



# Resolution of infertility following hydatid cyst removal: a case report

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**Introduction and importance:** Human echinococcosis, also known as ‘hydatid cyst,’ constitutes a zoonotic parasitic disease attributed to *Echinococcus granulosus* or *Echinococcus multilocularis*. Primary afflictions occur in the liver and lungs, whilst the implication of alternative organs remains infrequent. The clinical presentation can vary, and large cysts may cause compression symptoms and complications. This case involved a 21-year-old Palestinian female with three large hydatid cysts in the liver, causing compression of adjacent structures and unique clinical manifestations.

**Case presentation:** A female aged 21 arrived with signs of heart palpitations, shortness of breath, tiredness, and pain in the right upper abdomen, alongside a background of infertility. During routine pre-IVF ultrasound, three liver hydatid cysts were unexpectedly found, leading to a referral to the surgical department. Laboratory tests and imaging confirmed this diagnosis. The surgery involved cyst aspiration, hypertonic saline injection, and marsupialization. Post-surgical complications were managed as they occurred. The symptoms lessened afterward, and a successful pregnancy was achieved 9 months post-surgery.

**Clinical discussion:** Zoonotic infection by cystic echinococcosis (CE) primarily engages the liver and lungs. The progression of symptoms is contingent upon cyst placement and resultant pressure on adjacent tissues. Diagnosis requires imaging alongside serological assays, while large cysts necessitate surgical intervention.

**Conclusion:** The presented case underscores the complexity of managing multiple extensive liver hydatid cysts, highlighting the necessity to consider hydatid disease amidst patients presenting with indeterminate symptoms, particularly within endemic zones. An extensive surgical strategy produced positive outcomes, illustrating the significance of prompt intervention for symptom alleviation and sustained patient health.

**Keywords:** case report, *Echinococcus*, hydatid cyst, infertility, multiple systems

## Introduction

The medical condition referred to as human echinococcosis, widely recognized under the term ‘hydatid cyst,’ is a parasitic zoonotic pathology originating from infection with either *Echinococcus granulosus* or *Echinococcus multilocularis*. Marked by the presence of liquid-filled cysts, this affliction chiefly affects the hepatic organ (50–70%) and pulmonary system (20–30%), with infrequent occurrences in the cerebral, pelvic, skeletal, and abdominal regions. This particular disease maintains a notable prevalence within global sheep-rearing territories,

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## HIGHLIGHTS

- Large hepatic hydatid cysts: The presence of three sizable hepatic hydatid cysts, with the largest measuring up to 19 × 13 cm, poses diagnostic and management challenges due to their rarity and potential for compression of nearby structures.
- Extensive Compression Symptoms: The patient experienced rare and severe compression symptoms affecting multiple surrounding structures, indicating a significant impact of the cysts on anatomical function.
- Successful Pregnancy Post-Surgery: Despite a history of infertility and the presence of extensive cysts, the patient achieved a successful pregnancy without medical assistance nine months after surgery, highlighting a rare and favorable outcome following surgical intervention.

including the Mediterranean zone, the Middle Eastern area, and regions within Africa, Australia, and New Zealand. Transmission to humans, who serve as incidental hosts, ensues through the ingestion of food, water, or manual surfaces contaminated with canid excrement<sup>[1,2]</sup>. This work is in line with the SCARE 2023 criteria<sup>[3]</sup>. The clinical presentation varies depending on the cyst’s location, and it is typically solitary, asymptomatic, and accidentally discovered<sup>[4]</sup>. However, large cysts can cause compression symptoms and complications<sup>[5]</sup>. Patients predominantly pursue medical consultation for cysts with noteworthy dimensions<sup>[5,6]</sup>,

eliciting pain habitually situated in the right upper quadrant or epigastrium, lassitude, pyrexia, nausea, or dyspepsia, potentially preceding jaundice, acute pancreatitis, or acute cholecystitis in the initial phases of the ailment<sup>[7]</sup>. In our investigation, the subject exhibited three considerable hydatid cysts in the hepatic region, culminating in compression of the pericardium, diaphragm, kidneys, and stomach. Moreover, the cyst was found to be connected to the biliary tract.

