Original Article

Coarctation of the Aorta with Aortic Arch Hypoplasia: Midterm Outcomes of Aortic Arch Reconstruction with Autologous Pulmonary Artery Patch

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Abstract

Background: Coarctation of the aorta (CoA) with aortic arch hypoplasia (AAH) is a relatively common congenital heart disease in clinical practice. Nonetheless, the corrective surgical technique for infants and children is a clinical problem that remains controversial. In this study, we sought to evaluate the surgical effects of aortic arch (AA) reconstruction with coarctation resection and aortoplasty with autologous pulmonary artery patch for infants and young children with CoA and AAH.

Methods: Between January 2009 and December 2015, a total of 22 infants and young children with CoA and AAH who underwent coarctation resection and aortoplasty with autologous pulmonary artery patch were enrolled in this study. The median age of patients was 4.5 (Q1, Q3: 2.0, 14.0) months and the median body weight was 5.75 (Q1, Q3: 4.10, 9.38) kg. All patients were diagnosed with CoA and AAH, and concomitant cardiac anomalies were corrected in one stage. Perioperative and postoperative data were collected and analyzed using the paired sample *t*-test.

Results: No perioperative deaths occurred. No residual obstruction was detected by echocardiography. The postoperative pressure difference across the repaired segment of CoA was 14.05 ± 4.26 mmHg (1 mmHg = 0.133 kPa), which was smaller than the preoperative pressure difference (48.30 ± 15.73 mmHg; $t = -10.119$, $P < 0.001$). The median follow-up time was 29.0 (Q1, Q3: 15.5, 57.3) months. There was no death during the follow-up period, and all patients experienced obvious clinical improvement. Only one child underwent subsequent aortic balloon angioplasty due to restenosis. Computed tomography angiography showed that the AA morphology was smooth, with no aortic aneurysm or angulation deformity.

Conclusion: AA reconstruction with coarctation resection and aortoplasty with autologous pulmonary artery patch could effectively correct CoA with AAH, and the rate of reintervention for restenosis is low.

Key words: Aortic Arch Hypoplasia; Aortic Arch Reconstruction; Aortic Coarctation; Cardiac Surgical Procedures; Congenital Heart Disease

Introduction

Coarctation of the aorta (CoA) is defined as congenital narrowing of the aortic isthmus near the ductus arteriosus or arterial ligament. It can manifest as isolated stenosis or long-tubular hypoplasia of the transverse aortic arch (TAA). CoA, a common congenital heart disease (CHD) in clinical practice, accounts for 6–8% of CHD.[1] It may occur alone or in combination with other cardiac and vascular anomalies, such as atrial septal defect (ASD), ventricular septal defect (VSD), patent ductus arteriosus (PDA), bicuspid aortic valve, double outlet right ventricle (DORV), transposition of the great arteries (TGA), and other cardiac malformations.[2,3] CoA combined with aortic arch hypoplasia (AAH) is a serious cardiac abnormality that should be treated at an early stage to improve the

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long-term survival of patients. To date, the choice of corrective surgical technique for infants and children with CoA and AAH remains controversial. The commonly used surgical techniques include patch aortoplasty and extended end-to-end anastomosis (EEEA).[4] In our center, the commonly used patch material is pulmonary artery patch. In this study, we sought to evaluate the surgical effects of coarctation resection and aortoplasty with autologous pulmonary artery patch to provide advice for the treatment of CoA and AAH in infants and young children.

Methods

Ethical approval

This retrospective study was approved by the medical ethical review board, and informed consent was obtained from all patients.

