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Role of endovascular repair in the management of late pseudo-aneurysms following open surgery for aortic coarctation

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Abstract

Background: Coarctation of the aorta accounts for almost 5% of all congenital cardiac malformations, and it is usually treated by open surgical procedures. Despite the excellent primary results, many patients may develop anastomotic pseudo-aneurysms, associated with considerable morbidity and mortality rates. We investigated the role of endovascular repair as an alternative to open re-do surgery on the descending aorta. **Methods:** We retrospectively collected data of 11 consecutive patients who presented with pseudo-aneurysms of the descending aorta following previous surgical repair of congenital aortic coarctation. Nine patients developed a pseudo-aneurysm after patch aortoplasty, while an interposition graft repair was performed in two cases. Seven patients underwent an endovascular repair (26.9 years after the previous open surgery). Four patients with a pseudo-aneurysm after coarctation repair associated with arch hypoplasia underwent conventional open re-operation. **Results:** No complications or death were observed in the conventional surgical group. Technical success was obtained in all patients who received a stent-graft procedure. More than one stent graft was necessary only in one case. A left carotid–left subclavian artery bypass was performed in three patients. Type II endoleak was detected by angiography in one patient who had previously undergone subclavian re-vascularisation. The median follow-up (FU) was 44.5 months and was 100% complete. Neither re-operations nor complications or deaths occurred during FU. **Conclusions:** Endovascular repair of pseudo-aneurysms following open surgery for aortic coarctation is feasible and safe. However, more patients and longer follow-up are necessary to assess the efficacy of this promising less-invasive alternative to open re-do surgery. © 2009 European Association for Cardio-Thoracic Surgery. Published by Elsevier B.V. All rights reserved.

Keywords: Aortic coarctation; Pseudo-aneurysm; Descending thoracic aorta; Endovascular stent-graft repair

1. Introduction

Open surgical repair via left posterolateral thoracotomy is still considered the standard procedure for correction of simple aortic coarctation, although balloon angioplasty and stent placement are nowadays considered a suitable option after 3 months of age and in the adult population [1–5]. Different surgical strategies have been used to approach the aortic coarctation, such as subclavian flap angioplasty, patch angioplasty, interposition graft repair and end-to-end anastomosis [6–11]. Despite the primary success, 9% of patients can develop local aneurysms late after coarctation surgery, with an inherent risk of rupture and lethal outcome. Long-term follow-up (FU) is therefore strictly required to prevent complications of late pseudo-aneurysms at the

previous repair site [12–14]. Surgical re-intervention on the descending thoracic aorta, with its related morbidity and mortality rates, may be necessary in these cases. Recently, endovascular stent-graft treatment has been proposed as an alternative to open repair in various diseases of the descending aorta with encouraging outcomes. The aim of this study is to analyse the procedural feasibility, early and midterm results and the clinical outcome of patients undergoing endovascular treatment for aortic pseudo-aneurysm after the open surgical correction of aortic coarctation over a period of 7 years.

2. Materials and methods

2.1. Patients

This retrospective study has been approved by the local Ethics Committee and a signed informed consent has been obtained by all participating patients. From July 1997 to December 2007, 230 patients underwent endovascular repair

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at our institution. Of these, seven patients (3%; four males and three females; mean age: 45.7 ± 13.6 years; range: 29–66 years) received a stent graft for late pseudo-aneurysm following open surgery for adult aortic coarctation in the last 7 years (Fig. 1a and b). In one case, a bicuspid aortic valve was associated with aortic coarctation. Patch aortoplasty was previously performed in six cases (85.7%), while an interposition graft was used in one patient. Conventional open surgery was performed 25.5 ± 8.3 years before endovascular repair of pseudo-aneurysms. In one patient, a re-implantation procedure, sparing a bicuspid aortic valve,

was performed after coarctation repair, 13 months prior to stent-graft treatment. Systemic blood hypertension was detected in four patients (57.1%). Two patients were smokers. Atrial fibrillation was present in two cases, and type II diabetes in one patient. Clinical history of the patients is listed in Table 1.