There are no studies discussing if there is an effect of a hydatid cyst on female fertility. Inflammatory mechanisms play a significant role in the major processes of physiological reproduction in women, such as menstruation, ovulation, embryo implantation, and the onset of labor. Moreover, a remarkable feature of the female reproductive tract is the rapid resolution of these inflammatory events to re-establish the normal reproductive tract. However, any alteration or disruption of reproductive tract resolution capacity may lead to uncontrolled inflammation, or any cause of excessive inflammation, such as infarctions, may disrupt physiological reproductive function in females<sup>[8]</sup>. A hyperinflammatory status can disrupt the axis between the immune and endocrine systems in the female reproductive tract, which may impair pregnancy capacity or lead to pregnancy complications<sup>[9]</sup>. *Echinococcus* organisms are highly multicellular, immunogenic, stimulating pro-inflammatory cellular responses, significant antibody production, and T cell and other cell-mediated responses in humans, which lead to an inflammatory condition with the release of inflammatory mediators, cytokines, and other substances that magnify the inflammatory condition. Persistent inflammatory conditions may affect physiological reproductive functions, such as ovulation and implantation, and may lead to infertility, but the mechanism of inflammation or infection affecting reproductive function is not clearly understood<sup>[10,11]</sup>.

## Case presentation

A female individual aged 21 from Palestinian descent presented at a healthcare institution with three significant hydatid cysts. Over the course of one year, she exhibited a spectrum of symptoms, including palpitations and dyspnea, the latter exacerbating when in a supine position, along with general fatigue and discomfort in the right upper quadrant (RUQ). This RUQ discomfort was correlated with instances of nausea and vomiting, extending to the dorsal region and becoming more pronounced upon the consumption of fatty meals. However, none of these symptoms prompted the patient to seek medical advice. She had a 5-year history of infertility and was referred to an obstetrics and gynecology specialist for infertility evaluation, undergoing various tests; everything was normal and she subsequently opted for in vitro fertilization (IVF). An incidental pre-IVF ultrasound revealed hepatic cysts, leading to referral to the surgical department. There is a history of raising sheep and goats at home. The patient reported no significant past medical or surgical history and no notable family history. Physical examination revealed no signs of irritation, abdominal distension, or tenderness.

Laboratory tests revealed Hb: 9.1 g/dl (12–14 g/dl), platelets: 672/l (140–450 l/l), aPTT: 41.9 s (25–35 s), PT: 19.3 s (11–15 s), fasting blood sugar (FBS): 284 mg/dl (74–110 mg/dl), alkaline phosphatase: 256 U/l (30–105), leukocyte:  $9.1 \times 10^3/\mu\text{l}$  (4.8–10.8). Liver function tests showed normal values. Normal total bilirubin, negative HBsAg, and hormonal levels (prolactin,

