Endovascular treatment of bronchial aneurysms

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Disclosures: Authors have nothing to disclose with regard to commercial support.

Received for publication Sept 3, 2017; revisions received Dec 10, 2017; accepted for publication Dec 29, 2017; available ahead of print Feb 13, 2018.

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J Thorac Cardiovasc Surg 2018;156:e109-17

0022-5223/\$36.00

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<https://doi.org/10.1016/j.jtcvs.2017.12.121>

Bronchial artery aneurysm on 3-dimensional CT scan reconstruction.

Central Message

A total of 108 cases of bronchial artery aneurysms have been reported. Embolization must be considered as the treatment of choice. Aortic stent-graft implantation or open surgery is required in selected cases.

See Editorial Commentary page e119.

▶ Video clip is available online.

Bronchial artery aneurysms (BAAs) are a rare condition with a reported prevalence of less than 1% of all selective bronchial arterial angiograms.¹ Its rupture can cause a life-threatening hemorrhage. We report a case of a mediastinal BAA treated with embolization.² Because this disease is rare, the literature is reviewed, and several principles are suggested to improve the management.

CLINICAL SUMMARY

A 64-year-old man presented with an episode of acute chest pain. The patient had normal results on physical examination. His medical history included only arterial

FIGURE 1. A, CT scan and 3-dimensional reconstruction confirmed a BAA in the initial part of the bronchial artery with a short neck (arrow). B, CT scan confirmed a BAA (12.1 mm).

FIGURE 2. A, Angiography shows the aneurysm of the BAA. B, Coil and Onyx (ev3, Irvine, Calif) embolization of the BAA. C, Control arteriogram, showing exclusion from the systemic circulation.

hypertension. The blood cell count, biochemistry, and coagulation showed normal results. Electrocardiography and chest radiography did not show any abnormal findings.

Contrast-enhanced computed tomography (CT) angiography demonstrated a single BAA, 12.1 mm in diameter, that was seen originating from the aortic arch (Figure 1). Through a right femoral artery access, the selective catheterization of the left bronchial artery was achieved with a 5F catheter. A selected bronchial arteriogram was obtained (Figure 2, A). The BAA was identified originating off the proximal left bronchial artery and 2 ecstatic branches of the bronchial arteries in addition to the aneurysm. Subsequently, the BAA was embolized using a microcatheter (Cantata 2.5; Cook Medical Inc, Bloomington, Ind) first with 1 microcoil (Detach-18, Cook Medical Inc) and then in a retrograde fashion with Onyx 18 (ev3, Irvine, Calif) (Figure 2, B). A “buffer” injection technique was used to prevent reflux into the aorta. A postembolization arteriogram demonstrated no aneurysm permeability (Figure 2, C). After the procedure, no complications occurred. The patient had an uneventful recovery and was discharged the day after. The postoperative CT angiogram was satisfactory (Video 1), with thrombosis of the aneurysm sac. The patient was followed up for 12 months, with no signs of recurrence.

DISCUSSION

Although BAA is a rare occurrence, Medline, Embase, Ovid, and the Web of Science were searched for available articles using the following keywords: “bronchial artery,” “bronchial artery aneurysm,” “mediastinal aneurysm,” “pulmonary aneurysm,” and “pulmonary vascular malformation.” One case from the author’s hospital (Valladolid University Hospital) was added to the review. Information was extracted by 2 independent reviewers. A total of 108 cases have been reported in the literature

(Table 1)³⁻⁹⁶ with a high variety of follow-up (3 days to 7 years).

The mean age of presentation was 57.9 years (15 days to 89 years). They can be classified as mediastinal, intrapulmonary, or both. The cause of BAAs is unknown; nevertheless, they are associated with bronchiectasis (19.4%), infectious disease (9.3%), atherosclerosis (3.7%), and tuberculosis (3.7%). However, many patients with BAAs had a clean medical history (idiopathic cause, 49.1%). The normal diameter of the bronchial arteries is smaller than 1.5 mm at the origin and smaller than 0.5 mm more distally. The mean size at presentation is 25 mm (3-121 mm).

Clinical presentation depends on the size, localization, and presence of concomitant disease and whether the aneurysm has ruptured. The most frequent clinical appearance is hemoptysis (30.5%), chest pain (9.3%), and hemomediastinum (8.3%), although 11 published cases

VIDEO 1. Endovascular treatment of bronchial aneurysms. Video available at: [http://www.jtcvsonline.org/article/S0022-5223\(18\)30043-6/fulltext](http://www.jtcvsonline.org/article/S0022-5223(18)30043-6/fulltext).

