

Case Report

Near Lethal Hemoptysis Caused by the Recanalized Bronchial Racemose Hemangioma

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ABSTRACT

Bronchial racemose hemangioma (BRH) is a rare vascular malformation. We present a case of a 79-year-old female complaining of hemoptysis who had received the bronchial arterial embolization (BAE) for BRH twice. She was brought to our emergency room due to massive hemoptysis. Because fresh hemoptysis continued, she developed respiratory insufficiency. The bronchoscopy with suction showed the massive bleeding from the left bronchus. A dynamic and three-dimensional CT demonstrated the recanalized BRH. A selective bronchial angiography showed convoluted bronchial artery, and then consecutive BAE with Spongel was performed. She was extubated the next day of BAE, and then discharged after five days. She is doing well one year after the treatment.

Keywords: Bronchial racemose hemangioma, Hemoptysis, Bronchial arterial embolization

Abbreviation: BRH: Bronchial Racemose Hemangioma; CT: Computed Tomography; BAE: Bronchial Arterial Embolization

Introduction

Several pulmonary diseases, such as tuberculosis, bronchiectasis, and primary lung cancer, cause hemoptysis. Bronchial racemose hemangiomas (BRH) are rare vascular malformations, and frequently cause hemoptysis. We herein report a case of BRH with near lethal hemoptysis.

Case Report

A 79-year-old Japanese female first visited our hospital in January 2016 with hemoptysis. She was never smoker, nor family history of

malignancies. A chest X-ray did not detect an abnormal shadow, but a computed tomography (CT) showed infiltration of the left lung opacities. A dynamic CT revealed convolution and 3.4 -mm dilated of bronchial vessels, and three-dimensional CT demonstrated a bronchial racemose hemangioma (BRH) extending from descending aorta (Figure 1A). A selective bronchial angiography showed 3-mm dilated convolution and pooling of contrast injection in the left upper lobe (Figure 1B). Therefore, a bronchial arterial embolization (BAE) was performed with absorbable gelatin sponge (Spongel) consecutively. After BAE, the BRH could not demonstrate, and then she discharged two days after the treatment. In March 2017, she

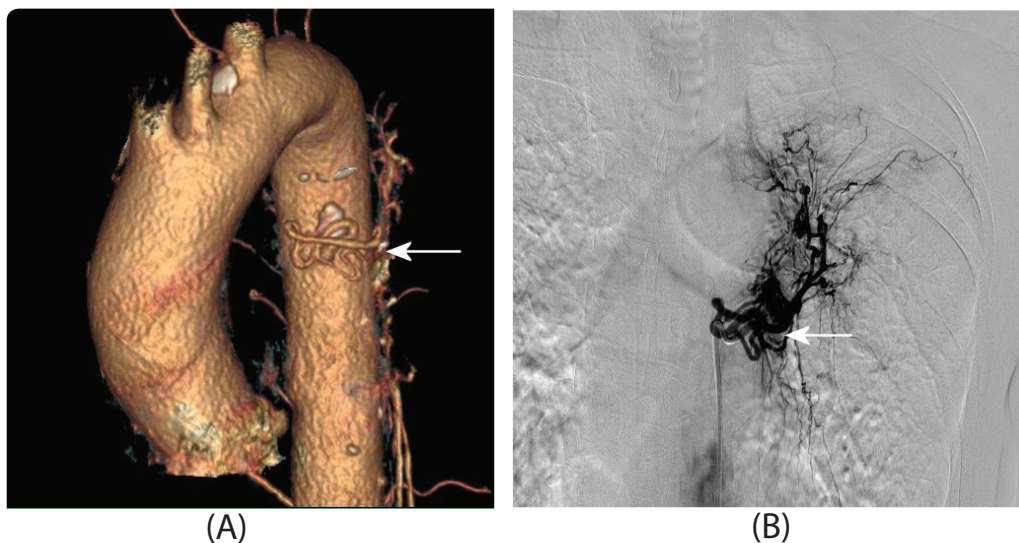


Figure 1A,B: A: three-dimensional CT demonstrates the dilated bronchial racemose hemangioma (BRH) from descending aorta (white allow). B: selective bronchial angiography shows the dilated convolution and pooling of contrast injection in the left upper lobe (white allow).

visited our hospital again with hemoptysis. At that time, the bleeding was stopped naturally. She was treated by an intravenous injection of a hemostatic agent, and then went home.

