

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 an infectious disease caused mainly by *E. Granulosus*, which is generally considered benign, however, the rupture of the hepatic Hydatid Cyst to the abdominal cavity is a life-threatening incident that requires urgent and multidisciplinary management (emergency physician, radiologists, aneashtetists and surgeons). This study aims to describe the various clinical and paraclinical features of ruptured liver hydatid cyst in the peritoneal cavity, and to detail the appropriate treatment.

Materials and Methods

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

Results

There were nine men and six women. The average age was 38 years. Two patients were admitted with abdominal trauma. All patients presented with acute abdominal pain. Only one patient had anaphylactic shock. Abdominal ultrasound showed discontinuous cyst wall and intraperitoneal fluid in 100% of cases. Abdominal computed tomography (CT) showed discontinuous cyst wall with intraperitoneal fluid in 100% of cases. Intraoperatively, the intraperitoneal fluid 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 considered as a differential diagnosis in every case of an acute abdominal pain, especially in endemic areas and in presence of an allergic reaction or signs of anaphylactic shock. Combined medical and surgical 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 human, as an accidental intermediate host via a fecal-oral route. The liver, which represents the first effective barrier to the spread of the parasite, represents the most frequently involved organ (75%). This disease represents a considerable public health issue in Tunisia, which remains a highly-endemic area. Although commonly considered "benign," its real morbidity is far from zero. Actually, this condition continues to have 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 the various clinical and paraclinical features of ruptured liver hydatid cyst in the peritoneal cavity and to detail the appropriate treatment.

Methods:

Our study is based on a descriptive retrospective analysis carried out over a period of eight years extended from January 2012 to December 2019, including patients hospitalized with hepatic hydatid cyst. We used as a data source: the register of the department of visceral and digestive surgery of the Jendouba Hospital and clinical records of patients. Fifteen cases of acute intraperitoneal rupture were collected out of 625 hydatid liver cysts. We excluded from our study: All records regarding another complication of the cyst, any rupture of the cyst in an organ or space other than the abdominal cavity and cases of peritoneal hydatidosis, which witnessed an old rupture in the peritoneal cavity that went unnoticed. Each patient enrolled in the study was assigned a file recording his clinical details, results of biological and radiological examinations, and therapeutic procedures used, as well as postoperative possible complications. All patients underwent urgent surgical intervention that focused on treating the primary cyst and its complications, combined with some necessary intensive care measures.

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

Results:

Among the fifteen patients were nine men and six women. The median age was 38 years (ranging 14 to 59 years). Three patients had a prior medical history of hydatid cysts. Two patients (13.33%) were admitted after an abdominal trauma. All patients had acute abdominal pain, twelve patients had generalized abdominal pain, while the three others had 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 manifestations such as urticaria and pruritus. Clinical findings are summarized in Table 1. Blood tests showed biological inflammatory syndrome in ten patients, Prothrombin ratio higher than 70% in 100% of cases, biological cholestasis in three cases. Functional renal failure with blood urea level around 19 mmol / l was observed in one patient. All patients underwent a chest X-ray that did not reveal a hydatid cyst of the lungs in any case. Abdominal ultrasound detected liver hydatid cysts associated with small-volume intraperitoneal free fluid in 12 cases and moderate volume in the other 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%). Dilatation of the common bile duct was noted in two patients. The size of the cysts varied from 7 cm to 16 cm. All patients underwent abdominal CT scan that showed ruptured hydatid cysts, 12 of them were located in the liver right lobe (80%) and 3 in the liver left lobe (20%). All ruptured cysts had a protruding dome and seven hydatid cysts spread to two or more liver segments. The CT scan demonstrated discontinuity in the cyst-wall with intraperitoneal fluid and showed the exact rupture site in the wall of the cyst in all cases (Fig. 1). Radiological data are summarized in Table 2. All patients

underwent urgent midline surgery. Intra-abdominal fluid was clear “spring water” in thirteen patients and purulent in two. Intra-peritoneal fluid specimen was taken in each case and sent to the laboratory for gram stain and culture. All patients were treated with an unroofing procedure associated with an irrigation of the cystic cavity using 30% hypertonic saline, a large-volume peritoneal lavage using hypertonic saline solution and an external drainage. In four patients whose cyst size exceeded 10 cm, an intraoperative cholangiogram was performed. It showed a major cysto-biliary fistula (> 5mm diameter) in two cases and a minor cysto-biliary communication of less than 5mm diameter in one patient. Choledochotomy did not find hydatid material in the common bile duct in any case. The major cysto-biliary fistulas were treated with internal drainage, and the minor fistula (< 5mm) was sutured. Only three patients had postoperative complications (01 residual cavity abscess, 01 wound infection and 01 pulmonary embolism). Intra-operative findings and postoperative complications are summarized in Table 3. The in-patient hospital stay ranged from 5 to 9 days, with an average of 6.11 days. All patients were treated with albendazole 15 mg / kg / day for 12 months after surgery. No cases of secondary peritoneal hydatidosis were observed during the median follow-up period of 19 months (5 to 36 months).

