New-generation stent grafts for endovascular management of thoracic pseudoaneurysms after aortic coarctation repair

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Objective: Late thoracic aortic aneurysms develop in 5% to 12% of patients having undergone open repair for coarctation of the aorta (CoA). We report our early results for thoracic endovascular aortic repair for pseudoaneu-rysms after CoA repair.

Methods: From 2008 to 2013, data regarding demographics, aneurysm morphology, procedure, and follow-up were collected prospectively on all patients treated for pseudoaneurysms after CoA repair. Retrospective analysis of identified patients was then performed.

Results: Thirteen patients (six men, seven women) were treated. Patients were a median age, 45 years (interquartile range [IQR], 39-56; range, 27-66 years, and the median time after CoA repair to aneurysm treatment was 34 years (IQR, 24-40 years). All patients had saccular pseudoaneurysms of the aortic arch, with a median aneurysm size of 4.1 cm (IQR, 3.4-5.1 cm). The left subclavian artery (LSCA) was involved in 10 patients and was occluded at presentation in three. Four patients had concurrent LSCA revascularization with carotid-subclavian bypass, one had aortic arch hybrid repair, and the LSCA was intentionally covered in two patients. Patients underwent thoracic endovascular aortic repair using the conformable TAG (6 of 13; W. L. Gore & Associates, Flagstaff, Ariz), Valiant device (4 of 13; Medtronic, Minneapolis, Minn), and a custom-made Relay endograft with LSCA scallop (4 of 13; Bolton Medical, Barcelona, Spain). Technical success was 100%, with satisfactory deployment of the stent grafts in all patients. There was no 30-day mortality, stroke, or paraplegia. Median follow-up was 15 months (IQR, 9-19 months; range, 1-67 months). Two type II endoleaks from an intercostal artery were managed conservatively, and one type Ib endoleak was treated successfully with distal stent extension.

Conclusions: In this cohort, new-generation stent grafts have good early clinical and radiologic outcomes, avoiding the need for redo open surgery. Management of the LSCA can be tailored to individual patients with new stent graft technology. Long-term follow-up of these patients is crucial to understanding whether endovascular management of this cohort is acceptable. (J Vasc Surg 2014;60:330-6.)

Late thoracic pseudoaneurysms develop in 5% to 12% of patients undergoing repair for coarctation of the aorta (CoA), the incidence varying with the method of repair.1-3 We report our early results using new-generation thoracic stent grafts for thoracic endovascular aortic repair (TEVAR) of the aortic arch and descending thoracic pseudoaneurysms after CoA repair.

CoA describes the congenital narrowing of part of the aorta. Narrowing of the isthmus around the insertion of the arterial duct is the most common site of obstruction and leads to thoracic CoA. CoA accounts for ~7% of congenital heart lesions, with an incidence of 0.3 to 0.4/1000 live births.4 In 87% of cases, CoA is associated with one or more cardiac anomalies, most commonly hypoplasia of the aortic arch (49%), ventricular septal defect (49%), patent ductus arteriosus (49%), and bicuspid aortic valve (46%).5 Almost all CoA is now identified and treated in the early years of life.

Open surgical repair was first described in 1945, balloon dilation in 1982, and stent implantation in the 1990s.6,8 Surgical repair is usually performed through a left lateral thoracotomy or median sternotomy. In recent years, the most common method of surgical repair is resection and end-to-end anastomosis.8 Other techniques include resection and prosthetic graft replacement, subclavian flap repair (ligation of the distal subclavian artery to use the proximal portion to overlay patch the coarctation segment), and synthetic patch aortoplasty, in which a
patch, usually Dacron (DuPont, Wilmington, Del), is placed across the coarctation segment to relieve the stenosis. The rationale for use of Dacron patch aortoplasty was that avoidance of a circumferential suture line would allow better growth at the repair site, thereby reducing the incidence of restenosis. In recent years, increasing numbers of patients have also been treated by endovascular methods for the management of primary and recurrent coarctation.

