CASE REPORT

Bronchial arterial embolisation for massive haemoptysis in cavitary sarcoidosis

Geoffrey Andrew Loh,¹ Christopher J Lettieri,² Anita A Shah³

¹Department of Internal Medicine, Walter Reed National Military Medical Center, Bethesda, Maryland, USA

²Department of Pulm/Critical Care/Sleep, Walter Reed National Military Medical Center, Bethesda, Maryland, USA

³Madigan Army Medical Center, Tacoma, Washington, USA

Correspondence to
Dr Geoffrey Andrew Loh,
geoffloah@gmail.com

SUMMARY

A 48-year-old non-smoking man with a 6-year history of pulmonary cavitary sarcoidosis presented with acute onset of haemoptysis of approximately 600 ml. Prior episodes of haemoptysis had resolved only after serial upper lobe wedge resections bilaterally and steroids. A chest CT identified bilateral upper lobe cavitary lesions with extravasation of contrast from a large right upper lobe cavity. The patient underwent urgent bronchial angiography and subsequent bronchial artery embolisation of a left bronchial artery and three right bronchial arteries. He was started on methotrexate and steroids for refractory sarcoidosis. Two years after embolisation, the patient remained haemoptysis-free with his sarcoid well controlled on methotrexate monotherapy.

BACKGROUND

This case is important for a number of reasons. Only a couple of case reports have been published previously about arterial embolisation for haemoptysis in sarcoidosis as it is a rare manifestation of the disease. However, there are no guidelines for management of this complication of this disease and the best management for this complication is also unclear. Arterial embolisation is becoming more and more favoured over wedge resections for massive haemoptysis due to being less invasive and offering fewer complications, especially to those who are poor surgical candidates or have less pulmonary reserve. Others will be able to learn from the case in thinking about the issues we present with haemoptysis in patients with underlying disease.

CASE PRESENTATION

A 48-year-old man, non-smoker, presented with 500 ml of bright red haemoptysis. He was diagnosed with pulmonary sarcoidosis 6 years earlier during evaluation for bilateral upper lobe cavitary lesions noted on chest roentogram. At the time of diagnosis, he was asymptomatic, denied prior episodes of haemoptysis and did not require medical therapy. He remained asymptomatic until 4 years prior to admission when he presented with his first episode of massive haemoptysis. Chest CT revealed progressive enlargement of a right upper lobe (RUL) cavitary lesion. Bronchoscopy revealed active haemorrhage from his right upper lobe. No infectious aetiology was identified. He underwent wedge resection of his RUL cavity and was initiated on prednisone therapy. The patient experienced

resolution of haemoptysis for approximately 2 years when he represented with massive haemoptysis. CT scan revealed progressive enlargement of his parenchymal cavities and active haemorrhage was noted from his left upper lobe (LUL) on bronchoscopy. Wedge resection of his LUL cavity lesion and reinstituting corticosteroid therapy resulted in resolution of haemoptysis. His current presentation with acute onset of 500 ml of haemoptysis was 18 months after his second wedge resection and in the setting of recently discontinuing his steroids. He denied fevers or recent illnesses.

INVESTIGATIONS

On presentation, the patient was noted to be anxious and dyspnoeic with haemoptysis. He had tachycardia and tachypnoea with an oxygen saturation of 90% on supplemental oxygen. Blood pressure was normal. Bilateral diffuse crackles were auscultated.

The patient's haemoglobin, platelet count and coagulation factors were within normal limits. A contrast-enhanced chest CT revealed thickening and expansion of cavitary lesions in both upper lobes and interval development of new lower lobe cavities bilaterally. Focal extravasation of contrast into an upper lobe cavity was noted (figure 1). Bronchoscopy identified active haemorrhage from both upper lobes that could not be controlled through endobronchial modalities.

TREATMENT

Despite prior wedge resections of both upper lobes, the patient continued to have recurrent episodes of acute, massive haemoptysis involving multiple lobes of his lungs. Furthermore, the patient had

Figure 1 Multiple bilateral cavitary lesions are shown with extravasation of contrast in the right lung. Bronchoscopy confirmed active haemorrhage from bilateral upper lobes.

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Figure 2 This blushing of parenchyma represented the source of his haemorrhage from his right lung. Embolisation of three intercostobronchial trunks on the right lung and one on the left lung resulted in resolution of haemoptysis.

progression of his existing cavities and developed new lower lobe lesions. Although another wedge resection would be possible in a person with pulmonary haemorrhage, parenchymal-sparing strategies had to be considered given the concern that he would require future resections which could probably compromise his remaining pulmonary function.

Angiography of bronchial arteries and the thoracic aorta revealed blushing of the parenchyma in the right mid-lung (figure 2) and extravasation of contrast from a left-sided intercostal bronchial trunk. Successful particle embolisation was performed of three right intercostal bronchial trunks and one left-sided bronchial trunk. The patient tolerated the procedure well and there were no obvious complications identified. His haemoptysis resolved and repeat bronchoscopy did not reveal further haemorrhaging. No infectious aetiologies were identified and all cultures were negative.

