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Case Report

Arterial Embolization and Cone-Beam Computed Tomography-Guided Lung Resection for Anomalous Systemic Arterial Blood Supply to Normal Lung: Two Case Reports

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Systemic arterial blood supply to a normal lung is a rare anatomical abnormality. Surgery is usually indicated because this abnormality leads to pulmonary hypertension. Herein, we report our experience and ideas for safe vessel dissection. Case 1 was a woman in her 50s. We performed a left lower lobectomy following percutaneous coil embolization. The aberrant artery with emboli was confirmed intraoperatively by cone-beam computed tomography (CBCT) to safely dissect under thoracoscopic surgery (TS). Case 2 was a man in his 40s. Following percutaneous endovascular plug occlusion, we performed a left partial resection using indocyanine green fluorescence navigation. Intraoperatively, CBCT imaging demonstrated the aberrant artery and exact position of the emboli. This combination technique of interventional radiology and TS with CBCT imaging was considered safe and more secure for the treatment of anomalous systemic arterial blood supply to a normal lung.

Keywords: cone-beam computed tomography, systemic arterial blood supply to a normal lung, coil embolization, vascular plug, aberrant artery

Introduction

In 1962, Campbell et al. reported systemic arterial blood supply to a normal lung, which was distinguished from Pryce's type I intralober sequestration.[1)] In this anatomical abnormality, systemic circulation from thoracic descending aorta, ascending aorta, abdominal aorta, or celiac artery flows into the normal lung parenchyma that is lacking pulmonary artery flow.[2,3)] Blood pressure in the aberrant arteries is high compared with that of the pulmonary circulation; thus, the abnormal flow usually leads to pulmonary hypertension, heart failure, hemoptysis, and arterial rupture.[1)] Surgery is usually required to remove the aberrant artery and the lung perfused by the aberrant artery, although some reports by suggesting embolization to occlude the aberrant artery without lung resection.[4)] In the last decades, thoracoscopic surgery (TS) rather than open thoracotomy has been more widely practiced for treatment of anomalous systemic arterial supply to basal segment of the lung. However, there was some reports about complications of TS with direct dissection of the aberrant artery using a stapling device, for example bleeding from the aberrant artery.

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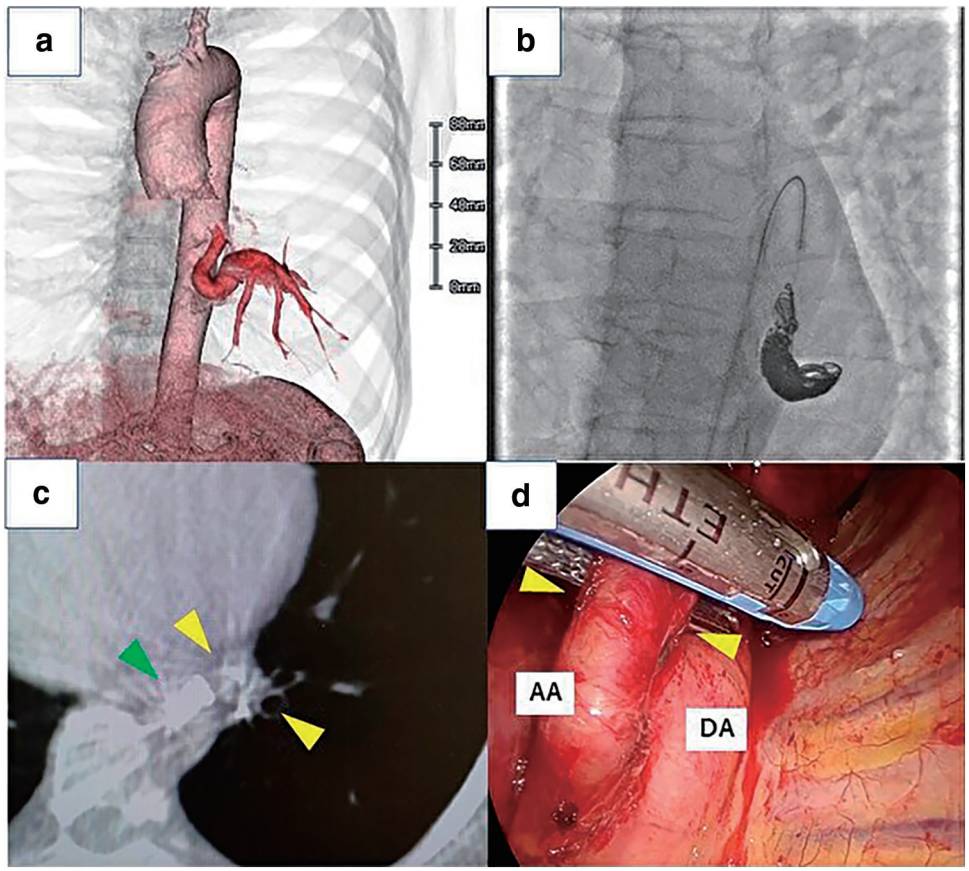

