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RESEARCH ARTICLE

CASE REPORT OF OBSTRUCTIVE CHOLANGITIS CAUSED BY HYDATID CYST MEMBRANES

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Abbreviations
NKDFA, not known drug or food allergy; ERCP,endoscopic retrograde cholangiopancreatography; RUQ, right upper quadrant; PPI, proton pump inhibitor; PO, per oral; CBD, common bile duct; LFT's, liver function tests; IV, intravenous; IVD, intravenous drip; PMH, past medical history; PSH, past surgical history; BMI, body mass index; US, ultrasonography; MRCP, magnetic resonance cholangiopancreatography.

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ABSTRACT

Introduction: Biliary obstruction, that affect around 5 cases per 1000 people in the united states, are due to several causes, with the Gallstones presenting the most common cause. Of parasitic causes, adult Ascaris lumbricoides, eggs of certain liver flukes (e.g. Clonorchis sinensis, Fasciola hepatica), echinococcosis, can obstruct the smaller bile ducts within the liver, resulting in intraductal cholestasis (1). Hydatid disease is a worldwide zoonosis produced by the larval stage of the Echinococcus tapeworm (2). In humans, hydatid disease involves the liver in approximately 60 to 75% of the cases (3). Echinococcal cysts of the liver can cause complications in about 40% of cases and manifest linically with acute abdominal pain. The most common complications in order of frequency are infection then rupture mainly into the biliary tree. Only 3-17% of the patients have a frank rupture, which has an overt passage of intracystic material to the biliary tract and, among hem, only 8 to 11% occur in the common bile duct (CBD) or cystic duct (4,5). We report this case of an abdominal pain due to an intrabiliary frank rupture of a hepatic hydatid cyst, occurring in the common bile duct causing a cholangitis. Case presentation 30 years lady presented with picture of cholangitis, found on ultrasound abdomen to have a cholelithiasis with dilation of common bile duct and possible CBD stone without abnormalities in the liver. While doing ERCP, incidentally a brown to white thick amorphous membrane was discovered in the common bile duct instead of a stone, that have been extracted by balloon technique. The finding raised the suspicion of a parasitic infection, so MRCP was done that showed remnant of complicated hydatid cyst in the right hepatic lobes. **Discussion:** Based on the previous results, an intrabiliary ruptured hydatid cyst of the liver was suspected in the common bile duct, causing an bstructive jaundice/cholangitis due to the intracystic hydatid material (membrane) that was found on ERCP. In front of an obstructive jaundice, the first test to do is an ultrasound abdomen to differentiate intra and extrahepatic causes of cholestasis according to the presence or absence of bile duct dilation. Once choledocholithiasis suspected, ERCP should be done to drain the common bile duct. (6) Most patients with Echinococcosis infection are asymptomatic and hydatid cyst is discovered incidentally during imaging. However, in around 25% of the cases, patient presents with an obstructive jaundice due to the rupture of hepatic hydatid cyst into the biliary tree (7). The diagnosis of Echinococcosis is usually established by radiologic tests and serology (8). ERCP is indicated when the results of these tests are unconclusive in patients with biliary colic associated with cholangitis. 3 different aspects may be seen on ERCP: round lucent filling defects due to daughter cysts or filiform wavy material in the common bile duct due to laminated hydatid membranes or brown thick amorphous membrane also due to hydatid membranes (9). Treatment of hydatid disease usually involves a combination of an anti-helminthic therapy and surgical resection / percutaneous aspiration of the cyst with endoscopic treatment in some cases (10,11,12). **Conclusion:** Physician must raise attention about parasitic disease-causing abdominal pain in the mergency departments. The diagnosis of abdominal parasitosis can be delayed because of the clinical similarities with other more frequent causes of abdominal pain. US abdomen may miss the diagnosis of a hydatid cyst due to several factors (obesity, location of the cyst, and the physician skills). A hydatid cyst may rupture in the biliary tract with migration of intracystic material into the CBD, such as hydatid membranes, mimicking CBD stone on US and leading to an obstructive jaundice / cholangitis.

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INTRODUCTION

Biliary obstruction, that affect around 5 cases per 1000 people in the united states, are due to several causes, with the Gallstones being the most common one.

Of parasitic causes, adult Ascaris lumbricoides, eggs of certain liver flukes (e.g., Clonorchis sinensis, Fasciola hepatica) and echinococcosis can obstruct the smaller bile ducts within the liver, resulting in intraductal cholestasis (1).

