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Case Report

Cholecysto-Hydatid Fistula Complicated by Cholecystitis and Acute Rupture Hydatid Cyst into Peritoneal Cavity- A Case Report

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Abstract

Introduction: Cholecysto-hydatid fistula is a very rare complication of liver echinococcosis. We report the case of a patient with hydatid cyst fistula to the gallbladder accompanied by cholecystitis.

Patient information: 50-year-old patient admitted to emergency department due to pain in the upper right abdominal quadrant. Imaging studies (ultrasound, CT) showed parasitic cyst in the IV segment of the liver coexisting with a fistula with an inflamed gallbladder, as well as two other smaller cysts. Intra operatively rupture of the cyst into the peritoneal cavity was observed. Excision of the gallbladder and the adjacent cyst en bloc was performed. Histopatological examination revealed protoscolices of Echinococcus granulosus. In the postoperative period complicated such as severe bacteremia, wound suppuration and intraperitoneal abscesses developed, despite the use of antibiotic therapy. Spectacular clinical improvement was achieved after using polyclonal immunoglobulin.

Conclusion: Echinococcosis is disease with an insidious course. Communication of hydatid cysts with the biliary tract can cause a severe clinical course with numerous complications.

INTRODUCTION

Hepatic cysts can be both hereditary and secondary. Cystic echinococcosis is a common cause of secondary hepatic cysts in areas endemic to Echinococcus granulosus such as the Mediterranean, South America, and the Middle east [1]. The human being can become an intermediate host after consuming food contaminated by feces of the definitive host (canine) containing eggs of the parasite. These eggs transform in oncospheres in the duodenum, which then migrate through the wall of the intestine via the venous system to various organs where they form single cell parasitic cysts [2]. They can invade almost any tissue yet they most often form in the liver (50-75%) or lungs (25%) [3]. In the liver echinococcus cysts form in the right lobe in 81.9% of patients [4]. The wall of the cyst is made of a fibrous capsule composed of two concentric layers; the inner layer made of parasitic tissues while the other layer is made up of compressed tissues of the host which undergoes fibrosis due to a local inflammatory response. The interior of the cyst is surrounded by a germinal membrane which is connected with daughter cysts containing protoscoleces in a clear liquid. It is estimated that 40% of hydatid cysts found in the liver will cause complications, of which the most common is migration into the respiratory and biliary tract [5]. Secondary bacterial infection and cystic rupture into the peritoneal cavity is less common. Fistulas between the gallbladder and hydatid cyst occur occasionally. In Poland Echinococcosis cysts is a rare disease with a prevalence of 0,026 / 10x5 population among women in rural areas [6]. We report a case of a patient with a fistula between the hydatid cyst and gallbladder with coexisting inflammation of the gallbladder.

CASE PRESENTATION

A 50 year old obese female patient was seen in the emergency department with severe abdominal pain located in the upper right abdominal quadrant. Onset of symptoms was associated with a heavy meal. The patient had never travelled abroad and she lived in an urban area. Patient history was negative of trauma and concomitant diseases. The physical examination showed pain in the upper right quadrant with rebound and guarding. Laboratory tests showed a mildly elevated white blood count of 10,560/ul and C-reactive protein of 0.56 mg.dl. Ultrasound found an uneven hyperechogenic cystic oval structure located near the gallbladder suspected of having a fistula with the gallbladder. Another cystic structure was described in the right lobe of the liver. The patient was admitted to the surgical department for observation towards cholecystitis. A CT scan was then performed which revealed 3 cysts in the liver suspect of being parasytic cysts. The largest cyst, 63 mm in diameter, was located in the near vicinity of the gallbladder in the IV segment of the liver

(Figure 1). The smaller cysts, 50 mm and 58 mm in diameter were located between the VI/VII and V/VIII segment of the liver. Due to an increase in the white blood count and C-reactive protein the patient was qualified for surgery the next day. Laparotomy was performed. Intraoperative findings showed an enlarged inflamed gallbladder without gallstones which was connected by a fistula to a cyst over 60 mm in diameter. About 100 ml of clear liquid was aspirated from the cyst and sent to histopathological examination. Cholecystectomy with radical dissection of the cyst was performed. The peritoneal cavity was flushed with 3% saline solution. Due to technical difficulties the other two cysts were left untouched. The histopathological examination showed E. granulosus protoscolices (Figure 2).

In the postoperative period the patient received broad spectrum antibiotic therapy (ceftriaxone 2.0g/d and gentamicin 2 mg/kg) for 10 days and albendazole 10 mg/kg during a period of 6 months. Despite treatment, the patient's condition did not improve pneumonia and wound infection developed. 20 days after the primary operation the white blood count rose to 35,000/ul and the C-reactive protein rose to 31.8 mg/dl. A CT scan was performed which showed intra-abdominal abscesses. The patient was once again qualified for surgery. Relaparotomy was performed during which two abscesses, located below the diaphragm and in the right iliac fossa, were evacuated. Obliteration of one of the remaining cysts located between the VI/VII segments was performed using 3% saline solution and electro coagulation. Control swab from the peritoneum did not show the presence of E. granulosus, as well as repeated serological tests were negative for Echinococcus. The patient condition continued to be severe. Blood culture was positive for Staphylococcus haemolyticus, while swabs from the wound were positive for Staphylococcus Aureus MRSA and multi resistant strain of Acinetobacter baumannii. Targeted antibiotic therapy using meropenem and vancomycin was given for a period of 14 days, and negative-pressure wound therapy was implemented. Infusion of polyclonal immune globulins was added due to an unsatisfactory response to treatment. Afterwards a systematic decrease in inflammatory parameters, resolution of fever, good wound healing was observed. On the 45th day of hospitalization, the patient was discharged home in good condition with a recommendation of control in the Department of Infectious Diseases.

