ORIGINAL RESEARCH PAPER

Cardiovascular Surgery

RUPTURED PULMONARY HYDATID CYST PRESENTING AS HYDROPNEUMOTHORAX WITH EOSINOPHILIC PLEURAL EFFUSION AND TRAPPED LUNG SUCCESSFULLY MANAGED BY VIDEO-ASSISTED THORACOSCOPIC SURGERY (VATS): A CASE REPORT AND LITERATURE REVIEW

KEY WORDS: Pulmonary Hydatid Disease; Echinococcus Granulosus; Hydropneumothorax; Trapped Lung; Decortication; VATS; Albendazole.

Dr Tejwaswi Yadavalli — Intern, Mater Hospital, Townsville, Australia

Dr Manjunath Bale — Senior Consultant Thoracic Surgeon, Apollo Institute of Medical Sciences and Research, Jubilee Hills, Hyderabad.

Dr Hitesh Billa Lakshmi — Consultant Allergy Specialist and Assistant Professor, Department of Interventional Pulmonology, Apollo Institute of Medical Sciences and Research, Jubilee Hills, Hyderabad.

Dr Nithya Reddy Kalluri — Post Graduate 1ST Year, Department of Interventional Pulmonology, Apollo Institute of Medical Sciences and Research, Jubilee Hills, Hyderabad.

Dr Indhu Mallipeddi — Post Graduate 1ST Year, Department of Interventional Pulmonology, Apollo Institute of Medical Sciences and Research, Jubilee Hills, Hyderabad.

ABSTRACT

Background: Pulmonary hydatid disease caused by Echinococcus granulosus remains an important parasitic disease in endemic regions. Although the liver is the most commonly affected organ, pulmonary involvement accounts for approximately 10–30% of cases. Rupture of pulmonary hydatid cysts may lead to life-threatening complications including pneumothorax, hydropneumothorax, empyema, anaphylaxis, and respiratory failure. Development of trapped lung following rupture is uncommon and often necessitates surgical intervention. **Case Presentation:** A 48-year-old male farmer presented with acute worsening of dyspnea, pleuritic chest pain, and cough of three days duration on a background of progressive exertional breathlessness over four months. Clinical examination revealed respiratory distress and features suggestive of a large left-sided hydropneumothorax. Intercostal drainage was performed. Pleural fluid analysis demonstrated an exudative eosinophilic effusion. Cytological examination unexpectedly revealed hooklets of Echinococcus granulosus, establishing the diagnosis of ruptured pulmonary hydatid cyst. Contrast-enhanced computed tomography demonstrated a multiloculated hydropneumothorax with detached endocyst membranes and non-expansion of the left lung despite adequate drainage, suggestive of trapped lung. Hepatic involvement was excluded. Following initiation of albendazole therapy and comprehensive pre-operative assessment, the patient underwent video-assisted thoracoscopic surgery (VATS) with removal of hydatid membranes and decortication. Postoperatively, complete lung re-expansion was achieved with satisfactory recovery. Albendazole therapy was continued postoperatively. **Conclusion:** Ruptured pulmonary hydatid cyst should be considered in the differential diagnosis of eosinophilic pleural effusion and spontaneous hydropneumothorax, especially in endemic regions. Early recognition, appropriate anti-helminthic therapy, and definitive surgical management can achieve excellent outcomes even in complicated cases associated with trapped lung.

INTRODUCTION

Hydatid disease is a zoonotic parasitic infestation caused predominantly by Echinococcus granulosus. It remains endemic in many developing countries including India, the Middle East, South America, Africa, and Mediterranean regions. Humans become accidental intermediate hosts through ingestion of parasite eggs.

The lungs represent the second most common site of hydatid involvement after the liver. Pulmonary hydatid cysts may remain asymptomatic for prolonged periods and are often discovered incidentally. Symptomatic disease usually results from enlargement, infection, or rupture of the cyst.

