A Rare Case of Pneumothorax Caused by Ruptured Pulmonary Hydatid Cysts Accompanied by Deep Vein Thrombosis: A Case Report

Reza Eslamian[1], Sam Moslemi[2], Sina A Sharghi[3*], Amir Reza Fazeli[4]

[1] *Division of Thoracic Surgery, Department of Surgery, Shariati Hospital, Tehran University of Medical Sciences, Tehran, Iran.*
[2] *Division of Endocrine Surgery, Department of Surgery, Shariati Hospital, Tehran University of Medical Sciences, Tehran, Iran.*
[3] *Division of Endocrine Surgery, Emam Ali Hospital, Alborz University of Medical Sciences, Karaj, Iran.*
[4] *Division of Colorectal Surgery, Department of Surgery, Imam Khomeini Hospital Complex, Tehran University of Medical Sciences, Tehran, Iran.*

Received: 2024-08-17; Received in revised form: 2024-09-20; Accepted: 2024-10-03

Abstract

Hydatid disease is mostly found in endemic areas such as the Middle East, particularly in patients with a certain history of exposure. Signs and symptoms depend on the organ infected by the cysts. Hepatic cysts may cause abdominal pain and jaundice. Pulmonary hydatid cysts, on the other hand, usually occur in the lower lobe of the right lung and present with chest pain, cough, or shortness of breath. Rupture occurs in about one-third of patients, releasing a highly antigenic fluid, which can result in anaphylaxis when ruptured into the bronchus. Alternatively, although not common, pulmonary hydatid cyst rupture may result in pleural effusion or, rarely, pneumothorax. Our patient was a young man who presented with pneumothorax resulting from a ruptured pulmonary hydatid cyst and concurrent extensive DVT. The cysts were completely resected, and the underlying parenchyma was repaired through a posterolateral thoracotomy, while DVT was managed by anti-thrombotic therapy.

Keywords: Hydatid Cyst, Echinococcus Granulosus, Pneumothorax, Deep Vein Thrombosis

Citation: Eslamian R, Moslemi S, A Sharghi S, Fazeli AR. **A Rare Case of Pneumothorax Caused by Ruptured Pulmonary Hydatid Cysts Accompanied by Deep Vein Thrombosis: A Case Report.** *Acad J Surg*, 2024; 7(3): 59-64.

Introduction

Hydatid cysts caused by the cestode *Echinococcus granulosus* are mostly found in the Middle East, Asia, Central and South America, and Europe. Humans serve as accidental intermediate hosts as the metacestodes mature into tapeworms in the definitive host's gastrointestinal (GI) tract, and released eggs are ingested into the human GI tract where they can transform into metacestodes. The liver is the most affected organ where the cysts are formed, followed by the lungs. However, the spleen, brain, eyes, heart, bone marrow, kidneys, and other rare organs may also be affected by the cysts [1]. Hydatid disease might have an indolent course as the cysts grow slowly [2]. Signs and symptoms depend on the location of the cysts. Hepatic cysts may cause abdominal pain or jaundice. Pulmonary hydatid cysts mostly occur in the lower lobe of the right lung and present with chest pain, cough, or shortness of breath [3]. Cyst rupture occurs in about one-third of patients, releasing a highly antigenic fluid, which can result in anaphylaxis, particularly when they rupture into the bronchus [4, 5]. Rupture into the pleural cavity may result in pleural effusion or, rarely, pneumothorax [6]. Here we report a young man who presented with pneumothorax resulting from a left ruptured pulmonary hydatid cyst.

Case Presentation

A 23-year-old man from Afghanistan presented to the ED with shortness of breath and cough, which started about a month ago and gradually became more severe. The patient, who was a construction worker, was forced to withdraw from work since the onset of symptoms. He was tachypneic and had decreased blood oxygen saturation. Vital signs were as follows: PR: 70, RR: 30, BP: 120/80, T: 36.5, O_2 sat: 87%, which reached 90% with O_2 mask ventilation support. On physical examination, he was an average height, thin young man, with general malaise and weakness.

