SURGERY FOR CONGENITAL HEART DISEASE

RECURRENT AORTIC COARCTATION: IS SURGICAL REPAIR STILL THE GOLD STANDARD?

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Objective: We reviewed our experience with surgical repair compared with balloon aortoplasty of recurrent coarctations of the aorta. **Methods:** This is a retrospective review of 1 institution's 27-year experience with surgical repair of recurrent aortic coarctation. A thorough chart review was performed of all pediatric patients undergoing surgical repair for recurrent aortic coarctation ($n = 56$) from January 1970 through July 1996. **Results:** The vast majority of recoarctations were repaired with a prosthetic patch technique, with a greater than 96% success rate. No deaths or major complications occurred in the 56 patients. Although a direct comparison with balloon aortoplasty cannot be done, we have reviewed the data available in the literature and found higher complication rates and lower success rates than we obtained in our series. **Conclusions:** Although the treatment of aortic coarctation has improved significantly during the past decades, persistent hypertension after repairs at an older age and recurrent coarctation after repairs in neonates occur in all institutions. Surgeons have not agreed on the optimal approach to primary coarctation repair, and invasive cardiologists have challenged operative intervention for both recurrent and primary coarctation. This study demonstrates that surgical repair of recurrent coarctation of the aorta can be performed safely and with excellent results. We believe it is still the gold standard in the management of recurrent coarctation of the aorta. (J Thorac Cardiovasc Surg 1998; 116:560-5)

Coarctation of the aorta, one of the most common cardiovascular anomalies in children, was first described in the mid-1700s by both Morgagni¹ and Meckel.² The natural history of this congenital disorder, through progressive hypertension, leads to heart failure and premature death.³ As the disease progresses various complications can occur, such as aortic dissection or rupture, hemorrhagic cerebrovascular accident, or bac-

terial endocarditis.⁴ Gross and Crafoord were early pioneers in the development of surgical repair of aortic coarctations in the mid-1940s.³ Since then, great progress has been made in the diagnosis and treatment of these anomalies. Perioperative morbidity and mortality have been greatly reduced, particularly in cases of uncomplicated coarctation not associated with complex congenital heart disease.⁵

Persistent arterial hypertension in repairs done at an older age and recurrence of coarctation in repairs done in neonates, however, continue to challenge our management strategies.⁶ Patients with long-standing coarctation have an increased risk for the development of coronary artery disease, as well as aortic dissection and pseudoaneurysm formation, even after successful repair.⁷ Postoperative hypertension, which occurs in 7% to 33% of patients, may be a result of recurrent or residual coarctation or may be idiopathic.^{8,9} The incidence of postoperative recurrent coarctation of the aorta is

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reported as ranging from 5% to 50%, depending on the criteria and methods used in making the diagnosis.^{8,9} The rate of recurrence varies significantly, depending on the age at initial repair.^{9,10} The specific surgical method of repair, suture material used, and sewing technique do not appear to be as important as once suggested in determining recurrences.⁹

Balloon aortoplasty was introduced in the last several years as an alternative method of managing aortic coarctation, particularly recurrent coarctation. This is certainly a less invasive method of intervention, although well-described complications do occur and recurrence rates are relatively high.¹¹⁻²⁰ The role of balloon aortoplasty in the management of coarctation needs to be defined further. Is surgical repair of recurrent coarctation of the aorta associated with high complication rates and deaths, or can it be performed with acceptable morbidity and mortality?

Patients and methods

During the past 27 years, from January 1970 to July 1996, 645 patients with coarctation of the aorta underwent surgical repair at Riley Children's Hospital in Indianapolis. During that same time period, 56 children had recurrent coarctation of the aorta necessitating intervention (8.7% overall recurrence rate). At our institution, patients with recurrent coarctation very seldom have undergone balloon aortoplasty. In this study we have focused on the patients who underwent surgical repair of recurrent coarctation in this retrospective chart review. The records of these patients were available until they were 18 years old, at which time they no longer received medical follow-up at Riley Children's Hospital.

