Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

# Successful transcatheter embolization of an unruptured idiopathic peripheral pulmonary artery aneurysm ☆

*Chiaki Sato, MD[a], Takatoshi Kubo, MD, PhD[a,*], Eisuke Shibata, MD, PhD[a], Masahiro Yanagiya, MD, PhD[b], Jun Nakajima, MD, PhD[b], Osamu Abe, MD, PhD[a]*

[a] *Department of Radiology, The University of Tokyo Hospital, 7-3-1 Hongo, Bunkyo-ku, Tokyo, 113-8655, Japan*
[b] *Department of Thoracic Surgery, The University of Tokyo Hospital, 7-3-1 Hongo, Bunkyo-ku, Tokyo, 113-8655, Japan*

ARTICLE INFO

*Article history:*
Received 11 September 2022
Revised 19 September 2022
Accepted 25 September 2022

*Keywords:*
Transcatheter embolization
Pulmonary artery aneurysm
Coil embolization

ABSTRACT

Pulmonary artery aneurysms (PAAs) occurring in the distal branch of the pulmonary artery are rare, and there are only a few reports of transcatheter embolization of unruptured idiopathic peripheral PAAs. We report a case of a 66-year-old woman with a history of surgery for pulmonary arteriovenous malformations who presented with an idiopathic peripheral PAA of A10 confirmed by contrast-enhanced computed tomography and pulmonary artery angiography. Transcatheter embolization was performed to prevent rupture, and the aneurysm was successfully embolized. Although there is no consensus on the indications and treatment for unruptured idiopathic peripheral PAAs, transcatheter embolization may be a favorable treatment option.



## Introduction

Pulmonary artery aneurysms (PAAs) are rare, with autopsies reporting an incidence rate of approximately 0.007% [1]. Peripheral PAAs occurring in the distal branch of the pulmonary artery are more infrequent [2]. PAAs are classified as either congenital or acquired. Among acquired aneurysms, those without a history of secondary PAAs, such as pulmonary hypertension, connective tissue abnormalities, vasculitis, or trauma, are classified as idiopathic PAAs [2]. Idiopathic peripheral PAAs are very rare, and there are few reports of transcatheter treatment [3,4]. In this report, we present a case of transcatheter embolization of idiopathic peripheral PAA.

## Case

A 66-year-old woman had a history of multiple telangiectasias of the gastrointestinal tract and 2 partial left pulmonary resections for left pulmonary arteriovenous malformations (AVMs). The patient had no history of pulmonary hypertension, connective tissue abnormalities, vasculitis, or trauma.

☆ Competing Interests: The authors declare no conflicts of interest associated with this manuscript.

* Corresponding author.
E-mail address: kubo.tky@gmail.com (T. Kubo).

https://doi.org/10.1016/j.radcr.2022.09.087

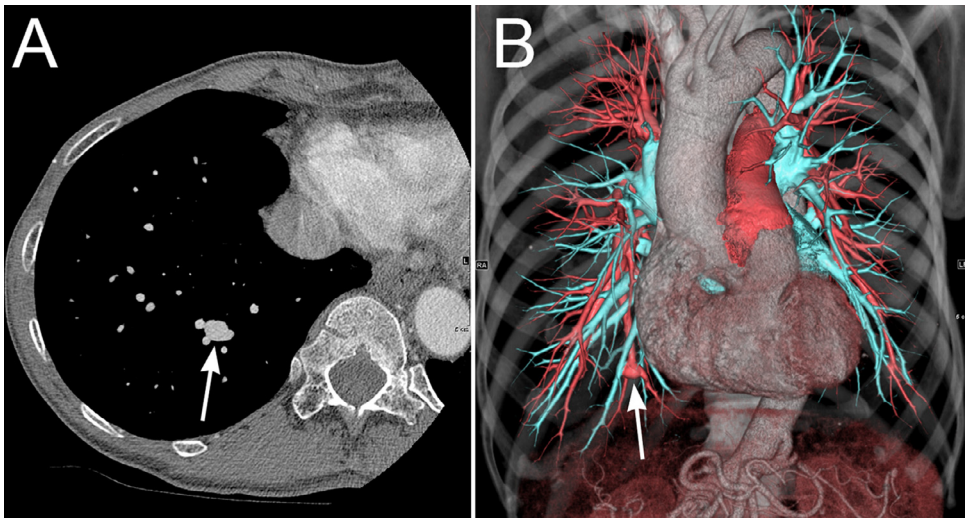


**Fig. 1 – Contrast-enhanced computed tomography (CT) before transcatheter embolization. (A) Contrast-enhanced CT showed a 0.9 × 0.7 cm aneurysmal dilatation in the right pulmonary lower lobe (arrow). (B) Volume rendered image in 15-degree left anterior oblique view showing aneurysm dilatation connecting to the A10b/c bifurcation (arrow).**

A 0.7 × 0.6 cm right lower lobe pulmonary nodule was incidentally detected on follow-up computed tomography (CT), which showed mild enlargement about 3 years of follow-up. Contrast-enhanced CT showed a 0.9 × 0.7 cm aneurysmal dilation adjacent to the right pulmonary artery A10b/c bifurcation without a distinct draining vein (Fig. 1). Therefore, PAA was suspected rather than a pulmonary AVM. Although the patient was asymptomatic, due to the increase in size, transcatheter embolization was planned to prevent rupture.

