

## A Case of Pseudoaneurysm Due to Fungus Ball within the Progressive Massive Fibrosis in a Patient with Pneumoconiosis: Computed Tomography-Pathologic Correlation

진폐증 환자에서 진행성거대섬유화증 내에 생긴 진균구에 의한  
가성동맥류 일 예: 전산화단층촬영과 병리소견

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We reported a case of pseudoaneurysm in the upper lobar branch of the right pulmonary artery, which was caused by a fungus ball within the progressive massive fibrosis (PMF) in a patient with pneumoconiosis. Coil embolization of the pseudoaneurysm initially stopped the bleeding. After right upper lobe lobectomy to prevent the recurrent hemoptysis, pathology confirmed pseudoaneurysm within the PMF due to aspergilloma.

### Index terms

Aneurysm  
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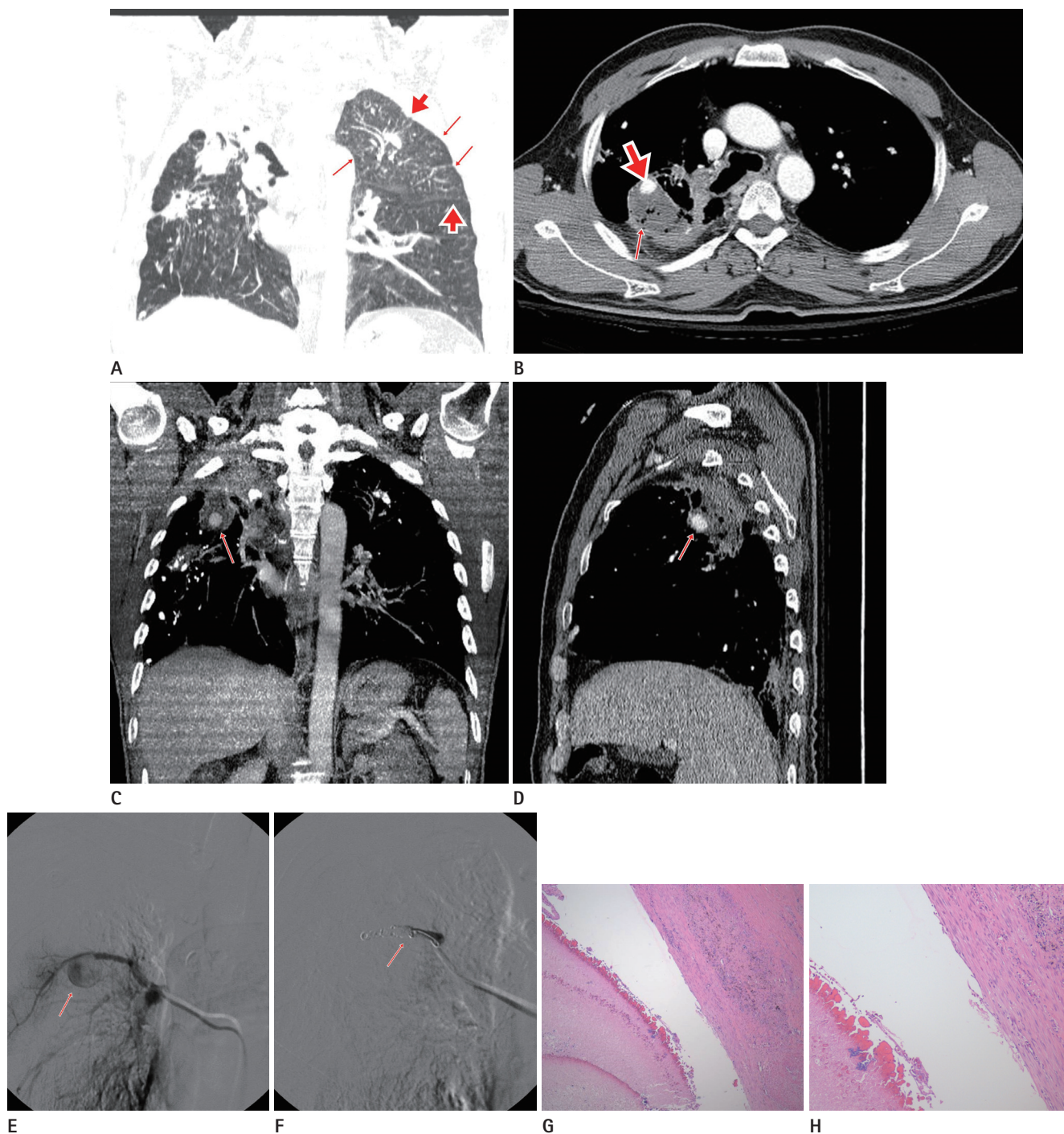
## INTRODUCTION

