Acute intraperitoneal rupture of hydatid cysts of the liver
Case series

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Abstract

Hydatid cyst is a parasitic infection caused mainly by Echinococcus granulosus, which is generally considered benign. However, the hepatic hydatid cyst rupture in the abdominal cavity is a life-threatening incident that requires urgent and multidisciplinary management (emergency physicians, radiologists, anesthetists, and surgeons). This study describes clinical and paraclinical liver hydatid cyst rupture in the peritoneal cavity and details the appropriate treatment.

A retrospective review of clinical records of patients hospitalized in Jendouba Hospital for liver hydatid cyst was performed over 8 years, from January 1, 2012 to December 31, 2019. Fifteen cases of liver hydatid cyst complicated with acute rupture into the abdominal cavity were collected out of 625 hydatid liver cysts. All patients underwent emergency laparotomy allowing conservative unroofing procedure associated with peritoneal lavage and external drainage combined with necessary intensive care measures. Clinical features, therapeutic procedures’ details as well as postoperative outcomes are reported. Statistical analysis was performed using the Statistical Package for the Social Sciences for Windows version 20.

There were 9 men and 6 women. Patients’ age ranged from 14 to 59 years, with an average of 38 years. Two patients were admitted with abdominal trauma. Acute abdominal pain was the most common complaint. Only 1 patient had an anaphylactic shock. Abdominal ultrasonography and computed tomography scan showed discontinuous cyst wall associated to intraperitoneal fluid in all cases. Intraoperatively, the intraperitoneal effusion was clear in 13 cases and purulent in 2. All patients underwent unroofing procedures associated with intra-operative peritoneal lavage and external drainage. The mean hospital stay was 6.11 days, and the mean follow-up was 19 months. No case of recurrence was reported among the patients.

In endemic areas, rupture of a hepatic hydatid cyst in the abdominal cavity should be considered in every case of acute abdominal pain, especially if associated with anaphylaxis signs. Early management starting in the emergency room is needed to ensure good outcome.

Abbreviation: CT = computed tomography.

Keywords: anaphylaxix, emergency, hydatid cyst, intraperitoneal, surgery

1. Introduction

Hydatid disease is a cosmopolitan zoonotic infection caused by the larval stage of Echinococcus granulosus.1,2 It affects humans as an accidental intermediate host via a fecal-oral route.1 The liver, which represents the first effective barrier to the parasite spread, represents the most frequently involved organ (75%).1,3 This disease represents a significant public health issue in Tunisia.
which remains a highly endemic area.\textsuperscript{[4]} Although commonly considered “benign”, its real morbidity is far from 0. This pathology continues to have a devastating impact on people’s health due to its potentially serious related complications, especially the rupture into the abdominal cavity.\textsuperscript{[1,4]}

The acute intraperitoneal rupture of hepatic hydatid cyst represents a life-threatening complication and its management still not codified worldwide. We aimed through this study to describe an endemic area institutional experience.

2. Methods

Our study is based on a descriptive retrospective analysis including patients hospitalized for a hepatic hydatid cyst. The data were retrieved from the register of the visceral and digestive surgery department of the Jendouba Hospital and patients’ clinical records. In this data source, we searched for all patients operated on for an acute intraperitoneal rupture of liver hydatid cyst between January 1, 2012 to December 31, 2019.

Fifteen cases of acute intraperitoneal rupture were collected out of 625 cases of liver hydatid cysts diagnosed and surgically managed in this period in our department. We excluded from our study: other liver hydatid cyst complications, cyst rupture into an organ or space other than the abdominal cavity, and cases of peritoneal hydatidosis, which witnessed an old and unnoticed rupture in the peritoneal cavity. The flowchart of patients’ enrollment is presented in Figure 1. Each patient enrolled in this study was assigned a file recording his demographic data, clinical details, results of biological and radiological examinations including cyst location, number, size, Gharbi classification, rupture signs, and therapeutic approach used, as well as morbidity, mortality, hospital stay and follow up visits’ clinical findings. During the preoperative evaluation, all patients underwent routine blood tests, chest X-ray, abdominal ultrasound, and an abdominal computed tomography (CT) scan. The treatment protocol included necessary intensive care measures (vigorous resuscitation, monitoring, intravenous analgesics, and antibiotic therapy) combined with an emergency laparotomy surgery based on the unroofing procedure. The unroofing technique involves the removal of the cyst contents including laminated membrane, germinal layer, daughter cyst, hydatid fluid, and scolices without removal of the pericyst.\textsuperscript{[5]} The residual cavity was then irrigated with a hypertonic saline solution for 10 minute and then managed with external drainage or omentoplasty. A peritoneal lavage using a hypertonic saline solution associated with another external drainage was performed.