Study population

Clinical data were retrospectively reviewed to screen for patients with CoA and AAH, with AAH diagnosed according to the CHD database.[5] Between January 2009 and December 2015, a total of 22 infants and young children with CoA and AAH underwent coarctation resection and aortoplasty with autologous pulmonary artery patch were enrolled in this study. The median age of patients was 4.5 (Q1, Q3: 2.0, 14.0) months and the median body weight was 5.75 (Q1, Q3: 4.10, 9.38) kg. The common associated cardiac anomalies in the study population included VSD, ASD, and PDA [Table 1]. Patients with associated complex CHD such as TGA, complete atrioventricular septal defect, and DORV were excluded from the study. Feeding intolerance, recurrent pulmonary infections, diminished femoral artery pulses, and congestive heart failure were common clinical manifestations among the studied patients. Conventional preoperative examinations included electrocardiography, chest X-ray, and echocardiography. Aortic computed tomography angiography was also necessary for accurate diagnosis.

Table 1: Clinical characteristics of enrolled patients (n = 22)

Patients characteristics	Results
Sex, *n*	
Male	14
Female	8
Infants, *n*	17
Children, *n*	5
Age (months)	
Median (Q1, Q3)	4.5 (2.0, 14.0)
Range	1–72
Weight (kg)	
Median (Q1, Q3)	5.75 (4.10, 9.38)
Range	3.2–18.0
Clinical manifestations, *n*	
Feeding intolerance	6
Recurrent pulmonary infections	8
Reduced femoral artery pulses	12
Congestive heart failure	7
Blood pressure (mmHg)	
Right upper limb systolic pressure	105.14 ± 17.30
Right lower limb systolic pressure	87.00 ± 21.54
Echocardiographic gradient (mmHg)	48.30 ± 15.73
Associated cardiac and vascular anomalies, *n*	
VSD	17
PDA	8
ASD	4
PFO	6
Bicuspid aortic valve	3
MI	5
MS	2
TI	4
PLSVC	3
PI	1
PAH	11
Noncardiac anomalies, *n*	
Bronchial stenosis	1

Data were presented as mean ± SD or median (Q1, Q3), or *n*; Blood pressure was measured at rest; 1 mmHg = 0.133 kPa; SD: Standard deviation; VSD: Ventricular septal defect; ASD: Atrial septal defect; PDA: Patent ductus arteriosus; PFO: Patent foramen ovale; MI: Mitral valve incompetence; MS: Mitral valve stenosis; TI: Tricuspid insufficiency; PLSVC: Persistent left superior vena cava; PI: Pulmonary insufficiency; PAH: Pulmonary arterial hypertension.

Operative technique

All patients underwent aortic arch (AA) reconstruction via median sternotomy with coarctation resection and aortoplasty with autologous pulmonary artery patch. Additional intracardiac malformations were corrected in one stage. The procedures were performed under extracorporeal circulation with deep hypothermic circulatory arrest and anterograde selective cerebral perfusion. To optimize surgical exposure, most of the thymus was removed intraoperatively.

To facilitate extensive dissection of the AA and its three branches, the descending thoracic aorta also had to be mobilized as much as possible. Once the stenotic segment was resected, the PDA and its surrounding tissues were also sufficiently resected. An incision was then made along the undersurface of the TAA. The incision was extended proximally to the origin of the innominate artery to effectively cut off the proximal congenital ring constriction [Figure 1a]. The posterior wall anastomosis consisting of the AA and descending thoracic aorta was made wide enough to reconstruct the posterior wall and part of the upper and lower walls of the AA.

After the narrowed segment was removed, an appropriately sized patch was cut from the pulmonary artery wall to reconstruct the AA defect – created by the resected coarctation. Care was taken to ensure that the patch dimensions exceeded those of the proximal AA narrowing to reduce the risk of proximal restenosis. For some patients with severe AAH, extensive removal of coarctation tissue required two or more pulmonary artery patches to reconstruct the tubular TAA structure and avoid annular anastomosis formation [Figure 1b and 1c]. All incisions to cut patches

