

CASE REPORT

Aortic Isthmus Pseudoaneurysm After Coarctation Repair as a Source of Thromboembolism

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ABSTRACT

A 60-year-old male smoker with a past medical history of moderate hypertension, hypercholesterolemia, bronchial asthma and surgically corrected aortic coarctation with interposition grafting at the age of 17, presented with four episodes of post-exercise lower limb thromboembolism within a period of two years. The electrocardiogram was normal and multiple Holter recordings showed no rhythm abnormalities. Transthoracic echocardiography showed normal left ventricular dimensions and systolic function, normal right ventricle, bicuspid aortic valve with moderate insufficiency and mild stenosis, ascending aorta with a diameter of 46 mm, and a pressure gradient across the aortic isthmus of 20 mmHg. Transesophageal echocardiography revealed no intracardiac thrombi or shunts and neither dissection nor thrombus could be detected in the descending thoracic aorta. Although the patient was subjected to multiple diagnostic imaging examinations, it was a dual source computed tomography (CT) with three-dimensional image reconstruction of the aorta that disclosed the detachment of the graft's wall inner surface at the site of its proximal anastomosis with the descending thoracic aorta, just distal to the origin of the left subclavian artery, that resulted in the formation of a pseudoaneurysm which served as the source of distally embolizing thrombi. Moreover, in the distal thoracic aorta just after the graft's distal anastomosis, a mild stenosis was noted due to intense intramural calcification. Although various therapeutic approaches were considered, the patient was finally taken to the operating room, where, via a left lateral thoracotomy, the preoperative findings were confirmed and the lesions successfully repaired.

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KEY WORDS: aortic isthmus; coarctation of the aorta; pseudoaneurysm; thromboembolism; computed tomography; magnetic resonance imaging; surgical repair; endovascular treatment

ABBREVIATIONS:

CoA = coarctation of the aorta
CT = computed tomography
DTA = descending thoracic aorta
ECG = electrocardiogram
LCCA = left common carotid artery
LSA = left subclavian artery
MRI = magnetic resonance imaging
TEE = transesophageal echocardiogram
TTE = transthoracic echocardiogram

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INTRODUCTION

The clinical application of computed tomographic (CT) imaging has recently been broadened with the advent of dual source CT permitting the use of two X-ray sources and two detectors at the same time at two different kV levels simultaneously. The advantage of this new imaging modality is that it enables double temporal resolution, double speed, and twice the power, while lowering dose even further. We recently employed dual source CT and obtained images of excellent quality which allowed us to identify a pseudoaneurysm at the site of previous surgical repair of coarctation of the aorta, as the embolic source in a 60-year-old patient suffering from recurrent systemic thromboembolic events.

Conflict of Interest: none declared

CASE REPORT

A 60-year-old male smoker presented with post-exercise pain in his left calf. Before this happened, the patient had been in good overall condition. His height was 180 cm and his weight 75 kg. His past medical history was significant for moderate hypertension managed with an angiotensin-converting-enzyme inhibitor and a calcium-channel blocker; hypercholesterolemia treated with statins; bronchial asthma for which he was receiving a β -agonist inhaler; and he had a history of surgically corrected aortic coarctation at the isthmus, with resection of the stenosed segment and insertion of an interposition graft, performed at the age of 17. Four years after the operation, the patient suffered a transient ischemic attack with right pyramidal distribution for which he had received anticoagulant therapy. This episode was attributed at that time to a cerebral embolus originating either from the aortic valve or the left carotid artery.

Upon presentation, the patient's left lower extremity was found cold and pale with non-palpable peripheral pulses. A Doppler ultrasound examination was performed that showed absent blood flow. He was managed conservatively with heparin therapy and vitamin K antagonists for his lower extremity occlusive disease with satisfactory restoration of blood flow thereafter.

