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Case Report

Gallbladder Hydatid Disease Complicated with Multiple Hepatobiliopancreatic Fistulae: A Case Report

*Gökhan Pösteki

Kocaeli State Hospital, Kocaeli, Turkey

<p>Received 15 Apr 2023 Accepted 11 Jul 2023</p>	<p>Abstract Gallbladder is a rare localization for hydatid disease. Complications are even rarer and precise diagnosis is quite difficult even with radiological assistance. We report a rare case of 41-yr-old male patient presenting with the rupture of a gallbladder hydatid cyst with multiple fistulae to intra and extrahepatic bile ducts and pancreas, at Kocaeli State Hospital, Turkey in 2021. The patient had abdominal pain and abdominal CT scan reported a bizarre “contrast enhanced cholangiography” sign - radiopaque contrast substance in gallbladder, intra and extrahepatic bile ducts and pancreatic ducts, with concomitant acute cholecystitis. Surgery was performed and intraoperatively gallbladder hydatid cyst with multiple fistulae was noted. Cholecystectomy with total cyst excision was performed. Endoscopic retrograde cholangio pancreatography (ERCP) was utilized to irrigate and eradicate the parasite in the fistulae tracts localized near pancreas and intra/extrahepatic ducts. Postoperative period was uneventful, antiparasitic treatment was started, and in the yearly follow-up patient had no recurrence. Multidisciplinary and minimal invasive management is crucial in such bizarre, complicated cases.</p>
<p>Keywords: Gallbladder; Hydatid disease; Parasitology</p>	
<p>*Correspondence Email: gokhanpostekidr@hotmail.com</p>	

Introduction

Hydatid disease (HD) usually affects liver and lungs, caused by the parasite *Echinococcus granulosus*. Liver localization may lead to rupture into the bile

ducts as seen in some cases (1). Such situations can cause major clinical problems during the follow-ups like prolonged biliary fistula. Likewise, infrequently hydatid cysts may also



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invade gallbladder and pancreas, affect the organs, and lead to organ dysfunctions. Concomitant intrahepatic bile duct, gallbladder, pancreatic duct and common bile duct invasions of the hydatid disease are extremely rare (1). This combination will most likely deter the situation in a swift fashion, leading to cholangitis, jaundice, cholangiosepsis and most importantly anaphylaxis. Therefore, proper diagnosis in biliary tract related hydatid disease is crucial.

In this case report, we aimed to present a bizarre and extreme case of gallbladder hydatid disease with unusual radiologic findings and its surgical treatment to widen the literature with an exceptional case. Informed consent and ethics approval were granted from hospital and the patient.

Case presentation

A 41-yr-old male patient without any chronic diseases, presented to Emergency Depart-

ment of Kocaeli State Hospital, Turkey with a complaint of severe abdominal pain in 2021. His history revealed that he had abdominal discomfort and nausea symptoms for the last 2 months.

Physical examination revealed tenderness in the upper abdomen. White blood count and C-reactive protein levels were elevated. Abdominal CT scan reported radiopaque contrast substance in gallbladder, intra and extrahepatic bile ducts and pancreatic ducts with concomitant acute cholecystitis (Fig. 1).

These findings were noted as contrast excretion to the bile ducts by the radiology consultant, which was usually seen after contrast enhanced cholangiography. The patient had no history of prior radiological imaging or IV/oral contrast administration. After this dilemma, radiology and hepatobiliary surgery teams had a meeting for this case and the common decision was surgical exploration, as acute cholecystitis with positive examination findings was the preoperative diagnosis.

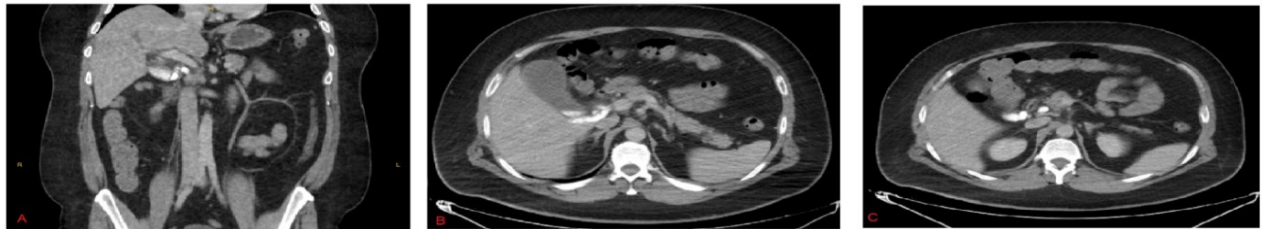


Fig. 1: Gallbladder, intrahepatic/extrahepatic biliary tract and pancreatic ducts filled with radiopaque “contrast” like hydatid cyst contents, mimicking “contrast enhanced cholangiography” – Preoperative CT scan

Diagnostic laparoscopy confirmed the acute cholecystitis, with severe gallbladder distension and wall thickening. During cholecystectomy, the gallbladder wall was ruptured by the laparoscopic manipulations. As it was ruptured, viscous white cystic contents and tissues with bile sludge were seen inside the gallbladder (Fig. 2). A frozen section was performed and according to evaluation, the cystic lesion from gallbladder was hydatid cyst. This explained the pre-operative radiopaque view in the gallbladder and bile ducts. Cholecystecto-

my was performed without further spillage. The operation lodge was irrigated with 20% NaCl solution. Preoperative endoscopic retrograde cholangiopancreatography (ERCP) was performed and common bile duct was irrigated with 0.9% NaCl solution.

