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Coronary-Bronchial Artery Fistula Manifested by Hemoptysis and Myocardial Ischemia in a Patient with Bronchiectasis

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A coronary-bronchial artery fistula is a very rare congenital anomaly of the coronary artery whose etiology and pathogenesis have not yet been clarified. Most patients with coronary-bronchial fistulas are asymptomatic; however, some patients present with congestive heart failure, infective endocarditis, myocardial ischemia induced by a coronary steal phenomenon, or rupture of an aneurysmal fistula. Furthermore, patients with a coronary-bronchial artery fistula rarely manifest life-threatening hemoptysis due to the associated bronchiectasis. We report herein the case of a patient with a coronary-bronchial artery fistula who had bronchiectasis and a history of massive hemoptysis and myocardial ischemia.

Key words: 1. Fistula
2. Hemoptysis
3. Myocardial ischemia

CASE REPORT

A 57-year-old man presented at our hospital with a massive hemoptysis. The patient had received medical treatment due to a mild fever, cough, and yellowish sputum that developed 10 days prior, but the symptoms had not improved. Furthermore, his oral intake had become worse 5 days before and hemoptysis started 2 days before admission to our hospital. At first, about a 10 mL of bloody sputum developed, but the amount progressively increased and eventually approximately 100 mL of bloody sputum was produced per hour. The dyspnea and chest pain became aggravated after the hemoptysis occurred. The patient was a white-collar worker, who had smoked 1 pack of cigarettes per day for 40 years and had consumed 1 alcoholic drink per day, 4 times a week, for 40 years. He had been diagnosed with diabetes mellitus at a regional hospital 3 years earlier and was still on medication. His pertinent surgical history was a bilateral knee replacement 2 years earlier. He was also diagnosed with bronchiectasis at a regional hospital 3 years earlier, and received medical treatment for the bronchiectasis 1 year earlier. At the time of that treatment, he did not undergo a bronchial artery embolization, but continued to receive drug therapy. The three months prior to admission at our hospital, episodic chest tightness developed, which became aggravated especially after intensive exercise. He reported that a reevaluation of his chest tightness and dyspnea at a regional hospital had not revealed any specific findings other than bronchiectasis.

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When admitted to our hospital, his blood pressure was 117/59 mmHg, heart rate was 87 beats/min, respiratory rate was 19 breaths/min, and body temperature was 37.6°C. The laboratory test results were within normal limits except the WBC (12,400/μL) and CRP (10.08). The electrocardiogram on admission showed a normal sinus rhythm and a chest radiograph showed a bronchiectatic change in the left lower lung (Fig. 1). Contrast-enhanced multidetector chest computed tomography (MDCT) revealed a severe cylindrical bronchiectatic change in the left lower lung field, and tortuous and anomalous communicating vascular crowding around the left hilum that was communicating with the right coronary artery (RCA) (Fig. 2). Since the transthoracic echocardiography revealed decreased wall motion in the RCA territory, a coronary angiography was performed to rule out coronary vascular disease. Coronary angiography via the radial artery revealed a coronary-bronchial artery fistula originating from the RCA and extending to the bronchial artery. And there was no change in the coronary artery system (Fig. 3). In this situation, we considered percutaneous transcatheter embolization and bronchial artery embolization. However, we performed surgical treatment in an attempt to treat the underlying bronchiectasis. Under general anesthesia, a posterolateral thoracic incision was made. A left lower lung lobectomy was performed and the coronary-bronchial artery fistula was removed. His clinical symptoms including chest tightness and dyspnea improved, and he was discharged from the hospital. At a 24-month follow-up, his postoperative course was found to be uneventful without any recurrence.

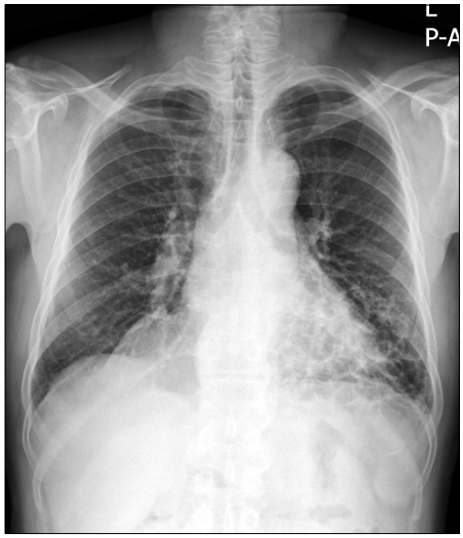

Fig. 1. Preoperative chest PA image shows a severe bronchiectatic change in the left lower lung field.

