

Liver Hydatid Cyst Rupture Into the Peritoneal Cavity After Abdominal Trauma: Case Report and Literature Review

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The aim of this study was to review the literature regarding the rupture of hydatid cysts into the abdominal cavity after trauma. We present both a new case of hydatid cyst rupture that occurred after blunt abdominal trauma and a literature review of studies published in the English language about hydatid cyst rupture after trauma; studies were accessed from PubMed, Google Scholar, EBSCO, EMBASE, and MEDLINE databases. We identified 22 articles published between 2000 and 2011 about hydatid cyst rupture after trauma. Of these, 5 articles were excluded because of insufficient data, duplication, or absence of intra-abdominal dissemination. The other 17 studies included 68 patients (38 males and 30 females) aged 8 to 76 years who had a ruptured hydatid cyst detected after trauma. The most common trauma included traffic accidents and falls. Despite optimal surgical and antihelminthic therapy, 7 patients developed recurrence. Complications included biliary fistula in 5 patients, incisional hernia in 2 patients, and gastrocutaneous fistula in 1 patient. Death occurred from intraoperative anaphylactic shock in 1 patient and gastrointestinal bleeding and pulmonary failure in 1 patient. Rupture of a hydatid cyst into the peritoneal cavity is rare and challenging for the surgeon. This condition is included in the differential diagnosis of the acute abdomen in endemic areas, especially in young patients.

Key words: Acute abdomen – Diagnosis – Peritonitis – Accident

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Hydatic cyst, also known as cystic echinococcosis, is a serious infection in endemic countries such as Turkey, where the prevalence was 585 per 100,000 people in 1991 and 291 per 100,000 people in 1999. It is a parasitic infection caused by the larval stage of *Echinococcus granulosus*.^{1–23} The primary carriers are dogs and wolves, and intermediate hosts are sheep, cattle, and deer. Humans are accidental hosts that do not contribute to the normal life cycle of this microorganism. Humans are infected by ingesting ova from soil or water contaminated by the feces of dogs.^{10,12,23} Hydatid cysts may develop in any body organ but occur most frequently in the liver (50%–77%) and lungs (18%–35%), and occasionally in other organs.^{1,3,9,10,17}

Hydatid cyst is a chronic, latent disease that often is asymptomatic. It may be found during routine clinical examination and serologic, radiographic, or ultrasonographic screening.^{1,3,7,15,18} The clinical signs and symptoms of hydatid cysts depend on location, size, relation to adjacent organs, and complications such as cyst rupture. A cyst may rupture into the biliary tree, a hollow organ, through the diaphragm into the pleural cavity, or directly into the peritoneal cavity.⁶ Cysts may rupture after trauma or spontaneously as a result of increased intracystic pressure.^{1,19} A hydatid cyst rupturing into the peritoneal cavity may cause complications including abdominal pain, urticaria, anaphylaxis, and sudden death.^{1,7,12} Therefore, hydatid cyst rupture requires both emergency surgery and careful postoperative care. In this report, we present the case of a patient who had a liver hydatid cyst that ruptured into the peritoneal cavity after blunt abdominal trauma.

Case Report

A 19-year-old man was admitted to our emergency clinic with intractable pain in the abdomen, nausea, and vomiting. The patient stated that he had been assaulted by 2 people 2 hours earlier, and had been kicked in the abdomen many times. The patient appeared exhausted and had multiple bruises, especially at the abdomen. The abdomen was slightly distended and did not move in tandem with respiration. Palpation of the abdomen showed guarding and rebound tenderness, primarily in the right upper quadrant.

The arterial blood pressure was 106/55 mmHg, pulse was 94 beats/min, respiratory rate was 26 breaths/min, white blood cell count was $17.6 \times 10^9/L$, hemoglobin was 148 g/L, and hematocrit was 42%.

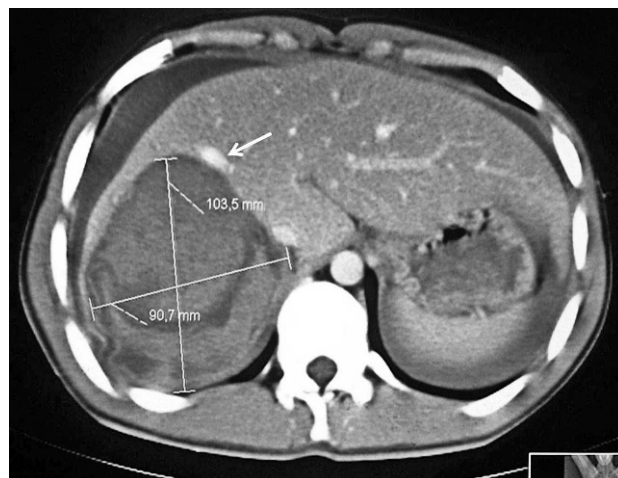


Fig. 1 Contrast-enhanced CT scan in a 19-year-old patient with a ruptured hydatid cyst after trauma. The large, high-density, cystic lesion is located in the right lobe of the liver and contains a floating germinative membrane (lotus flower sign). The lesion is adjacent to the posterior sectional branch of the right portal vein (white arrow). Free fluid levels are present throughout the abdomen, including the perihepatic and perisplenic areas.

