OLGU SUNUMU / *CASE REPORT*

A Serious Complication of Liver Hydatid Cyst: Intra-abdominal Rupture

Karaciğer Hidatik Kistinin Ciddi Bir Komplikasyonu: Karın İçi Rüptür

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ABSTRACT

Intra-abdominal rupture is a severe and rare complication of a hydatid cyst. This case report is aimed to present the diagnosis and treatment algorithm for spontaneous intra-abdominal rupture of liver hydatid cyst. A 46-year-old lady was followed up in the gastroenterology service with a cholangitis diagnosis. Endoscopic retrograde cholangiopancreatography (ERCP) was applied to the patient. The patient's abdominal pain and vital signs worsened on the first day after ERCP. After the patient was hemodynamically stabilized, intravenous contrast-enhanced computed tomography (CT) was performed. On CT, there was a 130 mm diameter air-containing hydatid cyst in the liver dome accompanied by intraperitoneal free air. Liver hydatid cyst perforation was considered in the patient, and the patient was taken for an emergency laparotomy. Cystotomy and unroofing were performed with cholecystectomy. Control thorax CT was performed on the patient due to low saturation (90% under nasal oxygen support) on the sixth day of follow-up. Due to pleural effusion in the right hemithorax on the CT scan, thoracentesis was performed on the patient, and 500 cc of seropurulent fluid was aspirated. On the 14th postoperative day, the patient was discharged without any problem and was prescribed 10 mg/kg/day of albendazole. No pathology was detected on the control CT in the first-month follow-up.

Key words: *albendazole; anaphylactic reaction; echinococcosis; spontaneous rupture*

ÖZET

Karın içi rüptür hidatik kistin ciddi ve nadir bir komplikasyonudur. Bu vaka raporunun amacı, karaciğer hidatik kist spontan intraabdominal rüptürünün tanı ve tedavi algoritmasını sunmaktır. Kırk altı yaşında bayan hasta kolanjit tanısı ile gastroenteroloji servisinde takibe alındı. Hastaya endoskopik retrograd kolanjiyopankreatografi (ERCP) uygulandı. Endoskopik retrograd kolanjiyopankreatografi sonrası 1. günde hastanın karın ağrısı ve vital bulguları kötüleşti. Hasta hemodinamik olarak stabilize edildikten sonra intravenöz kontrastlı bilgisayarlı tomografi (BT) çekildi. Bilgisayarlı tomografide karaciğer kubbesinde intraperitoneal serbest havanın eşlik ettiği 130 mm çapında hava içerikli hidatik kist mevcuttu. Hastada karaciğer hidatik kist perforasyonu düşünüldü ve hasta acil laparotomiye alındı. Kolesistektomi ile kistotomi ve unroofing yapıldı. Hastaya takibin 6. gününde satürasyonun düşük olması (%90 nazal oksijen desteği altında) nedeniyle kontrol toraks BT çekildi. Bilgisayarlı tomografide sağ hemitoraksta plevral efüzyon nedeniyle hastaya torasentez yapıldı ve 500 cc seropürülan sıvı aspire edildi. Postoperatif 14. günde hasta komplikasyonsuz taburcu edildi ve 10 mg/kg/gün albendazol tedavisi verildi. Birinci ay takibinde kontrol BT'de patoloji saptanmadı.

Anahtar kelimeler: *albendazol; anafilaktik reaksiyon; ekinokokkoz; spontan rüptür*

Introduction

Hydatid disease, also known as cystic echinococcosis, is a zoonotic disease with an annual incidence rate of 1–200 per 100,000[1]. Cystic echinococcosis causes significant public health problems and severe economic losses in Turkey[2]. It is endemically seen in the Eastern and South-Eastern Anatolia regions. The causative agent of this disease is a parasite called Echinococcus granulosus[3]. The primary source of this parasite is meat-eating animals such as dogs, cats, wolves, and foxes. However, often the etiological source is dogs. The parasite lives in the small intestines of dogs. The disease is transmitted to humans by eggs excreted in dog faeces[4].

Cysts are most commonly seen in the liver and lungs[5]. They can also be located in the spleen, peritoneum, kidney, bone, eye, brain, heart, and ovaries. While hydatid cysts localized to the liver may progress

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asymptomatically, symptoms such as pain in the upper right of the abdomen, nausea, vomiting, and sometimes itching and jaundice are observed in symptomatic cases[1]. Surgeons should know that hydatid cysts can spontaneously rupture after surgery or trauma. Although hepatic hydatid cysts most commonly rupture into the biliary system, they can also rupture into the peritoneal cavity, bronchi, or blood vessels. Spontaneous intra-abdominal rupture of a hydatid cyst is a rare (1–8%) and life-threatening condition[6]. Patients with hydatid cyst rupture into the peritoneal cavity often present to the emergency department with acute abdomen. In addition, patients can apply to the emergency service with anaphylactic or allergic reactions up to 25%[7].

