CASE REPORT

Massive haemoptysis secondary to mycotic pulmonary artery aneurysm in subacute invasive aspergillosis

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SUMMARY

A 68-year-old man, presented with 3 week history of infective symptoms and mild haemoptysis. Past medical history included severe emphysema and a chronic right upper lobe (RUL) cavity. He was discharged from follow-up a year ago in view of clinical and radiological stability; previous bronchoscopic examinations yielded no specific diagnosis. CT scan on admission confirmed complex cavitary consolidation of RUL. He developed massive haemoptysis requiring intubation and ventilation. CT pulmonary angiogram (CTPA) revealed 16 mm RUL pulmonary artery (PA) aneurysm which was successfully embolized. Sputum cultures, aspergillus antigen and rapidity of clinical progression suggested a diagnosis of subacute invasive aspergillosis (SAIA), prompting treatment with Voriconazole. Bronchoscopy showed blood ooze from RUL even after embolization. Unfortunately, patient continued to deteriorate and succumbed to profound septicaemia.

BACKGROUND

SAIA represents the severe form of aspergillosis; angioinvasion being the hallmark and haemoptysis as one of the most common symptoms. Haemoptysis in SAIA is usually from bronchial circulation. Our case was different as he developed massive haemoptysis secondary to PA aneurysm. To the best of our knowledge, very few case reports of significant haemoptysis secondary to PA aneurysm in invasive aspergillosis have been reported in the literature.

CASE PRESENTATION

A 68-year-old man presented with shortness of breath and productive cough with mild haemoptysis. He was followed up in the respiratory clinic up to a year ago for severe emphysema and chronic RUL cavity. Serial CT scans showed a radiologically stable cavity (figure 1). Bronchoscopy was performed twice during the follow-up period but yielded no specific diagnosis. He was discharged from follow-up a year ago in view of clinical and radiological stability.

During this admission, he had raised inflammatory markers with patchy opacities throughout the right lung on chest x-ray (CXR). Blood cultures, HIV, vasculitis profile and atypical pneumonia screen were all negative. CT scan showed complex cavitary consolidation of RUL. He was treated

with broad spectrum intravenous antibiotics and a bronchoscopy was requested. He developed massive haemoptysis on the fifth day of admission treated initially with Tranexamic acid. He remained haemodynamically stable and haemoptysis settled temporarily. Meanwhile, initial sputum cultures came back positive for aspergillus fumigatus and treatment was escalated to Voriconazole.

He suffered a cardiorespiratory arrest next day necessitating transfer to critical care. Bedside bronchoscopy by critical care team revealed a large clot completely occluding the right upper lobe and partially obstructing the bronchus intermedius. An emergency CTPA revealed a 16 mm PA aneurysm arising from posterior segmental branch of RUL PA as a likely source of haemoptysis (figure 2). Embolization of the PA aneurysm (figure 3) was performed on the same day using a 6 mm Amplatzer four plug with good results (figure 4).

On seventh day of admission, bedside bronchoscopy was performed by the respiratory team that showed fresh blood oozing from the posterior segment of right upper lobe; haemostasis was achieved with cold saline and intrabronchial instillation of epinephrine. Aspergillus antigens were reported as positive by that time.

On eighth day of admission, he developed worsening hypoxaemic respiratory failure despite ventilatory support and further bloody secretions were aspirated from endotracheal tube. CTPA was repeated which did not show any contrast extravasation from the previously embolized PA but worsening of right sided consolidative changes along with new left lower lobe consolidation. Unfortunately, patient continued to deteriorate and succumbed to profound septicaemia.

INVESTIGATIONS

Included in case presentation.

DIFFERENTIAL DIAGNOSIS

1. Cavitating pneumonia
2. Mycobacterial infection
3. Necrotizing lung cancer
4. Pulmonary infarction
5. Pulmonary vasculitis
6. Uncommon fungal infections including chronic cavitary pulmonary histoplasmosis, paracoccidioidomycosis and coccidioidomycosis.

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Figure 1 CT Thorax November 2014.

TREATMENT

Included in case presentation.

OUTCOME AND FOLLOW-UP

The patient was transferred to critical care after suffering a cardiorespiratory arrest on the ward. He continued to deteriorate despite optimal management and succumbed to profound septicæmia likely due to aspiration pneumonitis.

