

Loculated, Ruptured Primary Pleural Hydatid Cyst Mimicking Empyema: A Case Report

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Received: 07 January 2026

Revised: 30 May 2026

Accepted: 24 June 2026

Abstract

Primary pleural hydatid cyst is exceptionally rare, accounting for fewer than 1% of all hydatid disease cases. We reported the case of a 62-year-old woman from a rural area who presented with dyspnea, cough, and weight loss. Initial imaging suggested loculated empyema, and a tube thoracostomy drained purulent material without clinical resolution. Computed tomography (CT) demonstrated pleural thickening and effusion. Video-assisted thoracoscopic surgery (VATS) was performed under the presumptive diagnosis of fibrinopurulent empyema. Intraoperatively, a ruptured primary pleural hydatid cyst was identified and removed. Severe inflammation and fibrosis precluded immediate decortication, necessitating staged management with intrapleural irrigation followed by a subsequent thoracotomy. A superimposed infection with *Pseudomonas aeruginosa* and *Klebsiella pneumoniae* complicated the clinical course, requiring repeated interventions. A definitive posterolateral thoracotomy with decortication achieved recovery, and the patient returned to work. This case highlighted that ruptured pleural hydatid cyst can mimic empyema, elude detection by CT and ultrasound, and require clinical vigilance in endemic regions. Although VATS provided essential diagnostic confirmation, open thoracotomy might be required for definitive management.

Please cite this article as: Davari HR, Akbari Asbagh R. Loculated, Ruptured Primary Pleural Hydatid Cyst Mimicking Empyema: A Case Report. Iran J Med Sci. doi: 10.30476/ijms.2026.110437.4609.

Keywords • Hydatid cyst • Echinococcosis • Pleural diseases • Empyema • Thoracoscopy

What's Known

- Pleural hydatid cysts usually arise secondarily from hepatic or pulmonary lesions. Primary pleural cyst is rare and may be missed on sonography or computed tomography (CT).

What's New

- Primary pleural hydatid cyst can mimic empyema. Video-assisted thoracoscopic surgery (VATS) is invaluable for diagnosis and management, while resistant infection in the residual pleural space may require decortication even in the absence of frank pus.

Introduction

Hydatid disease, caused by the larval stage of *Echinococcus granulosus*, remains a significant public health concern in endemic regions, most commonly affecting the liver and lung.¹ Although hepatic and pulmonary hydatidosis are well documented, primary pleural hydatid disease is exceedingly rare, accounting for less than 1% of all cases.²⁻⁴ Since pleural involvement is usually secondary to transdiaphragmatic spread or rupture of adjacent pulmonary cyst, true primary pleural hydatidosis is often overlooked in differential diagnoses.^{3,4}

In this study, we reported a case of a ruptured primary pleural hydatid cyst presenting with clinical and radiologic features mimicking loculated empyema. Video-assisted thoracoscopic surgery (VATS) revealed a large ruptured pleural hydatid cyst as the underlying cause of chronic empyema. The diagnostic challenges, imaging pitfalls, and staged surgical management are discussed.

