Case Report

Ruptured Bronchial Artery Aneurysm Associated with Bronchiectasis: A Case Report

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A massive hemothorax from a ruptured bronchial artery aneurysm (BAA) is very rare. Only 12 cases of ruptured mediastinal BAA have been reported. This case study describes a 77-year-old female with bronchiectasis who presented with anemia, hypertension, hemothorax, and a mediastinal mass. A chest tube was inserted through which 2 liters of unclotted blood was drained from the left pleural cavity. An enhanced computed tomography scan revealed a ruptured 3-cm diameter mediastinal aneurysm of a bronchial artery supplying the left lower lobe. Transcatheter artery embolization (TAE) with multiple microcoils was performed successfully. Although the patient needed a transfusion, the subsequent course was uneventful. In the absence of trauma or other causes for hemothorax and mediastinal hemorrhage, the possibility of a BAA should be considered. TAE is the treatment method of choice as a minimally invasive strategy in patients with ruptured BAA. (Ann Thorac Cardiovasc Surg 2009; 15: 115–118)

Key words: bronchial artery aneurysm, ruptured, embolization, hemothorax

Introduction

A mediastinal bronchial artery aneurysm (BAA) is a rare condition; fewer than 40 cases have been reported in the literature. Once the bronchial artery ruptures, the clinical presentation is acute and life-threatening, with the most common symptoms being severe chest/back pain mimicking acute aortic dissection and symptoms of shock. To our knowledge, at least 12 ruptured BAAs have been reported in the English literature (Table 1).[1–12] We reviewed these cases in terms of their clinical presentation and therapeutic management.

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Case Report

A 77-year-old woman was hospitalized because of severe dyspnea and sudden chest pain with signs of severe anemia. She had tachycardia (120 beats per minute), hypertension (systolic blood pressure of 170 mmHg), and tachypnea (25 breaths per minute). Her hemoglobin level was 7.5 g/dl. There was no trauma, no sign of infection, and no hemoptysis. Her history included an episode of bronchiectasis (2 years previously) and chronic hepatitis C. Chest radiography showed left pleural effusion with slight shifting of the mediastinum to the right. A computed tomography (CT) scan showed left pleural effusion and bilateral lower lobe fibrosis. A chest tube was inserted through which 2 liters of unclotted blood was drained from the left pleural cavity. An enhanced CT scan revealed a mediastinal hemorrhage, a left hemothorax, and an aberrant enhanced lesion in the mediastinum 3 cm in diameter, which aroused suspicion of an aneurysm (Fig. 1). A 3-dimensional reformation of the CT angiogram revealed an aneurysm of an aberrant artery arising from the descending aorta (Fig. 2).

Table 1. Published cases of ruptured mediastinal bronchial artery aneurysm in the English literature

Author	Year	Age	Gender	Cause	Size (mm)	Region	Symptoms	Treatment	Outcome
Hall et al.[1]	1977	56	M	Bronchiectasis	30	Pleural cavity	Chest pain/shock	Surgery	Successful
Osada et al.[2]	1986	56	F	Unknown	12	Lung	Hemoptysis/shock	TAE + Surgery	Successful
Shaer et al.[3]	1989	79	M	Atherosclerosis	10	Esophagus	Hematemesis	None	Died
Cearlock et al.[4]	1995	22	M	Traumatic	15	Lung	Hemoptysis	TAE	Successful
Ishizaki et al.[5]	1995	25	F	Osler-Weber-Rendu	5	Pleural cavity	Chest pain/shock	TAE + Surgery	Successful
Hoffmann et al.[6]	1996	70	M	Atherosclerosis	33	Mediastinum	Shock	TAE + Surgery	Successful
Kalangos et al.[7]	1997	50	M	Degenerative	15	Pleural cavity	Back pain/shock	Surgery	Successful
Pugnale et al.[8]	2001	72	M	Unknown	15	Mediastinum	Epigastric pain	TAE	Successful
Suen et al.[9]	2003	50	F	Sarcoidosis	8	Pleural cavity	Back pain	Surgery	Successful
Fukunaga et al.[10]	2003	60	M	Unknown	40	Esophagus	Chest pain	TAE	Re-TAE
Chatterjee et al.[11]	2004	59	M	Unknown	NA	Pleural cavity	Chest pain	TAE	Successful
Karmy-Jones et al.[12]	2005	78	F	Tuberculosis	16	Pleural cavity	Myalgia	TAE	Successful
Present case	2009	77	F	Bronchiectasis	30	Pleural cavity	Chest pain	TAE	Successful

