Cardiology in the Young

cambridge.org/cty

Repair of iatrogenic left pulmonary artery to left atrial appendage fistula using a covered CP stent: a case report

Hisako Kakimoto, Takanari Fujii and Hideshi Tomita

Pediatric Heart Disease and Adult Congenital Heart Disease Center, Showa University Hospital, Shinagawa-ku, Tokyo, Japan

Brief Report

Cite this article: Kakimoto H, Fujii T, and Tomita H (2024) Repair of iatrogenic left pulmonary artery to left atrial appendage fistula using a covered CP stent: a case report. *Cardiology in the Young* **34**: 922–923. doi: 10.1017/S1047951123004481

Received: 3 July 2023
Revised: 16 November 2023
Accepted: 22 December 2023
First published online: 22 January 2024

Keywords:
complication; balloon angioplasty; three-dimensional printing

Corresponding author:
Hisako Kakimoto; Email: kgchsk@gmail.com

Abstract

We report a case of iatrogenic left pulmonary artery–left atrial appendage fistula following percutaneous transluminal angioplasty for residual pulmonary artery stenosis in an 11-year-old boy. This rare complication could have been predicted by understanding the anatomical relationship of these structures. In this study, simulation using three-dimensional printing greatly contributed to successful stent placement.

Case report

An 11-year-old boy was admitted for treatment of recurrent pulmonary artery stenosis complicated by pulmonary atresia with a ventricular septal defect. He underwent ventricular septal defect patch closure, trans-annular monocusp-valve outflow patch procedure, and atrial septal defect closure at 3 years of age.

Pulmonary artery stenosis gradually deteriorated, while cardiac catheterization at 10 years of age revealed a right ventricular to left ventricular pressure ratio of 1.1. Three-dimensional CT revealed stenosis of the main pulmonary artery bifurcation and long-segment right pulmonary artery stenosis was associated with compression by an enlarged ascending aorta. Furthermore, the right pulmonary artery was closely aligned with the left coronary artery. Consequently, bilateral PA augmentation using the expanded polytetrafluoroethylene patch on the anterior wall, elongation of the right ventricular outflow tract with the expanded polytetrafluoroethylene graft as a strip for the posterior wall and as a patch for the anterior wall, and extension of the ascending aorta with an interposition graft were performed. Angiography after eight months demonstrated residual stenosis of the proximal left pulmonary artery with a pressure difference of 22 mmHg.[1] The minimum lumen diameter was 5.9 mm, while the distal reference vessel diameter was 18.6 mm. Following angioplasty using two ultra-high-pressure balloons (Conquest® →Bard® 12 mm/4 cm, 20 atmospheres; Becton Dickinson), the "waist" of the balloon completely disappeared. Subsequent left pulmonary artery angiography demonstrated partial dilation of the left pulmonary artery to 10.2 mm; however, a tear was seen in the middle of the stenotic area at the floor of the left pulmonary artery which was communicated with the left atrial appendage (Fig 1a).

Although the patient remained haemodynamically stable, his oxygen saturation decreased from 100 to 90% soon after the procedure. Protamine was administered to reverse heparin, while a Z-MED balloon (15 mm/4 cm; NuMED) was dilated to seal the communication for 30 min. There was no pericardial effusion and bronchial bleeding. Despite oxygen desaturation to approximately 80% during exercise, the patient had no symptoms, such as dyspnea, and was discharged one week later.

As oxygen desaturation to 90% persisted, we planned covered CP stent implantation for left pulmonary artery stenosis four months later after approval by the ethics committee of the Showa University (IRB No. 1907-K-^00026). Before the procedure, CP stent implantation was simulated using three-dimensional printing (Fig 2a and b). Based on this simulation, a 28 mm covered CP stent was mounted on a 14 mm/3.5 cm BIB® (Balloon in Balloon) catheter and was implanted in the proximal left pulmonary artery. Transcutaneous oxygen saturation increased from 91 to 100% just after the procedure. Angiography demonstrated complete closure of the left pulmonary artery–left atrial appendage communication with preserved flow to the left upper lobe and resolution of proximal left pulmonary stenosis (Fig 2c).

Discussion

The anatomical relationship between the left atrial appendage and the left pulmonary artery is thought to be a major factor in complication of fistula. When using the WATCHMAN left atrial appendage closure system, a previous study showed that pulmonary artery perforation was

