One-stage repair of transposition complex and interrupted aortic arch in children

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Abstract

Background/Aim: A transposition complex with an interrupted aortic arch (IAA) is rare and surgically challenging because of its anatomical diversity and complexity. Herein, we aimed to present our 20-year experience with one-stage arterial switch surgery associated with IAA repair. Methods: From January 2000 to April 2017, 11 patients were diagnosed with transposition complex and IAA and underwent one-stage repair at our center. These patients were retrospectively reviewed. Two patients had transposition of the great arteries, while the others had double outlet right ventricles, of whom eight had subpulmonary ventricular septal defects (Taussig-Bing anomalies), and one had a non-committed ventricular septal defect. In terms of the IAA, three patients underwent repair by extended end-to-end anastomosis, and one 16-mm prosthetic vascular graft was replaced in an elder patient. The remaining patients underwent autologous pericardial patch enlargement. All the variables were summarized and reported with descriptive statistics. Results: Three early deaths occurred in this study. The median follow-up time was approximately 5 years (range: 3 – 14 years). No late deaths were reported. Only one patient required percutaneous re-intervention for recurrent coarctation. Moderate aortic regurgitation was observed in three patients. However, there was no requirement for aortic valvuloplasty or valve replacement. One patient had more than moderate tricuspid regurgitation. The other survivors are presently healthy. Conclusions: Although one-stage repair for transposition complex and IAA still has non-negligible mortality even in older children, the late outcomes of survivors are acceptable. Owing to the high rate of valve regurgitation, closer follow-up is necessary for these patients.

Original article

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Conclusions: Although one-stage repair for transposition complex and IAA still has non-negligible mortality even in older children, the late outcomes of survivors are acceptable. Owing to the high rate of valve regurgitation, closer follow-up is necessary for these patients.

Keywords: Congenital heart disease, transposition complex, interrupted aortic arch (IAA), one-stage repair, cardiac surgery

Introduction:

Patients with transposition complex and interrupted aortic arch (IAA) are very rare. It has been reported that only 6–10% of infants with IAA have transposition of the great arteries (TGA).[1] In the past, a staged repair was preferred. Primary IAA repair was performed via a left thoracotomy, followed by an arterial switch operation (ASO) after several months, which has been reported to be associated with high mortality. During the last two decades, a one-stage repair of IAA at the same time as ASO has been generally accepted as the treatment of choice.[2]

However, even in the recent era, these patients still represent a high-risk subgroup of candidates, and only a few reports have been published on this group. Herein, we report our surgical experience using several approaches since the year 2000 with one-stage ASO and IAA repair.

Materials and Methods:

Subjects

Between January 2000 and June 2017, 11 patients (seven boys and four girls) underwent complete one-stage repair of the transposition complex with IAA through midline sternotomy at Beijing Fuwai Hospital. The median age at the time of surgery was five months (range: 10 days–71 months), and only one patient was less than one month old. The median body weight was 6.2 kg (range: 2.2–15.0 kg). The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013) and was approved by the Ethics Committee of Beijing Fuwai Hospital. The requirement of individual consent for this retrospective analysis was waived.

There were two patients with TGA and ventricular septal defect (VSD) and seven patients with Taussig-Bing anomaly (TBA). One patient had double outlet right ventricle (DORV) and non-committed VSD, in which the VSD was below the septal leaflet, but the overriding tricuspid valve was absent. One patient with TGA had both perimembranous and muscular VSDs. Right ventricular outflow tract obstruction (RVOTO) was present in four patients and moderate mitral valve regurgitation in two patients. All patients except one had IAA type A, and the other had type B IAA. The relationships between the great arteries were anteroposterior in eight patients and side-by-side in the other three patients. The pulmonary-aorta ratio was observed to be more than 1.5:1 in all the patients, while the most discrepant one was 4:1. The coronary anatomies were normal (1L, Cx; 2R) in nine patients, and a single coronary artery was present in two patients. Intramural coronary arteries were not observed. Prostaglandin E_1 was administrated to the only newborn in this group preoperatively. No balloon atrial septostomy, mechanical ventilation, or inotropic support was required before definitive repair. Clinical data are presented in Table 1.

