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Original Article / Özgün Makale

Comparison of different surgical techniques for repair of aortic coarctation in childhood

Çocukluk çağı aort koarktasyonu tamirinde farklı cerrahi tekniklerin karşılaştırılması

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ABSTRACT

Background: This retrospective study aims to present the mid-term results and complications of different surgical techniques performed in our clinic due to aortic coarctation.

Methods: One hundred and six patients (61 males, 45 females; mean age 2.0±3.7 years; range 2 days to 16 years) who underwent repair of aortic coarctation between January 2005 and December 2013 were included in the study. Patients were divided into four according to age groups: newborns (n=36, 34%), infants (n=42, 39.6%), 1-5 years (n=13, 12.2%), and >5 years (n=15, 14.2%). Patients were performed 122 surgical procedures. Patients were operated following moderate hypothermia (34-35 °C) through left posterolateral thoracotomy with incision performed at fourth intercostal space.

Results: Mean follow-up duration was 34±11 months (range 1 to 64 months). Of the patients, end-to-end anastomosis was performed in 45.9%, extended end-to-end anastomosis in 26.2%, patch plasty in 22.1%, and graft interposition in 5.7%. During follow-up, recoarctation developed in 16 (15.1%) patients. All patients who developed recoarctation were younger than six months. Recoarctation was statistically significant in newborn age group (27.8% vs. 8.5%, p<0.05). Recoarctations which developed in newborn period were performed surgery and those that developed in later period were first performed balloon angioplasty and afterwards surgery. Twenty patients (18.8%) died in early postoperative period.

Conclusion: A comparison of the surgical techniques according to age groups revealed that extended end-to-end anastomosis was preferred at higher rates in newborn period, while end-to-end anastomosis was preferred at higher rates in other age groups. Recoarctation and mortality rates were higher in patients who were performed surgery during newborn period.

Keywords: Aortic coarctation; cardiac surgery; hypertension; mortality.

ÖZ

Amaç: Bu retrospektif çalışmada, kliniğimizde aort koarktasyonu nedeniyle uygulanan farklı cerrahi tekniklerin orta dönem sonuçları ve komplikasyonları sunuldu.

Çalışma planı: Ocak 2005 - Aralık 2013 tarihleri arasında aort koarktasyonu onarımı yapılan 106 hasta (61 erkek, 45 kız; ort. yaş 2.0±3.7 yıl; dağılım 2 gün-16 yıl) çalışmaya dahil edildi. Hastalar yaş gruplarına göre dörde ayrıldı: yenidoğan (n=36, %34), infant (n=42, %39.6), 1-5 yıl (n=13, %12.2) ve >5 yıl (n=15, %14.2). Hastalara 122 cerrahi işlem uygulandı. Hastalar orta derece vücut soğutulmasını takiben (34-35 °C) sol posterolateral torakotomi yoluyla dördüncü kaburgalar arası aralıktan yapılan kesi ile ameliyat edildi.

Bulgular: Ortalama takip süresi 34±11 ay (dağılım 1-64 ay) idi. Hastaların %45.9'una uç uca anastomoz, %26.2'sine uzatılmış uç uca anastomoz, %22.1'ine yama ile plasti ve %5.7'sine greft interpozisyonu uygulandı. Takipte 16 hastada (%15.1) rekoarktasyon gelişti. Rekoarktasyon gelişen bütün hastalar altı aydan küçüktü. Rekoarktasyon yenidoğan yaş grubunda istatistiksel olarak anlamlı idi (%27.8'e karşın %7.5, p<0.05). Yenidoğan döneminde gelişen rekoarktasyonlara cerrahi, sonraki dönemde gelişenlere önce balon anjiyoplasti, sonra cerrahi uygulandı. Yirmi hasta (%18.8) erken ameliyat sonrası dönemde kaybedildi.

Sonuç: Yaş gruplarına göre cerrahi teknikler kıyaslandığında; uzatılmış uç uca anastomozun yenidoğan döneminde; uç uca anastomozun ise diğer yaş gruplarında daha yüksek oranda tercih edildiği görüldü. Rekoarktasyon ve mortalite oranları yenidoğan döneminde cerrahi uygulanan hastalarda daha yüksekti.

Anahtar sözcükler: Aort koarktasyonu; kalp cerrahisi; hipertansiyon; mortalite.

