Advances in the management and surveillance of patients with aortic coarctation

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Chapter 3

Angioplasty for coarctation in different aged patients

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Abstract

Background:

Differences in indication and outcome of balloon angioplasty for coarctation in children and adults have not been elucidated sufficiently. Results of balloon angioplasty for coarctation are compared between pediatric and adult age groups.

Methods:

Balloon angioplasty for coarctation of the aorta was performed in 85 patients, classified according to age and native / recoarctation. Groups A (n=32) (<16 years) and B (n=17) (>16 years) included native coarctations. Groups rCoA A (n=33) (< 16 years) and rCoA B (n=3) (>16 years) included recoarctations. Follow-up included 2D Doppler echocardiography and additional angiography or MRI. Gradient reductions in groups were compared using independent-samples T-test. Kaplan-Meier and Logrank analyses were performed to compare long-term outcome.

Results:

No mortality occurred. Immediate success was equal in groups A, B and rCoA A (94%). Dilatation was unsuccessful in two patients in group rCoA B. Pressure gradients decreased 23 mmHg in group A, 31 in B, 18 in rCoA A and 11 in rCoA B. Pressure gradient drops, compared between groups A and B, showed a significant difference ($p<0,001$). Hospital stay ranged from 12-48 hours. Follow-up ranged from 6 months to 12 years (mean 4,9 years). Kaplan-Meiercurves of groups A and B are not different as determined by log rank analysis. No aneurysm formation was encountered.

Conclusions:

Results of balloon angioplasty for native coarctation in both selected children and adults are excellent. In recoarctation, we recommend balloon angioplasty in the pediatric age.

Introduction

In 1945 surgery was first described as treatment for coarctation of the aorta,[1] and various surgical techniques have been advocated and proved successful since then. Since 1982,[2] balloon angioplasty for coarctation has been added as viable alternative to surgery for treatment of coarctation of the aorta and it is currently considered as a safe and effective treatment for coarctation.[3-5]

Balloon angioplasty for coarctation has been used as treatment for both children and adults, in the literature as well as in our clinical setting.[4,6-9] Whether indication for and outcome of this treatment are the same in these different age groups remains to be elucidated more thoroughly. This study is explicitly limited to the comparison of results and follow-up of balloon angioplasty for coarctation in children and adults to test our hypothesis that the role of balloon angioplasty in pediatric and adult coarctation can be the same.

Methods

Patients

Balloon angioplasty for coarctation of the aorta was performed in 85 patients in our institute from April 1988 to July 2000. Native coarctation was treated in 49/85 patients (37 males and 12 females; age 3 months to 67 years; median 8,3 years). Recoarctation was treated in the other 36/85 patients (22 males and 13 females; age from 3 months to 43 years; mean 8,1 years). All patients with a localized type of coarctation or recoarctation and older than 3 months were included. We classified the patients with native coarctation in 2 groups according to age 16 years. Group A comprised 32 children (mean age 5 years, range from 3 months to 15 years). Group B included 17 adults (mean age 36 years, range from 16 to 67). The age of 16 years was chosen according to the patient population in two centers within our institute. 24/49 patients (49%) had associated heart defects (table I).

Table I. Associated heart defects in patients with native coarctation of the aorta, classified in two groups according age, treated with balloon angioplasty (n=49).

Group A: patients < 16 years old (n=32)	
Bicuspid aortic valve and/or aortic valve stenosis	6
Aortic valve stenosis and mitral valve insufficiency	2
Aortic valve stenosis and PDA	1
VSD (hemodynamic insignificant)	3
Isolated PDA	2
ASD (hemodynamic insignificant)	1
Group B: patients > 16 years old (n=17)	
Bicuspid aortic valve and/or aortic valve stenosis	6
VSD	2
Aortic insufficiency	1

ASD = atrial septum defect, PDA = patent ductus arteriosus, VSD = ventricle septum defect

Additionally, we classified the 36 patients with recoarctation angioplasty in a similar fashion. Group rCoA A included 33 patients (mean age 6,1 years; range from 3 month to 15 years) and rCoA B included 3 patients (mean age 30 years; range from 23 to 43 years). 30/36 patients (83%) had associated heart defects (table II).

