Advances in the management and surveillance of patients with aortic coarctation

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Chapter 5

Comparison of surgical repair with balloon angioplasty for native coarctation in patients from 3 months to 16 years of age

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**Abstract**

**Objective:**
Surgery and balloon angioplasty for coarctation of the aorta have shown comparable short-term results, but long-term follow-up remains unclear. Comparison of surgical repair and balloon coarctation for native coarctation of the localised membranous form is performed retrospectively. To allow a valid comparison between both techniques, identical inclusion criteria were applied.

**Methods:**
Results of surgery (group A, 18 patients, age 0.30-14 years, median 0.63 years) and balloon angioplasty (group B, 28 patients, age 0.25-15 years, median 5.8 years) for isolated, native coarctation in children > 3 months, performed in a 10-year-period, were compared. Kaplan-Meier analysis was performed in both groups. Mean follow-up ranged from 2.5 to 11 years (mean 7.2 ± 2.4 years) in group A and from 1.4 to 10 years (mean 5.4 ± 2.8 years) in group B.

**Results:**
Immediate success was obtained in all patients following surgery and 27/28 patients (96%) following balloon angioplasty. No statistical difference between surgery and angioplasty with respect to resultant pressure gradient decreases were found. Mortality was not encountered. Hospital stay varied from 6 to 20 days in group A and was 48 hours for all patients in group B. Recoarctation occurred in 1 patient (5.6%) in group A and in 2 patients (7%) in group B. Log-rank test reveals no statistical difference in freedom from reintervention probabilities between surgery and angioplasty. Aneurysm formation was not encountered.

**Conclusions:**
Both surgical repair and balloon angioplasty for native coarctation yield low reintervention probabilities in comparable patients. Aneurysm formation was not encountered following different treatment types.
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Introduction

The treatment of coarctation of the aorta, a malformation first described by Johannes Baptista Morgani in 1761, has started in the past few decades. Surgical repair has provided adequate treatment for patients with coarctation since Crafoord described a successful resection and end-to-end anastomosis (RETE) in 1945. Although long-term results of surgical intervention are reported satisfactory, balloon angioplasty has been proposed as a viable alternative to surgery in primary treatment of coarctation in 1982.

With advancing experience, indication for balloon angioplasty has become more defined, patients with long-segment coarctation and age less than three months generally being excluded from this type of intervention. Postoperative recoarctation and aneurysm formation are encountered in both surgery and balloon angioplasty. Whether balloon angioplasty should be first-choice therapy instead of surgical repair in a selected group of patients has not been established yet. Data comparing surgical versus balloon therapy are limited. This study contains a retrospective evaluation of 46 patients with coarctation of the localised membranous form, classified according surgical repair and balloon angioplasty, in respect to risk factors for recoarctation, management and outcome. Additionally, the outcomes of surgery and balloon angioplasty are compared, performed as primary treatment modes for native coarctation in comparable patient groups. Objectives are to establish and compare the long-term results of different types of management of native coarctation, in respect to immediate success, complications and long-term follow-up in the past decade.

Materials and methods

Demographics. This study includes 46 consecutive patients with coarctation of the localised membranous form. Neither isthmus hypoplasia, defined as isthmus diameter less than 40% of diameter of ascending aorta, nor arch hypoplasia, defined as proximal or distal transverse arch diameter less than 60% or 50% respectively of the diameter of the ascending aorta, were present in any of the patients. All patients were older than three months of age. This study covers a 10-year period of coarctation management, ending in February 2000. We classified these patients in two groups according to the kind of treatment performed. Group A consists of 18 patients who had a surgical repair, Group B of 28 patients who underwent balloon angioplasty. In group A, 16 patients were treated with RETE, one with patch aortoplasty (PA) and one was treated with a combination of a RETE and PA. Four children (14%) in group A needed additional surgery, including a VSD closure, an ASD closure, a TGA correction and an aortic valvotomy. These surgical procedures followed coarctation repair in three patients and was simultaneously performed in one patient. The patients’ ages at intervention ranged from 0.30 to 14 years of age (median 0.63 years) in group A and from 0.25 to 15 years of age (median 5.8 years) in group B (p=0.001, Mann-
Whitney test for non-parametric independent two-group comparisons). Associated congenital heart defects were present in 14/18 patients (78%) in group A and in 17/28 patients (61%) in group B (see Table 1). Non-cardiac malformations were present in two patients with Turner’s syndrome.

Table 1. Additional cardiac lesions in patients of age > 3 months to 16 years, with coarctation of the localised membranous form in group A (surgical repair) and B (balloon angioplasty).

