Surgical strategy for pulmonary sequestration: Focus on precautions for aberrant vessels under minimally invasive surgery

Tomoki Nakagawa¹, Atsushi Wada¹, Naohiro Aruga¹, Hajime Watanabe¹, Ryota Masuda¹, Shunsuke Yamada¹ and Masayuki Iwazaki²

Abstract

Background: Recently, thoracoscopic resection of pulmonary sequestration has become more common, since resection of an aberrant artery using an end-stapler is a safe maneuver in many cases. However, injury of the vessels can lead to major hemorrhage. We reported our surgical experience based on thoracoscopic surgery, with five cases of interlobar pulmonary sequestration, focusing on precautions for aberrant arterial vessels.

Object and methods: We performed pulmonary resections for five patients with interlobar pulmonary sequestration in a lower lobe (left, n = 4; right, n = 1) between April 2004 and May 2020. All aberrant vessels were derived from the lower thoracic artery. Two patients had a single aberrant artery and three had multiple. In four patients, these vessels were detected before surgery, and pulmonary sequestration was diagnosed in four. In one elderly patient, the aberrant vessel was overlooked, and lung cancer was suspected before surgery. Angiography or multidetector-row computed tomography was subsequently performed in four cases. The surgical plan was determined according to the location and size of the pulmonary lesion and three-dimensional images of aberrant vessels.

Result: In all patients, approaches were made thoracoscopically. Hemorrhage from an anomalous vessel was encountered in one case. Pulmonary resections included two lobectomies and three limited resections. Angioplasty for the root of anomalous branches was performed following pulmonary resections under converted minimal lateral thoracotomy in two cases.

Conclusion: Preoperative assessment of the anatomical variations in abnormal vessels is essential to achieve safe surgical procedures. According to the situation of the aberrant vessels, selecting surgical procedures with consideration of potential subsequent complications arising over a long period of time is important.

Keywords

pulmonary sequestration, stapling resection, minimum invasive surgery

Introduction

Pulmonary sequestration (PS) is a congenital anomaly of the lung, involving anomalous non-communication of parenchyma with normal bronchial trees, and aberrant arteries derived from systemic arteries. PS is sometimes treated by surgical resection, for reasons such as recurrent infection or bleeding.¹² Since these lesions show a nodular or cystic formation, differentiation of PS from tumor or granulomatous nodules is also important.¹² Detection of abnormal vessels derived from a systemic artery is essential to confirm PS. Recently, thoracoscopic resection of PS has become more common since resection of an aberrant artery using an end-stapler is a safe maneuver in many cases. However, injury of the vessels can lead to major hemorrhage. Preoperative...
assessment of the anatomical variations in abnormal vessels using multidetector-row computed tomography (MDCT) is essential to achieve safe surgical procedures. In this study, we evaluate our thoracoscopic procedures dealing with aberrant arterial vessels in 5 cases of PS from a single institution.

**Patients and Methods**

Between April 2004 and May 2020, pulmonary resection was performed for five patients with PS in our institution. All cases underwent thoracic computed tomography (CT). Angiography or MDCT was used to obtain further information and assess the structure of aberrant vessels. Based on the preservation of lung parenchyma, surgical plans were determined according to the location and size of the sequestered area, and the diameter of aberrant vessels. Surgery was started under a thoracoscopic approach, with a focus on the management of aberrant vessels.

**Results**

Characteristics of the five patients are listed in Table 1. Two patients presented with dry cough, and the remaining three patients were asymptomatic. The pulmonary lesions ranged in size from 26 mm to 32 mm and were in the left lower lobe in four patients and the right lower lobe in one patient. CT detected aberrant vessels in four cases, by angiography in the first case and MDCT in the other three cases. Aberrant arterial vessels were seen to originate from the lower thoracic aorta in all four detected cases, confirming PS in these patients. A draining vein to the azygos system (Figure 1(a)) was apparent in two cases. Thoracoscopic limited resection was planned in three cases. In Case 2, the site of the aberrant arterial branch from the thoracic aorta was slightly swollen like an aneurysm, and was about 24 mm in diameter in multidetector-row and axial computed tomography (CT) findings (Figure 1(b)). Angioplasty of a branching site of the aberrant vessel from the descending aorta was planned after thoracoscopic limited pulmonary resection, under conversion from thoracoscopic surgery to minimal lateral thoracotomy. The thoracotomy site was installed to be suitable for angioplasty. In Case 3, the abnormal vessel was overlooked in an elderly patient, and lung cancer was therefore suspected. Bronchoscopic biopsy could not confirm a definitive diagnosis. Thoracoscopic lobectomy was planned for diagnostic and therapeutic purposes. All cases were stated with thoracoscopic surgery. Surgical result and outcome of the five patients are listed in Table 2. The abnormal vessels and systemic draining vein were exposed in the pulmonary ligament under video-view in all cases. Preoperative evaluations of aberrant vessels including draining veins were almost the same as the surgical findings, except for the existence of two...