Rupture into the pleural cavity is relatively uncommon but may result in hydropneumothorax, eosinophilic pleural effusion, empyema, bronchopleural fistula, anaphylactic reactions, and trapped lung. Surgical intervention remains the cornerstone of treatment, while perioperative albendazole therapy reduces recurrence risk and addresses occult disease.

We report a rare case of ruptured pulmonary hydatid cyst presenting with eosinophilic hydropneumothorax and trapped lung requiring VATS decortication.

Case Presentation

A 48-year-old male farmer presented with acute worsening of shortness of breath for three days associated with pleuritic chest pain, cough, occasional expectoration, and intermittent low-grade fever.

Detailed history revealed progressive exertional dyspnea for four months. The patient also reported loss of appetite for three months and unintentional weight loss of approximately 5 kg over two months.

There was no history of tuberculosis, diabetes mellitus, hypertension, chronic respiratory disease, previous thoracic surgery, or malignancy. He denied exposure to household pets or cattle. There was no history suggestive of connective tissue disease or occupational exposure to toxic fumes.

On arrival, the patient was conscious and oriented.

Vital parameters: Heart rate: 110 beats/minute, Respiratory rate: 28 breaths/minute, Blood pressure: 100/70 mmHg, SpO2: 91% on room air and afebrile

Respiratory examination revealed markedly reduced air entry over the left hemithorax with hyperresonance in upper zones. Tracheal deviation toward the right side and mediastinal shift were noted.

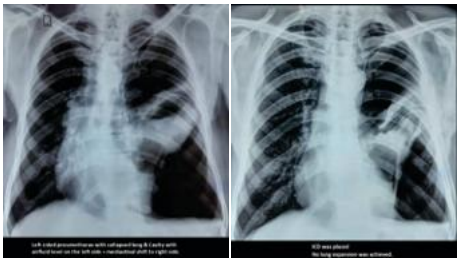
Laboratory Investigations

Initial blood investigations demonstrated mild leukocytosis

with peripheral eosinophilia, Inflammatory markers were mildly elevated , Liver and renal function tests were within acceptable limits.

Chest Radiography

Chest radiograph demonstrated a large left-sided hydropneumothorax with partial collapse of the underlying lung and contralateral mediastinal shift.



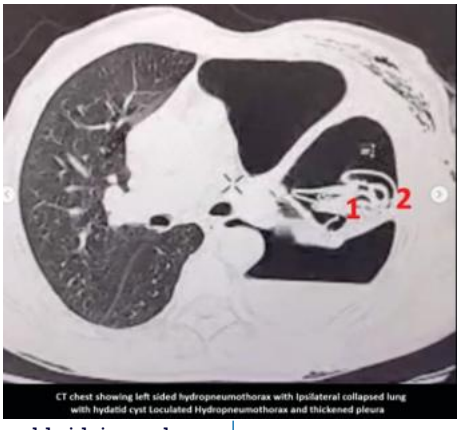
Management in Emergency Department -Oxygen supplementation was initiated.Nebulized bronchodilator therapy was administered. An intercostal chest drain was inserted resulting in evacuation of air and pleural fluid.

Pleural Fluid Analysis revealed: Exudative effusion, Predominant eosinophilic inflammation, Negative bacterial cultures, Negative malignant cytology. Remarkably, microscopic cytological examination demonstrated hooklets consistent with Echinococcus granulosus.

