

# Rupture of liver hydatid cysts into the peritoneal cavity

## A challenge in endemic regions

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### ABSTRACT

**الأهداف:** تقييم الظهور السريري والنتائج المبكرة وعلى المدى الطويل للمرضى الذين خضعوا للعلاج الجراحي لتمزق أكياس الكبد العدارية.

**الطريقة:** تم تقييم الملفات الطبية لعدد 21 مريض والذين تعرضوا لتمزق في أكياس الكبد العدارية في دراسة استيعادية خلال الفترة ما بين يناير 2000م وأبريل 2009م. نظراً لكون عدد ملفات المصابين بأكياس عدارية في الكبد 368 مريض. تم تقييم العمر، ونوع الجنس، والأعراض والنتائج المخبرية، والإجراءات التشخيصية، وأنواع المعالجة الجراحية، ومدة البقاء في المستشفى، ومعدل الوفيات، والحالة المرضية، ومعاودة ظهور المرض.

**النتائج:** من بين أولئك المرضى كان هنالك 16 مريض (76.2%). بلغ متوسط العمر 43.8 عام. كانت الأكياس مفردة لدى 16 حالة (76.2%). كانت الأكياس متموضعة في الفص الأيسر لدى 14 حالة (66.7%). كما كان هنالك كيس في الطحال لدى أربعة مرضى (19%). كان التدهور البسيط أو الإصابة المباشرة في البطن قابلة للاستجابة لدى 8 من المرضى (38.1%) وأشكال راجلة لدى 4 حالات (19%). تعرضت 9 حالات (42.8%) لتمزق عفوي. أجريت عملية التصريف الخارجي وتقويم الثرب لدى 12 مريض (57.1%) ولدى 9 مرضى (42.8%) لعلاج تجويف الكيس. تعرض 9 مرضى للحالة المرضية من بين 11 شخص (42.8%). لم يكن هنالك نسبة وفيات بعد العملية الجراحية. بلغت متوسط فترة المتابعة 63.1±31.3 شهر ومعاودة الظهور 23.8%.

**خاتمة:** تعتبر معدلات معاودة الظهور والحالة المرضية للتدخلات الجراحية من أجل أكياس الكبد العدارية عالية. على الرغم من ندرة حدوث هذا المرض إلا أنه ينبغي إدراجه في مختلف التشخيصات في حالات البطن الحادة في المناطق المستوطنة.

**Objective:** To evaluate the clinical presentation and early and long-term outcomes of patients treated surgically for intraperitoneal ruptured liver hydatid cysts.

**Methods:** The medical records of 21 patients with rupture of hydatid cysts were evaluated retrospectively between January 2000 and April 2009 at Izmir Atatürk Training and Research Hospital, Izmir, Turkey, as were the records of 368 patients with hydatid cysts. Age, gender, symptoms, laboratory findings, diagnostic procedures, surgical treatment modalities, in-hospital stay, morbidity, mortality, and recurrence were evaluated.

**Results:** Sixteen of these patients (76.2%) were women. The mean age was 43.8 years. Cysts were single in 16 cases (76.2%). In 14 cases (66.7%) cysts were localized only in the right lobe. There was also a cyst in the spleen in 4 patients (19%). Simple falls or direct abdominal minor trauma was responsible in 8 patients (38.1%), and pedestrian mishaps in 4 (19%). Nine (42.8%) patients experienced spontaneous rupture. External drainage was performed in 12 (57.1%) patients and omentoplasty in 9 patients (42.8%) to manage the cyst cavity. A total of 11 morbidities developed in 9 patients (42.8%). There was no postoperative mortality. The mean hospital stay was 11.3±7.4 days, and mean follow-up was 63.1±31.3 months. The recurrence rate was 23.8%.

**Conclusions:** The morbidity and recurrence rates of surgical interventions for ruptured hydatid cysts are high. This pathology, although rare, should be included in the differential diagnosis of an acute abdomen in endemic areas.

