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Case Report

# Postpartum maternal death resulting from complications of a large hydatid cyst of the lung in a resource-constraint setting: A rare case report and review of literature ☆,☆☆

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ABSTRACT

Cystic echinococcosis (CE), a parasitic zoonotic infection caused by the larval stage of Echinococcus granulosus, predominantly affects the liver and lungs but can occur in any organ or tissue. Hydatid cysts during pregnancy are rare, with an incidence of approximately 1 in 20,000-30,000 pregnancies, and are often associated with more severe symptoms due to the physiological changes of pregnancy. In endemic areas, early recognition and prompt management are critical to improving outcomes.

This case report from Northern Tanzania highlights a 30-year-old pregnant woman at 30 weeks of gestation who presented with respiratory distress, cough, fever, and chest pain. Imaging studies, including contrast-enhanced computed tomography (CT) of the chest, identified a large complex cystic lesion occupying the right hemithorax, consistent with a pulmonary hydatid cyst. Serological testing supported the diagnosis of CE. Following interdisciplinary team deliberation, labor was induced at 30 weeks, she delivered a healthy baby but subsequently developed postpartum hemorrhage, which required an emergency total abdominal hysterectomy due to uterine atony. Despite aggressive management, the patient developed severe respiratory complications and succumbed 10 days later in the surgical intensive care unit. This case underscores the challenges of diagnosing and managing CE in pregnancy, particularly in resource-limited settings.



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## Introduction

Echinococcosis, commonly referred to as hydatid disease, is caused by the larval stages of tapeworms belonging to the Echinococcus genus. The most prevalent cause of human infections is Echinococcus granulosus, which leads to cystic echinococcosis (CE). This disease is endemic in regions such as the Mediterranean, Eastern Europe, the Middle East, Africa, South America, Australia, and New Zealand [1,2]. While hydatid cysts most commonly affect the liver and lungs, they can develop in any organ or tissue, causing significant morbidity and mortality [2]. According to the World Health Organization (WHO), CE affects at least one million people globally each year and is classified as one of 7 neglected zoonotic diseases of global importance [1,3]. In Africa, CE is particularly prevalent, with regional estimates ranging from 1% to 11% [4].

Pregnancy-associated CE is rare, with a global incidence of approximately 1 in 20,000-30,000 pregnancies annually [5,6]. While many individuals remain asymptomatic, pregnant women are at an elevated risk of cyst growth and associated complications due to immune suppression during pregnancy [7]. Managing CE during pregnancy presents unique challenges, including increased respiratory demands, the risk of cyst rupture, potential miscarriage or preterm labor, and restrictions on certain medications [7,8].

Although case reports describe a range of medical and surgical management strategies, standardized guidelines for managing CE in pregnancy remain lacking [7,8]. Reports from Tanzania are particularly scarce, highlighting the need for well-documented best practices for diagnosis and treatment in resource-limited settings. Here, we present a rare case of symptomatic pulmonary hydatid disease in a pregnant woman during her third trimester.

## Case presentation

A 30-year-old Maasai woman, G4P 3L3, at 30 weeks of gestation, presented with progressive shortness of breath, dry cough, right-sided chest pain, nausea, vomiting, and fever. Her symptoms had worsened over 3 months before admission to a community hospital. She had attended 2 antenatal clinic visits during this pregnancy, with no significant medical or obstetric history.

On examination, the patient appeared ill, pale, and cachectic. Notable findings included facial edema, grade 2 pitting edema in the lower limbs, dilated superficial veins on the right side of the neck, and traditional scarification marks on her back. Vital signs showed blood pressure of 110/67 mmHg, pulse rate of 120 bpm, respiratory rate of 24 bpm, oxygen saturation of 84% on room air, and a temperature of 36°C. Respiratory examination revealed dullness to percussion at the right lung base, diminished breath sounds across the right lung, and reduced chest wall movement on the same side. Abdominal examination showed a fundal height of 30 cm, tenderness in the right upper quadrant, and caput medusae. Other systemic examinations were unremarkable.