![Flowchart of patients’ enrollment.](https://example.com/flowchart.png)
Albendazole therapy was given to all patients postoperatively. A minimal standard total follow-up period of 3 years was assigned to all patients. Initially, a follow-up visit was scheduled twice a month for 3 months followed by a visit every 3 months thereafter. During these visits, details about medical history, physical examination (searching for fever, jaundice, abdominal pain, abdominal distension) ultrasonography findings, and indirect hemagglutination serology test results were noted to search for any other case of residual cavity infection, recurrence, or secondary peritoneal hydatidosis.

Statistical analysis was performed using the Statistical Package for the Social Sciences for Windows version 20.

Ethical approval for the study was obtained from the Committee for Medical Ethics at the Jendouba Regional Hospital under CE.HRJ. 86/20. The study is reported in line with the STROBE guidelines.[6]

3. Results

Among the 15 patients, 9 were men and 6 were women. The median age was 38 years (ranging from 14 to 59 years). Three patients had a past medical history of hydatid cysts. Two patients (13.33%) were abdominal trauma victims. All patients had acute abdominal pain, 12 patients had generalized abdominal pain, and 3 had localized pain in the right upper abdominal quadrant. Five patients (33.33%) reported vomiting. One patient had hemodynamic instability with hypotension (blood pressure was 80/40 mm Hg), tachycardia (heart rate was 118 bps), polyplegia, and cutaneous allergic manifestations such as urticaria and pruritus. Clinical findings are summarized in Table 1. Blood tests showed biological inflammatory syndrome in 10 patients, higher than 70% prothrombin ratio in 100% of cases, and biological cholestasis in 3 cases.

A prothrombin ratio higher than 70% was found in all cases. Three patients had biological cholestasis, and 1 patient had an acute kidney injury with a blood urea level of around 19 mmol/l. All patients underwent a chest X-ray, and none revealed an associated lung hydatid cyst. Abdominal ultrasonography detected liver hydatid cysts associated with a small volume of intra-peritoneal free effusion in 12 cases and a moderate volume in 3 cases. According to the Gharbi classification (Table 1), there were 4 type I-cysts (26.66%), 6 type II-cysts (40%), and 5 type III-cysts (33.33%). The size of the ruptured cysts varied from 7 cm to 16 cm. 73.33% of the cysts were around 7 to 10 cm in size and 26.66% of them reached a size of 10 to 16 cm. We noted a common bile duct dilatation in 2 patients. An abdominal CT scan was performed in all cases and it revealed 12 ruptured hydatid cysts located in the right liver lobe (80%) and 3 ruptured hydatid cysts located in the left lobe (20%). All ruptured cysts had a protruding dome. Seven hydatid cysts spread to 2 or more liver segments. The CT scan showed cyst-wall discontinuity with intra-peritoneal fluid in all cases (Fig. 2). In all patients the rupture concerned only 1 hydatid cyst. Radiological data are summarized in Table 2.

All patients underwent urgent laparotomy using a large midline incision. The intra-abdominal fluid was clear “spring water” in 13 patients and purulent in 2 cases. The intra-peritoneal effusion sample was sent to the laboratory for gram stain and culture. All patients were treated with the unroofing procedure associated with cystic cavity irrigation using 30% hypertonic saline, a large-volume peritoneal lavage using a hypertonic saline solution, and external drainage. An intraoperative cholangiogram was performed in 4 patients who had a cyst with a diameter exceeding 10 cm. It showed a significant cysto-biliary fistula (>5 mm diameter) in 2 cases and a minor cysto-biliary communication of less than 5 mm diameter in 1 case. There was no hydatid material found in choledochotomy.

### Table 1

| Clinical findings incidence among patients. | Frequency | Percentages |
|------------------------------------------|-----------|-------------|
| Fever                                    | 11        | 73.33       |
| Vomiting                                 | 5         | 33.33       |
| Tenderness in the right upper quadrant    | 10        | 66.66       |
| Generalized abdominal tenderness         | 5         | 33.33       |
| Cutaneous allergic reaction               | 1         | 6.66        |
| Jaundice                                 | 0         | 0           |
| Hemodynamic instability                  | 1         | 6.66        |

CT = computed tomography.

