

# Intraperitoneal rupture of the hydatid cyst disease: Single-center experience and literature review

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## ABSTRACT

**BACKGROUND:** The primary aim of this study was to present our experience in the intraperitoneal rupture of the hydatid cyst in the guidance of literature data.

**METHODS:** Demographical, clinical, radiological and postoperative follow-up data of 29 patients who underwent surgical treatment in our institution with the diagnosis of intraperitoneal rupture of the hydatid cyst from January 2003 to July 2020 were analysed retrospectively in this study.

**RESULTS:** Among the 29 patients with an age range of from 16 to 79 years (median= 39, IQR=25.5), 16 were male (55.2%), and 13 were female (44.8%). Intraperitoneal rupture of the hydatid cyst was spontaneous in 21 (72.4%), traumatic in seven (24.13%) (two of them were iatrogenic) and was due to shotgun in one patient. Vast majority of the patients were admitted to the emergency department in the first 24 hours after the onset of sign and symptoms. WBC varied from 8.600 to 30.900/mm<sup>3</sup> (median=12.100, IQR=5.7). Ruptured cysts were localised in liver (n=25, 86.2%), in spleen (n=2, 6.89%) or in pelvis (n=2, 6.89%) and diameter varied from 40 to 200 mm (median=90, IQR=50). Among the cysts ruptured in liver, 19 (76%) of them were localised in the right lobe. Among the ruptured cysts of the liver, 20 (80%) of them underwent conservative surgery (e.g., partial pericystectomy), the remaining five patients (20%) underwent a radical surgical treatment (e.g., pericystectomy). A biliary orifice was diagnosed during surgical exploration in either cyst cavity or cut surface of the liver in 12 (48%) of the patients. Patients with a dilated common bile duct or a visible biliary duct orifice in cystic cavity underwent common bile duct exploration and T-tube drainage procedure (n=6, 24%). One patient died on postoperative day one, due to cardiopulmonary complications secondary to cyst rupture. All along the postoperative follow-up of median 1416 days, seven (24.1%) patients were diagnosed for diseased recurrence.

**CONCLUSION:** Intraperitoneal rupture of the hydatid cyst is a life-threatening complication of hydatid cyst disease, for which diagnosis without delay and timing of surgical treatment is essential. Anaphylactic shock require rapidly initiation of medical treatment against allergic reactions. Despite scolocidal agents, vesicular spread into peritoneal cavity accounts for the major risk factor for disease recurrence. Hence, abdominal cavity should be explored cautiously.

**Keywords:** Acute abdomen; anaphylactic reactions; Hydatid cyst; rupture; perforation; recurrence

## INTRODUCTION

Echinococcus granulosus belong to Taenidae family and cystic echinococcosis, also known as hydatid cyst disease is a zoonotic disease which affects mainly the liver, lung, and various other tissues and organs. Cystic echinococcosis accounts for approximately 95% of hydatid diseases.<sup>[1,2]</sup> Following initial in-

vasion of an organ or tissue, hydatid cysts remain silent for a long time. Most of the hydatid cysts are diagnosed incidentally during a radiological evaluation performed for another indication.<sup>[3]</sup> However, some of the patients may present with abdominal pain, nausea, vomiting, jaundice depending on the dimension and localisation of hydatid cysts.<sup>[3]</sup> Internal rupture-perforation (cysto-biliary fistula, rupture into the

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hollow viscus, broncho-biliary fistula, bronchopleural fistula, intrapericardial rupture, intrapleural rupture, intraperitoneal rupture), external rupture (cysto-cutaneous fistula), bacterial super-infection, anaphylactoid reaction, vascular compression, biliary compression and neighboring organ compression are among hydatid cystic disease related complications.<sup>[4-6]</sup> Intraperitoneal rupture of the hydatid cyst is a rare and life-threatening complication of hydatid disease for which rapid diagnosis and treatment is essential to reduce mortality.<sup>[4,7,8]</sup> This study aims to present demographical, clinical and long-term follow-up results of 29 intraperitoneal rupture of the hydatid cyst cases in guidance of current literature.

## MATERIALS AND METHODS

Between January 2003 and July 2020, demographic, clinical and follow-up data of 29 patients who were admitted to Inonu University Faculty of Medicine Emergency Unit (n=28) and General Surgery Outpatient Clinic (n=1) with signs and symptoms of intraperitoneal rupture of the hydatid cyst were analyzed retrospectively. Some of the patients with intraperitoneal rupture of the hydatid cyst included in this study were previously published.<sup>[1,3,9]</sup> In this study, we aimed to present all of our published and unpublished cases as single-center experience.

The following parameters were investigated for this study: age (year), sex (male, female), complaints/signs/symptoms (e.g., abdominal pain, nausea and anaphylactic reactions) possible cause of cyst rupture (spontaneous, traumatic), history of hydatid cyst, history of anthelmintic treatment (e.g., albendazole), interval between the onset of symptoms and the admission to the hospital, diagnostic tools (abdominal computed tomography [CT], ultrasonography [US]), last white blood count (WBC) before the operation, ruptured cyst location (e.g., liver, spleen and pelvis), ruptured cyst diameter (mm), pre or postoperative serologic tests (anti-echinococcus IgG ELISA and IHA), surgical incision (e.g., midline, right subcostal and J-shaped), surgical approaches (e.g., conservative and radical surgery), presence of cysto-biliary communication, postoperative complications, recurrence, follow-up (day) and outcomes (alive, dead).

