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Primary racemose hemangioma with bronchial-pulmonary arterial fistula

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Key message

Primary racemose hemangioma with bronchial-pulmonary arterial fistula is a very rare abnormality. We herein report an asymptomatic case of primary racemose hemangioma with no significant size change in 5 years.

Clinical Image

Primary racemose hemangioma of the bronchial artery is a very rare abnormality and it has been reported as one of the causes of hemoptysis. It is characterized by enlarged and convoluted bronchial arteries, and shunts between the bronchial and pulmonary arteries. Racemose hemangioma has been reported since the end of 1970s and is classified into primary and secondary forms, respectively, due to congenital malformation and bronchial–pulmonary inflammation [1]. We herein report an asymptomatic case of primary racemose hemangioma with bronchial–pulmonary arterial fistula.

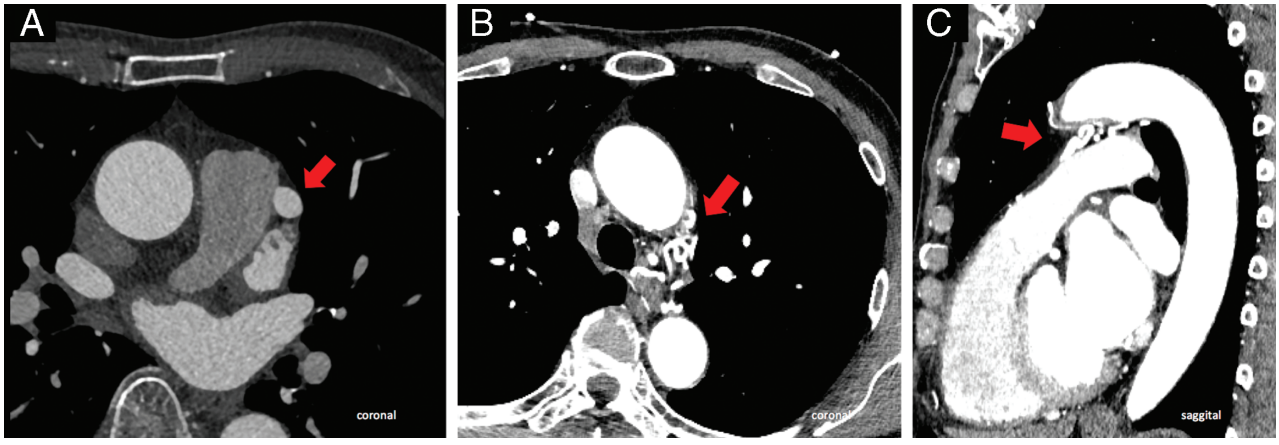

Figure 1. (A) Chest computed tomography (CT) shows a well-demarcated nodule adjacent to pulmonary trunk (arrow). (B, C) Contrast-enhanced CT reveals convolution and dilatation of bronchial arteries (arrow).

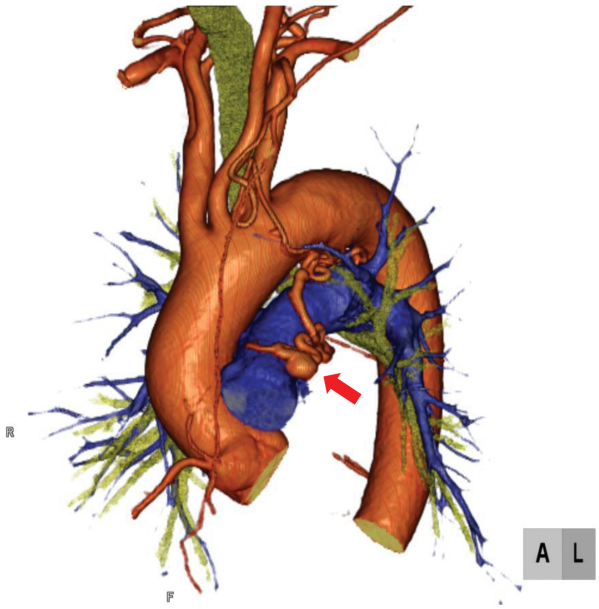

Figure 2. Three-dimensional computed tomography (CT) reveals the aneurysm connected to the pulmonary trunk and the dilated bronchial arteries (arrow).

