

CASE REPORT

Hydropneumothorax caused by complete rupture of a pulmonary hydatid cyst: A rare case report

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Key Clinical Message

Hydatid disease is a zoonotic disease endemic in developing regions. It is usually caused by infection with the tapeworm *Echinococcus granulosus* due to contaminated food or drinks or by close contact with dogs. The lungs are the second most affected organ (25%) after the liver (>65%). Cyst rupture is the most frequent complication. Enlarging pulmonary cysts are more vulnerable to rupture, with rupturing in the bronchial tree being the most common (20%–40%). Hydropneumothorax is a consequence of complete cyst rupture into the pleural cavity and occurs in rare cases (2%–4%). Superinfection is a common complication of the ruptured cyst, which might lead to empyema. A 26-year-old male presented to our clinic with dyspnea that had progressed recently and made him unable to walk a few meters. He had a history of cough and exertional dyspnea and was examined by three different clinics without performing a chest X-Ray. Physical examination revealed fever and a sick appearance. Chest X-ray revealed complete pneumothorax with an air-fluid level in the left hemithorax. Computerized tomography demonstrated two cysts, and one of them was ruptured, causing hydropneumothorax and empyema. The patient was treated with surgery, and follow-up showed recovery with a fully re-expanded lung. Rupture of pulmonary hydatid cyst is seen in the most of cases, the clinicians must be aware of such presentation and management of the pulmonary hydatid disease.

KEYWORDS

case report, echinococcosis, empyema, hydropneumothorax, pulmonary hydatid cyst

1 | INTRODUCTION

Hydatid disease, also known as human echinococcosis, is an uncommon disease caused by infection with *Echinococcus*, a type of tapeworm. Many species of *Echinococcus* have been identified. Two of them are pathogenic to humans, *Echinococcus granulosus* (*E. granulosus*)

which causes cystic echinococcosis (CE), and *Echinococcus multilocularis*, which causes alveolar echinococcosis. The other species have no potential for zoonotic transmission.¹

Human infection usually results from the ingestion of *Echinococcus* eggs, either from contaminated food or drinks or by close contact with dogs or other infected carnivores. When the eggs reach the intestine, they hatch into

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larvae, which spread via the blood and infect the organs, forming hydatid cysts.² The lungs are the second most common site of hydatid cysts (25%) after the liver (>65%). The kidneys, heart, bones, and central nervous system are rarely involved.³

Pulmonary hydatid cysts tend to grow slowly and asymptotically. Usually, they are diagnosed incidentally on a chest X-ray or computerized tomography (CT) that is performed for another reason.⁴ Symptoms often result from the mass effect of the extended cyst on the surrounding structure and include cough, chest pain, dyspnea, and hemoptysis. Fatigue, nausea and vomiting, and chest malformations are less frequent symptoms.

Cyst rupture is a serious complication, and the development of new symptoms or worsening of the existing symptoms can be clues for rupture. Cysts can rupture within the bronchial tree or, in rare cases, within the pleural cavity, which can lead to anaphylaxis, hydropneumothorax, and empyema.⁵ According to a cross-sectional study of pediatric patients with pulmonary hydatid cysts, the rupture rate for patients was 39.5%, and 29.5% for cysts, this make cyst rupture a common complication.⁶ However, The clinical features of pulmonary hydatid cysts are nonspecific, thus we report a case of pulmonary hydatid cyst rupture which might help the clinicians to get a better understand of its presentation.

2 | CASE PRESENTATION

A male in his 20s came to the clinic with progressed dyspnea and pain on the left side of the chest. The patient had a 3-month history of cough and exertional dyspnea. He was previously examined at another clinic, but a chest X-ray was not performed. As a result, his symptoms were attributed to bronchitis, and he was treated with anti-inflammatory agents and antibiotics. However, he continued to deteriorate, and the symptoms progressed recently to the point that he could not walk a block without stopping to catch his breath. The patient confirmed that he is a heavy smoker and lives in a rural area. He denied any other medical or family history of significant diseases. Physical examination revealed fever, sick appearance, resonance to percussion in the left hemithorax, and decreased breathing sounds. Laboratory evaluation showed leukocytosis and elevated CRP. Full results are presented in Table 1. A chest X-ray showed complete pneumothorax with an air-fluid level in the left hemithorax (Figure 1) and a wide patch of density over the left lung (Figure 2). A chest tube was inserted to relieve symptoms, and approximately 1 L of purulent fluid was drained, with previous findings strongly indicating empyema. After the procedure, a drainage tube was placed, and a course of

