

PEDIATRIC CARDIOLOGY

Aortic Aneurysm After Patch Aortoplasty Repair of Coarctation: A Prospective Analysis of Prevalence, Screening Tests and Risks

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Twenty-nine children were evaluated prospectively for the presence of an aortic aneurysm at the repair site 1 to 19 years after patch aortoplasty repair of coarctation of the aorta. In each child, noninvasive evaluation included a chest X-ray film, computed tomography of the chest and two-dimensional echocardiography. The presence and size of an aortic aneurysm were determined quantitatively by measuring the ratio of the diameter of the thoracic aorta at the repair site to the diameter of the aorta at the diaphragm (aortic ratio). An aortic ratio of ≥ 1.5 was judged abnormal and was shown to be significantly greater than the aortic ratio of a normal control group. An aortogram was obtained in each child if any noninvasive screening test was found to be abnormal.

As assessed by the aortogram, the prevalence of aortic aneurysm was 24% in this patient group. The sensitivity of echocardiography and chest computed tomography for

detecting an aneurysm was 71% and 66%, and the specificity 76% and 85%, respectively. The chest X-ray film was 100% sensitive and 68% specific in determining the presence of an aneurysm. Although the data are not statistically significant, they suggest that children undergoing patch aortoplasty as the primary procedure (rather than a reoperation after earlier resection), and children in whom a Dacron patch is utilized may be at increased risk for aneurysm formation.

Thus, in a prospective manner, this study has documented that aortic aneurysm occurs commonly after patch aortoplasty for coarctation in childhood, that the chest X-ray film provides a sensitive screening test and that the aneurysm may be evaluated quantitatively (by measuring the aortic ratio) with echocardiography or chest computed tomography.

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Patch aortoplasty repair of coarctation of the aorta was first described by Vosschulte (1) in 1957 as an alternative to resection and end to end anastomosis. The rationale for its use was that avoidance of a circumferential suture line would allow better growth at the repair site, thereby reducing the incidence of restenosis. However, 2 decades later, reports appeared (2,3) describing the development of aortic aneurysm at the repair site after Dacron patch aortoplasty.

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Although most patients described in these initial reports underwent surgical repair of a progressively enlarging aneurysm, one died suddenly from rupture of an aneurysm 17 years after patch aortoplasty (3). Efforts to define the prevalence of aortic aneurysm after patch aortoplasty for coarctation have yielded varying results. A prevalence rate of 27% was reported (4) in a series of 68 adults, including 2 who died after rupture of an aneurysm. The reported prevalence in the pediatric population has varied from 5% to 26% (5-7).

Several deficiencies in previous studies prompted the current investigation. First, earlier studies were based primarily on an untested assumption that the chest X-ray film is a sensitive screening test for detecting the presence of an aortic aneurysm after coarctation repair. Second, previous studies have not used objective criteria to define an aneurysm and have instead relied on subjective evaluations. Such methodology makes it virtually impossible to evaluate and compare findings. Third, earlier reports have failed to evaluate the effectiveness of noninvasive imaging techniques for

detecting the presence of aneurysmal aortic dilation at the coarctation repair site. The role of computed tomography and ultrasound, well established for the evaluation of abdominal aortic and coronary artery aneurysms (8-13), has not been defined in relation to thoracic aortic aneurysms after repair of coarctation. The present study, therefore, was designed to systematically evaluate a population of patients who had undergone patch aortoplasty repair of coarctation in childhood. The purpose was to define the prevalence of aortic aneurysms using objective criteria and to determine the sensitivity and specificity of the chest X-ray film, chest computed tomography, and two-dimensional echocardiography for detecting the presence of an aneurysm in this patient population. Furthermore, we sought to clarify potential clinical or surgical risk factors for the development of a late aortic aneurysm, and to develop a strategy for long-term follow-up of these patients.

Methods

Study patients. During a 12 month period, 29 consecutive outpatients who had had a prosthetic patch repair for aortic coarctation at least 1 year earlier were prospectively evaluated at the C. S. Mott Children's Hospital. The study was approved by the Institutional Review Board of the University of Michigan Medical Center and informed consent obtained in each case. All patients were evaluated for potential clinical risk factors including hypertension and a residual coarctation pressure gradient measured by right arm and leg blood pressure during the clinic visit. Operative notes were reviewed for potential surgical risk factors, including type of prosthetic patch, suture material, resection of the posterior shelf, surgeon and prior resection with end to end anastomosis (if patch aortoplasty constituted a second surgical procedure).

