

Hydatid Cysts of the Liver Rupture into the Peritoneal Cavity: Fifteen Cases and Review of the Literature

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Abstract

Background

Hydatid cyst is a parasitic infection caused mainly by *E. 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 physician, radiologists, anesthetists, and surgeons). This study describes clinical and paraclinical liver hydatid cyst rupture in the peritoneal cavity and details the appropriate treatment.

Materials and Methods

Fifteen liver hydatid cyst cases ruptured into the abdominal cavity that underwent urgent surgery were collected over eight years.

Results

There were nine men and six women. Patients' age ranged from 14 to 59 years, with an age average of 38 years. Two patients were admitted with abdominal trauma, and acute abdominal pain was the common consultation's reason. Only one patient had an anaphylactic shock. Both abdominal ultrasound and CT scan showed discontinuous cyst wall and intraperitoneal fluid in 100% of cases. Intraoperatively, the intraperitoneal effusion was clear in thirteen cases and purulent in two. All patients underwent unroofing procedure 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.

Conclusion

Rupture of a hydatid cyst in the abdominal cavity should be evoked in front of acute abdominal pain, especially in endemic areas, and in the presence of an allergic reaction or anaphylactic shock signs. Combined surgical and medical care starting in the emergency room is the only guarantee of a good outcome.

Introduction

Hydatid disease is a cosmopolitan zoonotic infection caused by the larval stage of *Echinococcus Granulosus*. It affects humans as an accidental intermediate host via a fecal-oral route. The liver, which represents the first effective barrier to the parasite spread, represents the most frequently involved organ (75%). This disease represents a significant public health issue in Tunisia, which remains a highly-endemic area. Although commonly considered "benign," its real morbidity is far from zero. 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.

This study aims to describe various clinical and paraclinical features of a ruptured liver hydatid cyst in the peritoneal cavity and detail the appropriate treatment.

Methods

Our study is based on a descriptive retrospective analysis performed over eight years, from January 2012 to December 2019, including patients hospitalized for a hepatic hydatid cyst. We used as a data source: the register of the visceral and digestive surgery department of the Jendouba Hospital and patients' clinical records. Fifteen cases of acute intraperitoneal rupture were collected out of 625 hydatid liver cysts. We excluded from our study: other liver hydatid cyst complications, cyst rupture in 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. Each patient enrolled in this study assign a file recording his clinical details, results of biological and radiological examinations and therapeutic procedures used, as well as possible post-operative complications. All patients underwent urgent surgical intervention that focused on treating the primary cyst and its complication, combined with necessary intensive care measures.

Ethical approval for the study was obtained from the Committee for Medical Ethics at the Jendouba Regional Hospital under CE.HRJ. 86/20.

Results

Among the fifteen patients were nine men and six 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, twelve patients had generalized abdominal pain, and three had localized pain in the right upper abdominal quadrant. Five patients (33.33%) reported vomiting. A single patient had hemodynamic instability with hypotension (Blood pressure was 80/40 mmHg), tachycardia (heart rate was 118 bps), polypnea, and cutaneous allergic manifestations such as urticaria and pruritus. Clinical findings are summarized in Table 1. Blood tests showed biological inflammatory syndrome in ten patients, a higher than 70% Prothrombin ratio in 100% of cases, and biological cholestasis in three cases. One patient had a functional renal failure with a blood urea level of around 19 mmol/ l. All patients underwent a chest X-ray, and none reveals a lung hydatid cyst. Abdominal ultrasound detected liver hydatid cysts associated with a small intraperitoneal free effusion in 12 cases and moderate volume in other cases. According to the Gharbi classification (Table 1), there were four types I cysts (26.66%), 6 type II cysts (40%), and five type III cysts (33.33%). The size of the cysts varied from 7 cm to 16 cm. We note a common bile duct dilatation in two patients. An abdominal CT scan was performed on all patients and revealed ruptured hydatid cysts; 12 lesions were in the right liver lobe (80%) and three in the left lobe (20%). All ruptured cysts had a protruding dome, and seven hydatid cysts spread to two or more liver segments. The CT scan showed cyst-wall discontinuity with intraperitoneal fluid and the rupture site in all cases (Fig. 1). Radiological data are summarized in Table 2.

Table 1
clinical findings incidence among patients

All patients, n = 15		
	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

Table 2
radiological findings incidence among patients

All patients, n = 15		
	Frequency	Percentages
Abdominal ultrasound findings		
Greater diameter		
7cm < \varnothing < 10 cm	11	73.33
\varnothing > 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
\geq 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

All patients underwent urgent midline surgery. The intra-abdominal fluid was clear "spring water" in thirteen patients and purulent in two cases. The intra-peritoneal effusion specimen was sent to the laboratory for gram stain and culture. All patients were treated with an unroofing procedure associated with cystic cavity's irrigation using 30% hypertonic saline, a large-volume peritoneal lavage using a hypertonic saline solution, and external drainage. An intraoperative cholangiogram was performed in four patients whose cyst size exceeded 10 cm. It showed a significant cysto-biliary fistula (> 5mm diameter) in

two cases and a minor cysto-biliary communication of less than 5mm diameter in one case. There was no hydatid material found in Choledochotomy. The major cysto-biliary fistulas were treated with internal drainage, and the minor fistula (< 5mm) was sutured. Only three patients had post-operative complications (01 residual cavity abscess, 01 wound infection, and 01 pulmonary embolisms). Intra-operative findings and post-operative complications are summarized in Table 3. The patient hospital stay ranged from 5 to 9 days, with an average of 6.11 days. After surgery, all patients were treated with albendazole 15 mg/kg/day for 12 months. No secondary peritoneal hydatidosis cases were observed during the median follow-up period of 19 months (5 to 36 months).

