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Percutaneous Embolization for Ruptured Ectopic Bronchial Artery Aneurysm

A Case Report

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Abstract: Bronchial artery aneurysm (BAA) is an uncommon disease, and spontaneous rupture of an ectopic BAA can be difficult for diagnosis and life-threatening. This case study describes a 52-year-old man who presented with acute onset of right chest pain, mild tachycardia, and hypertension. The initial diagnosis of acute myocardial infarction was made, and the patient was given nitroglycerin prior to admission to our hospital. However, the patient's symptoms deteriorated. An enhanced computed tomography scan revealed a ruptured 25-mm diameter mediastinal aneurysm under the tracheal bifurcation when he was admitted to our hospital. Bronchial arteriography further demonstrated a ruptured mediastinal BAA of a bronchial artery originated from the left subclavian artery, supplying the right lobe. Transcatheter artery embolization with polyvinyl alcohol particles and microcoils was performed successfully. The patient's symptoms were gradually relieved, and without recurrence on 1 year follow-up.

This case highlights the rare variation of mediastinal BAA and the role of interventional radiology in diagnosing and treating this critical condition.

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Abbreviations: BAA = bronchial artery aneurysm, BAG = bronchial arteriography, CT = computed tomography, PVA = polyvinyl alcohol.

INTRODUCTION

Bronchial artery aneurysm (BAA) is a rare but potentially lethal condition, and is reported less than 1% of all patients underwent selective bronchial arteriography (BAG).[1–3] The clinical presentation of a BAA, which depends on its size, location, and the presence of concomitant disease, can range from an incidental imaging finding to hemoptysis and shock caused by aneurysm rupture.[1,4,5] Correct diagnosis of this condition may be delayed because of its rarity and nonspecific presentation. In addition, it can be easily misdiagnosed as cardiovascular diseases, which accounts for approximately 30% of all death.[6] In this case study, we described a ruptured mediastinal BAA fed by an ectopic bronchial artery, which was originated from the contralateral subclavian artery. To our knowledge, this condition has not previously been reported in English literature. Transcatheter artery embolization (TAE) was successfully performed immediately after the confirmation of this condition with enhanced computed tomography (CT) scan and BAG.

CASE REPORT

A previously healthy 52-year-old man was referred to our hospital on August 2012 with a 24-hour history of right chest pain. He was diagnosed as myocardial infarction prior to admission to our hospital, based on acute chest pain, mild tachycardia, and hypertension. Nitroglycerin was given to the patient, however, the symptoms was deteriorated.

A physical examination revealed absent breath sounds in right lower lung, tachycardia (106 beats/minute) and hypertension (147/92 mmHg) when he was admitted to our hospital. Blood tests revealed normal biochemical and hematological parameters. Chest radiograph showed widening of the mediastinum and a large right pleural effusion (Figure 1). CT scanning of the chest showed a high-attenuation fluid collection at the mediastinum and right pleural cavity, suggesting mediastinal hematoma and right hemothorax (Figure 2A). Contrast-enhanced CT demonstrated a round, contrast-enhancing mass measuring 25 mm in diameter under the tracheal bifurcation, suggesting an aneurysm (Figure 2B). Initial aortic arch angiography showed an aberrant vessel arising from the left subclavian artery, and selective cannulation of this artery turned to be actually a right bronchial artery, which is a rare anomaly (Figure 3A, B). There was an aneurysm at the distal trunk of this ectopic right bronchial artery (Figure 3A, B). The imaging findings and clinical symptoms suggested that the aneurysm had ruptured, supporting the need for endovascular treatment.

With the co-axial technique, a 2.7-F Progreat catheter (Terumo, Leuven, Belgium) was advanced through the 5-F Cobra catheter (Terumo, Leuven, Belgium) into the main trunk of the right bronchial artery. Because of the tortuosity of the artery, the tip of the microwire could not be placed in the distal neck of the aneurysm. The decision was made to embolize the entire artery along with the aneurysm using polyvinyl alcohol (PVA) particles (Alicon, Hangzhou, China) (350–560 μm). The distal trunk of the bronchial artery and the aneurysm were occluded after embolization with 1 bottle of PVA (Figure 3C), then 2 microcoils (Cook, Bloomington, IN) (2 mm × 6 cm and 4 mm × 8 cm) were deployed above the proximal neck of the aneurysm to prevent recanalization (Figure 3D). A final BAG

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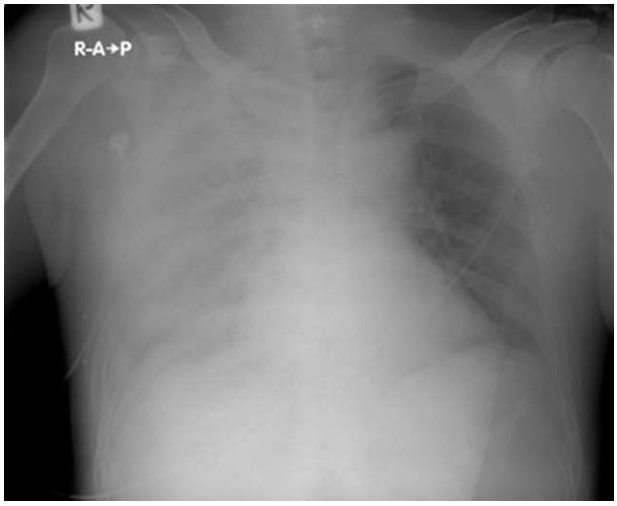

