Discrete supravalvular aortic stenosis in children: Is it necessary to reconstruct the whole aortic root?

Çocuklarda diskret supravalvüler aort darlığı: Tüm aort kökü rekonstrükte edilmeli mi?

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Abstract

Objective: Discrete supravalvular aortic stenosis (SAS) is known to involve the whole aortic root. Some surgeons have therefore changed their approach from relief of obstruction using a single-patch to symmetric reconstruction of the whole aortic root – three-patch technique. The advantages are said to be preserved long-term aortic valve function and allowance for growth. This is unproven. We compare growth and aortic root geometry in patients who have undergone relief of discrete SAS using either single-or three-patch technique.

Methods: Twenty-five patients (14 male, 11 female, mean age of 11± 4 years, range 4-18) underwent surgery for discrete SAS. No patients with diffuse SAS were included in this retrospective analysis. Twelve patients had features of Williams syndrome. Five patients had other concomitant procedures. A single-patch was inserted into the longitudinal incision, which passed across the stenosis into the non-coronary sinus in 14. A three-patch technique was used in 11 patients. Changes in aortic root following repair were documented in patients using both echocardiography and magnetic resonance imaging (MRI).

Results: There were no operative deaths. The mean preoperative gradient was 66 ±17 mmHg (range 50 – 100 mmHg), which decreased to 14±7 mmHg (range 4-18 mmHg) early postoperatively. The late mean gradient was 15±5 mmHg. There was no significant difference in the incidence of postoperative aortic regurgitation or gradient across the repair between two techniques according to the echocardiograms and MRI findings.

Conclusion: According to our study, we cannot demonstrate any benefit in reconstructing the whole aortic root for discrete SAS. A single-patch technique is easy, safe and appears durable. *(Anadolu Kardiyol Derg 2009; 9: 311-7)*

Key words: Discrete supravalvar aortic stenosis, left ventricular outflow tract obstruction, surgical procedures

Özet

Amaç: Diskret supravalvüler aort darlığı (SAS) tüm aort kökünü tutar. Bu sebeple bazı cerrahlar obstrüksiyonu gidermek için kullanılan tek yama tekniğinden; simetrik rekonstrüksiyon sağlayan üç yama tekniğine doğru, yaklaşımlarını değiştirmişlerdir. Bunun avantajının uzun dönemde aort kapağının fonksiyonunu koruduğu ve buna bağlı da büyümeyi sağladığı söylenmektedir. Bu henüz ispatlanmamıştır. Bu çalışmamızda diskret SAS sebebiyle tek ya da üç yama teknikleriyle ameliyat edilmiş hastalardaki aort kökü büyümesini ve geometrisini inceledik.

Yöntemler: Yirmi beş hasta (14 erkek, 11 kadın, ortalama yaş 11±4 yıl, dağılım 4-18 yıl) diskret SAS sebebiyle ameliyat edildi. Difüz SAS hastaları bu retrospektif çalışmaya dâhil edilmedi. On iki hastada Williams sendromu bulguları vardı. Beş hastaya ek prosedürler yapıldı. On dört hastaya darlık boyunca yerleştirilen ve non-koroner sinüse uzanan tek yama kullanıldı. Üç yama tekniği 11 hastaya uygulandı. Aort kökü değişiklikleri ekokardiyografik ve manyetik resonans görüntüleme (MRI) ile değerlendirildi.

Bulgular: Operatif mortalite sıfır idi. Ortalama preoperatif gradiyent 66 ±17 mmHg'dan (range 50–100 mmHg), postoperatif olarak 14±7 mmHg'ya (range 4-18 mmHg) inmiştir. Geç ortalama gradiyent ise 15±5 mmHg olarak tespit edilmiştir. Ekokardiyografi ve MRI bulgularında postoperatif aort yetmezliği ve düzeltme yapılan bölge boyunca alınan gradiyent açısından her iki cerrahi teknik açısından anlamlı bir fark gösterilememiştir. İki hastaya reoperasyon yapılmıştır.