### Table 2

| Radiological findings incidence among patients. | Frequency | Percentages |
|-----------------------------------------------|-----------|-------------|
| Abdominal ultrasound findings                 |           |             |
| • Greater diameter                            |           |             |
| 7 cm < dia < 10 cm                            | 11        | 73.33       |
| ≥10 cm                                        | 4         | 26.66       |
| • Gharbi classification                       |           |             |
| Type 1                                        | 4         | 26.66       |
| Type 2                                        | 6         | 40          |
| Type 3                                        | 5         | 33.33       |
| Type 4                                        | 0         | 0           |
| Abdominal CT scan findings                    |           |             |
| • Total number of hydatid cysts               |           |             |
| 01 hydatid cyst only                          | 6         | 40          |
| 02 hydatid cysts                              | 4         | 26.66       |
| ≥03 hydatid cysts                             | 5         | 33.33       |
| • Site of ruptured hydatid cyst               |           |             |
| Liver right lobe                              | 12        | 80          |
| Liver left lobe                               | 3         | 20          |
| • Site of hydatid cysts                       |           |             |
| Liver right lobe only                         | 10        | 66.66       |
| Liver left lobe only                          | 3         | 20          |
| Liver both lobes                              | 2         | 13.33       |

CT = computed tomography.

Figure 2. CT scan axial (A: view showing intraperitoneal fluid. B: view showing a hepatic hydatid cyst with a discontinuity in the cyst wall signing the rupture). CT = computed tomography.
major cysto-biliary fistulas were treated with internal drainage, and the minor fistula (<5 mm) was sutured.

Postoperatively, patients were closely monitored. Parameters assessed were level of consciousness, body temperature, respiratory rate, oxygen saturation, pulse rate, systolic blood pressure, diuresis, drainage fluid characters, and abdominal pain score. Only 3 patients (20%) had postoperative complications (1 residual cavity abscess, 1 wound infection, and 1 sub-segmental nonfatal pulmonary embolism). The postoperative course was uneventful in 12 cases. The residual cavity abscess was diagnosed on postoperative day 4 in 1 patient who had initially been surgically treated for a ruptured cyst of 9 cm in size located in the liver right lobe with a non-declivitous residual cavity. The presenting symptoms of abscess formation were fever, vomiting, worsening of the upper quadrant abdominal pain associated with elevated white blood cell count and C-reactive protein level. An abdominal CT scan confirmed the diagnosis and an emergency CT-guided percutaneous drainage associated with intravenous antibiotic therapy allowed a good outcome. As for the wound infection case, it was successfully treated with appropriate local wound care associated with oral antibiotics. Intra-operative findings and postoperative complications are summarized in Table 3. The patient hospital stay ranged from 5 to 9 days, with an average of 6.11 days. It was longer for the group of the 3 patients who had postoperative complications and they were discharged on postoperative day 9. There was no postoperative mortality case reported. After surgery, all patients were treated with Albendazole 15 mg/kg/d for 12 months. All patients were followed up for a median period of 19.13 months (5-36 months). Only 60% of them (n = 9) continued to show up until the 18th-month visit. No recurrence or other complication was encountered during follow-up.

4. Discussion

Hydatid cyst disease is caused by E. granulosus and less frequently with Echinococcus multilocularis. Tunis is an endemic country of liver hydatid cyst. Although commonly considered “benign”, this disease continues to have a devastating impact on people’s health due to its potentially serious complications. The most severe complication remains the rupture. Three types of ruptures were described: “contained rupture” when the ruptured cyst remains surrounded and contained by the adjacent hepatic parenchyma, “communicating rupture” with bile or vascular duct, and the “direct or free rupture” into the peritoneal cavity. Intraperitoneal cystic rupture is a rare complication of hydatid disease and occurs in 1% to 16% of reported cases. This result corroborates with the current series with a prevalence estimated at 2.4%. Intraperitoneal cyst rupture can take place spontaneously and it may be explained by intra-cystic pressure increase. It may also occur during intra-operative manipulation due to an accidental injury or by an abdominal trauma. Three factors were incriminated in intraperitoneal cyst rupture: the young age of patients, as shown in this series (mean age 38 years), the increase in cyst diameter above 10 cm, and the superficial location unprotected by liver tissue. There are 2 distinct forms of Hydatid cyst rupture, the small cracking, which is the most common form, usually induced by minimal neglected trauma, and the large authentic rupture, which is often caused by severe abdominal trauma. The minor cracking may cause a cutaneous allergic reaction in 16% to 25% of cases, and the small hydatid liquid spilled in the peritoneal cavity may either encyst or evolve to a military hydatid form. The large rupture is rare and may cause either an acute form leading to immediate fatal anaphylaxis shock in approximately 1.4% of cases or a severe allergic reaction such as dyspnea, syncope, and circulatory collapse in 1% to 12.5% of cases. In our study, severe allergic reactions were reported in 6.6% of cases.

