

# A patient with life-threatening hemoptysis from a ruptured pulmonary artery aneurysm was successfully treated with endovascular coil interventions and a pulmonary lobectomy

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## Summary

Life-threatening hemoptysis due to ruptured pulmonary artery aneurysm is a rare emergency with a high mortality rate if not treated promptly. Computed Tomography Angiography (CTA) is an important diagnostic method to detect causes of hemoptysis, especially severe hemoptysis. Endovascular coil interventions and surgery are the cornerstones of management for pulmonary aneurysms and cessation of hemoptysis.

**Keywords:** Life-threatening hemoptysis, pulmonary artery aneurysm, emergency endovascular coiling, pulmonary lobectomy.

## I. BACKGROUND

Life-threatening hemoptysis is coughing up blood regardless of the amount of blood that causes hemodynamic disturbances or respiratory failure leading to death if left untreated<sup>1</sup>, there is no consensus on the amount of blood coughed up, but the authors suggest that coughing up at a rate of more than 100ml/hour or over 500ml/24 hours poses a life-threatening risk<sup>2</sup>. About 5-14% of patients who cough up blood will have life-threatening hemoptysis. The mortality rate from life-threatening hemoptysis is reported to be around 9-38%. About 90% of hemoptysis is from bronchial artery circulation due to high pressure. There are also other vascular sources from non-bronchial artery circulation such as: aorta, intercostal artery, subclavicular artery. In which the source from the pulmonary artery accounts for only about 5% of the total number of cases<sup>2</sup>. Pulmonary artery aneurysm

rupture is one of the very rare causes of hemoptysis. The prevalence according to a previous study based on an analysis of 109,571 autopsies found 8 cases of pulmonary aneurysms<sup>3</sup>, that is equivalent to a rate of about 0.0073%. The cause and pathogenesis are not completely clear. Pulmonary artery aneurysms are usually asymptomatic, some may manifest initially with hemoptysis. Massive hemoptysis and life-threatening hemoptysis are common symptoms, accounting for 20-60% of pulmonary aneurysm ruptures<sup>4</sup>. Currently, recommendations for surgery and definitive treatment of pulmonary aneurysms are not yet available. When there is a life-threatening hemoptysis, emergency endovascular intervention and surgery are important measures to save the patient's life. Below we report a clinical case of life-threatening hemoptysis caused by ruptured pulmonary aneurysm treated with endovascular interventions with coils and pulmonary lobectomy.

## II. CASE PRESENTATION

49 year-old male patient with a history of type 2 diabetes mellitus treated with metformin 1700mg/day and had no any previous lung disease

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and heart disease. Two weeks before admitted to the hospital, the patient had hemoptysis, about 2ml each time, 3-5 times a day and mild fever. The patient went to a local hospital and took a chest X-ray, revealed thick interstitial tissue and ground glass opacity in the base of the left lung. The initial diagnosis was pneumonia, which was treated with antibiotics and hemostatic medications. However, the hemoptysis continued.

After that, the patient suddenly coughed up a large amount of blood of about 200ml, then, he was admitted to the Emergency Department of 108 Military Central Hospital in a state of alertness, stable hemodynamic and no respiratory failure. The patient underwent airway control and took a pulmonary artery computed tomography scan, revealed a left lower lobe pulmonary aneurysm measuring 1.91 x 1.21 x 1.80cm, with ground glass opacity at base of the 2 lungs. The patient was quickly transferred to endovascular coilings.

During the procedure, the patient continued to cough up large amounts of blood again for approximate 300ml, causing respiratory failure. The patient was intubated to control airway and the procedure continued. After the coiling intervention the patient stopped hemoptysis. After the patient

was stable, he underwent a flexible bronchoscopy, the results showed no abnormalities in the airways, only little residual blood in the lower left lobe and the lumen was pushed inward corresponding to the coil placement. Patient had negative bacterial culture, negative microscopy of bronchial lavage fluid for AFB and QuantiFERON TB. Sars-CoV-2 and influenza test were negative. Blood clotting tests and echocardiogram were normal. Finally, the patient was diagnosed with life-threatening hemoptysis caused by an aneurysm of left branch of pulmonary artery.

