Repair of isolated aortic coarctation over two decades: impact of surgical approach and associated arch hypoplasia†

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Received 28 September 2011; received in revised form 25 February 2012; accepted 13 March 2012

Abstract

OBJECTIVES: A variety of surgical approaches and techniques are used for isolated coarctation repair. We have retrospectively reviewed our results of isolated repair of coarctation over the last 20 years, to establish whether the approach affects clinical outcome and the need for re-intervention.

METHODS: Two hundred and eighty-eight patients who underwent isolated repair for coarctation of the aorta at Birmingham Children’s Hospital between 1991 and 2010 were enrolled in this study. Chart review and the Departmental database were used to determine demographics, operative details and complications.

RESULTS: The majority of patients (n = 237, 82%) underwent surgical repair via thoracotomy techniques, whereas median sternotomy was used in patients where there was associated arch hypoplasia (n = 51, 18%). For all 288 patients, median age at operation was 24 days (range 0–14 years). Between 1991 and 2000, ten patients (6%) underwent repair through midline sternotomy, increasing to 41 patients (36%) between 2001 and 2010. Overall early mortality was 1% and late mortality was 3%. There was a statistically higher re-intervention rate (16%) in the decade 1991–2000, compared to 5% in the period 2001–10 (P = 0.02). In patients with hypoplastic arch, the midline approach has a lower re-intervention rate than thoracotomy (P < 0.001).

CONCLUSIONS: In our institution, there has been a trend in recent years towards increased use of median sternotomy to repair the aortic arch, which has been associated with a reduced rate of re-intervention. The midline sternotomy approach for coarctation with arch hypoplasia significantly reduces the risk of re-coarctation.

Keywords: Coarctation of aorta · Hypoplastic arch · Re-coarctation

INTRODUCTION

The management of coarctation of the aorta has changed since the first description of this operation in 1945 [1, 2]. The recent trend to one-stage repair of cardiac and associated arch anomalies via midline sternotomy has increased familiarity with aortic arch repair using this approach [3–6]. The midline approach provides good access to the proximal, distal and proximal descending thoracic arches, particularly in infancy. Furthermore, experience with patch repair of the aorta—in particular with Norwood procedure—has proved to be robust and long-lasting, giving surgeons greater confidence to augment the aortic arch in infancy.

We have reviewed our experience of repair of isolated aortic coarctation, highlighting especially the impact of surgical approach and re-intervention over two decades.

MATERIALS AND METHODS

Patients

Two hundred and eighty-eight patients who underwent isolated repair for coarctation of the aorta at Birmingham Children’s Hospital between 1991 and 2010 were enrolled in this study. The period 1991 to 2000, inclusive, was defined as the 1st decade and the years 2001 to 2010 as the 2nd decade. Patients with any kinds of additional procedures, such as VSD closure or arterial switches, were excluded from this study. Chart review and the database were used to determine demographics,
operative details and complications. Patients with small VSDs that did not require surgical intervention were included.

**Surgical technique**

Extended end-to-end anastamosis via thoracotomy was our procedure of choice and midline sternotomy was reserved for selected patients with a small transverse arch. Over the years, we have reduced our threshold for midline sternotomy in all borderline cases in which the aortic arch is hypoplastic. We define hypoplastic arch as: (i) diameter less than the size of the innominate artery (in mm), or (ii) diameter (in mm) less than body weight (in kilograms) + 1 kg [3]. Borderline cases were defined as having dimensions at these values. Subclavian flap aortoplasty was performed for only a small number of patients (12). The last subclavian flap aortoplasty was performed in 2001.

For patients with hypoplastic arch, the approach was through a standard midline sternotomy with cardiopulmonary bypass. During this study, deep hypothermic circulatory arrest was initially used. However, since 2003, selective cerebral perfusion technique, with a short period of deep hypothermic circulatory arrest, has been used during resection of the coarctation and ductal tissue. The arch was then repaired by either direct anastamosis was achieved by extended end-to-end, or end-to-side anastamosis between the aortic arch and descending thoracic aorta (direct anastamosis repair). If tension or residual narrowing was anticipated following the direct reconstruction of the back wall of the aortic arch and thoracic aorta, then repair was supplemented anteriorly with a patch of pulmonary homograft or, rarely, bovine pericardium (back wall anastomosis with patch repair).

**Follow up and re-intervention**

Patients were routinely followed up at our outpatient clinic after discharge. Systemic hypertension, reduced femoral pulse and persistent left ventricular hypertrophy prompted detailed ultrasound evaluation. Doppler flow peak velocities in excess of 3.2 m/s, or the finding of a diastolic tail, prompted further catheter study. Balloon dilatation was considered in cases of discrete re-coarctation with pressure gradient >10 mm Hg. Multilevel obstruction, in particular involving transverse arch, was preferentially treated by surgical intervention.

