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## Intraperitoneal rupture of the hydatid cyst: Four case reports and literature review

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### Abstract

#### BACKGROUND

Most patients with hydatid cysts are asymptomatic, and they are diagnosed incidentally during radiological evaluations performed for other reasons. However, some patients develop symptoms and complications due to cyst size, location, and the relationship between the cyst and adjacent structures. The most serious complications that can occur are rupture of the cysts into the biliary tract, vascular structures, hollow viscus, and peritoneal cavity. We aimed to describe the management of four cases of intraperitoneal rupture of hydatid cysts.

#### CASE SUMMARIES

Four patients aged between 27 and 44 years (two men and two women) were admitted to our clinic with sudden abdominal pain ( $n = 4$ ), hypotension ( $n = 3$ ), and anaphylaxis ( $n = 2$ ). Three of the perforated cysts were located in the liver, and one was located in the spleen. Two patients developed cyst rupture after minor trauma, and the other two developed spontaneous rupture. Enzyme-linked immunosorbent assay IgG results were positive for two patients and negative for the other two. All patients received albendazole treatment after surgical intervention (range: 2-6 mo). Two patients developed hepatic abscesses requiring drainage; one of these patients also developed hydatid cyst recurrence during postoperative follow-up (range: 25-80 mo).

#### CONCLUSION

Intraperitoneal rupture is a life-threatening complication of hydatid cysts. It is important to manage patients with surgical intervention as soon as possible with aggressive medical treatment for anaphylactic reactions.

**Key words:** Hydatid cyst; Complication; Intraperitoneal rupture; Spontaneous rupture; Traumatic rupture; Anaphylactic reactions; Case report

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**Core tip:** Spontaneous or traumatic intraperitoneal hydatid cyst rupture is a rare but life-threatening complication. Therefore, hydatid cyst rupture should be considered as a differential diagnosis in patients who have sudden onset abdominal pain and allergic reactions like urticaria, especially those in regions endemic to the disease. Deteriorated hemodynamic parameters due to anaphylactic reactions should be corrected quickly with subsequent emergent surgery for a life-saving procedure. Herein, we aimed to present the management algorithm of four patients diagnosed with intraperitoneal hydatid cyst rupture.

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## INTRODUCTION

Hydatid disease is a zoonotic disease caused by the echinococcus parasite, which is a member of the Taeniidae family. Most common echinococcus species that cause hydatid disease in humans are *Echinococcus granulosus* (cystic echinococcosis) and *Echinococcus multilocularis* (alveolar echinococcosis). Cystic echinococcosis, also known as hydatid cysts, constitute 95% of all hydatid diseases [1-3]. Hydatid diseases can develop in almost all organs and tissues of the human body, but the most frequently involved organs are the liver (50%-77%), lungs (15%-47%), spleen (0.5%-8%), and kidneys (2%-4%) [1-6]. Humans, who have no role in the biological life cycle of echinococcosis, can accidentally become intermediate hosts by ingesting tapeworm eggs [1,4]. Hydatid cysts can grow at an average of 10-50 mm/year depending on the location of the cyst, so most patients remain asymptomatic for years [3,7,8]. Usually, asymptomatic patients are diagnosed incidentally by radiological evaluations that are performed for other reasons. Nevertheless, some patients have signs and symptoms, including mild or severe abdominal pain, nausea, vomiting, jaundice due to size, location, and/or involvement of the adjacent structures and organs of the cystic lesions [1]. The most frequently reported complications are rupture (perforation), bacterial infection, anaphylactic reaction, compression of the vascular and biliary structures, and compression of the neighboring organs [4,5]. Hydatid cyst rupture can be internal (cysto-biliary fistula, rupture into the hollow viscus, broncho-biliary fistula, bronchopleural fistula, intrapericardial rupture, intrapleural rupture, intraperitoneal rupture) or more rarely, it can be external (cysto-cutaneous fistula) [3]. Intraperitoneal rupture is a rare but life-threatening, severe complication of hydatid cyst disease [3]. We aim to describe the cases of four patients who developed intraperitoneal rupture and review the medical literature.