This case report is aimed to present the diagnosis and treatment algorithm for spontaneous intra-abdominal rupture of liver hydatid cyst.

Case Report

A 46-year-old lady was admitted to the Gastroenterology Clinic of Erzurum Regional Training and Research Hospital in January 2022 with complaints of right upper quadrant pain, fever, and chills lasting for about three days. She had no other disease or history of abdominal surgery. She was normotensive (blood pressure 134/72 mmHg), tachycardia (pulse 108/min), and febrile (body temperature 38.2°C). Her oxygen saturation in room air was 96%. On abdominal physical examination, the patient had abdominal pain in deep palpation in the right upper quadrant. Other system examinations, including digital rectal examination, were routine.

The patient's pathological laboratory parameters were as follows: alanine aminotransaminase (ALT): 119 U/L [7–40], aspartate aminotransaminase (AST): 108 U/L [13–40], alkaline phosphatase (ALP): 637 U/L [46–116], gamma-glutamyl transferase (GGT): >1200 U/L [0–38], total bilirubin (TB): 1.92 mg/dL [0.3–1.2], direct bilirubin (DB): 1.64 mg/dL [0–0.3], and c-reactive protein (CRP): 120 mg/L [0–5]. Other laboratory parameters were not remarkable, including tumor markers. On magnetic resonance cholangiopancreatography (MRCP) scan, condensed bile content was observed in the gallbladder. In addition, the liver had a 90 mm diameter hydatid cyst (Fig. 1). Endoscopic retrograde cholangiopancreatography (ERCP) was applied to the patient. During ERCP, it was observed that there was an occlusive stone distal to the common bile duct. After stone removal by sphincterotomy, a stent was placed in the common bile duct. The stent was removed on the normalization of control liver function tests.

The patient was admitted to the gastroenterology clinic in February 2022 with similar symptoms and physical examination findings. The patient's pathological laboratory parameters were as follows: alanine aminotransaminase (ALT): 49 U/L [7–40], aspartate aminotransaminase (AST): 42 U/L [13–40], alkaline phosphatase (ALP): 556 U/L [46–116], gamma-glutamyl transferase (GGT): 835 U/L [0–38], total bilirubin (TB): 2.10 mg/dL [0.3–1.2], direct bilirubin (DB): 1.58 mg/dL [0–0.3], and c-reactive protein (CRP): 323 mg/L [0–5]. Other laboratory parameters were not remarkable. Endoscopic retrograde cholangiopancreatography (ERCP) was applied to the patient. During ERCP, common bile duct and intrahepatic bile ducts were observed as dilated. Bile sludge and pus were extracted from the common bile duct with a balloon. On the 1st day after ERCP, the patient's abdominal pain and vital signs worsened. The patient was consulted at the general surgery clinic. She was hypotensive (blood pressure: 94/42 mm Hg), tachycardia (pulse rate: 118 beats per minute), and febrile (body temperature: 38.5°C). Her oxygen saturation on room air was 94%; on abdominal physical examination, she had defense and rebound on deep palpation in all quadrants. Her vital signs were stabilized with aggressive fluid resuscitation. Intravenous steroid (hydrocortisone 200 mg) and antihistamine (pheniramine 45.5 mg/2 mL) treatments were administered to resolve the allergic reaction. After the patient was hemodynamically stabilized, intravenous contrast-enhanced computed tomography (CT) was performed. A 130 mm diameter hydatid cyst was on a CT scan with air in the liver dome with intraperitoneal free air (Fig. 2 and 3). Liver hydatid cyst perforation was considered in the patient, and the patient was taken for an emergency laparotomy.

A ruptured liver hydatid cyst was observed during laparotomy (Fig. 4). In addition, free infective germinative membranes with massive purulent fluid were observed in the intra-abdominal cavity (Fig. 5). Cystotomy and unroofing were performed with cholecystectomy. Three bile duct repairs were performed due to the biliary fistula. The abdominal cavity was washed and cleaned with warm saline. One drain was placed in the cyst pouch, and the other was placed in the pelvis. The patient was taken to the intensive care unit for follow-up.

