Case Report

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Endovascular Treatment of Incidentally Found Multiple Aneurysms Originating from a Bronchial Artery: A Case Report

우연히 발견된 하나의 기관지 동맥에서 발생한 다발성 동맥류에 대한 혈관내 치료: 증례 보고

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Bronchial artery aneurysm (BAA) is a rare disease, and multiple aneurysms of a single bronchial artery are rarer. Regardless of the size of the lesion, it is at risk of rupture and can cause massive hemoptysis or severe pain. We report a rare case of bronchial artery embolization (BAE) of multiple aneurysms of a single bronchial artery. During medical examination, a 64-year-old female was diagnosed with multiple BAAs and endobronchial lesions in the right lower lung on CT 10 years prior to presentation to our hospital. Further evaluation of the lesions was recommended; however, the patient was lost to follow-up. The patient complained of dyspnea and visited our hospital, and the size of the BAA had increased on CT. BAE was done successfully using N-butyl-2-cyanoacrylate and detachable coils. Follow up CT after BAE showed significant decrease in extent of inflammatory lesion in the right lung.

Index terms Bronchial Arteries; Aneurysm; Endovascular Procedures

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INTRODUCTION

Bronchial artery aneurysm (BAA) is a rare disease; additionally, multiple aneurysms arising from a single bronchial artery are extremely rare. BAA is found in < 1% of selective bronchial angiograms (1), and multiple aneurysms from a single bronchial artery have only been reported twice to date (2). It may be symptomatic or asymptomatic; however, rupture of a BAA can cause massive bleeding, which can be potentially life threatening. Immediate treatment is required for patients with a BAA, and interventional treatment is primarily performed (3). We report the case of a 64-year-old female patient with multiple BAAs from the right bronchial artery showing improvement in inflammatory lesions in the right lung after bronchial artery embolization (BAE).

