Case Report

Aspergillus colonization in hydatid cyst: Addition of a case

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ABSTRACT

Aspergillus is a common saprophytic fungus that causes invasive or non-invasive disease in humans. It commonly colonizes pre-existing lung cavities. It has been earlier reported to coexist in previously operated or ruptured hydatid cysts. However there have been only few case reports of its occurrence in previously unoperated cysts in immunocompetent hosts. The present case adds to this category.

KEY WORDS: Aspergillus, colonization, hydatid, lung

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INTRODUCTION

Hydatid disease is caused by the larval form of *Echinococcus* granulosus. It is common in all parts of India. Long standing cavities in lung can easily be colonized by saprophytic fungi. Colonization of hydatid cyst by filamentous fungi is uncommon, especially in an unruptured cyst. Deterioration of local defense mechanisms may be responsible for the co-existence.

CASE REPORT

A 52-year-old non-diabetic gentleman presented to this hospital with complaints of cough with productive sputum, anorexia and mild weight loss for the last four months. He had similar episodes of cough two years back for which he had been evaluated elsewhere. He did not have any other significant past medical history. General and systemic examination was unremarkable except for decreased breath sounds over the left lung fields. Hemogram was unremarkable. He was HBsAg and HIV negative.

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Sputum culture did not reveal any specific findings. Pulmonary function test showed a combined restrictive and obstructive ventilation defect with mild airflow limitation. Chest X-ray showed a circumscribed cystic lesion in right lower lobe of lung. Tomogram chest showed soft tissue opacity in left mid and right lower zone with thickening of fissure on left side. Plain and contrast CT revealed a well-defined peripherally enhancing thick-walled cystic lesion of size 65 × 52 mm in the apical segment of left lower lobe [Figure 1]. The lesion showed tiny air pockets within the cyst (meniscus sign). Another similar smaller lesion was seen on right side. CT abdomen showed enlarged spleen with a lobulated non-enhancing lesion of size 31 × 42 mm near splenic hilum and similar smaller lesions anterior to bladder and rectum. With these findings a diagnosis of disseminated thoraco-abdominal hydatid disease was made.

In view of the present respiratory complaints, the patient was taken up for surgery, first on the left side. The thorax was opened via postero-lateral incision and the lesion was approached via the fifth intercostal space. The cyst was identified, pericyst was incised and cyst was anucleated. The specimen was submitted for histopathological evaluation. Grossly, the specimen measured 8 × 9 × 6 cm. The external surface was shiny. It yielded 10 cc of slimy turbid fluid. The inner surface was yellowish with focal brownish black granules [Figure 2].

Histological examination revealed lamellated hyaline eosinophilic membrane of hydatid cyst. The inner wall of

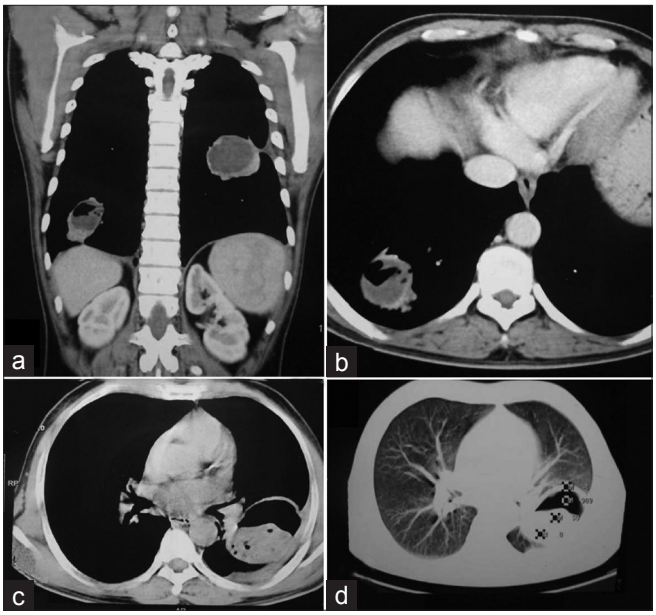

Figure 1: (a) CECT chest (coronal re-formatted) shows cysts in both lungs. Air is seen in upper most part of cyst on right side (Meniscus sign); Axial CECT shows well-circumscribed cyst in posterobasal and lateral basal segments of right lung (b) and postero-basal and lateral basal segments of left lung (c); (d) Axial CT scan-chest (lung window) showing different attenuation values of air and fluid components of mass lesion in left lung

