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Endovascular management of a rare complication of an aortic coarctation

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ABSTRACT

A 28-year-old pregnant woman presents with arterial hypertension of the upper limbs. The examination suggests an aortic coarctation. After a normal delivery, a contrast-enhanced computed tomography revealed a subocclusive aortic coarctation of the descending thoracic aorta and a 33-mm aneurysm developed from the left cervical-thoracic artery. The coarctation of the aorta was treated by a stent graft, and the aneurysm was treated by an injection of thrombin and glue.

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Introduction

Coarctation of the aorta (CoA) is a relatively common defect that accounts for 5%-8% of all congenital heart defects. CoA may occur as an isolated defect or in association with various other lesions. Late aneurysmal formation in the proximal or the distal aortic arch is a well-recognized sequela of untreated coarctation and is associated with increased risk of aortic rupture and death [1]. We present a rare complication of a CoA and its endovascular management.

Case report

During her obstetrical consultation, a 28-year-old pregnant woman presented with arterial hypertension of the upper limbs. She was not found to have proteinuria or other clinical or biological sign of pre-eclampsia. An aortic coarctation was suspected. After a normal delivery, she presented with persistent high blood pressure of the upper limbs and low blood pressure of the lower limbs with an ankle brachial index of >20 mm Hg.

Contrast-enhanced computed tomography (CT) (Fig. 1A and B) revealed a subocclusive coarctation of the descending thoracic aorta and a wide aneurysm (33 mm). During aortography, selective injection through the left subclavian artery (LSA) confirmed the diagnosis, revealing an enlarged cervical-thoracic artery afferent to the aorta (Fig. 2). A 6-mm balloon dilatation (Mustang 5F Boston Scientific Benelux, 75 cm 6 × 20 mm) of the CoA was performed through a percutaneous approach of the left brachial artery because the balloon did not pass the CoA by a right arterial femoral access. Thereafter, a 16 mm × 4 cm stent graft (Advanta V12; Atrium Europe B.V.

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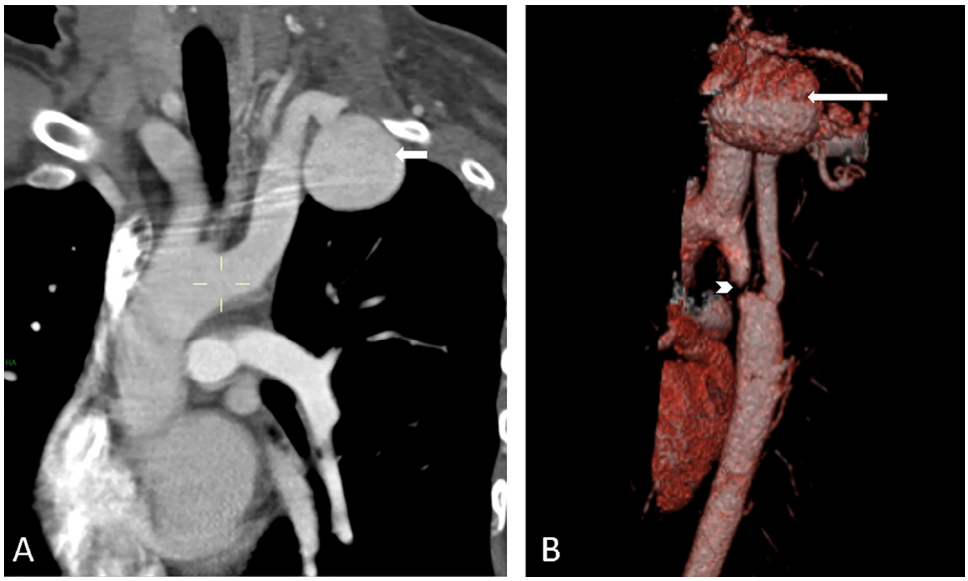

Fig. 1 – (A) Initial computed tomography with contrast enhancement. Coronal reconstruction shows a left large aneurysm (arrow). (B) Three-dimensional surface reconstruction demonstrates a subocclusive coarctation of the descending thoracic aorta (arrowhead) and the aneurysm (arrow).

Mijdrecht, The Netherlands) was introduced through an arteriotomy of the right femoral artery. The stent was placed at the aortic isthmus and covered the CoA. The postoperative CT 5 days later showed a well-deployed stent graft but a persistent flow into the aneurysm of the left cervical-thoracic artery. Angio-CT 3 months later revealed aneurysm patency despite the stent (Fig. 3).