<table>
<thead>
<tr>
<th></th>
<th>Group A (n=18)</th>
<th>Group B (n=28)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bicuspid aortic valve</td>
<td>6</td>
<td>7</td>
</tr>
<tr>
<td>Patent arterial duct</td>
<td>9</td>
<td>4</td>
</tr>
<tr>
<td>Aortic valve stenosis</td>
<td>2</td>
<td>6</td>
</tr>
<tr>
<td>Atrial septal defects</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Ventricular septal defect</td>
<td>5</td>
<td>3</td>
</tr>
<tr>
<td>Mitral valve stenosis</td>
<td>1</td>
<td>-</td>
</tr>
<tr>
<td>Transposition of the great arteries</td>
<td>1</td>
<td>-</td>
</tr>
<tr>
<td>Aortic valve insufficiency</td>
<td>2</td>
<td>-</td>
</tr>
</tbody>
</table>

Technique of balloon angioplasty
Balloon angioplasty was carried out under complete anesthesia. There were no important differences in technique or equipment over the 10-year period. Aortic arch angiography was performed and the aortic diameter at the level of the diaphragm was measured. The balloon catheter was advanced up to the aortic arch, deflated and then retracted until the balloon crossed the coarctation. Inflation with diluted contrast was performed until the waist in the balloon disappeared. This procedure was performed three times, to optimize the final result. To measure aortic pressures and perform an angiogram a catheter was passed over the guide wire. This procedure was repeated using a larger balloon diameter when the result was unsatisfactory. The size of the balloon was chosen depending on the aortic diameter as measured at the level of the diaphragm, in such a way that the balloon would not exceed that diameter initially, and not by more than 2 mm in a secondary stage, performed when necessary.

Immediate results
The immediate results of surgery were considered satisfactory if a pulsatile flow in the descending aorta was registered by palpation after surgery and the resultant pressure gradient was less than 20 mmHg. In angioplasty, satisfactory result was obtained on behalf of the angiography performed after angioplasty and if a gradient less than 20 mmHg was reached.

Follow-up
Follow-up ranged from 2.5 to 11 years (mean 7.2 ± 2.4 years) in group A and from 1.4 to 10 years (mean 5.4 ± 2.8 years) in group B. Follow-up included clinical evaluation every three months in the first year after intervention and yearly thereafter. Arm and
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Leg cuff pressures were registered and chest radiography and 2D / continuous-wave echocardiographic Doppler ultrasound studies performed. Follow-up angiography and MRI were performed in patients suspect for aneurysm formation. A gradient of 30 mmHg and a registered continuation of flow in diastole in echocardiographic Doppler ultrasound studies, with blood pressure gradients between upper and lower extremities exceeding 20 mmHg, were considered to correspond with recoarctation requiring reintervention. MRI was performed in group B in patients with a follow-up exceeding five years, to exclude aneurysm formation. Recoarctation was treated with repeat surgery or balloon angioplasty after additional angiography.

Statistical methods
Test results with a p-value < 0.05 were considered significant in all statistical analyses. Pre- and post-operative pressure gradients were analyzed with the T-test for paired results. Confidence intervals were computed for pressure gradient decreases in groups A and B. Resultant pressure gradients decreases were compared between groups A and B with the T-test (2-tail, independent samples). 95% confidence intervals of the difference were computed for this comparison also. Kaplan-Meier curves were constructed to determine the intervention free probabilities in groups A and B. Additionally, the log-rank test was used to compare intervention free probabilities between both groups.

Results
Immediate results
The mean pre-operative pressure gradient in group A decreased from 47 ± 21 mmHg to 15 ± 9.8 mmHg post-operative (p<0.001, 95% confidence interval ranges from 23 to 42 mmHg). In group B, the mean pressure gradient was reduced from 39 ± 10 mmHg to 12 ± 9.1 mmHg (p<0.001, 95% confidence interval ranges from 22 to 31 mmHg). In 27/28 patients (96%), procedures were considered successful. No immediate success was obtained in one patient, due to a persisting coarctation membrane. This patient underwent a second, this time successful, balloon angioplasty after three months. T-test (2-tail, independent samples) revealed no statistical difference between groups A and B with respect to resultant pressure gradient decreases. 95% confidence intervals for difference from the mean ranged from −1.96 mmHg (lower) to 16.18 mmHg (upper). Hospital stay varied from six to 20 days in group A, averaging 9.4 days. Hospital stay was 48 hours for all patients in group B. Artery-section was required to get access to the femoral artery in one patient following percutaneous intervention. No difference in the length of the legs was noted in subsequent follow-up.

