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GI/GU/Thoracic/Nonvascular Interventions *Case Report*

Coil embolization of a pulmonary artery-bronchial fistula associated with intra parenchymal pseudoaneurysm, endovascular approach, and bronchoscopy monitoring – A case report

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ABSTRACT

A 62-year-old man with a heavy background of the right upper lobe squamous cell carcinoma was admitted with massive hemoptysis. Computerized tomography (CT) showed a large right upper lobe lesion with infiltration of mediastinum and airways. A right pulmonary artery pseudoaneurysm slightly bulging into the right main bronchus was seen, suspicious for a pulmonary artery bronchial fistula. A combined endotracheal and angiographic approach was done. The right pulmonary artery angiogram showed a now occluded neck to the pseudoaneurysm which was selectively cannulated. Framing and packing coils were deployed. Contrast was endoscopically noted without bronchial bleeding. As the coils were entering the lung parenchyma, packing back to the neck was done carefully. No new bleeding was noted. A bronchial stent was inserted 6 weeks later and one coil protruding into the bronchus was seen. A 3 month CT follow-up did not show any residual pseudoaneurysm. Patient died 3 months after initial procedure of his progressive illness. Only three cases of endovascular management of bronchial-pulmonary artery fistula reported with different techniques including pulmonary artery stenting. Our case is the only one reported treated with only coil embolization. Limited data are available regarding the risk of pulmonary embolism after stenting. Anticoagulation required after stenting increases the risk of bleeding in patients with severe hemoptysis. Management with coils only facilitates further management. This preserves pulmonary circulation in patients with already compromised respiratory function. Coil only embolization of uncommon pulmonary artery bronchial fistula is feasible and facilitates further management as it does not require anticoagulation in patient with recurrent bleeding.

Keywords: Broncho vascular fistula, Lung cancer, Pulmonary artery

Herein, the authors present a case of a pulmonary artery bronchial fistula and pseudoaneurysm in a patient with massive hemoptysis and a right upper lobe squamous cell carcinoma.

INTRODUCTION

Massive hemoptysis secondary to a bronchovascular fistula is a potentially fatal condition requiring urgent intervention. Bronchovascular fistula formation is most commonly seen after lung intervention including endobronchial stent placement, anastomotic complications following sleeve resection, lung transplantation, and compromised stump after pneumonectomy. The

occurrence of such fistulas without intervention is relatively rare.[1]

Management of major hemoptysis is generally done one of four ways drug therapy, bronchoscopic intervention, catheter intervention, and lobectomy. Recent evidence has suggested coil embolization of arterial bleeding in hemoptysis is a well-recognized and relatively safe treatment.[2]

In this case, we present a patient with a pulmonary arterial-bronchial fistula presenting with massive hemoptysis secondary to a pulmonary arterial pseudoaneurysm connecting with the right main bronchus.

CASE REPORT

A 62-year-old man with a right upper lobe squamous cell carcinoma (SCC) undergoing chemotherapy presents with 500 mL hemoptysis. Two years before this, he was diagnosed with a right upper lobe SCC Stage IIIa, T4N2M0, with spread to mediastinal and hilar lymph nodes, after presentation with a clear productive cough. This was deemed not suitable for curative management and a combination of chemotherapy and radiotherapy was undertaken. His lung function tests at the initial presentation showed an FEV1 1.81L (47%) and a DLCO 6.8L (66.8%). His background included a 60 year pack history, with nothing else of note. He was treated with six cycles of carboplatin and gemcitabine which showed good response at the completion staging computerized tomography (CT). Subsequently, he underwent radical radiotherapy to the right upper lobe with further response seen on CT. A trial of durvalumab 1000 mg twice weekly was started 8 months after diagnosis as an adjunct. In June of this year, a CT showed complete right upper lobe collapse without further lung or distant lesions. A follow-up position emission tomography scan showed an flourodeoxyglucose avid right upper lobe tumor obstructing the right upper lobe bronchus, mediastinal invasion, and significant mass effect on the superior vena cava and pulmonary artery. Endoscopic bronchial ultrasound confirmed that the SCC had invaded a level 4R lymph node. Further carboplatin and gemcitabine were started and one cycle had been completed at the time of presentation. Just before the second treatment of carboplatin/gemcitabine, he was admitted to hospital with a ST-elevation myocardial infarction, found to be in atrial fibrillation, and had percutaneous coronary intervention to his right coronary artery and a pneumonia during this admission managed with antibiotics. He was discharged from that admission on aspirin and ticagrelor for 1 year but not anticoagulated due to tumor proximity to major vessels.

Two years post-diagnosis, the patient attended a peripheral emergency department with large volume (500 mL) hemoptysis and oxygen saturations of 80% on room air. He was transfused 2 units red blood cells in the emergency room and was transferred to a tertiary center. His case was discussed as an emergency in the lung cancer multidisciplinary meeting that morning and the decision was taken to bring the patient forward for simultaneous endobronchial examination and endovascular treatment of the pseudoaneurysm.

