Balloon dilation angioplasty of aortic coarctations in infants and children

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ABSTRACT Balloon dilation angioplasty (BDA) was attempted nine times in eight infants and children with aortic coarctation. In three infants (all with associated ventricular septal defect or atroventricular canal and marked hemodynamic instability) dilation was attempted at a site of aortic narrowing that had not been operated on previously. Although the coarctation gradient fell 40% or more over the short term in two of the three, there was no angiographic or late gradient evidence of improvement. All three underwent subsequent coarctation surgery. Five dilations were performed in four infants and children who had previously undergone coarctation surgery (end-to-end anastomosis, attempted jump graft, and subclavian flap) and had residual gradients. Dilation was successful in all five cases, resulting in an increase in the diameter at the coarctation site (4.7 ± 2.6 to 7.7 ± 4.0 mm, p < .05) and a decrease in the gradient measured 24 hr after dilation (42.0 ± 15.5 to 11.8 ± 11.2 mm Hg, p < .05). In one child with a long area of hypoplasia of the thoracic aorta and similar lesions of the brachiocephalic vessels, a preliminary attempt to dilate a severely narrowed subclavian artery was unsuccessful. Postdilation angiography demonstrated evidence of intimal tears in three of five successful dilations. Follow-up (1 to 6 months) has demonstrated continued gradient relief in four of five children. BDA is frequently, but not always, a successful treatment for human aortic coarctation. The chief determinant of success appears to be the nature of the lesion; short-term changes in coarctation gradient are unreliable indicators of success of failure. Although BDA was not associated with mortality or significant morbidity in this group of patients, its role in the management of children with coarctation is yet to be determined.


ALTHOUGH the first attempts to relieve congenital vascular narrowings with catheters were reported nearly 30 years ago, interest in this approach lay dormant until it was observed that the new generation of dilating catheters made of polyvinyl or polyethylene could relieve aortic coarctation in postmortem hearts of newborns who had not undergone heart surgery. Since then, several case reports, and two series of patients, have appeared describing the variable results of balloon dilation angioplasty of congenitally or operatively narrowed valves and vessels. Along with this tentative clinical experience, a number of studies in animals and excised specimens have supported the notion that certain forms of aortic coarctation may be amenable to balloon dilation. However, the results of these studies indicate that high dilating pressures may be required and that transvascular rupture can occur with the use of very large dilating balloons; these results reinforce the need for caution.

While these preliminary studies may be encouraging, they do not justify the use of dilation angioplasty in all infants and children with coarctation. In general, the results of operative management have been excellent, with very low morbidity and mortality rates in uncomplicated cases. Nonetheless, there are three groups of patients in whom traditional operative management has been less successful: infants with coarctation and associated cardiac defects, children in whom previous surgery had left a residual gradient, and patients with long segments of aortic narrowing. We therefore instituted a clinical trial of balloon dilation angioplasty in these types of patients; from July 1981 to December 1982, nine dilations were attempted in eight children.

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Materials and methods

Patients. Infants under 1 month of age were considered candidates for dilation only if an associated cardiac defect (large ventricular septal defect [VSD], severe valvar aortic stenosis, or cyanotic heart disease) was likely to complicate successful coarctation repair, and only if shock and acidosis were present in the 48 hr before catheterization. Older infants and children were considered candidates for dilation if the coarctation was long and tubular (greater than 2 cm in length), or if there was a significant residual gradient after a prior attempt at repair. Unless emergency therapy was required, those children who met the above requirements were referred to the combined Pediatric Cardiology/Surgery/Radiology conference at the University of Minnesota Hospitals. If the consensus was that further therapy was required, the patients’ parents were given a choice between the treatment courses of experimental dilation and conventional operative management. Informed consent was obtained from the parents and/or child when appropriate.

Assessment of effects of dilation angioplasty. Successful relief of a vascular obstruction should increase flow, increase diameter of the narrowed site, and decrease the pressure gradient. The presence of collateral vessels with coarctation complicates any noninvasive attempt to accurately measure flow across the dilated site. The diameter of the coarcted site and the diameter of the transverse aortic arch in both systole and diastole were measured directly on biplane aortograms before and after dilation. We corrected for magnification by using the known catheter diameter. Predilation and immediate postdilation systolic pressure gradients were measured by direct pullback across the coarctation with fluid-filled catheters connected to Statham 23Db strain gauges (“zeroed” to midchest level) and were recorded on an Electronics for Medicine VR 12 recorder. To avoid perforation, we never passed an unfurred catheter or wire across a previously dilated site. Thus, the predilation gradient was obtained before any contrast injection, whereas the postdilation gradient was measured after at least 3 ml/kg of meglumine diatrizoate had been administered for angiography.

