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Anterior Spinal Artery Syndrome After Bronchial Embolization for Hemoptysis: A Case Report and Literature Review

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ABSTRACT

Background Bronchial artery embolization (BAE) is the established first-line treatment for patients presenting with massive haemoptysis, a life-threatening condition that can occur because of numerous underlying diseases. BAE is a relatively safe procedure with control of haemorrhage achieved in 77%–90% of cases and rare occurrence of complications. Spinal cord infarction is one such rare complication, which can have severe implications in terms of morbidity.

Case presentations We present a case of a 70-year-old man who developed paraplegia with loss of pain and temperature sensation as well as sphincteric involvement following BAE for hemoptysis. MRI of the spine was suggestive of an ischaemic event involving anterolateral spinal cord segment T4–T6, so a diagnosis of anterior spinal artery syndrome post BAE was made. The patient was given corticosteroids, dual antiplatelet medications, pregabalin, supportive management and regular physiotherapy. Follow-up of the patient at 3 and 6 months failed to show any significant improvement in neurological function, although the patient did not report problem of significant hemoptysis afterward.

Conclusion Spinal cord infarct is a rare and disabling complication of BAE despite it being a safe procedure with good long-term outcomes. Detailed knowledge about the anatomy of bronchial arteries and spinal arteries with detailed preprocedure investigations may lower the risk of this disabling complication.

INTRODUCTION

Massive hemoptysis is a potentially life-threatening complication of several underlying lung disorders that require urgent medical attention. Bronchial artery embolization (BAE) is currently the standard treatment for massive hemoptysis, with a long-term survival rate of around 85%.[1] Anterior spinal artery syndrome (ASAS) is a rare complication following BAE, the reported incidence of which is as high as 5%.[2] ASAS can lead to paraparesis and can significantly impair the functional status of patients despite treatment. We encountered a case of a 70-year-old man who underwent BAE for hemoptysis and was referred to us for problems of paraplegia and sphincteric involvement. Imaging confirmed spinal cord infarct. The patient did not show any improvement despite medical management. Despite BAE being a safe procedure with good outcomes, it can lead to lifelong disability by causing spinal cord infarction. So, this rare complication should be kept in mind, and the patient should be well informed before the procedure. This case report is per CARE guidelines.[3] The ethical committee of Pakistan Kidney and Liver Institute and Research Center exempted the review board for this study but consent was taken from the patient.

CASE REPORT

A 70-year-old man presented to the Interventional Radiology Outpatient Clinic after being referred by a pulmonologist in private practice with the problem of two episodes of hemoptysis in the last one and a half months. The patient previously was a known hypertensive with end-stage renal disease, on maintenance haemodialysis twice per week for the previous 2 years. He was a known smoker and had no other comorbid diseases. The workup for pulmonary tuberculosis had been done before presentation and was negative. The patient had undergone a bronchoscopy in a private set-up for haemoptysis after he had been referred for BAE with a presumptive diagnosis of bronchiectasis in bilateral upper lobes following a lower respiratory tract infection and was being managed on oral antibiotics and tranexamic acid with no clinical improvement. Preoperative clinical examination was significant for right upper lobe coarse crepitations and bibasilar fine crepitations only.

The patient underwent CT aortogram and CT chest and abdomen in the arterial phase as part of the preoperative assessment for BAE. CT findings revealed a dense consolidation

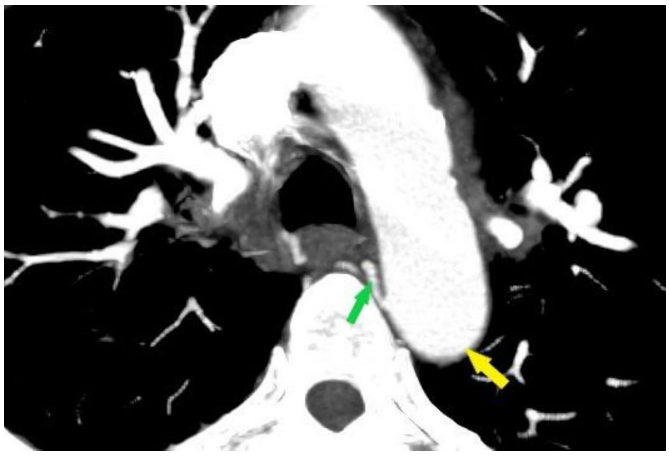

Figure 1 Preprocedural CT aortogram showing the origin of the right bronchial artery (green arrow) from the thoracic aorta (yellow arrow).