**Statistics**

Data were examined using the statistical software package R (version 2.13.0; R Foundation, Vienna, Austria). A probability value <0.05 was taken to represent a statistically significant difference between groups. Survival (freedom from re-intervention) was estimated by using the Kaplan-Meier product-limit method. Univariate analyses of actuarial outcome measures were made with the log-rank test.

**RESULTS**

**Preoperative data**

Preoperative data is shown in Table 1. There were 173 patients in the 1st decade and 115 patients in the 2nd decade. The median age at surgery was 16 days in the 1st decade and 35 days in the 2nd decade. However, there was no statistical difference between the 1st decade and the 2nd decade. One hundred and fifty-six (54%) patients had an operation during neonatal life. A higher number of patients with hypoplastic arch were identified in the 2nd decade: 20 patients in the 1st decade, 40 patients in the 2nd decade. Thirty-one patients had VSD that did not require attention at the time of aortic arch repair. Overall follow-up was 6.8 ± 8.3 years: 9.0 ± 9.8 years in the 1st decade and 3.5 ± 3.0 years in the 2nd decade.

**Surgical technique**

Table 2 summarizes surgical technique. Two hundred and thirty-seven patients (83%) had the repair through thoracotomy and 51 patients (18%) through midline sternotomy. In the 1st decade, the majority of patients (87%) had end-to-end anastomosis repair via thoracotomy. Eleven patients had subclavian flap aortoplasty in the 1st decade and 10 patients had extended

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**Table 1:**  Preoperative data in this study group

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<thead>
<tr>
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<tbody>
<tr>
<td>Number of patients</td>
<td>288</td>
<td>173</td>
<td>115</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>168 (58%)</td>
<td>100 (57%)</td>
<td>68 (59%)</td>
</tr>
<tr>
<td>Female</td>
<td>120 (41%)</td>
<td>73 (42%)</td>
<td>47 (40%)</td>
</tr>
<tr>
<td>Age at surgery</td>
<td>23 days (0–14 years)</td>
<td>16 days (0–14 years)</td>
<td>35 days (0–10 years)</td>
</tr>
<tr>
<td>Body weight at surgery</td>
<td>3.7 (1.1–66.3) kg</td>
<td>3.5 (1.4–66.3) kg</td>
<td>3.9 (1.1–29.9) kg</td>
</tr>
<tr>
<td>Age group</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Neonates (≤30 days)</td>
<td>156 (54%)</td>
<td>101 (58%)</td>
<td>55 (48%)</td>
</tr>
<tr>
<td>Infants (≥1 month–1 year)</td>
<td>87 (30%)</td>
<td>47 (27%)</td>
<td>40 (35%)</td>
</tr>
<tr>
<td>Children (≥1–15 years)</td>
<td>45 (16%)</td>
<td>25 (14%)</td>
<td>20 (17%)</td>
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<tr>
<td>Additional cardiac diagnoses</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hypoplastic arch</td>
<td>60 (21%)</td>
<td>20 (11%)</td>
<td>40 (35%)</td>
</tr>
<tr>
<td>VSD</td>
<td>31 (11%)</td>
<td>17 (10%)</td>
<td>14 (12%)</td>
</tr>
<tr>
<td>Follow-up</td>
<td>6.8 ± 8.3 years</td>
<td>9.0 ± 9.8 years</td>
<td>3.5 ± 3.0 years</td>
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end-to-end repair via midline sternotomy. One 14-year-old boy had a Gore-Tex patch augmentation repair through thoracotomy. In the 2nd decade, 41 patients (36%) had the repair through midline sternotomy and seventy-four (64%) through thoracotomy. Direct anastomosis (extended end-to-end or end-to-side) via midline sternotomy was performed in 21 patients (18%). Twenty patients (7%) had back wall anastomosis with patch. In recent years, there have been more patients who have had back wall anastomosis with patch repair. One patient with end-to-end anastomosis via thoracotomy required additional patch augmentation due to a pressure gradient across the anastomosis. Two patients had a patch augmentation via midline sternotomy without resection of coarctation: one had isolated hypoplastic arch without coarctation shelf and, in the other, the coarctation shelf involved the origin of the aberrant right subclavian.

Figure 1 demonstrates a trend toward more midline sternotomy than thoracotomy in our series, especially after 2001. There were no neurological complications associated with the surgery in either group. Four patients (8%) in the median sternotomy group had left recurrent laryngeal nerve palsy, confirmed on laryngoscopy.

