Case Report

Aberrant aneurysm of adult-type pulmonary sequestration: report of a case

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Abstract: Aberrant pulmonary artery exposed by high-blood pressure over a long period in adult-type pulmonary sequestration can be susceptible to arteriosclerotic change and aneurysmal formation. Dividing this aneurysmal artery has a risk of stump leakage and aneurysm. We herein report a 64-year-old man with an aberrant aneurysm of intralobar pulmonary sequestration. Thoracoscopic resection of the sequestrated lung was performed uneventfully by prior endovascular occlusion of the aberrant aneurysm.

Keywords: Pulmonary sequestration; adult; aberrant aneurysm; endovascular plug; video-assisted thoracic surgery (VATS)

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Introduction

Pulmonary sequestration is an interesting malformation of the lung, consisting with non-functional lung parenchymal supplied by systemic arterial flow. Pulmonary vessel in the adult-type pulmonary sequestration is affected over decades by much higher blood pressure than the ordinary pulmonary circulation. The vessels might be degenerated under such a high blood pressure. Surgical division of the aneurysmal artery has the risk of stapling failure and postoperative stump aneurysm. These complications can be avoidable by controlling the systemic blood flow prior to surgical procedure.

Case report

A 64-year-old man was referred to our hospital for evaluation of abnormal chest shadow detected at medical checkup. He had a 65-pack year of smoking history, and past medical history of hypertension. A chest computer tomography revealed intralobar sequestration in the right lower lobe and an aberrant aneurysm accompanied by dense atheromatous intima, 24 mm × 33 mm × 55 mm in size, arising from the celiac artery trunk. The blood flow of the sequestration drained into the right inferior pulmonary vein (*Figure 1*).

The patient was generally anesthetized under supine position. Intra-abdominal endovascular embolization at the proximal site of the aberrant artery to hemostat the blood flow into the aberrant aneurysm was performed by Seldinger technique via right femoral artery. A 16-mm diameter Amplatzer vascular plug (AVP) II (12-mm in length, AGA Medical Corporation, Plymouth, MN, USA) which was placed at the proximal site of the aberrant artery, 6 mm above its origin, completely occluded the blood flow into the aberrant aneurysm. Video-assisted thoracic surgery (VATS) was subsequently performed under a lateral position. Pulmonary ligament was divided carefully to expose the aberrant artery after peeling off the severe adhesions around the pulmonary pleura caused by chronic infection. The artery was uneventfully exposed, divided by an endoscopic linear stapler (Echelon flex; Echicon Endosurgery; Johnson & Johnson, Cincinnati, OH, USA) distally to the AVP. The sequestrated lung was then resected from the right lower lobe using endoscopic linear staplers (*Figure 2*). The surgical time of endovascular procedure and VATS were 91 and 136 minutes each. Total blood loss was 330 mL.

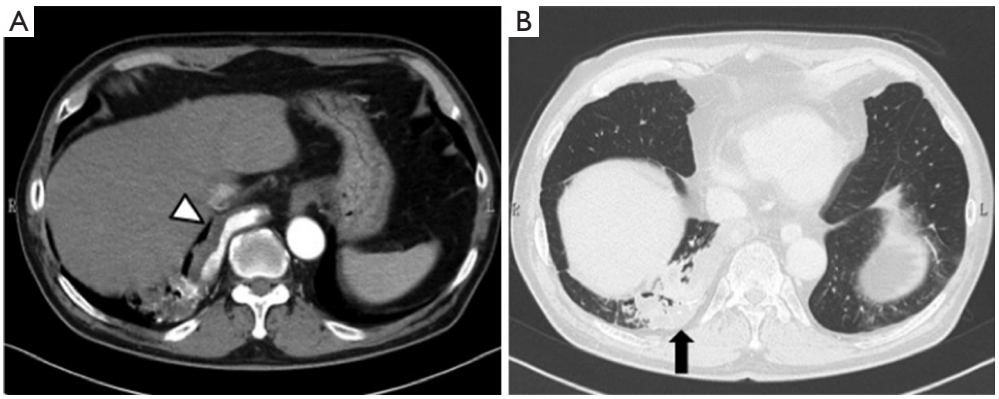

Figure 1 (A) Chest computer tomography showing an aberrant aneurysm with highly atherosclerotic change of the intimal. The artery is penetrating the diaphragm, running into the right lower lung (white arrow head); (B) intralobar sequestration is observed as a consolidation at the right lower lung (black arrow).

Figure 2 (Endovascular procedure) Arteriography showing an aberrant artery arising from the celiac artery trunk. The systemic blood flow is completely blocked by AVP placement. (VATS procedure) Diffuse adhesion around the sequestrated lung was observed. Pulmonary ligament was carefully separated to expose the aberrant aneurysm. The artery was safely divided using the stapling device (1). AVP, Amplatzer vascular plug. Available online: http://www.asvide.com/articles/466

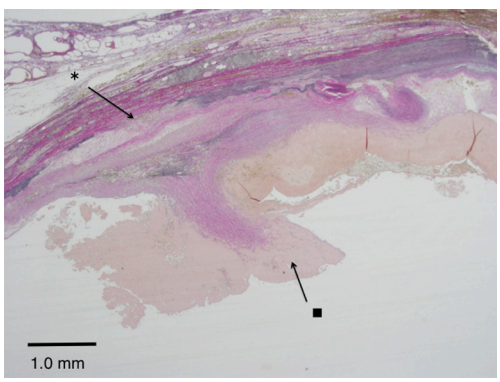

Figure 3 Pathological image of the aberrant aneurysm (elastica-van Gieson stain). Elastic layer of the tunica media is destroyed (*), accompanied with formation of the intraluminal plaque (■).