Table 3
Intraoperative and post-operative clinical features

All patients, n = 15		
	Frequency	Percentages
Intraoperative findings		
Intraperitoneal fluid characteristics		
<i>Clear</i>	13	86.66
<i>Purulent</i>	2	13.33
Intraperitoneal fluid location		
<i>Supra-colic compartment only</i>	10	66.66
<i>Supracolic compartment + pelvis</i>	5	33.33
Cysto-biliary communication		
<i>Major fistula > 5mm diameter</i>	2	13.33
<i>Minor fistula < 5mm diameter</i>	1	6.66
Postoperative Complications		
Specific complications		
<i>Residual cavity abscess</i>	1	6.66
non-specific complications		
<i>Surgical wound infection</i>	1	6.66
<i>Pulmonary embolism</i>	1	6.66

Discussion

Hydatid cyst disease is caused by *E. Granulosus* and less frequently with *E. Multilocularis* (1, 2). Tunisia is an endemic country of liver hydatid cyst (1). Although commonly considered "benign," this disease

continues to have a devastating impact on people's health due to its potentially serious complications, especially 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" in the peritoneal cavity (3, 4). Intraperitoneal Cystic rupture is a rare complication of hydatid disease and occurs in 1–16% of reported cases (2, 5, 6). This result corroborates with the current series with a prevalence estimated at 2.4%. Intraperitoneal cyst rupture can take place spontaneously and may be explained by intra-cystic pressure increase (7). It may also occur during intra-operative manipulation due to an accidental injury or by an abdominal trauma (8, 9). 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 (4, 9, 10). There are two 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 (7). The minor cracking may cause a cutaneous allergic reaction in 16–25% 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 (9) or a severe allergic reaction such as dyspnea, syncope, and circulatory collapse in 1–12.5% of cases (11). In our study, severe allergic reactions were reported in 6,6%.

The clinical features vary greatly, from an utterly asymptomatic form discovered late after secondary peritoneal hydatidosis to the authentic anaphylactic shock, a patient life-threatening (8). Either way, the nonspecific clinical presentation should not cause a diagnosis delay, which may be a risk factor for a poor prognosis (3, 12). The most frequent symptom is a sharp pain in the right upper abdominal quadrant associated with nausea and vomiting (9). However, peritoneal symptoms may be the first developed and can be more severe if bile leakage occurs or the cyst is infected (13, 14). Clinical examination typically reveals tenderness in the right upper abdominal quadrant, and diffuse abdominal tenderness may be objectified. In the current series, five patients had diffuse abdominal tenderness.

Ultrasound is the examination of choice (1). It allows detection of the cysts and precise staging according to Gharbi classification (15) with a sensitivity of 85% (9). It also allows suspecting cyst rupture by showing a floating membrane with intraperitoneal fluid (7, 10). However, CT with a sensitivity of 100% and high-resolution multiplanar reconstruction images is a powerful tool that provides an accurate assessment of the cyst, location, vascular and biliary connexions and detects any other concomitant cysts in the abdomen (1, 3, 13). It confirms rupture by showing a collapsed cyst wall with a reduced cyst size compared to old CT findings, a detached membrane, a wall discontinuity, or daughter cysts and fluid in the peritoneal cavity (7). 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 (8). Urgent surgery is

the basis of treatment. It has two basic goals: First, to treat the primary liver hydatid cyst, and secondary, correct its complication to prevent local and peritoneal recurrences (4, 16). Approaches of the primary liver cyst's surgical treatment can be divided into the unroofing procedure, a conservative modality, and radical methods that include pericystectomy, pericystoresection, and hepatectomy (1). In the emergency context, conservative modality seems to be the choice method since it did not require high surgical skills, it is associated with low bleeding risk and shortens the operative time (14, 17). Moreover, the unroofing prevent the morbidity of more invasive methods that would be less tolerable, especially by a patient already weakened by anaphylaxis. Our results favor this technique with a zero mortality rate and low specific post-operative morbidity (6.66%). The large volume intraoperative lavage is the second step of surgical treatment (5). Hypertonic saline solution (3%-30%) is the widely used solution for this purpose (10, 16, 17). Some authors choose not to use hypertonic saline solution due to its possible complications, such as hypernatremia (1, 18). The scolicedal 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 scoliced potential (5, 16). This surgical step aims to end the peritoneum and hydatid antigens contact, causing the allergic reaction and removing all cyst contents, especially the protoscolex, source of late disseminated recurrence (4, 8, 9). 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 (19–21). Anthelmintic treatment based on albendazole (15 mg/kg/day) should be initiated as soon as possible after surgery (8). There is no consensus on the treatment's duration. Besides, several experimental studies have demonstrated the superiority of the combination of chemotherapy and cytokines over albendazole alone (22). In our center, a medical habit of maintaining Albendazole treatment for at least 12 months was respected in all patients. The effectiveness of this procedure is confirmed by none secondary peritoneal hydatidosis was observed in our series. Considering recurrence risk, correlated to insufficient peritoneal lavage or medical treatment (6), patients with treated hydatid cysts should have appropriate follow up based on hydatid serology (indirect hemagglutination test) associated with abdominal ultrasound every 3 to 6 months. We estimate that those two methods allow recurrence early checking and treatment. The CT scan may be a part of the subsequent investigations in doubtful cases only 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. (1). In the present series, no recurrence case was detected with a median follow-up period of 19 months.

