

Case Report

Rare presentation of multi-organ abdominal echinococcosis: report of a case and review of literature

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Abstract: Hydatid disease, which is also known as cystic echinococcosis, is a zoonotic infection caused by the cestode tapeworm *Echinococcus granulosus* and rarely by *Echinococcus multilocularis*. In this report we describe an unusual case of a 19-year-old woman who was admitted to our hospital for abdominal pain, nausea, and vomiting. Computed tomography revealed multi-organ abdominal echinococcosis. The patient recovered after undergoing surgery to excise the cyst. The diagnosis, clinical features, treatment, and prevention in this case of multi-organ abdominal echinococcosis are discussed, in light of the relevant literature.

Keywords: Hydatid disease, echinococcosis, multiple abdominal organs

Introduction

Hydatid disease (HD) is endemic in the cattle grazing areas of Australia, New Zealand, the Middle East, India, Africa, South America, and Turkey. In humans, HD commonly involves the liver (75%) and lungs (15%), followed by other regions of the body (10-15%) [1-4]. Multi-organ abdominal HD is the most serious form of HD and is potentially fatal. Here, we describe the case of a 19-year-old patient with multi-organ abdominal HD and review the relevant literature.

Case report

A 19-year-old Tibetan woman was admitted to China-Japan Friendship Hospital on July 14, 2014 due to periumbilical pain accompanied by nausea and vomiting, constipation, and inability to pass flatus for over a week, without any obvious predisposing causes. Abdominal computed tomography (CT) revealed cysts of various sizes distributed throughout the liver parenchyma, capsule of the right liver lobe, and peritoneal cavity. Also visible in the liver and peritoneum were lower-density daughter cysts and soft-tissue nodules, of which the largest was detected in the right liver lobe (approx-

mately 11.1 × 9.6 × 12.0 cm in size). The CT also revealed an ileus with no obvious mechanical obstruction. Despite being placed nil-by-mouth and administration of gastric decompression and fluid perfusion, the patient's condition failed to improve. Thus, the patient sought additional medical treatment at our hospital. Since the onset of her illness, the patient exhibited a conscious mental state and experienced fatigue and poor sleep, but did not experience irregular urination or weight change.

Patient epidemiology and history

The patient was born and lived for many years in a Tibetan cattle-grazing community in which HD was endemic. The patient often drank tap water and ate raw meat during her childhood while living in this community.

When she was 12 years old, the patient experienced acute abdominal pain and was locally diagnosed with HD. At that time, she was treated with traditional Tibetan medicine, which helped to alleviate her symptoms; however, she continued to experience intermittent abdominal pain. At the age of 15, the patient suffered from repeated episodes of abdominal pain and was treated with an oral vermifuge drug.

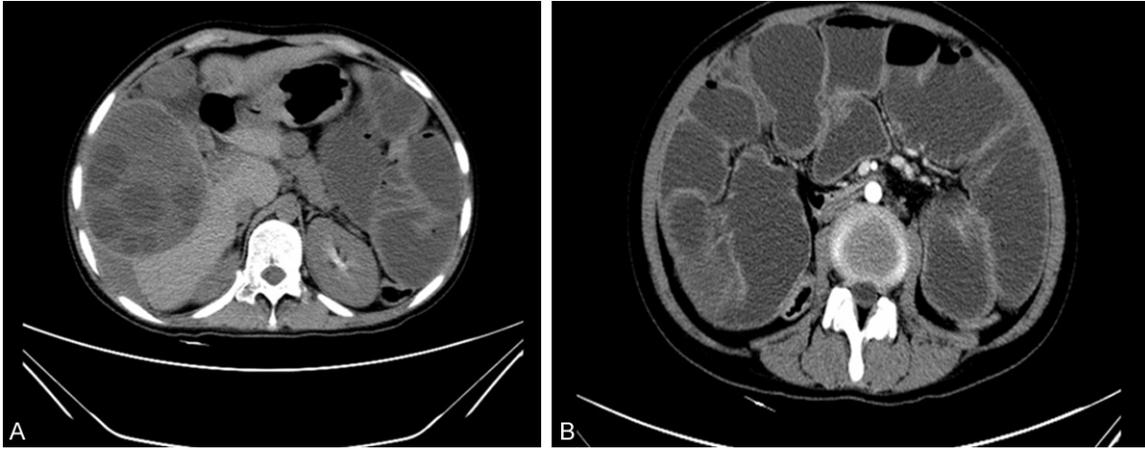


Figure 1. Abdominal and pelvic helical computed tomography for hepatic echinococcosis and pelvic and abdominal echinococcosis. A. Within the liver, multiple round, low-density echogenic lesions were visible, with the largest lesions measuring approximately 12.0 × 9.5 cm. B. Within the pelvis, several cystic lesions containing multiple small vesicular structures were visible.

