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Case Report

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Liver Hydatid Cyst Presenting as a Cystocutaneous Fistula in a 6-Year-Old Child

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Abstract

Hydatid disease, or cystic echinococcosis, is a zoonotic parasitic infection caused by the larval stage of *Echinococcus granulosus* [1]. While the liver and lungs are the most commonly affected organs, hydatid cysts in atypical locations such as the abdominal wall are rare and often misdiagnosed. This case report describes a six-year-old male from Alsahol, Ibb, Yemen, presenting with a painless epigastric mass persisting for three weeks. Initial imaging suggested an abdominal wall abscess, but further investigation with Computed Tomography (CT) revealed hydatid disease complicated by fistula formation. The patient was treated with Abandazol preoperatively and scheduled for surgical intervention. This case emphasizes the importance of high clinical suspicion, detailed imaging, and a multidisciplinary approach to manage rare presentations of hydatid disease in endemic regions.

Keywords: Hydatid cyst, Cystic echinococcosis, Echinococcus granulosus, Cystocutaneous fistula, Pediatric hydatid disease, Abdominal wall hydatid cyst, Zoonotic parasitic infection, Rare presentation, Albendazole therapy, Surgical ectocystectomy, Endemic parasitic diseases, Public health

Introduction

Hydatid disease, also known as cystic echinococcosis, is a chronic parasitic infection caused by the larval stage of *Echinococcus granulosus*. It remains a significant public health concern in endemic regions such as the Middle East, North Africa, Central Asia, and South America, where close human-livestock interactions and poor sanitation facilitate transmission [1]. Humans, as accidental intermediate hosts, acquire the infection by ingesting eggs excreted in the feces of infected dogs, the definitive host. Once ingested, the eggs hatch in the small intestine, releasing *oncospheres* that penetrate the intestinal wall and migrate *via* the bloodstream to various organs, where they develop into hydatid cysts [2].

The liver and lungs are the most frequently affected organs, accounting for approximately 60-75% and 15-20% of cases, respectively [3]. However, hydatid cysts in atypical locations, such as the epigastric region with *cystocutaneous fistula* formation, are exceedingly rare, accounting for less than 5% of cases and often mimicking abscesses, hernias, or tumors [4]. These atypical presentations are further complicated by a lack of specific symptoms, with clinical manifestations typically arising due to mass effect, rupture, or secondary infection [5].

The diagnosis of hydatid disease involves imaging modalities such as ultrasonography, Computed Tomography (CT) and Magnetic Resonance Imaging (MRI), combined with serological tests to detect *Echinococcus granulosus*-specific antibodies [1]. Albendazole, a benzimidazole compound, is often used preoperatively to reduce cyst viability and minimize the risk of recurrence [5]. Surgical excision remains the cornerstone of treatment, particularly for complicated cases with fistula formation or other secondary complications.

This case report highlights the diagnostic and management challenges of a rare presentation of hydatid cyst involving the abdominal wall in a six-year-old male from Yemen, emphasizing the importance of comprehensive evaluation and multidisciplinary care.

Case Presentation

Patient information

- **Age:** 6 years
- **Gender:** Male
- **Residence:** Alsahol, Ibb, Yemen
- **Presentation:** Painless epigastric mass persisting for three weeks.

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- **Chief complaint:** Painless epigastric swelling without associated symptoms, aggravating, or relieving factors.
- **Medical history:** No prior history of similar complaints or chronic conditions.
- **Family history:** No known family history of parasitic diseases or chronic illnesses.
- **Social history:** The patient resides in a rural area with frequent exposure to livestock and domestic dogs. Poor hygiene practices are likely risk factors for parasitic infections. The patient's family reported no recent travel outside of Yemen, and the child had no exposure to additional risk factors beyond the endemic environment.

Clinical examination

General appearance: Alert, cooperative six-year-old male with no acute distress.

Vitals:

- Blood Pressure: 100/65 mmHg.
- Pulse: 90 beats per minute (regular).
- Temperature: 36.8°C (afebrile).
- Respiratory Rate: 18 breaths per minute.
- Oxygen Saturation: 98% on room air.

Abdominal examination:

- Inspection: Visible swelling in the epigastric region without erythema or signs of inflammation.
- Palpation: Firm, non-tender, well-circumscribed mass in the epigastric region. No fluctuation or localized warmth.
- Percussion: Dullness over the mass.
- Auscultation: Normal bowel sounds.