While uncommon, pulmonary artery aneurysms and pseudoaneurysms are associated with high mortality. Overall they are considered rare entities with prevalence rates of around 1 case per 14000 to 100000 autopsies (1, 2). Most pulmonary artery aneurysms and pseudoaneurysms are acquired and associated with cardiovascular disease, infection, iatrogenic causes, trauma, neoplasm, and connective tissue disease. To our knowledge, while there are many cases of Rasmussen aneurysm in patients with cavitary tuberculosis, there have been no reports of pseudoaneurysm due to fungus ball in the progressive massive fibrosis (PMF) of a patient with pneumoconiosis. We described a case of pseudoaneurysm detected by contrast-enhanced chest CT with three-dimensional (3D) reconstruction. The pseudoaneurysm arose in

the upper lobar branch of the right pulmonary artery and caused by a fungus ball within the PMF in a patient with pneumoconiosis, who underwent transcatheter endovascular embolization of the aneurysm.

## CASE REPORT

The patient was a 51-year-old man with a history of pneumoconiosis, tuberculosis, and hypertension. He was a coal miner for 12 years and had a 30 pack-year smoking history. He initially presented to another hospital with an episode of pneumonia and was transferred to our hospital for recurrent episodes of massive hemoptysis of more than 1 liter per day. On hospital admission, the patient was afebrile and hemodynamically stable (134/85 mm Hg, 106 beats per minute, 20 breaths per minute, and tempera-



**Fig. 1.** Pseudoaneurysm due to fungus ball within the progressive massive fibrosis in a 51-year old patient, presenting with hemoptysis.

**A.** Coronal reconstruction image (lung window) shows multiple well-defined centrilobular nodules, mainly distributed in the corticomedullary junction of the lung (small arrows) suggestive of pneumoconiosis. Also in addition, multiple small costal pleural and fissural nodules (large arrows) indicate perilymphatic distribution of pneumoconiotic nodules.

**B.** Contrast-enhanced CT scan (mediastinal settings, 2.5 mm collimation) shows focus of enhancement suggesting pseudoaneurysm of segmental pulmonary artery in right upper lobe (large arrow). Spongiform air bubble suggesting a fungus ball within progressive massive fibrosis (PMF) (small arrow).

**C, D.** Coronal and sagittal reconstruction images show pseudoaneurysm (arrow) of right upper lobe by fungus ball in PMF.

**E.** Right pulmonary artery selective angiography shows a pseudoaneurysm (arrow) of segmental pulmonary artery in right upper lobe.

**F.** Transcatheter embolization was performed with use of 4-5 mm coils (arrow) via the right femoral venous approach.

**G, H.** Hematoxylin & eosin stain ( $\times 40$ ) and B ( $\times 100$ ) show fungus ball in the cavity of PMF.

ture of 36.8°C). The initial laboratory results showed anemia, mild leukocytosis (hemoglobin: 7.6 g/dL, hematocrit: 22.7%, and white blood cell count: 10300/ $\mu$ L) and negative three sputum acid fast bacillus smears and culture. Contrast-enhanced chest CT with 3D reconstruction (LightSpeed 16; GE Medical Systems, Milwaukee, WI, USA) showed multiple perilymphangitic nodules suggestive of pneumoconiosis (Fig. 1A), spongiform air bubbles indicating a fungus ball within a PMF, and a tuberculous scar with volume loss and traction bronchiectasis. There was no three-in-bud or centrilobular nodule, cavitary nodule, lymphadenopathy, or effusion that was suggestive of active tuberculosis. Within the fungus ball, a pseudoaneurysm was noted in the upper lobar branch of the right pulmonary artery (Fig. 1B-D). The lack of a draining vein ruled out an arteriovenous malformation. Emergent pulmonary artery angiogram and transcatheter embolization were performed with 4–5 mm coils (Tornado; Cook, Bloomington, IN, USA) via a right femoral venous approach (Fig. 1E, F). Particle embolization (Contour; Boston Scientific, Natick, MA, USA) of the right intercostobronchial trunk and right 1st–2nd intercostal artery arising from the costocervical trunk were also performed due to arterial hypertrophy. Following these procedures, the hemoptysis stopped, but recurred at not more than 20 cc per day after one week. Because of recurrent hemoptysis and deteriorating pulmonary function test results, a right upper lobe lobectomy was performed for the pulmonary aspergilloma with pulmonary artery pseudoaneurysm. Although the presence of cavitation in a conglomerate mass is an important indication of associated tuberculosis, the pathologic specimen of this case showed fungal hyphae within the PMF cavity without chronic granulomatous inflammation suggesting tuberculosis (Fig. 1G, H). Polarized light images showed scattered interstitial silica particles. The postoperative period was uneventful and the patient was discharged in the fourth postoperative week.