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

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 hydatid cyst of the liver (1). Although commonly considered "benign," this disease continues to have devastating impact on people's health due to its potentially serious related-complications, especially the rupture. Three types of ruptures have been described: "contained rupture" where the ruptured cyst remains surrounded and contained by the adjacent hepatic parenchyma, "communicating rupture" with a bile or vascular duct and the "direct or free rupture" which occurs to 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). These results corroborate with data in the current series showing that its prevalence is estimated at 2.4%. Intraperitoneal cyst rupture can take place spontaneously, and it may be explained in this case by an increase in the intra-cystic pressure (7). It may also be caused iatrogenically during intra-operative manipulation as a result of accidental injury, or by an abdominal

trauma (8, 9). The three leading incriminated factors in intraperitoneal cyst rupture are: The young age of the patients, as shown in this series (mean age 38 years) and it has a direct relationship with the activity (4, 9), the increase in cyst diameter above 10 cm and the superficial location unprotected by covering liver tissue (9, 10).

The clinical features vary greatly, from a completely asymptomatic form discovered late after secondary peritoneal hydatidosis to the authentic anaphylactic shock, which can immediately endanger the patient's life (8). It is often a sharp pain in the right upper abdominal quadrant associated with nausea and vomiting (9). Clinical examination typically reveals tenderness in the right upper abdominal quadrant and in a number of cases diffuse abdominal tenderness. In the current series, five patients had diffuse abdominal tenderness. Peritoneal signs and symptoms may develop earlier and can be more severe if bile leakage occurs or the cyst is infected (11, 12). Isolated cutaneous allergic symptoms, such as urticaria, macular eruption and pruritus, may occur in 16–25% of cases and indicate a minimal allergic reaction developed against the cystic content spilled into the abdominal cavity (13). The clinical presentation of intraperitoneal rupture of hydatid cyst is strongly related to the rupture size and the aspects of the cysts content (4). There are two distinct forms of Hydatid cyst rupture to the peritoneal cavity, the small fissuring which is the most common form, usually induced by a minimal neglected trauma and the large authentic rupture, which often takes place following a severe trauma (7). The minor fissuring results in cutaneous allergic reaction, the small amount of hydatid liquid spilled in the peritoneal cavity may either encyst or result in a military hydatid form. The large rupture is rare and may cause a real acute form leading to immediate fatal anaphylaxis choc (7). Anaphylactic shock incidence rate is approximately 1.4% (9). Severe allergic reactions such as dyspnoea, syncope and circulatory collapse occur in 1%-12.5% of cases (14). In our study, severe allergic reactions were reported in 6,6%. Either way, nonspecific misleading clinical presentation should not be a cause of diagnostic delay as the latter is a critical risk factor accounting for poor prognosis.

Hydatid serology associated with imaging modalities instead play a key role in establishing the diagnosis (3, 15). Ultrasound is the first examination of choice (1). It allows detection of the cysts as well as their precise staging according to Gharbi classification (16) with a sensitivity of 85% (9). It also helps to suspect the rupture of the cyst 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 more powerful tool that provides an accurate assessment of the cyst, its exact location, its vascular and biliary connexions and detects any other concomitant cysts in the abdomen (1, 3, 11). It confirms rupture by showing a collapsed cyst wall with a reduced cyst size compared to former CT findings, a detached membrane, a discontinuity in the cyst wall, or daughter cysts and fluid in the peritoneal cavity (7). In our series, CT scan allowed establishing the diagnosis in all cases. However, its main constraining factor remains the requirement of patient's hemodynamic stability.

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

should be considered. It has two basic goals: to treat the primary liver hydatid cyst and to treat its complication in order to prevent local and peritoneal recurrences (4, 13). Approaches of surgical treatment of the primary liver cyst can be divided into the unroofing procedure, which is a conservative modality and radical methods that include pericystectomy, pericystoresection and hepatectomy (1). In this emergency context, the conservative procedures seems to be the method of choice, since It does not require very high surgical skills, poses a negligible risk of bleeding, shortens the operative time (12, 17). Moreover, the unroofing prevents the morbidity of more invasive methods that would be less tolerable especially by a patients already weakened by anaphylaxis (1). The Zero mortality rate and low specific postoperative morbidity (6.66%) in our series strongly argues in favour of this technique. The second pillar of the surgical treatment is a large volume intraoperative lavage (5). Hypertonic saline solution (3%-30%) is the widely used solution in this purpose (10, 13, 17). Some authors choose not to use hypertonic saline solution due to its possible complications, such as hypernatremia (1, 18). This scolicidal 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 scolicid potential (5, 13). The aim of this surgical step is to put an end to the contact between the peritoneum and the hydatid antigens causing the allergic reaction and to remove all cyst contents especially the protoscolex, source of late disseminated recurrence (4, 8, 9). In case of large cysts greater than 10.5 cm in diameter and / or with bilious content and / or in 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 how long to continue this treatment. 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 the fact that no case of secondary peritoneal hydatidosis was observed in all patients of our series.