Focused assessment with sonography in trauma (FAST) revealed a hyperechogenic area (7.5×7 cm) with lobulated contours in the posterosuperior segment of the right hepatic lobe; there was free fluid noted in all abdominal quadrants, most pronounced in the perihepatic region. Abdominal computed tomography (CT) showed a mass (10.4×9 cm) consistent with a ruptured hydatid cyst in the right lobe of the liver and free fluid throughout the abdomen (Fig. 1).

The patient developed dizziness, hypotension, and itchiness, consistent with anaphylaxis. He was treated with corticosteroids, pheniramine maleate, intravenous hydration, and nasal oxygen. A laparotomy was made through a right subcostal incision. Yellow serous fluid (1500 mL) was noted, containing many daughter vesicles disseminated throughout the abdomen. A mass consistent with a hydatid cyst was noted at the sixth and seventh hepatic segments and adherent to the lateral abdominal wall and diaphragm. The lateral wall of the cyst had ruptured and released some of its contents, including daughter vesicles, into the abdominal cavity.

The abdominal cavity was washed twice with cetrimide-chlorhexidine solution at 10-minute intervals. A pericystectomy was done, followed by removal of both the germinative membrane lining the cystic cavity and the daughter vesicles from the abdominal cavity. Further inspection showed fistula formation between 2 large bile ducts and the cyst

Table 1 Clinical features of 68 patients who had hydatid cyst rupture after trauma^a

Reference	Age (y)	Sex	Causes	Preoperative duration	White blood cell count ($\times 10^9/L$)	Diagnostic tools	Cyst location	Cyst diameter (cm)	Complications	Duration of albendazole (mo)	Recurrence (No. of patients)	Follow-up (mo)
Dirican <i>et al</i> ¹	24	M	Fall	90 h	12	US, CT	Bilobar	11	None	3	None	40
	29	M	Fall	4 h	10.7	CT	Pelvic	8	None	3	None	30
	37	M	Shotgun	1 h	13	Laparotomy	Spleen	9	None	3	None	56
Elmali <i>et al</i> ²	15	M	Fall	30 min	NS	US	Right	4	Nonoperative management with albendazole			
Feleppa <i>et al</i> ³	8	M	Trauma	NS	18.5	US, CT	Left	6	None	3	None	24
Rami <i>et al</i> ⁴	10	M	Fall	NS	15	US	NS	NS	None	6	None	36
	11	F	Trauma	48 h	14.3	US	NS	NS	None	6	None	36
	12	M	Fall	NS	11.2	US	NS	12	None	6	None	9
	11	F	Trauma	NS	13	US, CT	NS	NS	None	6	None	6
Bari <i>et al</i> ⁵	40	F	Trauma (delayed)	3 y	NS	Laparotomy	Right	NS	None	NS	None	6
Erel <i>et al</i> ⁶	19	M	Trauma	2 h	17	US, DPL	Right	NS	None	2	None	18
Kara <i>et al</i> ⁸	54	M	Fall	48 h	23	US	NS	10	None	6	None	NS
Shah <i>et al</i> ⁹	45	F	Trauma (delayed)	5 y	NS	US, CT, laparotomy	Left	1–12	Died from anaphylactic shock			
Ozturk <i>et al</i> ¹⁰	35.7 (17–75)	M: 11; F: 9	Traffic accident; assault victim; minor trauma: 6	NS	11–23	CT: 13; US: 17; DPL: 7	Right: 18; left: 2	12 (10–20)	Bile fistula: 4; mortality: 1	2 (all patients)	4	47 (8–57)
Akcan <i>et al</i> ¹¹	37 (17–76)	M: 14; F: 13	Traffic accident; fall; pedestrian mishap: 6	4.4 h (0.5–24 h)	NS	US: 27; CT: 11	Right: 16; left: 5; bilateral: 6	NS	Bile fistula: 1; hernia: 2; gastrocutaneous fistula: 1	3	3	722 (4–131)
Gulalp <i>et al</i> ¹²	18	F	Fall	15 min	18.1	US	Right	7.2	None	6	None	NS
Eren <i>et al</i> ¹³	40	F	Fall	NS	Normal	US, CT	Right	20	NS	3	NS	NS
Papazioglu <i>et al</i> ¹⁴	39	M	Traffic accident	9 y	21	CT	Multiple	5–15	None	6	None	12
Kurt <i>et al</i> ¹⁵	8	F	Fall	8 h	17.1	US, DPL	Left	7	None	3	None	56
	14	M	Fall	7 h	9	CT	Left	4	None	3	None	4
Koyuncu <i>et al</i> ¹⁶	25	M	Assault victim	4 h	16.2	US	Bilobar	5–10	None	Yes	None	4
Doganay <i>et al</i> ¹⁷	12	F	Trauma	NS	NS	US	Bilobar	7.4, 5.6	None	1	None	12
Velitchkov <i>et al</i> ¹⁸	13	M	Fall	72 h	12.1	US	Right	8	None	2	None	34

