I. Introduction

Coronary-bronchial artery fistula (CBAF) is a very rare anomaly of the coronary artery. There have been few reports regarding the pathology of CBAF and the mechanism of hemoptysis. Although the optimal treatment for CBAF has not been established, associated fistula should be closed in symptomatic patients. Here, we describe a case involving a patient with CBAF that caused hemoptysis that was successfully treated via surgical repair.

II. Case report

A 75-year-old woman who experienced hemoptysis every morning for one year was admitted to our hospital. She had been treated for hypertension at a local hospital and had no obvious history of Kawasaki disease or any other infectious diseases. First, she visited the Department of Respiratory Medicine. However, she was referred to a cardiologist because contrast-enhanced computed tomography revealed both a coronary artery aneurysm and coronary-pulmonary artery fistulae. Bronchography was not performed, since computed tomography revealed no respiratory lesions in either of her lungs. Initial laboratory data were as follows: hemoglobin level, 11.4 g/dl; hematocrit level, 32%; C-reactive protein level, 0.02 mg/dl; white blood cell count, $4.6 \times 10^3/\mu l$; and brain natriuretic peptide level, 51 pg/dl. A chest radiograph revealed that the patient had no remarkable lung lesions. Electrocardiography indicated a normal sinus rhythm and no remarkable ischemic changes. Echocardiography revealed normal cardiac function and little pericardial effusion. Coronary angiography revealed the presence of multiple coronary arteriovenous fistulae that had arisen from the right coronary (Fig. 1a) and left anterior descending (Fig. 1b) arteries, which drained into the pulmonary artery. The coronary arteriovenous fistula of the left anterior descending artery also revealed a saccular aneurysmal change that was 10 mm in diameter (Fig. 1b). Contrast-enhanced multidetector computed tomography (MDCT) was performed for further examination and revealed CBAF with mild dilatation of bronchial arteries (Fig. 2a, b). CBAF consisted of a tortuous vessel that originated from the left anterior descending artery, and passed laterally between the ascending aorta and pulmonary artery (Fig. 2b, c). Then, it turned to the lesser curvature of the aortic arch to Anastomose with the bronchial arteries (Fig. 2b). Because MDCT showed dilatation of bronchial arteries, CBAF was determined to be the cause of hemoptysis. Preoperative cardiac catheterization revealed a pulmonary-to-systemic flow ratio of 1.1, and systolic and mean pulmonary artery pressures were 20 and 11 mmHg, respectively.

Open surgical repair was used to treat CBAF and multiple coronary-pulmonary artery fistulae with saccular aneurysmal changes. After median sternotomy, cardiopulmonary bypass was established. A saccular aneurysm and multiple coronary arteriovenous fistulae draining into the pulmonary artery were detected. Fistulae were closed using continuous sutures, and aneurysmal...
Fig. 1
a: Right coronary angiogram shows multiple coronary artery aneurysms with pulmonary artery fistulae.
b: Left coronary angiogram reveals a saccular aneurysm (white arrow) and multiple coronary artery aneurysms with pulmonary artery fistulae arising from the left anterior descending artery.

c: Preoperative multidetector computed tomography shows a connection (white dotted arrow) between the bronchial artery and the left anterior descending artery (white arrow).

Fig. 2
a: Preoperative multidetector computed tomography shows a mildly dilated bronchial artery (white arrow head).
b: Preoperative multidetector computed tomography shows a coronary-bronchial artery fistula (black dotted arrow) arising from the left anterior descending artery (black arrow) draining into the bronchial artery (white arrow head).
c: Preoperative multidetector computed tomography shows a connection (white dotted arrow) between the bronchial artery and the left anterior descending artery (white arrow).

d: Postoperative multidetector computed tomography reveals no abnormal flow of coronary-bronchial artery fistula (black dotted arrow).
walls were ligated using mattress sutures without cardioplegic arrest (Fig. 3a). CBAF detected between the pulmonary artery and ascending aorta was closed using hemostatic clips (Fig. 3b). After the procedure, we confirmed the closure of fistulae via the elimination of Doppler signal via Doppler ultrasonography (Direction Volume Meter®, Hayashi Denki, Kanagawa, Japan).

The patient was successfully weaned from cardiopulmonary bypass, and there was no need for blood transfusion. Her postoperative course was uneventful, and hemoptysis disappeared immediately. She was discharged on postoperative day 20. Postoperative MDCT revealed no abnormal blood flow through the fistulae and aneurysm, and no significant stenosis of the coronary arteries. In addition, MDCT did not detect a connection between the left anterior descending and bronchial arteries (Fig. 2d). To avoid ischemic events from thromboembolism, antiplatelet therapy with aspirin was administered for a one-month period following surgery and anticoagulant therapy consisting of warfarin was administered for a three-month period following surgery until low-level control of the prothrombin produced an international normalized ratio of 1.5–2.0. Six months later, the patient was free of hemoptysis and cardiac events.