TABLE 1. Reported cases of endovascular treatment of bronchial aneurysms in the literature

Author	Year	Age, y	Clinical appearance	Ruptured	Diameter (mm)	Cause	Treatment	Success	Follow-up
Hirano and Maeda ³	1976	58	–	–	9	–	–	–	–
		54	–	–	5	–	–	–	–
Hall and colleagues ⁴	1977	56	Aortic dissection	Yes	9	Bronchiectasis	Lobectomy	Yes	–
Chantepie and colleagues ⁵	1980	15 days	Hemoptysis	–	15	Mycotic	Open surgery	No	Died
Watanabe and colleagues ⁶	1981	55	–	–	11	–	–	–	–
		70	–	–	12	–	–	–	–
Abet and Pietri ⁷	1981	50	Aortic dissection	–	9	Atherosclerosis	Thoracotomy	No	Died
Osada and colleagues ⁸	1986	62	Hemoptysis	Yes	30	Bronchiectasis	Aneurysm ligation and sclerosis	Yes	–
		56	Shock	Yes	12	–	Aneurysm extirpation + lobectomy	–	13 d
Katoh and colleagues ⁹	1987	–	Hemoptysis	No	–	–	Embolization	–	–
Tagushi and colleagues ¹⁰	1987	10	Hemoptysis	Yes	–	Mycotic	Pneumonectomy	Yes	–
Shaer and Bashist ¹¹	1987	79	Hematemesis	Yes	10	Atherosclerosis	–	No	Died
Remy-Jardin and colleagues ¹²	1991	50	Hemoptysis	–	–	Bronchiectasis	Embolization (dura-mater particles)	No	2 y
Fujita and colleagues ¹³	1991	66	–	–	25	Atherosclerosis	Lobectomy	Yes	–
Servois and colleagues ¹⁴	1992	34	Hemoptysis	–	10	Mycotic	Embolization (dura-mater particles)	Yes	4 wk
Connolly and colleagues ¹⁵	1994	2	Hemoptysis	–	–	Pseudoaneurysm (hyperimmunoglobulinemia E)	Embolization (PVA)	Yes	–
Komoda and colleagues ¹⁶	1994	57	Asymptomatic	–	15	–	Embolization	Yes	2 y
Cearlock and colleagues ¹⁷	1995	22	Hemoptysis	Yes	15	Posttraumatic	Embolization (particles)	Yes	–
Ishizaki and colleagues ¹⁸	1995	25	Massive hemothorax	Yes	50	Vascular malformations	Embolization (coil)	Yes	–
Nakajima and colleagues ¹⁹	1995	41	Asymptomatic	–	57	–	Embolization (coils)	Yes	13 mo
Hoffmann and colleagues ²⁰	1996	70	Acute superior vena cava obstruction	Yes	–	Atherosclerotic	Surgery clipping	Yes	2 mo
Siegler and colleagues ²¹	1996	60	Hemoptysis	–	7	Mycotic	Embolization (gel foam)	No	–
Kalangos and colleagues ²²	1997	50	Aortic dissection	Yes	15	Degenerative	Surgical ligation	Yes	–
Oka and colleagues ²³	1997	54	Tumor	–	10	–	Embolization (gel foam)	Yes	1 mo
Sakai and colleagues ²⁴	1998	72	Hemoptysis	–	25	Mycotic	Embolization (coils) + aortic stent-graft	Yes	7 mo
Sancho and colleagues ²⁵	1999	50	Chest pain	–	40	–	Embolization (gel foam + coils)	Yes	6 mo
Vernhet and colleagues ²⁶	1999	57	Hemoptysis	No	–	Bronchiectasis	Embolization (gel foam + coils)	Yes	2 y

(Continued)

