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Original Article / Özgün Makale

The Ross and Ross-Konno operations for congenital aortic valve diseases in infants and children

Bebekler ve çocuklarda doğuştan aort kapak hastalıklarında Ross ve Ross-Konno ameliyatları

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ABSTRACT

Background: In this study, we present early and mid-term outcomes of Ross and Ross-Konno operation in pediatric patients with congenital aortic stenosis.

Methods: Between January 2011 and December 2015, a total of eight patients (4 boys, and 4 girls; median age 4.5 years; range 3 to 17 years) with a preliminary diagnosis of congenital aortic stenosis underwent either Ross or Ross-Konno operation were retrospectively analyzed. Duration of cardiopulmonary bypass and cross-clamp, and length of intensive care unit and hospital stays were recorded.

Results: Seven patients had previous interventions [balloon valvuloplasty (n=4), ventricular septal defect repair (n=1), aortic valve repair (n=1), and aortic coarctation repair (n=1)]. The Ross-Konno operation was performed in five patients, while the remaining three patients underwent the Ross operation. Additional subaortic resection was required in two patients and mitral valve repair in one patient. Only one adult-sized patient had pulmonary autograft wrapping with Dacron conduit. There was no mortality and major morbidity. The mean cardiopulmonary bypass and cross-clamp times were 234±6 and 177±4 min, respectively. Two patients underwent delayed sternal closure. The median length of intensive care unit and hospital stays was 4.5 and 13.5 days, respectively. The mean follow-up was 29.3±15.6 months (range, 3 months to 4 years). Only two patients had mild-to-moderate pulmonary conduit stenosis. There was no moderate or severe aortic valve regurgitation in the final follow-up visit.

Conclusion: Our study results show very good pulmonary autograft and pulmonary conduit functions in the mid-term follow-up. Based on these findings, the Ross and Ross-Konno operations remain a good option for aortic valve replacement in infants and children with low mortality rates.

Keywords: Aortic valve stenosis; autograft; congenital heart disease.

ÖZ

Amaç: Bu çalışmada doğuştan aort darlığı olan çocuk hastalarda Ross ve Ross-Konno ameliyatının erken ve orta dönem sonuçları bildirildi.

Çalışma planı: Ocak 2011 - Aralık 2015 tarihleri arasında doğuştan aort darlığı ön tanısı ile Ross veya Ross-Konno ameliyatı yapılan toplam sekiz hasta (4 erkek, 4 kız; medyan yaş 4.5 yıl; dağılım, 3-17 yıl) retrospektif olarak incelendi. Kardiyopulmoner baypas ve kros-klemp süresi ve yoğun bakım ünitesi ve hastanede yatış süresi kaydedildi.

Bulgular: Yedi hastaya daha önce girişim uygulanmıştı [balon valvuloplasti (n=4), ventriküler septal defekt tamiri (n=1), aort kapak tamiri (n=1) ve aort koarktasyon tamiri (n=1)]. Beş hastaya Ross-Konno ameliyatı yapılırken, geri kalan üç hastaya Ross ameliyatı yapıldı. İki hastada ilave subaortik rezeksiyon ve bir hastada mitral kapak tamiri gerekli oldu. Erişkin boyutundaki hastaya Dacron kondüit ile pulmoner otogreft wrapping yapıldı. Mortalite ve majör morbiditeye rastlanmadı. Ortalama kardiyopulmoner baypas ve kross klemp süresi sırasıyla 234±6 ve 177±4 dk idi. İki hastanın sternumu sonradan kapatıldı. Medyan yoğun bakım ünitesi ve hastane kalış süreleri sırasıyla 4.5 ve 13.5 gün idi. Ortalama takip süresi, 29.3±15.6 ay (dağılım, 3 ay - 4 yıl) idi. Yalnızca iki hastada hafif ila orta düzeyde pulmoner kondüit darlığı vardı. Son takip vizitinde hiçbir hastada orta veya ciddi aort kapak yetmezliği görülmedi.

