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Case report

Treatment of a large pulmonary artery pseudoaneurysm secondary to fungal infection using Amplatzer plugs: New embolisation devices for the management of haemoptysis

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ABSTRACT

Pulmonary artery pseudoaneurysms (PAP) may result in life threatening haemoptysis but are fortunately uncommon. Most are caused by trauma, iatrogenic injury or infection. We describe a case of large PAP secondary to fungal infection in an immunocompromised patient, which was successfully treated percutaneously using Amplatzer embolisation plugs. The technical considerations and advantages of these new devices are explained.

1. Case report

A 76-year old male with a three-year history of Myelodysplasia presented with symptoms and signs of right upper lobe pneumonia. Initial investigations revealed neutropenia (neutrophil count 1.0) for which he was commenced on Clarithromycin, Tazocin and Gentamicin. Blood and sputum cultures for bacteria including acid-fast bacilli and urinary antigen for Legionella were negative. A chest x-ray confirmed right upper lobar pneumonia with a bulging horizontal fissure (Fig. 1). On going high-grade pyrexia and haemoptysis prompted a change of antibiotic regime to include antifungal agents after repeat sputum cultures grew *Stenotrophomonas maltophilia* and *Candida melibiosica*. A contrast enhanced CT chest was performed to investigate the cause of haemoptysis. This demonstrated right upper lobe pneumonia as well as a large pulmonary artery pseudoaneurysm (Fig. 2).

Due to the size of the pseudoaneurysm and potential for continued or massive haemoptysis, a decision was made to proceed to embolisation. Informed consent was obtained prior to arterial access and placement of a 5 French sheath in the right common femoral artery. Right upper lobe pulmonary angiography demonstrated the feeding pulmonary artery (Fig. 3), which was crossed using a Terumo (Terumo Corporation) hydrophilic guide wire and 4 French Bernstein (Cook medical) catheter. A 6 French straight destination sheath was exchanged at the groin and advanced to just beyond the pseudoaneurysm origin. A 6 mm Amplatzer 1 (AGA medical) plug was deployed distally and a second 8 mm Amplatzer 1 plug proximally to occlude both the inflow and any potential for back flow into the pseudoaneurysm. Completion angiography (Fig. 4) demonstrated complete cessation of filling of the pseudoaneurysm. There were no procedural complications.

Following embolisation and commencement of antifungal agents there was immediate rapid resolution of haemoptysis and within two days, resolution of pyrexia. A follow up chest x-ray at 8 weeks showed continued satisfactory resolution of the consolidative changes with some right upper lobe volume loss. The Amplatzer plugs were noted to be unchanged in position (Fig. 5).

2. Discussion

Pulmonary artery pseudoaneurysms (PAP) may result in potentially life threatening haemoptysis but are fortunately uncommon. In our case, the causative organism resulting in pseudoaneurysm formation is unclear, however severe lobar pneumonia with repeated infection/inflammation and heavy coughing with initially suboptimal antibiotic treatment, may have all played a part. Arterial phase contrast enhanced multislice CT was the key in making the diagnosis and enabled planning of endovascular intervention as well as embolisation device selection.

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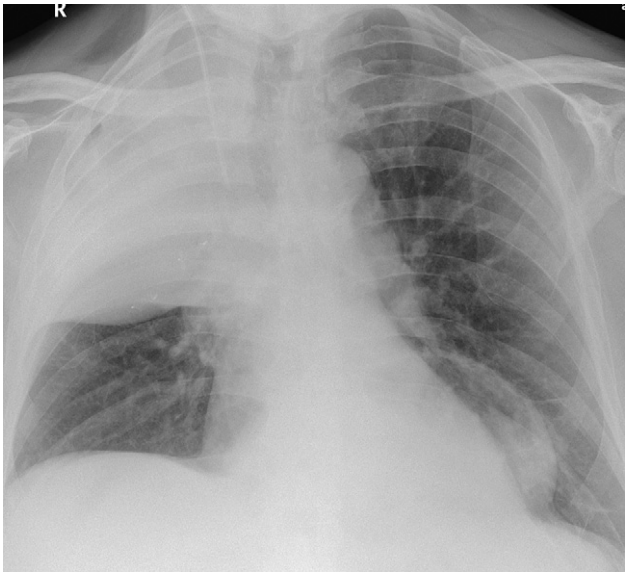

Fig. 1. Initial AP erect chest radiograph showing right upper lobe consolidation and a bulging horizontal fissure consistent with lobar pneumonia. There is an incidental hiatus hernia behind the heart.

Pseudoaneurysms by definition do not have a covering of all three layers of the arterial wall and are effectively contained arterial leaks that are considered to be at a high risk of rupture. When occurring within a consolidated or infected lung, the potential for tissue breakdown, at the margins of the pseudoaneurysm and communication with the airways, may result in massive haemoptysis or even death. Early recognition and minimally invasive endovascular treatment may help to reduce the associated mortality rate of PAP.

