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Haemoptysis due to an atypical right bronchial artery branching from the left subclavian artery evaluated by four-dimensional CT and bronchial arteriography

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SUMMARY

Haemoptysis can be a life-threatening condition, and bronchial artery embolisation (BAE) is the most effective treatment. A 19-year-old man who had been followed up for neutrophil migration failure and bronchiectasis since birth was admitted to our department with repeated haemoptysis. Contrast-enhanced CT revealed hyperplasia and tortuosity of the bronchial artery; however, it could not reveal the origin and running by the axial views. Three-dimensional (3D) and four-dimensional (4D) CT revealed branching of the right bronchial artery from the left subclavian artery and a bronchial artery–pulmonary artery shunt. Bronchial angiography confirmed these findings. Based on these findings, BAE was successfully performed. 3D and 4D CT can be useful for determining the cause of haemoptysis. Furthermore, evaluating the anatomy of bronchial arteries prior to BAE may be useful for performing safer procedures.

BACKGROUND

Haemoptysis can be a life-threatening condition, and the bronchial artery is the most common source of haemoptysis.[1] Bronchial artery embolisation (BAE) is the most effective nonsurgical treatment for the management of massive and recurrent haemoptysis.[2–4] Moreover, evaluating the anatomy of the bronchial arteries is essential for performing safer BAE. However, the bronchial arteries are highly variable in terms of origin and branching distribution.[5] Furthermore, in cases associated with chronic airway inflammation, hyperplasia and tortuosity of blood vessels can occur, which makes the anatomy of the bronchial arteries more complicated.[4 6]

Herein, we present a case of haemoptysis due to an atypical right bronchial artery branching from the left subclavian artery evaluated by four-dimensional (4D) CT and bronchial arteriography.

CASE PRESENTATION

A 19-year-old man was admitted to our department with repeated haemoptysis. He had been followed up for neutrophil migration failure and bronchiectasis at the paediatrics department in our hospital since birth. Five months before the admission, the patient was referred to our department with haemoptysis for 2 weeks. Although airway bleeding associated with bronchiectasis was suspected, his symptom was improved with carbazochrome sodium sulfonate hydrate and tranexamic acid. Three months before the admission, he visited again because of recurrence of haemoptysis. Plain CT showed lobular central granular shadows in the right middle lobe, left lingular segment and bilateral lower lobes. He was admitted for evaluation of the source of bleeding. On admission, his height was 168.5 cm, and his weight was 53.5 kg. His vital signs were blood pressure, 103/49 mm Hg; pulse rate, 81 beats/min; pulse oximetry, 99% in room air; and body temperature, 36.7°C. Physical examination showed coarse crackles in both lung fields. Laboratory examinations showed no abnormality. Chest X-ray showed bronchial wall thickening in the right middle and left lower lung field and the surrounding ground-glass opacity and consolidation (figure 1A). Plain chest CT also showed bronchial wall thickening and dilation in bilateral lung fields, partial mucoid impaction and multiple centrilobular granular shadows in the right middle lobe, lingular segment and bilateral lower lobe (figure 1B–D). Contrast-enhanced CT showed hyperplasia and tortuosity of the bilateral bronchial artery; however, it was difficult to obtain a detailed evaluation of the origin and running by the axial views (video 1). The three-dimensional (3D) image showed that the right bronchial artery branched from the left subclavian artery and was running on the dorsal side of the aortic arch (video 2 and figure 2A,B). On the other hand, the left bronchial artery branched from the aortic arch and perfused to the left lingular segment. In 4D CT, the pulmonary artery was imaged again after the bronchial artery was imaged, indicating a bronchial artery–pulmonary artery shunt on both sides (video 3 and figure 2C–E). Bronchial angiography confirmed the branching of the right bronchial artery from the left subclavian artery and shunting to the pulmonary artery (video 4 and figure 3A,B). Additionally, the left bronchial artery branched from the aortic arch and perfused to the left lingular segment consistent with the CT findings.

