

A Ruptured Cystic Echinococcosis in the Gallbladder and Intra/Extrahepatic Biliary Tract, Radiological and Surgical Imaging Findings

Safra Kesesi ve İntra/Ekstrahepatik Safra Yollarında Rüptüre Kistik Ekinokokkozisin Radyolojik ve Cerrahi Görüntüleme Bulguları

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ABSTRACT

Cystic echinococcosis is a common parasitic infestation that can still cause serious complications in endemic areas. Intrahepatic rupture is a well-defined complication, but rupture into the gallbladder is rare. The disease may present with cholecystitis and cholangitis. Clinicians and radiologists working in the emergency room will find the management of the disease much easier if they become familiar with the clinical and radiological findings of the cyst. In this article, a 28-year-old male admitted to the emergency department with acute abdominal pain who was examined for suspected acute cholecystitis and diagnosed with a rupture of the hydatid intra/extrahepatic bile ducts and gallbladder is presented. Our aim is to present the clinical findings and surgical images of the case (ultrasonography, computed tomography, magnetic resonance imaging) and compare them with the literature.

Keywords: Cystic echinococcosis, gallbladder, rupture, ultrasonography, computed tomography, magnetic resonance imaging

ÖZ

Kistik ekinokokkozis, endemik bölgelerde hala ciddi komplikasyonlara neden olabilen yaygın bir parazit enfeksiyonudur. İntrahepatik rüptür iyi bilinen bir komplikasyondur, ancak safra kesesine rüptür nadirdir. Hastalık kolesistit ve kolanjit ile kendini gösterebilir. Acil serviste çalışan klinisyenler ve radyologlar, kistin klinik ve radyolojik bulguları iyi bildikleri takdirde hastalığın tedavisini çok daha kolay yapacaklardır. Bu yazıda, akut karın ağrısı şikayeti ile acil servise başvuran, akut kolesistit şüphesiyle muayene edilen ve hidatik intra/ekstrahepatik safra yolları ve safra kesesi rüptürü tanısı alan 28 yaşında erkek hasta sunulmaktadır. Bu çalışmanın amacı olgunun klinik bulgularını ve cerrahi görüntülerini (ultrasonografi, bilgisayarlı tomografi, manyetik rezonans görüntüleme) sunmak ve literatürle karşılaştırmaktır.

Anahtar Kelimeler: Kistik ekinokokkozis, safra kesesi, rüptür, ultrasonografi, bilgisayarlı tomografi, manyetik rezonans görüntüleme

INTRODUCTION

Cystic echinococcosis (CE) is a parasitic infestation most commonly seen in endemic areas and it is caused by *Echinococcus granulosus*. Infection develops when parasitic eggs thrown into the external environment by final host dogs and canines are ingested by humans

and natural intermediate hosts, sheep, goats, cattle (1). These cysts are commonly seen in Europe, Mediterranean countries, Asia, the Middle and Far East, South America, and Australia (2).

The clinical signs and symptoms caused by CE depend on the location, size, developmental stage, pressure on

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the surrounding tissues and structures of the cyst and the defense mechanism of the infected person (3).

We present a case of hepatic CE intra-/extrahepatic biliary rupture, which causes obstructive jaundice. It was successfully treated surgically. In this case, our aim is to present the radiological findings and surgical management of parasitic disease, and images used to diagnose it.

CASE REPORT

A 28-year-old male presented to the general surgery clinic two weeks before with abdominal pain. His physical examination revealed pain and hepatomegaly in the epigastric region and palpation in the right upper quadrant of the abdomen. According to the patient's history, he lived in a village and was engaged in animal husbandry. The patient also reported nausea, vomiting, and anorexia. Since this region is endemic in terms of CE, it was suspected in the preliminary diagnosis, and his serum was tested for CE antibodies. Indirect hemagglutination assay (IHA) result was CE positive.

The computed tomography (CT) examination showed a type 3 CE lesion with multiple daughter vesicles and septa in the posterior segment of the right lobe of the liver (Figure 1a). Upon this, we decided to perform elective surgery due to the Coronavirus disease-2019 pandemic and his medical treatment started with albendazole 10-15 mg/kg/day.