Noninvasive screening tests. A chest X-ray was obtained in all 29 patients, and a two-dimensional echocardiogram and contrast-enhanced computed tomographic scan of the chest were obtained in all patients with the exception of two who had recent angiograms. The chest computed tomography was also omitted in one patient with allergy to contrast media. All studies were read independently and without knowledge of other data. Chest X-ray films were evaluated by two cardiologists (R.H.B. and A.P.R.) for the presence of mediastinal widening suggestive of aortic dilation at the site of repair. Echocardiograms and chest computed tomographic scans were evaluated (by A.R.S. and E.B., respectively) in the following manner. The diameter of the aorta was measured at the repair site and at the level of the diaphragm, and the ratio of the two was determined (aortic ratio). Echocardiography utilized the suprasternal and subcostal views and chest computed tomography the cross-sectional view. If the aortic ratio by echocardiography differed in the suprasternal and subcostal views, the larger

ratio was taken. A study was defined as positive if the aortic ratio was ≥ 1.5 .

Aortography. An aortogram was performed if any one of the noninvasive studies (chest X-ray film, echocardiography or chest computed tomography) suggested the presence of an aortic aneurysm (i.e., positive test). In one patient who was allergic to contrast agents, nuclear magnetic resonance imaging was used as the definitive imaging study in lieu of an aortogram and clearly delineated the anatomy of the aortic arch and descending aorta. The presence of an aneurysm was defined only by positive findings on aortography (or nuclear magnetic resonance imaging in one case) when the ratio of the diameter at the repair site to the diameter of the aorta at the diaphragm was ≥ 1.5 in the anteroposterior or lateral plane (9,10). The absence of an aneurysm was defined by negative findings on aortography (aortic ratio < 1.5) or by negative findings on chest X-ray film, echocardiogram and chest computed tomographic scan, in which case we believed that an aortogram was not justified. Twenty-two of the 29 patients underwent aortography, 13 because of an abnormal chest X-ray film, chest computed tomographic scan or echocardiogram, and 9 for reasons unrelated to the aneurysm study.

Statistical analysis. Data are presented as mean values \pm 1 SEM. Comparisons between groups were performed with a two-tailed Student's *t* test for continuous data or the Fisher exact test for categorical data. A *p* value < 0.05 was required as evidence of a significant difference between groups.

Results

Patient characteristics (Table 1). The 29 children ranged in age from 1 month to 15 years (mean 5.8 years) at the time of surgery. Their ages ranged from 2 to 26 years (mean 11.3) at the time of the present evaluation, and the duration of follow-up after surgery ranged from 1 to 19 years (mean 5.6). Thirteen patients had systolic hypertension, defined as a systolic blood pressure exceeding the 90th percentile for age and gender, and five were taking antihypertensive medication. Five had a residual systolic pressure gradient > 20 mm Hg between the right arm and leg.

Aneurysm prevalence (Table 2). Seven of 29 patients satisfied the criteria for the presence of an aneurysm, yielding a prevalence rate for aortic aneurysm of 24% in this patient group (95% confidence interval: 12% to 42%). In these children the aortic ratio determined by angiography ranged from 1.6 to 1.8. The seven children had undergone patch aortoplasty 2 to 11 years earlier, six with a Dacron patch. One child was hypertensive, and one had a residual systolic coarctation gradient > 20 mm Hg. None had Turner syndrome and none had symptoms related to the aortic aneurysm.

To determine if aneurysmal aortic dilation is a normal postoperative finding, we compared our study patients with

Table 1. Comparison of Pertinent Clinical and Surgical Data in 7 Children With and 22 Children Without an Aneurysm After Patch Aortoplasty

| | Aneurysm Present (7) | Aneurysm Absent (22) | p Value |
|---------------------------------|----------------------|----------------------|---------|
| Clinical data | | | |
| Age at repair (yr) | 5.7 ± 1.5 | 5.8 ± 1.1 | 0.99 |
| Follow-up years | 5.9 ± 1.1 | 5.5 ± 0.9 | 0.82 |
| Turner syndrome | 0 | 1 | 1.0 |
| Systolic hypertension | 1 | 12 | 0.09 |
| Postop gradient > 20 mm Hg | 1 | 4 | 1.0 |
| Surgical data | | | |
| Patch material* | | | 0.15 |
| Dacron | 6 | 11 | |
| PTFE (Gore-Tex, Teflon, Impira) | 1 | 9 | |
| Previous resection | | | 0.17 |
| Yes | 0 | 5 | |
| No | 7 | 17 | |
| Posterior shelf reaction | | | 0.94 |
| Yes | 2 | 6 | |
| No | 5 | 16 | |

