Journal of Pulmonary & Respiratory Medicine

Sathyasudish et al., J Pulm Respir Med 2014, 5:1
DOI: 10.4172/2161-105X.1000227

Case Report Open Access

Rare Presentation of Primary Pulmonary Aspergillosis

Sathyasudish N, Vishak KA*, Sydney DS, Santosh R, Anand R and Preetam AP

Department of Chest Diseases, Kasturba Medical College Hospital, Attavara, Mangalore, Karnataka, India

***Corresponding author:** Vishak Acharya K, Department of Chest Diseases, Kasturba Medical College Hospital, Attavara, Mangalore - 575 001, Karnataka, India, Tel: 9448331570; E-mail: achvish@gmail.com

Received date: Oct 20, 2014, **Accepted date:** Dec 25, 2014, **Published date:** Dec 30, 2014

Summary

A middle aged female presented with symptoms of chronic cough, breathlessness, fatigue and weight loss of 4 months duration. Chest X-ray (CXR) was suggestive of bilateral upper lobe mass lesion with a differential diagnosis in favour of malignancy. Contrast enhanced computed tomography (CECT) chest showed collapse of right upper lobe and left upper lobe. Bronchoscopy showed thick gelatinous globular plugs occluding both upper lobes and guided biopsy showed granular necrotic debris and dense eosinophilic infiltrates. Her serum immunoglobulin-E (IgE) levels were very high (18,887 IU/ml) and Gomori Methenamine Silver staining was suggestive of Aspergillus sps. She was diagnosed to have Primary pulmonary aspergillosis and treated with itraconazole for 2 months. She responded very well and lung lesions cleared drastically on follow up.

Introduction

Primary pulmonary aspergillosis usually occurs in the background of a chronic lung disease. Our patient's presentation initially mimicked malignancy on radiography which was later confirmed bronchoscopically as endoluminal gelatinous blobs secondary to primary pulmonary aspergillosis causing bilateral lung collapse. She did not have any underlying structural lung disease like fibrosis, cavitation or systemic tissue invasion.

Such presentation with very high serum IgE levels cannot be explained by semi-invasive aspergillosis or hypersensitivity reaction alone. Pulmonary aspergillus overlap syndrome is a possible explanation where in different patterns of aspergillosis either co-exist or progress from one pattern to another. Unless suspected such varied and rare presentations can be easily missed and can prove fatal to the patients.

Case Presentation

A 45 year old female presented with complaints of fever, chronic cough and breathlessness of 4 months duration. It was a dry cough with episodic bouts more in supine position and at nights. Shortness of breath worsened from grade I to grade III Modified Medical Research Council dyspnoea scale (MMRC) over 4 months [1].

She also had a history of fatigue, weight loss and loss of appetite. However, there was no history of any hemoptysis, chest pain, orthopnea or paroxysmal nocturnal dyspnoea. There was no apparent history of exposure to mould/ hay, asthmatic symptoms, allergies or lung disease in past. She has 10 years history of diabetes mellitus on treatment with oral hypoglycemic agents. Respiratory examination revealed decreased breath sounds with impaired note in both right and left upper lobe areas suggestive of bilateral upper lobe collapse. Other systemic examination was normal.

Investigations

Initial differential diagnosis of malignancy was considered as chest X-ray PA view was showing a right upper lobe mass with golden 'S' sign. Chest X-ray and Computerised tomogram (CT) scan chest did not show any of the radiological signs of aspergillosis (CT halo sign/ hypodense sign/ air crescent sign). Contrast enhanced CT (CECT) chest showed complete cut off of right upper lobe bronchus with collapse of both right and left upper lobes and enlarged right paratracheal/ subcarinal lymph nodes (Figure 1).

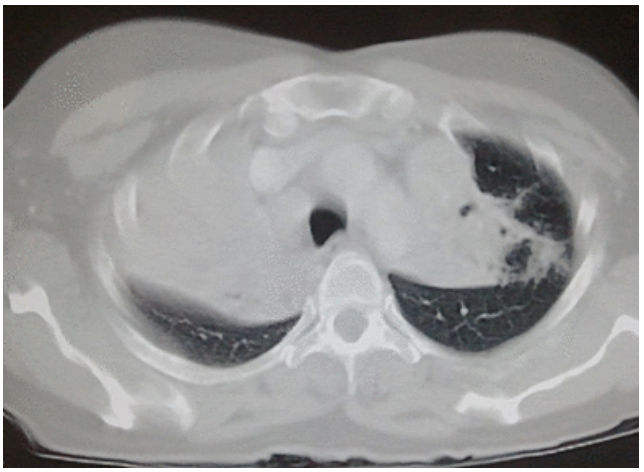

Figure 1: CECT chest showing complete cut off of right upper lobe bronchus with collapse of right upper lobe and collapse consolidation of left upper lobe with right paratracheal and subcarinal enlarged lymph nodes.

