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Case Report

A Surgical Case of Bronchial Artery Aneurysm Connecting to a Pulmonary Artery and Vein Complicated by Racemose Hemangioma

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We report a surgical case of bronchial artery aneurysm (BAA) that directly connected to a pulmonary artery and a pulmonary vein through an abnormal vessel. It was complicated by racemose hemangioma. This is a rare vascular malformation. An 82-year-old female had a large BAA that was found incidentally. First, we consider treating the BAA with embolization by interventional radiology (IVR). However, because of strong meandering of the bronchial artery, we could not advance a microcatheter into the BAA. Therefore, a surgical operation was performed through a standard posterior lateral thoracotomy. The BAA was located between the upper and lower lobes and directly connected to the pulmonary artery. Some bronchial artery branches that provided inflow to the aneurysm were ligated, and the abnormal vessel that connected the BAA to the upper pulmonary vein was ligated easily. A fistula between the BAA and pulmonary artery was sutured by the cardiovascular surgeon using an artificial cardiopulmonary device, with permissive stenosis of A2b (ascending A2).

Keywords: bronchial artery aneurysm, vascular malformation, racemose hemangioma

Introduction

Although bronchial artery aneurysms (BAAs) may be rare, they are sometimes life threatening and require treatment to avoid rupture. Recently, several cases of BAA have been incidentally found by computed tomography (CT) performed for another purpose. In the past, a few cases of BAA that connected to a pulmonary artery have been reported in the literature.[1–3] However, there are no reports of BAA that connected to both a pulmonary artery and a vein.

We present a surgical case of BAA that directly connected to the right pulmonary artery and pulmonary vein through an abnormal vessel. It was also complicated by racemose hemangioma.

Case Report

An 82-year-old female came to our hospital because of dyspnea, and an abnormal shadow was found on her chest X-ray. Chest contrast CT showed multiple pulmonary artery emboli (**Fig. 1A**) and a highly enhanced nodule 28 mm in diameter at the right hilum, which suggested BAA. The diagnosis for dyspnea was acute pulmonary

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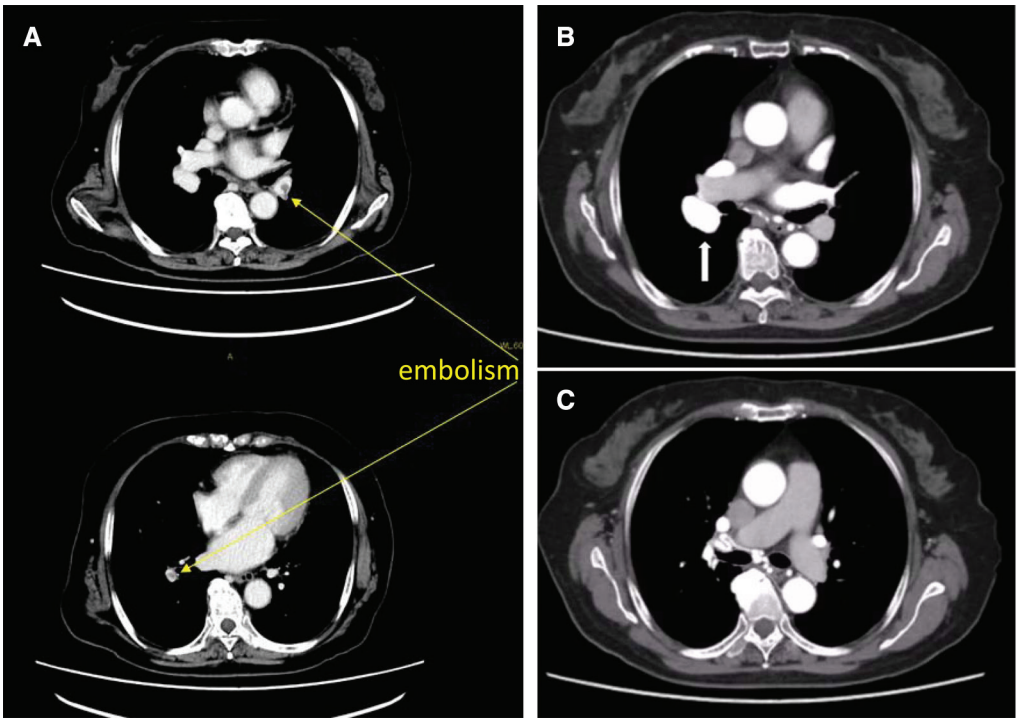