Dilation protocol. The protocol design was based on our experience in over 50 lambs with experimental aortic coarctations that were dilated in our laboratory. These studies indicated that normal aortas could be stretched by at least 30% without producing any visible vascular injury, that balloon diameters less than twice the diameter of the coarctation had a minimal beneficial effect, and that balloon diameters greater than 3 times the coarctation diameter would often produce transvascular tears and mediastinal hemorrhage. Accordingly, we chose polyethylene dilating balloons (Meditech, Inc., or Cook, Inc.) with inflated diameters 2.5 times the size of the coarctation diameter. In all but one patient (No. 2), a pigtail catheter was advanced percutaneously from the femoral artery into the ascending aorta and the pressure gradient and angiographic diameter of the coarctation were measured. An appropriately sized Teflon-coated guidewire (0.021 inch for infants and 0.038 inch for older children) was advanced through the pigtail to the ascending aorta or left ventricle. The deflated dilating catheter (Nos. 4.5F to 5F for infants and Nos. 8F to 9F for older children) was then advanced percutaneously, without a sheath, into the ascending aorta over the guidewire. The balloon was then withdrawn slowly under the area of coarctation and inflated. Adequate balloon size and position were confirmed in each case by inflating the balloon (with 60% contrast diluted 1:1 with saline) to 1 to 2 atm and recording on cineangiogram the presence of a typical waist (figure 1). At this point the balloon was inflated to progressively higher pressures until either the waist disappeared or the known pressure limits of the balloon were reached. Inflation was held for 25 to 60 sec, the balloon was deflated and removed (leaving the guidewire in position), and a pigtail catheter was again advanced over the guidewire into the ascending aorta for postdilation angiography and pressures.

Dilations were performed in the cardiac catheterization laboratory. Three of the four infants were hemodynamically unstable at the time of dilation so that the procedure was performed while they were intubated and being artificially ventilated. The other children were presedated with secobarbital (2 mg/kg im) and morphine (0.1 mg/kg im). Additional sedation (diazepam or additional morphine) was administered as required through venous catheters. At least 2 units of blood were available for each dilation, cardiovascular surgical standby was arranged for each child, and the chest was fluoroscoped for evidence of extravascular bleeding. Since in our previous study several lambs had had ill-defined episodes of instability associated with experimental dilation, each patient was monitored overnight in an intensive care unit. Systemic heparinization was used only in those patients (Nos. 7 and 8) in whom a carotid artery was occluded by the dilating balloon during the procedure. On late follow-up, pressure gradients were determined clinically with appropriately sized arm and leg blood pressure cuffs.

FIGURE 1. Predilation lateral aortogram (Pre), fully inflated balloon in position (middle panel), and postdilation lateral aortogram (Post) from patient No. 3. Note that waist in balloon persists even at very high dilating pressures.
Results

Patients. A total of nine dilations were attempted in eight children. The clinical summaries, dilation techniques, and results of dilation are summarized in table 1. In each case an appropriate dilating catheter could be advanced across the narrowing and in each the initial balloon diameter was 1.7 to 2.9 times the diameter of the coarctation.

Two infants (patients 1 and 2) with large muscular or atrioventricular canal-type VSDs had shock, aci- demia, and anuria at the time of attempted dilation. Both had evidence of active disseminated intravascular coagulation (thrombocytopenia and prothrombin time > 1.5 times normal). Two other infants (Nos. 3 and 4) had undergone previous subclavian flap and pulmonary artery banding procedures, and had intractable failure along with a significant residual gradient. In one the gradient was due to a residual narrowing at the original operative site; in the other (in whom the first operation was performed without prior catheterization) the gradient was due to an area of isthmic hypoplasia proximal to the original coarctation (figure 1).

