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Chest

Massive hemoptysis due to multiple bronchial artery aneurysms and multiple aneurysmal dilations: A case report

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ABSTRACT

Bronchial artery aneurysm (BAA) is a rare but potentially life-threatening clinical entity. Patients with multiple BAAs and multiple aneurysmal dilations are even rarer. In this case report, we will investigate a case of multiple BAAs and multiple aneurysmal dilations arising from 2 right bronchial artery branches presenting with hemoptysis. The patient was successfully treated with transcatheter arterial embolization. We should be vigilant for the possibility of BAA when encountering patients presenting with hemoptysis. Transcatheter arterial embolization is safe and effective to solve this condition.

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Introduction

Bronchial artery aneurysm (BAA) is a rare but potentially life-threatening clinical entity, reportedly accounting for less than 1% of cases selected for bronchial arteriography. Patients with multiple BAAs and multiple aneurysmal dilations are even rarer. Here, we report a case of multiple BAAs and multiple aneurysmal dilations arising from 2 right bronchial artery branches presenting with hemoptysis. The patient was successfully treated with transcatheter arterial embolization (TAE).

Case report

A fit 21-year-old girl with intermittent hemoptysis for 3 days amounting to 600 mL was admitted to our Respiratory Intensive Care Unit. There was no associated history of hemoptysis. Before coming to our hospital, the patient took some Yunnan Baiyao (a traditional Chinese hemostatic agent) prescribed by a local clinic, but her hemoptysis was not relieved.

On admission, the patient was conscious, tachypneic (25 breaths/min), and tachycardiac (108 beats/min). Her blood

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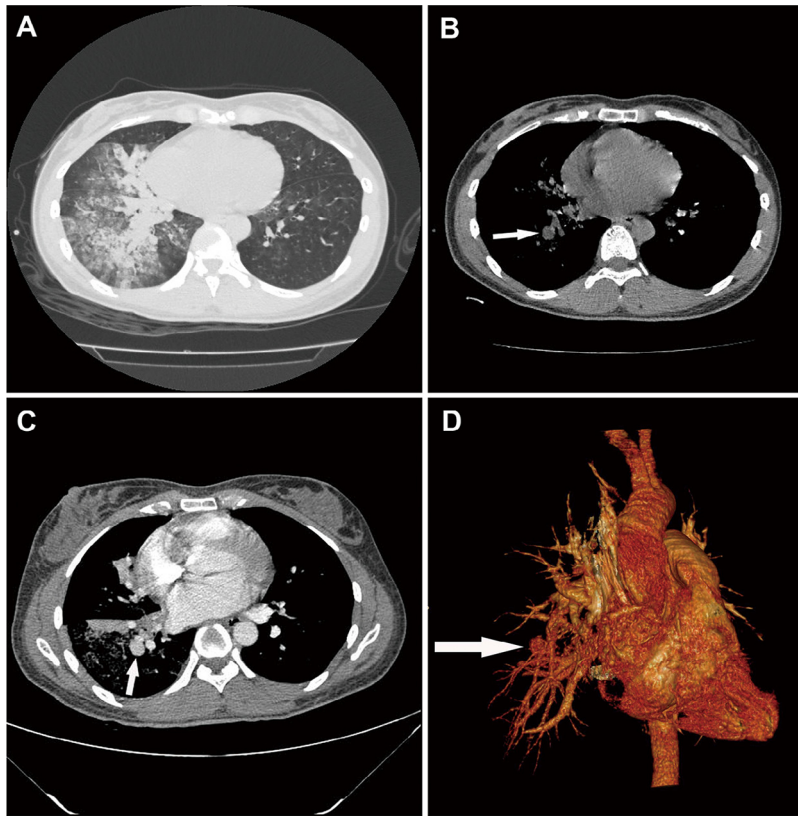


Fig. 1 – (A) A chest computed tomography (CT) image showed patchy ground-glass opacity and consolidation in the middle and lower lobes of right lung. **(B)** A small mass shadow adjacent to posterior basal segment of the right lower lobe can be seen in the soft tissue window (white arrow). **(C)** A contrast-enhanced CT and **(D)** 3D CT displayed clearly a small mass suspicious for BAA (white arrows).

pressure was normal (131/75 mm Hg). Rales were detected in the right lung through lung auscultation. The laboratory results were as follows: white blood cell count was 7210 cells/ μ L (normal range: 4000-10,000 cells/ μ L) with 77.09% (normal range: 40%-75%) neutrophils, C reactive protein level was 1.53 mg/L (normal range: 0-10 mg/L), erythrocyte sedimentation rate level was 11 mm/h (normal range: 0-20 mm/h), and procalcitonin level < 0.05 ng/mL (normal range < 0.05 ng/mL). Liver and renal functions were normal. Coagulation profile was within normal limits. Serum tumor markers and D-dimer were negative. Further examinations for possible connective tissue disease, such as antinuclear antibody, extractable nuclear antigen, and antineutrophil cytoplasmic antibody, revealed normal results.