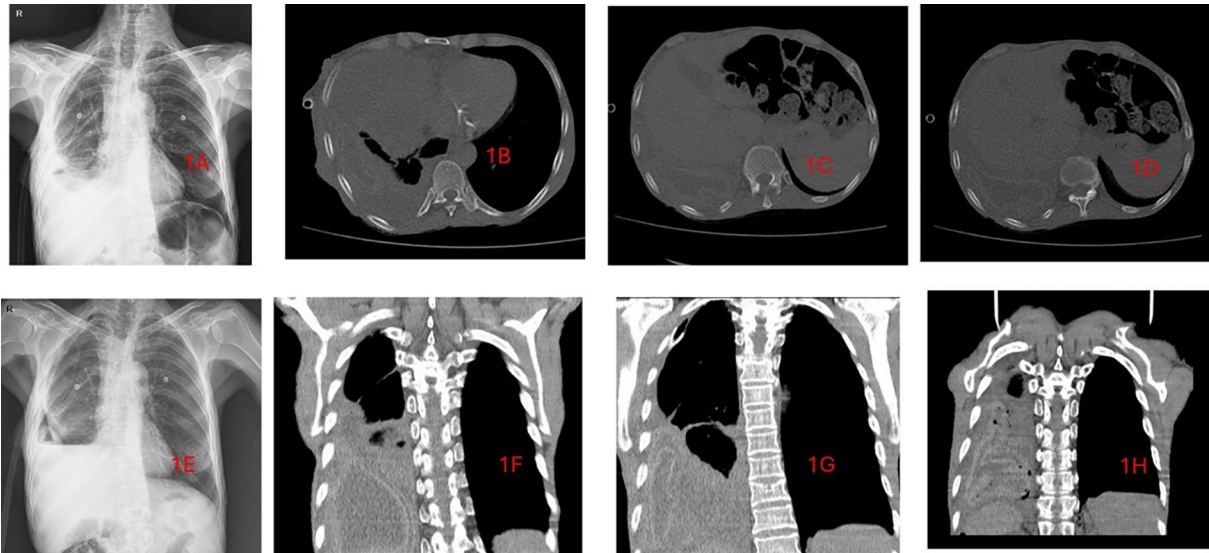


Figure 1: Chest radiography and computed tomography (CT) scan demonstrate a loculated pleural effusion with multiple septations before surgical intervention. 1A: The figure shows the initial chest radiograph obtained on admission, revealing an initially placed tube thoracostomy. 1B–1D: The figures display axial mediastinal window images in which a faint, overlooked double-layer cystic structure is visible. 1E: The figure shows a follow-up chest X-ray demonstrating a change in the tube thoracostomy position and the resulting residual pleural space. 1F–1H: The images present coronal mediastinal window images that again reveal the subtle, previously overlooked double-layer density.

Case Presentation

In September 2024, a 62-year-old woman from a rural area presented with dyspnea, cough, and significant weight loss. She was admitted to Namazi Hospital, Shiraz, Iran. Chest ultrasonography demonstrated a mild to moderate loculated pleural effusion with septations. An initial tube thoracostomy drained purulent material yet failed to achieve adequate resolution. She was referred to thoracic surgery and managed for empyema. Following a spiral computed tomography (CT) scan, a new dependent tube thoracostomy with an irrigation catheter was inserted (figure 1).

Despite empirical antibiotics and daily irrigation with 0.5% povidoneiodine (Povidone Iodine-Behvazan 10%, 60 mL Solution, Iran) in normal saline using a Gamco® suction device (FABSCO Medical Company, Iran), her condition did not improve. CT imaging showed marked pleural thickening and significant residual pleural space (figure 2). With a provisional diagnosis of fibrinopurulent empyema, VATS was performed.

Intraoperatively, a large, unilocular, ruptured primary pleural hydatid cyst was unexpectedly identified and removed. The pleural cavity was irrigated with copious normal saline and

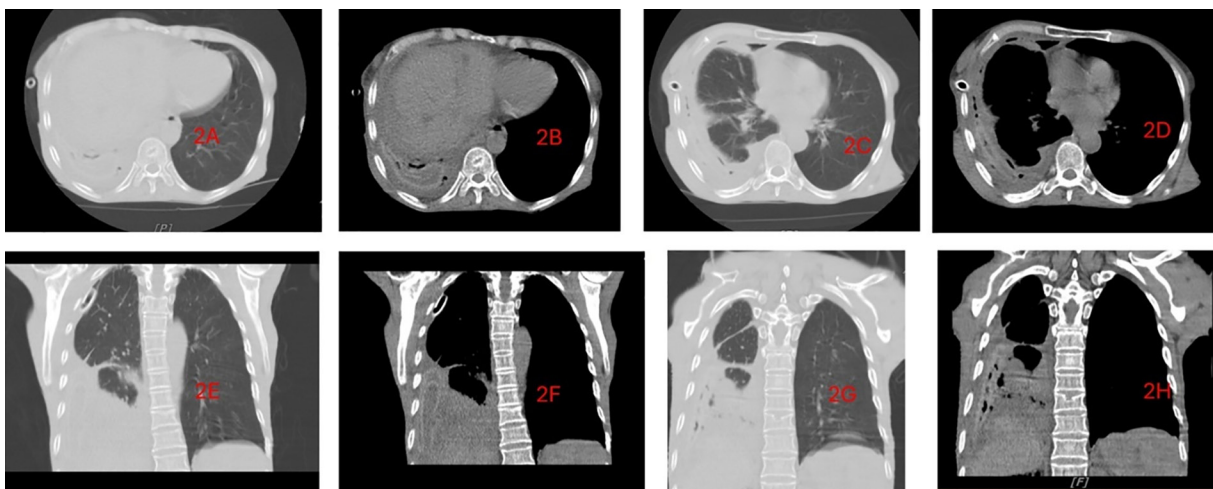


Figure 2: CT scan demonstrates persistent pleural thickening and extensive pleural space involvement after placement of a tube thoracostomy with an irrigation catheter, Gomco suction device, and Retiplate instillation. 2A–2D: The images show axial lung and mediastinal window images with a faint double-layer cystic structure visible on the mediastinal windows. 2E–2H: The figures show coronal lung and mediastinal window images obtained after repositioning the tube thoracostomy and initiating irrigation, Gomco suction, and Retiplate, again demonstrating the overlooked faint double-layer appearance without radiologic improvement.