Follow up
No mortality was encountered in this series. In group A, recoarctation occurred
in 1/18 (5.6%) patient, following PA at six months of age. This recoarctation was found to be localised, limited to the site of the anastomosis. Hypoplasia of the aortic isthmus and/or arch was not encountered. Subsequently, balloon angioplasty was performed successfully in this patient. In group B, recoarctation occurred in 2/27 (7%) patients in which primary treatment was considered successful immediately. Both recoarctations were localised at the site of the former coarctation ridge and

**Figure 1.** Kaplan Meier curves for coarctation of the localised membranous form in patients from three months to 16 years of age, managed with surgical repair (group A, n=18) and balloon angioplasty (group B, n=28).

**Figure 2.** Flow diagram of all patients, managed in a 10-year period.
CoA = coarctation of the aorta
BA = balloon angioplasty
RETE = resection and end-to-end anastomosis
PA = patch angioplasty
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membraneous. Neither isthmus nor arch hypoplasia were found. These children were three and eight years old at primary treatment. Both were re-operated using RETE. No aneurysm formation was encountered in this series. MRI was performed in 13/28 patients (46%) following balloon angioplasty for more than five years. Figure 1 shows the Kaplan-Meier curves for groups A and B. Log rank analysis reveals no significant difference between surgery and balloon angioplasty. Follow-up of all patients is summarized in figure 2.

Discussion

Surgical repair has provided adequate treatment for patients with this malformation since Crafoord described a successful RETE in 1946.² Balloon angioplasty has been proposed as a viable alternative to surgery in primary treatment of coarctation in 1982.⁴,¹⁴ In spite of disappointing results of balloon angioplasty in initial reports, subsequent experience appears favorable, although the necessity of long-term follow-up continues to exist. However, the number of studies comparing balloon angioplasty with surgical repair is limited. Shaddy et al. reported a prospectively randomized study including 36 patients in 1992.¹¹ They reported a higher risk of aneurysm formation and possibly higher risk of restenosis after angioplasty, although risks of other complications were similar. In 1993, Johnson et al. presented a review of literature on treatment of native coarctation in infants with both techniques.¹² Although early mortality rate was similar, balloon angioplasty revealed a much higher rate of recoarctation (57%) in infants as compared with those who underwent surgical repair (14%) in reviewed literature. Subsequent experience appears more favorable on behalf of balloon angioplasty, recoarctation rates being comparable to surgical repair in recent reports.⁵,¹⁵,¹⁶ Aneurysm formation is encountered after both balloon angioplasty and all types of surgical repair performed in this series: not solely after percutaneous intervention.

Several remarks should be made on comparing surgical repair to balloon angioplasty. First, as was pointed out by Hanley,¹⁷ a comparison can not be directed towards a single ideal form of therapy, considering the complex physiologic and morphologic variability of this lesion. Instead thereof, a more defined indication for both surgery and angioplasty should be established. Second, individual studies span different time periods, focusing on particular age groups and particular morphologic subsets, which undermines a meaningful comparison between different techniques. Third, controversy about an ideal type of surgical repair continues to exist, impairing comparison of balloon angioplasty with surgery in general.

To meet these considerations, variability in age and morphology has been compensated for in the present study, including a selected group of patients managed surgically. Although median age varies in both groups, age borders are identical. To our knowledge, no reported or hypothetic indications exist for different outcomes within this age group between younger and older patients. Since our study focuses exclusively on
a localised and membraneous type of coarctation morphology, excluding associated isthmus and/or arch hypoplasia, it allows a more valid comparison between both treatment strategies. Furthermore, the different surgical techniques involved in this comparison were limited to RETE (16/18 patients), PA (1/18 patient) and a combination of these techniques (1/18 patient). Nevertheless, we are aware that this retrospective approach poses an important limitation, in which this study resembles the major part of literature.

**Immediate results**

Significant reduction of peak-to-peak systolic gradients can be accomplished with both surgery and balloon angioplasty, as shown in literature.\(^{16,18}\) Reduction of pressure gradients was more pronounced in group A than in group B, but in both groups these results were clinically sufficient, certainly since decrease of the residual gradient can be expected when growth occurs and flow increases proportionally after the various procedures.\(^{19}\) Therefore, we consider the upper limit of the 95% confidence interval for the difference of the mean, 16 mmHg, not high enough to choose for surgery instead of balloon angioplasty. The finding of smaller resulting pressure gradient following balloon angioplasty compared to surgery accords with several other reports.\(^{11-13}\) In 1 patient in group B the procedure was not successful immediately. This was a patient with Turner’s syndrome, for which secondary balloon angioplasty was performed successfully after 3 months. Patients with Turner’s syndrome are notorious for a fragile aortic wall structure which can cause problems in both surgical intervention and balloon angioplasty.\(^{20}\) The unfavorable outcome in this patient can probably be explained by this particular morphology and a first-choice therapy in this class of patients has not been established.

