large, symptomatic hydatid cyst and cholelithiasis. During the surgery, a hydatid cyst was detected that almost completely filled the right lobe of the liver and was consistent with the preoperative imaging findings. One stone was palpated in the gallbladder. A scolocidal agent, 10% NaCl, was instilled into the cyst for the cyst sterilization and waited for 10 minutes. Cyst cavity opened via a cystostomy. It was observed that there were

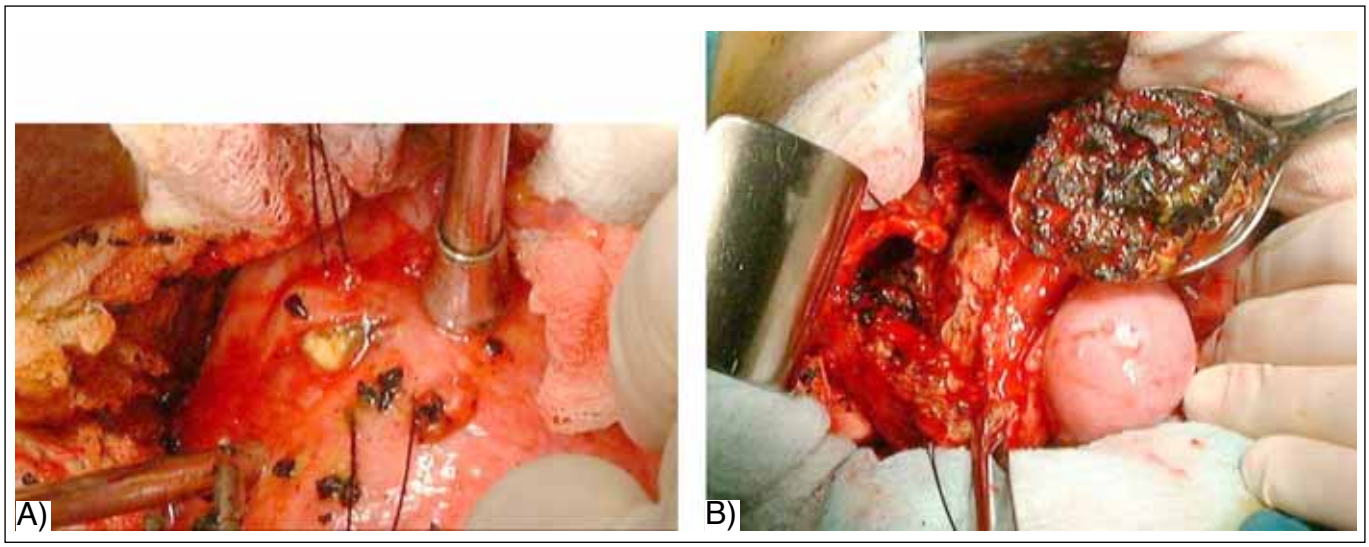

Fig. 1: A) Hydatid cyst before opening; B) Content of hydatid cyst.

