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Abdominal hydatidosis: Unveiling a rare clinical entity – A case report and review of literature

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ABSTRACT

Hydatid disease (HD), or echinococcosis, is a zoonotic parasitic infection caused by the larval stage of the *Echinococcus* tapeworm. It is endemic in several regions globally, particularly in rural areas of southern South America, Central Asia, China, parts of Africa, the Mediterranean basin, and the Middle East. The liver is the most affected organ, followed by the lungs. However, the disease can involve almost any organ, including the brain, bones, and, in rare instances, the pelvic region, as highlighted in this case report. HD often progresses silently, remaining asymptomatic during the initial stages and sometimes even for years. The clinical presentation is usually non-specific, with common symptoms including abdominal pain, hepatomegaly, and, in severe cases, anaphylaxis following cyst rupture. Extrahepatic, intra-abdominal hydatid cysts are particularly rare, occurring in only 6–11% of cases, making this presentation an unusual and noteworthy clinical finding. In this case, a 32-year-old female involved in animal husbandry and farming presented with a progressively enlarging lower abdominal lump, accompanied by intermittent abdominal pain and constipation. Hydatid cysts involving multiple abdominal structures, including the liver, spleen, omentum, broad ligament of the uterus, and pelvis, are uncommon occurrence. Such complex cases pose considerable challenges in diagnosis and management, highlighting the importance of a careful and collaborative multidisciplinary approach. This case report highlights the complexities associated with diagnosing and managing abdominal hydatidosis, particularly in its atypical presentations. Accurate diagnosis relies on a comprehensive approach, integrating clinical assessment, serological investigations, and advanced imaging techniques.

Key words: *Echinococcus*; Extrahepatic; Hydatid cyst; Intra-abdominal; Exploratory laparotomy

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INTRODUCTION

Hydatid disease (HD) is a parasitic infection caused by the larval form of the *Echinococcus* species. The cystic variant is primarily induced by *Echinococcus granulosus*, a tapeworm that infects humans as an inadvertent intermediate host within its life cycle. In this cycle, dogs and other canines act as the definitive hosts, whereas herbivorous animals serve as intermediate hosts. HD is a significant health problem in areas where animal husbandry is common. Definitive hosts become infected by ingesting contaminated flesh, which leads to the excretion of eggs or gravid proglottids through their feces. Intermediate hosts, including humans, contract the infection by consuming these eggs. Human infections typically occur through exposure to contaminated food or water, often linked to unwashed products, infected animals, or contaminated soil.[1,2]

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Cystic HD is widespread globally, with prevalence rates varying from 1 to 200 cases/100,000 individuals. However, it is particularly endemic in regions such as South and Central America, the Middle East, Mediterranean countries, and certain areas of sub-Saharan Africa. While it can affect any organ in the body, the liver is the primary site of involvement in 52–77% of cases, followed by the lungs in 10–40%.[1,3] Isolated extrahepatic intra-abdominal lesions are rare.

This case report seeks to emphasize the rare occurrence of abdominal hydatid cysts in multiple abdominal structures, including the liver, spleen, omentum, broad ligament of the uterus, and pelvis. This unusual presentation necessitates a comprehensive evaluation of a wide array of differential diagnoses and highlights the need for a range of treatment approaches to effectively address this potentially fatal disease.

CASE REPORT

A 32-year-old female engaged in animal husbandry and farming presented with an 8-month history of a progressively enlarging lump in the lower abdomen, accompanied by intermittent abdominal pain and constipation. Bowel and bladder habits were normal.

However, the patient reported a reduced appetite and exhibited poor physical build. On clinical examination, multiple firm lower abdominal lumps, and splenomegaly were noted, with no tenderness upon palpation. Vital signs were stable.

Laboratory investigations revealed an absolute eosinophil count of 1.28 × 10^9/L, an erythrocyte sedimentation rate of 110 mm/h, and a C-reactive protein level of 32 mg/L. Enzyme-linked immunosorbent assay (ELISA) for native antigen B was positive. Routine hematological and biochemical tests were within normal limits. Abdominal ultrasonography and contrast-enhanced computed tomography (CT) identified multiple cystic lesions of varying sizes involving the liver, spleen, omentum, and pelvis (Figure 1a and b).

The patient was initiated on a 21-day course of albendazole 400 mg twice daily before undergoing exploratory laparotomy. The abdomen was accessed through a midline incision.

