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# Original articles

## Coarctation repair with prosthetic material: surgical experience with aneurysm formation

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**Key words:**

Aortic aneurysm;  
Coarctation of the aorta;  
Prosthesis.

**Background.** Late aneurysm formation is a common complication after repair of an aortic coarctation with prosthetic material; its incidence varies between 5 and 46%. We reviewed our experience with the management of this complication and propose a radical surgical treatment, which has proved to be free from severe complications; furthermore, we suggest the possibility of a new percutaneous management of this complication.

**Methods.** From September 1974 to November 2002, 195 patients underwent primary repair of an aortic coarctation with prosthetic material (Dacron, polytetrafluoroethylene or heterologous pericardium), with patch aortoplasty as the most common technique. During the follow-up period, reoperation for aneurysm formation was required in 13 asymptomatic patients. The diagnosis was made at angiography in 3 patients and at magnetic resonance imaging in 10. The indication for reoperation was an isthmio-diaphragmatic aortic diameter ratio > 1.5. Aneurysmectomy and tube graft interposition was performed in 12 patients; femoro-femoral cardiopulmonary bypass with a period of deep hypothermic circulatory arrest was carried out in 7 cases while 5 patients were submitted to normothermic atrio-femoral bypass; 1 patient underwent endovascular prosthesis implantation.

**Results.** There were no in-hospital deaths. Three patients experienced postoperative complications: bleeding (n = 1), left phrenic nerve paresis (n = 1), and chylothorax (n = 1). The mean follow-up period was 51.8 ± 46.2 months; all patients were asymptomatic without clinical or instrumental evidence of recurrence.

**Conclusions.** Aneurysm formation after primary repair of an aortic coarctation using prosthetic material is a potentially worrisome late complication and lifelong surveillance of these patients by means of magnetic resonance is mandatory. Surgical management, when indicated, has proved to be a definitive treatment and free from major complications. In highly selected patients, interventional management by percutaneous techniques may provide promising results.

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### Introduction

The surgical strategy for aortic coarctation is greatly influenced both by the anatomy of the lesion and by the age of the patient. While coarctectomy with end-to-end anastomosis is the procedure of choice, this technique may prove to be excessively difficult in the presence of a long segment of hypoplastic aorta or in older children and adult patients, due to the limited mobility of the aorta. Patch graft isthmoplasty, proposed in 1957 by Vosschulte<sup>1</sup>, is an effective alternative approach in such patients. However, despite the excellent early clinical results, long-term follow-up reports of patients who have undergone this procedure have shown a great number of aneurysms at the site of isthmoplasty. The incidence of this complication is reported to range between 5 and 46%<sup>2-10</sup>. The aim of

the present study was to report our experience concerning coarctation repair by patch graft aortoplasty. We endorse the use of a radical surgical approach, which has proved to be free from important complications; furthermore, we explored the possibility of an interventional treatment of post-isthmoplasty aneurysms.

### Methods

From September 1974 to November 2002, 345 patients (mean age 24.0 ± 16.2 years) underwent primary repair of aortic coarctation at our institution. One hundred and fifty patients (43.5%) underwent repair with coarctectomy and end-to-end anastomosis, while the remaining 195 (56.5%) underwent repair with the use of prosthetic material, such as Dacron, polytetrafluoro-

roethylene or heterologous pericardium and form the study population. Among these patients, 133 (38.5%) underwent patch isthmoplasty, 49 (14.2%) received a thoracic aortic replacement with a tubular graft, while an extra-anatomic bypass was performed in 13 patients (3.8%). All patients operated upon for primary repair of aortic coarctation were followed up by means of routine clinical examinations including arterial blood pressure measurements in both the upper and lower limbs, chest X-ray and echocardiography. Cardiac catheterization and angiography was routinely performed until 1990, whereas magnetic resonance imaging (MRI) was the method of choice from 1991 onwards. Among the patients who have been submitted to primary repair using prosthetic material, 13 (6.5%) (8 females and 5 males, mean age at the time of primary repair  $24 \pm 16.2$  years, range 5-54 years) had to be reoperated due to the formation of an aneurysm at the repair site. In this subgroup, the repair was performed by Dacron patch isthmoplasty in 8 patients (61.5%), by isthmic aortic replacement with a tube graft in 3 (23.1%), and by subclavian-aortic bypass in 2 (15.4%). Among the 8 patients treated by patch aortoplasty, the aneurysms developed in the aortic wall opposite to the patch, whereas false aneurysms occurred at the suture line in the other 5 patients. All these patients were asymptomatic with a normal systemic blood pressure and a good quality of life. Routine chest X-ray showed a widening of the mediastinal shadow in all cases. In 3 patients the diagnosis was made at angiography, whereas in the last 10 cases at MRI. The indication for re-operation was, in accordance with the literature<sup>3,11,12</sup>, an isthmic-diaphragmatic aortic diameter ratio  $> 1.5$ . The mean age at the time of reoperation was  $37.8 \pm 13.7$  years (range 14-52 years) while the mean interval between the initial coarctation repair and reoperation was  $170.7 \pm 13.7$  months (range 92-276 months).

