

# Massive Hemoptysis due to Right Inferior Phrenic Artery-to-Right Pulmonary Artery Fistula in the Right Middle Lobe of the Lung

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## Abstract

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Massive hemoptysis is a medical emergency and needs immediate treatment. It occurs in a wide variety of pulmonary diseases and typically originates from the bronchial arteries. We herein report a very rare case of a patient bleeding from a right phrenic artery-to-pulmonary artery fistula accompanied with focal bronchiectasis in the right middle lobe of the lung. In this case, multi-detector computed tomography was useful for clarifying the etiology and the abnormal anastomosis and facilitated effective angiographic embolization.

**Key words:** hemoptysis, bronchiectasis, embolization, fistula, non-bronchial artery

(Intern Med 56: 687-689, 2017)

(DOI: 10.2169/internalmedicine.56.6783)

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## Introduction

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Massive hemoptysis is a life-threatening pulmonary emergency and has a variety of underlying conditions. Transcatheter bronchial artery embolization (BAE) is a well-established and effective non-surgical procedure for the management of massive hemoptysis (1). Recently, non-bronchial systemic arteries have been reported as an important source of bleeding with massive hemoptysis. Computed tomography (CT) and computed tomography angiography (CTA) are useful for assessing the cause and origin of hemoptysis (2). We herein report a rare case of massive hemoptysis in a patient with focal bronchiectasis and right inferior phrenic artery-to-right pulmonary artery fistula diagnosed by CT and CTA.

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## Case Report

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An 82-year-old woman was transferred to our hospital because of dyspnea and massive hemoptysis. The patient had a history of bronchial asthma that was well-controlled with bronchodilator medications. She had no history of tuberculosis, nontuberculous mycobacterial infection, or smoking.

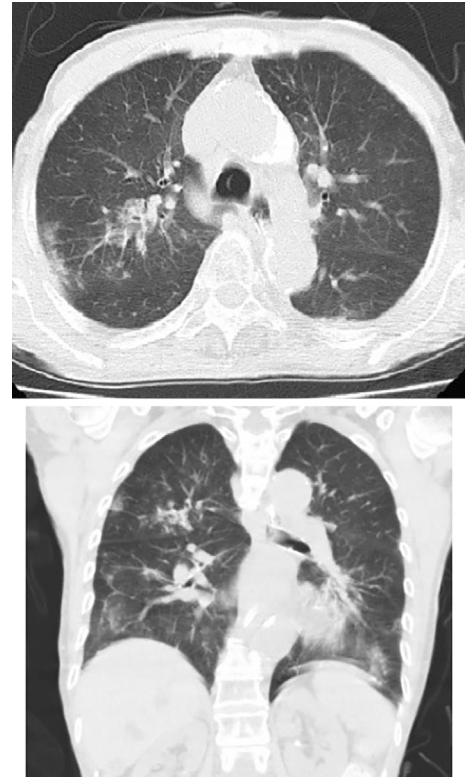
The physical examination revealed diffuse bilateral crack-

les. She suffered from severe hypoxemia (pH 7.362, PCO<sub>2</sub> 35.1 mmHg, PO<sub>2</sub> 61.0 mmHg, HCO<sub>3</sub><sup>-</sup> 20.1 mmHg, BE -5.5 mmHg, SpO<sub>2</sub> 90.5%, under 10 L O<sub>2</sub>/min, reservoir mask). After the tracheal intubation, 100 mL of bright-red blood was aspirated. A chest radiograph showed bilateral infiltrates (Fig. 1). A chest CT further demonstrated multiple consolidations and ground glass opacity and focal bronchiectasis in right segment 4 (S4) (Fig. 2). There were no space-occupying lesions. Four days after admission, her respiratory condition was improved. Since there was no active hemorrhaging from the tracheal tube, she was then extubated. After that, only a small amount of bloody sputum was coughed up.