Table II. Associated heart defects in patients with re-coarctation of the aorta, classified in two groups according age, treated with balloon angioplasty (n=36).

Group rCoA A: patients < 16 years old (n=33)	
Bicuspid aortic valve and/or aortic valve stenosis	9
VSD (hemodynamic insignificant)	7
VSD and PDA	5
Isolated PDA	1
ASD (hemodynamic insignificant)	2
TGA (associated with VSD and/or PDA)	3
Mitral valve stenosis	2
Group rCoA B: patients > 16 years old (n=3)	
VSD	1

ASD = atrial septum defect, PDA = patent ductus arteriosus, TGA= transposition of the great arteries, VSD = ventricle septum defect

Primary management consisted of surgery in 34/36 patients, in whom the period in between primary and secondary intervention ranged two months to 17 years, median being 5 years. Balloon angioplasty was primary treatment in 2/36 patients, intervention free period following, being 3 and 15 months.

Technique balloon angioplasty. Balloon angioplasty was carried out under complete anesthesia. The technique used for percutaneous balloon angioplasty was described previously.[10] There were no important differences in technique or equipment over the 12-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, de-aired 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 3 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.

A different technique was used in 3 patients in group B with a local occlusion at the coarctation site, in whom the brachial, anterograde route was chosen and in which after perforation of the vessel with a stiff (coronary) probe, angioplasty with a 4 mm coronary angioplastic balloon was performed. In 1 patient (group B) an 18-mm wall-stent was deployed.

The immediate result of angioplasty was considered successful if angiography showed no residual stenosis, indented vessel walls, and the intra-vascular pressure gradient was reduced to less than 20 mmHg. In those cases in which the angiographic result was satisfactory but pressure gradient lower than 20 mmHg could not be reached, procedures were considered successful consequently.

Follow-up

Follow-up included clinical evaluation and echocardiographic Doppler ultrasound studies, in which a gradient of 30 mmHg and a registered continuation of flow in diastole, with blood pressure gradients between upper and lower extremities exceeding 20 mmHg were considered to correspond with recoarctation requiring reintervention. Local evaluation of the coarctation site by Doppler echocardiography assessed the aortic flow and morphology directly. Consequently, in deciding to perform reintervention, echocardiographic results were considered informative in addition to arm/leg pressure gradients, determining the influence of collateral vasculature. Additional angiography or MRI was performed to exclude possible aneurysm formation. Vascular complications were accounted for by clinical evaluation, including local palpitation and auscultation. A vascular surgeon was consulted and peripheral Doppler performed following all dubious findings.

Statistical analysis

Due to a limited number of patients in group rCoA B, statistical comparison was limited to groups A and B. Decreases in pressure gradients before and after dilatation of the native coarctation between groups A and B were compared using independent-samples T-test (2-tail, unequal variances). A p-value $< 0,05$ was considered significant. Chi-square test was performed to compare recoarctation rates between groups. Kaplan-Meier analysis was performed to display the incidence of reintervention after balloon angioplasty. Logrank analysis was used to compare time to reintervention between child and adult populations. Two patients in whom aortic valve stenosis was encountered, were excluded from the independent-samples T-test. The patient in group B who received a stent was excluded from statistical analysis as indicated above.

Results

Immediate results

The peak to peak systolic gradient across the coarctation decreased from 37 (13 SD) to 12 (9,5 SD) ($p<0.001$) in group A, from 50 (22 SD) to 5,4 (8,6 SD) ($p<0.001$) in group B. Independent-samples T-test (2-tail, unequal variances) determined a significant difference in decrease of pressure gradients between both groups ($p<0,001$). Although no gradient reduction was encountered in some patients, angioplasty was considered successful on basis of angiography. Since registered pressure gradients are