TABLE 1

Test	Result	Normal value
Hemoglobin	15.6 g/dL	13–17 for male
Hematocrit	46%	42–52 for male
Erythrocytes (RBCs)	$5.72 \times 10^6/\mu\text{L}$	4.6–5.6 for male
Leucocytes (WBCs)	$17.2 \times 10^3/\mu\text{L}$	5–10
Neutrophil	85%	40–70
Lymphocyte	6%	25–45
Monocyte	9%	2–8
Eosinophil	00%	1–5
Basophil	00%	0–1
Platelet	$274 \times 10^3/\mu\text{L}$	150–400
MCV	78	72–100
MCH	27	28–33
MCHC	35	32–36
CRP	67 mg/L	Up to 6

Abbreviations: CRP, C-reactive protein; MCH, mean corpuscular hemoglobin; MCHC, mean corpuscular hemoglobin concentration; MCV, mean corpuscular volume.

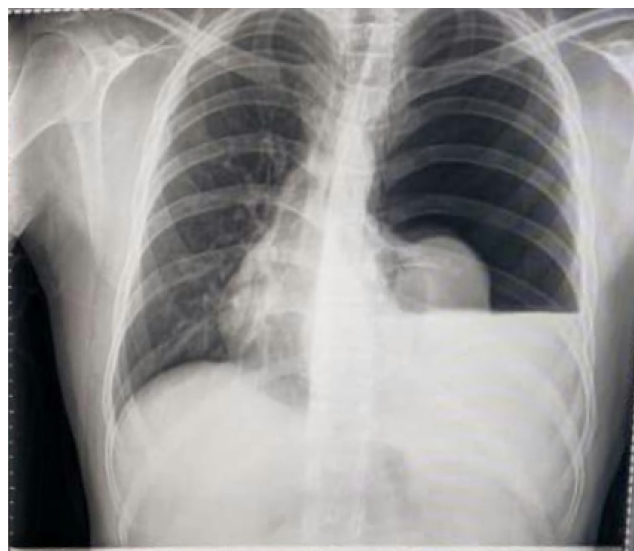


FIGURE 1 Chest X-Ray of the patient at come shows complete pneumothorax with air-liquid level in the left hemithorax.

antibiotics including vancomycin and meropenem was started. Post-procedure chest X-ray showed a wide patch of density over the left lung (Figure 2).

A CT scan of the chest was performed, and it demonstrated a 62×79 mm intact homogenous cyst with a thin and regular wall located in the left-upper lobe and a 65 mm cyst with signs of rupture (air inside the cyst and detached germinal layer floating), located in the left-lower lobe with atelectasis in this lobe. It also revealed signs of superinfection (Figures 3 and 4). There was no other lesion on CT. An abscess has a very similar pattern to the

complicated cyst, which might cause confusion about the diagnosis. However, having another intact cyst that had the typical characteristics of a hydatid cyst was very helpful in making the diagnosis.

Surgery was the treatment of choice, and the patient was prepared with albendazole therapy (400 mg), which continued for 6 months after the surgery. An open incision was made, and the intact cyst was treated by enucleation. A concentrated salt solution was infused inside the ruptured cyst and then aspirated. The inner germinal layer was excised, and then the bronchial fistulas and cyst cavity were closed. During the procedure and due to empyema, the surgeon observed a layer of fibrous tissue overlying the lung, which resulted in “restrictive lung”. Therefore, decortication of the lung was performed. No lobectomy was needed.



FIGURE 2 Chest X-Ray of the patient after thoracentesis shows a wide patch of density over the left lung.

Serial chest X-ray and clinical examinations were undertaken for follow-up after surgery. They showed recovery with a fully re-expanded lung (Figure 5).