Hydatid disease is a worldwide zoonosis produced by the larval stage of the Echinococcus tapeworm. There are 4 types of Echinococcus infections. E**.** granulosu**s** is the most common type(2).The definitive hosts for *E. granulosus* are canines (dogs, wolves, and foxes) in which the adult worm resides in the intestines. The parasite forms a hydatid cyst in humans and other species, who serve as intermediate hosts (16).Infection occurs following ingestion of tapeworm eggs excreted in the faeces of infected dogs. Once ingested by human, the eggs release larvae into the duodenum that migrate through the intestinal mucosa and reach the liver via the mesenteric vessels. In humans, hydatid disease involves the liver in approximately 60 to 75% of the cases and less frequently the lung, the spleen, the kidney, the bones, and the brain (3). Echinococcal cysts of the liver can cause complications in about 40% of cases and manifestclinically with acute abdominal pain. The most common complications in order of frequency are infection then rupture mainly into the biliary tree. Only 3-17% of the patients have a frank rupture, which has an overt passage of intracystic material to the biliary tract and, among them, only 8 to 11% occur in the common bile duct (CBD) or cystic duct, the other 90% occurring at right hepatic and left hepatic duct (4,5). We report this case of an abdominal pain due to an intrabiliary frank rupture of a hepatic hydatid cyst, occurring in the common bile duct causing a cholangitis.

Case description

The patient discussed in this report is a30-year-old, NKDFA,presenting with severe epigastric pain radiating to RUQ of 3 days duration associated with severe nausea and increasing progressive jaundice. History goes back to 3 weeks ago when the patient started to have multiple attacks of acute onset RUQ pain increasing in intensity, radiating to right shoulder,associated with nausea and decrease po intake, relieved by symptomatic treatment. In the last 3 days, a new similar attack but this time continuous colicky pain, associated with jaundice, in addition to intermittent fever minimally relieved on antipyretic despite starting antibiotics. She noted having pale stool in those 3 weeks with fatigue but without weight loss.

PMH: Anemia not investigated, obese (BMI= 36,3Kg/m2)

PSH: Uncomplicated C-section twice (in 2012 and 2014)

Social history: Living in poor conditions (low socio-economic status, poor sanitation, bad hygiene, with frequent street dogs and cats in the area where she lives).

Habits history: Non-smoker, non-alcoholic, ingestion of raw meat on weekly basis, ingestion of non-purified water on daily basis.
Family history: Cardiovascular disease in father, diabetes mellitus in mother.

On physical examination, she was febrile with mild tachycardia but hemodynamically stable (table 1). Icteric conjunctiva with jaundice covering all her body, the abdominal exam was remarkable for RUQ tenderness with equivocal murphy sign, voluntary guarding. The rest of the physical exam was otherwise normal. The patient's laboratory studies showed leukocytosis with a left neutrophil shift, associated with an elevated CRP, elevated LFT's and elevated total bilirubin with direct fraction predominant (Table 2). HAV and HCV serology turned negative, and HBsAg was also negative. An ultrasound abdomen pelvis was performed, and it showed a multiple stones in the contracted gallbladder with a dilated CBD of 14mm and possible CBD stone. Based on clinical findings and work up results, a cholangitis was diagnosed, and IV antibiotic started. An ERCPperformed: duodenoscopy was normal with normal papilla; cannulation was done and while doing sphincterotomy we were surprised to find a suspicious mucoid glistening thick structure. This finding was extracted by balloon technique (Picture 1).

The patient stayed febrile with continuous elevation of the bilirubin, a laparoscopic cholecystectomy was done, where the gallbladder was removed and showed a few microlithiasis, cholangiogram was impossible because of the short cystic duct but the CBD was not dilated as by the surgeon. An MRCP done and showed remnants of complicated hydatid cyst of bare area of the liver with a well-defined capsulated 7x4cm cyst in right hepatic lobe revealing thick low signal intensity capsule, that was not been detected on the US abdomen. Albendazolewas started in addition to the escalation of antibiotics to piperacillin/tazobactam for possible secondary bacterial infection.The patient improved clinically in the following days with normalization of the lab measurement and weaning of the fever.