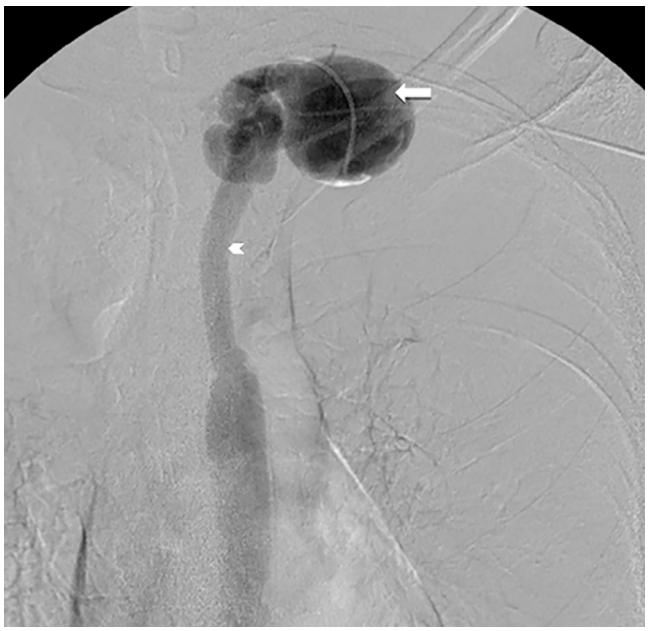

Fig. 2 – Opacification of the aneurysm (arrow) of a developed left cervical-thoracic artery (arrowhead) through the left axillary artery.

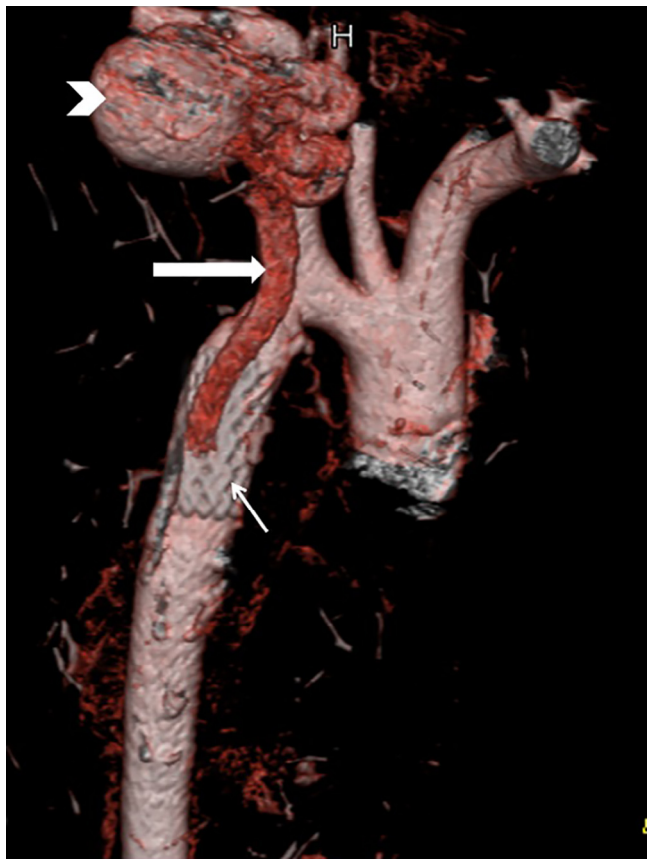

Fig. 3 – Angiocomputed tomography 3 months after stenting. Three-dimensional surface reconstruction demonstrates the aneurysm patency (arrowhead) despite the stent (thin arrow) and the collateral (arrow).