In search for the source of the embolus responsible for the patient's symptoms, an extensive evaluation was performed. The electrocardiogram (ECG) was normal and there were not detected any rhythm abnormalities on multiple Holter recordings. The transthoracic echocardiogram (TTE) showed normal left ventricular dimensions and systolic function, with an ejection fraction of 60% without regional wall motion abnormalities, normal right ventricular dimensions and function, and a bicuspid aortic valve with moderate insufficiency and mild stenosis. No other intracardiac pathology could be identified. The ascending aorta had a diameter of 46 mm and the pressure gradient across the aortic isthmus was 20 mmHg. A cardiac magnetic resonance imaging (MRI) examination was subsequently performed that showed the distal thoracic aorta (DTA) with the existent interposition graft in place. A concentric stenosis, with a diameter of 10 mm, was shown just after the graft's distal anastomosis with the DTA, 70 mm distal to the origin of the left subclavian artery (LSA); in addition, a regional dissection of 35 mm in length was revealed originating in the distal aortic arch and extending to the proximal graft's anastomosis with the DTA, just after the origin of the LSA (Figures 1 & 2).

At that point, only the diagnosis of residual isthmic stenosis had been made and it was not still clear which was the source of emboli responsible for the patient's symptoms.

Eighteen months later, and while receiving therapy with a vitamin K antagonist, the patient presented again with a lower extremity post-exercise thromboembolic occlusive event. The Doppler ultrasound examination at that time showed absence of blood flow in both left anterior and posterior tibial arteries

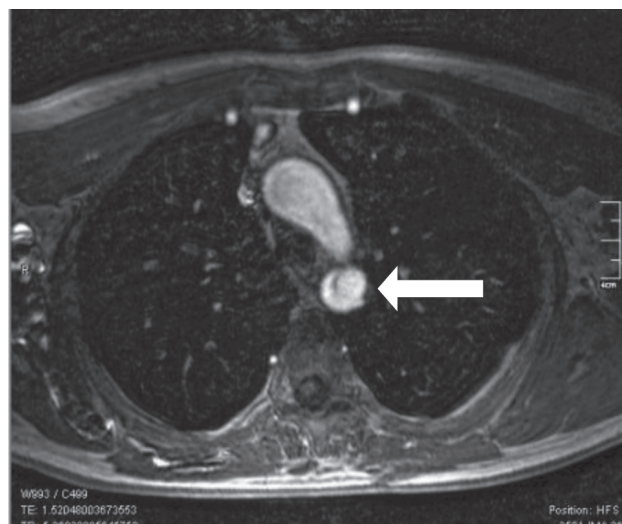


FIGURE 1. Cardiac MRI image showing the dissection of the DTA (arrow).

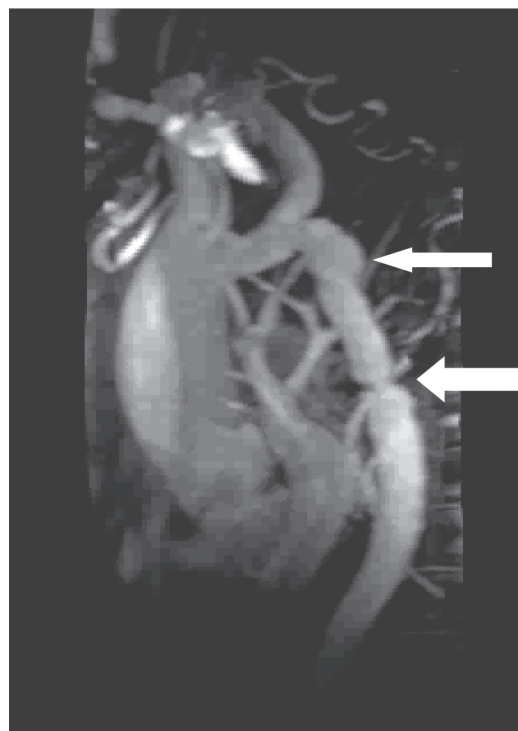


FIGURE 2. Cardiac MRI image which shows the dissection of the DTA (thin arrow) and the ring-like stenosis (thick arrow).

and once again he was managed conservatively.

That episode was followed by another one 5 months later. Arteriography was performed that showed total occlusion of the right popliteal artery (Figure 3). The patient underwent surgical revascularization by embolectomy with successful

CONGENITAL CORONARY ANOMALIES

thrombus removal and subsequent restoration of blood flow. At that time, a chest computer tomography (CT) was obtained that showed the dissection in the DTA, as it had already been imaged by the previous cardiac MRI; additional findings now included the intraluminal thrombus visualized at the site of the graft (Figure 4).

Eleven months later, the patient developed a new episode of lower extremity thromboembolism. Arteriography was performed and this time occlusion of the right common femoral artery with restricted peripheral blood flow was demonstrated (Figure 5).