CASE REPORT

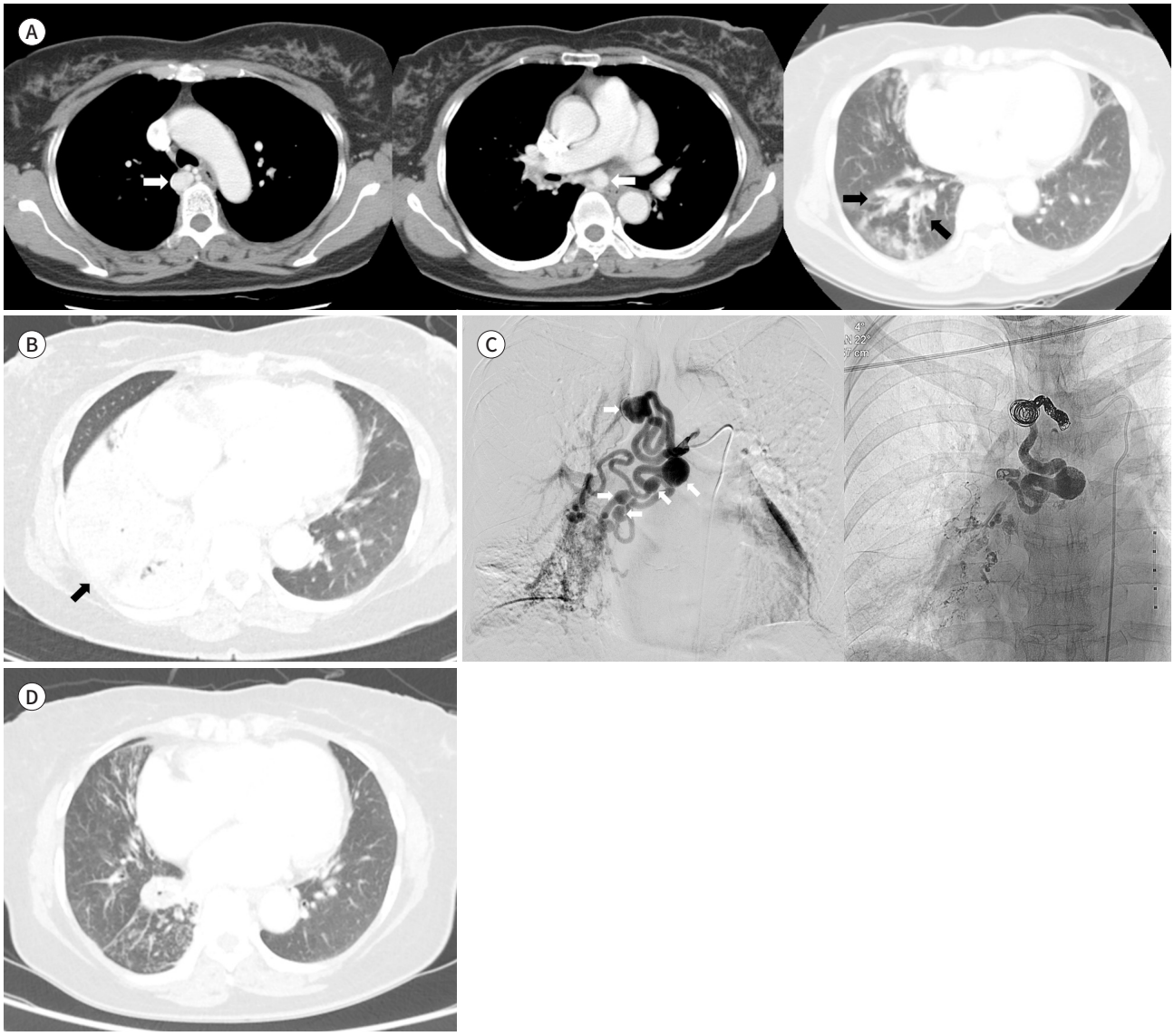
A 64-year-old female visited the pulmonary clinic in our hospital with complaints of dyspnea on exertion that had started a month prior to presentation. She had no other symptoms, such as hemoptysis, and had no underlying diseases, significant family history, history of smoking, or occupational factors. She had undergone contrast-enhanced chest CT 10 years prior to presentation because of abnormal findings during a regular checkup. The CT at that time revealed multiple BAAs, bronchiectasis, endobronchial soft tissue, bronchial wall thickening, and mucus plugging in the right lower lobe; the largest BAA measured 1.7 cm in diameter (Fig. 1A). Bronchoscopy revealed purulent secretions and nodular lesions with hyperemic mucosa. An acid-fast bacilli smear and culture with bronchial washing were performed, and the results were negative. The Department of Pulmonology recommended further evaluation and follow-up of the lesions. However, the patient was lost to follow-up for approximately 10 years. Chest CT performed during the present visit showed an increase in the number and size of the BAAs. More than five BAAs were identified, with the largest BAA showing an increase in diameter from 1.7 cm to 1.9 cm. In addition, the extent of obstructive pneumonitis and bronchiolitis in the right lower lung was significantly increased (Fig. 1B). The white blood cell count and erythrocyte sedimentation rate were increased (12570 cells/μL, 27 mm/h). Other laboratory test results were within the normal ranges. After obtaining written informed consent, the patient was referred to an interventional radiologist for BAE to prevent rupture.