Sonuç: Çalışma bulgularımız, pulmoner otogreft ve pulmoner kondüit fonksiyonlarının orta dönem takipte çok iyi olduğunu göstermektedir. Bu bulgulara dayanarak, bebeklerde ve çocuklarda aort kapak replasmanı için Ross ve Ross-Konno ameliyatları, düşük mortalite oranları ile iyi birer seçenektir.

Anahtar sözcükler: Aort kapak darlığı; otogreft; doğuştan kalp hastalığı.

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The Ross operation can be defined as the use of the autogenous pulmonary valve for aortic valve replacement and reconstruction of the right ventricle outflow tract (RVOT) with a valved conduit.[1,2] It was first performed by Donald Ross in 1967.[1] The management of congenital aortic stenosis during infancy and neonatal period is challenging. For children who require aortic valve replacement, the Ross procedure may be the only option.[2,3] The Ross procedure has some advantages including potential of autograft growth, freedom from anticoagulation, and resistance to infections.[2,3] In patients with left ventricular outflow tract (LVOT) obstruction, particularly in small aortic roots, the Ross procedure can be combined with Konno operation. Despite the advantages of Ross procedure, the long-time durability of pulmonary autograft and pulmonary conduit varies widely among the series.[2] In this study, we present the early and mid-term results of the Ross and Ross-Konno procedures in pediatric patients with congenital aortic stenosis.

PATIENTS AND METHODS

The study protocol was approved by the Acibadem University, School of Medicine Ethics Committee (ATADEK). The study was conducted in accordance with the principles of the Declaration of Helsinki.

This retrospective study included eight patients (4 boys, 4 girls; median age 4.5 years; range, 3 months to 17 years) who underwent Ross or Ross-Konno operation by a single surgeon in two centers (İstanbul Mehmet Akif Ersoy Training and Research Hospital and Acibadem University Atakent Hospital) between January 2011 to December 2015. Four patients were under one year of age. A standard set of perioperative data was collected retrospectively for all patients including medical history and clinical examination, electrocardiography (ECG) and chest X-ray, complete laboratory analyses, and Doppler echocardiography (ECHO) of the heart and valves. We also recorded the data for time of mechanical ventilation, need of inotropic support and need of dialysis, organ failure and other postoperative complications. All patients were contacted by phone calls and their last echocardiographic examination was taken. Primary endpoints were survival and freedom from re-intervention.

Operative technique

Following median sternotomy and full heparinization, cardiopulmonary bypass was commenced with aortic and bicaval cannulation. Under moderate hypothermia, pulmonary artery bifurcation and main pulmonary artery were separated from the aorta with a sharp dissection. After aortic clamping, the ascending aorta was opened with a transverse incision 1 cm above the coronary ostia. Isothermic blood cardioplegia was given directly through the coronary ostia for cardiac arrest. The coronary artery ostia were explanted with large coronary buttons comprising almost the entire wall of the sinus of Valsalva. Aortic valve was, then, resected. The pulmonary artery was divided at the bifurcation level, and the pulmonary annulus diameter was measured using the Hegar's dilators. Infundibulum of the right ventricle was opened 5 to 15 mm below the pulmonary valve and, then, the incision was extended. The pulmonary autograft was prepared. An incision to the interventricular septum was made in patients who needed the Ross-Konno procedure (Figure 1). Pulmonary infundibulum was sutured to the Konno incision during autograft implantation (Figure 2).

Figure 1. An intraoperative view of Konno incision.

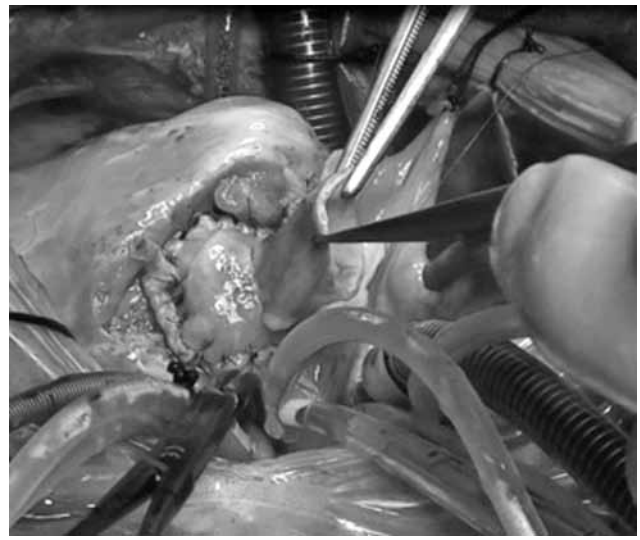

Figure 2. An intraoperative view of autograft implantation during Ross-Konno procedure.

