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## Research Article

### Long-term Outcomes of Ruptured Intraperitoneal Hydatid Cysts

Sakcak I<sup>1,2\*</sup>, Erdoğan A<sup>1</sup>, Aydoğan I<sup>1</sup>, Ismail E<sup>1,3</sup>, Doganay M<sup>1</sup>, Akat AZ<sup>1</sup>

<sup>1</sup>Department of General Surgery and Hepatobiliary Surgery Clinic, Ankara Numune Education and Research Hospital, Ankara, Turkey

<sup>2</sup>Department of General Surgery Clinic, Medicalpark Hospital, Ankara, Turkey

<sup>3</sup>Department of General Surgery Clinic, Memorial Hospital, Ankara, Turkey

\*Corresponding author: Dr. İbrahim Sakçak, MD, Assoc Prof., Ankara Numune Education and Research Hospital, Talatpasa Avenue, Sıhhiye, Ankara, Turkey, Tel: +90 507 7608030; Fax: +90 312 2946886; Email: [ibrahimsakcak@yahoo.com](mailto:ibrahimsakcak@yahoo.com), [ibrahimsakcak66@gmail.com](mailto:ibrahimsakcak66@gmail.com)

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## Abstract

**Aims:** Hydatid cysts may cause major complications. In this study, we investigated intraperitoneally ruptured hydatid cysts, and analyzed surgical treatment, cyst diameter and type, postoperative complications, and mortality.

**Materials and Methods:** Sixteen patients older than 18 years of age that underwent surgical treatment of ruptured intraperitoneal hydatid cysts were analyzed. Age, gender, diagnostic procedures, cyst location, type of cyst, surgical procedures, morbidity, mortality, and cyst recurrence were retrospectively evaluated.

**Results:** Nine (66.2%) of the patients were males and 7 (43.8%) of them were females. Eleven (68.7%) patients had history of abdominal trauma. Thirteen (81.3%) patients had nonspecific abdominal symptoms before cyst rupture, and three (18.8%) patients were asymptomatic. There were 10 (67.5%) patients with type 3 cysts, four with type 4 cysts, one with a type 1 cyst, and one with a type 2 cyst. Cyst diameter was >10 cm in nine (56.3%) patients, and <5 cm in only one patient. The patients underwent partial cystectomy and external drainage.

**Conclusions:** Ruptured intraperitoneal hydatid cysts may recur, especially after conservative surgery. Cyst rupture is most likely in patients with a cyst diameter >10 cm, and/or in case of type 3 cysts.

**Keywords:** Peritonitis; Hydatid Cyst; Rupture; Recurrence; Trauma

## Introduction

Human hydatid disease is generally caused by larval or metacystode forms of the tapeworm *Echinococcus granulosus* and sometimes *Echinococcus multiloculari* [1]. Predators such as dogs, foxes, and coyotes are the main hosts, and harbor the adult stage of the parasite while ruminants such as sheep, cattle, and camels are the intermediate hosts [2,3]. Humans are sometimes unintended hosts. Hydatid cysts develop in liver in 50–75%, and in lungs in 20–30% of infected humans. Nails and hair may also be the sites of infection [1,4].

Hydatid cysts are usually asymptomatic and benign, however they may cause fatal complications. An emergency operation may be necessary if the cyst ruptures [5]. Lewall and McCorkell reported that ruptured cysts could be classified as contained, communicated, and direct cysts [6]. In the contained type, the cystic contents are limited to the pericystic cavity. In the communicated type, the contents spread to the biliary system or other major organs. Finally, in the direct type, the contents spread to the pleural or peritoneal cavity.

Primary intraperitoneal hydatid cysts are rare, occurring in only 2% of cases. Generally, intraperitoneal hydatid cysts develop secondary to a cyst in another location [7,8]. Trauma or spontaneous rupture of the cyst may cause complications [9]. Although spontaneous rupture of intraperitoneal hydatid cysts occurs in only 1–8% of the patients, it may lead to anaphylactic reactions, and is potentially fatal. The main risk factors for cyst rupture include younger age, a cyst diameter >10 cm, and superficial cyst location.

