CHAPTER V

IS THERE A PLACE FOR PEDIATRIC VALVOTOMY IN THE AUTOGRAFT ERA?

Is there a place for pediatric valvotomy in the autograft era? A.J.J.C. Bogers, J.J.M. Takkenberg, A.P.Kappetein, P.L. de Jong, A.H. Cromme-Dijkhuis, M. Witsenburg. *Eur J Cardiothorac Surg 2001;20:89-94*

Abstract

Objective. Valvotomy and the autograft procedure are the most common surgical treatment options for children with valvular aortic stenosis. We evaluated the results of these surgical procedures in our institution.

Methods. Retrospective analysis was done of all patients presenting with aortic stenosis and operated upon before the age of 18. In 11 patients a valvotomy was performed and in 36 an autograft procedure.

Results. There was no hospital mortality. Mean follow-up in the valvotomy group was 4.8 years (SD 3.3), in the autograft group 4.5 years (SD 3.3). During follow-up 1 patient died suddenly 2 months after valvotomy. Two patients in the autograft group died (not valve-related). After valvotomy 3 patients underwent a balloon valvotomy, in 1 followed by an autograft procedure and 1 patient had a repeat valvotomy. In the autograft group 1 patient was reoperated for severe aortic regurgitation and moderate pulmonary stenosis. At last echocardiography after valvotomy (8 remaining patients) in only 2 patients (25%) no aortic stenosis or regurgitation was present. In the remaining 6 patients aortic stenosis is mild in 2 and moderate in 3, including 1 with moderate aortic regurgitation. In 1 patient without stenosis, moderate aortic regurgitation was seen. No pulmonary stenosis or regurgitation is present. Echocardiography after autografting (33 remaining patients) showed no aortic stenosis. Aortic regurgitation was mild in 7 patients, moderate in 2, severe in 1. Pulmonary stenosis was present in 2 patients (16%). Pulmonary regurgitation was mild in 3 patients and moderate in 1.

Conclusions. In selected patients with valvular aortic stenosis who are beyond infancy, valvotomy may be adequate and may postpone further surgery for a significant length of time. After valvotomy the main problem is residual aortic stenosis while after autografting a shift occurs to aortic regurgitation and problems related to the pulmonary valve. Careful clinical and echocardiographic follow-up is therefore warranted in young patients after the autograft procedure.

Key words

Aortic stenosis

Valvotomy

Autograft procedure

Introduction

Currently balloon valvotomy is considered the initial treatment of choice for valvular aortic stenosis in the pediatric age group, especially in neonates and infants. In addition balloon valvotomy is also used in selected cases of valvular stenosis in multilevel disease and in restenosis[1]. However, in a number of patients the balloon valvotomy is not indicated, unsuccessful or even failing[1-5]. These patients require surgical treatment. The most common surgical alternatives are valvotomy and the autograft procedure. The number of comparative studies evaluating the results of these two methods is limited and there are no randomized investigations[3-7]. In order to provide further information in this regard we present our experience with valvotomy and autografting for aortic stenosis in children.

Materials and methods

Study population. All patients younger than 18 years of age, who initially presented with aortic stenosis and who were consequently operated between September 1988 and May 2000, were analyzed. In 11 patients a valvotomy was performed and in 36 an autograft procedure. In 4 more pediatric patients an autograft procedure was done for primary isolated aortic valve regurgitation and in 11 additional pediatric patients an allograft root replacement was performed because no adequate autograft was present. No mechanical or biological aortic valve prostheses were implanted in this era in this age group. In the same time span 80 percutaneous balloon valvotomy procedures were done for valvular aortic stenosis in 72 patients in this age group. We limited our analysis to those patients presenting with aortic stenosis who underwent either valvotomy or an autograft procedure.

