Case Report: Spontaneous Liver Hydatid Cyst Rupture in a Child

Sumer A[a], Caglayan K MD[b], Altinli E[c], Koksal N[c]

[a]Department of General Surgery, Kas State Hospital, Antalya,
[b]Department of General Surgery, Kars State Hospital, Kars, and
[c]Second General Surgery Unit, Department of General Surgery, Haydarpasa Numune Training and Research Hospital, Istanbul, Turkey

Abstract

Hydatid disease is a parasitic infection caused by *Echinococcus granulosus*. Hydatid cysts can rupture either spontaneously or following trauma. Ruptured hydatid cyst of the liver is an infrequent clinic entity. Ultrasonography and computerized tomography are the main diagnostic tools, with 85% and 100% sensitivity, respectively. Treatment with surgery is mandatory. We report a case of spontaneous rupture of hydatid cyst of the liver in a 5-year-old boy who presented to the emergency department with a history of abdominal pain. The germinative membrane was removed from the cyst pouch, and the peritoneal cavity was lavaged with hypertonic saline solution, followed by omentoplasty. Albendazole was administered for three months postoperatively. The recovery course was uneventful. In endemic regions, ruptured hydatid cyst of the liver should be included in the differential diagnosis of abdominal pain.

MeSH Words: hydatid cyst, liver, rupture, acute abdomen

Introduction

Hydatid cyst is caused by infection of larvae of the parasite *Echinococcus granulosus*. It is endemic to Turkey, the Middle East, Far East, South America, Australia, New Zealand, China, northern Kenya, and other sheep-raising areas [1], where it constitutes an important medical and public health problem. With immigration, the prevalence of the disease has increased in developed countries [2-4]. Dogs are the definitive hosts. Humans are intermediate hosts, infected by oral ingestion of excrement from animals [5,6]. Hydatid cysts may form in any organ. The liver is most commonly involved (70% of cases) [7], followed by the lungs (10–15%), and less frequently, the spleen, kidneys, heart, bones, and central nervous system [1,2]. Primary peritoneal hydatid cyst infection is rare (2%), and its mechanism is unknown [1].

Rupture of hydatid cysts occurs infrequently. Free intraperitoneal rupture has been reported in approximately 3.2% of all cases of liver hydatid disease. Intraperitoneal cysts may rupture spontaneously, due to increased intracystic pressure, or as a consequence of trauma [5,7], leading to the spread of hydatid fluid in the intraperitoneal cavity [8,9].

The clinical signs and symptoms of hydatid cyst depend on its localization, size, and relationship to adjacent organs, and complications [1,5]. The diagnosis is based on laboratory tests, including immunology and radiological imaging, particularly ultrasonography and computed tomography (CT). Ultrasonographic classification has provided a good, widely accepted, morphologic description of the disease [3]. The treatment of hydatid cyst of the liver is complicated [10]. For ruptured cysts, surgery is mandatory.

We report a case of spontaneous rupture of a hydatid cyst of the liver in a 5-year-old boy.

Case Report

A 5-year-old boy presented to the emergency department with abdominal pain associated with abdominal distention, nausea, and vomiting. His past medical history revealed lung surgery performed 2 years previously for hydatid cyst, followed by the administration of albendazole.

On clinical examination, blood pressure was 90/60 mmHg and pulse rate was 110 beats/min. No signs of trauma were visible. Palpation revealed a sensitive, tender abdomen with muscular defense and rigidity. Axillary temperature was 38.2^{o}C, and rectal temperature, 38.8^{o} C. On blood analysis, hemoglobin measured 12.6 g/dl, hematocrit 38.2%, and white blood cell count, 15200 cells/mm^{3}. Urine analysis revealed no abnormalities. Findings on plain X- ray of the abdomen and chest were inconclusive.

Abdominal CT scan demonstrated a 60x60 mm cyst in the right lobe of the liver and a detached germinative membrane, in addition to free fluid in the subhepatic area (Figure 1). Because signs of acute abdomen were present, laparotomy was performed. A right subcostal approach was used on the basis of the CT findings. Exploration revealed approximately 200 cc of hydatid fluid in the subhepatic area and a ruptured hydatid cyst in the fifth segment of the liver (Figure 2). The germinative membrane was removed from the cyst pouch (Figure 3), and the pouch and peritoneal cavity were irrigated with 3% hypertonic saline for 10 to 15 minutes, followed by omentoplasty. Postoperatively, treatment with albendazole 10mg/kg/day (Andazol; Biofarma, Istanbul, Turkey) was continued for 3 months. The course of recovery was uneventful.

