

Unusual Fungal Infection in Macleod's Syndrome in an AdultDr. Jyothiratha Bangera MD^{1*}, Dr. Lavina Mirchandani, MD², Dr. Mugdha Bhide, MD³, Mrs. Sheela Naicker, M.Sc⁴¹Professor and Head, Department of Microbiology, K.J Somaiya Medical College & Research Centre, Ayurvihar, Mumbai India²Professor and Head, Department of Respiratory Medicine, K.J Somaiya Medical College & Research Centre, Ayurvihar, Mumbai India³Assistant Professor, Department of Respiratory Medicine, K.J Somaiya Medical College & Research Centre, Ayurvihar, Mumbai India⁴Tutor, Department of Microbiology, K.J Somaiya Medical College & Research Centre, Ayurvihar, Mumbai India***Corresponding author**

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**Abstract:** *Aspergillus nidulans* is one of the few species of *Aspergillus* with low pathogenicity. The significant infections of *Aspergillus nidulans* in humans have rarely been reported. It has been reported in chronic granulomatous disease which is a defect of phagocytic cells leading to recurrent bacterial and fungal infections. Here we report a case of *Aspergillus nidulans* infection which is isolated from bronchoalveolar lavage in a patient with Macleod's Syndrome. Macleod's Syndrome is a rare lung disorder having unilateral hyperlucency of lung with hypo plastic pulmonary artery.**Keywords:** *Aspergillus nidulans*, Macleod's Syndrome, Bronchoalveolar lavage.**INTRODUCTION***Aspergillus nidulans* is one of the few species of *Aspergillus* with low pathogenicity. The significant infections of *Aspergillus nidulans* in humans have rarely been reported. It has been reported in chronic granulomatous disease which is a defect of phagocytic cells leading to recurrent bacterial and fungal infections. Here we report a case of *Aspergillus nidulans* infection which is isolated from bronchoalveolar lavage in a patient with Macleod's syndrome. MacLeod's syndrome is a rare lung disorder having unilateral hyperlucency of lung with hypo plastic pulmonary artery.**CASE REPORT**

A 49 year old housewife was admitted in the Respiratory Medicine Department of tertiary care Hospital with complaints of 2 episodes of hemoptysis around 15 to 20 cc per episode since one week. There was no complaint of breathlessness, hematemesis, and hematuria or chest pain.

She was recently diagnosed with systemic hypertension. No other significant family or past medical history. No history of any addiction.

On examination patient was conscious, oriented, Pulse -80/min, BP - 140/80 mmHg, Respiratory rate -16/min, SPO2 off O2- 98%, No Clubbing or oedema feet, breath sounds decreased in left infra scapular area, no other adventitious sounds. Chest X-Ray (Fig 1) showed hyperlucency on left side in mid & lower zone. High Resolution CT Scan (Fig 2) showed hyperlucent left lung with cystic and tubular bronchiectatic changes in anteromedial

segment of left lower lobe and the periphery of lingular segments and mucoid impaction in a few subsegmental bronchi. CT Pulmonary angiography confirmed hypoplastic left pulmonary artery (PA diameter 13mm), hence a diagnosis of Macleod's syndrome was made.

Sputum was negative for acid fast bacilli and fungus. Bronchoalveolar lavage was collected by bronchoscopy and cultured in Sabourauds dextrose agar with and without chloramphenicol and was incubated at 25–30 °C. After five days of incubation, the colonies were powdery with dark green color surrounded by white brim. (Fig.3) The slide culture techniques were put up for detailed morphological study. LPCB mount of slide culture showed short smooth brown colored conidiophores, biserial phialides, short columnar headed conidia (Fig 4) and cleistothecium (Fig.5).

Bronchoalveolar lavage also sent for aerobic culture, genexpert, MGIT for tuberculosis and found to be negative.

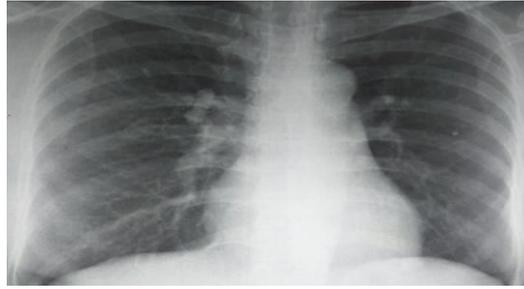


Fig-1: Xray Chest PA View: Left lung hyperlucency with small left hilar shadow.

