



Right completion pneumonectomy for impending rupture of a pulmonary artery pseudoaneurysm

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Abstract

We present a case of right completion pneumonectomy for a pulmonary artery pseudoaneurysm after right upper lobectomy and radiotherapy for metachronous multiple right lung cancers. An 80-year-old man was referred to our hospital because of repeated hemoptysis. Computed tomography revealed a pseudoaneurysm in an interlobar pulmonary artery. We considered hemoptysis as a sign of impending rupture. Radiologists could not recommend embolization because of a high risk of rupture or pulmonary infarction, and re-hemoptysis due to coil movement; therefore, we performed right completion pneumonectomy because of severe adhesion. At last, the patient has completely recovered from impending rupture status.

Keywords Surgical treatment · Pulmonary artery pseudoaneurysm

Introduction

A pulmonary artery pseudoaneurysm (PAP) is an extremely rare, but potentially lethal condition requiring emergent interventions for life-saving treatment [1]. The decision to treat PAPs by surgery or endovascular repair is a matter of some debate and it is necessary to examine therapies depending on each case [2]. We report a case in which right completion pneumonectomy was urgently performed for impending rupture of a PAP after right upper lobectomy and radiotherapy for previous metachronous multiple right lung cancers.

Case

An 80-year-old man was referred to our hospital because of repeated hemoptysis. He previously underwent right upper lobectomy for lung cancer of the right upper lobe (13 years ago) and radiation therapy (48 Gy) for lung cancer of the right lower lobe (6 years ago). Chest radiography did not show an abnormal shadow causing hemoptysis in the lung

field. However, chest contrast-enhanced computed tomography (CT) revealed a saccular aneurysm 18 mm in size, which was thought to cause hemoptysis, arising directly from the interlobar pulmonary artery in front of the right middle lobe pulmonary artery divergence (Fig. 1a, b). Bronchoscopy showed a clot, which was compatible with a sign of impending rupture of a PAP, in the right middle lobe branch. We considered hemoptysis as a sign of impending rupture. Radiologists did not recommend embolization because of a high risk of rupture or pulmonary infarction, and re-hemoptysis due to coil or embolic material movement, even though we considered severe adhesion, which made surgical treatment difficult, because of previous surgery and radiotherapy for the right lung cancer. Therefore, we decided to perform surgical treatment urgently to save his life.

Median sternotomy with right neck transverse incision was performed with the patient in a supine position. Anesthesiologists maintained low blood pressure of the patient to avoid perioperative rupture of the PAP. First, we secured the right main pulmonary artery and both the right superior and inferior pulmonary veins in preparation for perioperative aneurysm rupture. Because of the influence of previous treatment, the right main pulmonary artery was secured between the superior vena cava and ascending aorta near the transcatheter sac, and the right pulmonary vein was secured, respectively, in the cardiac sac space. As the aneurysmectomy was difficult because of the severe and tight adhesion of the thoracic cavity, we decided to

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Fig. 1 Chest (a) contrast-enhanced CT and (b) three-dimensional CT revealed a saccular aneurysm 15 mm in size (arrow), which was thought to cause hemoptysis, arising directly from the interlobar pulmonary artery in front of the right middle lobe pulmonary artery divergence. **a** Anterior view. **b** Posterior view. *CT* computed tomography

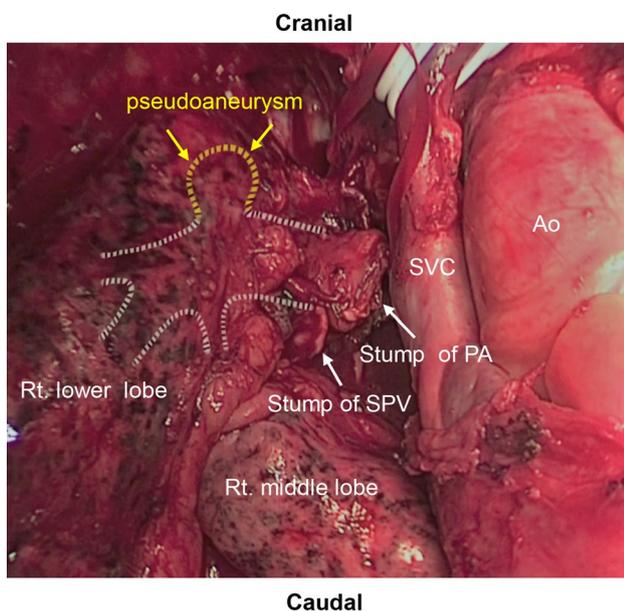
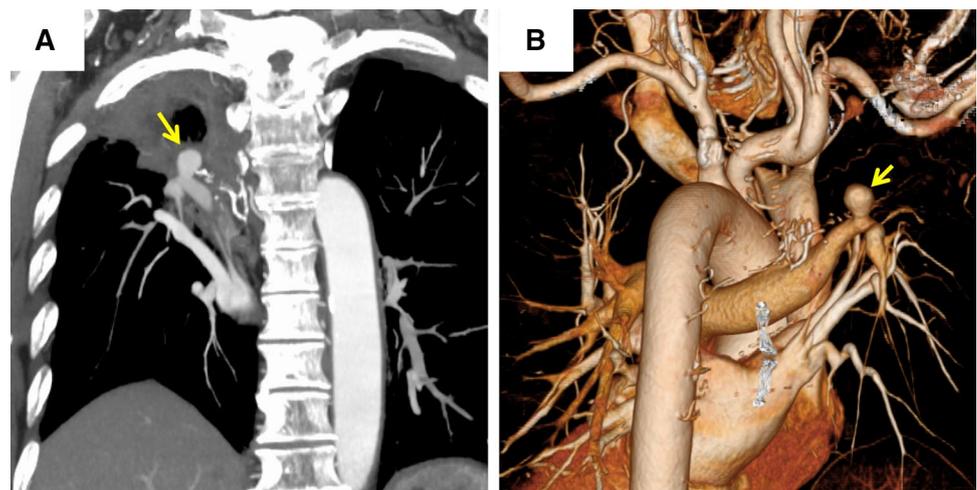