patch with perilesional ground glass infective nodular infiltrates, atelectatic bands in the right apex and fibrotic bands in the bilateral apical regions, right middle lobe and lingular segment of the left lung. The right bronchial artery had a separate origin from the thoracic aorta, with a diameter of 3.2 mm (figure 1). There was no common intercostobrachial trunk on the right side. The right bronchial artery was directly arterialising the consolidated area in the right apex, and some of the small branches were also seen coming from the subclavian artery with evidence of right apical bronchopulmonary shunting. Left bronchial arteries were of normal calibre and had a separate origin from the left anterolateral aspect of the thoracic aorta, just inferior to the arch of the aorta, having a diameter of 1 mm. Specks of atherosclerotic calcification were seen in the arch of the aorta, abdominal aorta and bilateral common iliac arteries. Other notable CT findings included bilateral small renal arteries with right distal renal artery aneurysms, cholelithiasis and CBD (common bile duct) stones.

BAE was performed under fluoroscopic guidance after puncturing and cannulating the right common femoral artery. The catheter-guidewire combination was advanced up to the right bronchial artery, and an angiogram showed diffuse vascularity towards the apex; however, there was no evidence to suggest that any radicular artery or the anterior spinal artery was arising from the right bronchial artery. Embospheres and microcoils were used to embolise the branches of the right bronchial artery. Subsequently, the right second intercostal artery was engaged, which showed an arteriopulmonary fistula. Embosphere (100–300 µm) was also used to embolise the branch of the second intercostal artery. An angiogram through the right subclavian artery did not show any supply.

Postprocedure angiogram showed adequate stasis. The patient developed sudden onset bilateral lower limb jerky movements during the procedure, followed by right lower limb weakness. After 6 hours, the patient gradually developed left lower limb weakness as well.

PHYSICAL EXAMINATION

Physical examination showed afebrile, vitally stable male GCS 15/15, with intact cranial nerves. Fundus examination only revealed grade 2 hypertensive retinopathy with no papilloedema. The cerebellar examination was normal. Neurological examination of the upper limbs showed power 5/5 in both limbs, with intact sensory system and reflexes +3 (Hoffman sign positive). A note was made of an AV(arteriovenous) fistula on the left arm. The lower limb motor examination was notable for the power of 0/5 on the right and 1/5 on the left sides. Bilateral lower limb reflexes were 2+ with upgoing plantars. Sensory examination showed decreased pain and temperature sensation bilaterally up to T5–T6 dermatomal level, while proprioception and vibration were intact bilaterally. There was loss of sphincter control with urinary retention and constipation. The localised spine exam showed no tenderness, and signs of meningeal irritation were negative.

INVESTIGATIONS

MRI of the brain without contrast and lower cervical and upper dorsal spine with intravenous contrast was performed, followed by haemodialysis. MRI brain showed no evidence of acute brain infarct, whereas, in MRI of the spine, T2-weighted images demonstrated high signal area in anterolateral spinal cord segment T4–T6, with subtle diffusion restriction on diffusion-weighted imaging and restricted diffusion on axial apparent diffusion coefficient, suggestive of an ischaemic event involving anterolateral spinal cord segment T4–T6 (figures 2 and 3).

Following imaging, a diagnosis of ASAS post BAE was made.

DIFFERENTIAL DIAGNOSIS

A diagnosis of ASAS post BAE was made based on clinical presentation, examination and relevant investigations.

TREATMENT

The patient was started on corticosteroids, which were given for 2 weeks, pregabalin 50 mg at night, and dual antiplatelets (aspirin 75 mg and clopidogrel 75 mg) one time a day. A bowel regimen with lactulose was started, the patient was catheterised and regular physiotherapy with nursing care was instituted. Despite these measures, the patient failed to report any improvement in motor power and sphincter control. He was eventually discharged home on a single antiplatelet (aspirin 75 mg one time a day) and regular physiotherapy. Corticosteroids were continued for 2 weeks and then tapered off.