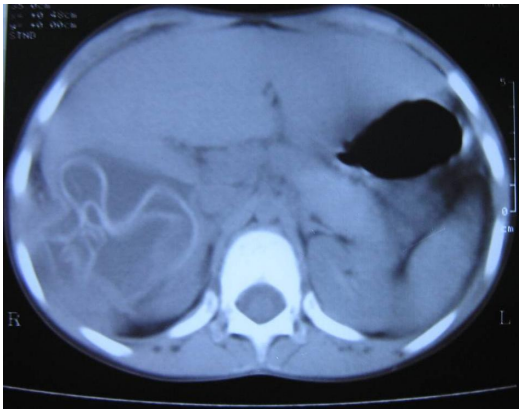

Figure 1: Computed tomographic scan shows the ruptured hydatid cyst.

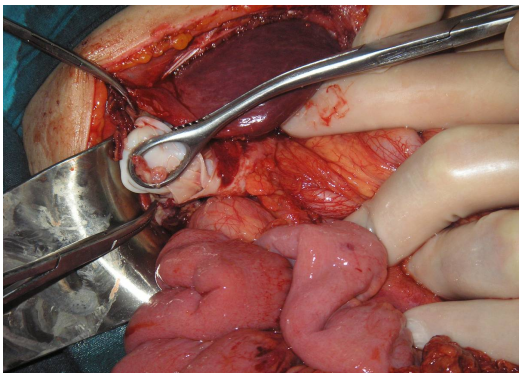

Figure 2: Panoramic view of the ruptured HC at operation.

Figure 3: Removed germinative membrane.

Discussion

Most patients with hydatid cysts are asymptomatic, and the diagnosis is usually made

incidentally during clinical or radiological examination for unrelated reasons [11].

Intrabiliary rupture is the most common complication of hydatid cyst, with an incidence of 5% to 25% of patients [7]. Rupture into the peritoneum occurs less often [7,11], with reported rates ranging from 3.2% to 16% [5,12,13]. Akcan et al. [10] noted a 7.8% rate of peritoneal rupture of hydatid cysts in Turkey, where the disease is endemic. Trauma due to traffic accidents, which occur at a high rate in Turkey, was the leading cause [10]. Other studies implicated mainly trauma due to falls [14] and sports activities [7]. Sozuer et al. [1] noted a 19% rate of blunt abdominal trauma. Our patient, however, had no history of trauma prior to admission.

Significant risk factors for hydatid cyst perforation include younger age, cyst diameter of >10 cm, and superficial cyst location [10]. Milicevic [15] found that developing cysts increase by about 1 to 1.5 mm in diameter per month. Our patient had already undergone surgery for a lung hydatid cysts at age 3 years, and the size of the present hepatic cyst was 60x60 mm.

Intraperitoneal rupture of a hydatid cyst is considered an urgent clinical event. The dissemination of the cyst contents poses a 1.0-2.5% risk of severe, life-threatening anaphylactic reaction [9,10].

The most common complaints associated with hydatid cyst of the liver are abdominal pain, nausea, vomiting, and jaundice [1,5]. The signs and symptoms of ruptured cyst are nonspecific. Our patient presented with acute abdomen, which is unusual in this setting [9].

Ultrasound is considered the first diagnostic option in the examination of patients with suspected uncomplicated hydatid cyst of the abdomen. Its reported sensitivity is 85%. CT is currently the most sensitive tool for detection of hydatid cyst rupture in the liver and spleen, with 100% sensitivity [9,10].

Hydatid cyst may be treated by medical means or percutaneous drainage. The combined use of these methods is on the rise. However, surgery remains the main treatment modality, either radical (total pericystectomy with liver resection) or conservative (unroofing and omentoplasty). The choice of operative approach depends on the experience of the surgeon and the localization of the cyst. Although laparoscopy has been performed for hydatid cysts of the liver in most major surgical centers [16], we found no reports of its application in cases of perforated cyst [1,8,16]. In general, the specific management of patients with peritoneal perforation has not been evaluated sufficiently, and no clear guidelines are available [10].

Lavage of the peritoneal cavity with scolicidal agents is well accepted, although the risk of inducing sclerotizing cholangitis should be borne in mind [8,9]. Koyncu et al. [9] reported that povidone iodine was more effective than hypertonic solution, but experimental studies have found it to have high toxicity [1]. In complicated cases, profuse peritoneal lavage with hypertonic solution is indicted to limit recurrence [10]. In the present patient, we used the 3% hypertonic saline solution for lavage of the peritoneal cavity.

Albendazole has proven prophylactic activity in perforated hydatid cysts [1]. Its safety and efficacy for 2-3 months of postoperative use is well recognized. Our patient was treated with albendazole 10mg/kg/day for 3 months postoperatively.