Initial differential diagnoses included superior vena cava syndrome, thrombosis, and lung metastases from hepatocellular carcinoma. Serological testing for Echinococcus was performed, revealing elevated serum immunoglobulin E (778 KU/L; normal: 0-114) and positive enzyme-linked immunosorbent assay (ELISA) for Echinococcus IgG. Other laboratory findings included hemoglobin of 8.5 g/dL, erythrocyte sedimentation rate (ESR) of 67 mm/hr (normal: 0-35), C-reactive protein of 100,000 ng/mL, alpha-fetoprotein (AFP) of 310 IU/mL, and hypoalbuminemia (1.12 g/L). Hepatitis serologies, renal function tests, liver enzymes, coagulation profiles, and platelet counts were normal. Obstetric ultrasound confirmed a single live intrauterine pregnancy at 30 weeks and 5 days gestation, with a normal biophysical profile.

Due to financial constraints, a noncontrast chest and abdominal CT was performed. It revealed a large, complex cystic lesion occupying the right hemithorax, causing atelectasis and mediastinal shift to the left. Dilated, tortuous superficial veins were also observed on the anterior abdominal wall and right neck. The findings raised strong suspicion of pulmonary hydatid disease.

Management required a multidisciplinary team, including surgeons, obstetricians, anesthetists, pediatricians, and internists. After counseling, labor was induced at 30 weeks while providing high-flow oxygen via nasal cannula. The patient delivered a healthy male infant vaginally without respiratory deterioration during labor. However, 20 minutes postpartum, she experienced excessive vaginal bleeding exceeding 500 mL. Despite uterine massage and first-line medical therapies (oxytocin, misoprostol, and tranexamic acid), bleeding persisted. An emergency laparotomy revealed an atonic uterus, necessitating a total abdominal hysterectomy (TAH). Estimated blood loss was 900 mL, and the patient stabilized postoperatively.

The patient was transferred to the surgical intensive care unit (SICU), where her respiratory condition deteriorated, requiring intubation and mechanical ventilation. Postoperative care included sedation, pain management, antibiotics, and albendazole administered via a nasogastric tube. Plans for thoracotomy to address the pulmonary cyst were deferred pending her recovery from the TAH. Unfortunately, on postoperative day 10, she succumbed to respiratory complications. The neonate, monitored in the neonatal unit, was later discharged without complications.

## Discussion

Hydatid cyst is a zoonotic disease found worldwide, particularly in regions where cattle rearing is a common occupation. In humans, hydatid cysts are most commonly caused by the larvae of the tapeworm echinococcus granulosus, while infection with echinococcus E.multilocularis is less common but more severe [9]. The life cycle of E. granulosus involves an alternation between carnivores and herbivores, such as dogs and sheep, with humans acting as accidental intermediate hosts. In humans, ingested ova penetrate the intestinal mucosa and travel via the portal vein to the liver, where they develop into hydatid cysts [10]. The liver, particularly its right lobe, is the most frequent site of infection [11].

