



Case report

Successful transcatheter arterial embolization in an asymptomatic patient with primary racemose hemangioma of the bronchial artery

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ARTICLE INFO

Keywords:

Primary racemose hemangioma
Bronchial artery
Bronchial arteriography
Bronchial arterial embolization
N-Butyl-2-cyanoacrylate

ABSTRACT

An asymptomatic 70-year-old woman presented with a nodular lesion overlapping the pulmonary artery at the right hilar region on a chest X-ray. Bronchial arteriography revealed an aneurysmal dilation of the long segment of the right bronchial artery and a shunt from the right bronchial artery to the right lower pulmonary artery. She was diagnosed with primary racemose hemangioma of the bronchial artery (RHBA). Considering the risk of hemoptysis, we performed a bronchial arterial embolization (BAE) using coils and N-butyl-2-cyanoacrylate. She had no complication after the BAE and no recurrences of hemoptysis at the 36-month follow-up. RHBA should be considered in case of aneurysmal dilation in the long segment of the bronchial artery, and BAE should be considered as a treatment strategy despite the absence of symptoms.

1. Introduction

Primary racemose hemangioma of the bronchial artery (RHBA) is a rare abnormality of the bronchial artery [1]. While RHBA usually presents with hemoptysis, it is difficult to diagnose in asymptomatic patients [2]. Bronchial arterial embolization (BAE) is the first choice for treatment for RHBA; however, recurrence is a common complication after BAE [2]. Other treatment strategies include surgical resection and bronchial arterial ligation [3].

Here, we describe a case of asymptomatic RHBA with no complications or recurrence following a successful transcatheter arterial embolization.

2. Case report

A 70-year-old woman was admitted to our hospital with an abnormality found on a routine chest X-ray. She had no symptoms or history of lung disease. A physical examination showed no abnormalities. Her chest X-ray revealed a nodular lesion overlapping the pulmonary artery at the right hilar region (Fig. 1A). Contrast-enhanced computed tomography (CT) of the chest revealed hyperplasia, dilatation, and

tortuosity of the right bronchial artery which branched from the aortic arch. No other abnormalities, such as lung field or heart abnormalities, were observed (Fig. 1B, C, D). Four-dimensional reconstruction of the CT image revealed that aneurysmal dilatation of the long segment of the right bronchial artery, which was suspected of RHBA, was contrasted during the early arterial phase, but enhanced during the pulmonary arterial phase (Fig. 1E). Bronchial arteriography revealed an aneurysmal dilatation in the long segment of the right bronchial artery (Fig. 2A) and a shunt from the right bronchial artery to the right lower pulmonary artery (Fig. 2B and C). Thus, she was diagnosed with RHBA.

In view of the risk of hemoptysis, she underwent BAE of the RHBA. A bilateral femoral artery catheterization was performed under local anesthesia. The right bronchial artery was approached using a 4-Fr guiding sheath from the right femoral artery and a 5-Fr guiding sheath from left femoral artery. Triaxial systems insertion was performed using a small (1.7/2.4-Fr) microcatheter (Excelsior SL-10; Stryker, Michigan, USA; internal diameter (ID) 0.016 inch) and a large (4.2-Fr) catheter (Asahi Fubuki; Asahi Intecc, Tokyo, Japan; ID 0.043 inch), via the right-sided guiding sheath, and a small 1.9-Fr microballoon catheter (Scepter XC; Terumo, Tokyo, Japan; ID 0.0165 inch) and a large (2.1/2.8-Fr) microcatheter (Sniper; Terumo, Tokyo, Japan; ID 0.027 inch) via the

Abbreviations: BAE, bronchial arterial embolization; CT, computed tomography; ID, inner diameter; NCBA, N-butyl-2-cyanoacrylate; RHBA, primary racemose hemangioma of the bronchial artery.

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<https://doi.org/10.1016/j.rmcr.2020.101060>

Received 3 February 2020; Received in revised form 16 April 2020; Accepted 17 April 2020