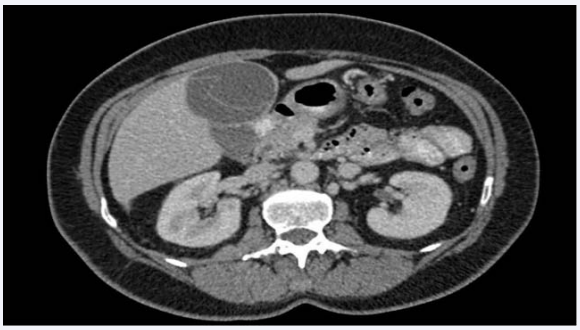

Figure 1 CT imaging Hydatid cyst communicates with gall bladder.

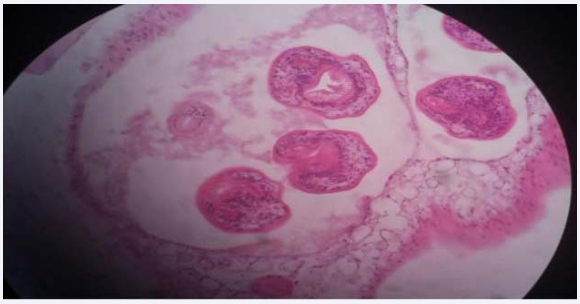

Figure 2 Protoscolices of Echinococcus granulosus from peritoneal fluid.

DISCUSSION

Hydatid disease is often symptomless and slow growing. The onset of symptoms is dependent on the size and location of the cysts. The most common symptom is pain in the upper right abdominal quadrant [8]. It is said that cysts grow at a speed of 1-5 mm/year [9]. Risk factors associated with rupture include size over 10 cm and young age of patients [6]. Our patient was middle aged while the diameter of the cyst did not exceed 7 cm. A factor, such as the inflammation of the gallbladder may have caused the cyst to rupture into the gallbladder. The question remains whether cholecystitis was primary or secondary to the cystic fistula.

An ongoing inflammatory process induced by pro-inflammatory cytokines (IL-1, IL-6, TNF-α) leads to an increase in production of metalloproteinase [9]. The degradation of the extracellular matrix which causes damage to the structure of the collagen fibrous capsule of cysts can increasing the likelihood of their rupture. Regardless of the cause, the incidence of hydatid liver cysts rupture into the peritoneal cavity ranges between 1-16% [10]. This complication can lead to allergic reactions and even anaphylaxis. Our patient did not present such symptoms.

The rupture of the contents of a hydatid cyst into the peritoneal cavity requires emergency surgery [11]. There are two possible approaches to surgery, radical and conservative. In practice, the choice of the approach depends on the anatomical location and size of cysts, also the condition of the patient as well as surgeon experience is very important [13]. Depending on the method postoperative mortality range between 0.5-7.5% [12]. Regardless of the approach, intra operative care should be taken to avoid dissemination of the parasite [14].

Intraoperatively linen soaked with concentrated saline solution can be used in the prevention of dissemination. In order to inactivate protoscoleces in the peritoneal cavity, lavage can be performed using hypertonic saline solution, hydrogen peroxide, povidone iodine, chlorhexidine, cetrimide, and silver nitrate. It seems safe and effective to use a saline solution of 3% [15]. In the above presented case we rinsed the peritoneal cavity abundantly with approximately 2.5 L of 3% saline solution. The saline solution did prevent dissemination yet it did not prevent secondary abdominal infection which was probably associated with the patient's general state and lowered immunity. Hypernatremia was not observed in the postoperative period.

Bacterial infections, incidence of approx. 3-7%, are fairly frequent complications of echinococcosis [16]. Despite the

use of antibiotics our patient developed multi pathogenic and multi-resistant infections with co-existing bacteremia and intraperitoneal abscesses. We achieved significant improvement and stabilization of the patient condition after administering polyclonal immunoglobulin.

Despite positive histopathology for Echinococcosis, serological tests [ELISA (+), western blot (-)] were negative. This may have been due to an early phase of invasion and lack of specific response due to minimal contact between host and parasite. Treatment of cyst rupture to the peritoneal should be carried out using albendazole at a dose of 10-15mg / kg / day for a period of 3-6 months and in the case of intolerance to albendazole, mebendazole should be administered at a dose of 40-50 mg / kg / day. The use of these drugs should be monitored monthly by liver tests and blood count, since neutropenia, and liver damage can occur. Contraindications to treatment are pregnancy, liver failure and bone marrow suppression [8]. In this case no side effects were noted.

The proper diagnosis and treatment of echinococcosis is at times difficult especially when the disease affects an uncommon localization. These patients are at a high risk of developing concomittant bacterial infections and should be monitored closely in the postoperative period.

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