4 days after the procedure, the patient coughed up blood again with the amount of about 150ml. The patient was re-evaluated by pulmonary artery CT scan immediately, suspected another small pulmonary aneurysm on the periphery of the left lung and had an extensive consolidation in lower lobe of the left lung due to blood clot. Therefore, the patient underwent surgery to remove the lower lobe of the left lung to solve both radically residual pulmonary aneurysm and large consolidation. After surgery, the patient had hemoptysis ceased for 7 days and had no complication from surgery and was discharged from the hospital.



Figure 1a

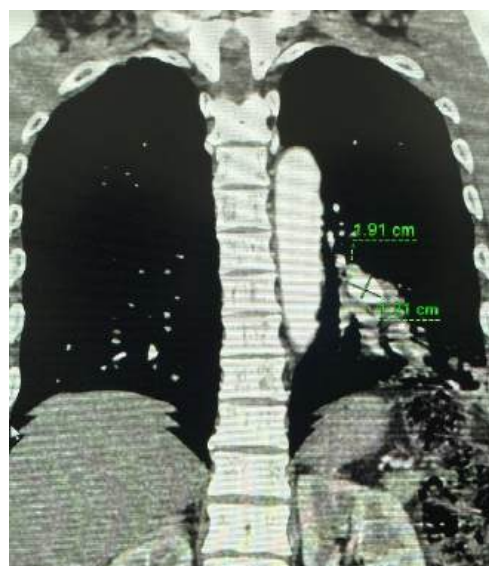


Figure 1b

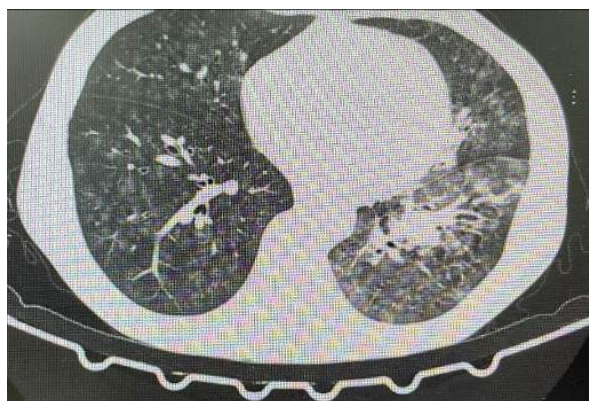


Figure 1c



Figure 1d

**Figure 1.** 1a: Left basal alveolar opacity on posteroanterior chest X-ray; 1b: Left lower lobe pulmonary artery aneurysm on coronal plane thoracic CT-scan. 1c: Mass in left lower lobe and ground-glass opacities in lower lobes of lung on chest-CT scan; 1d: left lower lobe pulmonary artery aneurysm on axial plane chest CT-scan.



Figure 2a



Figure 2b



Figure 2c

**Figure 2.** Endovascular intervention with coil into aneurysm. 2a: DSA image before the coiling; 2b: DSA image after coiling. 2c: Chest X-ray image after intervention (DSA: Digital Subtraction Angiography).

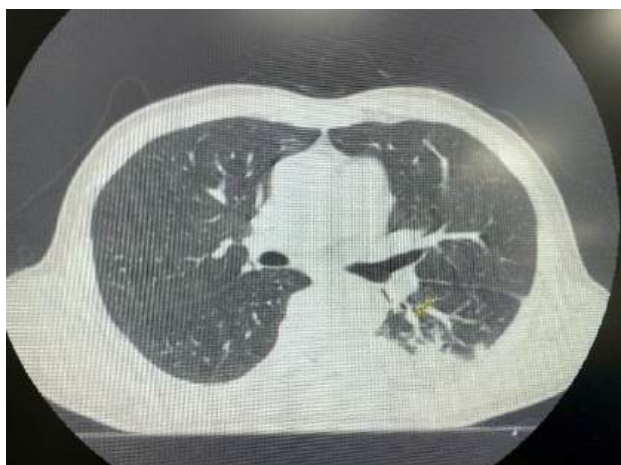


Figure 3a



Figure 3b

**Figure 3.** 3a: A suspicious small pulmonary aneurysm on the periphery of the left lung after being reevaluated by chest-CT scan. 3b: Extensive consolidation in the lower lobe of left lung due to blood clot.