Investigations

Ultrasound (Initial):

- **Findings:** The ultrasound, performed on 05/11/2024, revealed a hypoechoic lesion in the epigastric region, initially interpreted as an abdominal wall abscess. The liver, kidneys, spleen, pancreas, and other abdominal structures appeared normal. Further evaluation with CT was recommended.

CT scan (Follow-up)

Findings:

- Multiple cystic lesions within the abdominal wall, with evidence of daughter cysts suggestive of hydatid disease.
- Cysts display the "water lily sign," characteristic of hydatid cysts, indicating detachment of the germinal layer.
- No involvement of liver, lungs, or other visceral organs.
- Fistulous tract observed during follow-up evaluation, connecting the cyst to the external skin surface.
- The cystic lesions are well-defined, multilocular, and show rim enhancement on contrast.
- Adjacent inflammatory changes noted, likely due to the presence of the fistula and secondary infection.

Laboratory findings

- White Blood Cell (WBC) Count: 5×10^9/L (within normal range).
- Eosinophil Count: 8% (elevated, consistent with parasitic infection).
- Neutrophils: 60% (normal range: 40-70%).
- Basophils: 0.5% (normal range: 0-1%).
- Lymphocytes: 30% (normal range: 20-40%).
- Hemoglobin (HB): 13 g/dL (within normal range).
- Platelet Count (PLT): 420×10^9/L (slightly elevated, likely reactive to inflammation).

Clinical correlation: The laboratory results, notably the elevated eosinophil count, align with a parasitic etiology consistent with hydatid disease.

Diagnosis

- Initial Diagnosis: Abdominal wall abscess.
- Final Diagnosis: Hydatid cyst involving the abdominal wall with fistula formation.

Management

Pharmacological Treatment:

- Albendazole: Administered at 15 mg/kg/day for one week to reduce cyst viability, minimize the risk of intraoperative spillage, and prevent recurrence [5].

Surgical management:

Procedure: Complete resection of the cyst along with its fistulous tract was performed. The operation involved ectocystectomy, ensuring no residual cyst cavity remained to manage.

Post-surgical steps: The liver edge was primarily sutured to restore anatomical integrity and prevent bile leakage or complications.

Outcome: The surgical intervention successfully removed the hydatid cyst and its complications, including the fistulous connection, with no immediate postoperative complications.

Follow-Up

Post-albendazole follow-up: Formation of a fistula prompted additional imaging studies, confirming the diagnosis of a hydatid cyst.

Post-surgical follow-up: After complete resection and ectocystectomy, the patient demonstrated stable recovery with no bile leakage or postoperative complications. The surgical site is healing appropriately, and follow-up imaging has been scheduled to monitor for recurrence. The patient's family was educated on hygiene, deworming household pets, and preventive measures to minimize the risk of reinfection.

Current Status: The patient remains stable with no signs of recurrence or postoperative complications during follow-up (Figure 1-7).

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Figure 1: Abdominal wall hydatid cyst with fistula formation.

Note: Clinical photograph of the patient's abdomen showing a visible epigastric swelling with discoloration, central ulceration, and evidence of fistula formation. This presentation is consistent with a complicated hydatid cyst involving the abdominal wall.

Figure 2: Excised hydatid cyst with fistulous connection.

Note: (A) The surgically excised hydatid cyst showing an intact structure with visible inflammation and a fistulous tract extending from the cystic mass. (B) The hydatid cyst post-excision with visible evidence of the attached fistula and inflammatory changes. (C) A detailed view of the cyst, highlighting the complex structure, including fibrotic and infected components.

