

Case Report

Chronic cavitary aspergilloma in an old emaciated patient with review of three cases

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ABSTRACT

Aspergilloma is aspergillus fungus overgrowth, most frequently in upper respiratory passages or lung, in a spatial configuration. Chronic cavitary or cystic parenchymal disease, causing damage to bronchial tree, predisposes to aspergilloma. It produces significant morbidity and requires early surgical intervention. The controversies still exist regarding medical and surgical management of aspergilloma. With decreasing incidence of tuberculosis, surgery is now less challenging for aspergilloma, with improved outcome, during past three decades. Presenting here is a case of giant aspergilloma involving the left upper and middle lobe, which had been progressively increasing since 15 years, in a patient with no history of tuberculosis, with two more cases, along with symptoms and signs, operative and postoperative management, as well as literature review of this rare, clinically important entity.

Keywords: Aspergilloma, Cavity, Tuberculosis

INTRODUCTION

Pulmonary aspergilloma (fungal ball or intra-cavitary mycetoma) is aspergillus infection of pre-existing cystic or cavitary lung disease. The saprophytic fungal hyphae grow on the cavity wall, peel off and form a fungal ball, mixed with blood products and cellular debris.¹ Residual tubercular cavities are most commonly involved,² though pulmonary cavity of sarcoidosis, lung cysts, bullae, cavitated bronchogenic carcinoma, pulmonary infarction and apical fibrosis of ankylosing spondylitis may also harbour aspergilloma.³ Cases of aspergilloma with no previous lung disease have also been reported. Other fungi causing aspergilloma are *Allescheria boydii*, *Fusarium* species and *Zygomycetes*. Identification of fungi is important for medical treatment, as *Allescheria boydii* does not respond to amphotericin B.⁴ Presented here is a case of aspergilloma with chronic cavitary

lesion developing in a patient with no previous lung disease.

CASE REPORT

Case 1

A 70 years female, non-diabetic, non-hypertensive, farmer by occupation presented with progressive breathlessness, off and on, since 15 years, along with productive cough, fever and occasional hemoptysis. The breathlessness was initially on exertion, but later progressed to rest. The cough was productive, with thick, mucoid discharge, not responding to routine medications and with occasional hemoptysis. The fever was mild grade, continuous type. The patient also had occasional chest pain, loss of appetite and weight and easy fatigability. She had multiple hospitalization for such episodes in last 15 years, and was diagnosed outside as

cavitary pulmonary Koch's, non-responsive to anti Koch's treatment.

On examination, patient was afebrile, thin built with pulse rate 100/min, respiratory rate 34/min and blood pressure 100/60 mmHg. She was pale, anicteric, had clubbing with no cyanosis or lymphadenopathy. Respiratory system examination showed bulge over left side of chest with tracheal shift over right. There was decreased tactile vocal fremitus, decreased vocal resonance, dull percussion note and decreased breath sounds on left side at supraclavicular, mammary, axillary and suprascapular and upper interscapular regions. There were occasional rhonchi and crepts due to bronchospasm. Rest of the systemic examination was normal.

Investigations revealed hemoglobin of 8.8 gm% and total leucocyte count of 15000 cells/mm³. Her ESR and kidney function test were normal and profile for tuberculosis (Sputum AFB, BACTEC) was negative. Chest radiograph showed a large cavitary lesion, involving left upper and middle lobe, with shift of trachea, mediastinum and heart to opposite side (Figure 1). The cavitary lesion was moving on supine and erect X rays. CT thorax revealed large, well defined, fluid density, septated lesion with air fluid level in left upper and middle lung lobe, abutting and displacing the mediastinum to right likely to be moving fungal cavitary lesion with abscess within. There was consolidation in left anterior segment of superior lobe and superior lingular segment of middle lobe of left lung (Figure 2). USG guided FNAC from left lung mass revealed aspergilloma. Pulmonary function test could not be done as the patient was dyspnoeic. In view of increasing size of lesion, recurrent bronchospasm and reduced quality of life, relatives were explained about possible benefits of surgery, to which they agreed.