## CASE PRESENTATION

The summary information of the four cases described below is given in [Table 1](#).

### Case 1: Chief complaints

A 44-year-old man was admitted to the emergency department with sudden onset abdominal pain, skin flushing at some areas of the abdomen and hypotensive shock without any history of trauma.

### History of present illness

The patient was diagnosed with obstructive jaundice complicated by a hydatid cyst approximately one month ago. The connection between the biliary tract and hydatid cyst was visualized by radiological examinations. Endoscopic retrograde cholangiopancreatography (ERCP) was performed to decompress the biliary tract before surgical intervention. Albendazole treatment was initiated before the surgery.

### History of past illness

The patient had no medical disease other than the hydatid cyst.

### Laboratory examinations

**Table 1** Demographic and clinical characteristics of four patients with hydatid cyst perforation

Parameters	Case 1	Case 2	Case 3	Case 4
Age	44	37	27	40
Gender	Male	Male	Female	Female
Cause of perforation	Spontaneous	Trauma	Spontaneous	Trauma
Clinical presentation	Pain (sudden)	Pain (sudden)	Pain (sudden)	Pain (sudden)
	Hypotension	Hypotension	Hypotension	Anaphylaxis
	Anaphylaxis	Anaphylaxis		Not clear
Serology	ELISA IgG (+)	ELISA IgG (+)	Negative	Negative
History of HD	Known	Unknown	Unknown	Known
Cyst location	Liver (Bilobar)	Spleen	Liver (Bilobar)	Liver (Bilobar)
		Retroperitoneum		
		Retrovesical		
Perforated cyst location	Liver (VI)	Spleen	Liver (III)	Liver (II-VI)
Cysts size (mm)	165 × 100 × 80	50 × 45	80 × 75	110 × 85
	130 × 100 × 95	50 × 40	40 × 10	30 × 25
	85 × 70 × 65	60 × 50		
Preop antihelminthic	ALBZ (1 mo)	No	No	ALBZ (? mo)
Postop antihelminthic	ALBZ (6 mo)	ALBZ (2 mo)	ALBZ (2 mo)	ALBZ (2 mo)
Diagnostic tools	US + CT	US	US + CT	CT
Surgical approach	Partial Cystectomy+	Splenectomy+	Partial Cystectomy+	Partial Cystectomy+
	CBD exploration+	Retroperitoneal+	Omentopexy	Omentopexy
	T-tube insertion+	Retrovesical		
	Omentopexy	Cystectomy		
Recurrence	Yes	No	No	No
Postop complication	Liver abscess	Bleeding	No	Liver abscess
Follow up (mo)	80	34	25	36

HD: Hydatid disease; ALBZ: Albendazole; US: Ultrasonography; CT: Computed tomography; CBD: Common bile duct.

Complete blood count (CBC) was as follows: White blood cells (WBCs): 12900/mL, neutrophils: 80%, and eosinophils: 0.9%. Test results for biochemical analysis were as follows: Aspartate aminotransferase (AST), 152 (5-34 U/L); alanine aminotransferase (ALT), 58 (5-34 U/L); alkaline phosphatase (ALP), 260 (40-150 U/L); gamma-glutamyl transferase (GGT) 125, (12-64 U/L); and total bilirubin, 1.03 (0.2-1.2 mg/dL).

### Imaging examinations

Abdominal ultrasonography (US) and contrast-enhanced abdominal computed tomography (CT) showed that multiple cystic lesions compatible with hydatid cyst disease (largest size 165 mm × 100 mm × 80 mm) were localized in the liver. CT scans also showed that the ruptured cystic lesion was localized in the VI segment of the liver (Figures 1 and 2).

## FINAL DIAGNOSIS

The final diagnosis of the present case was intraperitoneal liver hydatid cyst rupture.