DISCUSSION

The patient clinical presentation (with RUQ pain, fever and jaundice) and laboratory results (cholestatic pattern elevation of liver enzyme with conjugated hyperbilirubinemia) go with an obstructive jaundice/cholangitis. Testing in this group of patients typically starts with right upper quadrant ultrasonography to assess the hepatic parenchyma and bile ducts. According to the presence or absence of bile duct dilation, ultrasound will differentiate between extrahepatic and intrahepatic cause of cholestasis respectively, but it will rarely identifies the cause of obstruction (6). The presence of common bile duct dilatation on ultrasonography in this reported case, in a context of a lithiasic gallbladder suggests extra hepatic cholestasis mainly due to a choledocholithiasis that represent the most common cause. Once ultrasonography suggests obstruction due to a stone endoscopic retrograde cholangiopancreatography (ERCP) should be carried out to confirm the diagnosis and facilitate biliary drainage (6), as in this case. The findings on ERCP of a brown to white, thick, glistening amorphous membranes (picture 1) instead of a stone suggest to look for other causes of common bile duct obstruction with persistence of low grade fever and mild abdominal pain in the patient despite laparoscopic cholecystectomy that revealed an inflamed gallbladder with microlithiasis. So MRCP was done and the result was surprising to detect a remnant of complicated hydatid cyst that ultrasound abdomen did not reveal it at the beginning.

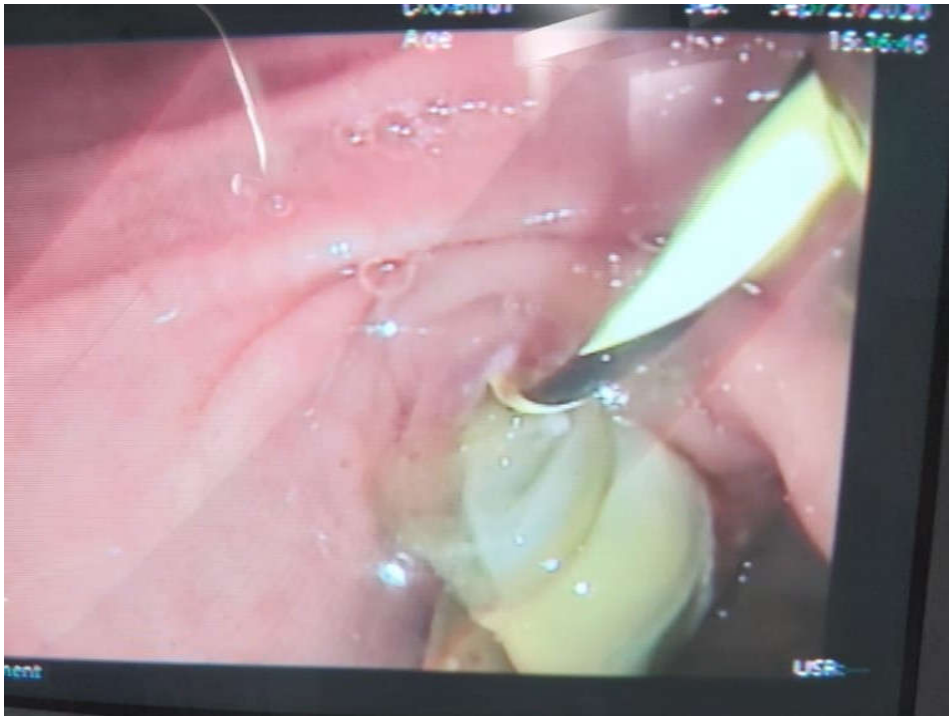
Most patients with Echinococcosis are asymptomatic and hydatid cyst is discovered incidentally during imaging. However, in around 25% of the cases, patient present with an obstructive jaundice due to the rupture of hepatic hydatid cyst into the biliary tree caused by the higher pressure in the cyst, often up to 80 cm H_2O (7). Three types of rupture may occur, a contained endocyst rupture, a simple communication that is discovered usually during surgery in a asymptomatic patient, and a frank intrabiliary rupture with the contents of the cyst

Table 1. Showing vitals sign during hospital stay

Date	20	21	22	23	24	25	26	27	28	29	30	1	2
Temp (*c)	38,3	38,5	38,2	38,5	38	38	38,3	38,2	38,3	37,4	36,5	36,6	36,7
BP (mmHg)	135/70	130/50	140/70	110/60	120/80	130/80	120/70	130/80	110/60	130/70	130/70	120/60	120/50
Heart rate	105	88	105	110	104	90	104	112	96	90	84	80	81