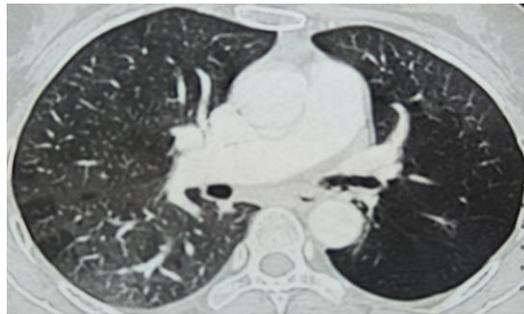


Fig-2: CT scan chest: Hyperlucent left lung with hypoplastic left pulmonary artery



Fig-3A: nidulans growth on SDA



Fig-4: LPCB mount A.nidulans

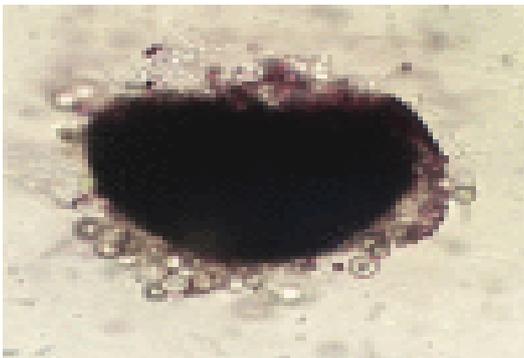


Fig-5: LPCB mount of Cleistothecium

DISCUSSION

Macleod's syndrome also known as unilateral hyperlucent lung syndrome which was first described in 1953 by Swyer and James and in 1954 by Macleod is a rare entity occurring in 3.8% of patients with bronchiolitis obliterans [1]. It is characterized by hyperlucency of one lung, lobe or part of a lobe and is associated with childhood infections most commonly post-infectious bronchiolitis obliterans and pneumonitis due to various viruses, bacteria and fungi. In most cases, both the small bronchi and bronchioles are affected, and the pulmonary damage that occurs during childhood prevents normal development of the alveolar ducts. Airways develop submucosal fibrosis leading to luminal irregularity and occlusion. Pulmonary vasculature is hypoplastic while the lungs distal to diseased bronchioles become hyperinflated with pan-acinar emphysematous changes in some cases [2] Most patients have a unilateral involvement primarily of the left lung, as seen in this patient. Presentation usually occurs in adulthood with an asymptomatic history in many cases or with a history of childhood infections.

Diagnosis is made radiographically by chest x-ray, CT scan and is an incidental finding in some cases. The characteristic radiographic findings include a unilateral hyperlucent lung along with decreased broncho-vascular markings, a small hilar shadow and slight displacement of the mediastinum to the affected side [3], as is also seen in this case (fig 1). CT scan is more sensitive than radiography in detecting hyperlucent lung regions and assessing the extent and distribution of the disease [4] revealing areas of low attenuation signifying air trapping, along with bronchiectasis, atelectasis and scarring, as was seen in this patient also. Bronchiectasis on CT scan is evident in only 30% of patients but those with saccular bronchiectasis usually have more severe exacerbations than those without [5]. Pulmonary angiography is not an essential criterion for the diagnosis of the entity although it can reveal hypoplasia and diminished size of the affected pulmonary artery. Pulmonary angiography however has its limitations, as it cannot discern congenital from acquired etiologies of hypoplastic pulmonary vasculature.

Invasive fungal infections are a major threat for patients with chronic lung disease. Common *Aspergillus* species causing respiratory infections are *Aspergillus fumigatus*, followed by *Aspergillus flavus* and *Aspergillus terreus*. Rarely *Aspergillus niger* has been reported as a cause for pneumonia [6-8]. Infections by *Aspergillus nidulans*, a fungus of extremely low pathogenicity, are relatively rare even in immunocompromised hosts. It has been reported almost exclusively in patients with chronic granulomatous disease and in one case of Pseudomembranous necrotizing bronchial aspergillosis⁹. Our patient was not immunocompromised and never had repeated

respiratory infections, but had a rare chronic lung disease known as Macleod's Syndrome.

Isolation of *Aspergillus nidulans* carries more severe implications than that of *Aspergillus fumigatus* as it is generally refractory to intensive antifungal therapy like amphotericin B, suggesting early surgery may be indicated in such cases [10]. However our patient responded clinically to itraconazole 200 mg twice a day given for four weeks along with supportive therapy with chest physiotherapy, low-dose inhaled corticosteroids, and inhaled bronchodilators.

In patients with underlying diseased lung, unusual presentations, lung infections not responding to antibiotics, there must be a high index of suspicion for *Aspergillus* fungal infection even if the patient is otherwise immunocompetent; as this ubiquitous fungus needs to be identified early and treated aggressively; as otherwise progressive lung damage can occur.

CONCLUSION

In patients with underlying lung infections not responding to antibiotics, there must be a high index of suspicion for *Aspergillus* fungal infection even if the patient is otherwise immunocompetent. Since it is ubiquitous fungus needs to be identified early and treated aggressively. So progressive lung damage can be prevented if treated at the earliest.

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