

[ CASE REPORT ]

## A Tuberculous Bronchial Artery Aneurysm with Abnormal Findings on Autofluorescence Imaging Bronchoscopy

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### Abstract:

Pulmonary tuberculosis is a common disease that may result in hemoptysis. Fetal hemoptysis is known to be related to the rupture of a pulmonary aneurysm formed in the cavity wall. We herein report a case of non-cavity pulmonary tuberculosis that developed with massive hemoptysis following bronchial artery aneurysm. Bronchial artery embolization was performed, and autofluorescence imaging bronchoscopy was conducted one month after the anti-tuberculosis treatment. Bright-green color was observed in the ulcerative lesion with a white coat, corresponding to the bronchial artery aneurysm. This is the first report of the autofluorescence imaging observation of an ulcerative lesion caused by bronchial tuberculosis.

**Key words:** pulmonary tuberculosis, hemoptysis, bronchial artery aneurysm, autofluorescence imaging bronchoscopy

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### Introduction

Hemoptysis is a common respiratory emergency in daily practice, and the frequency of hemoptysis owing to pulmonary tuberculosis is 6.8% (1). An active pulmonary cavity lesion with the formation of pulmonary aneurysms (Rasmussen aneurysms) is a causative factor of massive hemoptysis owing to pulmonary tuberculosis (2). However, massive hemoptysis rarely but occasionally occurs in non-cavity pulmonary tuberculosis through the formation of an aneurysm.

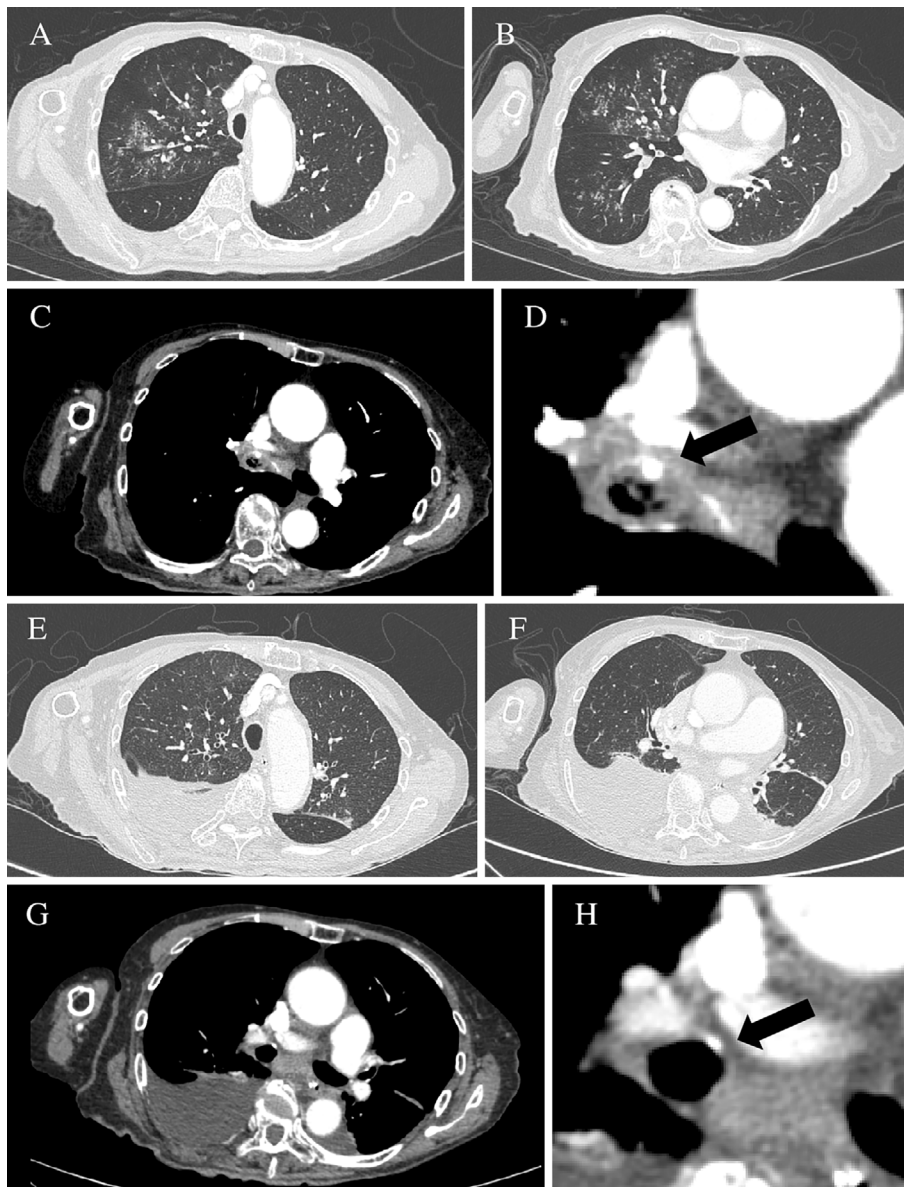
Autofluorescence imaging bronchoscopy, which combines autofluorescence with two reflected light signals, is useful for detecting early lung cancer and bronchial dysplasia (3). However, the epithelial layer changes associated with other diseases have not been completely investigated.