TREATMENT

BAE was initially performed for the right bronchial artery. It was approached via the left brachial artery with a 5 Fr sheath. A 5 Fr internal mammary artery (IMA) catheter (Cerulean; Medikit, Tokyo, Japan) was placed in the right bronchial artery branching from the left subclavian artery. Triaxial insertion through the catheter was performed by advancing a small, 1.9 Fr microcatheter (Carnelian Marvel; Tokai Medical, Kasugai, Japan) and a large, 2.7 Fr microcatheter (Carnelian HF; Tokai Medical, Kasugai, Japan). The microcatheter was advanced

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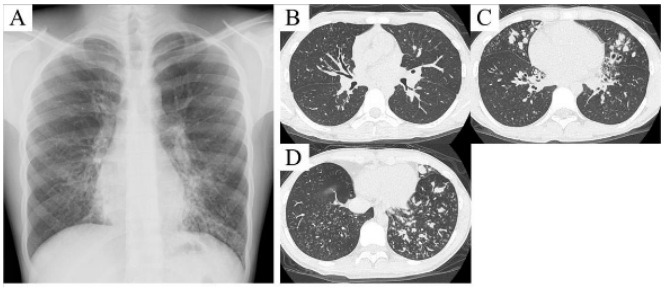


Figure 1 (A) Chest X-ray at the patient's admission to our department reveals bronchial wall thickening in the right middle and left lower lung field and the surrounding ground-glass opacity and consolidation. (B–D) Chest CT also reveals bronchial wall thickening and dilation in bilateral lung fields, partial mucoid impaction and multiple centrilobular granular shadows in the right middle lobe, lingular segment and bilateral lower lobe.

into the feeding artery as deep as possible, and embolisation was performed with spherical porous gelatine particles 1 or 2 mm in diameter (Gelpart; Nippon Kayaku, Tokyo). Postprocedural angiography revealed successful embolisation of the bronchial artery–pulmonary artery shunt (figure 3C,D).

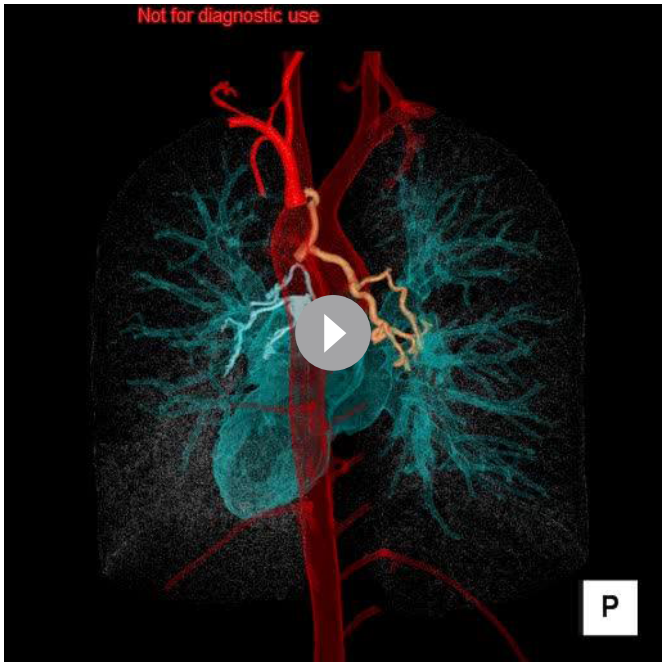
OUTCOME AND FOLLOW-UP

The BAE for the right bronchial artery was successfully completed without any complications. BAE for the left bronchial artery was also performed successfully without any complications. Twenty-four months after treatment, there was no recurrence of haemoptysis.

DISCUSSION

There were two notable clinical findings in this case. First, the bronchial arteries causing haemoptysis may communicate with various arteries, including the subclavian artery. Second, 3D and 4D CT can be useful for evaluation and understanding of the anatomy of bronchial arteries to perform safer BAE.

Video 1 Contrast-enhanced CT reveals hyperplasia and tortuosity of the bronchial artery.



Video 2 Three-dimensional CT images reveal that the right bronchial artery branches from the left subclavian artery and is running on the dorsal side of the aortic arch. The blood vessel coloured red is the left subclavian artery, orange is the atypical bronchial artery perfusing the right lung and blue is the normal bronchial artery, which branches from the aorta and perfuses the left lung.

The bronchial arteries causing haemoptysis may communicate with various arteries other than the aorta, including the subclavian artery. Approximately 70%–83.3% of bronchial arteries arise between the lower margin of T4 and the upper margin of T6 along the major bronchi.[7] In addition, the origins of bronchial arteries include the inferior aortic arch, distal descending thoracic aorta, subclavian artery, brachiocephalic trunk, thyrocervical trunk and IMA as well as the coronary artery.[8 9] An evaluation of the aortic arch should be performed when the significant bronchial artery, which can cause haemoptysis, is not demonstrated in the aortography of the descending aorta.[10] Our patient had a complicated bronchial artery running in which the right bronchial artery branches from the left subclavian artery. Cauldwell *et al* reported that only 3 (2%) of 150 cadavers had a subclavian bronchial artery origin, and among these three cases, two cases originated from the left subclavian artery and one from the right subclavian artery.[11] Hartmann *et al* evaluated multidetector-row helical CT angiography in 214 patients and reported that 13 (10.5 %) of 124 origins of ectopic bronchial arteries were subclavian arteries.[8]