Regardless of the type of surgical approach, all patients were treated using albendazole (Andazol; Biofarma, Istanbul, Turkey) at a dose of 10–15 mg/kg/day in the postoperative period. The type of surgical procedure, antiscloidal agent, and the duration of the postoperative anthelmintic treatment varied depending on the experience of the surgical team.

All patients except the patient who died in the postoperative day one were invited for periodic outpatient clinic control in the postoperative period. The surveillance protocol was as follows; patients were requested to come to outpatient clinic control every three months in the first year, every six months in the second year, and once a year afterwards. During the

controls, the abdominal US and serological tests were studied routinely. Abdominal CT was additionally performed to patients in whom there was a suspicion of recurrence.

## Statistical Analysis

Statistical analyses were performed using Statistical Package for Social Sciences (SPSS) v25.0 (IBM Corp., Armonk, NY, USA). The continuous variables were expressed as median and Interquartile range (IQR). The categorical variables were reported as number and percentage. This retrospective study was approved by the Inonu University Institutional Review Board for Non-interventional Studies (Approval No:2020/1010).

## RESULTS

Among the 29 patients with an age range of from 16 to 79 years (median=39, IQR=25.5), 16 were male (55.2%) and 13 were female (44.8%). Age of females varied from 22 to 79 years (median=50, IQR=30), and the age of males varied from 16 to 59 years (median=33.5, IQR=24.5). Intraperitoneal rupture of the hydatid cyst was spontaneous in 21 (72.4%), traumatic in seven (24.13%) (two of them were iatrogenic) and was due to penetrating abdominal trauma (shotgun injury) in one patient. Traumatic perforation cases were due to falling from height (n=3), assault (n=2) and iatrogenic injury (n=2). Time from incident to admission into the emergency department was determined in 24 cases, which varied from one hour to 30 days. However, 17 patients were admitted in the first 24 hours. The US and CT (n=20), US (n=4) or CT (n=3) was the diagnostic tool in 27 of the patients, the remaining two patients were directly admitted to the operating theater (Fig. 1a–c). Upon admission to the emergency room, WBC varied from 8.600 to 30.900/mm<sup>3</sup> (median=12.100, IQR=5.7). Major complaints and symptoms upon admission were abdominal pain, nausea/vomiting, fever, anaphylaxis, hypotension, urticaria and dyspnea in the decreasing order of frequency. Localization of perforated cysts were in liver (n=25, 86.2%), spleen (n=2, 6.89%) and pelvis (n=2, 6.89%). Localization of cysts perforated in liver were right lobe (n=19, 76%) or left lobe (n=6, 24%). Perforated cyst diameters varied from 40 to 200 mm (median=90, IQR=50). Preoperative or postoperative echinococcal ELISA or IHA results were achieved in 17 cases, among which 14 (82.3%) were seropositive.

## Surgical Management

Patients underwent surgical treatment due to emergent conditions, except one case who was admitted to the emergency department with non-specific symptoms and underwent elective surgical treatment. Midline (n=18), right subcostal (n=7), J-shaped (n=3) or laparoscopic trocar incisions (n=1) was used. Among the perforated cysts of the liver, 20 (80%) of them underwent conservative surgery (e.g., partial pericystectomy), the remaining five patients (20%) underwent a radical surgical treatment (e.g., pericystectomy). Following



**Figure 1.** (a) Coronal section of the contrast-enhanced CT scan shows that one of the hydatid cysts localized in the right lobe of the liver is ruptured into the peritoneal cavity. (b) Coronal section of the contrast-enhanced CT scan shows that hydatid cyst rupture causing free fluid accumulation in the pelvic cavity. (c) Coronal section of the contrast-enhanced CT scan shows hydatid cyst rupture causing free fluid accumulation in the right paracolic gutter.

partial or total pericystectomy, a biliary orifice was diagnosed during surgical exploration (with either leakage test or visual inspection) inside the cyst cavity or on the cut surface of the liver in 12 (48%) of the cases. All visible bile duct orifices were sutured. Common bile duct exploration and t-tube insertion was performed to six (24%) patients with a dilated common bile duct or a wide bile duct orifice inside the cystic cavity. Partial cystectomy was performed to the pelvic cyst, considering adhesions to surrounding tissues. An iatrogenic injury due to dissection was diagnosed in a patient during splenectomy. Following chest-tube insertion, diaphragm was sutured primarily. A patient admitted with abdominal pain and dyspnea was diagnosed with cystic perforation located in the right liver lobe with right pleural effusion. Surgical exploration of this patient revealed empyema as a result of cystic perforation into the right hemithorax. Following drainage, chest-tube was inserted, and diaphragm was sutured primarily. All the patients included were treated with one of the NaCl (3%), Cetrimide-Chlorhexidine (1/30), Povidone-iodine (10%) or NaCl (0.9%) solution for cystic cavity and intraabdominal lavage.