During the same period, four patients (mean age: 28.2 ± 8.4 years; range: 18–37 years) with pseudo-aneurysm after coarctation repair and concomitant arch hypoplasia, and/or a short proximal neck (distance from the left carotid artery and pseudo-aneurysm) and/or diameter of the left subclavian artery >20 mm, were surgically treated. Aortic arch hypoplasia was defined as an arch diameter ≤ 0.9 times the aortic diameter at the level of the diaphragm. Three patients had undergone patch aortoplasty, and one had a bypass graft.

2.2. Stent-graft procedure and conventional surgery

Routine examination of the heart, lung, liver and kidney functions as well as contrast-enhanced computed tomography (CT), magnetic resonance imaging (MRI) and/or angiography was carried out on all patients. Anatomic conditions allowing endovascular treatment were considered: no aortic wall alteration (thrombus, severe atherosclerosis, intramural haemorrhage) as well as a diameter ≤ 42 mm and ≥ 20 mm at the proximal and distal necks, diameter of the femoral or iliac arteries ≥ 8 mm and no extension of the aortic lesion to the aortic arch. All procedures were performed in the operating room. Patients were under general anaesthesia receiving mechanical ventilation. Blood pressure was monitored by right radial artery cannulation. Ceftriaxone (2 g intravenously) was administered prior to the procedure. The common femoral artery was used for access after surgical exposure in all patients. After exposition of the artery, a 6F sheath was inserted and 2500 UI of heparin administered. In patients with active bleeding into the pleural and/or mediastinal space, no systemic heparin was administered. Angiography and trans-oesophageal echocardiogram (TEE) were performed to identify the lesion, landing zones and its relation to side branches in all patients. Cerebro-spinal fluid drainage was never used in this series of patients. Eight thoracic stent grafts (five Talent and three Valiant Medtronic, Santa Rosa, CA, USA) were loaded on an extra-stiff guide-wire and delivered under fluoroscopic and TEE control with induced hypotension (systolic pressure less than 80 mmHg) to prevent inadvertent downstream displacement of the stent graft during delivery. The proximal end of the endografts was always an uncovered stent (free-flow end). On the basis of CT/MRI measurement, an over-sizing of 10–15% was applied in the choice of stent-graft diameter. Postprocedural angiography and TEE control were performed to reveal the final result. Preoperative imaging and intra-operative data are reported in Table 2.

Conventional open surgery was performed in four patients with an associated arch hypoplasia through a median sternotomy by a single-stage transmediastinal re-repair using moderate hypothermic cardiopulmonary bypass and antegrade selective cerebral perfusion. The left recurrent laryngeal nerve was identified and protected. The pseudo-aneurysm and the proximal descending aorta were exposed