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Hydatid disease is a parasitic infestation caused by *Echinococcus granulosus* and less frequently by *Echinococcus multilocularis*. Hydatidosis is endemic in the Mediterranean region. The globally found parasite causes considerable socioeconomic consequences in endemic areas. However, the incidence is relatively low in Western industrial nations. Hydatid disease is still an important public health problem in Turkey, with a prevalence of 585 per 100,000 population in 1991, and 291 per 100,000 population in 1999.<sup>1</sup> Following oral intake of the cestode's eggs (diameter 40 µm) the oncospheres are released in the upper gastrointestinal tract, penetrate the gut wall, enter the portal vein, and reach the liver. In humans, 50-75% of hydatid cysts occur in the liver, 25% are found in the lungs, and 5-10% are distributed along the arterial system.<sup>2</sup> In principle, all tissues can be affected, however, in around 80% of patients a solitary cyst occurs in one organ.<sup>3</sup> Symptoms are caused by compression or displacement of other structures or organs. The most common complaints of hydatid disease are abdominal pain, nausea, vomiting, or jaundice. However, hydatid disease may be asymptomatic or have few symptoms for many years and is commonly diagnosed alongside other illnesses.<sup>4</sup> Ultrasonography (US) and computerized tomography (CT) are the major diagnostic methods in use.<sup>5</sup> The differential diagnosis comprises dysontogenetic cysts, abscesses, metastasis, and occasionally alveolar echinococcosis. Complications of hepatic hydatid cysts are perforation of the cyst, either internally or externally as a result of increased intracystic pressure and secondary bacterial infection.<sup>4,6</sup> In hydatid cysts of the liver, spontaneous or traumatic perforation into the peritoneum can occur in 2-7% of the cases, followed by secondary infection, jaundice, and an anaphylactic reaction.<sup>2</sup> Systemic anaphylactic reactions may be a life-threatening complication, and have been reported in 1-12.5% of the patients with intraperitoneal perforations.<sup>6,7</sup> The treatment of intraperitoneal rupture of the hydatid cyst requires emergency surgical intervention, however, there is no consensus for the treatment of cyst rupture.<sup>8</sup> In this study, we evaluated 21 patients with ruptured liver hydatid disease into the peritoneum with regard to surgical treatment modalities and postoperative morbidity, mortality, and recurrence rates.

**Methods.** We retrospectively evaluated the medical records of 21 patients with intraperitoneal perforation of liver hydatid cysts. The patients were admitted to the 1st and 4th General Surgery Clinics, Izmir Atatürk Training and Research Hospital, Izmir, Turkey between January 2000 and April 2009. In the same interval, 368 cases of hydatidosis were treated in our clinics; the

frequency of perforation was 5.7%. Recorded patient data in the electronic charts and the clinic's information systems during treatment and follow-up was collected for this study. When analyzing the patients' medical records, the following criteria were assessed: age, gender, initial complaints, physical findings, time to surgery from the onset of the symptoms, laboratory findings, diagnostic procedures, surgical procedures, in-hospital stay, morbidity, mortality, and recurrences. The preoperative evaluation included blood tests (complete blood count, liver function tests), chest x-ray, abdominal US, abdominal CT, and magnetic resonance imaging (MRI). A chest and abdominal x-ray and abdominal US were performed in all patients at admission. Computed tomography was performed in 11 patients, and MRI scans in 2 patients. The upper normal limits for liver function test results in our laboratory were as follows: aspartate aminotransferase (AST), 37 U/L; alanine aminotransferase (ALT), 40 U/L; alkaline phosphatase (ALP), 133 U/L; gamma-glutamyl transpeptidase (GGT), 50 U/L; total bilirubin level, 1.2 mg/dL (20 µmol/L); and leukocyte count, <10000 /mm<sup>3</sup>. A midline incision was performed in all patients. Besides managing peritoneal dissemination, conservative treatment of intact and perforated cysts was applied. The cystic cavities were irrigated with hypertonic saline (3%) for 10-15 minutes. Any orifice of bile ducts observed on the inner surface of the cavity was sutured with nonabsorbable sutures. The peritoneum was lavaged with povidone-iodine solutions followed by isotonic saline and multiple drains were placed before the abdomen was closed. Albendazole treatment 10 mg/kg per day (Andazol; Biofarma, Istanbul, Turkey) was given to all the patients for 3 months postoperatively to prevent recurrence. The patients were seen every 3 months during the first postoperative year, every 6 months for the second year, and annually thereafter. During follow-up, US and indirect hemagglutination tests were performed to detect recurrences. Computed tomography scan was performed as required. The observation of a cyst in patients with an indirect hemagglutination test result of 1/256 or above was reported as a recurrence.