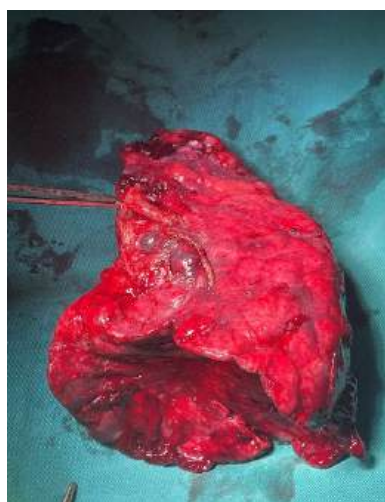


Figure 4a



Figure 4b



Figure 4c

**Figure 4.** 4a. A picture of the lower lobe of the left lung that has been surgically removed; 4b. The image of the coil was removed. 4c. Chest X-ray after removal of the left lower lobe of lung.

### III. DISCUSSION

Pulmonary aneurysm is defined by pulmonary artery dilation at least 1.5 times the normal size and damage to all 3 layers of the walls of blood vessels. Distinguishing from pulmonary pseudo-aneurysms when damage to less than 3 layers of artery walls<sup>5</sup>. About 89% of the aneurysm occurs in the pulmonary artery trunk, only 8% of the damage in the right pulmonary artery branch, and 3% of the damage in the left branch<sup>6</sup>. Our clinical case has a left lower lobe pulmonary aneurysm.

**Etiology and classification:** Pulmonary artery aneurysms can be divided into 2 main groups: Those with high endovascular pressure and those with low endovascular pressure<sup>6</sup>.

*High pressure pulmonary artery aneurysms include:*

Pulmonary artery aneurysms with pulmonary hypertension associated with heart disease, most commonly due to ductus arteriosus, interventricular septal abnormalities, and atrial septal abnormalities<sup>7</sup>. Generally these abnormalities are caused by left-right shunt formation that increases pressure on the vascular wall and forms an aneurysm<sup>8</sup>.

Pulmonary artery aneurysms with idiopathic pulmonary hypertension and pulmonary artery aneurysms with pulmonary hypertension due to other causes (such as chronic pulmonary arterial thrombosis)<sup>6</sup>.

*Low pressure pulmonary artery aneurysms include:*

Due to infections: Untreated syphilis and tuberculosis were often associated with pulmonary artery aneurysms. Pulmonary artery aneurysms in syphilis are usually in large pulmonary arteries. In tuberculosis, it usually forms in the lung parenchyma, called Rasmussen aneurysm<sup>5</sup>. Today, pyogenic bacteria such as *Staphylococcus* increase the risk of pseudo-aneurysms.

**Vasculitis:** Behcet is an unexplained systemic vasculitis, characterized by damage to the mucosa and skin, presenting in the eyes, and systemic vasculitis of both arteries and veins of all sizes. Pulmonary artery aneurysms are feature of this disease<sup>9</sup>. Hughes-Stovin syndrome, this is a very rare vasculitis that affects mainly large blood vessels, consisting of 3 main stages: stage I is thrombophlebitis. Stage II is the formation of multiple pulmonary artery aneurysms and stage III is the rupture of the aneurysms causing massive hemoptysis<sup>10</sup>.

Connective tissue diseases include Marfan syndrome and Loeys–Dietz syndrome (or osteoarthritis-aneurysm syndrome).

Pulmonary artery dilation after pulmonary artery valve stenosis.

Idiopathic pulmonary artery aneurysms: Being a rare form, this is the final diagnosis after excluding possible causes.

Patients we report had a normal echocardiogram should be in the low-pressure pulmonary artery aneurysm group. There were no signs of infection, tests for tuberculosis were negative. There were no other systemic manifestations of vasculitis or connective tissue disease. So, there was a high probability of an idiopathic aneurysm.