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**Table 1.** Patient Characteristics.

| Case | Age/ Sex | Symptom | Shadow | R/L | Location | Diagnosis | Reoperative images of aberrant vessel | Draining vein to systemic vein | Surgical plan |
|------|----------|---------|--------|-----|----------|----------|-------------------------------------|---------------------------------|----------------|
| 1    | 41/M     | (-)     | PN (36mm) | L   | S10      | PS       | Lower thoracic artery               | 1                               | 10(mm) (-) Limited resection |
| 2    | 19/M     | (-)     | PN (32mm) | L   | S10      | PS       | 1                                   | 24(mm)                         | 1 Limited resection + angioplasty Lobectomy |
| 3    | 79/F     | Cough   | PN (28mm) | R   | S7S10    | LC       | (1)                                 | 4(mm) (-)                      | Lobectomy |
| 4    | 47/F     | Cough   | PN (26mm) | L   | S10      | PS       | 1                                   | 13(mm) (-)                     | Limited resection |
| 5    | 18/M     | (-)     | Radiolucency; hyper-areated area | L   | S10      | PS       | 3                                   | 8.0(mm)                        | Limited resection |

PN: pulmonary nodule, PS: pulmonary sequestration, LC: lung cancer.
*remarking image findings

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**Figure 1.** (a–b) Multidetector-row and axial computed tomography (CT) showing slightly swollen like an aneurysm (arrowed head; 1b) at the site of the aberrant arterial branch from the thoracic aorta, and a systemic draining vein (arrowed head; 1a) in Case 2. (c–d) Tomography 3 years after angioplasty of an aberrant vessel branch showing repaired the descending aorta (arrowed head; 1c) in Case 2.
small, thin-walled aberrant vessels in Case 1. In the case where the aberrant vessel was overlooked (Case 3), adhesions were seen from the dorsal side of the lower lobe. The thickened pulmonary ligament became very bloody during division of this tissue. CT findings were then re-checked, and the presence of an abnormal branch arising from the thoracic aorta was recognized (Figure 2). This aberrant vessel was finally exposed in the pulmonary ligament after processing the pulmonary vessels and lower bronchus. Surgical hemorrhage was encountered in Case 1. The safety of end-staplers for arterial procedures was not confirmed until 2004, so stapling division was planned after proximal ligation of the aberrant artery using 2-0 silk.3 In surgery, tension on the vessel during proximal ligation using a knot pusher led to injury at the branching point from the lower thoracic aorta. Fortunately, hemostasis was achieved by maintaining pressure on the bleeding point throughout the whole lung with Narke Type Thoraco-Cotton (Kenzmedic co, Saitama, Japan), without needing conversion to thoracotomy. All aberrant vessels were eventually divided using the end-stapler.

In this study, aberrant arteries arose from the thoracic aorta in all cases, as single vessels in three cases, and multiple in two. The draining vein was a systemic draining vein (azygos system) in two. As for management of aberrant vessels and draining veins to the hemi-azygos vein, stapling resection was safely completed in four cases. In a large-diameter case (Case 2), as a temporally maneuver, the distal branches of aberrant vessels were ligated using the knot pusher and divied (Figure 1(a)). Pulmonary resections included two lobectomies and three limited resections. In the case with limited resection, the margin of sequestered parenchyma between normal tissues was clear. Wide-wedge resection of sequestered lung was easily achieved using the end-stapler. On the other hand, in the lobectomy cases (Cases 3 and 4), massive adhesion made the boundary between normal and sequestered lung unclear.

After limited pulmonary resection, angioplasty was performed in two cases. One case (Case 1) was the case with injury of the proximal branch, while the second (Case 2) involved a stump of large-diameter branch (24 mm). Following conversion to minimal lateral thoracotomy, lateral mattress suturing of the vessel stump at its origin with pledges, under lateral clamping of the thoracic aorta, was performed under direct vision. Reasons for choosing this technique were that injury to the intima of the proximal branch (Case 1) or a remnant large aberrant vessel stump (Case 2) may subsequently result in delayed complications, such as aneurysmal formation. The pathological diagnosis was interlobar sequestration in all cases. All patients displayed an uneventful postoperative course. Follow-up for patients ranged from 6 to 57 months. Follow-up CT did not detect abnormal findings such as aneurysmal formation at the vascular stump. During the follow-up period, left pneumothorax developed 3 months after surgery in Case 5, and thoracoscopic bullectomy was performed. Looking at the site where the abnormal vessels were divided, the stump of the aberrant vessels was shortened and the surrounding area was organized (Figure 3).

