SURGERY FOR CONGENITAL HEART DISEASE

LEFT HEART HYPOPLASIA AND NEONATAL AORTIC ARCH OBSTRUCTION: IS THE RHODES LEFT VENTRICULAR ADEQUACY SCORE APPLICABLE?

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Objective: Although the influence of small left heart structures on outcome of a biventricular repair in neonatal critical aortic stenosis is well documented, little is known about its effect in neonates with aortic arch obstruction and coarctation. The purpose of this study was to evaluate the influence of small left heart structures on early and late results of repair and the ability to achieve a biventricular repair in neonates with coarctation and aortic arch obstruction. **Patients:** Neonates included in this study had a left ventricular adequacy score (as proposed by Rhodes and associates for critical aortic stenosis) that would have predicted a need for a univentricular (Norwood) repair. All were ductus dependent but had antegrade ascending aortic flow and a small but nonstenotic aortic valve (<30 mm Hg gradient). Twenty neonates aged 10 ± 9 days were identified for the study with weights averaging 3.1 ± 0.6 kg. Selected left heart measurements obtained by preoperative echocardiography included the following: aortic anulus 5.3 ± 0.3 mm, mitral anulus 8.4 ± 1.0 mm, transverse aortic arch 3.4 ± 0.6 mm, and left ventricular volume 25 ± 4 mL/m². All patients underwent coarctation repair by resection and extended end-to-end anastomosis to enlarge the transverse arch as needed. Three patients underwent simultaneous pulmonary artery banding because of a hemodynamically significant ventricular septal defect. These 3 patients have subsequently had their defects successfully closed without mortality. **Results:** There were no early or late deaths at a follow-up of 38 ± 16 months after the operation. Three patients (3/20, 15%) have had to undergo reintervention with balloon aortoplasty because of recurrent coarctation (gradient > 20 mm Hg) in 2 and resection of subaortic stenosis in 1. Late follow-up in the remaining patients reveals 1 with moderate subaortic stenosis (gradient = 43 mm Hg), 2 with mild aortic stenosis (gradient < 30 mm Hg), and 2 with mild to moderate mitral stenosis. At late follow-up, 16 patients (16/20, 80%) are completely free of symptoms and 4 (4/20, 20%) have mild residual symptoms. **Conclusions:** Biventricular physiology can be successfully achieved in neonates with small left heart structures and aortic arch obstruction with minimal mortality and excellent late function-

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al results. Standard echocardiographic measurements used to predict the need for a univentricular repair in critical aortic stenosis are not valid for the neonate with aortic arch obstruction. (J Thorac Cardio-vasc Surg 1999;118:81-6)

Neonatal coarctation of the aorta and aortic arch obstruction are frequently associated with other forms of left heart obstruction including aortic stenosis (AS), aortic annular hypoplasia, mitral stenosis, and left ventricular hypoplasia.¹ These coexisting abnormalities have generally been thought to have a negative effect on coarctation repair and, in extreme circumstances, may necessitate conversion to single ventricle physiology or even cardiac transplantation.² Rhodes and colleagues³ have described a left ventricular (LV) adequacy score that uses measurements of body size and left heart structures to predict biventricular repair in critical AS.³ Others have described a variety of other factors predictive of success with biventricular repair with critical AS and hypoplasia of left heart structures.⁴ These factors have been suggested by some to predict success for infants with coarctation of the aorta and aortic arch obstruction, but with very little objective data supporting this extension of their use.^{2,3,5}

The purpose of this study was to examine the surgical outcome and long-term survival of a subgroup of neonates and infants with coarctation of the aorta and aortic arch obstruction and varying degrees of left heart hypoplasia.

Patients and methods

We reviewed the hospital records of all neonates (≤ 30 days of age) with the diagnosis of coarctation of the aorta between January 1993 and April 1997. The following inclusion criteria were used to identify study patients: (1) diagnosis of coarctation of the aorta leading to surgical repair in the first month of life, (2) patency of all 4 cardiac valves, (3) normally related great vessels, (4) absence of a true atrial septal defect or anomalous pulmonary venous drainage, (5) absence of significant aortic valve or subaortic stenosis (>20 mm Hg Doppler echocardiographic gradient), (6) ductal dependency, and (7) an LV adequacy score, according to the criteria of Rhodes, that would have predicted the need for a univentricular repair in the setting of critical AS (score < -0.35). Patients with a coexisting ventricular septal defect (VSD) or bicuspid aortic valve were included in the study. The medical records of each patient were reviewed to determine patient characteristics, including age at diagnosis and operation, weight, body surface area, outcome, and functional status at follow-up. All patients underwent a complete preoperative 2-dimensional, M-mode, and Doppler echocardiogram with the

