Patient Safety in Surgery

CASE REPORT

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Safe surgical management of two concomitant giant hepatic hydatid cysts: a case report

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Abstract

Background Hydatid disease (HD) is a parasitic infection caused by the larval stage of the tapeworm Echinococcus granulosus. The liver is primary affected, accounting for up to 70% of cases, but it can also affect other organs such as the spleen, pelvic cavity, and bones. Patients can remain asymptomatic for 10 to 15 years. This report, accompanied by an extensive literature review, aims to contribute to the understanding of the diagnostic and surgical management of giant hepatic hydatid cysts, given their rarity and the variety of locations in the body where they occur.

Case presentation A 32-year-old male presented with a 3-day history of diffuse abdominal pain. Examination revealed a tense abdomen, and laboratory tests showed mild leukocytosis. Ultrasound and CT scan demonstrated two giant hepatic hydatid cysts. The patient underwent elective open surgery with aspiration of both cysts, instillation of a scolicidal solution, removal of the germinative membranes, and partial pericystectomy with preservation of hepatic tissue. A drain was placed, and the postoperative course was uneventful. He was discharged after 3 days of hospitalization. Albendazole therapy was initiated 2 weeks postoperatively and continued for 6 months. At 6-month follow-up, the patient was asymptomatic with normal liver function tests and no evidence of recurrence on imaging.

Conclusion Our case highlights that despite the limited medical resources in conflict-affected Syria, hepatic hydatid cysts can be successfully managed effectively through a combination of radiological evaluation, open surgical intervention, and antiparasitic drug therapy, resulting in a significant improvement in patient outcomes.

Clinical trial number Not applicable.

Keywords Hepatic hydatid cyst, Echinococcus granulosus, Albendazole, Abdominal pain, Syria

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Background

Hydatid cysts are a parasitic disease caused by the larval stage of the tapeworm called Echinococcus granulosus, belonging to the genus Echinococcus [1]. Humans become accidental intermediate hosts when they ingest the parasite's eggs, usually through contaminated food or direct contact with infected dogs [2, 3]. Incidence rates are as high as 50 per 100,000 person-years with an estimated prevalence of 10% of the population in highly endemic areas relying on agriculture and animal husbandry - including Syria, where it still poses a serious public health problem [4]. While it primary targets the liver, making up to 70% of cases, it may be present in other organs, such as the lung, spleen, and pelvic cavity [5]. The right lobe of the liver is affected in 60–70% of hepatic cases [6]. Bone involvement is observed in a small number of cases, ranging between 0.5% and 2.5% of all hydatid disease cases. Spinal involvement may account for about 50% of these cases [7]. Patients are usually asymptomatic for a long period, which may extend from 10 to 15 years [8]. Diagnosis is a significant challenge because the morbidity rate is low, and surgical procedures vary depending on the location of the lesions [9].

Within this report, we're going to highlight a rare case of two giant hydatid cysts located on the inferior surface of the liver, through disclosing the clinical presentation, diagnosis, and a detailed treatment approach. The aim is to consider atypical conditions, to prevent fatal conditions from spreading.

Case presentation

A 32-year-old male presented to the general surgery clinic at Al-Mowasat University Hospital, one of the largest academic teaching hospitals in Damascus, Syria, with a history of abdominal pain for three days without any other accompanying symptoms. The pain was vague and diffuse, not accompanied by nausea, vomiting, fever, or jaundice. The patient weighs 61 kg and has a smoking history of 8 pack-years. In the patient's medical history, a right lower limb deep vein thrombosis (DVT) was mentioned, treated with Apixaban, with no other medical, surgical, or allergic history. Physical examination showed a tense abdomen with generalized dullness, without other accompanying signs. Vital signs were stable. Laboratory tests were within normal limits except for a slight increase in white blood cell count (see Table 1). Abdominal ultrasonography demonstrated two large hepatic hydatid cysts measuring 16 × 17 cm and 18 × 20 cm, respectively.

A computed tomography scan (CT scan) was performed to determine the relationship of the cystic lesions to the surrounding structures, revealing low-density, well-defined cystic formations within the liver that were radiologically consistent with hydatid cysts (see Fig. 1).