Surgical Technique

All surgeries were performed using a routine sternotomy incision. Except for arterial cannulation in the ascending aorta, ductal cannulation was used to ensure distal perfusion. Bicaval venous cannulation was performed to avoid hypothermic circulatory arrest while repairing the intracardiac anomaly. Myocardial protection was accomplished with a single dose of histidine-tryptophan-ketoglutarate solution, regardless of the length of aortic clamping.

These 11 procedures were completed by four surgeons at our center. Therefore, the sequence and details of the repair might vary depending on the surgeon's preference. Except for the first patient in which the VSD was closed before aortic arch reconstruction in order for the temperature to reach 18°C, the repair of the IAA was routinely performed after aortic clamping at 25–28°C core temperature. At that point, aortic cannulation in the ascending aorta was transferred into the innominate artery, and systemic arterial perfusion was stopped while selective antegrade cerebral perfusion was used. Extended resection, including the ductal tissue, was performed, and three different approaches were used for arch repair. The first three patients underwent extended end-to-end anastomosis before 2012. Subsequently, aortic arch reconstruction, including the first posterior hemi-anastomosis between the posterior walls of the distal arch and the proximal descending aorta, and then enlargement of the arch with glutaraldehyde-treated autologous pericardial patches was performed in seven patients. A 16-mm intergard woven vascular graft (Maquet, La Ciotat, France) was used for a 6-year-old child.

After IAA repair was completed, arterial cannulation was moved back into the ascending aorta, and extracorporeal perfusion was resumed. Subsequently, VSD closure and ASO were performed. The VSD was closed with a Dacron patch through the tricuspid valve in two patients, through the tricuspid valve and original pulmonary valve in seven patients, or even through an infundibulotomy incision in two patients, among whom a non-committed VSD was present in one. An approximate 5 mm fenestration was left in the VSD patch in two older patients (4-year and 6-year-old), in case of persistent postoperative pulmonary hypertension (PHT).

After harvesting from the original aortic root, the coronary buttons were anastomosed to the original pulmonary root with the most favorable orientation to avoid significant angulation of the coronary arteries using the trap-door technique. When the discrepancy between the great vessels was remarkable, part of the wall of the neoaortic non-coronary sinus was stitched together using a continuous 5/0 or 6/0 Prolene suture to

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diminish the size of the neoaortic root to suit the proximal ascending aorta. The Lecompte maneuver was used in all patients except one whose great vessel relationship was side by side. Resection of the prominent parietal and septal bands was accomplished carefully through the original pulmonary root in two of the four patients with RVOTO to avoid injury to the neoaortic valve. In the other two patients, an infundibulotomy incision was required. Two patients were diagnosed with moderate mitral valve regurgitation preoperatively; posterior annulus constriction with 5/0 Prolene suture was performed to complete mitral valvuloplasty.

Modified ultrafiltration is routinely performed after cardiopulmonary bypass. A left atrial pressure monitoring line was inserted intraoperatively through the atrial septum. Only two patients were transferred to the intensive care unit (ICU) with an open sternum. Operative data are presented in Table 2.

Postoperative Care

Inotropic support was initiated through the combined use of dopamine and dobutamine. Adrenaline was also needed in most patients, combined with vasodilators such as milrinone to reduce afterload. In three patients aged >1 year, inhaled nitrous oxide and oral sildenafil and bosentan were used to treat postoperative pulmonary hypertension and prevent pulmonary hypertension crisis. Peritoneal dialysis was used in seven patients to remove excessive water. The median ICU stay was 23.4 days (range: 2–35 days).

Follow-up

All hospital and follow-up data were retrospectively collected from patients' clinical records. Patients who survived the operation would return to our center for follow-up examinations at certain intervals when direct contact was made with the patients' caretaker. Echocardiograms were available for all patients. The median follow-up time was 58.5 months (range: 37–163 months).

Results:

Early Mortality

There were three early deaths (27.3%) among the 11 patients. The first patient was patient 2, a 4-month-old female infant weighing 3.5 kg with TGA, type A IAA. The relationship between the great arteries was anteroposterior, with a remarkable discrepancy of 4:1. The patient had both perimembranous and muscular VSDs and normal coronary artery anatomy (1L, Cx; 2R), and underwent an uneventful surgery. However, an immediate postoperative echocardiogram demonstrated a low left ventricular ejection fraction of approximately 25%, without explicit segmental wall motion abnormality. Severe pneumonia followed, and a high-frequency oscillatory ventilator was used. The patient died of sepsis on postoperative day (POD) 34.

