

Necessity of life-long follow-up after surgery for coarctation of the aorta: a case series of very late false aneurysm formation

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Background

Coarctation of the aorta accounts for 5–7% of congenital defects of the heart and great vessels. It requires treatment in the form of open surgical or percutaneous repair. Common long-term complications include re-stenosis and aneurysm formation. The formation of a false aneurysm is a complication with a significant morbidity and mortality.

Case summary

We reviewed six cases of late false aneurysm after repair of a coarctation of the aorta. Our six cases developed a false aneurysm after an open surgical repair of a coarctation more than 30 years after initial surgical repair. All aneurysms were located at the aortic repair site.

Discussion

The symptoms or risk factors in the described cases are not uniform and are difficult to include in a general follow-up protocol. Guidelines recommend frequent evaluation, but do not specify duration or intervals of imaging follow-up. Our cases support the necessity of life-long follow-up in patients with open aortic repairs irrespective of symptomatology.

Keywords

Coarctation • False aneurysm • Follow-up • Case series

ESC Curriculum

2.1 Imaging modalities • 2.4 Cardiac computed tomography • 7.5 Cardiac surgery • 9.1 Aortic disease • 9.7 Adult congenital heart disease

Learning points

- False aneurysms can be diagnosed decades after initial surgical aortic coarctation repair.
- Aortic false aneurysms have a high morbidity and mortality burden and therefore life-long imaging follow-up after surgical repair of aortic coarctation is warranted.
- Serial imaging (preferably computed tomographic angiography or magnetic resonance angiography) should be considered in these patients irrespective of symptoms.

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Introduction

Coarctation of the aorta is a congenital defect of the heart and great vessels that accounts for 5–7% of these congenital defects and has an incidence of between 0.3 and 0.4 per 1000 live births.¹ Concomitant defects include mostly bicuspid aortic valve, congenital aortic valve stenosis, ventricular septal defect (VSD), patent ductus arteriosus, transposition of the great arteries, left ventricular outlet tract stenosis, mitral valve malformations, or hypoplastic left heart syndrome.^{2–5}

Therapeutic options include surgical intervention and balloon angioplasty, which is usually combined with stent placement. Surgical interventions tend to be performed in newborns and infants, whereas stent placement is the preferred intervention in adolescents and adults.⁶ These surgical interventions have developed from end-to-end anastomosis, subclavian flap technique and patch aortoplasty to extended end-to-end anastomosis, which is currently the most performed procedure.⁶ Long-term postoperative complications include re-stenosis and (false) aneurysm formation. The life-time risk of these complications varies between 1% and 38% and depends on the primary type of repair.^{1,7,8}

A false aortic aneurysm is a rare complication with a described incidence of 1.3–3.0% after surgery, 8% after redo thoracoabdominal or

descending aortic aneurysms, and >10% after open repair for coarctation of the aorta. These reported incidences are within a median follow-up period from 5.9 to 13.1 years.^{7,9–13}

While much is known about more common complications such as a dysfunctional bicuspid aortic valve, less is known about the very long-term follow-up (e.g. more than 20 years) of aortic repairs and the occurrence of false aneurysms. This case series reports six cases of false aneurysm that occurred more than three decades after open repair for coarctation of the descending aorta.

The purpose of this case series is to raise awareness about false aneurysms that can appear decades after the primary operation and to underscore the importance of lifetime follow-up. Studies have shown very good long-term survival up to 60 years in patients with coarctation who reach adolescence.¹⁴ However, long-term comorbidity and complications are common, including arterial hypertension, left heart failure, intracranial haemorrhage (from berry aneurysm), infective endocarditis, aortic rupture/dissection, premature coronary and cerebral artery disease, and associated heart defects such as re-coarctation and stenotic or regurgitating bicuspid aortic valve disease.^{5,14,15}

