Primary repair of coarctation of the thoracic aorta by patch graft aortoplasty. A three-decade experience and follow-up in 60 patients

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Abstract Objectives. The present report is a critical review on primary repair of aortic coarctation by patch aortoplasty on the basis of over 30 years surgical experience. Methods. Since 1962, 60 patients (mean age 9.4±4.8 years, range 2–25 years), affected by aortic coarctation, underwent patch aortoplasty repair. During the operation protective guidelines were adopted: additional external Dacron was placed around the repaired site in cases of friable host tissue, the aortic ridge was not excised to leave the posterior aortic wall intact, and the patent ductus arteriosus or ligamentum arteriosum was transected and sutured. Prophylactic measures of neurologic sequelae were: dual pressure monitoring, sequential aortic clamping, surgical shunt or left heart bypass associated with moderate hypothermia when the distal aortic pressure was less than 50 mmHg. Results. No early deaths occurred. The overall survival rate was 92.77±4.04% at 31 years from surgery. Three late deaths occurred. Pressure gradients across the patch ranged between 9 and 20 mmHg. Late aneurysm occurred in one patient (1.3%), 2 years after bacterial endocarditis had developed on a bicuspid aortic valve. Conclusions. Patch aortoplasty is an effective and safe surgical procedure for primary repair of isthmic aortic coarctation when other surgical techniques cannot be performed.

Key words Aortic coarctation • Patch aortoplasty • Recoarctation • Aortic aneurysm

Introduction

Resection of the thoracic coarctation and primary repair by end-to-end anastomosis to restore vascular continuity has been the most common surgical procedure used since its introduction by Crafoord and Nylin [13]. Modifications in this surgical technique were based on the anatomical characteristics of the coarctation. Surgical alternatives for aortic coarctation are numerous and include direct repair techniques as well as the use of supplemental autologous tissue or synthetic materials [8, 13, 14, 25, 29, 47, 49, 50, 52]. However no technique has proven free from complications [5, 9, 11, 16, 20, 23, 30, 32, 33, 35–39, 42, 43, 54].

Patch aortoplasty, first described by Vossschulte [49, 50], was commonly used in many medical Centers until an alarming incidence of late aneurysm formation involving the remaining aortic wall was reported [5]. Although some recent investigations have supported patch aortoplasty [1, 30, 44], this issue remains controversial [9, 44].

The aim of this study was to report over 30 years’ experience on patch aortoplasty for primary repair of coarctation of the thoracic aorta, successfully performed in selected cases at our Institution since 1962.
Material and methods

Patient population

From May 1962 to December 1992, 266 patients underwent primary repair of thoracic coarctation. The Crafoord technique was the standard surgical method used. This report concerns 60 patients of the 266, who underwent the patch aortoplasty technique. The latter was used in children and young adults with both thoracic aortic walls as lumen continuity, in whom, on the basis of aortography or intraprocedural, the extent of coarctectomy might cause tension on the suture line with end-to-end anastomosis. The mean age was 9.4 ± 4.8 years (median 8.5 years, range 2–25 years), with a female predominance of 3:1. Thirty-six (60%) patients had simple isolated aortic coarctation, 24 (40%) patients had associated cardiac malformations. The distribution of patients, with respect to some clinical and operation-related characteristics is presented in Table 1.

Operative technique

All operations were performed through a left posterolateral thoracotomy using the 4th intercostal space. After dissection of the thoracic aorta, the left subclavian artery and two to three intercostal arteries were prepared for patch enlargement. The patent ductus arteriosus or ligamentum arteriosum was transected and sutured. Intercostal arteries were not transected. Temporary loop ligatures were used to control bleeding, when required. A patch of adequate size was sutured into place after the longitudinal incision of the thoracic aorta into the root of the left subclavian artery and below the coarctation. The aortic ridge was not excised to leave the posterior aortic wall intact for further growth, and to avoid damage of the aortic wall. The patch was fixed by continuous 4-0 silk suture in the early years [48], afterwards Prolene monofilament was the suture material used. In the early years synthetic material was used (Dacron, Teflon) [48], which has recently been substituted by reinforced) polytetrafluoroethylene (Gore-Tex). An additional external Dacron graft (cuff) was applied around the repaired site in older patients with friable host aortic tissue in the earlier surgical experience [45]. Total cross-clamping time of the thoracic aorta was reduced to prevent or minimize spinal cord injury by sequential cross-clamping of the thoracic aorta [48], while monitoring the proximal and distal aortic pressures. Somatosensory evoked potentials were not monitored. Percutaneous cannulation was performed in both the right radial and dorsalis pedis arteries. In some cases the left femoral artery was used to record the distal aortic pressure. In patients with poor collaterals (incomplete coarctation, heart failure) and a distal aortic pressure of less than 50 mmHg, a surgical shunt or left heart bypass associated with moderate hypothermia was routinely inserted [31, 55]. The repair of other congenital defects was postponed, particularly in children and infants, with the relief of congestive heart failure symptoms following correction of the coarctation.