Figure 3: The chest radiographs and postoperative imaging demonstrate findings following the second video-assisted thoracoscopy (VATS) procedure and subsequent thoracotomy. 3A: The figure shows post-irrigation chest X-ray demonstrating persistent pleural space despite adequate drainage. 3B and 3C: The images show follow-up posteroanterior and lateral chest X-rays demonstrating complete radiographic recovery.

0.5% povidoneiodine. Severe inflammation and dense fibrotic pleural thickening prevented decortication. Two thoracostomy tubes with irrigation catheters were placed, and intrapleural Retiplase (Mirel 10.4 IU Powder for injection, Royan Daru, Iran) was administered over 3 days with continuous suction. Cultures initially grew *Pseudomonas aeruginosa* ([Supplement Video: Spiral CT Scan post exchange tube thoracostomy with irrigation catheter.mp4](#)).

She was discharged with clear drainage and instructed to continue saline irrigation at home, with postponed elective decortication planned once her general condition improved. However, she was severely emaciated and initially unable to tolerate open thoracotomy. Nonadherence to the irrigation protocol resulted in readmission with recurrent purulent discharge, with cultures now growing *Klebsiella pneumoniae*. Given this, repeat VATS with irrigation was performed. After 2 weeks of daily irrigation, drainage became clear. However, cultures remained positive, with persistent pleural space (figure 3A).

A classic right posterolateral thoracotomy with decortication was ultimately performed. Due to her frailty and the risk of wound dehiscence, rib approximation was achieved using two wires. Her postoperative recovery was uneventful, and she eventually returned to work as a farm laborer (figures 3B and 3C).

This study was approved by the Ethics Review Board of Shiraz University of Medical Sciences (code: IR.SUMS.MED.REC.1404.503). Written informed consent was obtained from the patient for the publication of this case.

Discussion

Hydatid disease most frequently affects the liver and lung.^{1,2} Pleural involvement is rare, and true

primary pleural hydatid cyst is exceptionally uncommon, representing fewer than 1% of all cases.²⁻⁴ In most patients, the pleura is affected secondarily through transdiaphragmatic extension of hepatic cysts or rupture of adjacent pulmonary lesions.^{2,4}

Primary pleural hydatidosis refers to hydatid disease arising within the pleural cavity without hepatic or pulmonary involvement. Rupture of cyst in this location may release parasitic material into the pleural space, resulting in pleural effusion, pneumothorax, hydropneumothorax, empyema, pleural thickening, or secondary lung collapse. Although uncommon, rupture of pleural or pulmonary hydatid cyst can also trigger anaphylaxis, a rare and serious complication; and in rare instances, may erode through the chest wall as hydatid necessitans, similar to empyema necessitans.¹⁻⁸

These cysts are categorized among extrapulmonary intrathoracic hydatid lesions, which also include cysts of the parietal pleura, mediastinum, pericardium, diaphragm, interlobar fissures, and chest wall. Pleural hydatid cysts originate within the parietal pleura while they remain external to the visceral pleura layer. Clinically, primary pleural hydatidosis might present as solitary or multiple pleural cysts or as a parasitic pleural effusion. Up to 15% of patients may remain asymptomatic. Uncomplicated cysts are usually asymptomatic and discovered incidentally.^{4,5}

Complicated cysts become symptomatic as a result of enlargement or rupture, which may occur spontaneously or following trauma. Rupture may extend into the pericardium, lung parenchyma, mediastinum, pleural cavity, or even the peritoneal cavity. Clinical manifestations include chest pain, cough, hemoptysis, fever, and dyspnea. Intrathoracic rupture may lead to

empyema, pneumothorax, or pleural effusion. Secondary infection is possible, most commonly with *Haemophilus influenzae*. Pleural fluid typically shows eosinophilic predominance. Therefore, hydatid disease should be considered in the differential diagnosis of eosinophilic pleural effusion in endemic regions.⁶