Multiple reports appeared describing the development of aortic pseudoaneurysms at the repair site related to all methods of repair. A large study evaluating the long-term results from 891 patients after open CoA repair 1 to 24 years previously showed 48 (5.4%) developed pseudoaneurysms at the site of repair; 89.6% in those undergoing patch aortoplasty, 8.3% after end-to-end anastomosis, and 2.1% with prosthetic graft replacement. Pseudoaneurysm formation is most frequent with patch aortoplasty in several reports (24%-90%), with a spontaneous rupture rate of 31% and rupture-related mortality of 78%-100%, thereby necessitating timely intervention.

As a result of these findings, the patch aortoplasty technique was abandoned at most centers in the late 1980s, but a number of patients are presenting with anastomotic pseudoaneurysms secondary to a previous repair. Late pseudoaneurysm formation is also seen with endovascular techniques, at 43% after balloon angioplasty and 5.4% after stent insertion.

METHODS

The National Research Ethics Service (United Kingdom) confirmed that ethical approval was not required for this study.

Patients. Data were collected using a prospectively maintained in-house database on all patients treated from 2008 to 2013 for pseudoaneurysms after CoA repair at the Imperial Vascular Unit, London. Retrospective analysis of identified patients was then performed. The unit receives direct referrals of these patients from the adult Congenital Heart Disease Centre at the Royal Brompton and Harefield NHS Foundation Trust.

Imaging. Computed tomography (CT) images were acquired using the Brilliance iCT 256-slice scanner (Philips Healthcare, Best, The Netherlands) in accordance with standard clinical protocol with the following parameters: 200 mA, 120 kEV, 1-mm slice thickness, and 1024 x 1024 matrix. Intravenous iodinated contrast was administered in conjunction with image acquisition, and the image series was timed for opacification of the aorta. Advanced software on dedicated Extended Brilliance Workspace 3.5.0.2254 CT work stations (Philips Medical Systems) enables multiplanar three-dimensional image reconstruction, evaluation of aortic diameters measured perpendicular to aortic center line, and whole-vessel morphology to facilitate case planning; these aspects are particularly relevant for planning of custom-made scallop endografts. For saccular aneurysms, the aneurysm size is measured across the whole aorta.

Operative technique. We attempt to minimize proportion of aortic coverage to preserve spinal cord function, and therefore, a single stent graft is used where possible. Spinal cord protection with preoperative spinal drain insertion is used only if the patient has undergone previous aortic replacement, such as abdominal aortic aneurysm repair, or if the endograft will cover a significant length of the aorta. Drinkwater et al demonstrated that spinal cord ischemia is more likely to occur at a mean percentage of aortic coverage of 76%, with 270 mm as the minimum length of aortic coverage resulting in spinal cord ischemia.

All cases are performed with the patient under general anesthesia with prophylactic intravenous antibiotics (1 g vancomycin and 750 mg cefuroxime, unless contraindicated) and 5000 U intravenous heparin, with further administration if required to maintain an activated clotting time of 250 to 350 seconds.

Extra-anatomical bypass, followed by stent insertion, is usually performed as a single-stage procedure where necessary to preserve circulation to the left subclavian artery (LSCA) or left common carotid. A proximal scalloped endograft is used when the proximal landing zone is short (<2 cm) or there is significant angulation of the aortic arch and further stent graft apposition to the aortic wall would be beneficial to prevent seal failure, bird-beaking, and migration, without the need for extra-anatomical bypass. Intraoperative transcranial Doppler monitoring of bilateral middle cerebral arteries is performed to monitor cerebral embolization rates during the procedure.

A surgical groin cutdown is performed and a 22F introducer sheath placed in the common femoral artery (CFA). Contralateral percutaneous CFA access is obtained with a 5F sheath for insertion of an imaging catheter. Pharmacological blood pressure manipulation is performed with glyceryl trinitrate infusion to achieve controlled systemic hypotension to a systolic blood pressure of 70 mm Hg to counter the windsock effect and enable precise stent deployment. Balloon molding of the proximal and distal stent and any endograft overlap is performed when required, and arch angiography using a pigtail catheter is done to check graft position and patency of arch vessels. After this, the mean arterial pressure is maintained >80 mm Hg using vasopressors where required to ensure adequate perfusion of the spinal cord.