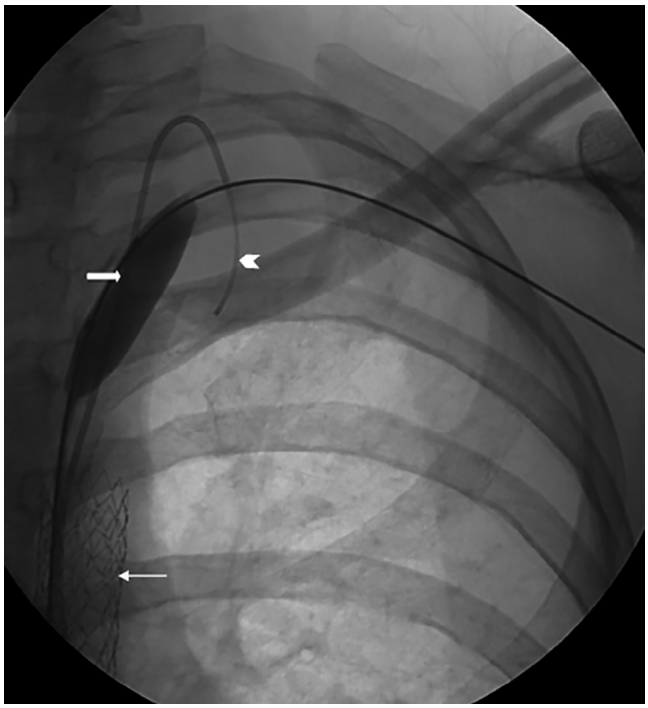

Fig. 4 – Thoracic radiography showing the balloon deployment into the left subclavian artery (arrow), as well as a microcatheter into the aneurysm (arrowhead) and the stent graft into the descending thoracic aorta (thin arrow).

The arteriography confirmed the persistent flow into the aneurysm and several collaterals.

Consequently, the aneurysm was treated through a percutaneous bilateral approach of the femoral arteries. After an intravenous injection of 5000 UI of heparin (heparin, LEO Pharma A/S), a 12 mm × 2 cm balloon (Mustang 5F Boston Scientific Benelux, 75 cm 12 × 20 mm) was placed into the LSA at the ostium of the left costocervical artery (Fig. 4). A microcatheter was introduced into the aneurysm, through the contralateral femoral artery. After deployment of the balloon, 1000 UI of thrombin (protamine sulfate; LEO Pharma A/S, Ballerup) were injected in the aneurysm and a partial thrombosis was obtained.

The procedure continued with an injection of 4 mL of glue (Glubran Nootens H. Rodenbach) and lipiodol (Lipiodol Ultra Fluide Guerbert) in a proportion of one-third.

A complete thrombosis of the aneurysm was obtained (Fig. 5). During the immediate postprocedure observation, the patient presented with a subischemia of the left upper limb characterized by acute pain, which completely resolved within 72 hours after initiation of an intravenous heparin therapy. A CT angiography performed 2 weeks after the intervention confirmed the complete thrombosis of the aneurysm (Fig. 6).

Discussion

CoA corresponds to a congenital narrowing of the lumen of the aorta that occurs in the upper thoracic aorta just below the origin of the LSA. Sometimes, CoA can reach the abdominal aorta. CoA is present at birth and men are more often affected than women (male-to-female ratio, 1:5) [2]. Some congenital heart abnormalities may also be associated. CoA represents 5%-7% of the congenital cardiovascular diseases, and

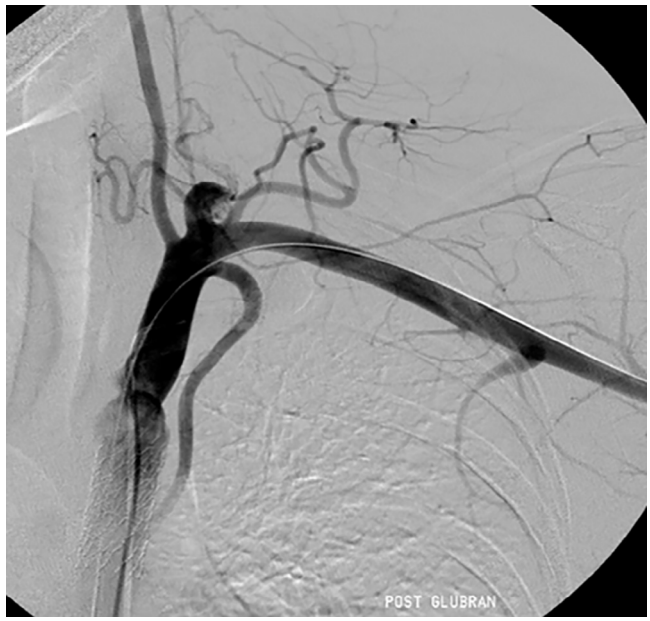

Fig. 5 – Opacification of the left subclavian artery showing a complete thrombosis of the aneurysm after the injection of thrombin and glue.