Timeline

Patient number	1	2	3	4	5	6
Age at time of surgical repair of coarctation	19 years	16 years	27 years	21 years	2 months	10 days
Type of surgery	Surgical bypass from left subclavian artery (LSA) to descending aorta	Interposition graft LSA to descending aorta	Interposition graft LSA to descending aorta	Subclavian flap technique	End-to-end anastomosis	Patch angioplasty
Time from primary to false aneurysm repair	31 years	48 years	34 years	43 years	34 years	30 years
Concomitant cardiac lesions and/or previous cardiovascular surgery	Ventricular septal defect (VSD), pulmonary artery stenosis	Bicuspid aortic valve (BAV), paroxysmal atrial fibrillation, mechanical aortic valve replacement (AVR)	None	BAV, mechanical AVR	Balloon angioplasty	VSD, reoperation patch angioplasty and stenting
Symptoms	Haemoptysis	Dyspnoea	Collaps and haemoptysis	Fatigue and chest pain	None	None
Hypertension (>140 mmHg)	Yes	Yes	No	No	No	Yes
Systolic and/or diastolic left ventricular function	Good systolic function	Good systolic function, diastolic dysfunction	Good systolic function	Good systolic function	Good systolic and diastolic function	Good systolic and diastolic function

Continued

Continued

Patient number	1	2	3	4	5	6
Diagnostics	Computed tomographic angiography (CTA)	CTA	CTA	CTA	CTA	CTA
Dimensions of aneurysm	40 mm	9 mm	46 mm	16 mm	21 mm	19 mm
Setting of surgery	Emergency	Elective	Emergency	Elective	Elective	Elective
Type of repair	Interposition graft left carotid artery (LCA) to thoracic level (Th) 7 with separate graft to LSA	New interposition graft LSA to descending aorta	Interposition graft distal of lusorian artery to Th6	Interposition graft LCA to descending aorta with separate graft to LSA	Interposition graft LSA to Th5	Interposition graft LCA to Th7 with separate graft to LSA
Duration of hospital admission (days)	18	10	10	14	5	23
Peri- and postoperative complications	None	Hoarseness	None	Hoarseness	None	Respiratory insufficiency and posterior ischaemic optic neuropathy

Clinical series

Six patients were referred to our academic center with a false aneurysm of the aortic repair site. All patients had undergone surgical repair of an aortic coarctation more than 30 years earlier.

Patient 1

The first patient is a 49-year-old male with Alagille syndrome with a coarctation of the aorta that was surgically bypassed at 19 years of age. Concomitant cardiac lesions or defects included VSD and pulmonary artery stenosis. His medical history also included hypertension and chronic renal disease (as part of the Alagille syndrome). Current medication was candesartan 16 mg once daily. He was admitted to the emergency room because of haemoptysis. At presentation, he was hypertensive. Physical examination revealed no cardiac soufflé but crackles were heard with pulmonary auscultation. Peripheral pulsations were strong and there were no signs of mottled skin. The acute presentation, haemoptysis, and his medical history made an aortic problem very likely. Therefore, a computed tomographic angiography (CTA) was performed, which revealed a 40 mm big false aneurysm at the distal anastomosis site without an active blush (see [Figure 1A–C](#)). Laboratory studies showed a haemoglobin (Hb) level of 7.3 mmol/L (normal range 8.4–10.8), creatinine of 170 µmol/L (normal range 60–110), modification of diet in renal disease-glomerular filtration rate (MDRD-GFR) of 37 (normal >60), and lactate (arterial blood sample) of 1.6 mmol/L (normal range 0.8–2.1).

The patient was directly brought to the operation theatre, where a left posterolateral thoracotomy was carried out. Using deep hypothermic circulatory arrest (DHCA), the false aneurysm was resected and an aortic interposition graft was placed from the common carotid artery to thoracic level (Th) 7 with a separate interposition graft to the subclavian artery ([Figure 1C](#)). Subsequently, a lobectomy of the left upper lobe was deemed necessary, because bleeding from the false aneurysm into the lung had completely destroyed the lung tissue. The patient fully recovered and was discharged 18 days after surgery. At discharge, he was on aspirin 80 mg once daily, amlodipine 10 mg once daily, lisinopril 5 mg once daily, and metoprolol 200 mg with modified release once daily. In the first year of follow-up, the performed CTAs showed no new false aneurysm formation and the maximal diameter of the aorta was 31 mm. Further follow-up was performed by the referring hospital.

