A Fatal Case of Immunocompetent Aspergilloma: Bronchial Artery Embolization Fail to Halt Recurrent Hemoptysis

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Abstract: Aspergillus is an organism responsible for aspergilloma. Massive hemoptysis, which is a life-threatening symptom in a patient with pulmonary aspergilloma, is difficult to treat. Options for definitive treatment include bronchial artery embolization and surgical removal by thoracotomy. We report a 40-year-old housewife with no past medical illness, who presented with recurrent hemoptysis caused by multiple bilateral pulmonary aspergilloma who underwent repeated embolization in combination with systemic antifungal therapy. Prompt diagnosis and bronchial artery embolization was successful in embolizing the feeding vessel; however, it failed to reduce morbidity and mortality caused by recurrent pulmonary hemorrhage.

Keywords: Aspergilloma, bronchial artery embolization, hemoptysis, massive hemoptysis, HIV.

1. INTRODUCTION

Aspergilloma is a term describing a fungus ball within a pre-existing cavity that develops in the lung. It is mainly associated with immunocompromised patients. Patients with a history of chronic lung disease such as tuberculosis, lung carcinoma and sarcoidosis and immunocompromised state, are at a high risk for aspergilloma [1, 2]. Patients are usually asymptomatic and do not require specific treatment. However, some may present with massive recurrent hemoptysis which requires intubation for airway protection [2]. Lung resection is the main option for definitive treatment. Antifungal therapy has been tried with limited success, except for cases with evidence of local invasion with systemic manifestation. Embolization is an option for those who are not fit for surgery. However, the success rate for treatment is not assured because collateral vessels will regenerate soon after embolization [2].

2. CASE REPORT

A 40-year-old housewife with no past medical illness presented with hemoptysis. She denied any high-risk behavior and had a history of contact with pulmonary tuberculosis. On examination, she was of average size and build for a woman. Further examination found nothing remarkable. Her blood culture did not grow any organisms. Screenings for Human Immunodeficiency Virus (HIV), Hepatitis B, C and tuberculosis were all negative. Her chest radiograph showed soft tissue opacity on the right upper zone associated with air crescent sign (Fig. 1). Initial CT-thorax showed bilateral upper zone consolidation and soft tissue opacity on the right upper lobe with air crescent sign in keeping with aspergilloma (Fig. 2). She was extubated after 3 days but required re-intubation within the next 24 hours to secure her airways following the second episode of massive hemoptysis. Bronchoscopy findings revealed the bleeding mainly originated from the left lung (Fig. 3). The main feeding vessels of the right and left lung lesion were embolized by an interventional radiologist (Fig. 4). Second embolization was performed because of subsequent episodes of hemoptysis. The patient showed no clinical improvement.
Subsequent repeat CT thorax showed features suggestive of local invasion to the surrounding structure. Galactomannan assay and Aspergillus PCR performed on the patient’s broncho-alveolar lavage (BAL) were positive. She was started on intravenous voriconazole. Unfortunately, she succumbed to the illness due to hypercapnic and hypoxaemic respiratory failure as a result of recurrent hemoptysis and ventilator-associated pneumonia.

3. DISCUSSION

Aspergillosis is an infection commonly encountered in immunocompromised patients. The most common species causing aspergillosis is *Aspergillus fumigatus*. Aspergillosis can be classified into three major groups: invasive aspergillosis (IA); chronic forms of aspergillosis; and allergic forms of aspergillosis [3]. The most common clinical presentation is hemoptysis.

The treatment for massive hemoptysis in aspergilloma patients remains controversial and difficult. There is a lack of proper guidelines for treatment of this condition. The complexity and variability of the disease process make it difficult to produce comprehensive guidelines. Generally, treatment can be divided into medical, radiological embolization, and surgical approaches. In our case, during her first presentation, the physician faced a dilemma in how to proceed with a surgical approach because of the variability of the survival rate postoperatively, and a lack of experience at our centre.

Given the limited surgical experience, the complexity of the disease, and the presence of comorbidities and poor surgical outcome, not many cardiothoracic surgeons in tertiary hospitals in Malaysia offer surgical intervention for such cases. Instead, the treating clinicians treat such patients conservatively. If the hemoptysis is massive, the patient will be intubated to prevent aspiration of blood. This complication occurred in our patient, in whom intubation was performed during the first episode of massive hemoptysis (with 300-400 ml blood). The causes of hemoptysis unknown [2]. It has been reported to occur following the release of trypsin and endotoxin which hydrolyse the adjacent vessels. Mechanical irritation of vasculature in the aspergilloma cavity may also contribute to bleeding during our patient’s early presentation. In the majority of cases, failure of medical treatment warrants radiological intervention approach to stop the bleeding as a temporary measurement [4]. Our patient had undergone embolization of the feeding vessels of the lesion and bleeding stopped for one week. Nevertheless, episodes of recurrent bleeding and development of hospital-acquired infection rendered our patient ventilated in the intensive care unit for another three weeks. Repeat CT thorax revealed multiple aspergilloma of various sizes in both lungs with evidence of local invasion, which was not seen in the earlier CT thorax. Bronchial artery embolization outcome was not great [5].

The local invasion could explain the possible pathophysiology of hemoptysis in our patient. The other possible mechanism could be superimposed bacterial infection. Multidisciplinary discussions have discussed the best available treatment options in view of failed
embolization. Unfortunately, few options were left because the patient had already developed ventilated-associated pneumonia. The patient would not be able to withstand one lung ventilation owing to poor left lung function. Bronchial artery embolization was only a temporary measure in our case. This can be useful as a method to reduce risk of massive bleeding prior to surgical resection. The collateral vessel will develop as early as 4-7 days in our patient. However, one case reported that Bronchial artery embolization was able to remove fungal ball radiologically in 2 weeks [6].

Early surgical intervention in selected patients with complex aspergilloma has been found promising [7]. Even though there has been no consensus regarding the timing of surgery [2], the multidisciplinary discussion should be initiated as early as possible. If the resident surgeon has inadequate experience, the patient should be referred to established centres to ensure the better surgical outcome. However, the mortality rate has been found to be 25%, with the morbidity of 60% in cases of excessive bleeding [8].

Three types of surgery can be offered for aspergilloma cases, including wedge or segmentectomy, anatomical resection such as pneumonectomy or lobectomy and cavernostomy and thoracoplasty [2]. Though anatomical resection is the gold standard [2], in some cases such as our patient, surgical removal is not suitable in view of involvement of both lungs.

CONCLUSION

Bronchial artery embolization successfully embolized the feeding vessel; however, failed to prevent recurrent bleeding in our patient 4-7 days post embolization. Recurrent massive hemoptysis caused by multiple bilateral lung aspergilloma is very difficult to manage. Early multidisciplinary discussion and referral may improve morbidity and mortality in the future.

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CONFLICT OF INTEREST

The authors declare no conflict of interest, financial or otherwise.

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