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Case Report

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Hepatic Hydatid cyst Cystobiliary Communication, Case Report & Literature Review

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ABSTRACT

Hepatic hydatid cysts are benign cysts in the liver that are the result of parasites infection. They are caused by echinoccocus granulosis or multilocularis. They caused several symptoms like pain, obstructive jaundice, and sepsis. Hydatid cyst can be complicated to cystobiliary communication (CBC) which can be frank CBC or occult CBC. Medical, endoscopic, percutaneous and surgical treatments are different approaches to treat hydatid cyst.

Here we report a case of hepatic hydatid cyst with cystobiliary communication, causing obstructive jaundice and treated with sphincterotomy and insertion of biliary stent through endoscopic retrograde cholangiopancreatography (ERCP).

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Abbreviation: CBC: Cystobiliary Communication. ERCP: Endoscopic Retrograde Cholangiopancreatography

Introduction

Hepatic hydatid disease is a parasitic zoonosis caused by the Echinococcus tapeworm. Echinococcus granulosus & Echinococcus multilocularis are the parasites that cause hydatid cysts in the liver.

E. granulosus can lead to endemic infection with varying prevalence and distribution (mainly in the Mediterranean). These parasites form a spherical, fibrous-rimmed cyst with little, if any, surrounding host reaction. Typically, it has a large parent cyst which contains numerous peripheral daughter cysts. Satellite daughter cysts (outside the parent cyst) are seen frequently (~16% cases).

There are two forms of E. granulosus: the most common form is pastoral, whose main host is domestic dog. The other form is sylvatic; who's the main host is wolf or dog.

The definitive host of E. multilocularis is the red fox (Vulpes), cats and dogs. Sometimes, humans can be intermediate host for this type of parasites. It is widely distributed throughout the Northern hemisphere.

The eggs enter the small intestine of the human emerging the vasculature, then liver and lungs.

After 3 to 4 weeks of incubation, the cysts become mature and visible in the liver. Mature cyst has a germinal layer surrounding a fluid-filled central hydatid cavity.

Its clinical presentation varies. Small cysts, <5 cm, are asymptomatic. However, larger cysts cause an inflammatory reaction and may lead to abdominal pain.

Infected or ruptured cysts result in severe acute abdominal pain. The rupture of the cyst incidentally or iatrogenically can cause a severe allergic reaction, leading to ascites, peritonitis, and anaphylactic shock.

Rarely, the cysts can extrude into the biliary tree, leading to jaundice and cholangitis.

Diagnosis is made based on the combination of the 3; clinical presentation, serological testing (ELISA IgG, IHA or IEP) and imaging (U/S, CT or MRI).

Case presentation

Presents to our ED a 30 year old smoker, Caucasian male patient with no significant PMH or PSH, complaining of RUQ pain and increasing jaundice since 1 week, associated with decreased appetite, early satiety, nausea and occasional vomiting. Laboratory findings suggested a cholestatic obstructive pattern with elevated GGT (450), A.P (560) as well as direct and indirect hyperbilirubinemia (3, 8). Normal INR, leukocytosis (13k), elevated CRP (90).

Initial U/S imaging showed a dilated CBD with no visualized stone and a hepatic lesion. CT scan was then done showing dilated CBD Citation: Hallal Mahmoud (2020) Hepatic hydatid Cyst Cystobiliary Communication, Case Report & Literature Review. Journal of Gastroenterology & Hepatology Reports. SRC/JGHR-103. DOI: doi.org/10.47363/JGHR/2020(1)103

with obstruction at the level of the Cystic duct along with a large 6-7 cm hepatic cyst with multiple septations (daughter cysts) and a communication between the hepatic cyst and the cystic duct. So patient was planned for ERCP, were selective cannulation was done along with a wide sphincterotomy, when whitish material started draining out of the CBD. A plastic stent was placed and patient was then referred for surgery for cyst drainage, were an external cyst drainage was placed. IEP was positive for hydatid cyst, patient was started on albendazole 400mg twice daily. clinical improvement with a significant drop in LFTs was observed the next day which continued so over the following days/weeks.

Discussion

Cysto-biliary communication (CBC), Communication between the biliary tree and the hydatid cyst can be frank or occult [1]. Frank intrabiliary cystic rupture can be manifested by colicky right hypochondrial pain and obstructive jaundice [2]. Occult rupture, on the other hand, usually has no clinical signs, and the most common symptom, if present, is abdominal pain. Occult CBC can lead to fistulas post biliary surgery if the bile duct is not detected and properly sutured during surgery [3].

There are two theories that have been proposed for pathogenesis. First, hydatid cyst compresses biliary duct wall and lead to progressive necrosis, hence, a cyst. Second, there is an intracystic small biliary radical that do a pressure on pericystic membrane and becomes atrophied [4].

Frank CBC is a 5 mm (wide) communication between the cyst and the biliary tree. When intracystic pressure is more than biliary system, cystic contents like daughter cysts or membrane fragment invade biliary tree. It causes in 3 to 17 % cholestasis due to obstruction, cholangitis or even the most severe signs is anaphylaxis [4,5]. If the communication is more than 5 mm, hydatid cyst materials are seen in 65% of patients in preoperative imaging, hence the treatment is decided accordingly [4].

The other type of CBC is occult CBC that has less than 5 mm communication between the cyst and the biliary tree. It is seen in 10 and 37% of hydatid cyst patients. It is asymptomatic, thus difficult to diagnose [5,6]. Earlier the diagnosis of hydatid cyst, better the prognosis and lesser the morbidity and biliary leak post-operative [4,7]. MRCP and ERCP has low sensitivity to diagnose frank CBC.