III. Discussion

CBAF was first reported by Moberg3 as the presence of small extracardiac anastomoses to the heart through the bronchial artery. Several mechanisms are involved in the pathogenesis of CBAF: various cardiovascular diseases, Takayasu arteritis, pulmonary thromboembolism, bronchiectasis, and pulmonary tuberculosis4–6. Localized bronchiectasis is the most common condition associated with CBAF; however, CBAF of the present case was combined with a coronary-pulmonary artery fistula. CBAF has been suggested to be present in all patients from birth, but remains closed as a result of similarities between filling pressures of coronary and bronchial circulations4. In the present case, CBAF may have opened because of filling pressure changes produced by coronary-pulmonary artery fistula. Coronary-pulmonary artery fistula might be involved in increase in filling pressure of coronary artery.

In the literature, CBAF has typically been shown to originate from the left circumflex artery via the left atrial branch and cross the pericardium by means of pericardial reflections within retrocardiac spaces2–4. However, in the present case, CBAF originated from the left anterior descending artery which connected to the pulmonary artery and was anastomosed to bronchial arteries. Most patients with CBAF are asymptomatic, and some present with myocardial ischemia induced by a coronary steal phenomenon or rupture of an aneurysmal wall8. Hemoptysis is extremely rare4–6,7 but sometimes extremely life-threatening. A previous report has shown that pressure differences between coronary and bronchial arterial circulation patterns may result in artery enlargement8. Bronchial steal is rarely diagnosed because it is generally not detected via coronary angiography. Bronchial steal has been mainly reported in patients with proximal stenosis of the coronary arteries in whom distal coronary artery pressure is decreased relative to bronchial artery pressure8. Therefore, bronchial steal is clinically less problematic than coronary steal.
The diagnosis of coronary artery anomalies is usually established during invasive coronary angiography. However, coronary angiography often fails to clearly indicate the relationship between the anomalous coronary artery and surrounding structures. Contrast-enhanced MDCT reveals trivial coronary artery anomalies with high diagnostic accuracy. In the present case, contrast-enhanced MDCT was a reliable technique for identifying CBAF and confirming the abolition of abnormal blood flow.

If hemoptysis remained after open surgical repair, we planned to perform coil embolization to treat CBAF. Currently, coil embolization is regarded as the best treatment of coronary steal phenomenon. Embolization can be safely performed after anatomical relationships between the fistula and surrounding structures are completely assessed. However, after CBAF embolization, a few studies have reported the occurrence of myocardial infarction. Moreover, the anatomy and characteristics of some fistulae do not allow for catheterization and various complications should be considered including residual flow, arrhythmia, distal embolism, and coil detachment. In case of multiple aneurysms or fistulae, open surgical repair is preferable to other treatment methods. In our literature review, we found various reports regarding the use of treatment methods; however, simple ligation is the only strategy used in surgical treatment for CBAF. Closure using hemostatic clips was effective in the present case, but other strategies used should be improved.

There are no official guidelines regarding surgical indications of coronary artery aneurysm and fistula. To summarize various reports, surgical indications for coronary artery aneurysm in the present case include the symptomatic nature of the aneurysm and the presence of saccular aneurysmal change and surgical indications for coronary artery fistula include the presence of aneurysmal change. In the present case, we chose open surgical repair to treat CBAF as a result of the surgical indications of coronary artery aneurysm and coronary-pulmonary artery fistula. If the patient had no surgical indications, the appropriate strategy for CBAF treatment would have been controversial. A treatment strategy for CBAF should be established, which considers the severity of hemoptysis and other concomitant clinical features.

IV. Conclusion

We successfully surgically treated CBAF that caused hemoptysis. MDCT can provide anatomical orientation of the vascular anomaly that is superior to that of conventional angiography.

Acknowledgment

We thank the Honyaku Center Inc. for reviewing and editing the manuscript. We also thank our colleagues for their helpful comments.

Disclosure Statement

Kayo Sugiyama, Yasuhiro Futamura, Masaho Okada, Hirotaka Watanuki, Makio HagiHaraba, Masahiro Shimoda, and Katsuhiko Matsuyama declare no conflicts of interest, and the authors did not receive any financial support for this study.

References