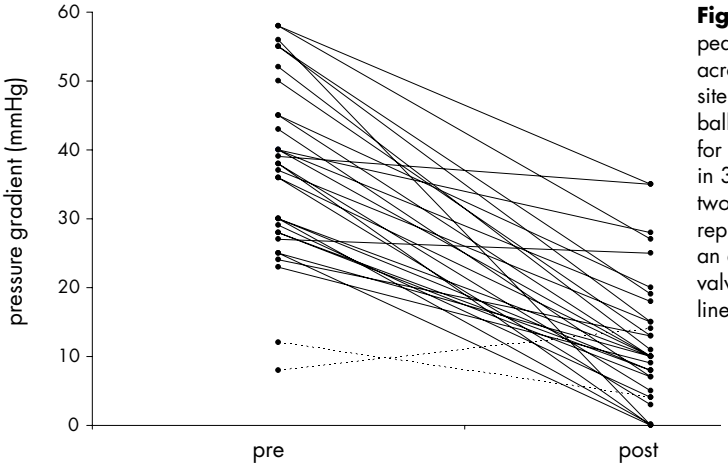

Figure 1. Peak to peak systolic gradient across coarctation site before and after balloon angioplasty for native coarctation in 32 children. The two dotted lines represent patients with an associated aortic valve stenosis. Several lines overlap.

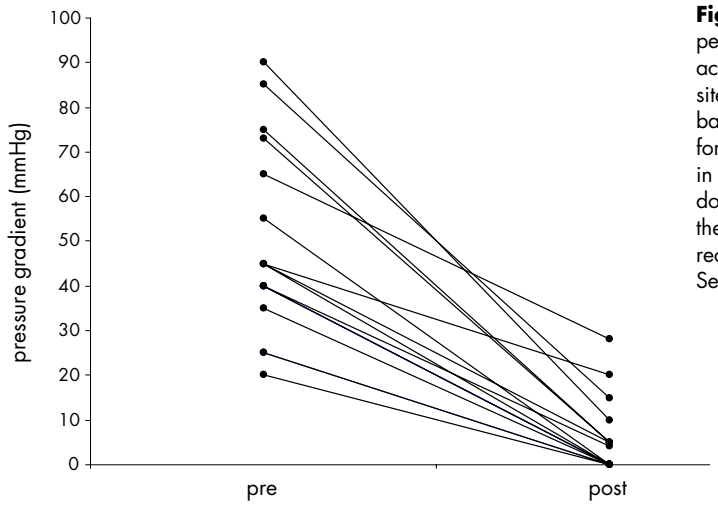

Figure 2. Peak to peak systolic gradient across coarctation site before and after balloon angioplasty for native coarctation in 17 adults. The dotted line represent the patient who received a stent. Several lines overlap.

meaningless addressing immediate success in patients with aortic valve stenosis, these values are excluded in the analysis. These two patients are depicted by dotted lines in the first figure (figures 1,2).

In 46 patients (94%), treatment was considered immediately successful. Three patients (6%) had an unsuccessful immediate result: two patients in group A. One of these children had a successful surgical repair after 1 month and the other was successfully redilatated after 3 months. One procedure was only partial successful in group B, low pressure gradient and a satisfactory clinical presentation in follow-up prevented re-intervention. Hospital stay varied from 12 to 48 hours for all patients.

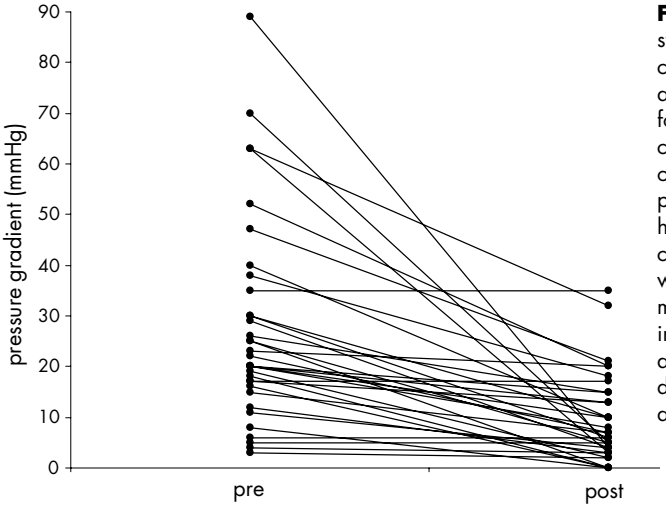

Figure 3. Peak to peak systolic gradient across coarctation site before and after balloon angioplasty for recoarctation in 33 children. Several lines overlap. A number of patients in this group had developed extensive collateral vasculature which leads to a misrepresentation of the initial pressure gradients and the pressure gradient drop during balloon angioplasty.

