

Percutaneous embolization of a caroticoazygous fistula with the Amplatzer Vascular Plug 1

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ABSTRACT

We present the case of a five-year-old boy with a carotico-azygous fistula, which is an extremely rare congenital vascular malformation. The patient also had patent ductus arteriosus, a common cause of continuous murmur. The murmur continued despite successful coil embolization of the ductus. Repeated catheter-angiography revealed a large carotico-azygous fistula. The fistula was successfully embolized using the Amplatzer® Vascular Plug 1. Transcatheter occlusion of caroticoazygous fistula was performed easily, safely, and efficiently with a vascular plug.

Key words: • patent ductus arteriosus • therapeutic embolization • arteriovenous fistula

Carotico-azygous fistula is an extremely rare form of arteriovenous fistula, resulting from an abnormal communication between the carotid artery and azygous vein (1–4). As a result of continuous murmur, which is a typical physical finding of these vascular malformations, a caroticoazygous fistula could be confused with patent ductus arteriosus. Transcatheter coil embolization of these fistulas has been reported previously (5).

Here, we describe a five-year-old boy with caroticoazygous fistula, who also had patent ductus arteriosus, the main cause of misdiagnosis, and successful transcatheter embolization with the Amplatzer® Vascular Plug 1 (AVP 1).

Case report

A five-year-old boy was referred to our clinic with cardiac murmur. He had no history of any complaint. During physical examination, a continuous murmur was heard in the left chest area. Electrocardiogram and telecardiography were normal. Color and continuous wave Doppler echocardiography showed patent ductus arteriosus with dilated left atrium and left ventricle. Left atrium-to-aortic root diameter ratio was 1.7. He underwent cardiac catheterization and descending aorta angiography, focusing only on possible patent ductus arteriosus arising from the aorta. Patent ductus arteriosus was detected by angiography. The narrowest diameter of patent ductus arteriosus was measured as 2.3 mm. A detachable coil, 5 mm in diameter and having five loops (William Cook Europe, Bjaeverskov, Denmark), was used for occlusion of patent ductus arteriosus. No residual shunt was observed in control angiography (Fig. 1). During examination on the next day, a continuous murmur was still heard in the same area, but no residual shunt was detected by echocardiography. A chest X-ray confirmed that the coil had not migrated. We then highly suspected an arteriovenous fistula but it was not revealed by echocardiography. Therefore, other reasons for the continuous murmur were investigated. Initially, cranial computed tomography with contrast and abdominal Doppler ultrasound were performed to reveal a fistula in those areas, but the results were normal. We then decided to perform further cardiac catheterization. During the second cardiac catheterization procedure, aortic angiography detected a fistula tract between the carotid artery and azygous vein (Fig. 2).

Aortic and subsequent selective angiography in anteroposterior and lateral views showed multiple branches of the vertebral arteries linking the fistula tract. It had a tortuous course, and the narrowest part measured 3.4 mm (Fig. 2). The calculated pulmonary to systemic blood flow ratio was 1.7.

Lateral selective descending angiography was performed to delineate the fistula tract (Fig. 2a). The fistula tract had several connections to

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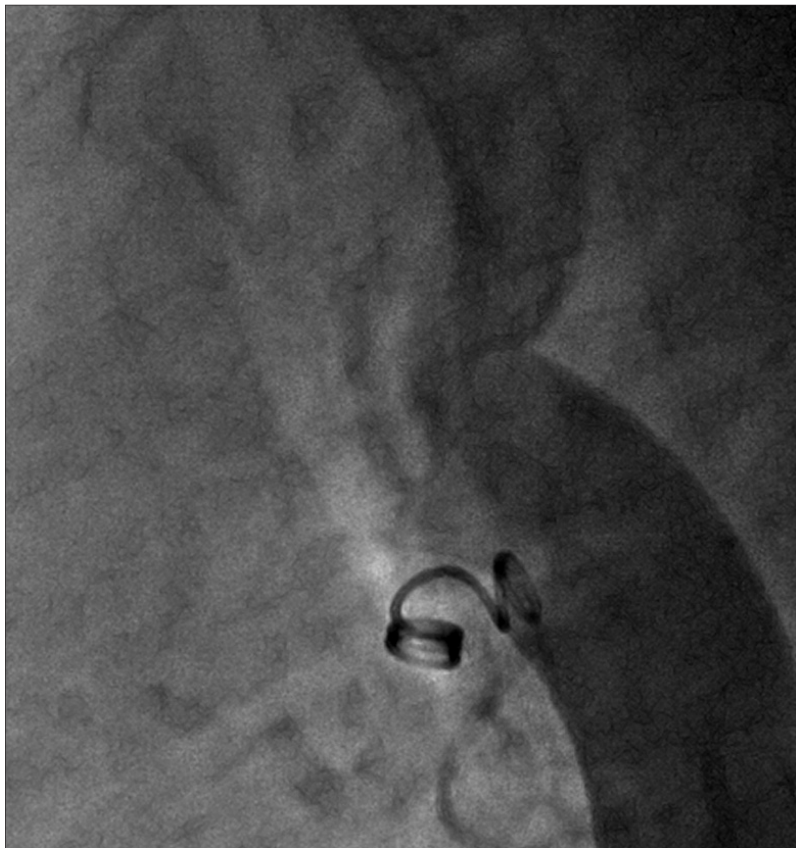


Figure 1. Lateral selective descending aorta angiogram after coil occlusion of patent ductus arteriosus. No residual shunt across the ductus was observed.