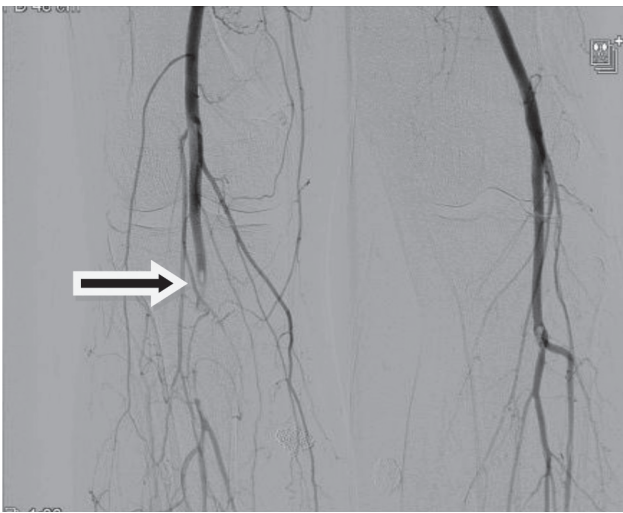


FIGURE 3. Angiographic image showing the total occlusion of the right popliteal artery (arrow).

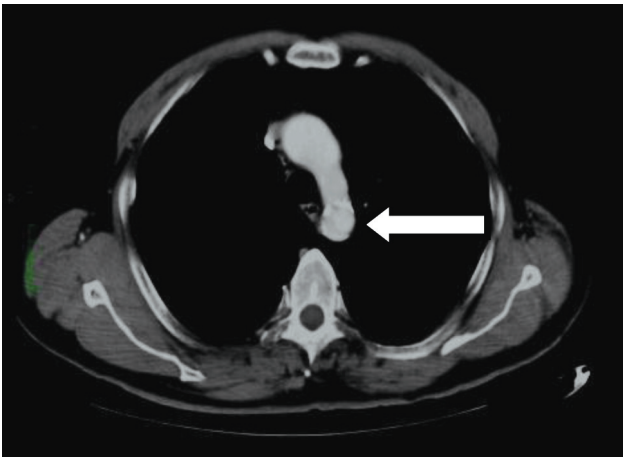


FIGURE 4. Chest CT image showed the dissection of the DTA just after the origin of the LSCA and contrast deficits compatible with thrombosis (arrow). CT = computed tomography; DTA = descending thoracic aorta; LSCA = left subclavian artery.

After that last event, it was speculated that somewhere within this patient's arterial tree there must have been a nidus for continuing thrombus formation which was acting as a source of distal emboli responsible for his recurring symptoms. It was felt that this needed to be further elucidated and the anatomy accurately delineated before it could be made possible to properly and satisfactorily manage the patient's problem, rather than continuing with a conservative approach. Clearly, the imaging modalities which had been employed so far during the diagnostic work up had all failed to achieve that and the problem had still remained largely undiagnosed and recurring.

With that in mind, a further attempt was made at identifying the embolic source by performing a dual source chest CT with thin slices. The obtained images were reconstructed in the sagittal, horizontal, and coronal planes and additionally, three-dimensional reconstruction of the aorta was performed. It was thus shown that at the graft's proximal anastomosis with the DTA, the outer surface of its wall was appropriately attached to the aortic wall while its inner surface was not; instead, it was protruding inside the aortic lumen. In this way a tear had been formed which led to a chamber with characteristics of a pseudoaneurysm. Moreover, within this pseudoaneurysm, small hypodense formations attached to the graft's wall were observed, with some of them "hanging" inside the aortic lumen. These, most probably, represented thrombi. Also, heavy intramural calcification was shown just after the graft's distal anastomosis resulting in the ring-like stenosis of the DTA