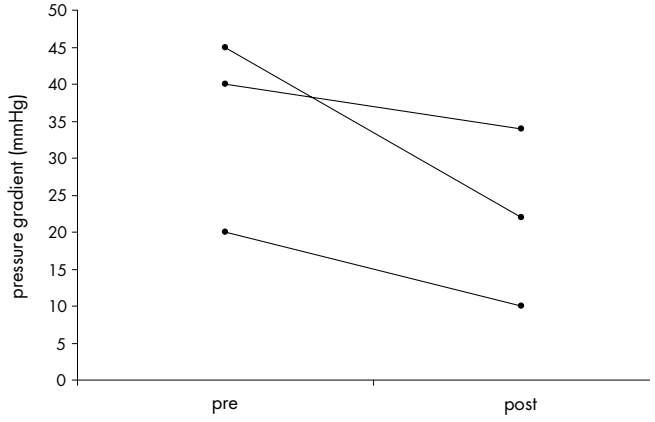

Figure 4. Peak to peak systolic gradient across coarctation site before and after balloon angioplasty for recoarctation in 3 adults.

Mean pressure gradient decreased from 27 (20 SD) to 9,5 (8,9 SD) ($p<0.001$) in group rCoA A and from 35 (13 SD) to 17 (6,1 SD) ($p<0,05$) in group rCoA B. Associated aortic valve stenosis prevented the registration of a meaningful gradient in one of the patients in group rCoA A. This patient is represented by a dotted line, these pressures were not included in analysis (figures 3,4).

Treatment was considered immediate successful in 31 (94%) patients in group rCoA A and failed to occur in two patients (6%). These both patients were managed surgically; a patch angioplasty was performed in both.

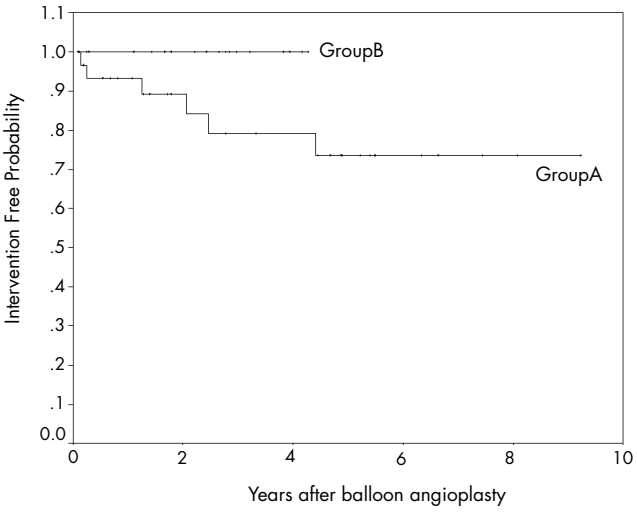

Figure 5. Graph showing re-intervention free survival after balloon angioplasty for native coarctation of the aorta in children and adults. No re-intervention took place in group B, the adult group. The child population is depicted by the lower line: A. In this group, the ratio of 78% at 2,5 years remained intervention free throughout 9-year follow-up period.