<table>
<thead>
<tr>
<th>Patient No./age</th>
<th>Diagnoses</th>
<th>Balloon pressure (atm)</th>
<th>Diameter of proximal aortic arch (mm diastole/mm systole)</th>
<th>Predilation</th>
<th>Postdilation</th>
</tr>
</thead>
<tbody>
<tr>
<td>1/1 wk</td>
<td>Complete AV canal; shock; juxtaductal coarctation</td>
<td>4/8</td>
<td>3.8/4.0</td>
<td>14</td>
<td>2.3</td>
</tr>
<tr>
<td>2/4 wk</td>
<td>Muscular VSD; juxtaductal coarctation; renal failure</td>
<td>5/8</td>
<td>3.1/3.4</td>
<td>30</td>
<td>2.1</td>
</tr>
<tr>
<td>3/6 wk</td>
<td>Muscular VSD; S/P subclavian flap; isthmic hypoplasia</td>
<td>5/9½</td>
<td>4.3/4.3</td>
<td>74</td>
<td>2.2</td>
</tr>
<tr>
<td>4/15 wk</td>
<td>VSD; S/P subclavian flap; PA band</td>
<td>5/8</td>
<td>3.9/4.2</td>
<td>30</td>
<td>1.8</td>
</tr>
<tr>
<td>5/6 yr A</td>
<td>Coarctation, S/P end-to-end anastomosis</td>
<td>8/8</td>
<td>9.8/10.8</td>
<td>67</td>
<td>3.5</td>
</tr>
<tr>
<td>5/7 yr</td>
<td>S/P prior dilation</td>
<td>12/7</td>
<td>10.0/10.8</td>
<td>28</td>
<td>5.2</td>
</tr>
<tr>
<td>6/9 yr</td>
<td>Coarctation, S/P end-to-end anastomosis; S/P jump graft</td>
<td>12/8</td>
<td>9.2/10.5</td>
<td>42</td>
<td>4.3</td>
</tr>
<tr>
<td>7/16 yr</td>
<td>Supravalvar AS; multiple great vessel stenoses and hypoplasias</td>
<td>8/6; 10/4</td>
<td>—</td>
<td>74</td>
<td>2.7</td>
</tr>
<tr>
<td>8/22 yr</td>
<td>Coarctation, S/P end-to-end anastomosis; Turner’s syndrome</td>
<td>20/3</td>
<td>15.0/16.5</td>
<td>43</td>
<td>8.9</td>
</tr>
</tbody>
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AV = atrioventricular; S/P = status post; PA = pulmonary artery; AS = aortic stenosis.

Patient No. 5 underwent two separate dilation procedures 6 months apart.

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Two older patients (6 and 22 years old) presented with residual gradients after an end-to-end resection had been performed during infancy. The 22-year-old patient, who had Turner’s syndrome, had the coarctation just distal to the left carotid artery; subsequent angiography showed that any attempt to cross-clamp the coarctation site would necessitate occlusion of the left carotid. A third patient, who had an end-to-end resection in infancy, had an unsuccessful jump graft from the transverse arch to the descending aorta attempted at age 6. A 16-year-old girl had a familial form of supravalvar aortic stenosis with multiple areas of discrete arterial hypoplasia. Although her supravalvar aortic stenosis had been successfully relieved when she was 9, she also had 2 to 6 cm long areas of obstruction in both proximal subclavians, both midcarotid arteries, and her midthoracic aorta such that systolic aortic arch pressures were 210 mm Hg, but the pressures in her distal carotids, subclavians, and aorta below the diaphragm ranged between 110 and 130 mm Hg. We speculated that the aortic obstruction would be structurally similar to the other vascular obstructions, and therefore attempted to dilate her right subclavian artery before dilating her diffusely hypoplastic thoracic aorta.

Protocol for dilation. No changes were made in the dilation protocol during the course of this study; the approach we used is summarized in table 2.

Effects of dilation. If only short-term effects of dilation on coarctation gradient are considered, then dilation angioplasty was an extremely successful maneu-