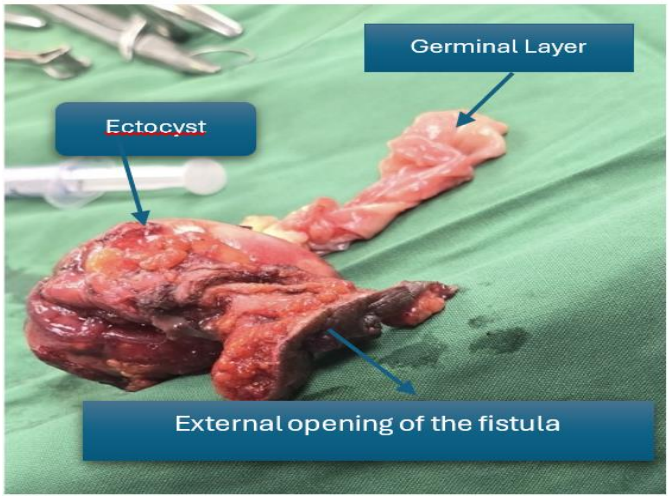


Figure 3: Annotated image of the excised hydatid cyst and fistula.

Note: Surgical photograph of the excised hydatid cyst showing:

- Ectocyst (outer layer): Marked for visualization.
- Germinal layer: Highlighted to demonstrate internal structural integrity.
- External opening of the fistula: Clearly visible, marking the site of fistulous communication.

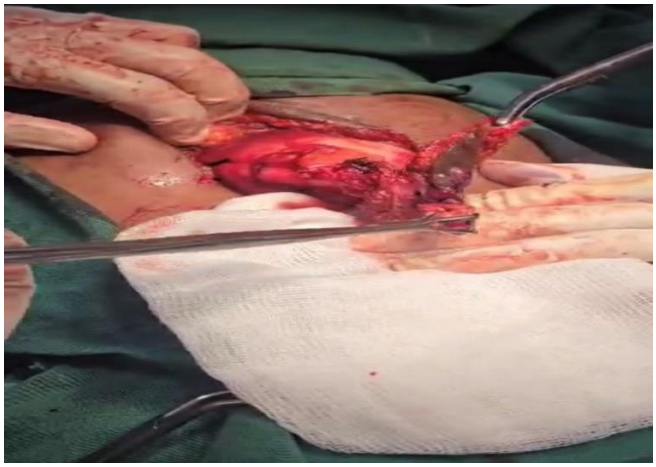
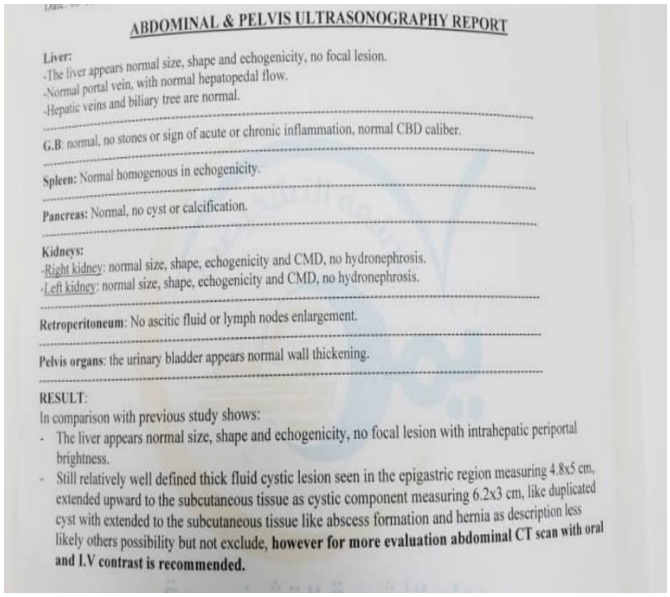


Figure 4: Intraoperative visualization of the hydatid cyst and fistula.

Note: Intraoperative photograph showing the surgical exposure of the hydatid cyst within the abdominal wall. The image highlights the connection between the cyst and the fistulous tract, with surrounding tissue dissection and meticulous surgical handling.



ABDOMINAL & PELVIS ULTRASONOGRAPHY REPORT

Liver:
-The liver appears normal size, shape and echogenicity, no focal lesion.
-Normal portal vein, with normal hepatopedal flow.
-Hepatic veins and biliary tree are normal.

G.B: normal, no stones or sign of acute or chronic inflammation, normal CBD caliber.

Spleen: Normal homogenous in echogenicity.

Pancreas: Normal, no cyst or calcification.

Kidneys:
-Right kidney: normal size, shape, echogenicity and CMD, no hydronephrosis.
-Left kidney: normal size, shape, echogenicity and CMD, no hydronephrosis.

Retroperitoneum: No ascitic fluid or lymph nodes enlargement.