Patient characteristics, prior treatment, hemodynamic diagnosis and clinical condition at surgery are displayed in Table 1. In the autograft group 13 patients had prior balloon dilatation of the aortic valve. Prior surgery in the left ventricular outflow tract was performed in 17 patients in the autograft group and consisted of single or combined enucleation of discrete subaortic stenosis in 4 patients, of valvotomy in 14 patients and of aortic valve repair in 2 patients. In the valvotomy group 3 patients had a previous unsuccessful balloon dilatation. In 1 of the 3 patients in the valvotomy group who had coarctation repair this procedure was combined with closure of a patent arterial duct. Other prior cardiac surgery

Table 1. Patient characteristics, prior treatment, hemodynamic diagnosis and clinical condition at surgery.

	Valvotomy (n=11)	**Autograft (n=36)**	**p-value**
Patient characteristics			
Mean age (years)	2.7 (1w-13.7)	9.8 (0.3-16.2)	<0.001
Mean weight (kg)	10.5 (3.2-34)	44 (4.0-16.2)	<0.001
Female/male	6/5 (55/45%)	10/23 (33/67%)	ns
Prior treatment			
Balloon valvuloplasty	3 (27%)	13 (36%)	ns
LVOT surgery	-	17 (47%)	<0.003
Coarctation repair	3 (27%)	6 (18%)	ns
Other cardiac surgery	1 (9%)	2 (5%)	ns
Hemodynamic diagnosis			
Valvular AS	9 (82%)	7 (19%)	<0.001*
Valvular AS/AR	0	20 (56%)	
Valvular AR	0	4 (11%)	
Multilevel AS(AR)	2(0) (18%)	5(1) (11%)	
Clinical condition			
Surgery urgent/elective	3/8 (27/73%)	8/28 (22/78%)	ns
NYHA class I-II/III-IV	8/3 (73/27%)	32/4 (88/12%)	ns

LVOT: left ventricular outflow tract. AS: aortic stenosis. AR: aortic regurgitation. NYHA: New York Heart Association. * Pearson Chi-square test.

consisted in the valvotomy group of one-stage repair of an atrial septal defect and aortic arch interruption in 1 patient and in the autograft group of closure of ventricular septal defect in 2 patients.

Peri-operative data (Table 2). All patient were operated with standard cardiopulmonary bypass techniques using bicaval cannulation, moderate hypothermia and cardioplegic arrest. Cross-clamp and perfusion times were significantly longer in the autograft group, and are displayed in Table 2. Additional circulatory arrest (mean duration 29 minutes; SD 14, range 28-36 minutes) was used only in the valvotomy group in the 3 patients who had one-stage procedures. Valvotomy was done sharply through the opened ascending aorta and according

Table 2. Peri-operative data

	Valvotomy (n=11)	Autograft (n=36)
Cardio-pulmonary bypass		
Cross-clamp time	36 minutes (SD 18;range 16-71)	127 minutes * (SD 24;range 90-203)
Perfusion time	75 minutes (SD 38;range 31-151)	183 minutes * (SD 58;range 129-465)
Concomitant procedures		
VSD closure	3	0
Enucleation discrete subaortic stenosis	1	2
Coarctation repair	3	0
LVOT enlargement	0	4
Other	0	2
Allograft characteristics (n=33)		
Cryopreserved/fresh		32/1
Mean diameter (SD; range)		22 mm (SD 4; range 13-28 mm)
Aortic/pulmonary		4/29
Postoperative		
Hospital mortality	0	0
Permanent pacemaker implantation	0	1
Bleeding requiring reoperation	0	0

* P<0.001; VSD = ventricular septal defect; LVOT = left ventricular outflow tract.

to the anatomy of the valve at the discretion of the attending surgeon. The modified Ross procedure with autograft aortic root replacement with reimplantation of the coronary arteries and pulmonary artery replacement with an allograft was used in all autograft patients.

