Case Report

ANNALS OF MEDICINE & SURGERY

OPEN

Acute abdomen due to rupture of a hydatid cyst of the liver: a rare complication – a case report

Subodh Adhikari, MBBS, Madhur Bhattarai, MBBS*, Sushil Gyawali, MS, Sudhan Subedi, MS, Abhishek Bhattarai, MS, MCh, Laligen Awale, MS, MCh, Prasan B.S. Kansakar, MS, MCh

Introduction and importance: Hydatid cyst is a parasitic disease that is transmitted from animals to humans caused by the larval stage *Echinococcus*, especially *Echinococcus granulosus*. A complication of a hydatid cyst of the liver is its rupture, either traumatic or spontaneously.

Case presentation: A 19-year-old male presented with an acute abdomen for 12 h. After clinical assessment, contrast-enhanced computed tomography showed a rupture of the anterior wall of the hepatic hydatid cyst with intra-abdominal and pelvic dissemination. Exploratory laparotomy was performed with the evacuation of the daughter cyst and peritoneal lavage. The patient recovered well and was discharged with albendazole therapy.

Clinical discussion: Hydatid cyst rupture is a rare but serious complication. Computed tomography has high sensitivity in demonstrating cyst rupture. The patient underwent laparotomy, where disseminated cysts were evacuated, and the anterior wall of the cyst was deroofed, along with the removal of a ruptured laminated membrane. Emergency surgery plus albendazole therapy are recommended protocols for cases like ours.

Conclusions: A patient from an endemic region with acute presentation of right upper quadrant pain can have spontaneously ruptured hydatidosis as a differential diagnosis. Intraperitoneal rupture and dissemination of hydatid cysts of the liver can be life-threatening if intervention is delayed. Immediate surgery is life-saving and prevents complications.

Keywords: acute abdomen, disseminated hydatidosis, *Echinococcus*, Nepal, ruptured hydatid cyst

Introduction

Hydatid cyst disease, also called cystic echinococcosis (CE) or hydatidosis, is a zoonotic parasitic disease that occurs in humans due to infection of the larval stage (metacestode) of a parasite belonging to the genus *Echinococcus*, especially *Echinococcus granulosus*[1]. In developing countries like Nepal, the burden of the hydatid cyst is significant but a neglected public health problem[2].

The liver is the most frequently involved organ (50–77%), followed by the lungs (18–35%); however, it can potentially impact any organ of the body[3,4]. Hydatid cysts grow at an average of 10–50 mm/year, depending on the site of the cyst, for which patients remain asymptomatic for years. Intraperitoneal rupture can happen due to trauma, or it may occur spontaneously as a result of increased pressure within the cyst[5]. This can lead to complications ranging from mild to fatal such as abdominal pain, urticaria, anaphylaxis, and even sudden death[5]. Thus it often warrants both emergency surgery and careful postoperative care[5]. In this report, we present the case of a patient with a liver hydatid cyst with spontaneous intraperitoneal rupture. Spontaneous intraperitoneal rupture is known to be a rare complication. This case report has been reported in line with the SCARE (Surgical CAse REport) criteria[6].

HIGHLIGHTS

- Hydatid cyst is endemic in sheep-rearing parts of the world but a neglected public health problem.
- Spontaneous intraperitoneal rupture is a rare complication of hydatid cysts.
- Clinical evaluation and radiological imaging confirm the intraperitoneal rupture and its extension.
- Appropriate, timely intervention, along with medical management, can treat the condition.

Case presentation

A 19-year-old male presented to the emergency department with the chief complaint of pain abdomen around the umbilical region and right upper quadrant for 12 h. The pain was acute in onset, gradual in progression, severe in intensity, aggravated with movement, and relieved slightly with intravenous analgesia. It was associated with nausea but without fever or vomiting. He had no abdominal trauma prior to the presentation. The patient had similar nature of pain 6 months back, and he was suspected

Institute of Medicine, Tribhuvan University, Maharajgunj, Nepal

Sponsorships or competing interests that may be relevant to content are disclosed at the end of this article.