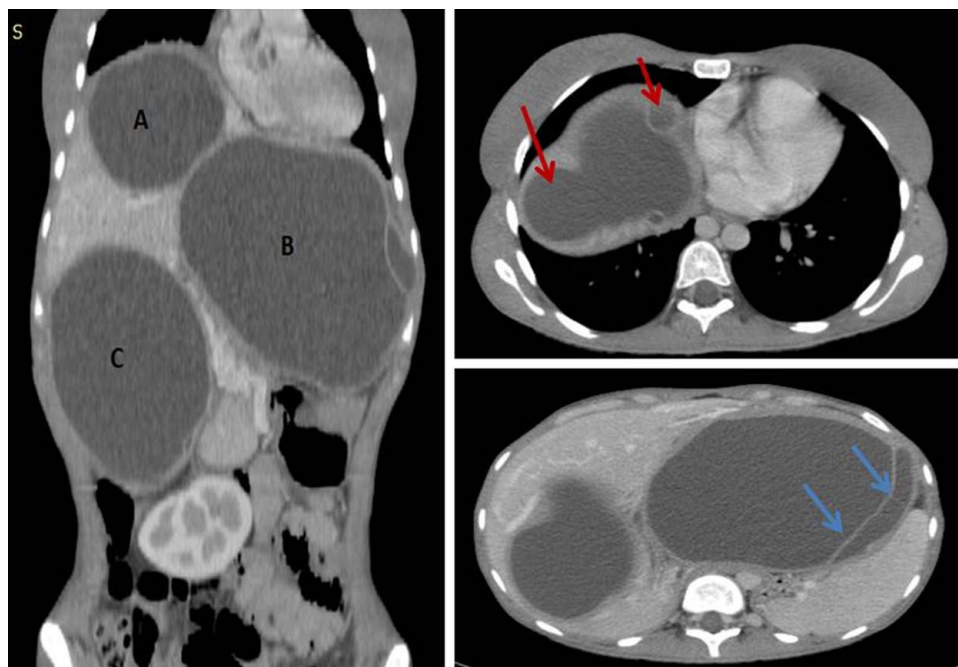
LH, FSH, 17-beta estradiol) were within the normal range, except for elevated TSH: 6.9  $\mu\text{IU/ml}$  (0.4–4  $\mu\text{IU/ml}$ ). Notably, specific serological tests for hydatid disease were not performed during admission. Further workup for high sugar reading was performed, and all results were within the normal range; therefore, a high FBS reading is considered a laboratory error.

Upon radiological examination, the ultrasound images depict three conspicuous hepatic cysts, in which the right liver lobe shows two separated thick-walled cystic masses, measuring about  $17 \times 13$  and  $13.5 \times 11.5$  cm, in anterior-posterior (AP) and transverse (TC) dimensions, respectively, and the left liver lobe shows the largest one, measuring about  $19 \times 13$  cm, also in AP and TC dimensions, respectively. Subsequently, chest and abdominal computed tomography (CT) with intravenous contrast was performed, which confirmed the ultrasound findings (Fig. 1). It also showed marginal daughter cysts, each measuring up to 2.3 cm. CT scan also revealed mild intrahepatic biliary dilatation attributed to the mass effect exerted on these ducts, and midline structures shifted to the contralateral side, including the heart and major vessels, with leftward displacement of the intestinal loops and stomach in the abdomen. The gallbladder appeared slit-like owing to massive compression, and the right kidney was malrotated with inferolateral displacement (Figs 2, 3). Furthermore, mild splenic enlargement was noted during CT scan evaluation as an incidental nonspecific finding.

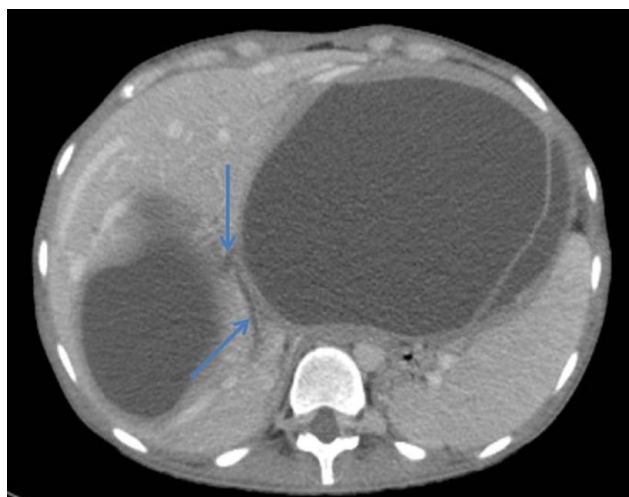
Upon receiving diagnostic conclusions, surgical involvement was resolved. A tri-monthly duration preceding the surgery appointment, oral administration of albendazole at a dosage of 400 mg was administered twice daily in conjunction with 5 mg of bisoprolol once daily. A whole-body CT scan was performed prior to surgery to check for additional sites, and only a hydatid cyst was observed in the liver. Postoperatively, albendazole was continued for an additional 3 months, and the surgical procedure, performed by a general surgeon, involved a Kocher incision and isolation of the operative field using abdominal packs saturated with 23.4% hypertonic saline. The largest hydatid cyst was aspirated with a needle, and the cystic cavity was instilled with 23.4% saline solution 15 min before aspiration. This process was repeated for the remaining two cysts.

