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## Original Article

# A Troublesome Complication of Hydatid Cysts; Intra-Abdominal Rupture: A 10-Year Study from a High-Volume Center

\*Hüseyin Yönder, Mehmet Sait Berhuni, Hasan Elkan, Abdullah Özgönül, Ahmet Bertan, Vedat Kaplan, Ali Uzunköy

Department of General Surgery, Faculty of Medicine, Harran University, Şanlıurfa, Turkey

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### \*Correspondence Email:

hyonder@hotmail.com

### Abstract

**Background:** Hydatid cyst is a parasitic infection, often caused by *Echinococcus granulosus*. Although it is classified as a benign disease, cyst ruptures in the abdomen can be fatal. Ruptures occur spontaneously or after trauma. We aimed to report data from patients who underwent emergency surgery due to spontaneous intra-abdominal hydatid cyst rupture.

**Methods:** Upon a retrospective review of the records at Department of General Surgery, Faculty of Medicine, Harran University, Şanlıurfa, Turkey, we found that 34 cases were operated on due to hydatid cyst rupture between January 2012 and October 2022. All patients were operated on in an emergency, and partial cystectomy, intra-abdominal irrigation, and drainage were performed using laparotomy. The patients were evaluated in terms of age, sex, symptoms, radiological findings, laboratory results, intraoperative findings, and postoperative follow-ups.

**Results:** Twenty-two (64.7%) female and 12 (35.3%) male patients were enrolled. The mean age was 39.1 ( $\pm 17.58$ ) years. All patients experienced spontaneous rupture. The ruptured cyst was found in the liver in 32 patients (94%), the spleen in 1 patient (3%), and the pelvis in 1 patient (3%). The diagnosis was determined using ultrasonography in 12 (35.3%) patients, computed tomography in 21 (61.8%) patients, and magnetic resonance imaging in 1 (2.9%) patient. All patients exhibited acute abdomen and leukocytosis. The average length of hospital stay was 5.14 ( $\pm 1.37$ ) days.

**Conclusion:** Hydatid cyst rupture should be considered in cases of acute abdomen, particularly in regions where the disease is endemic, as in our region. The ruptured hydatid cyst was primarily observed in the liver (94.11% of cases).



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## Introduction

**H**ydatic cyst disease, also known as echinococcosis or hydatidosis, was first described in dogs by Hartman in 1695. In most cases, *Echinococcus granulosus* is the causative microorganism. Turkey is a community where the disease is common. The disease affects 70% of the liver, 20% of the lungs, and 5%–10% of other organs in people (1-3).

Hydatid cysts are rarely symptomatic unless they become large enough to induce compression symptoms. Ultrasonography (USG) and computed tomography (CT) are primarily used in diagnosis (1).

The most common symptom of cyst perforations into the peritoneal cavity is right upper quadrant pain. This pain can also be felt widely throughout the abdomen and is often accompanied by nausea and vomiting. Peritoneal findings are more common in perforations with infected cyst content (1).

We aimed to remind people that hydatid cyst rupture is a clinical condition that should be considered in cases of acute abdomen in our region, where the disease is endemic, and to present data from patients with ruptured cysts.

## Methods

Between January 2012 and November 2022, 34 patients were operated at Department of General Surgery, Faculty of Medicine, Harran University, Şanlıurfa, Turkey due to intra-abdominal hydatid cyst rupture. Patients were evaluated in terms of age, sex, reason for admission, reason for rupture, number of cysts, location and diameter of the ruptured cyst, diagnosis method, preoperative white blood cell and bilirubin values, whether they had previously received hydatid cyst diagnosis and treatment, length of hospital stay, and perforated cyst. Hydatid cyst rupture was examined in terms of its relationship with bile ducts, postoperative bile leakage, the Gharbi classifi-

cation of the ruptured cyst, nature of the intra-abdominal fluid, and whether or not it indicated preoperative allergic reactions.

Under general anesthesia, all patients underwent a laparotomy with a midline incision or subcostal incision, depending on the location of the cyst. All intra-abdominal fluid was aspirated. Partial cystectomy was performed in 33 patients, and total pericystectomy was performed for pelvic cysts. The abdomen and cyst cavities were irrigated with diluted povidone-iodine and 10% hydrogen peroxide solution. Primary repair with 4/0 prolene was performed in cases when a bile duct opening was detected. Drains were placed into the abdomen and the cyst lumen for drainage purposes at the end of the surgery, and the surgery was terminated.