Patients are monitored postoperatively in the high-dependency unit (HDU) and vasopressors weaned over 24 to 48 hours, with gradual reintroduction of antihypertensive medications. We aim to discharge patients on postoperative day 3, and all patients are discharged with prophylactic aspirin therapy for 6 weeks.

RESULTS

Thirteen patients have been treated to date using endovascular techniques. Patient demographics are detailed in Table. Two patients presented with Ortner’s syndrome: left recurrent laryngeal nerve palsy due to compression by
Coarctation repair technique

Patient comorbidities

Table. Patient demographics

<table>
<thead>
<tr>
<th>Variables</th>
<th>Value (N = 13)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Female sex</td>
<td>7 (54)</td>
</tr>
<tr>
<td>Age at time of pseudoaneurysm repair, years</td>
<td>45 (39-56)</td>
</tr>
<tr>
<td>Range, years</td>
<td>27-66</td>
</tr>
<tr>
<td>Pseudoaneurysm size, cm</td>
<td>4.1 (3.4-5.1)</td>
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Aneurysm characteristics

Saccular distal aortic arch pseudoaneurysm

Involving origin of LSCA

No patent LSCA present due to use in CoA subclavian flap repair

Just distal to LSCA

Patient comorbidities

Hypertension

Hypercholesterolemia

Multiple sclerosis, in remission

Aortic valve with or without root replacement

Bicuspid aortic valve

Patent ductus arteriosus ligation

Turner syndrome

Epilepsy

Diabetes mellitus

Pulmonary stenosis

Severe left ventricular impairment with cardiac pacemaker

Revision surgery for CoA

Previous surgical management of post-CoA repair pseudoaneurysm

Coarctation repair technique

Dacron patch plasty

Subclavian flap repair

End-to-anastomosis bypass

Extra-anatomical bypass

Age at time of CoA repair, years | 7 (1-19) |

Time to treatment of aneurysm after CoA repair, years | 34 (24-40) |

CoA, Coarctation of the aorta; LSCA, left subclavian artery.

aContinuous data are shown as median (interquartile range), unless indicated otherwise, and categoric data are shown as number (%) or number.

bIncludes 1 patient with Dacron (DuPont, Wilmington, Del) patch (age 18 years) then revision with interposition graft for pseudoaneurysm (age 36 years) and 1 patient with subclavian flap repair (age 9 weeks), followed by revision Dacron patchplasty (age 9 months).

a Distal aortic arch aneurysm measuring 6.7 cm in one patient and 6 cm in the other. The remaining 11 patients were asymptomatic, with the pseudoaneurysm detected after CoA repair surveillance imaging. Four patients had concurrent LSCA revascularization with carotid-subclavian bypass, and one patient had an aortic arch hybrid procedure with carotid-carotid crossover and carotid-subclavian bypass. The LSCA was intentionally covered in two patients with a nondominant left vertebral artery; one with significant comorbidity, including severe left ventricular impairment and cardiac pacemaker, and the other was the first patient treated as part of this series in 2008.

Patients underwent TEVAR using the conformable TAG device (6 of 13; W. L. Gore and Associates, Flagstaff, Ariz), the Valiant device (4 of 13; Medtronic Vascular, Minneapolis, Minn), and a custom-made Relay endograft with LSCA scallop (3 of 13; Bolton Medical, Barcelona, Spain). The proximal landing zone was zone 2 of the thoracic aorta in 12 of 13 and zone 1 in one patient.

Access was through a CFA surgical cutdown in 12 of 13 patients, with an iliac conduit required in one patient. Five patients required additional percutaneous access through the left brachial artery to obtain angiographic images in the three patients with scallop device insertion to ensure accurate orientation of the scallop with respect to the LSCA and in two patients for embolization of the covered LSCA with an Amplatzer plug (St. Jude Medical, Minnesota, Minn). Completion angiograms revealed two type Ia endoleaks and one type Ib endoleak, all successfully excluded by stent extension in two patients and balloon molding in one patient.