*** Corresponding author**: Sina A Sharghi
Division of Endocrine Surgery, Emam Ali Hospital, Alborz University of Medical Sciences, Karaj, Iran.
Tel.: +98 939 779 9157 Email: sina_ea23@yahoo.com

Chest wall movement and lung sounds were notably decreased in the left hemithorax. Additionally, his left leg appeared swollen below the knee, with mild to moderate non-pitting edema, making distal pulses difficult to palpate. His VBG revealed respiratory acidosis: pH: 7.28, PCO_2: 57, HCO_3: 25, BE: -1.8. He also had high levels of CRP (CRP: 29.9 mg/L) and D-dimer (D-dimer: 5.3 µg/mL). Other laboratory findings were within normal limits.

A chest X-ray showed a significant left pneumothorax and mild pleural effusion, so tube thoracostomy was performed using a 32 Fr chest tube (Figure 1). The tube had significant oscillation and minor bubbling, and its location was confirmed by another CXR. Pleural fluid analysis suggested exudative fluid with no growth of microorganisms. Mycobacterium PCR from the pleural fluid was negative. The patient's O_2 saturation reached 90-91% without O_2 ventilation support.

After the patient was stabilized, a lower extremity color Doppler ultrasound showed thrombosis in the left popliteal vein extending to the superficial femoral and common femoral veins. An abdominal CT scan was normal. Treatment with IV heparin was initiated.

By the next day, he was semi-seated, with incentive spirometry and respiratory physiotherapy performed regularly. Although reduced in severity, he still felt shortness of breath and needed respiratory support with an O_2 mask. Chest tube oscillation and bubbling were still present by day 5. Due to the persistent leak, bronchoscopy was performed, which showed no endotracheal pathologic findings. There was no significant change in the patient's condition, so we performed a thoracic CT scan. There was a near-complete collapse of the left lung, with significant amounts of debris (Figure 2).

We decided to perform diagnostic video-assisted thoracoscopic surgery (VATS) as the patient's condition had failed to improve. The pleural space was filled with debris, the left lung was collapsed and almost completely overlaid by thickened peel and debris, and three ruptured hydatid cysts were found

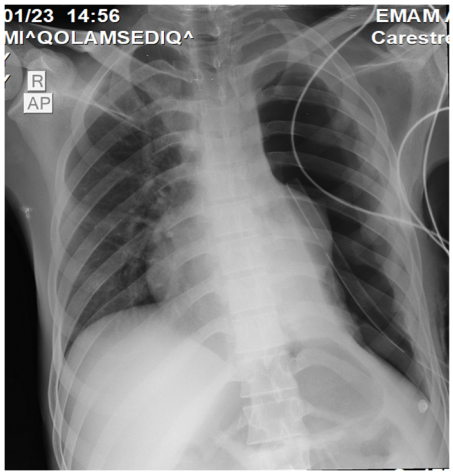

Fig. 1: Left pneumothorax and left lung collapse, a 32-fr chest tube is placed in the plural space.

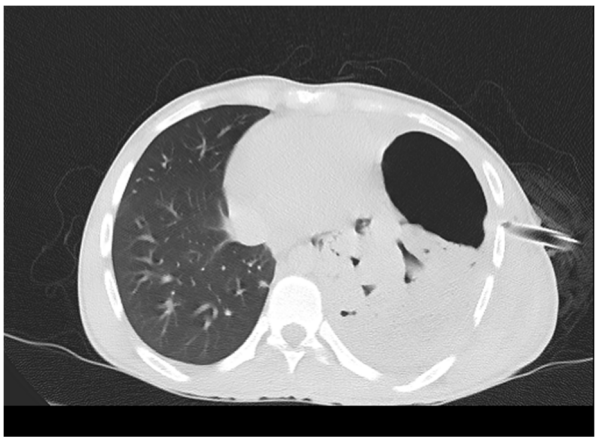

Fig. 2: Near-complete collapse of the left lung, with significant amounts of debris.

within the lung peripheral parenchyma in the left lower lobe. The debris was suctioned and irrigated, and the cyst walls with their germinal layers were carefully detached from the lung parenchyma and extracted (Figure 3). Finally, a 32 Fr chest tube was placed in the pleural space.