Two weeks later, the patient presented to the emergency outpatient clinic with a sudden onset of pain in the right upper quadrant, where physical abdominal pain, tenderness in the abdomen, and cold sweating were observed.

The laboratory findings were as follows: White blood cell count (21.96×10^9) high, serum aspartate transaminase (85 U/L) and alanine aminotransferase (74 U/L) slightly high, direct bilirubin slightly high (0.43 mg/dL), amylase (40 U/L) within normal limits, and gamma-glutamyl transferase high (320 U/L).

Contrast-enhanced CT showed in the right lobe of the liver a connection between the intrahepatic bile ducts in the medial and inferior sections with daughter vesicles (Figure 1b).

Ultrasonographic (US) imaging revealed multiloculated hepatic cystic lesions and dilatation of the intra-/extrahepatic bile ducts. Increased gallbladder size and daughter vesicles were observed in the lumen and common biliary tract (Figure 2a, 2b).

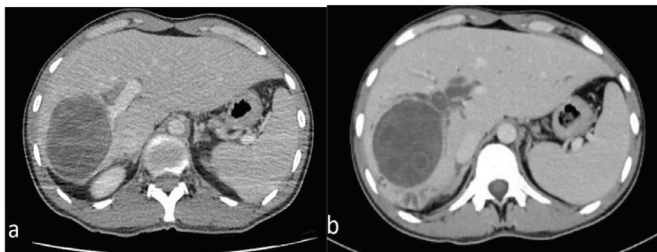


Figure 1a, b. Contrast-enhanced CT image of a 28-year-old male patient admitted to the emergency department with sudden onset abdominal pain two weeks before and at the time of presentation; type 3 cyst hydatid in the liver, before (a) and after rupture (b) into the intrahepatic biliary tract

CT: Computed tomography

Magnetic resonance imaging (MRI) showed a type 3 CE in the liver and daughter vesicles in the common hepatic duct lumen, as well as daughter vesicles in the gallbladder and common bile duct lumen (Figure 3a-c).

The patient underwent emergency laparotomy in the general surgery unit as well as cystectomy, cyst drainage, cholecystectomy, common bile duct exploration, and T tube drainage. During the operation, we observed CE rupture into the bile ducts and daughter vesicles in the common bile duct. In addition, there were multiple daughter vesicles and cysts in the hydropic gallbladder (Figure 4a, b).

After the operation, all laboratory findings returned to normal values and the patient recovered and was discharged.

DISCUSSION

CE is still an important zoonotic parasitic infection that threatens public health in the world and in our country. It has been reported that there has been a dramatic decrease in the incidence and prevalence of CE throughout the world, especially in recent years. In addition, it is stated that the negative effects of infection on public health and economy continue, especially in developing countries where animal husbandry is common (4,5).

The liver is the most common site of involvement of CE followed by the lungs. All other sites of involvement are considered as unusual, and they include the peritoneal cavity, spleen, kidney, uterus and adnexa, retroperitoneum, pancreas, brain, gallbladder