Follow-up study

All patients were reviewed in two subsequent follow-ups. The first follow-up concerned 46 patients [48] who were clinical examined once a year. The second follow-up data were obtained during a 3-month period in 1993. The mean follow-up was 20.1 ± 9.2 years, range 1–31 years (Table 1). Recurrence of coarctation was detected at rest by Doppler examination at the coarctation site. Pressure gradients of more than 20 mmHg were considered suggestive of recoarctation. Patient screening to detect thoracic aortic aneurysm was initially performed by plain chest radiography; in the case of cardiac silhouette abnormalities, aortograms were performed to confirm the validity of the surgical procedure and assess a) the morphology of the thoracic aorta and b) pressure gradients across the patch aortoplasty.

Table 1 Distribution of patients according to clinical and operation-related characteristics

<table>
<thead>
<tr>
<th>Clinical and operation-related characteristics</th>
<th>No. of patients</th>
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<td>Aortic valve lesion</td>
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<td>Pulmonary stenosis</td>
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<td>Mitral valve lesion</td>
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<tr>
<td>Aneurysm of membranous septum</td>
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<tr>
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Year of operation

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Duration of follow-up (years)

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Age at operation (years)

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Statistical analysis

All quantitative data are presented as the mean ± SD, unless specified. Survival data were examined using the Kaplan-Meier survival analysis to calculate the cumulative survival at different times after surgery. Patients still alive at follow-up were included in the analysis and considered as censored. The independent effect of age at surgery and sex and of follow-up on pressure gradients detected at the time of the follow-up was assessed by multiple linear regression analysis. A simple linear regression analysis was applied to evaluate the relationship between pressure gradients at follow-up and year of operation and age at surgery. The significance of regression coefficients was assessed by the t-test. The significance level was considered as P<0.05. Statistical analysis was performed by BMDP package [17].

Results

There were no operative deaths. Three late deaths occurred. In one patient no follow-up could be obtained. The over-
Fig. 1 Kaplan-Meier survival curve. Steps correspond to the 3 patients who died 14, 18 and 20 years after primary patch repair. Vertical bars represent 1 standard error.

Fig. 2 The aortogram shows aortic aneurysm at the site of primary patch aortoplasty repair in the patient who had developed endocarditis on a bicuspid aortic valve 2 years previously.

all survival rate was 92.77 ± 4.04% (S.E.) at 31 years from surgery (Fig. 1). Eighteen years postoperatively, one patient developed bacterial endocarditis on a native bicuspid aortic valve, and was treated successfully with specific antibiotic therapy in combination with aortic valve replacement. Two years later, aortic aneurysm at the level of the aortic patch aortoplasty was detected by plain chest radiography and confirmed by aortograms (Fig. 2). The patient underwent resection of the aorta involved in the false aneurysm. Aortic continuity was reestablished with an interposed tubular Dacron graft, using left atrial bypass and moderate hypothermia. The patient died suddenly 6 months after hospital discharge. Acute myocardial infarction was the cause of death in the second patient. In the last patient death occurred following cerebral hemorrhage.