TABLE 1. Continued

Author	Year	Age, y	Clinical appearance	Ruptured	Diameter (mm)	Cause	Treatment	Success	Follow-up
Yanagihara and colleagues ²⁷	1998	50	Hemoptysis	No	20	Bronchiectasis	Embolization (coils)	Yes	1 wk
		64	Hemoptysis	–	22	Bronchiectasis	Embolization (gel foam + coils)	Yes	6 y
		64	Hemoptysis	–	30	Bronchiectasis	Embolization (gel foam + coils)	Yes	3 y
Braks and colleagues ²⁸	1999	41	–	–	–	Bronchiectasis	Embolization	Yes	–
Saito and colleagues ²⁹	2000	33	Tumor	–	20	–	Lobectomy and aneurysmectomy	–	–
Shimokawa and colleagues ³⁰	2000	25	Back pain and shock	–	–	–	Embolization (coils)	Yes	7 y
Pugnale and colleagues ³¹	2001	72	Epigastric pain	Yes	15	–	Embolization (cyanoacrylate)	Yes	1 mo
Sakuma and colleagues ³²	2001	63	Asymptomatic	–	30	–	Embolization (coils)	Yes	18 mo
Tringali and colleagues ³³	2002	48	Dysphagia	–	40	–	Surgical resection	Yes	6 mo
Fukunaga and colleagues ³⁴	2003	60	Chest and back pain, dysphagia, hematemesis	Yes	50	–	Embolization (coils)	Yes	4 y
Kasashima and colleagues ³⁵	2003	79	Worsening hoarseness	–	60	Tuberculosis	Aortic stent-graft	Yes	18 mo
Suen and colleagues ³⁶	2003	50	Hemothorax	Yes	–	Sarcoidosis	Surgical excision	Yes	6 mo
Tanaka and colleagues ³⁷	2003	78	Tumor	–	40	–	Surgical excision	Yes	–
Chatterjee and colleagues ³⁸	2004	59	Chest pain, shock	Yes	–	–	Coil embolization	Yes	8 d
Haddad ³⁹	2004	62	Hemoptysis	–	–	Bronchiectasis	Surgical resection	–	–
Karmy-Jones and colleagues ⁴⁰	2005	78	Hemothorax	Yes	16	Tuberculosis	Embolization (coils)	Yes	11 mo
Aburano and colleagues ⁴¹	2006	30	Asymptomatic	–	20		Embolization (cyanoacrylate)	Yes	3 mo
Kalva and Wicky ⁴²	2006	29	Hemoptysis	–	10	Cystic fibrosis	Embolization (PVA + coils)	Yes	2 y
		36	Atelectasis		13	Cystic fibrosis	Embolization (coils)	Yes	5 mo
Sanchez and colleagues ⁴³	2007	69	Dysphagia		40	Tuberculosis	Aortic stent-graft + embolization (fibrin)	Yes	3 d
Tsolaki and colleagues ⁴⁴	2007	76	–		–	Bronchiectasis and interstitial fibrosis	Aortic stent graft + embolization (microspheres + coils)	No	10 mo
Cecka and colleagues ⁴⁵	2008	50	Epigastric pain		10	–	Embolization (coils)	Yes	2 y
Lin and Wood ⁴⁶	2008	37	Chest discomfort		40	–	Surgical resection	Yes	1.5 mo
Lioulis and colleagues ⁴⁷	2008	22	Hemoptysis		3	Bronchotomy, mycotic	Surgical resection	Yes	4 mo
Yajima and colleagues ⁴⁸	2008	69	Cough		70	–	Embolization (cyanoacrylate + coils)	No	2 y
Mizuguchi and colleagues ⁴⁹	2009	77	Shock	Yes	30	Bronchiectasis	Embolization (coils)	Yes	9 mo

(Continued)

