Clinical Case Report

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Endovascular treatment of ectopic bronchial artery aneurysm with brachiocephalic artery stent placement and coil embolization

A case report and literature review

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Abstract

Background: Bronchial artery aneurysm (BAA) is an uncommon but potentially life-threatening disease, and multiple BAAs are even rarer. Clinically, the tortuous and short neck of a BAA may present significant challenges for invasive intervention.

Methods: This report describes the detailed process of diagnosis and treatment and includes a literature review of the etiology, clinical presentation, and therapeutic management of BAA.

Results: A rare case of multiple BAAs, with one having an inflow artery arising from the brachiocephalic trunk, was referred to our hospital. The patient was successfully treated with coil embolization and brachiocephalic artery stent placement. In addition, we conducted a literature review involving 63 cases of BAA. BAA was most commonly associated with bronchiectasis and was located predominantly in the mediastinum. There was no significant difference between the diameters of the ruptured aneurysms and those of the nonruptured aneurysms ($P=0.115$). Transcatheter arterial embolization was the most commonly adopted technique to treat BAA, while thoracic aortic endovascular repair was selected if the neck between the aneurysm and the aorta was short. Subgroup analysis suggested that patients with >1 BAA were significantly more likely to be female than male (χ^2 test, $P=0.034$).

Conclusion: Transcatheter coil embolization combined with stent placement could be a reasonable treatment option for BAAs with a tortuous and short neck. According to our literature review, patients with multiple BAAs display distinctive clinical characteristics compared with patients with a single BAA.

Abbreviations: BAA = bronchial artery aneurysm, CT = computed tomography, TAE = transcatheter arterial embolization.

Keywords: brachiocephalic artery stent, brachiocephalic trunk, bronchial artery aneurysm, coil embolization

1. Introduction

Bronchial artery aneurysm (BAA) is a rare but potentially deadly disease, reportedly accounting for <1% of all aneurysms, revealed by selective bronchial arteriography.[1] Patients with multiple BAAs are even more rarely seen. Here, we report a case of multiple BAAs arising from an ectopic bronchial artery. The patient was successfully treated with coil embolization combined with stent placement. The etiology, clinical manifestations, and therapeutic treatment of BAA are herein discussed, with reference to 63 cases reported previously.

2. Consent

This study adhered to the tenets of the Declaration of Helsinki. Informed consent was signed by the patient for publication of this report and its related images.

3. Case presentation

A 50-year-old female with chronic chest pain radiating to her back was referred to our hospital for further examination. She had been treated for hypertension for 7 years with felodipine (Plendil; 5 mg daily). The patient was normal on physical examination. Her laboratory results were generally nonsignificant, except mildly elevated high-sensitivity C-reactive protein 9.36 mg/L, gamma-glutamyl transferase 83 U/L, aspartate aminotransferase 38 U/L, and erythrocyte sedimentation rate 25 mm/h.

A contrast-enhanced computed tomography (CT) scan revealed multiple aneurysms with the largest lesion adjacent to the brachiocephalic artery. By carefully studying a 3-dimensional reconstruction of a CT scan, we found 3 aneurysms, 2 of which were bronchial artery aneurysms, and one was in the phrenic artery. The largest one, 2.5×2.2 cm, was located in the mediastinum and was fed by an abnormal bronchial artery branching from the brachiocephalic trunk (Fig. 1); the other 2 were inside the right lung, with 1 (1.0×0.9 cm) fed by right bronchial artery and 1 (1.1×1.1 cm) fed by phrenic artery.

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The authors report no conflicts of interest.