M, male; F, female; NA, not available; TAE, transcatheter arterial embolization; Re-TAE, repeat transcatheter arterial embolization.

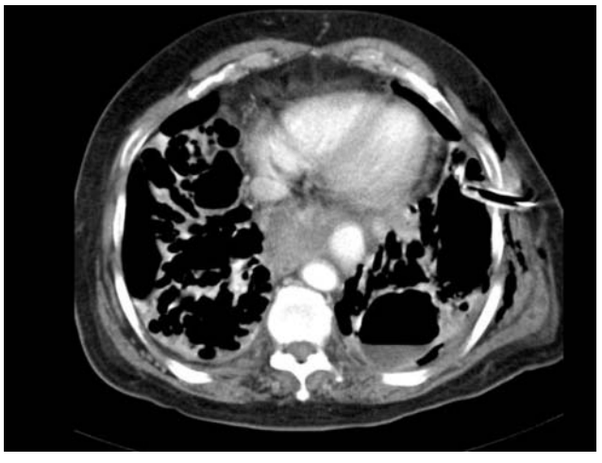

Fig. 1. Enhanced chest computed tomography revealed a low-density, partially enhanced mass anterior to the descending aorta.

We performed angiography to assess this malformation and find the feeding vessel. The angiogram revealed an aneurysm arising from the bronchial artery supplying the left lower lobe (Figs. 3A and 3C). A leakage of contrast medium proved bleeding from this aneurysm into the mediastinum and left pleural cavity. The aneurysm was immediately occluded with 2 interlocking detachable coils (Boston Scientific Corp., Watertown, MA) and 3 tornado coils (Cook Medical Inc., Bloomington, IN) (Figs. 3B and 3C), and there was no further contrast effusion from it. The angiogram also revealed bronchial artery dilatation and 5-mm diameter aberrant arteries arising from the bronchial artery and inferior phrenic artery to the bilateral lower lobes.

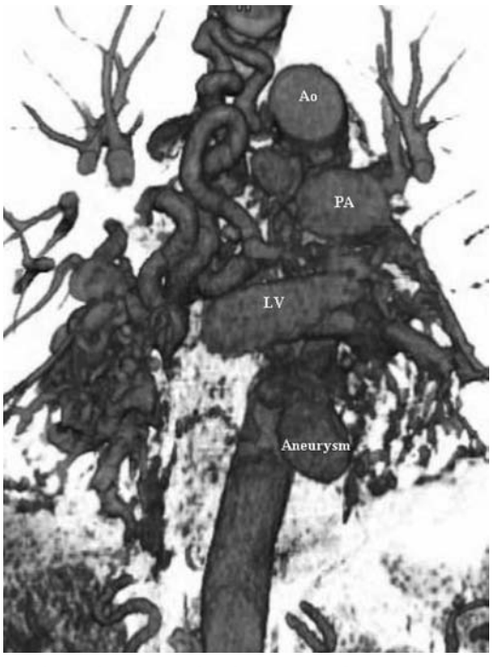

Fig. 2. Three-dimensional reformation of CT angiogram with volume-rendering technique revealed an aneurysm of a bronchial artery arising from the aorta.
Ao, aorta; PA, pulmonary artery; LV, left ventricle.

Because of her blood loss, the patient needed a transfusion (four units of packed red cells). The remaining course was uneventful; the drainage tube was removed on day 5, and she left the hospital on day 14. Nine months after transcatheter artery embolization (TAE), she was asymptomatic, and a chest CT revealed no sign of bleeding.