**Mortality**

Three patients died within the first 30 postoperative days (early mortality): two patients in the 1st decade and one in the 2nd decade (Table 3). One patient died of sudden arrhythmia, one of low cardiac output syndrome, and one of respiratory infection soon after discharge. Overall hospital mortality was 1%: 2% in the 1st decade, 1% in the 2nd decade. Overall late mortality was 3%: 3% in the 1st decade and 2% in the 2nd decade. One patient diagnosed with Shone’s complex had repair of mitral valve and relief of left ventricular outflow tract, and died due to low cardiac output syndrome after the age of 15 months. One patient returned to the theatre for a Norwood-type operation due to borderline LV and died after the operation at the age of 2 months. One patient died of low cardiac output syndrome after requiring VSD closure (initially assessed as being small) at 34 days. Of the non-cardiac deaths, one patient died of septic shock at 11 months, one from respiratory infection at 4 months and another from acute lymphoblastic leukaemia at 17 months. Five patients died of unknown causes. Overall 1-, 5- and 10-year survival rates were 95%, 94% and 92%, respectively. There were no significant differences in survival between the 1st decade and the 2nd decade.

### Table 2: Surgical technique

<table>
<thead>
<tr>
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<tbody>
<tr>
<td>Thoracotomy</td>
<td>237 (83%)</td>
<td>163 (94%)</td>
<td>74 (64%)</td>
</tr>
<tr>
<td>End-to-end anastomosis repair</td>
<td>223 (77%)</td>
<td>151 (87%)</td>
<td>72 (63%)</td>
</tr>
<tr>
<td>Subclavian flap aortoplasty</td>
<td>12 (4%)</td>
<td>11 (6%)</td>
<td>1 (1%)</td>
</tr>
<tr>
<td>Patch repair</td>
<td>2 (1%)</td>
<td>1 (1%)</td>
<td>1 (1%)</td>
</tr>
<tr>
<td>Midline Sternotomy</td>
<td>51 (18%)</td>
<td>10 (6%)</td>
<td>41 (36%)</td>
</tr>
<tr>
<td>Direct anastomosis repair</td>
<td>31 (11%)</td>
<td>10 (6%)</td>
<td>21 (18%)</td>
</tr>
<tr>
<td>Back wall anastomosis with patch repair</td>
<td>20 (7%)</td>
<td>0</td>
<td>20 (17%)</td>
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### Table 3: Postoperative results

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<tr>
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<tbody>
<tr>
<td>Mortality</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Early mortality</td>
<td>3 (1%)</td>
<td>2 (1%)</td>
<td>1 (1%)</td>
</tr>
<tr>
<td>Late mortality</td>
<td>8 (3%)</td>
<td>6 (3%)</td>
<td>2 (2%)</td>
</tr>
<tr>
<td>Morbidity</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Re-intervention for arch</td>
<td>33 (11%)</td>
<td>27 (16%)</td>
<td>6 (5%)</td>
</tr>
<tr>
<td>Surgical intervention</td>
<td>12 (4%)</td>
<td>9 (5%)</td>
<td>3 (3%)</td>
</tr>
<tr>
<td>Catheter intervention</td>
<td>24 (8%)</td>
<td>19 (11%)</td>
<td>5 (4%)</td>
</tr>
<tr>
<td>Multiple intervention</td>
<td>6 (2%)</td>
<td>4 (2%)</td>
<td>2 (2%)</td>
</tr>
<tr>
<td>Median interval for intervention</td>
<td>110 days (1–5035 days)</td>
<td>85 days (1–5035 days)</td>
<td>143 days (68–820 days)</td>
</tr>
</tbody>
</table>
Re-intervention for recurrent arch obstruction

Thirty-three patients (11%) required re-intervention for arch obstruction (Table 3). Twelve patients had single surgical intervention (4%), 24 had catheter intervention and six had multiple interventions. There was a statistically higher re-intervention rate in the decade 1991–2000, compared to 2001–10 (16% vs 5%). Figure 2 shows Kaplan-Meier re-intervention-free survival curves comparing the 1st and 2nd decades. Patients in the 2nd decade had higher intervention-free survival, compared to the 1st decade (92% vs 85% at 5 years, \( P = 0.02 \)).