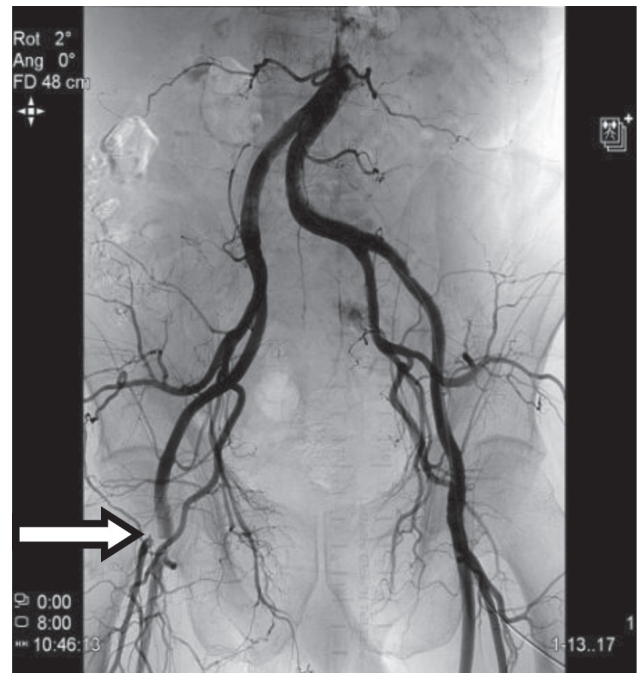


FIGURE 5. Angiographic image depicting the occlusion of the right common femoral artery (arrow).

which had already been revealed by the previously performed cardiac MRI (Figures 6, 7, and 8).

These findings demonstrated that it had been a pseudoaneurysm at the site of previous surgical repair of the coarctation of the aorta, the place where thrombi were produced and then distally embolized resulting in the patient's ischemic symptoms in the lower extremities.

It was then decided to proceed to surgical repair of this pseudoaneurysm and further preoperative evaluation was planned. A transesophageal echocardiogram (TEE) was performed which showed similar findings with those of the TTE and also provided some additional information. Particularly, the bicuspid aortic valve's non-coronary/right coronary cusp appeared calcified and immobile, while the left coronary cusp was freely mobile, thus resulting in a normal overall opening with mild stenosis and moderate insufficiency; the ascending aorta was mildly dilated with a diameter of 43 mm and a left ventricular outflow tract of 30 mm; slight angulation of the aorta at the area of origin of the LSA was shown, and the graft was demonstrated with a diameter of 12 mm and the DTA proxi-



FIGURE 7. Chest CT image showing the pseudoaneurysm in close proximity to the left subclavian artery (arrow).

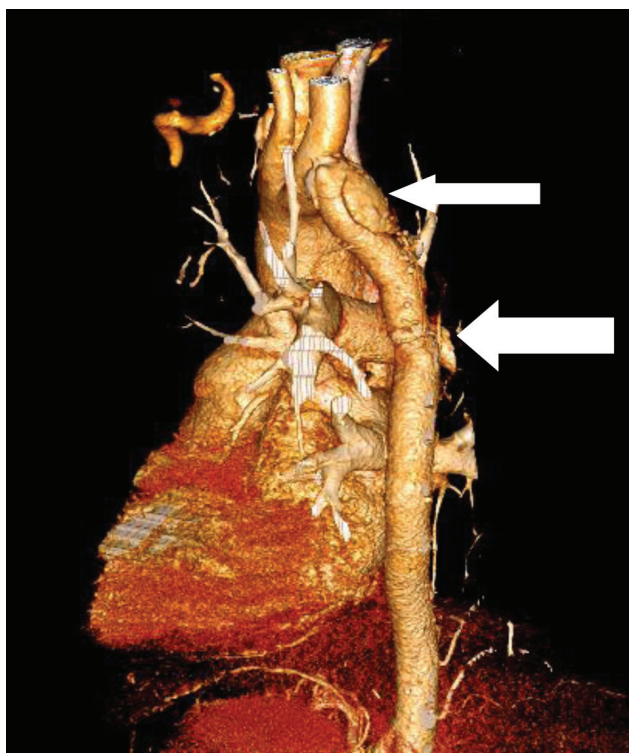


FIGURE 6. Dual Source chest CT image with three-dimensional reconstruction of the aorta. The thin arrow points to the aortic dilatation corresponding to the pseudoaneurysmal chamber which was formed due to detachment of the graft's wall inner surface at its proximal end just after the origin of the left subclavian artery. The thick arrow points to the ring stenosis just after the graft's distal end.