Twenty-nine cryopreserved allografts were obtained from the heart valve bank in Rotterdam (Heart Valve Bank Rotterdam, Rotterdam, The Netherlands), 4 from Barcelona (Transplant Services Foundation, Hospital Clinic I Provincial de Barcelona, Barcelona,

Barcelona, Spain), and 2 from Berlin (Homograft Laboratory, Deutsches Herzzentrum Berlin, Berlin, Germany). One fresh allograft was obtained from London (National Heart Hospital, London, UK). Allograft characteristics are displayed in Table 2.

Concomitant procedures in the valvotomy group consisted of closure of ventricular septal defect in 3 patients, 1 of which was a one-stage procedure including coarctation repair, an enucleation of discrete subaortic stenosis was performed in 1 patient and coarctation repair in 2 more patients. In the autograft group concomitant procedures were enucleation of discrete subaortic stenosis in 2 patients, enlargement of the left ventricular outflow tract in 4 patients, enlargement of the ascending aorta in 1 and ligation of a patent arterial duct in 1.

Operative mortality was absent. There were no bleeding complications necessitating rethoracotomy in either group. One patient in the autograft group needed a permanent pacemaker postoperatively because of total heart block.

Follow up. Clinical follow up was obtained by collecting information from the patient records. Data on death, reoperation, occurrence of valve-related events[8] and NYHA classification were extracted. Autograft failure was defined as observed valve failure at death, reoperation or at echocardiographic examination.

Echocardiography during follow up was compiled from all records of postoperative echocardiographic examinations. Valve function was assessed by echo Doppler studies including color flow mapping. Aortic regurgitation was scored using a jet length grading system of 0 to 4. Grade 0 was defined as no regurgitation, grade 1 as trivial, grade 2 as mild, grade 3 as moderate and grade 4 as severe. Aortic stenosis was scored in 4 grades. No or trivial stenosis was defined as a gradient up to 10 mmHg, mild as gradient of 10 to 30 mmHg, moderate as gradient of 30 to 50 mmHg, severe as a gradient over 50 mmHg. Autograft failure was defined as aortic regurgitation of more than grade 2 or aortic stenosis of more than 50 mmHg. Pulmonary regurgitation was also scored using a jet length grading system of 0 to 4. Grade 0 was defined as no regurgitation, grade 1 as trivial, grade 2 as mild, grade 3 as moderate and grade 4 as severe. Pulmonary stenosis was scored in 4 grades. No or trivial stenosis was defined as a gradient up to 10 mmHg, mild as gradient of 10 to 30 mmHg, moderate as gradient of 30 to 50 mmHg, severe as a gradient over 50 mmHg.

Statistical analysis. All data were entered into a relational database, MS Access 97 (Microsoft, Redmond, WA) and all analyses were done using SPSS 9.0 for Windows (SPSS, Chicago, Ill). Means were compared using one way analysis of variance. Proportions were compared using the chi-square test. Cumulative survival analysis was conducted according to the method of Kaplan and Meier[9].

To study the progression of echocardiographic aortic regurgitation over time after autograft procedure, for each patient (N=32) who had at least 2 post-operative echocardiographic examinations a linear regression function was constructed of the relationship between time and the grade of their aortic regurgitation. In addition an overall linear regression function was constructed for all 32 patients combined, in order to illustrate the overall progression of aortic regurgitation over time in the whole group.

Results

Follow-up. No patients died during hospitalization or within 30 days after the procedure. Follow-up was 100% complete at the closing date of the study (August 2000) with a total follow-up of 161 patient years. Mean follow-up in the valvotomy group was 4.8 years (SD 3.3, range 0.1-10.8 years, 53 patient years), in the autograft group 4.5 years (SD 3.3, range 1 week – 11.5 years, 161 patient years).

During follow-up 1 patient died suddenly 2 months after valvotomy. Two patients in the autograft group died, one after 1.5 month as a result of a systemic Candida infection (not valve-related), one after 21 months due to progressive congestive heart failure (aortic regurgitation 1+ after 17 months; not valve-related).

