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Chapter 10

Summary, conclusions and future perspectives

Adapted from:
Advances in the management and surveillance of patients with aortic coarctation

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The introduction of balloon angioplasty for aortic coarctation in children, adolescents and adults prompted the initiation of this thesis. Although the long-term results of surgical repair had been improving over the years, the less invasive character of angioplasty and encouraging initial results of this technique triggered its widespread introduction. However, uncertainty existed regarding long-term risks of aneurysm formation and recoarctation for these patients. More research on long-term results had been advocated in early reports on angioplasty for coarctation for this reason. Chapter 2 describes the single-institution results of balloon angioplasty as the primary treatment of coarctation of the aorta in 30 children, managed for this condition during a period of 10 years from its introduction in 1990. No mortality occurred and 28 of 30 procedures were successful immediately, with a significant reduction in mean pressure gradient of 23 mmHg. Four patients (13%) developed a recoarctation. No aneurysm formation was encountered. These excellent long-term results demonstrate that balloon angioplasty for the treatment of native coarctation of the aorta in children may be an efficient and safe solution. Whether angioplasty can be performed as effectively and safely at adult age has been a matter of debate. Results of angioplasty were therefore compared for different aged patients and are summarized and discussed in chapter 3, including those of angioplasty for recoarctation. Although pressure gradient drops were significantly higher for adults in native coarctation management, both immediate and long-term results appeared to be not different for adults as compared to children for native and recoarctation, with an average follow-up duration of 4.9 years. In conclusion, balloon angioplasty for native coarctation has a similar performance with an identical technical approach in children and adults. Due to the limited number of procedures performed, balloon angioplasty in recoarctation can only be recommended in the pediatric age group. These first studies, comprising chapters 2 & 3, were performed several years before the publication of this thesis. Since then, the authors did not analyse the follow-up results in these particular patients systematically thereafter. There has been an explosion of reports of endovascular treatment of both native and recoarctation in other institutions in the last 10 years. Most of these cite satisfactory results, although certain morbidity in most, and mortality in some of these series has been encountered. Based on findings in literature and those in our institutions, risks should be accounted for in the approach of patients with native and recurrent coarctation. The counselling of patients and their parents should be accurate in this respect. Before comparing angioplasty to surgical repair in selected patients, long-term results of the main two types of surgical repair were evaluated for an unselected population of 262 patients with aortic coarctation during a 28-year period. The results thereof are described in chapter 4. Not surprisingly, the presence of complex intracardiac malformations appeared to be associated with mortality in these patients. Another, probably more remarkable finding was that if ridge resection was involved in surgical repair by patch aortoplasty, using polytetrafluoroethylene material, this technique appeared to be associated with more aneurysm formation and late hypertension than resection and end-to-end anastomosis. Arch hypoplasia and age up to one month at repair were associated with recoarctation. Chapter 5 comprises the comparison of
surgical repair with balloon angioplasty for the specific group of patients between 3 months to 16 years of age with an isolated, discrete type of coarctation. Although satisfactory long-term results can be expected for both management types in this relatively benign patient population, comparative data had been limited. During follow-up, which averaged a period of longer than 5 years, immediate success rates and late reintervention probabilities were not found to be different between these techniques. Obviously, hospital stay was longer following surgical repair. Chapter 6 describes the finding of an aortic aneurysm in an asymptomatic 43-year-old male, who was managed by Dacron patch aortoplasty for native coarctation of the aorta 25 years before. This case demonstrates the need of lifelong surveillance after coarctation repair. Furthermore, the role of MR angiography is highlighted. Acute and late aneurysm formation has been an important concern about balloon angioplasty performed in adolescents and adults with native aortic coarctation. Chapter 7 reports a series of adult patients that were managed by balloon angioplasty. Although intima tearing was encountered in 8 of 29 procedures immediately, no aneurysm formation was found in follow-up angiography and MRI.

High cardiovascular complication rates have been reported in patients with native coarctation, despite initial success of management. To elucidate whether this finding is associated with different vascular characteristics, aortic flow properties of such patients were investigated by applanation tonometry and MR-measurements. Chapter 8 focuses on 11 patients following successful initial coarctation management in whom pulse wave was analyzed using applanation tonometry. Results thereof were compared to those in 14 matched controls. The objective of this study was to identify flow characteristics that may be responsible for premature vascular damage that is encountered by these patients. An earlier return of aortic wave reflection was found in patients, compared to controls. Unfavorable augmentation of the maximum systolic pressure and lengthening of the systolic period were found, which corresponds to the premature wave reflection. These findings provide an additional explanation for high cardiovascular complication rates, despite anatomically satisfactory coarctation management. Chapter 9 reviews the measurement of proximal and distal aortic properties with magnetic resonance in adults after successful coarctation management with the same purpose as chapter 8. Again, results were compared between patients and matched controls. Although not found in distal segments of the thoracic aorta, lower distensibility and higher pulse wave velocity of the ascending aorta were found in patients. These unfavourable alterations have been described in experimental and native coarctation in neonates and those surviving neonatal coarctation repair. This study demonstrates these changes also in patients in whom coarctation has been managed successfully after childhood. This warrants a close and lifelong monitoring of the cardiovascular risks of these patients.
Chapter 10

Study limitations and recommendations for further research

The most important limitations of the studies outlined above are the non-randomized, retrospective study designs and relatively small study samples. Furthermore, all patients were recruited at one institution. So far, just one single-institution clinical randomized trial for patients with native coarctation has been reported. Possible barriers for a multicentre prospective trial relate to differences in treatment approaches between centres and the absence of a systematic centralized database. The creation and funding of a National registry and DNA-bank of adult patients with congenital heart disease (CONCOR) in the Netherlands in 2002 may facilitate future research in this respect. More prospective clinical trials are necessary to determine whether angioplasty compares favourably to surgical repair for selected patients with aortic coarctation and should be adopted as first-choice procedure due to its less invasive character. Meanwhile, balloon angioplasty may provide a safe and effective alternative to surgical repair of unoperated coarctation of the aorta in both adults and children beyond infancy, without hypoplasia of the aortic arch or isthmus. Another limitation of the studies in this thesis is the absence of aortic stenting for this condition in these series. This technique has been in vogue recently. Although satisfactory long-term results after stenting have been reported, no data are available at present to justify the choice of stenting in favour of balloon angioplasty. Again, the superior endovascular approach could probably be identified by a randomized trial. Another suggestion for future research would be the assessment of characterise aortic flow and jet velocity in ascending and descending aortic segments in patients after successful coarctation management with normal blood pressure at rest. The mechanism of exercise systolic hypertension that is frequently found is not completely understood. Doppler echocardiography during exercise and cardiac catheterization with catecholamine infusion, as a surrogate of exercise, have been applied for this purpose. Hypertension during exertion was found to be correlated to descending aortic peak systolic velocity at peak exercise. Magnetic resonance imaging and flow quantification with increased cardiac output have not been applied yet in this context. Its superior imaging quality and its non-invasive character would be appropriate for a study of aortic flow at rest and during catecholamine infusion. In addition to sophisticated measurements of vascular function by applanation tonometry and magnetic resonance, simpler and readily available indicators of cardiovascular risk can probably be developed. The role of ambulatory blood pressure monitoring in the assessment of vascular function, for example, may be particularly promising in this respect.