An emergency chest computed tomography (CT) scan showed patchy ground-glass opacity and consolidation in the middle and lower lobes of right lung (Fig. 1A). Of note, a small mass shadow adjacent to posterior basal segment of the right lower lobe could be seen in the soft tissue window (Fig. 1B). A contrast-enhanced CT clearly displayed the small mass suspicious for BAA whose density was the same as thoracic aorta (Fig. 1C). We found an aneurysm by carefully studying a 3-dimensional reconstruction of the CT scan (Fig. 1D). Subsequent selective bronchial arteriography confirmed the finding and revealed at least 3 aneurysms with varying sizes in the right upper bronchial artery branch and multiple aneurysmal dilations, tortuous-like beads in the 2 right bronchial

arteries (Fig. 2A and B). Considering the aneurysms and aneurysmal dilations were the cause of hemoptysis, we determined to perform embolization to prevent recurrence. We first filled aneurysm sacs with polyvinyl alcohol particles of varying sizes (Contour, Boston Scientific, MA) and then embolized the right bronchial artery trunk using 6 coils (Tornado, 3 mm/2 mm \times 3, 4 mm/2 mm \times 3, Cook Medical Inc, Bloomington, IN). A postprocedural angiogram confirmed embolization of the right bronchial arteries. The multiple BAAs and multiple aneurysmal dilations were completely occluded (Fig. 2C). The postprocedural CT pulmonary arteriography (CTPA) was satisfactory, with thrombosis formed in the aneurysm sac in artery phase (Fig. 2D).

The patient was discharged without any complications. Postoperative recovery was uneventful. Through the follow-up phone call, her condition remained stable.

Discussion

BAA is a rare but potentially life-threatening disease that is observed in less than 1% of patients selected for bronchial arteriography [1]. Patients with multiple BAAs are even rarer. Most published papers reported cases of single or double BAAs. To date, there was only 1 reported case of 3 occurrences of BAAs