Patient 2

The second case is a 63-year-old woman with a coarctation of the aorta that was replaced with an interposition graft at the age of 16 years and a mechanical aortic valve replacement at the age of 57 because of bicuspid aortic valve stenosis. Her medical history included hypertension and paroxysmal atrial fibrillation. She attended the outpatient clinic with complaints of dyspnoea and hypertension despite antihypertensive medication (quinapril 20 mg once daily, sotalol 40 mg twice daily, and phenprocoumon (dosage based on frequent international normalized ratio (INR) measurements)). Physical examination was normal and no difference was detected in left/right blood pressure. Laboratory

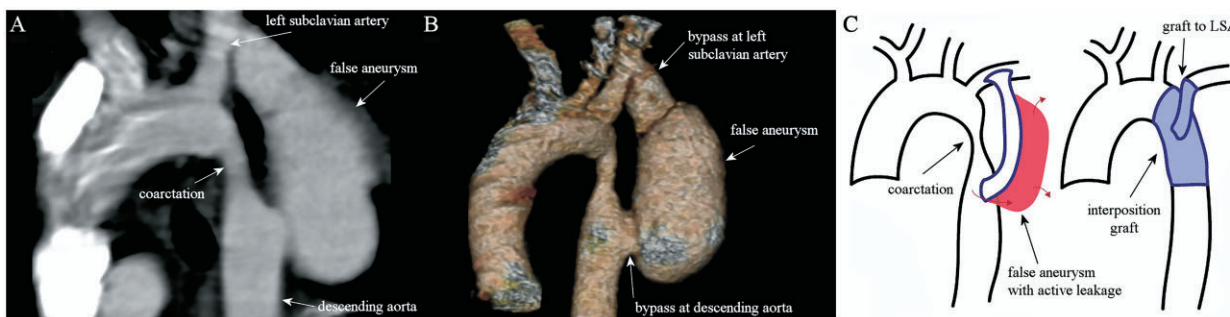


Figure 1 Illustration of the first patient with a false aneurysm after correction of a coarctation of the aorta using a bypass more than 30 years ago. (A) The computed tomography scan shows a false aneurysm of the bypass. (B) The three-dimensional reconstruction is shown. (C) A schematic drawing of preoperative situation (left) with the false aneurysm (shaded in red) and postoperative result (right). LSA, left subclavian artery.

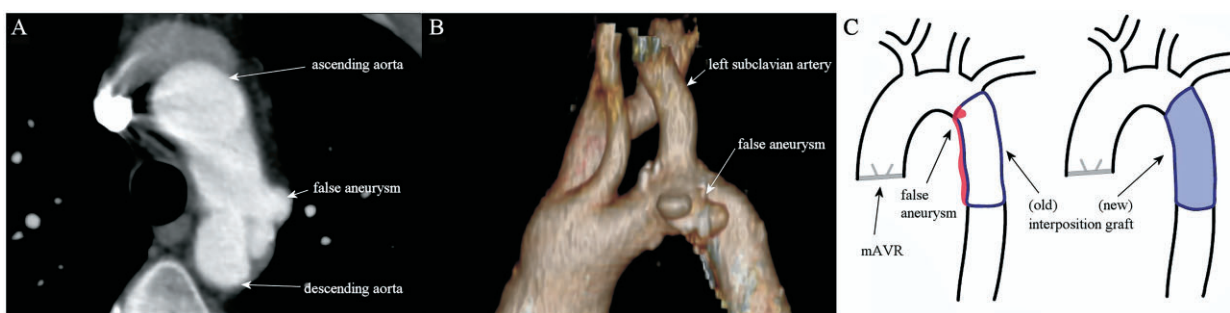


Figure 2 Illustration of the second patient with a false aneurysm after correction of a coarctation of the aorta using an interposition graft more than 30 years ago. (A) The computed tomography scan shows a false aneurysm of the interposition graft. (B) The three-dimensional reconstruction is shown. (C) A schematic drawing of preoperative situation (left) with the false aneurysm (shaded in red) and postoperative result (right). mAVR, mechanical aortic valve replacement.

examination showed a Hb of 8.0 mmol/L, MDRD-GFR >90, and a preoperative INR of 1.6 (reference to operate INR <2.0). Cardiac ultrasound showed no abnormalities. As her complaints could not be explained and she had hypertension with aortic disease in the past, a CTA was performed, which revealed a false aneurysm of 9 mm at the proximal anastomosis (Figure 2A–C).

