
CASE REPORT

Hydatid Cyst: Enteric Fistula in a Patient with Disseminated Abdominal Hydatidosis

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ABSTRACT

Disseminated peritoneal hydatidosis is rare. Infrequently hydatid cysts can rupture into adjacent organs. We report a case of secondarily disseminated peritoneal hydatidosis with rupture into the gut presenting as hydatidorrhoea. A 26-year-old man presented with passage of white 'grape'-like structures with stool. Ultrasonography showed disseminated hydatidosis. Magnetic resonance imaging further showed a large hydatid cyst arising from the parietal wall with a peripheral daughter cyst and localised air-fluid level, suggestive of a peritoneal hydatid cyst with enteric fistula. The fistulous site was demonstrated on magnetic resonance imaging. Due to the presence of disseminated disease, the patient was treated medically. Gastrointestinal complications of hydatid infestation are uncommon as the walls of the digestive tract offer major resistance to the formation of communication between the cystic cavity and the gastrointestinal lumen. Rare cases of rupture of hepatic hydatids into the stomach, duodenum, and colon have been reported. Peritoneal cyst rupture into the bowel is extremely rare. It is important to identify this uncommon complication and follow-up the patient for a prolonged period to ensure resolution of disseminated disease.

Key Words: Cysts; Echinococcosis; Peritoneal diseases; Rupture, spontaneous

中文摘要

彌散性腹包蟲囊病引致的腸瘻

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瀰漫性腹腔包蟲病很罕見。包蟲囊腫很少可破裂至鄰近器官。本文報告一例繼發性瀰漫性腹腔包蟲病破裂進入腸道的病例。患者為一名26歲男性，大便見白色葡萄狀的排泄物。超聲檢查顯示為瀰漫性包蟲病。磁共振成像進一步證實腔壁有一個大型包蟲囊腫，在其外圍另有一個小型囊腫，內有水-空氣界線，顯示腹膜包蟲囊腫引致的腸瘻。磁共振成像確定了腸瘻位置。此病例屬於彌散性，患者後接受藥物治療。由於消化道壁可以作為囊腔和胃腸管腔之間的一道屏障，因此包蟲能侵染胃腸道的機會很低。肝包蟲囊腫破裂而進入胃、十二指腸和結腸的情況很罕見，而腹腔囊腫破裂並進入腸道更為極其罕有。識別這種罕見的併發症並長期跟進患者以確保其完全消散至為關鍵。

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INTRODUCTION

Echinococcosis, also known as hydatid disease, is the most common infestation in humans worldwide.¹ Echinococcosis is caused by the larval stage of *Echinococcus granulosus*. Common sites include the liver (75%) and lung (15%).¹ Peritoneal echinococcosis (13%) is usually secondary to a primary hepatic cyst. Presentation in the form of disseminated intraperitoneal hydatid disease is extremely rare.² The cyst can rarely rupture into the peritoneal cavity, biliary tree, pleural space, or hollow viscus. We present a case of secondarily disseminated peritoneal hydatid cyst with rupture into the gut presenting as loose stool and passage of daughter cysts per rectum.

CASE REPORT

In January 2014, a 26-year-old man presented with abdominal distension and pain for 1 month followed by loose stools for 10 days. He also complained of two episodes of passage of small white ‘grape’-like structures with stool. He had a history of excision of hepatic hydatid cyst 14 years previously. General examination revealed cachexia, pallor, and abdominal distension.

Ultrasonography (USG) revealed multiple large thin-walled fluid-filled cysts with daughter cysts involving the right lobe of the liver, spleen, and peritoneal cavity, including the pelvis (Figure 1). A diagnosis of disseminated hydatidosis was made. All cysts belonged to the World Health Organization stage cyst type 2 of cystic echinococcosis.³ Due to severe distortion of the normal anatomical structures by the cysts, a second line of investigation by contrast-enhanced computed

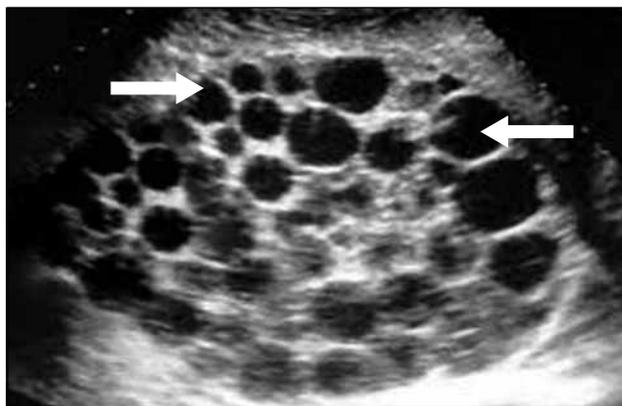


Figure 1. An ultrasonogram shows a hydatid cyst in the liver with multiple daughter cysts (arrows).