**Corresponding author. Address: Institute of Medicine, Tribhuvan University, Maharajgunj 44600, Nepal. Tel.: +977-9861678146. E-mail address: madhurbhattarai180@gmail.com (M. Bhattarai).*

Annals of Medicine & Surgery (2023) 85:1172–1176

Received 7 January 2023; Accepted 1 March 2023

Published online 28 March 2023

http://dx.doi.org/10.1097/MS9.0000000000000383

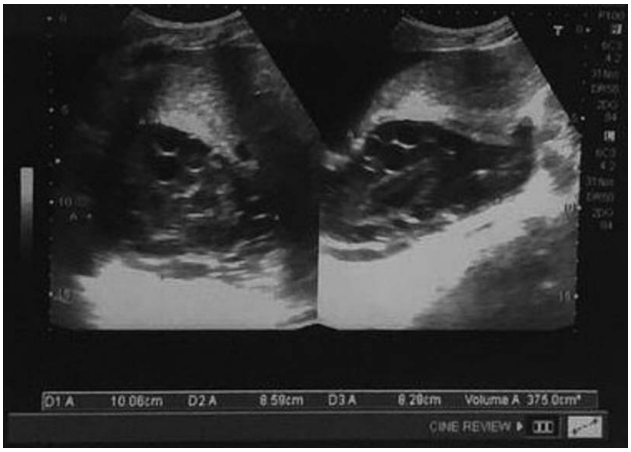

Figure 1. Ultrasonography showing heterogeneously hypoechoic area involving the right lobe of the liver with multiple septations.

of having some liver pathology in the local center and was advised for contrast-enhanced computed tomography of the abdomen and pelvis, but the patient failed to follow up. He had no systemic illnesses or prior abdominal surgeries. He used to consume alcohol occasionally and was a nonsmoker. He used to work as a shepherd for the last 3 years. On physical examination, he was tachycardiac and had lower blood pressure. Mild icterus was also present. Abdominal examination showed hepatomegaly and tenderness with guarding in the periumbilical and right upper quadrant region; later, it became generalized within 1–2 h. There were no features of allergic reaction. Blood investigation revealed an increased total leukocyte count of 14 500 with a neutrophilia count of 86%, decreased lymphocyte count of 3.2%, and an increased eosinophil count of 8%. The liver function test revealed a slightly raised aspartate aminotransferase at 62 U/l, with slightly raised bilirubin (total bilirubin 2.1 mg/dl and direct bilirubin at 1.4 mg/dl) and normal alanine transaminase at 40 U/l. Albumin and prothrombin time/international normalized ratio values were within normal limits. A serology test for *Echinococcus* was not performed.

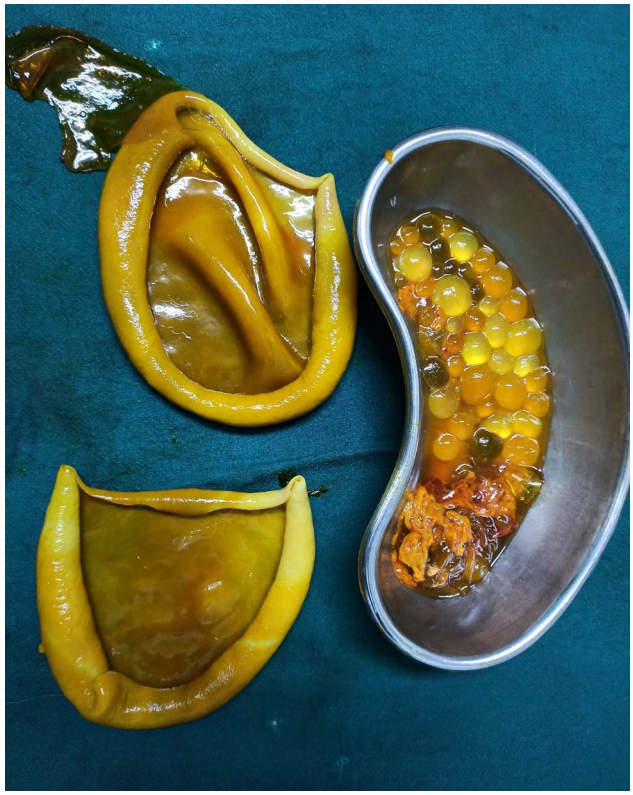

Figure 3. Daughter cysts and ectocysts.