OUTCOME AND FOLLOW-UP

Following the procedure, the patient was restarted on corticosteroids and methotrexate was eventually added to reduce his steroid dose. During serial follow-up evaluations over the next 2 years, the patient remained asymptomatic with no further episodes of haemoptysis. He reported significant improvements in exercise tolerance. A repeat CT at 2-year follow-up revealed a dramatic decrease in the size and wall thickness of his pulmonary cavities.

DISCUSSION

Sarcoidosis is an idiopathic, systemic disease hallmarked by non-caseating granuloma formation that can affect multiple organ systems. Pulmonary involvement is the most common manifestation and may lead to parenchymal fibrosis. While uncommon, cavitations may also form. In a series of 1060 patients with sarcoidosis with high resolution CT, 3.9% were found to have cavitary lesions.¹

Cavitary sarcoid is frequently complicated by recurrent haemoptysis, pneumothorax and an increased risk of secondary infections, especially aspergillus infection. On-going inflammation and progressive parenchymal cavitation can erode into bronchial vessels causing haemoptysis. Massive

haemoptysis, generally defined as between 300 ml and 500 ml,² may occur.

Haemoptysis can be managed conservatively or surgically. Intermittent, non-massive haemoptysis in sarcoids can often be treated successfully with conservative management and corticosteroids. During massive haemoptysis of all causes, death rates were high with death rates of 3 to 80% for those treated with conservative measures alone.³ Frequently, life-threatening massive haemoptysis may require urgent surgical resection of the involved cavitary lesion. Unfortunately, despite surgery, 11.5–23% of individuals do not survive.³

As in the presented patient, sarcoidosis may be progressive with enlargement of previously identified cavities and development of new lesions. As such, there is a potential need for multiple resections. In addition, the presence of fibrosis and already impaired lung function may further limit tolerance of repetitive resections. Given this, parenchymal sparing means to control haemoptysis are favourable for many patients.

Bronchial angiography involves catheterisation of the aorta and using contrast to visualise the intercostobronchial trunks. Bronchial circulation is a higher pressure system relative to pulmonary circulation and responsible for up to 90% of cases of massive haemoptysis.² Bronchial angiography can identify abnormalities such as hypervascularity, bronchial artery hypertrophy, vascular abnormalities or extravasation of contrast into bronchial lumens. Embolisation of these areas can mitigate haemoptysis and offer a potentially surgery-sparing and parenchymal-sparing approach to massive haemoptysis. The use of bronchial artery embolisation (BAE) for acute management of massive haemoptysis was first reported in 1973 and has emerged as an alternative therapy in individuals who are poor surgical candidates or when haemostasis cannot be achieved by alternative endobronchial modalities.⁴

For patients presenting with haemoptysis of all causes, BAE is frequently successful with 73–98% experiencing short-term resolution of haemoptysis.² Rates of long-term success have varied between 10% and 52%. However, this is based on relatively small sample sizes and markedly different observation periods ranging from 1 month to 4 years.² Given the successes reported for other causes of massive haemoptysis, BAE may represent a reasonable therapeutic option for patients with cavitary sarcoidosis presenting with massive haemoptysis. Not only can it resolve acute bleeding, it mitigates the need for resection and further loss of lung parenchyma. BAE should be considered in those who are poor surgical candidates, those with extensive parenchymal involvement and individuals with other cavitary lesions posing a risk for future episodes of haemorrhage. Furthermore, one case report identifies a patient with recurrent haemoptysis in cavitary sarcoidosis in which pulmonary artery embolisation, in addition to BAE, was successful in halting haemoptysis.⁵

Although frequently successful, BAE is associated with potential complications. Chest pain and dysphagia are common but generally transient events are probably secondary to ischaemia. Vessel dissection with subintimal dissection of the aorta or bronchial artery during BAE has a reported prevalence of 1–6.3%.⁶ The most significant complication reported with BAE is spinal cord ischaemia and infarction, which occurs in 1.4–6.5% of cases.⁶

Learning points

- ▶ Cavitory lesions are present in 3.9% of sarcoid patients and predispose patients to haemoptysis, as well as increasing risk for infection.
- ▶ Conservative management and corticosteroid therapy may be effective for intermittent bleeding. However, massive haemoptysis frequently requires surgical resection.
- ▶ BAE provides a parenchyma sparing option for management of massive haemoptysis and has successfully been used in cavitory sarcoidosis.
- ▶ Spinal cord ischaemia is the most significant adverse consequence of bronchial artery embolisation and occurs in 1.4–6.5% of cases.
- ▶ Bronchial circulation is a higher pressure system relative to pulmonary circulation and responsible for up to 90% of cases of massive haemoptysis.

Competing interests None.

Patient consent Obtained.

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