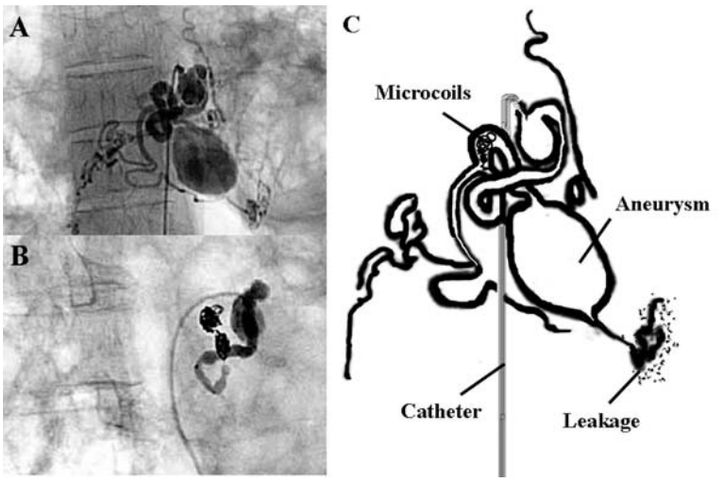

Fig. 3. Bronchial arteriography before (A) and after (B) transcatheter arterial embolization. A leakage from the aneurysm into the mediastinum and left pleural cavity was shown in schema (C). The aneurysm was successfully embolized with microcoils.

Discussion

Acute hemomediastinum and hemothorax are usually related to chest trauma, rupture of a thoracic aortic aneurysm, or aortic dissection. The causes of spontaneous mediastinal hemorrhage have been subdivided into four categories: (i) complication of enlarging mediastinal masses, (ii) transient increase in intrathoracic pressure, (iii) sudden sustained hypertension, and (iv) altered hemostasis.[8,13] A ruptured BAA is rarely the etiology of mediastinal hemorrhage.

BAA is a rare entity that is observed in less than 1% of all cases of selective bronchial arteriography.[14] BAA may present as asymptomatic mass, but it is more commonly diagnosed after complications occur.[12] It has been reported that BAA can be congenital, in the context of pulmonary sequestration or pulmonary agenesis, or acquired as a result of atherosclerosis,[3,6] inflammatory lung disease such as bronchiectasis,[1] systemic disease,[5,9] or trauma.[4] Most cases of ruptured BAA have chest/back pain and hemothorax resulting from rupture into the pleural cavity and mediastinum. There may also be epigastric pain, hematemesis, and hemoptysis resulting from rupture into the esophagus[3,10] and pulmonary parenchyma.[2,4] Although little is known about the process that leads to BAA rupture, the diameter of the aneurysm (5 to 40 mm) was not an incremental risk factor in previous cases. Detailed reviews[2,7,12,15] report that BAAs as large as 8–10 cm in diameter were occasionally detected as an incidental finding on radiological examination. A BAA should be treated whether it is symptomatic or not.

In patients with symptoms of chest pain, mediastinal hemorrhage, and hemothorax, enhanced CT should be performed to detect the bleeding point before treatment. In a hemodynamically stable patient like this one, the definite diagnosis must be confirmed by selective bronchial artery angiography, which enables concurrent therapeutic embolization.[8,12] In previous reports, TAE was performed in 9 cases (69%), and most of them were treated successfully by TAE alone. TAE to occlude the afferent and efferent arteries of the BAA is considered first-line management if the patient is stable. In this case, catheterization of the efferent branches was impossible because of the tortuosity of the left bronchial artery. Surgery should be reserved for patients with a contraindication to embolization, such as an allergy to iodinated contrast medium or a medullary artery.[11] The advantages and disadvantages of surgery and TAE should be recognized, and the appropriate procedure should be selected based on the patient's clinical status.

Conclusion

Massive hemothorax from a ruptured BAA caused by bronchiectasis is very rare. However, in the absence of trauma or other causes for hemothorax, its possibility should be considered. TAE is the treatment of choice as a minimally invasive strategy for patients with a ruptured BAA.

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