tomography (CT) or magnetic resonance imaging (MRI) of the abdomen was advised to look for any evidence of complications or involvement of sites poorly accessible by USG. As the patient had marginally elevated serum creatinine levels (serum creatinine, 124 $\mu\text{mol/l}$ [reference range, 53-106 $\mu\text{mol/l}$]) with an estimated glomerular filtration rate of 38 ml/min/1.73 m² (reference range, >60 ml/min/1.73 m²), MRI was preferred to avoid the nephrotoxic effects of iodinated contrast medium. MRI

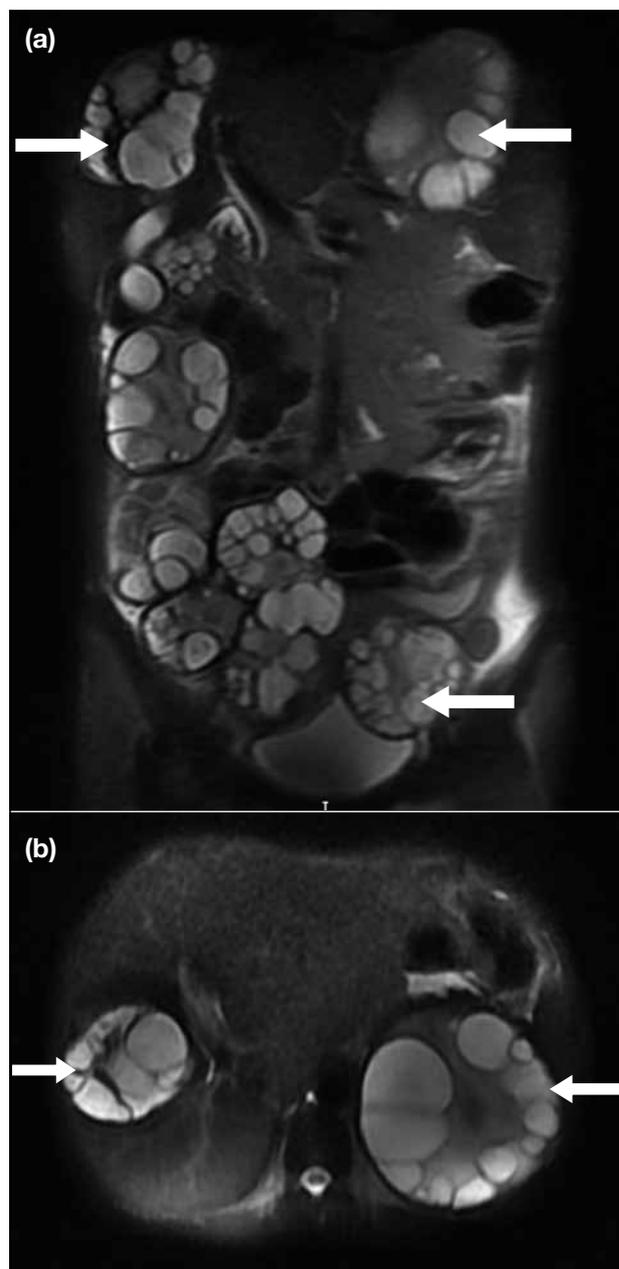


Figure 2. T2-weighted magnetic resonance images: (a) coronal and (b) axial images show multiple disseminated hydatid cysts (arrows) involving the liver, spleen, and peritoneal cavity.

showed multiple cysts, which were hypointense in T1-weighted and hyperintense in T2-weighted images, with daughter cysts and an inner hypointense and outer hyperintense peripheral rim representing the ectocyst and pericyst, respectively (Figure 2). No evidence of spinal cord involvement, biliary communication, or pleural involvement was noted. However, triplanar images revealed a large hydatid cyst arising from the parietal wall, with peripheral daughter cysts and localised air-fluid level (Figure 3). Repeated USG showed multiple small hypoechoic cysts arising from the inner surface of the parietal wall. This was suggestive of a peritoneal hydatid cyst with enteric fistula. As the ruptured cyst adhered to the anterior parietal wall in the pelvis, topographically away from the anterior, transverse and descending colon, it was

suspected that the fistula was connected to the adjoining jejunal / ileal loops. On careful examination, a possible fistulous site with adjacent small bowel was identified (Figure 4). CT enteroclysis for confirmation could not be done due to altered renal function and financial constraints.

Due to the presence of disseminated disease, surgery could not be performed. The patient was treated with long-term oral albendazole (400 mg twice daily for 6 weeks). At follow-up after 6 weeks, the patient's symptoms were alleviated and blood count and liver function tests were normal. The cysts, however, did not appear to have reduced in size or stage. A further 6-week course was prescribed. He was later lost to follow-up.