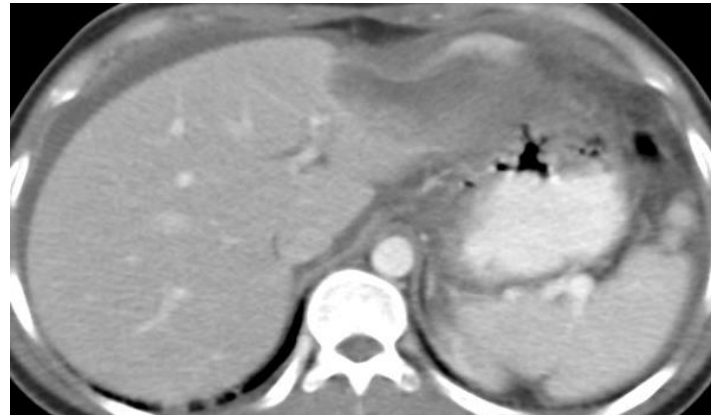
In this study, we performed a retrospective analysis of 16 patients with hydatid disease who experienced rupture into the peritoneum. We characterized the patients in terms of their surgical treatment, cyst diameter and type, postoperative complications, and mortality.

## Methodology

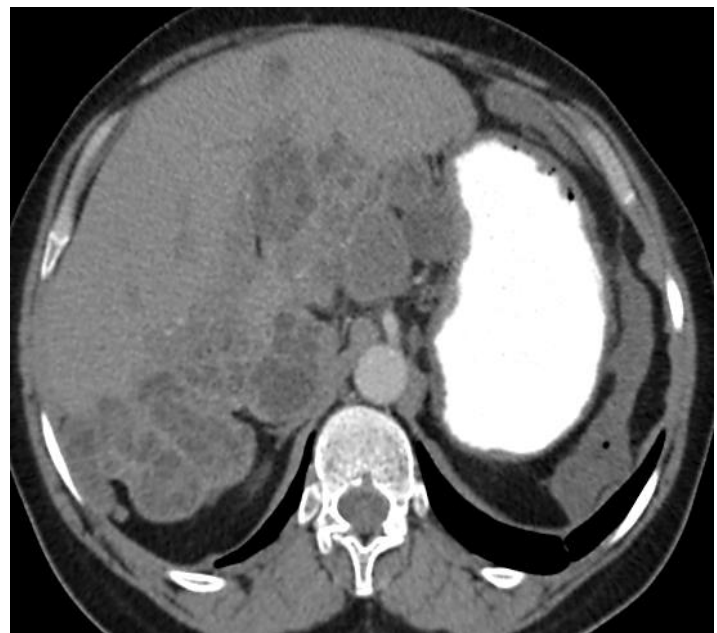
Between May 1996 and December 2013, 756 patients with a hydatid cyst were treated in the emergency operating room of Ankara Numune Education and Research Hospital. Of those, 16 patients experienced cyst rupture. We performed a retrospective analysis by retrieving the following information from the medical records of the patients: age, gender, cyst location, cyst diameter, cyst type, laboratory findings, surgical procedures, complications, and mortality.

Abdominal ultrasound and abdominal computed tomography (CT) were performed for preoperative diagnosis. CT is able to reveal multiple cystic lesions in the liver and peritoneum, and the absence of intraperitoneal fluid (Figures 1, 2). All patients were preoperatively administered pheniramine maleate and/or steroids to prevent allergic reactions. Seven patients were

given intravenous ceftriaxone (1 g) for prophylaxis, which was continued for 24 h after surgery to prevent postoperative infection. The cystic cavities were irrigated with scolicidal agents such as hypertonic saline (10%), povidone-iodine, or chlorhexidine gluconate (0.05%) plus cetrimide (0.5%). Multiple drains were placed in the intraperitoneal area. Bile ducts or orifices in the cyst cavity were sutured with nonabsorbable sutures.



**Figure 1.** Perforated hydatid cyst in the left hepatic lobe and perihepatic fluid (white arrows show ruptured HC cavity, black arrows show perihepatic free fluid).



**Figure 2.** Recurrent multiple hydatid cysts after partial cystectomy and drainage operation.

All patients were prescribed albendazole (15 mg/kg/day) for 4 months after surgery to prevent recurrence. The patients were asked to return to the hospital for follow up visits 1, 3, 6, and 12 months after surgery, and thereafter yearly for ultrasound, CT, liver function tests, and indirect hemagglutination tests to

detect cyst recurrence.

