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Case Report

Pulmonary Artery Pseudoaneurysm Secondary to Lung Inflammation

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Pulmonary artery aneurysms (PAA) and pseudoaneurysms (PAP) are caused by infections, vasculitis, trauma, pulmonary hypertension, congenital heart disease, and connective tissue disease. Most cases of such aneurysm occur in the trunk or major branches of the pulmonary artery, while the peripheral type is less common. The treatment modalities are medical therapy, surgery, and percutaneous catheter embolization. The mortality rate associated with rupture is approximately 50%. We encountered a case of a 53-year-old man with a pulmonary artery pseudoaneurysm secondary to pneumonia and cavity formation during chemotherapy for acute myeloid leukemia (AML). In diagnosis, contrast-enhanced chest computed tomography (CT) scan and pulmonary angiography were very useful. He was treated with right middle and lower lobectomy. After 1-month follow-up, he could restart additional chemotherapy.

Keywords: pulmonary artery pseudoaneurysm, lung inflammation, pneumonia

Introduction

Pulmonary artery pseudoaneurysms are uncommon. Because of the risk of high mortality secondary to pseudoaneurysm enlargement and rupture, prompt therapy is needed. In this paper, we report a case of right pulmonary artery pseudoaneurysm in a patient with acute myeloid leukemia (AML).

Case Report

A 53-year-old man was admitted to the hematologic department of our hospital because of pancytopenia. After further examination, he was diagnosed with AML. He did not have a history of malignancy or illness except for anxiety disorder, which was treated with minor tranquilizers. Chemotherapy (idarubicin hydrochloride and cytarabine) for AML had been administered for 1 month, but it was stopped because of neutropenic fever and pneumonia of the right middle lobe (RML) that developed during treatment. No bacteria were detected in his sputum cultures. He was administered antibiotics (tazobactam/piperacillin), and the pneumonia resolved completely. However, after 1 month, he acquired pneumonia of the right lower lobe (RLL). He was treated with antibiotics (tazobactam/piperacillin and vancomycin) for 1 month. A chest X-ray and computed tomography (CT) scan showed pneumonia of the RLL with a pulmonary cavity. No infectious organs or tumor cells were detected on bronchoscopy. Although the patient was given antibacterial and antifungal medication (micafungin and amphotericin B), the pneumonia in the RLL and cavity formation was not cured. Subsequent contrast-enhanced chest CT scan after 1 month showed an enhanced mass inside the pneumonia lesion in the RLL (**Fig. 1**). Pulmonary angiography showed pulmonary artery

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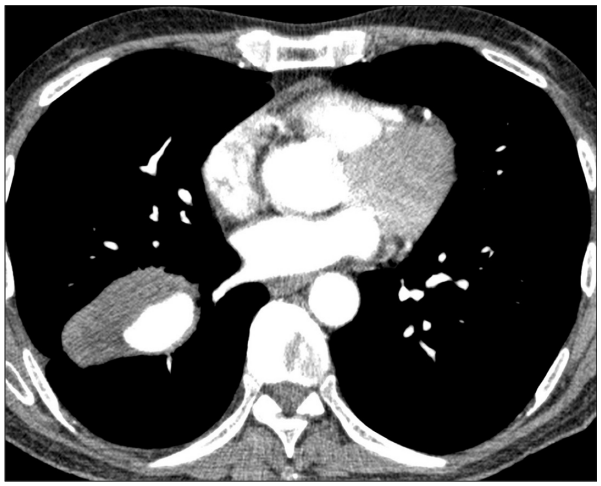

Fig. 1 Subsequent contrast-enhanced chest CT scan showed an enhanced mass inside the pulmonary cavity of the RLL. CT: computed tomography; RLL: right lower lobe

