INTRODUCTION

Pulmonary arterial aneurysms (PAAs) are very rare, the reported incidence being 0.007%.\(^1\) PAAs are classified according to causes as congenital, acquired, and idiopathic.\(^2\) Idiopathic PAAs are rare; however, several cases have been reported.\(^3\)\(^-\)\(^9\) Greene and Baldwin have proposed four pathological criteria for diagnosing an idiopathic PAA: simple dilatation of the pulmonary trunk with or without involvement of the rest of the arterial tree, the absence of intracardiac or extracardiac shunts, the absence of chronic cardiac or pulmonary disease, and the absence of arterial disease such as syphilis or more than minimal atheromatosis or arteriosclerosis of the pulmonary vascular tree.\(^10\) We here report a patient who underwent transcatheter pulmonary artery embolization for an idiopathic peripheral PAA and had a good clinical outcome.

CASE REPORT

A 53-year-old man was referred to our institution after a peripheral PAA had been discovered by contrast-enhanced computed tomography (CT) that demonstrated an 8.6 mm diameter aneurysm in the periphery of the right pulmonary artery A10 (Figure 1A,B). He chose follow-up observation. A CT one year later showed the diameter of the PAA had increased to 9.9 mm (Figure 1C-E). No abnormality was found on cardiac ultrasound examination, and his tricuspid valve pressure disparity was normal (14 mm Hg). He had the comorbidity of diabetes, which was well controlled. He had a smoking history (Brinkman index 3450); however, his respiratory function was normal. He had no history of infectious diseases such as syphilis or tuberculosis and no history of Behcet disease or Marfan syndrome.

2.1 Transcatheter embolization

Transcatheter pulmonary artery embolization was performed to prevent rupture of the peripheral PAA. After placement of a 4 Fr introducer sheath in the right femoral vein under local anesthesia, a right pulmonary artery angiogram confirmed the PAA at the branches of A10b and A10c (Figure 2A). To embolize the draining artery, an AMPLATZER™ Vascular Plug 4 and hydrogel-coated metallic coils were used. A10b, one of the draining arteries, was plugged with a 6-mm AVP 4, and A10c, the other draining artery, was plugged with a 7-mm AVP 4 (Figure 2B). The PAA was embolized with four

Key Clinical Message

The natural history of idiopathic peripheral pulmonary arterial aneurysms (PAAs) is unclear; however, they can cause sudden death by rupture. Our case illustrates the utility and low invasiveness of transcatheter embolization using an AMPLATZER™ Vascular Plug 4 and hydrogel-coated metallic coils in patients with idiopathic peripheral PAAs.

KEYWORDS

AVP 4, embolization, hydrogel-coated metallic coil, peripheral pulmonary arterial aneurysm
hydrogel-coated metallic coils, AZUR® CX35 (Azur peripheral hydrocoil; Terumo Medical Corporation) (Figure 2C). A10b+c, the feeding artery, was plugged with an 8-mm AVP 4. Occlusion of the PAA was confirmed by repeat angiography after embolization (Figure 2D).

3 | DISCUSSION

Pulmonary arterial aneurysms are very rare. Deterling and Clagett reported eight patients with PAAs in 109,571 consecutive postmortem examinations.1 Depending on the site of onset, PAAs are classified as central and peripheral. The term central PAAs denotes PAAs generated in the left and right main pulmonary arteries from the main pulmonary artery, whereas peripheral PAA denotes PAAs generated from a lobe branch in the periphery. Central PAAs are often secondary to pulmonary hypertension and vasculitis, whereas peripheral PAAs are often secondary to inflammation and trauma associated with arteriosclerosis.11 In the present patient, we considered the PAA to be idiopathic because he had none of these underlying diseases. Although secondary PAAs reportedly have a high incidence of secondary hemoptysis resulting from rupture,6,12,13 there are few reports of rupture of idiopathic peripheral PAAs. However,
According to Inaba et al, hemoptysis occurs in only one of 12 patients. In the past, it was recommended that pulmonary aneurysms should be treated as soon as the diagnosis had been made; however, the current recommendation is to resect only very invasive idiopathic peripheral PAAs, especially those considered at relatively high risk of rupture. In such patients, intravascular treatment is less invasive and therefore preferable to surgery. Embolization methods include embolization of the aneurysm, embolization of the feeding artery, embolization of both the feeding and draining arteries, occluding the aneurysm, and embolization of both the feeding and draining arteries. The procedure can be shortened by using an AVP to embolize both the feeding and draining arteries. Though it remains unclear which pulmonary aneurysms should be treated, transcatheter embolization is a minimally invasive means of treating for idiopathic peripheral PAAs. Accumulation of more cases is needed.

