Cardiology in the Young

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Isolated coarctation repair through a left thoracotomy in children

Shiraslan Bakhshaliyev[1], Serhat Bahadır Genç[1], Görkem Çitoğlu[1], Zeynep Gülben Kük Özalp[1], Servet Ergün[2], Hacer Kamali[3], Okan Yildiz[1], Onan İsmihan Selen[1], Alper Guzeltas[3] and Sertac Haydin[1]

[1]Pediatric Cardiac Surgery, Mehmet Akif Ersoy Thoracic and Cardiovascular Research and Education Hospital, Istanbul, Turkey; [2]Pediatric Cardiovascular Surgery, Erzurum Regional Training and Research Hospital, Erzurum, Turkey and [3]Department of Pediatric Cardiology, Mehmet Akif Ersoy Thoracic and Cardiovascular Research and Education Hospital, Istanbul, Turkey

Original Article

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Author for correspondence:
Shiraslan Bakhshaliyev, Pediatric cardiac surgery, Mehmet Akif Ersoy Thoracic and Cardiovascular Research and Education Hospital, Istanbul, Turkey.
E-mail: sh.bakhshaliyev@gmail.com

Abstract

Introduction: Isolated aortic coarctation performed through a left thoracotomy resection and end-to-end anastomosis results in low mortality and morbidity rates. Recoarctation and late hypertension are among the most important complications after such repairs. In this study, we reviewed the results of children who underwent left-side thoracotomy to correct an isolated aortic coarctation. *Method:* A consecutive sample of 90 patients who underwent resection and extended end-to-end anastomosis through a left-side thoracotomy in our centre between 2011 and 2021 was retrospectively analysed. The patients' preoperative characteristics, operative data, and post-operative early and long-term results were examined *Results:* All patients underwent resection and extended end-to-end anastomosis. A pulmonary artery band was applied simultaneously to three (3.3%) patients, and an aberrant right subclavian artery division was applied to one (1.1%) patient. The mean cross-clamp time was 29.13 ± 6.97 minutes. Two (2.2%) patients required reoperation in the early period. Mortality was observed in one (1.1%) patient in the early period. Eight (8.8%) patients developed recoarctation, of whom four (4.4%) underwent reoperation and four (4.4%) underwent balloon angioplasty. Twenty-two (26.8%) patients received follow-up antihypertensive treatment. The mean follow-up period was 41.3 ± 22.8 months. No mortality was observed in the late period. *Conclusion:* Isolated coarctation is successfully treated with left-side thoracotomy resection and an extended end-to-end anastomosis technique with low mortality, morbidity, and low long-term recoarctation rates. Long-term follow-up is required due to the risks of early and late post-operative recoarctation, which requires reintervention.

Aortic coarctation is the congenital narrowing of the aortic isthmus, usually (but not always) occurring between the left subclavian artery and the ductus arteriosus.[1] It has an incidence of 1 in approximately 2500 live births and is twice as common in males.[2] It is often associated with bicuspid aortic valve and ventricular septal defects, with or without left ventricular outflow tract obstruction.[3] Aortic arch hypoplasia is common in patients with neonatal aortic coarctation.[4]

Surgical treatment of aortic coarctation was first reported in 1945, and since then, a variety of techniques and modifications have been developed.[5] Today, the preferred method is resection and extended end-to-end anastomosis, the most important advantages of which are total excision of the coarcted segment with ductal tissue and extended anastomosis.[6,7] However, the resection and extended end-to-end anastomosis treatment is controversial if the patient also has a hypoplastic aortic arch. In such cases, the preferred surgical approach depends on the degree of the associated hypoplastic aortic arch and any other cardiac anomalies present. Recurrent aortic arch obstruction and persistent hypertension continue to be important problems in the long-term post-operative period.[8]

In this study, we reviewed the surgical treatment results of children who underwent a left thoracotomy to correct an isolated aortic coarctation.

Materials and method

This study is a retrospective analysis of a consecutive sample of patients who underwent left-side thoracotomy to correct an isolated coarctation in our centre from 2011 to 2021. Approval for this study was obtained from the Ethics Committee of the Istanbul Mehmet Akif Ersoy Training and Research Hospital.