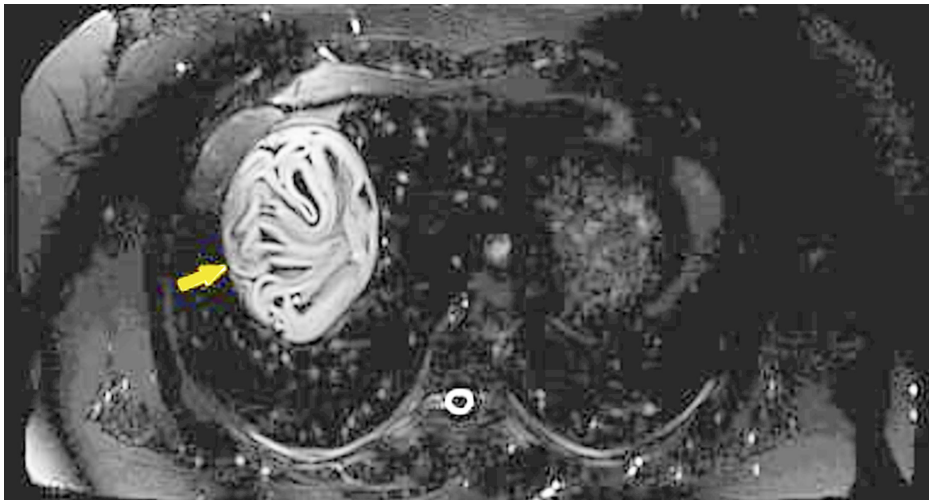

Figure 1. *Magnetic resonance imaging of liver hydatid cyst at first cholangitis attack (shown by a yellow arrow).*

Figure 2. *Preoperative computed tomography image of ruptured liver hydatid cyst: black arrows show ruptured hydatid cyst, and yellow arrows show intraperitoneal free air (axial image).*

Figure 3. *Preoperative computed tomography image of ruptured liver hydatid cyst: yellow arrow shows intraperitoneal free air due to perforation (sagittal image).*

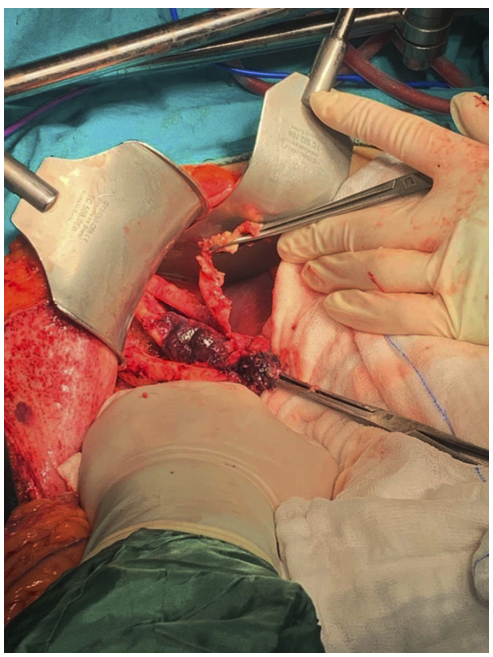

Figure 4. *Intraoperative view of ruptured liver hydatid cyst.*

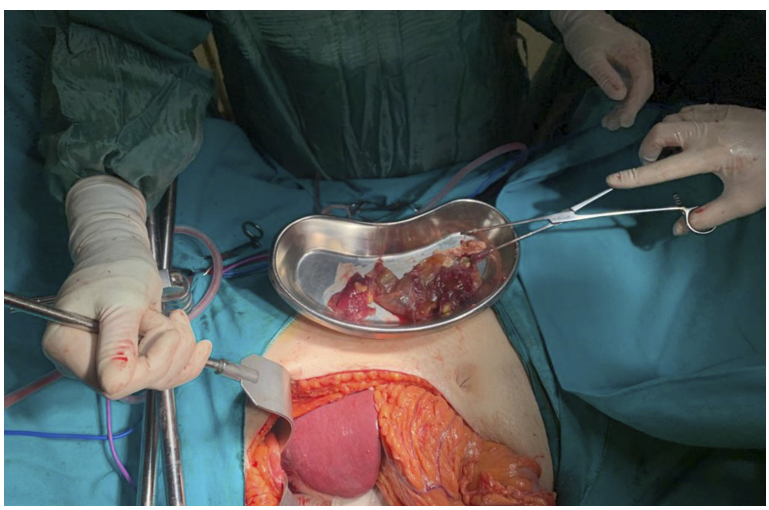

Figure 5. *Free infective germinative membranes.*

Piperacillin/Tazobactam (4.5 g vial every 6 hours), meropenem (1 g vial every 8 hours), and amikacin (500 mg vial every 12 hours) were started. In addition, treatment was administered with intravenous steroid (methylprednisolone 40 mg vial every 12 hours) and antihistamine (pheniramine 45.5 mg/2 mL vial every 12 hours). Control thorax CT was performed on the patient due to low saturation (90% under nasal oxygen support) on the 6th day of follow-up. Due to pleural effusion in the right hemithorax on the CT scan, thoracentesis was performed on the patient, and 500 cc of seropurulent fluid was aspirated. Both drains were removed on the 12 th postoperative day. On the 14th postoperative day, the patient was discharged without any problem and was prescribed 10 mg/kg/day of albendazole. No pathology was detected on CT in the first-month follow-up.