Examination at admission

The examination at admission resulted in the following: body temperature, 37°C; pulse, 150/min; respiration, 20/min; blood pressure, 95/53 mmHg; normally developed; dim consciousness; active position; no spider angioma; no swollen superficial lymph nodes; no generalized edema; normal breath sound; no rhonchi; normal apex beat; no palpated fibrillation; normal cardiac dullness border; no irregular heart beat; no murmur at the cardiac valve auscultation areas; and no pericardial rub. Her abdomen was soft and flat, and there was no visible peristalsis, positive right upper quadrant pressure pain, no rebound tenderness or rigidity, no shifting dullness, positive knocking pain over the liver, and no bowel sounds present during the one-minute auscultation. In addition, there was no lower limb swelling, and her limb extremities were warm.

She was diagnosed with acute mechanical intestinal obstruction, hydatid disease, and possible anaphylactic shock.

Auxiliary examinations

The routine blood examination revealed the following: white blood cell (WBC) count, $8.1 \times 10^9/L$; granulocyte (GR) count, $7 \times 10^9/L$; GR%, 86.5%; eosinophil (EO) count, $0.05 \times 10^9/L$; EO%, 0.6%; hemoglobin, 119 g/L; and platelet count, $119 \times 10^9/L$. The routine urine examination revealed the following: protein (-), urobilino-

gen (-), bilirubin (-), ketone bodies (-), occult blood (-), and WBC (-). The comprehensive metabolic panel revealed the following: alanine transaminase, 29 U/L; aspartate transaminase, 92 U/L; alkaline phosphatase, 72 U/L; gamma-glutamyl transpeptidase, 37 U/L; total protein, 64.9 g/L; albumin, 31.4 g/L; globulin, 33.5 g/L; total bilirubin, 5.06 $\mu\text{mol/L}$; direct bilirubin, 0.6 $\mu\text{mol/L}$; indirect bilirubin, 4.46 $\mu\text{mol/L}$; cholinesterase 3.99 KU/L; creatinine, 227.3 $\mu\text{mol/L}$; blood urea nitrogen, 22.91 mmol/L; UA, 438 $\mu\text{mol/L}$; calcium, 1.95 mmol/L; phosphorus, 0.63 mmol/L; cholesterol, 1.27 mmol/L; triglycerides, 0.18 mmol/L; high-density lipoprotein cholesterol, 0.89 mmol/L; low-density lipoprotein cholesterol, 0.11 mmol/L; glucose, 5.42 mmol/L; sodium, 160 mmol/L; chlorine 129.5 mmol/L; potassium, 5.74 mmol/L; CO_2 , 19 mmol/L; anion gap, 11.5 mmol/L; and osmolality, 334.9 mOsm/L. The screening for parasites resulted in a positive result for hepatic echinococcosis IgG antibody.

Abdominal and pelvic helical CT was performed. Within the liver, multiple round, low-density echogenic lesions were visible, with the largest lesions measuring approximately 12.0 × 9.5 cm (**Figure 1A**). Intracystic density was unevenly distributed. Multiple small vesicular structures were visible within the lesions, some with cyst walls of varying thickness, some with calcification, and some protruding from the liver surface. Upon perfusion, the lesion periphery