**Results.** Sixteen of the patients (76.2%) were women and 5 (23.8%) were men. The mean age was 43.8 (range 14-76 years). Seven patients (33.3%) had a history of chronic nonspecific abdominal pain and dyspepsia. Two patients (9.5%) had previous diagnosis of hydatid disease and one of them had previously undergone hydatid cyst surgery. Ten patients (47.6%) did not have any complaints prior to the rupture of the cysts. In patients with rupture, the most common complaint

**Table 1** - Demographic features, clinic appearance, and etiology of 21 patients with ruptured liver hydatid cyst.

Variable	n	(%)
Female/male ratio	16/5	
<i>Common complaints</i>		
Acute abdominal pain	20	(95.2)
Abdominal distension	16	(76.2)
Nausea and vomiting	10	(47.6)
Urticaria	3	(14.3)
Jaundice	2	(9.5)
<i>Etiology</i>		
Trauma	4	(19.0)
Spontaneously	17	(81.0)

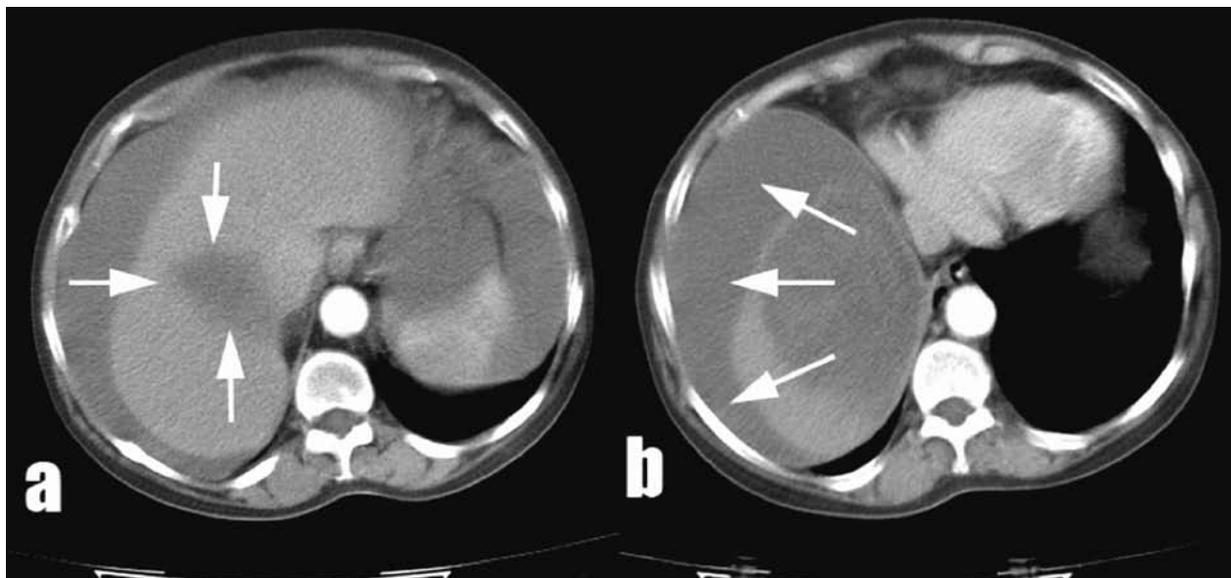
The mean age (years) was  $43.8 \pm 17.2$  with an age range of 14 -76