Sonuç: Çalışmamıza göre; diskret SAS hastalarında, tüm aort kökünün rekonstrükte edilmesinin bir avantaj sağladığını söyleyemeyiz. Tek yama tekniği; kolay, güvenli ve uzun ömürlü görünmektedir. *(Anadolu Kardiyol Derg 2009; 9: 311-7)*

Anahtar kelimeler: Diskret supravalvüler aort darlığı, sol ventrikül çıkım yolu darlığı, cerrahi işlemler

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Introduction

Supravalvular aortic stenosis (SAS) was first described by Mencarelli in 1930 (1). It exists in two forms; a discrete form in which the narrowing is localized to the supravalvar area of the ascending aorta, or a diffuse form in where the narrowing affects the whole length of the ascending aorta and a variable amount of the arch and brachiocephalic vessels (2-4). McGoon et al. (5) reported the first description of surgical correction of discrete SAS in 1961 using a tear dropped shape patch performed in 1956 by Kirklin. In between these dates, several other successful corrections using similar, or alternate techniques had been performed (6, 7).

Supravalvular aortic stenosis, however, is known to involve the whole aortic root and this has led to more intricate procedures being proposed as the optimal surgical approach (8). These techniques aim to restore the whole aortic root thus allowing for growth. The first of these procedures has been attributed to Brom and involves excision of the narrowing ridge and enlargement of each sinus using a triangular prosthetic patch (9). Myers et al. (12) modified this technique by patching of each sinus with advancement flaps from the ascending aorta.

It is not possible to determine from the published series whether the three-patch technique confers benefit over the tear dropped shape patch for several reasons. Firstly, the relative rarity of this condition, a total of 200 cases in published series, which extend over 40 years, make meaningful comparisons difficult. Secondly, the series include a wide spectrum of patients from those with more complex associated cardiac lesions. Thirdly, there are few meaningful and clearly comparable end points and when consideration has been given to these they have changed over the past 40 years.

Our practice has reflected the evolving understanding of the morphology of SAS. We review the experience of two pediatric cardiac surgery units at Ufuk University and Başkent University in Ankara between 1997 and 2006 and compare changes in root growth between patients who underwent a single-patch repair and those who underwent a three-patch technique repair.

Methods

Patient population and data collection

We retrospectively reviewed the records of 25 patients operated on discrete SAS. No patients with diffuse SAS were included in the analysis. The individual patient demographics and associated lesions are shown in Table 1. There were 14 male, 11 female with a mean age of 11±4 years (range 4-18). Among them 12 patients had features of Williams syndrome.

Nine patients had associated pulmonary artery stenoses, in only 3 however were these deemed significant enough to require enlargement. The other 6 patients with unoperated mild pulmonary branch stenosis remained free of symptoms. Two patients had other additional concomitant procedures, one aortic valvotomy and one-patch enlargement to a stenosed left carotid artery.

Follow-up ranged from 2 to 11 years (mean 6.8 ± 2.7 years) and was 100% complete in 25 patients.

Operation

Standard techniques for cardiopulmonary bypass were used. Cold potassium crystalloid or blood cardioplegia was used for myocardial protection. Although Dacron (CR Bard, Covington, GA) was used as patch material in the past, bovine pericardium (Periguard, Synovis Life Technologies St. Paul, MN, USA), which was our choice of interest, became available and has been used as a patch afterwards. Fourteen patients underwent a single-patch repair and 11 patients underwent a three-patch repair technique for the correction of SAS. A tear dropped shaped single patch was inserted in the ascending aorta after longitudinal incision through the stenotic region extending into the non-coronary sinus. For three-patch technique, after transecting the ascending aorta with complete excision of the fibrous ridge; a triangular shaped patch enlargement of all the three sinuses with three separate bovine pericardial patches were performed.

Echocardiography

Postoperative gradients and degree of aortic incompetence were assessed before discharge and in outpatients clinic 1 year after the operation using two-dimensional and Doppler echocardiography with a Vivid 7 (GE, Vingmed, Milwaukee, WI, USA) machine.

