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Fistulization between liver hydatid cyst and gallbladder. A case report and review of the literature

*The most serious complications of liver hydatid cyst disease are fistulization into biliary tract, compression of adjacent vascular structures, anaphylactic reaction, and perforation. Fistulization between liver hydatid cyst and gallbladder tract is an extremely rare complication with only a few cases reported so far. Herein, we aimed to report a 43-year-old man who was diagnosed as having a cholecysto-hydatid cyst fistula. The patient presented to emergency department with signs and symptoms of cholangitis. His biochemical tests revealed elevated AST, ALT, GGT, and bilirubin levels. The radiological examinations (CT, MRCP) revealed a lesion consistent with hydatid cyst (Hydatid cyst ELISA IgG +) with an approximate size of 90*65 mm, which was posterolateral to the gallbladder and fistulized into the latter. In order to relieve pressure within the biliary tract, ERCP with sphincterotomy was performed. He was taken to the operating room a few days later. After draping sponges soaked with 3% NaCl onto the surgical field, near-total pericystectomy + omentopexy + cholecystectomy + common bile duct exploration + T-tube drainage were performed. Bile duct opening to the posterior wall of the cyst was sutured with a prolene suture. Albendazole treatment was started on first postoperative day. After taking a cholangiogram on 21st postoperative day, the T-tube was removed without any complication. In conclusion, cholecysto-hydatid cyst fistula is an extremely rare complication of hydatid cyst disease even in endemic regions. The gold standard for the diagnosis is the combined use of characteristics of clinical presentation, biochemical parameters, and radiological studies. Treatment plan is designed on the basis of the relationship of a fistulized cyst with other bile ducts.*

KEY WORD: Cholecysto-Hydatid Cyst Fistula, Gallbladder, Hydatid Cyst, Liver

Introduction

Hydatid disease is a zoonotic disease caused by parasites of echinococcus species belonging to the taeniidae family of the cestode class. E. Granulosus causing Cystic echinococcosis is responsible for 95% of hydatid disease affecting humans. Having no role in the biological cycle of the disease, humans are mostly accidentally infected by ingesting echinococcus eggs found in dog feces. Oncospheres released from ruptured eggs in the intestinal lumen adhere to intestinal mucosa with their hooks and pass to the portal circulation. Larvae passing to hepatic sinusoids through the portal vein may reside in almost all human tissues and organs, most notably liver and lungs. Larvae passing through certain maturation stages form cystic lesions in various organs. As cystic lesions grow slowly, they are incidentally detected by radiological studies performed for other indications. However, complications such as infection, anaphylaxis, compression of adjacent organs, free perforation, and fistulization may develop in a minority of patients. Liver hydatid cysts may fistulize into biliary tract, gastrointestinal tract, bronchi, and pleural cavity. Cholecysto-

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hydatid cyst fistula development is an extremely rare complication of hepatic hydatid cyst disease [1-7]. In this paper we aimed to report a case of cholecysto-hydatid cyst fistula in the light of the relevant literature.

