

Pulmonary Hydatid Cyst with Aspergillous Fungal Co-Infection: A Case Report and Review of Literature

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ABSTRACT

Isolated pulmonary aspergilloma as well as cystic echinococcosis is known to occur in lungs. The co-existence of aspergillosis and echinococcal cyst is extremely rare. Active invasion and proliferation of the fungi in the laminated ectocyst of echinococcal cyst is very unusual. We report a case of a 33-year-old immunocompetent patient who presented with history of intermittent fever and cough, on radiological investigation suspected as a case of hydatid cyst, and histopathological examination of cyst wall revealed the laminated membrane of hydatid cyst and infiltration of wall with septate fungal hyphae with acute angle branching suggestive of aspergillosis.

KEYWORDS: Aspergillosis, Hydatid Cyst, Immunocompetent

INTRODUCTION

The larval form of genus *Echinococcus* causes hydatid disease or echinococcosis. Saprophytic fungal infection can colonize long standing cavities in lung. Aspergillosis is a saprophytic infection which can develop in cavities formed as a result of tuberculosis, sarcoidosis, bronchiectasis, and lung abscess and cavitary neoplasia. Aspergillous can co-exist with hydatid cyst more commonly in immunocompromised patients.¹ Such a co-existence is extremely rare, and to our knowledge, no more than ten cases have been reported in the English literature. Here we describe this rare entity of aspergilloma in a pulmonary echinococcal cyst with invasion into the ectocyst layer as well in an immunocompetent female patient.

CASE REPORT

A 33-year-old female presented with history of intermittent fever and cough since 6 months, without any past history of tuberculosis. The patient was a non-alcoholic and a non-smoker. Physical and laboratory examination showed normal. Chest examination revealed decreased chest movements and breath sounds on right side. Patient was non-reactive for human immunodeficiency virus and sputum examination was negative

for acid fast bacilli and fungi. Chest x-ray showed a cystic cavity with thickened-wall. CECT scan of thorax showed a large well defined cystic cavity with regular thickened-wall and multiple floating membranes like hydatid cyst in right lower lobe (Figure No.1). Other



Figure-1: CECT scan of thorax showing a regular well defined cystic lesion of size 4x5cm with presence of multiple floating laminated membranes.

organs or systems were normal. The case was clinicoradiologically diagnosed as hydatid cyst. Enucleation of the cystic cavity was done and specimen

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was sent for histo-pathological examination. Gross examination revealed a pearly white cystic wall measuring 8x8cm with few black spots on the surface (Figure No.2). Histo-pathological examination revealed laminated membrane of hydatid cyst along with Echinococcus hooklets and its wall is infiltrated by fungal septate hyphae with acute angle branching consistent with Aspergillus and presence of hooklets (Figure No.3). The Periodic Acid Schiff stain revealed positivity of these fungal hyphae (Figure No.4). The patient was prescribed with tablet Itraconazole 200 mg once daily for three weeks followed by tablet albendazole 400 mg twice daily for two months. Follow up x-ray of chest after 3 months of surgery showed normal.



Figure-2: Gross specimen showing cut-open cystic pearly white cyst wall with few black areas on the surface

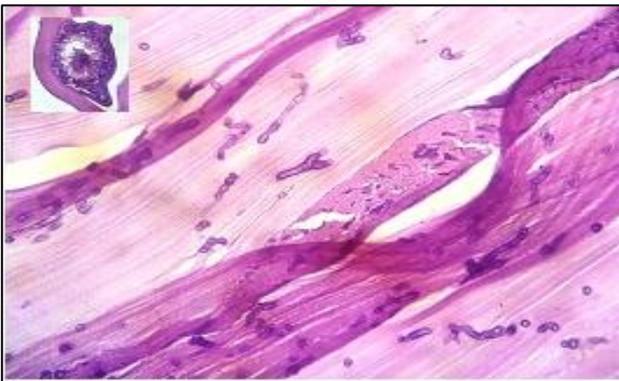


Figure-3: (H & E 400X) Microscopic examination showing acellular laminated membranes of hydatid cyst diffusely infiltrated with septate fungal hyphae with acute angle branching and presence of hooklet (inset) in left upper part