After valvotomy 1 patient had a balloon valvotomy after 1 month and eventually an autograft procedure after 1.3 years, 2 patients had a balloon valvotomy after 3.3 and 9.5 years respectively. In 1 patient valvotomy was repeated after 9.2 years. In the autograft group 1 patient was reoperated after 7.5 years for severe aortic regurgitation and moderate pulmonary stenosis. A bileaflet mechanical aortic valve prosthesis (St. Jude, St. Paul, MN) was implanted in the autograft root and the pulmonary allograft was replaced by a new pulmonary allograft (Heart Valve Bank Rotterdam, Rotterdam, The Netherlands). In addition 1 patient was reoperated after 9.8 years for severe pulmonary stenosis, a new pulmonary allograft (Heart Valve Bank Rotterdam, Rotterdam, The Netherlands) was inserted. One patient was re-operated after 6.9 years for mitral valve replacement and tricuspid valve repair. Neither thrombo-embolic events nor endocarditis were recorded after valvotomy or autografting.

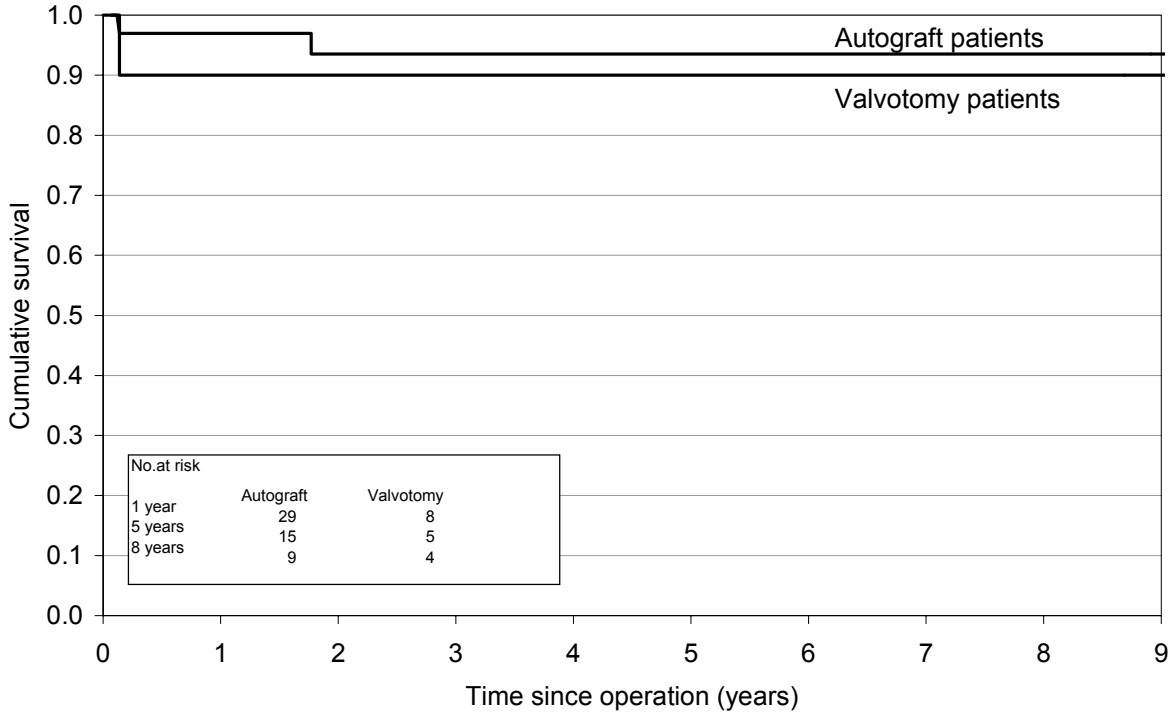

Figure 1. Cumulative survival in autograft and valvotomy patients who presented with the diagnosis of aortic stenosis.