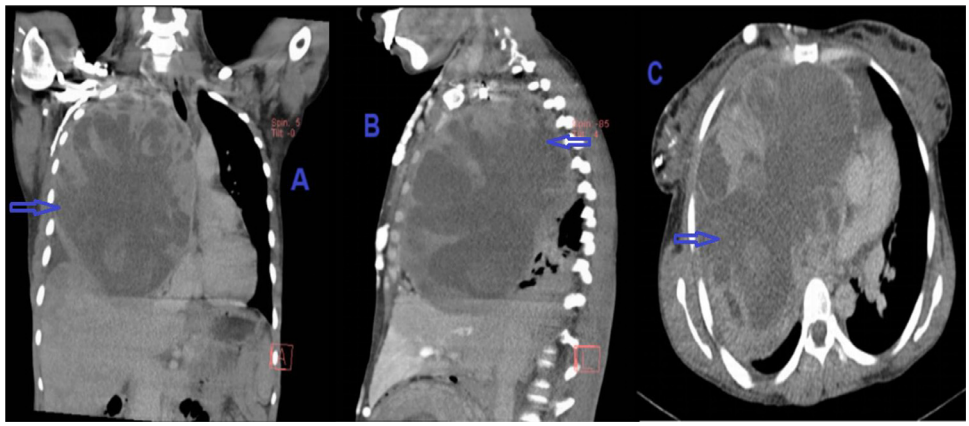


**Fig. 1 – Thoracoabdominal CT with coronal view (A), sagittal view (B) and (C) axial view showing a large complex cystic lesion indicated by an arrow occupying the right hemithorax and measuring 20 x 16 x 15 cm. The right sided lesion is associated with significant mediastinal shift to the contralateral side and atelectasis of the right upper and lower lobes.**

Pregnant women are disproportionately affected by cystic echinococcosis (CE), likely due to hormonal changes during pregnancy and lactation that suppress immune function [4]. Decreased cell-mediated immunity and the immunosuppressive effects of placental steroids create an environment conducive to accelerated parasitic growth and cyst enlargement [11]. These factors may explain why the Echinococcus cysts in our patient grew rapidly and suddenly became symptomatic. Consequently, pregnant individuals are at increased risk of developing severe complications, including cyst rupture and secondary bacterial infections [8].

Diagnosing CE during pregnancy is particularly challenging due to overlapping symptoms with other conditions, the need for careful assessment of risk factors, and the potential limitations on imaging. Imaging remains the cornerstone of hydatid disease diagnosis, yet radiation concerns during pregnancy may delay appropriate investigations. Providers must balance the risks of fetal exposure to imaging with the dangers posed to the mother by delayed or missed diagnoses [12,13]. In cases where a patient is highly symptomatic or clinically unstable, imaging should not be deferred or omitted, as timely diagnosis can significantly impact outcomes [12,13]. Serological tests, including direct hemagglutination, immunoblots, and enzyme-linked immunosorbent assays (ELISA), are valuable diagnostic tools for confirming CE [14].

Management of CE in pregnancy requires a tailored approach, considering the patient's clinical status, gestational age, and the risks associated with intervention. Treatment options include conservative monitoring, medical therapy, percutaneous aspiration, and surgery [6,8,11]. Surgery is often the treatment of choice in symptomatic cases; however, during pregnancy, surgical intervention carries risks of spontaneous abortion or preterm labor. Nonetheless, it may be necessary to prevent life-threatening complications such as cyst rupture or organ compression [8]. Medical management with albendazole is a cornerstone of CE treatment, but its safety profile during pregnancy remains inconclusive, limiting its use [15].

Obstetric complications associated with hydatid disease include abdominal pain, prolonged or obstructed labor, and the risk of uterine rupture. Additionally, cyst rupture during labor can lead to anaphylactic shock, posing a life-threatening situation [16].

In our case, albendazole was initiated postdelivery to facilitate healing and minimize recurrence risk before definitive thoracic intervention. While this strategy was intended to stabilize the patient before surgery, she unfortunately succumbed to respiratory complications before the planned thoracotomy. Had she survived, the treatment plan would have included continued albendazole for 1-3 months postoperatively to reduce the likelihood of recurrence.

This case highlights the complexities of managing CE in pregnancy, emphasizing the need for a multidisciplinary approach and careful consideration of individualized treatment strategies. It also underscores the importance of further research to guide evidence-based management and improve outcomes in this vulnerable population.

## Conclusion

Pulmonary hydatid disease during pregnancy is a rare but serious condition associated with significant risks of complications and mortality. Its management is particularly challenging due to the unique physiological demands of pregnancy and the lack of standardized treatment protocols. In endemic regions, maintaining a high index of suspicion is essential for timely diagnosis and intervention to optimize outcomes for both the mother and fetus. This case underscores the importance of a multidisciplinary approach and highlights the need for further research and guidelines to support the management of hydatid disease in pregnancy (Fig. 1).

## Author contributions

JL, and OM, were involved in data collection and analysis, drafting and editing of the final manuscript. All authors discussed the case and approved the final manuscript.

## Patient consent

I hereby give my consent for images or other clinical information relating to my case to be reported in a medical publication.

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