We present a surgical case of bronchial artery aneurysm (BAA) connecting pulmonary artery accompanied with racemose hemangioma. This is a third surgical case report of BAA directly connecting pulmonary artery in the English literature. A 63-year-old female was found a BAA, 2 cm in diameter, connecting right A4 pulmonary artery. The patient underwent two attempts for embolization. However, due to extensive collaterals, there was persistent flow in the aneurysm. Standard lateral thoracotomy was performed. A BAA was located between A4 and A5 PA. A small branch of A4 PA was separated, and the small vessel connecting to the BAA could be ligated. A5 PA was separated similarly, however BAA was ruptured not to identify the other small vessel connecting to the BAA. After a clamp of the BAA, middle lobe lobectomy was performed. We removed the aneurysm with dilated bronchial artery connecting to the aneurysm. The postoperative course was uneventful.

Keywords: bronchial artery aneurysm, connection to the pulmonary artery, racemose hemangioma

Introduction

Bronchial artery aneurysm (BAA) is rare, but they are potentially life threatening and require treatment to avoid rupture and other complications.

We present a surgical case of bronchial artery aneurysm connecting directly right pulmonary artery.

To our knowledge, a few cases of bronchial artery aneurysm connecting pulmonary artery has been reported in Japanese literature,\(^1\)\(^2\) and only three in English literature.\(^3\)\(^-\)\(^5\) This is a third surgical case report of bronchial artery aneurysm directly connecting pulmonary artery in the English literature.

Case Report

A 63-year-old female was found to have an abnormal shadow in a regular checkup chest X-ray film in 2005. She had no symptoms such as bloody sputum, hemoptysis or cardiac failure. Chest computed tomography (CT) scan revealed a round mass adjacent to the pulmonary artery in the right hilum, and bronchial artery dilated like vine; so called racemose hemangioma. Aortography, bronchial arteriography and pulmonary arteriography showed a bronchial artery aneurysm, 2 cm in diameter, connecting right pulmonary artery. The patient underwent two attempts for embolization at another hospital in 2006 and 2007. However, due to extensive collaterals, there was persistent flow in the aneurysm. Aortography revealed a submucosal elevated lesion in the right middle bronchus (Fig. 1). Three dimensional computed tomography (3-D CT) demonstrated a narrow vessel which is connected aneurysm and right A4 pulmonary artery (Fig. 2).

Standard lateral thoracotomy was performed via the fifth. A BAA 20 mm in diameter with thrill was located between...
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A4 and A5 pulmonary artery (Fig. 3). First, we ligated and cut winding dilated bronchial artery at the level of carina in the posterior mediastinum. However, the pulsation of the BAA did not diminish and a thrill was remained. Next, we dissected BAA from pulmonary artery carefully. A small branch of A4 PA was separated, and the small vessel connecting to the BAA could be ligated. A5 PA was separated similarly, however BAA was ruptured not to identify the other small vessel connecting to the BAA in taping of A5 PA with forceps. After a clamp of the BAA, middle lobe lobectomy was performed. We removed the aneurysm with dilated bronchial artery connecting to the aneurysm. We used autologous blood transfusion system during the operation. Intraoperative blood loss was 1000 ml without non-autologous transfusion.

The postoperative course was uneventful, and the patient was discharged on the seventh postoperative day.

The 35 days postoperative chest CT scans showed no evidence of BAA.

Discussion

Bronchial artery aneurysm (BAA) is rare, detected in fewer than 1% of all patients who undergo selective bronchial arteriography.6)

Bronchial artery aneurysm (BAA) can be classified according to the location as either mediastinal or intrapulmonary.7)

The chief complaints for mediastinal BAA is relative to compression or rupture into contiguous structures. Intrapulmonary BAA presents with hemoptysis, which is the most frequent symptom. BAA in this case could not be classified either type because of hilar lesion with compression of right middle bronchus.

It was located in the horizontal fissure of the right lung, and directly connected to the pulmonary artery, and presented no symptoms.

BAA connecting directly to the proximal pulmonary artery is extremely rare. The bronchial artery arises mainly from thoracic aorta or its branches, and empties mainly into the pulmonary vein and a lesser bronchial vein system that enters the azygos venous system on the right.8)

Existence of small anastomosis between the bronchial and pulmonary artery in the peripheral area were known.9) We thought the increasing blood flow of the bronchial artery caused to generate the aberrant anastomosis in this case.

BAA in this case is not only anastomosed with pulmonary artery but also accompanied with racemose hemangioma. Racemose hemangioma of the bronchial arteries is a rare abnormality and is characterized by enlarged and convoluted bronchial arteries arranged segmentally along the longitudinal axis of bronchus.

Since the term “racemose hemangioma” is firstly reported by v Babo H, et al. in Germany in 1976,10) some
have detected and ligated the aberrant dilated artery in the anterior side of the right main bronchus as possible as we can. Second, we could not detect the second aberrant small vessel connecting between BAA and pulmonary artery. If we had dissected the BAA more carefully after the ligation of first aberrant artery, we might remove the BAA without rupture.

**Conclusion**

We presented a surgical case of bronchial artery aneurysm connecting directly right pulmonary artery. In dissection of aneurysm, it is important to reduce a blood flow of the aneurysm with the ligation of winding dilated bronchial artery.

**Disclosure Statements**

There are no companies, etc. in a relation of conflict of interest requiring disclosure in relation to this manuscript.

**References**

10) v Babo H, Huzly A, Deininger HK, et al. [Angiomas and angioma-like changes of the bronchial arteries (author’s transl)]. Rofo 1976; 124: 103-10. (in German)