The second was patient 3, a 10-day-old and 2.1 kg premature newborn girl with TBA, type A IAA, side by side relationship of the great vessels with significant mismatch 3:1, and a single coronary artery ostium at sinus 1. The patient's surgical procedure was uneventful. Moderate aortic valve regurgitation was detected by echocardiography on POD three but did not evolve thereafter. The patient's chest was left open after surgery and was not successfully closed until POD 13 because of refractory oliguria and severe edema. The patient died of severe capillary leakage on POD 17.

The third patient was patient 8, a 2-month-old and 6.1 kg boy with TBA, type A IAA, anteroposterior great vessels, and typical coronary artery anatomy. The patient exhibited anuria one day after surgery and subsequently immediate renal failure. Soon after, multiple organ dysfunction developed. After conferring with his parents, therapy was stopped 26 days after the surgery; eventually, the infant died.

Early Morbidity

Two patients required tracheostomy and persistent ventilatory support for approximately 1 month. Cerebral hemorrhage and infarction occurred in one case each, but no permanent sequelae were observed.

Re-intervention and Late Events

No late deaths occurred during the follow-up. Recoarctation occurred only in one patient who underwent percutaneous aortic balloon angioplasty 13 months after the initial surgery. No false or true aortic arch aneurysms were detected in patients undergoing patch reconstruction (Fig. 1). Three neoaortic valves showed moderate regurgitation and were under close follow-up. Mild pulmonary stenosis was observed in one patient. None of the patients had an RVOT pressure gradient >30 mmHg. At the last follow-up, no reoperation was required for neoaortic valve dysfunction or right-sided obstructions. One tricuspid valve showed more than moderate regurgitation. This was noted in patient 7 who had DORV with a non-committed VSD and type B IAA. The patient has no significant symptoms and remained under close monitoring. For the two patients who had 5 mm VSD fenestrations, a 2 mm and 4 mm residual shunt could still be detected at the latest follow-up echocardiograms.

All surviving patients were in New York Heart Association (NYHA) class I, except for patient 10, who was 4-year-old and had reserved VSD fenestrations at the initial surgery. The patient's condition remained NYHA class II, and the patient was still receiving bosentan orally for postoperative PHT. No cardiac medication was required for the other patients.

Discussion:

Only a few case reports and series, including single-digit patient numbers, exclusively documented the surgical procedures in the transposition complex associated with IAA.[3-5] Pigott et al. first reported the single-stage strategy for TGA with aortic arch obstruction (AAO) in 1987.[6] Moreover, in 1993, Planché et al. demonstrated superior outcomes of the single-stage repair over the two-stage repair for TGA, VSD, and AAO.[2] The advantages of the single-stage repair may be due to the early reestablishment of the physiological circulation and aversion of detrimental effects of the pulmonary artery banding. Moreover, the hypoplastic aortic arch, coarctation, and interruption can be treated efficiently through a median sternotomy incision with mild tension on the anastomosis at the time of the ASO.[7] Since 2000, single-stage repair for ventriculoarterial discordance associated with IAA has become the treatment of choice at our center.

Compared to other series, the age of the patients at the initial repair was significantly older in this series, which is closely related to the social and economic developmental factors in our country. Because of poor medical and health conditions at the grass-root level, most of the patients in this series were beyond the neonatal period when they were first referred to our center; moreover, there were two patients, a 4- and 6-year-old. To some extent, these patients were somewhat "naturally selected." This may explain why the incidence of coronary artery anomaly was obviously low, and no right ventricle hypoplasia was detected preoperatively in our series, both of which remain important risk factors for this complex subgroup of patients, as stated in the report by Pocar et al.[8] To complete accurate coronary artery transfer during ASO, coronary reimplantation after neoaortic reconstruction was recommended by some previous studies.[9] In our series, the trap-door technique was used, but coronary malperfusion did not seem to be the direct cause of death. Of the three early deaths, only one had abnormal coronary artery anatomy, which was a single coronary artery originating from sinus 1. The premature neonate died of severe capillary leakage subsequently. In the other two patients, the direct causes of death were sepsis and treatment withdrawal because of multiple organ dysfunction.