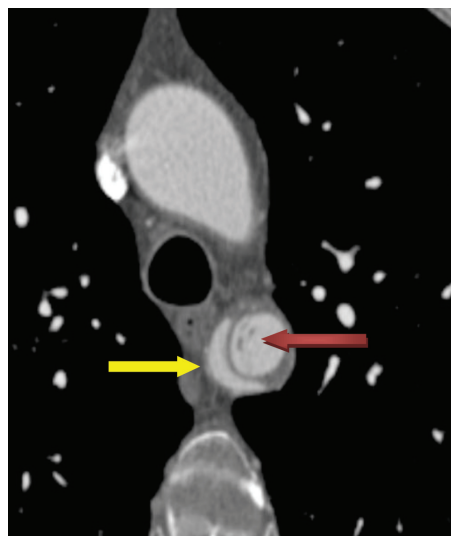


FIGURE 8. Chest CT image showing the small hypodense formations inside the aortic lumen probably representing thrombi (red arrow) as well as the pseudoaneurysm sac (yellow arrow).

mal to it with a diameter of 23 mm resulting in a mild residual aortic isthmus stenosis with a maximum pressure gradient of 18 mmHg; finally, neither intracardiac thrombi nor shunts were discovered. At coronary angiography, which was subsequently performed, no coronary artery disease was identified and the

aortography performed at the same time revealed moderate aortic insufficiency and mild residual aortic isthmus stenosis with a pressure gradient of 25 mmHg.

The surgical repair of the pseudoaneurysm was performed via a left lateral thoracotomy under femoro-femoral cardiopulmonary bypass for distal (abdominal) organ protection and mild hypothermia (32-34 degrees Celsius). The graft was dissected, tapes were passed around the distal aortic arch and the left subclavian artery which were individually cross-clamped and the pseudoaneurysm opened at the level of the proximal graft's anastomosis with the DTA peripheral to the origin of the LSA. The thrombi were removed through an incision on the graft's surface and the graft itself was resected and replaced by a straight 20 mm Vascutec graft (Figures 9 and 10). The postoperative period was complicated by development of subcutaneous emphysema and pneumothorax which were successfully treated with chemical pleurodesis with talc. The rest of the patient's hospitalization was uneventful and he was well at discharge. He remains well at one year follow-up.

DISCUSSION

The present case demonstrates several important aspects of coarctation of the aorta (CoA). From a historical point of view, this patient was among the first patients who had surgical repair of CoA over 40 years ago in the United Kingdom in 1967. At that time, surgery was the only option for such patients and he was treated successfully with resection and interposition grafting. Shortly after the operation he suffered from a transient ischemic attack which at that time was attributed to an embolic event. It could have been possible though that it represented a manifestation of a more widespread vasculopathy, characteristic of CoA, which involves the cerebral microcirculation as well and contributes to the increased rate of cerebrovascular events in such patients.¹

Over the ensuing years after the operation he remained in good overall health with hypercholesterolemia, moderate hypertension, and asthma being the medical conditions for which he was receiving treatment. Arterial hypertension after CoA repair can develop at various stages throughout patients' life. Late hypertension in such patients does not appear to be related to the mechanisms that are involved in the development of essential hypertension² and most probably reflects the generalized and persistent nature of the vasculopathy in CoA.

The non-invasive cross-sectional imaging techniques, including echocardiography, CT, and MRI, are used for post-treatment evaluation and surveillance. Considerations about the young age of most patients after CoA repair, the need for periodical examinations, and the hazards from repeated radiation exposure should be kept in mind. Since MRI has been shown to be superior to echocardiography for pre- and post-treatment evaluation of CoA,³ the most "cost-effective"

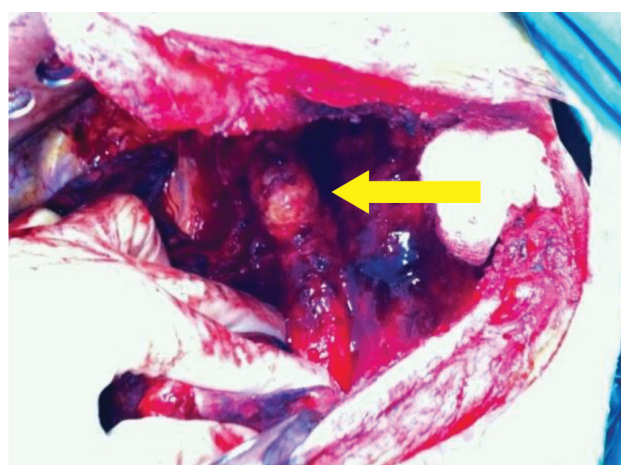


FIGURE 9. Photograph taken during the operation shows the aortic dilatation just after the origin of the LSA (arrow).