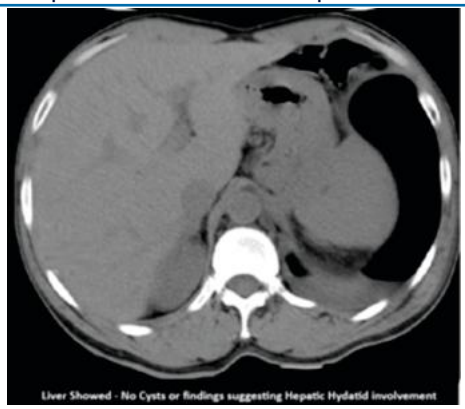
Hooklets of echinococcus in the pleural fluid cytology and microscopy

This finding established the diagnosis of ruptured pulmonary hydatid cyst with pleural contamination

Computed Tomography Findings - Large multiloculated left hydropneumothorax, Intercostal drain within one of the locules, Detached floating hydatid membranes, Pleural thickening ,Failure of left lung expansion despite adequate drainage *and* Features suggestive of trapped lung *with* No obvious contralateral pulmonary lesions



CT chest showing left sided hydropneumothorax with Ipsilateral collapsed lung with hydatid cyst Loculated Hydropneumothorax and thickened pleura



Liver Showed - No Cysts or findings suggesting Hepatic Hydatid involvement

Additional Evaluation

Ultrasonography and CT evaluation of the abdomen demonstrated no hepatic hydatid cysts.

Infectious disease consultation was obtained- Albendazole therapy was initiated pre-operatively.

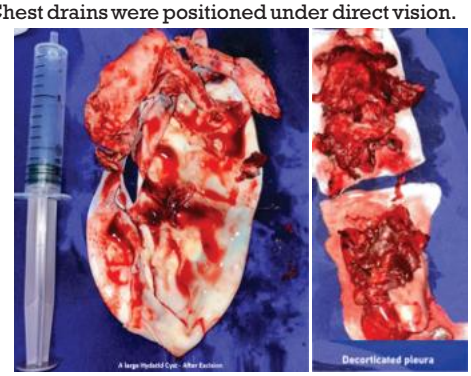
Cardiology evaluation and anesthetic assessment were completed. Flexible bronchoscopy was performed to exclude an endobronchial obstruction contributing to non-expansion of the lung and was unremarkable.

Definitive Surgical Management

Following multidisciplinary discussion involving thoracic surgery, pulmonology, infectious diseases, anesthesia, and radiology teams, definitive surgical intervention was planned.

Video-assisted thoracoscopic surgery (VATS) was performed.

Intraoperative findings included: Multiple detached hydatid membranes, Residual parasitic material, Dense visceral pleural peel, Entrapped left lung, Pleural inflammatory adhesions, No active bronchopleural fistula identified, Complete removal of hydatid membranes and parasitic remnants was performed., Visceral pleural decortication was undertaken to release the trapped lung.Thorough pleural cavity irrigation with scolicidal precautions was performed and Chest drains were positioned under direct vision.

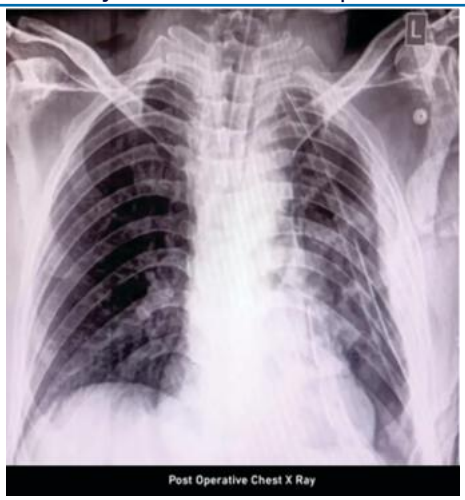


Decorticated pleura

Postoperative Course

The patient was extubated successfully, Aggressive pulmonary rehabilitation was instituted including: Early ambulation, Chest physiotherapy, Incentive spirometry, Adequate analgesia, Deep breathing exercises - Serial chest radiographs demonstrated progressive re-expansion of the left lung.

No persistent air leak was observed.Drain output gradually reduced and chest drains were subsequently removed.The patient remained hemodynamically stable throughout recovery. Postoperative imaging confirmed satisfactory lung expansion. Albendazole therapy was continued postoperatively and the patient was discharged in stable condition with planned outpatient follow-up.



