Case Report

An Uncommon Pulmonary Presentation of a Common Disease

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Introduction

We report a spontaneous hydatid cyst rupture, first misdiagnosed as pneumothorax. It was initially managed as a secondary pneumothorax with an Intercostal Drainage (ICD) tube. It was definitively managed with lung hydatid cyst excision and lung decortication.

Case history

We present a case of a 24-year-old female resident of central India who presented with complaints of right-sided chest pain with minimal expectoration for the last 6 to 7 months with no complaints of shortness of breath, palpitations, fever, or loss of appetite. She was initially evaluated and treated in a private hospital as a pneumothorax.

Based on a Chest X-ray (Figure1a), An ICD tube was inserted, which remained in situ for about a month. No radiographic resolution of pneumothorax developed, and the tube was removed. After that, she presented to our Medicine Outpatient Department with complaints of persistent cough and right-sided chest heaviness.

On examination, pallor was present. There were decreased breath sounds in the right supra- clavicular, mammary and supra-scapular areas and absent breath sounds in infra-mammary,axillary, infra-axillary and infra-scapular areas. Moderate hepatomegaly (liver span-17cm) was present. Routine blood investigations revealed mild normocytic normochromic anemia with normal liver and kidney profiles.The serology for the Echinococcus was negative twice.

Her radiological investigations revealed right-sided hydropneumothorax with air-fluid level with no mediastinal shift on Chest X-ray (Figure1b). Left side lung parenchyma was normal. On Ultrasound examination of the lungs, multiple well-defined anechoic cystic lesions in the right middle and lower zone with surrounding hypoechoic matrix, with some cystic lesions showing detached membranes folded and floating in the cyst lumen, were present. There was no free fluid in the pleural cavity (Figure 2).Contrast enhanced Computed tomography (CECT) thorax and abdomen revealed a large, well-defined, thick-walled cavity in the right hemithorax with internal air-fluid level and multiple non-dependent round to oval daughter cysts with a well-circumscribed hypodense lesion of

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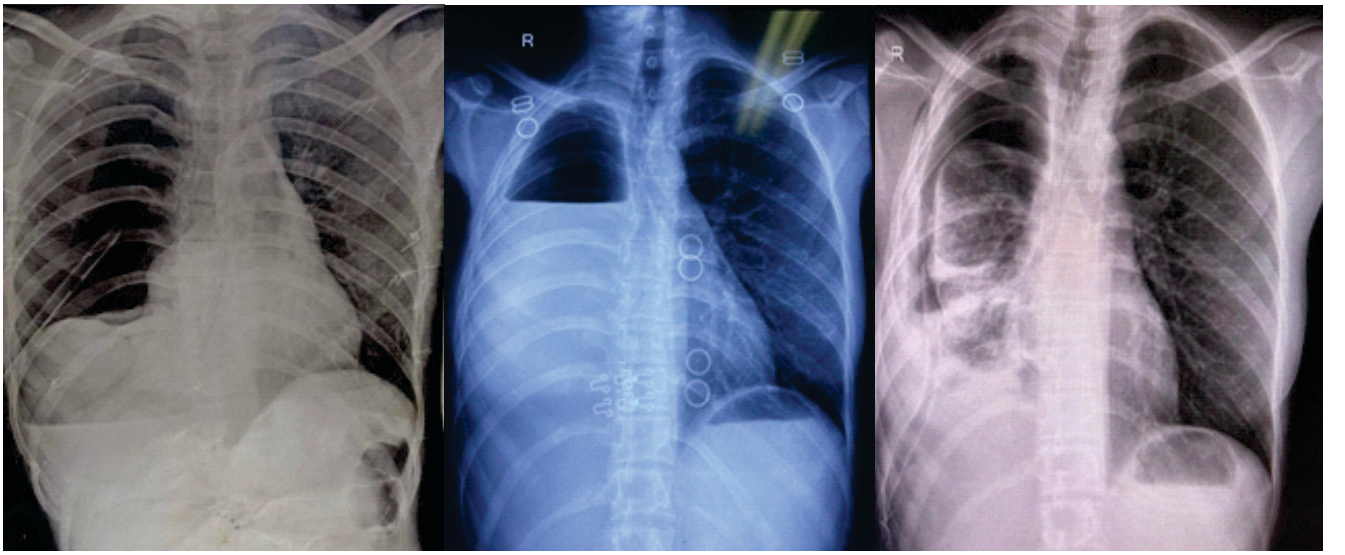