Pre-operative CT imaging showed a large right upper lobe lesion with infiltration of mediastinum, trachea, and airways with contrast blush into the right main bronchus with apical consolidation and opacities, bronchiectasis pulling the mediastinum to the right [Figure 1]. The presumptive diagnosis was infection and hemorrhage in combination with the likely sight of bleeding being an 11 × 9 mm right pulmonary artery pseudoaneurysm.

The procedure was performed under general anesthetic, with an endotracheal tube placed into the right main bronchus with a fiber optic scope at the carina to monitor any hemoptysis. This allowed the option of treatment from the bronchus if required, however, it was felt most appropriate to treat it intravascularly in the first place. Initially, an 8 French (Fr) 90 cm destination sheath (Terumo, Shebuya City, Japan) was placed into the right common femoral vein under ultrasound guidance and advanced into the right pulmonary artery, where an angiogram showed a distorted main right pulmonary trunk with a now occluded neck to the aneurysm [Figure 2]. At this point, the thoracic aorta was also imaged through the right groin showing tortuous hypertrophic bronchial arteries but no evidence of any pseudoaneurysm and unlikely to be the source of the patient's hemoptysis. A 0.038 5 Fr Bern Catheter (Merrit, Utah, USA) was used to hook the neck of the pseudoaneurysm. From this point, on attempts were made to try and pass a wire into the fistula. After a prolonged effort, a small neck was identified, and a 0.016 pre-shaped Fathom wire (Boston Scientific, Massachusetts, USA) was passed into the pseudoaneurysm. A 2.6 Fr 45° lantern microcatheter (Penumbra, California, USA) was advanced into the sac. Once in the aneurysmal space, a combination of framing and packing coils was placed with careful flushing and small injections of contrast. A combination of soft ruby coils (Medtronic, Minneapolis, USA) was used with 2 × 8 mm × 35 cm, 1 × 6 mm × 20 com, 1 × 6 mm × 30 cm, 2 × 4 mm × 15 cm, POD5 30 cm coils × 2 and J-pod 60 cm × 1 (Penumbra, California, USA). These were visualized by the anesthetist through the scope at the carina, but no hemoptysis was identified. It became apparent that the aneurysm had no real wall and that coils were entering the lung parenchyma [Figure 3]. It was then decided to pack back to the neck of the aneurysm which was done carefully.

Final angiogram showed no further communication with coils down to the neck of the aneurysm. A good dense ball of coils was achieved. The use of bronchoscopy in this instance was valuable in ruling out a primary bleed which could potentially have been treated with covered bronchial stent [Figures 3 and 4]. Direct visualization again allowed

Figure 1: Computerized tomography images of pseudoaneurysm: (a) Coronal view of right pulmonary artery pseudoaneurysm (yellow arrow), (b) sagittal view of right pulmonary artery pseudoaneurysm (yellow arrow), (c) axial view of right pulmonary artery pseudoaneurysm after embolization (white arrow), (d) showing proximity of right pulmonary artery pseudoaneurysm after embolization (white arrow) to the right stented main bronchus (green arrow), and (e) with stent in right main bronchus.

confirmation that coils had not been left in the bronchus potentially leading to bronchial collapse. A decision was made not to stent the pulmonary artery due to unknown behavior of a balloon expandable stent in a highly compliant vessel which also had tumor eroding into its wall.

Hemostasis of femoral artery was obtained with Starclose (Abbott, Illinois, USA) closure device and manual compression of the venous site. Total fluoroscopy time was 1:20:06 with a cumulative DAP 124.039Gy.cm^2 and exposure cumulative DAP 60.577Gy. cm^2. Total Dose was 1426mGy.

Postoperatively, the patient made an unremarkable recovery, he was extubated in the angiography suite and transferred to the ward. A post-procedural CT showed satisfactory coiling of the aneurysmal segment and no ongoing bleed, so his dual antiplatelet therapy (DAPT) was restarted. Five days following procedure, he had a repeat bronchoscopy which showed a large circumferential tumor invading the right main bronchus arising from the right upper lobe with erosion of the coils in to the bronchus. It was friable and bled easily; however, the patient had just restarted there DAPT. There was a small canal which could potentially be stented. After this, the patient was further discussed in the multidisciplinary meeting and decision to let everything settle and consider bronchial stenting at a future date if seen as appropriate by the medical oncologists. He was then transferred to a peripheral center for ongoing physical and occupational therapy input before discharge. Subsequently, he was brought back 6 weeks later for bronchial stent insertion [Figure 4]. He continued to remain without dyspnea, hemoptysis, or chest pain.