Fig. 1 (**A**) Chest contrast CT showed multiple pulmonary artery emboli that caused her dyspnea. (**B**) Contrast-enhanced CT showed a huge aneurysm 28 mm in diameter at the right hilum, and (**C**) there was racemose hemangioma with strong meandering. CT: computed tomography

artery embolism, and the right hilar nodule was an incidental finding. She was treated by anticoagulant drugs for embolism, and enhanced CT was performed again to obtain more detailed information on the nodule, especially during the pulmonary arterial phase. The second CT showed that the multiple pulmonary emboli had disappeared, and the highly enhanced nodule at the right hilum was diagnosed as BAA that was complicated racemose hemangioma with strong meandering (**Fig. 1B** and **1C**).

A few days later, bronchial arteriography was performed. As soon as contrast agent appeared in the aneurysm, it also appeared in the right pulmonary artery. Following that, CT angiography was performed. It showed that the aneurysm connected directly to the pulmonary artery, and the pulmonary vein connected to an abnormal drainage vein (**Fig. 2**). At first, bronchial arterial embolization by interventional radiology (IVR) was planned; however, the bronchial artery inflow to the aneurysm became racemose hemangioma with strong meandering, so we were not able to reach the aneurysm with a microcatheter. Because of the right-to-left shunt, embolization with n-butyl-2-cyanoacrylate (NBCA) and lipiodor was considered a high-risk procedure, since it might cause

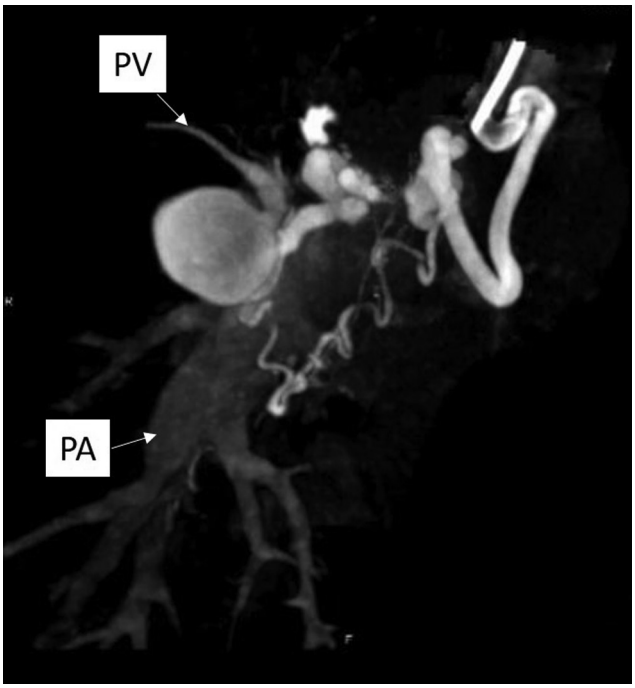

Fig. 2 CT angiography after bronchial arteriography demonstrated an aneurysm with racemose hemangioma located on the pulmonary artery, and the superior pulmonary vein was enhanced. CT: computed tomography