Days after the initial surgery, he remained stable, had minimal improvement in respiratory function, but no significant change in O_2 saturation as it remained 90-91%, and oscillation and bubbling persisted. Treatment with albendazole, 400 milligrams twice daily, was initiated while he also tested positive for echinococcal antibody. CXR demonstrated that the left lung was still completely collapsed, despite the chest tube being connected to two-bottle Gomco suction.

The patient was planned for thoracotomy to search for possible air leaks. A posterolateral left thoracotomy was done. The left lung was found completely collapsed with no expansion when two lung ventilation was tried, and the layer of debris and peel had gotten even more thickened and was attached all over it (figure 4). Complete decortication was done, and the lung parenchyma was freed and began to expand. A leak test with saline was performed and two major leak sites were identified in the lower lobe. Bronchopleural fistulas were repaired with 3,0 PDS suture. There were no leaks anymore, and the lung was partially expanded (figure 5). Two 32 fr chest tubes were placed and the patient was transported to the ICU.

Post operatively, no serious complications occurred, other than a surgical site infection which was treated with a combination of meropenem and vancomycin. Gomko suction continued while incentive spirometry and respiratory physiotherapy were done as frequently as possible. By day 5 post operation, CXR and chest CT scan showed a 60-70% left lung expansion (Figure 6), he had significantly improved, felt no more shortness of breath, the respiratory acidosis was corrected, and he had a blood oxygen saturation of 98-99%. Also, leg edema had almost resolved. Finally, he was discharged with albendazole 400mg BD and 80mg subcutaneous enoxaparin per day. He was able to go back to work two weeks after discharge.

Discussion

Hydatid disease, caused by *Echinococcus* organisms, is seen in developing countries, especially in patients with certain histories of exposure to domestic animals such as sheep, or consumption of raw vegetables[7]. Patients may present with a variety of signs and symptoms depending on the location of the cysts and their integrity. The lungs serve as the second most common site of involvement, and pulmonary hydatid cysts may cause respiratory symptoms such as cough, shortness of breath, or anaphylaxis, particularly when cysts rupture [1, 8].

Hamouri et al. studied 43 children (mean age 13±4 years) with pulmonary hydatid cysts retrospectively. Among them, 17 patients had at least one ruptured hydatid cyst. Most of them had intrabronchial ruptures, and only 4 patients (7%) had their cysts rupture into the pleural cavity, causing pneumothorax in 3 patients [9]. Karimi et al. reported a case of a

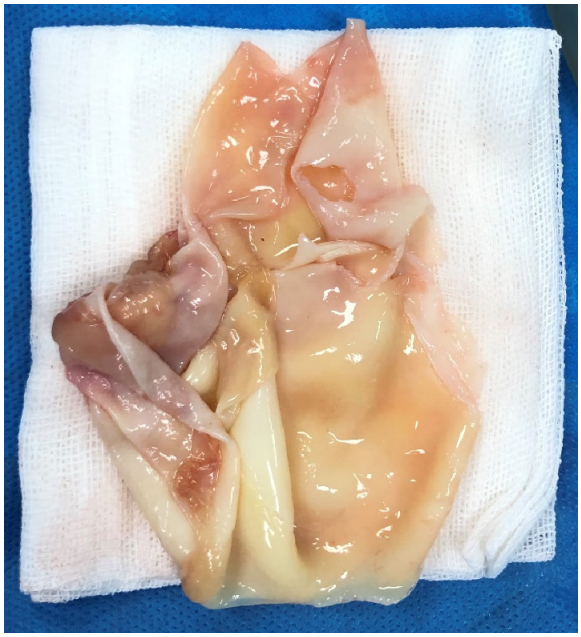

Fig. 3: Cyst walls were resected along with their germinal layers.

