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Case Report

Aspergilloma in a pre-existing unoperated pulmonary echinococcosis

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Introduction

Aspergillosis is one of the most common fungal infections associated with pre-existing pulmonary cavities. The most common causes for cavity formation are those secondary to tuberculosis, sarcoidosis, pulmonary infarction and bronchiectasis.¹ There are only rare reports of pulmonary aspergillosis with hydatid cyst in India.² The incidence is more common after surgical removal of these cysts, there being only two reports in unoperated cases till date to the best of our knowledge.³ We present here a case presenting with hemoptysis and unresolving cavity in right lung, which underwent lobectomy for the same and was later found to have co-existing aspergillosis and echinococcosis.

Case report

A 43-year-male patient presented with low grade fever, productive cough and right sided chest pain of 4 months duration. A history of breathlessness on exertion, streaky hemoptysis and weight loss of 3 kg over 3 months was elicited. The general and systemic examination was not contributory. His hematological and biochemical parameters were essentially within normal limit. Sputum examination was negative for acid fast bacilli (AFB) and culture for *Mycobacterium tuberculosis* showed no growth even after 6 weeks. Mantoux test showed an induration of 20 mm diameter. The chest radiograph showed an ill-defined cavity in the right middle zone closely abutting the right hilum. CT chest revealed a well-defined cavitory lesion in the right middle lobe, around 2 cm in diameter, mobile, with smooth walls, enhancing hyperdense contents and perilesional consolidation was evident, suggestive of aspergilloma. Broncho-alveolar lavage was done and Gram, Ziehl–Neelsen (ZN) and fungal stains revealed no organisms. Lavage fluid culture was negative. The patient was diagnosed as a case of ‘Smear negative Pulmonary Tuberculosis’ clinically and exhibited anti-tuberculous therapy (ATT) in February 2010 (HRZE for 6 months). The patient responded to the ATT and gained weight of 7 kg. The chest radiograph also showed features of partial resolution.

In February 2011, the patient reported back with history of hemoptysis. The constitutional symptoms were however absent. Chest X-ray showed partial resolution of the cavity and

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the surrounding consolidation. Sputum and broncho-alveolar lavage fluid were subjected to Gram, ZN and Grocott's stains, which were all negative. The aerobic and anaerobic cultures on blood agar and McConkey agar, Saboraud's dextrose agar and Lowenstein–Jensen media showed no growth. As the patient continued to have hemoptysis, right bronchial artery was embolised and the patient was taken up for bilobectomy of the right middle and lower lobes.

The gross examination of the bilobectomy specimen showed a cavity measuring around 2 cm in diameter in the right middle lobe (Fig. 1A). The wall of the cavity was collapsed. Histopathological examination revealed a cyst wall which was laminated and hyaline (ectocyst of *Echinococcus*) surrounded by thick and fibrous pericyst (Fig. 1B). The germinative layer was denuded and sloughed off. The cyst wall showed invasion by numerous septate fungal hyphae showing acute-angled branching. The mycelia were seen to form a fungal ball at places. The surrounding lung showed features of bronchopneumonia (Fig. 1C and D). A final diagnosis of pulmonary hydatid cyst with aspergilloma (operated) was given.

Discussion

Aspergilloma is the most common form of pulmonary aspergillosis, which is one of the commonest opportunistic infections in immunocompromised patients. There are few reports of aspergillosis in cavities formed by hydatid cysts

post-surgical resection till date. The cause for this rarity is the difference in the elective sites which is upper lobe for aspergilloma and lower lobe for echinococcosis.¹ In our country, where tuberculosis is so common, it forms the first differential diagnosis for cavitory lesion with perilesional consolidation. Hence, our patient, despite negative microbiological work-up, was put on ATT to which the patient responded partly with partial resolution of the lesion. But he reported back with hemoptysis and unresolving cavity, for which bilobectomy of the right lung was done. In view of the presence of radiological features suggestive of aspergillosis, positive Mantoux test and response to ATT, tuberculosis could have been a pre-disposing factor in this case leading to immune-suppression. The presence of hydatid cyst was a surprise when the bilobectomy specimen was received. The symptoms of hemoptysis, chest pain and fever could have been due to complicated hydatid cyst. *Aspergillus*, being a saprophytic fungus, was flourishing on the laminated and chitinous cyst wall of the hydatid.

Echinococcosis or hydatid disease is caused by larvae of the tapeworm *echinococcus*. Four species are recognized and the vast majority of infestations in humans are caused by *Echinococcus granulosus*. In case of clinical suspicion of pulmonary echinococcosis and absence of contraindications, a CT guided fine needle aspiration cytology of the cavitory lesion can be done to see watery fluid with scolices, hooklets and membrane. However, due to risk of anaphylaxis, this is generally not done. Protoscolices or degenerated hooklets can be shown in sputum, bronchial washings or pleural fluid specimens.