In August 2017, she was brought to the emergency room of our hospital due to massive hemoptysis. On arrival at our hospital, she was no loss of consciousness and had a Glasgow Coma Scale (GCS) score of 15. Her initial systolic blood pressure was 120 mmHg with a pulse of 70/minute. Because a fresh hemoptysis continued, she developed respiratory insufficiency and required endotracheal intubation for artificial ventilation. A bronchoscopy showed the massive bleeding from the left side of the bronchus (Figure 2A), therefore we performed the right-side intubation to save her healthy right lung. Chest CT showed bilateral lung opacities compatible with blood aspiration (Figure 2B). A dynamic CT revealed convolution and dilation of bronchial vessels, and three-dimensional CT evidenced the BRH from the same point of descending aorta (Figure 2C). A selective bronchial angiography showed the dilated convoluted bronchial artery and corroborated recanalization of previously embolized artery had recanalized (Figure 2D), so a consecutive BAE was performed with Spongel using the same procedure. After BAE, the BRH could not be demonstrated. She was extubated the next day of BAE, and then discharged after five days. She is doing well one year after the treatment, and three-dimensional CT cannot detect the recurrence of the BRH (Figure 3).

Discussion

Bronchial and non-bronchial systemic arteries are now considered to be the main source of hemoptysis [1-3]. BRH is characterized by not only a dilated and convoluted bronchial artery but also a vascular hyperplasia causing abnormal anastomosis to adjacent vessels [4]. Bavo, et al. [5] firstly described BRH as angioma-like changes in 19761. BRH has been classified into two types, congenital vascular malformation as a primary form and subsequent vascular malformation by broncho-pulmonary inflammation as a

secondary form. Narato et al. [6] reported 34 Japanese cases of BRH, and the authors demonstrated that 30 cases caused hemoptysis and 25 cases possessed lesions in right lungs among 16 male and 18 female population with a mean age of 49 years (range 19-80). The present case demonstrated as a primary BRH in the upper lobe of the left lung.

There have been a lot of reports about bronchoscopy at the time of hemoptysis [7,8]. Bronchoscopy produces more information about the bleeding point causing hemoptysis. Furthermore, it can use for intubation at the time of massive bleeding as a sucker, and for confirmation the tube position directly. In our case, bronchoscopy was very useful for intubation of the healthy lung in a short time.

BAE is well accepted in the management of hemoptysis, however, of the patients treated by BAE successfully, 20 percent rebled during a six-month follow-up [9]. The reason of recurrent hemoptysis seems to be a recanalization and recruitment. There were two peak times of recurrent hemoptysis, the first being from 1 to 2 months post-BAE and the second being from 1 to 2 years post-BAE. The first peak reflects the incomplete BAE such as a case of hemoptysis from the other non-bronchial arteries. In addition, the effect of absorbable gelatin sponge seems to continue for only a limited time, therefore, recurrence in the first peak could be caused by recanalization of the embolized vessels. The second peak reflects a recruitment of vasculature by the underlying pulmonary disease and indicates a relapse. The present case occurred recurrence of hemoptysis 20 months after an initial BAE. The embolized artery had recanalized, therefore, embolized absorbable gelatin sponge particles should be resorbed. In this way, absorbable gelatin sponge particles have a weak point. However, both permanent embolized particles and microcoils have a risk of neighborhood organ's necrosis and spinal cord paralysis. Recent technologies such as BAE with a super-selective microcatheter provide a long-term effect in the management of hemoptysis.