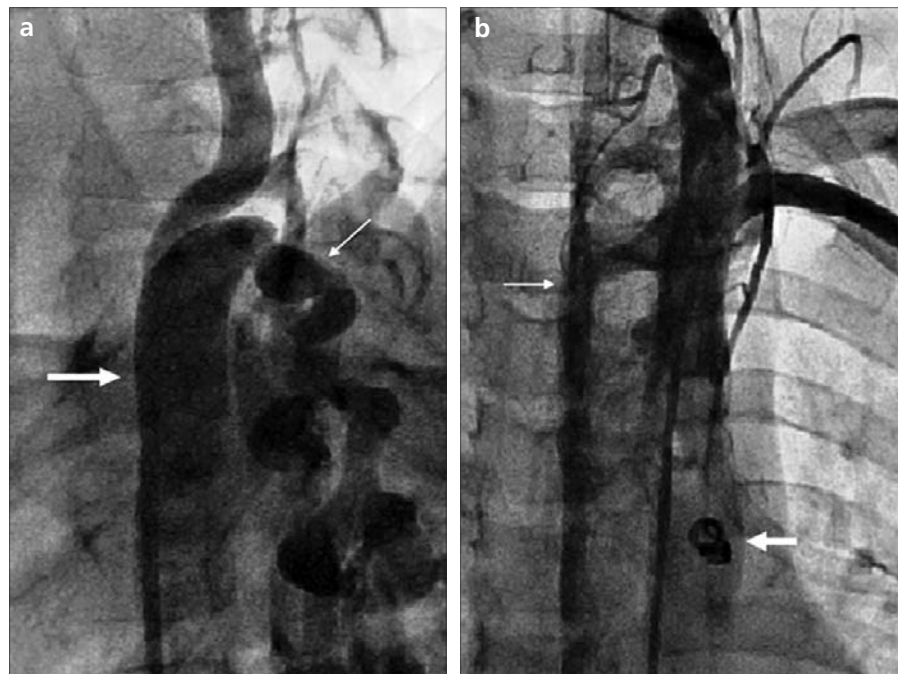


Figure 2. a, b. Lateral selective descending aorta angiogram (**a**; *thin arrow*, fistula tract; *thick arrow*, descending aorta) and ascending aorta angiogram (**b**; *thin arrow*, hemizygous vein; *thick arrow*, patent ductus arteriosus coil).

paravertebral veins. The orifice of the fistula was occluded with a Berman balloon angiographic catheter. After that, aortography was performed to confirm an exact fistula tract and blood supply of the subclavian and left common carotid arteries (Fig. 3a). Therefore, we decided to embolize the fistula with the AVP 1 (6 mm in diameter). A right Judkins guiding catheter and device were inserted through the fistula tract. Test injections were performed to verify the correct position of the device before it was released (Fig. 3b). Ten minutes after deployment of the plug, repeat aortography showed complete occlusion of the fistula tract. According to the aortogram, no flow decrease occurred in the left subclavian and left common carotid arteries (Fig. 3c). Continuous murmur also had disappeared. The patient was discharged on the following day.

Discussion

Thoracic arteriovenous fistulas are rare congenital malformations. Their clinical presentation is closely related to the size of the fistula. Initial symptoms and signs may include congestive heart failure or continuous or innocent murmurs. Several case reports of an arteriovenous fistula simulating patent ductus arteriosus have been published (6). The present patient also had a patent ductus arteriosus, which was the main cause of late diagnosis.

In the second procedure, we particularly focused on the fistula. Although our patient had no complaint, we decided to occlude the fistula tract because of its large size and high calculated pulmonary to systemic blood flow ratio. Although a traditional surgical approach was possible, we chose percutaneous interventional treatment because of its several advantages over surgery, such as shortened hospitalization, performance without thoracotomy, and short procedure time. However, few reports in the literature have described transcatheter embolization of an aorto-azygous fistula. Gianturco coils and Gianturco-Grifka vascular occlusion devices have been used to occlude arteriovenous malformations (5, 7).