Although coronary artery anomalies were uncommon, there were other risk factors in our series. Compared to patients with noncomplex TGA or TGA with arch coarctation, PHT is more common and persistent in this population, with complex forms of TGA associated with IAA. Postoperative PHT has been believed to be correlated with a prolonged stay in the ICU among patients with coexisting VSD,[8] and implantation of a fenestrated atrial or ventricular septal patch or a one-way valved atrial patch has been advocated in patients with hypoplastic right ventricle (RV) or PHT.[10,11] In our series, postoperative inhaled nitric oxide and oral sildenafil and bosentan were used to reduce pulmonary artery pressure in three patients who were >1 year old, among whom VSD fenestration was performed in two older children. In two of these three children, medications were stopped for a satisfactory decrease in pulmonary artery pressure and good exercise tolerance. This shows that PHT is reversible, even in older children with complex anomalies. However, a 4-year-old child at the time of operation required continued oral bosentan for persisting PHT.

This demonstrates the complexity and diversity of PHT.

Size discrepancy between the great arteries has been reported to significantly complicate surgery and influence early and late outcomes, especially neoaortic valve regurgitation. The fate of the neoaortic root and valve is of particular concern after the ASO.[12] A higher postoperative neoaortic root/ascending aorta ratio may be a risk factor for neoaortic regurgitation evolution at follow-up.[13] Anastomosis of a dilated neoaortic root with a diminutive ascending aorta may cause supravalvular aortic stenosis, which has been reported as a cause of early death after an ASO.[14] In previous studies, several procedures have been performed to overcome the mismatch in size, such as V-shaped resection of the posterior sinus,[13] extensive patch enlargement of the entire arch and ascending aorta,[7] and relocation of the right coronary above the aortic anastomosis.[15] Herein, sinus plasty after transfer of the coronary arteries was performed in 7 of the 11 patients. Without using additional material in the anastomosis of the neoaorta, the probability of major distortion and bleeding of the reconstructed sinotubular junction was minimized through a simple procedure. The integrity of the neoaortic root and apparent diminutive size of the ascending aorta could pose a concern. However, no supravalvular aortic stenosis or neoaortic root dilation was found on postoperative or follow-up echocardiography. Although three patients developed moderate neoaortic valve regurgitation, no reoperation was necessary.

Transpulmonary VSD closure has also been reported to be a significant risk factor for neoaortic valve regurgitation.[13] Moreover, in a previous study, it was believed that it was possible to close the VSDs through the tricuspid valve in nearly all cases.[16] However, in our series, we closed the VSD through the right atrium in two patients with TGA, through the right atrium and the original pulmonary valve in seven patients with TBA, and through an infundibulotomy incision in one patient with TBA and one patient with DORV and non-committed VSD. The high probability of transpulmonary VSD closure in our series may be the reason for the relatively high rate of aortic valve regurgitation (3 of 8 surviving patients). However, we believe that a decent approach route depends on anatomic features, and irrespective of the route used, it is essential to carefully trim the VSD patch to avoid interference with the neoaortic valve.

In a report from Australia, the end-to-side arch repair was performed in four of five patients with transposition complex and IAA.[5] The authors suggested that pericardial patch augmentation of the arch should be avoided, because it may result in dilation of the neoaorta and subsequent neoaortic valve insufficiency.[5] However, in a series from France, extended end-to-end anastomosis was reported to be a risk factor for recoarctation; in another report from England, augmentation of the arch with a homograft patch was advocated for IAA repair.[7] Repair procedures performed for IAA at our center varied over time. In the first three cases before 2012, extended end-to-end anastomosis was performed. Since then, glutaraldehyde-treated autologous pericardial patch enlargement has been performed in the following eight patients, except for the 6-year-old child in whom a 16-mm interposition graft was implanted; therefore, this approach is our preferred choice for IAA repair in patients with associated anomalies. We believe that this approach may significantly reduce the tension of the anastomosis and the distortion of the neoaortic root.