Fig. 1 Radiographic images for Case 1. (**a**) Volume rendering CT image showing an aberrant artery arising from the descending aorta entering the left lower lung. (**b**) Experienced interventional radiologists performed coil embolization to occlude the blood flow of the aberrant artery on aortography. (**c**) Intraoperative CBCT showing the embolizing coils (green arrow) and metal clip marking (yellow arrows) to highlight the cut line of the aberrant artery and avoid cutting over the coils. (**d**) The aberrant artery was safely cut on the side peripheral to the embolizing coils using an automatic stapler referring to the metal clip markings (yellow arrows). CT: computed tomography; CBCT: cone-beam computed tomography; AA: aberrant artery; DA: descending aorta

As you know, direct surgical dissection can carry the risk of bleeding because of high systemic systolic blood pressure. To safely treat an aberrant artery, there was several publications of transcatheter arterial embolization (TAE) or aortic stent graft that were performed in advance. However, it is still controversial how to ensure safety when dissecting a high-pressure aberrant artery. Herein, we propose a new technique with two cases of aberrant arteries that were dissected under cone-beam computed tomography (CBCT) guidance after TAE. Written general consent forms were obtained for both patients before surgery.

Case Reports

Case 1

An abnormal shadow on a chest X-ray was detected during a medical checkup in a woman in her 50s. Chest computed tomography (CT) revealed a 30-mm local consolidation surrounded by ground-glass opacity in her left lower lung. The vessels were widely dilated and the parenchyma of the lower lobe was congested. No bronchus pulmonary sequestration abnormalities were observed. CT angiography showed that the left basal segment of the lung was perfused by an aberrant artery, which was dilated to 15 mm in diameter, directly branching from the descending aorta (**Fig. 1a**). This patient was diagnosed with systemic arterial blood supply to a normal lung. Although the patient did not have any symptoms and adverse events associated with pulmonary hypertension, surgery was indicated to avoid to be going to pulmonary hypertension with some symptoms, such as hemoptysis and pulmonary infection, as patient's expectation. Blood flow reduction was necessary before surgery to avoid interoperative massive bleeding because the diameter of the aberrant artery was 15 mm and blood

flow was from the descending aorta with systemic blood pressure.

TAE was performed by radiologists experienced in interventional radiology (IVR). Aortography showed an aberrant artery perfusing the left basal segments. The aberrant artery branched from the aorta and drained into the left lower pulmonary vein. The periphery of the aberrant artery was occluded with a balloon catheter to stabilize the embolizing coils in the vessel. Metallic coils (POD Packing Coil 8 mm × 60 cm; Penumbra, Alameda, CA, USA) were used to embolize the aberrant artery (**Fig. 1b**).

After the TAE, she was transferred to a hybrid operating room (hOR) equipped with CBCT (Allura Xper FD 20, Philips, Amsterdam, The Netherlands). First of all, the absence of left bronchus abnormalities was confirmed by a bronchoscope under general anesthesia. Then, the patient was placed in the right decubitus position in the hOR. First, CBCT scanning was performed to confirm the exact position and length of the intravascular embolus. TS was started, and then the aberrant artery was carefully exposed. Referring to the CBCT image, we placed two metal clips (Ligaclip ER420; Ethicon, Tokyo, Japan) to soft tissue on the distal side of the embolus as a landmark of cutting line (**Fig. 1c**). After confirming the spatial relationship between the metal clips and intravascular metallic coils on second CBCT scanning, the aberrant artery was safely cut on the distal side of the embolus by an automatic stapler (ENDOPATH Powered ECHELON FLEX; Ethicon Endo-Surgery Inc., Cincinnati, OH, USA) (**Fig. 1d**). Afterward, the left lower lobe was removed. The total blood loss was 10 ml without blood transfusion. The operating time which structured coil embolization and surgery was 236 min. The postoperative course was uneventful. The patient was discharged on the fourth day after surgery. Pathological examination revealed degeneration of the aberrant artery media with lymphocyte infiltration and hemorrhagic and congestive lung parenchyma to the periphery. No complication, such as aneurismal changes, coil migration, or systemic embolism, occurred until the two-month follow-up.