Case Report

A 43-Year-Old Man Presented to emergency department with right upper quadrant pain that had started 5 days earlier. He stated that he had undergone right nephrectomy for an unknown indication years ago but denied having any other chronic health condition. His biochemical analysis in the emergency department revealed the following: AST: 137 U/L (normal range: 5-34), ALT: 237 U/L (normal range: 0-55), GGT: 605 U/L (normal range: 9-64), ALP: 132 U/L (normal range: 38-155), T Bil: 1.37 mg/dL (normal range: 0.2-1.2), BUN: 12 mg/dL, Creatinine: 0.97 mg/dL and WBC: 8500/ ml (4.3-10.3). A contrast-enhanced dynamic abdominal computed tomography (CT) taken in the emergency room revealed a hydropic gall bladder with increased mural thickness, a common bile duct diameter of 14 mm, a defect in the posterolateral position of the bladder neck, and a cystic lesion with septae with a diameter of 77*65 mm related to that defect (abscess formation?) (Fig. 1 A-B). A dynamic magnetic resonance imaging (MRI) and a magnetic resonance cholangiopancreatography (MRCP) were performed to evaluate the relationship of the cyst with both liver parenchyma and biliary tract. T2A weighted images of MRCP revealed a septated cystic lesion with a size of 90*60*65 mm and a hyperintense-hypointense heterogenous content; the lesion showed an anatomic relationship with gall bladder neck at posterolateral localization (Fig. 2 A-B). When both images and the previously obtained abdominal ultrasonogram were evaluated together, a conclusion was reached that the lesion in question may be a hydatid cyst fistulized into gallbladder. Additionally, an Echinococcus ELISA Ig G test was performed, which returned positive (12.83). As the cyst fistulized into the gall bladder and the diameter of the common bile duct was increased, an ERCP was performed. The latter showed no daughter vesicles in the biliary tree, and thus the procedure was terminated after a sphincterotomy was performed. In the light of the available information, the patient was put on andazole treatment and preoperative preparations were arranged for surgery. The abdominal cavity was entered through a right subcostal incision. Sponges soaked in 3% NaCl solution were draped around the surgical field. As bile was noted to ooze upon cyst puncture, NaCL was not injected into the cyst. A hole was opened in the cyst's anterior wall and whole cyst content was evacuated, followed by near-total pericystectomy. A fistula tract of at least 3 cm in length was noticed between the cystic lesion and the gallbladder. Additionally, a bile duct with a diameter of 4-5 mm opened into the cyst's posterior inferior wall. First, the orifice of the bile duct opening into the cystic lesion was sutured with prolene sutures, and then cholecystectomy was performed. A leak test done through the cystic canal confirmed the absence of any other bile leakage. After performing choledochotomy, both proximal bile ducts and distal common bile duct were explored and the absence of any daughter vesicle in the biliary tract was confirmed. Then, a T-tube was placed into the common bile duct and the operation was completed. Albendazole was begun on the first postoperative day.

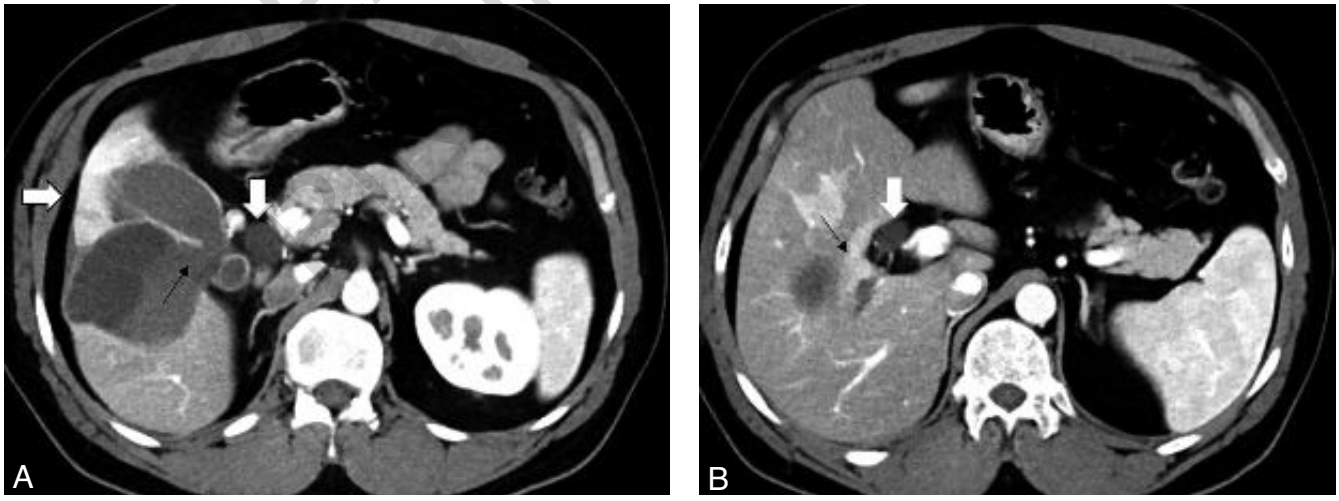

Fig. 1: Contrast –enhanced CT images taken in portal phase. A) the close relationship of the gall bladder and the hydatid cyst and the loss of integrity in the medial wall of the neck of the gall bladder (thin black line). The enhanced contrast uptake due to inflammation in segment five of the liver was shown with thick white arrow. Additionally, there is increased contrast uptake in the gall bladder and cystic canal's wall; B) The hydatid cyst opens into posterior segment biliary tract and there is increased contrast enhancement due to cholangitis in liver parenchyma in that region (thin black arrow). Dilated common bile duct is shown by thick white arrows.