On ultrasonography, intra-abdominal and pelvic fluid collection and a heterogeneously hypoechoic area measuring $10 \times 8 \times 8$ cm involving the right lobe of the liver with multiple septations within the abdominal collection was seen (Fig. 1). With suspicion of ruptured hydatidosis, a contrast-enhanced computed tomography scan of the abdomen and pelvis was done and showed the rupture of the anterior cyst wall with the presence of multiple cysts in the right liver disseminated to the pelvic cavity (Fig. 2).

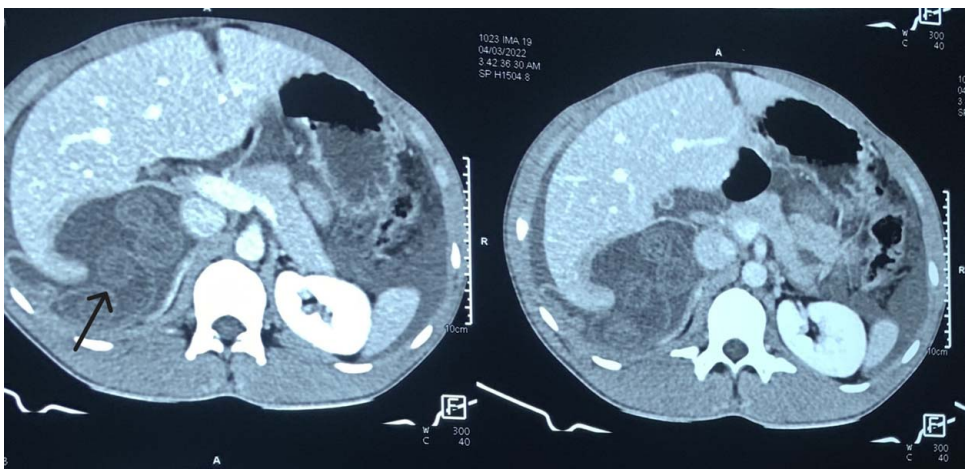

Figure 2. Contrast-enhanced computed tomography of the abdomen (arrow shows the cyst).

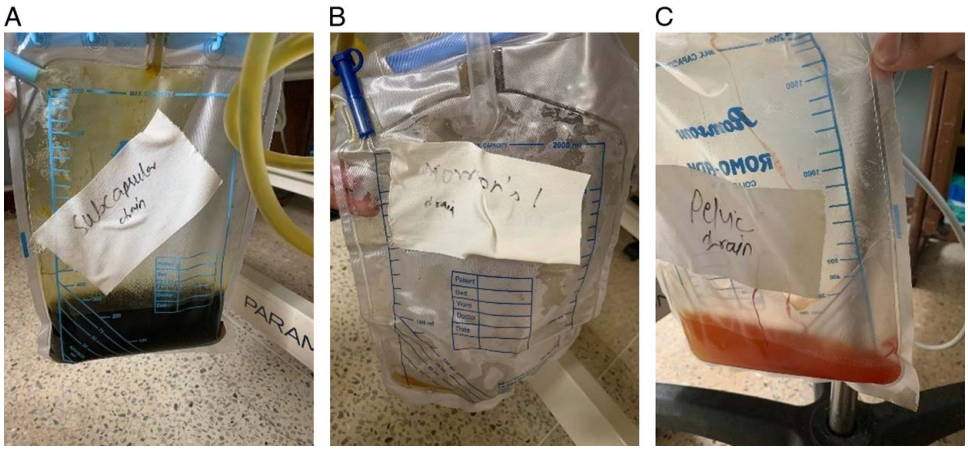

Figure 4. (A) Subcapsular drain with bilious content; (B) Morrison's drain with serous content; (C) pelvic drain with serosanguinous content.