### Case Report

An 80-year-old never-smoker woman had a history of stroke. In May 20XX, she developed dry cough that gradually worsened. In August 20XX, she suddenly developed massive hemoptysis and was rushed to our hospital. Her body temperature was 37.0°C, and her percutaneous arterial

blood oxygen saturation was 99% on inhaling 10 L/min of oxygen via a reservoir mask. Breathing sounds in the right side of the chest were attenuated. Contrast-enhanced chest computed tomography on admission revealed centrilobular granular shadows with ground-glass opacities around the entire right lung field; however, no cavity lesions were found (Fig. 1A, B). A 4-mm-bronchial artery aneurysm was found in the anterior wall of the right main bronchus (Fig. 1C, D). Laboratory test results revealed a white blood cell count of 5,900/ $\mu$ L with 71.7% neutrophils and 20.3% lymphocytes, C-reactive protein level of 0.2 mg/dL (normal <0.3 mg/dL), and serum lactate dehydrogenase (LDH) level of 148 IU/L (normal, 119-229 IU/L). Electrolytes, creatinine, liver function tests, and coagulation were normal.

Tracheal intubation was performed on the day of hospitalization, and ventilation was initiated. Angiography revealed a dilated and meandering right bronchial artery; however, the aneurysm was unclear (Fig. 2A). After embolization with gelatin sponge strips, the dilated tortuous blood vessel shadows disappeared (Fig. 2B). A large amount of clotted blood obstructed the right bronchus, and the bronchoscope was used to aspirate as much as possible, but the obstruction of the middle lobe could not be released. No active hemorrhaging was detected after bronchial artery em-



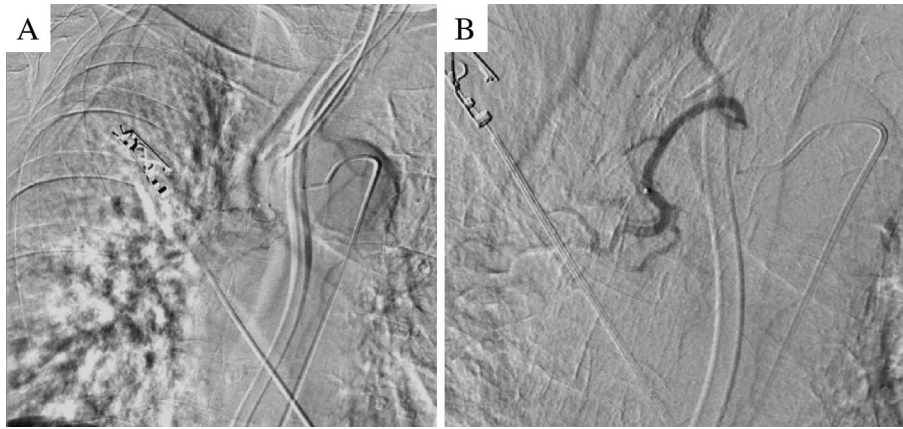
**Figure 1.** Contrast-enhanced chest computed tomography on admission revealed centrilobular granular shadows around the entire right lung field (A, B) and a 4-mm-bronchial artery aneurysm (arrow) in the anterior wall of the right main bronchus (C, D). After bronchial artery embolization and tuberculosis treatment, the shadow in the lung field improved, and the bronchial aneurysm shrank (E-H, arrow).

bolization, and ventilation was discontinued at four days after the disease onset.

