


Pregnancy and delivery after percutaneous embolization with a combination of microvascular plugs and hydrogel-coated coils for unilateral diffuse pulmonary arteriovenous malformations: a case report

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Background

Pulmonary arteriovenous malformations (PAVMs) are abnormal communications between the pulmonary arteries and veins; right-to-left shunts can cause hypoxaemia, emboli to systemic circulation, and brain abscesses. Cyanosis during pregnancy may increase the probability of premature birth or spontaneous abortion and may increase maternal cardiac complications.

Case summary

We describe a case of a 24-year-old woman with diffuse multiple PAVMs localized to the left inferior lobe and chronic cyanosis. She had increased exertional fatigue and chronic headaches and was New York Heart Association class II, although her rest sitting peripheral oxygen saturation (SpO₂) had remained unchanged at 83% over the past 20 years. She underwent percutaneous embolization with microvascular plugs and hydrogel-coated coils. A microvascular plug was placed as an anchor near the venous sac, followed by hydrogel-coated coil embolization of the proximal pulmonary artery. A total of six sessions of catheter intervention were performed. The embolization was successful, her hypoxaemia was relieved, and she was able to conceive and deliver. Three years have passed since the last session, and SpO₂ 97% has been maintained.

Discussion

In the treatment of complex PAVMs, the combination of microvascular plugs and hydrogel-coated coils resulted in shorter procedure time, lower risk of migration of the embolus to the pulmonary veins, and less recanalization and revascularization. Percutaneous embolization of PAVMs resulted in safe delivery for the mother and child.

Keywords

Pulmonary arteriovenous malformation • Arteriovenous fistula of pulmonary vessels • Percutaneous embolization • Microvascular plug • Hydrogel-coated coil • Case report

ESC curriculum

7.4 Percutaneous cardiovascular post-procedure • 9.8 Pregnancy with cardiac symptoms or disease • 9.7 Adult congenital heart disease

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Learning points

- Cyanosis during pregnancy may increase the probability of premature birth or spontaneous abortion and may increase maternal cardiac complications.
- The combination of microvascular plugs and hydrogel-coated coils can safely occlude multiple feeding arteries in complex pulmonary arteriovenous malformations.