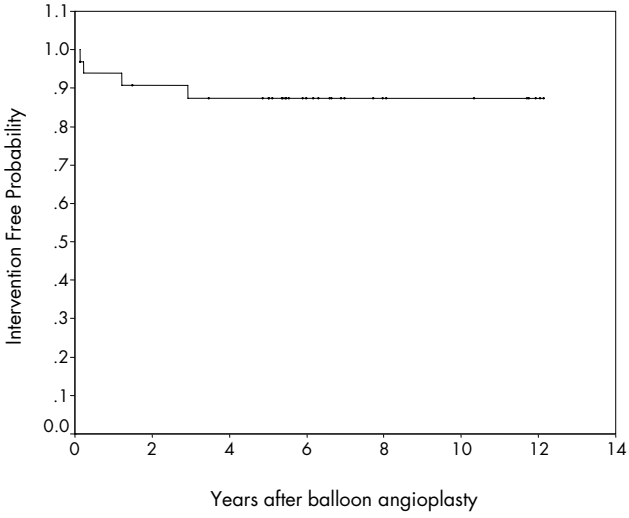

Figure 6. Graph showing re-intervention free survival after balloon angioplasty for recoarctation of the aorta in children.

One of 3 procedures turned out successful immediately in group rCoA B. In 1 patient resultant pressure gradient exceeded 40 mmHg. This patient was managed with patch angioplasty successfully. Low pressure gradients and a satisfactory clinical presentation in follow-up prevented re-intervention in the other patient after an insufficient angiographic result initially.

Follow-up

Results are summarized in an organogram (figure 7).
Follow-up of all 85 patients ranged from 6 months to 12 years (mean 4,9 years). Neither diastolic flow nor a pressure gradient exceeding 30 mmHg were registered in echocardiographic study in patients free from reintervention in follow-up. Of the 46 patients treated successfully initially in groups A and B, four (9%) developed a recoarctation. All recoarctations developed in the 30 patients (13%) who were immediate successful in group A. No recoarctations were encountered in group B. Patients with recoarctation underwent surgery (2 patients), or balloon angioplasty (2 patients). These reinterventions were all successful. Chi-square test for restenosis yielded no significant difference between groups A and B. The probabilities of reintervention after balloon angioplasty in groups A and B are depicted in Kaplan-Meier curves. The curves of groups A and B are not significantly different as determined by log rank analysis ($p = 0,089$) (figure 5).
One patient (3%) was treated for hypertension at angioplasty in group A, but after angioplasty blood pressure normalized and medication could be reduced. In group B, 13 patients (68%) were treated for hypertension at the time of balloon angioplasty. In 6 patients (46%) anti-hypertensive medication could be stopped, in 1 the medication was reduced and in another 6 (46%) anti-hypertensive medication had to be continued.
One of the 31 patients treated successfully in group rCoA A developed recoarctation in follow-up. Patch angioplasty was performed successfully in this patient. The probability of reintervention after balloon angioplasty in group rCoA A is depicted in a Kaplan-Meier curve (figure 6).

Aneurysms

All patients in groups A and B had ultrasound investigations, 17 (35%) had MRI, 19 (39%) conventional chest-röntgengraphy, 7 DVI (14%) and 8 (16%) had angiographic studies in follow-up. In groups rCoA A and rCoA B, all patients had ultrasound studies, 16 (44%) had MRI, 22 (61%) conventional chest-röntgengraphy and 9 (25%) had angiographic investigations in follow-up. No aneurysm formation was encountered.

Mortality

One death occurred in group rCoA A, not related to the angioplastic procedure. A Waterhousen-Friedrigsen sepsis developed in this patient 3 months after balloon angioplasty for re-coarctation.

Complications

Complications occurred in 2 of the 49 patients in groups A and B. In 1 patient artery-section was required to get access to the femoral artery. The second patient developed femoral artery occlusion, sufficient collateral circulation prevented further treatment and no difference in length of the legs was noted in follow-up.
No complications occurred in groups rCoA A and rCoA B. In 3/36 procedures balloon rupture occurred without consequences for the patients.

