

# The Factors Affecting Recoarctation Development After Surgical Correction in Patients with Aortic Coarctation

Aort Koarktasyonu Tanısıyla Cerrahi Düzeltme Uygulanan Hastalarda Rekoarktasyon Gelişimi Üzerine Etki Eden Faktörler

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## ABSTRACT

Aim: In this study, the factors affecting the development of recoarctation in patients who had been operated for aortic coarctation (AC) were investigated. It was aimed to contribute to the reduction of recoarctation rates in the light of the results obtained.

**Materials and Methods:** In this retrospective study, 217 patients out of 358 patients, who underwent surgical correction for the diagnosis of AC at Hacettepe University Faculty of Medicine, Department of Cardiovascular Surgery between 1977 and 2010, who were followed up regularly, and whose data could be reached, were included. Age, body weight and results of surgical correction techniques used during surgical correction were compared in cases having recoarctation. Data obtained were evaluated statistically. Descriptive statistics for categorical variables were stated as numbers (n) and percentages (%). The results were presented in mean±standard deviation and/or percentage (frequency). The alpha significance value was accepted as <0.05.

**Results:** One hundred forty seven (67.7%) of the patients were male and 70 (32.3%) were female. Ninety (41.4%) patients were operated for simple coarctation while 127 (58.6%) for coarctation and accompanying complex intra cardiac anomalies. Recoarctation was detected in 36 (16.5%) of 217 patients. Of 36 patients with recoarctation, 21 (58.3%) were treated with balloon angioplasty, 10 (27.7%) surgically, and 5 (13.8%) with stent implantation.

**Conclusion:** Development of recoarctation was found to be statistically significant in the patient group whose body weight was <3 kg and age was <1 year at the time of the first coarctation repair. No statistical significance was found among the surgical techniques that were used in the first coarctation repair. It is necessary to be more careful in terms of recoarctation during follow-up of the patients whose body weight was under 3 kg and whose age was under 1 year during the first coarctation repair.

Keywords: Aortic coarctation, coarctation repair, recoarctation, surgical technique

# ÖΖ

Amaç: Çalışmamızda, aort koarktasyonu (AK) tanısıyla opere edilmiş hastalarda rekoarktasyon gelişimi üzerine etki eden faktörler araştırılmış ve elde edilen sonuçlar ışığında, rekoarktasyon oranlarının azaltılmasına katkıda bulunulması hedeflenmiştir.

**Gereç ve Yöntem:** Bu retrospektif çalşmaya, Hacettepe Üniversitesi Tıp Fakültesi Kalp ve Damar Cerrahisi Kliniği'nde 1977-2010 arasında AK tanısıyla cerrahi düzeltme yapılan 358 hastadan, verilerine tam olarak ulaşılabilen ve düzenli olarak takiplerine gelmiş olan 217 hasta dahil edildi. Rekoarktasyon gelişen olgularda cerrahi düzeltme esnasındaki yaş, vücut ağırlığı ve kullanılan cerrahi tekniklerin sonuçları karşılaştırıldı. Veriler istatistiksel olarak değerlendirildi. Kategorik değişkenler için tanımlayıcı istatistikler sayı (n) ve yüzde (%) olarak belirtildi. Sonuçlar ortalama±standart sapma ve/veya yüzde (frekans) cinsinden sunuldu. Alfa anlamlılık değeri <0,05 olarak kabul edildi.

**Bulgular:** Hastaların 147'si (%67,7) erkek, 70'i (%32,3) kadındı. Doksan (%41,4) hasta basit koarktasyon, 127 (%58,6) hasta ise koarktasyon ve eşlik eden kompleks intrakardiyak anomaliler tanıları ile opere edildi. Otuz altı hastada (%16,5) rekoarktasyon tespit edildi. Rekoarktasyon tespit edilen 36 hastadan 21'i (%58,3) balon anjiyoplasti, 10'u (%27,7) cerrahi, 5'i stent implantasyonu (%13,8) ile tedavi edildi.

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**Sonuç:** Çalışmamızda vücut ağırlığı <3 kg ve yaşı <1 yaş olan hasta grubunda rekoarktasyon gelişimi istatistiksel olarak anlamlı bulunmuştur. Kullanılan cerrahi teknikler arasındaysa istatistiksel bir anlam tespit edilmemiştir. AK nedeni ile opere edilecek hasta gruplarında vücut ağırlığı <3 kg ve yaşı <1 yaş olan hasta gruplarının post operatif takiplerinde rekoarktasyon gelişimi açısından daha dikkatli olunması gerekmektedir.