Table 1 Laboratory tests

Test	Result	Normal Range
WBC	12.3×10^9/L	$4–11 \times 10^9$/L
GRA	72.6%	40–70%
HGB	13 g/dl	13–16 g/dl
PLT	186×10^3/ml	$150–450 \times 10^3$/ml
Na	138 mmol/l	135–145 mmol/l
K	3.9 mmol/l	3.5–5.0 mmol/l
ALT	44 U/l	7–55 U/l
AST	79 U/l	8–48 U/l
Urea	16 mg/dl	10–50 mg/dl
Creatinine	0.8 mg/dl	0.6–1.3 mg/dl
PT	88%	70–100%
TB	0.68 mg/dl	0.1–1.2 mg/dl
DR	0.12 mg/dl	0–0.3 mg/dl
ALP	125 U/l	44–147 U/l
Amylase	35 U/l	30–110 U/l

The chest X-ray was unremarkable. Based on the surgeons' extensive experience in managing hydatid cysts, given their prevalence in Syria, and due to their large size in our patient, an appointment was scheduled for the patient to undergo open surgery after obtaining a vascular surgery consultation to manage venous thrombosis. A midline incision extending above and below the umbilicus was performed. Intraoperatively, a large cyst was found occupying the entire abdominal cavity (see Fig. 2). The gallbladder was dissected from the wall of the first cyst, which was then aspirated, yielding approximately 700 cc of cystic fluid. Aspiration also allowed us to assess for cystobiliary communication by identifying the color of the aspirated fluid. The cavity was injected with

Fig. 1 A CT scan image shows the presence of two hydatid cysts in the liver

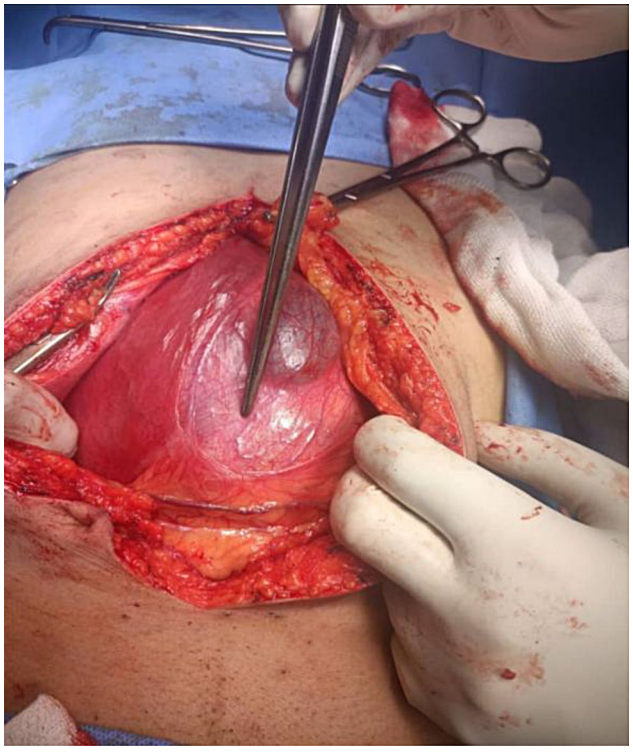

Fig. 2 Findings are consistent with a large inferior hepatic cyst inciting a surrounding fibrotic reaction, forming a distinct fibrous wall. The gallbladder demonstrates adherence to this fibrotic capsule

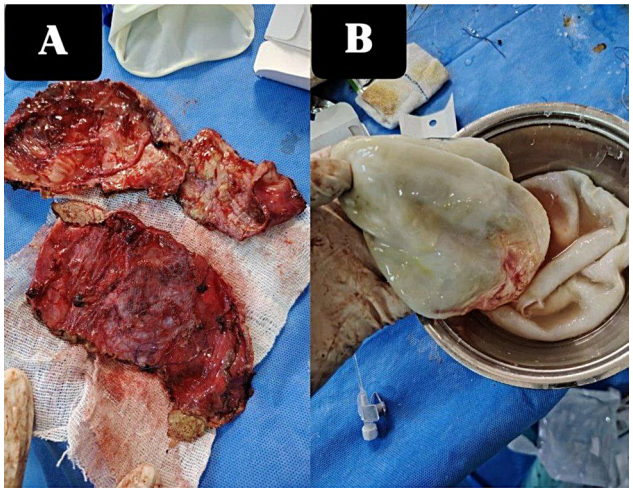

Fig. 3 **(A)** Intraoperative photograph showing the fibrotic reactive tissue surrounding the hepatic hydatid cysts. A large portion of this fibrotic capsule was excised to prevent postoperative fluid collection and abscess formation. **(B)** Intraoperative photograph demonstrating the excised germinal membranes of the cysts, which are responsible for producing daughter cysts. Their removal was performed to minimize the risk of recurrence