Pressure gradients at rest varied between 9 and 20 mmHg at follow-up with a mean pressure gradient of 14 mmHg. An inverse relation of pressure gradients at rest was found with regard to age at primary repair (multiple linear regression coefficient = -0.1938 ± 0.0792, t = -2.45, P = 0.02; multiple r = 0.33). As no effect was found on pressure gradients due to either duration of the follow-up (multiple linear regression coefficient = -0.0226 ± 0.0421, t = -0.54, P = 0.59), or to sex (multiple linear regression coefficient = -0.4175 ± 0.8555, t = -0.49, P = 0.63), a simple linear regression analysis was applied to pressure gradients and age at operation. The results were similar to those obtained by multiple regression analysis (simple linear regression coefficient = -0.1825 ± 0.0759, t = -2.40, P = 0.02; r = -0.31). Two patients had pressure gradients (9 and 19 mmHg) far from the expected values in relation to age at surgery (2 and 25 years, respectively). When excluding these patients ("outliers") coefficients, although smaller than 0.5 (multiple linear regression coefficient = -0.3595 ± 0.0736, t = -4.89, P < 0.001; multiple r = 0.58, and simple linear regression coefficient = -0.3596 ± 0.0722, t = -4.98, r = -0.57, P < 0.001), an inverse relation between pressure gradients and age at surgery was shown (Fig. 3). Nevertheless no pressure gradient was suggestive of recoarctation.

Discussion

Surgical treatment of coarctation of the thoracic aorta with patch enlargement is still controversial, prosthetic aortoplasty being the most widely reported technique [1, 9, 35,
The subtraction aortography shows normal aortic growth at the site of patch aortoplasty repair (arrow) 10 years after primary patch repair.

Late aortic aneurysm formation

The late outcome of patch aortoplasty may be complicated by true and false aneurysms. A true aneurysm may occur late postoperatively and develops from the aortic wall opposite the patch [2, 5, 24, 25, 37, 50]. False aneurysms (suture line) are usually uninfected and they have etiology and incidence similar to the false femoral aneurysm that occurs at the distal anastomosis of an aortofemoral prosthetic graft [10, 25, 46] and can be mycotic when they occur in the postoperative period [5]. True aneurysms after primary surgical repair of the coarctation have always been associated with a Dacron onlay patch repair technique [11, 16, 19, 25], and were emphasized by Bergdahl and Ljungqvist in 1980 [5]. However, the development of aortic aneurysms after patch aortoplasty is still a major concern for many surgeons [2, 10, 11, 16, 19, 20, 26, 50] because of the increased incidence, from 5% to 38%, occurring in late follow-up [6, 12, 16, 19, 25, 42]. Conversely, studies by Walter et al. [54] and Reul et al. [35] led to opposite conclusions. More recently Ahmadi et al. [1], in a long-term follow-up of 66 patients undergoing patch aortoplasty, correlated the lack of this complication with the retention of the intimal ridge of the aortic wall.

We paid special attention to those surgical techniques able to protect the operation site. First, an external cuff around the site of the repair was introduced to protect the suture line of the thoracic aorta, as previously carried out in patients with isolated patent ductus arteriosus operated on by section/suture technique or in patients who received vascular homografts [45]. This procedure was limited during the first decade of surgical experience and was, of course, performed in the older patients with friable host aortic tissue. Second, as also recommended by other authors [15, 16, 19, 26, 32, 35, 39], the aortic ridge was not excised to avoid wall damage and prevent late aneurysm formation, leaving the posterior aortic wall intact for further growth. Third, the patent ductus arteriosus or ligamentum arteriosum was transected in all patients to eliminate a traction point on this side: in fact, it may contribute to kinking anastomosis of the thoracic aorta and cause tension on the suture line of the patch.

We did not have neurologic damage in our patients. Neurologic sequelae during cross-clamping of the thoracic aorta occur between 0.5 and 3.1% as reported by Brewer et al. [7], who reviewed over 12,532 cases of aortic coarctations operated on the United States. They found 66 cases of paraplegia, with an incidence of 0.41%. To avoid or reduce potential spinal cord damage as a result of ischemia, Hughes and Reemtsma [22] first monitored the distal aortic pressure at the time of cross-clamping and described...
the indication for left heart bypass on low distal aortic pressure. There is no doubt that aortic cross-clamping produces important hemodynamic changes proximal and distal to the coarctation. This is even more enhanced when the subclavian artery is totally occluded during performance of the proximal anastomosis. An additional drop in distal aortic pressure from 10 to 15 mmHg occurs when clamping the aorta below the coarctation, including two to three intercostal arteries. The ligation of collateral vessels (intercos- tal) was associated with a decrease in distal aortic pressure of mean 9.6 mmHg in eight patients [31, 55].