High incidence of recoarctation in children younger than three months has been described,\(^{5}\) although Rao reported a similar recoarctation rate in patients less than three months in comparable groups of balloon angioplasty (n=15) and surgery (n=14).\(^{11}\) Nevertheless, other report’s proportions reintervention following balloon angioplasty, varying from 50-83% in this age group, justify the exclusion of this age group in our study.\(^{5,15}\) In part, these adverse results may be caused by associated arch and/or isthmus hypoplasia in this infant population. The presence of aortic hypoplasia usually increases the left ventricular obstruction created by the coarctation, thus raising signs and symptoms before the age of three months in most patients. Consequently, the age minimum of three months overlaps with the exclusion of most patients with aortic hypoplasia.

**Recoarctation**

Recoarctation as a late complication is found to occur in 10 to 30% after surgery.\(^{3,6-9}\) Varying occurrence of restenosis has also been encountered after balloon angioplasty, ranging from 11 to 60%.\(^{11}\) This variation may depend on specific aspects of surgical/angioplasty technique, including patch material, extension of resection and balloon diameter. Additionally, the rates of associated aortic hypoplasia included and age at surgery/ balloon angioplasty may attribute to different outcomes in various studies.
Recoarctation rates for balloon angioplasty (19%) and surgery (11%) were similar in a recent review of 58 reports by Rao. In our study comparable results were established after surgery (5.6%) and balloon angioplasty (7%) in patient groups, comparable in respect to age and coarctation morphology. With respect to these recoarctation rates, we presume that different recoarctation mechanisms may play a role. Type of coarctation management may influence the prevalence of residual or recurrent coarctation by (incomplete) resection of ductal tissue, suture material and the width of the anastomosis. The possible mechanisms in the process of recoarctation related to these different techniques consist of inadequate growth of the anastomosis, active fibrosis and narrowing at the anastomotic site, thrombosis at the suture line, and retention of abnormal, possibly ductal, tissue. In our series, a failure to grow at the site of surgical anastomosis may have caused the recoarctation following RETE. Localised recoarctations following balloon angioplasty, located at the site of the former coarctation ridge, may be caused by the failure to remove this material completely. Kaplan-Meier curves accomplished to compare the intervention free probability of surgery and balloon angioplasty show no significant difference in intervention free probability between groups A and B. None of the therapy strategies was identified to be favorable in respect to reintervention probability. The relative advantages of the less invasive character and the shorter hospital stay might tip the balance in favor of balloon angioplasty in this patient category. A thorough and informative counseling of the parents on this topic however is in this situation obligatory to obtain informed consent.

Aneurysm formation

The incidence of postoperative aneurysms reported by several investigators varies between 0% and 24%. Aneurysm formation has not been encountered in this series. This observation corresponds with other series. Echocardiography played a central role in the screening for aneurysm formation of our patients. When optimal imaging is feasible, being the case in infants and children, the literature supports combined two-dimensional and Doppler color flow echocardiography to image the aortic arch, isthmus, and coarctation site. A sensitivity of 100% for aneurysm formation in adults can thus be achieved by echocardiography and clinical visits, as was pointed out by Therrien et al. Since we consider echocardiography more sensitive and specific at lower patient’s ages, we believe that this policy is appropriate for children as well. To rule out any doubt, MRI was systematically performed in 13/28 patients (46%) following balloon angioplasty for more than five years.

Conclusions

Both surgical repair and balloon angioplasty for native coarctation yield low reintervention probabilities in patients with localised and membranous coarctation morphology and age from three months to 16 years. Pressure gradient decreases appear to be higher following surgical repair. Balloon angioplasty, may remain, despite this result, a justifiable option for coarctation of the aorta of the localised
membranous form in the paediatric age group older than 3 months, since it is a safe and less invasive procedure. Our data suggest that aneurysm formation should not be reason to counsel in favour of surgical management, since no aneurysms were encountered after balloon angioplasty.

To allow a proper parental decision it is of crucial importance that they should be well informed on the considerations mentioned above to allow them to make a deliberate choice.

References

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