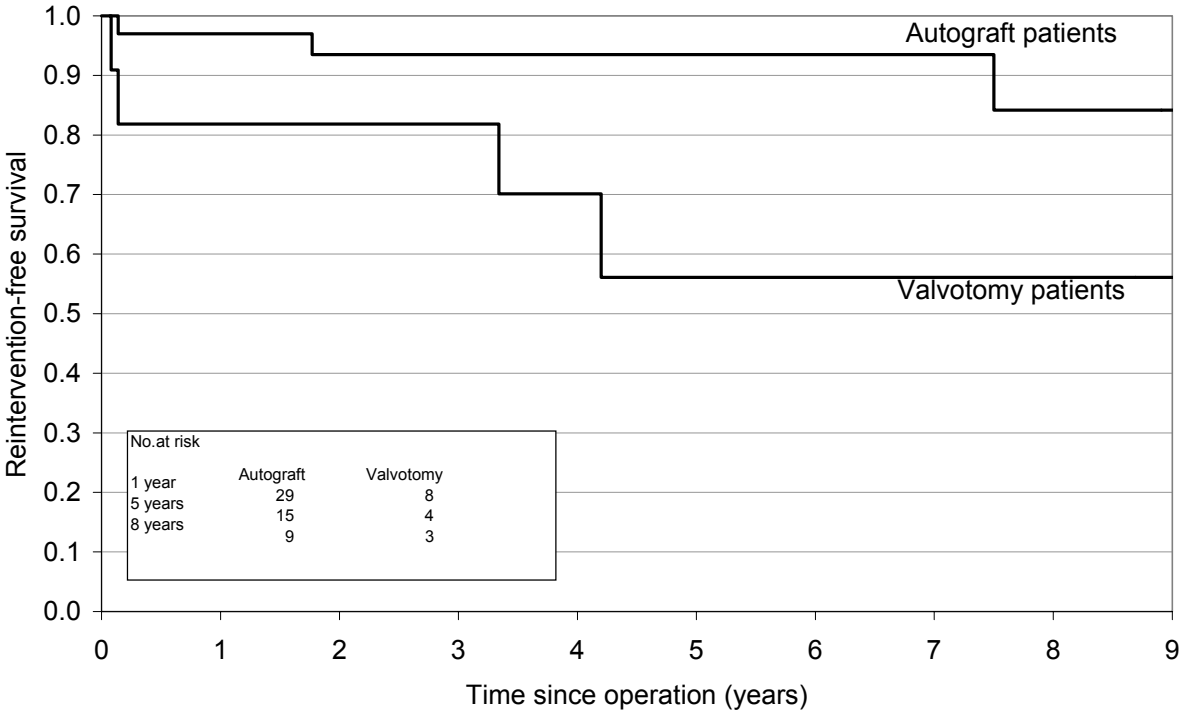

Figure 2. Reintervention-free survival in autograft and valvotomy patients who presented with the diagnosis of aortic stenosis.

Cumulative survival (Figure 1) after valvotomy was 90% at 5 and 8 years (se 9%), reintervention-free survival was 56% at 5 and 8 years (se 17%. Survival after autografting was 94% at 5 and 8 years (se 4%), reintervention-free survival was 94% at 5 years (se 4%) and 84% at 8 years (se 10%) see Figure 1 and 2.

Functional class. At last follow up all patients after valvotomy are in NYHA class I. After autografting all patients are in NYHA class I, except for 1 patient in class II.

Table 3. Echocardiographic findings at last examination

	Valvotomy (n=8)	**Autograft (n=34)**
Aortic regurgitation		
0-1+	5	12
2+	2	8
3+	1	3
4+	0	1*
Aortic stenosis		
No	3	34
Mild	2	0
Moderate	3	0
Pulmonary regurgitation		
0-1+	8	13
2+	0	3
3+	0	1
4+	0	1
Unknown	0	16
Pulmonary stenosis		
No	8	16
Mild	0	0
Moderate	0	2*
Unknown	0	16

* One patient underwent replacement of the autograft and allograft shortly hereafter.