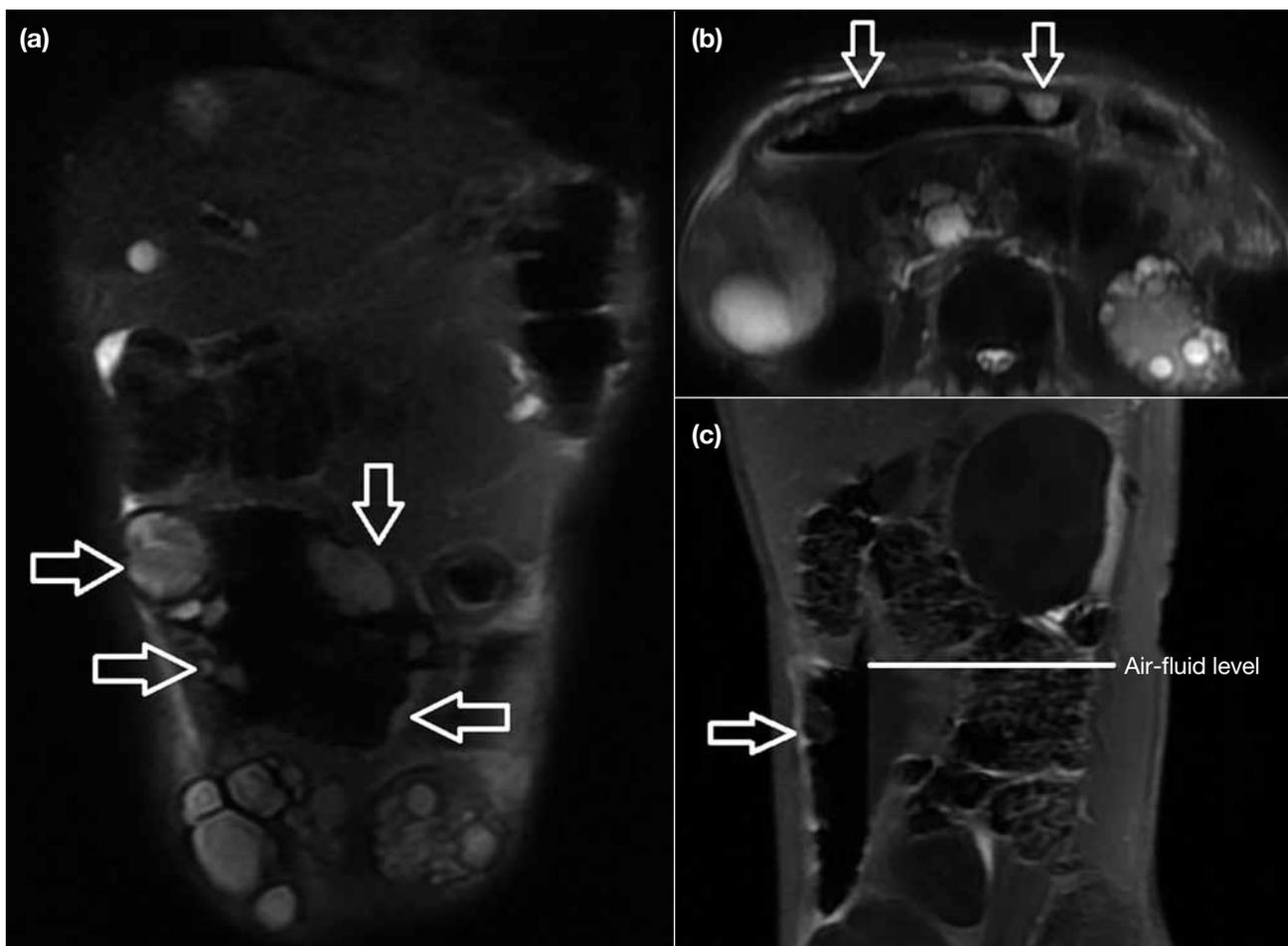


Figure 3. Magnetic resonance images: (a) coronal T2-weighted image shows a well-defined thick-walled rounded cavity containing air with peripheral daughter cysts (arrows) just below the anterior abdominal wall; (b) axial T2-weighted image shows the daughter cysts within the collapsed mother cyst (arrows); and (c) post-contrast image (sagittal T1 fat-saturated sequence) demonstrates the air-fluid level and enhancing cyst wall (arrow).