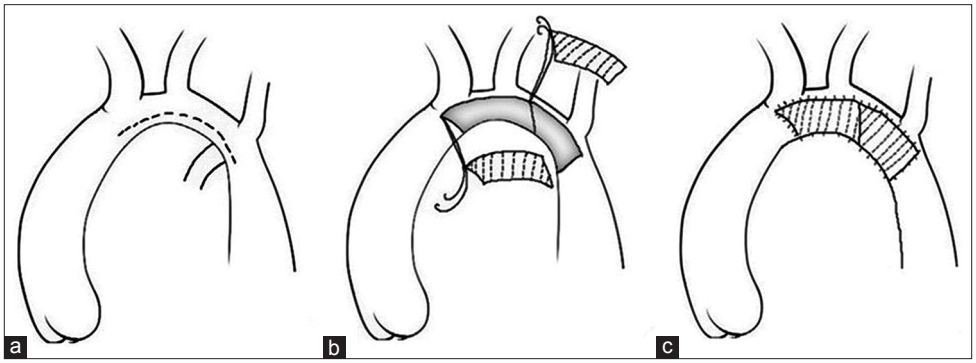

Figure 1: Surgical techniques for aortic arch repair. (a) An incision is made parallel to the TAA on the inferior surface, extended proximally to the origin of the innominate artery to sufficiently excise the proximal ring constriction; (b) After removing the narrowed segment, the AA defect is reconstructed with an autologous pulmonary artery patch. Occasionally, two or more pulmonary artery patches are necessary to reconstruct the AA in cases of severe AAH to reconstruct the tubular TAA structure and avoid the formation of an annular anastomosis; (c) The tubular AA structure of AA has been reconstructed with smooth morphology and no angulation deformity. TAA: Transverse aortic arch; AA: Aortic arch; AAH: Aortic arch hypoplasia.

from the pulmonary artery wall were located at least 5 mm away from the pulmonary artery sinuses and did not extend beyond the origin of the pulmonary artery branch. The pulmonary artery wall defect was reconstructed with fresh autologous pericardium.

Postoperatively, the peak pressure gradient across the repaired segment was measured by transesophageal echocardiography. A postoperative pressure <15 mmHg (1 mmHg = 0.133 kPa) was considered to be satisfactory.

Statistical analysis

All statistical analyses were performed using PASW Statistics version 18.0 (SPSS Inc., Chicago, Illinois, USA). Categorical variables were presented as numbers or percentages. Continuous variables were presented as the mean ± standard deviation (SD) for normally distributed values. Nonnormally distributed data were presented as median (Q1, Q3). The differences between the preoperative and postoperative data were compared using the paired sample *t*-test for normally distributed values. Nonnormally distributed data were compared using the nonparametric Wilcoxon rank-sum test. $P < 0.05$ was considered statistically significant.

Table 2: Operative data, postoperative results, and follow-up data (n = 22)

Characteristics	Results
Operative data (min)	
Cardiopulmonary bypass time	131.2 ± 33.1
Aortic cross-clamp time	78.6 ± 23.5
Selective antegrade cerebral perfusion time	38.0 ± 13.7
Postoperative results, median (Q1, Q3)	
Mechanical ventilation time (h)	27.5 (18.0, 90.0)
ICU monitoring time (days)	3.7 (1.5, 9.5)
Postoperative hospital stay (days)	14.5 (9.0, 25.0)
Blood pressure (mmHg)	
Right upper limb systolic pressure	89.00 ± 9.49
Right lower limb systolic pressure	100.57 ± 14.49
Echocardiographic gradient (mmHg)	14.05 ± 4.26
Follow-up time (months)	29.0 (15.5, 57.3)
Complications, *n*	
Severe pulmonary infection	6
Reintubation	1
Re-intervention, *n*	1
Overall mortality, *n*	0

Data were presented as mean ± SD, median (Q1, Q3), or *n*; Blood pressure was measured at rest; 1 mmHg = 0.133 kPa. SD: Standard deviation; ICU: Intensive Care Unit.

