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Case Report

# *First Successful Yasui Operation in Thailand with Early and Mid Term Results*

**Sonthakit Leelahanon***
**Kornprom Saengaram**†
**Tanarat Layangool**‡
**Vorakarn Promphan**‡

**Pediatric Cardiovascular-thoracic Surgeon, Division of Cardiovascular-thoracic Surgery, Department of Surgery, Queen Sirikit National Institute of Child Health, Bangkok, Thailand*
*†Invited Cardiovascular-thoracic Surgeon & Airbus 330 Captain at Thai Airways International Public Company Limited, Vibhavadi, Jatujak, Bangkok, Thailand*
*‡Pediatric Cardiologist, Division of Cardiology, Department of Pediatric, Queen Sirikit National Institute of Child Health, Bangkok, Thailand*

*Abstract* **The Yasui operation is a complex biventricular repair for a patient with adequately sized ventricles, ventricular septal defect, severe hypoplastic or atretic aortic valve and severe hypoplastic or interrupted aortic arch. This operation is rarely performed due to its complexity and rarity of cases. We report the first successful Yasui operation in Thailand which was performed on 21st April 2010. Early to mid term results and a review of the literature are presented in this report. In conclusion, one stage primary Yasui operation in neonate can be performed safely with good results.**

## Introduction

Yasui operation is an operation for neonates who have adequately sized ventricles, severe hypoplastic aortic valve or aortic atresia, hypoplastic ascending aorta, hypoplastic or interrupted aortic arch, ductal dependent systemic circulation and a ventricular septal defect (VSD). In the past, this group of patients was recognized as one of the variants of hypoplastic left heart syndrome (HLHS)[1] due to the similarity of the lesions and the similarity of treatment by a palliative Norwood operation. At the present, the treatment of choice for this group of patients has changed to a more definitive biventricular repair such as Yasui or Ross/Konno operation. Due to atypical lesions, an adequately sized left ventricle (LV), a presentation of VSD and different management, this group of patients categorized as HLHS variant was reclassified into a critical left ventricular outflow tract obstruction (LVOTO) group. In this study, we report our first successful Yasui operation in Thailand with early to mid term results.

**Correspondence address:** *Sonthakit Leelahanon, MD, Division of Cardiovascular-thoracic Surgery, Department of Surgery, Queen Sirikit National Institute of Child Health, 420/8 Rajathevi Road, Bangkok 10400, Thailand; Telephone:/Fax: +66 2354 8095; E-mail: sonthakit@gmail.com*

## Case Report

A 16-day-old girl, presented with cyanosis and in coma. She was admitted and treated at a local hospital. Physical examination revealed high fever, dyspnea, and central cyanosis with crepitation in both lungs. A systolic ejection murmur was heard at the left parasternal border. Hepatomegaly was also found. Chest X-ray revealed cardiomegaly, increased vasculature and infiltration of both lungs. EKG revealed right atrial enlargement and biventricular hypertrophy. Complete blood count showed leukocytosis with lymphocytic predominance. She was initially treated at a local hospital for pneumonia, congestive heart failure and septicemia by intubation, diuretics and antibiotics administration. After three days, her clinical condition improved and she was able to be extubated. Her hemodynamics had also improved and she was transferred to our hospital at the age of 23 days.

Physical examination at our hospital showed a normal body temperature, her heart rate was 170/min, blood pressure was 69/33 mmHg and her respiratory rate was 55/min. Body weight was 3,000 gram (birth weight 3,200 gram) and height was 51 cm. Central cyanosis was found with oxygen saturation about 80%. There was no differential cyanosis. Blood pressure was not significantly different in all extremities. An active precordium was found with systolic ejection murmur grade 3 at the left parasternal border. Chest sounds were clear. The liver edge was 2 cm below right costal margin. Other examinations were unremarkable. Chest X-ray showed cardiomegaly and increased vasculature (Figure 1). Echocardiogram showed a small patent foramen ovale (PFO) with left to right shunt, large subpulmonic VSD (Figure 2), normal size LV, severe hypoplastic aortic valve (4 mm, z-score -5.04) (Figure 3), severe hypoplastic ascending aorta (5 mm), severe hypoplastic aortic arch (3.2 mm) and severe hypoplastic aortic isthmus (2.7 mm) (Figure 4). Ductal dependent systemic circulation was found from a large patent ductus arteriosus (PDA) of 6 mm. The descending aorta was normal size (8 mm). Pulmonary valve, pulmonary artery and pulmonary branches were normal. Echocardiographic diagnosis for this patient was a HLHS variant.