The bronchial arteries become dilated and tortuous due to chronic inflammatory diseases such as bronchiectasis and tuberculosis.[6] Angiogenic growth factors released with inflammation promote angiogenesis and pulmonary vascular remodelling.[12] The resulting new collateral vessels have thin walls and are prone to rupture, which can cause haemoptysis.[13] In this case, chronic inflammation of the bronchi was accompanied by neutrophil migration failure and bronchiectasis, and chronic inflammation could have contributed to the complexity and proliferation of bronchial arteries. However, it was unclear whether the new blood vessels grew from congenital atypical bronchial arteries as a result of inflammation or the originally existing systemic arteries grew this way.

3D and 4D CT evaluation can be useful as non-invasive methods to evaluate the anatomy of bronchial arteries. Multidetector CT

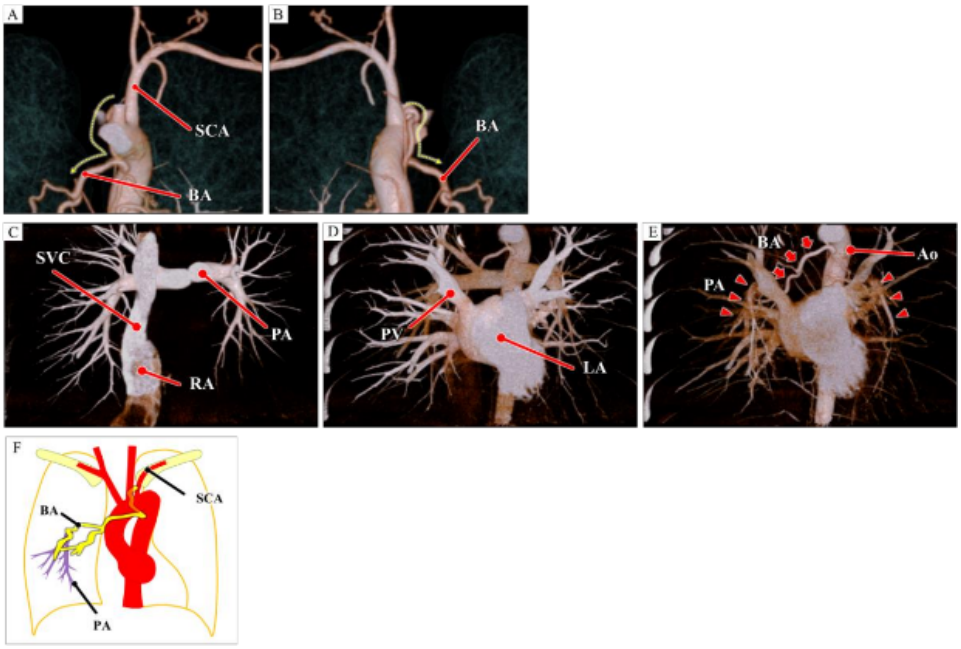
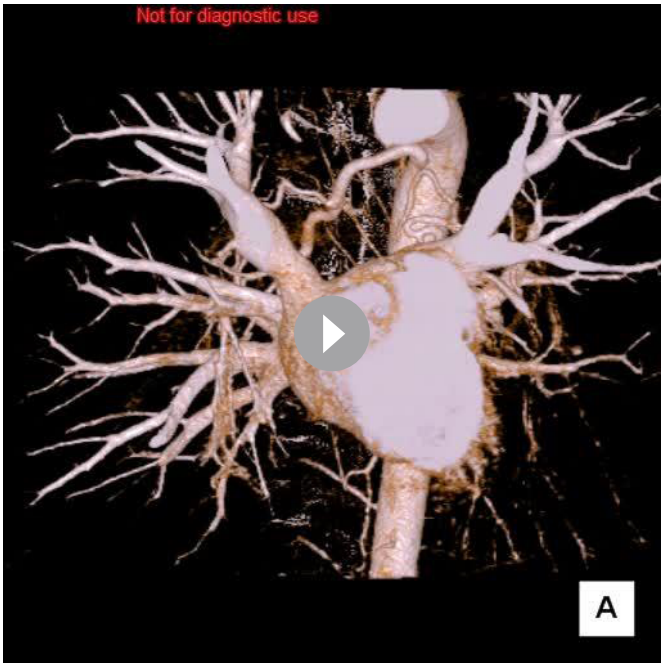


