Clinical Medicine Insights: Case Reports

CASE REPORT

Pulmonary Hydatid Cyst with Complicating *Aspergillus* Infection Presenting as a Refractory Lung Abscess

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Abstract

Background: Hydatid disease is rare in the United States. Rarely the hydatid cyst can become infected with mycotic organisms, such as *Aspergillus*. We describe a young male who presents with clinical features of suppurative lung abscess whose workup diagnosed hydatid cyst complicated by *Aspergillus* co-infection.

Case presentation: A 27-year-old Peruvian male was hospitalized because of fever, chills, and productive cough of three months' duration. Clinical features were consistent with a suppurative lung abscess. Significant findings included leukocytosis with eosinophilia and a chest x-ray showing a large lingular lobe thick-walled cavity with a wavy irregular fluid level. The patient ultimately underwent surgical resection of the lingular lobe. Examination of the surgical specimen revealed the cavity to be a hydatid cyst. Histologic examination of the cyst wall showed intense inflammation and several septate hyphae of *Aspergillus* species. The patient recovered fully and has remained in good health.

Conclusion: A thick-walled cavity and a wavy meniscus constitute unusual features for an ordinary pyogenic lung abscess and suggests other possibilities. Endogenous cases of hydatid disease are uncommon in the United States, with the majority of cases occurring in immigrants. There are few published case reports describing incidental findings of *Aspergillus* in a hydatid cyst. The rare occurrence of such a condition can lead to a delay in diagnosis and treatment.

Keywords: lung cavity, lung abscess, echinococcosis, hydatid cyst, *Aspergillus*, aspergilloma

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Introduction

Hydatid disease or echinococcosis is a parasitic infection caused by the larval stage of the tapeworm genus *Echinococcus*. There are four recognized species. *Echinococcus granulosus* accounts for the vast majority of cases afflicting humans presenting as cystic echinococcosis.[1] There are two biotypes of *E. granulosus*, pastoral and sylvatic.[2] The pastoral variety is distributed worldwide, and concentrated mostly in sheep and cattle areas. Though sheep and cattle serve as intermediate hosts, humans can acquire the infection by accident. *Echinococcus multilocularis* also affects humans, causing alveolar echinococcosis which carries a poorer prognosis.[1,3,4]

E. granulosus is a minute tapeworm with two hosts. Canines, especially dogs, are the definitive hosts. Warm-blooded vertebrates, such as sheep and cattle, are the intermediate hosts. Humans are an accidental intermediate host for the parasite. Normally, the adult worm is found in the small intestine of canine animals and produces millions of ova that are discharged in the fecal matter. From the feces, the ova are released into pastures where these ova are ingested by grazing sheep and cattle or adhere to the fur of these animals. The parasite does not make its intermediate or definitive hosts ill. Once ingested, the ovum releases the hexacanth embryo which migrates through the duodenal mucosa into the lymphatics and the blood stream. These embryos enter the portal circulation and eventually develop into cystic metacestodes in the liver or the lung. The parasite grows to form a cyst filled with fluid. Within the cyst, there are protoscolices that becomes the adult tape worms in canines. When a fertile cyst or its contents are ingested by canines, the life cycle is renewed.

Though widespread in many temperate and subtropical areas of the world, hydatid disease is relatively uncommon in North America.[5] Human infection is due to contact with infected dogs, food from contaminated soil or inhalation of airborne ova.[1,2] Most cases in the United States are from immigrants from endemic areas.[1]

Rarely the hydatid cyst can become infected with mycotic organisms such as *Aspergillus*.[6] There are approximately 650 species of *Aspergillus*, but the predominant cause of human infection is *Aspergillus fumigatus*.[7] There are only few previously published case reports describing incidental findings of *Aspergillus* in a hydatid cyst.[8,9]

Case Report

A 27-year-old Peruvian male was admitted to our hospital because of fever, chills, productive cough, and chest pain. The patient had been in his usual state of general health until three months earlier when he began to notice some chest discomfort which was pleuritic in nature. The pleuritic chest pain was soon followed by the development of fever and productive cough. After a couple of weeks, the patient presented to a different hospital. At that hospital he was treated for lung abscess of the lingular lobe with clindamycin and an aminogylcoside for a period of four weeks, with little radiographic or clinical improvement. The patient was eventually discharged and presented to the emergency room several weeks later with no resolution in his symptoms.