Pelvis organs: the urinary bladder appears normal wall thickening.

RESULT:
In comparison with previous study shows:
- The liver appears normal size, shape and echogenicity, no focal lesion with intrahepatic periportal brightness.
- Still relatively well defined thick fluid cystic lesion seen in the epigastric region measuring 4.8x5 cm, extended upward to the subcutaneous tissue as cystic component measuring 6.2x3 cm, like duplicated cyst with extended to the subcutaneous tissue like abscess formation and hernia as description less likely others possibility but not exclude, **however for more evaluation abdominal CT scan with oral and I.V contrast is recommended.**

Figure 5: Abdominal and pelvis ultrasonography report.

Note: Ultrasound report highlighting a well-defined thick fluid cystic lesion in the epigastric region, measuring 4.8×5cm, with an extension into the subcutaneous tissue, mimicking an abscess or hernia. Further evaluation with CT and contrast was recommended for confirmation.

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Figure 6: CT image showing the "water lily sign" of hydatid cyst. **Note:** Axial computed tomography (CT) scan of the abdomen displaying the "water lily sign," a characteristic feature of hydatid cysts. The sign represents the detachment of the germinal layer from the pericyst, forming floating membranes within the cyst cavity. This finding is pathognomonic of hydatid disease and confirms the diagnosis.

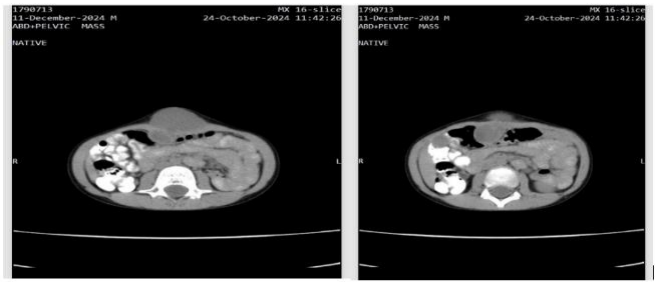

Figure 7: CT image showing the anterior abdominal wall cyst with communication to the liver cyst.

Note: Axial Computed Tomography (CT) scans of the abdomen reveal a cystic lesion in the anterior abdominal wall, with evidence of communication to an adjacent liver cyst. This finding confirms the extension of the hydatid cyst and its involvement across multiple anatomical structures, illustrating the complexity of the disease.

Discussion

This case of hydatid cyst in the abdominal wall demonstrates the diagnostic complexity of atypical hydatid disease. Abdominal wall involvement is a rare manifestation, accounting for less than 5% of cases, and is often misdiagnosed as abscesses or neoplastic conditions [4]. The absence of systemic symptoms further complicates early identification.

The patient's residence in Alsahol, Ibb, Yemen, is significant as it highlights the endemic nature of hydatid disease in rural Yemen. Factors such as close interaction with livestock, poor sanitation, and limited access to healthcare contribute to the high prevalence of hydatid disease in the region. In Alsahol, reliance on livestock farming, coupled with poor hygiene practices and close contact with 7.

domestic dogs, facilitates the transmission of *Echinococcus granulosus*. Additionally, inadequate public health education and limited veterinary care perpetuate the risk of infection.

Albendazole plays a critical role in preoperative preparation, reducing cyst viability and minimizing intraoperative complications [5]. Surgical intervention remains the definitive treatment, particularly for complicated presentations with fistula formation or secondary infections [6].

This case underscores the need for heightened clinical suspicion in endemic areas, particularly in children presenting with atypical cystic lesions. Public health measures, including deworming programs, improved hygiene practices, and health education, are critical to mitigating the burden of hydatid disease in rural communities like Alsahol.

Conclusion

Hydatid disease in atypical locations, such as the abdominal wall, presents unique diagnostic and therapeutic challenges. This case illustrates the importance of thorough evaluation and appropriate management, particularly in endemic regions. Public health measures, including education, improved sanitation, and deworming programs, remain crucial in reducing the disease burden.

Ethical Considerations

Informed consent was obtained from the patient's parent for the publication of this case report. The consent includes permission to share anonymized details of the patient's medical history, diagnostic findings, and treatment outcomes. All identifying information in the clinical and surgical images has been anonymized to ensure patient confidentiality.

Competing Interests

The authors report no conflicts of interest in this work.

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