Case Report

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Radiology Section

Ruptured Pulmonary Hydatid Cyst with Multiple Small Bronchopleural Fistula Formations: A Case Report

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ABSTRACT

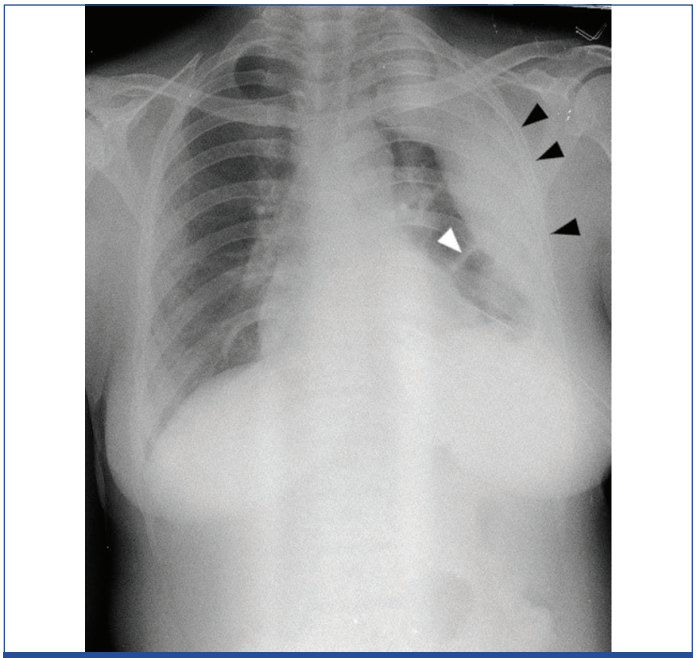
Hydatid disease is a zoonosis caused by *Echinococcus granulosus*. It manifests as cystic lesions, most commonly found in the liver and lungs, and rarely in other parts of the body. The disease occurs when a human host accidentally ingests contaminated food containing eggs from canine waste products. Since the clinical characteristics of this disease are nonspecific, radiological investigations play a crucial role in diagnosis. Diagnostic challenges arise due to atypical imaging features of complicated hydatid cysts. A 40-year-old female patient presented with fever, breathlessness, chest pain, and an unproductive cough persisting for eight days. Upon examination, she was in hypovolaemic shock, and an intercostal drainage tube had been inserted in the left sixth intercostal space. The chest radiograph revealed a dense, homogeneous opacity in the left hemithorax, partially compressing the left lung medially. An High-resolution Computed Tomography (HRCT) thorax showed a thick-walled cavity with an air-fluid level and layered membranes in the dependent part of the cavity, indicating a water lily sign in the left lower lobe of the lung, suggesting a ruptured hydatid cyst. The patient underwent cystectomy with suturing of a small bronchopleural fistula. Computed Tomography (CT) is the preferred imaging modality, especially for assessing associated complications and ruling out differentials in cases of ruptured pulmonary hydatid cysts.