PAP are often associated with trauma and most commonly result secondary to complications from Swan-Ganz catherisation.[1] Amongst the infective causes, the most common is due to pulmonary tuberculosis. These pseudoaneurysms, known as Rasmussen aneurysms arise from small to medium sized pulmonary arterial branches in the vicinity of a tuberculous cavity.[2] Other cases of PAP have been described with fungal infections, pyogenic bacteria and Mucormycosis.[3] It is important to remember that the majority of cases of haemoptysis occur due to bronchial arterial bleeding, most often occurring in chronically infected or inflamed lung tissue such as in patients with bronchiectasis or cystic fibrosis. This is traditionally investigated with bronchoscopy for localization of the

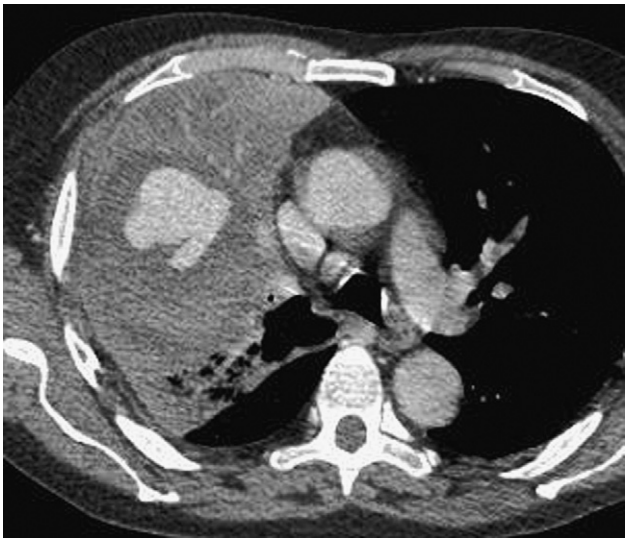

Fig. 2. Contrast enhanced CT chest in arterial phase reveals a large pseudoaneurysm of the right upper lobe pulmonary artery with surrounding upper lobe consolidation.

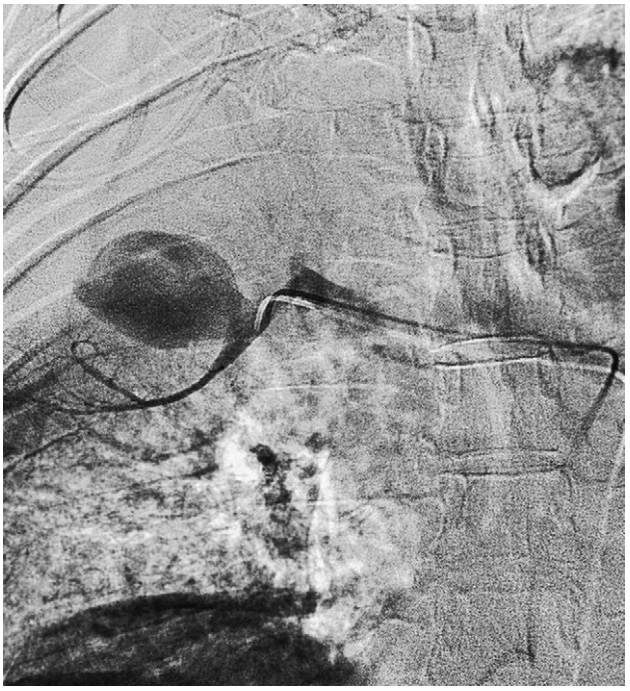

Fig. 3. Selective angiogram of the right upper lobe pulmonary artery reveals the pseudoaneurysm arising from the feeding right upper lobe pulmonary artery segmental branch.

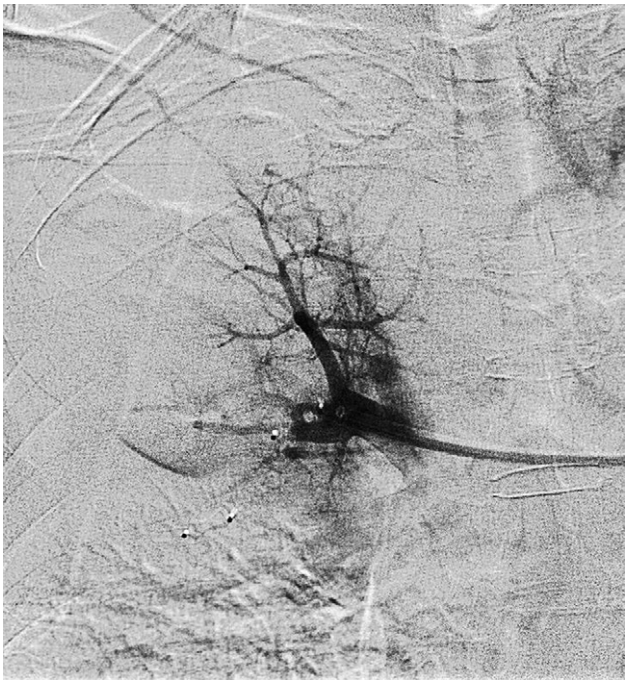

Fig. 4. Final angiogram post embolisation demonstrating absence of filling of the pseudoaneurysm. The end markers of the two Amplazter plugs are visible close to the tip of the guide catheter. There is preservation of an adjacent pulmonary segmental artery.