## DISCUSSION

Pulmonary artery aneurysms and pseudoaneurysms can be congenital or acquired. Common congenital causes include vessel wall deficiency, valvular and postvalvular stenosis, and increased flow due to left-to-right shunts. Common acquired causes include trauma, pulmonary artery hypertension, vasculitis (Behçet's syndrome and Hughes-Stovin syndrome), infection (my-

cotic aneurysms and pseudoaneurysms), neoplasm, iatrogenic causes (malpositioned Swan-Ganz catheters), and connective tissue abnormalities such as Marfan syndrome, Ehlers-Danlos syndrome, and cystic medial necrosis (3, 4). There have been many reports of pulmonary artery pseudoaneurysm due to tuberculosis or fungus infection. Also, fungus infection is well known in patients with pneumoconiosis. To the best of our knowledge, there have been no reports of pseudoaneurysm due to fungus ball in the PMF of a patient with pneumoconiosis. Therefore, we reported the first case of pseudoaneurysm due to fungus ball in the PMF of a patient with pneumoconiosis.

The cavity formation within the PMF suggests ischemic necrosis due to deficient blood supply (5). PMF greater than 4 cm may show cavitation due to ischemic necrosis. Tuberculosis is another possible cause of cavitation in PMF. The presence of cavitation in a conglomerate mass (PMF) is an important indication of associated tuberculosis (6). Kato et al. (7) proposed that chronic persistent or progressive upper lobe infiltrates and cavities in patients with pneumoconiosis should raise the possibility of chronic necrotizing pulmonary aspergillosis. Preexisting diseases associated with aspergilloma include tuberculosis, bronchiectasis, pneumoconiosis, and sarcoidosis (8). Aspergillomas occur predominantly in the upper lobes, reflecting a predilection for cavity formation at this site, because the relative imbalance in perfusion and ventilation in the lung apices provide an oxygen-rich environment for micro-organisms (9).

Our case showed spongiform air bubbles in the PMF cavity suggestive of fungus ball. Ischemic necrosis of PMF due to deficient blood supply might cause cavitation of PMF and subsequent fungal hyphae growth in the PMF cavity. Mycotic pulmonary artery pseudoaneurysms are caused by several mechanisms such as direct extension of inflammation to involve the vessel wall, endovascular seeding of the vessel wall from bronchial arteries in septicemia and intimal invasion of the vessel wall from septic embolism. In our case, it is likely that the fungal hyphae eroded and weakened the branch of the pulmonary artery, leading to the vessel pseudoaneurysm.

CT is useful for diagnosing diseases that cause massive hemoptysis, localizing bleeding sites, and selecting vessels to be embolized (10). Contrast-enhanced chest CT with 3D reconstruction scan helped us to detect pseudoaneurysm of the pulmonary artery in PMF, because the enhancement density of the pseudoan-

eurysm of pulmonary artery was similar to that of the aorta. After detecting the cause of massive hemoptysis such as pseudoaneurysm, we suggest that embolization of pseudoaneurysm due to fungus ball within the PMF is necessary to stop abrupt massive pulmonary bleeding. After that, surgical removal of PMF with fungus ball is the best way to prevent recurrent hemoptysis.

In conclusion, we reported a case of pseudoaneurysm due to fungus ball in the PMF of a patient with pneumoconiosis. Assessment with contrast-enhanced CT with coronal reconstruction images allowed accurate evaluation of pulmonary artery pseudoaneurysm, facilitating prompt diagnosis and treatment.

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## 진폐증 환자에서 진행성거대섬유화증 내에 생긴 진균구에 의한 가성동맥류 일 예: 전산화단층촬영과 병리소견

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저자들은 진폐증 환자에서 진행성거대섬유화증 내에 생긴 진균구에 의한 우상엽 폐동맥의 가성동맥류 증례를 보고하는 바이다. 코일 색전술로 가성동맥류에 의한 객혈은 일시적으로 지혈되었다. 진행성거대섬유화증 내에 국균종에 의해 생긴 동맥류는 반복적으로 재발한 객혈치료를 위해 시행된 우상엽 절제술을 통해 병리적으로 확진되었다.

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