Available online 21 April 2020

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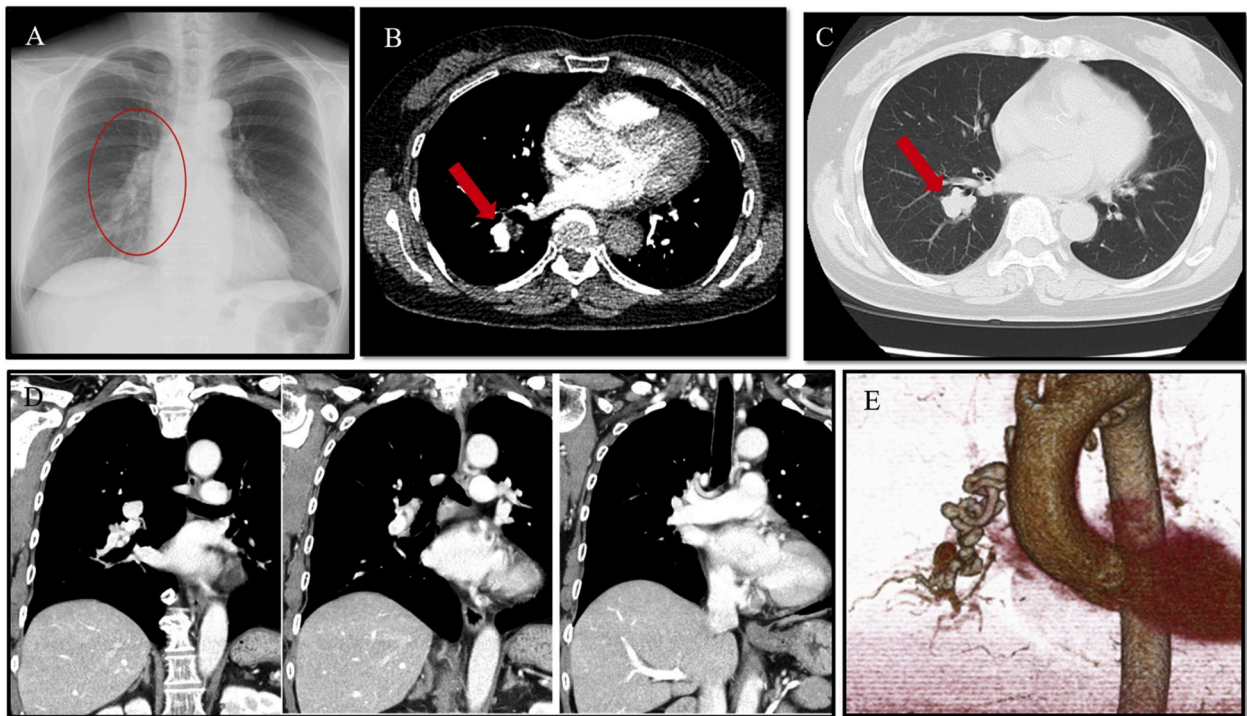


Fig. 1. (A) Chest X-ray showing a nodular lesion overlapping the pulmonary artery at the right hilar region. (B) Chest contrast-enhanced computed tomography (CT) showing convolution and dilatation of the bronchial artery (arrow). (C) Chest CT showing a nodular lesion in the right lower lobe of the lung (arrow). (D) Coronal image of enhanced lung CT showing hyperplasia, dilatation and tortuosity of the right bronchial artery, which has originated from the aortic arch. (E) Four-dimensional reconstruction of the CT revealing that the primary racemose hemangioma of the bronchial artery was not shown during the early arterial phase, but enhanced during the pulmonary arterial phase.

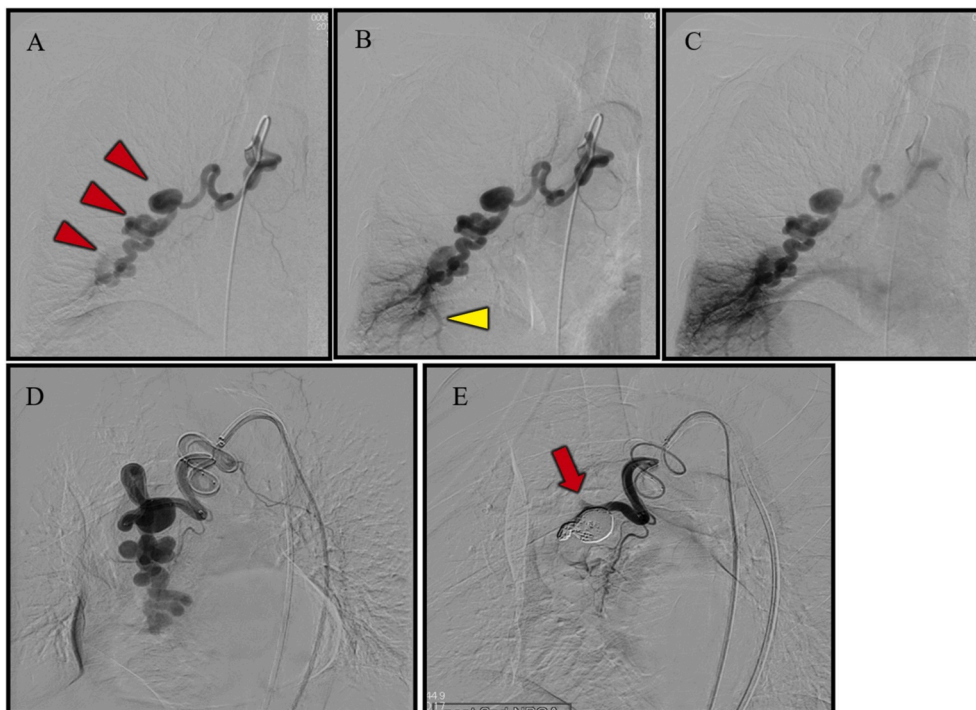


Fig. 2. A bronchial arteriography revealing (A) an aneurysmal dilatation in the long segment of the right bronchial artery (arrows). (B) A shunt from the primary racemose hemangioma of the bronchial artery (RHBA) to the right lower pulmonary artery (arrow). (C) An enhanced lung due to the pulmonary artery via a bronchial artery. (D) RHBA before bronchial arterial embolization (BAE) in the right anterior oblique at a 45-degree-angle. (E) Absent opacification of RHBA after BAE, and the right bronchial artery (arrow).