In our series, we often completed IAA repair prior to ASO; hence, it is extremely important to tailor the patch to provide less of an acute angle of the arch, especially after performing the Lecompte maneuver and moving the ascending aorta posteriorly. Sometimes, two separate patches are required, with one moving up the ascending aorta and one moving down the descending aorta to create a smoother curve. Re-intervention for recoarctation was necessary for only one patient in whom percutaneous balloon angioplasty was performed uneventfully 13 months after the initial surgery, in contrast to the findings of several previous series with a high rate of re-intervention.[7,13] Differences in surgical technique and thresholds of balloon dilation for recoarctation may explain such differences.

Limitations

This was a retrospective study. It only included 11 consecutive patients with a rare complex congenital heart disease and had limited statistical analysis. The operation is technically challenging, and selection bias may not have been completely avoided. Moreover, larger patient numbers and longer follow-up times are needed to enable survival and potential risk factor analyses.

Conclusions

One-stage repair of the transposition complex and interrupted aortic arch remains technically challenging and has non-negligible mortality, even in older children. However, the follow-up outcomes of the survivors were acceptable. Several procedures can be performed to repair the interrupted aortic arch with a low recoarctation rate. Continuous follow-up is required for the high incidence of long-term valve regurgitation.

Author contributions:

Study concepts and design: LZ and QW; Literature research: LZ; Clinical studies: LZ and ML; Statistical analysis: LZ; Manuscript preparation: LZ and ML; Manuscript editing: SL, JY and QW.

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Table 1. Clinical patient data

No.	Age (m)	Weight (kg)	Sex	Transposition complex	IAA type	Relation of GAs	Coronary artery	RVOTO	Follow-up (m)	Prognosis
1	2	4.5	M	TBA	A	anteroposterior	1L or Cx;2R	No		119
2	4	3.5	F	TGA	A	anteroposterior	1L or Cx;2R	No		
3	10 d	2.2	F	TBA	A	side by side	2L, Cx,R	No		
4	5	6.2	F	TBA	A	anteroposterior	1L or Cx;2R	Yes		45
5	71	15	F	TBA	A	anteroposterior	1L or Cx;2R	No		33
6	6	6.6	M	TBA	A	anteroposterior	1L or Cx;2R	No		25
7	28	11	M	DORV (ncVSD)	B	side by side	1L, Cx;2R	Yes		24
8	2	6.1	M	TBA	A	anteroposterior	1L or Cx;2R	Yes		
9	8	6.8	M	TBA	A	side by side	1L, Cx;2R	No		13
10	50	15	M	TBA	A	anteroposterior	1L or Cx,R	Yes		16
11	4	4.8	M	TGA	A	anteroposterior	1L or Cx;2R	No		6

d, days; DORV, double outlet right ventricle; GAs, great arteries; IAA, interrupted aortic arch; m, months; ncVSD, non-committed ventricular septal defect; RVOTO, right ventricular outflow tract obstruction; TBA, Taussig-Bing anomaly; TGA, transposition of great arteries.

Table 2. Perioperative patient data (n=11)

Characteristic	Number of patients
VSD repair through	
TV	2
TV+PV	7
RVOT	2
IAA repair	

Characteristic	Number of patients
Extended end-to-end anastomosis	3
Patch enlargement	7
Interposition graft	1
RVOTO repair through	
Infundibulotomy incision	2
PV	2
VSD fenestration	2
Mitral valvuloplasty	2
Lecompte	10
Delayed sternal closure	2
Peritoneal dialysis	7
Variables	**Mean±SD**
CPB time (min)	263.1±44.9
ACC time (min)	177.2±24.3
HCA time (min)	31.2±8.2
ICU stay (days)	23.4±10.1

ACC, aortic cross-clamp; CPB, cardiopulmonary bypass; HCA, hypothermic circulatory arrest; IAA, interrupted aortic arch; ICU, intensive care unit; PV, pulmonary valve; RVOT, right ventricular outflow tract; TV, tricuspid valve; VSD, ventricular septal defect.

Figure 1. Three-dimensional computed tomography images of a 3-month-old infant with Taussig-Bing anomaly and interrupted aortic arch (IAA). (a) Preoperative anterior view showing the transposition of the great arteries. (b) Preoperative posterior view showing the IAA. (c) Two years after the procedure