Conclusion

Although rare, the rupture of a hydatid cyst of the liver should be considered a differential diagnosis in every case of acute abdominal pain in an endemic area, significantly if associated with allergic reactions or signs of anaphylactic shock. 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 repair complication damage. 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 and through strict compliance with hygiene measures and the early surgical management of every operable hydatid cyst to avoid tragedies.

Abbreviations

CT
Computed tomography
E.Granulosis
Echinococcus Granulosus
E. Multilocularis
Echinococcus Multilocularis

Declarations

Ethics approval and consent to participate: An ethical approval was obtained from the Jendouba Regional Hospital Medical Ethics Committee N° CE.HRJ. 86/20: We confirm that all methods were performed by the 1975 Declaration of Helsinki's ethical guidelines.

Consent for publication: A written informed consent for the publication of personal/ clinical data has been obtained from each patient.

Availability of data and materials: There are no additional data available to share with the readers. The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Competing interest: The authors declare that they have no conflicts of interest.

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Authors' contributions: AM contributed to the work conception, JR and SS collected data, KA and MAM analyzed clinicopathological data, and AO wrote the manuscript. The final version of the manuscript was read and approved by all authors.

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Figures

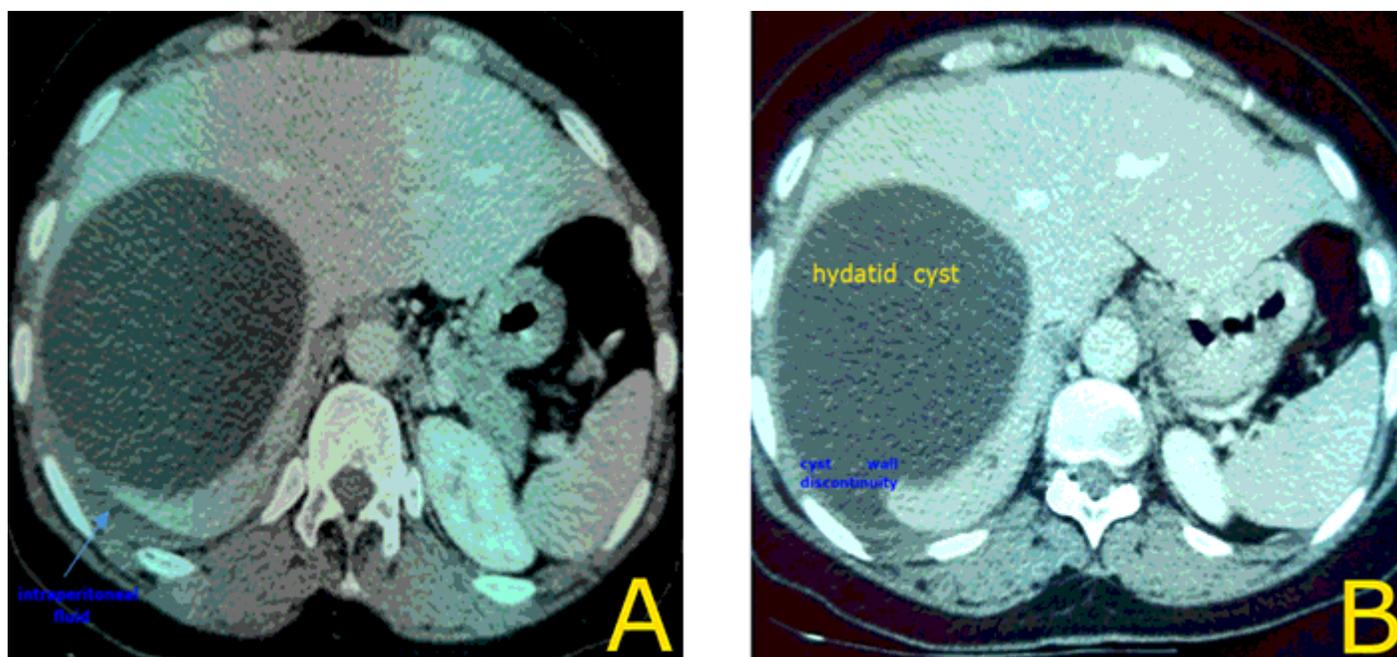


Figure 1

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)