Results

Clinical characteristics

A total of 22 infants and young children (14 males and 8 females) were enrolled in this study. All patients underwent coarctation resection and aortoplasty with autologous pulmonary artery patch. The patients' characteristics are listed in Table 1. The mean postoperative right upper limb systolic blood pressure was 89.00 ± 9.49 mmHg, representing a significant decrease from the preoperative mean systolic blood pressure of 105.14 ± 17.30 mmHg (t = −4.582, $P < 0.001$) [Tables 2 and 3]. The mean postoperative right lower limb systolic blood pressure was 100.57 ± 14.49 mmHg, representing a significant increase from the preoperative mean systolic blood pressure of 87.00 ± 21.54 mmHg (t = 5.973, $P < 0.001$). The mean pressure difference across the repaired segment of CoA was 14.05 ± 4.26 mmHg, representing a significant decrease from the preoperative result of 48.30 ± 15.73 mmHg (t = −10.119, $P < 0.001$).

Morbidity

No inhospital death, renal failure, or neurological complications occurred among the 22 patients studied. Six patients with increased airway secretions were found to have serious pulmonary infections, all of which responded well to drug therapy [Table 2]. One infant with a serious postoperative pulmonary infection required repeated tracheal intubation. This patient was 2 months old with ASD, VSD, and severe pulmonary artery hypertension (PAH). The intensive care unit stay for this patient was 16 days, including 13 days of ventilator support. This patient eventually

recovered and was discharged from the hospital 45 days postoperatively.

Follow-up and recoarctation

Postoperative follow-up data were obtained from 20 patients, with a median follow-up time duration of 29.0 (Q1, Q3: 15.5, 57.3) months. No deaths occurred during the follow-up period, and there were no aortic aneurysms, bronchial compression, or aortic angular deformities [Figure 2]. Blood pressures and echocardiography were checked regularly during clinical follow-up. No obvious hypertension was detected, and pulmonary valve function was found to be normal in all patients without regurgitation or stenosis. Restenosis was defined as a resting echocardiographic peak pressure gradient across the repaired segment of >20 mmHg. During the follow-up, the echocardiographic peak pressure gradient increased to >20 mmHg in three patients, 1 of whom required reintervention by aortic balloon angioplasty. Further medical history reviewed showed that this patient underwent corrective surgery at 4 months of age, after which the echocardiographic peak pressure gradient clearly decreased postoperatively from 48 mmHg to 18 mmHg. However, 30 months later, the peak pressure gradient rose from 18 mmHg to 47 mmHg and rose further to 85 mmHg during the subsequent 12 months. When this patient underwent aortic balloon angioplasty at our hospital, angiography revealed that restenosis had occurred primarily at the distal anastomosis site. After this patient underwent aortic balloon angioplasty, the peak pressure gradient fell from 85 mmHg to 40 mmHg. Follow-up was continued for two other patients because their peak pressure gradients were found to be 22 mmHg and 27 mmHg, respectively, with no obvious symptoms.

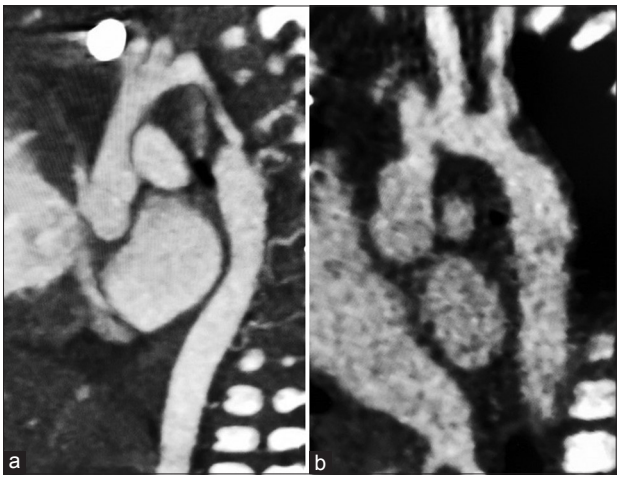

Figure 2: Pre- and post-operative computed tomography angiography images. (a) Preoperative CTA image of a 1-month-old infant that was diagnosed with CoA and AAH; (b) Postoperative CTA was examined 13 days after autologous pulmonary patch aortoplasty. CTA: Computed tomography angiography; CoA: Coarctation of the aorta; AAH: Aortic arch hypoplasia.