After the initial coarctation repair, the patients routinely returned to the clinic for measurement of resting blood pressures and echocardiograms. Resting arm-leg gradients were also obtained and were considered abnormal if greater than 20 mm Hg. Exercise arm-leg gradients were also obtained in some patients and, if more than 40 mm Hg, were likewise considered abnormal. The patients who had persistent hypertension, elevated arm-leg gradients, or significant pressure gradients on echocardiogram underwent echocardiography or catheterization or both. The patients were then discussed among pediatric cardiologists and cardiothoracic surgeons before a decision regarding therapy was made. Among the patients who ultimately underwent repair of recurrent coarctation, the resting pressure gradient across the coarctation ranged from 10 to 80 mm Hg, with a mean of 42.7 mm Hg.

Procedure. All patients scheduled for repair of recurrent coarctation are studied with somatosensory evoked potentials during their preoperative visit the day before their scheduled operation. The intraoperative management is standard and does not vary significantly from a primary coarctation repair. The patients are placed in a right lateral decubitus position. The same skin incision is used as was used for the initial repair, but the intercostal space entered may be above or below the intercostal space used for the initial repair if that

intercostal space is fused. The condition of the patients is monitored during the operation with somatosensory evoked potentials. Intrathoracic adhesions are divided so that the left lung can be retracted anteriorly and inferiorly. The pleura overlying the aorta is elevated, with care taken to avoid violating the adventitial layer of the aorta or any of the intercostal vessels. No intercostal arteries are ligated or divided in most instances. Care is taken to mobilize the vagus and recurrent laryngeal nerves so that they are protected throughout the procedure. After the transverse aortic arch proximal to the left subclavian artery, the left subclavian artery, the previously repaired coarctation, and the descending thoracic aorta have been dissected from their surrounding adventitia, vascular clamps are applied to the aorta and the evoked potentials are monitored for 7 minutes.

All patients have a proximal and distal pressure line so that the distal arterial pressure can be monitored. Our goal is to keep the distal arterial pressure above 40 mm Hg while the aorta is clamped. If the distal pressure falls below 40 mm Hg, then crystalloid or colloid solution, 10 to 30 mL/kg, is administered in an attempt to obtain a distal arterial pressure of greater than 40 mm Hg. It is occasionally necessary to raise the proximal pressure to above 170 mm Hg but less than 200 mm Hg to accomplish this goal. During the initial phases of the surgical procedure, the patient's systemic temperature is allowed to drift to between 34°C and 35°C. If the patient has not reached this temperature by the time the aortic clamps are initially applied, cool saline solution is placed in the intrathoracic cavity to help cool the patient systemically. If the distal arterial pressure is greater than 40 mm Hg, and if the evoked potentials during a 7-minute period remain stable, then the clamps are removed. A 5- to 7-minute period of reperfusion is allowed before the clamps are reapplied and a definitive repair initiated.

Our preferred prosthetic patch material for recurrent coarctation is a collagen-impregnated knitted Dacron graft, which is fashioned in a diamond shape to allow the widest part of the patch to be in the narrowest area of the recurrent coarctation. The prosthetic patch is sewn into place with monofilament polypropylene suture. The patch can usually be inserted in a 15- to 25-minute time period. In the rare patient whose distal arterial pressure is marginal (ie, 35 to 45 mm Hg mean), a 15 mg/kg dose of methylprednisolone sodium succinate (Solu-Medrol) is given before application of the vascular clamps for the definitive repair. If for any reason the patient becomes hyperthermic (above 37°C), a vigorous attempt should be made to reduce the temperature before any definitive repair is attempted.

In the rare instance in which the distal arterial pressure falls below 35 mm Hg mean, or if the evoked potentials flatten during the initial 7-minute test clamp period, then a bypass shunt is inserted between the ascending aorta and descending aorta. In most instances the shunt is 2 arterial perfusion cannulas placed end to end, and the patient is heparinized while the shunt is in place. In only 1 instance in the past 25 years has cardiopulmonary bypass been necessary for the treatment of recurrent coarctation of the aorta.

Fig 1. Time interval between repairs for primary and recurrent coarctations.