FIGURE 1. Chest radiograph showed widened mediastinum and large right pleural effusion.

demonstrated cessation of flow within the bronchial artery without refilling of the aneurysm (Figure 3D). The patient's symptoms were gradually relieved after procedure and pleural drainage, and a follow-up contrast-enhanced CT scanning of the chest at day 14 (Figure 4) showed that the aneurysm was no longer present, with nearly complete resolution of the hemomediastinum and hemothorax. The patient was followed up for 1 year by phone call, with no recurrence of symptoms.

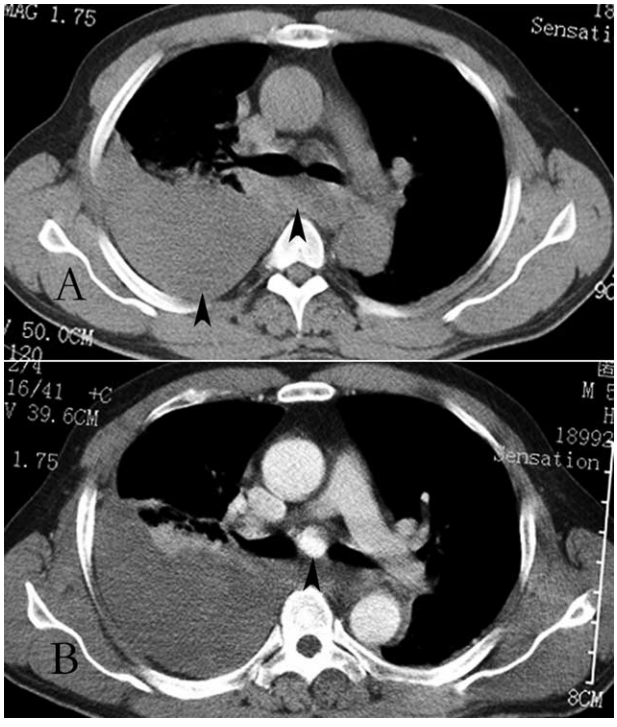

FIGURE 2. (A) Computed tomography (CT) scanning of the chest without IV contrast demonstrated high-attenuation fluid (arrowhead) at the mediastinum and right pleural space. (B) Contrast-enhanced CT scanning demonstrated a round, enhanced mass (arrowhead) under the tracheal bifurcation, measuring approximately 25 mm in diameter.

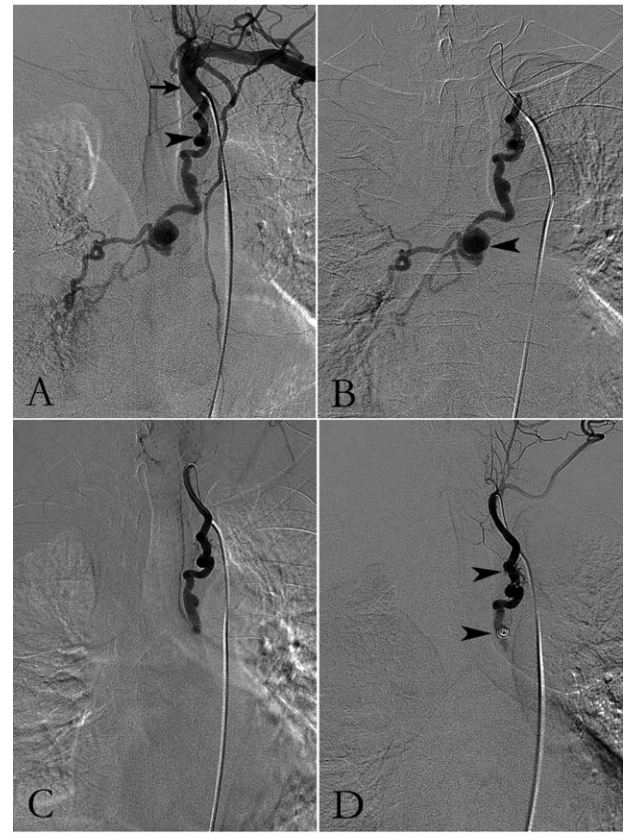

FIGURE 3. (A, B) Selective arteriography demonstrated the right bronchial artery (arrowhead) arising from the left subclavian artery (arrow). There is an aneurysm (arrow) at the distal trunk of the ectopic right bronchial artery. (C) Arteriography after embolization with polyvinyl alcohol (PVA) confirmed cessation of flow to the bronchial artery; the aneurysm is not visualized. (D) Two microcoils (arrowhead) were deployed above the proximal neck of the aneurysm to prevent recanalization.