Laboratory investigations were Hb 11.9 g/dl, TC 8700 cells/mm3, ESR-20 mm/hr, Fasting blood sugars were 135 mg/dl, Sputum AFB by Ziehl neelsen stain was negative. Serum IgE level was 18,887 IU/ml (normal value 200 IU). Bronchial lavage did not show any fungal elements or acid fast bacilli.

Bronchial brushing for cytology study was negative for malignancy and was suggestive of pulmonary mycosis in view of predominant eosinophilic infiltration. Bronchoscopic biopsy showed granular necrotic debris, mucinous material and dense eosinophilic infiltrate (Figure 2).

Periodic acid Schiff (PAS) stain for fungus showed occasional fungal hyphae and Gomori Methenamine Silver (GMS) staining showed septate fungal hyphae with acute branching suggestive of Aspergillus sps (Figure 3).

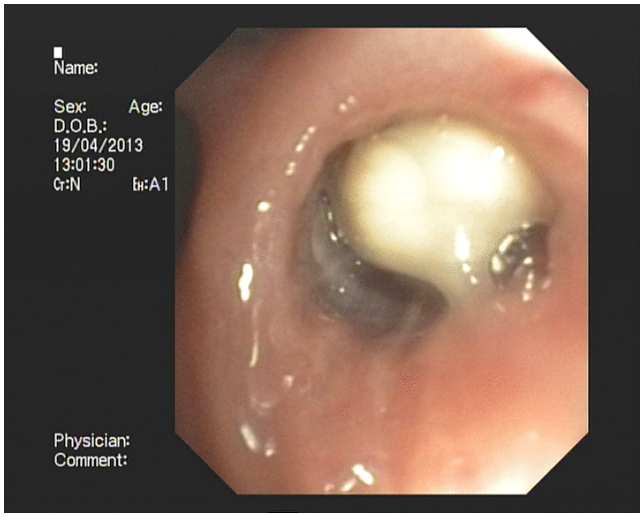

Figure 2: Primary pulmonary Aspergillosis presenting as endoluminal gelatinous blobs causing lung collapse.

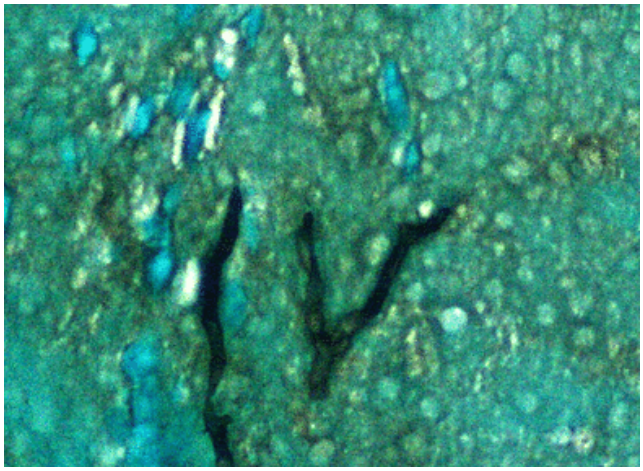

Figure 3: Gomori Methenamine Silver staining of biopsy specimen showed septate fungal hyphae with acute branching suggestive of Aspergillus sps.

Treatment

She was treated with Itraconazole for 2 months and she responded to treatment very well. Chest X-ray on follow up after one month showed drastic resolution of lung lesions (Figure 4).

Discussion

Being a tertiary care centre, our hospital is a major referral centre with high patient burden. We have seen a wide variety of presentations of Aspergillosis over the last few years. Most of the patients are either chronically ill or immunocompromised. Aspergillus fumigatus is the major isolate from such patients [2].

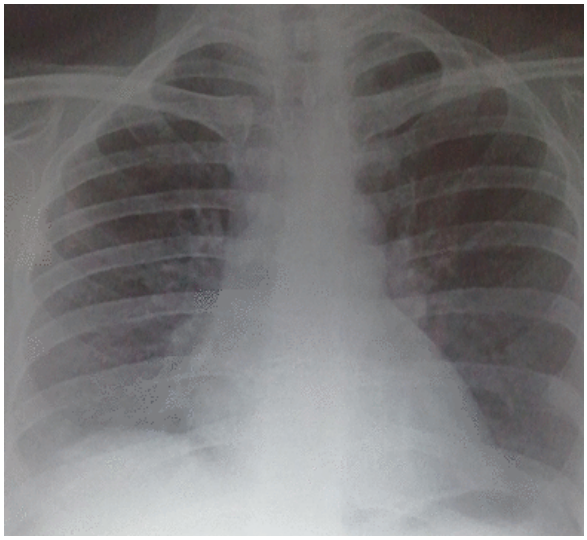

Figure 4: Chest X-ray on follow up showing drastic improvement in lung lesions.