Results

The patients comprised 25 girls and 31 boys whose ages ranged from 2 months to 18 years. Fifty-seven percent (32/51) had had their initial operation in the newborn period (within 1 month of birth), and 73% (41/56) had undergone their initial operation by 6 months of age. Only 18% (10/56) had their initial repair after 6 months of age. In 5 patients who had their first coarctation repair at other institutions, the exact age at first repair could not always be determined (Table I).

Fifty-nine percent of patients (33/56) had associated cardiac defects but only 20% (11/56) had complex congenital heart disease. The most commonly associated anomaly was patent ductus arteriosus in 34% of the patients (19/56). Ventricular septal defect was present in 27% of patients (15/56) and atrial septal defect and patent foramen ovale were present in 13% (7/56).

The surgical techniques used during the native coarctation repair were as follows: resection with primary end-to-end anastomosis in 53% (30/56); prosthetic patch aortoplasty in 23% (13/56); subclavian flap angioplasty in 18% (10/56); and tube interposition graft in 4% (2/56); in 1 patient, initially operated on overseas, the operative technique could not be discerned from the records (Table II). Regarding end-to-end anastomoses, 13% (4/30) were closed with interrupted sutures anteriorly and running suture posteriorly; 10% (3/30), running sutures anteriorly and posteriorly; and 17% (5/30), interrupted sutures anteriorly and posteriorly; in 60% (18/30) this detail is unknown.

Roughly half the patients (25/56) had an associated procedure performed when their coarctation was ini-

Table I. Age at first coarctation repair

Age	No. of patients	Percent
<1 mo	32	57%
1-6 mo	9	16%
>6 mo	10	18%
Unknown	5	9%

Table II. Surgical technique of coarctation repair

	Primary		Recurrent	
	Percent	No.	Percent	No.
Resection and end-to-end anastomosis	53%	30	4%	2
Prosthetic patch angioplasty	23%	13	80%	45
Subclavian flap angioplasty	18%	10	7%	4
Tube graft	4%	2	9%	5
Unknown	2%	1		

tially repaired. Thirty-four percent (19/56) had division of a patent ductus arteriosus, 20% (11/56) had pulmonary artery banding, and 11% (6/56) had both.

These patients underwent reoperative coarctation repair 2 weeks to 18 years after their first repair. The temporal pattern of recurrence appeared to have a bimodal distribution (Fig 1). Roughly one third of all patients (18/56) underwent a second operation within 2 years of the first operation, and half (28/56) underwent a second operation from 7 to 15 years after the first operation.

We performed subgroup analyses for age and technique during the first repair. Although the total number of patients in each group is relatively small, we found that in the early recurrence group (within 2 years) the mean age during the initial operation was 15 days. In the late recurrence group (7 to 15 years later) the mean age during the initial corrective operation was 67 days. We also found an overrepresentation of subclavian flap repairs in the early recurrence group, 55% compared with 16% in the whole series. Only 2 patients with previous subclavian flap repairs had recurrence proximal to the previous repair. Otherwise, recurrence was at the site of or just distal to the previous repair.

The surgical technique used during the second corrective operation was a prosthetic patch aortoplasty in 80% of patients (45/56). Nine percent of patients (5/56) required a tube interposition graft, 7% (4/56) underwent a subclavian flap angioplasty, and 2 patients had a resection with end-to-end anastomosis (Table II). Only 1 patient with a mid-transverse arch defect required circulatory arrest and cardiopulmonary bypass through a median sternotomy.