An elective surgical repair was scheduled. The aorta was approached through a left posterolateral thoracotomy with the use of a left-heart bypass. The repair involved replacing the old graft with an interposition graft from the left subclavian artery to the descending aorta (Figure 2C). The patient's postoperative course was marked by hoarseness due to left-sided recurrent nerve injury. She was discharged 10 days after surgery whilst on acenocoumarol (dosage based on frequent INR measurements) and the same antihypertensive drugs as preoperative with addition of hydrochlorothiazide 12.5 mg once daily. After vocal cord augmentation her voice fully recovered. She had one follow-up CTA 3 months after surgery that revealed no new false aneurysm. All further follow-up scans were performed in the referring hospital, and there have been no further complications.

Patient 3

The third patient is a 60-year-old man with coarctation of the aorta that was surgically replaced with an interposition graft at the age of 27. Further medical history only included hypertension. He had collapsed after physical exercise and had haemoptysis. At presentation, he was pale, sweaty, and experiencing intense pain. His hypertension was adequately treated with oral medication at the outpatient clinic. The types of antihypertensive drugs he was on and their respective dosage were unknown in the emergency setting. He was not on any anticoagulant therapy. Physical examination revealed crackles with pulmonary auscultation and normal heart murmurs. In this hypotensive setting, it was impossible to measure oxygen saturation levels because of the poor peripheral blood flow (shock status). Laboratory results showed low levels of Hb (5.1 mmol/L), elevated levels of D-dimer (6080, normal <500 ng/mL), creatinine (100 μmol/L), and lactate (arterial blood sample) of 9.9 mmol/L. A CTA was performed to find the focus of bleeding or shock. It revealed a saccular aneurysm of the graft with active leakage to the lung and a hemothorax and coincidentally an aberrant right subclavian artery (lusoria) without dilatation (Figure 3A–C).

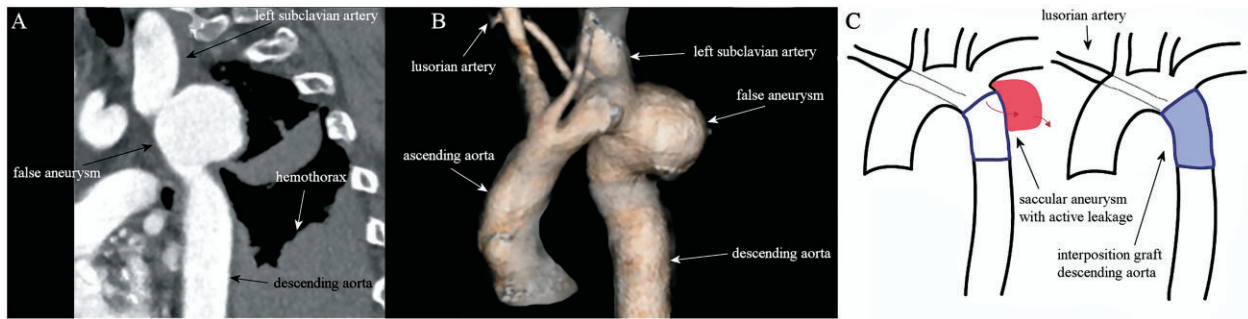


Figure 3 Illustration of the third patient with a false aneurysm after correction of a coarctation of the aorta using interposition graft more than 30 years ago. (A) The computed tomography scan shows a false aneurysm of the interposition graft. (B) The three-dimensional reconstruction is shown. (C) A schematic drawing of preoperative situation (left) with the false aneurysm (shaded in red) and postoperative result (right).