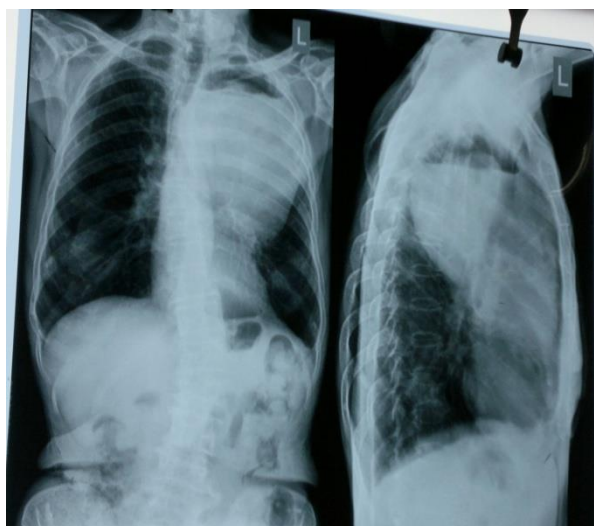


Figure 1: Showing chest X ray PA and lateral view, depicting large cavitary lesion, occupying whole of upper and middle lobe, with lung compressed inferiorly.

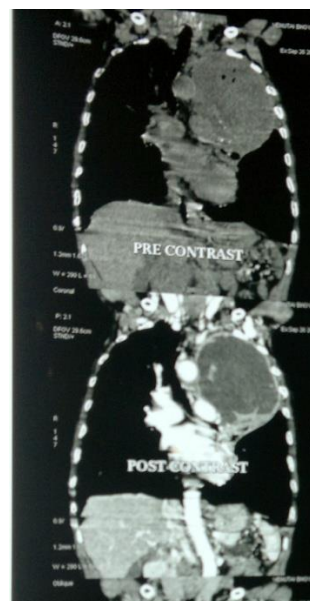


Figure 2: Showing CT scan thorax depicting large, well defined, non-enhancing, septated lesion with air fluid level in left upper and middle lung lobe, abutting and displacing the mediastinum to right.

A left posterolateral thoracotomy through 5th intercostal space for excision of the cavitary lesion was planned. On retracting the ribs, a large cavitary lesion was seen occupying whole of upper and middle lobe, with lung compressed inferiorly (Figure 3). Due to chronicity, there were dense adhesions. The posterior and superior adhesions with pleura were separated by blunt and sharp dissection. An attempt to deliver the cavitary lesion, out of the incision, was made. During this attempt, because of chronic fibrosis, around pulmonary vein and hilar bronchus, one of the pulmonary vein was torn with torrential bleeding. Immediate hemostasis was achieved. The cavitary lesion was opened and pus along with necrotic material was drained (Figure 4). The cavity was communicating with bronchus. The bronchial openings were sutured with silk 2-0 and wall of cavitary lesion was used as a membrane to seal the leakage. After confirming no air leak and complete hemostasis, an underwater seal drain was put into thoracic cavity and closure done in layers.



Figure 3: Showing intra-operative photo of large aspergilloma occupying left upper and middle lobe displacing lower lobe inferiorly.

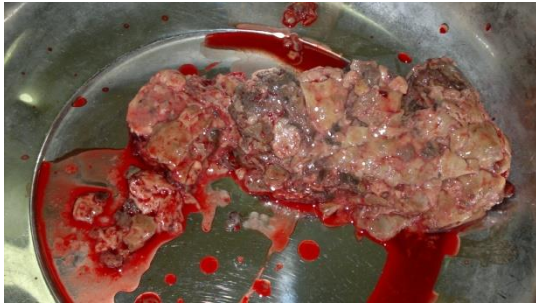


Figure 4: Showing necrotic material from within the aspergilloma cavity containing pus and cellular debris.

The patient required ventilator support postoperatively along with inotropes and blood transfusion. The patient developed subcutaneous emphysema and bronchopleural fistula, which was managed conservatively. Bronchopleural fistula sealed spontaneously by 4th day. The drain charting was 800 ml, serohemorrhagic, on first day followed by subsequent decrease and finally, the drain was removed on 7th day. The patient was started orally through Ryle's tube on 3rd day to prevent postoperative sepsis and improve nutritional status. The patient was kept on ventilator for 5 days, after which she was gradually weaned and ventilator support was removed on 7th day. Immediately, post-operative CXR showed expansion of remaining lung and subsequent X rays showed further partial expansion of lung. Intermittently, the patient went into respiratory acidosis due to bronchospasm, which was managed with positive pressure ventilation and an increasing dose of steroids. Sutures were removed on 12th day. However, there was wound gaping of 4 cm. Wound swab culture sensitivity showed growth of *Pseudomonas aeruginosa*, *E. coli* and *Citrobacter koseri* sensitive to gentamycin and amikacin, which were used topically to heal the wound. The patient was discharged on 25th day on itraconazole 20 mg BD lifelong and is under regular followup. Histopathology of the friable tissue revealed aspergilloma (Figure 5).