TABLE 1. Continued

Author	Year	Age, y	Clinical appearance	Ruptured	Diameter (mm)	Cause	Treatment	Success	Follow-up
Shih and colleagues ⁵⁰	2009	60	Back pain	Yes	—	—	—	—	—
Kamper and colleagues ⁵¹	2010	89	Chest pain	No	—	—	No	—	—
Takahashi and colleagues ⁵²	2010	74	Asymptomatic	No	60	Bronchiectasis	Embolization (coils) + aortic stent-graft	Yes	6 mo
Misselt and colleagues ⁵³	2010	53	Asymptomatic	No	—	—	Embolization (coils)	Yes	3 mo
Guerra and colleagues ⁵⁴	2011	65	Shock	Yes	8	—	Surgical resection	Yes	2 y
Hu and colleagues ⁵⁵	2011	67	Chest pain	Yes	40	—	Embolization (gelatin sponge) + aortic stent-graft	Yes	1 y
Karmakar and colleagues ⁵⁶	2011	56	Hemoptysis	—	5	Tuberculosis	Embolization (PVA)	Yes	2 mo
Lü and colleagues ¹	2011	43	Hemoptysis	—	3	Bronchiectasis	Embolization (PVA + Histoacryl, TissueSeal, Ann Arbor, Mich)	Yes	—
		52	Hemoptysis	—	8	Bronchiectasis	Embolization (PVA + Histoacryl)	Yes	—
		62	Hemoptysis	—	14	Bronchiectasis	Embolization (PVA + Histoacryl)	Yes	2 mo
		50	Hemoptysis	—	12	Bronchiectasis	Embolization (PVA + Histoacryl)	Yes	—
		63	Hemoptysis	—	4	Bronchiectasis	Embolization (PVA + Histoacryl)	Yes	4 mo
Quero-Valenzuela and colleagues ⁵⁷	2011	82	Hemomediastinum + hemothorax	Yes	—	—	Embolization (coils + microparticles)	Yes	8 d
Seo and Kwak ⁵⁸	2011	41	Hemomediastinum + hemothorax	Yes	—	—	Embolization	Yes	16 d
Tsuboi and colleagues ⁵⁹	2011	82	Hemoptysis	No	33	Bronchiectasis	Embolization (gelatin sponge)	Yes	11 d
De Dominicis and colleagues ⁶⁰	2011	44	Chest pain	No	6	Right pulmonary agenesis Coronary-to-BAA	Embolization (coils) + pneumonectomy	Yes	6 mo
Cao and colleagues ⁶¹	2012	57	Swallowing discomfort	No	—	—	—	—	—
Guzzardi and colleagues ⁶²	2012	66	Dysphagia	No	40	Bronchiectasis	Embolization (coils) + aortic stent-graft	Yes	6 mo
Hori and colleagues ⁶³	2012	78	Chest pain	Yes	10	—	Embolization (coils)	Yes	1 wk
Tan and Thirugnanam ⁶⁴	2012	53	Cough	No	42	—	Surgical resection	Yes	—
Arici and colleagues ⁶⁵	2013	64	Asymptomatic	No	18	Mycotic	Aortic stent-graft	Yes	30 d
Iida and colleagues ⁶⁶	2012	70	—	No	—	—	Embolization (coils)	Yes	—
Kawai and colleagues ⁶⁷	2013	62	—	No	19	—	Embolization (cyanoacrylate)	Yes	6 mo
Kim and colleagues ⁶⁸	2013	79	Hemomediastinum	Yes	7	—	Embolization (gel foam + coils)	Yes	1 y
Martinez and colleagues ⁶⁹	2013	70	Hemomediastinum	Yes	15	—	Embolization	Yes	—

(Continued)

Adult: Other: Case Report

TABLE 1. Continued

Author	Year	Age, y	Clinical appearance	Ruptured	Diameter (mm)	Cause	Treatment	Success	Follow-up
Cheng and Ly ⁷⁰	2014	59	Cough	No	4	Cirrhosis	–	No	–
Kaufman and colleagues ⁷¹	2014	61	Hemomediastinum	Yes	57	–	Embolization (coils + spheres)	Yes	3 wk
Kim and colleagues ⁷²	2014	73	Hemoptysis	No	32	Bronchiectasis	Embolization (PVA + gel foam) + aortic stent-graft	Yes	2 y
Nakamura and colleagues ⁷³	2014	63	Tumor	No	25	–	Surgical resection (VATS)	Yes	6 mo
Patel and colleagues ⁷⁴	2014	12	Hemoptysis	No	–	Mycotic	Embolization (PVA)	Yes	1 mo
Vosse and colleagues ⁷⁵	2014	76	Chest pain Hemomediastinum	Yes	55	–	Embolization (coils)	Yes	6 wk
Watanabe and colleagues ⁷⁶	2014	71	Hemoptysis	No	–	–	Embolization	–	–
Xiaowei and colleagues ⁷⁷	2014	44	Hemoptysis	Yes	–	–	Surgical resection	Yes	–
Divisi and colleagues ⁷⁸	2015	75	Hemothorax	Yes	6	–	No	Yes	2 y
Kim and colleagues ⁷⁹	2015	55	Hematemesis hemomediastinum	No	–	–	Embolization (glue + coils)	Yes	1 y
Kitami and colleagues ⁸⁰	2015	63	Tumor	No	–	–	Surgical resection	Yes	35 d
Mahmood and colleagues ⁸¹	2015	73	Hemoptysis	No	20	–	Embolization (PVA + coils)	Yes	1 y
Latip and colleagues ⁸²	2016	61	Hemoptysis	No	–	Mycotic	Surgical resection	Yes	–
Rognoni and colleagues ⁸³	2015	58	Asymptomatic	No	44	–	Embolization (Amplatzer [St Jude Medical, St Paul, Minn] + coils)	Yes	1 wk
Samura and colleagues ⁸⁴	2015	77	Tumor	No	53	–	Embolization (coils) + aortic stent-graft	Yes	2 y
Shi and colleagues ⁸⁵	2015	52	Chest pain	Yes	25	–	Embolization (PVA + coils)	Yes	1 y
Di and colleagues ⁸⁶	2016	50	Asymptomatic	No	25	–	Embolization (bare metal stent + coils)	Yes	4 mo
Fiorucci and Simonte ⁸⁷	2016	89	Asymptomatic	No	38	–	Aortic stent-graft	Yes	6 mo
Stewart and Norris ⁸⁸	2016	65	Horner syndrome	No	–	–	–	–	–
Izaaruene and colleagues ⁸⁹	2016	59	Dysphagia Cough	No	55	–	Embolization (Onyx [ev3, Irvine, Calif] + coils)	Yes	1 mo
Mahmoud and colleagues ⁹⁰	2016	70	–	–	–	–	Embolization (Amplatzer)	Yes	–
Sato and colleagues ⁹¹	2016	59	Hemoptysis	Yes	–	Tracheal stent migration	Embolization	No	–
Song and colleagues ²	2016	59	Chest pain Dysphagia	No	18	–	Embolization (coils) + aortic stent-graft	Yes	3 mo
		63	Chest pain Hemomediastinum	Yes	5	–	Embolization (coils) + aortic stent-graft	Yes	3 mo
Wang and colleagues ⁹²	2016	66	Hemomediastinum	Yes	–	–	Embolization (coils)	Yes	–