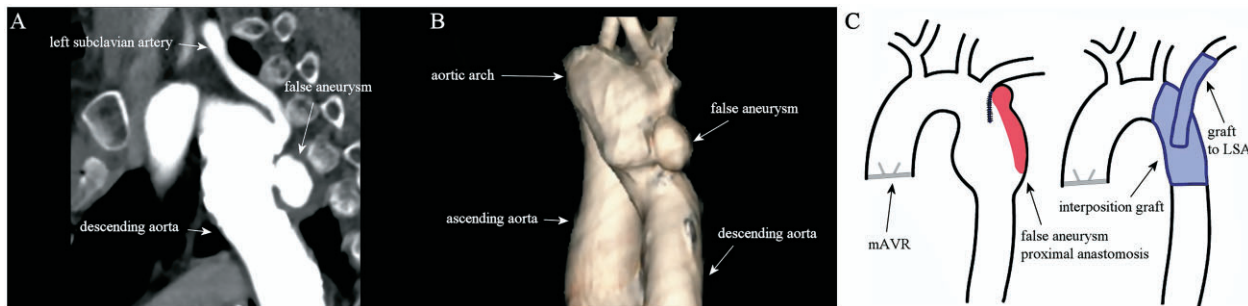


Figure 4 Illustration of the fourth patient with a false aneurysm after correction of a coarctation of the aorta using subclavian flap technique more than 30 years ago. (A) The computed tomography scan shows a false aneurysm of the repair site. (B) The three-dimensional reconstruction is shown. (C) A schematic drawing of preoperative situation (left) with the false aneurysm (shaded in red) and postoperative result (right). LSA, left subclavian artery; mAVR, mechanical aortic valve replacement.

The patient underwent an emergency operation that involved a left posterolateral thoracotomy with the use of extracorporeal circulation (ECC). An interposition graft was placed just distal of the lusorian artery to level of Th6 (Figure 3C). The patient recovered well and was discharged on the 10th postoperative day only receiving labetalol 400 mg three times a day as antihypertensive medication. Computed tomographic angiography was performed during follow-up 1 month after surgery and has continued on an annual basis. Four years after surgery, the follow-up scan interval was prolonged and transferred to the cardiologist as no complications had arisen and the aortic diameter remained stable (maximally 34 mm).

Patient 4

The fourth case is a 64-year-old female with a surgical subclavian flap technique repair of a coarctation of the aorta at the age of 21. When 48 years old, her severely stenotic bicuspid aortic valve had been replaced. Her medical history also included a stroke from which she had fully recovered and chronic obstructive pulmonary disease GOLD II. At the time of presentation, she was on phenprocoumon (dosage based on frequent INR measurements), ramipril 10 mg once

daily, diltiazem 300 mg with modified release once daily and several pulmonary inhalers. She presented with complaints of fatigue and atypical chest pain. The patient was normotensive (systolic blood pressure <140 mmHg). A clear click of the mechanical aortic valve was heard upon a further normal physical examination. Laboratory results were also normal (Hb 8.9 mmol/L; MDRD-GFR 65; sodium 137 mmol/L (Na, normal range 135–145), and potassium 4.9 mmol/L (K, normal range 3.5–4.7)). On account of her symptoms and normal physical and laboratory examination, a CTA was expedited which showed a false aneurysm of the proximal anastomosis (Figure 4A–C). Compared to her previous computed tomography (CT) scans, performed at a 6-month interval after the first appearance of the aneurysm and extended to every 2 years after stable diameters, the false aneurysm grew in size (from 9 to 16 mm). Pulmonary preoperative function testing showed a forced expiratory volume in one second of 1.32 L (65% of predicted) and a Tiffeneau of 51%.