About clinical manifestations: Usually no clinical symptoms or nonspecific clinical symptoms, which could be seen in many different diseases such as:

Symptoms caused by aneurysms compressing the surrounding structure such as bronchial compression causing shortness of breath, cough, post-obstructive pneumonia, bronchiectasis.

Hemoptysis due to rupture of an aneurysm: Amount of blood of 100-1000ml per day are considered massive<sup>1</sup>. There are many causes of hemoptysis such as bronchiectasis, tuberculosis, fungal infections, lung cancer. In which, vascular abnormalities are one of the possible causes of life-threatening hemoptysis. However, hemoptysis caused by vascular abnormalities accounts for only about 0.2% of all causes of hemoptysis<sup>13</sup>. Our clinical case had life-threatening hemoptysis from a ruptured pulmonary artery aneurysm.

Symptoms of aneurysm dissection: This is a rare symptom that can cause chest pain, cardiogenic shock and sudden death.

Symptoms of the cause of the aneurysm: depends on each cause.

Diagnosis: Diagnosis of pulmonary artery aneurysm by pulmonary angiography, however this is an invasive technique and only lumen of aneurysm is identified. Currently, pulmonary artery CT scans have helped confirm the diagnosis, assess thrombosis and other abnormalities of the walls of blood vessels.

Treatment: There are currently no clear recommendations for the treatment of pulmonary artery aneurysm, treatment methods may include:

Conservative treatment can be applied to asymptomatic cases, the absence of significant pulmonary hypertension and stable size. Some authors suggest conservation for cases of aneurysm diameter < 6cm<sup>12</sup>. Conservative treatment is the medical treatment of underlying causes such as immunosuppressants for vasculitis, anti-tuberculosis drugs, antibiotics for syphilis.

Endovascular intervention: Various types of intervention materials such as coils, glues, covered stents, detachable balloons, vascular plug and PDA closure devices, depending on the anatomical position and characteristics of the aneurysm, there are many different endovascular embolization procedures<sup>13</sup>. Treatment of endovascular intervention is one of the fairly new approaches, especially applied to small peripheral branches. The advantage of the aneurysm coiling is that it can retain many peripheral pulmonary artery branches and preserve lung function in the distal segment of the aneurysm<sup>14</sup>. Our patients had an aneurysm in the left pulmonary artery branch, so initial treatment option with the aneurysm coiling was reasonable and feasible. According to the medical literature, the initial success rate was 95%. However, placing coils within an aneurysm carries a potential risk of rupture. Care must be taken to avoid overpacking or advancing the catheter and coils through the aneurysm wall<sup>14</sup>. In our case, the picture after surgery showed, coiling still worked well.

Surgical treatment remains the foundational treatment for pulmonary artery aneurysms, and there is insufficient evidence of a pulmonary trunk aneurysm diameter threshold for surgery<sup>7</sup>. Surgery is usually performed in cases of large aneurysms (> 6cm), which are particularly symptomatic in patients with signs of right ventricular dysfunction and pulmonary hypertension<sup>12</sup>. In cases of focal pulmonary artery dilation, the aneurysm may be surgically removed. In cases of diffuse dilation, the entire trunk of the pulmonary artery is replaced by an artificial or allogeneic artery segment. However, peripheral pulmonary aneurysms are more difficult

and often require a pulmonary resection and the risk of death is higher. The mortality rate of medical and surgical treatment for massive hemoptysis can range from 35-100%<sup>14</sup> which should be performed in facilities equipped with experienced surgeons. However, our patient was still coughing up large amounts of blood after 4 days of endovascular intervention due to a suspicious small pulmonary aneurysm and a total consolidation of lower lobe of left lung by blood clot, so surgery planned. The patient underwent a safe left lower lobectomy, which completely stopped hemoptysis after that with no complication.

#### IV. CONCLUSION

Pulmonary artery aneurysm is a rare medical condition with many potential risks. Massive hemoptysis caused by a ruptured pulmonary artery aneurysm is one of the life-threatening complications that can cause rapid death. Endovascular intervention with the coils is a method that can be performed quickly in cases of peripheral aneurysms, with a high initial success rate. Surgery is still the foundational method for treatment of pulmonary artery aneurysms, stopping hemoptysis.

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