Friedman's test was used to analyze data, and p value <0.05 was considered as statistically significant.

## Results

The demographic and clinical characteristics of the patients and the type of trauma are shown in Table 1. All patients had evidence of peritoneal irritation, including extensive tenderness and guarding. Thirteen (81.3%) patients had nonspecific abdominal symptoms before cyst rupture, and three (18.8%) patients were asymptomatic. Only one (6.3%) patient had prior history of hydatid disease.

**Table 1.** Demographic characteristics of the patients.

	Mean±SD
Age (Mean±SD)	39,0±19,9
Sex	
Female (%)	7 (43.7 %)
Male (%)	9 (56.3 %)
History of trauma	
Yes (%)	5 (31.3%)
Falling down	2
Traffic accident	2
Assault	1
No (%)	11 (68.8%)
Follow-up (Months)	83.7±69.7

SD: Standard deviation.

Cyst location, cyst type, surgical treatment, and morbidities are summarized in Table 2. None of the patients died. Anaphylactic reactions were found in two (12.5%) patients. Six (37.5%) patients had intrabiliary rupture together with cyst rupture. The diameter of the biliary fistula was >0.5 cm in five of those patients. Those patients underwent cholecystectomy and T-tube drainage. An enterocutaneous fistula developed in two (12.5%) patients, despite T tube drainage. Five (31.3%) patients had the signs of cyst infection when the cyst cavity was opened. They underwent partial cystectomy and external drainage. One (6.3%) of the patients with an infected cyst also had an abscess cavity, while the other had an abscess cavity and wound infection. Of 16 patients included in this study, two (12.5%) had disease recurrence at 28 and 32 months after surgery.

There were 10 (67.5%) patients with type 3 cysts, four with type 4 cysts, one with a type 1 cyst, and one with a type 2 cyst. None of the patients had a type 5 cyst. These results indicate that type 3 cysts are more likely to rupture.

**Table 2.** Sites of the primary cysts, surgical procedures, and postoperative morbidities.

	Number of patients	%
<b>Localization</b>		
Liver right lobe	6	(37.5%)
Liver left lobe	6	(37.5%)
Liver right lobe +Omentum	1	(6.3 %)
Liver both lobes	1	(6.3%)
Spleen+mesocolon	1	(6.3%)
Omentum	1	(6.3%)
<b>Type of Cyst*</b>		
Type 1	1	(6.3%)
Type 2	1	(6.3%)
Type 3	10	(62.5%)
Type 4	4	(25.0%)
Type 5	0	
<b>Cyst Size</b>		
>10 cm	9	(56.3%)
5–10 cm	6	(37.5%)
<5 cm	1	(6.3%)
<b>Surgery</b>		
Partial cystectomy + ED**	8	(50.0%)
Partial cystectomy + Omentopexy+ ED**	5	(31.3%)
Partial cystectomy	1	(6.3%)
Capittonage	1	(6.3%)
Total cyst excision	1	(6.3%)
<b>Morbidity</b>		
Cavitary abscess	2	(12.5%)
Biliary fistula	2	(12.5%)
Eventration	1	(6.3%)
Wound infection	1	(6.3%)
Recurrence	2	(12.5%)

\* Classified according to Gharbi et al.[21], ED: External drainage.

## Discussion

Hydatid disease is endemic in various regions, including the Far East, Middle East, India, the Mediterranean region, and South America [10]. Cysts are generally asymptomatic, and signs and symptoms usually develop if the cyst increases in size, becomes infected, and ruptures into neighboring organs or the peritoneum. Intraperitoneal rupture may be associated with anaphylactic reactions, which may be life threatening [11]. The prevalence of direct cyst rupture varies considerably among studies. Sozuer et al. [12] Beyrouiti et al. [13], Ozturk et al. [1], and Malik et al. [14] reported the prevalence of cyst rupture as 8.6%, 1.75%, 3%, and 3%, respectively. In our study, 16/756 patients had a ruptured hydatid cyst, corresponding to a prevalence of 2% in a region of Turkey thought to be endemic for hydatid disease [15].

