



CASE REPORT

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Proximity predicament: a rare case of hydatid cyst in the porta hepatis region

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Abstract

Hydatid cysts near the porta hepatis present notable medical challenges due to their intricate positioning and potential influence on crucial structures within the liver and its vicinity. While relatively rare, their presence can lead to intricate medical scenarios because of their proximity to vital liver and bile duct structures, but their occurrence can present complex medical situations due to their proximity to critical structures in the liver and bile ducts. We present an unusual case of a 33-year-old male with no known comorbidities presented to the outpatient department of general surgery with pain, insidious in onset and gradually progressive, in the right hypochondrial region for the past 3 months, along with mild fever, reduced appetite, and weight loss of 3 kg. Managing hepatic cysts, especially when near the porta hepatis, poses significant diagnostic challenges. Imaging scans revealed multiple hypodense lesions suggestive of hepatic cysts, with the largest one measuring 5 cm × 8 cm in segment VI. Native post-contrast lesions were also observed in the right lower lobe. Although hepatic duct dilation was not observed, the presence of a cyst near the porta hepatis an atypical site for hydatid cyst, complicated potential PAIR procedures (puncture, aspirate, injection, re-aspirate), raising concerns about the risk of damaging vital structures like the hepatic artery or major bile duct. A multidisciplinary approach engaged interventional radiology to evaluate PAIRS feasibility. PAIRS was performed successfully, except for the cyst near the porta hepatis to avoid complications. Despite this, the patient continued to experience abdominal discomfort post-operation, although liver function tests improved in the postop period. This case underscores the uncommon occurrence of hydatid cysts near the porta hepatis and highlights the need for additional research to comprehend its prevalence and causes. Further investigation is crucial to enhance our understanding of the epidemiology and optimal management strategies involving PAIRS in such challenging cases.

Keywords Case report, Hydatid cyst near porta hepatis, PAIRS procedure

Introduction

A zoonosis called hydatidosis is caused by the larval form of *Echinococcus*. Humans serve as an unintended intermediate host for the development of cystic lesions, so-called hydatid cysts [1]. In 75% of cases, hydatid cysts infect the liver, making it the most vulnerable organ in the body. However, when hydatid cysts develop, the right lobe of the liver is affected more often than the left [2]. It is distributed throughout the world but is particularly common in endemic regions including the Mediterranean, Australia, New Zealand, North Africa, Eastern Europe, the Balkans, the Middle East, and South America

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[3]. Clinical suspicion is required for the diagnosis of an uncomplicated hydatid cyst in the liver. The two primary diagnostic techniques, computed tomography, and ultrasound are useful for detecting complications and developing a treatment plan [3, 4]. Diffusion-weighted magnetic resonance imaging (MRI) is helpful in distinguishing between simple and hydatid cysts in the liver [5].

Medical therapy, percutaneous drainage, or surgery may be used to treat hydatid liver cysts [6]. Usually, when surgery is contraindicated, drug therapies are used, but a proven drug such as albendazole is effective in adjuvant therapy before surgery and reduces the cyst recurrence rate [6, 7]. Ultrasound-guided percutaneous drainage of the cyst has become very reliable and widely used following the introduction of percutaneous aspiration injection and re-aspiration (PAIR) and its modifications [8]. Laparoscopic, conservative, and radical approaches are available for surgical procedures [6]. All interventions are considered safe and effective. However, the recurrence and complication rate with the radical approach is significantly low, making it a treatment of choice over other procedures [9, 10].

Hydatid liver cysts, if left untreated, can progress to frequently occurring complications like hydatid cyst superinfection, intra-biliary rupture, and direct rupture into the thoracic or abdominal cavities [11]. Rupture of the cyst into hollow viscera, abdominal wall invasion, and hepatic vasculature-related complications such as portal vein thrombosis, Budd-Chiari syndrome, peritoneal seeding, and hematogenic spread are serious but rare complications of hydatid liver cyst [11, 12].

Timeline

2023-07-23 33-year-old male, RUQ pain for 2–3 months. CT shows multiple liver lesions, largest in segment VI, likely hepatic hydatid cysts. Advised SIP excision; considering PAIRS for further evaluation

Narrative

Patient information

A 33-year-old male with no known comorbidities presented to the outpatient department of general surgery with pain, insidious in onset and gradually progressive, in the right hypochondrial region for the past 3 months, along with mild fever, reduced appetite, and weight loss of 3 kg.

The patient's symptoms continued to worsen despite undergoing treatment with multiple antibiotics. There was no significant past medical or surgical history.

Clinical findings

Normal height and build were noticed upon physical examination. The patient exhibited no signs of irritation apart from severe abdominal distension and minimal tenderness.

Laboratory findings: a leukocyte count of $7.5 \times 10^3/\text{UL}$ (within the range of 4.3–10.3), eosinophil percentage at 5.8% (within the range of 0.9–6), aspartate aminotransferase level at 78 U/L (normal range 5–34), alanine aminotransferase level at 133 U/L (normal range 0–55), gamma-glutamyl transferase at 63 U/L (normal range 9–64), alkaline phosphatase at 97 U/L (normal range 40–150), total bilirubin at 2.2 mg/dL (normal range 0.2–1.2), Ca 19–9 at 8.5 U/mL (normal range 0–35), and HBsAg at 3201 S/CO. Subsequently, the ELISA for antiEchinococcus antibodies returned positive titers.