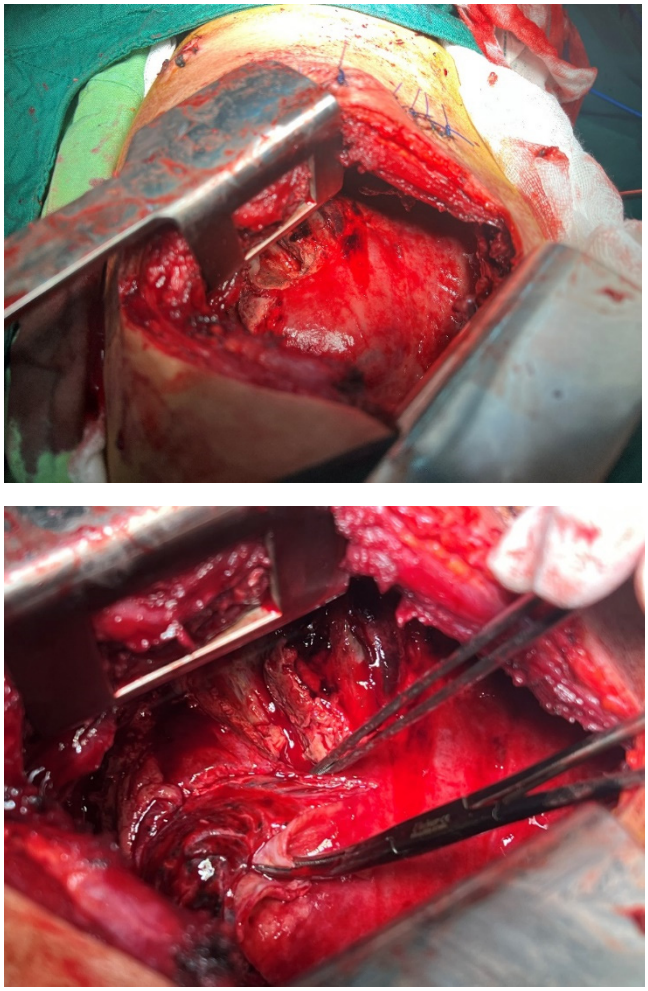

Fig. 4: A: Complete collapse of left lung overlayed by thick peural peel; B: Decortication of left lung

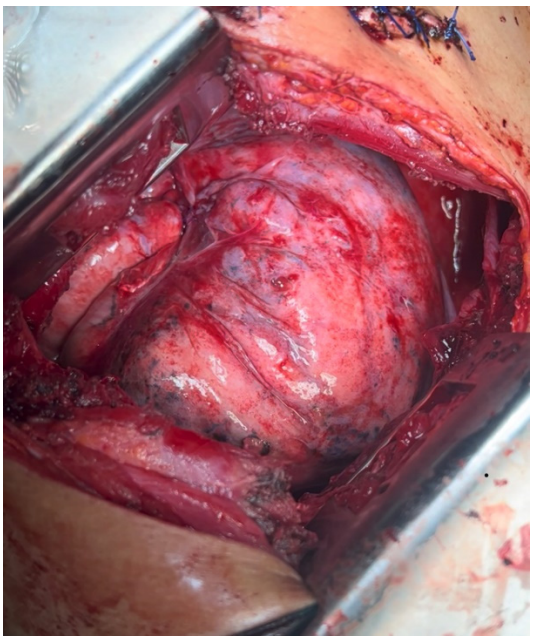

Fig. 5: re-expansion of the left lung after complete decortication.