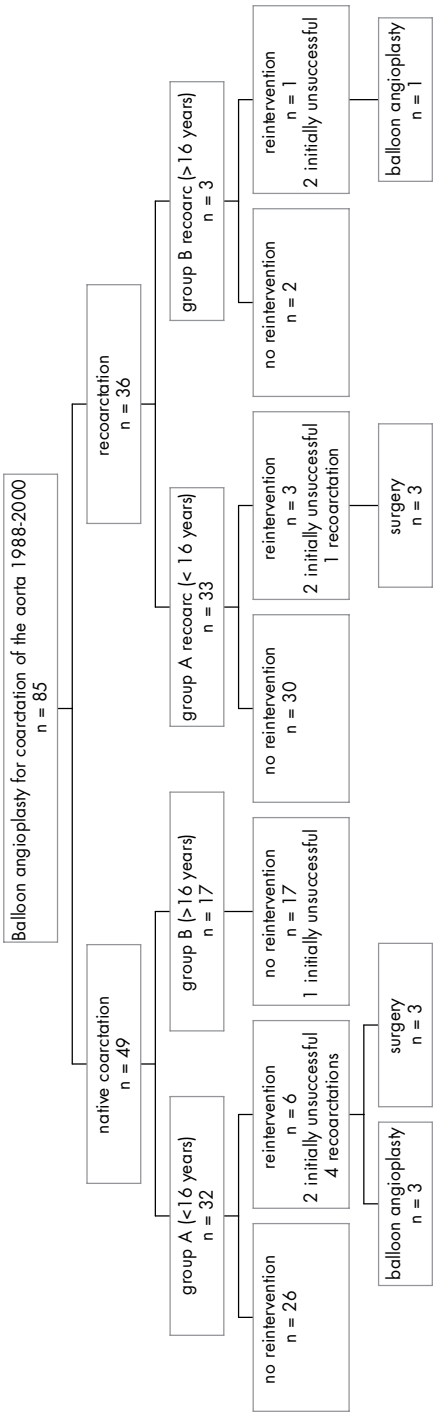

Figure 7. Organogram showing outcome of various types of interventions for coarctation of the aorta in various groups.

Discussion

Balloon angioplasty has proven to be an alternative for surgery in coarctation of the aorta in a selected group of patients, although concerns exist on the long-term follow-up in regard to recurrent or residual aortic coarctation. A number of investigators have suggested that age has an important impact on outcome. Some series have shown an increased risk of recurrence in very young patients.[4,11] Hellenbrand et al. however suggested that the age at angioplasty was inversely related to adequacy of angioplasty.[12] We propose that long standing aortic obstruction could be associated with increased fibrotic change in the aorta and therefore better results were to be obtained in younger patients in this series. Excellent results for angioplasty in adults were reported by Fawzy et al. and they suggested that advanced age at dilatation could explain the low incidence of coarctation recurrence.[8] These different reports evoke the question whether the management of coarctation in children and adults should be different.

Immediate results

In attributing success to procedures, angiography was considered more important than the resulting pressure gradient. Different from gradients post dilatation, angiographic results cannot be underestimated in the presence of extensive collateral vasculature. Furthermore, the decision to perform reinterventions could not be based on temporarily excessively high gradients, as were encountered in some of the patients in our series. Instead thereof, in those cases in which the angiographic result was satisfactory but the pressure gradient remained over 20 mmHg, the procedures were considered successful and were not repeated with a larger balloon in the absence of remaining waist which could be dilated further. These decisions proved to be justified by the follow-up studies in which no further residual gradients were encountered in these patients, probably secondary to remodelling, as proposed by Ebels.[13] Gradient reduction ranging from 31-35 mmHg in children[3,4,11] and from 34-57 mmHg in adults[8,9,14] are reported. Significantly larger pressure gradients decreases after balloon angioplasty in adults (44 mmHg), compared to children (23 mmHg) were also encountered in this study. The existence of both higher gradients before, as well as lower gradients after dilatation, enhances the pressure gradient drop in adults. This is, at least partially, caused by higher blood pressures in adults, so that percent reduction is limited. The resulting lower gradients after dilatation may be explained by the larger absolute surface gain that is obtained in adults in the angioplasty procedure, which can be attributed to the elastic recoil of the aortic wall, occurring predominantly in children and less in adults who have probably more fibrotic tissue in the aortic wall.