Postoperative final pathology analysis supported the frozen section results. There were no postoperative complications after surgery. Eight hundred mg/day albendazole treatment was started on postoperative day 1. The patient was discharged on the postoperative day

10. Albendazole was continued for 12 months. There were no radiological and clinical recur-

rence, therefore it was decided to follow up the patient without any medications.

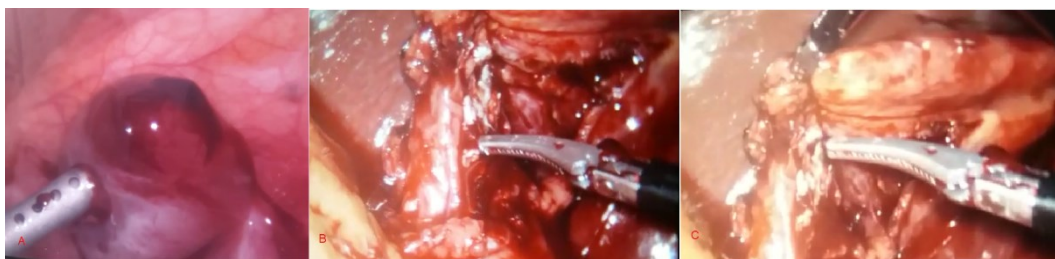


Fig. 2: A: Peroperative view of the gallbladder cystic lesion B: Cholecysto-choledocal fistula of the hydatid disease C: Gallbladder and cystic duct from a wide angle - preoperative laparoscopic images

Discussion

Clinical history, radiology and blood test results may help diagnosis of the hydatid cyst ruptures. Although most cases with gallbladder HD are asymptomatic, abdominal mass, nausea, vomiting, flank pain may be seen among some cases (2). Some of these were also the main complaints of our patient.

Gallbladder hydatid cyst is unusual with an incidence of 0.3–0.4% of all atypically located hydatid cysts (2). In our case, the hydatid cyst originated from the gallbladder, and even rarer, it invaded the intrahepatic/extrahepatic ducts and pancreatic duct, and made fistulae between them.

Diagnostic tools such as MRI and CT scans are very supportive in the diagnosis of hydatid cysts (3). Sometimes exact localization of gallbladder related hydatid cysts may be challenging (2, 4). The precise localization and extensions of complicated hydatid disease in some cases is confirmed only intraoperatively, for example in this case. Invasions and fistulae from the gallbladder to intrahepatic/extrahepatic ducts and pancreatic duct made our case an extreme anatomical challenge for the radiologists, therefore the initial ultrasound and computed tomography scans may be misleading as in our case (4). The radiopaque characteristics of the hydatid cyst contents may be the explanation. Radiologic findings may be inconsistent and the diagnosis

may be have to correct during surgery (5). This was our first case regarding a hydatid cyst with multiple fistulae in our practice of 112 patients with hydatid disease.

Preoperative differential diagnosis is crucial. Complicated cholecystitis, common bile duct neoplasms, cholangiocarcinomas, bile duct cystic neoplasms, pancreatic tumors, hepatic abscess, gallbladder polyps should all be considered in the differential diagnosis of biliary tree hydatid cysts (6, 7). Serology is also a beneficial tool for differential diagnosis as well as follow-up after treatment (8, 9). ELISA and indirect hemagglutination antibody assay are among the most universally used techniques. Our case presented in an emergency setting and had the pre-operative diagnosis of acute cholecystitis, so we could not utilize such serological tests. The final diagnosis is confirmed by surgical and histopathological examination, whereas frozen section pathology assessment can also be useful when there is pre-operative confusion. In our case, both frozen section and final histopathological evaluation after excision showed that the lesion was hydatid cyst.

Surgery is the desired treatment for complicated biliary tree hydatid diseases (8, 10, 11). The most common therapeutic approach in the present review is open surgery but recently, laparoscopic surgeries are shown to be safe and feasible for the surgical treatment of hepatobiliary hydatid disease with the ad-

vantage of minimal invasive surgery (11, 12). We performed a minimal invasive method and supported it with other minimal invasive methods like ERCP, to prevent radical and harsh treatment protocols. Before, during or after surgery, ERCP may be chosen as a minimal invasive method to irrigate and eradicate the residue parasite, with an additional good effect of preventing possible obstruction in the biliary tract (13). In surgery, it is essential to eradicate the parasite without spillage of the cyst content, in a meticulous manner (2). Hypertonic saline may be used for irrigation during surgery as in our case (14).

