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Unusual Pneumothorax Complicating Pulmonary Aspergilloma in an Immunocompetent Patient

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Abstract **Case Report**

Spontaneous pneumothorax is a common disease. However, its association with pulmonary infection due to Aspergillus fumigatus is very rare [1]. Few studies have reported an association to suggest that Aspergillus may have been responsible for pneumothorax. In this article, we report the case of a man 30 years with a history of genetic myopathy associated with pulmonary aspergilloma and a locally septal pneumothorax. Therapeutically, the decision was difficult to manage between drainage, surgery, embolization and antifungal treatment. The prevention of aspergillosis risk in patients with sequellar lung lesion seems necessary to discuss.

Keywords: Pneumothorax, aspergilloma, embolization, antifungal treatment.

INTRODUCTION

Spontaneous pneumothorax is a common disease. However, its association with pulmonary infection due to Aspergillus fumigatus is very rare [1].

Aspergilloma is the localized form of infection resulting from colonization of a pre-existing lung cavity, usually by Aspergillus fumigatus [1]. It is a rare condition, with a prevalence of only 18/100,000 worldwide, mainly affecting patients with an underlying immunocompromised state. Its primary and main symptom is hemoptysis, and very few cases have been associated with pneumothorax [1, 2]. Few studies have reported an association to suggest that Aspergillus may have been responsible for pneumothorax.

In this article, we report the case of a man with a history of genetic myopathy associated with pulmonary aspergilloma and a locally septate pneumothorax**.**

CASE PRESENTATION

The patient was a 30-year-old male, nonsmoker and never treated for pulmonary tuberculosis, he was followed for a familial genetic myopathy since the age of 10 years, with two deaths in the siblings. The patient presented for 18 months before his admission episodes of hemoptysis of low abundance, fractionated and intermittent, with recent aggravation 1 day before his admission becoming of great abundance, he also mentioned that he felt a thoracic pain and oppression evolving in a context of altered general state.

On admission, vital signs were respiratory rate at 20 cycles per minute, heart rate of 89 beats per minute, blood pressure of 160/60 mmHg with an oxygen saturation of 97% on room air. On inspection, the patient had a chest deformity, pulmonary auscultation revealed a decrease in vesicular murmurs in the right pulmonary hemi field. The left lung breath sounds were clear on auscultation, with no crackles or wheezes. The patient did not stand up with functional impotence of both lower limbs due to his myopathy.

The chest X-ray on admission showed a right axillary pleural thickening with excavated axillary opacity and bilateral basal bronchial syndrome (Figure 1), on the control X-ray two days later, we observed an axillary nodular opacity surmounted by a gaseous crescent, with the appearance of a right clearness without a vascular framework evoking a low-abundance spontaneous pneumothorax with axillary detachment (Figure 2).

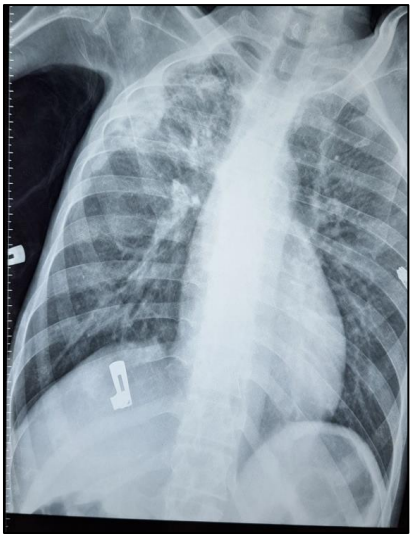

Figure 1: Axillary opacity with pleural thickening

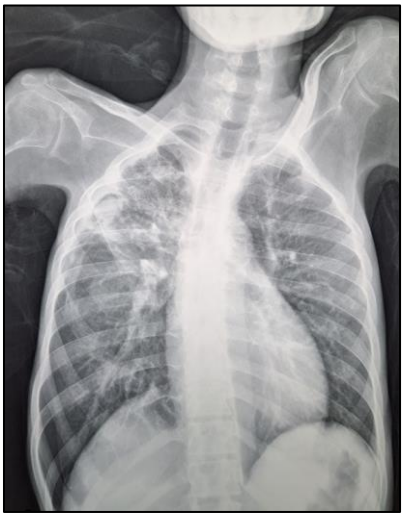

Figure 2: Axillary image as a bell with right pneumothorax

The chest CT angiography did not show any pulmonary embolism and revealed a cavity image of the posterior segment of the right upper lobe with tissue content probably related to an aspergilloma associated with diffuse ground glass areas related to alveolar hemorrhage, right pneumothorax of medium abundance partitioned in places (Figure 3), which was not apparent on a thoracic CT scan performed 18 months earlier (Figure 4).