Acuson 128XP/10 ultrasound system (Acuson, Mountain View, Calif) recorded on 0.5-inch videotape. An LV adequacy score was calculated according to the method of Rhodes and colleagues.³ This method combines 4 factors to determine a score that is predictive of death after biventricular repair for neonatal critical AS (< -0.35) and includes body surface area (BSA), aortic root dimension indexed to BSA, the ratio of the length of the LV to the length of the heart (long-axis ratio, or LAR), and the mitral valve area indexed to the BSA (MVAi):

$$\text{Score} = 14.0 (\text{BSA}) + 0.943 (\text{ROOTi}) + 4.78 (\text{LAR}) + 0.157 (\text{MVAi}) - 12.03$$

In addition to the echocardiographic measurements taken for the calculation of the LV adequacy score, the aortic annulus diameter, mitral annulus diameter, distal transverse arch diameter measured between the left carotid and subclavian arteries, and LV volume, calculated by the hemisphere cylinder or bullet method,⁶ were measured. The LV outflow tract was evaluated by Doppler ultrasound, and any patient with an aortic valvular or subvalvular gradient greater than 20 mm Hg was excluded from the study, as defined previously. The echocardiogram was also used to evaluate ductal shunting, associated intracardiac defects, and the direction of flow in the ascending aorta.

Surgical technique. Standard surgical techniques were performed by means of a left thoracotomy approach and extended end-to-end anastomosis.⁵ The anastomosis was routinely carried proximal to the left subclavian artery onto the transverse arch between the left carotid and subclavian arteries. Even more extension was done proximal to the left carotid artery, when dictated by the arch anatomy and hypoplasia. Concomitant pulmonary artery banding was performed in the case of multiple VSDs or a single large VSD that seemed to be in a difficult location to close (mid or low muscular). Patients with small to moderate-sized VSDs or large VSDs in accessible areas (perimembranous) were not banded and underwent subsequent VSD closure when the patient status indicated.

Follow-up. Postoperative follow-up was obtained in all patients by direct visits with attending cardiologists. At least 1 year's follow-up was available for all patients. Complete 2-dimensional and Doppler echocardiograms were obtained in all patients at late follow-up. Postoperative residual coarctation gradients were obtained by Doppler estimation of the aortic arch gradient and measurement of arm and leg blood pressures. A gradient greater than 20 mm Hg by either method was used to define recurrent coarctation. Late functional status was determined by patient examination and parental questioning at the most recent follow-up visit with

Table I. Patient characteristics and echocardiographic measurements

Patient No.	BSA (m ²)	MV area (cm ² /m ²)	Aortic root (cm/m ²)	LAR	LV adequacy score	Aortic anulus (mm)	Mitral anulus (mm)	Transverse aortic arch (mm)	LV volume (mL/m ²)
1	0.24	2.2	3.4	0.98	-0.49	6.3	8.2	3.5	13.8
2	0.20	2.0	3.2	1.0	-1.12	4.7	7.2	4.2	14.5
3	0.21	2.3	3.7	1.0	-0.45	5.6	7.8	3.5	14.8
4	0.23	3.1	3.3	0.9	-0.91	6.5	9.5	4.0	37.3
5	0.22	3.0	3.9	0.92	-0.43	6.3	9.3	3.8	17.7
6	0.19	3.8	3.9	0.95	-0.51	6.2	9.7	4.5	63.2
7	0.21	2.4	3.2	0.99	-0.92	5.0	8.1	2.5	17.9
8	0.21	3.0	3.8	0.89	-0.77	5.0	9.1	3.1	23.2
9	0.16	2.4	3.2	0.98	-1.73	4.0	7.0	2.3	19.5
10	0.16	3.0	4.3	0.91	-0.95	4.2	7.9	3.3	18.1
11	0.23	2.8	3.0	0.96	-0.95	5.5	9.1	4.0	23.5
12	0.26	2.1	2.8	0.98	-0.69	5.4	8.5	3.1	27.7
13	0.20	2.0	3.9	1.01	-0.41	5.3	7.2	3.4	30.6
14	0.19	3.6	3.2	0.92	-1.37	5.5	9.4	3.3	—
15	0.24	1.9	3.5	0.99	-0.37	6.3	7.7	3.0	29.3
16	0.21	3.7	3.1	1.0	-0.82	5.0	10.0	4.0	31.9
17	0.20	1.9	3.9	1.0	-0.48	4.8	6.9	3.0	14.5
18	0.18	2.1	3.2	0.89	-1.94	4.5	7.0	2.5	36.7
19	0.25	3.0	3.0	0.9	-0.92	5.7	9.9	5.1	35.4
20	0.30	2.1	2.6	0.93	-0.64	5.7	9.0	3.5	19.0
X ± SD	0.21 ± 0.03	2.6 ± 0.6	3.4 ± 0.4	0.96 ± 0.04	-0.84 ± 0.4	5.3 ± 0.8	8.4 ± 1.0	3.5 ± 0.6	25 ± 12