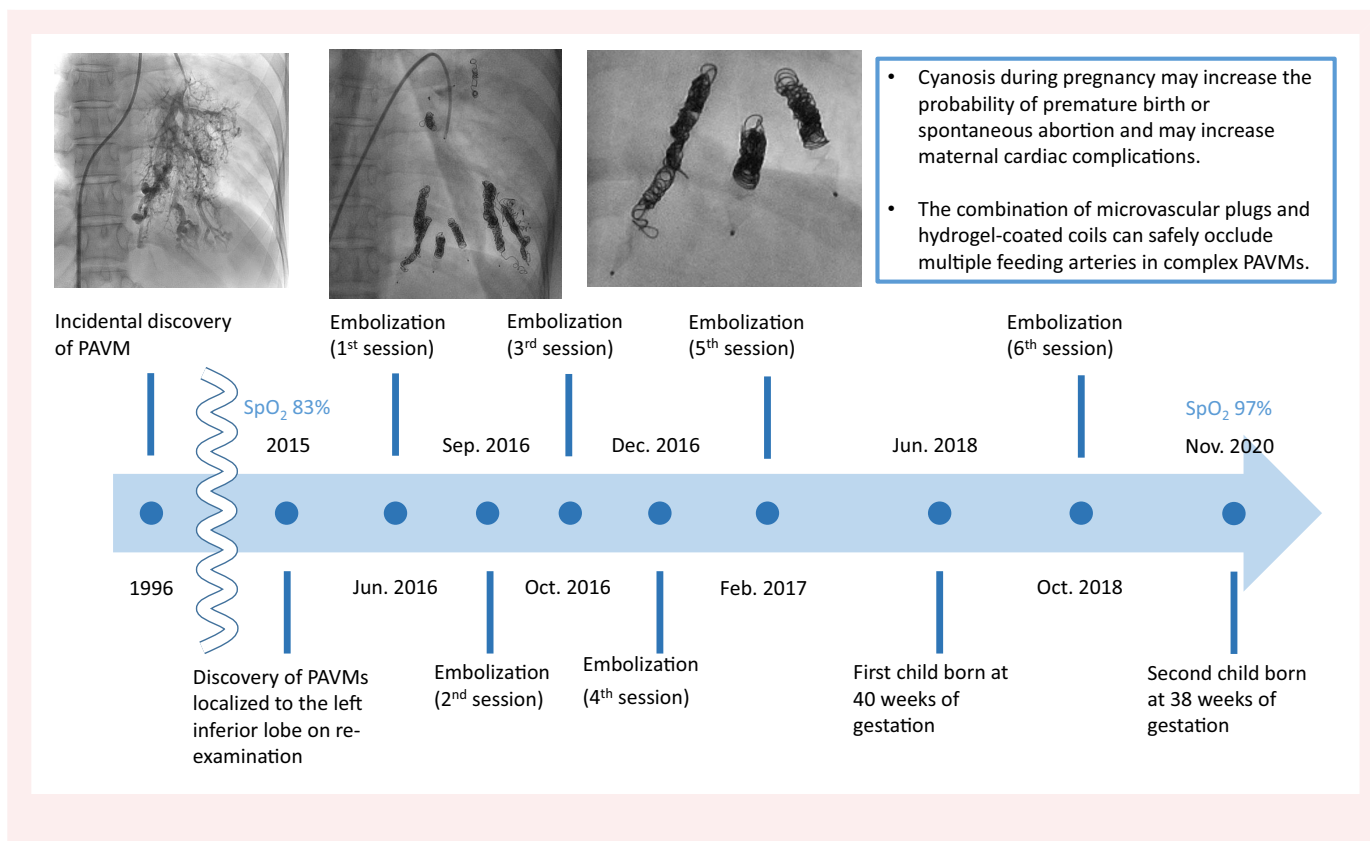
Introduction

Pulmonary arteriovenous malformations (PAVMs) are abnormal communications between the pulmonary arteries and veins; right-to-left shunts can cause hypoxaemia, emboli to systemic circulation, and brain abscesses. Pulmonary arteriovenous malformations with feeding artery diameters ≥ 2 –3 mm, increased measurable size of PAVMs, paradoxical embolism, or symptomatic hypoxaemia are indications for embolization.^{1,2} Moreover, cyanosis during pregnancy may increase the probability of premature birth or spontaneous abortion and frequency of maternal cardiac complications; thus, PAVMs should be treated before pregnancy whenever possible.^{3,4} We describe a 24-year-old woman with chronic cyanosis and diffuse multiple PAVMs localized to the left inferior lobe, who conceived and carried pregnancy to term after percutaneous embolization with microvascular plugs and hydrogel-coated coils.

and chronic headaches and had New York Heart Association class II, although her resting sitting peripheral oxygen saturation (SpO₂) remained unchanged at 83% over the past 20 years. She had a history of allergic rhinitis, for which she received antileucotriene agents. She also occasionally used nonsteroidal anti-inflammatory drugs for chronic headaches. We reconsidered the treatment indications because of the patient desired to conceive.

Physical examination revealed cyanosis and digital clubbing. Auscultation revealed normal cardiac and pulmonary sounds. Electrocardiography depicted normal sinus rhythm, whereas echocardiography showed normal biventricular function without any structural abnormalities. Chest radiography and computed tomography revealed PAVMs in the left inferior lobe (Figure 1). Left pulmonary angiography identified diffuse and multiple segmental feeding artery branches measuring 2–4 mm in diameter (Figure 2). All segmental arteries in the left inferior lobe were involved. No obvious lesions were observed in the right or left superior lobe. In the supine position, the arterial blood oxygen saturation (SaO₂) was 91.8% (reference: 94–99%), and partial pressure of arterial oxygen (PaO₂) was 54.3 mmHg (reference: 80–100 mmHg). The mean pulmonary arterial pressure was 11 mmHg. The SpO₂ increased from 89 to 95% after the occlusion test

Summary figure



Case presentation

The patient was a 24-year-old woman with PAVMs diagnosed at 1 year of age because of hypoxaemia. She experienced increased exertional fatigue

for the descending branch of the left pulmonary artery. The pulmonary arterial pressure remained unchanged. The haemoglobin and haematocrit levels were 17.7 g/dL (reference: 11.6–14.8 g/dL) and 53.2% (reference: 35.1–44.4%), respectively.