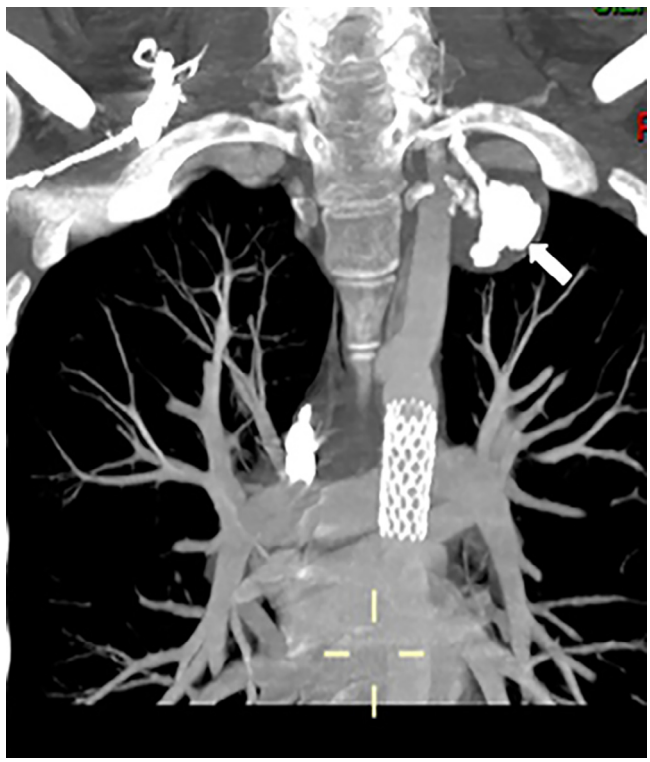

Fig. 6 – Angiocomputed tomography 2 weeks after intervention shows complete thrombosis of the aneurysm (arrow).

its incidence is estimated at 0.3-0.4 per 1000 [3]. CoA is a solitary lesion in 82% of cases, but multiple associations are described, such as the Turner syndrome, a bicuspid aortic valve, an intracranial aneurism (10%), a ventricular septum defect, and an atrial septal defect among others [4]. Because of mechanical outflow obstruction and extensive collateral formation, a number of hemodynamic changes are seen in CoA. The most notable is a differential hypertension produced in the segment of the aorta above the site of narrowing, leading to increased dilatation of cervicocephalic arteries. Aneurysms can develop upstream or downstream of the stenosis. It has been reported that 32% of aneurysms are proximal to the coarctation, 51% are distal, and 17% involve the LSA rather than the aorta [1].

Clinical symptoms depend on the position, the degree, and the extent of the stenosis. Commonly, the diagnosis is evoked by discovering a difference of 20 mm Hg in systolic blood pressure between the arms and the legs in favor of the arms. The pathogenesis of CoA remains unclear. Some hypotheses are proposed, such as a hypoperfusion of the aortic isthmus during fetal life, an extension of the ductal tissue into the aortic wall, or an abnormal migration of neural crest cells [5]. If unrepaired, average survival with CoA is 31 years, with a reported mortality of 75% by 46 years of age [3]. The treatment improves life expectancy and quality of life by reducing the incidence of aortic and cardiac disabling conditions, such as aneurysm of the ascending aorta, coronary artery disease, high blood pressure, and aortic and mitral valvular diseases [6]. Mortality mainly occurs as a result of congestive heart failure (25.5%), aortic rupture (21%), endocarditis (18%), or an intracranial hemorrhage (11.5%).

Initially, CoA was treated by open surgery. The first operation was performed by Crafoord in 1945 and consisted of a complete resection of the CoA and an end-to-end anastomosis. Other surgical approaches have been attempted, such as subclavian flap angioplasty or patch aortoplasty [4]. Balloon angioplasty became available as an alternative treatment in the 1980s, but stenosis recurrences, aneurysms, and aortic dissections were found to be disadvantages of this treatment [7]. In the early 1990s, endovascular stents were introduced and became a less invasive alternative to open surgical repair [8,9]. Potentially, stent grafts reduce the risk of hemorrhage due to postangioplasty aortic rupture. However, no study has compared open surgery and stent placement for the treatment of the CoA [7]. The treatment remains a challenging procedure and long-term analyses are needed to identify the best therapeutic strategies.

Conclusion

The association of a CoA with aneurysm of the left cervicalthoracic artery is rare and the management is challenging. The percutaneous route should be considered, if feasible and safe, and where expertise is available.

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