### Follow-Up

Liver abscess (n=2), haemorrhage (n=1) and cardiopulmonary arrest (n=1) were events on postoperative course. Patients with liver abscess underwent percutaneous US-guided drainage, patient with haemorrhage underwent explorative laparotomy. Albendazole treatment with a dose of 10–15 mg/kg/day was administered to 28 patients postoperatively. Duration of anti-helminthic treatment varied from two to six months according to clinical status, disease recurrence and personal experience of the surgeon. During the postoperative follow-up of median 1416 days (min-max=1–3456, IQR=1625), seven (24.1%) patients were diagnosed for dis-

eased recurrence. One of the patients died on postoperative first day due to cardiopulmonary complications of cyst perforation. Another patient died due to cardiopulmonary disease on postoperative day 60. Demographic and clinical characteristics of 29 patients with intraoperative hydatid cyst rupture are summarized in Table 1.

### DISCUSSION

Depending on the size, location, stage, relationship of the cyst with neighboring organs, 0.6–40% of patients with abdominal hydatid cyst disease may develop various complications, such as cystobiliary communication, superinfection, bronchobiliary fistula, intrapericardial rupture, intrapleural rupture, intraperitoneal rupture, Budd-Chiari syndrome, portal hypertension, gastric outlet obstruction, allergic reactions and rupture into the hollow viscus organs.<sup>[1,3,4,7,10,11]</sup>

Current studies on complications of the hydatid cysts suggest that intraperitoneal rupture (1–16%) is the third most common complication of the disease following intrabiliary rupture (overall: 2–50%, occult: 10–37%, frank: 3–17%) and allergic reactions (1–25%).<sup>[1,3,7,12–15]</sup> We have summarized the results of the literature search on PubMed and Medline databases using the key words hydatid cyst, echinococcosis, rupture ve perforation in different combinations in Table 2. The current literature suggests that intraperitoneal rupture develops in 0.4–18.1% of the cases. In the second period of the present study (2009–2020), 11.2% of the patients who underwent abdominal hydatid cyst surgery were operated due to hydatid cyst rupture, which is consistent with the current literature.

Intraoperative rupture of the hydatid cyst may develop as a result of trauma or spontaneously due to increased intracystic pressure. Occasionally, intraperitoneal rupture of the hydatid

**Table I.** Summary of the demographic and clinical characteristics of 29 patients with intraperitoneal hydatid cyst rupture

Age	Sex	Cause of cyst perforation	Preoperative duration (hour)	Diagnostic tools	Preop WBC	Complaint/Sign/Symptoms	Perforated cyst location	Cyst diameter (mm)	Serology
32	F	Spontaneous	3	US+CT	18.000	Abdominal pain + Urticaria	Seg VI-VII	130	IHA (+)
24	M	Trauma (Fall)	96	US+CT	12.000	Abdominal pain Nausea/Vomiting	Seg. IV-VI	110	NA
20	M	Spontaneous	10	US+CT	11.800	Abdominal pain + Nausea	Seg VI-VII	140	NA
22	F	Spontaneous	24	US+CT	12.400	Abdominal pain + Nausea + Blindness	Seg VI	60	NA
30	M	Spontaneous	24	Laparotomy	11.400	Abdominal pain	Seg VI	90	NA
29	M	Trauma (Fall)	4	US+ CT	10.700	Abdominal pain+ Nausea/ Vomiting+ Urticaria	Pelvis	80	NA
79	F	Spontaneous	12	US	9.200	Abdominal pain	Seg VI	80	NA
47	M	Spontaneous	90	US+CT	19.900	Abdominal pain	Seg VI-VII	100	NA
37	M	Shotgun	1	Laparotomy	13.000	Abdominal pain	Pelvis	100	NA
22	M	Spontaneous	30	US	11.200	Abdominal pain + Nausea + Urticaria	Spleen	90	NA
52	F	Trauma (Fall)	24	US+CT	11.800	Abdominal pain	Seg VI	55	ELISA (+)
29	F	Trauma (percutaneous)	NA	US+CT	11.900	Abdominal pain	Seg VI-VII	85	ELISA (+)
44	M	Spontaneous	24	US+CT	8.600	Abdominal pain + Hypotension + Anaphylaxis	Seg VI	70	ELISA (+)
50	F	Spontaneous	48	US+CT	14.900	Abdominal pain + Fever + Urticaria	Seg VIII	50	ELISA (+)
59	M	Spontaneous	24	CT	12.500	Abdominal pain	Seg VI-VII	150	ELISA (+)
16	M	Spontaneous	1	US	13.100	Abdominal pain + Cyanosis + Urticaria	Seg V-VI	79	ELISA (+)
39	F	Spontaneous	NA	US+CT	12.100	Abdominal pain	Seg III	100	ELISA (-)
37	M	Trauma	24	US	11.600	Abdominal pain + Hypotension + Anaphylaxis	Spleen	50	ELISA (+)
70	F	Spontaneous	3	US+CT	11.900	Abdominal pain	Seg II- III	140	NA
58	M	Spontaneous	24	US+CT	16.700	Abdominal pain + Dyspnea	Seg VI-VII	200	ELISA (+)
50	F	Spontaneous	72	US+CT	10.300	Abdominal pain + Fever	Seg II- III	70	ELISA (+)
47	M	Trauma (percutaneous)	15	US+CT	30.900	Abdominal pain + Fever	Seg II- III	110	ELISA (+)
20	M	Spontaneous	120	US+CT	18.800	Abdominal pain + Fever	Seg V-VIII	98	NA
54	F	Spontaneous	168	US+CT	9.800	Abdominal pain + Hypotension + Anaphylaxis	Seg VI-VII	75	ELISA (+)
41	M	Spontaneous	NA	US+CT	11.000	Abdominal pain	Seg VII-VIII	160	ELISA (+)
27	F	Spontaneous	24	US+CT	20.100	Abdominal pain + Hypotension	Seg III	40	ELISA (-)
67	F	Spontaneous	NA	CT	17.300	Abdominal pain	Seg VII-VIII	200	NA
19	M	Trauma	2	US+CT	14.300	Abdominal pain + Nausea/Vomiting	Seg VI-VII	75	ELISA (+)
40	F	Spontaneous	30 (day)	CT	18.500	Abdominal pain + Anaphylaxis	Seg II-VI	50	ELISA (-)