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exhibited a pronounced banded morphology. Several cystic lesions containing multiple small vesicular structures were visible within the pelvis (**Figure 1B**). The cystic lesions contained multiple small vesicular structures. Perfusion imaging increased the cyst wall density. The CT also displayed noticeable expansion of the small intestine, with pneumatosis and effusion. The left hepatic lobe was slightly enlarged; no noticeable widening of the liver fissure was observed, and the porta hepatis was clear. Gallbladder morphology and size were normal, with no noticeable irregular density, and abnormal enlargement of the bile duct was not apparent. Spleen morphology and size were normal, with no noticeable irregular density. The pancreatic border was clear, and the morphology and size were normal. A small echogenic cyst was seen in front of the pancreas. Several echogenic small lymph nodes were observed throughout the abdominal cavity and posterior to the peritoneum, with the larger of these nodes measuring approximately 0.8 cm at the narrowest point. No irregularities were observed within the kidneys. The uterus position and morphology were normal, and no calcified shadows were observed. Low-density lesions were observed in the uterine appendages. Bladder filling was poor, and a density shadow of gases was seen within the bladder. A density shadow of a thin layer of fluid was observed in the pericardium. The lower lobes of the left and right lungs exhibited consolidation and ground-glass opacity.

Of these observations, the expanded small intestine was commensurate with the diagnosis of intestinal obstruction. There were shadows of multiple abdominal and hepatic echogenic cystic structures. Further observation of the abdominal cavity and post-peritoneal lymph nodes might be needed. In addition, there was a small accumulation of fluid in the pericardium, low-density lesions in the bilateral uterine appendages, accumulation of fluid in the abdomen and pelvis, striped shadows on the bilateral lungs, and ground glass opaque lesions in the bilateral lungs.

After admission, the patient was administered albendazole (20 mg/kg/d, 3 times/d) nasogastrically for treatment of the primary disease. For the intestinal obstruction and shock, gastric tube drainage, fasting, gastro-intestinal decompression, bowel relaxing, and anti-infection and

anti-shock rehydration were administered. On July 24, 2014, after the patient's condition was stabilized, we performed an exploratory laparotomy, enterolysis, ileocecal resection, partial enterectomy, and internal cystectomy of the hydatid cyst under general anesthesia. Through the routine exploration, we detected mild ascites in the abdominal cavity, obvious enlargement of the jejunum and ileum, and edema and congestion of the intestinal wall. Multiple cystic space-occupying grayish-white lesions of varying sizes adhered to each other to form gobbets in the pelvic cavity. Multiple cystic space-occupying lesions were also observed in the greater omentum and ascending colon. The liver was soft, red-brown, and sharp-edged with cystic space-occupying grayish-white lesions (10 × 7 cm) in segment V, protruding to the liver surface and adhering to surrounding tissues to form gobbets. Scattered cystic space-occupying lesions were observed in the stomach periphery and ligamentum teres. The gall bladder measured 8 × 3 × 3 cm with a wall thickness of approximately 0.2 cm and no noticeable stones. There were no noticeable swollen lymph nodes at the porta hepatis. No abnormalities were found in the other organs of the abdominal cavity.

Several cystic space-occupying lesions were completely removed from within the pelvis. Given that the hydatid cysts of the liver were located mostly in the parenchyma, we used fine needle aspiration to draw a small amount of thick, turbid fluid. The cyst wall was then incised, and several internal septa were removed with oval forceps. The green-yellow color of the internal septa suggested that the hydatid cyst was interconnected with the bile duct. The abdominal cavity was washed thoroughly with plenty of metronidazole, and a drainage tube was placed in the right intrahepatic space and another in the pelvis to drain through the right abdominal wall.

Postoperative course

The diameter of the pile of cystic dermoid substances was 8 cm, and they were hepatic hydration cyst walls. There were 13 pelvic nodular cysts of 2-11 cm in diameter, and the sections were cystic and contained clear and jelly-like fluid. The dyatid cyst walls were microscopically confirmed (**Figure 2A**). An excised ileum fragment (length, 8.5 cm; circumference, 10 cm)