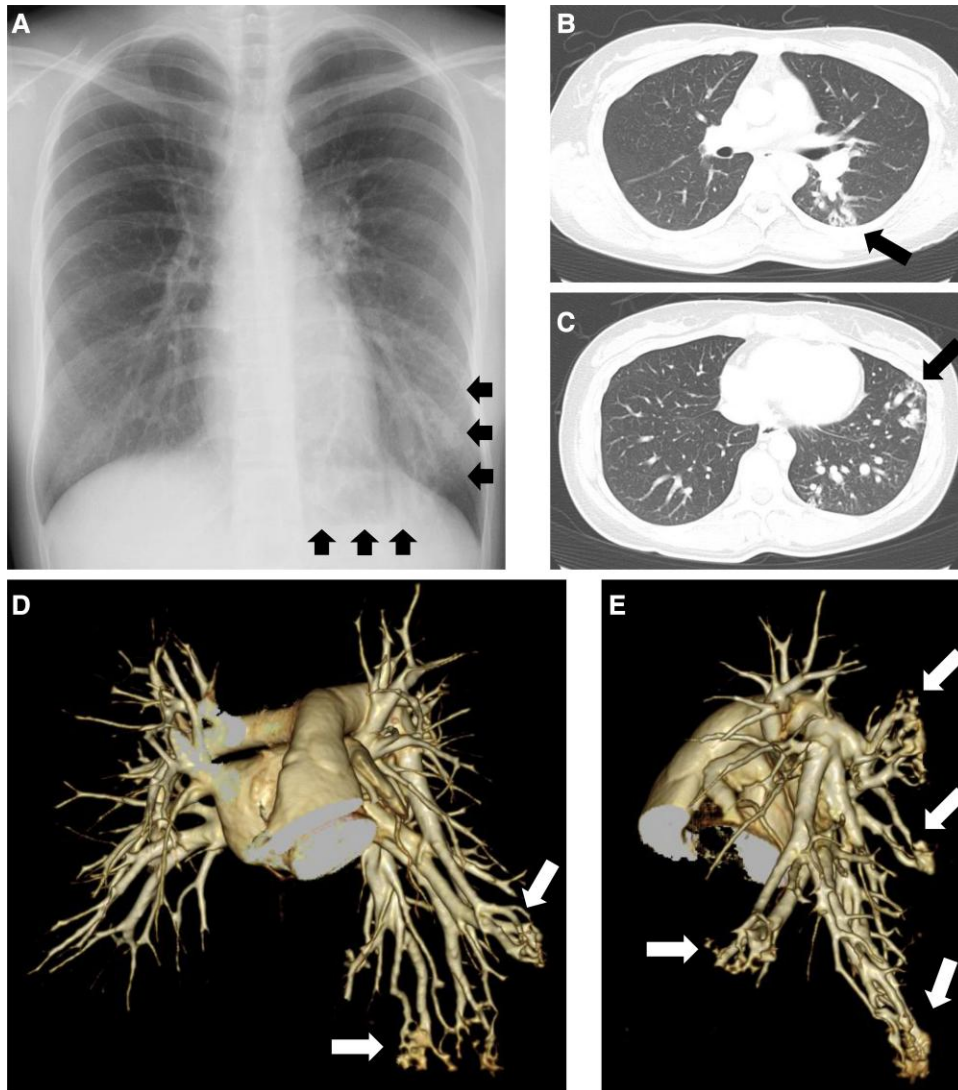


Figure 1 Presentation of pulmonary arteriovenous malformations (arrows). A chest radiograph (A), axial computed tomography images (B, C), and 3D reconstruction images (D, E) showing diffuse multiple pulmonary arteriovenous malformations localized to the left inferior lobe.

Subsequently, five sessions of percutaneous embolization were performed over 8 months (Figure 3) via the femoral vein under local anaesthesia. Heparin 2500 IU was administered intravenously for prophylactic anticoagulation immediately after sheath insertion, followed by an additional dose of 1500 IU hourly. A 4–6-F angiographic catheter with a 0.035-inch-diameter guidewire was advanced into the feeding artery as close to the venous sac as possible. An Amplatzer Vascular Plug 4 (AVP 4; AGA Medical, Plymouth, MN, USA) measuring 130–150% of the vessel diameter size was placed. An Amplatzer Vascular Plug II (AVP II; AGA Medical) was used if the guiding catheter could be inserted into the target site. Subsequently, a microcatheter with a 0.014-inch-diameter guidewire was advanced to the AVP and completely embolized with a hydrogel-coated coil (AZUR Peripheral HydroCoil Embolization System; Terumo Medical Corporation, Tokyo, Japan). Device diameters of 4–7 and 6 mm were selected for AVP 4 and AVP II, respectively. A loop diameter of 2–6 mm and coil length of 4–20 cm were used for hydrogel-coated coils. Overall, 12 AVPs and 29 AZURs were used. No intra-procedural adverse events occurred. However, chest pain and C-reactive protein elevation occurred 6

weeks after the first session. One week after the second session, chest and back pain, C-reactive protein elevation, fever, and pleural effusion were observed. Mild chest pain occurred 2 days after the third session. All symptoms resolved with antipyretic medications. After the fifth session, oxygenation improved to an SaO₂ of 97.7% and a PaO₂ of 79.4 mmHg.