Patients who underwent aortic coarctation repair through a left thoracotomy were included. Patients who underwent arch reconstruction with median sternotomy to correct a hypoplastic aortic arch had more complex cardiac anomalies, such as transposition of great arteries,

double-outlet right ventricle, or complete atrioventricular canal, and were excluded. The preoperative, demographic, operative, and post-operative data we evaluated were gathered from our clinic's database, including patient anamneses, operation notes, perfusion database, clinical follow-up notes, preoperative and post-operative echocardiography and cardiac catheterisation reports.

Surgical indications

Patients with low birth weight, severe proximal arch hypoplasia, additional intracardiac defects (such as ventricular septal defect, congenital aortic stenosis, and complex intracardiac defects) underwent arch reconstruction with a median sternotomy. Patients with isolated coarctation and no intracardiac defects were treated through a left thoracotomy.

Surgical technique

Cerebral and somatic near-infrared spectroscopy monitoring and femoral and right radial artery cannulation were performed on all patients. To reduce the risk of paraplegia, passive cooling was used to reduce the body temperature to 34°C. The thorax was entered through the 3rd or 4th intercostal space with a muscle-sparing left-side posterolateral thoracotomy. After the left lung was retracted from the anterior, the mediastinal pleura was opened and hung. Considering the vagus and phrenic nerves, the aortic arch, supra-aortic branches, and the patent ductus arteriosus, the descending aorta, and the first two rows of collaterals were released. The patent ductus arteriosus was divided by double ligation. After administration of 100 u/kg heparin, proximal clamping was performed with the side clamp after the left carotid, and distal clamping was performed with a 90° cross-clamp. The coarcted segment was excised extensively. The small curvature of the arch was opened to the left carotid artery. A cut-back incision was made to the posterior of the descending aorta, which was then anastomosed end-to-end to the aortic arch. Depending on the surgeon's preference, 7/0 (>5 kg 6/0 was used) Maxon (Covidien, United States), Polidioksanon (Ethicon, United States), and Prolene (Ethicon, United States) were used as sutures with the continuous or posterior continuous, anterior interrupted technique. The patients radial and femoral pressures were recorded after the clamps were removed. Direct descending aortic pressure was measured in patients without a femoral artery cannula. After the bleeding was controlled, the mediastinal pleura was closed in all patients. One drain was placed in the left thorax and closed in accordance with the thoracotomy layers. All patients were hospitalised in the post-operative ICU, transthoracic echocardiography was performed both in the post-operative period and before discharge from the hospital, and routine follow-ups were performed in the outpatient clinic.

Follow-up

Early outcomes were defined as outcomes that occurred within 30 days after surgery or during hospitalisation. Late results were defined as results occurring after this period. Patients were followed up with routine echocardiography. Gradient patients exceeding 25 mmHg and extending to the diastole along the repair site and patients with a difference of more than 20 mmHg between the upper and lower extremity arterial pressures received cardiac catheterisation with a diagnosis of haemodynamically significant recoarctation. During cardiac catheterisation, a gradient above 20 mmHg was considered an indication for reintervention. Reintervention decisions were made by the patients' treating physicians based on the general clinical picture. Patients who developed recoarctation in the first 3 months post-operatively underwent surgery and balloon angioplasty after 3 months. Surgical intervention was performed again for patients who did not benefit from balloon angioplasty.

Statistical Analyses

Statistical analyses were performed using SPSS software (version 25.0, IBM, Armonk, NY, United States). The continuous data were expressed as medians (interquartile range) due to their non-normal distribution, while the categorical variables were expressed as frequencies and percentages.

Results

Preoperative variables

In our clinic, 90 patients underwent surgery to correct an isolated aortic coarctation. The patients' mean age was 280 ± 683.4 days, their mean weight was 6.42 ± 5.46 kg, and their mean body surface area was 0.33 ± 0.21 m^2. Thirty (33.3%) were newborns, 46 (51.1%) were infants, and 14 (15.5%) were children, 51 (56.7%) were male, and 39 (43.3%) were female.