OUTCOME AND FOLLOW-UP

Follow-up of the patient at 3 and 6 months failed to show any significant improvement in neurological function,

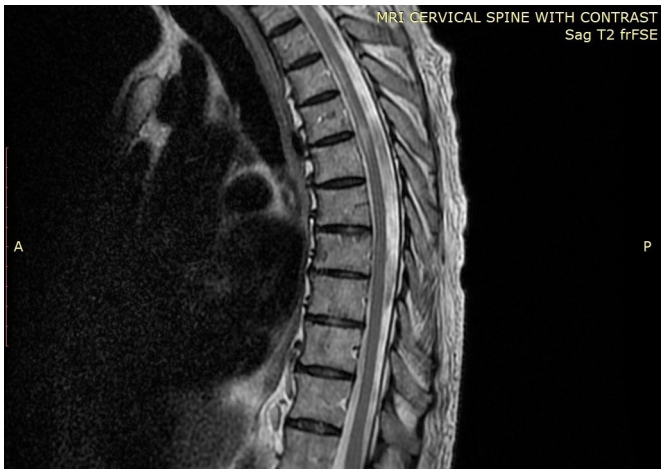

Figure 2 Sagittal T2-weighted Image showing hyperintense signal in T4–T6 region of the anterior spinal cord.

and he also developed complications due to long-term disability in the form of bed sores and depression. However, the patient did not have any problems of major haemoptysis afterward.

DISCUSSION

Massive haemoptysis is often a consequence of chronic inflammatory lung conditions, which lead to lung tissue destruction and local hypoxia. This triggers the recruitment of angiogenic growth factors and hypertrophy and proliferation of blood vessels, which can rupture because of friable walls and vascular inflammation, leading to the expectoration of large amounts of blood.[1] BAE is currently the established first-line treatment for massive haemoptysis and is generally a safe procedure with high efficacy.[1 2]

Spinal cord ischaemia or infarction as a complication of BAE has been sparsely reported, with a prevalence ranging from 1.4% to 6.5%.[4] Nevertheless, this is one of the most severe complications of BAE, which can lead to lifelong neurological morbidity, as reported in our case. While iatrogenic complications can occur with every procedure, the main risk factor compounding spinal cord infarction is the presence of a radiculomedullary artery at the thoracic level, which follows the trajectory of a corresponding spinal nerve till it reaches the anterior spinal artery.[5] This artery can rarely originate directly from the bronchial artery in 5% of the cases or more commonly originate from the intercostobrachial trunk or an intercostal artery; visualisation of this artery is important especially when it arises directly from the bronchial artery, as the high risk for embolisation would then be an absolute contraindication for BAE.[6]

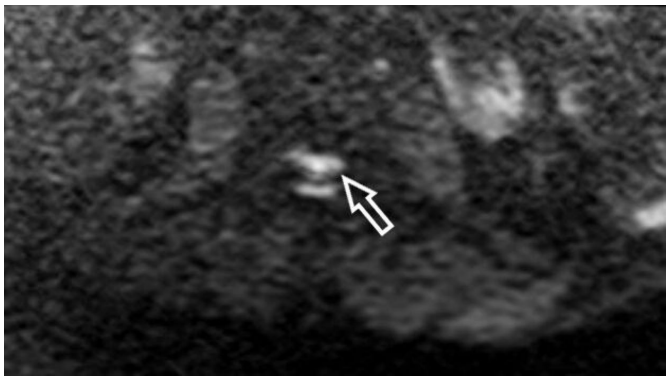

Figure 3 Diffusion-weighted imaging (DWI) at T4 level showing diffusion restriction (hyperintense signal).

According to existing evidence, the anterior spinal artery and radicular arteries typically do not arise from the bronchial artery alone. Still, they are more commonly associated with the intercostal artery alone or the intercostobronchial trunk involving the intercostal artery. This is consistent with the results of Brown and Ray.[1] Therefore, it seems likely that the spinal cord infarction in this case was related to the embolisation of the right second intercostal artery rather than the right bronchial artery. It has been hypothesised that haemodynamic changes during BAE can eventually occlude the distal outflow vessels, with resultant redirection of flow to the undetected medullary artery/anterior spinal artery—this could have been a possible cause of ASA embolisation in our patient.[1] The size of embosphere is important in this regard as well, and generally, embolisation particles larger than 350 µm in size are recommended as they are too large to penetrate the smaller end vessels that supply the cord.