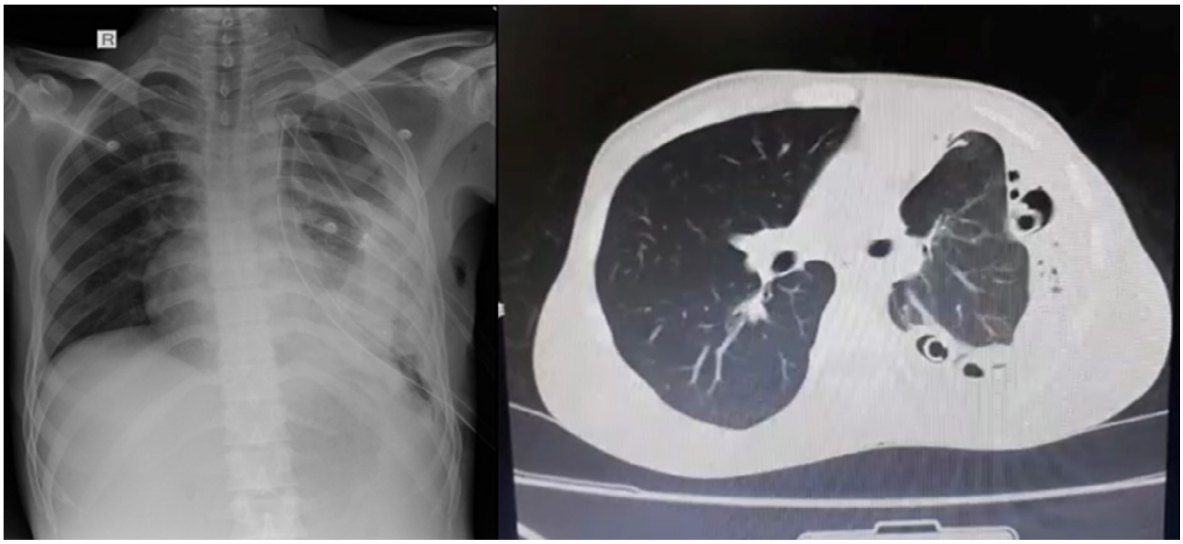

Fig. 6: Left lung expansion demonstrated in CXR (left) and Chest CT scan (Right)

ruptured pulmonary hydatid cyst presenting with fever, cough, vomiting, shortness of breath, and weight loss. They also reported an itchy rose spot on the patient's throat and chest. Radiographic evidence of a multilobulated cyst was evident on CXR and confirmed by thoracic CT scan, and the cyst was evacuated through a posterolateral thoracotomy[2]. Acharya et al. also reported a ruptured cyst that caused tension pneumothorax, managed by pigtail insertion and surgical cyst removal after confirmation of the diagnosis with a thoracic CT scan[6].

Our patient presented with a history of shortness of breath which gradually worsened, leading him to present to the OR. Radiographic diagnosis of the cysts was not possible due to the complete collapse of the affected lung, as they became evident during thoracoscopy. Our management was consistent with Hamouri et al., as they also stated that pleural decortication is vital in cases of long-standing empyema, hemothorax, or pleural tumors, for the lung parenchyma to be able to expand [10]. In cases of pulmonary hydatid disease, it is always important to suspect a leak from the lung parenchyma after cyst removal. In our case, leak sites were mostly overlaid by the thick layer of peel and debris and became apparent after complete decortication was done.

Some authors have also mentioned vascular complications due to ruptured hydatid cysts. Venous thrombosis is thought to result from direct compression by the cysts, particularly hepatic cysts near the IVC, or even transmission of cyst contents into the venous system. Rasheed et al. reported a case of hepatic hydatid cysts, where the patient presented to the ER with right upper quadrant abdominal pain, jaundice, vomiting, fever, and ascites. An abdominal CT scan revealed a filling defect in the IVC suggesting a thrombus, which was later confirmed by Doppler study, with involvement of the right and main portal vein, causing Budd-Chiari syndrome. They managed the extensive thrombosis with an intravenous drip of heparin [10]. Kirmizi et al. also reported a 33-year-old male with hepatic hydatid cysts and portal vein thrombosis (PVT), which was visualized in CT scan, MRI, and Doppler study. They managed the PVT with a meso-caval shunt [11]. Even pulmonary thromboembolism is reported by Poyraz et al., resulting from a hepatic cyst rupture into the IVC [12]. Our patient, on the other hand, had pulmonary hydatid cysts and his liver was intact. He also presented with left leg swelling, and Doppler study revealed extensive thrombosis in the left popliteal vein, extending to the superficial femoral and common femoral veins, involving the ipsilateral common iliac vein and infrarenal IVC. Extensive deep vein thrombosis in this case could result from cysts rupturing into pulmonary veins, or even long-term immobilization, as he had been bedridden for about a month. We also managed this complication with IV heparin and discharged the patient with subcutaneous enoxaparin as his symptoms were relieved.