An abnormal shadow in the left mediastinum was found by chance on computed tomography (CT) during follow-up after resection of bladder cancer in a 73-year-old man without any symptoms. Contrast chest CT revealed a round aneurysm, 1 cm in diameter, adjacent to the pulmonary trunk and dilated bronchial arteries around the aneurysm, suggesting the presence of fistula between bronchial and pulmonary arteries (Fig. 1). The aneurysm was derived from the bronchial arteries from the distal aortic arch and the left subclavian artery and connected to the pulmonary trunk (Fig. 2). It had been slowly growing in size as revealed by the CT image obtained 5 years before (Fig. 3). As he had no medical condition that may cause secondary bronchial artery aneurysms, such as chronic inflammatory lung diseases and mitral valve stenosis, we diagnosed it as primary racemose hemangioma. The diameter of his aneurysm was 1 cm with no significant size change in 5 years; we, therefore, decided to observe him conservatively with follow up CT once every 6 months.

Primary racemose hemangioma of the bronchial artery has been considered to be a congenital abnormality, but its genesis and natural course remain uncertain. Although the classical diagnostic approach involves angiography, this case was diagnosed by contrast chest CT with three-dimensional (3D) image reconstruction. According to the reports of racemose hemangioma, bronchial-pulmonary arterial fistula is formed via the pulmonary parenchyma, which is the cause of symptoms that lead to discovery such as hemoptysis [2]. On the other hand, in the present

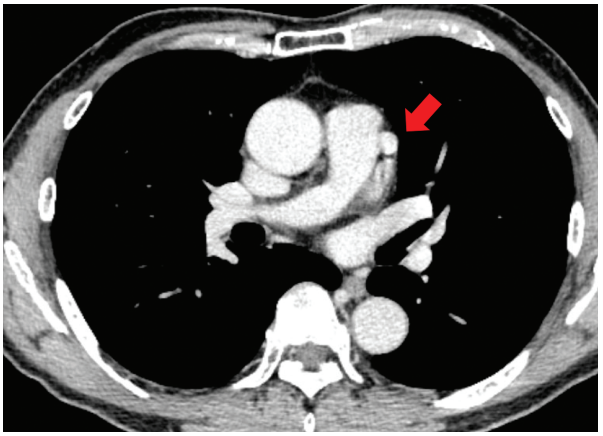

Figure 3. Chest computed tomography (CT) obtained 5 years before shows the aneurysm, 1 cm in diameter, almost with no size change (arrow).

asymptomatic case, the bronchial artery directly communicated with the pulmonary trunk without connection of the pulmonary parenchyma. The processes that lead to an aneurysm rupture are unknown and diameter is not the sole incremental risk factor [3]. Therapeutic candidates for racemose hemangioma include bronchial arterial embolization, bronchial arterial ligation, and surgical resection. However, the treatment strategy for primary racemose hemangioma of the bronchial artery has not been established. According to a review by Narato et al., in Japan, whereas lobectomy was performed most often approximately at 38%, bronchial arterial embolization was carried out at 29% for racemose hemangioma [1,4]. If therapeutic intervention is required to prevent rupture of this aneurysm, surgical intervention is necessary because effective embolization is difficult due to multiple blood supply. However, we plan to continue follow up with CT as the patient is asymptomatic with no significant size change of the aneurysm in 5 years.

Disclosure Statement

Appropriate written informed consent was obtained for the publication of this case report and accompanying images.

References

1. Kato M, Morio Y, Matsunaga T, et al. 2016. Bronchial-pulmonary arterial fistula with primary racemose hemangioma. Respirol. Case Rep. 4:e00148.
2. Pillai R, Mitchell A, and Jackson JW. 1982. Arterial fistula between the left internal mammary artery and left pulmonary artery. Thorax 37:386–387.

3. Kalangos A, Khatchatourian G, Panos A, et al. 1997. Ruptured mediastinal bronchial artery aneurysm: a dilemma of diagnosis and therapeutic approach. J. Thorac. Cadiovasc. Surg. 114:853–856.

4. Narato R, Enomoto T, Ono H, et al. 2006. A case of successful bronchial artery embolization for primary racemose hemangioma with massive hemoptysis. Nihon Kokyuki Gakkai Zasshi 44:641–646.