Table III. Recurrent coarctation of the aorta treated with balloon aortoplasty: Literature review

First author	No. of patients	Ages	Follow-up after balloon dilatation	Consecutive series?	Residual or recurrent coarctation after dilatation	Patients eventually requiring aortic surgery	Postdilatation aortic aneurysms	Femoral artery tear or occlusion
Lock (1983)	5	1 mo–22 y	1–6 mo	—*	60%	40%	0%	—*
Allen (1986)	8	7 mo–20 y	—*	—*	37%	—*	0%	—*
Lorber (1986)	5	3–9 mo	2–30 mo	—*	80%	—*	0%	20%
Saul (1987)	27	3 mo–22 y	—*	—*	26%	—*	7%	19%
Cooper (1989)	44	2 mo–20 y	2–24 mo	—*	14%	—*	14%	9%
Rao (1990)	9	6 mo–7 y	4–5 mo	Yes	11%	11%	0%	11%
Anjos (1992)	26	2 mo–18 y	2 mo–6 y	—*	42%	19%	8%	15%
Witsenburg (1993)	24	3 mo–16 y	18 mo–6 y	—*	17%	12.5%	4%	13%
Hellenbrand (1990)	190	1 mo–26 y	—*	No	22%	—*	—*	9%
VACA Registry								
McCord (1996)	548	1 mo–54 y	—*	No	25%	—*	—*	4%
VACA Registry								
Yetman (1997)	90	1 mo–20 y	3 mo–12 y	Yes	12%	—*	1%	25%

VACA, Valvuloplasty and Angioplasty of Congenital Anomalies.

*No data available.

The overall mortality, including late deaths, for all coarctation repairs for the past 27 years has been 8.1% (52/645) at our medical center. However, no deaths have occurred in patients who underwent recurrent coarctation repair. In addition, no major perioperative complications were encountered, such as phrenic nerve injuries, recurrent laryngeal nerve injuries, chylothorax, or spinal cord ischemia. One child with a hypoplastic aortic arch required a third coarctation repair a few weeks after the second because of residual coarctation in the transverse arch that was not recognized during the second operation. Overall, 3 patients had a hypoplastic aortic arch and 2, who had primary repair by a subclavian flap, had recurrent coarctation caused by the arch disease.

Follow-up was available in 71% of patients (40/56) and ranged from 1 to 11 years. Ninety-five percent of patients who were followed-up (38/40) are normotensive. Eighteen percent (7/40) have medically controlled hypertension. Five children are normotensive (2/5 medically controlled) but have echocardiographic evidence of a gradient across the coarctation repair site. The range of the gradient is 15 to 44 mm Hg with an average of 27.2 mm Hg. Two of these children have no echocardiographic evidence of gradient across the coarctation repair site.

Aneurysm formation is a concern when aortic patches are used with primary coarctation repairs. An aneurysm developed opposite the prosthetic patch in only 1 patient, an adolescent boy in whom a prosthetic patch was used during the initial repair. During the course of that operation, the patient had extensive debridement of the coarctation web without any attempt to reinforce

this area. The aneurysm that developed necessitated resection and graft replacement about 15 years after the initial repair. No other patient in the series has an aneurysm, and during the course of the initial coarctation we routinely avoid extensive debridement of the coarctation web or reinforce the coarctation web with absorbable sutures after the web has been debrided.

Comment

During the past 27 years, 645 children underwent primary repair of aortic coarctation and 56 children underwent repair of recurrent coarctation at our institution. Given that the number of native coarctation repairs over the years has remained fairly constant at our institution, we have an 8.7% overall recurrence rate, which is within the 5% to 50% range found in the literature.^{5–10}

Seventy-three percent of the children in our series had their initial repair performed when they were 6 months of age or younger and two thirds had their initial repair performed when they were neonates. This confirms the previously described relatively high recurrence rate when aortic coarctation is repaired during the first 6 months of life, and especially in the neonatal period.^{7–10} The age at repair appears to be inversely proportional to the rate of recurrence.

The question of which surgical techniques predispose to recurrence has been the subject of much debate. Initially resection with end-to-end anastomosis was the preferred surgical method (Gross and Crafoord). This technique then fell into disfavor as the subclavian flap angioplasty and prosthetic patch techniques gained wider acceptance.^{21,22} Subsequently, the extended end-to-end

anastomosis technique was reappraised and the subclavian flap angioplasty was implicated with higher recurrence rates.^{23,24} More recently, it has been shown that only subclavian flap angioplasties performed within 2 months of life have high recurrence rates, but if this technique is used in older children, the recurrence rate is no higher than that with other techniques.^{9,25} In our series, we did not clearly find that the particular technique of repair affected the overall incidence of recurrence.