The operation was performed on 21$^{st}$ April 2010 at the age of 36 days. The intra-operative findings were the same as the echocardiographic findings except that there was an aberrant right subclavian artery. Cardiopulmonary bypass was accomplished by placing two aortic cannulas in the right common carotid artery (via PTFE graft) and main pulmonary artery (MPA). The MPA cannula was passed into the descending aorta via the PDA connection. A venous cannula was placed into the right atrium. Cardiac arrest was achieved by blood cardioplegia. The right ventricular outflow tract (RVOT) was opened and a Dacron patch was used as an intracardiac baffle to redirect blood from the LV through the VSD and to the pulmonary valve (making double outlet left ventricle). Pulmonary artery bifurcation was cut off from main pulmonary artery and anastomosed to a Bovine jugular venous valve conduit (Contegra$^{TM}$) size number 14. Under hypothermic circulatory arrest with antegrade cerebral perfusion from right common carotid artery, the patent foramen ovale was closed. The ascending aorta and aortic arch were anastomosed to the MPA by a side-by-side technique with a xenograft patch augmentation (Damus-Kaye-Stansel anastomosis). Total circulatory arrest time with antegrade cerebral perfusion was 75 minutes. During the rewarming and with the heart beating, a proximal anastomosis of the valve conduit to RVOT was performed. Total cardiopulmonary bypass time was 341 minutes and the aortic cross clamp time was 151 minutes. The operative picture explanation is shown in Figure 5.

Her recovery was uneventful. She could be extubated on the sixth day after her operation. Postoperative echocardiogram showed no residual VSD, no right nor left ventricular outflow tract obstruction (Figure 6). Her native pulmonary valve showed mild regurgitation and the conduit valve showed mild to moderate regurgitation (Figure 7). No stenosis of neo-ascending, aortic arch and descending aorta were found (Figure 8). Ventricular function was good and she was discharged from hospital on the 19$^{th}$ day after her operation.

Follow up at the 35$^{th}$ day after the operation; she was in good health with New York Heart Association (NYHA) classification class 1. Her body weight had increased to 4 kg. A chest X-ray showed mild cardiomegaly with normal pulmonary vasculature (Figure 9).

At 3 years and 4 months old, she was in very good health without any symptoms. Her body weight was 12 kg and height was 96 cm. Routine follow up echocardiogram showed a new supravalvular pulmonic