<table>
<thead>
<tr>
<th>TABLE 2</th>
<th>Dilation protocol: aortic coarctation</th>
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<tr>
<td>Balloon diameter (inflated)</td>
<td>2.25 to 3 times coarctation diameter</td>
</tr>
<tr>
<td>Balloon length</td>
<td>2 to 4 cm</td>
</tr>
<tr>
<td>Dilating pressure</td>
<td>3 to 9 atm (42 to 126 psi), depending on balloon tolerance and pressure at which waist disappears</td>
</tr>
<tr>
<td>Dilating fluid</td>
<td>Half-strength contrast medium</td>
</tr>
<tr>
<td>Duration of dilation</td>
<td>30 to 60 sec</td>
</tr>
<tr>
<td>No. of dilations</td>
<td>1</td>
</tr>
<tr>
<td>Medications</td>
<td>Routine precatheterization sedation; extra analgesia (morphine) just before actual dilation (heparinization appears to be optional)</td>
</tr>
<tr>
<td>Catheter approach (size)</td>
<td>Percutaneously from femoral artery, No. 4.5F (infants), No. 8 or 9F (children)</td>
</tr>
<tr>
<td>Precautions</td>
<td>Never pass unguided wire or catheter across recently (less than 2 wk) dilated coarctation; monitor each patient overnight in ICU setting</td>
</tr>
</tbody>
</table>

FIGURE 2. Pressure gradient across aortic coarctation (or right subclavian artery, patient No. 7) before dilation, immediately after dilation, 18 to 24 hr after dilation, and on late follow-up.

ver, causing gradients to fall in eight of nine attempts (figure 2). However, this short-term change in gradient did not correlate well with changes in angiographic appearance or with the late clinical effects of dilation. In patients 3 and 7 the gradient fell by more than 40 mm Hg over the short term, but the angiograms showed very little change (figures 1 and 3) and in each case the gradient had returned to predilation levels within 24 hr. Conversely, after the first dilation in patient 5 there was only a 19 mm Hg fall in the gradient, despite improvement in the angiographic appearance and further improvement in the gradient over the subsequent 12 hr. These findings indicate that an early gradient fall is an unreliable indicator of successful dilation of an aortic coarctation.

Relying on angiographic appearance and gradient and pulse quality 24 hr after dilation, and labeling as a success those procedures resulting in an increase in coarctation diameter of more than 30% and in a gradient of less than half that before dilation, the procedure was successful in five of nine dilation attempts (patients 4, 5, 5', 6, and 8; figure 2).

Of the four infants with coarctation, three (patients 1, 2, and 3) had not had surgery previously. In two of the three the coartations were typical; the third had an area of isthmic hypoplasia proximal to the left subclavian. In all three dilation was unsuccessful, and in each the procedure failed because the waist in the balloon could not be eliminated, despite very high dilating pressures (figure 1). In the fourth infant dilation was
only moderately successful, but it did reduce the gradient enough to allow the child to successfully undergo VSD closure 2 days later.

In the child with multiple arterial hypoplasias dilation was also unsuccessful despite an early dramatic fall in the gradient across the right subclavian (figures 2 and 3). Since the subclavian dilation was clearly unsuccessful, no attempt was made to dilate the diffusely hypoplastic aorta. In this case dilation failed because the site was very compliant, stretching but not tearing even when a balloon more than three times the diameter of the vessel was used (figure 3).

In the children with previous end-to-end anastomoses and residual gradients, dilation was uniformly successful, with the gradient at late follow-up value in each patient being less than 10 mm Hg. In the child with the tightest coarctation and highest resting gradient, dilation was performed as a staged procedure with an 8 mm balloon the first time and a 12 mm balloon the second time, with excellent results (figure 4). After two of the four dilations in this group of children, irregularities in the margins of the contrast column could be seen after dilation, presumably because of small intimal tears (figures 4 and 5).

Morbidity and mortality. Only one patient (No. 1) in this series died, an infant with an atrioventricular canal and massive mitral regurgitation. At the time of dilation, he had uncontrollable shock despite pressors, prostaglandin therapy, and continuous bicarbonate infusions. When no apparent benefit was derived from dilation, he underwent an operation to attempt coarctotomy and pulmonary artery banding. At operation, the coarctation site was noted to be slightly edematous. He died during banding of the main pulmonary artery.

Two infants who had had unsuccessful dilations underwent subsequent successful coarctation surgery; in each, the coarctation site was carefully inspected before incision and only mild edema was noted.

The dilation procedure itself was well tolerated, even in infants with severe hemodynamic instability. One infant had an episode of ventricular fibrillation before dilation when the guidewire was advanced through a pigtail into the left ventricle. He underwent cardioversion and subsequent dilation was without incident. There was no evidence of cardiac irritability, bradycardia, or hypotension in any of the children at the time of aortic occlusion. Similarly, no episodes of instability were seen in the postdilation period during the monitoring of each child in the intensive care unit.