3 | DISCUSSION

Hydatid disease is a zoonotic disease endemic in developing regions worldwide. It results from the larval stage of infection by types of tapeworms. *E. granulosus* belongs to the family Taeniidae and genus *Echinococcus* and causes CE, which is the most common species of human disease.¹ The life cycle of *Echinococcus* includes dogs or related species as the definitive host and sheep, goats, or swine as intermediate host, and humans are secondary hosts. Human infection results from the consumption of food or drinks contaminated by *Echinococcus* eggs, or by close contact with dogs or other infected carnivores.²

After ingestion, the eggs hatch and release the embryo in the small intestine, and they invade the mucosa,

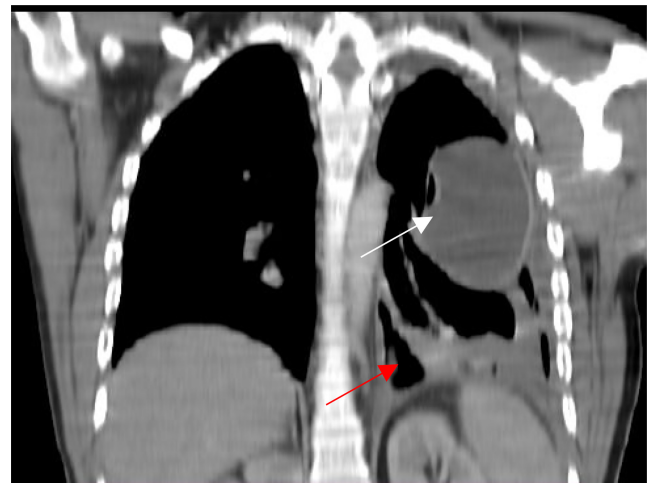


FIGURE 4 CT with Coronal section scan shows of the ruptured cyst (red arrow). And the intact cyst (white arrow).

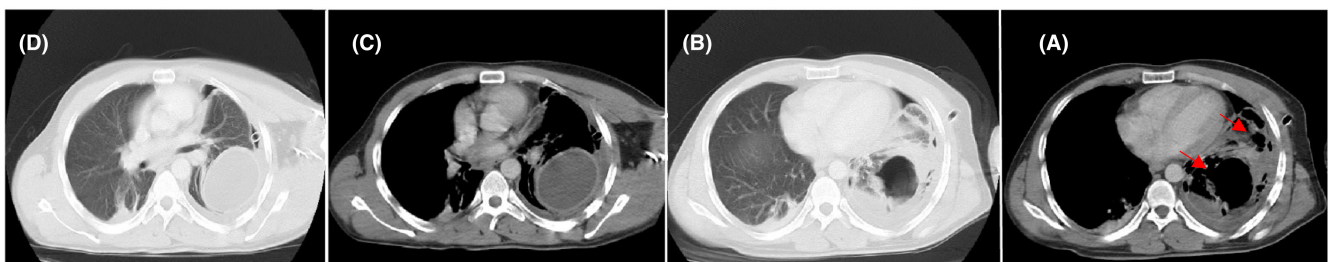


FIGURE 3 CT with axial sections scan of the chest. (A) An axial-section in the Mediastinal window shows signs of the ruptured cyst (air inside) and signs of empyema (air bubbles with the red arrow). (B) the same section in A with the lungs window. (C) axial -section in the Mediastinal window shows the intact cyst homogenous opacity with sharp contours with moderate density. (D) the same section in C with the lungs window.