In our series the recurrent coarctations appeared in 2 temporal clusters after the initial repair. One third appeared within 2 years of the first repair and half appeared 7 to 15 years later. Subgroup analysis revealed that in the early recurrences there was an overrepresentation of subclavian flap repairs: 55% of the patients had undergone subclavian flap repairs compared with 16% in the whole series. Interestingly, Ralph-Edwards and colleagues⁷ also found that subclavian flap failures tend to occur early and end-to-end anastomosis failures tend to become evident later. Could this be because some ductal tissue is not excised with subclavian flap angioplasty and this residual tissue predisposes to recurrence? Perhaps the failure of the end-to-end anastomoses and patch aortoplasties is caused by lack of growth rather than true restenosis.

We also found the mean age during the initial operation was 15 days in the early recurrence group and 67 days in the late recurrence group. Hence the recurrence rate is high in newborn infants, and coarctations also tend to recur early, within 2 years of the operation.

The vast majority of repairs for recurrent coarctation were performed with a prosthetic patch technique. This technique has been very satisfactory in most cases, as others have reported.^{7,9} Only 1 child required a third coarctation repair because of a residual transverse arch coarctation, unrecognized during the second repair. No untoward sequelae occurred with the third procedure, and no further gradient can be identified across the surgical repair site. Another patient has a 25- to 30-mm Hg gradient across the repair site, is being managed medically, but will likely require another intervention. Therefore, after the second operation, the gradient across the coarctation repair was either trivial or completely absent in 96% of patients (54/56).

No deaths occurred in our series of recurrent coarctations repaired surgically. The absence of deaths is undoubtedly influenced by the modest number of associated complex cardiac anomalies. Furthermore, there have been no major complications in this series, namely, no instances of phrenic nerve injury, recurrent nerve injury, chylothorax, or spinal cord ischemia. Spinal cord ischemia has been preventable by techniques previously described in this article. Careful test clamping of the

aorta combined with monitoring of evoked potentials has decreased the incidence of this complication in our series. Similar results have been obtained in other surgical series by Kron,⁹ Sweeney,²⁶ and their associates.

Because balloon aortoplasty is seldom performed at our institution, a direct comparison cannot be made. However, balloon aortoplasty is being used as the first line of treatment with recurrent aortic coarctation in several centers.^{19,20} In the literature, the various institutional series of balloon dilatations for recurrent coarctation appear to be relatively small, selective, and non-consecutive.¹¹⁻¹⁵ Follow-up is limited, however, and a considerable proportion are dilated repeatedly and finally corrected surgically.^{16,18} Success rates vary considerably, but with increased invasive cardiology experience and longer follow-up, recurrent or residual coarctation rates range from 20% to 40% (Table III).

Complications after balloon dilatation are not infrequent. Postdilatation aortic aneurysms develop in up to 14% of patients, and cerebrovascular accidents, although usually transient, also occur in a small percentage of patients.^{16,18} Femoral artery thrombosis and tears, which are more common in younger infants, occur in 9% to 20% of cases. Many balloon dilatation series do not report the incidence of persistent hypertension, but it appears to be at least 20%,^{18,20} compared with 7% in our series. Finally, several isolated cases of aortic rupture and death have been reported.^{20,27}

Balloon aortoplasty may have a role in the management of selected patients with recurrent aortic coarctation, but it appears to be unsuitable, at least for young infants, in whom recurrences and complications are high.^{8,16} The role of balloon angioplasty and stent placement is yet to be determined. This treatment also appears to be ineffective in the presence of associated arch hypoplasia.^{16,17} Although theoretically reoperative surgery is more hazardous than primary coarctation repair and older surgical series reported high death rates and complications,^{12,16} this series demonstrates that surgical repair can actually be performed with very high success and low morbidity and mortality rates. In light of these results, the role of balloon aortoplasty should be reevaluated.²⁸ We believe that surgical repair is still the gold standard for the management of recurrent aortic coarctation.

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