Considering the risk of recurrence, sometimes 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 periodically conducted every 3 to 6 months. We estimate that those two methods combined allows recurrence to be early checked and acted upon. 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 a higher rate at 14% was reported by Sosuer et al (1). In the present series, no case of recurrence was detected with a median follow up period of 19 months.

Conclusion

Although rare, rupture of a hydatid cyst of the liver should be considered as a differential diagnosis in every case of acute abdominal pain in an endemic area, especially, if it is associated with allergic reactions or signs of anaphylactic shock. It is a real emergency that can be life threatening. Close

collaboration between emergency physicians, anaesthetists and surgeons is the cornerstone of a right management. Emergency surgery is undeniably mandatory. It aims to treat the primary cyst and repair the damage caused by its complication. 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 its complications through strict compliance with hygiene measures and the early surgical management of every operable hydatid cyst in order to avoid easily avoidable tragedies.

Abbreviations

CT
computed tomography
E. Granulosis
Echinococcus Granulosis
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 in accordance with the ethical guidelines of the 1975 Declaration of Helsinki.

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 analysed 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 analysed clinicopathological data and AO wrote the manuscript. The final version of manuscript was read and approved by all authors.

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Figures

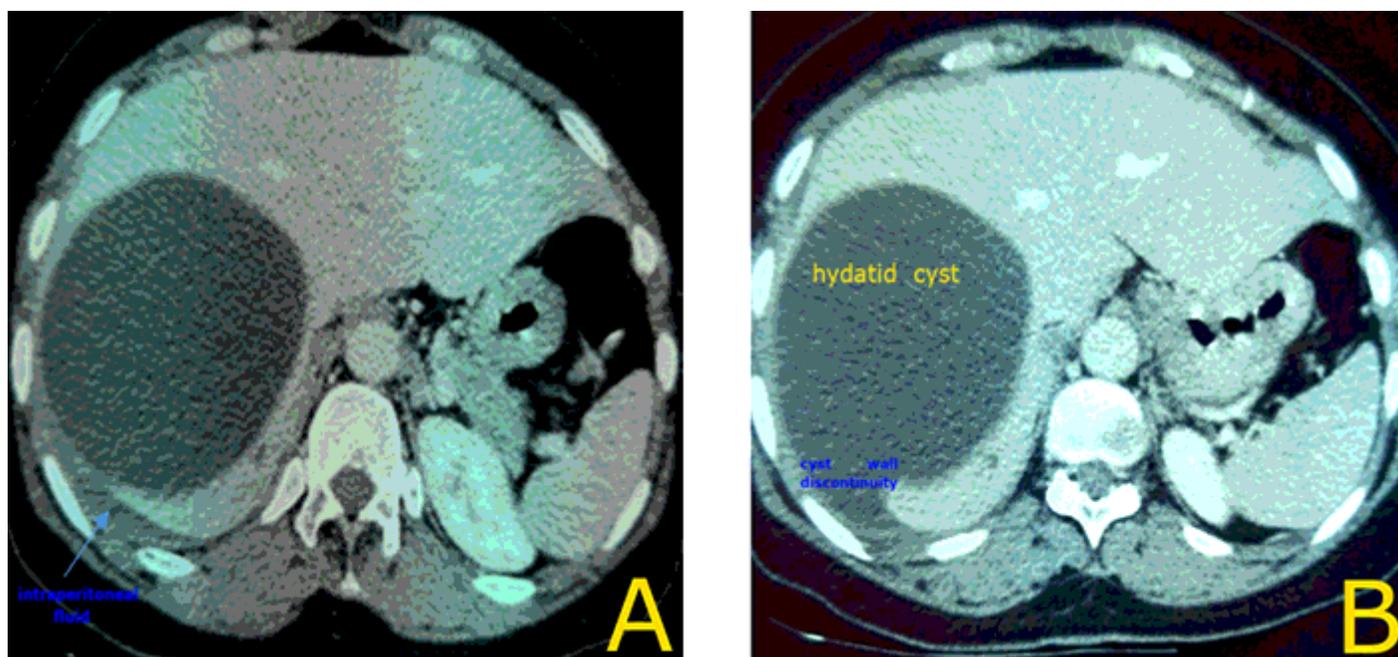


Figure 1

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)