Diagnostic assessment

The imaging scans with contrast revealed multiple hypodense lesions pleading the presence of hepatic cysts, with the largest one measuring 5 cm × 8 cm in segment VI as shown in Fig. 1, a few native lesion post-contrast lesions were visible in abdominal slices, primarily located in the right lower lobe of the liver.

Dilation of the hepatic ducts was not noted. However, one of the cystic lesions was in close proximity to porta hepatitis, an atypical site for hydatid cysts as shown in Fig. 2, which posed a challenge for PAIR procedures (puncture, aspirate, injection, respire) with a significant risk of damaging the hepatic artery or major bile duct.

Diagnosis

The imaging scans with contrast revealed multiple hypodense lesions pleading the presence of hepatic cysts, one of the cystic lesions was in close proximity to porta hepatitis, an atypical site for hydatid cyst.

Therapeutic interventions

The patient was initially started on albendazole 4 weeks; however, deterioration of liver functions was anticipated as a side effect of albendazole; an ultrasound was done to rule out intrahepatic or perihepatic collections due to cyst rupture, which was normal. At this stage, a multidisciplinary approach was taken and interventional radiology was taken on board to assess for possible PAIRS. PAIRS was possible for cysts other than one that was in close proximity to porta hepatitis to avoid any complications. The patient proceeded with a PAIR procedure for definitive management as per the plan. PAIRs were done by utilizing ultrasound to guide puncture of the cyst, destruction of the germinal layer with protoscolices using chlorhexidine-cetrimide

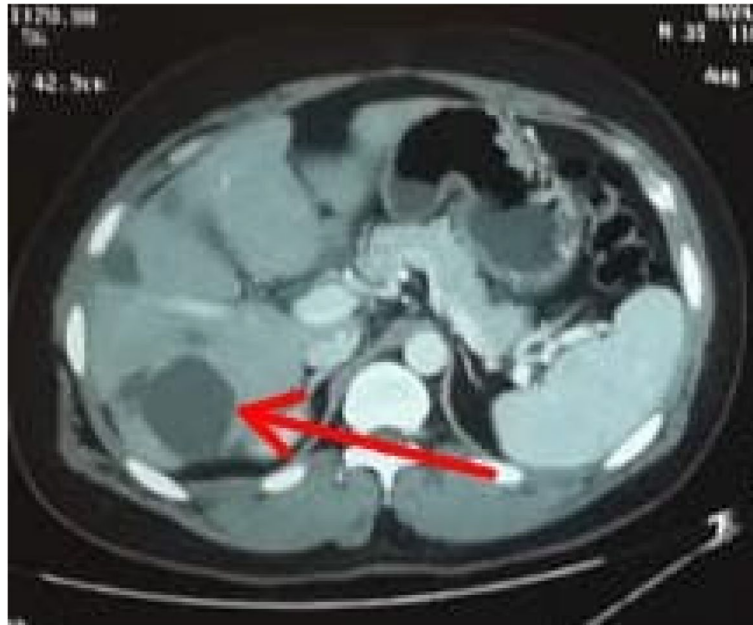


Fig. 1 Axial CT showing hypodensity (red arrow) showing 5 cm × 8 cm on segment VI hydatid cyst

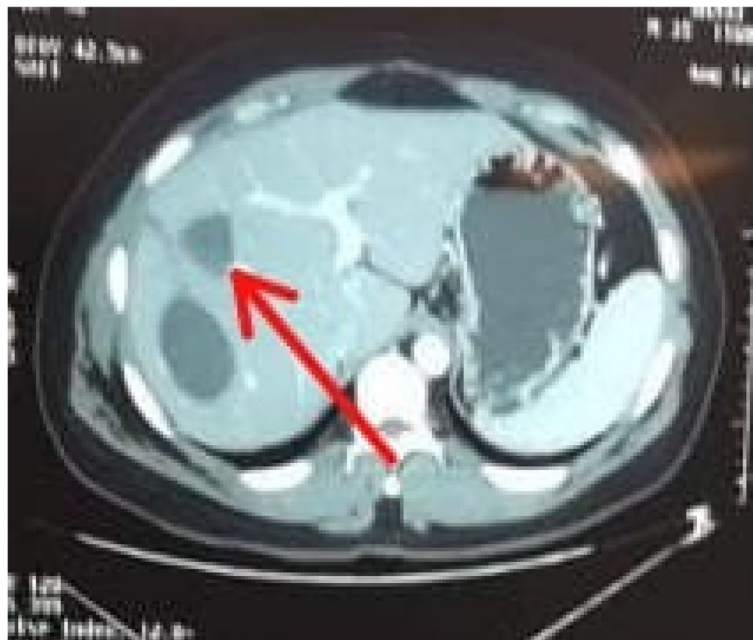


Fig. 2 Axial CT showing hypodensity (red arrow) near porta hepatis depicting hydatid cyst

solution was used as the scolical agent, and reaspiration of this fluid after 15–20 min. Following the procedure, the patient continues to experience persistent abdominal distress, which is inconsistent with the normalization of the liver function test.