However, some gallbladder-originated cysts may be adjacent to duodenum and large vascular structures, reliant on the site; therefore, it may not be probable to eliminate the disease. In such cases complete per cystectomy is not always possible, and therefore partial per cystectomy or other conservative approaches are generally used among complicated cases. In selected cases, the cyst content can be drained with putting an omentum patch for the cavity, which can help absorption of the liquid content and reducing the risk of secondary infections (2). Percutaneous (PC) treatment is likewise a minimal invasive method, it can support surgery or it can be used as the sole method for the treatment, in selected cases (15). PC methods severely depends on radiology consultants' experience, and may not be applicable in most settings. Complicated cases may sometimes require conservative strategies; this may be the most adequate scenario where PC fits best. Antiparasitic treatment (albendazole, mebendazole) can also be given to reduce recurrence (8).

Conclusion

Primary hydatid disease in gallbladder with multiple fistulae to biliary tree and pancreas is an unusual setting even in endemic areas. Clinical assessment consisting of history, physical examination and serological tests can be valu-

able. Imaging modalities can provide useful data, as management of such patients requires collaboration between radiology, infectious diseases and surgery departments. Percutaneous methods and ERCP are minimal invasive approaches, which are supportive in applicable cases. Conservative ways and means like "watch and wait" may be an option but in complicated cases, contemporary options include minimal invasive surgery - which is proven effective as traditional open and radical surgery is in most cases.

Conflict of interest

The author declares that they have no conflict of interest.

References

1. Bilgi Kırmacı M, Akay T, Özgül E, et al. Cholecysto-Hydatid Cyst Fistula: A Rare Cause of Cholangitis. *Am J Case Rep.* 2020;21:e921914.
2. Yagnik VD, Dawka S, Patel N. Gallbladder Hydatid Cyst: A Review on Clinical Features, Investigations and Current Management. *Clin Exp Gastroenterol.* 2020;13:87-97.
3. Polat P, Kantarci M, Alper F, et al. Hydatid disease from head to toe. *Radiographics.* 2003;23(2):475-94.
4. Kumar R, Reddy SN, Thulkar S. Intrabiliary rupture of hydatid cyst: diagnosis with MRI and hepatobiliary isotope study. *Br J Radiol.* 2002;75(891):271-4.
5. Aghajanzadeh M, Ashoobi MT, Hemmati H, et al. Intrabiliary and abdominal rupture of hepatic hydatid cyst leading to biliary obstruction, cholangitis, pancreatitis, peritonitis and septicemia: a case report. *J Med Case Rep.* 2021;15(1):311.
6. Anderson MA, Bhati CS, Ganeshan D, et al. Hepatobiliary mucinous cystic neoplasms and mimics. *Abdom Radiol (NY).* 2023;48(1):79-90.
7. Saldaña C, Bolado F, González de la Higuera B, et al. Biliary stenosis secondary to fistulised hydatid cyst that mimics Klatskin tumour. *Gastroenterol Hepatol.* 2020;43(8):455-6.

8. McManus DP, Zhang W, Li J, et al. Echinococcosis. *Lancet*. 2003;362(9392):1295-304.
9. Sachar S, Sachar S, Goyal S, et al. Uncommon locations and presentations of hydatid cyst. *Ann Med Health Sci Res*. 2014;4 (3):447–52.
10. Kumar A, Upadhyaya DN, Singh S et al. Cholecysto-hydatid cyst fistula. *Indian J Gastroenterol*, 2004; 23(2): 76–7.
11. Bektasoglu HK, Hasbahceci M, Tasci Y, et al. Comparison of laparoscopic and conventional cystotomy/partial cystectomy in treatment of liver hydatidosis. *BioMed Res Int*. 2019; 2019:1212404.
12. Ertem M, Aytaç E, Karaduman Z. Cystic hydatid disease of the gallbladder. *Turk J Gastroenterol*. 2012;23(6):825–6.
13. Hamza A, Krasniqi A, Sada F, et al. ERCP treatment of obstructive jaundice caused by hydatid cyst in extrahepatic ducts 13 years after liver hydatid endocystectomy. A case report. *Int J Surg Case Rep*. 2020;74:38-41.
14. Sharafi SM, Sefiddashti RR, Sanei B, et al. Scolicidal agents for protoscolices of *Echinococcus granulosus* hydatid cyst: Review of literature. *J Res Med Sci*. 2017;22:92.
15. Mönnink GLE, Stijnis C, van Delden OM, et al. Percutaneous Versus Surgical Interventions for Hepatic Cystic Echinococcosis: A Systematic Review and Meta-Analysis. *Cardiovasc Intervent Radiol*. 2021;44(11):1689-96.