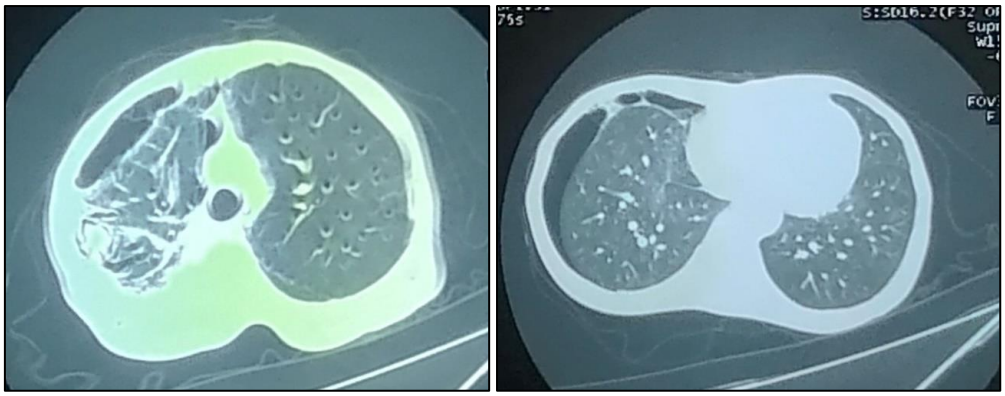

Figure 3: Chest CT scan transverse view showing a right pneumothorax of moderate size with localized partitioning associated with image as a bell

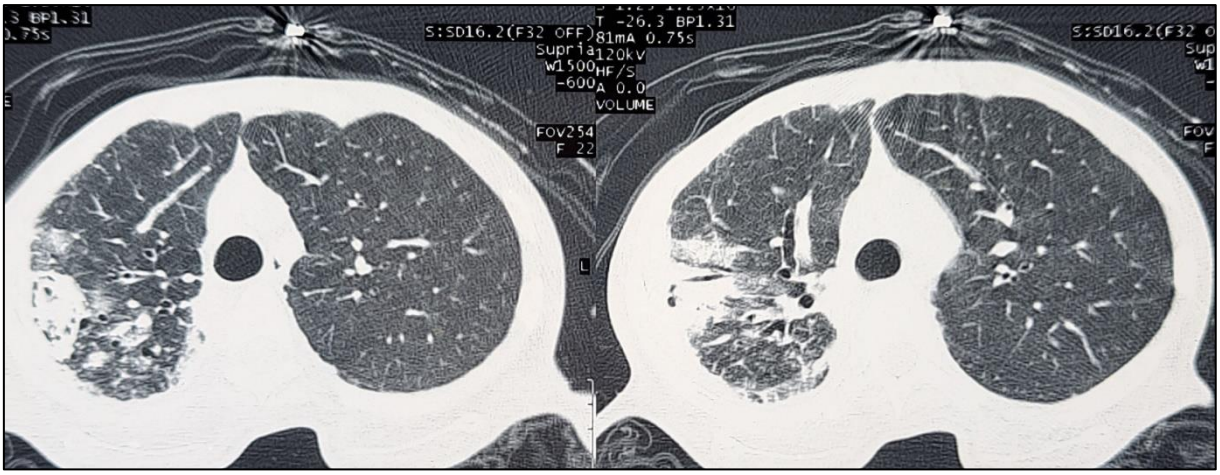

Figure 4: Chest CT scan transverse view showing a right pneumothorax of moderate size with localized partitioning associated with image as a bell

The laboratory tests showed that the research of BK and geneXpert in the sputum were negative, the mycological examination did not objectify mycelial filaments nor yeast, the aspergillary serology was positive, the prothrombine rate was at 75%, platelets count at 214000 elements/ul and the hemoglobin at 13 g/dl, he had 1% of eosinophil. The bronchoscopy performed on the patient was unremarkable, the genXpert in the bronchial aspiration fluid was negative, the bronchoalveolar lavage was inflammatory neutrophilic, the Gold score was equal to 40, with absence of malignant cells.