Intraoperative exploration revealed multiple encapsulated cystic masses located in the omentum, spleen, liver, broad ligament of the uterus, and pelvic cavity (Figure 2a and b). Each cyst was meticulously excised through careful dissection to avoid spillage. To further minimize the risk of intraperitoneal dissemination, a surgical mop soaked in 10% povidone-iodine was

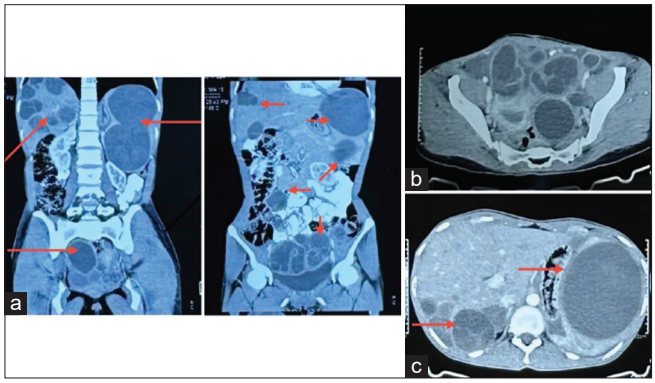

Figure 1: Contrast-enhanced computed tomography of the abdomen and pelvis in coronal (a) and axial sections (b) shows multiple cystic lesions in the liver, spleen, peritoneal cavity, and the pelvic cavity

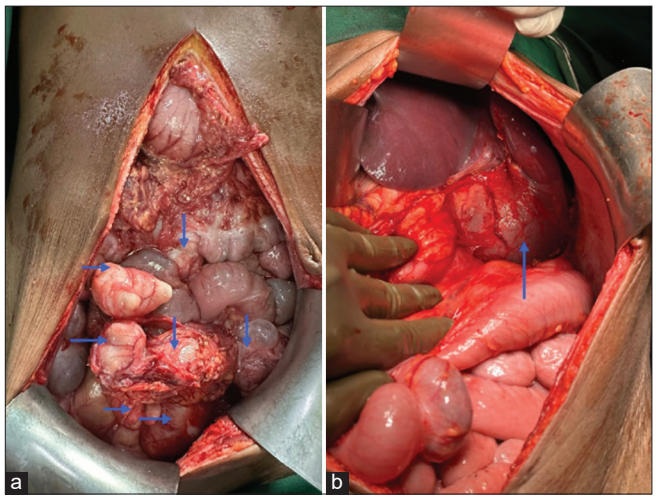

Figure 2: (a) Intraoperative images show multiple intra-abdominal masses (b) Intraoperative images show a giant splenic hydatid cyst

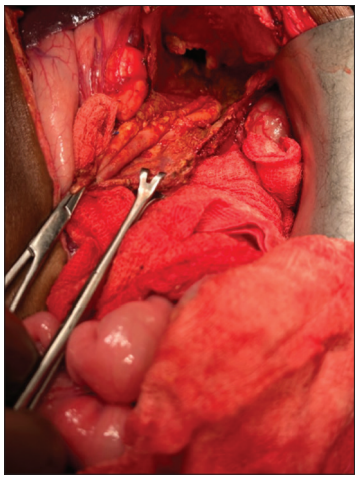

Figure 3: Drained and deroofed giant splenic hydatid cyst

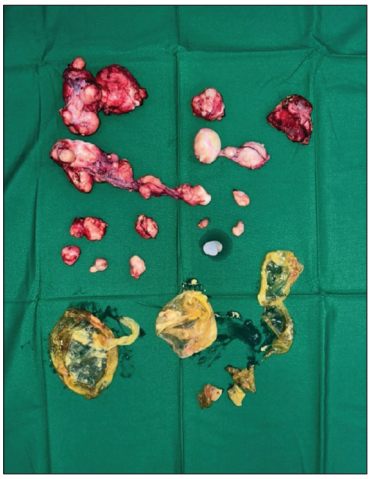

Figure 4: Excised specimen of multiple hydatid cysts

strategically placed around the cystic lesions in the liver and spleen. Scolicidal 20% hypertonic saline was injected into the cyst cavities and allowed to act for 15 min before the cysts were opened. The liver and splenic cysts were carefully drained, and their roofs were excised without any peritoneal contamination (Figure 3). Surgical drains were inserted into the hepatic, splenic, and pelvic regions to facilitate post-operative drainage. Excised specimen (Figure 4) sent for histopathological examination.

The patient experienced an uneventful post-operative recovery and was discharged on the 7th post-operative day following drain removal. She was prescribed a 4-month course of albendazole and praziquantel therapy to prevent recurrence. Histopathological examination confirmed the diagnosis of HD. Follow-up with abdominal ultrasonography was recommended every 3 months for 2 years to monitor for recurrence or complications.

At the 1-year follow-up, the patient remained asymptomatic, demonstrating favorable post-operative outcomes and disease control.

DISCUSSION

Hydatidosis is a zoonotic parasitic infection caused by tapeworms within the class Cestoda, family Taeniidae, and genus *Echinococcus*. The species *E. granulosus*, responsible for cystic hydatidosis, exhibits a nearly global distribution. Regions including South America, Central Asia, and the Mediterranean basin are recognized as highly endemic areas for this infection.[1,3]

The adult *Echinococcus* tapeworm resides within the small intestine of its definitive hosts, primarily canines such as dogs and other related species. Gravid proglottids, containing infective eggs, are excreted daily in the feces of these hosts. Upon ingestion by an appropriate intermediate host – typically herbivores such as sheep, goats, swine, cattle, horses, camels, and occasionally humans – the eggs hatch in the small intestine, releasing a larval form known as the oncosphere. The oncosphere, armed with six hooks, penetrates the intestinal mucosa and migrates through the circulatory system to various organs, with the liver and lungs being the most common sites of infestation.[4] Definitive hosts, including canids, become infected by ingesting the cyst-containing organs of parasitized herbivores.[4-6]