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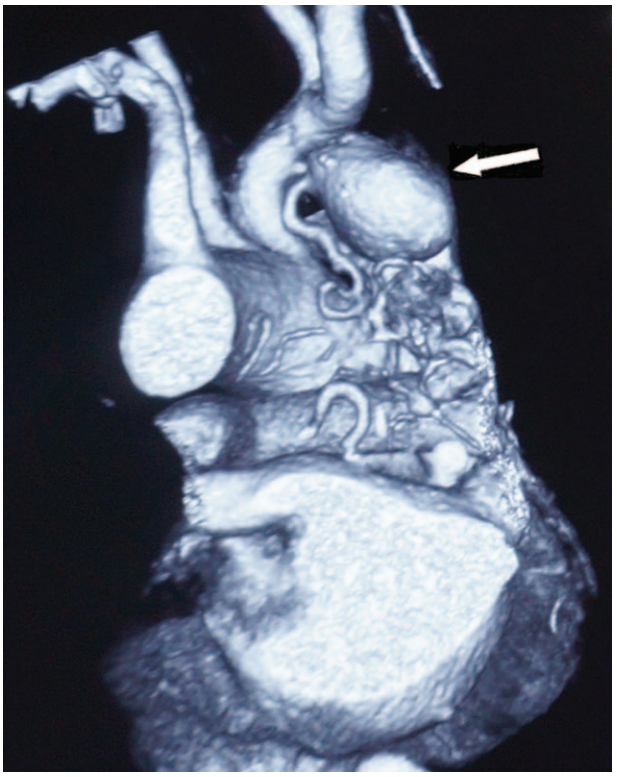

Figure 1. Three-dimensional reconstruction of contrast-enhanced computed tomography scan. The white arrow indicates the bronchial artery aneurysm adjacent to the brachiocephalic trunk. Note the ectopic bronchial artery branching from the brachiocephalic trunk and the extremely short neck of the aneurysm.

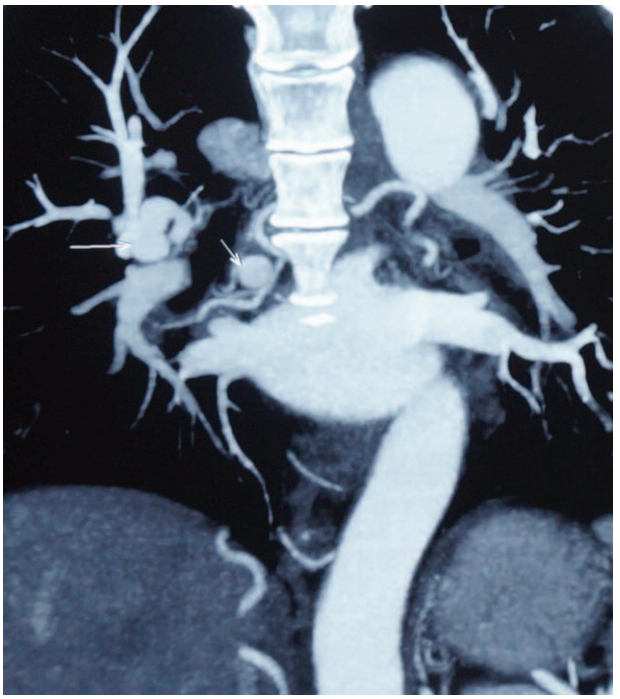

Figure 2. Coronary reconstruction of contrast-enhanced computed tomography scan. The 2 small white arrows from left to right show the other bronchial aneurysm and phrenic artery aneurysm, respectively.

Enlargement of the right bronchial and phrenic artery were also observed (Fig. 2).

We decided to treat only the ectopic bronchial artery aneurysm localized in the mediastinum via an endovascular approach. Considering the short neck of the aneurysm, we combined transcatheter arterial embolization with stent placement. Following aortic arch aortography via a femoral approach (Fig. 3A), a selective brachiocephalic arteriography demonstrated the ectopic BAA arising from 1 abnormal branch of brachiocephalic trunk (Fig. 3B). We managed to advance into the outflow artery and released several interlocking coils (Cook Medical, Bloomington, IN) into the outflow artery, the aneurysm sac, and then the inflow artery. A bare-metal stent (Cordis, 12–40 mm) was then successfully deployed across the origin of the abnormal bronchial artery at the brachiocephalic trunk. Owing to the length of the brachiocephalic trunk, the origin of the right subclavian artery was covered.

A postprocedural angiogram confirmed embolization of the aneurysm and patent right common carotid and subclavian arteries (Fig. 3C). The postoperation CT angiogram was satisfactory, with thrombosis formed in the aneurysm sac (Fig. 4).

The patient returned 4 months after the procedure. The CT angiogram showed a patent right subclavian artery, and the completely thrombosed aneurysm (Fig. 5).