Keywords: Complicated, Cystectomy, Hydatidosis, Lung

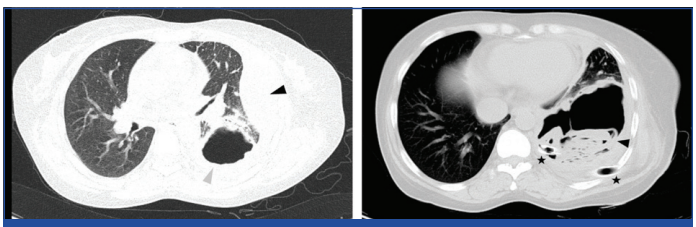
CASE REPORT

A 40-year-old female patient developed an acute onset of complaints, including fever, breathlessness, chest pain, and an unproductive cough for eight days. The patient had previously been admitted to a peripheral hospital where a chest radiograph suggested left pleural effusion. Despite repeated drainage attempts, the patient's condition did not improve and instead worsened. She was referred to us in a drowsy state with an intercostal drainage tube inserted into the left sixth intercostal space. She reported the presence of stray dogs near her home in her native village. She had no prior medical history. Upon admission, she was afebrile, with a pulse rate of 136/minute, blood pressure of 76/50 mm Hg, and a respiratory rate of 28 cycles/minute. Her oxygen saturation was 72% on room air and 86% on high-flow oxygen. She presented with hypovolaemic shock and was initiated on norepinephrine. Her complete blood profile showed a haemoglobin level of 9.1 milligrams/decilitre, a white blood count of 15,700 leucocytes per microlitre, with a mild eosinophilia at 5%, and a platelet count of 218,000 platelets/microlitre.

Upon examination, decreased air entry was noted on the left-side, while normal air entry was observed on the right-side. A clinical diagnosis of left pleural effusion was made. Once the patient was stabilised, she was referred for further diagnostic evaluation. A chest radiograph [Table/Fig-1] was performed, revealing partial replacement of the left hemithorax with a dense homogeneous opacity that compressed the left lung medially. An HRCT thorax [Table/Fig-2,3] showed a thick-walled cavity with an air-fluid level within and layered membranes in the dependent part of the cavity, indicative of a water lily sign in the left lower lobe of the lung, possibly indicating a ruptured hydatid cyst. The right lung parenchyma appeared normal. Two intercostal drainage tubes were noted, one within the cavity of the lesion and the other in the left pleural space. Additionally, a loculated pleural collection with air foci along the lateral aspect of the apico-posterior segment of the left upper lobe, likely caused by previous instrumentation, was observed. An abdominal ultrasound was performed, revealing no cystic lesions in the liver or elsewhere.

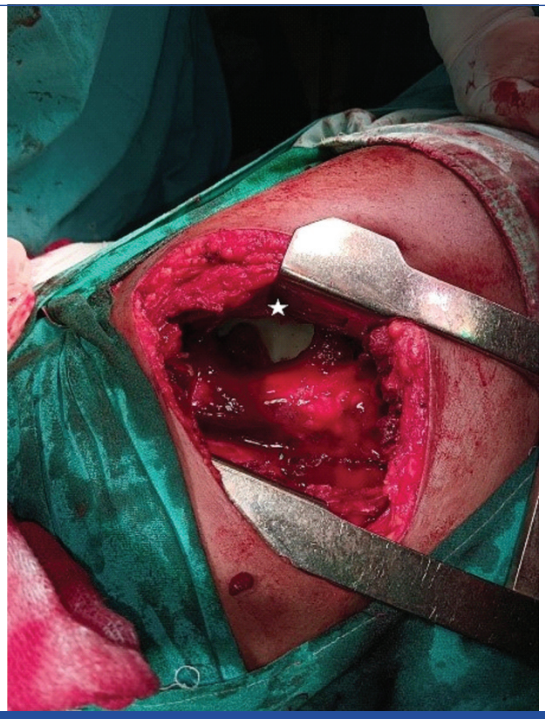


[Table/Fig-1]: Chest radiograph Postero-anterior (PA) view shows a homogenous radio-opacity in the left superolateral hemithorax (black arrowheads) and a cystic lesion in the left lower lobe (white arrowhead).