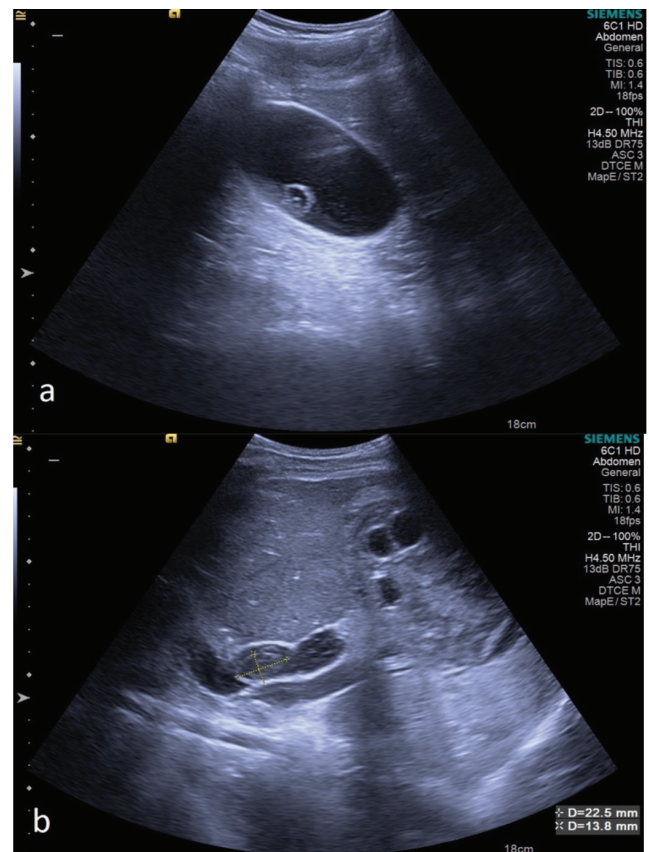


Figure 2a, b. On the ultrasonographic examination, the gallbladder is distended and cholecystitis is observed. Image showing daughter vesicles in the gallbladder lumen (a) and common bile duct lumen (b)

(<1%) and others (6). CE in the gallbladder is extremely unusual, even in endemic regions. CE may primarily be seen the gallbladder only in rare cases, or secondary invasion may be seen by daughter cysts of a previously infected liver. Depending on whether the cyst develops in the lumen of the gallbladder or on its external surface, the pathogenesis may vary. If the cyst is in the gallbladder lumen, the cysts have spread through the biliary duct, and if the cyst is on the gallbladder's wall, there has been spread through the lymphatic circulation (7). During the operation, we observed CE rupture into the bile ducts and daughter vesicles in the common bile duct. In addition, there were multiple daughter vesicles and cysts in the hydropic gallbladder.



Figure 3a. On MRI coronal examination (a); type 3 cyst hydatid (long arrow) in the liver and daughter vesicles (short arrow) in the gallbladder

MRI: Magnetic resonance imaging



Figure 3b. MRI sagittal T2 WI (b) shows daughter vesicles in the gallbladder and liver in images (arrow)

MRI: Magnetic resonance imaging

This parasitic disease can infect all tissues and organs and cause fatal complications. As an acute complication, it causes rupture due to pressure increase in the cyst due to blunt abdominal traumas, stressful situations, or severe cough or defecation causing intraabdominal pressure increase. With biliary rupture of liver hydatid cysts, the most obvious signs and symptoms are right upper quadrant pain, jaundice, and high temperature (8). In our case, sudden onset abdominal pain, nausea, vomiting, and laboratory white blood cell elevation were notable.

The most important clue for recognizing CE disease is the patient's history and clinical suspicion in endemic areas. Various radiological and serological methods can help in the diagnosis (9). Preoperative diagnosis of hepatic CE complications before ultrasonography (USG) or CT was difficult and was based on clinical and laboratory findings. It has been reported that the use of only radiological methods as a diagnostic method may be

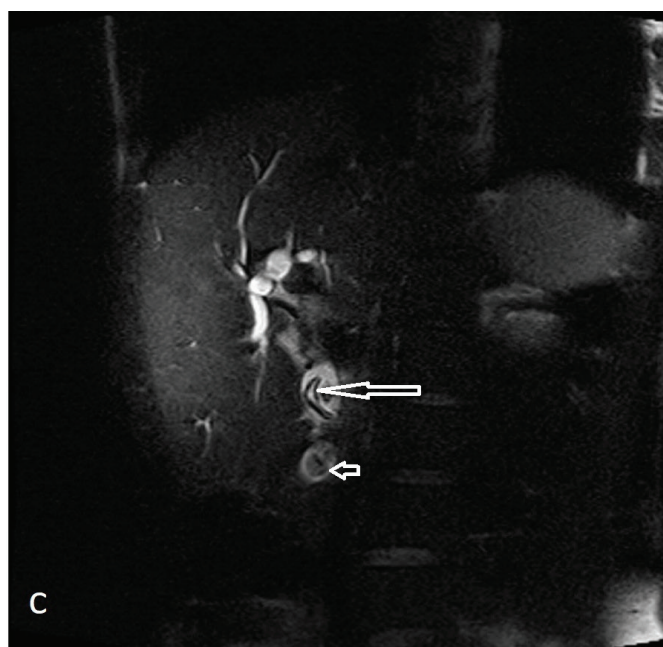


Figure 3c. In the coronal image (c) dissociate germinative membrane of the CE (long arrow) and daughter vesicles (short arrow) are observed in common bile duct lumen