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Aortic coarctation was first described by Morgagni in 1760 as a stenotic segment of the descending aorta.[1] Aortic coarctation is congenital narrowing of aorta which frequently occurs in the juxtaductal position and may be associated with long segment stenosis, aortic arch hypoplasia or stenosis of the abdominal aorta.[2] It is one of the most common causes of secondary hypertension which can be treated surgically. If untreated, it appears to be associated with premature morbidity and mortality. Aortic coarctation is the fifth most common congenital cardiovascular anomaly which needs treatment during infancy and childhood. The occurrence rate is 0.4 per 1000 live births and represents 5 to 8% of all cases of congenital heart disease. It is three times more common in males.[3]

Coarctation often has coexisting congenital heart defects. It was previously reported that approximately 40% of coarctation patients have bicuspid aortic valves.[4] Other congenital heart anomalies seen with coarctation are ventricular septal defects, subaortic stenosis, mitral valve abnormalities, patent ductus arteriosus, sinus venosus type atrial septal defects, left persistent superior vena cava, right aortic arch, transposition of great arteries, supravalvular pulmonary stenosis, truncus arteriosus, Taussig Bing anomaly, double inlet left ventricle with tricuspid stenosis, and hypoplastic left heart syndrome.[5]

The first successful repair of coarctation was performed in Stockholm, Sweden in 1944 by Clarence Crafoord and this technique is still ideal for infant coarctations.[6] The surgical management of aortic coarctation has evolved considerably during the past 60 years since the first description of this operation with the aim of minimizing recoarctation. Despite all these efforts, systemic hypertension, cerebrovascular events and aneurysm formation are still challenging morbidities. Mortality rate for repair of isolated aortic coarctation without other complex heart anomalies was reported to be less than 1% in recent studies.[7] The best surgical technique is still controversial. Thus, in this study, we aimed to present the mid-term results and complications of different surgical techniques performed in our clinic due to aortic coarctation.

PATIENTS AND METHODS

This retrospective study included 106 patients (61 males, 45 females; mean age 2.0±3.7 years; range 2 days to 16 years) who underwent repair of aortic coarctation between January 2005 and December 2013 at Dr. Sami Ulus Maternity and Children's Health and Diseases Training and Research Hospital. Patients were divided into four according to age groups: newborns (n=36, 34%), infants (n=42, 39.6%), 1-5 years (n=13, 12.2%), and >5 years (n=15, 14.2%). These 106 patients underwent 122 surgical procedures. Patients with interrupted aortic arch and who underwent open heart surgery with repair of aortic coarctation were excluded. The study protocol was approved by Dr. Sami Ulus Maternity and Children's Health and Diseases Training and Research Hospital Ethics Committee. The study was conducted in accordance with the principles of the Declaration of Helsinki.

Patient data were collected from electronic medical records, Cardiology Department's charts, and the echocardiogram database. Demographic distribution of the patients is shown in Table 1.

All operations were performed under moderate hypothermia (34 to 35 °C) by topical cooling with water-flow cooling blanket and through left

Table 1. Preoperative demographic data of patients

	n	%	Mean±SD	Min.-Max.
Age groups				
Newborn	36	34		
Infant	42	39.6		
1-5 years	13	12.2		
>5 years	15	14.2		
Gender				
Female	45	42.4		
Male	61	57.6		
Body weight (kg)	9.1			1.6-70
Mean gradient (mmHg)			40±16.9	
Hypertension	64	60.4		
Heart failure	53	50		

SD: Standard deviation; Min.: Minimum; Max.: Maximum.

posterolateral thoracotomy at fourth intercostal space. Four different types of surgical techniques were used after dissection of aortic arch vessels, descending aorta, and ductus arteriosus. End-to-end anastomosis was preferred for discrete coarctation. When aortic arch was diminutive, extended end-to-end anastomosis was performed. In the setting of long segment stenosis, patch plasty technique was achieved, while tubular graft interposition was used for extreme stenosis.

We conducted follow-up examinations in the postoperative period in first, third, sixth, and 12th months, and then once a year with routine physical examination and echocardiography. Patients who had 20 mmHg or above gradients between upper and lower extremities and/or "diastolic tail" in color Doppler were evaluated by computed tomographic angiography or catheter-based aortography. In invasive imaging techniques, more than 50% of luminal narrowing was accepted as recoarctation.

Statistical analysis

Statistical analyses were performed with SPSS version 16.0 software (SPSS Inc., Chicago, IL, USA). Continuous variables were expressed as mean and standard deviation, while categorical variables were defined as numbers and percentages in brackets. The categorical variables were compared with chi-square and Fisher's exact test. All *p* values less than 0.05 were considered statistically significant.