Based on the history, examination and imaging, the patient was diagnosed with a ruptured hydatid cyst of the liver [stage CE-3A; WHO-IWGE (Informal Working Groups on Echinococcosis) classification] with dissemination without anaphylaxis. Along with resuscitation, the patient underwent emergency exploratory laparotomy where 2000 ml of bile-stained fluid with multiple disseminated daughter cysts (Fig. 3) in the peritoneal cavity, with omental deposits and multiple thickened separated laminated membranes in the liver were present. The daughter cysts were evacuated. The anterior wall of the cyst was deroofed. The laminated membrane was removed from the liver. Peritoneal lavage with a diluted povidone-iodine solution followed by 15 l of normal saline was done. Three closed system drains were kept, namely Morrison's drain (24 Fr), subcapsular drain (28 Fr), and pelvic drain (24 Fr) (Fig. 4A–C). The cystobiliary communication was suspected due to bilious intra-abdominal collection. However, cystobiliary communication was not clearly appreciated intraoperatively. So suturing was not done. The right Morrison's drain on the first, second, and third days was 50 ml per day, serous nature, and later its amount decreased. The left pelvic drain on the first day was 400 ml serosanguinous nature, 300 ml on the second day, 200 ml on the third and fourth days and minimum beyond and was removed later. The right subcapsular drain on the first day was 850 ml bilious, which decreased progressively being 300 ml, with very slight bile stained on the 12th postoperative day. The patient was discharged with a right subcapsular drain on the 12th postoperative day with albendazole therapy and follow-up counseling. The patient revisited the hospital after 4 days due to abdominal pain. The ascitic collection was present in the ultrasonographic evaluation. The right subcapsular drain was repositioned by the intervention radiology team. After this, the drain amount decreased and the drain was removed after 1 week. The patient underwent endoscopic retrograde cholangiopancreatography (ERCP) guided biliary sphincterotomy with common bile duct stenting 1 month later. The patient was kept on albendazole therapy. At 6 months of follow-up, no issues were present.

Discussion

Hydatid cyst is caused by a tapeworm, *Echinococcus granulosus*. The primary carriers of this tapeworm are dogs and wolves, and intermediate hosts are sheep, cattle, and deer. Humans are not part of the usual life cycle of this microorganism and are therefore considered accidental hosts. Humans tend to acquire this by ingesting ova from soil or water contaminated by the feces of infected dogs. It can occur in any organ of the body; however, the liver is the most frequently involved (50–77%), followed by the lung (18–35%)[3,4]. Hydatid cysts grow at an average of 10–50 mm/year, depending on the site of the cyst, for which patients remain asymptomatic for years[7]. Symptoms like abdominal pain and discomfort can manifest due to an increase in cyst size[8]. Some frequently reported complications in literature are rupture (perforation), bacterial infection, anaphylactic reaction, compression of the neighboring organs and compression of the vascular and biliary structures[9].

Hydatid cyst rupture is a rare but serious complication. It can be internal (cystobiliary fistula, rupture into the hollow viscus, bronchobiliary fistula, bronchopleural fistula, intraperitoneal rupture, intrapericardial rupture, intrapleural rupture) or, more rarely, it can be external (cystocutaneous fistula)[10]. Rupture into the peritoneal cavity can be traumatic, or it may occur spontaneously due to increased intracystic pressure[7]. The predisposing factor for the rupture of the cyst includes young age, cyst diameter greater than 10 cm, and superficial location of the cyst[11]. Rupture of cyst in the peritoneum is usually symptomatic, with constant abdominal symptoms (sharp pain in the right upper quadrant, vomiting, abdominal tenderness, and/or rebound)[12,13]. Hydatid fluid or free bile can cause peritonitis. If bile leakage occurs or the cyst is infected, the peritoneal sign and symptoms may develop earlier with increased severity. Allergic reactions to the cyst content may develop urticaria and maculopapular eruption of the skin. Life-threatening anaphylactic shock may also be a possible event[7,14].

The rupture of hydatid cysts is of three types according to the classification published by Lewall and McCorkell: contained, communicating, and direct[15]. Contained rupture is considered when only the endocyst of the parasite ruptures and the cyst contents remain within the pericyst derived from the host. Rupture

is called communicating if cyst contents escape through the biliary or bronchial radicles that are incorporated in the pericyst. Direct rupture occurs when both the endocyst and the pericyst tear release cyst contents directly into the peritoneal or pleural cavities or occasionally into other structures[15]. Ultrasound has a sensitivity of 85% and allows the detection of the cyst as well as to suspect cyst rupture by showing a floating membrane with intraperitoneal fluid[16]. Computed tomography (CT) has a sensitivity of 100% in demonstrating cyst rupture. CT is mostly preferred because it allows imaging of the entire abdomen and pelvis. In direct rupture, CT shows various findings like a detached membrane, cyst wall discontinuity, change in the architecture of the cyst or presence of daughter cysts, and fluid in the peritoneal cavity[8,16].