Hydatid cyst in some cases can be treated percutaneously (with injection of hypertonic saline or alcohol), or laparoscopically as alternative of surgery.

Hydatid cyst can be complicated rarely to a ruptured cyst (in 20-50%) or can do secondary bacterial infection (in 5-8%). There are 3 types of ruptured cyst, contained, communicating (most common up tp 64%) and direct rupture.

Contained rupture or Endocyst rupture is when hydatid fluid comes out between the pericyst and the endocyst resulting collapse of the endocyst [8].

It is seen as undulating membranes inside the hydatid cyst without decrease in the cyst size. It is is called the "waterlilly sign" or "snake or serpent sign" [8,9,10]. The other type of ruptured cyst which is the most common type is communicating rupture seen in 44-64% of cases. This communication can be done by small fissure or bile cyst fistulas (10) or by a wide perforation of the cyst into a main biliary radicle [11]. Then hydatid cyst content like

fluid, sand and daughter cysts enter the biliary system in 3-17% of patients [11]. Usually hydatid cyst ruptured in right hepatic duct (55-60% of cases) and less frequently in left duct (25-30%) or gall bladder [12,13].

Hydatid cyst rupture can do right upper quadrant pain, obstructive jaundice, fever, cholangitis or sepsis and even death. Diagnosis should be rapid and accurate.

On CT scan characteristic findings of hydatid cyst rupture include wall calcification discontinuity, dilatation of the biliary tree with air or air-fluid level or containing high density linear materials. [8,11].

Direct rupture of cyst is when both the endocyst and pericyst are damaged,thus, cyst content invades peritoneal cavity, thorax and lungs through the diaphragm, mediastinum or occasionally a hollow organ such as the colon. It can lead to anaphylactic shock if not managed rapidly (11).

The other complication is secondary bacterial infection which is very rare due to lack of vascularization and connection of cyst and vascular system [8]. It is caused by a bacteria entering the cyst after rupturing both pericyst and endocyst [8]. Air fluid level, no change of cyst size and hepatic abcess are the result of this infection. Medical, endoscopic intervention, minimally invasive surgery are all options to treat hydatid cyst. Albendazol is an antiparasite to treat echinococcus granulosus. It is poorly absorbed and should be ingested with food, albendazole, 10-15 mg/kg/d in 2 divided doses (the usual dose for an adult is $2 \times 400 \text{ mg}$)] during fat-rich meals to increase bioavailability [14,15]. In the absence of albendazole, mebendazole may be used as an alternative therapy; it is less well absorbed than albendazole.

Albendazole should be given for 1 to 3 months and may be up to 6 months. It depends on patient clinical manifestations (WHO stage CE1 and CE3a cysts). For cyst more than 5 cm o WHO stage Ce2 or CE3b cyst, medical treatment alone is not effective, it reduces the recurrence of the disease. It should be started at least 4 days before surgery to 1 month (for albendazole) or 3 months (mebendazole) post-surgery. Albendazole is generally well tolerated but it can do nausea, hepatotoxicity, neutropenia, and occasionally alopecia. Monitoring leukocyte count, liver function tests should done regularly in all patients. Pregnant, patients with chronic liver disease, and bone marrow depression can't take albendazole [16]. Other type of management is ERCP that can detect many biliary duct abnormalities including diagnosing biliary rupture preoperative [17,18].

It can decrease post operative morbidity by 7.6 to 11.1 % while detecting fisula and cystic hydatid content draining into biliary tree. So ERCP has 86–100% sensitivity to diagnose frank CBC. It treats acute hydatid cyst to defer it for elective surgery, hence decreases postoperative complication and hospital stay. Endoscopic sphincterotomy (ES) in ERCP is the first line of treatment in obstructive icteric patients [4]. This sphincterotomy decreases the risk of post-operative fistula and 25% of the patients with frank CBC are cured with ERCP without the need for further surgery. Extracting with balloon or basket catheter, nasobiliary draining and inserting biliary stent are different therapeutic methods that are done by ERCP [19].

Patients who have hydatid cyst more than 5 cm and who are not candidate for surgery, can do PAIR, is a percutaneous treatment. It can't be done in patient with biliary fistulas or communications with the biliary tree because of the risk of biliary sclerosis. PAIR

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is contraindicated also in patients with inaccessible cysts, or complicated, multivesiculated cysts [20,21]. PAIR can rarely do (in 0.03%) lethal anaphylaxis shock [22].

Complicated cyst (ruptured, infected, bleeding, compressing or with fistula, multivesicular cysts and cyst that have contraindication to percutaneaous treatment (WHO stage CE2 and CE3b)) can be managed surgically [23]. Cyst with diameter >10 cm, superficial cyst at risk of rupture due to trauma, and extrahepatic disease are managed surgically [24].

Conclusion

Hydatid disease of the liver is still endemic in certain parts of the world. The diagnosis of non-complicated hydatid cyst of the liver depends on clinical suspicion. Ultrasonography and computed tomography, the most important diagnostic tools, are helpful for determining the complications and planning treatment. The modern treatment of hydatid cyst of the liver varies from surgical intervention to percutaneous drainage or medical therapy. Surgery is still the treatment of choice and can be performed by the conventional or laparoscopic approach. Percutaneous drainage and treatment of the cyst with hypertonic saline or alcohol seems to be a good alternative to surgery in selected cases.

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