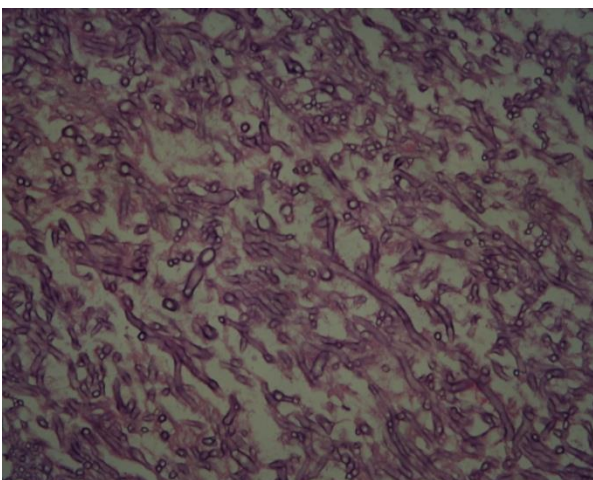


Figure 5: Photomicrograph (H & E, 40X) showing fungal hyphae.

Case 2

A 50 years female, staff nurse with previous history of tuberculosis, had presented with breathlessness, productive cough, fever and occasional hemoptysis. The mean duration of symptoms was 2 years post anti Koch's treatment. CT scan showed a cavitary lesion in right lower lobe (Figure 6). A right posterolateral thoracotomy, with lobectomy of lower lobe, containing fungal ball in post tubercular cavity was performed (Figure 7). The patient was discharged and later, after two years, developed aspergillosis, which was treated with antibiotic and anti-fungal treatment. The patient is now under regular follow up and free of complications.

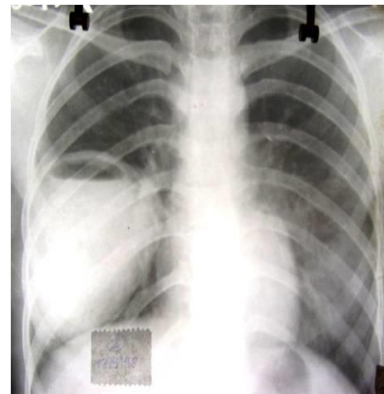


Figure 6: Showing CXR PA view, with large cavitary lesion, in right lower lobe, in a patient of tuberculosis.

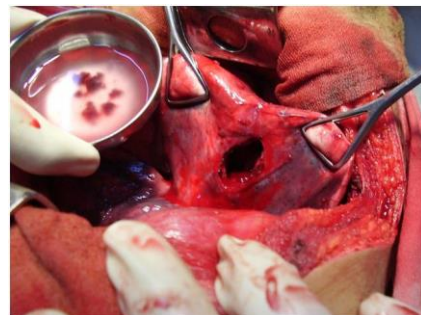


Figure 7: Intraoperative photo showing aspergilloma in right lower lobe, in which lobectomy was performed.

Case 3

A 60 years old, female, farmer by occupation, presented with breathlessness on exertion, productive cough and hemoptysis. This patient had previously taken treatment for bronchiectasis. CT scan revealed cavitary lesion in left upper and middle lobe. Left posterolateral thoracotomy was performed with left upper and middle lobectomy. There was fibrosis all around and sections from healthy lung portion have to be taken. The patient is under regular follow up with improved pulmonary function tests.

DISCUSSION

Aspergillus species is a ubiquitous, saprophytic fungus. Pulmonary involvement of *Aspergillus* species includes aspergilloma, allergic bronchopulmonary aspergillosis, chronic necrotizing pulmonary aspergillosis and invasive aspergillosis.¹ Preexisting lung cavity favours colonization of *Aspergillus* to form pulmonary aspergilloma (fungal ball). Patients with asthma develop hypersensitivity reaction with *Aspergillus* antigen known as allergic bronchopulmonary aspergillosis.² Chronic lung disease associated with diabetes mellitus, corticosteroid therapy and malnutrition favours local invasion into lung tissue to produce chronic necrotizing pulmonary aspergillosis.⁵ Invasive aspergillosis is severe and fatal disease seen in severely immunocompromised patients.⁴ Endobronchial aspergilloma, a rare entity, is growth of *Aspergillus* species into bronchial lumen.