One stent graft was used in nine of 13 patients, with median length of stent 130 mm (interquartile range, 100-150 mm). In patients requiring more than one stent, the length of aortic coverage was measured along a central luminal line on a postoperative CT aortogram (CTA). Three patients required two stents each, with aortic coverage lengths of 93, 100, and 126 mm. In one patient, three stents were used due to an angulated arch and size discrepancy between the proximal and distal landing zones, where a type Ib endoleak resulted after deployment of the second stent, and therefore requiring a third stent for distal extension. The resultant aortic coverage length was 191 mm. No patients required spinal drain placement due to the relatively short lengths of aortic coverage.

Technical success was 100%, with satisfactory deployment of the stent grafts in all patients, with no retrograde type A dissection, stent collapse or deformation, stent migration during deployment, or conversion to open procedure. All patients were extubated immediately postoperatively and monitored in the HDU for 24 to 48 hours. There was no 30-day mortality, stroke, or paraplegia. Postoperative complications in one patient each included carotid-to-carotid graft infection requiring revision right-to-left carotid bypass with long saphenous vein, lymph leak from a LSCA revascularization neck wound requiring reexploration, and partial visual field defect due to left retinal artery branch occlusion (single embolus seen on funduscopy).

Median follow-up was 15 months (interquartile range, 9-19 months; range, 1-67 months). If completion angiography is satisfactory, our standard thoracic aortic surveillance protocol includes CTA performed 6 weeks postoperatively. Of the 13 surveillance CTAs performed at this time, 10 showed stable graft position with no evidence of endoleak or graft migration, and no further procedures were required.

Imaging is then repeated at 1 year and at annual intervals thereafter. Our experience with long-term surveillance indicates that once the stent graft is in a stable position with no endoleak at 1 to 3 years, a magnetic resonance aortogram is acceptable thereafter, particularly in this cohort of young patients. Evidence of type II endoleak was seen 6 weeks postoperatively in two patients, so CTA was repeated at 6 months. Both patients had a small persistent type II endoleak from an intercostal artery with decreasing
sac size, which was therefore managed conservatively with annual imaging surveillance. In one patient, a type Ib endoleak was evident on postoperative scanning but was not seen on the completion angiogram. This was successfully treated with distal stent extension 3 months after the primary procedure.

**DISCUSSION**

Nine of 13 patients in our cohort had undergone CoA repair with Dacron patch plasty. In our experience these patients present with two discrete aneurysm morphologies: aneurysmal degeneration involving the Dacron patch or a pseudoaneurysm in the form of a saccular blowout at the anastomotic site (Fig 1). A proposed theory to explain aneurysm formation is that coarctation may represent a diffuse aortopathy because it is associated with subsequent aneurysm formation at the site of previous repair and in contiguous and remote segments of the aorta and branch arteries such as the ascending aorta and LSCA.

Different ways that post-CoA repair anastomotic aneurysms may occur include the synthetic patch repair causing increased wall strain and substantial wall stress opposite the patch, with aneurysm formation resulting from progressive degeneration, pseudoaneurysms resulting from inflammation of the suture line between the aortic wall and the Dacron patch, and degeneration of the Dacron patch fibers themselves. Studies have suggested that saccular aneurysms may be more prone to rupture than fusiform aneurysms of similar diameter and that these post-CoA repair pseudoaneurysms have a particularly malignant course, providing a rationale for the repair of saccular descending thoracic aneurysms at a smaller diameter.

Endovascular stent implantation for management of CoA is gaining popularity because stent therapy has resulted in significant angiographic improvement and gradient reduction of the coarctation with low morbidity and mortality. Forbes et al compared surgical, stent, and balloon angioplasty treatment of CoA in a multicenter study, concluding stents have significantly lower acute complications although they are more likely to require planned reintervention, and 5.4% of stent patients undergoing intermediate imaging follow-up had pseudoaneurysm formation. Prevention of an intimal tear has been proposed as the reason why stent implantation may protect against later aneurysm formation compared with other methods of repair.