To the best of our knowledge, this is the first report on transcatheter embolization of a caroticoazygous fistula using the AVP 1. This is a

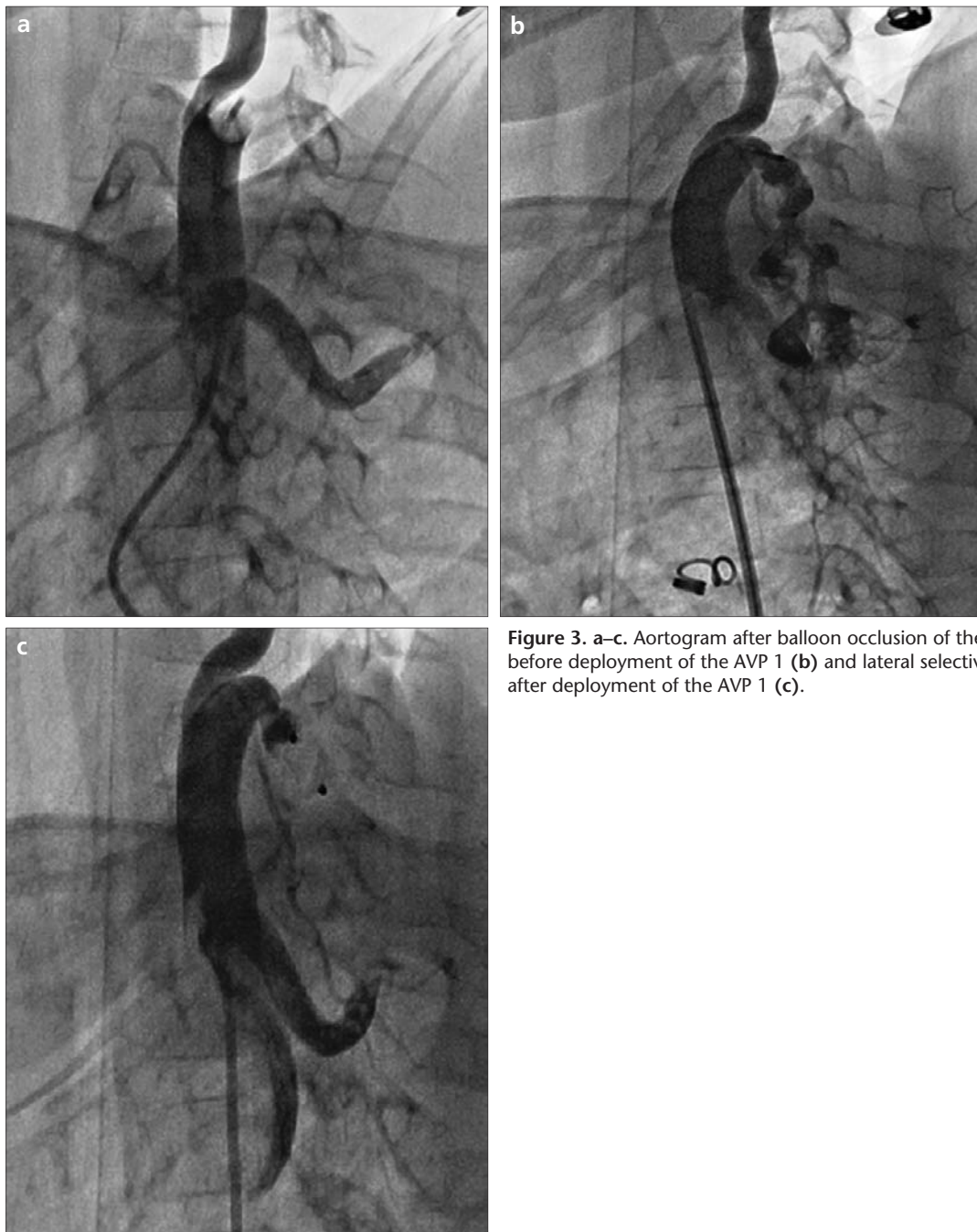


Figure 3. a–c. Aortogram after balloon occlusion of the fistula (a). Test injection before deployment of the AVP 1 (b) and lateral selective descending aorta angiogram after deployment of the AVP 1 (c).

self-expandable device made of nitinol mesh wire. Vascular Plug I can be used for several types of arteriovenous malformations, such as coronary and pulmonary arteriovenous fistulas. The potential advantages of vascular plugs are the ease of delivery, low profile, wide range of device sizes, fast and safe positioning, and possibility of repositioning or removal before final delivery (8–10). The manufacturer recommends selecting a device approximately 30%–50% larger than the vessel diameter. In this particular

case, we safely and effectively used a larger-diameter AVP 1 (176% of the narrowest part of the vessel), which has also been applied in previous cases (11). We prefer the AVP 4 over AVP 1. This new device can be deployed through a 4 F diagnostic catheter, which is a major advantage of this device, especially in pediatric populations (12). However, it was not available at our hospital.

In conclusion, a caroticoazygous fistula is an extremely rare arteriovenous malformation and should be

differentiated from patent ductus arteriosus. In patients with continuous murmur, one should keep in mind that these two conditions may be present at the same time. Transcatheter embolization of these malformations can be performed safely, more comfortably, and efficiently with the AVP 1 as compared to surgery.

Conflict of interest disclosure

The authors declared no conflicts of interest.

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