Six (6.7%) patients were intubated and followed up in the preoperative period. Three (3.3%) patients underwent intervention in the preoperative period. All of these patients underwent balloon angioplasty in the external centre during the neonatal period two. Low cardiac output syndrome was present in 16 (17.8%) patients before the operation, 14 (15.6%) of whom received inotropic support while 1 (1.1%) received extracorporeal membrane oxygenation support in the preoperative period. Extracorporeal membrane oxygenation was continued for the latter patient during surgery, separate from post-operative extracorporeal membrane oxygenation. This patient then received dialysis treatment due to acute renal failure for two weeks; however, the patient died of sepsis on the 30th post-operative day. Three (3.4%) patients received a prostaglandin E1 infusion to correct patent ductus arteriosus-dependent distal circulation (Table 1).

Operative variables

All patients underwent extended end-to-end anastomosis. Three (3.3%) patients underwent pulmonary artery banding simultaneously, and 1 (1.1%) patient underwent aberrant right subclavian artery division. The mean cross-clamp time was 29.13 ± 6.97 minutes (Table 2).

Post-operative variables

The most common complication in our sample was pulmonary complications. Two (2.2%) patients were reattached due to pleural effusion and seven (7.9%) were reintubated due to atelectasis. Recurrent nerve paralysis was observed in one (1.1%) patient. With the exception of seizures, no major neurological deficits were observed. Two (2.2%) patients required reoperation in the early period: one patient underwent surgery the following day due to a technical problem in the anastomosis and the anastomosis was redoned, while the other patient underwent ductus ligation the following day due to excessive chylous drainage. Major adverse events were observed in two (2.2%) patients and mortality in

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Table 1. Demographics and preoperative variables

Variable	Mean ± SD (%) (n = 90)
Age (days)	280 ± 683.4
Weight (kg)	6.42 ± 5.46
Body surface area	0.33 ± 0.21
Sex	
Male	51 (56.7)
Female	39 (43.3)
Preoperative intubation	6 (6.7)
Pulmonary hypertension	3 (3.3)
Preterm	5 (5.6)
Preoperative intervention	3 (3.3)
Low cardiac output syndrome	16 (17.8)
Bovine arch	1 (1.1)
Preoperative extracorporeal membrane oxygenation	1 (1.1)
Non-cardiac anomaly	5 (5.6)
Bicuspid aortic valve	41 (45.6)
Aberrant right subclavian artery	2 (2.2)
Prostaglandin E1 infusion	3 (3.4)
Preoperative inotropic agents	14 (15.6)
Proximal arch Z-score	−2.30 ± 1.53
Distal arch Z-score	−1.66 ± 1.56
Aristotle comprehensive complexity score	7.29 ± 1.76
Mortality score	0.48 ± 0.62
Mortality category	1.48 ± 0.73

Table 2. Operative variables

Variable	n (%) (n = 90)
Procedure	
Extended end-to-end anastomosis	90 (100)
Additional procedure	
Pulmonary artery banding	3 (3.3)
Aberrant right subclavian artery division	1 (1.1)
Suture material	
7/0 Prolene	16 (19.8)
7/0 Polydioxanone	40 (49.4)
6/0 Prolene	4 (4.9)
6/0 Polydioxanone	2 (2.5)
5/0 Prolene	1 (1.2)
7/0 Maxon	18 (22.2)
Clamp time (min)	29.13 ± 6.97

one (1.1%) patient. The patients' mean intubation time was 23.00 ± 34.15 hours, their mean ICU stay was 5.22 ± 8.73 days, and their mean hospital stay was 8.64 ± 2.63 days (Table 3).

Table 3. Post-operative variables

Complications	n (%) (n = 90)
Pulmonary	
Pleural effusion	2 (2.2)
Reintubation due to atelectasis	7 (7.9)
Wound complication	1 (1.1)
Infection	1 (1.1)
Arrhythmia	–
Recurrent nerve paralysis	1 (1.1)
Diaphragm paralysis	–
Renal	1 (1.1)
Neurological deficit	–
Extracorporeal membrane oxygenation	1
Early reoperation	2 (2.2)
Major adverse events	2 (2.2)
Mortality	1 (1.1)
Ventilation time (h)	23.00 ± 34.15
Length of ICU stay (d)	5.22 ± 8.73
Length of hospital stay (d)	8.64 ± 2.63

Table 4. Long-term follow-up variables

Variable	Mean ± SD / n (%) (n = 28)
Echocardiography gradient	18.8 ± 8.14
Hypertension	22 (26.8)
Reoperation	4 (4.4)
Reintervention	4 (4.4)

Long-term follow-up variables

The mean follow-up time was 41.3 ± 22.8 months. The mean echocardiographic gradient last measured in the long-term follow-ups was 18.8 ± 8.14 mmHg. Twenty-two (26.8%) patients were followed up under antihypertensive treatment due to hypertension.