Intraperitoneal rupture of a hydatid cyst caused by scolices or daughter vesicles may lead to relapse. In earlier studies, the recurrence rates ranged between 0% and 28.5%, and may be related to factors such as operative technique, cyst location, and intracavitary or intraabdominal lavage during surgery. Kurt

et al. [16] reported intracavitary recurrence in 2/7 (28.6%) patients who underwent surgery due to an intraabdominal hydatid cyst. They suggested that recurrence was related to daughter cysts that were possibly left in the intracavitary space due to difficulty of exposure. Gunay et al. [17] did not report any recurrence in their follow-up of nine patients with ruptured intraabdominal hydatid cysts. Derici et al. [5] followed 17 patients for a median of 78 months (1–145 months), and reported only one (5.9%) intraabdominal recurrence 32 months after surgery. Yilmaz et al. [18] reviewed 68 patients, of which seven (10.3%) had postoperative recurrence. In our study, two (12.5%) patients had recurrence. Recurrence may be due to inadequate surgical or medical treatment after rupture of a hydatid cyst, or when intraabdominal area is not fully examined during surgery or in the postoperative period. However, some studies have reported recurrence beyond 2 years after surgery. Derici et al. [5] and Unalp et al. [9] reported the earliest recurrences 32 and 33 months after surgery, respectively. In our study, the recurrence was seen in two patients, 28 and 32 months after surgery.

Although surgery is essential for treatment of ruptured hydatid cysts, there is no consensus on the most appropriate surgical procedure. The surgical procedures used in the treatment of hydatid disease include radical (pericystectomy and hepatic resection) and conservative (unroofing, partial pericystectomy, and partial cystectomy combined with other procedures to manage the residual cavity) procedures [1]. Radical procedures carry higher operative risks compared to other procedures. Although conservative procedures are safer and easier to perform, they are associated with greater postoperative morbidity compared to radical procedures [1, 8, 9,18]. Dziri et al. [19] suggested that total pericystectomy should be the treatment of choice. Although Sozuer et al. [13] performed pericystectomy in some of their patients, they preferred conservative procedures such as partial cystectomy and drainage with or without omentopexy. We used conservative procedures in all of our patients except one patient who underwent left total cystectomy. We have preferred conservative approaches because they are easy to perform and we feel confident with those procedures, which means that these procedures are more appropriate for patients requiring urgent treatment.

The risk of direct cyst rupture increases as the diameter of cyst increases [20]. Large cysts with a diameter of >10 cm are thought to be more likely to rupture compared to smaller cysts [9]. In a series of 20 patients, Ozturk et al. [1] reported that all of the cysts had diameters  $\geq 10$  cm, and the mean diameter of the ruptured cysts was 12 cm. Mouaqit et al. [8] reported that nine (64.3%) of 14 ruptured cysts had cyst diameters >10 cm. In our study, nine (56.3%) patients had cyst diameters >10 cm. In our review of the English literature, we found no reports describing cyst rupture in cysts <4 cm.

In our study, 10 (67.5%) patients had type 3 cysts but none of them had type 5 cysts. Type 3 hydatid cysts appear to be more likely to rupture compared to other cyst types. According to the classification proposed by Gharbi et al. [21], the germinal membranes of type 3 cysts are defective, allowing the scolex to form daughter vesicles. Defects in the membrane can therefore weaken the cyst wall. Since type 5 cysts are calcified hydatid cysts, they are unlikely to rupture.

Hydatid cysts usually rupture due to trauma, or they may rupture spontaneously due to increased intracystic pressure. The intracystic pressure may reach very high levels (up to 61 cm H<sub>2</sub>O). In a series of 21 patients, Unalp et al. [9] reported that the cyst rupture followed abdominal trauma in 13 (57.2%) patients while rupture was spontaneous in eight (42.8%) patients. Kurt et al. [16] reported that intraperitoneal cyst rupture followed trauma in two (28.6%) of seven consecutive patients. Overall, five (31.3%) of our patients had a history of trauma, including traffic accident, fall, and assault. Our patients were mostly middle-aged people, with a mean age of 39.0 $\pm$ 19.9 years.

## Conclusion

Hydatid cysts are generally asymptomatic, and signs and symptoms usually develop when cyst size increases, it becomes infected, and ruptures into neighboring organs or the peritoneum. Large cysts, especially those with a diameter >10 cm as well as type 3 cysts are associated with a high rupture risk.

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