## TREATMENT

Fluid resuscitation, antihistaminic, and corticosteroid treatment was initiated at the emergency department. Laparotomy was performed due to the patient's general condition. Intraoperatively, the cyst located in the liver ruptured, and 2500 mL of fluid containing cyst material was aspirated from the peritoneal cavity. The peritoneal cavity was washed with saline solution and partial cystectomy, omentoplasty, and common bile duct exploration with T-tube placement were performed.



**Figure 1** Coronal reformatted contrast-enhanced computed tomography shows that fluid collection and daughter vesicles adjacent to the hydatid cyst located in segment VI of the liver. This finding is consistent with a perforated hydatid cyst.

## OUTCOME AND FOLLOW-UP

The patient developed an abscess that required drainage in the postoperative period. Six albendazole treatments with 3-wk cycles were administered as medical treatment. The T-tube was removed when normal biliary tract anatomy was visualized during cholangiography, approximately three weeks postoperative.

### **Case 2: Chief complaints**

A 37-year-old man presented in our emergency unit with severe abdominal pain and anaphylactic shock signs/symptoms, such as hypotension and allergic reaction.

### **History of present illness**

The patient suddenly felt sharp abdominal pain after playing with and tossing his nephew and simultaneously felt like something was leaking inside him. Subsequently, dizziness and diffuse flushing occurred.

### **Laboratory examinations**

CBC analysis revealed leukocytosis (18800/mL), with predominant neutrophils (86.4%) and normal eosinophils (0.1%). Biochemical blood test results were within normal limits except for total bilirubin (1.5 mg/dL).

### **Imaging examinations**

US showed massive intraabdominal fluid collections in the perihepatic and perisplenic area. Additionally, a ruptured and degenerated cyst with a germinative membrane collapse near the spleen was visualized.

## FINAL DIAGNOSIS

The final diagnosis of the present case was intraperitoneal splenic hydatid cyst rupture.

## TREATMENT

Fluid resuscitation, antihistaminic, and corticosteroid treatment was initiated at the emergency department. The patient was then taken to the operating room, and laparotomy was performed with a midline incision. An exophytic, ruptured hydatid



**Figure 2** Axial plane computed tomography of the same patient shows exophytic extension of the giant hydatid cyst located in the right lobe of the liver.

cyst originating from the spleen, which had adhesions to adjacent structures, was identified. Total splenectomy was required (Figures 3 and 4). An iatrogenic left diaphragm injury due to intraabdominal adhesions occurred during surgery. A left-sided chest tube was inserted after the primary repair of the diaphragm. Additionally, other cystic lesions located at the posterior side of the urinary bladder (60 mm × 50 mm) and at the retroperitoneum (50 mm × 40 mm) were completely excised. A chest tube was removed postoperatively on the fourth day.

## OUTCOME AND FOLLOW-UP

The patient had an uneventful postoperative clinical course. *Pneumococcus* vaccination was administered 14 d after surgery. Continuous albendazole treatment was administered for two months after surgery.

### Case 3: Chief complaints

A 27-year-old woman experienced sudden onset abdominal pain at another medical center. She was diagnosed with a ruptured hydatid cyst and was referred to our medical center. She had hypotension during her first evaluation at the emergency unit.

### History of present illness

She had no history of hydatid cyst.