Case 2

Abnormal findings on CT were detected during a systemic survey after a trauma in a main in a man in his 40s. CT showed a 60-mm in diameter focal consolidation in the left lung. The lung parenchyma was congestive and mucoid impaction was observed in the left basal bronchi (**Fig. 2a**). CT also showed an aberrant artery branching from the descending aorta into the left lung basal segments (**Fig. 2b**). He was diagnosed with systemic arterial blood supply to a normal lung. C-reactive protein was slightly elevated at the time of diagnosis, and he had treated pneumonia couple of times before. Thus, surgery was indicated due to a possible pulmonary infection.

Similar to the strategy in Case 1, TAE was performed by an experienced radiologist before surgery. A guide wire was inserted from the right femoral artery. Aortography showed an aberrant artery branching directly from the descending aorta into the left basal segments. An endovascular plug (AMPLATZER Vascular Plug II; AGA Medical Corp, Golden Valley, MN, USA) was placed in the aberrant artery to occlude blood flow.

TAE was followed by TS after he was transferred to an hOR. First of all, we confirmed the exact position of endovascular embolus demonstrated by the first CBCT scanning. Then we placed metal clip markings to the distal side of the embolus. The aberrant artery was ligated safer and accurately just on the peripheral end of the endovascular plug referred second CBCT scanning (**Fig. 2c**). After cutting the aberrant artery, indocyanine green (ICG) was injected into the patient's peripheral vein to reveal the area perfused by the aberrant artery. A demarcation line between the normal area and the aberrant artery-perfused area in the left lower lobe was visualized and the left lower lung perfused by the aberrant artery was partially harvested (**Fig. 2d**). The total blood loss was 152 ml without blood transfusion. The operating time that structured vascular plug embolization and surgery was 213 min. Macroscopically, some cystic areas filled with pus were observed in the lung parenchyma. Microscopically, abundant inflammatory cells including neutrophils, macrophages, and lymphocytes infiltrated the interstitial tissue. A fibrotic lesion, calcification, and thickness of the intrapulmonary vessels were also observed. The pathological findings suggested pulmonary hypertension with plexiform lesions. After caring for a surgical site infection, the patient was discharged on the 14th day after surgery. No mid-term complications, such as aneurismal change, coil migration, and systemic embolism, were detected on CT six months after the surgery.

Discussion

We resected anomalous systemic arteries using a combination of TAE and CBCT-guided surgery. The technical options for surgical treatment of aberrant arteries