a hypertonic saline and left for 12 min before removing the germinative membrane. The fibrous wall covered by visceral peritoneum was excised as much as possible, while preserving the hepatic tissue. The second cyst was aspirated, yielding about 100 cc of cystic fluid, followed by injection of hypertonic saline and a 12-minute wait. The germinative membrane was then removed, and as much of the fibrous wall as possible was excised (see Fig. 3). A drain was placed, and the incision was closed. The patient was monitored with analgesic medications for three days post-surgery to ensure there was no biliary leakage. The patient was also prescribed albendazole at a dose of 10 mg/kg/day, starting two weeks after surgery. The treatment regimen lasted approximately six months, divided into three-week courses of treatment followed by a one-week break. The purpose of this treatment plan was to prevent recurrence, particularly in cases of minor leakage during surgery. We scheduled regular follow-up appointments with ultrasound scans every 6 months to detect any relapse or complications, such as abscess formation. In addition, liver enzyme tests were performed to assess the drug's effect on liver function, and all results were within normal limits.

Discussion

One of the leading zoonotic parasites within the Middle East is the genus Echinococcus, a tapeworm, which is responsible for hydatidosis. It's also prevalent in farm-rearing countries including Mediterranean countries, New Zealand, Australia and North Africa [10]. The World Health Organization (WHO) has included cystic echinococcosis in the list of neglected tropical diseases [11]. Recently, the scientific revolution and advances in artificial intelligence tools have led to the development and improvement of the accuracy of differential diagnoses, particularly the use of deep learning in predicting and differentiating liver cancers from liver cysts and other lesions [12]. The prevalence of hepatic cysts ranges between 0.1% and 0.5%, and less than 10–15% of patients suffer from significant clinical symptoms [13]. Our case differed from that of Adadi et al., whose patient presented with a gluteal muscle mass [14], and also differed from the case of Jellali et al., which showed symptoms of abdominal pain, jaundice, and fever due to rupture of the cyst into the biliary ducts [15].

In adults, the liver is the most common site of infection, while in children, the lungs are the most common [16] (see Table 2). The hydatid cyst can also affect the heart through the coronary circulation, with the left ventricle being the most commonly affected site, although other locations such as the interventricular septum or the right ventricular myocardium may also be involved [17]. A small proportion of hydatid cysts has also been observed in the subcutaneous tissue, which can be either secondary or primary. In secondary cases, the subcutaneous cyst should be associated with the presence of a hydatid cyst in one or more of the common sites of hydatid disease, such as the liver or the lungs [18]. The

Table 2 Representative literature-reported cases of hydatid cysts across different anatomical sites

Author	Sex	Age	Location	Cystic Size	Surgical Procedure
Our report	Male	32 years old	Two hydatid cysts in the liver	16×17 cm / 18×20 cm	Cystectomy + albendazole
Chtira et al. [30]	Female	3 years old	Intra-orbital	28×18 mm	Cyst puncture + microscopic extirpation
Wu et al. [9]	Female	28 years old	Pancreas	6.0×5.3 cm	Pancreaticoduodenectomy + albendazole
Agholi et al. [31]	Female	40 years old	Triceps brachii	6.5 cm × 5.5 cm	Cystectomy + albendazole
Kothari et al. [32]	Male	28 years old	Pericardial cysts	Largest: 12 cm × 11 cm × 10 cm	Cystectomy + albendazole
Salih et al. [18]	Female	34 years old	Thigh	8×9×11 cm	Cystectomy + albendazole
El Alaoui et al. [33]	Male	29 years old	Gluteal muscle	97×65 mm	Cystectomy + albendazole
Babiker et al. [34]	Female	40 years old	Cystic lesions spread in the lungs, spleen and liver	A 6 cm cyst in the anterior segment of the right upper lobe of the lung, a 5 cm cyst in segment V/VI of the liver, and a 7.5 cm cyst in the upper pole of the spleen.	Albendazole + Cystectomy
Al Taei et al. [35]	Female	37 years old	Lower pole of the left kidney	12.3×8.6 cm	Albendazole + total left nephrectomy
Hanifa et al. [36]	Female	28 years old	Intrapleural cavity	---	Cystectomy + albendazole
Jallali et al. [37]	Female	33 years old	Spleen	12×9 cm	The patient underwent splenectomy and was vaccinated against pneumococcus and haemophilus.
Kakaei et al. [38]	Female	46 years old	Uterus	135×110 mm	Hysterectomy with left salpingo-oophorectomy + albendazole
Thakur et al. [39]	Female	13 years old	Right submandibular region	5×4.7×3.2 cm	Cystectomy
Ghanem et al. [40]	Female	26 years old	Neck	7.5×5×5 cm	Cystectomy and total thyroidectomy
Chemlali et al. [41]	Female	45 years old	Appendix	9 cm	Appendectomy + complete excision of all hydatid cysts
Al Sharei et al. [42]	Female	38 years old	Breast	3.2×3×3 cm	Albendazole + Cystectomy
Ahmed et al. [43]	Male	45 years old	Brain	3.0×3.0×1.0 cm	Albendazole + the cyst was microsurgically excised without rupture