Following the successful evacuation of the three cysts, including daughter cyst removal (Figs 4, 5), a uniform marsupialization technique was applied. Each excised cyst cavity received a tube drain, with an additional drain strategically placed in the hepatorenal pouch. Posterior excision of the right cyst resulted in iatrogenic diaphragmatic injury, which was promptly repaired with non-absorbable sutures (nylon) in a continuous fashion. A right-sided chest tube was used to address any potential complications. In addition, the patient received two units of packed RBCs during surgery, and the contents were sent to the Department of Pathology (for histopathological examination). A histopathological examination confirmed the diagnosis. The patient underwent open surgery because the hospital lacked facilities for a laparoscopic approach, and there was no surgeon experienced in the laparoscopic technique. Additionally, due to the large size of the cyst, open surgery was deemed more appropriate in this case.

Postoperatively, the patient exhibited manifestations of bile leak from the cyst-biliary fistula, leading to ERCP referral. A pigtail stent was placed in the biliary tree, and over the subsequent weeks, the bile leak abated, allowing systematic removal of the abdominal drain until complete recovery.



**Figure 1.** Coronal (right) and axial (left) views, post-contrast images of the patient's abdomen show daughter cells (red arrows) and internal septations (blue arrows).



**Figure 2.** Axial view post-contrast CT scan shows intrahepatic biliary dilatation (blue arrows).

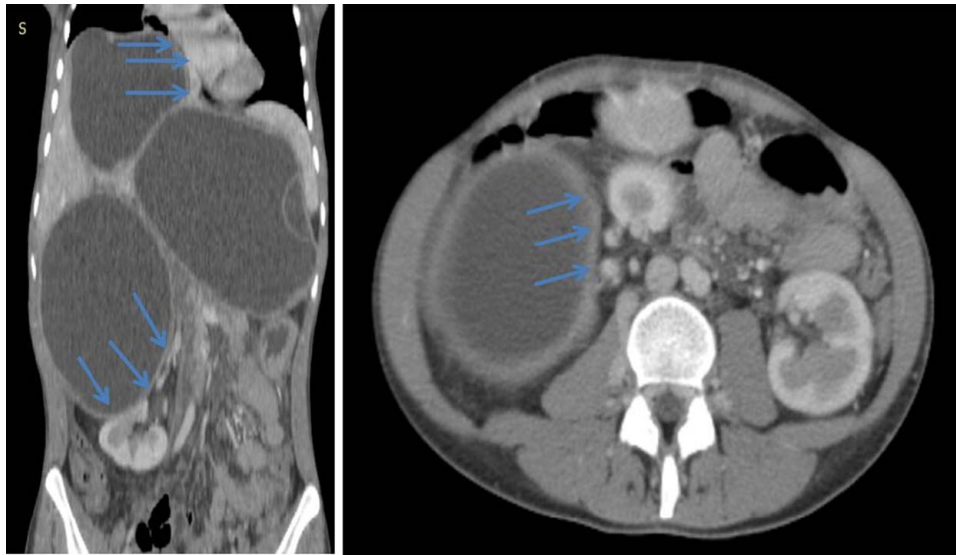
Notably, the comprehensive resolution of dyspnea, palpitations, and pressure symptoms facilitates the resumption of daily activities after surgery. Six-month follow-up CT scan (Fig. 6) showed no recurrence, and the patient reported a successful pregnancy without medical assistance (naturally) at 9 months postoperatively.