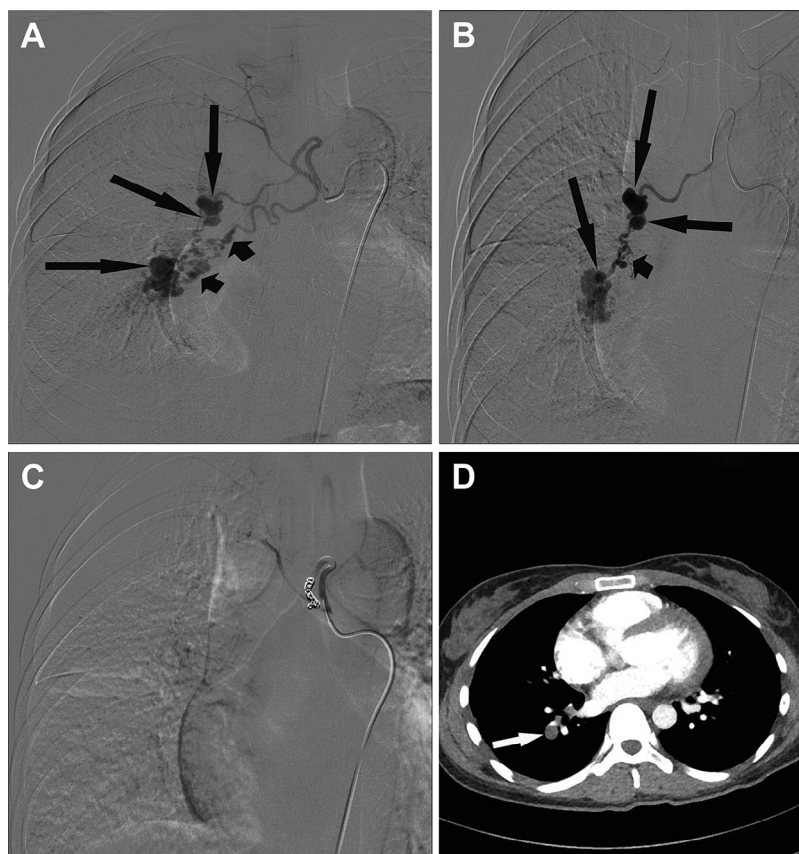


Fig. 2 – (A, B) Selective bronchial arteriography demonstrated multiple bronchial artery aneurysms (black arrows) and multiple aneurysmal dilation (black arrowheads) arising from 2 right bronchial artery branches. (C) Bronchial arteriography after embolization with polyvinyl alcohol and 6 coils confirmed cessation of blood flow to the bronchial arteries; the aneurysms are not visualized. (D) A postprocedural computed tomography pulmonary arteriography image showing the aneurysm thrombotic change in the artery phase.

with unknown etiology [2]. In this paper, we reported a case of at least 3 BAAs and multiple aneurysmal dilations in right bronchial arteries.

The underlying etiology of BAA has not been clarified. It has been widely accepted that increased bronchial arterial flow and focal weakening of vessel wall contribute to the formation of BAA [3]. BAA development can be congenital or acquired. An analysis by Di et al. revealed that BAA was most associated with bronchiectasis, followed by other factors, including hypertension, chronic obstructive pulmonary disease, vasculitis, tuberculosis, chronic bronchopulmonary infection, and trauma [4]. However, many cases with BAA had no definite cause. Taking our case as an example, the patient was a fit young girl, without evidence of infection, drug abuse, trauma, or autoimmune diseases. After ruling out known factors, we considered the BAAs and tortuous dilations of bronchial arteries congenital.

BAAs can be anatomically classified as intrapulmonary, mediastinal, or both. The clinical manifestations of BAAs may vary, depending on where the aneurysm is located and whether the aneurysm has ruptured. The condition may manifest as hemoptysis [1,2,5], mediastinal hemorrhage [2,6,7], hemothorax [8], or hematemesis [2,7,9]. Our case presented with intermittent hemoptysis for 3 days. For some cases with intact aneurysms, BAA was usually identified incidentally on thoracic

scanning. The risk factors that lead to aneurysm ruptures are unknown. There is no correlation between the diameter of the aneurysm and the likelihood of rupture [4,10]. To reduce the risk of life-threatening complications, the incidental finding of a BAA of any cause should be assumed a “time bomb,” and immediate management should be undertaken regardless of the presence of symptoms.

Contrast-enhanced chest CT, in the arterial phase, discloses a high index of suspicion for BAA, but it cannot lead to an explicit diagnosis. Definitive diagnosis should be confirmed by selective bronchial artery angiography. Moreover, selective bronchial arteriography plays a therapeutic role. Nowadays, endovascular techniques such as TAE have been increasingly applied in clinical practices, as it is less invasive than surgical extirpation and able to provide satisfactory exclusion of the aneurysm, with a shorter hospital stay [11]. TAE was employed in our case with the aim to totally occlude the aneurysm and tortuous bronchial arteries. All sorts of embolic materials have been successfully used in clinical practices, such as detachable coils, gelatin sponges, or *N*-butyl cyanoacrylate [12]. Among these, coils are most commonly used to treat BAA. They can be positioned at the target site with fewer complications than other agents [13,14]. To prevent retrograde filling of the aneurysm, both the afferent and the efferent of BAA need

to be occluded [15]. In our case, we first filled aneurysm sacs with polyvinyl alcohol particles of varying sizes and then embolized the right bronchial artery trunk using 6 coils in view of multiple BAAs and multiple aneurysmal dilations of the 2 right bronchial artery branches. Arteriography after embolization confirmed cessation of blood flow to the bronchial arteries, and the aneurysms were no longer visualized.

For some patients with severe complications such as rupture [6], patients who have complex anatomy [16], or patients allergic to contrast medium, surgical extirpation may be an alternative or a remedial measure.

Conclusion

We reported a rare and unusual case of multiple BAAs and multiple aneurysmal dilations arising from 2 right bronchial artery branches, which were successfully treated with TAE. When encountering patients presenting at an emergency room with hemoptysis, we should be vigilant for the possibility of BAA. Endovascular treatment such as TAE is safe and effective for this potentially life-threatening condition.

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