Conclusion

Pneumothorax is a rare complication of ruptured pulmonary hydatid cysts. It is managed by early tube thoracostomy, followed by surgical intervention to evacuate the cysts. Diagnosis of hydatid disease may be more complicated in cases of lung collapse, as the cysts may not be evident in early imaging studies but must always be kept in mind when facing a collapsed lung that does not expand despite thoracostomy, particularly in patients with a suggestive exposure history.

References

1. Rostami A, Mozafari M, Gholipourmalekabadi M, Caicedo HH, Lasjerdi Z, Sameni M, Samadikuchaksaraei A. Optimization of fluoride-containing bioactive glasses as a novel scolicidal agent adjunct to hydatid surgery. Acta Trop. 2015.148:105-14. https://doi.org/10.1016/j.actatropica.2015.04.021
2. Karimi M, Rostami A, Spotin A, Rouhani S. Ruptured pulmonary hydatid cyst: a case report. J Parasit Dis. 2017 Sep;41(3):899-902. https://doi.org/10.1007/s12639-017-0880-z
3. Aydin Y, Ulas AB, Ahmed AG, Eroglu A. Pulmonary Hydatid Cyst in Children and Adults: Diagnosis and Management. Eurasian J Med. 2022 Dec;54(Suppl1):133-140. https://doi.org/10.5152/eurasianjmed.2022.22289
4. Karimi M, Rostami A, Spotin A, Rouhani S. Ruptured pulmonary hydatid cyst: a case report. J Parasit Dis. 2017 Sep;41(3):899-902. https://doi.org/10.1007/s12639-017-0880-z
5. Shameem M, Akhtar J, Bhargava R, Ahmed Z, Khan NA, Baneen U. Ruptured pulmonary hydatid cyst with anaphylactic shock and pneumothorax. Respir Care. 2011 Jun;56(6):863-5. https://doi.org/10.4187/respcare.00821
6. Acharya AB, Bhatta N, Mishra DR, Verma A, Shahi R. Rare Cause of Tension Pneumothorax: Hydatid Disease of Lung: A Case Report. JNMA J Nepal Med Assoc. 2020 Apr 30;58(224):265-268. https://doi.org/10.31729/jnma.4693
7. Rostami A, Ebrahimi M, Mehravar S, Fallah Omrani V, Fallahi S, Behniafar H. Contamination of commonly consumed raw vegetables with soil transmitted helminth eggs in Mazandaran province, northern Iran. Int J Food Microbiol. 2016 May 16. 225:54-8. https://doi.org/10.1016/j.ijfoodmicro.2016.03.013
8. Santivanez S, Garcia HH. Pulmonary cystic echinococcosis. Curr Opin Pulm Med. 2010 May;16(3):257-61. https://doi.org/10.1097/MCP.0b013e3283386282
9. Hamouri S, Odat H, Syaj S, Hecker E, Alrabadi N. Rupture of pulmonary hydatid cyst in pediatrics: A cross-sectional study. Ann Med Surg (Lond). 2021 Jan 8;62:31-36. https://doi.org/10.1016/j.amsu.2021.01.001
10. Rasheed K. Hydatid cyst of liver complicated with budd-Chiari syndrome and portal vein thrombosis. N Am J Med Sci. 2013 Mar;5(3):242-4. https://doi.org/10.4103/1947-2714.109206
11. Kirmizi S, Kayaalp C, Yilmaz S. Hydatid liver cyst causing portal vein thrombosis and cavernous transformation: a case report and literature review. Gastroenterol Hepatol Bed Bench. 2016;9(4):331-335.
12. Poyraz N, Demirbaş S, Korkmaz C, Uzun K. Pulmonary Embolism Originating from a Hepatic Hydatid Cyst Ruptured into the Inferior Vena Cava: CT and MRI Findings. Case Rep Radiol. 2016; 2016:3589812. https://doi.org/10.1155/2016/3589812