Fig. 1 – (A) Gross view – cut open lobectomy specimen showing the pulmonary hydatid cyst with surrounding consolidation of the lung parenchyma. (B) The germinative layers of the hydatid cyst (H & E stain, $\times 400$). (C) Bronchopneumonia in the surrounding lung parenchyma ($\times 200$). (D) PAS stain showing *Aspergillus* mycelia invading germinative layers of hydatid cyst wall ($\times 400$). Inset shows *Aspergillus* filaments (Grocott's stain).

Routine laboratory investigations are usually normal except for eosinophilia in few cases. Serological methods which detect antibodies and echinococcal antigens can be adopted. An enzyme-linked immunosorbent assay or indirect hemagglutination test detects around 50% cases. A negative serological test, however, does not rule out echinococcosis. Histopathological evaluation is essential for the diagnosis and for the planning of management.^{4,5} Numerous stains have been reported to be of utility in identifying hooklets of hydatid cyst – Ziehl–Neelsen and Masson trichrome stain have shown to be consistent in the staining character. Of particular interest would be the use of the universally available Ziehl–Neelsen stain as the hooklets are acid fast and stain pink.

Aspergilloma (fungus ball), a non-invasive form of aspergillosis, occurs usually singly, but bilateral involvement is seen in 5–10% of cases. It is composed of fungal hyphae, inflammatory cells, fibrin, mucus, and tissue debris. The most common species of *Aspergillus* recovered from such lesions is *Aspergillus fumigatus*. The fungus ball may move within the cavity, but does not usually invade the surrounding lung parenchyma or blood vessels. In the majority of cases, the lesion remains stable, but in 10% of cases the aspergilloma may decrease in size or resolve spontaneously without treatment.^{5,6}

Semi-invasive aspergillosis and chronic necrotizing aspergillosis occur in patients with mildly depressed immune system. In this case, the fungus develops its own cavity and then grows in it in a non-invasive form.⁵ Radiological presentation is as a mobile, intracavitary mass usually in the upper lobes. The establishment of a definitive diagnosis of aspergillosis, from either sputum or tissue culture, is often difficult because of frequent contamination by ubiquitous airborne spores of *Aspergillus*. The chances of isolating *Aspergillus* species from sputum and BAL in patients with aspergilloma are about 50% and 43% respectively.⁷ The low recovery rates are due to its intracavitary location and non-communication with the bronchus. This explains the reason for the sputum and broncho-alveolar lavage fluid being negative for fungal elements in our case.⁶

Radiologically an unruptured hydatid cyst is usually well-defined and round. More peripheral cysts conform to pressure from adjacent structures, and cysts in the interlobar fissures resemble loculated effusions. Cyst enlargement may produce pleural reaction and atelectasis, resulting in loss of definition of its contours. Calcification is rare. However, open (ruptured) cysts produce several different appearances. As the cyst enlarges, it erodes bronchioles. Air is introduced between the pericyst and laminated membrane and appears as a thin, lucent crescent in the upper part of the cyst: the crescent or meniscus sign or perivesicular pneumocyst. It is a sign of impending rupture and is an indication for urgent thoracotomy. As more air enters the cavity between the pericyst and the laminated membrane, the two separate completely and the cyst shrinks and ruptures. When the parasite itself ruptures and air enters it, the endocyst is outlined by a double

arch of air: Cumbo's sign. Tomography may show an onion-peel appearance. When completely collapsed, the crumpled endocyst floats freely in the most dependent part of the pericyst cavity, the iceberg, water-lily, or camalote sign. Daughter cysts may, rarely, be present, and the appearance of a round daughter cyst in the lowest part of the cavity resembles the rising sun.⁸

Surgery remains the treatment of choice for cystic pulmonary echinococcosis. The efficacy of anti-fungal therapy for aspergilloma is still doubtful. Recommended surgical treatment for aspergilloma includes lung resection for patients with adequate pulmonary function tests. The prognosis seems to be better if aspergilloma occurs within hydatid cyst than tuberculosis cavities.¹

Conclusion

We conclude that in areas where hydatid cyst is common, echinococcosis must be considered a possibility before labeling a cavity as post-tuberculous or aspergilloma. The co-existence of aspergillosis and hydatid cyst, though an unusual entity, must be kept in mind as delay in treatment may prove fatal.

Conflicts of interest

All authors have none to declare.

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