## Discussion

The two primary types of echinococcosis, which hold considerable importance for human medical practice and public health,

are cystic echinococcosis (CE) and alveolar echinococcosis (AE)<sup>[12]</sup>. CE emerges as a zoonotic affliction instigated by the larval forms of the cestode *Echinococcus granulosus*<sup>[13]</sup>. This condition chiefly depends on dogs and various other carnivores to act as definitive hosts, wherein the adult parasites reside within their small intestines. Eggs excreted in the feces of infected dogs pose a risk of transmission to humans, even in the absence of close contact with carrier animals<sup>[1]</sup>. The echinococcosis life cycle is reinitiated when humans unintentionally ingest *Echinococcus* eggs and become intermediate hosts<sup>[14]</sup>. Upon hatching, the oncosphere, a hooked-end larva, uses six hooks to penetrate the intestinal wall. Subsequently, it travels through the circulatory system, predominantly affecting organs such as the liver and lungs. In these organs, the oncosphere sheds its hooks and transforms into a slow-growing cyst<sup>[15]</sup>.

Cystic echinococcosis (CE) exhibits a global presence except in the continent of Antarctica. In endemic regions, reports indicate occurrences exceeding 50 cases per 100 000 person-years. In contrast, incidence rates are pronounced, fluctuating between 5 and 10% within certain geographical locations such as Peru, Argentina, East Africa, Central Asia, and China. Conversely, alveolar echinococcosis (AE) is geographically limited to the Northern Hemisphere, with a specific incidence in regions of China, the Russian Federation, and various countries across continental Europe and North America<sup>[12]</sup>. The causative agents for these conditions are distinct: *Echinococcus granulosus* causes CE, *Echinococcus multilocularis* causes AE, and polycystic forms are attributed to *Echinococcus vogeli* or *Echinococcus oligarthrus*<sup>[16]</sup>. CE predominantly affects the liver (70–75%)<sup>[12]</sup> and lungs (10–20%), with occasional involvement of the brain, bones, and heart<sup>[17]</sup>. While hydatid cysts are a common occurrence worldwide, our case is particularly intriguing because it shows uncommon manifestations of cystic echinococcosis (CE). With three cysts affecting various adjacent structures, such as the



**Figure 3.** Coronal (right) and axial (left) views, post-contrast images of the patient's abdomen show mass effects.

diaphragm, pericardium, stomach, right kidney, and spleen, the uniqueness of this presentation adds to our understanding. Notably, the patient's 5-year history of unexplained infertility remarkably resolved following the surgical removal of the cysts. This raises questions regarding the potential connection between hydatid disease and infertility.

The strengths of this case report include the comprehensive details of the patient's symptoms, diagnostic process, treatment, and follow-up, which offer valuable insights into the clinical management of hydatid cysts. An interdisciplinary approach involving multiple specialties such as obstetrics, gynecology, surgery, and radiology, enhances the credibility and depth of the report. Additionally, a successful outcome, including the resolution of infertility and subsequent successful pregnancy, underscores the effectiveness of treatment. However, the study's limitations include the absence of specific serological tests for hydatid disease during admission, which may have affected the comprehensiveness of the initial evaluation. A follow-up period of 6 months may not be sufficient to fully assess the long-term outcomes and potential recurrence. To overcome these difficulties, future research should include larger sample sizes and multicenter studies to validate the findings and enhance their generalizability. Incorporating specific serological tests for hydatid disease and extending follow-up periods are also recommended to provide a more robust understanding of patient outcomes.

Reports indicate that hepatic hydatid cysts experience an initial growth of ~1 cm within the first 6 months, followed by an annual increase of 2–3 cm, depending on tissue resistance in the surrounding area<sup>[18]</sup>. Progression often involves an asymptomatic incubation phase lasting several years before clinical symptoms become apparent. The presentation of symptoms depends on both the hydatid cyst location and the pressure applied to the neighboring tissues. Common symptoms include loss of appetite, fatigue, and weight loss. In cases where hydatids develop in the liver, additional symptoms such as abdominal discomfort, nausea, and vomiting are commonly observed<sup>[13]</sup>.