*Data not available in two patients. Postop = postoperative; PTFE = polytetrafluoroethylene.

a set of control patients who had levophase aortography performed during evaluation of right-sided lesions and were believed to have a normal aorta (Fig. 1). The aortic ratio of the control subjects was similar to that of the 22 study patients defined as having no aneurysm (1.16 ± 0.05 versus 1.19 ± 0.12 , $p = 0.59$) but differed significantly from that of the seven patients with an aortic aneurysm (1.71 ± 0.36 , $p < 0.001$). Thus, aneurysmal aortic dilation as defined in this study is not a normal finding after prosthetic patch aortoplasty. It is absent in the majority of patients after patch aortoplasty (in whom the aortic ratio does not differ from controls).

Noninvasive screening tests (Table 3). The chest X-ray film proved to be a very sensitive test (100% sensitivity) for detecting the presence of an aortic aneurysm at the site of

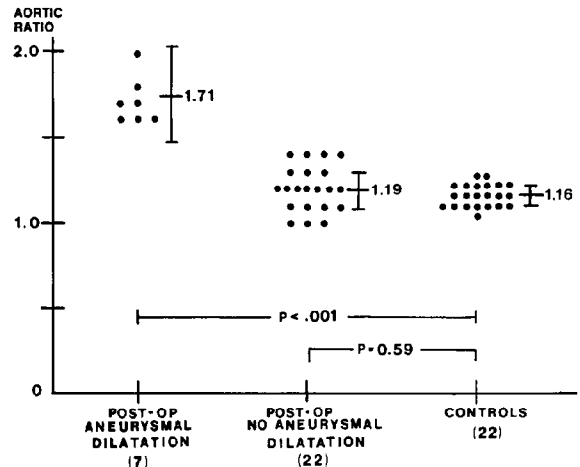


Figure 1. Aortic ratio (ratio of the aortic diameter at the repair site to the diameter of the aorta at the diaphragm) in 29 children after patch aortoplasty with ($n = 7$) and without ($n = 22$) aneurysmal aortic dilation and in 22 children with a normal aorta (controls). Post-op = postoperative.

patch aortoplasty; it correctly identified all seven patients with an aortic aneurysm proved by angiography. Its specificity, however, was 68% and its positive predictive value was only 50%. The chest X-ray film was interpreted as positive in 14 children, but the aortogram documented the presence of an aneurysm in only 7 of the 14. The sensitivity of echocardiography was 71% and that of chest computed tomography was 66% if used independently; the sensitivity of these two tests increased to 86% if used in combination. Each failed to identify two of seven patients with an aortic aneurysm proved by angiography. The aneurysm seen on X-ray film in one child (Patient 5, Table 2) was not detected by either echocardiography or chest computed tomography.

To determine the ability of echocardiography and chest computed tomography to quantitate aortic dilation at the

Table 2. Pertinent Clinical Data Regarding Seven Children With an Aortic Aneurysm After Patch Aortoplasty Repair of Coarctation

| Case | Age at Repair (years) | Follow-Up (years) | HTN | Gradient (mm Hg) | Patch Material | Previous Repair | Angiographic Aortic Ratio | Positive Screening Test | | |
|------|-----------------------|-------------------|-----|------------------|----------------|-----------------|---------------------------|-------------------------|------|----------|
| | | | | | | | | CXR | Echo | Chest CT |
| 1 | 7 | 6 | No | 4 | Dacron | No | 1.6 | Yes | Yes | No |
| 2 | 7 | 2 | No | 6 | Dacron | No | 1.6 | Yes | Yes | Yes |
| 3 | 8 | 11 | Yes | 8 | Dacron | No | 2.0* | Yes | Yes | — |
| 4 | 11 | 3 | No | 14 | Dacron | No | 1.7 | Yes | No | Yes |
| 5 | 0.1 | 7 | No | 12 | Gore-Tex | No | 1.7 | Yes | No | No |
| 6 | 1 | 5 | No | 25 | Dacron | Yes | 1.6 | Yes | Yes | Yes |
| 7 | 6 | 7 | No | 15 | Dacron | No | 1.8 | Yes | Yes | Yes |