Anahtar Kelimeler: Aort koarktasyonu, koarktasyon tamiri, rekoarktasyon, cerrahi teknik

# INTRODUCTION

Aortic coarctation (AC) is a discrete narrowing at the entry of the ductus arteriosus of the proximal thoracic aorta, that is, in the aortic isthmus region, with a frequency of 98%. AC constitutes 5-8% of all congenital heart diseases and is more common in males<sup>1</sup>. It is the second most common cardiovascular anomaly requiring surgical intervention in the newborn period<sup>2</sup>. AC was first described by Morgagni in an autopsy in 1760. The first successful AC repair was performed by Craaford and Nylin<sup>3</sup> in 1945.

Recoarctation is defined as the presence of a resting gradient greater than 20 mmHg in the area of AC repair. Rates ranging from 4% to 20% have been reported in various series<sup>4</sup>. Recoarctation emerges as a complication of surgical correction and is directly related to long-term morbidity.

Age, body weight and the surgical technique used at the time of the first operation are among the main factors affecting the development of recoarctation<sup>5.6</sup>. In this study, the factors affecting the development of recoarctation in patients who had been operated for the diagnosis of AC were investigated and it was aimed to contribute to the reduction of recoarctation rates in the light of the results obtained.

# **MATERIALS AND METHODS**

### Selection and Description of Cases

Of 358 patients who had undergone surgical correction with the diagnosis of AC at Hacettepe University Faculty of Medicine, Department of Cardiovascular Surgery between 1977 and 2010, 217 patients whose data could be accessed and who had regular follow-up visits were included in this retrospective study.

Patients with a peak gradient >20 mmHg in transthoracic echocardiographic (TTE) and angiographic measurements were considered as the recoarctation group. Diagnostic cardiac catheterization was performed in all patients with recoarctation as TTE in the controls. After the cases with recoarctation were identified, these cases were categorized in terms of age, body weight and surgical techniques used at the time of the first surgical correction.

The patients were divided into 3 groups considering age distribution, as 0-1 years, 1-5 years and >5 years. In terms of body weight distribution, they were divided into 3 groups, as

0-3 kilograms (kg), 3-10 kg and >10 kg. Six different surgical techniques were used in the coarctation repair surgeries of the patients. Before starting the study, permission was obtained from the Ethics Committee of Hacettepe University Faculty of Medicine (decision number: 02/10, date: 07.01.2010).

## Surgical Technique

While 177 of the 217 patients included in the study were operated with left posterolateral thoracotomy, sternotomy was used in 40 patients. The patients operated with sternotomy were included in the group of patients with a hemodynamically and clinically significant and correctable intracardiac pathology accompanying coarctation, and all were operated on with the support of cardiopulmonary bypass (CPB).

Of these patients, simultaneous closure of ventricular septal defect was performed in 12 patients, intervention in congenital mitral stenosis in 9 patients, aortic arch reconstruction in 7 patients, aortic commissurotomy in 6 patients, partial pulmonary venous return anomaly correction in 3 patients, arterial switch in 2 patients and central shunt operation in 1 patient. All 7 patients who underwent aortic arch reconstruction were operated under total circulatory arrest (TSA).

Standard aortic and bicaval venous cannulation was performed in all patients operated with sternotomy. Following the transition to CPB, AC was first intervened. Following the division of the patent ductus arteriosus, all aortic tissue was released from the aortic arch to the descending thoracic aorta. Meanwhile, all intercostal arterial branches and the phrenic nerve were preserved. After the distal and proximal clamps were placed, the coarctation area and ductal tissue remnants were completely resected.

Following extensive mobilization, the distal end of the aortic arch was anastomosed to the proximal end of the descending thoracic aorta end-to-end with 7.0 polypropylene (pp) suture material. TSA was used in 7 patients with aortic arch hypoplasia. The mean TSA times were 24.8 minutes (range 18-33 minutes). In patients who underwent posterolateral thoracotomy, the thoracic cavity was entered through the left fourth intercostal space, and the mediastinal pleura was opened after retraction of the left lung upper lobe. The coarctate segment was explored, preserving all collateral lateral branches. For concomitant patent ductus arteriosus, division was used in 23 patients and double ligation-transfixion methods were used in 18 patients. After the resection of the coarctate segment and the placement of proximal and distal vascular clamps with the end-to-end anastomosis technique, the coarctate segment was resected and end-to-end anastomosis was completed with continuous suture in the posterior aortic wall and with interrupted suture in the anterior aortic wall, using a 7.0 pp suture material.