DPL, diagnostic peritoneal lavage; F, female; M, male; NS, not stated; US, ultrasonography.

^aReported in the English language between 2000 and 2011.

cavity. The orifices of both bile canals were sealed primarily with prolene suture, and an omentoplasty was performed. The common bile duct was explored, and a T-tube was inserted to decompress the biliary tree. The surgical procedure was completed after further irrigation of the entire abdominal cavity with cetrimide-chlorhexidine solution. After surgery, the patient was started on albendazole (10 mg/kg) for 8 weeks.

Literature Review

A search of the PubMed, Google Scholar, MEDLINE, EMBASE, and EBSCO databases was performed with combinations of the search terms "hydatid cyst," "liver," "rupture," "trauma," "blunt," and "peritoneal cavity." This provided 22 full-text English language articles published between 2000 and 2011.¹⁻²² Of these, 5 articles were excluded because of insufficient data about 15 patients, duplication of 1 case, and dissemination of the contents of the ruptured cyst into the biliary tree instead of the abdominal cavity in 1 patient.^{7,19-22} The other 17 studies included 68 patients (38 males and 30 females) aged 8 to 76 years who had a ruptured hydatid cyst detected after trauma (Table 1). The most common types of trauma were traffic accidents (25 patients) and falls from a height (18 patients). There were 2 patients who were admitted because symptoms developed long after the index trauma: 3 years in 1 patient and 5 years in 1 patient. Treatment included surgery in 67 patients, and 1 patient had nonoperative treatment with albendazole and supportive therapy.

Despite reportedly optimal surgical and medical therapy, 7 patients developed recurrence, either in the same hepatic lobe as the initial disease (6 patients) or in the lung (1 patient). Complications included biliary fistula (5 patients), incisional hernia (2 patients), and gastrocutaneous fistula (1 patient). The biliary drainage resolved after endoscopic sphincterotomy (4 patients) or spontaneously (1 patient). The patient with a gastrocutaneous fistula was treated with excision of the fistula tract, truncal vagotomy, and pyloroplasty. Death occurred from intraoperative anaphylactic shock in 1 patient and from gastrointestinal bleeding and pulmonary failure in the intensive care unit in 1 patient.

Discussion

Complications occur in 5% to 40% patients with hepatic hydatid cysts and include the formation of

small cystobiliary fistulas, cyst rupture into the biliary tree, biliary compression, cyst infection, hydatid allergy, membranous glomerulitis, and intraperitoneal rupture.^{12,15,18} The reported frequency of liver hydatid cyst rupture into the peritoneal cavity ranges from 1% to 16%.^{1,3,15,19,21,22} Rupture may result from trauma or may occur spontaneously from increased pressure of the cystic fluid. The main risk factors predisposing to rupture include young age, cyst diameter >10 cm, and superficial cyst location. Young age is a risk factor because of the greater frequency of traumatic events and higher prevalence of hydatid disease in children and adolescents than in adults. Increased cyst diameter, which increases the internal tension, and superficial location of the cyst are important risk factors for rupture, even with minor trauma.^{1,3}

The clinical signs and symptoms of hydatid cyst rupture occasionally are not severe, but hydatid fluid or free bile can cause peritonitis, as occurred in the present patient. Furthermore, peritoneal signs and symptoms may develop earlier and can be more severe if bile leakage occurs or the cyst is infected.^{1,7,10,11} Urticaria and macular eruptions on the skin may result from an allergic reaction to the cyst contents. The patient may develop anaphylaxis, including life-threatening anaphylactic shock.⁶