*Nuclear magnetic resonance imaging ratio. Angiography aortic ratio = ratio of diameter of aorta at the repair site to diameter at the diaphragm measured by angiography; Chest CT = chest computed tomographic scan; CXR = chest X-ray film; Echo = two-dimensional echocardiogram; gradient = residual systolic pressure gradient across the coarctation repair site; HTN = upper limb hypertension.

Table 3. Assessment of Noninvasive Screening Tests for the Detection of Aortic Aneurysms After Patch Aortoplasty in 29 Patients

| | Angiography | | Sensitivity (%) | Specificity (%) | Positive Predictive Value (%) |
|----------|------------------|-----------------|-----------------|-----------------|-------------------------------|
| | Aneurysm Present | Aneurysm Absent | | | |
| CXR | | | 100 | 68 | 50 |
| Positive | 7 | 7 | | | |
| Negative | 0 | 15 | | | |
| Echo | | | 71 | 76 | 50 |
| Positive | 5 | 5 | | | |
| Negative | 2 | 16 | | | |
| Chest CT | | | 66 | 85 | 57 |
| Positive | 4 | 3 | | | |
| Negative | 2 | 17 | | | |

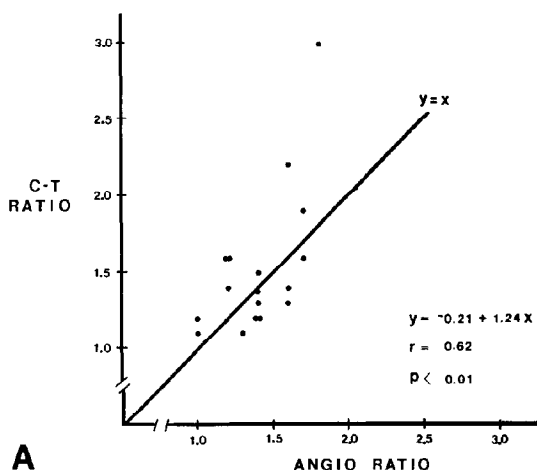
Abbreviations as in Table 2.

coarctation repair site, the aortic ratios measured by these tests were compared with those determined by angiography (Fig. 2). A fair correlation was found between the aortic ratio measured by each noninvasive test and by angiography. Both echocardiography and chest computed tomography tended to overestimate the angiographic aortic ratio in the higher range. Neither imaging technique tended to systematically underestimate the angiographic ratio.

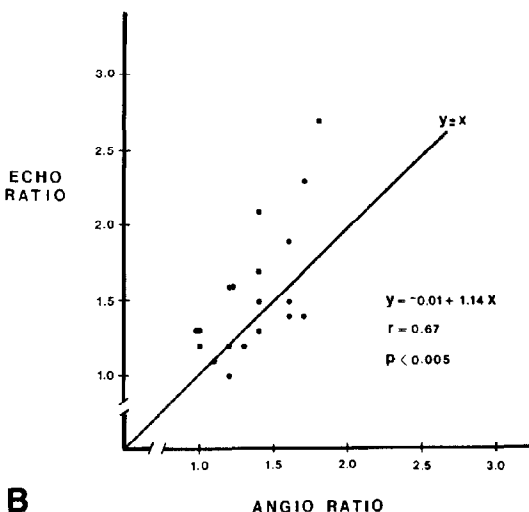
Figure 3 depicts serial chest X-ray films from Patient 7, who underwent a Dacron patch aortoplasty at 6 years of age. The films demonstrate mild prominence of the upper thoracic aorta 1.5 years postoperatively (Fig. 3A) that had enlarged considerably by 7 years after the repair (Fig. 3B), when an aortogram confirmed the presence of a large aneurysm at the site of coarctation repair (Fig. 4). At surgery, a large fusiform aneurysm was found encompassing the circumference of the aorta at the level of the patch. The aneurysm was excised and sections obtained adjacent to and opposite the patch demonstrated the wall to consist of dense fibrous tissue with scattered areas of mucinous degeneration, focal calcification and foreign body giant cells (Fig. 5). Interrupted remnants of an external elastic lamina found throughout both specimens supported the diagnosis of a true aneurysm.