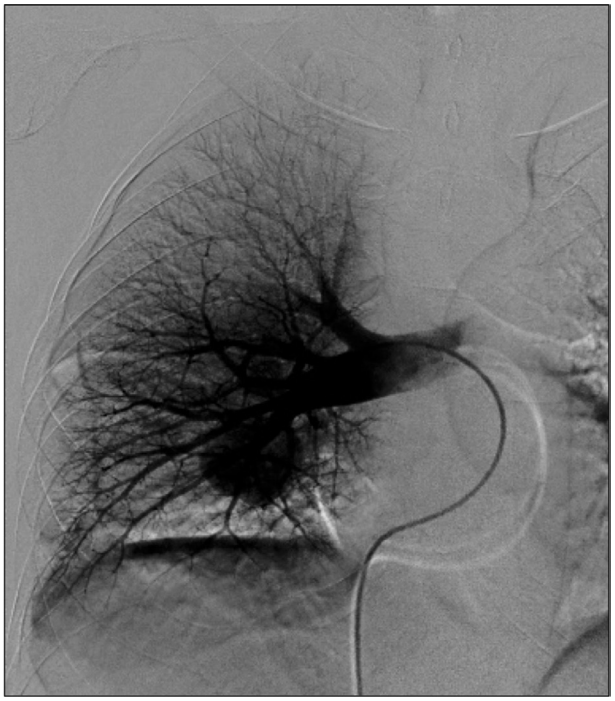

Fig. 2 Angiogram of right pulmonary artery and right lower pulmonary artery aneurysm/pseudoaneurysm.

aneurysm/pseudoaneurysm (PAA/PAP) in the RLL of the lung (**Fig. 2**).

Blood test results indicated that antibacterial and antifungal therapy decreased the inflammatory reactions, but vascular disease with mass shadow and localized pneumonia remained. To remove the antibacterial and antifungal therapy-resistant lung lesion, he underwent right middle and lower lobectomy. On thoracotomy, inflammation was found to have extended to the RML.

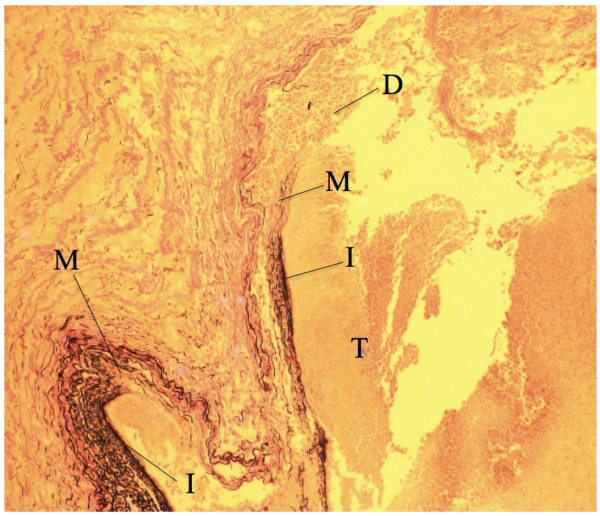

Fig. 3 Microscopically, on incision of the pulmonary artery, disruption of elastic fibers in the intima and media were noted (Elastica van Gieson staining × 40). I: intima; M: media; D: disruption of elastic fibers in the intima and media; T: thrombus

The postoperative course was uneventful, and he was discharged on the 10th postoperative day. After 1-month follow-up, he restarted additional chemotherapy at the hematologic department.

Macroscopic examination of the bilobectomy specimen showed a grossly consolidated lung, distorted by adhesions. The specimen had characteristics indicative of pneumonia together with necrosis and pulmonary cavity. Microscopically, on incision of the pulmonary artery, disruption of elastic fibers in the intima and media was noted using Elastica van Gieson staining (**Fig. 3**). As such, PAP was considered. Furthermore, filamentous fungi with thin and septate hyphae were detected. Pigmented fungi were also detected using Fontana-Masson staining. These findings were compatible with pneumonia caused by *Cladosporium* species.