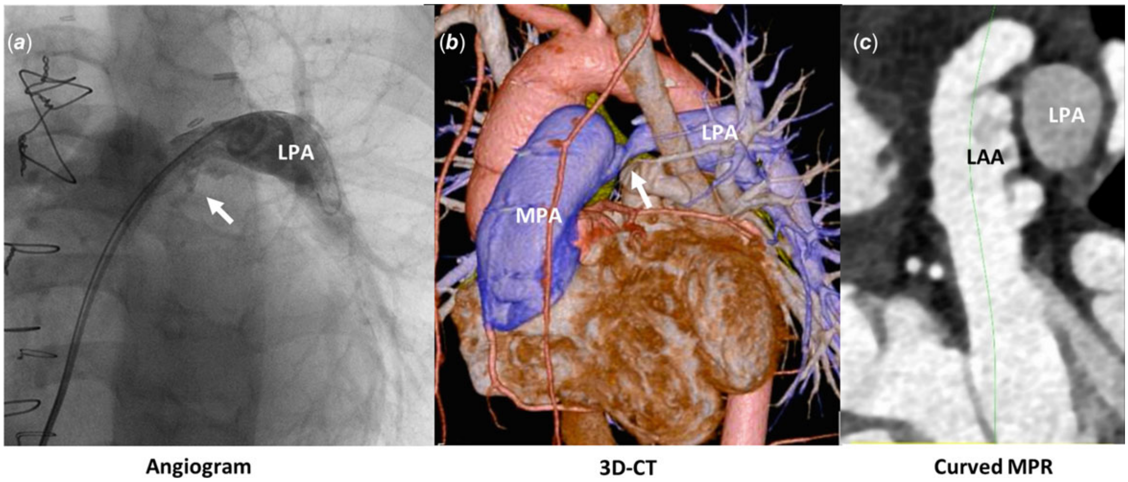

Figure 1. (***a***) A tear can be seen in the middle of the stenotic area at the underside of the left pulmonary artery with minimal contrast extravasation. (***b***) The distance between the LAA and the narrowest part of the LPA was small. (***c***) The proximal LAA was in contact with the LPA, and the connective tissue thickness between them was <1 mm.3D-CT: three-dimensional CT; LAA = left atrial appendage; LPA = left pulmonary artery; MPR = multiplanar reconstruction.

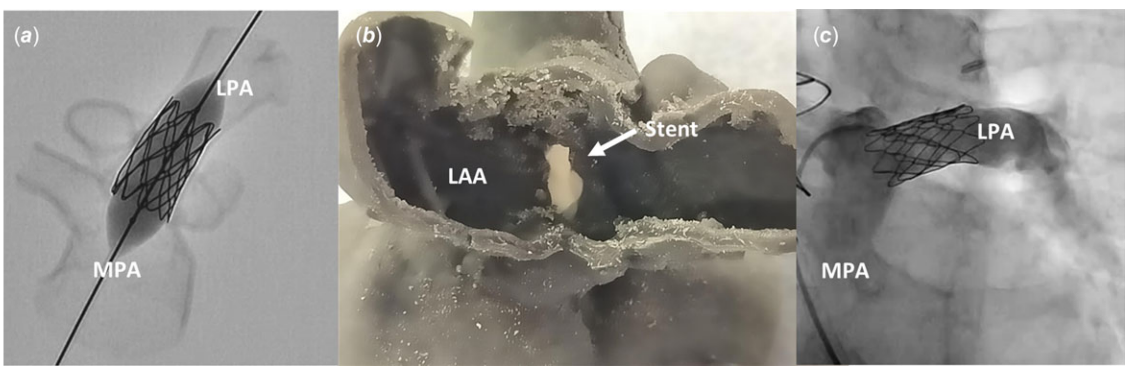

Figure 2. (***a***) The stent was placed securely in the three-dimensionally printed model. (***b***) The communication between the pulmonary artery and the LAA was successfully sealed using the polytetrafluoroethylene graft in the model. (***c***) Angiography demonstrating complete closure of the left pulmonary artery–left atrial appendage fistula with preserved flow to the left upper lobe and resolution of proximal left pulmonary artery stenosis.

related to the anatomical proximity between the left atrial appendage and the pulmonary artery.[2] In this case, the distance between the left atrial appendage and the left pulmonary artery was small, with only a small amount of connective tissue between them (Fig 1b and c). In our case, the most stenotic part of the left pulmonary artery was in contact with the left atrial appendage, tearing occurred at that site, and the thin connective tissue could not separate these structures. And the fact that the target vessels were composed of polytetrafluoroethylene patches and native tissue, materials that differ in compliance, may have contributed to the wall tears.

Since the fistula developed in the narrowest portion of the left pulmonary artery in this case, we chose off-label use of the covered stent, not only to avoid device protrusion, which may exacerbate pulmonary artery stenosis, but also to avoid increasing the risk of left atrial appendage thrombus formation on the device.

The covered CP stent was useful, not only to close the fistula but also to resolve proximal left pulmonary artery stenosis. The stent was implanted using 14 mm balloon, which, we suppose, is almost similar to the adult left pulmonary artery. Even in case size mismatch develops following somatic growth, there is a report on post-dilation of covered CP stent.[3] Furthermore, CT angiography and three-dimensional printing helped to simulate the device landing zone and contact between the left atrial appendage and the pulmonary artery.

Financial support. The authors received no specific funding for this study.

Competing interests. The authors have no conflicts of interest to declare.

Ethical standard. The treatment was approved by the ethics committee of the Showa University.

References

1. Tarui S, Miyahara Y, Oyama N, Fujii T, Ishino K, Tomita H. Double-outlet extension for narrowed retroaortic space. Asian Cardiovasc Thorac Ann 2020; 28: 123–125.
2. Lu C, Zeng J, Meng Q, Zeng Z. Pulmonary artery perforation caused by a left atrial appendage closure device. Catheter Cardiovasc Interv 2020; 96: E744–E746.
3. Ewert P, Schubert S, Peters B, Abdul-Khaliq H, Nagdyman N, Lange PE. The CP stent – short, long, covered – for the treatment of aortic coarctation, stenosis of pulmonary arteries and caval veins, and Fontan anastomosis in children and adults: an evaluation of 60 stents in 53 patients. Heart 2005; 91: 948–953.