Figure 6 demonstrates the ability of echocardiography and chest computed tomography to detect a smaller aortic aneurysm (aortic ratio = 1.6) that was present in Patient 6 (Table 2) and confirmed by angiography. This was the smallest aneurysm detected in the present series. The non-invasively derived aortic ratio will be followed up longitudinally in this child as a quantitative measure to detect progression in aneurysm size.

Risk factors. To assess possible risk factors that may predispose to the development of aneurysmal aortic dilation, the seven patients with an aneurysm were compared with the 22 children without this complication (Table 1). No clinical



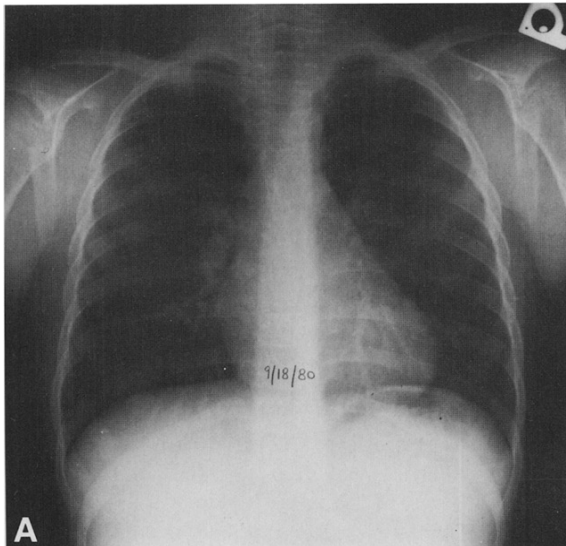
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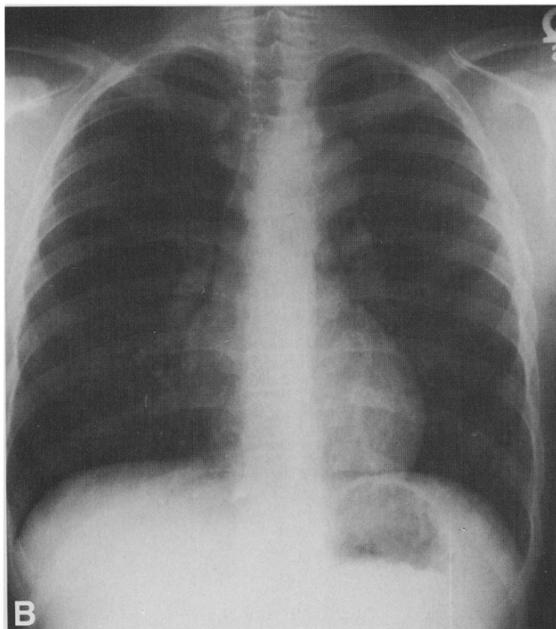
B

Figure 2. Comparison of aortic ratio measured by angiography (angio) with the ratio measured by two-dimensional echocardiography (echo) (B) and by computed tomography (C-T) of the chest (A).

factors correlated with the presence of an aortic aneurysm including age at repair, length of follow-up, presence of hypertension or a residual pressure gradient. Similarly, no surgical factor emerged as a clear risk factor. Posterior shelf resection, suture material and surgeon had no apparent bearing on late aneurysm formation. Two surgical factors, however, showed trends that might have become statistically significant with a larger sample size. First, previous coarctation resection (performed as the initial operation in five children who subsequently underwent patch aortoplasty for restenosis) appeared to have a protective effect after angioplasty. All patients with an aortic aneurysm had patch aortoplasty as the initial surgical procedure. None of the five



A



B

Figure 3. Patient 7. Serial chest X-ray films obtained 1.5 years (A) and 7 years (B) after patch aortoplasty document progressive enlargement of an aortic aneurysm.

patients in this series with an earlier resection developed an aneurysm after patch aortoplasty ($p = 0.17$). Second, in six of seven patients with an aneurysm, the coarctation had been repaired with a textile patch (Dacron), whereas only one had a polytetrafluorethylene nontextile patch (Gore-Tex, Impira or Teflon) utilized for the aortoplasty procedure ($p = 0.15$).