Following surgery, all patients were administered 10 mg/kg albendazole for 6 months.

## Statistical analysis

The data were analyzed using the SPSS 22 (IBM Corp., Armonk, NY, USA) presented as number, percentage, mean, median, minimum, and maximum. The Shapiro-Wilk test was used as a normal distribution test. The Mann-Whitney *U* test, *t* test, Chi-square test, and logistic regression analysis were used in the study. A *P*-value of <0.05 was considered statistically significant.

## Ethics committee approval

This study was approved by Harran University Clinical Research Ethics Committee (Date: 21/08/2023—HRÜ. 23/15/27).

## Results

The study included 22 (64.7%) female patients and 12 (35.3%) male patients. The mean age was 39.14 ( $\pm 17.58$ ) years. Sixteen patients (47.1%) had a preoperative diagnosis of hydatid cyst. They had received hydatid cyst treatment. At the time of diagnosis, 21 (61.8%)

patients had a single cyst, and 13 (38.2%) patients had multiple cysts. However, only one cyst was perforated in all patients. All patients had spontaneous hydatid cyst perforation.

While all patients had acute abdomen along with abdominal pain, nausea or vomiting was reported by 26 of the patients (76.5%). The ruptured cyst was located in the right lobe in 21 patients (61.8%), the left lobe in 11 patients (32.3%), and the spleen in 1 patient. The pelvis was located independently of the surrounding organs in one patient.

Perforation was diagnosed using CT in 21 (61.8%) patients, USG in 12 (35.3%) patients, and magnetic resonance imaging (MRI) in 1 (2.9%) patient. The mean complaint onset time was 1.70 ( $\pm 0.70$ ) days. While bilirubin levels were normal upon admission in 27 (79.4%) patients, hyperbilirubinemia was detected in seven (20.6%) patients. All of our patients had leukocytosis. The segmental distribution of ruptured cysts is shown in Table 1.

**Table 1:** Ruptured cyst localization

| <i>Variable</i> | <i>Number</i> | <i>Percent</i> | <i>Cumulative percentage</i> |
|-----------------|---------------|----------------|------------------------------|
| Spleen          | 1             | 2.9            | 2.9                          |
| Segment 2       | 3             | 8.8            | 11.8                         |
| Segment 3       | 3             | 8.8            | 20.6                         |
| Segment 4       | 5             | 14.7           | 35.3                         |
| Segment 5       | 7             | 20.6           | 55.9                         |
| Segment 6       | 5             | 14.7           | 70.6                         |
| Segment 7       | 7             | 20.6           | 91.2                         |
| Segment 8       | 2             | 5.9            | 97.1                         |
| Pelvis          | 1             | 2.9            | 100.0                        |
| Total           | 34            | 100.0          |                              |

According to the Gharbi classification, 13 (38.2%) of the ruptured cysts were stage 1, 13 (38.2%) were stage 2, and 8 (23.5%) were stage 3.

While 24 (70.6%) patients had no allergic reaction at the time of diagnosis, 10 (29.4%) patients had an allergic reaction. While eight (80%) of the patients with allergic reactions had local findings, two (20%) patients had systemic abnormalities.

When the fluid aspirated from the abdomen was evaluated intraoperatively, it was found to be serous in 16 (47.1%) patients, bilious in 13 (38.2%) patients, and purulent but without bile in 5 (14.7%) patients. While no bile connection was found in 19 (59.37%) perforated

liver hydatid cysts, 13 (40.63%) patients had an intraoperative bile duct connection.

Bilious discharge was found in the drains of 10 (29.4%) patients during postoperative follow-up. Because the drain flow did not decrease during follow-up, eight (23.5%) patients underwent endoscopic retrograde cholangiopancreatography (ERCP). Due to conservative follow-up, biliary drain discharge was discontinued in two (5.9%) patients.

There was no mortality in 34 patients during follow-up. Recurrence of the perforated cyst was detected during follow-up in 8 of the 34 patients (23.5%) included in the study. Descriptive data on the enrolled patients are shown in Table 2.

