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Case Report

Thoracoscopic Bronchial Artery Resection for Multiple Bronchial Artery Aneurysms

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We describe a 36-year-old asymptomatic female with multiple bronchial artery aneurysms (BAAs) and a bronchial artery (BA) to pulmonary artery (PA) fistula. She was treated with thoracoscopic BA resection without lobectomy in lieu of catheter embolization as first-line treatment. The configuration of the BA and the location of the BAAs were clearly visualized using three-dimensional computed tomography (3DCT); therefore, the segment of the BA to resect was assessed preoperatively and complete resection of all BAAs was performed. Preoperative BA angiography delineated the BA to PA fistula, and guided surgical decision-making.

Keywords: aneurysm, bronchial arteries, mediastinum, thoracoscopy

Introduction

Bronchial artery aneurysms (BAAs) are rare, and multiple BAAs are rarer still. BAAs should be treated immediately even when asymptomatic because they may cause massive hemoptysis or lead to rupture.[1,2)]

Transcatheter embolization has become the first-line treatment of BAAs; however, both treatment failure and complications can occur that require lobectomy with standard thoracotomy.[2–4)] Minimally invasive video-assisted thoracic surgery (VATS) is preferable to a thoracotomy, but there is a lack of evidence of its safety and efficacy for the treatment of multiple BAAs.

We describe the case of a patient with multiple asymptomatic BAAs that were safely resected via thoracoscopic bronchial artery (BA) resection using preoperative three-dimensional computed tomography (3DCT) and arterial angiography.

Case Report

A 36-year-old asymptomatic female presented to our hospital for an abnormal right lung field shadow detected on a chest X-ray that was obtained as part of an annual medical checkup. All laboratory data were normal. A chest CT scan and 3DCT showed at least five right BAAs, and BA angiography showed that the BA directly branched off of the aorta and connected to the right peripheral pulmonary artery (PA) (**Fig. 1a** and **1b**). After discussion in a multidisciplinary conference, we performed BA resection using VATS rather than catheter embolization. The reasons for this were as follows. First, the BA was tortuous and catheter selection for all afferent and efferent branches would have been difficult. Second, catheter-based embolization could potentially lead to a pulmonary embolism via the BA-PA fistula. Finally, the part of the BA that needed to be removed was

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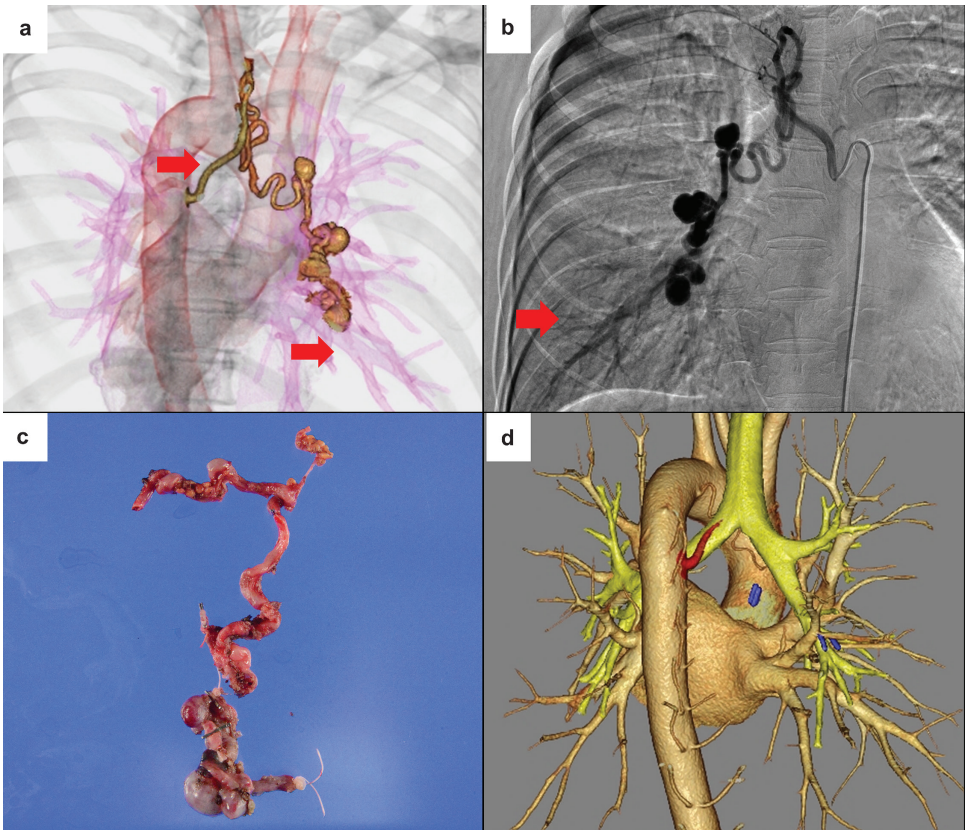

Fig. 1 (**a**) 3DCT shows the BA and associated aneurysms. The arrows show which portion we intended to ligate. (**b**) BA angiography shows a tortuous BA directly arising from the aorta, with connection to a peripheral PA (red arrow). (**c**) Gross specimen shows en bloc resection of the aneurysms. (**d**) 3DCT after surgery shows no recurrence of blood flow. 3DCT: three-dimensional computed tomography; BA: bronchial artery; PA: pulmonary artery

clearly identified preoperatively, and there were no inflammatory changes on CT; thus, BA resection without lobectomy seemed possible.