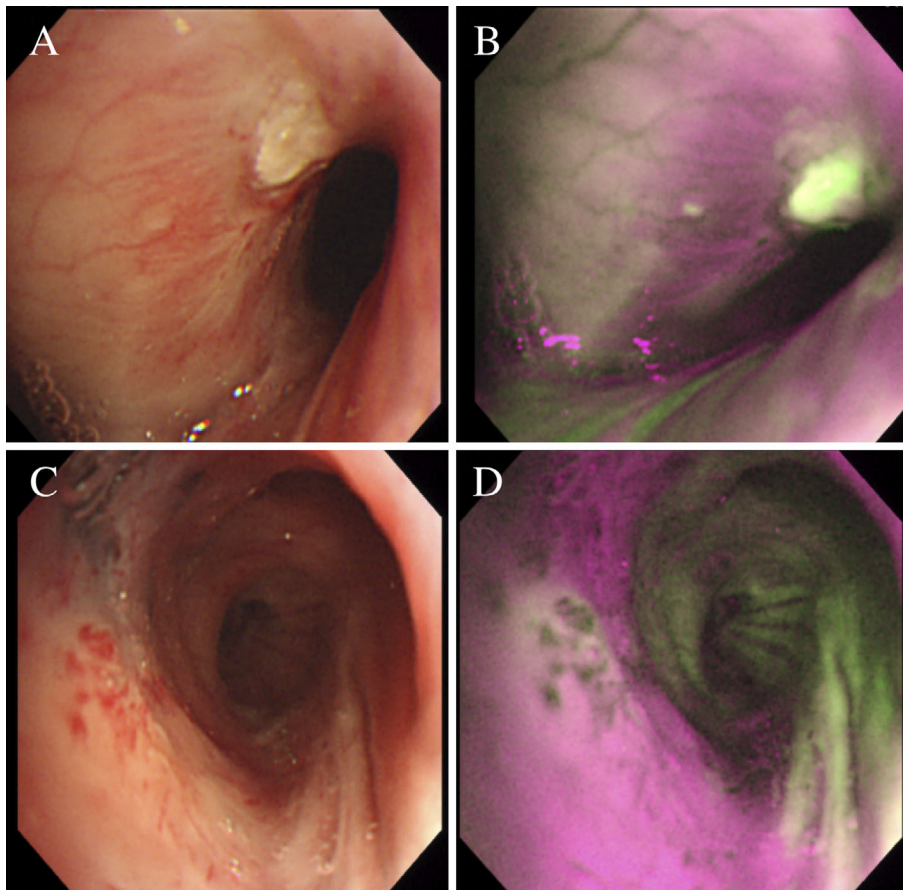
On the day of the hospital visit, examination results of intra-tracheal specimens were negative for acid-fast bacilli, but on the fifth hospitalization day, polymerase chain reaction revealed the presence of *Mycobacterium tuberculosis*, so rifampicin, isoniazid, ethambutol, and pyrazinamide were initiated. One month after initiating the anti-tuberculosis treatment, contrast-enhanced computed tomography revealed right-sided pleural effusion, which may have developed because of bronchial obstruction owing to blood clots; however, the centrilobular granular shadows disappeared from the entire right lung (Fig. 1E, F), and no aneurysm was observed in the right bronchus (Fig. 1G, H). The right pleural

effusion then gradually decreased.