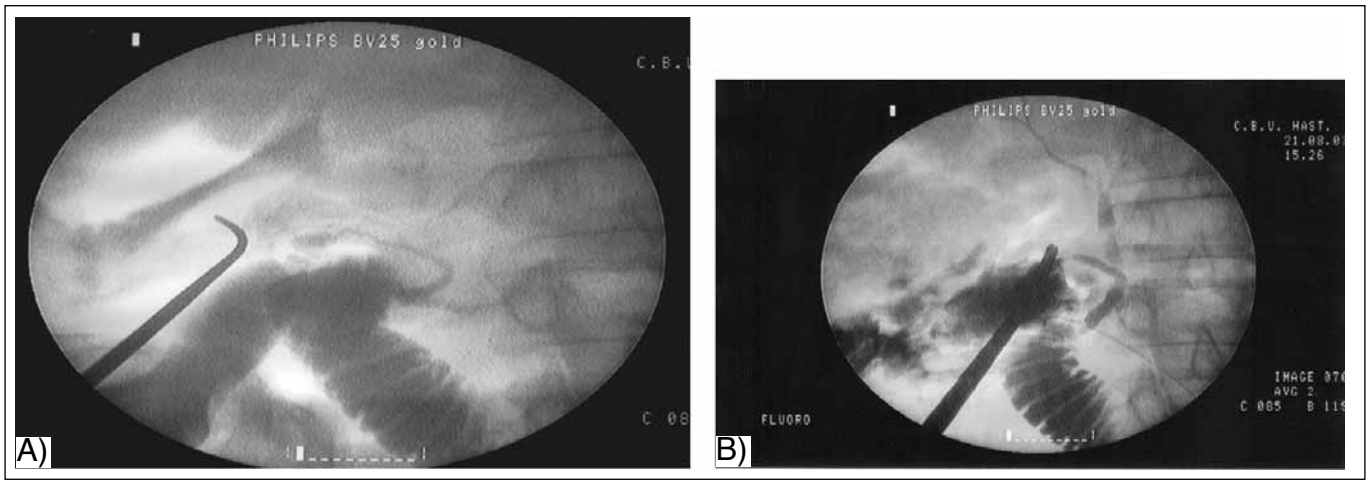

Fig. 2: A) Contrast agent fills the duodenum; B) Contrast agent fills the cyst cavity.

many stones of different sizes, biliary sludge and a liquefied germinative membrane of mucopurulent consistency stained with bile (Fig. 1A, 1B). The cyst content was evacuated. Peroperative cholecysto-cholangiography was performed through a catheter inserted through the fundus of the gallbladder in order to determine whether there was a relationship between the cyst and the gallbladder and bile ducts. After filling the bile ducts with contrast material, it was observed that the contrast first passed into the duodenum normally (Fig. 2A).
Continuing to administer the opaque material, we detected that the contrast material passed from the right hepatic duct into the cyst (Fig. 2B). On macroscopic examination, it was observed that the cyst had bile drainage through an opening of approximately 2 millimeters, in the wall of a bile duct at the base of the liver, so small that the stones in the cyst could not pass into the bile duct .This canal was repaired with primary sutures. The cyst pouch was filled with the omentum. Two drains were placed in the cyst. Cholecystectomy was also performed. The cyst material sent for pathological examination was reported as hydatid cyst with bacterial clusters and mixed inflammatory cell infiltration, and cholecystectomy material as chronic calculous cholecystitis with acute attack. In the postoperative period, 800 mg/day albendazole treatment was started. Afterwards, the patient was discharged with the recommendation of outpatient control. No problem was detected in the clinical or tomographic examination during the postoperative follow-up.

Discussion

Bile stones may occur in the intrahepatic bile ducts because of hemolytic diseases, parasites, biliary strictures,

sclerosing cholangitis, and biliary ectasia anomalies [3]. In this case we observed that multiple stones of different sizes developed in the echinococcal cyst in the liver. Most hydatid cysts are asymptomatic. However, depending on the size or location of the cyst, symptoms of compression or complications such as opening to the biliary tract and infection may occur.

The most common complication is rupture of the cyst into the bile ducts with a rate of 5-17% [4]. In our case, we observed that there was a bile duct opening to the cyst, but the contents of the cyst did not discharge into the bile ducts and did not cause obstruction in the biliary tract. Infection of cyst contents with pyogenic agents is an another major complication, which should be treated with drainage and appropriate antibiotherapy [4]. The main purpose of treatment in hydatid cyst is to remove the parasite from the body, thus preventing possible complications and recurrences [5]. For this purpose, various interventions such as interventional radiology, percutaneous aspiration- irrigation (PAIR) or surgery can be applied [6].

As surgical treatment; partial cystectomy, capitonage, marsupialization, introflexion, pericystectomy, partial liver resection or lobectomy can be performed. For the cysts that rupture into the biliary tract, common bile duct exploration with T-tube application or choledochoduodenostomy may also be preferred [7]. In our case, the hydatid cyst was of Garby type 3-4. In the perioperative imaging, there wasn't adequate liver parenchyma tissue between the cyst wall on the liver surface an. So, PAIR was not found appropriate for the treatment and open surgery was preferred. Following the cystotomy, liquefied germinative membrane was evacuated along with many stones that could not be detected clearly in preoperative imaging. The ruptured bile duct at the base of the cyst was repaired with a primary suture, and partial cystectomy and omentoplasty were performed. No postoperative complications developed in the patient and discharged with full recovery.

Conclusion

Hydatid disease is endemic in our country, Turkey. In our clinical practice, we encounter many cases of hydatid cysts and we perform these operations successfully. However, we have seen for the first time that stone formation in a liver hydatid cyst. In literature search, we found that no other similar case was reported.