Post Operative Chest X Ray

DISCUSSION

Hydatid disease remains a significant public health problem in many endemic regions including the Indian subcontinent, the Mediterranean basin, South America, the Middle East, and parts of Africa. The disease is caused predominantly by Echinococcus granulosus, a cestode parasite that completes its life cycle between canids as definitive hosts and herbivorous animals as intermediate hosts. Humans become accidental intermediate hosts through ingestion of parasite eggs, leading to the development of hydatid cysts in various organs. While hepatic involvement accounts for approximately 60–70% of cases, the lungs represent the second most frequently affected organ, accounting for nearly 20–30% of reported cases. The relatively compressible nature of pulmonary parenchyma permits considerable cyst enlargement before symptoms become clinically evident, explaining why many patients remain asymptomatic until complications occur.[1-4]

Pulmonary hydatid cysts may present with cough, chest pain, dyspnea, hemoptysis, or expectoration of cyst contents. However, rupture of a pulmonary hydatid cyst into the pleural space is an uncommon but potentially life-threatening complication. The incidence of pleural complications has been reported to range between 2% and 17% depending on the patient population and stage of disease. Rupture may occur spontaneously because of progressive enlargement and increased intracystic pressure, secondary infection, trauma, or erosion into adjacent structures. Following rupture, hydatid fluid, protoscolices, and membrane fragments may enter the pleural cavity, provoking an intense inflammatory response and leading to hydropneumothorax, pleural effusion, empyema, bronchopleural fistula formation, or even anaphylactic reactions.[5-8]

The present case demonstrates several unusual and clinically important features. First, the patient presented with a large hydropneumothorax associated with eosinophilic pleural effusion. Eosinophilic pleural effusion is defined by the presence of at least 10% eosinophils in pleural fluid and constitutes approximately 5–16% of all pleural effusions. Although air or blood in the pleural cavity, malignancy, tuberculosis, medications, and autoimmune diseases are more commonly implicated, parasitic infestations remain important causes in endemic settings. The presence of eosinophilic pleural effusion in the current patient likely reflected a robust immunological response to hydatid antigens released following cyst rupture.[9-11]

A particularly noteworthy aspect of this case was the identification of Echinococcus hooklets on pleural fluid cytology. While serological testing and imaging studies are commonly employed in the diagnosis of hydatid disease, direct visualization of hooklets or scolices remains pathognomonic. Such findings are infrequently encountered in routine clinical practice and provide definitive confirmation of the diagnosis. Reports describing diagnosis through pleural fluid cytology remain limited in the literature, making the present case educationally valuable for clinicians managing unexplained eosinophilic pleural effusions.[12,13]

Computed tomography played a crucial role in characterizing the extent of disease and guiding management. Classical radiological signs of ruptured pulmonary hydatid cysts include the water-lily sign, cumbo sign, serpent sign, and floating membrane sign. In our patient, CT imaging demonstrated hydropneumothorax, detached cyst membranes, loculated pleural collections, and failure of lung re-expansion despite adequate pleural drainage. These findings suggested the presence of significant pleural inflammation and organization. CT imaging remains the modality of choice for evaluating complicated pulmonary hydatid disease and identifying associated pleural complications requiring surgical intervention.[14-16]

One of the most remarkable findings in the present case was the development of trapped lung. Trapped lung refers to the inability of the lung to fully expand because of a fibrous visceral pleural peel that mechanically restricts expansion. Inflammatory injury within the pleural space stimulates fibroblast proliferation and collagen deposition, ultimately resulting in formation of a dense visceral rind. Persistent non-expansion of the lung despite appropriately positioned chest drainage should raise suspicion for this condition. While trapped lung is well recognized following empyema, hemothorax, and chronic pleural inflammation, reports associated with ruptured pulmonary hydatid disease are exceedingly rare. In the present patient, prolonged exposure of the pleural cavity to parasitic antigens and inflammatory mediators likely contributed to the development of significant pleural fibrosis necessitating surgical decortication.[17-18]