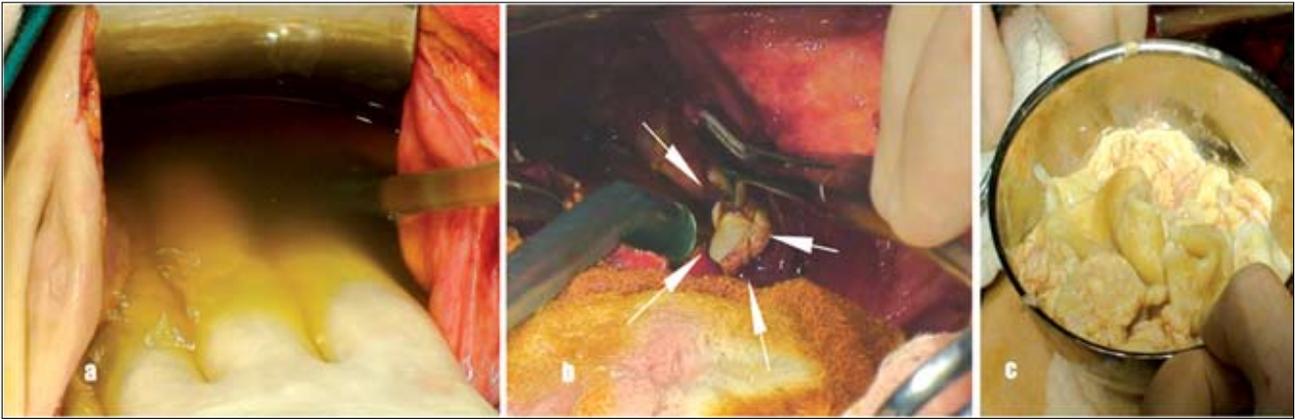
**Table 2** - Laboratory results of 21 patients with ruptured liver hydatid cyst.

Laboratory results	Mean $\pm$ SD	Range
Hemoglobin (gr/dL)	$11.6 \pm 1.2$	(8.7 - 14.2)
ALP	$110 \pm 21.8$	79 - 145)
ALT	$45.2 \pm 40.1$	(6 - 150)
AST	$45.4 \pm 38.4$	(7 - 168)
Bilirubin (mg/dL)	$1 \pm 0.3$	(0.2 - 3.1)
GGT	$117 \pm 25.6$	(84 - 161)
Eosinophil	$1.1 \pm 0.8$	(0.1 - 3.6)

ALP - alkaline phosphatase, ALT - alanine aminotransferase, AST - aspartate aminotransferase, GGT - gamma-glutamyl transpeptidase

was acute abdominal pain, followed by abdominal distension, nausea and vomiting, urticaria, and jaundice. All patients had signs of peritoneal irritation such as extensive tenderness and guarding (100%). Four patients had a history of minor blunt abdominal trauma (fall), but 17 patients did not describe any trauma. Demographic features, clinic and radiologic appearance, and laboratory results of patients are presented in Table 1 & 2. Eight patients (38.1%) had fever, 16 (76.2%) had elevated white blood cell count, and 2 (9.5%) had hyperbilirubinemia. Ultrasonography revealed free intraabdominal fluid in all cases. A heterogeneous cavity and/or multiple intraperitoneal cysts were visible in 21 (100%) cases. There was also a cyst in the spleen in 4 patients (19%) requiring splenectomy. Both CT (Figures 1a and 1b) and MRI confirmed US findings. Cysts were single in 16 cases (76.2%), and multiple in 5 cases (23.8%). In 14 cases (66.7%) cysts were localized only in the right lobe, in 2 cases (9.5%) cysts were only in the left lobe, and in 5 cases (23.8%) cysts were localized in both lobes. All ruptured cysts were located in the right lobe. There was also a cyst in the spleen in 4 patients (19%). Etiologic factors varied; simple falls or direct abdominal trauma was responsible in 8 patients (38.1%) and pedestrian mishaps in 4 (19%). Nine (42.8%) patients experienced spontaneous rupture. The mean interval between trauma and presenting at the hospital was 70 minutes (range 20-240), and it was 122 minutes (range 20-480) between presentation and surgery. Between 1-7 liters of hydatid fluid with floating daughter cysts and, purulent material was present in the

**Figure 1** - A CT image showing a) ruptured hydatid cysts of the liver, and b) intra-abdominal fluid.



**Figure 2** - Intraoperative appearance of a hepatic ruptured cyst showing, a) hydatid fluid in the abdomen, b) ruptured liver cyst, and c) cyst content.