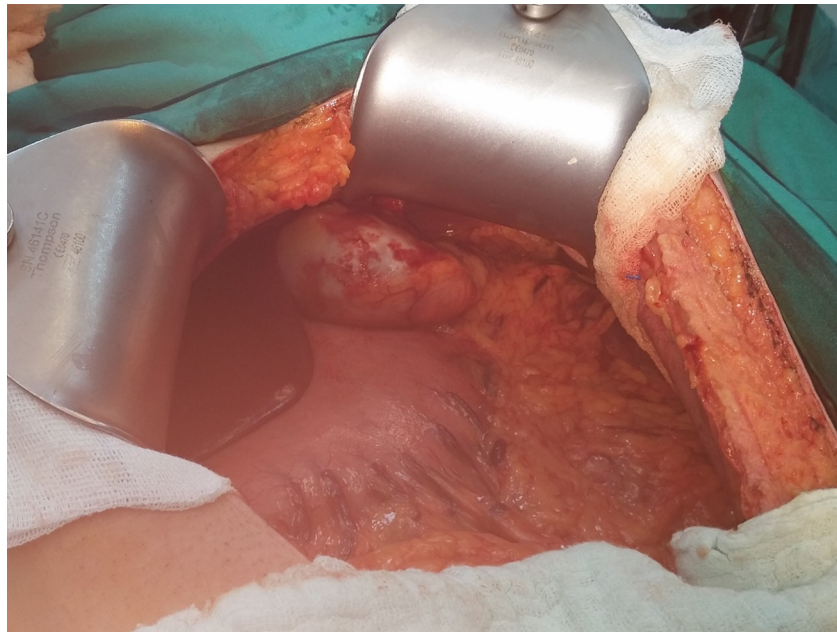
### Laboratory examinations

CBC analysis showed leukocytosis (24100/mL) with predominant neutrophils (94.5%) and normal eosinophils (0.2%). Biochemical blood tests were within normal limits.

### Imaging examinations

Contrast-enhanced abdominal CT revealed that one lesion was compatible with a hydatid cyst in the anterior sector of the liver (80 mm × 75 mm × 70 mm) and another one at segment III (40 mm × 10 mm). Additionally, several daughter cystic lesions (largest diameter 130 mm × 60 mm × 50 mm) secondary to the rupture of the cystic lesion near segment III were observed in the pelvic cavity (Figure 5).

## FINAL DIAGNOSIS



**Figure 3** Intraoperative appearance of the hydatid cyst compatible lesion that originated and ruptured from the spleen. This image was taken after aspiration of the hydatid cyst fluid in the abdominal cavity.

The final diagnosis of the present case was an intraperitoneal liver hydatid cyst rupture.

## TREATMENT

Fluid resuscitation was initiated at the emergency department. The patient was then taken to the operating room. The anterior wall of the cyst located in the anterior sector of the liver was excised, and the cystic components were completely evacuated. Three bile ducts that opened into the cystic cavity were closed with polypropylene sutures. Afterwards, the hydatid cyst wall localized next to segment III was excised, and the cyst components were completely evacuated. Cholecystectomy was performed to place high pressure saline solution into the common bile duct via cystic duct; there was no bile leak. Daughter vesicles located in the pelvic cavity were also removed, and the peritoneal cavity was washed with hypertonic saline solution.

## OUTCOME AND FOLLOW-UP

The patient had an uneventful postoperative clinical course. Postoperative albendazole treatment was administered for two months.

### Case 4: Chief complaints

A 40-year-old woman was suffering from abdominal pain for one month, and her pain became aggravated for two days prior to admission. Physical examination revealed widespread tenderness in all quadrants of the abdomen.

### History of present illness

The patient stated that she had received antihelminthic medical treatment several times.

### Laboratory examinations

CBC analysis showed leukocytosis (WBC: 23400/mm<sup>3</sup>, neutrophils: 90.6%, and eosinophils: 0.4%). Biochemical results were not altered except for ALP and GGT levels (AST: 18 U/L, ALT: 18 U/L, ALP: 197 U/L, GGT: 114 U/L, and total bilirubin 0.87 mg/dL).

### Imaging examinations

Contrast-enhanced abdominal CT revealed several hydatid cysts (largest diameter 10 mm × 85 mm × 60 mm) in the left lobe of the liver. Cysts located in the left lobe of the



**Figure 4** Appearance of the spleen and ruptured cyst specimens obtained from the same patient.

liver compressed the main bile duct, causing dilation of the intrahepatic bile ducts of the left liver lobe (Figure 6). Several cystic lesions were also observed in the pelvic cavity.