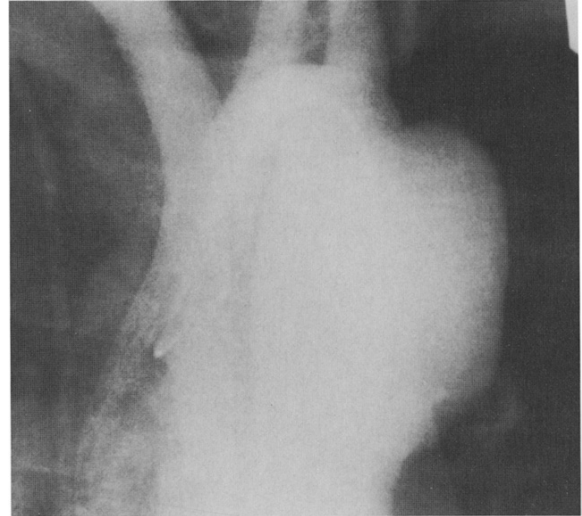


Figure 4. Patient 7. Aortogram demonstrating a large fusiform aneurysm at the site of the patch aortoplasty.

Discussion

Prevalence of aortic aneurysm after patch angioplasty. Using quantitative criteria we have documented a 24% prevalence rate of aortic aneurysms an average of 5.6 years after patch aortoplasty repair of coarctation in childhood. This finding is similar to the 26% prevalence rate found by Reuben et al. (5) and Clarkson et al. (7), but is higher than the 5% rate reported by Del Nido et al. (6). The discrepancy may relate in part to the subjective approach used to define an aneurysm in previous studies. Our study determined the prevalence of aneurysm prospectively on the basis of an objective measurable criterion. The aortic ratio of ≥ 1.5 (ratio of aortic diameter at the repair site to the diameter at the diaphragm) utilized to define the presence of an aortic aneurysm is not without precedent. It has been used to define aneurysms in other muscular arteries (9) and as a marker for intervention in adults with a peripheral arterial aneurysm (10). Our data (Fig. 1) indicate that an aortic ratio of ≥ 1.5 identifies a subgroup of children with patch aortoplasty repair of coarctation who have aneurysmal aortic dilation that is distinctly different from the aortic ratio of the majority of children after aortoplasty and from that of normal control subjects. Of practical importance, the aortic ratio provides an indexed measure for assessing aortic dilation in children of different ages and sizes, as well as longitudinally in the same child over time.

Noninvasive screening tests. The presence of a widened mediastinal shadow on chest X-ray film proved to be the most sensitive predictor (100% sensitivity) of the presence of an aortic aneurysm subsequently demonstrated by aortography. The chest X-ray film was also the least specific test,

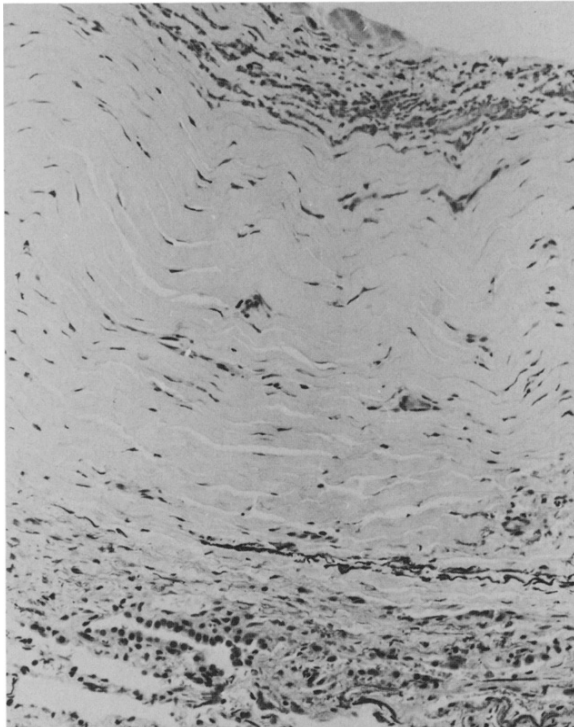


Figure 5. Patient 7. Biopsy of aneurysm wall opposite the Dacron patch. Surface fibrin and red cells overlie dense, broad, fibrous connective tissue with split disrupted peripheral elastic fibers. At the base, this is contained by adventitia. Biopsy specimen obtained at a site adjacent to the Dacron patch showed identical findings. (Movat pentachrome stain; original magnification $\times 330$, reduced by 10%.)