DISCUSSION

A coronary-bronchial artery fistula is a rare congenital anomaly that occurs at the coronary artery [1]. It has been reported that this anomaly is detected in 0.18% of patients who undergo coronary angiography [2]. With advances in diagnostic technologies such as MDCT, many cases of coro-

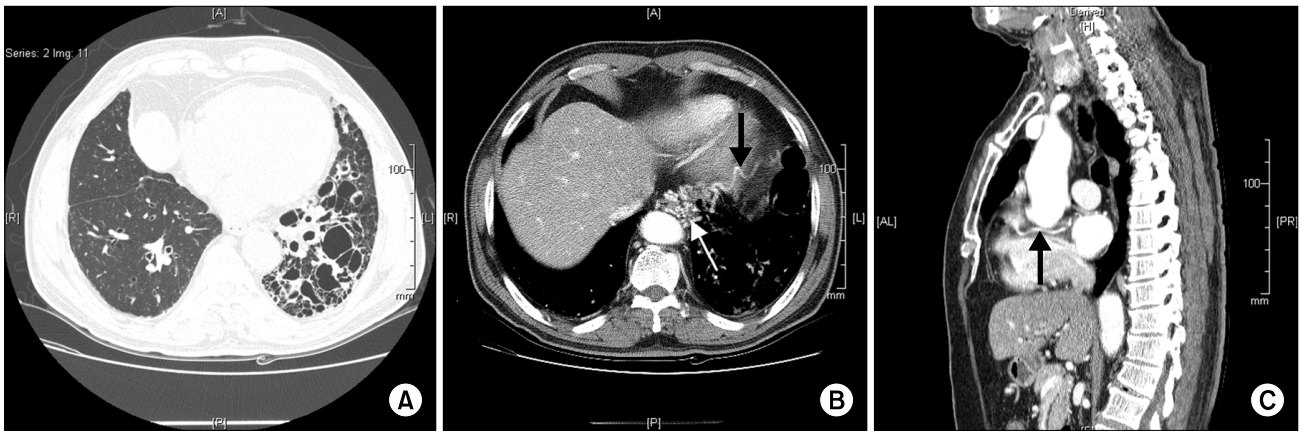

Fig. 2. (A) A chest computed tomography (CT) shows a severe bronchiectatic change in the left lower lung field. (B) An enhanced transverse chest CT shows multiple tortuous and vascular crowding of the bronchial artery (white arrow) and a coronary-bronchial artery fistula (black arrow). (C) An enhanced sagittal chest CT shows a coronary-bronchial artery fistula (black arrow).