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## FINAL DIAGNOSIS

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The final diagnosis of the present case was intraperitoneal liver hydatid cyst rupture.

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## TREATMENT

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There were several adhesions secondary to the ruptured cyst, and they were diagnosed during laparotomy as sclerosing encapsulated peritonitis (Figure 7). After adhesiolysis, the perforated wall of the cyst was excised, and the cyst components were completely evacuated (Figure 8). Additionally, cystic components located in the pelvic cavity were removed.

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## OUTCOME AND FOLLOW-UP

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The patient had an uneventful postoperative clinical course and was treated with continuous albendazole for two months. The patient developed left lobe liver abscess in the third postoperative year, which required percutaneous drainage.

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## DISCUSSION

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About 5%-40% of the hydatid cysts located in the liver and other organs of the body may cause various complications. The most common complications related with hydatid cysts include superinfection, cysto-biliary fistula (obstructive jaundice, cholangitis), allergic reactions, rupture into the gastrointestinal system (duodenum, small intestine, colon), Budd Chiari syndrome, portal hypertension, gastric outlet obstruction, membranous glomerulonephritis, broncho-biliary fistula, bronchopleural fistula, intrapericardial rupture, intrapleural rupture, and intraperitoneal rupture<sup>[1,3,7,9]</sup>.



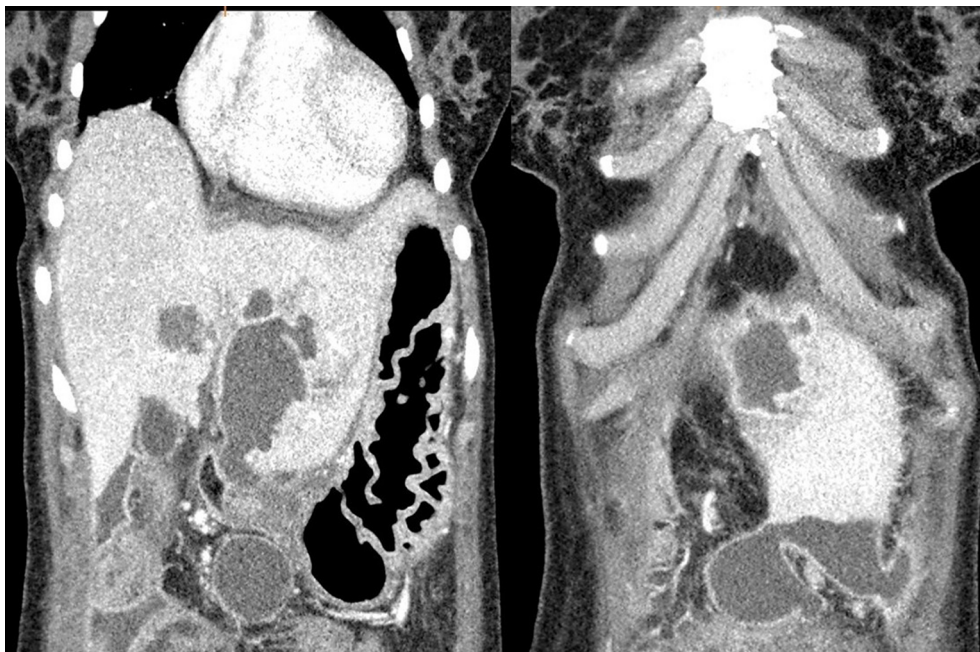
**Figure 5** Coronal reformatted images of the venous phase of computed tomography shows perforated hydatid cyst located in segment V of the liver. This image also shows fluid collection in the right paracolic area.

Intraperitoneal hydatid cyst rupture may result from trauma or may spontaneously occur due to increased intracystic pressure [1,10,11]. Sometimes, iatrogenic hydatid cyst rupture may occur during elective surgery. Some studies have concluded that most intraperitoneal ruptures develop after trauma; nevertheless, other studies pointed out that spontaneous ruptures may develop more frequently [6,7,12].