The autograft was, then, sutured to the native aortic annulus with an interrupted 5-0 polypropylene sutures. Three separate pericardial strips were used for buttressing the suture lines. Total root replacement technique was used in all patients. The coronary ostia were anastomosed to the appropriate sinuses of the autograft with a continuous 6-0 polypropylene suture. The proximal neoaorta was, then, sutured to the ascending aorta with a continuous polypropylene suture. The RVOT was reconstructed using a homograft- (n=1) or xenograft-valved conduit (Contegra, Medtronic Inc., Minneapolis, MN, USA) (n=7). All patients were weaned from bypass in the usual manner, and transesophageal echocardiography was performed. Pulmonary autograft-wrapping with a Dacron graft was performed in one patient who had an adult-sized aorta (patient aged 17 years) to prevent delayed dilatation of the neoaorta.

Transthoracic M-mode, two-dimensional, color-flow, and Doppler ECHO were obtained in all patients immediately postoperatively, before discharge, and during follow-up.

Statistical analysis

Statistical analysis was performed using internet based software (https://graphpad.com). Data were collected using the Microsoft Access database. The Pearson chi-square test was used to analyze non-parametric variables, while the Mann-Whitney U test was used to analyze parametric variables. All continuous variables were expressed in mean±standard deviation and categorical variables were expressed in median and range. A p value of <0.05 was considered statistically significant.

RESULTS

The indication for surgery was congenital aortic stenosis in all patients, while aortic regurgitation was coexisted in six patients. Four patients had a bicuspid aortic valve, while one patient had Shone's anomaly. Two patients had also subaortic stenosis. One patient suffered from moderate-to-severe mitral valve regurgitation. Seven patients had prior interventions including balloon aortic valvuloplasty (n=4), ventricular septal defect repair (n=1), aortic valve repair (n=1), and aortic coarctation repair (n=1). One patient needed mechanical ventilation preoperatively. Five patients (62.5%) with LVOT obstruction or aortic annular hypoplasia underwent the Ross-Konno procedure, while the remaining three patients had the Ross operation. An additional subaortic fibromuscular resection was required in two and mitral valve repair in one patient (Table 1).

Table 1. Baseline and perioperative data of study population

Patient no	Age	Previous intervention	Pathology	Operation	RVOT conduit size and type
1	16 years 9 months	-	AS+AR (Bicuspid aorta)	Ross+subaortic ridge resection+wrapping	No: 20 Contegra
2	7 years 3 months	Balloon valvuloplasty	AS+AR (Shone complex)	Ross-Konno+ subaortic ridge resection	No: 21 Pulmonary homograft
3	9 months	Balloon valvuloplasty	AS	Ross	No: 18 Contegra
4	3 months	Balloon valvuloplasty	AS+AR+Subaortic stenosis (bicuspid aorta)	Ross-Konno	No: 16 Contegra
5	10 years 2 months	VSD repair	AS	Ross	No: 20 Contegra
6	14 years 5 months	Aort coarctation repair	AS+AR+Subaortic stenosis (bicuspid aorta)	Ross-Konno	No: 20 Contegra
7	4 months	Balloon valvuloplasty	AS+AR (Bicuspid aorta)	Ross-Konno	No: 18 Contegra
8	1 year 11 months	Aortic valve repair	AS+AR+MR	Ross-Konno+mitral valve repair	No: 19 Contegra

RVOT: Right ventricle outflow tract; AS: Aortic stenosis; AR: Aortic regurgitation, VSD: Ventricular septal defect; MR: Mitral regurgitation.