4. Discussion

BAA occurs rarely. We searched the PubMed database with the search term "bronchial artery aneurysm" and found 56 articles with relevant data for 63 cases (Table 1). We reviewed these cases to enhance our understanding of the etiology, clinical presentations, and therapeutic management of BAA.

The etiology of BAA is poorly understood. It has been suggested that both increased bronchial arterial flow and focal weakening of the vessel wall contribute to the pathogenesis of BAA.[2] Our analysis of the relevant literature revealed that BAA was most commonly associated with bronchiectasis (Fig. 6), with other factors being hypertension, chronic obstructive pulmonary disease vasculitis, chronic bronchopulmonary infection, tuberculosis, and trauma. However, many patients with BAA had a clean medical history.

The clinical manifestations of BAA vary and depend on where the aneurysm is located and whether the aneurysm has ruptured. For patients who had a ruptured aneurysm, the most common symptom was chest pain, and then hemoptysis, back pain, epigastric pain, and symptoms related to shock. For patients with intact aneurysms, the BAA was usually identified incidentally upon thoracic scanning.

Other BAA-related symptoms included hemoptysis, dysphagia, chest pain, cough, hoarseness, dyspnea, pneumonia, and fever. There was no significant difference between the diameters of the ruptured aneurysms (mean 2.93 cm) and those of the nonruptured aneurysms (mean 2.18 cm; $P = 0.115$). Therefore, there may be no correlation between size of the aneurysm and the likelihood of rupture.

Our review of the literature also showed that most aneurysms (83.3%, 45/54) were located in the mediastinum, with either the left (20/50) or right (26/50) bronchial artery as the feeding artery. In addition, aneurysms arising from both bronchial arteries were observed (8.0%, 4/50), but intrapulmonary aneurysm of the left side was rarely encountered (3.7%, 2/54).

Because rupture can threaten life, BAA should be treated immediately after the diagnosis is established. With the

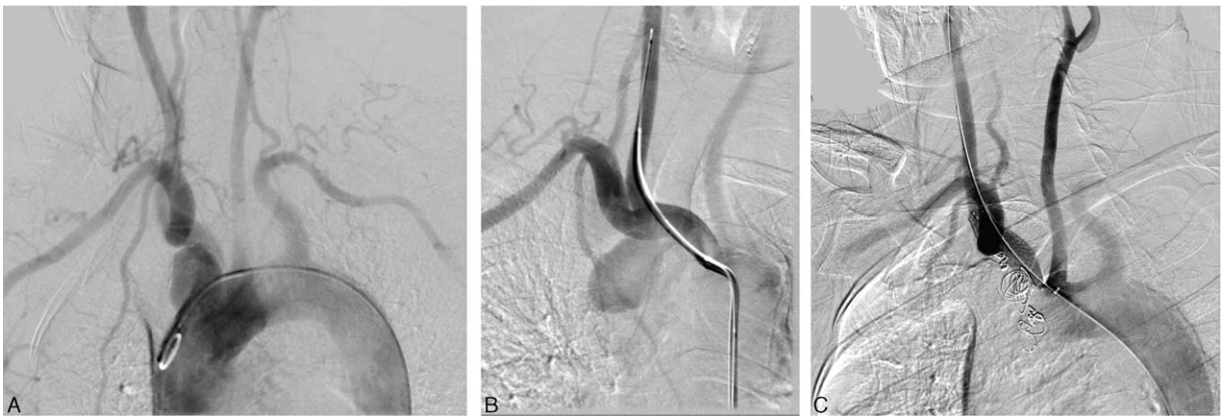

Figure 3. Images of aneurysm. (A) Aortic arch aortography in a frontal view. This angiogram shows an aneurysm adjacent to the brachiocephalic trunk. (B) Selective brachiocephalic arteriogrphy before embolization. The aneurysm was fed by an abnormal bronchial artery branching from the brachiocephalic trunk. (C) Postprocedure angiogram in a frontal view. The ectopic bronchial aneurysm had been successfully embolized by interlocking coils. The right common carotid artery and subclavian artery were patent.

improvement of endovascular technique, less invasive approaches are more commonly applied as initial treatments. These include transcatheter arterial embolization (TAE) of the bronchial artery, and occasionally thoracic aortic endovascular repair.