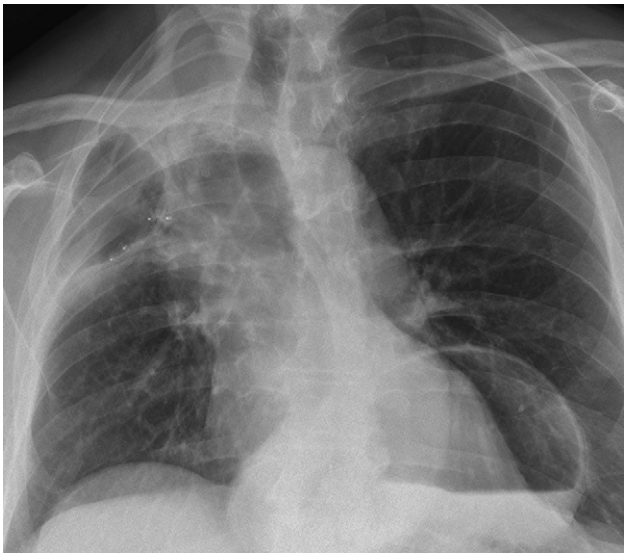

Fig. 5. Follow up chest x-ray shows volume loss of the right upper lobe with shift of the trachea to the right. The Amplatzer plugs are seen in the right upper zone and are unchanged in position. An incidental air fluid level is noted within the hiatus hernia. The patient's symptoms had resolved completely.

bleeding lobe followed by catheter angiography and embolisation. The improved spatial resolution and diagnostic capabilities of arterial phase contrast enhanced multislice CT using multiplanar reconstructions however, is likely to favour a non invasive diagnostic modality approach first, increasingly into the future.

Mycotic PAP are thought to be caused by several mechanisms such as direct extension of pneumonia to involve the vessel wall, endovascular seeding of the vessel wall from bronchial arteries in septicaemia and intimal invasion of the vessel wall from septic embolism. These all may lead to focal vessel wall damage or necrosis and subsequent dilatation and pseudoaneurysm formation.[4]

Contrast enhanced Multislice CT in the arterial phase allows accurate anatomical localization of the aneurysm and direct visualization of the feeding artery by its ability to acquire isometric volume data. This information is helpful for planning optimal angles for visualizing of the aneurysm during the selective arterial catheterization and embolisation.[5]

The mortality rate associated with massive haemoptysis is greater than 50% for patients who undergo conservative management.[6] Spontaneous regression of small, asymptomatic lesions has been observed.[7] Haranga et al., described a case of PAP secondary to lung abscess, which settled with antibiotic treatment alone.[8] Endovascular embolisation and resection of the affected pulmonary lobe are the most commonly performed treatment options for pseudoaneurysms. Postoperative complications are encountered in approximately 50% of these patients and a fatal outcome occurs in 20% especially when surgery is performed within the first 24 h after haemoptysis.[9] In our case the mortality risk was considered to be high due to the size of the PAP and associated co-morbidities.

With improving interventional vascular radiology techniques, transcatheter coil embolisation of the feeding artery or filling of the sac itself with coils has played a major role in the management of PAP in the past. Although many embolisation materials have been previously suggested as well like direct injection of sclerosant into the pseudoaneurysm sac, our case demonstrates a quick, safe and effective use of Amplatzer embolisation plugs for the treatment of PAP's. These nitinol wire mesh, self expanding plugs are oversized by 30–50% of the diameter of the intended vessel, thereby ensuring plug stability. A single plug is usually sufficient to occlude the vessel, which avoids the long procedural times and potential for non-target embolisation when using multiple conventional pushable coils. The ability to retract the plug following initial deployment allowing repositioning which is also helpful when dealing with multiple short branches and complex anatomy of the pulmonary arterial tree. This allows preservation of as much involved lung tissue as possible minimising the reduction of residual lung function. In this case, our patient was treated successfully by endovascular techniques, thereby avoiding major surgery. Furthermore he tolerated the procedure well and made an uneventful recovery.

Conflicts of interest

None of the authors has declared any conflict of interest within the last three years which may arise from being named as an author on the manuscript.

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