abdomen. An example of the intraoperative appearance of a ruptured liver cyst is shown in **Figure 2**. Unroofing the cyst and external drainage in 12 patients (57.1%), and omentoplasty in 9 patients (42.9%) were performed after removal of the intraperitoneal fluid to manage the cyst cavity. Cholecystectomy with T-tube drainage was required due to direct communication between the cyst and the gallbladder in one patient (4.7%) and cholecystectomy was performed for cholelithiasis in another patient (4.7%). No patients died in the early postoperative period. A total of 11 complications developed in 9 patients (42.8%). Low output biliary fistula (less than 300 mL/day) developed in 3 patients (14.2%) in whom fistulas terminated spontaneously, and high output biliary fistula developed in a patient (4.7%) who had undergone external drainage and required endoscopic sphincterotomy to control the fistula. Abscess in the cystic cavity developed in one case (4.7%) and required percutaneous abscess drainage. Wound infection and eventration developed in 4 (19%) patients. Other complications were pulmonary infection in one patient, and pleural effusion in one patient. The mean hospital stay was  $11.3 \pm 7.4$  days (range: 4-34 days), and the mean follow-up was  $63.1 \pm 31.3$  months (range 4-100 months). Recurrence developed in 5 patients (23.8%) in 33, 54, 71, 86, and 100 months after surgery. Recurrences were in the liver in 4 patients, and the peritoneum in one patient, and 3 of these patients underwent additional surgery. Total cystectomy was performed for the one recurred cyst in the peritoneum. Unroofing the cysts with external drainage was the operation of choice for recurrent liver hydatid cysts in 2 patients. The detected recurrences were too small (<4 cm) for surgical intervention in 2 patients with liver hydatid disease. These 2 patients are currently under our medical follow-up.

**Discussion.** Perforation of the hydatid cyst into the peritoneal cavity is a very rare event, even in areas where the disease is endemic.<sup>9</sup> Scolexes develop on the surface of the germinative membrane and secrete into the cyst a fluid that has both isotonic and antigenic properties. As more scolexes are produced, the cyst enlarges continuously filled by the secretion of more hydatid fluid. The intracystic pressure increases to more than 50 cm H<sub>2</sub>O with this growth, thereby leading to spontaneous or traumatic rupture.<sup>10</sup> In a large series, the rate of rupture of hydatid cysts into the peritoneum has been reported to be between 5.5-16%.<sup>11-13</sup> In the present study; the cyst rupture rate was 5.7%. Major or minor blunt abdominal trauma is the most frequent cause of perforation.<sup>6,14</sup> In our case series, 19% of the patients had a history of minor trauma to the abdomen, and 81% had spontaneous perforation. Abdominal pain is the most common presenting symptom of peritoneal perforation (76-100%).<sup>14,15</sup> Allergic reactions are not as frequent as once thought, and may be seen in 16.7-25% of the cases with ruptured hydatid cysts.<sup>6-8,16</sup> Jaundice may also occur after rupture of the cyst into the biliary system.<sup>9,17</sup> **Table 1** outlines the common complaints found in our patients.

Following rupture, anaphylaxis may result from drainage of the hydatid fluid into the peritoneal cavity.<sup>18</sup> Although fatal anaphylaxis after cyst rupture has been reported, anaphylactic reactions are rarer than expected, and present 1-12.5% among patients with ruptured hydatid cysts.<sup>6-9,14,19</sup> No anaphylaxis developed in our cases, but the potential for fatal anaphylaxis should always be kept in mind.<sup>8</sup> Rupture of a hydatid cyst requires emergency surgical intervention if the general condition of the patient allows. The goal of the surgical treatment is to eliminate disease, and to minimize morbidity, mortality, and recurrence rates.<sup>9,20</sup> There is no standardized treatment approach or clear

guideline for intraoperative management of ruptured liver hydatid disease. The main treatment modalities for noncomplicated cases are also valid for complicated ones, such as peritoneal perforation.<sup>8,11</sup> However, in complicated cases, profuse peritoneal lavage with hypertonic solutions appears to be more necessary to prevent recurrences in the abdomen.<sup>8</sup> Conservative methods such as external drainage, unroofing, and cavity filling are mostly preferred in endemic regions, because radical techniques such as pericystectomy and liver resection, are not always technically possible, involve longer surgical procedures, and require more experience.<sup>4,6,12</sup> Since the scolexes spread into the abdominal cavity following perforation, the cavity of the cyst should be identified and a suture should be placed, or closed with a clamp.<sup>10</sup> Followed by administration of scolical agents into the cyst cavity, and after a waiting period of 10-15 minutes, the cavity should be aspirated; partial or, if possible, total cystectomy should be performed.<sup>10</sup> In the current study, all of our patients received conservative surgical interventions. Partial cystectomy was performed in all patients. Both external tube drainage and omentopexy were performed for cavity management in our patients.