RESULTS

Mean follow-up duration was 34±11 months (range 1 to 64 months). Infants and newborns comprised most of the patients (39.6% and 34%, respectively). Mean body weight was 9.1 kg (range 1.6 to 70 kg). Most patients had additional pathologies that increased comorbidities (Table 2). Of the patients, 45.9% (n=56) underwent end-to-end anastomosis, 26.2% (n=32) underwent extended end-to-end anastomosis, 22.1% (n=27) underwent patch plasty, and 5.7% (n=7) underwent graft interposition. Mean aortic clamp times in end-to-end anastomosis, extended end-to-end anastomosis, patch plasty, and graft interposition were 16.7±5.8 minutes, 23.4±7.7 minutes, 18.3±5.9 minutes, and 26.3±7.2 minutes, respectively. Upon hospital discharge, detected postoperative gradient by transthoracic echocardiography was 9.5±6.2 mmHg (range 7 to 20 mmHg). Fourteen patients (eight >5 years old, four 1-5 years old, and two <1 year old) with persistent hypertension used antihypertensive drugs. However, eight of them discontinued medications in follow-up period and the remaining six patients were older than five years of age.

Twenty patients (18.8%) who had complex cardiac anomalies in addition to aortic coarctation died in early postoperative period. Additional pulmonary artery banding was performed in 14 of them. The other six patients did not need any additional procedure to coarctation repair. Of these 20 patients, 15 died because of pneumonia and sepsis, and the other five died because of heart failure. There was no mortality in isolated coarctation patients.

As shown in Table 3, patients experienced early postoperative complications such as pneumonia (n=16, 15.1%), sepsis (n=6, 5.7%), pneumothorax (n=3, 2.8%), paraplegia (n=1, 0.9%) and hoarseness (n=4, 3.8%). Flask paraplegia occurred in early postoperative period in a five-year-old boy with long segment tubular aortic coarctation who underwent patch plasty (cross clamp time was 21 minutes). Physiotherapy has been performed since early postoperative period and he is now walking with assistance.

There was no late death and 16 patients were lost to follow-up. Thus, 70 patients were followed-up for a minimum period of three years (mean 6.1 years). During follow-up, we observed recoarctation in

Table 2. Types of concomitant pathologies

Concomitant pathologies	n	%
Patent ductus arteriosus	55	51.9
Ventricular septal defect	48	45.3
Bicuspid aortic valve (no stenosis)	44	41.5
Atrial septal defect/patent foramen ovale	46	43.4
Pulmonary hypertension	32	30.2
Aortic arch hypoplasia	32	30.2
Prematurity	16	15.1
Double outlet right ventricle	13	12.2
Pulmonary stenosis	12	11.3
Atrioventricular septal defect	11	10.4
Aortic stenosis	8	7.5
Mitral valve disease	5	4.7
Turner syndrome	2	1.9
Vascular ring	1	0.9
Williams syndrome	1	0.9
Duodenal atresia	1	0.9

Table 3. Early postoperative complications

Complication	n	%
Pneumonia	16	15.1
Sepsis	6	5.7
Pneumothorax	3	2.8
Paraplegia	1	0.9
Hoarseness	4	3.8

Table 4. Recoarctation and mortality rates according to different surgical techniques

	End-to-end anastomosis (n=56)		Extended end-to-end anastomosis (n=32)		Patch plasty (n=27)		Graft interposition (n=7)		
	n	%	n	%	n	%	n	%	*p*
Recoarctation	7	12.5	4	12.5	4	14.8	1	14.3	0.93
Mortality	10	17.8	9	28.1	-	-	1	14.3	0.007

16 patients. The incidence of recoarctation was lower in extended end-to-end anastomosis without significant statistical difference compared to other techniques (p>0.5) (Table 4).

No recoarctation was seen in patients operated after six months of age. Recoarctation rate was statistically higher in newborn age group (n=10, 27.8% vs. n=6, 8.5%, p<0.05). Ten of the recoarctations were performed surgery during newborn period, and the other six during infancy (all younger than six months). Newborn recoarctations underwent surgery directly due to the risk of aortic wall complications and limited vascular access, while others underwent balloon angioplasty before surgery. Patients who underwent balloon angioplasty prior to surgery had congestive heart failure and/or multi-organ failure. They all underwent surgery after hemodynamic stabilization. Recoarctation did not develop after surgery in patients who underwent balloon angioplasty during infancy (49%).

Table 4 shows mortality rates according to surgical procedure types. No mortality was seen in patch plasty group while patients who underwent extended end-to-end anastomosis had the highest mortality rate (n=9, 28.1%). Mortality rates were 17.8% (n=10) in end-to-end anastomosis and 14.3% (n=1) in graft interposition groups.

Of the 106 patients, 21 (19.8%) had isolated aortic coarctation and no mortality was seen in this group. Pulmonary banding was performed in 32 (30.2%) severe pulmonary hypertensive (mean pulmonary artery pressure ≥35 mmHg) patients of 85 patients (80.2%) who had additional cardiac anomalies. Mortality rate of this group was 23.5%.

Mortality and recoarctation rates were compared according to age groups in Table 5. Although there was no significant difference between age groups in terms of recoarctation (p=0.55), mortality rate was significantly highest in newborns and decreased as patients grew older (p=0.001).