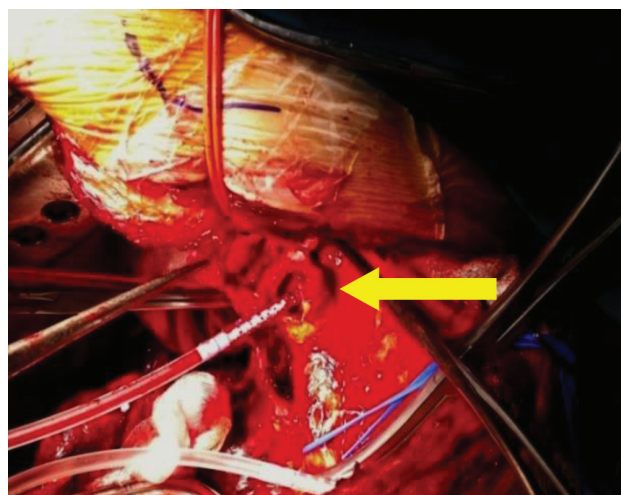


FIGURE 10. Photograph taken during the operation shows the pseudoaneurysm (arrow).

approach appears to be the combination of clinical assessment and MRI.⁴ An exception could be after stenting where signal drop-out artifact can obscure the obtained images so at least one contrast-CT study would be required for better assessment.⁵

These issues were evident in our patient's case. Initial TTE failed to show the pseudoaneurysm and only the gradient across the graft at the area of the isthmus was identified. The TEE which was performed later showed only some additional anatomic details about the intracardiac structures and the aorta. The cardiac MRI showed with sufficient detail the ring-like stenosis in the distal DTA but did not demonstrate the pseudoaneurysm as well. A plain chest CT performed later showed the intraluminal thrombi but failed to demonstrate the pseudoaneurysm. In the end, it was the dual-source CT

that showed clearly the pseudoaneurysm with the attached thrombi in it serving as the distal emboli source responsible for the patient's symptoms.

Aortic aneurysm formation at the site of repair of CoA can occur after either surgical or endovascular treatment. It complicates about 5.4% of surgical repairs for CoA, being most common after synthetic patch aortoplasty, and if left untreated it is universally fatal.⁶ Although various factors have been implicated as responsible for aneurysm formation, it seems that the structural abnormalities present in the aortic wall, which persist even after successful repair of the coarctation, play the most important role.⁷

Several complications have been described in association with such aneurysms. Rupture can occur and it is always fatal. An aortobronchial fistula presents with hemoptysis and CT is usually diagnostic. Surgical repair offers the definite treatment,⁸ whereas stent-graft repair can be used as a temporary stabilizing measure before surgery in emergency situations.⁹ Aortoesophageal fistula presents with chest pain, dysphagia, and hematemesis with contrast-CT being diagnostic.¹⁰ Esophageal repair can be offered but surgical repair of the aneurysm is usually not possible due to the contaminated field and conservative management results almost always in a fatal outcome.¹¹

In our case, distal embolization of thrombotic material that had accumulated within the pseudoaneurysm was described. Similar cases of thromboembolic leg ischemia after coarctation repair have been reported in the past.^{12,13} An interesting case of dual disability from a single thromboembolic source has been reported, whereby a pseudoaneurysm near the bifurcation of the brachiocephalic trunk was the source of thromboembolism to the right common carotid artery with resultant hemiplegia of the left arm, while at the same time thromboembolism to the right subclavian artery resulted to right arm claudication.¹⁴

Clearly, aneurysm formation at the site of previous repair of CoA is associated with serious complications and often fatal outcome and therefore requires an accurate diagnosis and optimal treatment. It seems that satisfactory results are achievable by all of the available therapeutic methods, open redo surgery, hybrid techniques, and endovascular repair.¹⁵ Considering that after repair of CoA, associated cardiovascular diseases, especially bicuspid aortic valve, are the most common cause for reoperation,¹⁶ it is important to carefully assess each patient and their comorbidities, which might require treatment as well, in order to provide the most appropriate therapy.