DISCUSSION

BAAs are observed in less than 1% of all cases of selective BAG.[2] The incidence of a bronchial artery arising from the left subclavian artery is reported to range from 0 to 2.4%.[1,7] To our knowledge, a ruptured aneurysm of an ectopic right bronchial artery arising from the left subclavian artery has not previously been reported in the English literature.

BAAs are classified as intrapulmonary or mediastinal according to their anatomic location. A mediastinal BAA, which is even less common than an intrapulmonary BAA, can present as an incidental finding without symptoms or can present with symptoms related to extrinsic compression from the mass. Once a mediastinal BAA ruptured, the clinical presentation is acute and life-threatening, with severe chest pain and dyspnea the most common symptoms.[8] In this case, it can be easily misdiagnosed as cardiovascular diseases. For our case, the initial misdiagnosis was mainly due to the clinical presentations of the patient, including acute chest pain and hypertension, which are also the most common symptom and risk factor among patients with initial myocardial infarction.[9] Dysphagia and hematemesis may also occur as a result of esophageal erosion.[10,11] On imaging, hemothorax and hemomediastinum are the most common findings;[12] acute superior vena cava

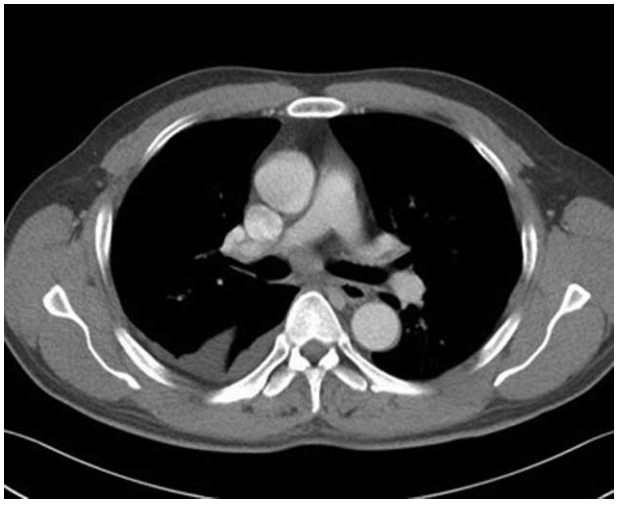

FIGURE 4. Follow-up contrast-enhanced computed tomography (CT) scanning of the chest at day 14 showed the aneurysm was no longer present, with nearly complete resolution of the hemomediastinum and hemothorax.

obstruction may also be seen.[13] CT angiography is the primary noninvasive diagnostic modality for BAAs. However, conventional angiography can be performed for both diagnostic and treatment purposes.[2]

Medical therapy has a limited role in treating this life-threatening condition. Surgical ligation and resection can reliably eliminate BAAs; however, because of the high risk and costs associated with this procedure, endovascular therapy should be attempted first. Surgery is also unsuitable for patients who cannot tolerate a thoracotomy.

With trans-catheter artery embolization, both the afferent and efferent arteries of the BAA need to be occluded to prevent retrograde filling of the aneurysm.[2,11,12] A variety of embolic materials has been used successfully for this purpose. Gelfoam, which is generally accepted as an absorbable embolic agent, is cost-effective and associated with a relatively low risk of tissue necrosis. However, the risk of recanalization of the aneurysm may be high when Gelfoam is used.[14] *N*-butyl cyanoacrylate has also been used to treat BAA,[15,16] but use of this agent has been associated with gluing of the catheter, tissue necrosis, and inadvertent embolization of the normal vessels secondary to uncontrolled reflux.[17] Coils are most commonly used to treat BAA. These permanent embolic agents can be positioned at the target site with fewer complications than other agents.[18,19] In our case, we reported a catheterization of the efferent branch was impossible because of the tortuosity of the ectopic right bronchial artery. Coils could not be deployed in the efferent branch, so PVA particles were chosen to embolize both the afferent and efferent vessels, including the aneurysm itself. PVA is a permanent embolic agent that has generally been used for bronchial artery embolization. Boushy et al[20] demonstrated occlusion of bronchial arteries with PVA without any clinically or radiologically detectable pulmonary infarction. In all cases, embolic materials that can pass through the bronchopulmonary anastomosis (325 μm in the human lung) must be avoided because of the possibility of paradoxical systemic infarction.[21] Complete embolization of the afferent artery with TAE alone is difficult when the segment between the aortic origin of the bronchial artery and the BAA is too short to directly embolize. In such cases, a combination of thoracic aortic stent-graft occlusion of inflow and coil embolization of outflow branches can be used.[22]

In our case, a follow-up contrast-enhanced CT scanning was only performed at day 14 after discharge, which is the main limitation. However, the patient was followed up by phone call for 1 year without recurrence of symptoms, which indicated the aneurysm was successfully embolized.

CONCLUSION

We reported a rare condition of ruptured BAA, which have not been described before. Correct diagnosis of this condition can be obtained by CT angiography and BAG. Endovascular treatment is safe and effective for this condition. In the case of severe tortuous bronchial artery described here, the combination of PVA particles and coils achieved an excellent therapeutic result.

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