There was no history of dyspnea, hemoptysis, seizure disorder, or alcohol or drug abuse. He denied any history of sick contacts. The patient was born and raised in Peru and had worked as a farm laborer prior to his migration to the United States five years prior to presentation. The patient had a history of contact with all domestic and farm animals while living in a farming village inland from Lima in his native Peru.

Physical examination on admission to the hospital showed a young, toxic-looking male with an oral temperature of 102 degrees Fahrenheit, a pulse rate of 120 beats per minute, respiratory rate of 20 breaths per minute, oxygen saturation of 95%, and a blood pressure of 110/70 mmHg.

Abnormal physical findings were confined to the chest, which showed bronchial breath sounds, an impaired percussion note, and few rales over the left lower chest anteriorly and laterally. There was mild clubbing of the fingers. Examination of the heart and the abdomen was unremarkable.

Initial white blood cell count was 10,900, with 60% polymorphonucleocytes, 7% band formations, 17% lymphocytes, 3% monocytes, and 13% eosinophils. A comprehensive metabolic panel, remaining complete blood count, and electrocardiogram were unremarkable. The chest radiograph (Fig. 1) showed a large thick-walled cavity involving the lingular lobe, containing an irregular wavy fluid level, suggestive of floating particulate matter ("waterlily sign"). A left lateral decubitus chest film confirmed mobility of the contents of the cavity.

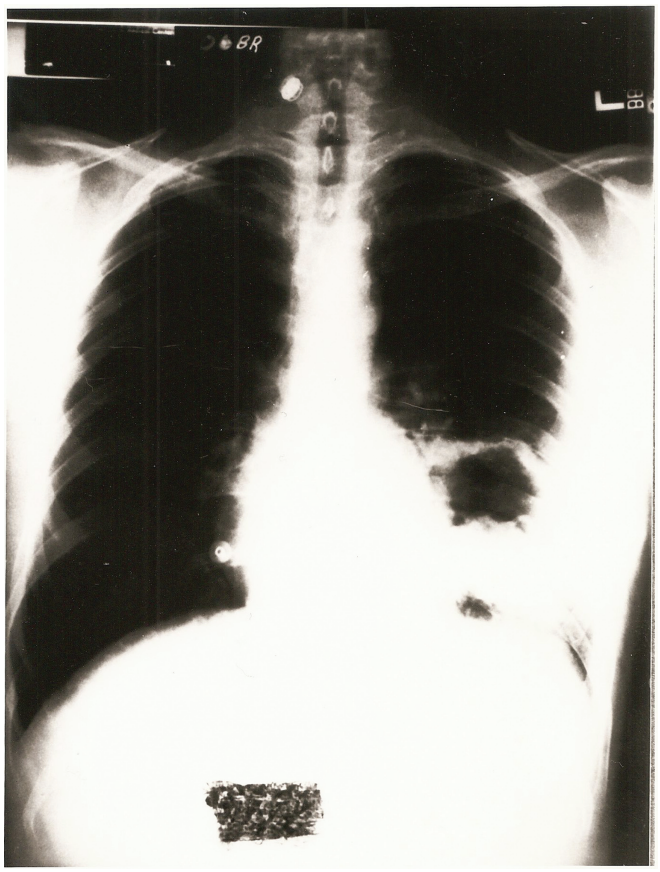

Figure 1. Chest roentgenogram (postero-anterior) view shows a thick-walled cavity with a wavy meniscus involving the lingula.

Eventual gram stain of the sputum showed polymorphonucleocytes with mixed bacterial flora but the culture was negative. Computed tomography of the chest also supported the findings noted on the chest radiograph and failed to disclose any intrahepatic or abdominal abnormalities.

A fiberoptic bronchoscopic examination showed mucosal inflammation and mucopurulent secretions in the lingular and left lower lobe bronchi. The airways were patent. A fistulous communication between the accessible airways and the cavity was not identified. Bronchial washings were negative for malignant cells and for acid-fast bacilli on smear examination. An intermediate strength purified protein derivative (tuberculosis) skin test showed a 15 mm induration.