left-sided guiding sheath for the angiography. We performed embolization using eight coils (Target XL 360 Soft; Stryker, MI, USA; 6 mm/20 cm, Orbit Galaxy Complex Fill; Codman Inc., MA, USA; 7 mm/21 cm and 7 mm/21 cm, DELTAMAXX, Codman Inc., MA, USA; 20 mm/60 cm, 20

mm/60 cm, 16 mm/50 cm, and 8 mm/35 cm, Target XL helical; Stryker, MI, USA; 5 mm/15 cm) via Excelsior SL-10. We additionally used a 2.0 ml of mixture of *N*-butyl-2-cyanoacrylate (NBCA; Histoacryl; B. Braun, Melsungen, Germany) and iodized oil (lipiodol; Guerbet, Tokyo, Japan);

NBCA:lipiodol 1:3) under balloon occlusion (Fig. 2D and E).

No complications occurred during or after embolization. Follow-up, extending to 36 months after the coil embolization, was satisfactory.

3. Discussion

This case report has two key findings. First, RHBA should be included in the differential diagnosis when a nodular lesion is seen overlapping the pulmonary artery at the hilar region. Thus, a contrast-enhanced CT should be performed, even in elderly asymptomatic patients. Second, in asymptomatic patients with RHBA, a treatment strategy that includes surgery and BAE should be used, considering the risk of hemoptysis.

RHBA is rarely diagnosed in asymptomatic patients. Narato et al. reported that in 34 patients with RHBA, the chief complaint was hemoptysis in 27 patients (79%), hemoptysis in 3 patients (8%), cough in one patient, dyspnea in one patient, and that there were no symptoms in 2 patients (5%). Asymptomatic patients were diagnosed with RHBA after a chest X-ray or CT was performed to rule out other diseases [2]. Chest X-ray findings for RHBA include hilar lymph node enlargement and a mass shadow at the hilum [4,5]. Contrast-enhanced chest CT and bronchial arteriography are useful for diagnosing RHBA [6]. As shown in our case, RHBA with hyperplasia, dilatation, and tortuosity of the bronchial artery were shown by CT and bronchial arteriography. Furthermore, bronchial arteriography can reveal the presence of a shunt from the RHBA to the pulmonary artery. Such a shunt is a risk factor for hemoptysis since the pressure in the bronchial artery, which is part of the systemic circulation, is higher than that of the pulmonary artery, which is part of the pulmonary circulation. In this case, the patient showed no symptoms, and was diagnosed because of a routine checkup.

Second, hemoptysis caused by a ruptured RHBA can be life-threatening [4]. There are no definite prognostic guidelines for asymptomatic RHBA [7]. Shinoda et al. reported that untreated RHBA grew over time [8]. In past reports, no correlation has been found between size and the risk of rupture [9]. Therefore, treatment of asymptomatic RHBA should be considered. Historically, RHBA was treated surgically by minimal resection of the lung and ligation of the bronchial artery. A BAE was performed in cases where surgery was expected to be difficult [6]. Remy et al. considered BAE efficient, as it is minimally invasive, quick, and preserves lung function. Therefore, BAE should be the first-choice treatment for RHBA [6]. In this case, considering the risk of hemoptysis, a BAE was performed although she had no symptoms. Due to the presence of a shunt from the RHBA to the right lower pulmonary artery, embolization to right bronchial artery was performed using coils and NBCA.

In this case, we used material coils and NBCA as embolization materials. Coil embolization is the best choice when a microcatheter can reach the aneurysm, because coils can be delivered directly into the sac of the aneurysm without migration of embolization materials. However, in this case, judging from the size and shape of the sac, we expected that a large number of coils would be required.

NBCA is a liquid embolization material whose time to coagulation

after injection can be controlled by dilution with Lipiodol, and the combined use of a coil and NBCA can decrease the number of coils required. For bronchial artery embolization, the disadvantage of NBCA is the risk of the materials escaping to pulmonary veins and systemic arteries. To prevent distal embolization, we used material coils and balloon occlusion for reduction of blood flow.

In summary, we present a case of successful BAE for RHBA in an asymptomatic patient. RHBA should be considered when an abnormal lung shadow is observed on a chest X-ray. Even in the absence of symptoms, BAE should be considered as a treatment strategy to prevent hemoptysis.

Funding

This case report did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Declaration of competing interest

The authors declare no conflict of interest.

Acknowledgments

We would like to thank Editage (www.editage.com) for English language editing.

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