First description of human aspergillosis was given by Virchow in 1856. Other fungi producing aspergilloma are *Zygomycetes*, *Fusarium* and *Allescheria boydii*. Identification and isolation of causative agent is important for medical treatment as *Allescheria boydii* does not respond to amphotericin B.⁵ Residual tubercular cavity is most important predisposing factor for aspergilloma, though sarcoidosis, bullae, lung cyst, bronchogenic carcinoma, bronchiectasis, pulmonary infarction and apical fibrosis of ankylosing spondylitis may also predispose.⁵ The unique finding in the present case was that no such predisposing factor was observed. Also, the fungus ball obstructing the major bronchus is a rare presentation observed in this case. Idiopathic pulmonary aspergilloma, without any predisposing factors, is rare (1-18% in different series).⁶ The mechanism is that a cavity, which is post tubercular or bronchiectatic, normally should collapse. Lung close to the chest wall is fixed and when, it is ventilated, the cavity remains open. This gives invitation to the fungus forming fungal balls. At times, these are seats of broncholiths in large dilated bronchi.

Patient with pulmonary aspergilloma may present asymptotically or with severe respiratory insufficiency.⁷ Hemoptysis and dyspnoea are most common presenting symptoms followed by fever and chest pain. Hemoptysis may be mild or severe especially with tuberculous cavity.⁶ Erosion of bronchial blood vessels lining the cavity by haemolytic exotoxin of fungus and mechanical friction by cavity wall against blood vessels are proposed mechanism.⁸ Fever indicates secondary bacterial infection.⁴

Most aspergilloma involve upper lobe owing to the affinity of tubercular bacteria, and thus cavity, for upper lobes. Middle and lower lobes are occasionally involved, especially in multiple and bilateral aspergilloma. A single cavity containing multiple fungal balls may form further cavities, areas of consolidation, empyema or emphysema.⁹

Chest X ray shows discrete, round or oval shadow, occupying upper lobe cavity, known as Monod's sign.¹⁰ Poorly defined intra-cavitary densities, intra-cavity air-fluid levels, empty cavities, pleural thickening, consolidation, empyema or, rarely, emphysema have also been described. Recent pre-existing cavity wall thickening or pleural thickening indicate early disease.⁹ CT depicts gas globules within the hyphal ball, either loose or attached to cavity granulation tissue.¹⁰ Fungal ball, partially filling cavity, are mobile within cavity and demonstrated on CT.

Unless cavity communicates with bronchus, patient do not expectorate and sputum is negative for mycelia.⁷ Serology utilizes serum anti-*Aspergillus* antibody detection for diagnosis.

Aspergilloma may either undergo spontaneous lysis¹⁰ or enlarge to cause life threatening hemoptysis, occurring in 20% cases.¹¹ Oral, intravenous and trans-cavitary instillation of antifungal drugs have limited success rate.¹² Simple aspergilloma is amenable to surgery and has low operative risk, if good pulmonary reserve is present, prior to surgery, but complex aspergilloma results in maximum post-surgical complications and even death.¹³ Preoperative pulmonary function test can indicate immediate outcome after surgery and on long term.

Surgery is indicated in all cases, by some, to avoid life threatening hemoptysis, while others prefer surgery only in symptomatic cases, due to high surgical morbidity and mortality (0-4.3%).¹⁴ Cavernostomy is performed in severely ill patients and complex aspergilloma. It is simple and effective and free of complications.¹⁵ Pulmonary resection is first choice procedure and includes lobectomies, segmentectomies, wedge resections and pneumonectomies. Surgery is technically difficult due to pleural adhesions and inflammatory changes around hilum. Best suitable treatment, to minimize complication, is thoracic plumbage (plastic balls are placed to support compliance of lung and prevent collapse of cavity) and/or thoracoplasty (in which ribs are cut and the cavitary segment is compressed). VATS have now been tried for simple aspergilloma with good success rate.¹⁴

Postoperative complications include haemorrhage, prolonged air leak due to bronchopleural fistula, empyema and subcutaneous emphysema and are commonly seen in complex cyst as was in our case.⁸ Pleural space problems occur after lobectomy, due to loss of elasticity of residual lung and fibrotic changes. Covering bronchial stump with bio-absorbable mesh or fibrin glue or packing thoracic cavity with pedicle muscle flap can prevent post-operative complications.¹⁰ Post-operative exercise for lung expansion is not undertaken, as overinflation of rest of the lung has danger of developing bronchiectasis.

CONCLUSION

The diagnosis of aspergilloma is based on radiological findings, sputum examination and percutaneous aspirations cytology. Saprophytic nature of *Aspergillus flavus* necessitates repeated isolation of organism, on culture, important. Early diagnosis prevents complications. Surgical resection of asymptomatic or symptomatic pulmonary aspergilloma is important to prevent recurrence and massive hemoptysis and has reasonable mortality and morbidity in early cases. However, surgery is technically challenging with high complication rate for complex cysts.

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