According to the literature, TAE was performed on 46 patients diagnosed with BAA. Among these cases, 5 procedures were unsuccessful and were succeeded immediately with surgical intervention or a second TAE.[3–6] After temporal success in another 5 patients, refilling of the aneurysm on follow-up imaging and recurrence of symptoms was documented.[7–11] The reasons for failure of a TAE were complicated, but may be related to the greatly enlarged bronchial artery and revascularization by collateral arteries.

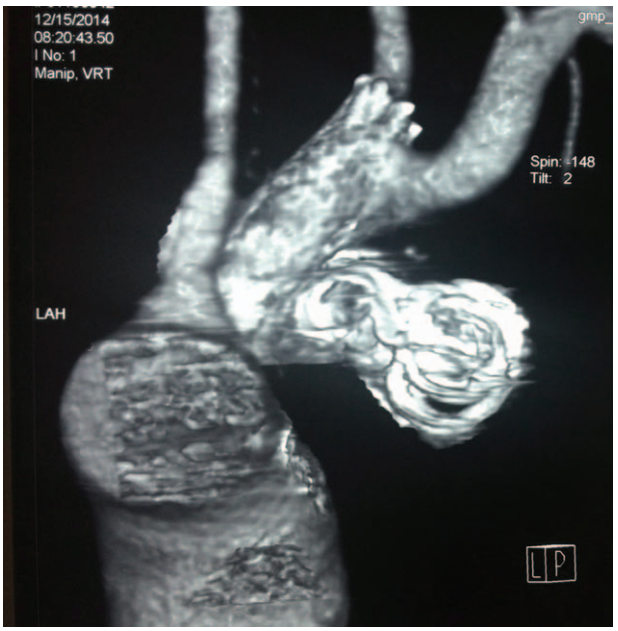

Figure 4. Three-dimensional reconstruction of contrast-enhanced computed tomography scan 1 week after operation. The aneurysm was successfully embolized by interlocking coils. The brachiocephalic trunk was patent and protected by a bare metal stent.

One of the challenging problems in treating some BAAs was the short neck between the aneurysm and aorta—in our case, between the aneurysm and the brachiocephalic trunk. Thoracic aortic endovascular repair was selected to solve the short neck issue, as reported in 9 articles.[4,12–19] In our case, we chose to implant a stent across the ectopic bronchial artery to avoid movement of the interlocking coils. To our best knowledge, this is the first time that combined TAE and stent was used in treating BAA. Long-term follow-up data are still needed, as thoracic aortic endovascular repair was reported to fail eventually.[17]

In addition, we conducted a subgroup investigation of patients with multiple BAAs, for which only 12 cases were available in the literature.[3,10,17,20–28] These patients were slightly younger (mean age, 50.7 years) than cases of single BAA reported in the literature (mean age, 59.3 years). We also observed that patients with >1 BAA were significantly more likely to be female (80%) than male (χ^2 test, $P=0.034$). Also, aneurysms arising

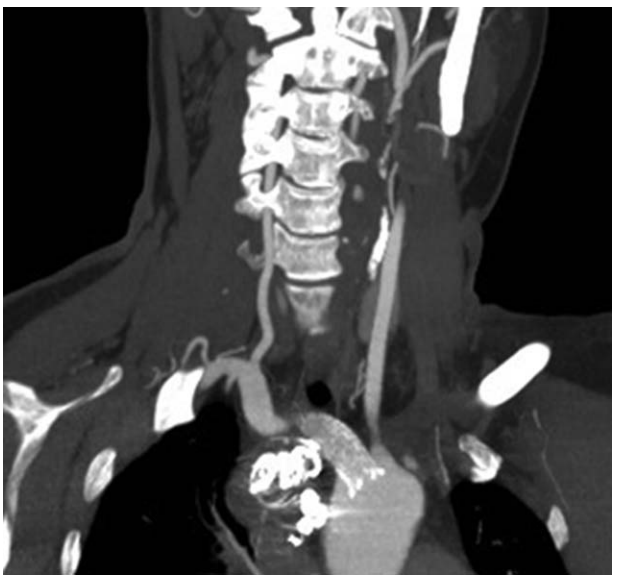

Figure 5. Coronary reconstruction of contrast-enhanced computed tomography scan 4 months after operation. The brachiocephalic trunk and right subclavian artery were patent and the aneurysm remained thrombosed.