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Figure 1: a-Chest X-ray showing right sided air fluid level suggesting hydropenumothorax with intercostal drainage tube in situ.b- Chest X-ray showing a large air fluid level on the right side consistent with Hydropneumothorax.c- Post operative Chest X-ray showing partially expanded lung with drainage tube in situ

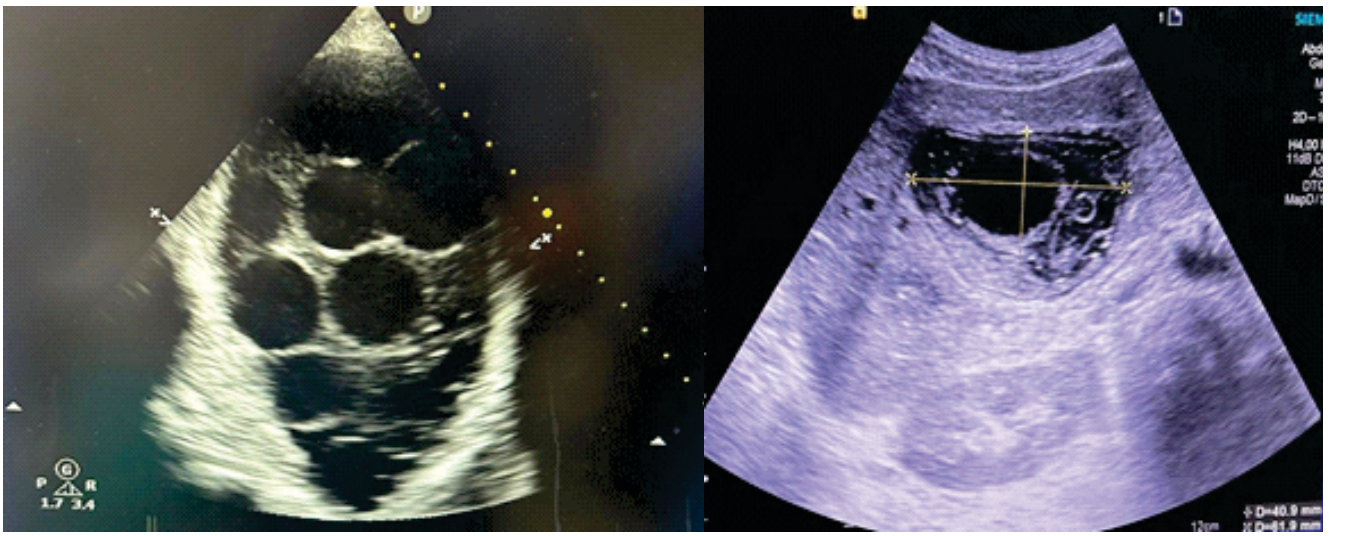

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Figure 2: a-USG Thorax showing multiple well defined anechoic cystic lesions noted in right upper and lower zone with intervening hypoechoic solid matrix. Some cystic lesions show detached membranes within folded and floating in cystic lesion. b-a large well defined cystic lesion with internal solid component measuring 11.7x7.7x6.3 cm in segment 6/7 of liver with wall thickness measuring 3.2mm showing two echogenic layers. There is hyperechoic solid component and few free floating laminated membranes in dependent portion.

8.6 cmx 6.7cm x 8.1cm in the right lobe of liver with multiple thin floating membranes (Water-lily sign) likely suggestive of hydatid cyst (WHO Class CE-3A) (Figure 3).

The pulmonary function test revealed abnormal ratio of Forced Expiratory Volume in the first second (FEV1) to Forced Vital Capacity (FVC) with reduced FVC, suggesting possible restriction. The bronchoalveolar lavage (BAL) study revealed no abnormality.