The cornerstone of treatment for pulmonary hydatid disease remains surgery. The primary objectives are complete elimination of parasitic elements, prevention of recurrence, preservation of functional lung tissue, and management of complications. Surgical approaches range from cystotomy with capitonnage to wedge resection, segmentectomy, lobectomy, or decortication depending on disease severity and the condition of the surrounding lung. Current thoracic surgical principles emphasize parenchymal preservation whenever feasible. In complicated cases involving pleural contamination, organized pleural disease, or trapped lung, decortication may be necessary to restore pulmonary expansion and function.[19-21]

Video-assisted thoracoscopic surgery has emerged as an attractive alternative to conventional thoracotomy in selected patients with pulmonary hydatid disease. Numerous studies have demonstrated reduced postoperative pain, shorter hospital stay, earlier ambulation, decreased respiratory complications, and faster recovery compared with open surgery. In the present case, VATS allowed excellent visualization of the pleural cavity, safe removal of hydatid membranes, effective decortication of the trapped lung, and complete re-expansion of the underlying pulmonary parenchyma. The successful outcome observed in our patient further supports the expanding role of minimally invasive thoracic surgery in the management of complicated pulmonary hydatid disease.[22-24]

Albendazole therapy constitutes an important adjunct to surgical treatment. Albendazole acts by inhibiting microtubule formation within parasitic cells, thereby impairing glucose uptake and energy metabolism. Current World Health Organization Informal Working Group on Echinococcosis recommendations support administration of albendazole at doses of 10–15 mg/kg/day in two divided

doses. Preoperative therapy is generally advised for one to four weeks before surgery to reduce parasite viability and minimize the risk of secondary implantation following inadvertent spillage. Postoperative continuation for at least one to three months is recommended, particularly in cases involving rupture, pleural contamination, multiple cysts, recurrent disease, or intraoperative spillage. Monitoring should include periodic liver function testing and complete blood counts because hepatotoxicity and leukopenia represent the most significant adverse effects.[25-28]

The successful outcome in our patient can be attributed to early recognition of a rare etiology of hydropneumothorax, prompt pleural drainage, accurate cytological diagnosis, multidisciplinary planning, perioperative antihelminthic therapy, and definitive thoracoscopic surgical intervention. This case highlights the importance of maintaining a high index of suspicion for hydatid disease in endemic regions when evaluating patients presenting with eosinophilic pleural effusion and spontaneous hydropneumothorax.

CONCLUSION

Ruptured pulmonary hydatid cyst should be considered an important differential diagnosis in patients presenting with hydropneumothorax and eosinophilic pleural effusion, particularly in endemic areas. Cytological demonstration of hydatid hooklets in pleural fluid provides definitive diagnosis and may obviate the need for additional invasive procedures. Persistent failure of lung expansion following chest tube drainage should prompt evaluation for trapped lung. Definitive management requires a multidisciplinary strategy integrating antihelminthic therapy and surgery. In selected patients, VATS decortication offers a safe and effective minimally invasive approach, resulting in complete lung re-expansion, symptom resolution, and excellent postoperative recovery.