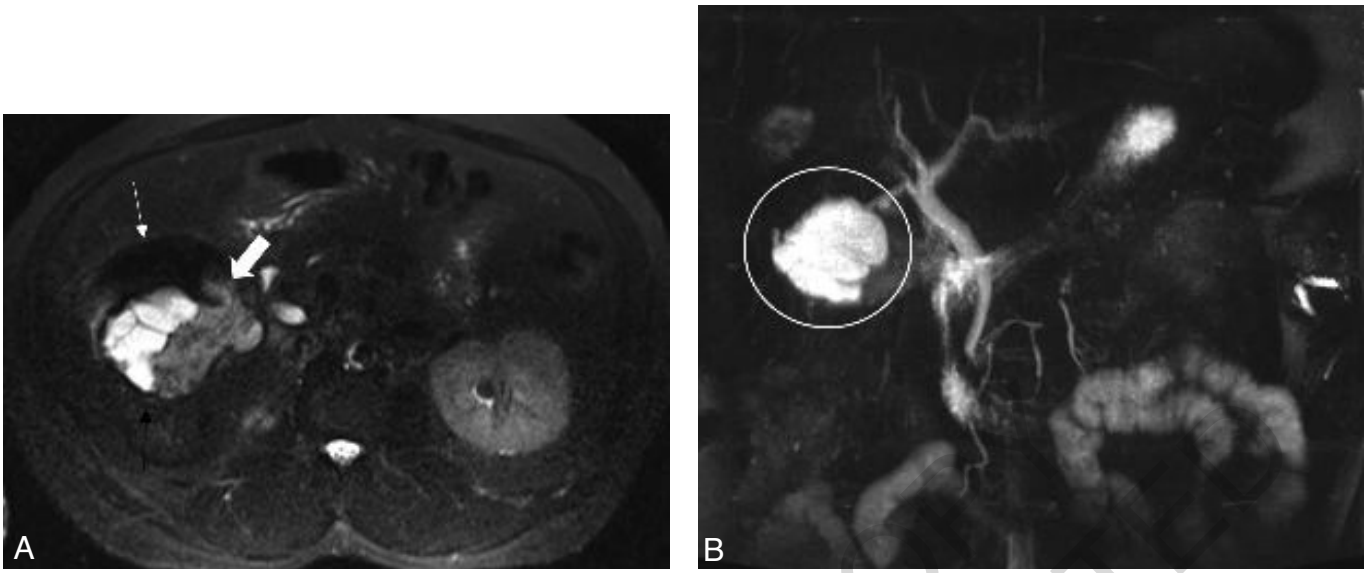

Fig. 2: T2 Haste and MRCP images. A) Gall bladder (thin white arrow) Hydatid cyst (thin black arrow) and the common content seen in both cavities (thick white arrow); BA) MRCP reformant image shows hydatid cyst cavity nearly totally filled up with bile (white circle).