Right bronchial artery angiography was performed via the right femoral artery using a 5-Fr catheter (Michaelson, Cook, Bloomington, IN, USA). Selective right bronchial artery angiography confirmed more than five BAAs and bronchial artery hypertrophy. Bronchial-to-pulmonary arterial shunting was observed in the right lower lung (Fig. 1C). After a 1.98-Fr microcatheter (Asahi Masters Parkway Soft, Asahi, Nagoya, Japan) was advanced into the main branch as distally as possible, N-butyl-2-cyanoacrylate (NBCA) with a lipiodol mixture at a ratio of 1:6 was administered distal to the most distal BAA. Because the most proximal BAA to the aorta was considered to be at risk of migration or reflux of embolic materials into the aorta, embolization was carefully performed using a total of 10 detachable coils (Concerto, Medtronic, Dublin, Ireland) (including two coils of 14 mm × 30 cm, one coil of 12 mm × 30 cm, one coil of 10 mm × 30 cm, one coil of 8 mm × 30 cm, four coils of 6 mm × 20 cm, and one

coil of 4 mm × 8 cm), instead of NBCA, from the aneurysm sac to the proximal feeding bronchial artery (Fig. 1C). The patient remained hemodynamically stable with no acute complications.

After BAE, the dyspnea on exertion improved. The 2-month and 8-month follow-up contrast-enhanced CT scans after BAE showed that the BAAs and right bronchial artery were filled with embolic material without contrast filling. The extent of post-obstructive pneumo-

Fig. 1. Multiple aneurysms originating from a bronchial artery in a 64-year-old female patient.
A. Initial chest CT images obtained 10 years ago show multiple BAAs arising from a right bronchial artery (white arrows) and focal bronchiectasis with peribronchial infiltrations in the right lower lung (black arrows).
B. Chest CT images at present illness show the increased extent of diffuse peribronchial consolidation with mucus plugging in the right lower lung, indicating post-obstructive pneumonitis (arrow).
C. Digital subtraction angiographic image shows more than five BAAs (arrows) originating from the right bronchial artery with bronchopulmonary shunting, and single-shot radiography after embolization showing multiple BAAs filled with N-butyl-2-cyanoacrylate and lipiodol mixture; the most proximal BAA was treated with detachable coils.
D. Follow-up chest CT, 2 months after bronchial artery embolization, shows a markedly decreased extent of post-obstructive pneumonitis and bronchiolitis in the right lower lung.
BAA = bronchial artery aneurysm



nitis and bronchiolitis in the right lower lung also markedly decreased during the follow-up (Fig. 1D). The patient had no complications or recurrent lesions during follow-up at 2 years and 8 months.

This study was approved by the Institutional Review Board of our institution (IRB No. 2022-11-011), and the requirement for informed consent was waived.

DISCUSSION

BAA is a rare, life-threatening disease that can cause massive hemoptysis and is found in < 1% of patients on selective bronchial angiography (1). Although little is known regarding the cause of BAA, increased blood flow to the bronchial artery and weakened blood vessel walls may contribute to aneurysmal dilatation of the bronchial artery (4). BAAs are classified as mediastinal, intrapulmonary, or both, according to their anatomical location, and as congenital or acquired, according to their etiology. It is mostly associated with atherosclerosis, bronchiectasis, bronchitis, inflammatory lung diseases, pulmonary sequestration, pulmonary aplasia, and systemic vascular abnormalities such as Osler–Weber–Rendu syndrome (5). In the present case, the multiple BAAs may have been caused by chronic bronchiectasis and obstructive pneumonitis.

Multiple BAAs are rarer than single or double lesions, and only 13 cases have been reported to date. The median age of patients with this disease is 57.5 years, and most patients are female (6). Our patient showed more than five BAAs, all of which originated from a single right bronchial artery. Cases of multiple BAAs arising from a single bronchial artery are extremely rare and have been reported only twice to date (2).

Patients with BAAs are usually asymptomatic before rupture or experience mass effects. The lesion is mostly found incidentally on CT scans. If the dilated artery ruptures, blood enters the bronchus and causes massive hemoptysis and hypovolemic shock, which are the most common symptoms (3). Rupture of such a BAA is irrelevant to its size and can have fatal consequences; therefore, it must be treated even if the patient is asymptomatic (1).

Treatments are largely divided into transcatheter arterial embolization (TAE) and surgery (3). Surgical treatments include aneurysm resection, pulmonary lobectomy, and bronchial artery ligation and are mainly used in ruptured BAA or in areas near the aorta that are difficult to access via catheters. TAE is less invasive than surgical treatment and can be performed in patients who cannot undergo surgery because of poor lung function or systemic conditions. However, there is a possibility of failure owing to limited access to the catheter depending on the shape of the blood vessels, and aneurysms may recur because of collateral blood flow after the procedure. Recently, embolization and catheterization techniques have been developed and have similar success rates to surgery, and the hospitalization period is short; hence, they are preferred in most cases (7).

It is important to select appropriate embolic materials for BAE in the case of BAAs. BAE has a high recurrence or recanalization rate (8). NBCA is better than polyvinyl alcohol in preventing recurrence and does not increase complications in BAE (9). We used NBCA to minimize the recanalization rate of the BAAs. NBCA is a powerful permanent embolic agent that can be infused distally, although its use requires considerable experience and skill. Diluted NBCA

(lipiodol mixture in a 1:6 ratio) was used to deliver NBCA to the most distally located BAA. However, the most proximal large BAA was embolized using detachable coils to prevent reflux or migration of the embolic agent into the aorta.

The right lower lung showed aggravated inflammatory lesions at the time of visit compared to the 10 years prior CT scan, which was the cause of dyspnea on exertion. Interestingly, there was an obvious gradual decrease in the extent of these inflammatory lesions on the follow-up CT after BAE. Considering the presence of bronchiectasis and the medication history of antitussives and expectorants, the probability of any association between this improvement and BAE was low. However, owing to the dramatic and persistent improvement of inflammation after BAE during follow-up, similar to transcatheter arterial microembolization (10), BAE might have contributed to the improvement of inflammation by reducing the blood flow, thereby decreasing the influx of inflammatory cells and proinflammatory cytokines to the diseased lung.

In conclusion, we report an extremely rare case of an incidental finding of more than five BAAs originating from a single bronchial artery that was successfully treated with BAE using NBCA and detachable coils. Endovascular embolization of BAAs associated with inflammatory lung lesions can prevent potential complications and may improve inflammation.