DISCUSSION

Despite excellent outcomes of advanced invasive methods, surgical repair is still an important treatment modality for aortic coarctation during childhood.[8] Surgical techniques include simple end-to-end anastomosis, extended end-to-end anastomosis, patch plasty, and graft interposition. Albeit none of the techniques used for aortic coarctation repair today was proved to be superior to the other,[9] our study showed significant statistical difference between surgical techniques regarding mortality. Patients who underwent extended end-to-end anastomosis had the highest mortality rate when compared to other techniques (Table 4). Since extended end-to-end anastomosis was mostly performed in newborns, high mortality rates may be related to low body weight and neonatal problems (Table 6).

Our group of patients demonstrated that 49% of the infants who underwent surgical repair of coarctation had balloon angioplasty prior to surgery. As a commonly used method recently, balloon angioplasty was related to high rate of recoarctation at early and late follow-up, but still considered useful for patients with heart failure and for stabilization of the preoperative status as a method of palliation.[10,11]

A comparison of mortality rates according to age groups revealed that mortality was highest in newborns and decreased as patients grew older (Table 5). Other

Table 5. Recoarctation and mortality rates according to age groups

	Newborn (n=42)		Infant (n=47)		1-5 years (n=15)		>5 years (n=18)		
	n	%	n	%	n	%	n	%	*p*
Recoarctation	6	14.3	8	17	1	6.7	1	5.6	0.55
Mortality	14	33.3	6	12.8	-	-	-	-	0.001

Table 6. Various surgical techniques for aortic coarctation compared according to age groups

	End-to-end anastomosis (n=56, 45.9%)		Extended end-to-end anastomosis (n=32, 26.2%)		Patch plasty (n=27, 22.1%)		Graft interposition (n=7, 5.7%)		
	n	%	n	%	n	%	n	%	*p*
Newborn	16	38.1	24	57.1	1	2.4	1	2.4	<0.001
Infant	19	40.4	8	17	18	38.3	2	4.3	0.009
1-5 years	11	73.3	-	-	4	26.7	-	-	0.053
>5 years	10	55.6	-	-	4	22.2	4	22.2	0.002

related factors regarding mortality were type of surgical technique and concomitant cardiac pathologies. In a previous multicenter retrospective review by St Louis et al.,[12] the greatest percentage of deaths occurred in patients who underwent surgery during the first week of life. They defined significant association of age at operation, type of surgical technique, and concomitant major cardiac anomalies with mortality similar to our results.

Although recoarctation rates were not statistically significant between age groups, it was higher in newborns. Similarly, recoarctation was shown to be higher in patients who underwent surgical repair during newborn period or early infancy.[13,14] Additionally, there was no significant difference between surgical techniques in terms of recoarctation.

The incidence of hypoplasia of aortic arch in our study was 30.2%. We defined hypoplasia as the segment being less than 50% of the diameter of the descending aorta and the aortic arch diameter less than a Z score of -2. Previous studies reported high coexistence of transverse arch hypoplasia with aortic coarctation.[15]

In our study, end-to-end anastomosis and extended end-to-end anastomosis techniques were performed in patients with discrete aortic coarctations. Coarctated segment was removed in these groups totally. Although it was not statistically significant, we can explain the low incidence of recoarctation in these techniques with complete removal of coarctated tissue (p=0.93).

Postoperative hypertension was shown to be a still-continuing and common problem after surgical repair.[16,17] Particularly, advanced age at surgery seems to be a risk factor for postoperative hypertension. Brown et al.[16] showed that risk of persistent postoperative hypertension was higher in patients in which coarctation was repaired after nine years of age. All of our patients with postoperative hypertension were older than four years of age at surgery. In our study, we observed postoperative hypertension in 14 (11.5%) patients. Eight (57.1%) of these were found to be normotensive during follow-up, and their antihypertensive medications were discontinued at the end of six months.

Paraplegia is a rare but serious complication of aortic coarctation repair. Spinal cord protection methods such as topical cooling is a useful and safe technique.[18,19] We cooled down our patients to 34-35 °C and did not observe any complications such as hypotension or bradycardia.

In this retrospective study, we observed that the mortality rates of coarctation repair in patients accompanied with intracardiac anomalies are higher than simple coarctation repair (p<0.01). These results are similar to recent publications.[20]

The limitation of this study is that it is a retrospective study including a small group of patients from a single centre. It may be possible to design comprehensive studies with larger sample sizes through the creation of a common national database.

In conclusion, despite the successful results of different types of surgical techniques used for pediatric aortic coarctation, particularly newborns and infants with additional cardiac pathologies have higher risk for mortality independent from the applied surgical technique.

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