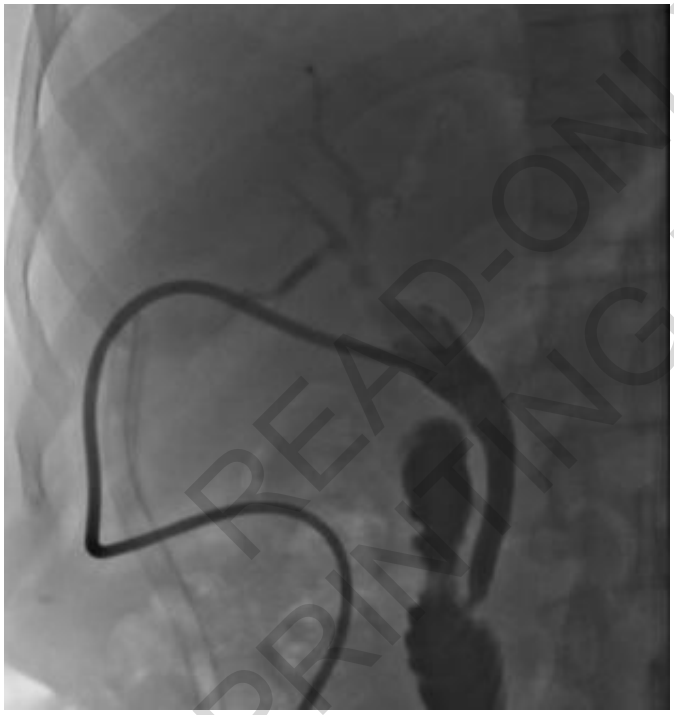

Fig. 3: Postoperative Cholangiographic image. A) cholangiogram taken by administering contrast agent through the T-tube shows cessation of biliary leak and the absence of filling of hydatid cyst cavity.

Bile leak was excluded by T-tube cholangiography on the 5th postoperative day, and the patient was discharged thereafter. The T-Tube was removed after a cholangiogram was taken on the 21st postoperative day (Fig. 3). The patient received albendazole for a total of 4 weeks. Graphic-1 depicts the changes in the serum laboratory measurements.

Discussion

Hydatid cyst disease is a zoonotic disease that poses a serious public health threat in the Middle East, Far East, Mediterranean, South America, Australia and Turkey, where agriculture and stockbreeding is the primary means of living. Hydatid cyst disease most commonly involve the liver, lungs, spleen, and kidneys [8-10]. However, pancreas, brain, retroperitoneal cavity, prostate, soft tissue, abdominal wall, and orbita are also involved, albeit to a lesser rate. Despite its close anatomic relationship with the liver, the gallbladder is one of the organs that are extremely rarely involved by hydatid cyst disease [8-10].
Hydatid cyst disease of the gallbladder mostly emerges secondary to the complications of liver hydatid disease, but it may also rarely occur as a primary disease. A number of hypotheses have been postulated for this subject. The first one hypothesizes that daughter vesicles, which pass into the biliary tree upon cyst rupture, enter the lumen of the gall bladder through the common bile duct. Secondary hydatid cyst disease of the gall bladder is most widely explained by this hypothesis. The second hypothesis is related to the passage of oncospheres into the lymphatic system through the intestinal wall, which then proceed upwards via lymphatic channels in the hepatoduodenal ligament [10]. Primary hydatid cyst disease of the gallbladder is most widely explained by this hypothesis. The third hypothesis involves the contamination of the gallbladder during hydatid cyst surgery. The fourth hypothesis states the development of a cholecysto-hydatid cyst fistula as a result of the erosion of gall bladder wall by hydatid cysts adjacent to the gall bladder.
Approximately 80% of hydatid cysts located in the liver tend to be localized to the right lobe of liver, most-

TABLE I - *A summary of fistulization between Liver Hydatid Cyst and Gallbladder reported in the English language literature until 2018*

References	Age/ Sex	Radiologic Tools	Total number of Liver Cyst	Fistulized Cyst Location	Fistulized Cyst Size (mm)	Surgical Approaches	Postop Treatment (mo)
Luks	50/F	US+CT	Three	Segment IV	63	Pericystectomy + Cholecystectomy	Albendazole (6 mo)
Benkoukous	31/F	US+CT	One	Segment IV	75*76	Evacuation + Cholecystectomy	NS
Wani*	65/F	US+MRCP+ERCP	One	Segment IV/V	45	Evacuation + Cholecystectomy + Choledochoduodenostomy	Albendazole (NS)
Sabat	35/F	US+CT	One	Segment IV/V	100*88	Partial pericystectomy + Cholecystectomy	Albendazole (9 mo)
Murtaza	32/F	US	One	Right Lobe	110*105	Evacuation + Subtotal cholecystectomy	Albendazole (NS)
Adaletli	46/M	US+MRI +MRCP+ERCP	One	Segment V	50*40	NS	NS
Kumar	27/F	CT	Two	Segment IV		Partial pericystectomy +Cholecystectomy	Albendazole (NS)
Present Case	43/M	US+CT+MRI	One	Segment V	90*65	Partial pericystectomy +Cholecystectomy CBD exploration with T-tube insertion	Albendazole (1 mo)