In our case, both imaging was done to confirm the intraperitoneal rupture and its dissemination. In hydatid cyst rupture, there is not much time before surgery as anaphylaxis is the most frequent cause of death[17]. However, anaphylaxis did not ensue in our case.

The American College of Gastroenterology Guidelines recommends surgery which can either be laparoscopic or open, based on available expertise in complicated hydatid cysts with multiple vesicles, daughter cysts, rupture, hemorrhage, fistulas, or secondary infection[18]. As in our case, direct rupture into the peritoneum requires emergent surgery. Immediate medical treatment against allergic reactions should also be initiated if necessary. Scolices can spill into the abdominal cavity, so the surgeon needs to address both the disease of the liver and the removal of protoscolices from the abdomen to decrease the possibility of metastatic hydatidosis[15]. Eliminating local disease, preventing complications, and minimizing morbidity, mortality, and recurrence risk is the main objective of surgery. However, there is controversy about the choice between radical and conservative approaches; radical operations include pericystectomy and liver resection, whereas conservative techniques include external drainage, unroofing, and cavity-obliterating methods. Conservative techniques are usually favored in endemic areas[19]. It is necessary to irrigate the peritoneal cavity with scolicidal agents and meticulously remove all the cystic contents. Cetrimide–chlorhexidine, povidone-iodine (10%), silver nitrate (0.5%), hypertonic saline solution (3–30%), chlorhexidine (0.4%), and praziquantel are some of the choices of scolicidal agents[3,14]. In our case, the extraction of laminated membrane and the peritoneal cavity was irrigated with povidone-iodine solution and washed with normal saline.

While performing surgery, surgeons should search for any connection between the cyst and the biliary tract and should treat it promptly, as bile leakage is the major problem after hydatid cyst surgery. Treatment may include suturing the bile duct, omentopexy, effective external drainage, and insertion of a T-tube for decompression. In case of postoperative detection of bile leakage, the biliary tree is decompressed via ERCP-guided sphincterotomy, nasobiliary drainage, or internal biliary stenting. Abdominal drains should be placed both into the cystic cavity and in the abdomen before surgery completion[7]. In our case, ERCP-guided biliary sphincterotomy with common bile duct stenting was performed postoperatively.

Antihelmintic treatment is started 2–3 weeks before elective hydatid cyst surgery, which is continued 6–8 weeks post-operatively. However, patients with traumatic or spontaneous cyst rupture have to undergo emergency surgery and cannot receive medical treatment prior to surgery. These patients are mandatorily started with medical treatment as early as possible after surgery, continuing it for 1–6 months in order to reduce the risk of recurrence[20]. Albendazole (10–15 mg/kg/day) is most prescribed antihelmintic drug. Intraperitoneal hydatidosis may occur in patients with ruptured hydatid cysts. Patients with uncomplicated hydatid cysts may require follow-up with ultrasonography and indirect hemagglutination test starting 6 months after surgery and subsequently repeated every 1–2 years. Patient with ruptured hydatid cyst requires scrupulous follow-up after surgery within a short interval[7,16,21]. Recurrence and secondary hydatidosis must be looked up.

Conclusions

The complication of hydatidosis must be included in the differential diagnosis of a patient with an acute abdomen in an endemic area. Intraperitoneal rupture of a hydatid cyst of the liver is a rare complication that is serious and life-threatening. Management and treatment depend on immediate surgery along with medical management. The patient should be kept on follow-up as there is a chance of high rates of recurrence.

Ethical approval

None.

Consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Sources of funding

None.

Authors' contribution

S.A., M.B., and S.S.: wrote the original manuscript, reviewed, and edited the original manuscript; S.G.: reviewed and edited the original manuscript; A.B., L.A., P.B.S.K.: senior author and manuscript reviewer.