**Table 1.** Summary of the demographic and clinical characteristics of 29 patients with intraperitoneal hydatid cyst rupture (continue)

Surgical approaches	Intracavitary lavage	Cysto-biliary communication	Postop complication	Recurrence	Follow up (day)	Outcomes
Partial pericystectomy + Drainage+ Omentopexy + T-tube	NaCl (3%) + NaCL (0.9%)	Yes (Sutured)	NA	Yes (CT)	2100	Alive
Partial pericystectomy + Drainage+ Falciformopexy	NaCl (3%) + NaCL (0.9%)	No	No	No	1200	Alive
Partial pericystectomy	Cetrimide–chlorhexidine (1/30)	NA	NA	NA	840	Alive
Partial pericystectomy + T-tube	Cetrimide–chlorhexidine (1/30)	NA	NA	NA	110	Alive
Partial pericystectomy	NaCl (3%) + NaCL (0.9%)	NA	NA	NA	780	Alive
Partial pericystectomy	Cetrimide–chlorhexidine (1/30)	NA	NA	NA	900	Alive
Total pericystectomy + T-Tube	NaCl (3%) + NaCL (0.9%)	Yes (Sutured)	Cardiac arrest	NA	60	Exitus
Partial pericystectomy + Drainage	NaCl (3%) + NaCL (0.9%)	NA	NA	NA	90	Alive
Partial pericystectomy + Drainage	NaCl (3%) + NaCL (0.9%)	NA	NA	NA	1680	Alive
Partial pericystectomy	Saline 1/10	NA	NA	NA	1170	Alive
Partial pericystectomy + Omentopexy	NaCl (3%) + NaCL (0.9%)	Yes (Sutured)	No	No	1273	Alive
Pericystectomy+ Drainage + Cholecystectomy	NaCl (3%) + NaCL (0.9%)	Yes (Sutured)	No	NA	457	Alive
Partial Cystectomy + Omentopexy + T-Tube	NaCl (3%)	No	Liver abscess	Yes (US+CT)	3044	Alive
Partial pericystectomy + Drainage + Cholecystectomy + T-tube	Cetrimide–chlorhexidine (1/30)	Yes (Sutured)	No	No	3357	Alive
Partial pericystectomy + Drainage + Cholecystectomy+Omentopexy	Cetrimide–chlorhexidine (1/30)	Yes (Sutured)	No	No	3456	Alive
Partial pericystectomy + Drainage	Povidone-iodine (10%)	No	No	Yes (US+CT)	2603	Alive
Partial pericystectomy + Drainage	NaCl (3%)	No	No	no	2267	Alive
Splenectomy+ Retroperitoneal + Retrovesical Cystectomy	NaCl (3%)	No	Bleeding	No	1694	Alive
Partial pericystectomy+ Drainage	NaCl (3%)	Yes (Sutured)	No	Yes (US+CT)	1590	Alive
Partial pericystectomy+ Drainage (right pleural empyema-chest tube)	NaCl (3%)	No	Cardiac arrest	No	1	Exitus
Partial pericystectomy + Drainage+ Omentopexy	NaCl (3%)	No	No	No	982	Alive
Partial pericystectomy + Drainage + Omentopexy	NaCl (3%)	No	No	Yes (US+CT)	1451	Alive
Pericystectomy + Drainage + Cholecystectomy	Povidone-iodine (10%)	Yes (Sutured)	No	No	573	Alive
Partial pericystectomy + Drainage	Cetrimide–chlorhexidine (1/30)	No	No	Yes (US+CT)	1639	Alive
Partial pericystectomy + Drainage	NaCl (3%)	Yes (Sutured)	No	NA	1385	Alive
Partial pericystectomy + Drainage + Cholecystectomy + Omentopexy	NaCl (3%) + NaCL (0.9%)	Yes (Sutured)	No	No	1416	Alive
Pericystectomy + Drainage + Cholecystectomy	NaCl (3%) + NaCL (0.9%)	Yes (Sutured)	No	NA	3116	Alive
Pericystectomy + Drainage + Cholecystectomy + T-tube	Cetrimide–chlorhexidine (1/30)	Yes (Sutured)	No	Yes (US+CT)	3125	Alive
Partial Cystectomy + Omentopexy	NaCl (3%)	No	Liver abscess	No	1701	Alive

cyst may develop iatrogenically during elective abdominal surgery or after percutaneous radiological procedures, such as in the two patients, presented in this study. There are many studies showing that the rupture of the hydatid cysts may

develop due to trauma and contradicting studies that show it is more frequently a spontaneous perforation.<sup>[1,16-18]</sup> The factors that increase the risk of cystic rupture include early age, the diameter of the cyst (>10 cm) and superficial lo-