although specificity is of less concern in a screening test. The false positive studies were believed to have resulted from poststenotic dilation or from postoperative mediastinal changes. The sensitivity of echocardiography and chest computed tomography for detecting an aortic aneurysm (71% and 66%, respectively) was disappointing, but this may be due in part to the arbitrary nature of our definition of an aneurysm. Had we used an aortic ratio of ≥ 1.4 (rather than ≥ 1.5) for the noninvasive evaluation, the sensitivity of echocardiography and chest computed tomography would have increased to 100% and 83%, respectively. There was reasonable agreement between angiography and echocardiography or chest computed tomography in measuring the aortic ratio, particularly in the 1.0 to 1.5 range (Fig. 2). In the higher ranges, larger discrepancies in measured aortic ratio occurred, with echocardiography and chest computed tomography correctly predicting the presence of an aneurysm but typically overestimating the angiographic aortic ratio.

Possible mechanisms of aneurysm formation. Earlier studies failed to demonstrate clear risk factors that may predispose to the development of an aortic aneurysm after patch

aortoplasty. Previous investigations (4-7) suggested that aortic aneurysm may develop after patch aortoplasty because of a compliance mismatch between the stiff synthetic patch and the native aortic tissue. These reports cited the evaluation by Kinley and Marble (14) of the mechanical properties of host arteries and vascular substitutes. However, the latter studies were intended to evaluate an end to end or end to side anastomosis, not an onlay patch of the type used in patch aortoplasty. Furthermore, the compliance mismatch described pertains to increased wall stress at the anastomosis site and not at the aortic wall opposite the patch where the occurrence of an aneurysm after patch aortoplasty was first described (2-4).

Two trends in our data suggest that factors other than a compliance mismatch may predispose to the development of an aortic aneurysm after patch aortoplasty. First, aneurysmal aortic dilation occurred in 6 of 17 patients with a Dacron patch but in only 1 of 10 patients with a nontextile polytetrafluoroethylene patch (Table 1). Because polytetrafluoroethylene is 20% less compliant than Dacron (15), this finding is the reverse of what would be expected if compliance mismatch were the major cause of aneurysms in these patients. Local tissue reaction to the synthetic patch may be a more pertinent variable because it has been implicated in the breakdown of peripheral vascular suture lines and may lead to false aneurysm formation (16-21). Experimental work has demonstrated abundant foreign body giant cell infiltration into Dacron grafts, whereas polytetrafluoroethylene develops no foreign body reaction (16). It is possible that the inflammatory response to a Dacron patch makes it more susceptible to aneurysm formation than a polytetrafluoroethylene patch despite their compliance differences. A false aneurysm is the predominant form of aneurysm in adults after synthetic graft implantation for peripheral artery disease (18-21). Previous studies have not resolved the question of whether aneurysms after patch aortoplasty are predominantly true or false aneurysms. A true aneurysm was found in the single surgically treated patient in our series and in the report by McGoldrick et al. (22); however, four of five patients undergoing surgery in the series of Clarkson et al. (7) were found to have a false aneurysm.

Second, we identified no aortic aneurysm in patients who had undergone coarctation resection before patch aortoplasty (Table 1), an observation also made by Clarkson et al. (7). Histologic analyses (23) have demonstrated persistence of ductal tissue in the resected aortic wall of patients undergoing surgical repair of coarctation. Prior resection of this abnormal aortic tissue may have a protective effect against the development of an aortic aneurysm after subsequent patch aortoplasty. These observations suggest that abnormal ductal tissue may be involved in the genesis of a postoperative aneurysm, either by providing a weak anchor for sutures (increasing vulnerability to a false aneurysm) or