A 4.5-F guiding sheath (Parent Plus 45; Medikit, Tokyo, Japan) was inserted from the left femoral vein into the right main pulmonary artery. A 5.2-F balloon catheter (Selecon MP Catheter II; Terumo, Tokyo, Japan) was advanced into A10b+c. A10b+c angiography under balloon dilation showed aneurysmal dilation at the A10b/c bifurcation and no early venous return, leading to the PAA diagnosis (Fig. 2A). A10b and A10c distal to the PAA, the PAA, and A10b+c proximal to the PAA were embolized with hydrogel coils (AZUR CX18; Terumo, Tokyo, Japan) and pushable coils (C-STOPPER 0.016″; Piolax Medical Devices, Kanagawa, Japan) using a 2.5-F microcatheter (Renegade; Boston Scientific, MA) under flow control by balloon dilation (Fig. 2B). After embolization, right inferior pulmonary artery angiography revealed the disappearance of the PAA and absence of non-targeted embolization (Fig. 2C).

Contrast-enhanced magnetic resonance imaging performed 3 months after embolization showed no recanalization of the PAA (Fig. 3). The patient has had no recurrence for 1 year after embolization.

## Discussion

PAAs are rare, with an estimated incidence of approximately 1 in 14,000 cases, based on autopsy reports [1]. Depending on the site, PAAs are classified as either central or peripheral. Central PAAs arise from large pulmonary arteries, such as the main pulmonary artery, whereas peripheral PAAs are in the distal branches [2]. PAAs are classified into congenital and acquired mechanisms. Acquired PAAs are further classified into secondary PAAs with causative diseases, such as pulmonary hypertension, vasculitis, infection, or trauma, and idiopathic aneurysms without such causes. Only approximately 40 cases of idiopathic PAAs have been reported, and approximately 1/3 of them are idiopathic peripheral PAAs, indicating that idiopathic peripheral PAAs are extremely rare [2].

In the present case, the PAA was located in the distal pulmonary artery, and there was no background disease known to cause secondary PAAs, which was classified by definition as idiopathic peripheral PAA. Hereditary hemorrhagic telangiectasia was suspected in this case because of the history of pulmonary AVMs and gastrointestinal telangiectasia. Still, genetic testing was not performed because only one category in the Curaçao diagnostic criteria was met [5]. In a retrospective study, PAAs were found in 0.7% of patients with hereditary hemorrhagic telangiectasia, a higher frequency than that in the general population [6]. The same study also suggested that AVMs unrelated to aneurysms and ACVRL1 mutations may be associated with aneurysm development. Although our case was classified as an idiopathic PAA, the history of pulmonary AVMs may have contributed to the development of the aneurysm.

Gupta et al. proposed rapid enlargement, hemoptysis, and compression of adjacent structures as criteria for treating PAAs [2]. However, there is no consensus on the treatment of PAAs. The frequency of rupture of idiopathic peripheral PAAs is unknown, but high mortality rates have been reported upon rupture of solitary peripheral PAAs [7]. Therefore, a treatment aimed at preventing rupture should be considered. In our case, embolization was performed because the PAA increased in size, and it was estimated that embolization would have little effect on pulmonary blood flow, considering the location of the aneurysm. Transcatheter embolization is a less invasive treatment than surgery for idiopathic peripheral PAAs. There