BSA, Body surface area; MV, mitral valve; LAR, long-axis ratio; LV, left ventricular; X ± SD, mean ± standard deviation.

the attending cardiologist. *Excellent functional status* was defined as no symptoms conveyed by the parents and no exercise intolerance. *Good functional status* was defined as no symptoms at rest and only mild symptoms with strenuous exertion. *Moderate functional status* was defined as symptoms with only minimal to moderate activity. *Poor functional status* was defined as symptoms at rest.

Statistics. All echocardiographic measurements were made in triplicate and averaged. All values are reported as mean ± standard deviation. Median and ranges for values are also included where applicable. Actuarial freedom from reintervention was calculated according to the method of Kaplan and Meier.⁷

Results

A total of 20 patients were identified who met the inclusion criteria during the study period (January 1993–April 1997). These study patients represented 22% (20/93) of all neonates who underwent repair of coarctation during this period (73 patients were excluded). Patient characteristics and echocardiographic measurements are shown in Table I. Eight patients had a VSD. All patients had bidirectional shunting at the ductal level and evidence of antegrade flow in the ascending aorta. In total, 8 patients had a mitral anulus diameter less than 8 mm, 8 patients had an aortic anulus diameter less than 5.5 mm, and 9 patients had a left ventricular volume less than 20 mL/m².

Seventeen patients underwent repair of the coarctation alone and 3 patients underwent simultaneous pulmonary artery banding. There were no operative deaths and no late deaths at a mean follow-up of 38 ± 16 months after the operation (range 11–64 months). Two patients (2/20, 10%) have recurrent coarctation or arch obstruction (>20 mm Hg gradient) and have required late balloon aortoplasty 4 and 44 months after the operation. Four patients have LV outflow obstruction on late echocardiographic follow-up, with severe subaortic stenosis in 1 (76 mm Hg gradient), mild AS in 2 (gradient < 30 mm Hg), and moderate subaortic stenosis in 1 patient (gradient = 43 mm Hg). The patient with severe subaortic stenosis has required late resection of subaortic stenosis and patch repair of mild residual arch obstruction 14 months after initial coarctation repair. Mild mitral valve stenosis is present in 2 patients (mean mitral diastolic gradient < 6 mm Hg). All 3 patients who underwent initial pulmonary artery banding have undergone successful band removal and VSD closure without early or late mortality. Freedom from reintervention for any reason (surgical or balloon aortoplasty) is 85% ± 9% at 1 year, 80% ± 10% at 2 years, and 70% ± 16% at 4 years after the operation.

Late functional status is excellent in 16 patients (16/20, 80%) and good in 4 patients (4/20, 20%).