Through a left thoracotomy and with the support of left-heart bypass, an interposition graft was placed between the left carotid artery and the descending aorta with a separate branch to the left subclavian artery (Figure 4C). The patient developed persistent hoarseness,

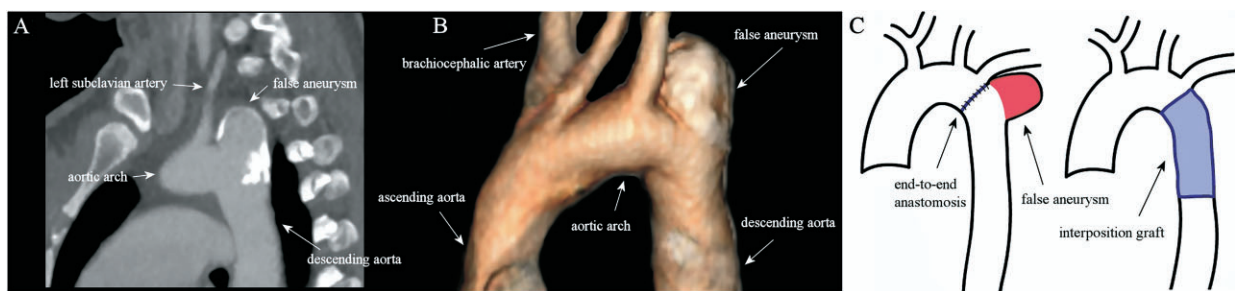


Figure 5 Illustration of the fifth patient with a false aneurysm after correction of a coarctation of the aorta using end-to-end anastomosis more than 30 years ago. (A) The computed tomography scan shows a false aneurysm of the anastomosis. (B) The three-dimensional reconstruction is shown. (C) A schematic drawing of preoperative situation (left) with the false aneurysm (shaded in red) and postoperative result (right).

probably caused by perioperative recurrent laryngeal nerve injury. After 2 weeks, the patient was discharged. Her voice improved after vocal cord augmentation. She was on acenocoumarol (dosage based on frequent INR measurements), lisinopril 5 mg once daily, metoprolol 100 mg with modified release once daily and her own pulmonary inhalers. Follow-up (at 6 months and 1 year) showed no postoperative complications.

Patient 5

The fifth case is a 34-year-old female with an end-to-end anastomosis repair of her coarctation when she was only 2 months old. Because of a re-coarctation, she had undergone balloon angioplasty at the age of 13. There was no further past medical history. At the age of 24 and even though she had been closely monitored and her blood pressure adequately controlled, a CTA revealed a false aneurysm of the proximal anastomosis of 21 mm, which was treated conservatively outweighing all risks (Figure 5A–C). Ten years later, with the surgeons experience of all previous patients including emergency settings, it was decided surgery outweighed all other risks and improved prognosis even in respect to stable diameters. Throughout this period her blood pressure was adequately controlled (systolic <140 mmHg) without need for medication. Preoperative laboratory examination was normal (Hb 8.0 mmol/L; MDRD-GFR >90).

In an elective setting, the end-to-end anastomosis site was replaced by an interposition graft from the left subclavian artery to Th5 (Figure 5C). This procedure was done through a left posterolateral thoracotomy with the use of a left–heart bypass. She recovered extremely well and was discharged 5 days after surgery. She only was on aspirin 80 mg once daily. One year postoperatively, the performed CTA showed no signs of complications and the aorta had a maximal diameter of 32 mm. For further follow-up, magnetic resonance angiography (MRA) will be used.

Patient 6

The sixth and final case describes a 30-year-old man with Klinefelter syndrome whose VSD was repaired with a pericardial patch when he was 9 days old, followed by a surgical patch angioplasty repair of his coarctation. He had undergone a balloon angioplasty and re-patch angioplasty at the ages of 4 and 5, respectively, but a severe stenosis was still left at the proximal site. The patient developed hypertension

during adolescence (>140 mmHg systolic blood pressure at outpatient clinics). He also had complaints of dyspnoea on exertion and during his physical examination a soufflé was heard on the back with a left/right blood pressure difference of >20 mmHg, so it was decided that a CTA should be performed. This showed a false aneurysm with a diameter of 19 mm at the distal anastomosis of the previous patch angioplasty (Figure 6A–C). Preoperative laboratory testing was normal (Hb 9.2 mmol/L; MDRD-GFR 86).