**Table 2.** Brief literature review on intraperitoneal ruptured hydatid cyst

First author's surname	References (Journal Information)	Country	Study period	Total case	Cyst perforation	Rate (%)
Tatli	Dicle Medical J 2017; 44:251-6	Turkey	2012-2016	218	12	5.5
Toumi	Eur J Trauma Emerg Surg. 2017;43:387-91	Tunisia	1990-2015	1350	12	0.9
Aghajanzadeh	Clin Surg. 2017; 2: 1820	Iran	2004-2015	352	4	1.1
Kloppers	S Afr J Surg. 2017;55:72	S. Africa	2012-2017	22	4	18.1
Bozdag	Dig Liver Dis. 2016;48:98-100	Turkey	2005-2015	NA	16	NA
Sakcak	J J Gastro Hepato. 2016. 3: 018	Turkey	1996-2013	756	16	2.1
Ozturk	J Surg Arts (Cer San D) 2016; 2: 61-6	Turkey	2008-2012	NA	13	NA
Symeonidis	Scand J Surg 2013;102:171-7	Greece	1980-2010	227	6	2.6
Mouaqit	World J Emerg Surg. 2013;8:28	Morocco	2008-2012	306	14	4.6
Rami	Pan Afr Med J 2011;8:27	Morocco	2004-2008	NA	5	NA
Malik	World J Gastrointest Surg 2010;2:78-84	India	2004-2005	69	2	2.9
Akcan	World J Gastroenterol 2010;16:3040-8	Turkey	1990-2008	372	28	7.5
Unalp	Saudi Med J 2010;31:37-42	Turkey	2000-2009	368	21	5.7
Agayev	Hepatogastroenterology 2008;55:1373-9	Azerbaijan	NA	484	6	1.2
Tekin	J Gastrointest Liver Dis. 2008;17:33-7	Turkey	1985-2005	700	14	2.0
Akcan	World J Surg 2007;31:1284-91	Turkey	1990-2005	347	27	7.8
Ozturk	Am J Surg 2007;194:313-6	Turkey	1979-2004	653	20	3.1
Derici	World J Surg 2006;30:1879-83	Turkey	1988-2005	306	17	5.6
Beyrouiti	Presse Med 2004;33:378-84	Tunisia	1990-2000	970	17	1.8
Puia	Chirurgia (Bucur) 2004;99:541-4	Romania	1993-2002	160	6	3.8
Kurt	J Gastrointest Surg. 2003;7:635-41	Turkey	1995-2001	99	7	7.1
Larbi	Ann Chir 2002;127:487-8	Tunisia	1993-1999	302	15	5.0
Sozuer	Am J Trop Med Hyg. 2002;66:575-7	Turkey	NA	242	21	8.7
Agayev	Khirurgiia (Mosk) 2001;32-6	Azerbaijan	NA	280	2	0.7
Gunay	J Trauma 1999; 46: 164-7	Turkey	1985-1997	NA	16	NA
Karydakis	J Chir (Paris). 1994;131:363-70	Greece	1972-1992	421	4	1.0
Chen	Zhonghua Wai Ke Za Zhi. 1994;32:166-8	China	1954-1990	907	50	5.5
Bilge	HPB Surg. 1992;6:57-64	Turkey	1978-1990	226	1	0.4
Erguney	Ann Chir. 1991;45:584-9	Turkey	1979-1989	328	7	2.1
Pulatov	Klin Khir. 1990;6:12-4	Russia	NA	NA	10	NA
Placer	Br J Surg. 1988;75:157	Spain	1965-1985	471	15	3.2
Androulakis	Eur Surg Res 1986;18:145-50	Greece	1964-1984	1310	7	0.5
Dedenko	Vestn Khir Im I I Grek. 1976;116:42-4	Russia	NA	231	35	15.2

calization.<sup>[1,7,18,19]</sup> Hydatid cysts are more frequently observed in younger people who are also more frequently involved in traumatic events. It is a common fact that intracystic pressure increases with increasing diameter. Once the intracystic pressure increases beyond 50 cm-H<sub>2</sub>O, the risk of spontaneous rupture or rupture following minor trauma increases.<sup>[1,9,16,17]</sup> In superficial cysts, the free surface of the cysts is a risk factor for intraperitoneal rupture and fistulization to adjacent hollow organs and cavities.<sup>[1,3,5]</sup> The present surgery was performed in our department of surgery, and we did not include pre-adolescent patients. Thus, we can not comment on this

age group; however, the more than half of the patients in our study were  $\leq 40$  years which seems to support the age range of the complication stated in the literature.<sup>[3]</sup>