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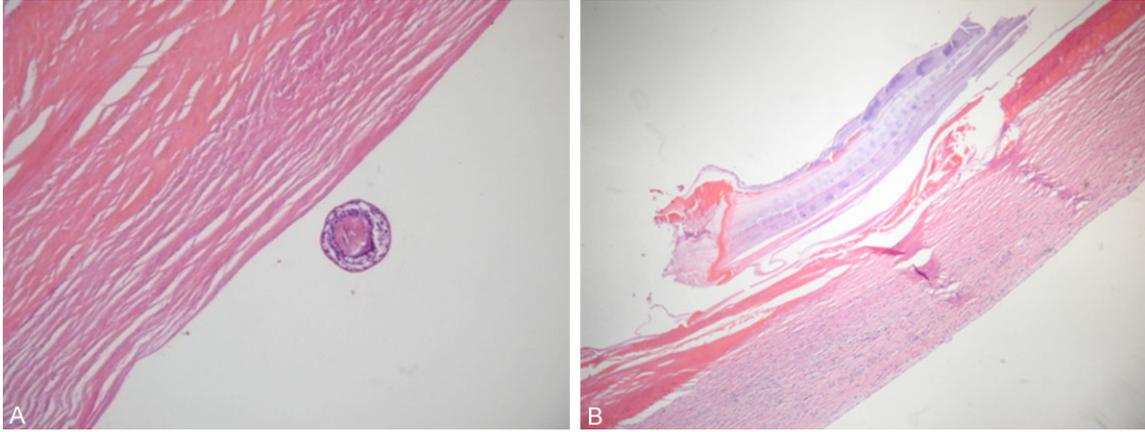


Figure 2. Postoperative pathology following treatment for hepatic echinococcosis and pelvic and abdominal echinococcosis. A. Dyatid cyst. B. Proliferation of serofibrous tissues and inflammatory cell infiltration.

was perforated (diameter, 2 cm) at a distance of 3 cm from one end and 5 cm from the other end. Microscopic observation showed formed ulcers at the perforation, with proliferation of serofibrous tissues and inflammatory cell infiltration (**Figure 2B**). Other mucosa showed chronic inflammation, serosa edema, and inflammatory cell infiltration. The terminal portion of the ileus (length, 7 cm; circumference, 7 cm), cecum (length, 2.5 cm; circumference, 6 cm), and appendix (length, 7 cm; diameter, 0.5 cm) were excised. Microscopic observation showed mucosal ulceration of the ileocecum, with partial intestinal wall necrosis, serosa edema, and inflammatory cell infiltration. Three mesenteric lymph nodes had lymphoid hyperplasia. There were no noticeable changes to the appendix.

After surgery, another course of albendazole was administered in addition to routine anti-infection treatment and fluid replacement. The patient recovered well; she had regular bowel movements and flatus, no bloating after meals, and no abdominal pain. After redressing, the patient was discharged.

At discharge, the diagnosis was acute mechanical intestinal obstruction, anaphylactic shock, hepatic echinococcosis, pelvic echinococcosis, acute hepato-renal injury, and acute myocardial injury.

Discussion

Hydatid disease is endemic in sheep farming and cattle farming areas of Asia, North and East Africa, South America, Australia, and the

Middle East. Dogs and other carnivorous animals are definitive hosts, while sheep, cattle, horses, and goats are intermediate hosts. Humans are an accidental and dead-end intermediate host. More than 80-90% of hydatid cysts occur in the liver, lungs, or both. Hydatid cysts have been infrequently reported in the spleen, kidney, peritoneal cavity, skin, and muscles and rarely involve the heart, brain, vertebral column, ovaries, pancreas, gallbladder, thyroid gland, breast, and bones [5-8]. Clinical presentation of HD depends upon the size, site, and depth of the lesion.

Most patients remain asymptomatic for years before presenting with vague abdominal symptoms such as nonspecific pain, abdominal fullness, dyspepsia, anorexia, and vomiting [9, 10]. Symptoms due to peritoneal hydatidosis commonly arise from complications of enlarging abdominal cysts or rupture into the peritoneum, which may present as acute abdominal pain [7, 11, 12].

The most useful diagnostic utility is abdominal ultrasound or CT, on which lesions appear well defined, with or without internal separation [13-15].

The principal treatment is surgery. However, pre- and postoperative courses of albendazole and praziquantel should be considered to sterilize the cyst and reduce the chance of anaphylaxis and recurrence [16-19].

This patient had hepatic echinococcosis and pelvic and abdominal echinococcosis, and she recovered favorably and was discharged follow-

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ing exploratory laparotomy, enterolysis, ileocecal resection, partial enterectomy, and hydatid cyst internal cystectomy under general anesthesia in addition to postoperative anti-infection therapy, anti-parasitic therapy, fluid infusion, and nutritional supplementation.

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Disclosure of conflict of interest

None.

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