No mortality or major morbidity was observed in the early postoperative period and during mid-term follow-up. The mean cardiopulmonary bypass and cross-clamp times were 234±64 and 177±38 min, respectively. Ultrafiltration was needed during the operation in three patients. Two patients needed high inotropic support. The mean mechanical ventilation time was 43.7±62.3 hours. The median length of intensive care unit and hospital stays was 4.5 (range, 1 to 23) and 13.5 (range, 5 to 28) days, respectively. Two patients underwent delayed sternal closure. Peritoneal dialysis was required in three patients in the postoperative period. Three patients had pneumonia. Postoperative arrhythmias (left bundle branch block) occurred in one patient. The mean follow-up was 29.3±15.6 months (range, 3 months to 4 years). According to the final ECHO findings during follow-up, two patients had mild-to-moderate pulmonary conduit stenosis (40 to 60 mmHg gradient), while one patient had mild mitral valve regurgitation. Moderate or severe aortic valve regurgitation was not seen in any of the patients in the final follow-up visit. Mild aortic regurgitation was seen in two patients. The neoaortic valve and aortic root grew with the patient, and aortic valve Z scores were in the normal range for all patients. No patient needed re-intervention.

DISCUSSION

Initial intervention for infants and children with congenital aortic stenosis is usually palliation, such as aortic valvotomy, either surgical or interventional.[2,3] The main goal is to achieve clinical improvement, until adult-sized mechanical aortic valve replacement can be done.[3] Although some residual stenosis or insufficiencies can be seen using this approach, many patients may reach adulthood.[3,4] However, some patients, particularly those with aortic annular hypoplasia or multi-level obstruction, need more extensive procedures.[3] Palliation is usually not enough for those patients to relieve the obstruction, even may worsen the pathology causing aortic regurgitation.[3,4] Unfortunately, treatment alternatives are limited in this age group. In recent years, aortic valvuloplasty techniques have been widely adopted. On the other hand, aortic valve repair has many technical challenges, particularly in small infants.[4] The durability of complex repairs are not well-described and reoperation rate is still too high, even in experienced centers.[4,5] In addition, patients with serious LVOT obstruction cannot be treated without the Konno procedure, which makes aortic valve repair impossible. In the present study, five of our patients (62.5%) had at least one initial palliative aortic valvular intervention. One of them had complex aortic repair in another center. Although early ECHO results were satisfactory, this patient suffered from severe aortic regurgitation six months after repair.

There is no ideal prosthetic valve for infants and children. Mechanical aortic valve replacement is far from being the ideal operation, even if it is possible to perform in selected cases with a large-sized aortic annulus. Although mechanical valves are durable, they have many disadvantages including the necessity for life-long anticoagulation, risks of endocarditis, bleeding, and thromboembolic events.[6,7] Even reoperation is required in at least 10% of mechanical valves by 20 years for endocarditis, paravalvular leak, thrombotic, or hemorrhagic complications or pannus formation with obstruction. Mechanical valves are also more problematic in female patients in childbearing age who need aortic valve replacement, leading to severe social and psychological distress.[6,7] Although bioprosthetic valves do not require anticoagulation, their long-time durability is not optimal in young patients.[4] The ideal valve substitute in children, therefore, should have somatic growth, no need for anticoagulation with excellent hemodynamic properties, resistance to infection, and long-term durability.[8] Besides, its implantation technique may be reproducible with low mortality and morbidity rates. To date, pulmonary autograft (the Ross) operation is still the only alternative which seems to fulfill the aforementioned criteria.[9,10] However, the major drawback of the Ross operation is the invasive nature of the operation, which jeopardize two valves for a single outflow tract disease.[11] In addition, extensiveness of the procedure necessitates experience. There have been also some concerns on the long-term performance of the Ross operation. Some reports describe dilation of the aortic annulus, sinus of Valsalva, and sinotubular junction, causing an aortic aneurysm and aortic valvular regurgitation, particularly in adults.[12-14] In those studies, the dilation of the sinotubular junction, which distracts the leaflets, preventing coaptation, is thought to be the primary pathology.[14,15] We did not observe any aortic annulus or sinotubular junction dilation in our patients in the mid-term follow-up. All autografts were in appropriate size to their ages. Another important concern is the durability of the pulmonary conduit. Original series of Donald Ross showed 80% freedom from re-operation for the right side at 20 years.[16] Pulmonary homografts are considered the optimal alternative for pulmonic reconstruction.[8,10,11] The orthotopic position of the conduit may also increase its longevity.[3] Since the availability of the homografts is limited in Turkey,