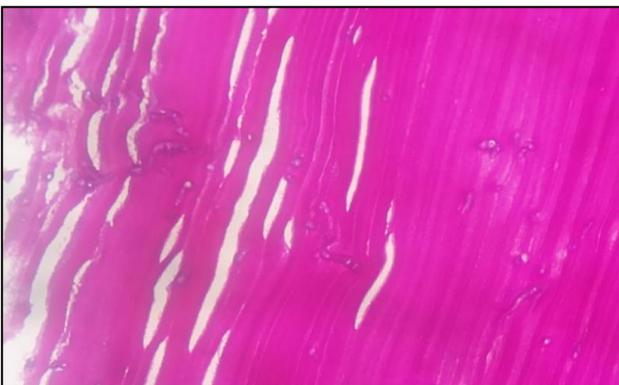


Figure-4: (H & E 400X) Periodic Acid Schiff showing laminated acellular ectocyst of hydatid cyst and collection of branching septate hyphae

DISCUSSION

Echinococcus commonly involves two organs, the lungs and liver, but can affect any organ of the body. Hydatid cyst is most commonly caused by Echinococcus granulosus, but pulmonary involvement is most commonly caused by Echinococcus multilocularis. Aspergillosis is a saprophytic fungal infection. Aspergillus fumigatus is the most common cause of aspergillosis in human beings.² Allergic pulmonary aspergillosis, aspergilloma, and semi-invasive and invasive aspergillosis may occur by Aspergillus infection.³ Pulmonary aspergillosis frequently complicates a pre-existing cavity, mostly due to tuberculosis. But, it can occur in other pre-existing cavities like sarcoidosis, neoplasia, hydatid cyst, etc. Development of aspergilloma in hydatid cyst cavity is very rare. Only few cases of the coexistence of aspergillosis and echinococcosis have been reported in the literature.³ Our case demonstrates aspergilloma involving the cavity as well as invasion of the echinococcal laminated ectocyst, by a filamentous fungus seen as hyphae branching dichotomously resembling as Aspergillus species.

Development of aspergilloma in a postoperative hydatid cyst cavity was reported after many years on one case and after 6 months in another.^{4,5} Approximately 60% of hydatid cyst occurs in right lung and out of which 50 to 60% are seen in lower lobe as in our case.⁶ Patient with immunocompromised states are prone to aspergillosis coexisting with hydatid cyst. Our patient was immunocompetent and aspergillosis was invading the wall of ruptured hydatid cyst without any underlying pathology in lungs predispose to aspergillosis.³ Hydatid cyst of larger size may be a predisposing factor for secondary co-infection.⁵ In the most cases, Escherichia Coli, Virridians group of Streptococci are seen in hepatic cysts, and Aspergillus are seen in lung cysts.¹ Hepatic cysts are coexisted in 20% of pulmonary hydatid disease.⁶

Aspergillus invading the wall of an active hydatid cyst is extremely rare and less than five cases has been reported in the literature to our knowledge. The clinical presentation of pulmonary hydatid cysts depends on site and size of the cyst and whether the cyst is intact or ruptured. Intact cysts are incidental finding and may present with cough, dyspnoea or chest pain. Cyst may rupture into a bronchus, pleural cavity or biliary tree. In ruptured cyst, the patient may present with expectoration of cystic contents, repetitive hemoptysis, productive sputum, fever, or anaphylactic shock. Our case was presented with fever, cough and hemoptysis. Whether ruptured of hydatid cyst is spontaneous or because of the invasion of aspergillosis is speculative. Wedge resection was done for recurrent hemoptysis in the present case which was recommended by others.⁷ In disseminated Echinococcosis, anti-helminthic therapy is an absolute indication.⁸ Our case was received 3 weeks of itraconazole 200 mg once daily followed by albendazole 400mg twice daily for 2 months.

CONCLUSION

The co-existence of aspergillosis should always be kept in mind in case of existing hydatid cyst for the better management. In areas where hydatid disease is still a common occurrence, pulmonary hydatid cyst should be kept in mind before labelling each cavity as post-tuberculous and aspergilloma as a secondary infection.

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