Discussion

Echinococcosis, also called hydatidosis, is a zoonotic infection that infects humans worldwide[8]. Hydatidosis results from an infection of the tapeworm *Echinococcus granulosus*. The disease characteristically shows hydatid cyst (metacestode) growth in the internal organs of intermediate hosts, including humans. The definitive hosts of the cestode are carnivores such as dogs. Humans and other hosts cause infection by ingesting eggs or pregnant proglottids excreted in the feces of the definitive host[9].

Liver hydatid cysts may be asymptomatic for many years and are often an incidental finding on ultrasound performed for another reason[10]. Although the diagnosis is often made when complications arise, jaundice is usually the first symptom, and rupture or secondary infection occurs as the first clinical presentation. The hydatid cyst rupture can occur in the biliary tract and the intra-abdominal cavity. When the cyst ruptures into the biliary tract, patients present with an attack of cholangitis. Intra-abdominal rupture, another condition, can cause life-threatening problems up to anaphylactic shock. The clinical picture was diffuse abdominal pain and mild anaphylactic shock findings in the presented case.

In laboratory evaluation, liver function tests are not specific in determining the severity of the disease, and pathological laboratory findings are seen only in approximately 40% of the cases. AST, ALT, and bilirubin levels typically remain within the normal range, while ALP is typically elevated. Eosinophilia may be seen in the complete blood cell count[11]. Detection of Echinococcus antibodies by ELISA is a standard method used for diagnosis. However, since this immune response depends on host immunity, positivity is not detected in every case[12]. In our case, there was an elevation in liver function tests, mainly due to a cholangitis attack. In addition, eosinophilia was not detected in the complete blood count, and the level of the Echinococcal antigen could not be determined due to the need for emergency surgery.

The most commonly used technique for diagnosis in radiology is USG. USG is the preferred method for cyst examination in elective conditions. On the other hand, CT imaging plays a vital role in situations where ultrasonography is difficult (e.g., obese patients) and is very important in the perioperative period. It can detect complications such as cyst rupture, underlying infection, and biliary or vascular involvement[13]. In the presented case, CT imaging was used as the first diagnostic tool and helped to establish the definitive diagnosis. CT scan revealed a ruptured hydatid cyst in the liver dome accompanied by intraperitoneal free air.

In treating a hydatid cyst, the stage of the cyst, the size of the cyst, and the presence of complications are important factors in making the treatment decision. Watch and wait, medical treatment with benzimidazoles, PAIR (puncture, aspiration, injection, re-aspiration), and surgery are the options for treatment. Surgery is the modality of choice for complicated cysts. Surgery is required for large cysts, superficial cysts prone to rupture, infected cysts associated with the biliary tract, and cysts pressing on adjacent organs[14]. Surgery was the first-line treatment because of intra-abdominal rupture and less severe allergic reaction findings.

Anaphylaxis due to intra-abdominal rupture is a rare and life-threatening condition. While there is an incidence of anaphylaxis up to 10 percent in the literature, less severe allergic reactions can be seen up to 25 percent[7,10]. Stabilization of vital signs should be prioritized in anaphylactic reactions affecting vital signs. It is essential to keep the airway open to ensure respiration continuity and protect cardiac functions. 0.01 mg/kg/dose of 1/1000 adrenaline solution (maximum dose=0.5 mg) should be applied to the anterolateral region of the middle part of the thigh in severe anaphylactic reactions[15]. However, intravenous steroid and antihistamine treatment is the first choice in cases with less severe allergic reactions. After stabilizing the patient's vital signs, the necessary surgical

intervention should be performed quickly. After discharge, all patients should be treated with albendazole for 2 to 3 months to decrease recurrences and followed with serologic and imaging tests for at least six months[16]. In this case, the patient had a less severe allergic reaction. She was operated on for rupture of the hydatid cyst after appropriate treatment for the allergic reaction, and albendazole was prescribed after discharge.

Conclusion

Intra-abdominal rupture is a severe and rare complication of a hydatid cyst and should be kept in mind in patients with a history of hydatid cysts. The patient's vital signs should be reviewed quickly, and appropriate resuscitation therapy should be applied during anaphylactic shock. After the patients are stabilized, a diagnosis should be made quickly, and surgical treatment appropriate for the diagnosis should be performed as soon as possible.

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