Our patient, apart from the pseudoaneurysm, had a bicuspid aortic valve and an aneurysmal dilatation of the ascending aorta. The association of CoA with a bicuspid aortic valve increased the prevalence of ascending aortic complications when compared to isolated bicuspid aortic valve, 8% versus 3.7%.¹⁷ Since our patient's mixed aortic valvulopathy was not severe enough to warrant valve replacement and the ascending aortic dilatation was not large enough to place him at risk for rupture it was decided to concentrate only on the pseudoaneurysm at

the site of previous repair of CoA.

Repeat surgery for repair of such pseudoaneurysms is associated with significant mortality and morbidity rates. On the other hand, open redo surgery, even via a repeat left thoracotomy, has been shown to be both feasible and effective with good late results.¹⁸ Moreover, when compared with endovascular therapy, surgery had resulted in a smaller percentage of patients who needed a second repeat procedure, 4% versus 28% at 5 years.¹⁹ In addition, approach via a median sternotomy provides the possibility to perform a single-stage repair of complex CoA, by the use of extra-anatomic ascending-to-descending aortic bypass, simultaneously with other associated cardiac diseases.²⁰

Endovascular techniques for treatment of pseudoaneurysm after previous repair of CoA are becoming increasingly popular and their use seems to have numerous supporters. Very promising short- and mid-term results favour the proposition of endovascular repair of such pseudoaneurysms as first line treatment option.²¹ Moreover, long-term follow-up of a small cohort of patients treated by endoluminal repair showed satisfactory initial results and long-term durability of the repair,²² similar to other long-term data from earlier studies.²³

Although endovascular repair of such pseudoaneurysms shows promise as an alternative to repeat surgery, various complications might occur, such as stent dislocation and fracture; aortic dissection and rupture; injury to the access vessels, peripheral emboli and cerebrovascular accidents.²⁴ Moreover, there are concerns regarding the incidence of cerebrovascular accidents, spinal cord injury, and left upper extremity ischemia after covering the origin of the left subclavian artery.

Hybrid repair of an aneurysmal dilatation of the thoracic aorta after surgical repair of CoA has been described, where percutaneous stent-graft implantation combined with LSA-to-left common carotid artery (LCCA) transposition achieving successful results.²⁵ Another case of staged hybrid treatment of ascending aortic and distal aortic arch pseudoaneurysm after repair of CoA has been reported.²⁶ In that case, the ascending aortic aneurysm was associated with a bicuspid aortic valve and was corrected during the first operation by a "modified Bentall - De Bono" with simultaneous explantation of the LCCA and connection with a graft to the ascending aortic conduit. During a second procedure, exclusion of the pseudoaneurysm in the DTA was performed by endovascular implantation of a stent-graft prosthesis and LSA coverage without complications.

In our patient's case, it was decided to proceed with surgical repair of the pseudoaneurysm instead of endovascular treatment. This was based primarily on the morphology of the involved region of the DTA. There was an interposition graft which had been inserted over 40 years ago with a pseudoaneurysm at its proximal anastomosis and a ring-like stenosis after its distal anastomosis. The wall of the DTA at that region was heavily calcified and it was questionable whether it could provide sufficient support for endovascular stent deployment. The risk of aortic rupture after stent dilatation was considerably high

and there were concerns about the proper seal of the stent on the weak aortic wall. The patient was operated on via a repeat left thoracotomy. The thrombi were removed from within the pseudoaneurysm and the old graft was excised and replaced by a new one with successful results. At one year follow-up he was well with no further complications up to that time. The mixed valvulopathy of the bicuspid aortic valve and the aneurysmal dilatation of the ascending aorta are under observation for further progression and possible intervention in the future.

It is possible that randomized control trials comparing the results from the various therapeutic interventions and their complications could result in more clear indications for the choice of the appropriate treatment in each patient. Considering though the fact that patients with late complications after previous repair of CoA represent a quite small population group who often present in an emergency setting with life threatening conditions, it could be difficult, if not inappropriate, to expect strict adherence to such trials' protocols. Collective experience by multiple centers and investigators specializing in this area provide a pool of data from which useful conclusions can be drawn. It seems that patients' characteristics and preferences as well as skill and experience of the interventional cardiologists and cardiac surgeons involved in the management of such patients determine the optimal treatment and outcome.

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