Four (4.4%) patients required balloon angioplasty during follow-up (Table 4). Three of this patient recoarctation developed in the early period and one patient after 7 years. Three of the patients underwent surgical repair in the neonatal period and one patient at the age of 1 year. Two (2.2%) of these patients received antihypertensive treatment in the follow-up period, while the other two patients did not develop any further problems.

Four (4.4%) patients needed reoperation in the long term. Of these four, three (3.3%) developed early recoarctation and one patient developed late recoarctation. Three patients underwent a second thoracotomy, two patients underwent resection and extended end-to-end anastomosis, and one patient received a second patch aortoplasty. The other patient underwent arch reconstruction with a median sternotomy. When we looked at the characteristics of the patients, two patients also had a hypoplastic aortic arch and one patient was premature, syndromic,

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Table 5. Patients with recarctation

Age at Surgery (d)	Weight at Surgery (kg)	Proximal arch Z-score	Operative Procedure	Intervention	Operation to reintervention (m)	Last ECHO aortic arch gradient (mmHG)	Late Hypertension
30	3	−3.4	REEA + PAB	Balloon angioplasty	3	17	–
1,800	18	−2.05	REEA	Patch aortoplasty	48	35	+
20	3.2	−1.7	REEA	Balloon angioplasty	5	17	–
35	4.6	−4	REEA	Repeat REEA	3	25	–
60	3.1	−1.86	REEA	Aortic arch reconstruction	2	40	+
17	3	−1.3	REEA	Balloon angioplasty	2	15	+
180	4.9	−0.89	REEA	Balloon angioplasty	5	38	+
15	3.3	−1.7	REEA	Repeat REEA	3	20	–

ECHO: echocardiography, PAB: Pulmonary artery band, REEA: Resection extended end-to-end anastomosis.

and had a low birth weight. Only one patient was prescribed medication due to late hypertension (Table 5).

Discussion

In this study, we examined the rates of mortality, morbidity, recoarctation, and late hypertension in patients who underwent resection and extended end-to-end anastomosis surgery in our centre to repair isolated aortic coarctation over the last 10 years. The resection and extended end-to-end anastomosis technique is applied with low mortality, morbidity, long-term reintervention, and reoperation rates in patients with an isolated coarctation.

Isolated coarctations are often accompanied by ventricular septal defect, bicuspid aortic valve, patent ductus arteriosus, and/or a hypoplastic aortic arch. Of the 90 patients we reviewed, 41 (45.6%) also had a bicuspid aortic valve and 18 (20%) also had a ventricular septal defect. Aortic arch hypoplasia is frequently accompanied by other defects, particularly in newborns. It is important to evaluate these patients preoperatively for surgical planning. Given the effect of the information on surgical planning, detailed evaluation of the aortic arch and associated cardiac anomalies is important. A patient's proximal transverse arch measurement and Z-score are key details in deciding the appropriate surgical approach for this population; however, distal transverse arch measurements are particularly important for patients with a bovine aortic arch. As echocardiographic measurements show subjective variability, CT angiography is recommended to evaluate borderline cases.[9] In our study, all patients were evaluated using both CT angiography and echocardiography. CT angiography is highly specific and sensitive, especially when used to evaluate extracardiac vascular pathologies. In their retrospective study on 226 simple aortic coarctation patients, Gropler et al. demonstrated that the resection and extended end-to-end anastomosis with a thoracotomy technique was performed with low mortality and a low recoarctation rate and suggested that the proximal arch Z-score is preferable for median sternotomy in patients with a distal arch score between −4 and −2.8.[10] The surgical technique used varies depending on the patient's characteristics and the preference of the centre (which is based on the experience of the surgeon and his or her team). In our centre, arch reconstruction with median sternotomy is used for patients with severe proximal and distal aortic arch hypoplasia, and the results of these cases have been reported previously.[11] All the patients included in our study were treated using resection and an extended end-to-end anastomosis technique through a left-side thoracotomy. In this technique, the aortic arch and descending aorta are mobilised and the first 2–3 rows of intercostal arteries are divided to reduce anastomotic tension. It is important that the patent ductus arteriosus tissue and coarcted segment are completely excised, that the aortic clamp is placed proximal to the left carotid artery, and that the incision is extended to the proximal along the small curvature of the arch. Two techniques were used for anastomosis, depending on the surgeon's preference: a continuous suture of the posterior wall, an interrupted suture of the anterior wall, and the full continuous technique.