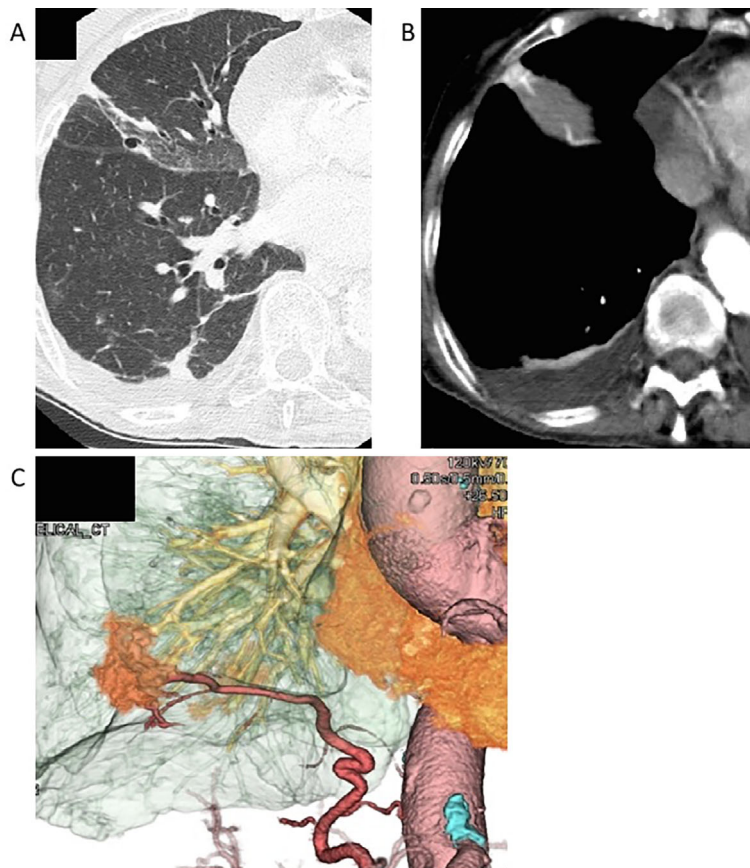
To determine the origin of bleeding, she underwent contrast-enhanced CT, which showed bronchiectasis in right S4 and regression of the infiltration. CTA revealed an abnormal vascular anastomosis between the right inferior phrenic artery and right pulmonary artery beside the focal bronchiectasis at the right middle lobe (Fig. 3), which led us to suspect it as the possible source of the massive hemoptysis. We therefore performed embolization by superselecting the right inferior phrenic artery with a 2.2-Fr. microcatheter (Fig. 4). An angiogram of the right bronchial artery showed no obvious active bleeding. Three weeks after the embolization, she was successfully discharged and has been free



**Figure 1.** A chest radiograph on admission showed bilateral infiltrates.



**Figure 2.** Computed tomography showed multiple consolidation and ground glass opacities.



**Figure 3.** A: A high-resolution computed tomography (CT) image showing focal bronchiectasis in the right middle lung (S4). B: CT angiography showing a vessel entering the bronchiectasis lesion arising from the right inferior phrenic artery. C: Three-dimensional CT depicting the right inferior phrenic artery-to-right pulmonary artery fistula.



**Figure 4.** Right inferior phrenic artery angiogram confirmed the right inferior phrenic artery-to-right pulmonary artery fistula (arrow) in an intrapulmonary portion.

from recurrent hemoptysis for three years.

## Discussion

The present case report describes an elderly woman who had right inferior phrenic artery-to-pulmonary artery fistula with focal bronchiectasis leading to life-threatening hemoptysis. BAE is widely performed for the treatment of hemoptysis, especially in severe cases. However, recurrent hemoptysis after successful BAE is not rare, and non-bronchial arteries can function as sources of bleeding in some cases (3). In the present case, plain CT demonstrated focal bronchiectasis in the right middle lobe (S4). We planned BAE to prevent recurrent hemoptysis and performed high-resolution CT and CTA before BAE. CTA revealed the right inferior phrenic artery-to-pulmonary artery fistula, not the bronchial arteries, as the source of the bleeding.

The bronchial arteries are responsible for bleeding in more than 90% of cases with massive hemoptysis. In the remaining 10% of cases, the internal mammary, intercostal, and inferior phrenic artery are typically involved in hemoptysis (3, 4). In most of the reported cases of inferior phrenic artery origin, the lower lobe of the lung was affected with chronic inflammation and found to be the site of bleeding (5-8). Transpleural systemic-pulmonary artery anastomoses may develop in patients with bronchiectasis, cystic fibrosis, tuberculosis, or chronic pneumonia (5). In the present

case, a right inferior phrenic artery-to-right pulmonary artery fistula was the origin of bleeding in the right middle lobe, instead of the lower lobe. She had focal bronchiectasis, but there was no sign of infection.

Hemoptysis is a respiratory emergency that can lead to a life-threatening condition. The present paper demonstrates that, for effective BAE in patients with massive hemoptysis, CT and CTA should be performed beforehand to precisely identify the culprit lesion for bleeding.

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

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