Discussion

Neonatal coarctation of the aorta is often associated with various left heart obstructive lesions including AS, aortic annular hypoplasia, subaortic stenosis, mitral valve stenosis or annular hypoplasia, and LV hypoplasia. The presence of one or more of these anomalies is associated with increased risk of early mortality in many series.^{1,2,5,8,9}

Rhodes and colleagues³ have emphasized the cumulative effect of these abnormalities in critical AS and the ability to predict death after a biventricular repair. In addition to the scoring system by Rhodes and associates, others have attempted to define critical values for left heart obstructive lesions such as critical AS that preclude biventricular repair, including aortic anulus size less than 6 mm, mitral anulus size less than 9 mm, and an LV volume less than 20 to 25 mL/m².¹⁰⁻¹² The alternative to biventricular repair in children with multiple left heart obstructive lesions including aortic arch obstruction is Norwood palliation and long-term conversion to single ventricle physiology. Despite improving results with this approach, long-term functional results are better with biventricular physiology, and the difficult dilemma is deciding which neonates can undergo successful biventricular repair.

In this study, we attempted to select the neonates with coarctation who had the most severe degrees of left heart hypoplasia and in some instances reflect the difficult decision-making process of single ventricle palliation versus biventricular repair. All of these neonates were ductus dependent and had an echocardiographic LV adequacy score on the Rhodes scale of less than -0.35, predicting a high mortality for biventricular repair in critical AS. We purposely excluded patients with significant intrinsic aortic valve disease, because this group has been extensively studied and we were attempting to determine whether there was a limit on the size of the left heart structure that might predict death or preclude biventricular physiology in coarctation of the aorta. Perhaps the critical factor in the success in this series was the fact that all of the neonates had evidence of antegrade flow in the ascending aorta and bidirectional shunting at the ductal level (rather than solely right-to-left shunting), indicating that hemodynamic and physiologic considerations are as important in determining success in biventricular physiology as left heart morphologic criteria.^{12,13}

The presence of left heart hypoplasia and coexisting left heart obstructive lesions has had a significant negative impact on operative mortality in some surgical series. In the Boston series, 12 of the 14 deaths in 98 neonates occurred in patients with "various left-sided

obstructive lesions," which were not specified as to type or severity.¹⁴ In the large multi-institutional study by the Congenital Heart Surgeons' Society, coexisting left heart obstructing lesions including mitral valve anomalies, aortic annular hypoplasia, and LV hypoplasia were significant contributors to death after neonatal repair of coarctation.¹ Lacour-Gayet and associates⁹ described 5 patients with a hypoplastic LV that they defined as an aortic anulus between 5.5 and 6.5 mm; three of the patients died. It is difficult to compare patients between surgical series, and we can only speculate that there may be differences in patient types explaining the 100% survival in our study as compared with other series of coarctation repair in the setting of multiple left heart obstructive lesions. Another explanation is that our study excluded infants with intrinsic AS, and it is possible that the presence of hypoplastic, but nonstenotic valves or LV hypoplasia does not necessarily have as poor a prognosis. The presence of right-to-left shunting at the ductal level, left-to-right shunting at the atrial level, and a hypertensive right ventricle may contribute to underfilling of the neonatal LV in coarctation and may alter all of the left heart measurements, including the mitral and aortic anulus LV volume measurements.⁶ We attempted to control for some of these left heart loading factors by excluding those neonates with real atrial septal defects. Similar parallels exist in interrupted aortic arch with VSD, in which loading conditions may alter LV volume and LV outflow tract size and biventricular repair is possible even with very small aortic or subaortic sizes.¹⁵ These same loading conditions may not be present in neonatal critical AS because of the hypertensive LV, making morphologic parameters and Rhodes criteria more applicable in the hemodynamic and physiologic conditions of neonatal critical AS than the conditions in neonatal coarctation.

Reintervention was needed in the relatively high number—6 reinterventions in 5 patients overall. Three patients underwent planned reintervention with debanding of the pulmonary artery and successful VSD closure. Although 1-stage coarctation repair and VSD closure has been advocated by some,⁵ we have been satisfied with the results of a selective approach of banding multiple or particularly difficult VSDs and later closure. In the large multi-institutional study from the Congenital Heart Surgeons' Society, this approach was associated with the highest survival for neonates with coarctation and VSD.¹ To date, 2 patients have undergone balloon dilation for recoarctation, 1 before debanding and VSD closure. Although this is not an inordinately high incidence of recoarctation, neonates

with relatively severe aortic arch obstruction and small transverse arches have a higher incidence of late recoarctation.⁵ One last patient has undergone successful reoperation for severe subaortic stenosis and recurrent coarctation 15 months after coarctation repair. Similarly, another patient has moderate subaortic stenosis and 2 others have mild to moderate degrees of mitral stenosis, with an unknown long-term prognosis. It is our impression to date that this selected group of neonates with left heart obstruction will have a relatively high need for late reinterventions for reasons related to development of LV outflow obstruction or mitral valve stenosis. Tchervenkov and associates¹⁶ have also seen a relatively high rate of reintervention in a similar group of neonates with aortic arch obstruction successfully undergoing biventricular repair.