*Fistulization occured both between hydatid csyt and right hepatic duct and between hydatid cyst and gallbladder, US: ultrasonography, CT: Computed tomography, MRCP: Magnetic resonance cholangiopancreatography, MRI: Magnetic resonance imaging, ERCP: Endoscopic retrograde cholangiopancreatography, NS: No-stated

ly the peripheral part of the latter. Hydatid cysts grow by approximately 5-10 mm per year, explaining why an important percentage of affected individuals remain asymptomatic for years. However, a smaller percentage of patients may become symptomatic after developing a number of complications such as secondary infection, anaphylaxis, vascular compression, compression of adjacent organs, and development of fistulae (rupture) [8]. Fistulization into biliary tract (5-25%), bronchi, peritoneal cavity, and pleural cavity is the most common major complication [6-8].

Cholecysto-hydatid cyst fistula is an extremely rare complication. A literature search in the PubMed, Google Scholar, and Google databases using the definitions "fistulization between gallbladder and hydatid cyst", "rupture of hydatid cyst into the gall bladder", and "cholecysto-hydatid cyst fistula" revealed a total 10 article titles. Table 1 summarizes the characteristics of 7 case reports with accessible full text versions [1-7].

Some patients with cholecysto-hydatid cyst fistula may remain asymptomatic or have nonspecific symptoms and signs. This is because for a patient to become symptomatic, the diameter of a bile duct with which there exists a communication should be ≥5 mm [7]. The majority of patients may present with signs and symptoms indicative of cholangitis, such as fever spikes after chills, upper right quadrant pain, and jaundice. The clinical presentation is much more severe when numerous cystic components pass into bile ducts, obstructing the latter. Similar clinical signs and symptoms may appear when a fistula develops between the gall bladder and a hydatid cyst. In cases where a cyst is in relation with a large bile duct, as the case we present in this paper, the signs and symptoms of cholecystitis and cholangitis may coexist. We would like to bring to the attention of the readers a question Luks *et al.* once asked: does cholecystitis develop as a result of a fistula or fistula develops secondary to the attacks of cholecystitis? [1].

As in other liver hydatid cyst diseases, cholecysto-hydatid cyst fistula is diagnosed with radiological tools, including US, CT, MRI/MRCP. ERCP is a very valuable tool that both demonstrates the relationship of cystic lesions with biliary tract and is used for decompression of biliary tract [5]. The treatment algorithm of cholecysto-

hydatid cyst fistula depends on the clinical condition of a patient (cholangitis, cholecystitis), comorbid conditions, relationship of a hydatid cyst with other bile ducts, the diameter of the common bile duct, and the presence of cyst elements in biliary system in preoperative MRCP/ERCP. We may summarize the treatment algorithm we recommend for patients with cholecysto-hydatid cyst fistula presenting with signs and symptoms of acute cholangitis/cholecystitis as follows:

(i) In patients with good overall condition in whom there are no cystic components in biliary tract in MRCP/CT/ERCP the most appropriate treatment approach is cholecystectomy + drainage of fistulized cyst + excision of cyst wall as much as possible. Whether a bile duct opens into cyst cavity should be confirmed with a leak test. In cases where a leak is found, the orifice of the bile duct should be closed with a prolene suture. When the diameter of a bile duct fistulizing into the cyst cavity is equal to or greater than 5 mm or when the diameter of the common bile duct is enlarged, common bile duct exploration and T-tube drainage should be carried out [5]. The most appropriate approach for patients with elevated postoperative bilirubin, ALP, GGT levels or drainage from the drainage catheter is reduction of bile tract pressure by performing sphincterotomy with ERCP.

(ii) In patients with confirmed presence of cyst elements in biliary tract by MRCP/CT, either the above-mentioned principles are applied or the biliary tract is decompressed with ERCP in order to prepare patients for a definitive surgery. The definitive surgical treatment to be performed under the coverage of neoadjuvant antihelminthic therapy is as described above.