Table 4 shows numbers of re-interventions for the surgical approaches and Figure 3 shows Kaplan-Meier re-intervention-free survival curves comparing midline sternotomy and thoracotomy approaches. There was no statistical difference between thoracotomy and midline approach (\( P = 0.11 \)). After midline sternotomy, no patient required surgical re-intervention. Figure 4 shows Kaplan-Meier re-intervention-free survival curves comparing midline sternotomy with thoracotomy among patients with hypoplastic arch. There was a significant difference between these groups (\( P < 0.001 \)). There was no recurrence of aortic arch obstruction after the use of a patch and there is also no evidence of aneurysm formation.

DISCUSSION

The management of coarctation of the aorta has changed since the first description of this operation in 1945 [1]. However, the optimal surgical management of coarctation still remains controversial [7]. The controversies concern several issues: method of repair, growth of the hypoplastic arch and incidence of re-coarctation.

The debate over the preferred surgical technique includes how extensively the proximal arch needs to be repaired, which relates to the degree of transverse arch hypoplasia. Part of the problem is that the definition of hypoplastic arch is also controversial. Mee and colleagues described a formula for hypoplastic arch as the transverse arch diameter (in mm) being less than the patient’s body weight (in kilograms) + 1 kg [3]. We extended this definition to include the comparison between the size of the innominate artery and the transverse arch. Brouwer believed that the arch grows after simple resection and end-to-end anastomosis, and that enlargement of the hypoplastic arch is not necessary [8]. Jahangiri reported that arch hypoplasia regresses after subclavian flap [9]. Liu and associates reported that moderately hypoplastic arches have adequate growth at long-term follow-up, but one third of them retain a small proximal arch [10]. As we presented in this paper, some degree of aortic arch hypoplasia is often present in many neonates presenting with coarctation. However, regardless of the definition of hypoplastic arch, the initial repair technique should completely relieve any obstruction of the entire aortic arch. Therefore, if there is any concern about the transverse arch, we prefer midline sternotomy for better and safer access to the proximal arch. The increasing trend towards midline sternotomy repair in this series reflects a lower threshold of adopting a more extensive repair approach in borderline cases.

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Some institutions have had excellent results by end-to-end anastomosis through thoracotomy. However, other series have reported a higher rate of recurrent coarctation by end-to-end repair (10–12% at 1 year) and cite hypoplasia of the transverse
arch as a risk factor [11, 12]. Additionally, it is difficult to relieve arch obstruction completely, in the setting of aortic arch hypoplasia, within limited aortic clamp time and clamping the proximal arch may compromise the cerebral perfusion during the anastomosis. Midline sternotomy allows us safe and good exposure with cardiopulmonary bypass support.

After the successful reports of one-stage repair of cardiac and arch anomalies, arch repair through midline sternotomy has become a more common procedure in many centres [3, 4]. Initially, one-stage approach technique was thought to allow complete repair of all lesions through one midline incision. However, in addition to this, recent reports have emphasized that this approach gave a reduced re-coarctation rate [4, 13]. Since we started one-stage repair for complex arch anomalies, we have seen an increase in the number of patients undergoing isolated arch repair via midline sternotomy (Figure 1). This may be due to increased experience and, hence, a lower threshold for performing this approach. Some authors argue against the use of cardiopulmonary bypass and DHCA for isolated arch repair, but many reports fail to demonstrate any long-term difference in neuro-developmental outcomes following the use of DHCA for repair of congenital heart disease [14]. In our series, no patient developed serious neurological complications during the study period.

Previous reports of patch repair of aortic coarctation were suboptimal because of higher incidence of re-stenosis of the aorta (27%); however, these techniques involved patching via thoracotomy, where the extent of the patch is much more limited [15]. Previous reports on midline approach to aortic coarctation preferred end-to-end type repair or end-to-side repair without foreign material [4, 13]. We also preferred direct anastomosis in the earlier decade but, with surgical experience of the Norwood stage 1 operation for hypoplastic left heart syndrome, the use of patch material for augmentation of the aortic arch in the Norwood procedure in neonates is widely accepted. With the use of a patch, the procedure is more reproducible and less likely to cause bronchial or pulmonary compression. In our midline approach series, no patients developed aneurysm formation or airway obstruction after the operation. However, we will continue to review long-term results.

Limitations of this study are that it is a retrospective, non-randomized design and the small number of events for re-intervention makes for limited statistical analysis. Some of the patients were lost to follow-up during these two decades. Additionally, age bias and changes in medical practice and surgical techniques could have contributed to the improved results of recent years. The small number of older patients in the study may have a different clinical course to the infant population—however, the numbers of older patients are small and a larger study may be required to evaluate this.