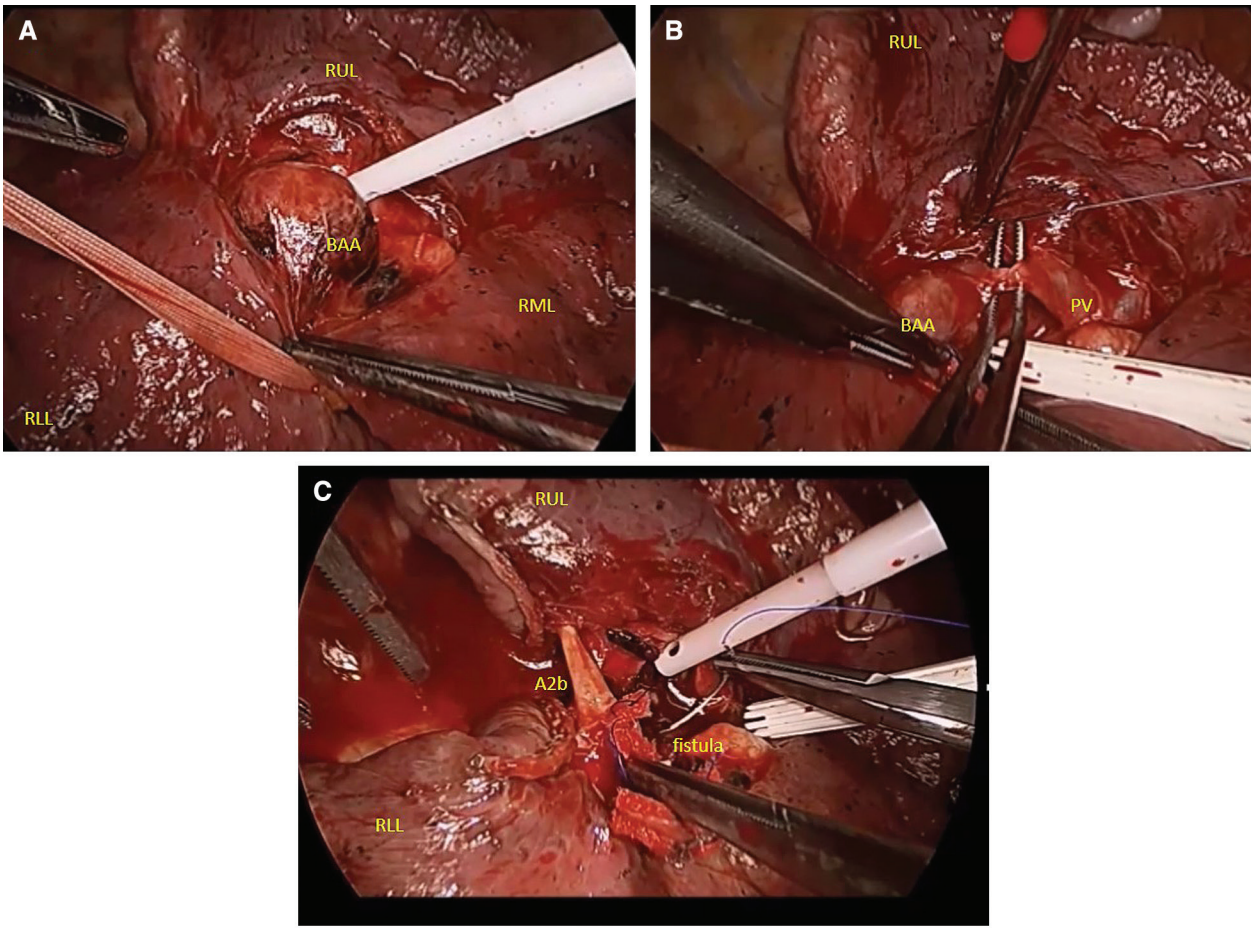

Fig. 3 There was a BAA between the upper and lower lobes (**A**). An abnormal vessel that connected the BAA to the pulmonary vein was found easily and ligated (**B**). The BAA connected directly to the pulmonary artery near the bifurcation of ascending A2 from the pulmonary arterial trunk. A fistula between the BAA and pulmonary artery was sutured (**C**). BAA: bronchial artery aneurysm

embolization of other organs. Furthermore, coil embolization of the BAA was thought to be too difficult from a remote vessel. Therefore, an operation was performed through a posterior lateral incision.

To decrease blood flow to the BAA, we planned to capture the superior and inferior pulmonary veins and the right main pulmonary artery. Moreover, preoperative thoracic endovascular aortic repair (TEVAR) was considered. However, CT revealed another small aneurysm under the bifurcation of the trachea, and we thought we would try IVR again for secondary treatment, so we did not perform TEVAR in this case. In fact, when we tried to secure the right main pulmonary artery, there was minor bleeding from the pulmonary artery. Although manual compression stopped the bleeding, we changed the operative strategy to the use of an artificial cardiopulmonary device by the cardiovascular surgeons. First, we approached the BAA from the interlobar pleura (**Fig. 3A**); however, the wall of the aneurysm was so vulnerable that the BAA was ruptured by our operative technique. After some branches of the bronchial artery that provided inflow to the aneurysm were ligated, we found that the aneurysm connected to V2 through an abnormal vessel (**Fig. 3B**), so we ligated it. Next, we ligated a main branch of the racemose hemangioma that provided inflow to the aneurysm. The BAA was directly connected to the pulmonary artery, so we tried to suture the fistula from the inside of the aneurysm; however, the bleeding was not controlled. To ensure the field of view, we resected the interlobar line between the upper and lower lobes, and then found the fistula located near the branch of A2b (ascending A2). Finally, we sutured the fistula with permissive stenosis of A2b (**Fig. 3C**).