Magnetic resonance imaging

Aortic root geometry in the operated roots was assessed using magnetic resonance imaging (MRI). In 2008, all patients were invited for an MRI scan to assess the late-term results of the supravalvular repair. All patients accepted our invitation. The MRI examinations were performed on a 1.5 T MR scanner (Signa Excite HD, GE Medical Systems, Milwaukee, WI, USA) and the aortic root measurements were performed at four levels: at the annulus of the aortic valve, at the sinus of Valsalva, at the sinotubular junction, and at 1 cm above the sinotubular junction (Fig. 1). The ratios of the annulus diameter, sinus of Valsalva

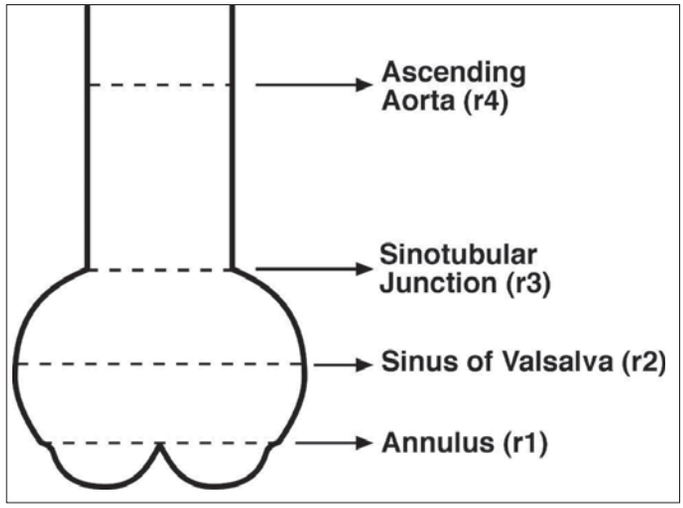

Figure 1. Four levels of measurement from magnetic resonance imaging scan

Tablo 1. Preoperative demographics, operative and postoperative details

Patient No	Year of operation	Age at operation	Sex	Williams syndrome	Associated lesion	Preop gradient (mmHg)	Preop AR/AS	Repair technique	Concomitant procedure	Early postop gradient (mmHg)	Late postop gradient (mmHg)	Postop AR	Gradient reduced (mmHg)
1	1997	4	M	N		65	No	1		10	10		55
2	1997	4	F	N		60	AS	1		15	15		45
3	1997	5	F	N		100	No	1	Left carotid patchplasty	10	10		90
4	1998	6	M	Y		100	No	1		15	15		85
5	1998	4	M	Y	Mild RPA stenosis	100	No	1		25	25		75
6	1999	5	M	Y		50	AR	1		15	15	Trivial AR	35
7	1999	4	M	Y	Mild PS	90	No	1		20	20		70
8	1999	4	M	N		90	AS	1	Aortic valvotomy	30	30		60
9	2000	4	F	Y		50	No	3		10	10		40
10	2000	3	M	N	Mild RPA stenosis	55	No	3		15	15		40
11	2000	3	M	Y		65	No	3	Enlarge PA stenosis	15	15		50
12	2001	3	M	N		50	No	3		10	10		40
13	2001	10	M	Y	Mild LPA stenosis	70	No	3		20	20		50
14	2002	6	M	N		50	AR	3		10	10	Trivial AR	40
15	2002	5	F	Y		70	No	3		15	15	Trivial AR	55
16	2002	5	F	N		75	AR	3		25	25		50
17	2002	1	F	Y	Valvar PS, L and R PA stenosis	50	No	3	Enlarge PA stenosis	20	20		30
18	2003	4	F	N		50	No	3		0	0	Trivial AR	50
19	2003	13	F	N	Mild supravalvar PA stenosis	60	No	3		10			50
20	2004	3	F	Y		50	No	1		10			10
21	2004	5	M	N		80	No	1		0		Trivial AR	80
22	2005	1	M	N	Mild supravalvar PA stenosis	50	AR	1	Enlarge PA stenosis	10		Trivial AR	40
23	2005	2	F	Y	Mild LPA stenosis	60	No	1		15			45
24	2005	3	M	Y	Mild LPA stenosis	50	No	1		20			30
25	2006	4	F	N		60	No	1		10			50