Hydatid cysts are associated with 12%-63% morbidity. The recurrence rate after open surgery for hydatid cyst of the liver ranges from 3% to 10% [8], and for perforated hydatid cyst of the liver from 0 to 13% [10]. The reported operative mortality rate of perforated liver hydatid cyst is 0 to 11.8% [10].

Although rupture of a hydatid cyst of the liver is an uncommon clinical entity, it may be fatal. Therefore, in endemic regions, clinicians should maintain a high index of suspicion in patients who present with abdominal pain.

References

1. Sozuer EM, Ok E, Arslan M. The perforation problem in hydatid disease. Am J Trop Med Hyg. 2002;66:575-577.

2. Khuroo MS, Wani NA, Javid G, Khan BA, Yattoo GN, Shah AH, Jeelani SG. Percutaneous drainage compared with

surgery for hepatic hydatid cysts. N Engl J Med. 1997;337:881-887.

3. Goktay AY, Secil M, Gulcu A, Hosgor M, Karaca I, Olguner M, et al. Percutaneous treatment of hydatid liver cysts in children as a primary treatment: long-term results. J Vasc Interv Radiol. 2005;16:831-839.

4. Safioleas MC, Misiakos EP, Kouvaraki M, Stamatakos MK, Manti CP, Felekouras ES. Hydatid disease of the liver: a continuing surgical problem. Arch Surg. 2006;41:1101-1108.

5. 5.Ozturk G, Aydinli B, Yildirgan MI, Basoglu M, Atamanalp SS, Polat KY, et al: Posttraumatic free intraperitoneal rupture of liver cystic echinococcosis: a case series and review of literature. Am J Surg. 2007;194:313-316.

6. Sielaff TD, Curley SA. Liver. In Brunicardi FC, Andersen DK, Billiar TR, Dunn DL, Junter JG, Pollock RE, eds. Schwartz's Principles of Surgery, eighth edition. New York: Mc Graw-Hill. 2005;1139-1186.

7. Gulalp B, Koseoglu Z, Toprak N, Satar S, Sebe A, Gokel Y, et al. Ruptured hydatid cyst following minimal trauma and few signs on presentation. Neth J Med. 2007;65:117-118.

8. Ertem M, Karahasanoglu T, Yavuz N, Erguney S: Laparoscopically treated liver hydatid cysts. Arch Surg. 2002;137:1170-1173.

9. Koyuncu A, Aydin C, Turan M, Taş F, Gökgöz S, Sen M: Traumatic pelvic hydatid cyst rupture: report of a case. Ulus Travma Acil Cerrahi Derg. 2003;9:212-214.

10. Akcan A, Akyildiz H, Artis T, Ozturk A, Deneme MA, Ok E, Sozuer E. Peritoneal perforation of liver hydatid cysts: clinical presentation, predisposing factors, and surgical outcome. World J Surg. 2007; 31:1284-1291.

11. Lo Casto A, Salerno S, Grisanti M, Mastrandrea G: Hydatid cyst of the liver communicating with the left colon. Br J Radiol. 1997;70:650-651.

12. Prousalidis J, Tzardinoglou K, Sgouradis L, Katsohis C, Aletras H. Uncommon sites of hydatid disease. World J Surg. 1998;22:17-22.

13. Karavias DD, Vagianos CE, Kakkos SK, Panagopoulos CM, Androulakis JA. Peritoneal echinococcosis. World J Surg. 1996;20:337–340.

14. Gunay K, Taviloglu K, Berber E, et al. Traumatic rupture of hydatid cysts: a 12-year experience from an endemic region. J Trauma. 1999;46:164-167.

15. Milicevic M. Echinococcal cysts: Cause, diagnosis, complications, and medical and surgical treatment. In: Fischer EJ, ed. Mastery of Surgery, fifth edition. Philadelphia: Lippincott Williams& Wilkins. 2007;1043-1069.

16. Altinli E, Saribeyoglu K, Pekmezci S, Uras C ,Tasci H, Akcal T. An effective omentoplasty technique in laparoscopic surgery for hydatid disease of the liver. JSLS. 2002;6(4):323-6

Competing Interests: None declared.

Funding: None declared.

This manuscript has been peer reviewed

Correspondence:

Aziz Sumer, MD
General Surgeon & Instructor of Trauma Surgery
Kaş Devlet Hastanesi Başhekimlik
07580,Kaş
Antalya, Turkey.
Tel: +90 05059257142
Fax: +90 2428361604
e-mail: azizsumer2002@yahoo.com