The patient was initially treated with clindamycin 600 mg intravenously every six hours for a period of ten days without a substantive clinical response. *A. fumigatus* had been isolated from bronchial washings. This information, coupled with a positive serum precipitin test for *A. fumigatus*, supported the diagnosis of an aspergilloma in a chronic abscess cavity. A Casoni test was not done. The patient underwent thoracotomy and resection of the lingular lobe.

Gross examination of the surgical specimen (Fig. 2) revealed the cavity to be a hydatid cyst that was 7.5 cm in its largest diameter. The cyst consisted of an outer shell, the ectocyst, with a glistening white inner surface. Contained within was a pearly membranous structure, the endocyst, lying in a pool of turbid brownish fluid. The surrounding lung parenchyma was compressed and firmly adherent to the outer surface of the ectocyst. Microscopic examination of the fluid sediment (Fig. 3) disclosed several hooklets and other remnants of scolices diagnostic of cystic echinococcosis. Histologic examination of the pericyst (Fig. 4) showed inflammatory changes and infiltration by numerous septate hyphae consistent with *Aspergillus* species. *A. fumigatus* was isolated on culture from the cystic fluid. Bacterial culture of the cystic fluid was negative.

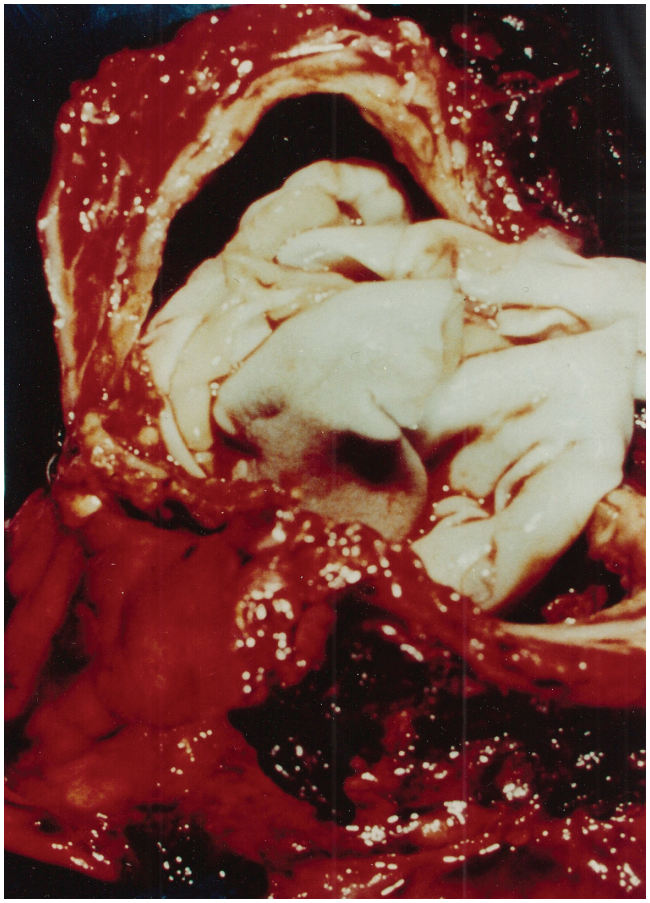

Figure 2. Surgical specimen containing dissected hydatid cyst shows a convoluted mass of glistening white endocyst within the ectocyst cavity. Wavy outline of the detached endocyst and the crescentic space above correspond to the wavy meniscus on the chest roentgenogram.