**CONCLUSION**

Surgical repair of isolated coarctation has excellent early and late outcomes but there remains a significant need for re-intervention. The need for re-intervention is frequently associated with a degree of hypoplasia of the transverse arch. In our institution, there has been a trend towards increased use of median sternotomy to repair isolated coarctation with hypoplasia of the aortic arch. This has been associated with a reduced rate of re-intervention in recent years. A midline sternotomy approach in coarctation with borderline transverse arch dimensions may give better long-term outcomes.

**Conflict of interest:** none declared.

**REFERENCES**

APPENDIX. CONFERENCE DISCUSSION

Dr T. Spray (Philadelphia, PA, USA): This large series of coarctation repair attempts to compare different techniques. It gets very confusing when you look at the two groups because the incidence of arch hypoplasia by your definition is higher in the second series. For some reason, I think we are seeing the same thing, that arch hypoplasia seems to be more common now. Maybe it is partly the definition of arch hypoplasia. But when you look at your first group, a proportion of patients ended up with a midline sternotomy and a repair, and I presume those were patch repairs through a midline sternotomy. For example, 11 out of the 20 patients that had arch hypoplasia in Group I had a midline approach. Was the surgical approach the same as in the second group? Was it patch augmentation, or did you do a direct anastomosis through a midline approach in that group?

Dr Sakurai: So the question is, did we do more patch repair recently?

Dr Spray: If you have arch hypoplasia, if you have made that diagnosis, it looks like in the first group you also did a midline approach for a significant number. Did you patch those patients?

Dr Sakurai: Well, we started the direct anastomosis with patch after 2003, so until that time we just extended it into a direct type anastomosis.

Dr Spray: So there were two years in which you might have done a patch in the first group; is that right?

Dr Sakurai: Yes. We have one patient in the first group, but the patch is done by a thoracotomy. This is a 14 year old patient, and the surgeon tried to do the direct anastomosis at that time, but he thought it might be difficult to do and keep the Gore-Tex patch only in that case.

Dr Spray: So what you are really trying to say is that extensive patch augmentation of the arch is good in arch hypoplasia, without a high recurrence rate, in fact, no recurrence if I understand your paper correctly; is that right?

Dr Sakurai: Yes. Well, this technique is coming from the Norwood operation, so after starting the Norwood with the use of a homograft patch, it does look like it is working okay so far, so then we extended the arch hypoplasia also. But in case we cannot do a direct anastomosis even from the midline sternotomy, we still do the direct anastomosis, but most of the time recently we tended to use a patch rather than the direct anastomosis.

Dr Spray: And why do you add a patch if you can do a direct anastomosis from the front?

Dr Sakurai: Well, if we can, we do direct anastomosis. But as I said here, if we have any concern about tubular narrowing of the transverse arch, we rather tend to use a patch. And so far, the patch works very well.

Dr Spray: Obviously our centre has used a very similar approach for years, especially with associated VSD, which we fix at the same time. But adding a patch has the advantage of taking some tension off the suture line and decreasing bleeding and potential for creating narrowing when you try to control bleeding, so I would certainly support that approach.

It is also a little difficult to sort out the reinterventions in Group I. It was stated in your paper that the surgical reinterventions were largely for arch problems.

Dr Sakurai: Yes.

Dr Spray: And if that is the case, how many of those patients had hypoplasia of the arch that did not grow?

Dr Sakurai: That is a very good question, but I do not have – well, I cannot find a hypoplastic arch from the operation note in this series. So I do not have the exact number of the size of the transverse arch or the size of the ascending aorta in this series.

So as you said initially, we may have more attention to the hypoplastic arch recently. And this is also very difficult to say. We do not know how much hypoplasia is acceptable, so this is why if we have any concerns about the transverse arch, we have a discussion with the cardiologist. But most of the time, if we have any concerns about the transverse arch, we prefer, whether it is with that patch, we prefer a midline sternotomy because it is the safe option.

Dr Spray: So we really do not have the answer to whether the hypoplastic arch grows or not, right? One other final question, and that is really related to concern about patching the aortic arch. I mean, patching of coarctations has sort of a bad reputation because of late aneurysm formation, especially opposite the patch. And we see in the Norwood operations and other patch repairs of the arch that the arches tend to dilate over time. Do you think that is going to be a concern for these patients as they get older?

Dr Sakurai: Well, I have to mention that in our Norwood series, we do have a 25% recoarctation rate, so it is not very low. But in terms of aneurysm formation, we do not see the aneurysm – well, it will be a significantly dilated ascending aorta, but so far we do not see aneurysm formation. And also, in another series with a complex arch, complex intracardiac anomaly, there is one patient with the autologous pericardial patch which is not untreated. It became an aneurysm. I think this is the only reoperation after using the patch.