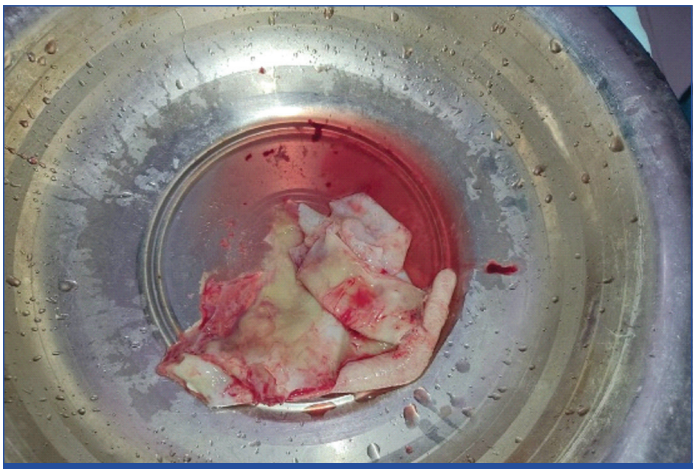


[Table/Fig-2]: Axial HRCT chest showing a thick walled cavitary lesion in left lower lobe (grey arrowhead) and loculated pleural collection (black arrowhead) compressing underlying lung parenchyma. **[Table/Fig-3]:** Axial HRCT chest showing layered membranes (black arrowhead) within the left lower lobe cavity giving waterlily appearance with minimal fluid within the cavity. Two intercostal drainage tubes are noted, one in the cavity and other in the left pleural space (star). (Images from left to right)

The patient underwent a left thoracotomy [Table/Fig-4,5] at the sixth intercostal space. A large germinal membrane was found, involving the entire left lower lobe and forming the hydatid cyst with daughter cysts inside. Cystectomy was performed, and a thorough scoliocidal wash was given to the pleural space after cystectomy. Multiple small bronchopleural fistulae were present in the remaining cavity of the left lower lobe, which were meticulously sutured. Two intercostal drainage tubes were inserted, one in the cyst cavity and the other in the pleural space. The patient remained intubated for four days after surgery. She did not experience any complications during this time and remained stable. On the fourth postoperative day, she was successfully extubated. Albendazole therapy was prescribed for three months. At the five-month follow-up, her symptoms had significantly regressed.



[Table/Fig-4]: Left sixth thoracotomy shows glistening white germinal membrane in the left lower lobe of lung (Star).



[Table/Fig-5]: Cystectomy specimen showing germinal membranes of the hydatid cyst.

DISCUSSION

Hydatidosis is a parasitic infection caused by larvae of species of the *Echinococcus genus*, commonly *E. granulosus*, and less commonly but with an aggressive clinical course, *Echinococcus multilocularis*. Cystic echinococcosis is caused by *E. granulosus*, whereas *E. multilocularis* causes alveolar echinococcosis [1,2]. Diagnosis is made based on clinical history, microbiological study, serological analysis, and radiological investigations [3]. It commonly involves the liver and less commonly affects the lungs, bones, brain, kidneys, and rarely other parts of the body. Extrapulmonary thoracic hydatidosis can develop in the pleural cavity, fissures, diaphragm, mediastinum, heart, pericardium, and vascular structures.

Pulmonary hydatid cysts often remain asymptomatic for years due to slow growth. They only become symptomatic when they rapidly enlarge or rupture [1,4]. Symptoms such as chest pain, dyspnoea, dry cough, and haemoptysis can occur due to the mass effect caused by larger cysts [5]. Acute-onset chest pain, coughing, haemoptysis, and anaphylactic reactions like urticaria, asthma, or anaphylactic shock may suggest cyst complications such as rupture or infection [5,6].

The degeneration of cyst membranes is responsible for rupture, which can be influenced by several factors, such as the age of the cyst, chemical pneumonitis, the host's immune system, or the use of Albendazole [7]. Cyst rupture can occur in the pleural cavity or bronchus. When cysts rupture into the bronchus, it can manifest as cough, haemoptysis, and fever, with the expectoration containing abundant hydatid sand and membrane fragments, known as hydatid vomica. Cyst rupture can also occur into the pleural cavity, leading to the formation of bronchopleural fistula, pneumothorax, hydropneumothorax, effusion, pleural thickening, and empyema [4,6,8,9]. Patient in the present report presented with acute complaints of fever, breathlessness, chest pain, and an unproductive cough, likely due to the pleural rupture of the cyst. A case described by Ataya J et al., involving a giant ruptured hydatid cyst, reported acute coughing with yellow watery sputum, likely due to bronchial rupture caused by erosion, as the cyst was of large size [10].