Recoarctation

Recoarctation is the prevailing long-term complication after angioplasty and surgical repair. Especially in children and infants recoarctation rates after balloon angioplasty may range from 15-30%. For this reason we excluded this procedure in infants younger than 3 months, in whom the risk is unacceptably large.[3,4] This resulted in an

overall recoarctation rate of 14% in our population. But even when this precaution is taken, recoarctation is less common in adults, in whom no recoarctations were encountered in follow-up. This is in accordance with several studies reporting that the risk of recoarctation decreases gradually with increasing age,[3,11,15,16] probably secondary to the different build up of the vessel wall with decreasing elasticity.
A number of patients in this category had developed extensive collateral vasculature which leads to a misrepresentation of the initial pressure gradients and the pressure gradient drop during balloon angioplasty.

Aneurysm formation

Aneurysm formation at dilatation site has remained a long-term concern, although a gradual decrease in the incidence of aneurysm formation has been reported. Early studies noted a significant incidence of 50%,[17] which decreased significantly, varying from 0-6% in later studies.[7,18,19] No aneurysm was encountered in the follow-up. Accordingly, all groups appear to have comparable low incidence of aneurysm formation. This finding would coincide with Ritter et al., who postulate that the incidence of formation of aneurysms is similar in children and adults.[20]
No indication exists that the natural history of aneurysms occurring after balloon dilatation of coarctation in adults is different for that in children. Media tear and cystic medial necrosis have been postulated as cause of aneurysm formation.[6] Sohn et al. demonstrated remodeling of dilated vessels.[21] It has been suggested that aneurysm development may be due to use of an overstressed balloon.[18] This probably explains the decrease in aneurysm formation over time. The larger the balloon, the better the gradient relief, but the higher the chance on medial tear. We therefore use a catheter of which the size 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 by more than 2 mm.

Hypertension

Reduction in blood pressure will be smaller and take longer if the procedure is carried out at increasing age.[22] This has been illustrated evidently in our series. Hypertension persisted in none of the children, whereas medication had to be continued in 6 out of 18 adults (33%). Systemic hypertension postoperatively, in the absence of residual coarctation, appears to be related to the duration of preoperative hypertension, which can be explained by an insufficient resetting of the baroreceptor postoperatively.

Treatment for recoarctation

Satisfactory results have been reported on angioplasty for recoarctation in adults.[23] Nevertheless, angioplasty failed in adult recoarctation in our patient population, which included 3 patients who underwent surgery. Trying to explain this failure, we could think of two unfavorable conditions, compromising balloon angioplasty in these patients: We hypothize that the adult restenosis has a stiff and fibrotic aortic wall, since the adult aorta would be expected to have less elasticity than an growing aorta. Additionally, scarring is probably more extensive following surgery, compared to angioplasty.

Patients with a recoarctation after primary angioplasty underwent surgery (2 patients), or secondary angioplasty (2 patients). The choice for either surgery or balloon angioplasty was based upon the gravity of stenosis: patients with a severe restenosis were operated on, while in less prompt and severe restenoses, patients underwent angioplasty. Both surgery and angioplasty turned out successful in long term follow-up. Although we did not obtain data on this subject, we propose that severe restenosis should be managed surgically, like a long-segment coarctation indicates surgical management in native coarctation.

Stent-implantation

Two possible mechanisms of recurrence include elastic recoil of tissue after angioplasty and the presence of long segment narrowing. Stents provide support to recoil of the dilated segment.[24] Implantation of stents in older children and adults in whom the aorta size has attained adult size is not problematic, although such may be problematic in infants and children, whose aorta is not totally developed. The indications for stent implantation are not clearly defined.[25] Possible indications are long segment coarctation and tortuous coarctation. We used 1 stent in an adult patient on request of the referring physician only.

One child developed femoral artery occlusion. This may be associated with intimal trauma as the balloon is withdrawn from the femoral artery. Care during the intervention will minimize this complication.

Limitations of the study

This study has several limitations: the small number of patients, the retrospective nature of this study and a limited mean follow-up of 4,9 years. Two patients in group rCoA A were excluded from balloon angioplasty because of severe restenosis, thereby introducing selection of patients in this group.

Conclusions

Balloon angioplasty for native coarctation works with a similar technical approach equally well in children and adults and warrants its role as first-choice therapy for this diagnose group. Balloon angioplasty in recoarctation can only be recommended in the pediatric age group.

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