we used the Contegra conduit in the majority of our patients thanks to its easy-to-use and readily available nature. Several studies comparing the homografts with this conduit have also demonstrated similar results.[14]

Interventional methods are also effective for the treatment of conduit failure. Balloon dilation, stent implantation and percutaneous pulmonary valve implantation have a great potential for reducing the rate of conduit reoperation.[17] Our policy is to use the largest possible conduit for RVOT reconstruction at the time of the operation to prevent early conduit failure. In children with aortic valve disease associated with annular hypoplasia or LVOT obstruction, the Ross procedure, combined with the Konno-type aortoventriculoplasty, may be the only choice.[3] In our study, five patients underwent the Ross-Konno operation. Surgeons have reported this operation in neonates and infants with an increasing frequency in the recent years.[18] Although the Ross operation was recommended in children and young adults earlier, the indications are increasingly expanding in all age groups currently. In addition, some reports have shown that the Ross operation in infants has a higher mortality and should be delayed, until the child reaches the late infancy;[19] however, we found that this operation was a safe and effective method for aortic valve replacement during infancy and childhood. In the present study, three patients were under one year old and no mortality was observed. Neonatal age, congenital aortic arch defects, mitral regurgitation, and preoperative severe hemodynamic instability are the main baseline factors associated with high mortality rates.[2,12,20] In this study, one patient who underwent additional mitral valve repair had mild mitral valve regurgitation. In our opinion, associated mitral valve disease is not a contraindication for the Ross operation, provided that a durable mitral valve repair is feasible. In a meta-analysis, the rate of autograft deterioration with the need for reoperation in pediatric patients ranged from 0.24 to 2.82% per patient-year with a pooled mean of 1.38% per patient-year.[13] For an ideal Ross operation, diameter of the pulmonary and aortic annuluses should be similar. If the diameter of the aortic annulus is 3 mm larger than the pulmonary annulus, it may affect the competence of the pulmonary autograft. Of note, one of the most common indications for reintervention is autograft dilatation, particularly in adults. On the basis of these data, we performed autograft wrapping using a Dacron graft in one patient who was 17 years old to prevent late dilatation of the neoaorta. In a recent study, the authors used a Hemashield graft for wrapping in 52 patients aged older than 10 years and five year freedom from reintervention rate on LVOT improved from 81 to 91%.[14] In our study, the total root replacement technique was used in all patients, and in some cases, an additional reinforcement of the aortic annulus with a strip of autologous pericardium. We believe this technique might help preventing annular dilatation preserving the somatic growth.[21] Some authors have reported a 10-year freedom from significant neoaortic regurgitation ranging from 75 to 90% following the Ross procedure with re-intervention rates of up to 10%.[17] In our study, we observed no moderate or severe aortic regurgitation. Furthermore, several studies have suggested that early postoperative dilation of the neoaortic root is stabilized over time.[22] Pulmonary autograft preparation should be taken into consideration, and septal artery should be protected during incision. Coronary re-implantation following the enlargement of the LVOT is also an important issue. Malposition of a coronary button may result coronary flow obstruction and cause serious problems.[23] However, we observed no such complications in our patient population. Considering that there is no operative mistake, the autograft is a permanent valve and will be subjected to the normal aging process. The neoaortic annulus and root will also increase in size proportionately to the somatic growth.

In conclusion, our study shows that the Ross and Ross-Konno procedures may be safely accomplished with very low morbidity and mortality rates and excellent mid-term outcomes. Of note, the Ross operation may be the choice of procedure in infants and children, when palliative interventions fail.

Declaration of conflicting interests

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