Cysts can last for years, if not decades, and can enlarge to a

diameter of 5–10 cm in a single year. Usually, there are no symptoms and the infection is often discovered accidentally when imaging is performed for other reasons. The compressive force of the cyst in a small area can result in symptoms, if any. Cysts in these areas can also cause pain and swelling of the belly and pelvis. In this instance, the patient's symptoms include hypotension, right-sided soreness, nausea, and vomiting<sup>[1]</sup>.

Imaging techniques (US, CT, and MRI) in conjunction with serology (usually enzyme-linked immunosorbent assays) are the mainstay for the diagnosis of *E. granulosus*. The best initial modality is ultrasound because it is easy to perform, inexpensive, and has 90–95% sensitivity<sup>[19,20]</sup>; a CT scan has a higher sensitivity (95–100%)<sup>[15]</sup>; and MRI has no major advantage over CT. Serology is useful for primary diagnosis and follow-up after treatment; it has a sensitivity of 80–100% and a specificity of 88–96% in the case of liver cysts but only 50–60% in the case of lungs and 25–56% in the case of other organs<sup>[21]</sup>. Serology is less sensitive than imaging and a negative result does not rule out the potential for a hydatid cyst. In this instance, radiographic findings indicate that surgery is the optimal course of treatment; however, serological research has not been conducted. US and CT scans revealed stage CE3B and stage three according to the WHO and Gharbi classifications, respectively. The treatment was surgical removal with albendazole.

Therefore, the removal of these hydatid cysts can resolve infertility through a variety of mechanisms. One mechanism is decreased immunological reactions and inflammation, which may be continued by hydatid cysts. Ovulation and implantation are two common reproductive processes that can be affected by persistent inflammation. In addition, restoration of normal anatomical structure and function can be achieved by the removal of these hydatid cysts, which may improve fertility. The various factors considered to ascertain that the resolution of infertility may be a result of the elimination of hydatid cysts rather than other factors, including the timing of fertility enhancement in the removal of cysts. No other interventions or treatments were introduced simultaneously, which can affect fertility, gives a high suspicion of the effectiveness of cyst removal. We could not find





**Figure 4.** Postoperative images. Coronal (right) and axial (left) views, as well as post-contrast images of the patient's abdomen, show postoperative changes (white arrow), such as an axial cut of the abdominal drain.