In conclusion, neonates with coarctation and aortic arch obstruction can have severely hypoplastic, but nonstenotic left heart anomalies and relatively small LV volumes as evaluated by traditional or standard morphologic criteria. Morphologic measurements and the LV adequacy score used for critical AS are not necessarily helpful in predicting a successful biventricular outcome in these neonates. Decisions regarding surgical options should also include hemodynamic and physiologic considerations such as the presence of antegrade flow in the ascending aorta and the absence of significant LV outflow tract gradients in considering biventricular repair in neonates with coarctation.

Within the range of values we examined in this study, we did not find minimum values of mitral or aortic valve size or LV volume that were predictive of death with a biventricular approach. Although there undoubtedly is a minimum value of left heart sizes compatible with biventricular physiology, we believe that a biventricular approach is possible in almost all neonates with coarctation, no significant LV outflow gradient (<20 mm Hg), and antegrade ascending aortic flow. The sizes of left heart structures incompatible with a biventricular approach are likely to be associated with a significant LV outflow gradient, in which the Rhodes LV adequacy score has been proven to be of predictive value.³ Although the short- and intermediate-term functional results are good with simple relief of aortic arch obstruction in these neonates, cautious optimism should be adopted for the long-term results and the development of late significant left heart obstructive lesions.

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Discussion

Dr Vaughn A. Starnes (*Los Angeles, Calif*). Dr Hawkins and his group should be congratulated on calling our attention to yet another subset of infants with extremely difficult lesions to treat—coarctation of the aorta and left heart hypoplasia. Although formulas are important, when we look at issues of neonatal critical AS, they are not totally applicable to the group being analyzed here.

The two functional parameters that Dr Hawkins has pointed

out, antegrade aortic flow and bidirectional shunting at the ductal level, have defined ventricles that are adequate for biventricular repair. Rhodes' formulas do not take this into account.

I have some questions for Dr Hawkins. One of the issues that we have when we evaluate this group of children, as we did when we looked at Rhodes' group, is that these are all retrospective analyses. Some selection does occur in retrospective analyses. You had 93 infants with coarctation of the aorta and hypoplasia from whom you selected 20, and you based your selection on the Rhodes adequacy score.

Did this group of children have combined aortic and valvular disease? Did the majority of these children have inadequate aortic sizes or inadequate mitral sizes as a combination?

Dr Hawkins. We primarily selected these patients on the basis of the so-called Rhodes score because we frequently quote this paper and attempt to base our decisions on it. However, whether the patients had both aortic and mitral hypoplasia, I cannot say. I do not have the data available to answer that question appropriately.

Dr Starnes. That group of children is difficult to treat and probably is better served with some form of palliative operation. One of the things that we face, of course, is trying to predict mortality with a palliative or transplant operation versus biventricular repair. We all like to perform a biventricular

repair because we think that offers the best functional outcome. I have done biventricular repairs in this high-risk group of children. What parameters are you following to determine the accuracy of repair? For example, we can have a small LV, small mitral valve, and small aorta, but we also can have a noncompliant LV, severe left atrial hypertension, and severe pulmonary hypertension. That would not necessarily be reflected in the mortality scores in the intermediate follow-up, but such a child with severe pulmonary hypertension might die 5 or 6 years later. Are you following these parameters?

Dr Hawkins. Yes, we are following these parameters. I think you are correct. I think that may be the limiting factor. In the conclusion to the article, I stated that the jury is not in yet on these patients over the long term. In the 1 patient who underwent artery construction and subaortic resection, left atrial pressures were very high for several days, but ultimately slowly came down. These patients do have restricted LVs, but at least my personal philosophy is that an LV that is small or somewhat restrictive may be better than univentricular palliation, at least in the high altitude of Salt Lake City.

Dr Starnes. This was a thoughtful paper. You brought to us yet two more parameters that we can use to make decisions on children with a small LV, that is, antegrade aortic flow and bidirectional shunting at the ductal level.