Intraperitoneal rupture is the third most common complication (0.9%-16%) after intrabiliary rupture (5%-25%) and allergic reactions (1%-25%) [1,4,7]. Most studies regarding intraperitoneal hydatid cyst rupture have been published as case reports. To our knowledge, four or more cases were reported in a limited number of studies [4,6,9,13-22] (Table 2). Risk factors for rupture include young age, cyst diameter (> 10 cm), and superficial localization [1,4,7]. Hydatid cysts are more common in younger individuals because they are exposed to traffic accidents more often than older individuals. Intracystic pressure increases with increasing cyst dimension. When intracystic pressure increases to more than 50 cmH<sub>2</sub>O, spontaneous or traumatic rupture risk also increases [6,10]. Most superficial cyst walls are not protected by the liver parenchyma. This is a facilitating factor for rupture of the cyst into peritoneal cavity or hollow gastrointestinal organs [1,4]. The aims of emergent surgical treatment include prevention or minimization of anaphylactic reactions in the early term and prevention of the development of long-term secondary peritoneal hydatidosis [4,7].

Clinical signs and symptoms of intraperitoneal cyst ruptures may vary widely among patients. The most frequent symptoms are mild or severe abdominal pain, vomiting, nausea, and some allergic reactions that span a wide range. If the content of the ruptured cyst is purulent or associated with the biliary tract, it may cause peritoneal irritation. Therefore, clinical signs of acute abdominal pain may occur [1,4]. A wide spectrum of life-threatening immunologic reactions, such as allergic reactions and/or anaphylactic shock, develop against the cyst content, which spread into the peritoneal cavity [1,4]. Therefore, some patients may have complaints such as hypotension, tachycardia, and respiratory distress [1].

While 16.7%-25% of patients with hydatid cyst rupture develop minor allergic reactions, such as urticaria and macular eruption, 1%-12.5% of patients develop more severe allergic reactions such as peripheral edema, syncope, and anaphylaxis [1,6]. The life-threatening anaphylactic shock incidence rate is approximately 1.4%. Allergic



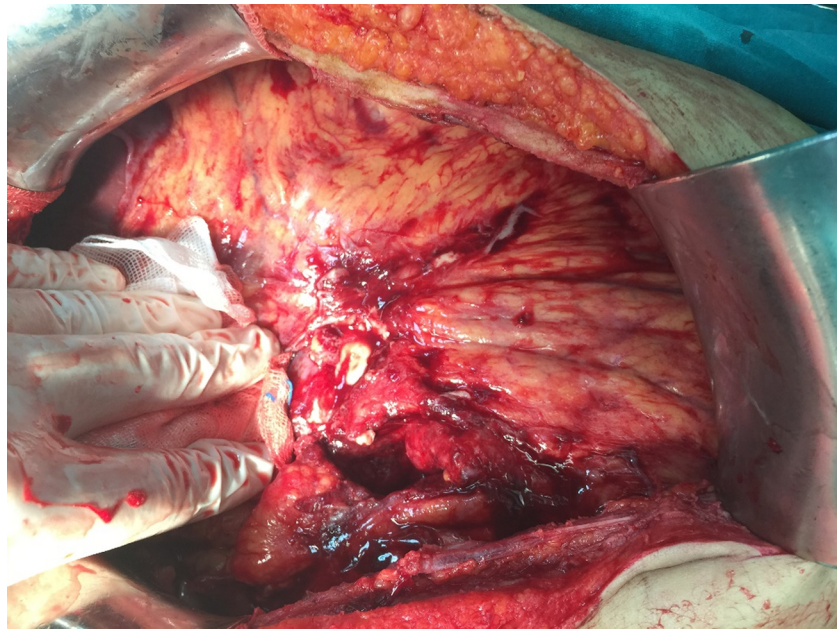
**Figure 6** Two different coronal reformatted contrast-enhanced computed tomography images of the same patient show a perforated hydatid cyst located in segment III of the liver and fluid collection in the perihepatic/pelvic area.

reactions may develop in macroscopic ruptures or when the cyst contents pass into the biliary tract after trauma, iatrogenic interventions, or spontaneously. Even allergic reactions may occur after direct communication when the cyst contents enter systemic circulation [1]. Anaphylactic reactions during cyst hydatid surgery may develop in 0.2%-3.3% of patients who do not experience rupture [1].