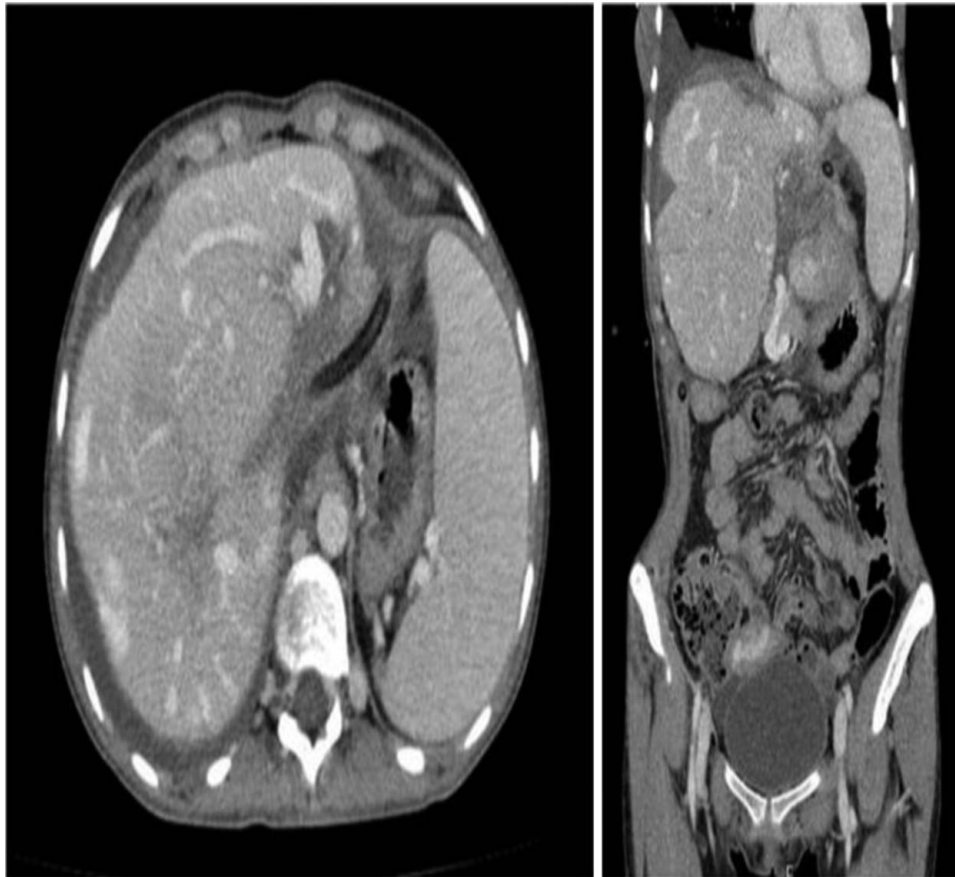
any articles directly relating hydatid cysts to infertility, although there is one on the immune response and inflammation concerning hydatid cysts<sup>[10]</sup>. As addressed earlier, this inflammation and immune response may lead to the physiological causes of infertility. In general, the effect of hydatid cysts on infertility may be due to mechanical obstruction, inflammatory responses, or hormonal imbalance<sup>[22]</sup>. Successful resolution of infertility in patients undergoing hydatid disease often requires a multi disciplinary approach. These include surgical intervention, medical management, and fertility treatment<sup>[22]</sup>.

Generally, the management of a hydatid cyst is based on the Al-Gharbi and WHO classifications, which evaluate the cyst ultrasonographically and usually involve antiparasitic therapy combined with removal or aspiration of the cyst<sup>[2]</sup>. Small stages (CE1 and CE3a cysts < 5 cm) were treated with antiparasitic agents alone for 3 or 6 months, depending on the clinical factors. If medical therapy fails, percutaneous aspiration is performed<sup>[23]</sup>, whereas stages (CE2 and CE3B) need more intervention, such as surgical removal or percutaneous aspiration-injection-reaspiration (PAIR) aspiration of the cysts, and medical therapy alone is not adequate<sup>[23]</sup>. Stages (CE4 and CE5) are inactive and treated conservatively<sup>[24–26]</sup>. The best option for large cysts is surgery, especially if they cause compression symptoms in the vital organs, as in our case<sup>[27]</sup>. Albendazole is the primary antiparasitic agent used to treat *E. granulosus*<sup>[28]</sup> and is useful as an adjunctive therapy before and after surgery or percutaneous therapy to sterilize cysts and prevent the spread of daughter cysts during or after surgery<sup>[1,29]</sup>. The albendazole dosage was 10–15 mg/kg/day for 4 weeks before the surgery; after each period, there was a rest period of one week<sup>[1]</sup>. After surgery, the patient continued albendazole for 6–8 weeks to clear any remaining content<sup>[29]</sup>.

Follow-up imaging should be performed because of the possibility of relapse of these cysts. Imaging with ultrasound or other modalities (CT and MRI) for 5 years is usually recommended<sup>[29]</sup>.



**Figure 5.** The three hydatid cysts after their surgical removal.



**Figure 6.** Axial (left) and coronal (right) views of abdomen CT with intravenous contrast. Follow-up after 6 months.

A thorough explanation of hydatid disease, including its parasitic origin linked to livestock such as sheep and goats<sup>[30]</sup>, and a focus on the significance of early detection and treatment are crucial first steps in ensuring effective follow-up health education for the patient and her family. A thorough set of instructions for post-surgical care should be provided. An important element is to follow specific wound care instructions, especially to identify infection symptoms. In addition, albendazole continues to prevent recurrence<sup>[31]</sup>.