The most important problem in patients who undergo aortic coarctation repair is the development of recoarctation. Different studies have reported the following factors affecting recoarctation: proximal arch hypoplasia,[12] suture type,[12,13] aberrant right subclavian artery, low weight,[14] and surgical technique.[15,16] Recently, Heremans et al.[17] reported a recoarctation rate of 13.5%, while Farag et al.[18] reported a rate of 9.9% and IJsselhof et al.[19] reported a rate of 10.3% in the neonatal group and 4.6% in the infant group. In our study, eight (8.8%) patients developed recoarctation: four of these patients underwent balloon angioplasty and four underwent reoperation. When we evaluated the characteristics of the patients who developed recoarctation, we found no statistically significant contributing factor. All the patients weighed over 3 kg. In the patients repaired with polypropylene sutures, higher rates of recurrent stenosis compared to absorbable polydioxanone sutures were observed both experimentally and clinically.[20] Polypropylene sutures were used in 4 patients, and polydioxanone sutures were used in four patients, depending on the surgeon's preference. Two patients had an aberrant right subclavian artery, which was divided among one patient and not divided in the other.

Recent studies have shown that major neurological complications are rare in these patients in the early period. Subarachnoid haemorrhage and ischaemic stroke due to permanent hypertension develop in many patients, especially at a young age.[21,22] Spinal cord

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ischaemia is also an important problem, especially in adult patients.[23] In our study, seizures were the only major complication observed in the early and late post-operative periods. In order to prevent neurological complications, the patients' body temperature was reduced to 34°C with a cooling bed during surgery, blood pressure was strictly monitored with a radial artery cannula during clamping of the aortic arch, they received cerebral and somatic near-infrared spectroscopy follow-up during surgery, antihypertensive medication was administered to those patients who developed late hypertension, and we closely and regularly followed up all patients after surgery.

The long-term complication of late hypertension is an important problem in this population. In a systematic review of 26 studies, 32.5% of patients were found to have developed late hypertension. The factors affecting the prevalence of hypertension were reported to include age during surgery, follow-up age, the method used to measure blood pressure, and the type of surgical intervention performed.[24] Recoarctation is another important risk factor.[25] In our series, 22 (26.8%) patients were using antihypertensive drugs due to hypertension. The mean age of surgery was 280 ± 683.4 days, 30 (33.3%) of the patients were neonates, 46 (51.1%) were infants, and 14 (15.5%) underwent intervention in childhood. The mean follow-up period was 41.3 ± 22.8 months. The mean echocardiographic gradient of the aortic arch was 18.8 ± 8.14 mmHg at the last control. Three of the patients who underwent intervention due to recoarctation were using antihypertensive drugs.

Studies conducted in the last decade have reported low rates of mortality in coarctation patients. One recent study presenting the results of isolated coarctation with thoracotomy reported a mortality rate of 1.4%.[26] In our study, mortality was observed in one (1.1%) patient, who underwent surgery under extracorporeal membrane oxygenation in the preoperative period, left-side extracorporeal membrane oxygenation in the post-operative period, and died of sepsis 30 days after surgery. The exclusion of patients with complex disease and severe transverse arch hypoplasia is one reason for the low mortality rate observed in the present study.

Study limitations

The primary limitation of this study is its retrospective design. There were no control groups treated with alternative techniques or a progressive operation to compare recurrence rates. Another limitation is the absence of long-term complications due to the short average follow-up period.

Conclusion

Isolated coarctation is successfully treated with left-side thoracotomy resection and extended end-to-end anastomosis technique and results in low mortality, morbidity, and long-term recoarctation rates. Long-term follow-up is necessary due to the risks of early and late post-operative recoarctation, which requires reintervention.

Data availability statement. Data sharing is not applicable to this article as no new data were created or analysed in this study. Institutional review board approval or waiver. The study protocol was approved by hospital management. This retrospective study was conducted in accordance with the principles of the Declaration of Helsinki.

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Conflicts of interest. None.

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