The signs and symptoms of intraperitoneal rupture of the hydatid cyst depend on the underlying cause, the interval between onset of symptoms and admission to the emergency department and the medical history of the patient. The common symptoms of the intraperitoneal rupture of hydatid cysts are mild to severe abdominal pain, nausea, vomiting, urticaria and hemodynamic instability (hypotension and tachy-

cardia) and dyspnea.<sup>[1,3,5]</sup> The cystic fluid is highly toxic and allergic. Thus, cystic fluid leakage to the peritoneal cavity, biliary system or systemic circulation can cause mild allergic reactions to severe anaphylactic shock depending on the amount of cyst fluid.<sup>[1,3,4]</sup> The review of the literature suggests that 16.7% to 25% of the cases develop minor allergic reactions, whereas 1–12.5% of the cases have life-threatening allergic reactions that require preoperative or intraoperative intervention, such as epinephrine, steroid and ventilatory support.<sup>[1,3,16,19]</sup> It should be kept in mind that 0.2–3.3% of the patients develop anaphylactic reactions during hydatid cyst surgery without obvious perforation.<sup>[20,21]</sup> Thus, allergic reactions can be encountered in any elective or emergency interventions for hydatid cysts and necessary precautions should be obtained. The authors of the present study have a consensus that any patients without prior antihelminthic therapy that will undergo hydatid cyst surgery should receive antihistaminic and 1 mg/kg corticosteroid during the anesthesia induction phase.

In the diagnosis of intraoperative rupture of the hydatid cysts anamnesis, physical examination and radiological evaluation of the patients should be made altogether. There is leukocytosis in most of the patients as presented in our study, but this is mainly due to peritoneal irritation rather than cyst rupture. There may be eosinophilia in certain patients. US is the first option for imaging due to the easy access and non-invasive nature of the technique. On the other hand, if the patients are hemodynamically stable, the best imaging modality that should be applied is the abdominal CT. CT can show the disruption of the cyst wall, reduction in the size of the cyst and intraabdominal rupture of the cyst as well as the exact localization of the cyst in the liver. The sensitivities of the US and CT are reported to be 85% and 100%, respectively.<sup>[5,7,9,19]</sup>

The patients with confirmed intraoperative hydatid cyst rupture should be treated with an evaluation of the signs and symptoms. The hemodynamic stability of the patients should be achieved and the operation should be performed as soon as possible to reduce the morbidity and mortality of the patients. The primary goal of medical therapy, which includes antihistaminics, fluid resuscitation, corticosteroids and oxygen support, is to provide hemodynamic stability and prevent anaphylactic reactions in the patients. Depending on the experience of the surgical team, laparoscopic or open surgical approaches may be preferred in patients who are hemodynamically stable. On the other hand, the open surgical approach is more suitable for patients who are hemodynamically unstable. During emergency operations, complete evaluation of the abdominal cavity should be performed through a midline incision. However, the type of incision (e.g., right subcostal or J-shaped incision) can change according to the preference of the surgical team.

The content of the cyst should be removed from the abdominal cavity as soon as possible and the peritoneal surface

should be irrigated with saline to minimize the contact with content, which will reduce the severity of anaphylactic reactions. After this step the abdominal cavity and cavity of the cyst can be irrigated with certain scolicidal agents, such as hypertonic saline (3–10–15–30%), silver nitrate (0.5%), cetrimide (0.5%), chlorhexidine (0.05%), cetrimide + chlorhexidine (1.5%+0.15%=10% Savlon), hydrogen peroxide (1.5–3%), povidone-iodine (10% betadine), ethyl alcohol (70–95%) and formalin (2%) according to the preference and experience of the surgical team.<sup>[3,6,7,18,19]</sup> Each scolicidal solution has a different duration of action.

We usually prefer hypertonic saline and cetrimide+chlorhexidine and we irrigate the abdominal cavity twice with 10 minutes interval and we wait for 10 to 15 minutes as we apply the fluid to the abdominal and cystic cavities.<sup>[1,3,9]</sup> We believe that we remove all the factors that cause allergic reactions. The cystic cavity should be evaluated as all these procedures are performed. This will enable the surgeon to see the cystic cavity thoroughly and any bile leakage can be determined.

The ruptured cystic cavity can be evaluated directly for any cystobiliary communication, and in suspected cases, transcystic or transcholedochal saline can be injected to perform bile leakage test.<sup>[22,23]</sup> Bile duct orifices in cyst cavities should be sutured. If the bile orifices are wide or there is a high output biliary fistula, than common bile duct exploration and T-tube drainage should be performed. However, since the intracystic pressure is higher than the intrabiliary pressure, the exploration of the cavity may not yield any bile leakage and biliary fistula may develop in the early postoperative period. Drainage of the cystic cavity enables early diagnosis and control of the biliary fistula. In patients with postoperative bile leakage, early ERCP (sphincterotomy±stenting) may be an effective diagnostic and therapeutic option. In the present study, 12 (48%) patients had cystobiliary communication; however, only four underwent common bile duct exploration+ T-tube drainage. Besides, two patients underwent common bile duct exploration and T-tube drainage due to the presence of cystic content in the biliary ducts and presence of dilatation in the bile ducts. Only one patient underwent postoperative ERCP due to biliary fistula, and in this patient biliary fistula was determined in the first operation. In splenic cyst ruptures, spleen preserving procedures should be selected to preserve the immunologic function of the spleen.<sup>[1,6,24]</sup>

Recurrence due to missed daughter vesicles is prevented by starting the antihelminthic therapy as soon as possible after the operation. There is no generally accepted protocol for the dose and duration of antihelminthic therapy (continuous or cyclic). Although there are controversial data in the literature, it is suggested that antihelminthic therapy should be continued for one to 12 months after the operation.<sup>[1,3,5,16,25]</sup> In the present study, we started antihelminthic therapy for 2–6 months after the operation to all the patients (except the patients who died on the postoperative first day).