One of the four infants had a diminished femoral pulse as a result of the dilation attempt.

Each of the three older children in whom dilation was successful complained of chest pain at the actual moment of dilation. In two patients the pain radiated to the throat and in one child a dull ache persisted for 1 to 2 hr.

Late follow-up. Of the five patients in whom dilations were initially successful, the improvement has persisted in four (figure 2). In the infant, a gradient of 40 mm Hg was documented on examination 4 months after dilation. In the other four the gradients have, if anything, fallen further in the postdilation period. Late postdilation angiography in one patient (figure 4) demonstrated no evidence of restenosis or aneurysm formation.

Discussion

Although previous case reports and brief re-
FIGURE 4. Series of four lateral aortograms from patient No. 5. Note area of irregularity in the contrast column's posterior margin after second dilation.

ports\textsuperscript{17, 18} have implied that balloon dilation angioplasty may successfully relieve the obstruction of aortic coarctation, its actual role in the management of patients with coarctation remains to be established. The number of patients reported here is too small, and the duration of follow-up too short, to allow any but the most tentative conclusions to be drawn. Nonetheless, certain observations can be made that may prove useful in directing the further use of this technique.

First, despite the implications of previous reports,\textsuperscript{4, 7} balloon dilation angioplasty of aortic coarctations is not a uniformly successful procedure. Three of our patients had unequivocal evidence of a technically adequate attempt at angioplasty (figure 1), and yet clearly did not benefit. At this time there is no reason to suspect that any simple technical modifications would have improved the results in these patients. This observation raises two further questions. Would the use of newer angioplasty techniques (e.g., balloons capable of dilating pressures higher than 10 atm) prove successful in the more difficult cases? Which forms of human aortic coarctation are dilatable with currently available techniques and which are not?

While the first question is, at present, unanswerable, we can at least begin to address the second question. Balloon angioplasty was attempted in three infants at a coarctation site that had not been operated on previously. In all three the procedure was unsuccessful. While these results do not indicate that infantile aortic coarctations are undilatable under all circumstances, they do imply that the native infantile aortic coarctation can be a very rigid lesion and that a higher
failure rate can be anticipated in this group of patients. Angioplasty was also attempted in five infants and children who had previously undergone coarctation surgery. Since these children had had three different types of coarctation surgery and were older than the infants in whom dilations were unsuccessful, no firm conclusions can be drawn about the relationship between coarctation dilatability and previous surgery. At first glance, our ability to dilate the transvascular "scar" of an end-to-end anastomosis appears at variance with the reports that fresh transvascular scars in animals are undilutable lesions. While this discrepancy may be due to some ill-defined species differences, it is perhaps more likely that the compositions of the vascular wall in 2-month-old and 5-year-old vascular scars are markedly different. At present, we cannot identify the precise mechanism of dilation in such end-to-end anastomoses, but it may turn out that the time interval between coarctation surgery and subsequent dilation attempts is a very important variable.

Second, it is clear that a short-term fall in the gradient across a coarctation site is an unreliable indicator of the success of dilation angioplasty. This should not be surprising; a coarctation gradient is the product of a complex interaction of numerous variables, including blood flow across the coarctation, flow through collateral vessels, the caliber of the coarctation, the vascular resistances above and below the coarctation, and the heart rate. We have therefore used, as tentative criteria for a successful dilation, the combination of a 30% or larger increase in the diameter at the dilated site and a greater than 50% fall in the gradient 24 hr after dilation. These criteria appear reasonably accurate in predicting clinical response.

Third, successful dilation of human coarctation is usually, if not invariably, associated with evidence of an intimal tear (figures 4 and 5). As is the case with dilations performed in experimental animals, these tears appear to be well tolerated, even in a patient with Turner's syndrome. Nonetheless, they do indicate that the potential for late morbidity and mortality from balloon dilation of aortic coarctation exists and that these patients will require close clinical and perhaps angiographic follow-up for a prolonged period of time.

In summary, the results of this study indicate that balloon dilation angioplasty of human aortic coarctations is frequently, but not invariably, a successful treatment, with minimal early morbidity. The chief determinant of success would appear to be the nature of the lesion itself. While these results clearly do not warrant the widespread use of dilation angioplasty, they do help identify those questions that will require answers before the optimal role of dilation angioplasty in the management of childhood coarctation can be outlined.

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