In the extended end-to-end anastomosis technique, the aortic arch and the distal thoracic aorta were radically dissected and released. In this way, it was ensured that there was no tension in the anastomosis line. After the division of the first intercostal artery was completed, proximal and distal vascular clamps were placed. After complete resection of the coarctate segment, anastomoses were completed with 7.0 pp suture material.

In the subclavian flap aortoplasty method, the left subclavian artery was ligated proximal to its all branches in order to preserve the collateral circulation feeding the left upper extremity. A vascular clamp was placed proximally to control the transverse aortic arch, left common carotid artery and left subclavian artery. The distal vascular clamp was placed as far as possible from the coarctate segment.

The left subclavian artery was opened longitudinally along its anterior wall, and after this incision was extended to 1 cm distal to the coarctate segment, the left subclavian artery was transected. Anastomosis was completed with continuous suture technique using 7.0 pp suture material.

In the modified subclavian flap aortoplasty technique, all branches of the left subclavian artery except the internal thoracic branch were performed ligation and division. After making sure that the left subclavian artery could extended up to 2 cm distal to the coarctate segment, the transverse aortic arch, left subclavian artery and distal aorta were clamped. Beginning proximal to the left subclavian artery, an arteriotomy was performed on its anterior face, and this arteriotomy was extended up to 2 cm distal to the coarctate segment. Anastomosis was completed with continuous suture technique using 7.0 pp suture material.

In the patch aortoplasty technique, after the proximal and distal thoracic aorta was released, proximal and distal control was achieved with the help of vascular clamps. After the coarctate segment was opened longitudinally, the anastomosis was completed with a continuous suture technique, using 7.0 pp suture material and a polytetrafluoroethylene patch material of suitable geometry.

In the graft interposition technique, following the control of the proximal and distal thoracic aorta with vascular clamps, the coarctate segment was resected, and then the anastomoses were completed with the continuous suturing technique, using 7.0 pp suture material, with both proxies of the graft of appropriate diameter end-to-end. After it was observed that there was no residual gradient between the right radial artery and the left femoral artery in all surgical techniques, bleeding controls were completed. Following the placement of the surgical drainage tubes, the layers were closed in accordance with the anatomical planning.

## **Statistical Analysis**

The obtained data were analyzed using the Statistical Package for the Social Sciences version 17.0 (INC., Chicago, IL, USA) software. Descriptive statistics for categorical variables were expressed as numbers (n) and percentage (%). Results are presented as mean±standard deviation, median, and/or percentage (frequency). In the categorical comparisons between the groups, the significance levels were checked with the Pearson correlation test using cross-table statistics. The Mann-Whitney U test was used for pairwise group comparisons and One-Way ANOVA test was used for multi-group comparisons of data that did not have a normal distribution. The stepwise logistic regression analysis was performed to evaluate risk factors. Alpha significance value was accepted as <0.05.

# RESULTS

A total of 217 patients were included in the study. Of the patients, 147 (67.7%) were male and 70 (32.3%) were female. The mean age of the patients included in the study was  $5.1\pm4.4$  years (range: 2 months-16 years), and the mean body weight was  $6.2\pm3.7$  kg. The patients were followed up for a median period of 78.8 months postoperatively. Gradient measurements were made after surgical correction by performing simultaneous right radial and left femoral artery catheterization in all patients.

In any of the 217 patients included in the study, no residual gradient was detected in the measurements performed on the operating table after the correction operation was completed. The mean peak systolic gradient value detected in the recoarctation region of the patients was  $56\pm17$  mmHg. The time between the first coarctation correction and the diagnosis of recoarctation was determined as  $5.1\pm2.1$  months. The demographic and operative characteristics of the patients included in the study are presented in Table 1. Of the patients, 90 (41.4%) were operated with the diagnosis of simple coarctation, and 127 patients with the diagnosis of coarctation and accompanying complex intracardiac anomalies. Intracardiac anomalies accompanying coarctation are presented in Table 2.

When patients who developed recoarctation were grouped according to their body weight, it was detected that 20 of 86 patients operated between 0 and 3 kg (p=0.013), 14 of 108

patients operated between 3 and 10 kg and 2 of 23 patients operated at >10 kg had recoarctation.