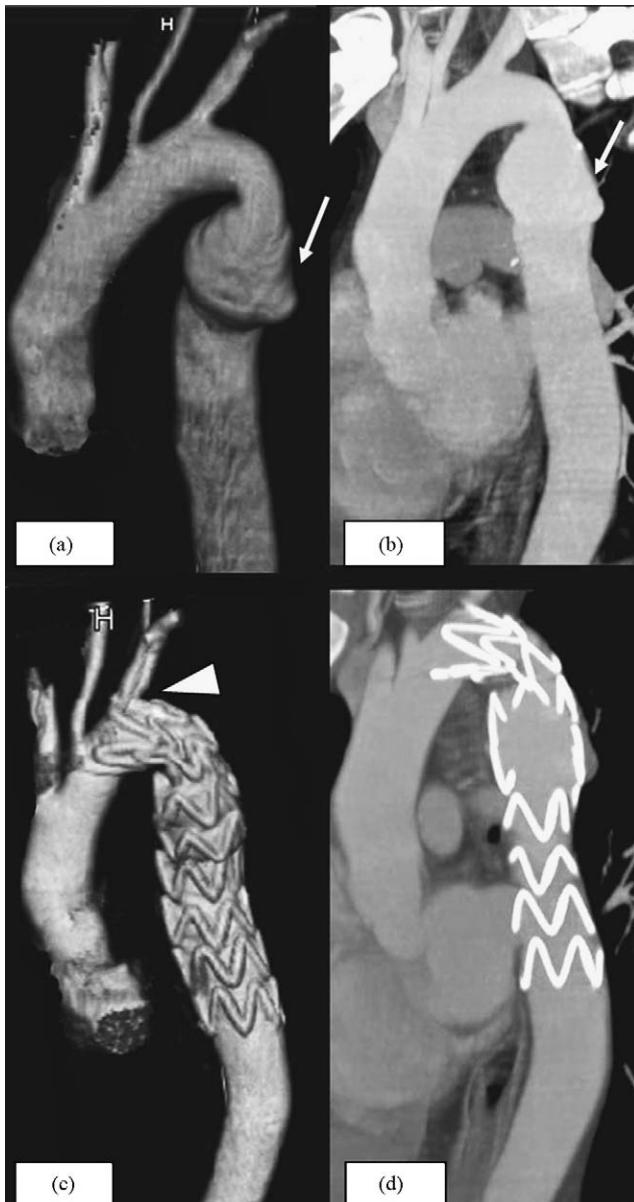


Fig. 1. Multidetector CT volume rendering (a) and MIP images (b) of a pseudo-aneurysm (white arrow) at level of a previous open coarctation repair. Corresponding images [VR (c) and MIP images (d)] show a correct position of the stent-graft and the absence of endoleak three months after the procedure. The uncovered proximal end of the stent-graft allows to land in the distal aortic arch without any obstruction to blood flow in the carotid artery (white arrowhead). CT, computed tomography; VR, volume rendering; MIP, maximum intensity projection.

Table 1
Clinical history of patients who underwent endovascular repair.

Patient	Gender/age at surgical repair	Previous surgical interventions	FU interval after surgery (years)	Dimension of pseudo-aneurysm (mm)	Age at SG implantation	FU interval after SG (months)
1	M/42 years	Patch aortoplasty	26	60	66	51
2	M/17 years	Interposition graft	32	42	48	92
3	F/16 years	Patch aortoplasty	14	45	29	52
4	F/13 years	Patch aortoplasty	28	35	39	44
5	M/22 years	Patch aortoplasty	28	40	50	39
6	F/22 years	Patch aortoplasty	36	37	57	25
7	M/17 years	Patch aortoplasty	15	46	31	2

Values are presented as absolute numbers. SG, stent-graft; FU, follow-up; mm, millimetres.

through the left pleural space with a downward gentle retraction of the left lung. The surgical technique consisted of graft replacement of the involved segments of the aorta in all cases, with re-implantation of the supra-aortic vessels. These vessels were re-implanted using separated graft technique in three patients and *en bloc* technique in one.

2.3. Statistical analysis

All statistical analyses were performed using the SPSS software package (version 14.0; SPSS, Chicago, IL, USA). Incidence rates of events are reported by giving the number of patients experiencing the event followed by the corresponding percentage. Continuous data are reported by giving the mean \pm standard deviation and/or median and the range of values observed. A paired-sample *t*-test was used to compare the mean preoperative and FU aortic diameters.

3. Results and follow-up

All patients were treated in elective conditions. The endovascular procedures were performed 25.5 ± 8.3 years

(range: 13.9–35.6 years) after previous aortic repair. Technical success (stent-graft insertion and deployment) was obtained in all patients. Four patients (57.1%) had an inadequate proximal neck (less than 5 mm from the left subclavian artery (LSA)). The LSA was completely covered without previous re-vascularisation in one patient, while a left carotid to left subclavian bypass (LCSB) with proximal ligation of the LSA was pre-emptively performed in three patients (7, 20 and 89 days before stent-graft treatment). All patients with subclavian re-vascularisation had the LSA origin later covered by the stent graft. Postprocedural angiography detected a type II endoleak in one patient, revealing an incomplete closure of the proximal subclavian stump associated with a slow flow inside the LCSB previously performed. A supra-clavicular re-entry to approach the LSA proved impossible in this patient 14 days later, because of the presence of strong adhesions, and a median sternotomy was necessary to close the proximal segment of the LSA. A bypass between the ascending aorta and the distal part of the vascular prosthesis previously used for the LCSB was performed to re-vascularise the mid- and distal segments of the LSA. Blood loss was limited to less than 200–300 cl in all cases and no patient required transfusions. Only four patients (first four cases in order of time) were transferred to the intensive care unit (ICU) after the procedure. Mean ICU stay was 13.7 h in these patients. Three patients were extubated in the operative theatre and sent directly to the ward. No other stent-related complications or deaths were observed. There were no cases of paraparesis or paraplegia or neurological deficits of the left arm. In-hospital results are summarised in Table 3.