Although the reported frequency of minor allergic reactions after traumatic or spontaneous rupture of hydatid cysts ranges from 16.7% to 25%, the incidence of more severe reactions is 1% to 12.5%.^{3,11,21} Anaphylactic shock, albeit rare, can develop with no overt evidence of macroscopic rupture²⁰ and may be explained by rupture of the cyst into the bile canals spontaneously or after trauma, or after indirect release of the cyst contents into the blood circulation.²⁰ In the literature search, we found anaphylactoid reactions manifesting as urticaria, peripheral edema, dyspnea, and syncope in 13 of 68 patients (19%). Timely surgery and appropriate medical intervention prevented the death of all except 1 patient with anaphylaxis. The incidence of life-threatening anaphylactic shock was 1.4%. These findings suggest that allergic reactions may be infrequent, but it may be prudent to maintain awareness of the potential for developing anaphylaxis. Therefore, immediate medical treatment for allergic reactions is advised.¹¹

There is no optimal treatment option for cystic echinococcosis, and no clinical trial has compared the different treatment modalities. Treatment indications are complex and based on cyst characteristics, available medical and surgical expertise and

equipment, and adherence of patients to long-term monitoring.²⁴ Surgery is carefully evaluated against other options, and it is the first choice for complicated cysts. The surgical management of a patient with a ruptured hydatid cyst is more complex than an unruptured cyst. Scolices can spill into the abdomen during emergency surgery for ruptured cysts, and the surgeon addresses both the disease in the liver and the removal of intra-abdominal protoscoleces.^{1,15} The main goals of surgery are to eliminate local disease, prevent complications, and minimize the morbidity, mortality, and recurrence risk.¹³ In patients with uncomplicated hydatid cysts, there is controversy between the benefits of a radical or conservative approach because each method has advantages and disadvantages. Cavity complications and recurrence are the major problems with conservative methods, but radical procedures have greater operative risks, such as bleeding and morbidity.^{1,6,15,19} After intervention for a ruptured cyst, it is important to irrigate the peritoneal cavity with sufficient scolical agents and perform careful, meticulous removal of all cystic contents. Scolical agents include cetrimide-chlorhexidine, povidone-iodine (10%), silver nitrate (0.5%), hypertonic saline solution (3%–30%), chlorhexidine (0.4%), and praziquantel.^{1,11,16} For several years, we have used cetrimide-chlorhexidine diluted 1:30 for the cyst cavity and diluted 1:1000 for the intra-abdominal spaces.^{1,7}

Bile leakage is a potential problem after hydatid cyst surgery. This complication can be prevented if the connection between the cyst and the biliary tract is recognized and treated. Therefore, surgeons should search for such a connection during the initial operation.¹⁵ In the literature search, the incidence of postoperative biliary leakage was 7.3%. When a passage is identified intraoperatively between the biliary tract and the cystic cavity, treatment may include suturing the bile duct, omentopexy, effective external drainage, and insertion of a T-tube for decompression. When a bile leakage is detected postoperatively, optimal treatment to decompress the biliary tree may include endoscopic retrograde cholangiopancreatography (ERCP)-guided sphincterotomy, nasobiliary drainage, or internal biliary stenting.

Anthelmintic treatment is started 2 to 3 weeks before elective hydatid cyst surgery and is continued for 6 to 8 weeks postoperatively. However, patients with traumatic or spontaneous cyst rupture may require emergency surgery and cannot receive medical treatment before surgery; therefore, medical treatment may be started as early as possible after

surgery and continued for 1 to 6 months, depending on individual circumstances, to reduce the risk of recurrence. Albendazole (10–15 mg/kg/d) is the drug used most commonly in previous studies and was used in the present patient.^{1–20}

A ruptured hydatid cyst requires meticulous postoperative follow-up. Patients with uncomplicated hydatid cysts may be followed with ultrasonographic examination and indirect hemagglutination test starting 6 months after surgery and subsequently repeated every 1 to 2 years; those with ruptured cysts are followed at shorter intervals. A CT scan may show recurrence. Cysts that were overlooked during surgery may be erroneously interpreted as recurrences during long-term follow-up.^{1,19} Recurrences may also occur because of insufficient surgery or medical treatment after rupture of a hydatid cyst. Recurrence has been reported in 6.7% to 28.6% cases.^{15,19,21,22} In the 68 cases reviewed, the frequency of postoperative recurrence (10.2%) was similar to that previously reported.

The rupture of a hydatid cyst into the peritoneal cavity is rare but presents a challenge for the surgeon. This condition is included in the differential diagnosis of the acute abdomen in endemic areas, especially in young patients. Emergency surgery is the main treatment for intraperitoneal rupture of hydatid cysts, and medical treatment should be given postoperatively.

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