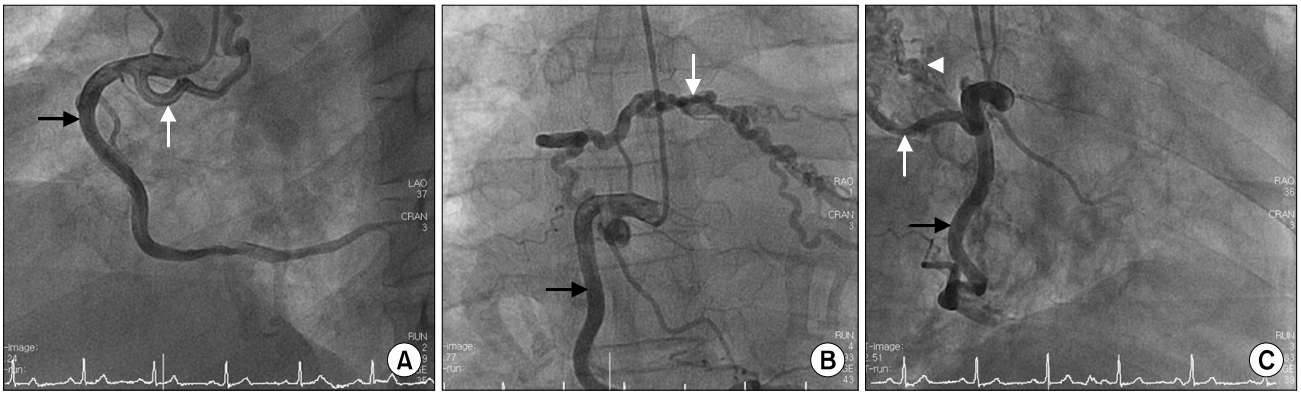

Fig. 3. A coronary angiograph shows a coronary-bronchial artery fistula (white arrow) originating from the proximal portion of the right coronary artery (black arrow). The coronary angiographic views in left anterior oblique view (LAO) 37° and cranial view (CRAN) 3° (A), right anterior oblique view (RAO) 1° and CRAN 3° (B), and RAO 36° and CRAN 3° (C) show a coronary-bronchial artery fistula (white arrow) originating from the proximal portion of the right coronary artery (black arrow). The white arrow head indicates multiple tortuous and vascular crowding of the bronchial artery.

nary-bronchial artery fistula have recently been reported. However, its pathogenesis has not yet been elucidated. Most cases of coronary-bronchial artery fistula have no clinical implications because they are asymptomatic, whereas some cases are related to the tetralogy of Fallot, a supraventricular aortic stenosis, aortitis syndrome. A previous study showed the relationship between bronchiectasis and coronary-bronchial artery fistula [3]. Further studies are needed to confirm this relationship. Several previous reports have proposed that patients with a coronary-bronchial artery fistula need aggressive treatment, including embolization, in order to prevent lethal complications such as infective endocarditis or an aneurysmal rupture. Aggressive treatment should be performed in cases of symptomatic coronary-bronchial artery fistula [4]. A comprehensive physical examination and thorough review of a patient's history are extremely important for determining the diagnosis of this fistula. Contrast-enhanced MDCT with retrospective electrocardiogram gating is an accurate and non-invasive screening test, which allows for more acceptable diagnostic outcomes with advances in CT resolution [5]. Echocardiography is essential to the evaluation of concurrent cardiac anomalies and left-to-right shunts. In our patient, a decrease in myocardial contraction was observed in the RCA territory. Shin et al. [6] have demonstrated that functional tests including the myocardial perfusion scanning are helpful in the diagnosis of coronary-bronchial artery fistula. A great variety of clinical features have been reported in patients who has coronary-bronchial artery fistula. Hackett and Hallidie-Smith [7] reported a case of spontaneous closure of a coronary artery fistula secondary to thrombosis. The severity of the clinical features of a coronary-bronchial artery fistula depends on the degree of the left-to-right shunt. As the degree of the left-to-right shunt increases, complications such as pulmonary hypertension or congestive heart failure occur more frequently. A few cases of rupture or thrombosis of coronary artery fistulas have been reported in association with arterial aneurysms or coronary steal phenomena [8]. Our patient presented with only chest pain and hemoptysis, and the diagnosis of a coronary-bronchial fistula was established by MDCT, which was performed to assess massive hemoptysis. It seems likely that recurrent life-threatening hemoptysis may be attributed to coronary-bronchial artery fistula associated with bronchiectasis, and that patients with such fistulas can be treated surgically. However, since a standard treatment method for coronary fistulas has not yet been determined, most patients have been managed in light of anecdotal case reports or small series of cases. Further studies with a larger number of cases are needed to understand the clinical features and to establish a standard treatment modality. Closure of coronary-bronchial artery fistula is mainly indicated for patients with heart failure, myocardial ischemia, or high-flow shunting. Shin et al. [6] have proposed prophylactic and ther-

apeutic embolization of coronary-bronchial artery fistula in patients with bronchiectasis. Embolization, which is less invasive, has recently been attempted. However, patients with such fistulas should be treated based on the presence of concurrent cardiac anomalies as well as the clinical evaluation of other organs. Embolization can be safely performed after the anatomical relationships between the fistula and the surrounding structures are completely assessed. Kang et al. [4] have indicated that contrast-enhanced MDCT coronary angiography is helpful in the identification of the course of a coronary-bronchial artery fistula. We reported the case of a patient with a coronary-bronchial artery fistula who presented with angina symptoms and bronchiectasis and was successfully treated. Further studies are needed to understand the precise pathogenesis of the fistula and the relationship between the fistula and bronchiectasis.

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