Fifteen months after the fifth session, she delivered her first child. Considering the residual right-to-left shunt, thromboprophylaxis with subcutaneous heparin calcium injection was initiated at 8 weeks of gestation. The exercise test showed SpO₂ of <90%; therefore, oxygen therapy was continued. Antibiotic prophylaxis (cefazolin 1000 mg) was administered during delivery. A 3195 g male infant was born at 40 weeks of gestation without complications. Catheterization was planned for evaluation 1 year after the fifth session but was postponed to 1 year after birth due to pregnancy. Preprocedural SaO₂ and PaO₂ were 96.5% and 86.2 mmHg, respectively, which had not decreased since the fifth session 2 years ago. The mean pulmonary arterial pressure was 10 mmHg. Pulmonary angiography revealed no evidence of reperfusion or recanalization. The remaining small PAVMs were embolized with AZUR. She conceived again and delivered her second child

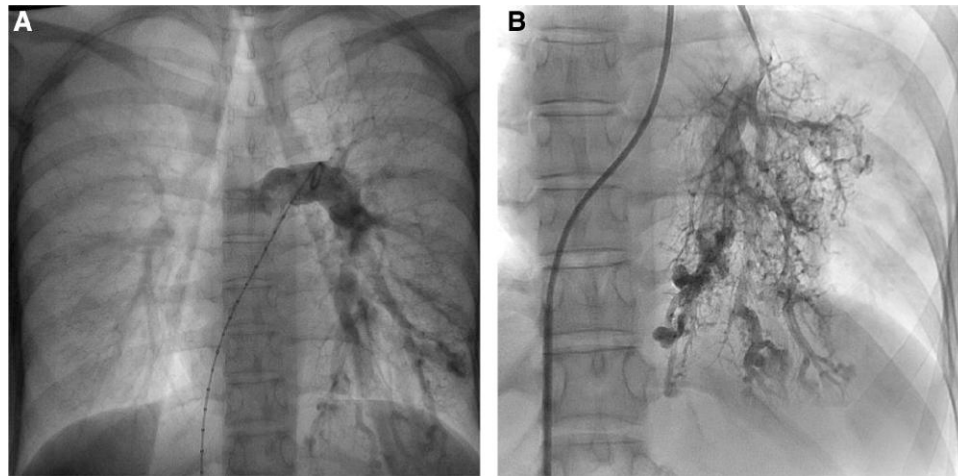


Figure 2 Left pulmonary arteriogram (A) and selective segmental arteriogram of the left inferior lobe (B) in the anteroposterior views showing diffuse multiple pulmonary arteriovenous malformations. Multiple segmental feeding artery branches are identified. All segmental arteries of the left inferior lobe are involved.