1- single-patch technique, 3 - three-patch technique,

AR - aortic regurgitation, AS - aortic stenosis, F - female, L - left, LPA: left pulmonary artery, M - male, PA - pulmonary artery, Preop. - preoperative, Postop. – postoperative, PS - pulmonary stenosis, R – right, RPA - right pulmonary artery

diameter, sinotubular junction diameter and the aortic diameter 1 cm above the sinotubular junction to the diameter of the descending thoracic aorta at diaphragmatic level were named as r1, r2, r3 and r4, respectively. Four observers recorded the measurements and mean values, standard deviations and standard errors were calculated to minimize the measurement errors (Fig. 2-3). Growth was potentially complete by age of 16 and therefore any patient older than 16 years at initial operation was excluded from growth analysis. Echocardiography was also performed during outpatients clinic visit.

Statistical analysis

The SPSS for Windows version 11.5 statistical package (SPSS Inc, Chicago, III, USA) was used to perform statistical analysis. Continuous variables are expressed as means ± SD throughout the article. Nominal variables are expressed as frequencies and percentages. Kolmogorov-Smirnov normality test was performed for gradients and R ratios. Gradient values were not normally distributed. The relation between age and gradients were analyzed by Spearman-Rank Correlation Analysis. As a significant relation between preoperative gradients and the age was identified, correction was performed to eliminate the effect of age ($y'=y-b*(age-mean_{age})$). In this formulate y' states the adjustment gradient where y is the true gradient and b is the regression coefficient (9, 10). After performing Kolmogorov-Smirnov normality test, the related gradient values were acceptable to normal distribution. Age, follow-up time, gradient and R values were compared for both surgical techniques by using Student *t* test. For independent samples, sex and concomitant William syndrome finding comparisons were performed by Fisher-Exact test. Time dependent gradient values for both techniques were compared by paired *t* test. The time dependent gradient changes between the two techniques were studied by repeated measurement of two-way ANOVA test was performed. The comparisons were evaluated at a significance level of 0.05.