The patient was planned for thoracic surgery and operated on for right lung hydatid cyst excision and lung decortication. The specimen was sent for histopatho-

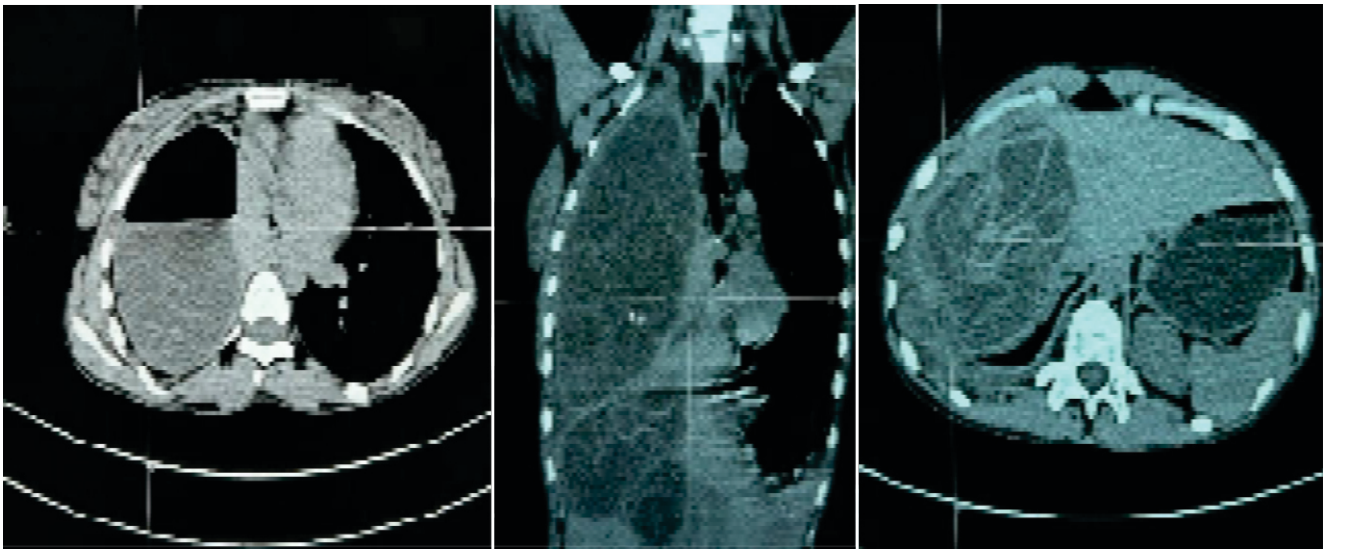

Figure 3: a-A large well defined thick walled cavity in the right hemithorax with internal air fluid level and multiple non dependent round to oval daughter cysts .b,c- A well circumscribed, thin walled round oval hypodense lesion seen in the subscapular location of right lobe of liver with multiple thin floating membranes (water lily sign)