When patients who developed recoarctation were grouped according to age distribution, recoarctation was found in 24 (p=0.016) of 102 patients operated at the age of <1 year, in 8 of 83 patients operated between the ages of 1 and 5 years, and in 4 of 32 patients operated at the age of >5 years.

In the evaluation made according to the surgical techniques applied to the patients, the development of recoarctation was detected in 26 of 131 patients who underwent resection and end-to-end anastomosis technique, in 3 of 24 patients who underwent patch aortoplasty, in 2 of 22 patients who underwent extended end-to-end anastomosis technique, in 4

Table 1. Demographic and operative patients	characteristics of the
Gender (male/female)	147/70 (67.7%/32.3%)
Body weight (kg)	6.2 <u>±</u> 3.7
Age (years)	5.1±4.4
Follow-up duration (months)	78.8
Peak systolic gradient in the recoarctation area (mmHg)	56±17
Duration of surgery (minutes)	68.7±10.2
Aortic clamp time (minutes)	17.5 <u>+</u> 2.1
Length of stay in intensive care (days)	4
Length of hospitalization (days)	9
Time between coarctation surgery and diagnosis of recoarctation (months)	5.1 <u>±</u> 2.1

Table 2. Concomitant cardiac anomalies (n=127)	
PDA	41 (32.2%)
Bicuspid aorta	22 (17.3%)
VSD	18 (14.1%)
Aortic stenosis	12 (9.4%)
Mitral stenosis	9 (7%)
Arcus aortic hypoplasia	7 (5.5%)
ASD	6 (4.7%)
PAPVD	3 (2.3%)
AVSD	2 (1.5%)
TGA	2 (1.5%)
PS	1 (0.7%)
c-TGA	1 (0.7%)
DORV	1 (0.7%)
ТА	1 (0.7%)
ALCAPA	1 (0.7%)

PDA: Patent ductus arteriosus, VSD: Ventricular septal defect, ASD: Atrial septal defect, PAPVD: Partial anomalous pulmonary venous drainage, AVSD: Atrio-ventricular septal defect, TGA: Transposition of the great arteries, c-TGA: Corrected-transposition of the great arteries, PS: Pulmonary stenosis, DORV: Doubleoutflow right ventricular TA: Tricuspid atresia, ALCAPA: Abnormal left coronary artery originating from the pulmonary artery

of 17 patients who underwent graft interposition technique, and in1 of 6 patients who underwent modified subclavian flap aortoplasty technique. Recoarctation did not develop in any of 17 patients who underwent subclavian flap aortoplasty technique.

According to the logistic regression analysis, there was no statistically significant difference between surgical techniques and the development of recoarctation. Early postoperative complications included bleeding revision in 8 patients, pneumonia in 7 patients, tracheostomy due to prolonged mechanical ventilation in 2 patients, infection in the thoracotomy incision in 2 patients, chylothorax in 1 patient, hoarseness due to n. laryngeus recurrens injury in 1 patient, left diaphragmatic paralysis due to n. phrenicus damage in 1 patient.

Of 36 patients who developed recoarctation, 21 (58.3%) were treated with balloon angioplasty, 10 (27.7%) with surgery, and 5 (13.8%) with stent implantation. Of the patients who underwent surgical intervention, graft interposition was performed between the ascending aorta and the descending aorta in 4, graft interposition between the left subclavian artery and the descending aorta in 3, and patch aortoplasty in 3 patients.

Late mortality was detected in 8 (3.6%) of 217 patients in the study group. It was found that one patient died due to early postoperative heart failure after correction surgery for the diagnosis of abnormal left coronary artery originating from the pulmonary artery that had emerged five years after the coarctation repair; one patient died due to infective endocarditis of the replaced mitral valve developing 23 years after the coarctation repair; one patient died due to hypertensive intracranial bleeding; and 5 patients died due to non-cardiac causes.

# DISCUSSION

If untreated, 75% of patients diagnosed with AC have an average life expectancy of 35 years<sup>7</sup>. In patients who are clinically stable in newborn or infancy, surgical correction is the primary treatment option as soon as the diagnosis is established<sup>8</sup>. Thus, life expectancy is prolonged and additional morbidities that may develop in the long term are prevented. It has been reported that recoarctation rates are low in patients diagnosed in adulthood and in those undergoing surgical correction<sup>9</sup>.