Aortic clamps both above and below the coarctation interrupt left collateral circulation. Subclavian steal syndrome may also develop during this manoeuvre, unless the vertebral artery is ligated [31]. In our experience sequential clamping of the thoracic aorta was used to reduce total cross-clamping time [48]. After proximal anastomosis of the upper half of the patch, the upper aortic clamp was displaced downwards, the homolateral circulation was restored through the unclamped left subclavian artery and a satisfactory perfusion pressure was reestablished in the distal aorta. This manoeuvre allowed us to reduce by half the clamping time of the thoracic aorta during proximal anastomosis. In a few patients with poor collateral, when distal aortic pressure dropped dangerously below 50 mmHg, surgical shunt was instituted.

Mechanical support with left heart bypass and moderate hypothermia was used only in the patient with late aneurysm, for graft interposition. However, spinal cord injury is unpredictable on the basis of the individual response to ischemia and protective techniques able to reduce its potential damage should be used [7, 31, 55]. Hypotension in the distal aorta may explain most of the neurologic complications following hypotensive drug infusion during aortic cross-clamping. Although some authors [27] used nitroprusside to protect the brain, neurologic damage of the spinal cord was demonstrated after infusion of this drug. Moreover, the drop in hypertension in the aortic arch has been shown to be responsible for spinal cord ischemia in some cases, since it reduces blood flow through the right collaterals as well as distal aortic perfusion [31].

Recoarctation

One of the major risks after coarctation repair in infancy is residual or recurrent coarctation, which may be dependent on age as well as on the surgical technique [2, 4, 9, 21, 25, 30, 38, 47]. Recoarctation is probably due to the lack of growth of the suture line or to the inadequate relief of the obstruction, or both [23, 47, 53]. Resection and end-to-end anastomosis in infants and children seems to be the most common cause of recurrent stenosis, due to failure of the circular anastomosis [8, 23]. For the Waldhausen technique, recoarctation rates range between 0% and 22% in recent series [3, 8, 26, 32, 40, 51]. Late results with the Maier-Men-doca technique are lacking [30], while van Heurn et al. [47] reported an actuarial freedom from recoarctation in 151 patients operated on with different techniques. Fifty-seven percent of patients were free from recoarctation at 4 years after subclavian flap angioplasty, 77% after end-to-end anastomosis, 83% after extended end-to-end anastomosis, and 96% after radically extended end-to-end anastomosis [47]. The growth of the aortic wall after patch aortoplasty in our series was evidenced by: 1) subtraction aortography (Fig. 4) and the 2) absence of any pressure gradient suggestive of recoarctation at follow-up. Therefore, even if significant gradients were not found, the inverse relation of pressure gradients with age at operation may indicate that aortoplasty patch repair is not the best surgical procedure in infants and younger children. However there was also no evidence of restenosis in a group of 156 children and adult patients 20 years postoperatively [54]. The recoarctation rate was 5% in 54 patients (15 younger than 1 week, with critical coarctation) followed for 5 years postoperatively [44]. In a review of 53 infants, restenosis occurred in 17.4% of patients younger than 1 year at operation [30]. In 158 patients operated on between 1973-1983, Rostad et al. [38] found that the frequency of moderate and severe recoarctation was higher (23%) in patients operated on with aortic resection and end-to-end anastomosis than in those with patch aortoplasty (6.7%).

Conclusions

Although the management of aortic coarctation has changed over the years, especially in relation to the optimal age for surgical repair, the long-term follow-up of all cases deserves more attention [42]. On the basis of a three-decade experience we conclude that patch aortoplasty for primary repair of thoracic aortic coarctation achieves several goals: 1) growth of the posterior wall opposite the patch, 2) a low incidence of recurrent coarctation, 3) a low mortality rate, and 4) a low incidence of late aneurysm formation when applying specific procedures ( cuff technique, section/suture of the ductus arteriosus or ligamentum arteriosum, retention of the intimal ridge to avoid changes in the aortic wall).

As already pointed out by Campbell and Waldhausen [9], emphasis should be placed on the need for a wide spectrum of validated techniques enabling the experienced surgeon to choose the best surgical treatment.

A three-decade surgical experience on aortic coarctation is a period of observation long enough to lead to definite and positive conclusions. Late aneurysm formation should not be given as a reason to avoid patch angioplasty for primary repair of coarctation of the thoracic aorta when end-to-end anastomosis cannot be performed, for it is an effective technique in the management of this complex disease.

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References


