

HEMOPTYSIS SECONDARY TO PULMONARY ARTERY-TO-INTERCOSTAL ARTERY FISTULA: A CASE REPORT

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ABSTRACT

Intercostal artery-pulmonary artery fistulas can be congenital or may occur due to trauma, neoplasms, and inflammation. These fistulas are usually asymptomatic, but they may occasionally give rise to emergencies presenting with symptoms such as hemoptysis. This case report presents a 35-year-old male patient with a history of acute tubular necrosis, chronic kidney failure, and tuberculosis who is receiving hemodialysis treatment. The patient, who was admitted to the hospital with an episode of hemoptysis lasting three days, underwent chest tomography and angiography examinations, and it was revealed that there was a fistula between the branches of the left pulmonary artery and the left eighth intercostal artery. This rare fistula could have caused potentially fatal complications in our dialysis patient receiving anticoagulation therapy. The interventional radiology team decided to choose surgical treatment after evaluating the patient in a council meeting, and the patient was taken into surgery. Surgical pulmonary resections can be successfully performed in the curative treatment of this rare disease.

Keywords: Arterio-arterial fistula, hemoptysis, tuberculosis

INTRODUCTION

Intercostal artery-to-pulmonary artery fistulas are congenital or acquired abnormal connections between the systemic artery and the pulmonary artery. This rare condition can be caused by trauma, neoplasms, or inflammation. Since the disease is usually asymptomatic, it is usually detected incidentally. In addition, the disease can cause symptoms in the form of hemoptysis, dyspnea, and heart failure (1).

A contrast-enhanced computed tomography (CT) scan, angiography, and radionuclide angiogram are used for a definitive diagnosis (2-4). Additionally, magnetic resonance velocity mapping, which is a non-invasive imaging technique of great interest in guiding the diagnosis of arteriovenous fistulas, can also be used. Selective arterial angiography is useful both in confirming the diagnosis and in treatment (5).

The treatment method alternatives include embolization of the responsible vessels, ligation of fistulas, or pulmonary lobectomy (2, 3). Post-treatment follow-up is necessary as the disease can

cause adverse hemodynamic effects, bacterial vegetation, and rupture (6). In this article, we aim to present a very rare case of connection between the intercostal artery and the pulmonary artery.

CASE REPORT

A 35-year-old male patient was admitted to our hospital due to massive hemoptysis that had been going on for three days. He also reported that he had been experiencing coughing and left-side chest pain for four to five months. Physical examination findings were normal, and the patient's history revealed that acute tubular necrosis had developed after six months of tuberculosis treatment, followed by the onset of chronic kidney failure. The patient stated that he had been receiving hemodialysis treatment for two years. Approximately 200 cc of hemoptysis not resulting in asphyxia was identified. A chest CT scan was performed on the patient who applied to the emergency department with massive hemoptysis. Considering



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the possibility of arterio-arterial fistula (AAF), the patient was admitted to the intensive care unit for follow-up. In the angiography, abnormal high-speed flow was detected in the fistula between the left pulmonary artery branches and the left eighth intercostal artery (Figure 1). Furthermore, a connection with the common basal artery was observed at the level of the intercostal artery fissure. Bleeding was controlled with embolization, then the patient developed pneumonia. During intensive care follow-up, a catheter was inserted to administer tazobactam for pneumonia treatment. Subsequently, due to the patient's persistent fever and progression observed in the posterior-anterior chest X-ray, along with a potential catheter infection, the antibiotic regimen was altered to linezolid and meropenem for two weeks. The interventional radiology team checked on the patient after their antibiotic treatment was discontinued. As the condition remained stable throughout follow-ups, the patient was discharged with a recommendation for a follow-up appointment at the outpatient clinic 10 days later. The case was evaluated in a multidisciplinary pulmonary council with the participation of a thoracic surgeon, pulmonologist, medical oncologist, nuclear medicine specialist, and radiologist. The interventional radiology team found re-endothelialization of the fistula risky due to the potential to cause lung infarction and decided that surgical treatment would be appropriate for the patient. Fiberoptic bronchoscopy was performed before surgery and hemoptysis was observed from the lower lobe bronchus of the left lung. In addition, lobectomy was preferred as a surgical procedure because the anastomosis was seen at the level of the common basal artery and intercostal artery fissure in the thorax CT.

Four months after the attack of hemoptysis, the patient was operated on under general anesthesia with left selective



Figure 1: Angiography view of the AAF between the left pulmonary artery branches and the eighth intercostal artery, marked with the blue arrow. AAF: Arterio-arterial fistula

intubation in the right lateral decubitus position. Using a left posterolateral thoracotomy incision, the thorax was accessed through the fifth intercostal space. During exploration, an intercostal artery connection was detected in the posterior part of the left lower lobe. When the artery was seen extending into the parenchyma, the fistula was closed using a LigaSure device, and it was decided to perform a left lower lobectomy. Adhesions in the lung were separated using blunt and sharp dissection techniques. Subsequently, the pulmonary ligament was released, completing the pneumolysis. The fissure was separated with blunt and sharp dissection. The inferior pulmonary vein was ligated and divided. The branches of the pulmonary artery supplying the lower lobe were ligated and divided. A laceration incurred during pneumolysis was repaired. After controlling bleeding and leaks, a thoracic drain was placed, and the layers were closed. Early complications were not detected. Furthermore, the pathological examination of the left lower lobectomy specimen was performed. The totally resected specimen, measuring 16.5x9x2.8 cm, had a pink cut surface. Macroscopically, no mass lesions were detected. Subpleural hemorrhagic areas measuring 4.2x3.5 cm were observed on the anti-hilar side of the outer surface. In microscopic examination, acid-fast bacilli were negative and there was no growth in culture. No fistula was detected in the pathological specimen. No neoplasm was found in the examination. The patient was discharged with satisfaction on the sixth postoperative day.