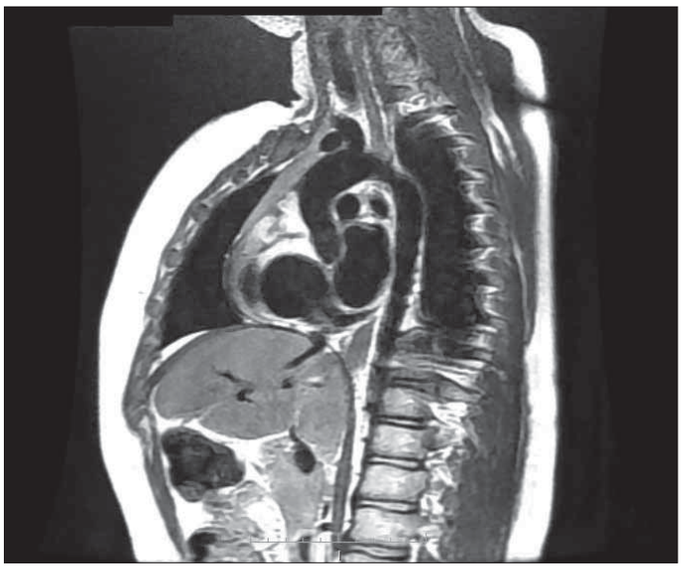

Figure 2. Sagittal magnetic resonance imaging of the thoracic aorta

Results

The individual operative and postoperative details are shown in Table 1. There were no operative deaths. All patients were alive at follow-up. Descriptive values for age, follow-up, sex and Williams syndrome are shown in Table 2. There was no significant difference between the two techniques. As age was found positively related with the gradient, the preoperative and postoperative gradient values were corrected (Table 3). The mean preoperative ventriculo-aortic gradient was 66±17 mmHg (range 50-100 mmHg), which decreased to 14±7 mmHg (range 4-18 mmHg) early postoperatively. The late mean gradient was 15±5 mmHg. The comparison of two techniques and time dependent gradient changes for both surgical techniques are shown in Table 4. According to this, the preoperative gradient values were significantly low in three-patch technique ($p<0.01$), while it was not significant for early postoperative and late postoperative period ($p>0.05$). After comparing the preoperative

Table 2. Demographic and clinical characteristics

Parameters	Single-Patch (n=14)	Three-Patch (n=11)	p*
Age at operation, years	3.86±1.29 (1-6)	5.18±3.46 (1-13)	NS
Age today, years	10.86±4.59 (4-16)	11.73±3.20 (7-18)	NS
Follow-up, years	7.07±3.61 (2-11)	6.55±3.61 (2-11)	NS
**Sex, n(%)			
Male	9 (64.3)	5 (45.5)	NS
Female	5 (35.7)	6 (54.5)	
**Williams syndrome, n(%)			
No	7 (50)	6 (54.5)	NS
Yes	7 (50)	5 (45.5)	

Data are expressed as means ± SD and proportion/percentage
*Student's unpaired t test and **Fisher-Exact test for sex and Williams syndrome
NS - not significant

gradient with the early and late postoperative gradients, there was significant reduction in gradient values for both techniques in early postoperative period. No statistically significant differences between the two techniques were identified in late postoperative gradient values. Although the time related changes between the two groups showed statistical difference ($p<0.05$), it was figured out that the difference was due to the preoperative values (Fig. 4).

Preoperatively 2 patients had mild aortic stenosis and 4 patients-aortic regurgitation. From the single-patch group, 3 out of 14 patients had postoperative trivial aortic regurgitation, 2 of whom had aortic regurgitation preoperatively. Postoperatively there was a trace of aortic regurgitation in 3 out of 11 patients who underwent three-patch technique repair, 1 of whom had aortic regurgitation preoperatively.

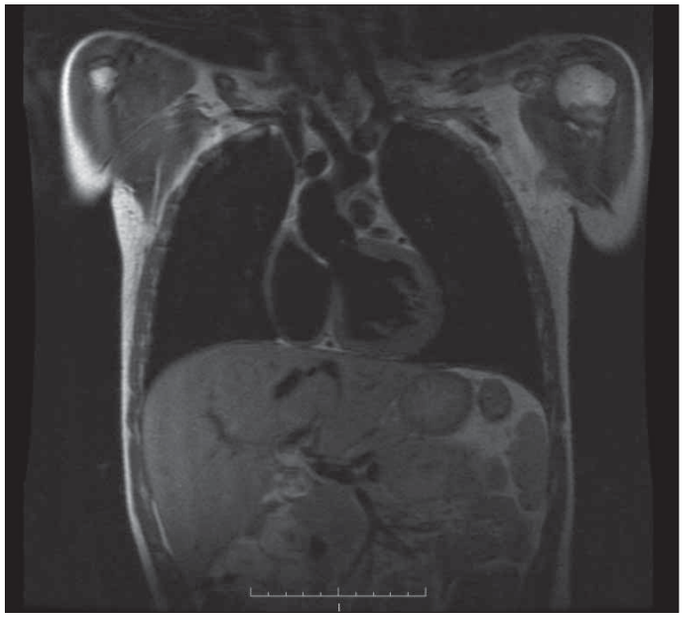

Figure 3. Coronal magnetic resonance imaging of the thoracic aorta

Table 3. Correlations between age and ventriculo-aortic gradient values

	Preop.	Postop.	Late
Age today	r=0.508	r=0.178	r=0.084
	p<0.01	NS	NS

Spearman-Rank correlation analysis
LATE - late postoperative, NS - not significant, PREOP - preoperative, POSTOP - postoperative