The operation time was 288 minutes, and the cardiopulmonary support time was 74 minutes.

CT scanning 6 and 12 months after surgery showed disappearance of the aneurysm, and the racemose

hemangioma and small aneurysm had faded away. Furthermore, the general condition of the patient was good, so we followed the patient carefully in the outpatient clinic.

Discussion

Some cases have reported BAA connecting to the pulmonary artery, and it has been noticed that bronchial-pulmonary arterial fistula is often complicated by racemose hemangioma.[4–6] However, we could not find any reports of BAA connecting to both a pulmonary artery and a vein, so this is an extremely rare case. In this case, because of racemose hemangioma and connection of the BAA to the pulmonary vein, we believed that treatment by IVR might not be possible, so we performed an operation. The operative method for complex BAA has usually been lobectomy as well as resection of the aneurysm. Narato et al.[7] reported that lobectomy was performed most often for racemose hemangioma (in approximately 38% of cases). In this case, the BAA was directly connected near the branch of A2b. If we had to perform lung resection in addition to aneurysmectomy, we might not have been able to prevent pneumonectomy. Nevertheless, we succeeded in closing the fistula to the aneurysm without lung resection using an artificial cardiopulmonary device. The patient's clinical course after surgery has been going well for 1 year.

We believe that control of blood flow is most important for the surgical strategy to treat complex BAA. In this case, blood flow from the bronchial artery and pulmonary vein were controlled rather easily without securing the central side. However, it was very difficult to control blood flow from the pulmonary artery. In fact, the fistula between the BAA and pulmonary artery was not sufficiently visible to suture due to massive blood flow, so we used an artificial cardiopulmonary device. As a result, we were able to preserve flow in the pulmonary artery, except for A2b, without lung resection.

Generally, BAA is classified according to its location as either mediastinal or intrapulmonary.[8] However, considering our treatment, we suggest that it should be classified as simple type or complex type. Simple bronchial aneurysm is generally treated by IVR at present; if surgical resection is needed, the operation is completed mainly by video-assisted thoracic surgery (VATS). However, treatment for a complex aneurysm needs more careful management, especially the surgical technique. Various methods should be considered depending on the type of BAA. A complex aneurysm is often accompanied by racemose hemangioma. Racemose hemangioma is a rare malformation and characterized by not only a dilated and convoluted bronchial artery but also vascular hyperplasia causing abnormal connections to adjacent vessels.[9] It is also said that racemose hemangioma creates a fistula to the pulmonary vasculature. The incidence of a fistula from bronchial racemose hemangioma to pulmonary arteriovenous malformation has been estimated at approximately 4%.[10]

There are some case reports of complex types of BAA and interesting strategies for its treatment. For example, TEVAR is one of the options to control inflow from the main bronchial artery.[11,12] The combination of TEVAR and embolization by IVR is considered feasible; however, TEVAR was not performed in our case because of the issues stated above. Also, Kawai et al.[13] reported a case of a large pulmonary arteriovenous malformation and BAA. They performed microcoil embolization of the pulmonary arteriovenous malformation, and 2 years later they performed NBCA and lipiodol embolization of the BAA. Thus, the strategy for a rare complex type of BAA is usually selected on a case-by-case basis, and we suggest that the strategy should be discussed adequately by the surgeon and IVR. Furthermore, if an operation is needed for complex BAA accompanied by a fistula to the pulmonary artery, we recommend the use of extracorporeal circulation.

Conclusion

We presented a very rare surgical case of BAA that connected to both the right pulmonary artery and pulmonary vein through an abnormal vessel. It was also complicated by racemose hemangioma. Control of blood flow was quite important in this surgical case.

Disclosure Statement

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

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