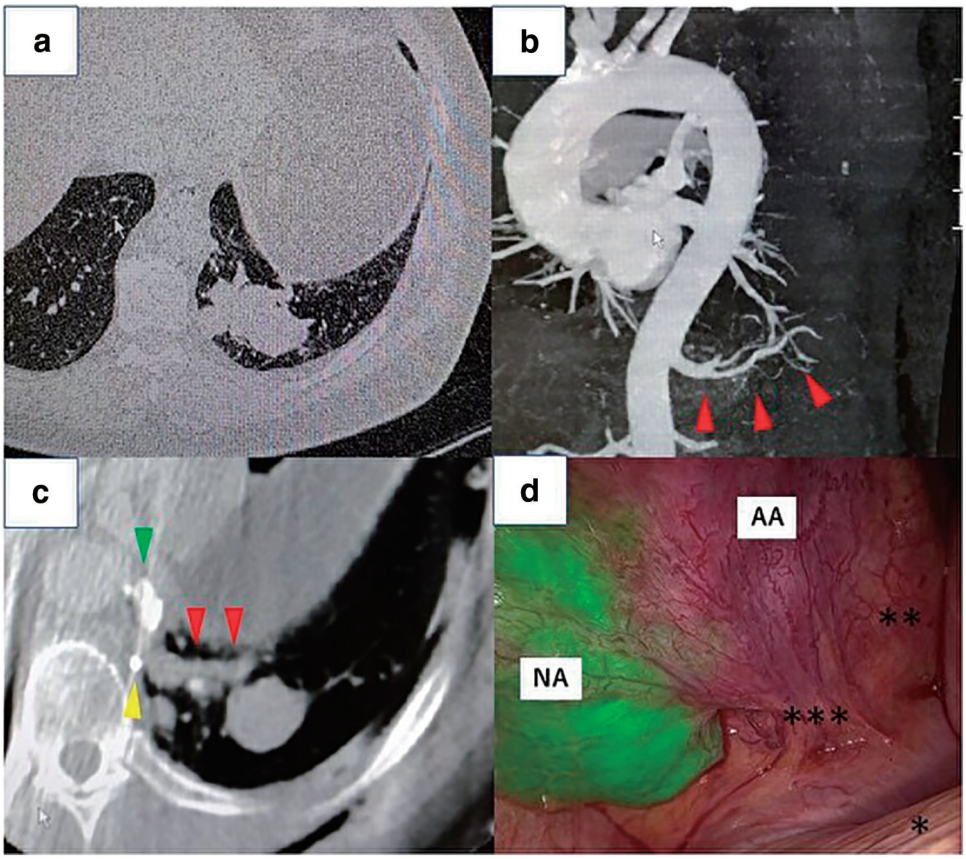

Fig. 2 Radiographical images in Case 2. (**a**) CT showing a local 60-mm consolidation in the left basal lung. (**b**) CT aortography revealing an aberrant artery arising from the descending aorta and feeding into the left basal lung (red arrows). (**c**) Intraoperative CBCT showing the endovascular plug (green arrow), metal clip marking (yellow arrow), and aberrant artery (red arrows) to safely cut the aberrant artery peripheral to the plug. (**d**) Under ICG fluorescence navigation, the normal area is green colored, and the aberrant artery-perfused area is a dark hue after cutting the artery. *: diaphragm; **: descending aorta; ***: pulmonary ligament; NA: normal lung area; AA: aberrant artery-perfused area; CT: computed tomography; CBCT: cone-beam computed tomography; ICG: indocyanine green

include simple ligation, stapler, ligation and stapler, or direct vessel suturing. No consensus on the appropriate technical approach has been reached because this anatomic anomaly is quite rare and few reports of adverse events associated with each technique have been published. Simple ligation is an easy conventional method but very occasionally, a knot might slip off the stump. The use of surgical staplers is now common in many surgical fields. General thoracic surgeons often use automatic stapling devices for resection of pulmonary vessels directly. Vessel stapling can endure up to 129.7 mmHg of pressure when not strained. However, under strain, such as lifting and/or twisting, the endurance pressure of stapling is less than 100 mmHg.[5] Therefore, stapling may not be adequate for sealing aberrant arteries. Direct vessel suturing is an alternative approach for cutting an artery. However, this method requires cardiovascular surgery techniques, such as partial clamping of the descending aorta while using a circulatory assist device. Furthermore, if that aorta is arteriosclerotic, the suturing may cause iatrogenic aortic dissection and postoperative bleeding.[6,7] In addition, minimally invasive surgery such as TS has been spread worldwide in the last twenty years. In our study, TAE was performed before TS for dissection of aberrant systemic arteries due to the reduction of high risk of bleeding when stapling. Interventional treatment with only TAE was described in several previous reports.[4,8,9] However, complications developed in the residual lung parenchyma in some cases, including pulmonary infection and inadequate blood flow.[4] Therefore, removal of lung segments perfused by an aberrant artery may result in fewer complications. Additionally, IVR using stent graft insertion to prevent intraoperative bleeding from the aorta was reported.[10] Stent grafts can cover emergency bleeding, such as the rupture of an aberrant artery.[11] However, aortic stent graft could be

avoided when a patient is not elderly because long-term outcomes are unclear.

We have already reported as a previous study that CBCT was originally adopted to resect pulmonary tiny nodules. We have also verified feasibility with X-ray dosimeter.[12] This time we used it to confirm accurate cutting line with metal clip markers to avoid running over emboli with the stapler.

In Case 1, we chose metallic coils for vascular embolization because the aberrant artery was meandering and the embolus was placed in a curved section of the artery. On the other hand, a vascular plug was chosen in case 2. Vascular plugs easily occlude blood flow with a high success rate and a low risk of migration. However, vascular plugs are not appropriate for meandering vessels due to the risk of migration.[13]

Since the aberrant artery perfused a larger area of normal left lower lung in case 1, a lower lobectomy was required in that patient. By way of contrast, a partial resection was performed in case 2 because the area perfused by the aberrant artery was relatively small. We used ICG fluorescence navigation to reveal the area perfused by the aberrant artery. ICG is dark green (water soluble) and the 774.96 molecular weight molecule is stabilized by plasma protein binding in the circulation. ICG is a fluorescent chromophore that excites at 750–810 nm. ICG navigation surgery is useful for lung segmentectomy during TS to distinguish the area of disrupted blood flow from the normal area.[14–16] In one recently reported case, this technique was employed to detect a specific-artery-perfused area.[17] We detected the area perfused by the aberrant artery, which showed us as an area of disrupted blood flow, using ICG navigation after ligation of the aberrant artery using a surgical endoscope (Image 1, D-light system; Karl Storz Endoscope, Japan K.K., Tokyo, Japan).

We consider this combined technique safe and useful, but there is a limitation. An hOR and an experienced IVR team are required to safely perform this procedure. Thus, the technique may not be highly versatile. However, this technique may become more widely available as the IVR technique becomes more popular.

Conclusion

We resected an aberrant artery with systemic arterial blood supply to a normal lung using a combination of TAE and CBCT imaging. This procedure is safe, useful, and minimally invasive.

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Disclosure Statement

All authors have no conflicts of interest to disclose.

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