Echocardiography (Table 3). At last echocardiographic examination after valvotomy (8 remaining patients) aortic stenosis is absent in 3 patients, including 1 patient with moderate aortic regurgitation. Aortic stenosis is mild in 2 and moderate (systolic Doppler gradient over 40 mmHg) in 3, including 1 with moderate aortic regurgitation. In only 2 patients no stenosis or regurgitation is present. No pulmonary stenosis or regurgitation is present in this group.

In the autograft group 34 patients had at least 1 echocardiographic examination postop. In total 129 echocardiographic examinations were performed. At last echocardiographic examination no aortic stenosis was observed. Aortic regurgitation was none or trivial in 22, mild in 8, moderate in 3, severe in 1 (this patient was reoperated shortly thereafter), not recorded in 2. Pulmonary stenosis was present in 2 patients (systolic Doppler gradient 40 mmHg). Pulmonary regurgitation was none or trivial in 13, mild in 3, moderate in 1, severe in 1, not recorded in 16.

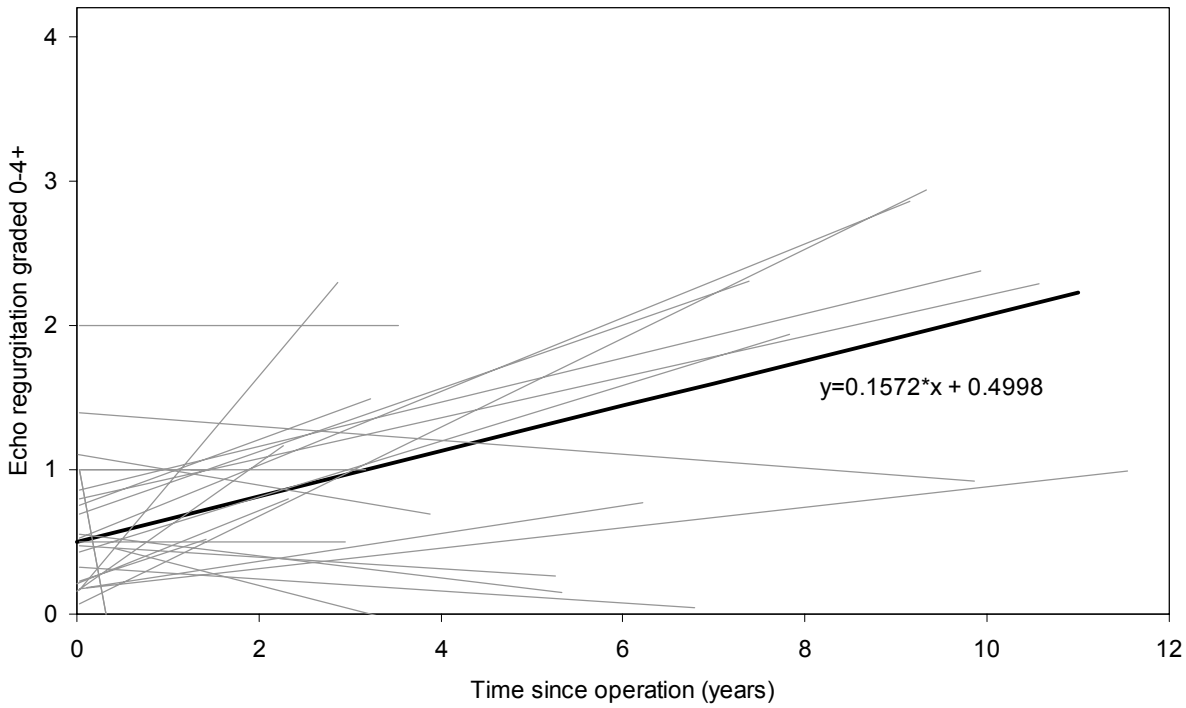

Figure 3. Progression of echocardiographic aortic regurgitation (jet length) over time in 32 patients after autograft procedure. For each patient the linear regression function that reflects the individual progression of aortic regurgitation over time is displayed (grey lines). The bold line represents the overall progression of echocardiographic aortic regurgitation for the whole group.