Radiologic findings such as daughter cysts, the waterlily sign, and the serpent sign are considered pathognomonic. However, collapsed or infected cysts might be radiologically occult, particularly on CT.^{3, 7} Serological tests may support diagnosis, yet they have high false positive rates.⁴

Although ultrasound is highly effective for hepatic hydatid disease, it is less frequently used for thoracic cyst, except when there is chest wall involvement. When performed, however, it can offer valuable complementary information to CT, particularly by demonstrating complex cystic structures or revealing the double-line sign in cases with collapsed membranes. Its diagnostic role in intrathoracic extrapulmonary hydatidosis has also been described, notably in a series of 14 patients reported by Gursoy and colleagues.⁴

Management of primary pleural hydatidosis follows the same surgical principles applied to other intrathoracic hydatid lesions. Complete evacuation of cysts and pleural fluid following scolical inactivation, commonly with hypertonic saline, is essential, complemented by chest drainage and postoperative albendazole therapy.⁴

The differential diagnosis includes empyema, dermoid cysts, mesothelioma, synovial cell sarcoma, bronchogenic cyst, lymphoma, and pleural metastases.^{1, 3}

Our patient presented late in the disease course, with chronic illness, cachexia, dyspnea, and cough. She was initially treated as a case of empyema with supportive care, pleural irrigation, and antibiotics. Definitive diagnosis was established only during VATS, which revealed a large ruptured primary pleural hydatid cyst with no pulmonary or hepatic involvement.

Several reports described unruptured primary pleural hydatid cysts, some associated with multiple hydatid cysts and mediastinal shift, while others presented as solitary lesions.¹⁻⁸ A 2023 report described a large calcified cystic lesion yielding 1000 mL of purulent fluid with negative cultures; histopathology confirmed *Echinococcus*, an unusual presentation for primary pleural hydatidosis.⁹

In our case, the cyst was confined entirely to the pleural space with no hepatic or pulmonary involvement, which was an exceptional finding. Similar rare presentations have been reported,

mostly as isolated case reports.^{1, 3, 4, 6, 8, 10} Misdiagnosis as empyema is common due to overlapping clinical features and nonspecific imaging findings.^{2-4, 6, 8} Retrospective CT review revealed subtle, partially collapsed faint two-layered membranes that were initially overlooked, a diagnostic pitfall well described in the literature.^{2, 3, 8} Although CT is generally reliable, it may fail to identify collapsed or infected cyst.^{3, 8} Although ultrasonography may demonstrate internal architecture and daughter vesicles, it can also be inconclusive, as in our case.^{2, 3}

Intraoperative VATS exploration was pivotal for both diagnosis and management. VATS confirmed rupture with intense inflammation and secondary bacterial infection, consistent with previous reports.^{1, 8} Isolation of *P. aeruginosa* and *K. pneumoniae* complicated the clinical course, reflecting the potential for superinfection and prolonged hospitalization.¹⁰

Noncompliance with postoperative irrigation highlighted the importance of structured follow-up. Ultimately, thoracotomy and decortication were required for definitive treatment, consistent with reports emphasizing the need for open surgery in cases with dense pleural peel.^{8, 10} Pleural irrigation with povidoneiodine has been described as an adjunct in empyema management and was applied in this case.¹¹

Conclusion

Ruptured primary pleural hydatid cysts are exceptionally rare and can closely mimic empyema, frequently resulting in diagnostic delay. In endemic regions, clinicians should maintain a high index of suspicion for hydatid disease when imaging findings and clinical presentation are discordant. Both CT and ultrasound may fail to adequately depict collapsed or ruptured cysts, underscoring the pivotal role of VATS in achieving definitive diagnosis and guiding initial management. Superimposed bacterial infection further complicates the clinical course and often necessitates staged interventions. Ultimately, thoracotomy with decortication remains the definitive treatment for patients with chronic, dense pleural peel. Vigilant postoperative care and structured follow-up are essential to optimize recovery and minimize the risk of recurrence.

Authors' Contribution

H.D. contributed to the conception and design of the work; the acquisition, analysis, and interpretation of data for the work; and drafting the work; R.A.A. contributed to the conception

and design of the work. Both authors reviewed the manuscript. H.D. has read and approved the final manuscript and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Declaration of AI

Microsoft Copilot, as an AI tool, was utilized to correct grammatical errors.

Conflict of Interest: None declared.

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