In the second month after surgery, the patient was admitted to the emergency department again with hemoptysis. During bronchoscopy, minimal bleeding due to irritation was observed in the upper lobe of the right main bronchial system. We did not see any bleeding areas around the bronchial stump that could cause hemoptysis. After confirming the absence of active bleeding, 0.5 g tranexamic acid was administered to the patient three times a day via infusion for five days. Subsequently, the administration was converted to an intravenous push as needed, and the patient was kept under supervision. Additionally, piperacillin-tazobactam 3x4.5 g antibiotic treatment was administered for seven days. Afterward, no pathology was found in the thorax CT. He was discharged due to the improvement of his medical condition. No complaints were observed in the postoperative period. The patient is still being followed up without hemoptysis in the sixth postoperative month.

DISCUSSION

In a typical situation, there is no direct connection between the intercostal artery and the pulmonary artery. However, abnormal connections called fistulas may occur due to trauma, infection, idiopathic factors, and congenital causes (6, 7). These triggering factors cause blood to form a fistula by following the fistula path with lower resistance instead of capillaries (8). These fistulas frequently cause no symptoms and are often found incidentally during medical investigations. Rarely, in cases of rupture of the fistula, massive hemoptysis can occur in the patient. Hemoptysis is a symptom that can be described

as bleeding from the trachea-bronchial system during coughing. Massive hemoptysis is defined as bleeding of more than 150 mL in 24 hours or bleeding rate >100 mL/hour (9). This may be caused by conditions, including tuberculosis, bronchiectasis, or arterio-venous malformations (10). This can lead to life-threatening complications such as airway obstruction, hypoxia, or hemodynamic instability (9). Therefore, it is necessary to take this situation seriously and start treatment immediately. We think that hemodialysis played an important role in the massive hemoptysis experienced by our patient with chronic renal failure. This was primarily attributed to the fact that the anticoagulants administered to the patient increased the blood flow rate.

To diagnose a pulmonary-intercostal artery fistula in a patient experiencing hemoptysis, chest X-ray and CT angiography are used as diagnostic methods. In these techniques, lung infiltration due to the indentation under the rib and signs of bleeding can be observed. However, these techniques cannot conclusively establish the diagnosis. Digital subtraction angiography is a safe diagnostic method that determines the correct location of intercostal-pulmonary artery fistulas and clearly visualizes the presence and size of these fistulas (7). Additionally, in life-threatening hemoptysis, the use of CT together with fiberoptic bronchoscopy can provide more effective results (11). Fiberoptic bronchoscopy allows visualization of the bleeding area and can provide treatment if bleeding continues (12). The use of fiberoptic bronchoscopy becomes even more important in ensuring airway control and in patients with bilateral lung disease (11).

In a study involving 348 patients treated for either moderate recurrent or life-threatening hemoptysis with bronchial artery embolization, active tuberculosis was found in 27% of these patients and tuberculosis sequelae were found in 29.9% (13). During angiography performed on these patients, it was observed that 14% of the patients had fistulas between the pulmonary artery and intercostal artery, as in our case (13).

When we examined the literature from a pathological perspective, we saw that patients with previous tuberculosis disease had conditions that resulted in fistula formation, recurrent hemoptysis, and death due to a decrease in functional lung parenchymal volume (14). These results confirmed that tuberculosis sequelae can cause fistula formation and recurrent hemoptysis, even if there is no active tuberculosis as in our patient (14).

In a literature review, 15 intercostal-pulmonary artery fistula cases were compiled. Tuberculosis played a role in three of these patients with known etiologies. The inflammatory reaction can cause increased pulmonary capillary permeability in the lungs and the infiltration of blood cells into the alveoli due to *Mycobacterium tuberculosis* toxins and massive sensitizers. Necrosis of lesion tissue due to caseous necrosis can lead to erosion and vessel damage. In the circular veins around the bronchial arteries, bronchiectasis and occlusion occur together (7).

As a result, inflammation and angiogenesis caused by tuberculosis, bleeding diathesis caused by hemodialysis, and heparin used during dialysis may have caused hemoptysis in our patient. However, the fact that fistula cases in the reported literature are detected at a young age also indicates that these may be rare congenital anomalies (15). In our patient, as a source of recurrent hemoptysis after surgery, no fistula or any cause was found. Since the condition did not recur after medical treatment, further examination could not be performed.

Embolization treatment is the preferred treatment method in AAFs because it is less invasive, does not require anesthesia, and causes less trauma. However, embolization has a high risk of recurrence. Various surgical treatments such as lobectomy, segmentectomy, and pneumonectomy have been reported. To preserve normal lung parenchyma, vascular dissection and peripheral lung resection can be performed (16). In life-threatening cases, wedge resection may be preferred as a fast and simple method to stop massive hemoptysis (17). Since our case carries a risk of recurrence, lobectomy was preferred as the treatment method. Surgical treatment is used in recurrent cases and multiple vascular abnormalities. This was the situation with our patient, taken into surgery and successfully discharged after undergoing a left lower lobectomy.

CONCLUSION

Pulmonary AAF is a very rare situation that can cause life-threatening complications. Selective arterial embolization is necessary both to confirm the diagnosis and to provide embolization to appropriate patient groups. Surgical pulmonary resections can be successfully performed in curative treatment.

Ethics Committee Approval: N/A

Informed Consent: Informed consent was obtained from the patient.

Conflict of Interest: The authors declared no conflict of interest.

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