While non-invasive aspergilloma was seen mainly in post-tubercular cavities, allergic bronchopulmonary aspergillosis (ABPA) was encountered in patients with an allergic/atopic background [3]. Our pulmonology and oncology units have encountered with HIV patients, cancer patients and COPD patients with a more debilitating presentations like invasive pulmonary aspergillosis with multi-organ involvement secondary to hematogenous spread of the fungus [3]. However, as Kousha [3] described there is an entity called "semi-invasive aspergillosis" which causes local destruction of the lung with a chronic disease course without invading into the systemic circulation [3]. David (2003) had suggested 3 distinct presentations of semi-invasive forms like "chronic necrotizing aspergillosis, chronic cavitary aspergillosis and chronic fibrosing aspergillosis" [4]. We had seen similar presentations mostly in patients with chronic lung disease and immunocompromised patients as was noticed in previous studies [3]. Although rare, there are few isolated case reports of patients without any previous lung pathology or immunocompromised state. 5-8 Risk factors reported are poor glycemic control in diabetics, old age, chronic alcohol consumption and nutritional deficiencies [3-6]. Tuberculosis and malignancy are the noted differential diagnoses in such cases of chronic pulmonary aspergillosis. Majority of our patients presented with fever, weight loss, chronic cough, and hemoptysis. Mannose-binding lectin gene polymorphism is the possible pathogenesis in most of these patients [3]. While bronchoscopic biopsy can prove local tissue invasion, culture positivity and serum IgG antibodies to aspergillus spp. can be used to confirm the diagnosis [3]. Computed tomograph scan may show either consolidation or mass like lesions mainly affecting the upper lobes [7,8].

Itraconazole or voriconazole is preferred for mild to moderate disease whereas intravenous amphotericin B or intravenous voriconazole is to be considered for severe disease [3]. Response to treatment can be monitored by improved symptoms, radiological improvement of lung lesions and reducing serum IgE levels. While some chronic cavitary forms may require lifelong suppressive therapy, some selective unresponsive patients might require surgical management. Our patient is a controlled diabetic on oral

hypoglycemic agents without any prior lung disease. Diabetes itself can be considered as mild immunocompromised state and one of the risk factors for aspergillosis as post prandial fluctuations of blood sugars are very common among diabetics. Semi-invasive aspergillosis forms do not usually present with such high serum IgE levels. High IgE levels could be secondary to hypersensitivity reaction. As described by Kousha [3] "Pulmonary aspergillus overlap syndrome" is a possible explanation for this where in different patterns of aspergillosis either co-exist or progress from one pattern to another [3].

Learning Points

Chronic pulmonary aspergillosis can present as mass causing lung collapse often mimicking malignancy especially in elderly and mildly immunocompromised patients.

Early diagnosis and treatment with anti-fungals can prevent irreversible lung damage and mortality.

Unusual presentations can delay diagnosis and complications ensued. Hence, high index of suspicion, knowledge of various risk factors and clinical presentations of aspergillosis is of paramount importance considering the high mortality if treatment is delayed.

References

1. Rabe KF, Hurd S, Anzueto A, Barnes PJ, Buist SA, et al. (2007) Global strategy for the diagnosis, management, and prevention of chronic obstructive pulmonary disease: GOLD executive summary. Am J Respir Crit Care Med 176: 532-555.
2. Franquet T, Müller NL, Giménez A, Guembe P, de La Torre J, et al. (2001) Spectrum of pulmonary aspergillosis: histologic, clinical, and radiologic findings. Radiographics 21: 825-837.
3. Kousha M, Tadi R, Soubani AO (2011) Pulmonary aspergillosis: a clinical review. Eur Respir Rev 20: 156-174.
4. Denning DW, Riniotis K, Dobrashian R, Sambatakou H (2003) Chronic cavitary and fibrosing pulmonary and pleural aspergillosis: case series, proposed nomenclature change, and review. Clin Infect Dis 37 Suppl 3: S265-280.
5. Kang EY, Kim DH, Woo OH, Choi JA, Oh YW, et al. (2002) Pulmonary aspergillosis in immunocompetent hosts without underlying lesions of the lung: radiologic and pathologic findings. AJR Am J Roentgenol 178: 1395-1399.
6. Anahita Si, Nahal Mi, Azadeh M (2010) Is Pulmonary Aspergillosis Common in Diabetes Mellitus Patients? Tanaffos 9: 69-74.
7. Yoon SH, Park CM, Goo JM, Lee HJ (2011) Pulmonary aspergillosis in immunocompetent patients without air-meniscus sign and underlying lung disease: CT findings and histopathologic features. Acta Radiol 52: 756-761.
8. Kim SY, Lee KS, Han J, Kim J, Kim TS, et al. (2000) Semiinvasive pulmonary aspergillosis: CT and pathologic findings in six patients. AJR Am J Roentgenol 174: 795-798.