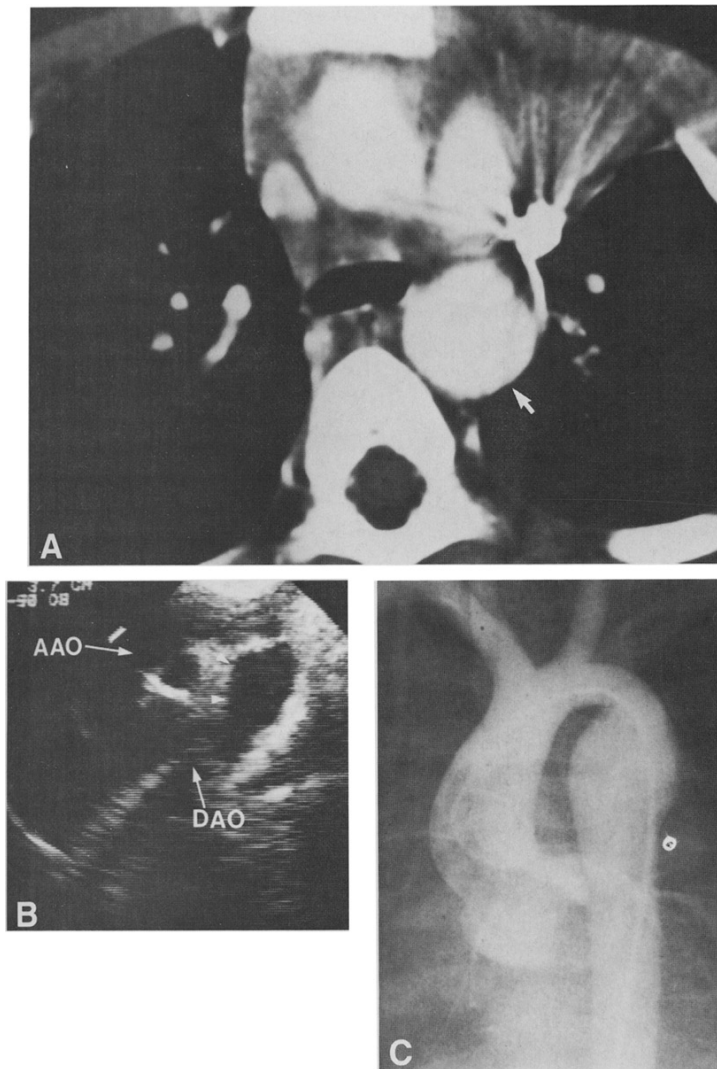


Figure 6. Patient 6. Multiple imaging of an aortic aneurysm, 5 years after Dacron patch aortoplasty repair of coarctation. **A**, Chest computed tomogram. The cross-sectional view demonstrates aneurysmal aortic dilation at the repair site (arrow). The aortic ratio measured 1.9. **B**, Two-dimensional echocardiogram. Suprasternal long-axis view of the aortic arch demonstrates marked aneurysmal dilation at the site of repair (arrowheads). The aortic ratio measured 2.3. AAO = ascending aorta; DAO = descending aorta. **C**, Aortogram demonstrating the fusiform aortic aneurysm at the site of patch aortoplasty. The aortic ratio was 1.6 by angiography.

as tissue more susceptible to increased wall stress (increasing vulnerability to a true aneurysm).

Conclusions. Aneurysmal aortic dilation occurs commonly after patch aortoplasty repair of coarctation in childhood. Historically, catastrophic rupture of such an aneurysm has been rare despite the relatively high prevalence of aortic aneurysm in this patient population. Because little is known regarding the natural history of aortic aneurysms after patch aortoplasty, however, we believe that all such patients must be followed up carefully. Our data indicate that aneurysmal aortic dilation at the site of coarctation repair can be imaged satisfactorily by echocardiography and chest computed tomography, and that these tools can provide a quantitative measure (the aortic ratio) with which to follow such patients. The potential role of nuclear magnetic

resonance imaging was not evaluated in this study, but our limited experience (Patient 3, Table 2) suggests that it also may provide an excellent noninvasive method for imaging postoperative aortic aneurysms. The chest X-ray film provides a sensitive screening test (although not specific) but not a quantitative method with which to follow these aneurysms longitudinally. The echocardiogram and chest computed tomography scan do provide such data and, in our opinion, should be repeated every 1 to 2 years in patients with chest X-ray evidence of an aortic aneurysm that is confirmed by aortography. If progressive aneurysmal dilation of the aorta is documented, surgical resection is probably indicated. It must be emphasized, however, that the natural history of such aneurysms remains unknown. For patients with a normal chest X-ray film, careful surveillance

every 2 to 3 years is warranted because of the progressive nature and sometimes late onset of aortic aneurysm in this patient group.

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