Figure 2 The anterior (A) and posterior (B) view of the three-dimensional image of multidetector CT angiography reveals that the right bronchial artery is branching from the left subclavian artery and is running on the dorsal side of the aortic arch (yellow dotted line). Four-dimensional CT of phases in which the pulmonary artery (C), pulmonary vein (D) and aorta (E) are contrasted shows a bronchial artery (arrow)–pulmonary artery (triangle) shunt (E). (F) The schema of the anatomy of the patient's right bronchial artery. The right bronchial artery branches from the left subclavian artery and forms the bronchial artery–pulmonary artery shunt. Ao, aorta; BA, bronchial artery; LA, left atrium; PA, pulmonary artery; PV, pulmonary vein; RA, right atrium; SCA, subclavian artery; SVC, superior vena cava. (F) was illustrated by HK.

(MDCT) angiography can provide a more accurate demonstration of bronchial and non-bronchial systemic arteries than conventional angiography[14] and detect 100% of bronchial and 62% of non-bronchial arteries causing haemoptysis.[15] Hwang reported that the evaluation for aberrant bronchial arteries by MDCT can be useful for planning therapeutic strategies such as BAE and surgery.[16] Furthermore, 3D images were superior to two-dimensional axial images in identifying ectopic origins of bronchial arteries, even with an enlarged and tortuous bronchial artery.[14 17] Furthermore, 4D CT can be used to evaluate the haemodynamic characteristics of the vasculature or the motion of organs, in addition to morphology.[18] In our case, 3D CT provided accurate anatomy of the bronchial artery,



Video 3 Four-dimensional CT revealed that the pulmonary artery was imaged again after the bronchial artery was imaged, indicating a bronchial artery–pulmonary artery shunt.

Video 4 Bronchial angiography confirmed the branching of the left subclavian artery to the right bronchial artery and shunting to the pulmonary artery.

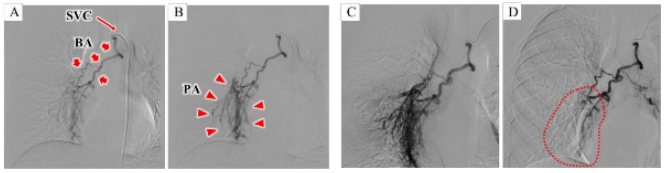


Figure 3 Bronchial angiography confirms the branching of the left subclavian artery to the right bronchial artery (arrow) and shunting to the pulmonary artery (triangle) (A, B). Bronchial angiography before (C) and after (D) bronchial artery embolisation. The pulmonary artery was no longer imaged after embolisation (the part surrounded by the red dotted line in panel (D)). BA, bronchial artery; PA, pulmonary artery; SVC, superior vena cava.

and 4D CT revealed a shunt of the pulmonary artery. Based on these findings, the appropriate type of catheter could be selected. Since the bronchial artery has many branches from the aorta and intercostal arteries, a shepherd's hook-type catheter whose tip hits the aortic wall at a right angle is usually used. However, in our case, since the bronchial artery was branched from the subclavian artery, an internal mammary-type catheter with an acute angle tip was used. Additionally, BAE is usually performed through the femoral artery.[1] However, in our case, the catheter was inserted from the left radial artery because there was a risk that the thrombus attached to the catheter would flow from the common carotid artery to the brain. As a result, the BAE was successfully performed without any complication. In fact, in a comparative study involving 200 patients, the use of MDCT before angiography reduced the rate of catheterisation failures and the number of patients needing surgical intervention.[19] Performing 3D and 4D CT prior to BAE makes it possible to evaluate the risk of complications, select the appropriate catheter and perform a safe and reliable embolisation.

Here, we present a case of haemoptysis due to an atypical right bronchial artery branching from the left subclavian artery. There are variations in the bronchial arteries in terms of origin and branching, including the branches from the subclavian artery, and it is important to evaluate the anatomy of the bronchial arteries prior to BAE. 4D CT is useful for evaluation of the anatomy of bronchial arteries.

Learning points

- Haemoptysis may be caused by the bronchial artery originating from the subclavian artery.
- Three-dimensional and four-dimensional CT can be useful modalities for evaluating blood vessel running of the bronchial arterial artery causing haemoptysis.
- Evaluating the blood vessel running of the bronchial artery before bronchial artery embolisation is useful for planning safer procedures.

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