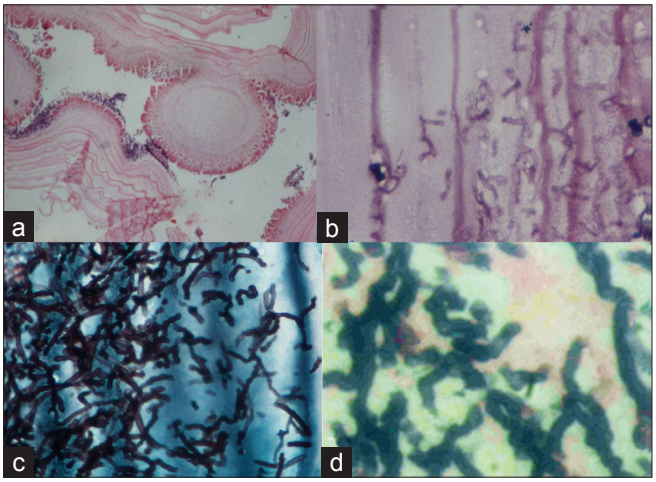

Figure 3: Hydatid membrane with Splendore–Hoeppli. Fungal hyphae are well stained by (b) PAS (c) Silver methanamine and (d) Masson's Fontana stains

the cyst showed dense lymphoplasmacytic inflammation and Splendore–Hoeppli composed of amorphous eosinophilic granular material with entrapped fungal hyphae. With Gomori's methanamine silver (GMS) stain, the fungal hyphae were slender, septate, branching at acute angles, morphologically compatible with *Aspergillus* species. The fungi were also stained with Masson's Fontana (MF) and Periodic Acid Schiff (PAS) stain [Figure 3]. The tissue was not submitted for culture.

The patient was prescribed 10 mg/kg/day of oral albendazole for 3 months.

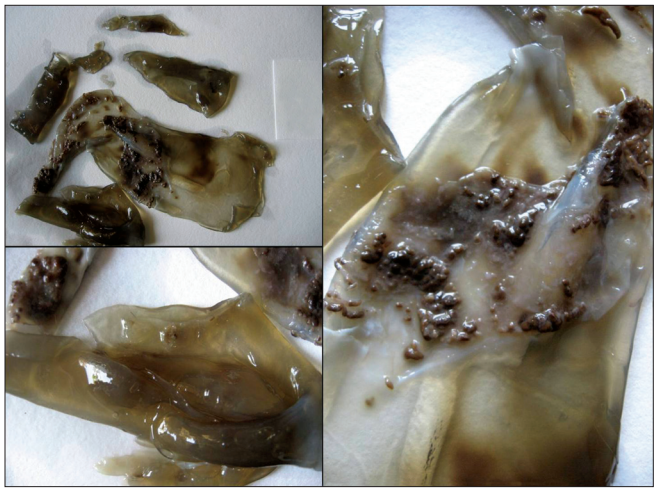

Figure 2: Gross appearance of cyst from left lung

DISCUSSION

Hydatid cyst is a zoonotic disease. The life cycle of *Echinococcus granulosus* requires an intermediate and definitive host. Though man may serve as an intermediate host, it implies a dead end for the parasite's life cycle. In man, the liver, lung and brain are the commonly involved organs; the parasite may be seen at other sites as well.

Aspergillosis in lung may be seen as non-invasive, semi and invasive forms.[1-3] It commonly colonizes diseased cavities of tuberculosis, bronchiectasis, sarcoidosis, malignancies or sometimes pulmonary infarcts.[4]

Co-existence of both is extremely rare. In a large retrospective analysis of 100 consecutive cases of hydatid cysts, colonization by *Aspergillus* sp. was seen in 2 cases.[5] Both these cases were seen in the lung in immunocompetent patients. The present case further adds to these findings. Another series showed Aspergillus colonization in two out of six cases.[6] But the spuriously high fraction may be just coincidental.

Pulmonary hydatidosis has been reported in association with cryptococcosis[7] and other saprophytic fungi.[8,9] These are generally immunocompromised patients. Fungal colonization is seen as a result of prior intervention or rupture of cysts. Larger size of hydatid cyst is a predisposing factor for secondary infection.[10] The responsible organisms as reported in a large series may be *Escherichia coli*, viridans group streptococci, in hepatic cysts and *Aspergillus fumigatus* in lung cysts.[11] Extensive colonization by Aspergillus in an unruptured cyst has been reported only once.[12] Deterioration of local defense mechanisms may result in such a complication. Cysts close to the hilum, prevent obliteration of the cavities, can cause colonization by the opportunistic *Aspergillus*.[13]

Splendore–Hoeppli phenomenon is defined by the presence of a thick proteinaceous core over an infectious

organism or inanimate particle.[14] The material comprises of proteinaceous antigen-antibody precipitate, tissue debris and fibrin formed in response to variety of fungi, parasite eggs or even suture material. Commonly associated organisms are blastomycosis, nocardiosis and actinomycosis. Aspergillus is not uncommon as was seen in the present case.[14] Strongyloidiasis, schistosomiasis and cutaneous larva migrans could also elicit the Splendore–Hoeppli phenomenon. It contributes to chronicity by evading host defense mechanisms such as phagocytosis and intracellular killing.[15] In the absence of Splendore–Hoeppli phenomenon, the presence of necrosis, granulomas with disproportionate number of giant cells and a neutrophilic infiltrate are morphological alerts to search for organisms.

There was disruption of the cyst at the time of surgical manipulation. Oral itraconazole 200 mg, once a day was given for 2 weeks. Disseminated echinococcosis is an absolute indication for anti-helminthic therapy.[16] Accordingly the patient has been on albendazole for two months now. He recovered well, was asymptomatic now for two months and is scheduled for the second surgery after one month.

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