It seems difficult to comment on the mechanism by which the stones detected in the hydatid cyst are formed. In our case, we did not have the chance to examine the chemical structure of the stones formed in the cyst. However, the detection of stones in the gallbladder in the patient can be interpreted as a predisposition for the formation of gallstones.

The relationship of the cyst with the biliary tract and the presence of bile in the cyst during the operation suggest that one of the mechanisms involved in the formation of gallstones may also play a role in the formation of hydatid cyst stones. On the other hand, although we have encountered many cases of hydatid cyst associated with the biliary tract, the fact that we have not seen any stone formation in the cyst before, so that it can be interpreted as a different mechanism from the classical gallstone formation mechanisms may also play a role. In conclusion, this is the first case in the literature stating that gallstones were detected in a hydatid cyst.

Riassunto

La malattia della cisti idatidea è una malattia parassitaria causata da un cestode, Echinococcus granulosus, e coinvolge più comunemente il fegato. Si tratta di aree endemiche come il bacino del Mediterraneo, il Nord Africa, l'Europa orientale, i Balcani e il Medio Oriente. La minaccia della cisti idatidea epatica è multimodale, che include tecniche mediche, chirurgiche e radiologiche interventistiche. Un paziente maschio di 48 anni è stato valutato al pronto soccorso a causa di dolore addominale e gli è stata diagnosticata una cisti idatidea epatica. Il paziente, risultato non idoneo all'intervento mininvasivo dopo gli esami, è stato portato all'intervento chirurgico. Durante l'intervento chirurgico sono stati osservati numerosi calcoli biliari nella cisti idatidea epatica. C'era un dotto biliare di 2 mm aperto nella cisti. Nella nostra revisione della letteratura, è stato visto che non c'erano altri casi di calcoli biliari nella cisti idatidea. La malattia della cisti idatica, causata da Echinococcus granulosus, è una malattia potenzialmente letale, che può essere diffusa ovunque nel mondo, ma soprattutto in aree endemiche come il bacino del Mediterraneo, il Nord Africa, l'Europa orientale, i Balcani e il Medio Oriente. Questo parassita si localizza principalmente nel fegato (3/4 dei casi), nella maggior parte dei casi è asintomatico e viene scoperto accidentalmente da un'ecografia addominale di routine o un'ecografia eseguita per la diagnosi di altre patologie. Il trattamento della cisti da echinococco del fegato è multimodale, e comprende trattamenti farmacologici, chirurgici e radiologici interventistici.

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Commento e Commentary

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La fistolizzazione dell'albero biliare in una cisti idatidea è un fenomeno abbastanza frequente, e spesso è la condizione che apre un quadro sintomatologico fin al momento silente. La bile è tossica per l'hydatide e quando entra in contatto con la membrana germinativa può provocarne la morte, con conseguente infezione suppurativa, distacco del pericistio e fenomeno della ninfea visibile con US. Raramente la fistolizzazione può avvenire con la colecisti come nell'articolo citato come reference, e in tal caso è comprendibile il pur raro ritrovamento di calcoli biliari anche all'interno della cavità cistica per trasposizione.
Il caso descritto dagli Autori non è invece facilmente spiegabile, perché data l'esiguità di soli 2 mm del dotto biliare fistolizzato, non si può che ipotizzare la formazione di una lenta raccolta biliare all'interno della cisti, che coagulandosi attorno dei nuclei litogeni abbiano dato la formazione di calcoli multipli dimostrati nell'iconografia. L'unicità della osservazione non consente di andare oltre un'ipotesi.

* * *

Fistulization of the biliary tree in a hydatid cyst is a fairly frequent phenomenon, and it is often the condition that opens the clinical symptomatology, otherwise total silent for a long time. Bile is toxic to hydatide and when it comes into contact with the germinative membrane it can cause its death, resulting in suppurative infection, detachment of the pericystic and water lily phenomenon visible with US. Fistulization can rarely occur with the gallbladder as in the article cited as a reference, and in this case it is understandable the rare finding of gallstones even within the cystic cavity by transposition.
The case described by the authors is not easy to explain, because given the smallness of only 2 mm of the fistulized bile duct, one can only hypothesize the formation of a slow biliary collection inside the cyst, which coagulating around lithogenic nuclei gave the formation of multiple calculations demonstrated in iconography. The uniqueness of the observation does not allow us to go beyond a hypothesis.

Reference

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