Most patients who have intraperitoneal rupture are admitted to the emergency unit with symptoms such as severe abdominal pain, hypotension, tachycardia, and allergic reactions [1,10,11]. Therefore, US is the most common radiologic tool for diagnosis. If patients are hemodynamically stable, contrast-enhanced CT may be used. The sensitivity of US and CT is 85% and 100%, respectively [1,8,9].

Medical treatment should be initiated as soon as possible at the emergency unit after confirming the diagnosis of intraperitoneal rupture. Moreover, it should be continued during surgery. To minimize morbidity and mortality, patients should be hemodynamically stabilized before surgery, and they should undergo surgery as soon as possible. Medical treatment is aimed at stabilizing hemodynamic status with fluid resuscitation and treating anaphylactic reactions with corticosteroids, antihistaminic, and vasopressor drugs.

There is no consensus in the literature about surgical treatment options for intraperitoneal cyst rupture [7,12]. We suggest that each case should be evaluated separately in accordance with the general principles of hydatid cyst surgery. Simply summarized, hemodynamically stable patients should undergo either laparoscopic or open surgery as soon as possible, and hemodynamically unstable patients should undergo open surgery. Cyst contents that trigger anaphylactic reactions should be removed from the abdominal cavity as soon as possible [1,5-7]. The peritoneal cavity should be washed with scolicidal solutions such as formaldehyde, hypertonic saline (3%-10%-15%-30%), silver nitrate (0.5%), cetrimide, chlorhexidine, cetrimide plus chlorhexidine (1.5%/0.15%), hydrogen peroxide (1.5%-3%), povidone iodine (10%-50%), or ethyl alcohol (70%-95%). Each solution has a different time frame for possible scolicidal effects [1,5-7]. We prefer hypertonic saline or cetrimide plus chlorhexidine to wash the peritoneal cavity at least two times within 10 min for 10-15 min each [1]. Allergens that lead to anaphylactic reactions can be removed in this manner. Perforated cystic cavities should be carefully evaluated. Remaining cystic contents should also be evacuated, and the free edges of the cystic cavity should be widely excised. Perforated cysts that are located in the liver should be examined via a leakage test, which can be performed with a saline solution administered through the common bile duct/cystic duct to observe the relationship between the cyst and the biliary tract. Bile duct orificies that lead to bile leakage should be repaired with different suture materials. Common bile duct and T-tube placement for high flow output bile fistulas can be performed simultaneously. Sometimes, ERCP can be selected instead of T-tube application to complete the surgery as soon as possible. The



**Figure 7** Intraoperative appearance of severe adhesions similar to sclerosing encapsulating peritonitis in the abdominal cavity secondary to hydatid cyst perforation.

biliary tract can be visualized via ERCP. If possible, spleen-preserving surgical interventions should be preferred [5]. Abdominal drains should be placed both into the cystic cavity and in the abdomen before surgery completion. Next, we would like to discuss the question “do we need any intervention for other unperforated cystic lesions identified during emergency surgery?”