Conflicts of interest disclosure

There are no conflicts of interest.

Research registration unique identifying number (UIN)

None.

Guarantor

Madhur Bhattarai.

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

[1] Mandal S, Deb Mandal M. Human cystic echinococcosis: epidemiologic, zoonotic, clinical, diagnostic and therapeutic aspects. Asian Pac J Trop Med 2012;5:253–60.

[2] Devleesschauwer B, Ale A, Torgerson P, *et al*. The burden of parasitic zoonoses in Nepal: a systematic review. PLoS Negl Trop Dis 2014;8:56.

[3] Akcan A, Sozuer E, Akyildiz H, *et al*. Predisposing factors and surgical outcome of complicated liver hydatid cysts. World J Gastroenterol 2010;16:3040.

[4] Shah NJ, Vithalani NK, Chaudhary RK, *et al*. Disseminated peritoneal hydatidosis following blunt abdominal trauma: a case report. Cases J 2008;1:118.

[5] Derici H, Tansug T, Reyhan E, *et al*. Acute intraperitoneal rupture of hydatid cysts. World J Surg 2007;31:1526–7.

[6] Agha RA, Franchi T, Sohrabi C, *et al*. The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines. Int J Surg 2020;84:226–30.

[7] Akbulut S, Ozdemir F. Intraperitoneal rupture of the hydatid cyst: four case reports and literature review. World J Hepatol 2019;11:318.

[8] Alexiou K, Mitsos S, Fotopoulos A, *et al*. Complications of hydatid cysts of the liver: spiral computed tomography findings. Gastroenterol Res 2012;5:139.

[9] Toumi O, Noomen F, Salem R, *et al*. Intraperitoneal rupture of hydatid cysts. Eur J Trauma Emerg Surg 2017;43:387–91.

[10] Akbulut S. Parietal complication of the hydatid disease: comprehensive literature review. Medicine (Baltimore) 2018;97:e10671.

[11] Akcan A, Akyildiz H, Artis T, *et al*. Peritoneal perforation of liver hydatid cysts: clinical presentation, predisposing factors, and surgical outcome. World J Surg 2007;31:1284–91.

[12] Majbar AM, Aalala M, Elalaoui M, *et al*. Asymptomatic intra-peritoneal rupture of hydatid cyst of the liver: case report. BMC Res Notes 2014;7:114.

[13] Bhattarai HB, Bhattarai M, Shah S, *et al*. Meckel's diverticulum causing acute intestinal obstruction: a case series. Clin Case Rep 2022;10:e6518.

[14] Yilmaz M, Akbulut S, Kahraman A, *et al*. Liver hydatid cyst rupture into the peritoneal cavity after abdominal trauma: case report and literature review. Int Surg 2012;97:239.

[15] Lewall DB, McCorkell SJ. Rupture of echinococcal cysts: diagnosis, classification, and clinical implications. AJR Am J Roentgenol 1986;146:391–4.

[16] Mejri A, Arfaoui K, Omry A, *et al*. Acute intraperitoneal rupture of hydatid cysts of the liver Case series. Medicine (Baltimore) 2021;100: e27552.

[17] Gulalp B, Koseoglu Z, Toprak N, *et al*. Ruptured hydatid cyst following minimal trauma and few signs on presentation. Neth J Med 2007;65:117–8.

[18] Marrero JA, Ahn J, Rajender Reddy K. Americal College of Gastroenterology. ACG clinical guideline: the diagnosis and management of focal liver lesions. Am J Gastroenterol 2014;109:1328–47.

[19] Pişkin T, Ara C, Dirican A, *et al*. Perforated hydatid cyst into peritoneum presented with urticaria: a case report. Dicle Med J Cilt 2010;37:71–4.

[20] Botezatu C, Mastalier B, Patrascu T. Hepatic hydatid cyst – diagnose and treatment algorithm. J Med Life 2018;11:203.

[21] Belli S, Akbulut S, Erbay G, *et al*. Spontaneous giant splenic hydatid cyst rupture causing fatal anaphylactic shock: a case report and brief literature review. Turk J Gastroenterol 2014;25:88–91.