Table 2
Preoperative imaging and intraoperative data of the endovascular group.

Diameter of pseudo-aneurysm (mm) \pm SD	43.6 \pm 8.3
Proximal landing zone diameter (mm) \pm SD	19.4 \pm 1.3
Distal landing zone diameter (mm) \pm SD	22.3 \pm 3.0
Interval between previous open repair and stent-graft procedure	25.5 \pm 8.3
LSA coverage without re-vascularisation	1 (14.3)
Pre-emptive LCSB	3 (42.8)
Interval between LCSB and stent-graft procedure ^a (days)	20 (7–89)
Technical success	7 (100)
Surgical access	
Right common femoral artery	7 (100)
Stent-graft per patient	1 (85.7)/2 (14.3)
>1 stent-graft used	1 (14.3)
Stent-graft length (mm) \pm SD	117.7 \pm 16
Fluoroscopy time (min) \pm SD	19.7 \pm 2.7
Contrast media (ml) \pm SD	114.3 \pm 26.4
Intraoperative use of TEE	7 (100)

Values are presented as mean \pm standard deviation (SD) or number of patients experiencing the event followed by the corresponding percentage in parentheses, unless otherwise noted. LSA, left subclavian artery; LCSB, left subclavian carotid bypass; mm, millimetres; min, minutes; ml, millilitres; TEE, transesophageal echocardiography.

^a Median (range).

Table 3
In-hospital results.

ICU stay (h) ^a	13.7 \pm 7.1
In-hospital stay (days)	7.8 \pm 2.5
Early endoleak (type II)	1 (14.3)
Need for median sternotomy ^b	1 (14.3)
Paraplegia	0
Stroke	0
Renal failure	0
Respiratory insufficiency	0
30-day mortality	0

Values are presented as mean \pm standard deviation (SD) or number of patients experiencing the event followed by the corresponding percentage in parentheses, unless otherwise noted. ICU, intensive care unit.

^a Values obtained on four patients.

^b Patient with type II endoleak required median sternotomy to close the proximal subclavian stump and to re-vascularise the distal vessel (see Section 2).

All patients underwent a strict FU protocol. Clinical examination, CT scan or MRI controls were performed at discharge, and 1, 3, 6 and 12 months after treatment, and every 12 months thereafter. The FU was 100% complete. The median FU period was 44.5 months (range: 3–92 months). A normal patency of the stent grafts and of the LCSB as well as a complete fibrotic remodelling of the aorta with retraction of the aneurysm (shrinkage) was observed in all patients (Fig. 1c and d). A reduction of mean diameters of the involved aortic segments was observed at last FU compared to preoperative mean values: 37.4 ± 9.1 mm vs 43.6 ± 8.3 mm ($P = 0.2$). Neither migration nor twisting of the stent graft nor any leakage was noted. No operative mortality occurred in patients submitted to open surgery. These patients were all transferred to the intensive care unit (ICU). No patient experienced stroke, paraplegia or other major complications.