A recent Cochrane review concluded there was insufficient evidence to conclude superiority with regards to stent placement vs surgery for the management of primary CoA, and United Kingdom National Institute for Health and Care Excellence (NICE) guidance supported the use of balloon angioplasty with or without stenting for coarctation and recoarctation in adults and children. The literature, however, demonstrates that postrepair pseudoaneurysm formation, along with restenosis, remains a disadvantage of all types of treatment and can occur several years after initial repair. As a result of these findings, and in accordance with the American Heart Association/American College of Cardiology guidelines for the care of adults with congenital heart disease, it is mandatory that all patients undergo regular follow-up and surveillance imaging after CoA repair.

We also treated a 42-year-old man who presented with hemoptysis as a result of an aortobronchial fistula at the site of Dacron patchplasty CoA repair 18 years previously. The fistula involved the origin of the LSCA, and urgent TEVAR was performed with a single Gore TAG stent and coverage of the LSCA origin without revascularization. He has been maintained on long-term oral antibiotic therapy with no signs of graft infection. At 9 years of follow-up, he remains well, with satisfactory stent position and no evidence of endoleak. This case highlights an additional post-CoA repair complication and how it was successfully managed conservatively with annual imaging surveillance.
managed with an endovascular technique in an infective field, with no complications at 9 years of follow-up.

The results presented here compare well with the literature on endovascular management of post-CoA repair pseudoaneurysms, which mainly comprises small case series, including:

- Ince et al\textsuperscript{19} (2003), six patients using Medtronic Talent grafts, no 1-year intervention-related morbidity or mortality;
- Bell et al\textsuperscript{20} (2003), five patients, all aneurysms remain excluded at median follow-up 7 months;
- Preventza et al\textsuperscript{21} (2006), eight pseudoaneurysms with or without restenosis using a combination of Palmaz stents and endoluminal grafts, two reinterventions 1 and 2 years postoperatively for stent migration and endoleak; and
- Marcheix et al\textsuperscript{22} (2007), four patients using Zenith TX2, Cook grafts, no stent-related complications at median follow-up 7.5 months.

These authors all reported results of the endovascular repair of pseudoaneurysms after previous surgical repair of CoA using first- and second-generation thoracic stent grafts, concluding that endovascular repair is safe, effective, and promising, with the potential to avoid repeat open surgery.

Of note, the series by Preventza et al\textsuperscript{21} had two patients and the series by Marcheix et al\textsuperscript{22} had one patient presenting with left arm ischemia after coverage of LSCA, thereby necessitating subsequent LSCA revascularization. In 2012, Roselli et al\textsuperscript{23} reported results for 25 patients undergoing management of post-CoA repair pseudoaneurysms, but results were presented as a mixed cohort undergoing treatment of primary and recurrent CoA and postrepair complications using open, hybrid, and endovascular methods, so no data are available for the specific subgroup of endovascular repair of post-CoA repair pseudoaneurysms. Preventza et al\textsuperscript{2} report the occurrence and treatment of post-CoA repair pseudoaneurysms where they treated 70% of patients with open surgical techniques. They managed 11 patients with TEVAR and four with carotid-subclavian bypass plus TEVAR, but again, specific outcomes for this subgroup were not reported.

Several anatomical challenges exist in the endovascular management of post-CoA pseudoaneurysms, which include angulation of the aortic arch, a size discrepancy between the proximal and distal thoracic aorta, and frequent involvement of the LSCA (Fig 1). Many of these patients have a significant degree of tortuosity of the aortic arch after previous surgery, which can lead to significant difficulty in graft planning and placement. Careful preoperative planning and adequate imaging is essential. The maximization of all available landing zones and accuracy of graft placement is essential given the young age of these patients. The size discrepancy between the proximal and distal landing zone is relevant for graft planning.