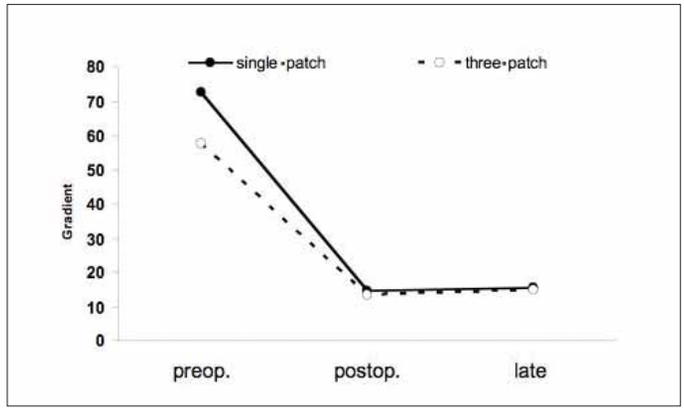

Figure 4. Time dependent changes in gradient values for single-patch and three-patch techniques by repeated measurements of two-way ANOVA test

There was no significant difference in the incidence of postoperative aortic regurgitation or early and late ventriculo-aortic gradient across the repair between the two techniques ($p>0.05$).

Two patients in single-patch group required reoperation. The first patient (No. 1) developed a significant gradient across the aortic valve associated with moderate regurgitation 8 years following correction of his SAS. He required an aortic valve replacement (AVR). The second patient (No. 8, age-4 years) who underwent an aortic valvotomy of a bicuspid valve at the time of SAS correction required relief of a juxtaductal coarctation 2 years later and AVR with enlargement of his aortic root 7 years after his SAS correction. The indication for the AVR was stenosis. The aortic root was enlarged in order to implant a large size mechanical valve to avoid further reoperation.

Aortic root geometry and growth were assessed by MRI scan. There were no significant differences between the MRI measurements of the created ratios and the operative technique (Table 5).

Postoperatively there were 19 patients (76%) in New York Heart Association class I and 6 patients (24%) in class II (Table 6).

Discussion

Congenital supravalvular aortic stenosis is a rare congenital cardiac anomaly occurring as a major feature of Williams syndrome. Despite the development of several surgical techniques aimed at an anatomic restoration of the entire aortic root, the

Table 4. The comparison of time dependent gradient changes for both surgical techniques

	Preoperative gradient	Early postoperative gradient	Late postoperative gradient	p* Pre. vs.Early P. gradients	p* Pre. vs Late P. gradients	p* Early P. vs Late P. gradients
Single- patch	72.61±16.20	14.76±7.22	15.39±4.54	<0.001	<0.001	NS
Three-patch	57.58±9.54	13.48±6.77	14.96±5.03	<0.001	<0.001	NS
p**	<0.01	NS	NS			

Continuous variables are expressed as means ± SD.
*Paired t test and **t test for independent samples
Early P - early postoperative, Late P - late postoperative, NS - not significant, Pre - preoperative

Table 5. Comparison of MRI ratios between the two surgical techniques

Diaphragmatic level	Single-patch (n=14)	Three-patch (n=11)	p*
r1	1.804±0.038	1.802±0.040	NS
r2	1.271±0.026	1.274±0.014	NS
r3	1.258±0.022	1.258±0.018	NS
r4	0.919±0.056	0.904±0.052	NS

Continuous variables are expressed as means ± SD
Student t test for independent samples
MRI – magnetic resonance imaging, NS - not significant, r - ratio

Table 6. Pre and post-operative NYHA class of discrete SAS patients

NYHA class	I	II	III	IV
Preoperative, n(%)	3 (12)	15 (60)	6 (24)	1 (4)
Postoperative, n(%)	19 (76)	6 (24)		

NYHA - New York Heart Association, SAS – supravalvular aortic stenosis

original simple single-patch enlargement of the sinotubular junction and the non coronary sinus of Valsalva remains the standard treatment option in many institutions (9, 12-18).