**Table 2:** Descriptive data

| <i>Variable</i>                        | <i>Mean</i> | <i>SD</i>  | <i>Median</i> | <i>Minimum</i> | <i>Maximum</i> |
|--|-------------|------------|---------------|----------------|----------------|
| Age (yr)                               | 39.147      | 17.5881    | 36.500        | 18.0           | 97.0           |
| WBC (White Blood Cell) level (10e3/uL) | 14,647.059  | 2,816.4275 | 14,500.000    | 11,000.0       | 23,000.0       |
| Length of hospital stay (days)         | 5.147       | 1.3955     | 5.000         | 3.0            | 8.0            |
| Detection time of recurrence (months)  | 1.559       | 2.9969     | 0.000         | 0.0            | 10.0           |
| Total follow-up period (months)        | 28.735      | 11.6185    | 28.000        | 6.0            | 48.0           |

According to the present data, no significant relationship was observed between the diameters of the patients' perforated cysts and the risk of developing allergic reactions ( $P = 0.528$ ). Moreover, no significant relationship was found between the diameter of the perforated cysts and the recurrence of these cysts during follow-up ( $P= 0.084$ ).

In pairwise comparisons, no correlation was observed between the location of the cysts in the patient (right lobe–left lobe–pelvis–spleen) and the number of cysts (single–multiple) and the frequency of allergic reactions at the time of diagnosis ( $P= 1.00$  and  $P= 0.755$ , respectively).

No correlation was found between hyperbilirubinemia detected at admission and the risk of allergic reactions upon diagnosis ( $P= 1.00$ ). Furthermore, there was no correlation between the opening of perforated cysts into the bile ducts and the risk of allergic reactions at diagnosis ( $P= 0.704$ ).

When the analyses results were evaluated in terms of recurrence in follow-ups, no correlation was noted between any of the criteria, such as cyst location, number of cysts, presence of hyperbilirubinemia at admission, and opening of the biliary tract, and recurrence in follow-ups. Additionally, the status of the bile

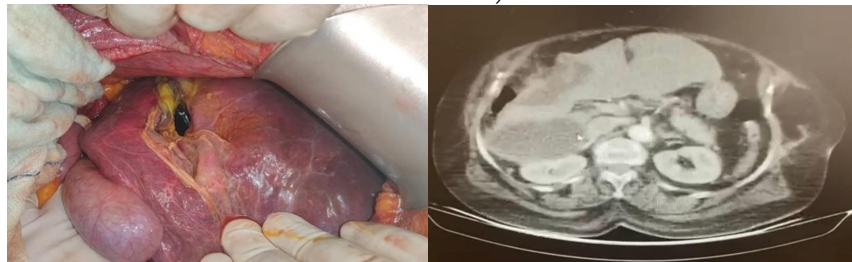
duct opening was unrelated to the diameter of the perforated cyst ( $P= 0.241$ ).

The logistic regression model developed using clinically important factors (bile duct opening, number of cysts, and diameter) was insignificant in terms of postoperative recurrence.

### Discussion

Hydatid cyst is a disease mainly caused by *E. granulosus*, which is commonly found in the liver. Although considered a benign disease, it has the potential for severe, life-threatening complications (1,2). The third most common complication in hydatid cysts, following bile duct opening and allergic reaction, is intraperitoneal perforation. Cyst perforation may occur due to trauma, spontaneously, or during surgical procedures (1-3).

In the literature, young age, increased cyst diameter (>10 cm), intracystic pressure, and peripheral cyst location are considered risk factors for the development of perforation (4). In our study, the mean age of patients who developed perforation was 39.14 (18–97) years. The perforated cysts had an average diameter of 10.8 cm and were all-adjacent to the subcapsular area where they were perforated (Fig. 1).



**Fig. 1:** Intraoperative and computed tomography images of the perforated cyst

### *Cyst perforation types*

It can occur in three forms: closed perforation, which does not disrupt the integrity of the host tissue because of partial rupture of the cyst, perforation by opening into the bile ducts, and free perforation into the peritoneal cavity (5). Our study included 34 patients with spontaneous perforation into the peritoneal cavity.

The most common symptom of cyst perforations into the peritoneal cavity is right upper quadrant pain. This pain can also be felt widely throughout the abdomen and is often accompanied by nausea and vomiting. Peritoneal findings are more common in perforations with infected cyst content (1). While all patients in our study had acute abdomen associated with abdominal pain, 26 patients (76.5%) had nausea or vomiting. The main factor determining the time between the onset of symptoms and the time of admission to the hospital is the severity of the symptoms, and in our study, this period was, on average, 1.7 ( $\pm 0.70$ ) days. Regarding the characteristics of the perforated cysts in our study, they were serous in 16 (47.1%) patients, bilious in 13 (38.2%) patients, and purulent but without bile in 5 (14.7%) patients. All of our patients had leukocytosis.