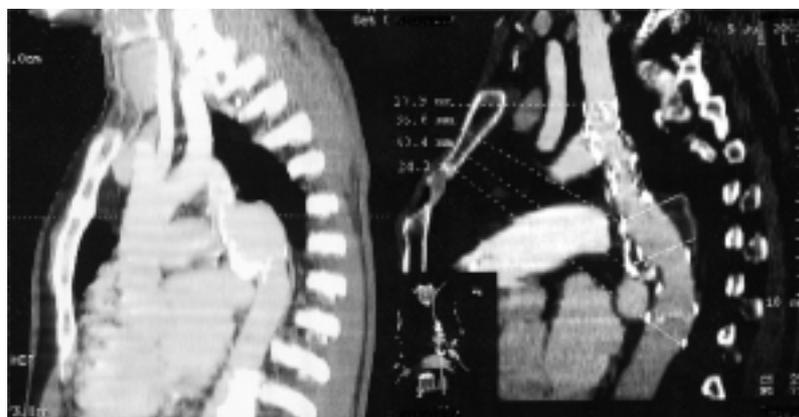
**Operative details.** In 11 patients (84.6%) a fourth intercostal left posterolateral thoracotomy was made and in 1 patient (7.7%) a median sternotomy. One other pa-

tient (7.7%) underwent percutaneous endovascular prosthesis implantation through the femoral artery. Femoro-femoral cardiopulmonary bypass with a period of deep hypothermic circulatory arrest was used in 7 cases (53.8%) since the cross-clamping of the aortic arch was considered unsafe or in case of a high risk of rupture of the aneurysm, whereas a normothermic left atrium to left femoral artery bypass was used in 5 cases (38.5%). In the 12 patients who underwent the conventional surgical approach (92.3%) an aneurysmectomy with the interposition of a tube graft was performed. One patient, also affected by severe bicuspid aortic valve stenosis with dilation of the ascending aorta, underwent one-stage repair, combining aortic valve and ascending aorta replacement with composite graft and tube graft interposition in the descending thoracic aorta. An endovascular prosthesis implantation (Talent, World Medical Manufacturing Corp., Sunrise, FL, USA) was performed in 1 patient (7.7%) in whom a giant false aneurysm had developed in the distal anastomotic site of the tube graft implanted during the primary repair (Fig. 1, Table I).

**Table I.** Operative details of aortic aneurysm repair.

	No. patients
Access	
Left posterolateral thoracotomy	11 (84.6%)
Median sternotomy	1 (7.7%)
Percutaneous endovascular prosthesis implantation	1 (7.7%)
Circulatory assistance	
Standard ECC with period of DHCA	7 (53.8%)
Normothermic atrio-femoral bypass	5 (38.5%)
No assistance	1 (7.7%)
Surgical strategy	
Aneurysmectomy with tube graft interposition	12 (92.3%)
Endovascular prosthesis implantation	1 (7.7%)

DHCA = deep hypothermic circulatory arrest; ECC = extracorporeal circulation.



**Figure 1.** Magnetic resonance images before (left) and after (right) aneurysm repair with an endovascular prosthesis.

## Results

There were no in-hospital deaths. Three patients presented with postoperative complications (23.1%): 1 patient required reoperation for bleeding, 1 patient had a left phrenic nerve paresis while the third presented with postoperative chylothorax. The mean follow-up period was  $51.8 \pm 46.2$  months (range 2-121 months); all patients were asymptomatic without clinical or instrumental evidence of residual complications.