A re-re-operation was performed with left posterolateral thoracotomy. With the use of ECC, DHCA, and antegrade selective cerebral perfusion, an interposition graft was placed distal from the left carotid artery to Th7 (Figure 6C). A separate graft was anastomosed to the left subclavian artery. The postoperative course was complicated by respiratory insufficiency and a posterior ischaemic optic neuropathy. Despite this complicated course, the patient was discharged 23 days after surgery and his vision fully recovered during follow-up. His medication at discharge included clopidogrel 75 mg once daily, amlodipine 10 mg once daily, atenolol 100 mg once daily, and lisinopril 10 mg once daily. Three months after surgery, a CTA showed no postoperative complications and a maximal aortic diameter of 25 mm.

Discussion

False aneurysm formation of the aortic repair site is not uncommon following aortic coarctation repair as dilatation of the repair site is seen in about 13% of the cases and aneurysm formation in about 9%.⁷ The overall survival of patients after aortic repair for coarctation is lower than the general population, and overall mortality is further increased in case of any secondary aortic intervention compared to initial aortic surgery.¹¹ However, early detection and management of the complications, especially but not limited to a false aneurysm, could lead to a better survival rate.⁷ This process would necessitate long-term follow-up, including serial imaging of the aorta (CTA or MRA). In that regard, MRA is preferred, especially in younger patients, because their relatively lower age results in a longer follow-up period which if CTA was used would lead to a higher cumulative radiation load.

There are several risk factors for developing false aneurysms of which previous aortic surgery is the most important.¹⁶ Even though this risk increases in the case of aortoplasty, coarctation repair does

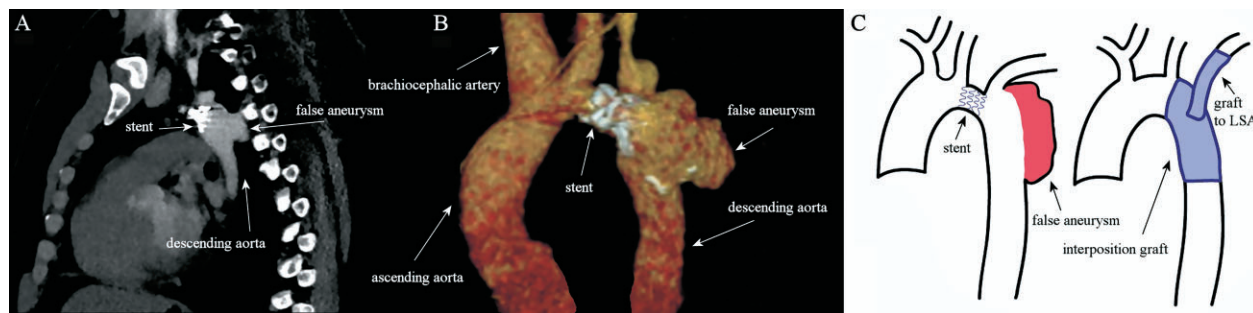


Figure 6 Illustration of the sixth patient with a false aneurysm after correction of a coarctation of the aorta using patch angioplasty more than 30 years ago. (A) The computed tomography scan shows a false aneurysm of the angioplasty. (B) The three-dimensional reconstruction is shown. (C) A schematic drawing of preoperative situation (left) with the false aneurysm (shaded in red) and postoperative result (right). LSA, left subclavian artery.

not seem to be a significant risk factor.^{17–20} This case series illustrates that it can occur regardless of the technique used (interposition graft, end-to-end, subclavian flap technique, or aortaplasty). Therefore, we recommend that all patients undergoing surgery for a coarctation of the aorta require life-long follow-up irrespective of the type of repair. It is advisable to remain cautious for the different types of complications that occur with each type of repair. This point is also clarified in the European Society of Cardiology (ESC) guidelines which caution that aneurysms, at the repair site in patch repairs or false aneurysms in interposition graft repairs, are a risk of rupture and death and so patients should be imaged regularly.⁵