The use of microcoils during BAE has classically been linked with a lower risk for spinal cord infarction. This can be explained if we consider the two main hypotheses for spinal cord infarction during BAE: the first one theorises backward reflux of embolic agents to proximal vessels such as the aorta, while the second one takes into consideration unintended embolisation of spinal arteries arising from distal part of target vessels. With microcoils, the potential for reflux is much lower due to their controlled embolisation nature, and they also embolise at the intended target instead of travelling further distally, as can be the case with other embolic agents. Only 1 out of 1577 patients had a spinal cord infarction when using coils, with significant differences compared with other embolic materials. The prevalence of spinal cord infarction after BAE with coils, GS particles and NBCA was 0.06% (1 of 1577 patients), 0.18% (12 of 6561 patients) and 0.71% (3 of 425 patients), respectively (p =0.04).[7]

Detection of spinal cord infarction can be tricky, as not all patients may present with weakness immediately after the procedure. The evolution of neurological symptoms can take time, and most reported cases had the onset of neurological symptoms on the first postoperative day.[6 8] Suspected spinal cord infarction needs to be investigated thoroughly to rule out other causes, such as cerebral stroke, and an MRI of the brain and spine needs to be carried out. In our patient, an unusual point was the rapidity with which symptoms developed—within 6 hours, the patient developed paraplegia, and while his MRI brain with contrast was normal, the MRI spine suggested ischaemia involving

Table 1 Different parameters of cases reported in the literature

Study	Indication of procedure	Symptoms of spinal cord syndrome	MRI findings	Treatment	Outcome
Brown *et al*, 2012[1]	Hemoptysis due to aspergilloma	Acute lower extremity weakness, skin temperature changes	Abnormal T2 signal at T1–T5 levels	Rehabilitation	Partial regain in function but died due to respiratory failure
Maramattom *et al*, 2016[6]	Massive hemoptysis	Left lower limb weakness and loss of sensation	Hyperintensity in the sagittal T2 image at D5–D6 levels	Intravenous steroids	Partial recovery
Chan *et al*, 2018[2]	Recurrent hemoptysis due to bronchiectasis	Left-sided weakness and numbness on belly and lower limb, decrease pain sensation in lower limb	T2/STIR hyperintensity within anterior spinal cord T6–T8	Rehabilitation	Good recovery
Balasubramanian *et al*, 2019[8]	Massive hemoptysis after taking biopsy from multiple lung masses	Flaccid paralysis of legs, sensory level at umbilicus	Complete paraplegia, impaired pinprick and light touch sensation below T4–T5 level	Supportive management (blood pressure control and rehabilitation)	Not reported
Gupta *et al*, 2019[12]	Hemoptysis due to pulmonary Koch's infection	Right-sided weakness and power on lower limb with autonomic dysfunction	T2 hyperintensity, extending from D1–D4	Steroids	Partial recovery, then lost to follow-up
Padgett *et al*, 2019[13]	Malignant hemoptysis from lung adenocarcinoma	Flaccid paralysis of legs, sensory level at umbilicus	Cord signal abnormality C5–C6 to T4–T5	Intravenous steroids, lumbar drain, blood pressure control	Persistent lower extremity paralysis
Torres *et al*, 2021[5]	Occasional hemoptysis due to pseudonodular lung lesion	Hypotonia of right lower limb	T2/STIR hyperintensity within anterior spinal cord T2–T7	Rehabilitation	Partial recovery
Walsh *et al*, 2023[14]	Massive hemoptysis due to anticoagulation d/t atrial fibrillation	Left lower limb weakness	Spinal infarct in midthoracic cord	Rehabilitation	Discharged to acute inpatient rehabilitation

anterolateral spinal cord segments T4–T6. Given that the patient had a history of ESRD (End stage renal disease) on haemodialysis, the possibility of a thromboembolic phenomenon causing his spinal cord infarction independent of the BAE could not be ruled out. Table 1 shows indications of BAE, clinical features, MRI findings, treatment given and outcomes of different reported studies.

Currently, no evidence-based treatment guidelines for spinal cord infarction post BAE exist, and it is generally treated as stroke with the addition of corticosteroids.[5] Isolated cases treated with fibrinolysis have been reported,[9] but in our patient involvement of the neurology team, arranging scans were done on the first postoperative day, and thus, conservative management was preferred. It must be noted that promptness in treating the underlying cause is the most important prognostic factor for anterior spinal cord syndrome[10]—risk factors for poor prognosis present in our patient included old age, initial severity and lack of improvement in the first 24 hours.[5] While up to 46% of patients can recover functional status and walk independently,[11] our patient did not unfortunately improve and is still undergoing rehabilitation and supportive care.

CONCLUSION

Despite BAE being a relatively safe procedure with beneficial good long-term outcomes as compared with its previous predecessors, the risk of spinal cord ischaemia can be a rare and disabling complication following the procedure. To some extent, spinal infarction can be preventable through knowledge about the anatomy of bronchial arteries and spinal arterial supply, angiography and investigations before embolization. But serious complication can happen following embolization despite taking all the necessary precautions. All patients undergoing emboliation should be well informed and counselled regarding this complication before undergoing such procedures.

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