Since 1945, when the first coarctation correction was made, many studies have been carried out in order to prevent complications that may occur after surgical correction, especially recoarctation. Age, body weight and the techniques used in surgical correction at the time of the first operation were evaluated as risk factors for the development of recoarctation.

Age and body weight are the most important parameters related to the somatic growth potential of the patient and therefore the native aorta. Recoarctation rates were found to be high in patients who were operated in the neonatal period<sup>10,11</sup>. In our study, it was found that recoarctation developed in 24 (p=0.016) of 102 patients who were operated under the age of one year.

Another factor blamed for the development of recoarctation is the patient's body weight at the time of surgery. It was found that the rate of development of recoarctation was significantly higher, especially in patients who were operated on with a body weight less than 1.5 kg<sup>12</sup>. Results of studies evaluating age and body weight simultaneously at the time of the first operation demonstrated the rates of recoarctation to be significantly higher<sup>13,14</sup>. In our study, it was found that 20 (p=0.013) of 86 patients who were operated under 3 kg developed recoarctation.

Many surgical techniques have been developed since 1945, when AC repair was first performed. Among these techniques, resection and end-to-end anastomosis, subclavian flap aortoplasty, patch aortoplasty, extended end-to-end anastomosis, graft interposition, and modified subclavian aortoplasty are used most commonly. Regardless of the surgical technique used, complete resection of the coarctate segment is the most important factor.

As the amount of residual tissue remaining in the aortic lumen increases, the probability of developing recoarctation also increases. In the first 40 years of the history of AC surgery, resection and end-to-end anastomosis technique was seen to be the most preferred technique especially in newborn and infant period<sup>15,16</sup>. With the increase in surgical experience, especially the high recoarctation rates of the resection and end-to-end anastomosis technique and use of the extended end-to-end anastomosis technique and use of this technique safely in newborns, infants and childhood in the following years<sup>17</sup>.

In this technique, the most important advantage is the complete resection of both coarctate tissue and ductal tissue remnants, and the ability to perform an extended and untensioned proximal anastomosis that preserves the growth potential of the native aorta<sup>18-20</sup>. In the subclavian flap aortoplasty technique, the use of autologous tissue with growth potential and surgical repair without creating a circular growth defect have caused this technique to be preferred more especially in infancy<sup>21,22</sup>.

Due to the possibility of impaired left upper extremity circulation in the subclavian flap aortoplasty technique, the

modified subclavian flap aortoplasty technique was developed and used with low recoarctation rates, especially in the newborn and infant period<sup>23</sup>. The patch aortoplasty technique has been used with low recoarctation rates, in which proximal and distal clamp times are much shorter, and thus postoperative mortality and morbidity are very low<sup>24</sup>.

In patients with AC diagnosed in adulthood, the possibility of recoarctation with the graft interposition technique is at the same rate as other techniques<sup>25</sup>. The long-term prognosis and possible morbidity of these patients following surgical repair of AC underline the need for effective long-term follow-up<sup>26</sup>.

When we compared our study with similar studies in the literature, similar recoarctation rates were found among surgical techniques. In our study, it was found that none of 17 patients who underwent subclavian flap aortoplasty technique developed recoarctation. However, the highest rate of recoarctation was found in the resection and end-to-end anastomosis technique applied in 26 of 131 patients. In our study, no statistical significance was found among surgical techniques in terms of the development of recoarctation.

## **Study Limitations**

The limitation of our study is that it was a retrospective and single-center study.

# CONCLUSION

In our study, the development of recoarctation was found to be statistically significant in patients with a body weight <3 kg and the age of <1 year and in those who were operated for the diagnosis of aortic coarctation. No statistical significance was found among the surgical techniques used in the correction surgery. In the patient groups to be operated for aortic coarctation, patients with body weight of <3 kg and age of <1 year should be more carefully monitored in terms of the development of recoarctation in post-operative follow-ups.

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### Ethics

**Ethics Committee Approval:** This retrospective study was approved by Ethics Committee of Hacettepe University Faculty of Medicine (decision number: 02/10, date: 07.01.2010).

Informed Consent: Retrospective study.

Peer-review: Externally peer-reviewed.

#### **Authorship Contributions**

Surgical and Medical Practices: B.Ş., A.Ö., M.Y., R.D., M.D., Concept: B.Ş., Design: B.Ş., A.Ö., Data Collection or Processing: B.Ş., Analysis or Interpretation: B.Ş., Literature Search: B.Ş., Writing: B.Ş.

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