The patient was placed in the left lateral decubitus position and VATS was performed using four access ports. There were no adhesions in the pleural cavity. The BA ran between the right bronchus and the PA. The PA was separated and secured before starting resection of the BA to visualize the BA clearly and not to injure the PA. The afferent artery was detected adjacent to the azygos vein and was divided after ligation (**Fig. 2a**). The BA was separated along with the bronchus and the most distal BAA was identified at the dorsal part of the inferior pulmonary vein. The BA was ligated and divided distal to the aneurysm. (**Fig. 2b**). The BA was completely separated from PA, pleura, bronchus, and other connective tissues. All the branches along the BA between the most proximal and the most distal BAA were ligated and divided, and all aneurysms with the associated BA were completely resected (**Fig. 1c**).

The patient was discharged from the hospital on postoperative day 4. There were no complications and she remained asymptomatic without recurrence after 2 years of follow-up (**Fig. 1d**).

Discussion

In this report, we described the case of a patient with multiple, asymptomatic BAAs treated with VATS BA resection without lobectomy as a first-line treatment. The configuration of the BA and the location of the BAAs were clearly visualized using 3DCT, ensuring safe and complete resection. Preoperative BA angiography demonstrated a BA-PA connection, the presence of which affected the treatment plan. Our patient had no signs of pulmonary inflammation as she was healthy and asymptomatic.

Recently, catheter embolization has become the first-line treatment for BAA, as it is a well-established and minimally invasive treatment. Risks of this strategy

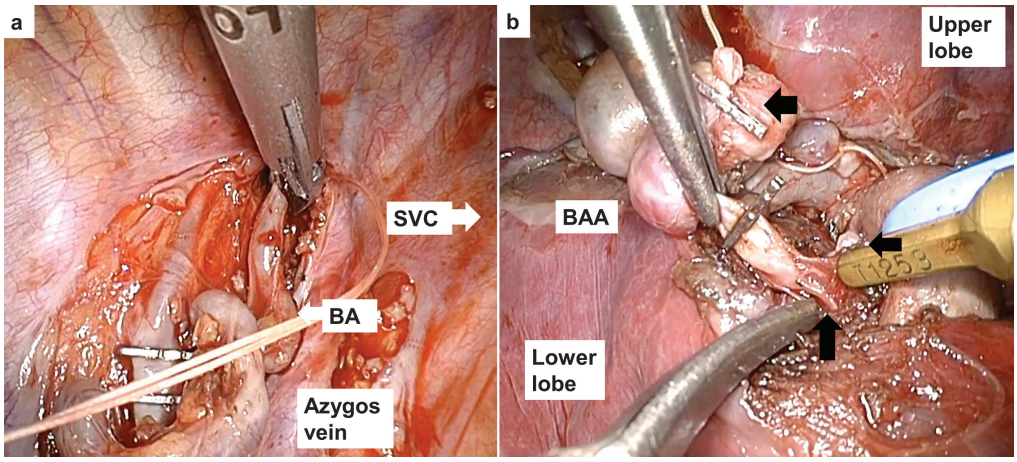

Fig. 2 (**a**) The most proximal BA aneurysm is detected in the upper mediastinum adjacent to the azygos vein. (**b**) The most distal BA aneurysm is adjacent to the interlobar PA. There were numerous branches off the artery and they were divided individually (black arrows). BA: bronchial artery; PA: pulmonary artery; BAA: bronchial artery aneurysm; SVC: superior vena cava

include both treatment failure and other complications. A review article[5] described that air emboli are reported in up to 2% of bronchial artery embolization (BAE) cases, and that spinal cord ischemia is reported in 1.4% to 6.5% of cases. If all afferent and efferent vessels cannot be embolized, treatment failure is possible.[2–4] In this report, there were many branches of BA found during the operation. Thus, there was a possibility of recurrence if embolization was selected.

Some of the common drawbacks after VATS are the operative wound and chest pain. Although BAE is in fact minimally invasive, the VATS procedure has also become less invasive as surgical instruments have improved. The perioperative risk of serious complications is lower if the patient has no signs of inflammation in the thoracic cavity, and if the part of the BA that needs to be removed is clearly identified preoperatively. Recurrence after surgical resection is rare if complete resection is achieved.

Surgery for BAAs includes surgical extirpation, BA ligation, resection of the BA with lobectomy, and BA resection without lobectomy.[2,3] In this case, we easily and successfully resected all BAAs and branches along with the BA. We could do this because the extent of resection was determined preoperatively using 3DCT, and VATS allowed clear visualization of the BA during the operation. Open lobectomy is invasive and can affect the hemodynamics of the patient; however, if the BA can be resected without lobectomy, the physiologic stress on the patient is less.

Indications of surgery for BAA treatment have been limited to cases wherein the patient presents with hemoptysis or hemothorax, or in cases where initial catheter embolization failed. Most previous reports describe resection with a thoracotomy.[2,3] In cases of life-threatening emergency, conventional thoracotomy may be preferable to VATS; however, when the patient is asymptomatic and catheter embolization is not straightforward or when complications are deemed likely, VATS BA resection as a definitive treatment should be taken into consideration.

Conclusion

Thoracoscopic BA resection without lobectomy as a first-line definitive treatment for asymptomatic multiple BAAs should be taken into consideration when the part of the BA that needs to be removed is clearly identified preoperatively and there are no inflammatory changes. Preoperative angiography and 3DCT can help to decide between catheter embolization and thoracoscopic surgery.

Disclosure Statement

The authors have declared that no conflict of interest exists.

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