DISCUSSION

Clinical manifestations of aspergillosis are very much host dependent. These range from simple aspergilloma, aspergillus nodule, chronic cavitary pulmonary aspergillosis and chronic fibrosing pulmonary aspergillosis in an immune competent subject; to subacute invasive aspergillosis and invasive aspergillosis in immunosuppressed individual.¹ Chronic obstructive pulmonary disease (COPD) and frequent use of both inhaled and systemic steroids during exacerbations has been considered a risk factors for invasive aspergillosis, as in our patient.²

In a patient at risk with symptoms of haemoptysis, aspergillosis should be suspected. Definitive diagnosis of invasive aspergillosis is made when aspergillus is grown from appropriate samples along with histopathological evidence of tissue invasion by fungal hyphae.³ This combination is not achieved frequently as was the case in our patient. Our patient had proven invasive fungal disease with positive mycological cultures from sputum samples. Necrotizing/cavitating pneumonia remains the main

Figure 2 CTPA showing pulmonary artery aneurysm.

Figure 3 Pulmonary artery angiogram showing aneurysm.

differential diagnosis and empiric therapy should involve appropriate broad-spectrum antibiotics.

Angioinvasion is an important characteristic of invasive aspergillosis.⁴ Lungs have both high and low pressure arterial circuits; bronchial and pulmonary arteries respectively. In more than 90% of the cases, haemoptysis is from bronchial or systemic arteries. Haemoptysis originating from pulmonary circulation constitutes less than 10% of the cases while a minor fraction is from microvessels and veins.⁵ Hence the initial work up of haemoptysis should include arterial phase CT and persistent bleeding should be investigated with CTPA.⁶ There are very few published case reports on significant haemoptysis secondary to PA aneurysm in invasive aspergillosis; these case reports are summarised in [table 1](#).

Voriconazole is considered the drug of choice for the treatment of invasive aspergillosis because of better response rate with improved survival (71% vs 58%) and less side effects when

Figure 4 Pulmonary artery angiogram – post embolization.

Table 1 Previous case reports of PA aneurysm associated with aspergillosis

Case report	Cause of haemoptysis and underlying conditions
Caulet <i>et al</i> , 1990 ¹³	Chronic necrotizing aspergillosis, diabetes mellitus.
Bowler <i>et al</i> , 1998 ¹⁴	Invasive aspergillosis, emphysematous bulla.
Kim <i>et al</i> , 2000 ¹⁵	Chronic necrotizing aspergillosis.
Sabayev <i>et al</i> , 2005 ¹⁶	Invasive aspergillosis, AIDS.
Sever <i>et al</i> , 2010 ¹⁷	Invasive aspergillosis, hairy cell leukaemia.
Kokkonouzis <i>et al</i> , 2011 ¹⁸	Chronic cavitary aspergillosis, COPD, non-tuberculous mycobacterial infection.
Cooper <i>et al</i> , 2013 ¹⁹	Invasive aspergillosis, Crohn's disease.
Didier <i>et al</i> , 2014 ²⁰	Invasive aspergillosis, acute myeloid leukaemia.
Reddy <i>et al</i> , 2014 ²¹	Invasive aspergillosis, myelodysplastic syndrome.

compared with Amphotericin B.⁷ Voriconazole was started in our patient as soon as the diagnosis of invasive aspergillosis was suspected.

Different modalities have been employed for the management of massive haemoptysis. Conservative treatment involves management of haemodynamic instability and correction of bleeding diathesis along with antifungal treatment but is associated with around 50% mortality.⁸ Bronchoscopy is an effective diagnostic and therapeutic tool once airway control has been achieved. Bronchoscopy can miss the bleeding site in around 50% of cases, and may not be useful due to tissue fragility.⁹ Emergency cardiothoracic surgery carries mortality of 40% in itself.¹⁰ CT angiogram is superior to arteriography in detection of bleeding from bronchial and non-bronchial systemic arteries, and also from inflammatory pulmonary artery aneurysms.¹¹ Pulmonary vascular embolization remains an important intervention. On-going bleeding after arterial embolization carries high mortality, as was the case with the subject under discussion.¹²

Our patient had CTPA that confirmed RUL PA aneurysm which was embolized successfully. The slow ooze of blood seen on the follow-up bronchoscopy was possibly from microvascular origin. Hence, massive haemoptysis observed in our patient had more unique and complex mechanisms. Most likely, this originated from a low pressure vascular system and even though major source of bleeding was controlled, it did not stop haemoptysis altogether. Unfortunately, due to ongoing septicaemia and blood loss, the patient could not survive.

Learning points

- ▶ Massive haemoptysis is a life-threatening emergency requiring airway protection and haemodynamic support.
- ▶ Early bronchoscopy and CT angiogram are required to identify and arrest the focus of bleeding.
- ▶ Haemoptysis originates from bronchial arteries in >90% of patients whilst <10% of the cases are from pulmonary circulation.
- ▶ High index of suspicion is needed to initiate early antifungal treatment in invasive aspergillosis to prevent such fatal complications.

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