(Continued)

TABLE 1. Continued

Author	Year	Age, y	Clinical appearance	Ruptured	Diameter (mm)	Cause	Treatment	Success	Follow-up
Hsieh and colleagues ⁹³	2017	63	Dyspnea	No	12	–	Embolization (cyanoacrylate)	Yes	1 y
Salamone and colleagues ⁹⁴	2017	63	Hemoptysis	No	9	COPD	Embolization (coils)	Yes	2 y
Ueda and colleagues ⁹⁵	2017	77	Hemoptysis		3	Mycotic	Embolization (cyanoacrylate)	Yes	7 mo
Dinis de Gama and Távora ⁹⁶	2017	65	Cough	No	60	–	Embolization	Yes	1 mo
Present case	2017	64	Asymptomatic	No	12.1	Bronchiectasis	Embolization (Onyx + coils)	Yes	1 y

PVA, Polyvinyl alcohol; BAA, bronchial artery aneurysm; VATS, video-assisted thoracoscopic surgery; COPD, chronic obstructive pulmonary disease.

have been asymptomatic (10%). For patients with a ruptured BAA (28.2%), the most common symptom is acute chest pain and then hemoptysis and back or epigastric pain. For patients with unruptured BAA (30.1%), it was usually identified incidentally on thoracic CT, unless thoracic pain, episodic hemoptysis, dysphagia, dyspnea, cough, or chest discomfort was present.

Diagnostic modes are CT scan and angiography. Magnetic resonance imaging has been used in a few cases. Chest radiography is considered as the initial examination, but its sensitivity is low.

There are 2 main treatment options described, open surgery (19.1%) and an endovascular approach (66.7%), with similar success rates (90.0% and 93.1%, respectively). Several surgical techniques have been used: aneurysm surgical excision/resection (12 cases), ligation (3 cases), lobectomy (4 cases), and pneumonectomy (2 cases).⁷³

Endovascular treatments are divided into selected embolization techniques (81.9%) or aneurysm exclusion by an aortic stent-graft (18.1%).^{87,89} Multiple materials have been used in the embolization of BAAs: coils (36.0%), cyanoacrylate (10.5%), polyvinyl acetate (10.5%), and gel foam (7.0%). The use of coils alone or in combination with gel foam, polyvinyl acetate, cyanoacrylate, or Onyx is the most popular technique described in the literature (15 cases).^{1,2,87}

CONCLUSIONS

An incidental finding of a BAA necessitates prompt treatment, regardless of the diameter, because its rupture can be life-threatening. Surgery should be reserved for patients with contraindications to embolization and should be selected on the basis of the patient's clinical status, comorbidities, and aneurysm's anatomy. An endovascular approach is considered the preferred way of treatment in patients with suitable anatomic features because of its safety, selectivity, efficacy, and low invasiveness.

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