Table 2. Showing laboratory results during hospital stay

Date september 2020	20	21	22	23	24	25	26	27	28	29	30	2 oct
WBC (x1000u/l)	5,7	8,97		14		9,36	13,35		17,3	13,7		8,97
Neutrophils (%)	69	75,8		83,9		72,7	79,6		82,7	76		66,6
Eosinophils (%)	2,9	3,5		3,4		4,7	2,2		1,7	5		6,1
Hemoglobin (g/dl)	12,5	12,3		11,3		10,3	11,7		9,7	9		9,4
MCV (fl)	83,8	84,1		83		81,6	81,4		81,3	82		84,5
Platelet (x1000u/l)	373	371		335		372	481		409	369		421
Creat (mg/dl)		0.3		0,3			0,2			0,3		0,3
Na (mmol/l)		138		135		137	133			135		136
K(mmol/l)		4		3,6		3,3	3,7			3,7		4,1
CO2(mmol/l)		22		23		25	21			24		23
Cl(mmol/l)		103		99		101	96			101		104
Total bilirubin (mg/dl)	6,98	7,4		10		10,6	13,5	13,6	12,9		7,9	4.8
Directbilirubin (mg/dl)	3,8	4,5		6,4		6,7	8,4	8,2	8		4	2.5
SGPT (IU/L)	100	389		273		179	251			174		147
SGOT (IU/L)	111	315		270						145		131
GGT (IU/L)		405		321		206	252			226		252
Phos Alkaline (IU/L)				474								
Amylase (IU/L)	34			15								
Lipase (IU/L)	54			19								
CRP				283								
INR		1										
APTT (sec)		26										



Picture 1. Mucoid glistening thick structure (hydatid membrane) while performing sphincterotomy during the ERCP

(scolices and daughter cysts) draining into the biliary ducts and causing obstruction of the bile duct, resulting in obstructive jaundice / cholangitis as seen in our patient. (9). Only 3-17% of the patients have a frank rupture, which has an overt passage of intracystic material to the biliary tract and, among them, only 8 to 11% occur in the common bile duct (CBD) or cystic duct. (4,5)

The diagnosis of Echinococcosis is usually established by radiologic tests and serology(8). Ultrasound (US) provides the clinician with important clinical information including the location, number, size and stage of cysts, with a higher sensitivity and specificity than serology. The two most common routes of exophytic growth are via the bare area of the liver and gastrohepatic ligament. HCs extend to lung and mediastinum when located at the bare area of liver. (15)

Although US is the modality of choice for determining cyst stage and number and the extent of disease , CT and MRI are valuable in certain circumstances such as patients in whom sonographic visualization is impaired because of bowel gas, obesity or previous surgical interventions, in disseminated disease, extra-abdominal location.In addition, MR cholangiopancreatography is preferred in complicated cases of communication or rupture into the biliary system. (13) Our patient, with a BMI of 36,3 Kg/m2 and a location of the cyst in the bare area of liver could explain the miss visualization of the cyst by ultrasound, without forgetting that this act is an operator dependent.

When the results of these tests (radiologic and serologic) are unconclusive in patients with biliary colic associated with cholangitis, ERCP is indicated. Other than the brown to white thick glistening membrane, seen in our patient, ERCP may show round lucent filling defects due to daughter cysts or filiform wavy material in the common bile duct due to laminated hydatid membranes (9). Treatment of hydatid disease usually involves a combination of Albendazole (an anti-helminthic highly effective) course and surgical resection / percutaneous aspiration of the cyst with instillation of scolicidal agents. A non-surgical endoscopic trans-papillary treatment of ruptured echinococcus liver cyst obstructing the biliary tree may be done, usually reserved for patients with suspected biliary complications occurring before or after surgery (10,11,12): A sphincterotomy is often needed in patients with an obstructive jaundice or cholangitis. The intracystic material are removed with the help of a basket or an occlusion balloon (9) with or without saline irrigation of the bile duct to flush out the hydatid sand and small daughter cysts. (14)

Conclusion

Physician must raise attention about parasitic disease-causing abdominal pain in the emergency departments. The diagnosis of abdominal parasitosis can be delayed because of the clinical similarities with other more frequent causes of abdominal pain. US abdomen may miss the diagnosis of a hydatid cyst due to several factors (obesity, location of the cyst, and the physician skills). A hydatid cyst may rupture in the biliary tract with migration of intracystic material into the CBD, such as hydatid membranes, mimicking CBD stone on US and leading to an obstructive jaundice / cholangitis.

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Author contribution

Writing the paper, Study Concept: Malek Michael Bouhairie, Racha Seblani Data collection, Study Concept: Malek Michael Bouhairie, Sabrina Nasredine

Supervision: Tarek Hijazi

Registration of research studies

- Name of the registry: N/A
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