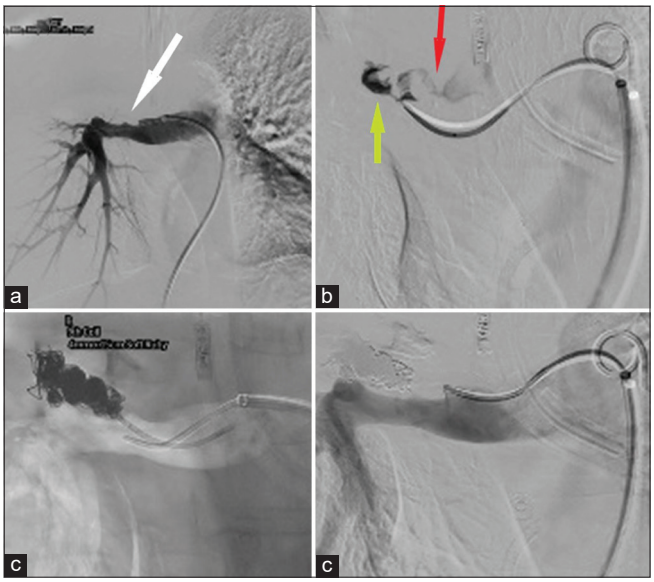

Figure 2: Angiography at time of coiling: (a) Pre-coiling showing a stump of the pseudoaneurysm neck (white arrow), (b) accessing the pseudoaneurysm and opacification of the sac (green arrow) and the pulmonary artery brochial fistula (red arrow), (c) shows coils in in the pseudoaneurysm, and (d) completion run showing exclusion of the pseudoaneurysm.

A follow-up CT scan was performed at 3 months. This showed no further hemorrhage with good placement of the coils. However, there was radiological progression of his lung cancer with increasing in size hilar lymphadenopathy and progressive collapse of the right upper lobe. He was subsequently admitted a few days following this CT scan with a productive cough and unfortunately succumbed to his illness a few days later.

DISCUSSION

Only three cases of endovascular management of bronchial-pulmonary artery fistula have been reported previously; occlusion of pulmonary artery with Amplatzer vascular plug (Abbott, Illinois, USA), stent graft placement in pulmonary artery, and bare stent and coil and N-butyl cyanoacrylate.[3-5]

This case is the only one to report treatment with only coil embolization with concomitant bronchial endoscopy. The alternative to this is pulmonary artery stenting, however, there is currently no long-term data about the outcome of pulmonary artery stents, either bare or covered. Pulmonary artery stents could theoretically increase the risk of pulmonary embolism and infection as well as requiring anticoagulation treatment. This is especially important in patients whose presentation is hemoptysis or those with lung cancer at it will give them an increased risk of bleeding from their primary pathology.

Management with coiling only facilitates further management, by not restricting further intervention if required. Coil embolization avoids blocking a proximal pulmonary artery allowing preservation of pulmonary circulation in patients with an already compromised respiratory function.

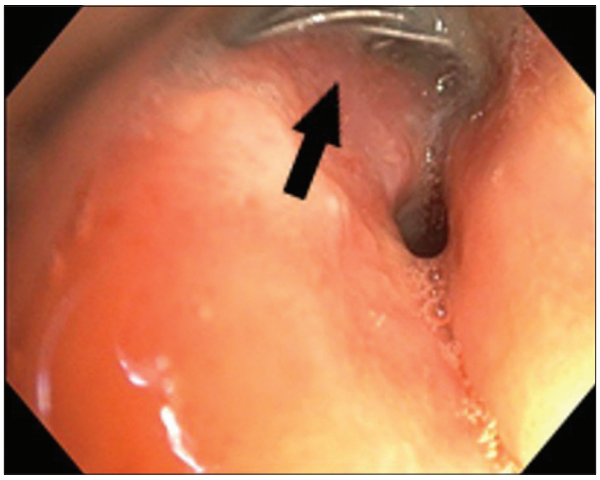

Figure 3: Endoscopic view of coil in the right main bronchus (black arrow).

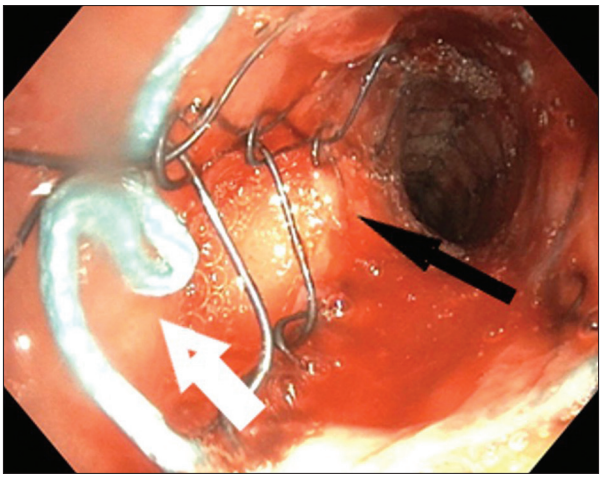

Figure 4: Endoscopically placed stent (black arrow) to the right mainstem bronchus trapping the coil (white arrow).

CONCLUSION

We have presented the case of a patient with massive and life-threatening hemoptysis on the background of lung cancer, successfully treated with coil embolization.

Acknowledgment

Waikato DHB Interventional radiology department.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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