Patients who develop perforation due to the toxic and allergic effects of the cyst fluid may experience allergic reactions ranging from a mild rash to anaphylaxis. Studies show minor allergic reactions in the range of 16%–25% and major allergic reactions in the range of 1%–12% (6). In our series, an allergic reaction was reported in 10 (29.4%) of the patients at the time of diagnosis. Local findings were observed in eight patients (23.5%), and systemic findings in two (5.9%) patients.

Ultrasonography, which also provides the Gharbi classification and has an 85% sensitivity, can be used for diagnosis (1,7). Although CT has 100% sensitivity, its use is limited, particularly in hemodynamically unstable patients (8). If the diagnosis remains doubtful, an MRI

may be recommended. The presence of a collapsed cyst wall, free-floating daughter vesicles in the abdomen, and a discontinuous cyst wall on CT is diagnostic. In the present study, CT was used to diagnose perforation in 21 (61.8%) patients, USG in 12 (35.3%) patients, and MRI in 1 (2.9%) patient. When the Gharbi classification of perforated cysts was evaluated, 13 (38.2%) were stage 1, 13 (38.2%) were stage 2, and 8 (23.5%) were stage 3.

Although there are case reports in the literature with nonoperative follow-up (9), surgery remains the gold standard treatment in perforated hydatid cyst patients due to the risk of anaphylaxis. Two surgical approaches can be used. The first is partial cystectomy, and the second is radical (total pericystectomy or hepatectomy) surgery. As a requirement of emergency surgery, conservative surgery (partial cystectomy) appears to be the preferred surgical procedure to decrease the risk of bleeding and the length of the operation (10). In our dataset, only one patient had a partial cystectomy with an open technique.

An important step of surgery is irrigating the cystic cavity with scolicidal agents. Many scolicidal agents are used for this purpose. Frequently used agents include hypertonic saline, hydrogen peroxide, povidone-iodine, and silver nitrate solutions (8,11,12). In our patients, the abdomen and cyst contents were irrigated with povidone-iodine and hydrogen peroxide.

If intraoperative bile ducts are visible opening into the perforated cyst lumen, they must be sutured. If there is high-flow bile leakage from the drain in the postoperative period, an ERCP procedure is an option for diagnosis and treatment (sphincterotomy and stenting) (13). The present study showed a gap between the cyst wall and the bile ducts in 13 (40.63%) patients during surgery. The connection was detected, and primary repair was performed. During postoperative follow-up, 10 (29.4%) patients had bilious discharge from their drains. An ERCP procedure was performed in eight patients (23.5%) because the drain flow



rate did not decrease during follow-up. Because of conservative follow-up, bile flow from the drain stopped in two (5.9%) patients.

There is no common consensus on the duration of anthelmintic treatment during postoperative follow-up (2). Patients were provided postoperative medical treatment for at least 6 months, along with hydatid cyst serology follow-ups, based on our clinical experience.

Recurrence rates after perforation range between 3% and 25% (14). In this study, follow-ups with USG were performed at 3- and 6-month intervals. Imaging was performed early for patients whose abdominal problems recurred in the postoperative period, and they were promptly evaluated for recurrence. These controls aimed to detect recurrences that may develop despite surgery or subsequent medical treatment. The mean follow-up period for the patients was 28.7 ( $\pm 11.44$ ) months. Recurrence in the perforated cyst was found in eight of them (23.5%). The mean time to detect recurrence postoperatively was 6.62 ( $\pm 1.86$ ) months. In the most recent case, recurrence was observed in the 10th month. Other cysts in patients with multiple cysts who only had the perforated cyst surgically removed were not included in detecting recurrence.

Despite previous studies reporting mortality rates between 0% and 23.5% (15,16), we had no patients with postoperative mortality in our study. However, a patient who, although excluded from the study, was brought to the emergency room with anaphylaxis and diagnosed with a hydatid cyst rupture, died before surgery could be performed.

Although our clinic is located in an endemic region, it should be noted that the number of patients was small, and the single-center, retrospective design of the study was a limitation.

## Conclusion

Although rare, hydatid cyst rupture should be considered in patients presenting with acute abdominal findings, particularly in endemic regions. Radical excisions should be

avoided in surgery, and conservative surgery should be performed in emergencies. After surgery, all patients should begin medical treatment. High-volume, multicenter studies are needed to support the results of the study.

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The data reported in this study were previously presented at a congress, the 10th National and third International Hydatidology Congress, held Online in Turkey in 2022.

## Conflict of interest

There are no conflicts of interest.

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