CE: Cystic echinococcosis

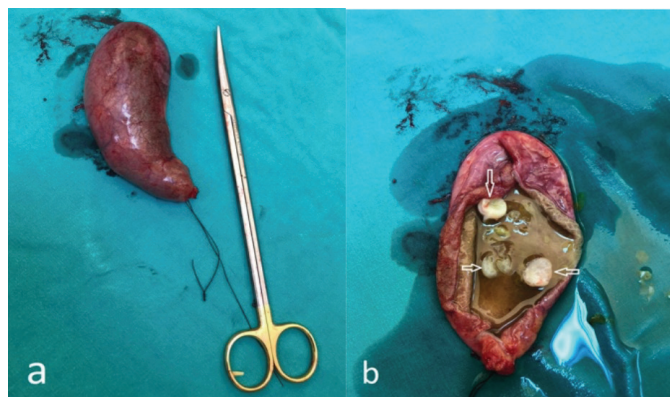


Figure 4a, b. Postoperative hydropic gallbladder specimen (a), showing hydatid daughter cysts in the gallbladder (b) (white arrow)

insufficient in the differential diagnosis of other lesions, and the use of only serological tests may be insufficient due to reasons such as individual immune response differences and cross-reaction, therefore it is necessary to use both diagnostic methods together (3). In our case, IHA from serological tests, USG, CT and MRI from radiological images were used for the diagnosis of CE. Nowadays, in the treatment of CE, interventional procedures appear to be less invasive and more comfortable in type 1-3 active cysts compared to Gharbi's US classification (10). If the cysts are very large or subcapsular or if complications have developed, the primary treatment is surgery.

CE is localized in many organs and tissues, especially the liver and lung. Many studies have been conducted on CE, which is located in different organs and tissues in our country. Some of these are as follows; Renal hydatid cyst was found in two patients who were admitted to the hospital with complaints of abdominal pain and swelling in the abdomen. In the USG evaluation of both cases, a cystic mass was observed and the cysts were surgically removed (11). Two pediatric patients admitted to the hospital with different complaints were diagnosed with hepatic echinococcosis by imaging methods and serological methods. Albendazole was followed by PAIR treatment in the treatment of the patients. No side effects and recurrences were observed in the treatment (12). In a study evaluating CE localizing in the pancreas, the patient with acute pancreatitis attack and splenomegaly was found to have pancreatic CE by imaging methods and IHA test. It has been reported that no postoperative complication developed in the patient who underwent albendazole and subsequent surgical intervention (13). In a study investigating whether CE could cause intra-familial infection; CE was detected in the lung and/or liver in a total of four individuals from the same family. A ruptured lung hydatid cyst and multiple cysts in the liver were found in the father from family members, and hydatid cysts in the liver were found in other children. It has been reported that genetic factors may affect the disease and that all family members of individuals living in risky areas should be evaluated for the disease (14). In another study; Although the patient with cough, fever and sputum complaints was initially interpreted as tuberculosis, as a result of the tests performed, a cystic lesion compatible with a hydatid cyst was detected in the left lobe of the liver in USG and CT. It was emphasized that the liver should also be evaluated with ultrasound in cases with suspected pulmonary CE, since the symptoms of ruptured lung CE are similar to tuberculosis (15).

In this study, the patient with abdominal pain, nausea, vomiting and loss of appetite was diagnosed with ruptured CE by CT, USG and MR imaging methods and IHA test. As in the literature reviewed (11,12,14,15), in this study, knowing the differential diagnosis features of CE and The importance of applying appropriate treatment was emphasized.

CONCLUSION

CE is still a common parasitic disease that can lead to serious complications in endemic areas. It can occur in many organs with different complications. Intrabiliary rupture is a well-defined complication, but rupture into the gallbladder is a rare and atypical finding. For clinicians and radiologists working in the emergency room management of the disease may be much easier if they become familiar with the clinical and radiological findings of the cyst.

* Ethics

Informed Consent: Informed consent was obtained.

Peer-review: Internally peer-reviewed.

* Authorship Contributions

Surgical and Medical Practices: S.T., Concept: İ.D., Design: L.T.Ç., Analysis or Interpretation: O.O., Literature Search: E.G., Writing: A.E.

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