The clinical features vary greatly, from an utterly asymptomatic form discovered late after secondary peritoneal hydatidosis to the life-threatening authentic anaphylactic shock. Either way, the nonspecific clinical presentation should not cause a diagnostic delay, which may be a risk factor of a poor prognosis. The most frequent symptom is a sharp pain located in the right upper abdominal quadrant associated with nausea and vomiting. However, peritoneal symptoms may be the first developed and can be more severe if bile leakage occurs or if the cyst is infected. Although clinical examination typically reveals tenderness in the right upper abdominal quadrant, diffuse abdominal tenderness may be found. In the current series, 5 patients had diffuse abdominal tenderness.

Ultrasongraphy is the examination of choice. It allows detection of the cysts and precise staging according to Gharbi classification with a sensitivity of 85%. It also allows suspecting cyst rupture by showing a floating membrane with intraperitoneal fluid. However, a CT scan with a sensitivity of 100% and high-resolution multiplanar reconstruction images remains the most powerful tool that provides an accurate assessment of the cystlocation, vascular and biliary connections and detects any other concomitant cysts in the abdomen. It confirms rupture by showing a collapsed cyst wall with a reduced cyst size compared to previous CT findings. It also shows a detached membrane, a wall discontinuity, or daughter cysts and fluid in the peritoneal cavity. In our series, the CT scan allowed establishing the diagnosis in all cases. However, its main constraining factor remains the patient’s hemodynamic stability. The patient’s prompt management must begin in the emergency room. Close monitoring associated with compulsory intensive care measures, the use of vasoactive drugs if needed, antihistamine medication, and corticosteroids are the first rescue...
measures to be taken in the emergency room. Urgent surgery is the basis of treatment. It has 2 basic goals: treating the primary liver hydatid cyst, and correcting its complication to prevent local and peritoneal recurrences. The primary liver cyst’s surgical approaches can be divided into the unroofing procedure, a conservative modality, and radical methods that include pericystectomy, pericystoresection, and hepatectomy. In the emergency context, the conservative modality seems to be the method of choice since it does not require high surgical skills, and is associated with low bleeding risk and shorter operative time. Moreover, the unroofing procedure prevents the morbidity of invasive methods that would be less tolerable, especially by a patient already weakened by anaphylaxis. Our results favor this technique with a 0 mortality rate and low specific postoperative morbidity (6.66%). The large volume intraoperative lavage is the second step of surgical treatment. Hypertonic saline solution (3%-30%) is the most widely used solution for this purpose. Some authors choose not to use hypertonic saline solution due to its possible complications, such as hypernatremia. The scleroidal solution was safely used in all cases in our study. Other experimental studies even advocate the use of povidone-iodine as a solution with a better scleroidal potential. This surgical step aims to end the contact between the peritoneum and hydatid antigens which is the cause of the allergic reaction. This step aims also to remove all cyst contents, especially the protoscolex, source of late disseminated recurrence. In case of large cysts greater than 10.5 cm or with bilious content or in the presence of clinical (cholestatic jaundice), biological (biological cholestasis) or radiological (bile ducts dilatation) signs of associated rupture into the bile ducts, intraoperative cholangiogram is indicated, and fistula treatment is required. Anthihelmintic treatment based on albendazole (15mg/kg/d) should be initiated as soon as possible after surgery. Bacteriological and parasitological analysis of the cystic content is mandatory. There is no consensus on the anthihelmintic treatment’s duration. In our center, the medical habit of maintaining Albendazole treatment for at least 12 months was respected in all patients. The 0 rates of secondary peritoneal hydatidosis observed in our series confirm the efficiency of this approach. Patients with treated hydatid cysts should have appropriate follow-up based on hydatid serology (indirect hemagglutination test) associated with abdominal ultrasonography every 3 to 6 months. This follow-up protocol allows early detection of recurrence correlated to insufficient peritoneal lavage or medical treatment. The CT scan may be requested only in doubtful cases to reduce patients’ exposure to radiation. Beyrouti et al reported a recurrence rate of 6.7%, and the higher rate at 14% was reported by Sosuer et al. In the present series, no recurrence case was detected with a median follow-up period of 19 months. The main limitations of the present study are the fact it was an observational study that was retrospectively conducted in a single center.

Although rare, the rupture of a liver hydatid cyst of the liver should be considered as a differential diagnosis in every case of acute abdominal pain in an endemic area, particularly if associated with anaphylaxis signs. It is a real emergency that can be life-threatening. Close collaboration between emergency physicians, anesthetists, and surgeons is the cornerstone of the right management. Emergency surgery is undeniably mandatory. It aims to treat the primary cyst and to repair complication damages. In these emergency circumstances, it would be more reasonable to go for a conservative technique. The best treatment remains, above all, the prevention of hydatid infestation through strict compliance with hygiene measures and early surgical management of every operable hydatid cyst to avoid tragedies.

**Author contributions**

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