Author Contributions

Conceptualization, C.J.G., P.J., L.S.Y.; data curation, H.J.; methodology, K.S.H.; project administration, P.B.; supervision, C.J.G.; writing—original draft, Y.M.; and writing—review & editing, C.J.G.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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REFERENCES

1. Mizuguchi S, Inoue K, Kida A, Isota M, Hige K, Aoyama T, et al. Ruptured bronchial artery aneurysm associated with bronchiectasis: a case report. *Ann Thorac Cardiovasc Surg* 2009;15:115-118
2. Hou Z, Wang J, Peng D, Li C, Yang J, Wang J. Massive hemoptysis due to multiple bronchial artery aneurysms and multiple aneurysmal dilations: a case report. *Radiol Case Rep* 2018;13:24-27
3. Tanaka K, Ihaya A, Horiuci T, Morioka K, Kimura T, Uesaka T, et al. Giant mediastinal bronchial artery aneurysm mimicking benign esophageal tumor: a case report and review of 26 cases from literature. *J Vasc Surg* 2003;38:1125-1129
4. Lü PH, Wang LF, Su YS, Lee DH, Wang SX, Sun L, et al. Endovascular therapy of bronchial artery aneurysm: five cases with six aneurysms. *Cardiovasc Intervent Radiol* 2011;34:508-512
5. Ishizaki N, Shimokawa S, Tanaka K, Taira A, Onohara S, Tabata M, et al. Ruptured bronchial artery aneurysm associated with pleural telangiectasis and tortuous portal obstruction: report of a case. *Surg Today* 1995;25:852-854
6. Li Y, Gu GC, Liu B, Shao J, Chen Y, Zheng YH. Endovascular treatment of multiple bronchial artery aneurysms with prominent fistula to pulmonary artery in a patient with interstitial lung disease: a case report and literature review. *Vasc Endovascular Surg* 2019;53:492-496
7. Tsolaki E, Salviato E, Coen M, Galeotti R, Mascoli F. Double right bronchial artery aneurysm treated with combined procedures. *Eur J Vasc Endovasc Surg* 2007;34:537-539
8. van den Heuvel MM, Els Z, Koegelenberg CF, Naidu KM, Bolliger CT, Diacon AH. Risk factors for recurrence of haemoptysis following bronchial artery embolisation for life-threatening haemoptysis. *Int J Tuberc*

JOURNAL of THE KOREAN SOCIETY of RADIOLOGY

Lung Dis 2007;11:909-914
9. Woo S, Yoon CJ, Chung JW, Kang SG, Jae HJ, Kim HC, et al. Bronchial artery embolization to control hemoptysis: comparison of N-butyl-2-cyanoacrylate and polyvinyl alcohol particles. *Radiology* 2013;269:594-602
10. Okuno Y, Iwamoto W, Matsumura N, Oguro S, Yasumoto T, Kaneko T, et al. Clinical outcomes of transcatheter arterial embolization for adhesive capsulitis resistant to conservative treatment. *J Vasc Interv Radiol* 2017;28:161-167.e1

우연히 발견된 하나의 기관지 동맥에서 발생한 다발성 동맥류에 대한 혈관내 치료: 증례 보고

윤민혁[1] · 차중근[1]* · 박종민[1] · 이상엽[2] · 김시형[1] · 홍지훈[3] · 박병건[3]

기관지 동맥류는 전 세계적으로 드문 질환으로 특히 한 개의 기관지 동맥에서 기인한 여러 개의 동맥류는 매우 드물다. 이는 병변의 크기에 상관없이 파열의 위험이 있고 대량 객혈과 심각한 통증을 유발할 수 있다. 저자들은 하나의 기관지 동맥에서 발생한 다발성 동맥류에 대해 성공적으로 색전한 증례를 보고하고자 한다. 64세 여성이 10년 전 건강검진에서 시행한 흉부 CT에서 다수의 기관지 동맥류와 우하폐의 기관지 병변이 발견되었다. 환자는 이에 대한 추가적 검사를 권유받았으나 거절 후 추적 소실되었다. 이후 호흡곤란을 주소로 내원 후 시행한 흉부 CT에서 기관지 동맥류의 크기가 증가한 소견을 보였고, 동맥류에 대해 분리코일, N-butyl-2-cyanoacrylate를 이용하여 성공적인 색전술을 시행하였다. 색전술이후 추적 흉부 CT에서 우하폐의 염증성 병변의 범위가 감소한 것이 확인하였다.

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