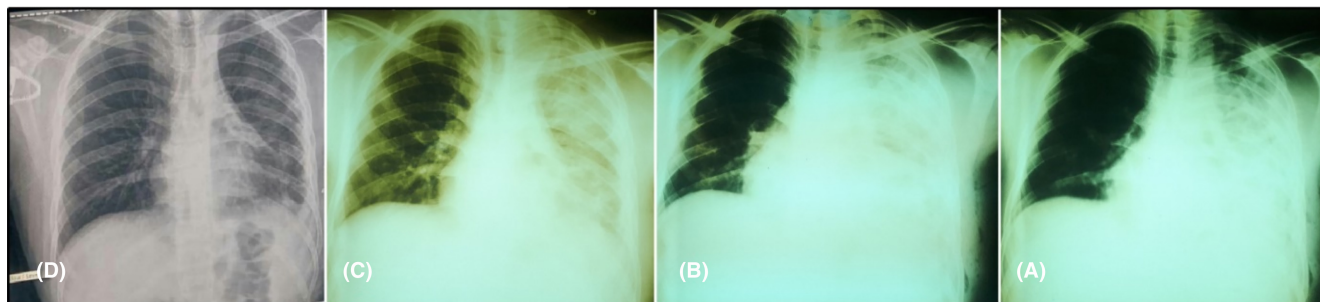


FIGURE 5 Post-surgery follow-up with chest X-Ray. (A) 2 days after surgery. (B) 4 days after surgery. (C) a week after surgery. (D) 3 weeks after surgery, shows recovery with full re-expansion pulmonary.

leading to the distribution of the larvae through the portal vein to the liver, while some of them pass through the capillaries and become lodged in the lungs and other organs, where the cyst develops. The lungs have the second-highest rate of organ location of the *Echinococcus* cyst (25%), after the liver (>65%). Involving other organs such as kidneys, brain, and bones were rarely observed. The solitary cyst is the most common form of the infection; however, multiple cysts and multiple organ cysts may occur.^{3,7} Symptoms depend on the site and size of the cyst; CE causes slowly enlarging cysts that often grow unnoticed and for years unless acute complications occur, and might discover accidentally during radiography on an unrelated occasion.⁸ Enlarging pulmonary cyst causes mechanical symptoms due to bronchial compression, which includes cough followed by chest pain, breathlessness, and expectoration. Fever, anorexia, and fatigue occur as a generalized toxic reaction due to the presence of the parasite itself.⁹

Spontaneous or trauma-induced lung cyst rupture is the most frequent complication. Rupture divides into contained and complete ruptures. In contained rupture, there is detachment of the pericyst from the endocyst but the pericyst is still intact with no perforation. Whereas in complete rupture, there is a connection with the bronchial tree or pleural cavity. Rupturing in the bronchial tree is most common (20%–40%). Hydropneumothorax is a consequence of the complete rupture of the cyst into the pleural cavity, which occurs in rare cases (2%–4%). Superinfection is a common complication of the ruptured cyst which might lead to empyema. An air-fluid level is an important sign of superinfection. The isolated ruptured cyst can resemble an abscess; therefore, catching the real cause might be difficult and needs advanced investigations. Chest X-ray can be diagnostic in many cases, especially with non-complicated cysts. However, this valuable simple investigation might be neglected due to the limited resources, poverty, and low income of the population in developing countries. CT is particularly useful in the case of complicated cysts. However, uncomplicated cysts are

visualized on a CT scan as rounded and of homogenous opacity with sharp contours with moderate density.^{10,11} In our case, having a ruptured cyst accompanied by an uncomplicated cyst in the same organ was helpful with the diagnosis.

Laboratory evaluations do not show typical changes. Eosinophilia usually occurs after cyst perforation but is not specific. Surgical treatment, with a variety of techniques, exists and is the method of choice in the treatment of pulmonary echinococcosis, particularly complicated cysts. The pathomorphological characteristics of the disease enable the removal of the cyst and preserve the lung tissue in many cases, including complicated cysts with reversible alterations. Albendazole chemotherapy is given in combination with surgery. Albendazole alone is reserved for patients with multiple intact cysts or who cannot tolerate surgery.¹¹

4 | CONCLUSION

Since rupture of pulmonary hydatid cyst is seen in the most of cases, the clinicians must be aware of such presentation and management of the pulmonary hydatid disease.

AUTHOR CONTRIBUTIONS

Maher Almousa: Conceptualization; data curation; investigation; resources; software; writing – original draft; writing – review and editing. **Ahmad Alhadla:** Data curation; formal analysis; resources; validation; visualization; writing – original draft. **Mohammad Aljomaa:** Data curation; formal analysis; resources; writing – original draft; writing – review and editing. **Tamim Abdalrazzak:** Data curation; investigation; methodology; project administration; resources; supervision; validation; visualization; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare that they have no competing interests.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study

CONSENT

A written consent form was obtained from the patient for all the information and pictures in this article.

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