## Discussion

The long follow-up time of our population allowed us to highlight the impact of different surgical techniques, especially with regard to the onset of late complications. In particular, the use of prosthetic material may be associated with an increased risk of late aneurysm formation, a potentially fatal complication. The overall prevalence of the development of a late aneurysm among our patients who underwent coarctation repair at our institution was 3.8%, corresponding to a prevalence of 6.7% among the subgroup of patients operated upon using prosthetic material. In the literature, the incidence of this complication is extremely variable. The prevalence of aneurysm formation varies from 6.8%, as reported by Hehrlein et al.<sup>4</sup> in their study on a series of 263 isthmoplasties performed in younger infants (< 1 year old), to the 28% reported by Clarkson et al.<sup>5</sup>. Moreover, in a series of 891 patients, Knyshov et al.<sup>13</sup> reported an incidence of aortic aneurysms of 89.9% among the patients treated by patch aortoplasty, of 8.3% in the end-to-end anastomosis group, and of 2.1% in the prosthetic graft replacement group.

In our series, the mean time interval between primary repair and the diagnosis of an aneurysm was  $14 \pm 7.4$  years (range 7.6-23 years). The extreme variability of the time interval between primary repair and aneurysm formation has been already described in the literature thus suggesting that lifelong surveillance of such patients is mandatory<sup>4,14</sup>.

The development of an aneurysm has been related to the different elastic properties of the aorta and of the synthetic patch<sup>15,16</sup>, to the wide resection of the intimal ring with consequent fragility of the aortic wall<sup>17</sup> and to the tissue abnormalities associated with aortic coarctation itself<sup>4,18</sup>. Furthermore, because transverse arch hypoplasia is commonly associated with aortic coarctation, a relationship between the flow acceleration and turbulence caused by the hypoplastic arch and the formation of an aortic aneurysm has been suggested<sup>3</sup>. Currently, the precise cause for late aneurysm formation after coarctation repair is still unknown, and a multifactorial pathogenesis cannot be ruled out.

Recent reports<sup>13,19</sup> have emphasized the problem encountered in detecting this complication. It is common experience that clinical examination is frequently

negative and that these patients are often asymptomatic. MRI proved to be the best diagnostic instrument for this kind of aneurysm, and the quality of images so obtained has lessened the importance of echocardiography, chest X-ray, computed tomography and angiography. The possibility of directly acquiring the interested planes (oblique sagittal, oblique coronal, oblique axial) together with the absence of radiation and contrast media, make MRI the gold standard diagnostic procedure for aneurysms developing after primary repair of coarctation. Moreover, MRI has proved to be highly effective even for the diagnosis of recoarctation<sup>20</sup>. Therefore, our experience adds further support to the recent recommendations that MRI be a routine imaging modality for all patients operated upon for coarctation repair<sup>13</sup>. At the beginning of our experience, MRI was not available in our institution. For this reason, the indication for reoperation in the first 3 patients was based on angiography. Since 1990, regardless of the patient's symptomatology or of the results of other instrumental techniques, MRI has been routinely used during the follow-up period of patients who had been submitted to primary repair of aortic coarctation. Our current follow-up policy is to perform MRI 4-5 years after repair in infants operated within 1 year of age or earlier if the operation was performed beyond infancy or with the use of prosthetic material. The indication for reoperation, as described in the literature, is an isthmio-diaphragmatic aortic ratio  $> 1.5^{2,3,21}$ .

Surgery was successfully performed in 12 cases whereas 1 patient was treated by means of percutaneous stent implantation. Resection of the aneurysm with the interposition of a prosthetic conduit was the surgical technique of choice; in 2 patients in whom the primary repair had been accomplished by an extra-anatomic subclavian-aortic bypass, the site of proximal tube graft anastomosis at the time of reoperation was the aortic arch in one case and the partially resected distal portion of the old prosthesis in the other one. The age of the patients who underwent reoperation led us to always use large size prosthetic conduits (14-22 mm), reducing the possibility that the patient outgrows this conduit.

In all the patients who were submitted to surgery, the use of circulatory support avoided major complications; specifically, there was no evidence of paraplegia or of postoperative renal failure.

Interventional management was used in a case in which a false aneurysm developed at the distal end-to-end anastomosis of a tube graft implanted 23 years previously; an endovascular prosthesis was implanted in general anesthesia with percutaneous entry through the femoral artery. It was possible to successfully perform the procedure owing to the presence of a good aortic wall ring between the origin of the subclavian artery and the proximal anastomosis of the graft.

In conclusion, the development of an aneurysm after primary repair of an aortic coarctation using pros-

thetic material is a relevant complication that may manifest even many years after surgery<sup>22</sup>. For this reason, lifelong surveillance of these patients is mandatory and MRI has proved to be the best diagnostic procedure for this purpose. Our current policy is to follow up the patients by means of MRI. Surgical management, when indicated, has proved to be a definitive therapy without major complications. In highly selected patients, the new interventional percutaneous management seems to provide promising results.

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