Concerning the assessment of the impact; a previous spirometry had objectified an attack of the small airways with FEV1 at 1,12 L /min that is to say 27%, the forced vital capacity was at 25%, probably in connection with a restrictive disorder due to his diffuse myopathy, we were not able to complete by a plethysmography seen the risk of aggravating the pneumothorax and the hemoptysis. The cardiac echography had objectified a hypokinetic in inferior with systolic ejection fraction at 42%, also in accordance with his myopathy, the 24H proteinuria was negative.

Therapeutically, the patient was put under double hemostatic treatment, strict rest and oxygen therapy, the pneumothorax was not drainable, it was localized, compartmentalized in places and without hemodynamic repercussions, an embolization was indicated but technically it was not feasible, A right upper lobar lobectomy was indicated but unfortunately the patient was not operable (FEMS at 27%, CVF at 25%, FES at 42). Failing that, the patient received as antifungal treatment: Voriconazol 200 mg tablet, twice a day. After one month of treatment, during monitoring, the patient was respiratory stable and presented an elevation of liver enzymes, with persistence of a right basal detachment on the thoracic radiography. The course of action was to stop the Voriconazol and to plan a follow-up assessment to decide on the balance of benefits and risks.

DISCUSSION

As reported in the literature, hemoptysis is the major complication of pulmonary aspergilloma, and pneumothorax is rare [1] especially in patients with tuberculosis where healing is associated with fibrosis and pleurodesis. Aspergilloma complex is usually adherent to the lung apex with a thick wall. This is very unusual. Few cases of spontaneous pneumothorax have been reported in patients with pulmonary aspergilloma or chronic pulmonary aspergillosis, Some authors have reported cases of pneumothorax associated with pleural and pulmonary aspergillosis but not with aspergilloma [2, 3].

Any causal relationship between pneumothorax and aspergillus infection was unclear. Sakuraba *et al*., [3] reported 11 cases of pneumothorax associated with Aspergillus infection without a direct causal link. We believe that the pneumothorax in our case was caused by rupture of a subpleural bulla.

The diagnosis of pulmonary aspergilloma in an immunocompetent patient can be difficult most of the time. Aspergillomas can be visualized on chest CT in a lung or pleural cavity or [4]. Radiography may show a rounded mass that usually moves into the cavity when the patient is repositioned, called Monod's sign, and provides a classic "crescent air sign" [5].

The mortality associated with ruptured aspergilloma complicated by pneumothorax has been very high as reported by Gupta *et al*., in a similar case where the immunocompetent patient died 5 days after admission [1]. Martino *et al*., reported 6 cases of pneumothorax in 46 immunocompetent patients after bone marrow transplantation, of which four died [2].

The decision to observe or treat with immediate intervention is guided by risk stratification that takes into account the patient's presentation and the probability of spontaneous resolution and recurrence. Although surgery for aspergilloma is very difficult, it is the treatment of choice. The European Respiratory Society recommends surgical excision of simple aspergilloma if technically feasible [6].

Non-surgical options exist for those unable to undergo surgery. Amphotericin B has a cure rate of approximately 10% but has a significant side effect profile. Itraconazole is the most tested antifungal agent, with two clinical trials showing cure rates above 60% with few side effects, in our country it is not marketed so it was contraindicated in our patient due to heart failure. Resistance to itraconazole makes voriconazole preferable. Voriconazole has been associated with liver toxicities in clinical trials, patients receiving voriconazole should be closely monitored. In addition, the effectiveness of the treatment focuses on both radiographic and clinical responses regarding follow-up.

CONCLUSION

Aspergilloma is rarely associated with pneumothorax. We reported one of the rare cases complicated by an unusual pneumothorax that was difficult to manage, the prevention of aspergillosis risk in patients with sequellar lung lesion seems necessary to discuss.

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