Discussion

The most typical site of presentation for hydatid disease, a zoonotic illness brought on by the *Echinococcus granulosus* species, is the liver [13]. Dogs and foxes are the disease's definitive hosts, whereas sheep, goats, camels,

and pigs are its intermediate hosts. The accidental hosts include humans, who may become infected by eating contaminated vegetables or by unintentional contact with hosts like pets [14]. Hydatid cysts can form slowly and in multiples; the mature cyst has three layers: an adventitial, a laminated, and an inner germinal layer [15].

Most of the time, cysts are asymptomatic or cause nonspecific symptoms in patients; however, the actual issue occurs when complications such as cyst rupture or perforation occur [16], which is why early management is typically advised to lower the morbidity. Hydatid disease symptoms vary depending on where the cysts are located. For example, pulmonary cysts can cause cough, dyspnea, chest discomfort, and hemoptysis [17], whereas hepatic cysts, like the one in our case, can cause right hypochondriac pain, weight loss, disordered LFTs, and other symptoms. Imaging modalities used for diagnosis are ultrasound abdomen and contrast-enhanced CT scans that show cystic lesions. Radiologically cysts can be classified into five types, including simple cysts (type I) with no internal architecture, and complex cysts with internal architecture. Type II cysts have daughter cysts and matrix, with three subtypes: type IIa featuring round daughter cysts at the periphery, type IIb featuring larger, irregularly shaped daughter cysts, and type IIc featuring oval masses with scattered calcifications and occasional daughter cysts. Type III cysts are calcified, indicating a dead or non-viable cyst, while type IV cysts are complicated and may have ruptured or undergone other significant changes [18].

There have been reports of four therapeutic modalities: (1) surgery; (2) puncture, aspiration, injection, and re-aspiration; (3) anti-worm medication chemotherapy using albendazole or mebendazole; and (4) observation and waiting [19], of which surgery has been shown to be the most successful technique; nevertheless, in cases in which the position of the cyst precludes surgery, medicinal treatment has nonetheless been shown to be a successful alternative [20]. The patient in our case had many hepatic cysts, one of which was next to the porta hepatis. This presented a problem for the PAIR technique since there was a chance that the cyst might rupture the hepatic artery or bile duct, increasing the patient's risk of death. The three traditional surgical approaches that are employed are hepatectomy, partial pericystectomy, and cystectomy. After these surgeries, additional modalities such as external drainage, bipolar drainage, cavity drainage via anastomosis with the stomach or jejunum, etc. are typically used to treat the residual cavity [3].

To make the parasite inactive, hypertonic saline, ethyl alcohol, hydrogen peroxide, or albendazole is injected before cavity opening [3]. A well-known side effect that our patient experienced was an albendazole-induced

idiosyncratic, hepatocellular pattern of hepatotoxicity, characterized by elevated bilirubin levels [21]. However, ultrasonography was performed to rule out other possible causes of increased LFTs, such as cyst rupture, and the results were negative, suggesting a pharmaceutical side effect. After that, the patient was treated according to the PAIR procedure's first treatment plan. A meta-analysis [21] indicates that combination therapy, such as albendazole plus praziquantel, yields superior outcomes. However, the data indicating this medication's efficacy is not yet convincing enough, and more studies are required to draw a firm conclusion. No post-treatment problems were noted in our patients; however, despite therapy, a persistent complaint of mild pain persisted, which may have been caused by the cyst that was left untreated because of its delicate placement.

Patient perspective

The patient, fully aware of their terminal illness, has willingly chosen palliative care, prioritizing comfort and quality of life.

Conclusion

This case underscores the exceptional nature of a hydatid cyst near the porta hepatis and its impact on treatment, notably complicating PAIRS procedures. It stresses the urgency for further research to refine approaches and elevate success rates, particularly in handling hydatid cysts positioned near the porta hepatis, ensuring improved patient outcomes and procedural efficacy. Advancements in understanding and tailored approaches are crucial to ensure higher success rates and better outcomes for patients encountering these atypical and typical hydatid cyst locations.

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Compliance with instruction to authors

We hereby affirm that this manuscript has been meticulously prepared in strict accordance with all prescribed instructions provided to the authors.

Publication status

We certify that this manuscript is entirely original and has not been published previously, nor is it currently under consideration by any other journal.

Authors' contributions

Abdul Raheem collected information. Abdul Raheem Aashish Kumar, Suhaib Ahmed, Syed Ali Aarsal, Syed Muhammad Sinaan Ali, and Syeda Areeba prepared the manuscript which has been thoroughly reviewed by Syeda Areeba and Abdul Raheem.

Declarations

Ethics approval and consent to participate

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

Consent for publication

We confirm that the authorship requirements have been diligently met, and the final version of the manuscript has been unanimously approved by all contributing authors.

Competing interests

The authors declare that they have no competing interests.

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