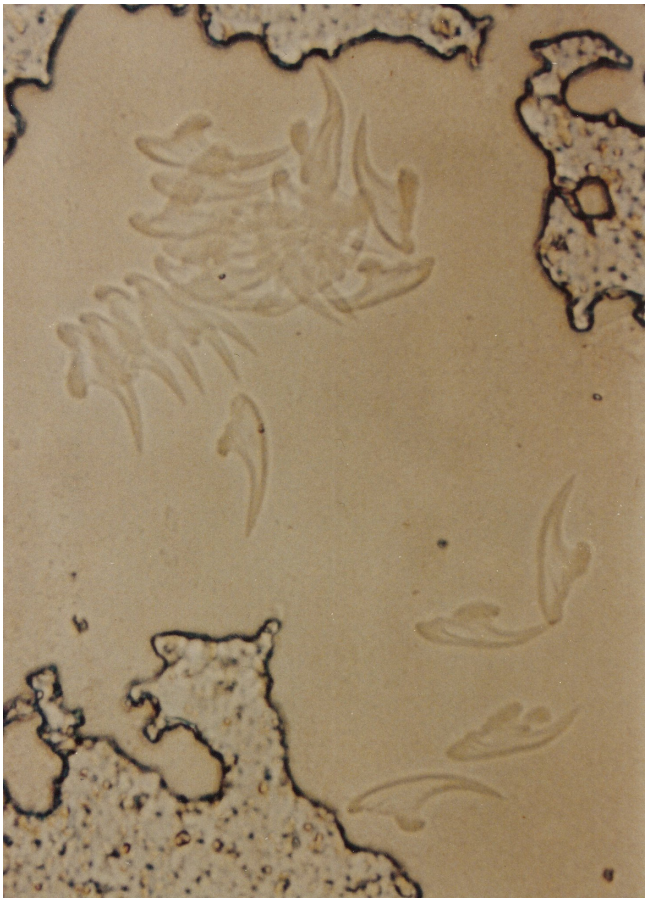

Figure 3. High power micrograph of the cyst-fluid showing several hooklets of the *Echinococcus*.

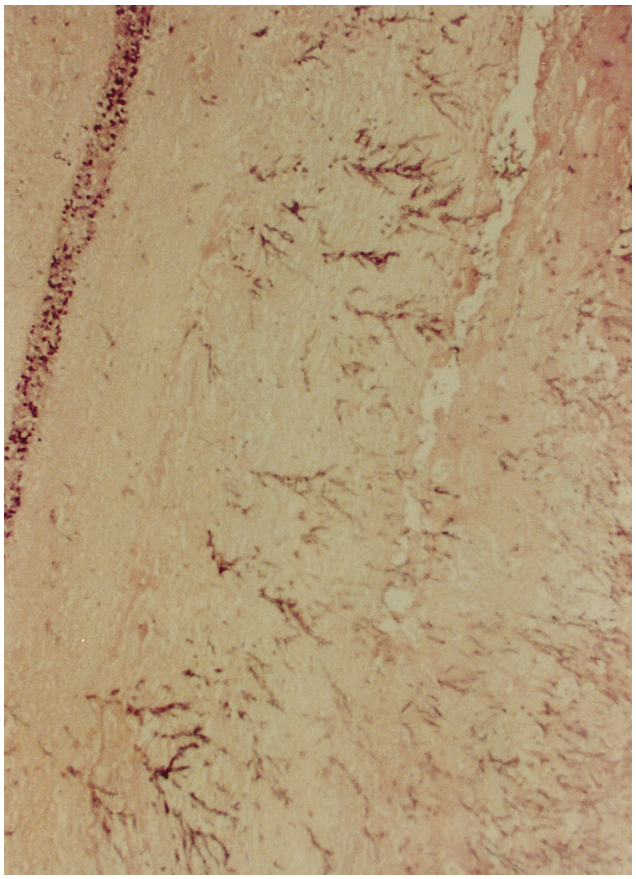

Figure 4. High power micrograph of the cyst wall (hematoxylin-eosin stain) showing infiltration with inflammatory cells and scores of branching hyphae that resemble *Aspergillus* species.

Following surgical resection of the hydatid cyst, the patient recovered uneventfully and became afebrile on the sixth postoperative day. There were no postoperative complications. The patient has remained in good health and has failed to show recurrence for three years after thoracotomy.

Discussion

A thick-walled cavity and a wavy meniscus constitute features that are unusual for an ordinary pyogenic lung abscess and suggest other possibilities. These include gangrenous lung, necrotizing neoplasm, aspergilloma in a pre-existing cavity complicated either by acute bacterial infection, intracavitary bleeding, or liquefaction, and a partially evacuated hydatid cyst.[10–12] In our patient, absence of an endobronchial tumor on bronchoscopic examination and negative bronchial washings for malignant cells eliminated the possibility of a necrotizing neoplasm. Peripheral eosinophilia noted in our patient is a feature shared by an intracavitary aspergilloma as well as a hydatid cyst. Presence of serum precipitins and a positive culture of the bronchial washings for *A. fumigatus* led us to an erroneous clinical diagnosis of an intracavitary aspergilloma complicated either by liquefaction or by an acute bacterial infection.