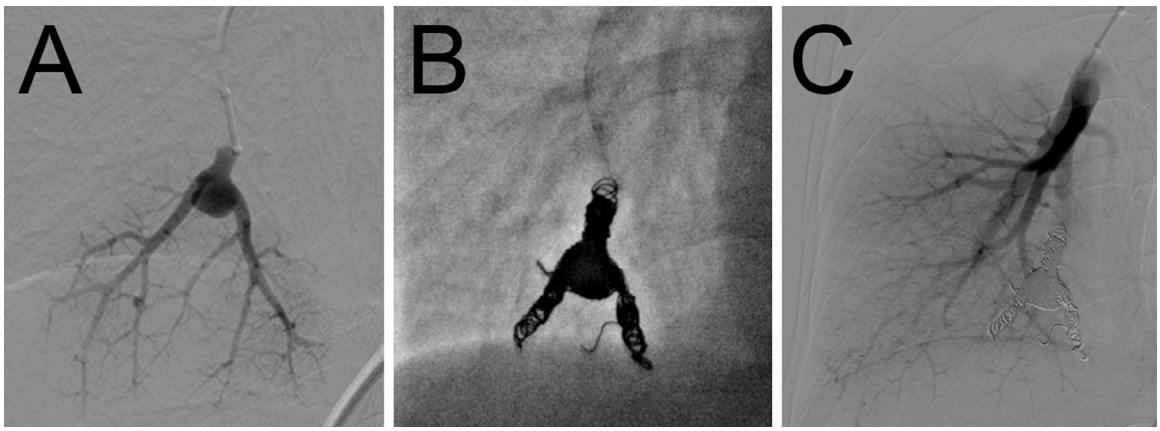


**Fig. 2 – Angiographic images obtained during the procedure. (A) Digital subtraction angiography under balloon occlusion from the right A10b+c showing a pulmonary artery aneurysm at the A10b/c bifurcation. (B) Fluoroscopy showing coils in A10b, A10c, aneurysm, and A10b+c after embolization. (C) Right inferior pulmonary artery angiography after embolization showing disappearance of the aneurysm and absence of nontargeted embolization.**

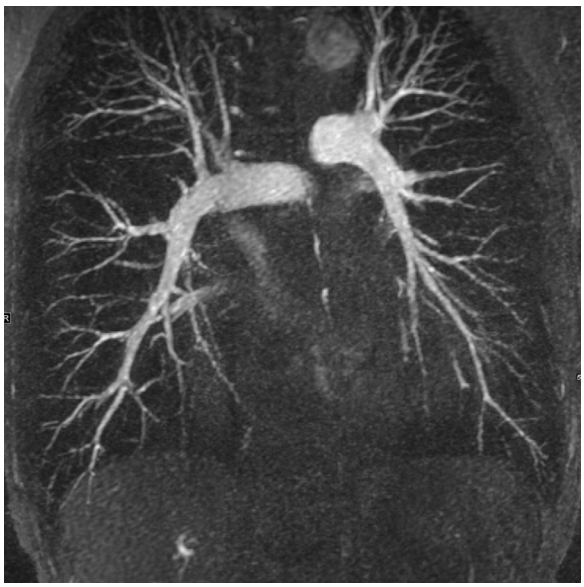

**Fig. 3 – Contrast-enhanced magnetic resonance imaging three months after embolization. Recanalization of the aneurysm was not observed.**

are only 2 reports of transcatheter embolization of unruptured idiopathic peripheral PAAs: one report of embolization of the aneurysm alone with coils, and the other report of embolization with vascular plugs distal to the aneurysm and coils within and proximal to the aneurysm, both with good clinical outcomes [3,4]. In the present case, the area distal, within, and proximal to the PAA was embolized with coils, and complete embolization was achieved without complications. Although there is no consensus on the indications and treatment for unruptured idiopathic peripheral PAAs, transcatheter embolization may be a favorable treatment option. Further accumulation of cases is required.

## Patient consent

Written informed consent for publication was obtained from the patient.

## 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;34:471–99. doi:10.1016/0002-8703(47)90527-9.
[2] Gupta M, Agrawal A, Iakovou A, Cohen S, Shah R, Talwar A. Pulmonary artery aneurysm: a review. Pulm Circ 2020;10:2045894020908780. doi:10.1177/2045894020908780.
[3] Rastogi N, Kabutey N-K, Kim D, Norbash A. Percutaneous management of segmental pulmonary artery aneurysm in a patient without pulmonary artery hypertension. Vasc Endovasc Surg 2011;45:283–7. doi:10.1177/1538574411399160.
[4] Tamura T, Yabuki T, Kawajiri T, Nishi T, Kudo K, Kuyama S. Transcatheter embolization for idiopathic peripheral pulmonary arterial aneurysm: a case report. Clin Case Rep 2019;7:1021–4. doi:10.1002/ccr3.2154.
[5] Shovlin CL, Guttmacher AE, Buscarini E, Faughnan ME, Hyland RH, Westermann CJ, et al. Diagnostic criteria for hereditary hemorrhagic telangiectasia (Rendu-Osler-Weber syndrome). Am J Med Genet 2000;91:66–7. doi:10.1002/(sici)1096-8628(20000306)91:1<66::aid-ajmg12>3.0.co;2-p.
[6] Ring NY, Latif MA, Hafezi-Nejad N, Holly BP, Weiss CR. Prevalence of and factors associated with arterial aneurysms in patients with hereditary hemorrhagic telangiectasia: 17-year retrospective series of 418 patients. J Vasc Interv Radiol 2021;32:1661–9. doi:10.1016/j.jvir.2021.08.018.
[7] 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. doi:10.1016/s0003-4975(10)65686-x.