4. Discussion

If natural history of untreated simple coarctation is poor, with mortality rate of 25%, 50%, 75% and 92% by the age of 20, 32, 46 and 60 years, respectively [1], surgical repair through left posterolateral thoracotomy is nowadays regularly performed in the first days of life with very low mortality. Long-term complications, such as re-coarctation or aneurysm formation, are still reported to be quite frequent, particularly after patch plasty of coarctation as shown by this series of patients. Conservative treatment of aneurysms after surgical coarctation repair remains unpredictable and is associated with a 100% rate of rupture within 15 years in the single-centre experience of Knyshov et al. [13] Consequently, aortic pseudo-aneurysms at the previous repair site have been usually managed by open surgical procedures with the help of cardiopulmonary bypass, hypothermic circulatory arrest or other methods of distal circulatory support [15,16], as our own group did in cases of associated hypoplastic aortic arch [17]. Despite the improvements in surgical techniques and postoperative care, conventional surgery for re-operations on the descending thoracic aorta is still associated with marked morbidity and mortality rates. In their review of 48 patients who developed aneurysms at the site of aortic repair, Knyshov et al. [13] reported an operative mortality of 13.3% and significant percentages of postoperative complications such as paresis of the left recurrent or left phrenic nerve and bleeding requiring re-thoracotomy. Although other groups do not refer to operative mortality [2,18], the incidence of perioperative complications remains high as well as the need for blood products and prolonged hospital stay [13,19]. Recently, endovascular stent-graft treatment has been proposed as an alternative to open repair in various diseases of the descending aorta with encouraging outcomes. The advantages of endovascular techniques could be theoretically even more evident when these procedures are performed in an alternative to open re-do surgery. For this reason, some authors, like us, have investigated the procedural feasibility, early and midterm results and clinical outcome of this minimally invasive approach in cases of pseudo-aneurysms following open repair of aortic coarctation. Our results are comparable with those of other groups [20–22] in terms of morbidity and mortality showing a

substantial durability of these procedures even at a longer FU. Nevertheless, in this small series of patients, we have observed a high incidence of inadequate proximal landing zones for stent grafts. Indeed, considering the immediate vicinity of the pseudo-aneurysm to the left subclavian artery, complete occlusion of the LSA was necessary in four cases (with previous re-vascularisation of the LSA in three patients) out of seven (57.1%). The presence of an uncovered proximal end of the stent graft is of paramount importance in this particular clinical scenario, since it allows to land in the distal aortic arch without any obstruction to the blood flow in the carotid artery. In fact, there is no general consensus regarding how best to handle the cases with proximal thoracic aortic pathology. A number of strategies have been described for managing the left subclavian artery origin during proximal aortic stenting in patients with a short juxta-subclavian neck, including intentional closure, coil embolisation, preoperative or postoperative subclavian-carotid bypass or transposition. The first case of pseudo-aneurysm with short proximal neck we faced in this series of patients was treated with intentional closure of the LSA without re-vascularisation because this patient presented with left vertebral artery hypoplasia. The increasing literature suggesting that selective re-vascularisation after endovascular repair with exclusion of the origin of the LSA may be required more frequently than previously reported [23], and the effective incidence of central and peripheral adverse neurological events after overstenting of the LSA without previous re-vascularisation [24], together with our ongoing experience in this field, induced us to avoid the intentional closure of the LSA except in case of pre-emptive re-vascularisation as in the last three cases of the series with a short proximal neck. In cases of LCSB, proximal ligation of the LSA seems to be mandatory to avoid type II endoleaks. However, at last FU, neither symptoms nor functional deficits were noted in all patients, either with or without subclavian re-vascularisation. However, we admit that the association of endovascular procedures and carotid to subclavian bypass in young patients could be debated, because both operations have no long-term FU regarding integrity and durability. The addition of an open surgical procedure (LCSB) reduces in part the advantages of the minimally invasive repair due to endovascular techniques.

5. Conclusions

Endovascular repair of pseudo-aneurysms following open surgery for aortic coarctation is feasible and safe. The LCSB may be pre-emptively required in cases with a short proximal neck. These procedures are a promising and less-invasive alternative to open re-do surgery even if more patients and longer FU are necessary to assess the efficacy of this therapeutic approach. Conventional surgery remains the treatment of choice in case of inadequate proximal aortic neck for stent graft or associated transverse arch hypoplasia.

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