Postoperative course was uneventful. He was discharged from the hospital 8 days after the operation. Pathological examination revealed an intralobar bronchopulmonary sequestration. Aberrant aneurysm was accompanied with highly arteriosclerotic change and collapse of the arterial tunica intima (*Figure 3*).

Postoperative 3D-CT showed a complete occlusion of the blood flow in the residual aneurysm by the endovascular plug (*Figure 4*). He is doing well 5 months after operation.

Discussion

Pulmonary sequestration is a rare anomaly of all congenital pulmonary malformations, having an incidence of 0.15% to 1.8%. It is an anomalous condition of the lung, consisting with non-functional lung parenchymal supplied by systematic arterial flow. Pulmonary sequestration is often diagnosed in childhood, whereas pulmonary sequestration discovered in adult is extremely rare. Moreover, pulmonary sequestration accompanied by aneurysmal artery is rarely reported (2-5).

Pulmonary artery is a vessel with a fragile structure composing of approximately 30 elastic layers likely the abdominal aorta. Such a fragile wall structure sometime

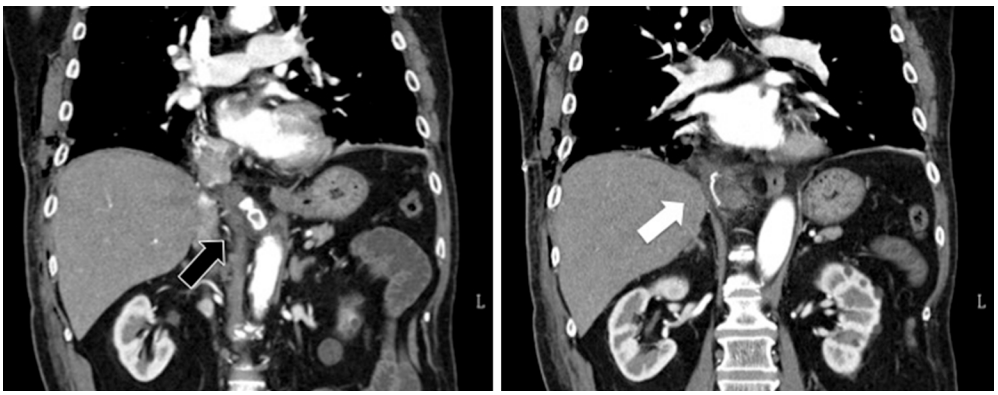

Figure 4 Postoperative contrast-enhanced computer tomogtraphy showing the AVP (black arrow) and the stapling line of the aberrant arterial stump (white arrow). Arterial stump is not imaged, demonstrating that AVP is successfully blocking the blood flow into the stump. AVP, Amplatzer vascular plug.

cause a life-threatening bleeding at the time of vascular division during surgery, even when the pulmonary artery is exposed by mean pulmonary artery pressure of 20 mmHg under ordinary pulmonary circulation. While the aberrant pulmonary artery is exposed to high blood pressure over a long time in the adult-type pulmonary sequestration, its intimal injury or intraluminal transportation of low-density lipoprotein can accelerate atherogenesis and aneurysmal formation likely the degenerated abdominal aorta characterized by atheroma and/or aneurysm (6). Pathological image of our case suggested degeneration of the vessel wall under high intraluminal pressure over decades. Division of this artery has a higher risk of rupture during surgical dissection or re-aneurysmal change of the arterial stump after operation. Such risks are avoidable by complete resection of the aberrant artery including its origin at the celiac artery, which requires additional laparotomy for arteries branching intra-abdominally. We herein reported that these troublesome could be substitutable by endovascular manipulation using the occlusion plug. The occluder blocks the systemic blood pressure and enables to complete safe division of the aberrant aneurysm by mechanical stapler under thoracoscopic surgery.

Previous articles report to perform endovascular occlusion few days to few weeks before surgery (7). However, performing vascular occlusion and surgery on the different day requires extra hospitalization and treatment costs, which is approximately 50,000-100,000 yen in Japan's National Health Insurance System. The complete occlusion of the high-pressure arterial circulation is reported to been achieved within 10-35 minutes using AVP (8). Our case had performed surgery just after the endovascular procedure, but no perioperative complication occurred. Therefore, these procedures are able to perform sequentially under single general anesthesia, which can reduce physical invasion and hospital costs.

In conclusion, adult-type sequestration may accompany a degenerated aberrant artery exposed by a high blood pressure. Arterial division during surgery is dangerous for its frailness, having a risk of stapling failure or development of stump aneurysm after surgery. Prior occlusion of the systematic blood flow by vascular plug reduces these risks, resulting in safe and secure thoracoscopic surgery.

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