Discussion

It is generally accepted that once diagnosed, CoA for which surgery is indicated should be corrected as early as possible.[6] Operative indications include AA diameter reduction of >50% or a resting echocardiographic peak pressure gradient of greater than 20 mmHg across the coarctation. To decrease the incidence of complications and increase the long-term survival rate, surgical correction should optimally be performed during infancy.[7] CoA combined with AAH is a complex form of CHD. Without timely and effective treatment, patients with this combined disorder always have a poor prognosis. Conte *et al*. reported that the early appearance of clinical symptoms correlated strongly with severe condition among newborns with CoA. In addition, up to 81% of these newborns had CoA combined with AAH.[8] For now, there are still no definite diagnostic criteria for AAH. In this study, the diagnostic criteria of AAH were based on the CHD database. According to these criteria, AAH is present if the AA diameter between the innominate artery and the left common carotid artery is <60% of the ascending aorta diameter, if the AA diameter between the left common carotid artery and the left subclavian artery is <50% of the ascending aorta diameter, or if the diameter of the aortic isthmus is <40% of the ascending aorta diameter.[5]

To reconstruct the AA and avoid the recurrence of restenosis, a variety of surgical procedures have been developed in clinical practice. At present, the common surgical techniques for CoA with AAH include patch aortoplasty and EEEA. EEEA is a modified end-to-end anastomosis technique. The first step involves dissection of the descending thoracic aorta, the AA and its three branches to mobilize the AA and descending aorta. The next step involves excision of the PDA and its surrounding coarctation tissues. Finally, an incision is created parallel to the undersurface of the TAA, with the open end of the descending aorta beveled to match the incision on the AA before it is sewn to the underside of the TAA. EEEA may be used to correct AAH. However, this procedure tends to result in high anastomotic tension due to limited mobilization of the AA and descending aorta. In turn, the resulting scar formation creates a potential risk factor for postoperative restenosis.[9] Furthermore, as mentioned above,

Table 3: Comparison of preoperative and postoperative data (n = 22)

Variables	Postoperative	Preoperative	t	P
Right upper limb systolic pressure (mmHg)	89.00 ± 9.49	105.14 ± 17.30	−4.582*	<0.001
Right lower limb systolic pressure (mmHg)	100.57 ± 14.49	87.00 ± 21.54	5.973*	<0.001
Echocardiographic gradient (mmHg)	14.05 ± 4.26	48.30 ± 15.73	−10.119*	<0.001

Data were presented as mean ± SD; 1 mmHg = 0.133 kPa. *Paired-sample t-test. SD: Standard deviation.

EEEA cannot effectively relieve the proximal obstruction of the hypoplastic AA, which also creates a potential risk factor for residual stenosis. In addition, the residual proximal AA obstruction would represent an important risk factor for postoperative restenosis and long-term hypertension due to the inconsistent growth rates of the distal and proximal AA.[10] The recent data suggest the restenosis rates of EEEA at 4–13% looking at patients 5–10 years postsurgery.[11-16] Accordingly, EEEA may be not always effective for the patients with CoA and AAH.

Patch aortoplasty was proposed in the 1990s as an operative technique for CoA. Potential patch materials include artificial materials, homograft vessels, autologous pericardium, and autologous pulmonary artery patches. Due to the high incidence of restenosis and aortic aneurysms, artificial materials have not been widely used in clinical practice.[17,18] Compared to other materials, autologous pericardium and autologous pulmonary artery patches have been widely used in clinical practice for the treatment of CoA with AAH. In 2013, Bernabei *et al*. reported a group of 39 newborns with CoA and AAH. No aortic aneurysms occurred among those who underwent AA reconstruction with autologous pericardium patches. However, restenosis occurred in 11 patients (28.2%), during mid- and long-term follow-up.[19] Compared to autologous pericardium, the autologous pulmonary artery patch has good elasticity, histocompatibility, growth potential, and high homology with the aortic wall.[20,21] According to the reported literature, no restenoses or aortic aneurysms occurred in the patients who underwent autologous pulmonary patch aortoplasty during midterm follow-up; the restenosis-free rate at 3 years after surgery was 100%.[9,22]