Humans, serving as accidental hosts, become infected by ingesting *Echinococcus* eggs. The World Health Organization-Informal Working Group on Echinococcosis classifies cystic echinococcosis (CE) into three categories based on ultrasonographic findings: Active (CE1 and CE2), transitional (CE3), and inactive (CE4 and CE5). CE1 and CE2 types are typically fertile and contain viable daughter cysts, whereas CE3 cysts often begin degeneration and may still contain daughter cysts. In contrast, CE4 and CE5 types are generally non-fertile. Given the fertility of CE1 and CE2 cysts, surgical or traumatic rupture poses a significant risk for peritoneal dissemination of the disease.[4,6]

HD most frequently involves the liver (60–70% of cases) and lungs (10–25% of cases), but it can affect any organ, including the brain, bones, and, in rare instances, the pelvic region, as exemplified by the present case report. Isolated extra-hepatic intra-abdominal hydatid cysts are an infrequent occurrence, accounting for 6–11% of cases.[5,6]

Clinical features are contingent upon factors such as the number, size, and anatomical location of the cysts. Typical clinical presentations include non-specific symptoms such as abdominal discomfort, hepatomegaly, splenomegaly, and, in the event of cyst rupture, anaphylactic reactions.[7]

The diagnosis of HD is based on a comprehensive approach that integrates clinical evaluation, serological tests, and advanced imaging modalities. Serological assays, such as the ELISA and indirect hemagglutination assay, detect specific antibodies against *Echinococcus*.[8] Imaging modalities, including ultrasound, CT, and magnetic resonance imaging (MRI), are pivotal in the diagnosis and evaluation of hydatid cysts.

Ultrasound is frequently the initial imaging technique of choice due to its widespread availability, gold standard, cost-effectiveness, and capability to distinguish between cystic and solid lesions.[9] Characteristic ultrasonographic features indicative of hydatid cysts include echogenic hydatid sand,

often referred to as the "snowflake sign," unilocular cysts containing daughter cysts, known as the "honeycomb sign," and cysts with a floating detached laminated membrane, referred to as the"water-lily sign."[9]

In contrast, CT scans are particularly effective in detecting calcifications and assessing bone involvement. MRI is considered the preferred imaging modality for HD due to its superior ability to reveal most of the disease's features, except for calcifications.[9,10]

The management of extrahepatic intra-abdominal HD requires a multidisciplinary approach that combines both medical therapy and surgical intervention. The cornerstone of medical treatment involves the use of benzimidazole derivatives, such as albendazole, to reduce the risk of recurrence and prevent systemic dissemination of the infection.[11] A typical treatment regimen includes a pre-operative course lasting 3 weeks to minimize the risk of anaphylactic reactions during surgery, followed by a post-operative course extending for several months to prevent recurrence of disease.[4]

Surgical intervention remains a critical component of the definitive treatment for hydatid cysts.[12] The selection of the surgical approach is primarily determined by the cyst's location, size, and involvement with adjacent structures.[12] Common surgical techniques include peri-cystectomy, cystectomy, and total organ resection, all aimed at achieving complete cyst removal while preserving the function of the affected organ. However, surgical management carries inherent risks, such as infection, cyst rupture, anaphylactic shock, and the potential for recurrence.[11]

Alternative treatment modalities include percutaneous approaches, such as the puncture, aspiration, injection, respiration (PAIR) technique, radiofrequency thermoablation, or, in some cases, conservative management.[13] In the PAIR method, the scolicidal agent is injected and aspirated. This technique is primarily indicated for type I and II cysts that are smaller than 6 cm and is not suitable for calcified or solid cysts.[14]

Reported recurrence rates for intra-abdominal hydatid cysts range from 2% to 30%. Regular follow-up, involving clinical evaluation and imaging, is essential for the early detection of recurrence. Follow-up typically includes imaging at 3-month intervals for a minimum of 3 years to monitor for cyst recurrence.[14,15]

CONCLUSION

This case highlights the importance of considering echinococcal disease in the differential diagnosis of any cystic mass, irrespective of its anatomical location, particularly in regions where the disease is endemic. This report emphasizes the diagnostic and therapeutic challenges associated with extrahepatic intra-abdominal hydatid cysts. A comprehensive approach, combining clinical evaluation, serological testing, and imaging techniques, is essential for accurate diagnosis. Surgical excision remains the treatment of choice, complemented by post-operative therapy with albendazole and praziquantel to prevent recurrence. Management requires a multidisciplinary strategy that integrates medical therapy and surgical intervention, aiming for complete cyst removal while minimizing the risk of complications.

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BLB- Literature search, prepared first draft of manuscript, implementation of study protocol, data collection, manuscript preparation; **ZH-** Literature search, data collection, manuscript preparation, editing, and manuscript revision; **SLG-** Review manuscript, manuscript revision, and submission of manuscript.

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