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CONFLICT OF INTEREST
The authors have no conflict of interests to disclose.

AUTHOR CONTRIBUTIONS
TT: conceived and designed the study, acquired the data, analyzed and interpreted the data, wrote the manuscript, and approved the final manuscript. TY, TK, TN, KK, SK: analyzed and interpreted the data, drafted the manuscript, and approved the final manuscript.

INFORMED CONSENT
Appropriate written informed consent was obtained for publication of this case report and accompanying images.

CONCLUSION
Transcatheter embolization using AVP 4 could become the standard means of treating idiopathic peripheral PAAs.
REFERENCES

1. Deterling RA Jr, Clagett OT. Aneurysm of the pulmonary artery: review of the literature and report of a case. Am Heart J. 1947;34:471-499.
2. Kreibich M, Siepe M, Kroll J, Hohn R, Grohmann J, Beyersdorf F. Aneurysms of the pulmonary artery. Circulation. 2015;131:310-316.
3. Shih HH, Kang PL, Lin CY, Lin YH. Main pulmonary artery aneurysm. J Chin Med Assoc. 2007;70:453-455.
4. Tsui EY, Cheung YK, Chow L, Chau LF, Yu SK, Chan JH. Idiopathic pulmonary artery aneurysm: digital subtraction pulmonary angiography grossly underestimates the size of the aneurysm. Clin Imaging. 2001;25:178-180.
5. Seguchi M, Wada H, Sakakura K, et al. Idiopathic pulmonary artery aneurysm. Circulation. 2011;124:e369-370.
6. Inaba H, Ohta S, Nishimura T, et al. Two cases of solitary peripheral pulmonary arterial aneurysm (In Japanese.). Nihon Kokyuki Gakkai Zasshi. 1998;36:384-388.
7. Haj-Yahia S, Shaib M, Bali K, et al. Case report and management approach in idiopathic pulmonary arteries aneurysm. J Cardiothorac Surg. 2018;13:110.
8. Greaves SW, Dye L 3rd, Aranda PS, et al. Perioperative management of a large idiopathic pulmonary artery aneurysm without pulmonary arterial hypertension. J Cardiothorac Vasc Anesth. 2018;32:2402-2408.
9. Sa-Kong H, Seol SH, No TH, et al. Huge idiopathic pulmonary artery aneurysm. Radiol Case Rep. 2017;12:236-239.
10. Greene DG, DeForest Baldwin E, Baldwin JS, Himmelstein A, Roh CE, Cournand A. Pure congenital pulmonary stenosis and idiopathic congenital dilatation of the pulmonary artery. Am J Med. 1949;6:24-40.
11. Ungaro R, Saab S, Almond CH, Kumar S. Solitary peripheral pulmonary artery aneurysms. Pathogenesis and surgical treatment. J Thorac Cardiovasc Surg. 1976;71:566-571.
12. Yanase K, Takizawa S, Nakamura M, et al. A case of tuberculous pulmonary artery aneurysm protruding into the bronchial lumen. J Japan Soc Respir Endoscopy. 1994;16:472-476.
13. Ianniello A, Carrafiello G, Nicotera P, Vaghi A, Cazzulani A. Endovascular treatment of a ruptured pulmonary artery aneurysm in a patient with behçet’s disease using the amplatzer vascular plug 4. Korean J Radiol. 2013;14:283-286.
14. Monchik J, Wilkins EW Jr. Solitary aneurysm of the middle lobe artery. A case report and review of solitary peripheral pulmonary artery aneurysms. Ann Thorac Surg. 1974;17:496-503.

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