The other therapeutic candidates for BRH include bronchial arterial ligation and surgical resection. Hamano et al. described that bronchial arterial ligation by surgery had an advantage for the

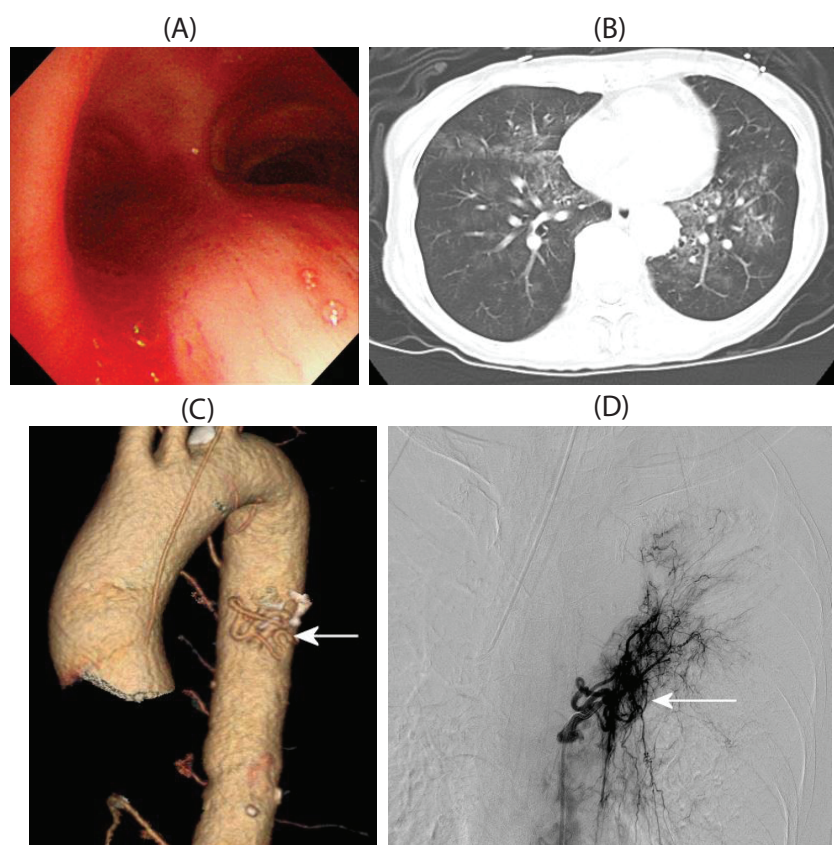


Figure 2A-B: A: bronchoscopy shows the massive bleeding from the left side of the bronchus. B: chest CT demonstrates bilateral lung opacities related to blood aspiration. C: three-dimensional CT demonstrates the recanalized BRH from descending aorta (white allow). D: selective bronchial angiography shows the same convolution and pooling of contrast injection in the left upper lobe (white allow).



Figure 3: Control three-dimensional CT after arterial embolization cannot detect the BRH from descending aorta now (white allow).

patient by means of certainty [10]. However, a general anesthesia must be essential, and the treatment may lead to be incomplete such as a case of hemoptysis from non-bronchial arteries. Furthermore, surgical resection of the bleeding region has a lot of complications; as an example, post-operative pneumonia, empyema, post-operative air-leakage, atelectasis, bleeding, and so on. Totally, about 20 percent

of the patients might have such complications, and the mortality was reported to 2 percent. Recently, Kanamori et al. described that bronchial arterial ligation by thoracoscopic surgery had an advantage for the patient by means of less invasiveness [11]. Kato et al. [12] reported that BRH also complicated bronchial-pulmonary arterial fistula, and the operation such as bronchial arterial ligation and left upper lobectomy was performed.

In conclusion, a primary BRH is an extremely rare vascular malformation, and it has a possibility to cause lethal hemoptysis. While BAE as an initial treatment of massive hemoptysis, this is a palliative procedure and potential for recurrence exists. We emphasize that both bronchoscopy and BAE is imperative for lethal hemoptysis.

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