Current literature suggests that morbidity and mortality due to operation performed for intraperitoneal rupture of the hydatid cysts are 10–63% and 0–23.5%, respectively.<sup>[1,3,7,18,19]</sup> The postoperative recurrence rates vary between 0 to 28.6%.<sup>[1,3,7,18,19]</sup> US, echinococcus IgG ELISA or IHA can be performed for the postoperative surveillance of these patients. In patients who are suspected of having a recurrence should be evaluated further by CT. The patients are evaluated every three months for the first year and every six months for the second year and annually thereafter.

In the present study, 24.1% of the patients developed recurrence. We do not have the data of all the cases with hydatid cysts and therefore we cannot perform an evidence-based evaluation of the results. However, the high recurrence rate in the present study can be due to variation in the surgical team, non-satisfactory exploration of the cystic cavity, inability to remove the daughter cyst from the abdominal cavity, absence of definitive scolical agent for irrigation and absence of definitive albendazole treatment scheme.

In conclusion, intraperitoneal, intrapericardial or intrapleural rupture is a rare but fatal complication of the hydatid cysts. In endemic areas, the presence of abdominal pain, urticaria and anaphylactic reactions should raise the suspicion of rupture of hydatid cyst. The timing of medical and surgical therapy is a significant factor that affects the morbidity and mortality of the patients. A thorough evaluation of the abdominal cavity is vital for the reduction of the recurrence in patients who are urgently operated for ruptured hydatid cysts.

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## REFERENCES

- Akbulut S, Ozdemir F. Intraperitoneal rupture of the hydatid cyst: Four case reports and literature review. *World J Hepatol* 2019;11:318–29.
- Akbulut S, Demircan F, Sogutcu N. Hydatid cyst disease of the thyroid gland: report of two cases. *Int Surg* 2015;100:643–7.
- Yilmaz M, Akbulut S, Kahraman A, Yilmaz S. Liver hydatid cyst rupture into the peritoneal cavity after abdominal trauma: case report and literature review. *Int Surg* 2012;97:239–44.
- Akbulut S. Parietal complication of the hydatid disease: Comprehensive literature review. *Medicine (Baltimore)* 2018;97:e10671.
- Toumi O, Noomen F, Salem R, Rabeh H, Jabra SB, Korbi I, et al. Intraperitoneal rupture of hydatid cysts. *Eur J Trauma Emerg Surg* 2017;43:387–91.
- Belli S, Akbulut S, Erbay G, Koçer NE. Spontaneous giant splenic hydatid cyst rupture causing fatal anaphylactic shock: a case report and brief literature review. *Turk J Gastroenterol* 2014;25:88–91.
- Mouaqit O, Hibatallah A, Oussaden A, Maazaz K, Taleb KA. Acute intraperitoneal rupture of hydatid cysts: a surgical experience with 14 cases. *World J Emerg Surg* 2013;8:28.
- Velitchkov NG, Losanoff JE, Kjossev KT, Mironov MB. Life-threatening traumatic rupture of a liver hydatid cyst. *Eur J Emerg Med* 2001;8:225–8.
- Dirican A, Yilmaz M, Unal B, Tatli F, Piskin T, Kayaalp C. Ruptured Hydatid Cysts into the Peritoneum: A Case Series. *Eur J Trauma Emerg Surg* 2010;36:375–9.
- Mehta V, Singh A, Sood A. An Unusual Cause of Gastric Outlet Obstruction. *Gastroenterology* 2017;152:e1–4.
- Kabiri EH, El Maslout A, Benosman A. Thoracic rupture of hepatic hydatidosis (123 cases). *Ann Thorac Surg* 2001;72:1883–6.
- Atli M, Kama NA, Yuksek YN, Doganay M, Gozalan U, Kologlu M, et al. Intrahepatic rupture of a hepatic hydatid cyst: associated clinical factors and proper management. *Arch Surg* 2001;136:1249–55.
- Yildirgan MI, Başoğlu M, Aramanalp SS, Aydinli B, Balik AA, Celebi F, et al. Intrahepatic rupture in liver hydatid cysts: results of 20 years' experience. *Acta Chir Belg* 2003;103:621–5.
- Ramía JM, Figueras J, De la Plaza R, García-Parreño J. Cysto-biliary communication in liver hydatidosis. *Langenbecks Arch Surg* 2012;397:881–7.
- Akkapulu N, Aytac HO, Arer IM, Kus M, Yabanoglu H. Incidence and risk factors of biliary fistulation from a hepatic hydatid cyst in clinically asymptomatic patients. *Trop Doct* 2018;48:20–4.
- Unalp HR, Yilmaz Y, Durak E, Kamer E, Tarcan E. Rupture of liver hydatid cysts into the peritoneal cavity. A challenge in endemic regions. *Saudi Med J* 2010;31:37–42.
- Erel S, Kilicoglu B, Kismet K, Gollu A, Akkus MA. Peritoneal hydatid cyst perforation: a rare cause of emergency abdominal surgeries. *Adv Ther* 2008;25:943–50.
- Tinsley B, Abbara A, Kadaba R, Sheth H, Sandhu G. Spontaneous intraperitoneal rupture of a hepatic hydatid cyst with subsequent anaphylaxis: a case report. *Case Reports Hepatol* 2013;2013:320418.
- Akcan A, Akyildiz H, Artis T, Ozturk A, Deneme MA, Ok E, et al. Peritoneal perforation of liver hydatid cysts: clinical presentation, predisposing factors, and surgical outcome. *World J Surg* 2007;31:1284–91.
- Davarci I, Tuzcu K, Karcioglu M, Yetim I, Aydogan A, Turhanoglu S. Anaesthetic Management of Anaphylactic Shock Caused by Nonruptured Hydatid Cyst of the Liver. *West Indian Med J* 2014;63:545–7.
- Khanna P, Garg R, Pawar D. Intraoperative anaphylaxis caused by a hepatic hydatid cyst. *Singapore Med J* 2011;52:e18–9.
- Kayaalp C. Intraoperative diagnosis of cystobiliary communications of hydatid liver cysts. *Clinics (Sao Paulo)* 2012;67:515.
- Kayaalp C, Aydin C, Olmez A, Isik S, Yilmaz S. Leakage tests reduce the frequency of biliary fistulas following hydatid liver cyst surgery. *Clinics (Sao Paulo)* 2011;66:421–4.
- Ran B, Shao Y, Yimiti Y, Aji T, Shayiding P, Jiang T, et al. Spleen-preserving surgery is effective for the treatment of spleen cystic echinococcosis. *Int J Infect Dis* 2014;29:181–3.
- Sozuer E, Akyuz M, Akbulut S. Open surgery for hepatic hydatid disease. *Int Surg* 2014;99:764–9.