The surgical specimen, instead, revealed a partially evacuated hydatid cyst containing a convoluted mass of endocyst in a pool of remaining cyst fluid, responsible for the radiographic features of a thick-walled cavity with the "waterlily sign". There was no fungus ball, but the cyst wall was found to be invaded by *A. fumigatus*. Otherwise, the cyst wall and fluid was devoid of any other pathogenic organisms on culture. This established *Aspergillus* infection as the cause of the protracted febrile illness.

Aspergillus is known to colonize pre-existing pulmonary cavities in immunocompetent patients.[13] The cavity of a dead hydatid cyst could have become colonized in the same manner as a remnant tuberculous

cavity or bronchiectatic cyst. Alternately, *Aspergillus* infection of the hydatid cyst wall could have led to the demise of the hydatid. In the case of this patient, it is more likely that the remnant cyst wall was colonized after the collapse and rupture of the hydatid cyst.

The majority of patients with hydatid disease of the lungs are asymptomatic, presenting radiographically with round or oval, solitary or multiple cystic densities measuring 1–20 cm in diameter.[14,15] As the cyst enlarges, compression of the surrounding structures may give rise to symptoms.[15] When a cyst ruptures into the bronchial tree, the patient presents with a sudden onset of cough with salty bronchorrhea, chest pain, or dyspnea.[14,16,17] Aspiration of the cyst fluid can cause an allergic consolidation or even anaphylaxis.[15] Contents of a smaller cyst may be coughed up in their entirety, leaving an empty ectocyst which may remain uninfected and undergo natural closure and collapse. Secondary infection with pyogenic bacteria, however, usually follows the rupture of a larger cyst, causing hydatid abscess formation with persistent purulent sputum, fever, and other constitutional signs of an acute lung abscess.[15] *Aspergillus* infection of a hydatid cyst is extremely rare.

Radiographically, a ruptured cyst with partial evacuation of its fluid contents presents with a cavity with wavy fluid level representing the endocyst membrane floating in the cyst fluid.[12,14,15] Uncontrolled bacterial infections cause continued disintegration of the membrane leading to the formation of an air fluid level with no demonstrable floating endocyst or daughter cysts. Clinically and radiographically this may be indistinguishable from an ordinary pyogenic lung abscess.[15]

Other clinical features of pulmonary hydatid disease include hemoptysis (60%), eosinophilia (40%), a positive complement fixation test (80%), and a positive Casoni intradermal skin test (80%).[15] Discovery of scoleces on cytological examination of the sputum is diagnostic.[15–17] Surgical resection is the treatment of choice for both simple as well as complicated pulmonary hydatid cysts.[14,18,19] Chemotherapy with an oral anthelminthic, mebendazole, is useful in unresectable, recurrent, or inoperable cases of hydatid disease.[20,21]

Conclusion

Endogenous cases of hydatid disease are uncommon in the United States, and the majority of such cases occur in immigrants.[1,3–5] Only 45 (7.3%) of the 611 patients reported in the United States literature by 1966 were native to the United States.[4] Hydatid disease of the lung is usually asymptomatic unless a large cyst leads to compression of lung structures or there is rupture of the cyst. Bacteria leading to abscess formation can infect the hydatid cyst. There have only been a few case reports describing the occurrence of *Aspergillus* coexistence with hydatid disease. As in this case, *Aspergillus* is discovered after removal of the cyst and histological analysis. The lack of data, except for occasional case reports, makes describing appropriate management difficult. In immunocompetent patients, the surgical removal of the cyst should be curative for both the hydatid cyst and *Aspergillus*. In patients who are immunocompromised for any reason, a course of antifungal medication would be beneficial to prevent complications.

Disclosure

The authors report no conflicts of interest in this work. Written consent was obtained from the patient to reproduce to reproduce information and photographs appearing in this work

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