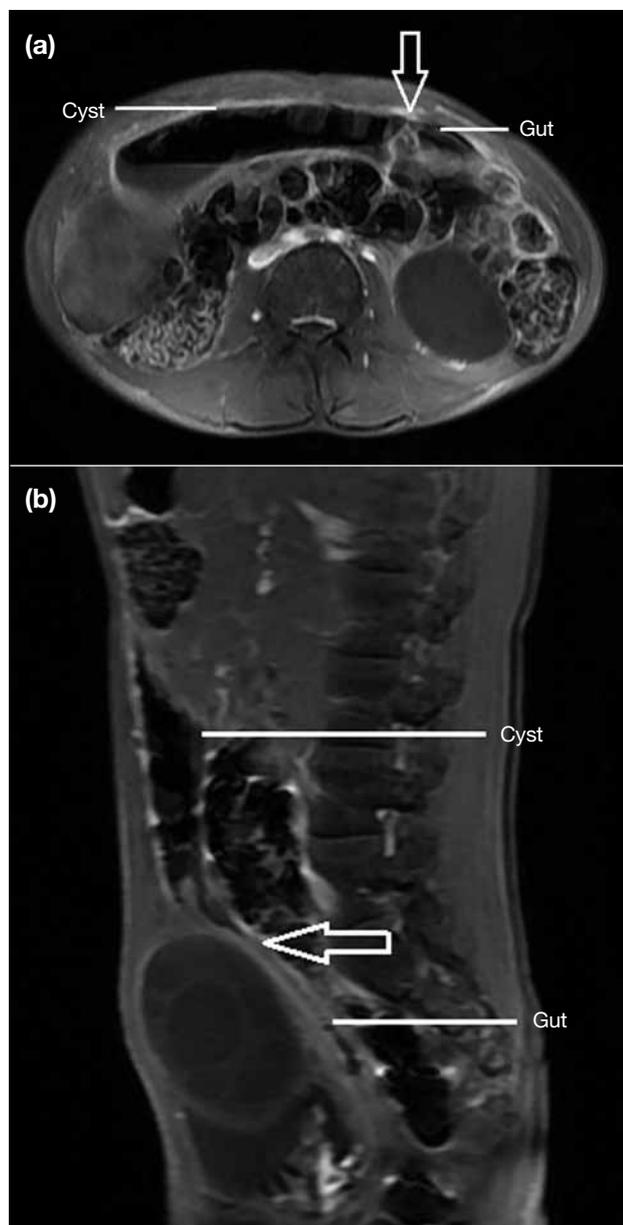


Figure 4. Post-contrast magnetic resonance images: (a) axial T1-weighted fat-saturated image shows a defect in the wall separating the cyst from the gut wall, the possible site of the fistula (arrow); and (b) sagittal T1-weighted fat-saturated image demonstrates a bowel loop posterior to the cyst, which is seen communicating with the cyst cavity (arrow).

DISCUSSION

Hydatid cysts can develop anywhere in the human body, but the liver is the most frequently involved organ (52%-77%), followed by the lungs (10%-40%), spleen (4%), kidney (3%), cerebrum (2%), and heart (0.02%-2%).⁴ Involvement of the small intestine is extremely rare and, to date, has been reported in only

one patient.⁴ Most patients are asymptomatic. Among the major complications of abdominal echinococcosis, intrathoracic or intra-abdominal rupture, communicating rupture into the biliary tree, infections, extrinsic compression of the bile ducts, systemic anaphylaxis, and peritoneal or pleural dissemination are the most frequent and life-threatening.²

Abdominal hydatid cysts, especially those in the liver, have the capacity for fistula formation, mainly in the biliary ducts, and thus present one of the classic complications with which the general surgeon is confronted during operation for hydatid disease. The gastrointestinal complications of hydatid infestation are less frequently seen, as the walls of the digestive tract offer major resistance to the formation of communications between the cystic cavity and the gastrointestinal lumen.⁵ Despite this, rupture into the hollow viscus has been reported in a few patients. Communication with the duodenum, stomach, and left colon from hepatic hydatid cysts has been documented.⁶ This patient represents a rare reported case of rupture of a peritoneal hydatid, possibly into a jejunal / ileal loop.

In the case of a rupture or a defect in the laminated membrane, the germinal layer passes through the membrane and creates a satellite hydatid cyst by a process known as ectogenic vesiculation.⁷ Primary intestinal involvement has been reported in only three patients to date. In this patient, MRI revealed the presence of daughter cysts in a large rounded cavity with enhancing walls and air-fluid levels. Secondary rupture was considered more likely than primary intestinal involvement because the morphology of the air-filled structure containing the daughter cysts seemed rounded in craniocaudal and transverse dimension instead of an elongated gut-like configuration. The patient could not undergo CT enteroclysis for confirmation due to altered renal function and cost constraints.

Gut involvement is usually treated by surgical resection along with pre- and post-operative albendazole therapy.⁴ In this patient, surgery was an unlikely option due to solid organ and disseminated peritoneal involvement. Anaphylactic shock due to spontaneous rupture may be a serious complication. There were no such issues with this patient, however, because of the contained cysto-enteric communication. It is important to identify this rare complication, and follow-up the patient for prolonged periods for resolution of disseminated disease.

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