The most important step is discarding the cystic content and germinative membranes and irrigating the peritoneal cavity with a sufficient amount of scolical agents and removal of all cyst content.<sup>8,10,17,20</sup> Although several scolical agents such as hypertonic saline solution, formalin, silver nitrate, povidone-iodine, chlorhexidine, and a combination of cetrimide and chlorhexidine, have been used for the purpose of inactivation, there is no consensus on which is the best agent.<sup>9</sup> We irrigated the perforated cysts cavity and abdomen with hypertonic saline (3% sodium chloride) and then with isotonic saline. We did not detect an increase in serum sodium and chloride concentrations following abdominal lavage in our patients. An alternative to conventional surgery, with the use of minimally invasive treatment methods such as laparoscopic treatment<sup>21</sup> or PAIR (puncture, aspiration, injection, respiration)<sup>22</sup> has gained interest during the last decade for uncomplicated hydatid disease. Although recent reports on PAIR are encouraging for management of uncomplicated cyst; presently, these techniques are not sufficient in the management of ruptured hydatid cysts with peritoneal spillage.<sup>8,22</sup>

The literature contains relatively few series of patients with perforated hydatid cysts. There are more associated organ injuries due to trauma or fall in these patients, so determining associations between surgical interventions and morbidity or mortality is difficult. It is well known that morbidity and mortality rates in patients with

perforated hydatid cysts are higher than in those with nonperforated cysts, and are reported to be 10-63% for mortality, and 0-23.5% for morbidity.<sup>5-12,23,24</sup> Our morbidity rate was 42.8% with no mortality in the present series.

It is well known that, albendazole, a broad spectrum anthelmintic drug, is effective for preventing recurrence and secondary hydatidosis.<sup>6</sup> The safety and efficacy of this drug have been shown in various studies, however, there is no consensus on the duration of use of the medication for cyst sterilization.<sup>6,9</sup> Side effects of albendazole such as allergic reactions and the elevation of liver transaminase levels require cessation of treatment. In the present study, albendazole 10 mg/kg/day was used for 3 months following the surgical procedure to prevent recurrence. Allergic reaction and elevated liver function test results were noted in one patient after a month of treatment. It has been reported that the recurrence rate in perforated cysts is between 0-28.6%.<sup>6,9,17,20</sup> In our study, there were 5 cases (23.8%) of recurrent disease; external tube drainage was performed in 4 cases and omentopexy in one. Recurrent disease is usually defined as reappearance of liver cysts at the site of a previously treated cyst or presentation with new extrahepatic disease.<sup>25</sup> It usually results from spillage of hydatid fluid containing daughter cysts during the operation or insufficient surgery leaving residual vesicles in place or medical treatment after rupture of a hydatid cyst.<sup>11</sup> In endemic regions, it may be difficult to differentiate whether this cyst is a recurrence or a re infestation. Akcan et al<sup>8</sup> reported that liver cysts grow as much as 2 cm per year, and nearly 5 years is required for recurrence. Recurrences before this period are probably cysts that had been missed during exploration. Accordingly, we may speculate that recurrences we detected may arise from remnant vesicles of previous operations.

In conclusion, peritoneal rupture of the hydatid cyst increases the postoperative morbidity and morbidity rates and the incidence of recurrence. The main treatment for these patients is emergency surgery, and medical treatment should be given postoperatively. In endemic areas, rupture of hydatid cysts into the peritoneal cavity, although rare, should be included in the differential diagnosis of acute abdomen. However, our study is a retrospective chart review and includes only 21 patients and has limitations that need to be acknowledged and addressed. It is difficult to determine a treatment guideline. Because of the relative infrequency of ruptured hydatid cyst, it is obviously difficult to plan a prospective clinical study. The present results raise the need to explore in further clinical investigations and experimental trials on this topic.

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