Bronchoscopy performed during artificial respiratory management did not allow sufficient observation due to hemoptysis, so one month after the start of tuberculosis treatment, bronchoscopy was performed again to check the condition of the airway epithelial layer at the bleeding source and the presence of airway scar stenosis. Bronchoscopy (EVIS LUCERA SPECTRUM, Olympus BF-F260 autofluorescence bronchovideoscope; Olympus, Tokyo, Japan) showed stenosis and an ulcerative lesion with a white coat in the right main bronchus. The site of the ulcerative lesion and the bronchial aneurysm coincided, and on autofluorescence imaging bronchoscopy, the red part of the epithelial layer was colored magenta, and the ulcerative lesion was



**Figure 2.** Angiography findings. Dilated and meandering right bronchial arteries were detected, but the aneurysm was unclear (A). After embolization with gelatin sponge strips, the dilated tortuous blood vessel shadows disappeared (B).



**Figure 3.** The bronchoscopy findings. The site of the ulcerative lesion and the bronchial aneurysm coincided, and on autofluorescence imaging bronchoscopy, the red part of the epithelial layer was colored magenta, and the ulcerative lesion was autofluorescent in bright green. Observation image of the right main bronchus from the trachea (A, B) and of bifurcation of right main bronchus (C, D).

autofluorescent in bright green (Fig. 3). The patient continued treatment without serious side effects but developed muscle atrophy associated with long-term bed rest and was transferred to another hospital for rehabilitation.

## Discussion

In Japan, pulmonary tuberculosis accounts for 6.8-10% of cases of hemoptysis (1, 4), and hemoptysis is not uncommon in patients with active pulmonary tuberculosis. Pseu-

doaneurysms were reportedly found in 7.1% of cases of pulmonary tuberculosis that caused hemoptysis, but the main cause of hemoptysis was inflammatory hypervascularity of the bronchial artery (5). In a study of 7 patients who died suddenly of massive hemoptysis due to pulmonary tuberculosis, all patients had a large cavity with a major axis of  $\geq 5$  cm (2). Fatal massive hemoptysis is known to be involved in the rupture of Rasmussen aneurysms, a pulmonary aneurysm formed in the cavity wall. A pathological examination of patients with chronic tuberculosis cavities confirmed the presence of Rasmussen aneurysms in 4% of cases (6).

Inflammation owing to tuberculosis leads to weakening of the arterial wall and is involved in the formation of pseudoaneurysms. Even in cases of non-cavity pulmonary tuberculosis, aneurysms can develop, and massive hemoptysis can occur, because of the spread of inflammation from pulmonary tuberculosis lesions to adjacent arteries, such as the pulmonary artery (7, 8), bronchial artery (9), and aorta (10) or the invasion of the arterial wall by tuberculosis through blood flow (11-13). In our case, dilated and meandering right bronchial arteries were confirmed on angiography, and embolization was performed. Subsequently, epithelial layer findings were observed at the site of the bronchial aneurysm on bronchoscopy. Bronchial aneurysm may have been caused by the inflammatory spread of bronchial tuberculosis and may have ruptured because of massive hemoptysis.

We examined the repair process of bronchial tuberculosis on electron fluorescence bronchoscopy. Electron fluorescent bronchoscopy is characterized by green autofluorescence emitted by the blue excitation light of tissues and red and blue reflected light of two kinds of irradiation (14) and detects abnormal epithelial layer thickening, such as that seen in early lung cancer. In our case, the epithelial layer around the bronchial ulcer was depicted in magenta. Similar to lung cancer findings, this resulted from the attenuation of green autofluorescence by the thickened epithelial layer and mixing of the red and blue reflected lights to show magenta. In addition, the area of the bronchial ulcer with a white coat showed strong green fluorescence. Green autofluorescence is enhanced in the extracellular matrix, such as with collagen and elastin (14), and is seen in porcine airway epithelia as evidence of ischemic changes in the airway (15). In this case, it was considered that the green autofluorescence enhanced by the extracellular matrix was not attenuated by the airway epithelial thinning due to the ulcer, and thus it was observed as strong green fluorescence. The change in autofluorescence is nonspecific, but confirming the extent of ischemic changes and inflammation of the airway epithelium by fluorescent imaging may make it possible to predict bronchial stenosis or obstruction which may occur during the healing process of bronchial tuberculosis. There have been no reports of such findings in the human airway epithelia, and this is the first report of ulcerative lesions caused

by bronchial tuberculosis observed by autofluorescence imaging.