Microbiological, serological, and imaging modalities are available for diagnosis. Serological tests like the Enzyme Linked Immuno-sorbent Assay (ELISA) test and Western blot tests can help in diagnosis, as humans are intermediate hosts and do not shed helminthic eggs in their faeces [3]. The visualisation of hydatid scolexes in sputum specimens is pathognomonic [11], as reported by Karimi M et al., in their case [4]. Radiographically, chest radiographs are a preliminary diagnostic modality for patients presenting with fever, dyspnoea, or other respiratory complaints. The presence of multiple rounded homogeneous radiodense lesions in the lung fields is pathognomonic for hydatid cysts [8]. However, HRCT is indispensable in cases with complicated cysts [4]. Various radiological signs on HRCT have been described, such as the air crescent sign (meniscus), inverse crescent sign, and air bubble sign in contained/partial rupture, and the cumbo sign, whirl sign, waterlily sign, rising sun sign, and dry cyst sign in complete rupture. Some imaging features have been described in cysts that rupture into the lung parenchyma, such as centrilobular nodular opacities, tree-in-bud opacities, and consolidation around the cyst wall [2,12]. Cyst wall calcification can also be seen on CT, which is strongly suggestive of hydatid cyst in appropriate clinical settings [13]. On HRCT, the present case showed rupture of the endocyst due to increasing air between the pericyst and endocyst, resulting in its collapse into the cavity and giving rise to the cumbo sign. Floating membranes were observed in the remaining cavity fluid, resembling a floating waterlily (Camellote) sign in the left lower lobe of the lung. Multiple loculated pleural collections in the left superolateral hemithorax were also present.

Differential diagnosis for hydatid cysts include tubercular cavities, bronchogenic cysts, abscesses, and necrotic pulmonary malignancies. The radiographic air crescent sign can also be observed in cavitating malignancy, Rasmussen's aneurysm, retracting blood clots, and Aspergilloma [8]. Ruptured pulmonary hydatid cysts can closely resemble pleural effusion, empyema, or hydropneumothorax, making it challenging to differentiate them in certain cases [10]. A case reported by Puri D et al., initially misdiagnosed and treated pyopneumothorax as tuberculosis [6]. Later, the diagnosis of hydatidosis was established during exploratory thoracotomy. Therefore, clinical correlation and a

high index of suspicion for hydatid disease are crucial for diagnosis, especially in endemic regions.

Surgery is the preferred treatment for patients with ruptured pulmonary hydatid cysts. Parenchyma-preserving surgical procedures are preferred over radical surgery to maintain normal post-operative pulmonary physiology. Conservative surgical options include cystectomy or cystotomy and capitonnage of the residual cystic space for uncomplicated cysts. More radical procedures such as segmentectomy, lobectomy, decortication, and pneumonectomy are reserved for complicated cysts [4,9,14]. In present case, cystectomy was performed along with closure of bronchopleural fistulae. She did not experience any post-operative complaints and showed symptom regression during the five-month follow-up. Radical surgery should be reserved for cases with complete pulmonary lobar destruction or severe bronchopleural fistulas, as observed in the case described by Rodríguez-Laiz G et al., [15].

Medical treatment with benzimidazole compounds can be initiated preoperatively in cases of pleural rupture of the cyst or disseminated hydatid cysts to prevent postoperative recurrences. It may be continued postoperatively for three months [9,10,14,16,17]. However, primary medical treatment of hydatid cysts carries the risk of albendazole-induced cyst rupture, as indicated by studies conducted by Koul PA and Kanjwal MK and Usluer O et al., [18,19]; as well as a recent case report by Sheikhy K et al., [7].

Limitation(s)

One limitation of the presented case is the absence of serological tests (Western blot or ELISA) before surgery, and postoperative histopathology was not performed, which could have aided in the definitive diagnosis of PHC.

CONCLUSION(S)

Computed Tomography is the imaging modality of choice, particularly in complicated hydatid cysts, as it can accurately diagnose the condition by revealing the internal characteristics and morphology of the lesion. Therefore, radiologists should be well aware of both the typical and atypical imaging features of the disease.

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