(iii) In patients who are deemed to have a poor medical condition to tolerate any urgent surgery, the most appropriate approach is the percutaneous decompression of both the gallbladder and the cystic lesion, followed by decompression of biliary tract by ERCP if possible. In this method the relationship of lesions with biliary tree can be clearly demonstrated. The definitive treatment is applied as described above once the overall status of a patient is improved. As it is impossible to administer any neoadjuvant antihelminthic therapy to patients undergoing urgent surgical or percutaneous drainage, only postoperative antihelminthic treatment can be applied. In patients who present with mild cholangitis or cholecystitis, who were administered antibiotherapy, and who were confirmed to have a cholecysto-hydatid cyst fistula, neoadjuvant antihelminthic therapy should be necessarily begun when an elective surgery is planned. Elective surgery is performed as described above. Administering neoadjuvant antihelminthic therapy [albendazole (10mg/kg/d), mebendazole (4,-50 mg/kg/d), praziquantel (20-75 mg/kg/d)] to all patients prior to surgical (2 weeks) or percutaneous drainage (1 week) reduces the risk of anaphylactic reaction during the procedure, reduces intracystic pressure, and facilitates the performance of the procedure, and reduces the postoperative recurrence rates [9]. Postoperative neoadjuvant antihelminthic therapy is unnecessary when there is no relationship between a cyst and bile ducts other than gallbladder and there is no cystic component in biliary tract on preoperative radiological tests (MRCP, ERCP, CT) and/or during exploration, or when a cyst is near-totally excised without leaving any germinative membrane behind. In all cases except the ones described above we suggest the administration of albendazole (at least 1 month) or mebendazole (at least 3 months), either continuously or in cycles. We routinely administer albendazole in our clinic.

In conclusion, cholecysto-hydatid cyst fistula is an extremely rare complication of hydatid cyst disease even in endemic regions. The gold standard for the diagnosis is the combined use of characteristics of clinical presentation (fever, chills, jaundice, upper right quadrant pain, biochemical parameters (WBC; bilirubin, ALP, GGT), and radiological studies (MRCP, CT). Treatment plan is designed on the basis of the relationship of a fistulized cyst with other bile ducts. The most appropriate approach is the excision of the gall bladder with the fistulized cyst after taking necessary measures.

Riassunto

Le complicanze più gravi dell'idatidosi epatica sono la fistolizzazione nelle vie biliari, la compressione delle strutture vascolari adiacenti, la reazione anafilattica e la perforazione.

La fistolizzazione tra cisti idatidea epatica e parete colecistica è una complicazione estremamente rara, con solo pochi casi segnalati finora.

Qui si presenta il caso di un uomo di 43 anni a cui è stata diagnosticata proprio una fistola tra cisti idatidea e colecisti, presentatosi al pronto soccorso con segni e sintomi di colangite.

I test biochimici presentavano livelli elevati di AST, ALT, GGT e bilirubinemia. Gli esami radiologici (TC, MRCP) hanno rivelato una lesione compatibile con cisti idatidea (confermata con ELISA IgG +) delle dimensioni di circa 90 x 65 mm, situata postero-lateralmente alla cistifellea e in essa fistolizzata. Per alleviare la pressione sulla vie biliari è stata eseguita una sfinterotomia mediante ERCP. Qualche giorno dopo il paziente è stato portato al tavolo operatorio. Dopo tamponamento con garze imbevute con soluzione di NaCl al 3%, è stata eseguita una peristectomia quasi totale, una omentopessi ed una colecistectomia con esplorazione dell'epato-coledoco e drenaggio con tubo di Kehr. Il dotto biliare aperto sulla parete posteriore della cisti è stato suturato con una sutura in prolene. Dal primo giorno postoperatorio è stato iniziato un trattamento con albendazolo. Previa colangiografia il 21° giorno postoperatorio è stato asportato il tubo di Kehr senza complicazioni.

In conclusione si conferma la rarità della fistola cisti idatidea-colecisti anche in regioni endemiche. La diagnostica gold standard è fondata sulle caratteristiche della presentazione clinica, sui parametri biochimici e sugli studi radiologici. Il piano del trattamento chirurgico va formulato sulla base dei rapporti della cisti fistolizzata con altri dotti biliari.

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