In conclusion, the present report provides important evidence that massive hemoptysis can be observed in non-cavity pulmonary tuberculosis through the formation of an aneurysm. In addition, ischemic lesions, such as bronchial ulcers, can be visualized as a strong green presence on electron fluorescence bronchoscopy.

**The authors state that they have no Conflict of Interest (COI).**

## References

1. Ishikawa H, Hara M, Ryuge M, et al. Efficacy and safety of super selective bronchial artery coil embolization for hemoptysis: a single-center retrospective observational study. *BMJ Open* **7**: e014805, 2017.
2. Tsukamura M. Background factors of death due to massive hemoptysis in lung tuberculosis. *Kekkaku (Tuberculosis)* **58**: 465-467, 1983 (in Japanese, Abstract in English).
3. Zheng X, Xiong H, Li Y, Han B, Sun J. RGB and HSV quantitative analysis of autofluorescence bronchoscopy used for characterization and identification of bronchopulmonary cancer. *Cancer Med* **5**: 3023-3030, 2016.
4. Minagawa Y. The clinical study of 60 hemoptysis patients over the past ten years at critical care center, Iwate Prefectural Kuji Hospital. *J Jpn Soc Emer Med* **16**: 607-612, 2013 (in Japanese).
5. Ramakantan R, Bandekar VG, Gandhi MS, Aulakh BG, Deshmukh HL. Massive hemoptysis due to pulmonary tuberculosis: control with bronchial artery embolization. *Radiology* **200**: 691-694, 1996.
6. Auerbach O. Pathology and pathogenesis of pulmonary arterial aneurysm in tuberculous cavities. *Am Rev Tuberc* **39**: 99-115, 1939.
7. Jemshad A, Ahammed S, Abdulla MC, Musambil M. Pulmonary artery aneurysm and thrombosis in active tuberculous consolidation. *Indian J Tuberc* **62**: 180-182, 2015.
8. Tebere H, Moifo B, Fongang E, et al. [Giant aneurysm of the pulmonary artery: a case report]. *J Mal Vasc* **38**: 201-205, 2013 (in French, Abstract in English).
9. Cheng YS, Lu ZW. Bronchial aneurysm secondary to tuberculosis presenting with fatal hemoptysis: a case report and review of the literature. *J Thorac Dis* **6**: E70-E72, 2014.
10. Sugane T, Takahashi N, Koura T, et al. A case of tuberculous aneurysm of the aorta. *Kekkaku (Tuberculosis)* **75**: 589-593, 2000 (in Japanese, Abstract in English).
11. Sakakibara K, Okano T, Kurane S, Kudoh S. A case of tuberculous aneurysm of subclavian artery occurred in the course of treatment for miliary tuberculosis. *Kekkaku (Tuberculosis)* **82**: 111-114, 2007 (in Japanese, Abstract in English).
12. Uchida Y, Tsukino M, Watanabe I. A case of miliary tuberculosis complicated by a tuberculous aneurysm of the thoracic aorta. *Kekkaku (Tuberculosis)* **88**: 629-632, 2013 (in Japanese, Abstract in English).
13. Matsutake T, Hashizume K, Kinoshita N, et al. A tuberculous pseudo-aneurysm of the abdominal aorta complicated by military tuberculosis. *Kekkaku (Tuberculosis)* **90**: 463-468, 2015 (in Japanese, Abstract in English).
14. He Q, Wang Q, Wu Q, Feng J, Cao J, Chen BY. Value of autofluorescence imaging videobronchoscopy in detecting lung cancers and precancerous lesions: a review. *Respir Care* **58**: 2150-2159, 2013.
15. Iga N, Oto T, Okada M, et al. Detection of airway ischaemic damage after lung transplantation by using autofluorescence imaging bronchoscopy. *Eur J Cardiothorac Surg* **45**: 509-513, 2014.

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