Discussion and Conclusion

An aneurysm clinically refers to any localized widening or outpouching of an artery, a vein, or the heart. A true aneurysm is one that involves all dilated layers of the vessel wall. Whereas a pseudoaneurysm is a collection of blood leaking completely out of the vessel wall, but confined next to the vessel by the surrounding tissues.[1)] PAA/PAP are rare but fatal disorders. Most cases of PAA/PAP occur in the trunk or major branches of the pulmonary artery due to pulmonary hypertension, congenital heart anomalies, endocarditis, or cardiac

surgery.[2] The peripheral type of PAA/PAP is less common but usually occurs with iatrogenic vascular injuries from Swan-Ganz catheters or infectious diseases, such as tuberculosis, bronchiectasis, or lung abscess.[3–5]

Presentation changes depend on the localization and size of PAA/PAP; symptoms include fever, chest pain, dyspnea, recurrent pneumonia, and hemoptysis. Major cause of hemoptysis is due to bronchial artery erosion. Hemoptysis is the most frequent and life-threatening symptom after PAA/PAP rupture. Although bleeding from the pulmonary artery is very rare, usually massive,[6] and is usually due to pseudoaneurysm rupture.

The diagnosis requires CT with intravenous contrast and pulmonary artery angiogram to exclude an infection and lung tumors. On imaging studies, the appearance of a PAA is described as round, regular-bordered mass and isodense to the central pulmonary artery with varying sizes.

Because there is a risk that PAA/PAP will enlarge and rupture, which leads to death in approximately 50% of patients,[7] it is necessary to treat promptly. The available treatments of PAA/PAP are medicine, surgery, and percutaneous catheter embolization. Several operation methods, such as aneurysmectomy, lobectomy, pneumonectomy, hilar clamping with direct arterial repair, and ligation, have been employed.

In our case, because excision of antibacterial and antifungal therapy-resistant lung lesion was required in addition to PAA, we performed lobectomy. Finally, *Cladosporium* species was detected in resected lung tissue samples.

Although small aneurysm lesions under low pressure often resolve spontaneously, the actual mechanisms of the formation of PAA/PAP are still unknown. 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]

Disclosure Statement

The authors state that they have no conflicts of interest.

References

1) Bartter T, Irwin RS, Nash G. Aneurysms of the pulmonary arteries. Chest 1988; **94**: 1065-75.
2) Deterling RA, Clagett OT. Aneurysm of the pulmonary artery; review of the literature and report of a case. Am Heart J 1947; **34**: 471-99.
3) Picard C, Parrot A, Boussaud V, et al. Massive hemoptysis due to Rasmussen aneurysm: detection with helicoidal CT angiography and successful steel coil embolization. Intensive Care Med 2003; **29**: 1837-9.
4) Ferretti GR, Thony F, Link KM, et al. False aneurysm of the pulmonary artery induced by a Swan-Ganz catheter: clinical presentation and radiologic management. AJR Am J Roentgenol 1996; **167**: 941-5.
5) Cantasdemir M, Kantarci F, Mihmanli I, et al. Emergency endovascular management of pulmonary artery aneurysms in Behçet's disease: report of two cases and a review of the literature. Cardiovasc Intervent Radiol 2002; **25**: 533-7.
6) Hoffmann RT, Spelsberg F, Reiser MF. Lung bleeding caused by tumoral infiltration into the pulmonary artery—minimally invasive repair using microcoils. Cardiovasc Intervent Radiol 2007; **30**: 1282-5.
7) Sirivella S, Gielchinsky I, Parsonnet V. Management of catheter-induced pulmonary artery perforation: a rare complication in cardiovascular operations. Ann Thorac Surg 2001; **72**: 2056-9.
8) Oguma T, Morise M, Harada K, et al. Pulmonary artery aneurysm/pseudoaneurysm, a delayed complication of lung abscess: a case report. Tokai J Exp Clin Med 2015; **40**: 86-9.