Thirty-two patients had 2 or more echocardiographic examinations. Figure 3 displays for each patient the linear regression line that reflects the progression of echocardiographic aortic regurgitation over time. The bold line represents the overall progression of echocardiographic aortic regurgitation of the whole autograft group over time, which can also be described by the equation: $y = 0.1572 * x + 0.4998$.

Discussion

This study describes our institution's experience with valvotomy and autografting in children with valvular aortic stenosis. When comparing the patients undergoing valvotomy or autografting, it is noted that the valvotomy group is much younger at the time of operation. The valvotomy group also had less prior surgery on the left ventricular outflow tract. Furthermore, in the valvotomy group the indication for surgery was exclusively aortic stenosis while in the autograft group most patients were diagnosed with combined aortic stenosis and regurgitation. This is in concordance with previous observations[7].

Concomitant procedures are also different between the two groups. In the valvotomy group resection of coarctation and closure of ventricular septal defect were done with some frequency. In the autograft group additional left ventricular outflow tract operations were done more frequently. Intra-operative analysis led to more of these interventions than preoperative diagnosis. Overall, autograft procedures took longer perfusion and cross-clamp times than aortic valvotomy.

There was no hospital mortality in either group. Late mortality occurred once after valvotomy and twice after autografting. In this respect the groups are comparable.

At medium term follow-up the remaining (n=9) patients in the valvotomy group are asymptomatic (all NYHA class I), but a substantial number will probably require reoperation or balloon valvotomy in the future mainly because of considerable residual or recurrent valvular aortic stenosis and in some patients because of post-interventional aortic regurgitation. This concurs with previous observations[6]. In our series after valvotomy a reoperation on the aortic root took place in 2 patients and balloon dilatation of the aortic valve in 2 more patients.

In the autograft group (n=33) most patients are also doing well (91% in NYHA class I), although 12 patients have mild or moderate aortic regurgitation at the last echocardiographic examination. In 1 patient a prosthetic aortic valve replacement of the

regurgitant autograft (echo aortic regurgitation 4+) was performed, while simultaneously the pulmonary allograft was replaced with a new allograft valved conduit because of moderate valvular and severe supravalvular pulmonary stenosis. Isolated pulmonary stenosis developed in 2 patients, requiring replacement of the allograft in 1.

Several groups have reported that the autograft conveniently increases in diameter in children, mostly concordant with growth[10, 11]. This diameter increase takes place for about half in the first few days after operation and for the last half in the further first year after operation[10, 12]. However, the diameter increase also takes place in adults where this is not appropriate and is associated with a higher incidence of aortic regurgitation[10, 12]. For these reasons close follow-up after autografting is necessary to monitor the value of this procedure. According to the serial analysis of echocardiographic aortic regurgitation over time in patients after the autograft procedure, there is indeed reason to be cautious. Although initial aortic regurgitation is minimal (0.5+ grade directly postop), there is clearly progression of aortic regurgitation over time. The clinical relevance of these observations will become clear in the next 5-10 years.

A limitation of the study is that this comparison of two surgical treatments is hampered by the retrospective nature of the study, by the difference in indication for surgery in both patient groups, and by the evolving experience with the autograft procedure during the study time period.

In conclusion, there is indeed a place for pediatric valvotomy in the autograft era. In children with valvular aortic stenosis, selected by unsuccessful balloon valvotomy or by concomitant cardiac anomalies such as ventricular septal defect, discrete subaortic stenosis or coarctation of the aorta, valvotomy may be adequate and will postpone further surgery for a significant length of time. After valvotomy the main problem is residual aortic stenosis while after autografting the problem shifts towards aortic regurgitation and problems related to the pulmonary valve. The autograft procedure may very well solve problems at the left ventricular outflow tract, but is not yet the perfect solution.

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