In our center, the commonly used surgical technique for patients with CoA and AAH is patch aortoplasty, and the pulmonary artery patch is commonly used. Through a comparative study with the pericardium patch aortoplasty group, we found that the pulmonary patch aortoplasty group had superior midterm outcomes. Kaplan–Meier curves revealed that for the pericardium patch aortoplasty group, the restenosis-free rates at 1 year and 3 years after surgery were 67.6% and 33.8%, respectively. Both of these rates were lower than those of the pulmonary patch aortoplasty group, which experienced restenosis-free rates of 95.5% and 72.1%, respectively (both $P < 0.05$).[23] No aortic aneurysms occurred in our study during the follow-up period, and only one infant (4.5%) underwent aortic balloon angioplasty 3.5 years postoperatively due to restenosis. We reviewed the medical history of this patient and considered that the reasons of restenosis for this patient might have been related to growth retardation of the AA and scar formation at the distal anastomosis.

The postoperative blood pressure is a critical factor which could influence long-term survival.[24] It has been reported that the incidence of hypertension in the patients who underwent EEEA was nearly 18%, and the long-term survival rate was 93%.[11,14,25,26] By comparison, no hypertension or deaths occurred in the patients who underwent autologous pulmonary patch aortoplasty during midterm follow-up.[9,22] In 2004, Ou *et al*. reported that postoperative hypertension was closely related to AA morphology and was clearly increased among patients with Gothic arch deformities.[27,28] Through comparative research on AA morphology, Seo *et al*. reported that 9 children (47.4%, $n = 19$) who underwent EEEA or end-to-side anastomosis with larger AA aspect ratios experienced reduced AA curvature, which made the AA look like a Gothic arch. However, Gothic arch deformities were not found in the children ($n = 12$) who underwent aortoplasty with autologous pulmonary artery patches.[29] In our study, we were very concerned about the reconstruction of the tubular AA structure during the surgery. During midterm follow-up, no deaths or hypertension occurred, and no bronchial compression or Gothic arch deformities developed in any patients.

PAH is most common in patients with CoA and AAH. The resultant dilation of the pulmonary artery provides favorable conditions for acquisition of a pulmonary artery patch. To avoid pulmonary valve regurgitation and coarctation of the pulmonary artery branch, we ensure that the surgical incision remains far from the sinuses and the pulmonary artery branch. Pulmonary arterioplasty may be performed concurrently, followed by reconstruction with fresh autologous pericardium. In our study, no pulmonary valve regurgitation or stenosis occurred during follow-up, and no aneurysmal dilation of the pulmonary artery and pulmonary branch arterial stenosis had been detected.

There were some limitations in our study. First, the number of patients in this study was small, and the time span was large. Moreover, the patients investigated were from a single center, which might not completely represent the general level of this surgical technique. Finally, although the operations were satisfactory with lower long-term reintervention rates, aortic aneurysm and restenosis represent long-term challenges which require long-term observation. Accordingly, longer follow-up may be necessary to evaluate subsequent developments with the pulmonary artery patch and AA.

In our study, patients with CoA and AAH can be effectively treated by coarctation resection and AA reconstruction using an autologous pulmonary artery patch. No aortic aneurysms, bronchial compression, or Gothic arch deformities occurred during midterm follow-up, and good follow-up results have been observed with low rates of reintervention for restenosis. Therefore, coarctation resection and AA reconstruction with autologous pulmonary artery patch could be a reliable and desirable operative technique for infants and children with CoA and AAH.

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Conflicts of interest

There are no conflicts of interest.

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