Antihelminthic treatment should be administered as soon as possible for patients diagnosed with intraperitoneal rupture to prevent disease recurrence due to overlooked cystic contents during surgery. The most preferred antihelminthic agent is albendazole (10-15 mg/kg per day). According to the literature, the treatment period should last between 1 and 12 mo [1,4,6,8]. Patients diagnosed with intraperitoneal hydatid cyst rupture should be followed-up more frequently than patients without complications. Morbidity and mortality rates after intraperitoneal rupture are 10-35.3% and 0-23.5%, respectively [7,9]. Clinical follow-up should be performed monthly, especially during the early period after surgery and once a year in the long-term. US, echinococcus IgG enzyme-linked immunosorbent assay, indirect hemagglutination, and CT can be selected during follow-up. If there is no recurrence after five years, clinical follow-up can be terminated. The recurrence rate after intraperitoneal hydatid cyst rupture is reported between 0-28.6% [1,4,6,7,9]. There was only one case of recurrence in our series.

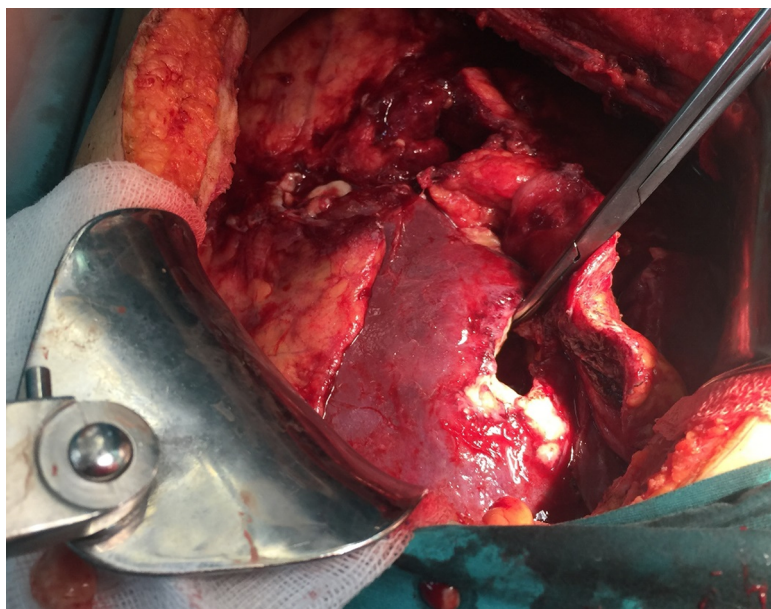
## CONCLUSION

Intraperitoneal hydatid cyst rupture is a life-threatening complication because it causes serious hemodynamic instability and allergic reactions. Therefore, hydatid cyst rupture should be considered a differential diagnosis in patients who have sudden onset abdominal pain and allergic reactions, like urticaria, especially in regions endemic for the disease. Deteriorated hemodynamic parameters due to anaphylactic reactions should be quickly corrected so that emergent surgery can be life-saving.

**Table 2** Short analysis of studies that published four or more cases of intraperitoneal hydatid cyst rupture

Authors	Years	Country	City	Study period	Perforated HC	Total HC surgery	Rate (%)
Toumi	2017	Tunisia	Monastir	1990-2015	12	1350	0,9
Sakcak	2015	Turkey	Ankara	1996-2013	16	756	2,1
Ozturk	2007	Turkey	Erzurum	1979-2004	20	653	3,1
Mouqait	2013	Morocco	Fes	2008-2012	14	306	4,6
Derici	2007	Turkey	Izmir	1988-2005	17	306	5,6
Unalp	2010	Turkey	Izmir	2000-2009	21	368	5,7
Kurt	2003	Turkey	Istanbul	1995-2001	7	99	7,1
Akcan	2010	Turkey	Kayseri	1990-2008	28	372	7,5
Bozdog	2016	Turkey	Diyarbakir	2005-2015	16	NA	NA
Ozturk	2016	Turkey	Izmir	2008-2012	13	NA	NA
Dirican	2010	Turkey	Malatya	2003-2008	10	NA	NA
Rami	2009	Morocco	Fes	NA	4	NA	NA
Gunay	1999	Turkey	Istanbul	1985-1997	16	NA	NA

NA: Not available; HC: hydatid cyst.

**Figure 8** Intraoperative image obtained after evacuating the cystic contents.

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