ORJİNAL ÇALIŞMA - ÖZET

## Hidatik kist hastalığının periton içine rüptürü: Tek merkez deneyimi ve literatür analizi

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**AMAÇ:** Bu çalışmanın amacı, intraperitoneal hidatik kist rüptürü gelişen hastalarla ilgili merkezimizin deneyimlerini literatür verileri ışığında sunmaktır. **GEREÇ VE YÖNTEM:** Ocak 2003 ile Temmuz 2020 tarihleri arasında kliniğimiz tarafından intraperitoneal hidatik kist rüptürü sebebiyle ameliyat edilen 29 hastanın demografik, klinik, radyolojik ve ameliyat sonrası takip verileri geriye dönük olarak incelendi.

**BULGULAR:** Yaşları 16 ile 79 yıl (median=39, IQR=25.5) arasında değişen 29 hastanın 16'sı (%55.2) erkek ve 13'ü kadın idi. Hastaların 21'inde (%72.4) spontan, yedisinde travmatik (iyatrojenik=2) ve birinde ateşli silah yaralanmasına bağlı hidatik kist rüptürü gelişti. Hastaların çoğu semptom ve bulguların başlamasından sonraki ilk 24 saat içinde acil servise başvurmuştu. WBC sayıları 8.600 ile 30.900/mm<sup>3</sup> (median=12.100, IQR=5.7) arasında değişmekteydi. Rüptüre kistlerin 25'i (%86.2) karaciğer, ikisi dalak ve geriye kalan ikisi pelvisteydi. Karaciğerde rüptüre olan kistlerin 19'u (%76) sağ lobtaydı. Rüptüre kistlerin çapları 40 mm ile 200 mm (median=90, IQR=50) arasında değişmekteydi. Karaciğer yerleşimli rüptüre kistlerin 20'sine (%80) konservatif cerrahi (parsiyel perisistektomi) işlem uygulanırken geriye kalan beş (%20) hastaya radikal cerrahi (perisistektomi) işlem uygulandı. Cerrahi işlem sırasında karaciğer yerleşimli rüptüre kistlerin 12'sinde (%48) kist kavitesinde veya karaciğer kesi yüzeyinde safra orifisi tespit edildi. Koledok çapı geniş olan veya kist kavitesinde geniş safra orifisi olan altı (%24) hastaya koledok eksplorasyonu ve t-tube drenaj işlemi uygulandı. Bir hasta kist rüptürüne ikincil kardiyopulmoner komplikasyonlar sebebiyle ameliyat sonrası birinci günde kaybedildi. Ameliyat sonrası median 1416 günlük takipte yedi (%24.1) hastada nöks gelişti.

**TARTIŞMA:** Intraperitoneal rüptür hidatik kist hastalığının hayatı tehdit eden bir komplikasyonudur. Hızlı tanı ve cerrahi tedavinin zamanlaması hayattır. Anafaktik komplikasyon gelişen hastalarda allerjik reaksiyonlara yönelik tıbbi tedaviye olabildiğince hızlıca başlanmalıdır. Her ne kadar skolisidal ajanlar kullanılsa bile rüptür sonucunda periton boşluğuna dökülen kız veziküller daha sonra nöks gelişmesi için en önemli risk faktörüdür. Bu yüzden tüm karın boşluğu itina ile kontrol edilmelidir.

**Anahtar sözcükler:** Akut karın; anafaktik reaksiyonlar; hidatik kist; nöks; perforasyon; rüptür.

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