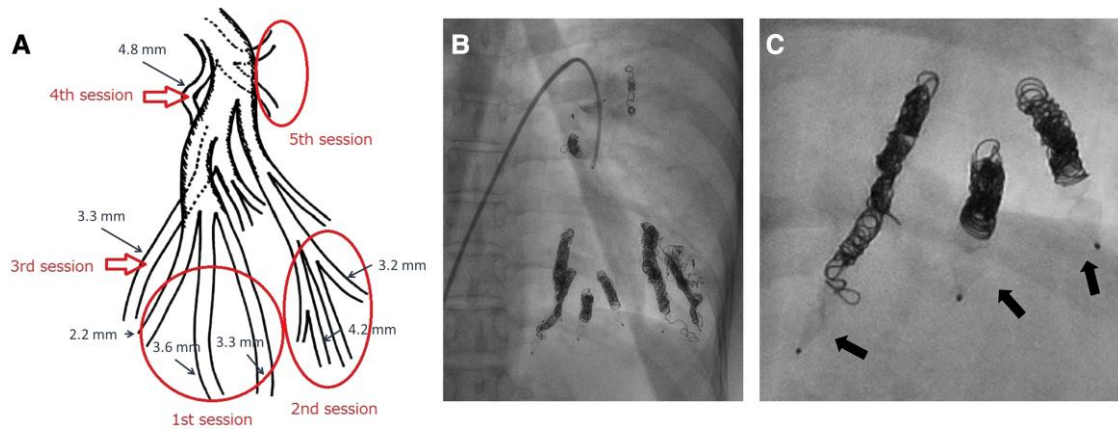


Figure 3 Percutaneous embolization with microvascular plugs and hydrogel-coated coils. An illustration (A) showing the feeding arteries of pulmonary arteriovenous malformations. The first session targeted the anteromedial basal segment (S7 + S8), the second session the lateral basal segment (S9), the third session the posterior basal segment (S10), and the fourth and fifth sessions the superior segment (S6). Fluoroscopic images (B, C) showing the postembolization state of the left lung. Pulmonary arteriovenous malformations were successfully occluded by microvascular plug embolization, followed by hydrogel-coated coil embolization. A magnified view of the left lung basal area (C) showing hydrogel-coated coils implanted proximal to the microvascular plugs (arrows).

(birth weight 2895 g) at 38 weeks of gestation, without complications. After 2 years, resting sitting SpO₂ was sustained at 97%.

Discussion

Cyanosis during pregnancy may increase the probability of premature birth or spontaneous abortion and frequency of maternal cardiac complications.^{3,4} The 2018 European Society of Cardiology guidelines state that maternal complications are more likely to occur in pregnant patients with cyanosis, and when oxygen saturation is <85%, foetal growth restriction, prematurity, and foetal death are common; thus, pregnancy should be discouraged.⁵ Our patient, who desired to bear a child but

had chronic hypoxaemia due to PAVMs with feeding arteries of ≥ 2 –3 mm, met treatment criteria and underwent successful percutaneous embolization of these malformations.

Percutaneous embolization is the first-line treatment for PAVMs. Although coils are standard, microvascular plugs were also used in the present case. Our five-point strategy stressed on less invasiveness, lower risk embolus migration to the pulmonary veins, high embolic effect, shorter procedural time, and no reperfusion or recanalization. We chose the combination of microvascular plugs and hydrogel-coated coils for two major reasons. First, microvascular plugs reduce the risk of inadvertent embolus migration to the pulmonary veins and shorten the procedure. Second, hydrogel-coated coils have a strong embolic effect and prevent

recanalization and revascularization. Microvascular plugs have a larger diameter than coils and can be implanted as anchors in lesions with rapid blood flow. The advantage of AVP 4 is that it can be implanted using an angiographic catheter and can easily reach the sac. In contrast, hydrogel-coated coils combine a platinum coil with an expandable hydrogel polymer that swells after implantation and occupies the space between the coils, yielding a greater embolic effect than conventional coils. The main mechanism of action of conventional coils is embolization by thrombus formation; once embolized, the thrombus may dissolve and recanalize. In contrast, hydrogel-coated coils are embolized by swelling of hydrogel, reducing the likelihood of recanalization. Shimohira *et al.*⁶ used hydrogel-coated coils in 57 PAVM cases and reported a technical success rate of 98% without recanalization at a mean follow-up period of 19 months. Similarly, a prospective study of 21 PAVM cases managed by venous sac embolization with hydrogel-coated coils reported technical success of 95% without recanalization or reperfusion.⁷ Trerotola *et al.*⁸ reported the embolic effect of microvascular plugs combined with coils. Type 1 AVPs were combined with fibred platinum coils to embolize 39 feeding arteries. An AVP was carefully placed into the feeding artery as close as possible to the venous sac to prevent the risk of recanalization. The procedure was successful for all PAVMs, without recanalization.

In conclusion, percutaneous embolization of PAVMs resulted in safe delivery for both mother and child. Microvascular plugs combined with hydrogel-coated coils produced long-term embolic effects in multiple PAVMs.

Lead author biography



Kayo Ogino is a general paediatrician and paediatric cardiologist at Kurashiki Central Hospital, Kurashiki, Japan. She graduated from Hokkaido University with an MD in 2005 and subsequently completed her residency at Yamanashi Prefectural Central Hospital and Kanagawa Children's Medical Center.

Consent: The authors confirm that written informed consent for the publication of this case report, including images and associated text, was obtained from the patient in line with the Committee on Publication Ethics guidelines.

Conflict of interest: None declared.

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Data availability

The data underlying this article are available within the article.

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