Our experience indicates the use of a single-patch repair in the correction of SAS leads to excellent long-term results, although this technique does not completely restore the geometry of the aortic sinuses. Brown et al. (19) pointed the use of a standard patch, which should be large enough to restore the aorta to a normal diameter. We believe that a single-patch repair provides a better anatomical repair than the three-patch technique in repairing SAS. And according to our results, there may not be a need for more extensive and complex operations like 3 patch technique. Hazekamp et al. (20) reported their experience with three-patch technique compared with 1- and 2- sinus reconstructions. Although they preferred Brom's technique, they did not also find a significant difference in long-term gradients or need for reoperation.

So-called supravalvular aortic stenosis is functionally and structurally a disease of entire aortic root. The underlying elastin arteriopathy is a generalized disease of both pulmonary artery and aorta. Furthermore, aortic valve function is affected and coronary blood flow is impaired due to the premature arteriosclerosis as a part of the aortic root malformation. Accepting the difficulties and pitfalls of a retrospective analysis, this series demonstrates that repair of discrete SAS can be performed with a near zero operative mortality, a low incidence of postoperative aortic incompetence and satisfactory relief of gradient. Unless co-existent cardiovascular abnormalities are present, reoperation rate is very low. Several other series report similar findings (12-14). This demonstrates why end points such as operative mortality, postoperative aortic incompetence, relief of gradient and reoperation rate are not suitable comparative purposes.

The age at operation has gradually decreased throughout the study period. As congenital cardiac surgeons have increased experience with operating on the great vessels and valvular function can be preserved. There is also evidence that early intervention may decrease the incidence of coronary artery disease and ventricular dysfunction thus conferring survival benefit (14).

Single-patch technique has been shown to be quite effective in reduction of the left ventriculo-aortic gradient. An advantage of reconstructing the whole aortic root has not been shown in reducing the aortic gradient. It is very well accepted clinically that echocardiography in the non-sedated patients show higher gradients across stenotic valves and vessels than are measured during cardiac catheterization, which is usually performed under sedation or general anesthetics in pediatric group. Interestingly, however, in our series, the postoperative gradient was extremely low in both groups of patients comparing with the other published data (15, 21).

In today's technological era, non-invasive cardiac imaging alternatives, such as MRI, can offer excellent image quality in various pathophysiological situations of the heart. MRI scanning is very attractive because of the lack of radiation. High-resolution imaging can give more robust information about cardiovascular anomalies comparing to conventional imaging techniques. To our knowledge, this study is the first in the published literature, which used MRI scanning in long-term follow-up of SAS patients. MRI findings and study results do not support the hypothesis that symmetric reconstruction of the left ventricular outflow tract will provide the best immediate relief of obstruction and long-term preservation of the aortic root. Our study demonstrates that standard single-patch aortoplasty provides good postoperative results with asymptomatic patients in the long-term period. It is probable that early relief of the obstruction restores flow in such a way that remodeling and growth of the aortic root can occur normally.

Limitations of the study

There are two limitations that need to be acknowledged and addressed regarding our study. The first limitation concerns the surgical preference for supravalvular aortic stenosis relief. Even though the findings of our study showed no difference between the two surgical techniques in aortic root geometry and the growth, different surgical reconstruction methods are available and in favor by different institutions. This study compares only two techniques which were used in participating institutions. Further study is needed to compare other options for the surgical treatment.

The second limitation has to do with the extent, which the findings can be generalized beyond the cases studied. The number of cases in our study is too limited for broad generalizations. However, the late-term gradient changes represent rather different aspects of the surgical repair. Therefore, patients will benefit from the two techniques equally but further studies with larger sample sizes are definitely needed.

Conclusion

In conclusion, we cannot demonstrate any benefit in reconstructing the whole aortic root for discrete SAS. A single

patch technique is easy, safe and appears durable. Three-patch technique, however, must always be remembered as an alternative technique thus; we believe that each aortic root should be treated on its own merit.

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