Fig. 2 Intraoperative findings: during surgery, the site of the pulmonary aneurysm could not be identified because of severe adhesion, but it seemed to be recognized in the region of the yellow dotted line (arrow)

perform right completion pneumonectomy. We separated the right main pulmonary artery and both the right upper and lower pulmonary veins and exposed and separated the right main bronchus (Fig. 2). Because adhesion in the apex area of the chest wall and the circumference of the azygos vein was particularly severe, anterior side thoracotomy was added after replacing the patient in the left lateral decubitus position. We separated the azygos vein nucleus at the level of the inflow section of the superior vena cava and exfoliated the hard adhesion of the chest wall. Finally, we performed right completion pneumonectomy.

Since intraoperative adhesion was robust and there was breakdown of the aneurysm wall in the excised specimen, it was impossible to identify the cause of the aneurysm (e.g., surgery effects, radiation therapy, inflammation).

We removed the mediastinal drain on postoperative day 2 and the thoracic drain on postoperative day 4. The patient recovered without re-hemoptysis and visited another hospital for rehabilitation 2 months after surgery.

Discussion

A PAP is an uncommon entity, but is important to recognize because of the associated morbidity [1]. A variety of causes have been proposed concerning its etiology and most suggest that pulmonary pseudoaneurysms are acquired [3]. The use of pulmonary artery catheter (iatrogenic) is the most common cause of PAP and is estimated to occur at a rate of 0.5% [4]. Vasculitis (Behçet's syndrome), infection (tuberculosis, pyogenic, and fungi), trauma, and neoplasms have been reported as other causes of PAPs [1, 3]. In this case, we could not pathologically identify the cause of the PAP in the resected specimens because of severe adhesion around the PAP. The most common presenting symptoms are hemoptysis, chest discomfort, cough, and dyspnea, but occasionally, they remain undetected and silent for many years [3]. In the literature, the prevalence of PAPs ranges from 8 to 11% in patients presenting with hemoptysis and who underwent bronchial or pulmonary angiography [5, 6]. The gold standard technique for diagnosis is pulmonary angiography, but this is now largely performed by non-invasive CT angiography and 3-dimensional reconstruction [7]. The actual mechanisms of the formation of PAP are still unknown, but one possible explanation is that increased focal tissue pressure due to the edema and soakage during the acute phase of inflammation decreases or stops the resolving phase and

eventually causes the dilation of vessels that have already been injured in the acute phase [8]. PAPs represent a life-threatening condition, causing spontaneous rupture, suffocation due to hemoptysis, infection, and distal embolization and thrombosis [9]; therefore, emergent interventions are needed for their treatment.

Treatment of PAPs is surgical (aneurysmectomy, pulmonary resection, or arterial branch ligation) or endovascular (embolization) [3, 10]. Surgery involves resection of the pseudoaneurysm itself; there is the advantage of decreasing the risk of recurrence, but depending on the etiology, the influence of intrathoracic adhesion or infection makes surgery extremely difficult, and lobectomy or pneumonectomy is required, not simple aneurysmectomy or arterial branch ligation. Therefore, embolization is associated with lower morbidity and mortality than surgical treatment and has become the first-line treatment [11]. However, there are limitations with embolization. First, the coil or embolic material may be a hindrance to pulmonary blood flow, leading to pulmonary infarction [12]. Second, as a pseudoaneurysm does not involve all layers of the arterial wall and is, therefore, at higher risk of rupture, embolization may cause aneurysmal rupture and lead to fatality [3]. Third, embolization prevents PAPs from rupturing, but in cases of complicated infections, it may have less efficacy in controlling inflammation [2]. Fourth, there is a possibility of recurrence due to movement of the coil or embolic material.

In the present case, the interventional approach seemed to be very difficult because the PAP with a sign of impending rupture existed in an interlobar pulmonary artery, and intra-aneurysmal embolization could provoke infarction of the right residual lung. In addition, there was the possibility of rupture or recurrence of the PAP. Therefore, after consultation with radiologists, we selected urgent surgical treatment of the PAP despite the patient being elderly with severe intrathoracic adhesion after previous surgery and radiotherapy for right lung cancer. There are few reports as treatment for the PAP, but one of the endovascular treatments includes stent graft insertion. There is a major merit that securing of peripheral blood flow is possible, and the stent graft insertion is used with wide-mouthed aneurysms like the disorder when a carp ring is difficult. Radiologists did not recommend embolization because of a high risk of rupture or pulmonary infarction and re-hemoptysis due to coil or embolic material movement. Although we performed right completion pneumonectomy because of severe adhesion in the thoracic cavity, treatment of blood vessels in the cardiac sac space could support us to avoid unexpected critical bleeding. The postoperative course was also uneventful. This is the first report of a patient with a PAP, who urgently underwent right completion pneumonectomy because of impending rupture.

Conclusion

Generally, embolization is a safe and effective treatment for PAP. However, surgical treatment after consultation with radiologists should be considered if a life-threatening status, like an impending rupture, is observed.

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Compliance with ethical standards

Conflict of interest We have no conflict of interest to disclose.

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