Both the American and European guidelines recommend that adult patients with aortic repair be followed yearly and that any cardiovascular risk factors, including hypertension, be managed aggressively. The American guideline on aortic diseases recommends different intervals for particular pathologies.²¹ They state that ‘the frequency of surveillance imaging is not clear as there are no data to accurately dictate surveillance intervals’. They thus advise that images be obtained at 1, 3, 6, and 12 months postoperatively and then annually for repairs of an aneurysm of the aorta. The European (ESC) guideline recommends starting with a 3- to 6-month interval and thereafter yearly. If stable disease is seen, the follow-up intervals can then be extended. The ESC guideline also states that all adult patients with aortic disease, regardless of initial therapy, require life-long follow-up.²²

The recently published ESC guideline on the management of congenital heart disease in the adult patient recommends yearly follow-up and advises having imaging intervals (preferable cardiac magnetic resonance imaging (MRI)) of 3–5 years.⁵ However, the exact interval depends on baseline pathology. The American guideline on congenital heart disease also recommends yearly follow-up, but they give no specific time interval for imaging.²³ The repair site should be evaluated every 5 years or less, depending on signs and symptoms at first presentation, using CT scan or MRI in adulthood.

The necessity of life-long follow-up, as recommended by the American and European guidelines for both aortic and congenital heart disease, is underlined here, where our patients were seen to develop false aneurysms over 30 years after the initial surgical intervention. However, no specific time interval can be concluded from our cases.

Recently, Abjigitova *et al.*²⁴ have published a single centre long-term (median follow-up of 27 years) outcome after open surgical repair of aortic coarctation in 90 adult patients. Re-intervention due to restenosis was necessary in four patients (one open and three stenting). However, no false aneurysm formation was seen, which might suggest that further follow-up is unnecessary. On the contrary, our case series clearly shows that a false aneurysm can form even after 30 years. Incidence numbers from this case series should be carefully appreciated as all patients were referred to our centre for surgical intervention.

All previously described studies agree on the necessity of long-term follow-up after aortic surgery (e.g. in the case of coarctation), yet there are still no clear schedules or methods. Our cases emphasize the necessity of life-long follow-up with imaging as potentially lethal complications can occur more than 30 years after the initial operation. Some patients had no complaints at all, which suggests that a clinical follow-up based solely on symptoms is insufficient and that imaging is a necessity. Time intervals between follow-up should slowly increase as the time since the initial surgery passes. However, we need to bear in mind that complications can occur at any time.

The appearance of a false aneurysm remains a rare complication after aortic coarctation repair. Other (more common) complications that could occur in a symptomless fashion include re-coarctation, endocarditis, and dissection.¹⁵ Moreover, patients with coarctation usually have a greater risk of developing other cardiovascular comorbidities that may also require potentially life-long follow-up, such as aortic valve stenosis or regurgitation (in the context of bicuspid aortic valve), coronary artery disease, cerebrovascular accidents, and systemic hypertension.¹⁵ These other cardiovascular events were also seen in our cases, as some had had prior aortic valve replacements due to bicuspid aortic valve stenosis (Cases 2 and 4) or multiple interventions regarding (re)coarctation (Cases 5 and 6).

Conclusion

Our case series shows that a false aneurysm can occur more than 30 years after aortic repair for coarctation. The symptoms or risk factors are insufficient alone to distinguish patients who require longer

or stricter follow-up. Therefore, we advocate life-long follow-up after aortic surgery with imaging modalities as CTA or MRA. This follow-up should not focus solely on the surgical site but should also include other complications or comorbidities seen after coarctation.

Lead author biography



Dr Tim Somers completed his medical training at Radboud University Medical Center, Nijmegen, The Netherlands. Dr Somers has special interest in aortic pathology and surgical approaches. He has been working under supervision of Prof. Morshuis. Recently, he spent time at the Oxford University, at the Department of Cardiac Surgery. Currently, he is working on his PhD regarding cardioplegia and cardio protection.

Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as [Supplementary data](#).

Consent: The authors confirm that written consent for submission and publication of this case series including images and associated text has been obtained from the patients in line with COPE guidance.

Conflict of interest: There are no conflicts of interest to declare.

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