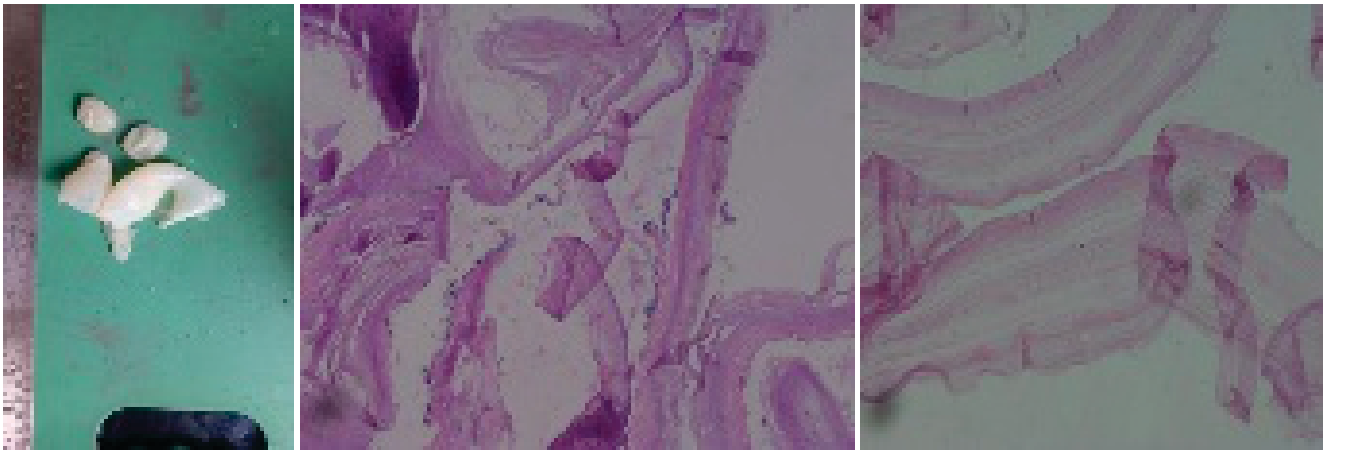

Figure 4: Gross examination-multiple white membrane like tissue pieces altogether measuring 9x8x2.2 cm.Microscopy-sections from cyst show laminated membranous structure lined by germinal epithelium. The laminated membrane is surrounded by dense inflammatory infiltrate comprising of neutrophils, lymphocytes, histiocytes and plasma cells. Granulation tissue formation is noted.

logical evaluation (Figure 4). The procedure went uneventful,and she had clinical stabilization and symptomatic relief.

Discussion

Hydatid disease is a zoonotic disease caused by the larval form of Echinococcus, most commonly Echinococcus Granulosus. Hydatid cysts can infect various organs, the liver being the most common (50-79%), followed by the lungs (10-30%)[1]. It can also involve other organs like muscles, kidneys, or heart. Pulmonary hydatid cysts are multiple in 30% of cases, bilateral in 20% and located in lower lobes, especially right in 60% of cases[2]. They are usually asymptomatic, grow slowly and are diagnosed incidentally on a chest radiograph. Structurally, hydatid cysts are made up of three layers. The outermost layer, pericyst, is formed by compressed host tissue and a fibrous reaction. The ectocyst, the middle laminated membrane layer, is an acellular structure. Finally, the endocyst, the innermost germinal layer, creates daughter protozoa[3,4]. The elastic capability of lungs and minimal resistance to expansion usually give rise to the growth of cysts,

leading to huge-sized cysts. These large cysts cause a space-occupying mass effect and have a high risk for rupture.

One unusual complication of pulmonary hydatidosis is spontaneous rupture, which can occur when the size reaches 7-10 cm in diameter or secondarily due to an infectious process, trauma to the chest, coughing or after needle aspiration. Ruptured hydatid cysts generally present withchest pain (49%), cough (46%), dyspnea (42%), hemoptysis (33%), fever (36%), and sputum production (33%),where as only 3% of patients with rupture are asymptomatic[5].

In this case, the giant cyst in the right lung (about 15 cm x 15 cm) and its rupture causing hydropneumothorax presented minimal clinical symptoms. The cyst and its contained fluid and associated lung collapse covered almost the entire hemithorax, creating an uncommon and intriguing appearance on Chest X-ray. As in this case, about 3-40% of human pulmonary hydatidosis cases are seronegative.

The treatment of ruptured pulmonary hydatid cysts is principally surgical. Albendazole is given as a complement to surgical treatment to avoid recurrence. Decortication and cyst extraction helped expand the collapsed lung, followed by an uneventful recovery in this case.

Conclusion

This case should raise awareness of an uncommon presentation of hydatid disease in medicine. Ruptured hydatid cysts can sometimes complicate as empyema, bronchopleural fistula and collapsed lung. These complications may mislead the diagnosis and treatment if prior evidence of cyst has not been documented before rupture.

Therefore, an ultrasound of the chest may be advisable before going for diagnostic and therapeutic thoracocentesis based on Chest X-ray alone. Unknowingly puncturing the cyst could have led to severe consequences like anaphylaxis.

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