majority of E. granulosus cysts are solitary (72%) with sizes ranging from 1 to 15 cm and a rate of growth from 1 to 30 mm per year [19]. This highlights an important distinction from our present case: 2 concomitant liver cysts that are 17 and 20 cm in diameter. Complications may also occur, so it is important to emphasize the potential danger of dealing with hydatid cysts. In our patient, no severe complications occurred, but another study reported the most serious complication, which is anaphylactic shock that can lead to death [20]. Ammar et al. also reported in their case vascular complications including portal vein thrombosis [6]. Although these disease-related complications are rare, they require rapid and decisive intervention because they may lead to the progression of liver cirrhosis [21]. Scans—like ultrasound or CT—usually give a much clearer idea about whether a hydatid cyst is actually there and what it looks like, which makes imaging the main way to confirm and assess the disease [22]. Unfortunately, despite the accuracy of CT scans in diagnosing many diseases, their frequent use is associated with the risk of cancer in patients due to DNA damage caused by absorbed radiation [23]. Furthermore, the IgG antibody ELISA test supports the diagnosis when positive, in accordance with the procedures of Peralta et al. [24], although this test was not used in our study. Another imaging option is magnetic resonance imaging (MRI), which is less frequently used due to expenses, but is useful for studying surrounding soft tissue [18]. Al-Asbahi et al. recommend magnetic resonance cholangiopancreatography (MRCP), as it is an important procedure for determining extrahepatic extension, vascular involvement, and biliary tree involvement [8]. The gold standard treatment remains surgical excision, especially for large hydatid cysts as in our case, or calcified hydatid cysts, in addition to the adjuvant chemotherapeutic agent albendazole, since it inhibits glucose absorption and consequently leads to glycogen depletion in the mitochondria and endoplasmic reticulum [25]. Many trials showed that the efficacy of albendazole (82%) is superior to that of mebendazole (56%), and that 25% of cases showed evidence of cyst recurrence [26]. Vomiting, abdominal pain, nausea, and elevated liver function tests are sometimes considered side effects of albendazole, and very rarely, toxicity has been reported in the medical literature [27]. Other therapeutic techniques and practices used for hydatid cysts that are recommended by the World Health Organization – Informal Working Group on Echinococcosis (WHO-IWGE) include

puncture-aspiration-injection-reaspiration (PAIR). However, recent studies are now focusing on and highlighting the importance of minimally invasive surgical procedures [28]. There are potential complications associated with the PAIR or surgical treatment, most notably infection at the surgical site, abdominal abscess, biliary fistula, or common bile duct injury [29].

Conclusion

Attention must be given to the differential diagnosis of cystic lesions in the liver. In such a case, laboratory tests usually do not provide any qualitative indicators useful for diagnosis. In contrast, both ultrasound and computed tomography revealed the presence of two hydatid cysts in the liver, which guided us to the correct diagnosis before surgery. As a result of appropriate surgical intervention and proper post-operative care, potential complications such as fluid leakage were avoided. For space-occupying cystic lesions with complex hepatic conditions, multiple diagnostic methods should be combined, including MRI, serology, and immunologic testing, to improve diagnostic accuracy. This case may serve as a reference for future management of cystic hepatic lesions.

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Author contributions
Conceptualization, H.H., M.A., B.A., R.A., and W.A.; Methodology, R.M., L.B., and M.A.; Data curation, M.A.; Writing—original draft preparation, M.A., K.K., B.A., R.A., and N.B.; Writing—review and editing, H.H., M.A.M., S.S., M.S., L.B., F.A., and S.Z.; Visualization, M.A.M., and K.K.; Supervision, Hamdah Hanifa, Samer Sara, and Mohammad Shafa'a. All authors contributed substantially to the conception and design of the work, participated in data acquisition and interpretation, critically reviewed the manuscript for important intellectual content, approved the final version to be published, and agreed to be accountable for all aspects of the work in accordance with ICMJE authorship criteria.

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Data availability
No datasets were generated or analysed during the current study.

Declarations

Ethical approval
Not applicable. As it's a case report, it is exempted from ethical approval by local institution responding on the case.

Consent for publication
Written informed consent was obtained from the patient to prepare the case for case report.

Competing interests
The authors declare no competing interests.

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