REFERENCES

1. Brunetti E, Kern P, Vuitton DA. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. Acta Trop. 2010;114(1):1-16.
2. Wen H, Vuitton L, Tuxun T, Li J, Vuitton DA, Zhang W, et al. Echinococcosis: advances in the 21st century. Clin Microbiol Rev. 2019;32(2):e00075-18.
3. Morar R, Feldman C. Pulmonary echinococcosis. Eur Respir J. 2003;21(6):1069-77.
4. Sarkar M, Pathania R, Jhobta A, Thakur BR, Chopra R. Cystic pulmonary hydatidosis. Lung India. 2016;33(2):179-91.
5. Aribas OK, Kanat F, Gormus N, Turk E. Pleural complications of hydatid disease. J Thorac Cardiovasc Surg. 2002;123(3):492-7.
6. Balci AE, Eren N, Eren S, Ulkü R. Ruptured hydatid cysts of the lung. Eur J Cardiothorac Surg. 2002;21(5):839-43.
7. Kuzucu A, Soysal O, Ozgel M, Yologlu S. Complicated hydatid cysts of the lung. Eur J Cardiothorac Surg. 2004;25(3):489-93.
8. Halezeroglu S, Celik M, Uysal A, Senol C, Keles M, Arman B. Giant hydatid cysts of the lung. J Thorac Cardiovasc Surg. 1997;113(4):712-7.
9. Light RW. Pleural Diseases. 7th ed. Philadelphia: Wolters Kluwer; 2022.
10. Ferreiro L, San José E, Valdés L. Eosinophilic pleural effusion. Arch Bronconeumol. 2013;49(11):484-9.
11. Oba Y, Abu-Salah T. The prevalence and diagnostic significance of eosinophilic pleural effusions. Eur Respir J. 2012;39(3):746-9.
12. Beggs I. The radiology of hydatid disease. AJR Am J Roentgenol. 1985;145(3):639-48.
13. Turgut AT, Altin L, Topçu S, Kiliço lu B, Aliinok T, Kaptano lu E, et al. Unusual imaging manifestations of hydatid disease. Eur J Radiol. 2007;63(1):84-93.
14. Akgul Ozmen C, Onat S, Yaldiz S. Imaging findings of pulmonary hydatid disease. Diagn Interv Radiol. 2015;21(1):28-33.
15. Do an R, Yüksel M, Cetin G, Süzer K, Alp M, Kaya S, et al. Surgical treatment of hydatid cysts of the lung. Ann Thorac Surg. 1989;48(4):495-501.
16. Burgos R, Varela A, Castedo E, Roda J, Montero CG, Serrano S, et al. Pulmonary hydatidosis: surgical treatment and follow-up of 240 cases. Eur J Cardiothorac Surg. 1999;16(6):628-35.
17. Huggins JT. Trapped lung and non-expandable lung. Clin Chest Med. 2013;34(1):165-72.
18. Feller-Kopman D, Light R. Pleural disease. N Engl J Med. 2018;378(8):740-51.
19. Dziri C, Haouet K, Fingerhut A. Treatment of hydatid cyst of the liver: where is the evidence? Br J Surg. 2004;91(1):1-13.
20. Kern P. Clinical features and treatment of echinococcosis. Adv Parasitol. 2003;52:1-52.
21. McManus DP, Gray DJ, Zhang W, Yang Y. Diagnosis, treatment, and management of echinococcosis. BMJ. 2012;344:e3866.
22. Isitmangil T, Sebit S, Tunc H, Gorur R, Erdik O, Kunter E. Clinical experience of thoracoscopic treatment of pulmonary hydatid cyst disease. Surg Endosc. 2002;16:189-92.
23. Sayir F, Cobanoglu U, Mergan D. Video-assisted thoracoscopic surgery for pulmonary hydatid disease. Ann Thorac Med. 2012;7(1):30-3.
24. Tuxun T, Zhang JH, Zhao JM, Tai QW, Abudurexti M, Ma HZ, et al. World review of cystic echinococcosis. Int J Infect Dis. 2015;42:11-9.
25. WHO Informal Working Group on Echinococcosis. Guidelines for treatment of cystic echinococcosis. Geneva: World Health Organization.
26. Horton J. Albendazole in treatment of human cystic echinococcosis: 12 years of experience. Acta Trop. 1997;64(1-2):79-93.
27. Vuitton DA, Brunetti E. Medical treatment of echinococcosis. Parasite. 2014;21:28.
28. Gómez i Gavara C, López-Andújar R, Belda Ibáñez T, Ramia JM, Moya Herraiz Á, Orbis Castellanos F, et al. Review of current management of cystic echinococcosis. World J Gastroenterol. 2015;21(1):124-31.