Table 1

Review of 56 articles involving 63 cases of bronchial artery aneurysms.

Year	Studies	Patients	First author, reference
2014	4	4	Cao et al[20], Kaufman et al[29], Watanabe et al[3], Nakamura et al[30]
2013	7	7	Kim et al[31], Kim et al[12], Hori et al[21], Iida et al[22], Kawai et al[32], Rognoni et al[33], Arici et al[13]
2012	1	1	Guzzardi et al[14]
2011	3	7	Tsuboi et al[34], Hu et al[15], Lu et al[2]
2010	4	4	Misselt et al[35], Lee et al[36], Takahashi et al[16], Kamper et al[37]
2009	2	2	Mizuguchi et al[38], Shih et al[39]
2008	5	5	Lin and Wood[40], Yajima et al[7], Cecka et al[41], Zhang et al[42], Kotelis et al[23]
2007	2	2	Sanchez et al[4], Tsolaki et al[17]
2006	2	2	Wilson et al[43], Aburano et al[44]
2005	1	1	Karmy-Jones et al[45]
2004	2	2	Chatterjee et al[46], Haddad[47]
2003	3	3	Fukunaga et al[8], Kasashima et al[18], Suen et al[48]
2002	1	1	Tringali et al[49]
2001	2	2	Pugnale et al[50], Sakuma et al[51]
2000	2	2	Saito et al[52], Shimokawa et al[24]
1999	3	5	Sancho et al[9], Yanagihara et al[5], Vernhet et al[53]
1998	2	2	Sakai et al[19], Herb et al[10]
1997	2	2	Kalangos et al[54], Oka et al[55]
1996	1	1	Hoffmann et al[6]
1995	2	2	Cearlock et al[56], Ishizaki et al[25]
1977–1994	5	6	Hall et al[59], Osada et al[26], Shaer and Bashist[58], Remy-Jardin et al[11], Connolly et al[57]
Total	56	63	

from the right bronchial artery were seen in 6 of 8 patients. There was no significant difference between multiple BBAs and single BBA (χ^2 test, $P=0.246$). In terms of etiology, 4 patients were associated with bronchiectasis, Hughes-Stovin syndrome, tuberculosis, and silicosis, respectively, whereas others were idiopathic. These findings suggest that the mechanism(s) underlying the development of multiple BAAs may be distinct from that of a single BAA.

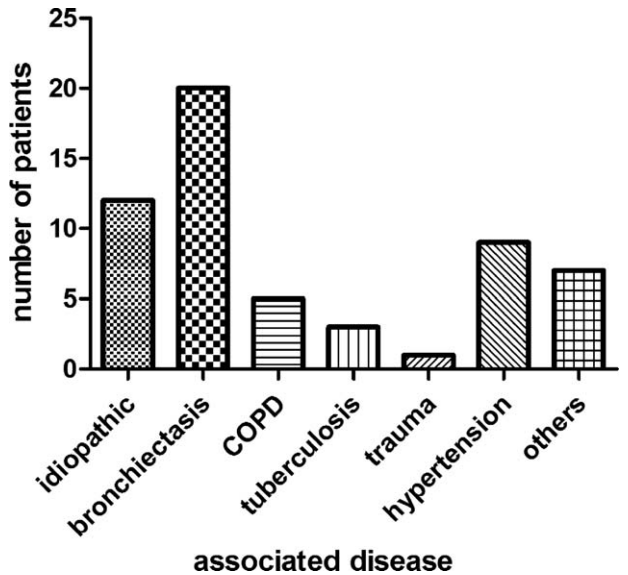

Figure 6. Common associated diseases of bronchial artery aneurysm. COPD=chronic obstructive pulmonary disease, No=no associated disease identified.

5. Conclusion

BAA is a potentially life-threatening lesion that requires prompt treatment, regardless of its diameter or patient symptoms. With the improvement of endovascular technology, TAE has become the major treatment for BAA. However, the tortuous and short neck of the BAA may still present significant challenges for clinical intervention. Long-term follow-up studies are needed. In addition, patients with multiple BAAs display distinctive patterns and need further clinical investigations.

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