Discussion

Thoracoscopic surgical resection for PS has recently been becoming more common. Stapling resection of aberrant vessels using an end-stapler has recently been considered a safe maneuver in many cases. Generally, the majority of aberrant vessels arise from the descending thoracic aorta (76%) or abdominal aorta (21%).5 Most cases involved a single supply (79%), with a smaller proportion showing two
or more supplies (21%). In our experience, two of five cases (40%) showed multiple branches. These results impressed that the PS with multiple aberrant systemic supplies were relatively common. The surgical problems in vascular management are as follows. First, the vessels are direct branches from the aorta, and are thus constantly exposed to high intra-luminal blood pressure. Second, these vessels are more fragile than normal arteries of similar size. An aberrant vessel wall histologically shows numerous elastic fibers and few smooth muscle layers, more closely resembling the pulmonary artery than the bronchial artery. As experienced in Case 1, such vessels are easily damaged by excessive tension or traction. Dissection of sufficient vessels and gentle maneuvering are required, especially when inserting an end-stapler for division of these vessels. Third, anatomically, most of these vessels enter the lung through the pulmonary ligament, which is deep below in the thorax. The pulmonary ligament is sometimes thickened and hemorrhagic due to chronic inflammation of sequestrated lung. These make surgical operability for aberrant vessels difficult. Intraoperative injury of these vessels, especially at the root of vessel, might lead to life-threatening hemorrhage, even during thoracotomy. Undiagnosed PS in right lower lobe, as in our Case 3, always carries a surgical risk of unexpected aberrant vessels. Especially, the right side approach in the left lateral decubitus position makes it anatomically difficult to secure the descending aorta as a last resort. Interlobar sequestration is diagnosed at or before 20 years old in approximately 50–60% of cases. Discovery in patients over 70 years old (as in Case 3) is rare. As a result, abnormal pulmonary vessels were overlooked in consideration of lung cancer since lung cancer is more prevalent than PS. As PS is predominantly seen in the posterior basal segment, care must be taken to avoid overlooking aberrant vessels associated with undiagnosed mass lesions located in the posterior basal segment. In this study, we determined the surgical plan according to the anatomy of aberrant vessels using MDCT-angiography. In our experience, two of five cases (40%) showed multiple branches. Most preoperative images were consistent with surgical findings, but two thin-walled small vessels in Case 1 were not visible on preoperative MDCT imaging. Assessment of the anatomical variations of the aberrant arterial vessels before surgery is important.

Management of aberrant vessels depends on the size of the vessels. In our experience from this study and the literature, division of aberrant arterial vessels using an end-stapler is safe and suitable for vessels up to 20 mm in diameter. Since the mean diameter of vessels associated with PS in the literature is 6.3–6.6 mm, most aberrant vessels can be managed using an end-stapler. Theoretically, cutting immediately after the aberrant vessel branches from the descending thoracic aorta is optimal. Advances in the technology of linear stapling devices have led to a narrower anvil and greater flexibility, allowing easier access beyond strictures in the thoracic cavity. However, in our experience, maneuvering a stapling device to perpendicular to the proximal aberrant vessel branch in the thoracoscopic procedure is difficult. The device is instead often inserted diagonally into the vessel, and the tip contacts the mediastinal surface. A medical accident report suggested that a risk of injury to the inner layer of the main artery appears immediately after grasping the branching artery using the end-stapler, resulting in the potential for serious bleeding consequences. The stapling site of an aberrant vessel using an end-stapler should thus be a certain distance from the branching point (Figure 3(a)). In such situations, we are concerned about subsequent complications at the cut-edge of fragile tissues. No reports have described aneurysm formation from remnant aberrant vessels after surgery. In Case 5, left pneumothorax surgery was able to reconfirm the stumps of three aberrant vessels (maximum diameter = 8 mm) 3 months after the initial surgery. The stumps of these vessels (Figure 3(a)) were shortened, and the surrounding area was organized (Figure 3(b)). The healing process for divided vascular stumps appeared good. On the other hand, aneurysmal dilatation with a diameter of 25 mm was reportedly seen in 0.25% of PS. A few cases have been reported of PS with aneurysmal formation of an aberrant artery. The question thus remains as to whether a large vessel diameter or injury to the root of a remnant vessel represents problems. The main point of concern is that vessel stumps in either of the above situations may result in subsequent aneurysmal formation under long-term exposure to high blood pressure.

In addition, after pulmonary resection, angioplasty was performed in two cases under minimum lateral thoracotomy. There are interesting surgical reports of PS with aneurysmal
dilatation at the origin of the aberrant vessel. The lumen of the aberrant vessel was closed by engraft, which involves application of aortic aneurysm treatment (thoracic endovascular aortic repair; TEVAR) in advance, and the aberrant vessel was divided using a stapling device. This method seems suitable for management in cases of dilated or aneurysmal aberrant vessels, as long as the endograft does not involve important blood vessels. The purpose of this surgery is in some way similar to two of our cases. The difference is whether to close the vessel of origin from the inside or outside, and this method also seems to be effective as an alternative treatment for a late-onset aneurysmal disorder of the vascular stamp.

Conclusion

We reported on our surgical experience with 5 cases of PS that we encountered at our hospital, focusing on the division of aberrant vessels. Multidetector-row computed tomography was used to provide further information to assess the structure of aberrant vessels. Almost all cases of aberrant vessels are ≤ 2 cm in diameter and could be managed using an end-stapler. However, the surgical stumps of these vessels are fragile and exist in a hypertensive environment. According to the situation of the aberrant vessels, selecting surgical procedures with consideration of potential surgical risk including subsequent complications arising over a long period of time is important.

Declaration of Conflicting Interests

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ORCID iD

Shunsuke Yamada https://orcid.org/0000-0001-5575-1061

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