Nutritional guidelines need to be modified to aid the recovery of patients, suggesting a cautious return to everyday activities to minimize overexertion and caution against fatty foods. Scheduling routine imaging tests, such as CT scans and follow-up appointments, is important to track the patient's progress and identify any recurrence indicators<sup>[31]</sup>. It is also important to discuss preventive actions, such as the significance of regular deworming of animals and pets and good hygiene practices to prevent reinfection<sup>[30]</sup>. In addition to offering resources for mental health support and encouraging people to join support groups to share experiences and strategies for coping, it is important to recognize the emotional impact that the illness and surgery that followed have had on the patient<sup>[32]</sup>. Finally, educational resources should be provided to the patient and her family, including brochures and links to reliable websites, along with contact details for medical professionals in case of any questions or problems. By taking these precautions, the likelihood of more issues

will decrease, and the patient and her family will be better prepared to manage the illness.

A case series observational study conducted by Palestinian authors demonstrated that cystic echinococcosis spread widely in 94% of cases in the West Bank and in 6% of cases in Gaza. That is why we encourage their recommendations for the Ministry of Health, General Administration of Veterinary Services and Animal Health, and local health authorities to decrease the burden of CE on humans by implementing control measures and creating a stringent surveillance system<sup>[33]</sup>. In addition, we recommend implementing preventive measures such as owners should make measures to ensure that the dogs do not come into contact with dead sheep or their waste, treating dogs with anti-tapeworm medications or worming regularly, deworming programs for livestock, proper disposal of infected dead animals, implementation and promotion of good hygiene practices among livestock handlers, educating communities, and creating health awareness campaigns about CE. These recommendations can be evaluated for their effectiveness through regular screening programs for livestock and humans in high-risk areas.

This paper describes the case of a young female patient whose infertility was resolved after hydatid cyst excision surgery. The patient presented with a variety of symptoms and underwent a range of diagnostic tests, which resulted in an unintentional finding of hepatic cysts during a pre-IVF ultrasound. This study followed the patients' clinical paths from start to end. In addition

to relief of her symptoms after surgery, the patient became pregnant naturally and did not require additional medical care. This example shows the effectiveness of surgical treatment in treating infertility linked to hydatid cysts and emphasizes the significance of ruling out this condition as a possible cause in cases of infertility that cannot be explained.

## Conclusion

In summary, this case highlights the importance of accurate and early diagnosis of hydatid cyst disease in patients who have been exposed to livestock. The patient's elaborate presentation showed an aspect of unexplained infertility and severe manifestation of cysts that necessitate a high index of clinical suspicion and a wide scope of diagnostic workup in endemic regions. Successful surgical intervention, together with preoperative and postoperative albendazole treatment, not only cured symptoms but also restored fertility. This case emphasizes that multidisciplinary management is mandatory for hydatid cyst disease, focusing on surgery and appropriate medical therapy for satisfactory outcomes. Moreover, it demonstrates how a lack of complete or targeted investigations may lead to misdiagnoses or delayed management. Therefore, healthcare workers require more education. Regular follow-up visits, including imaging, are necessary to monitor disease recurrence and maintain health status.

## Ethical approval

Not applicable.

## Consent

Written informed consent was obtained from the patient's family for reporting this case and its associated images. The consent is available for review on request.

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The study did not receive any funding.

## Author contribution

H.J.M.W., F.B.Y.S., B.Y.H.A., M.Y.H.A., and M.A.B.: writing the manuscript; K.N., S.N.F., and M.I.S.: imaging description; H.J.M.W. and M.Y.H.A.: reviewing and editing the manuscript.

## Conflicts of interest disclosure

The authors declare no conflict of interest.

## Research registration unique identifying number (UIN)

Not applicable.

## Guarantor

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## Data availability statement

Data is available upon request.

## Provenance and peer review

Provenance and peer review not commissioned, externally peer-reviewed.

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