First-generation and second-generation thoracic stent grafts have demonstrated good midterm outcomes in general. However, inadequate sealing and stent graft conformation in angulated necks, particularly when indications are extended to more proximal landing zones (such as that required in this post-CoA repair cohort) have led to the development of proximal endoleaks.\textsuperscript{24} New-generation stent grafts, such as the conformable TAG device and Valiant Captivia system (Medtronic Vascular), are particularly useful in these settings because the stents are flexible and conformable for use in an angulated arch and tortuous aorta and resist compression (Fig 2). The Gore device, in particular, has a generous oversizing range, and a single stent graft can treat a range of aortic diameters; for example, the 26-mm-diameter stent graft can be used in an intended aortic diameter of 19.5 mm to 24 mm. In addition, ready-made tapered devices are available with proximal and distal diameters of 26 mm × 21 mm and 31 mm × 26 mm to account for the frequent difference in diameter between the proximal and distal landing zones, especially when addressing smaller aortic diameters.

More recently, we have used the Relay custom-made devices with a scallop for the LSCA (Fig 3). The scalloped device is useful to gain extra proximal fixation distance to

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Fig 2. Angiogram shows (A) insertion and (B) deployment of the conformable TAG device (W. L. Gore and Associates, Flagstaff, Ariz) to exclude a distal arch aneurysm. This patient previously underwent aortic coarctation (CoA) repair with a subclavian flap technique, and therefore, no patent left subclavian artery (LSCA) is present.
increase seal and reduce the risk of long-term stent migra-
ition, without the need for extra-anatomical bypass. These
are especially suitable in young patients, and we also use
these devices for the treatment of other causes of descend-
ing thoracic pseudoaneurysm formation. The stent graft is
custom-made for each patient. A reinforced scalp is posi-
tioned proximally, allowing the stent to be deployed
beyond the origin of the LSCA. Radiopaque markers

Should we be stenting young patients? There are
specific concerns with endovascular intervention in young
patients. Management of the LSCA is important we believe,
particularly in this young cohort, to reduce the risk of posterior
circulation stroke due to dominant vertebral artery coverage
and to reduce paraplegia risk if further intervention is required
for subsequent aneurysmal dilatation, which these patients
have a tendency to develop. Treatment strategies include
extra-anatomical bypass with carotid-subclavian or carotid-
carotid crossover and carotid-subclavian revascularization,
followed by stent deployment. In suitable patients, use of the
Relay custom-made devices with a scallop for the LSCA avoids
extra-anatomical bypass. Appropriate management of the
LSCA provides a stable landing zone, and because there is
undiseased aorta at both the proximal and distal landing
zones, in contrast to patients with dissection or aneurysmal
disease, the risk of stent migration and type I endoleaks as a
result of future aortic dilatation should be minimized (Fig 4).

Although long-term results are awaited to evaluate the
durability of thoracic endovascular repair, particularly in
young patients, this is a similar cohort to those undergoing
TEVAR for management of aortic transection, where
midterm results have been satisfactory. Thirty-one patients
(mean age, 35 years) underwent emergency TEVAR for
traumatic aortic injury, with 100% technical and clinical
success of the procedures during the follow-up period
(mean, 26 months; range, 6-65 months) and follow-up
CTA demonstrating patent endografts with no endoleak or
graft migration.26

These young patients are at reduced risk of neurologic
injury because they have limited atherosclerotic disease,
thereby reducing the risk of intraoperative cerebral embolization, which we monitor intraoperatively using transcranial Doppler. Paraplegia is also reduced because extensive coverage of the thoracic aorta is usually not indicated, and single, short stent grafts are commonly used. In terms of long-term radiation exposure, the use of magnetic resonance imaging for long-term surveillance is advantageous.

CONCLUSIONS
In this cohort, new-generation stent grafts have good early clinical and radiologic outcomes, avoiding the need for redo open surgery. Management of the LSCA can be tailored to individual patients with new stent graft technology. Long-term follow-up of these patients is crucial to understanding whether endovascular management of this cohort is acceptable.

AUTHOR CONTRIBUTIONS
Conception and design: MM, AU, NC, CB
Analysis and interpretation: AP, NR, MH, EK
Data collection: AP, NR
Writing the article: AP, NR, CB
Critical revision of the article: MH, EK, MM, NC, CB
Final approval of the article: AP, NR, MH, EK, MM, AU, NC, CB
Statistical analysis: AP
Obtained funding: NC, CB
Overall responsibility: CB

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