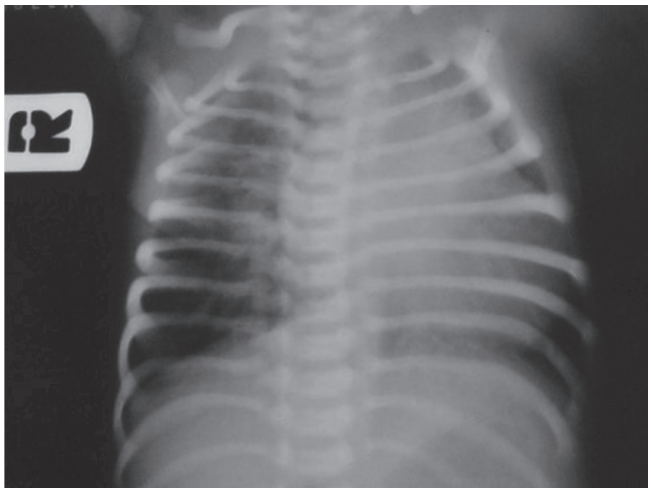


**Figure 1** Preoperative chest x-ray

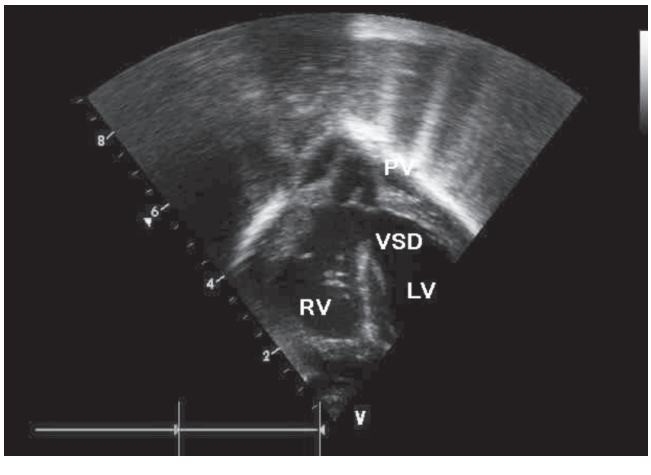



**Figure 2** Preoperative echocardiogram showing subpulmonic VSD

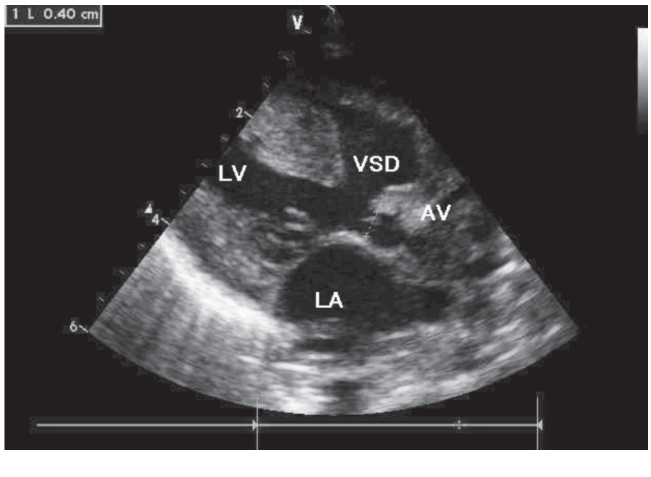



**Figure 3** Preoperative echocardiogram showing a small aortic valve

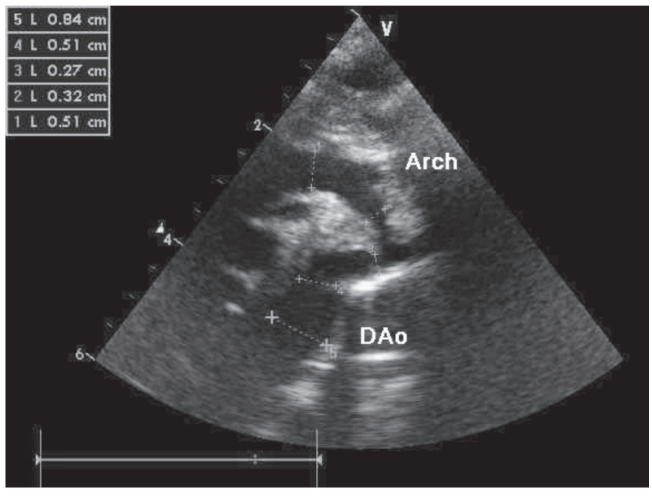



**Figure 4** Preoperative echocardiogram showing hypoplastic ascending aorta and arch

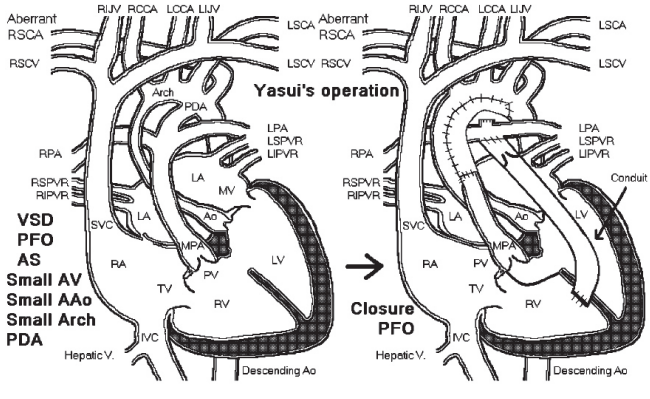



**Figure 5** Operative picture explanation of our case

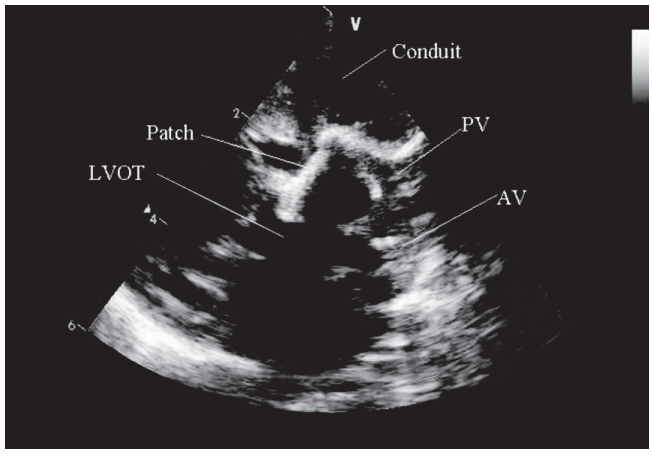



**Figure 6** Early postoperative echocardiogram showing no LVOTO

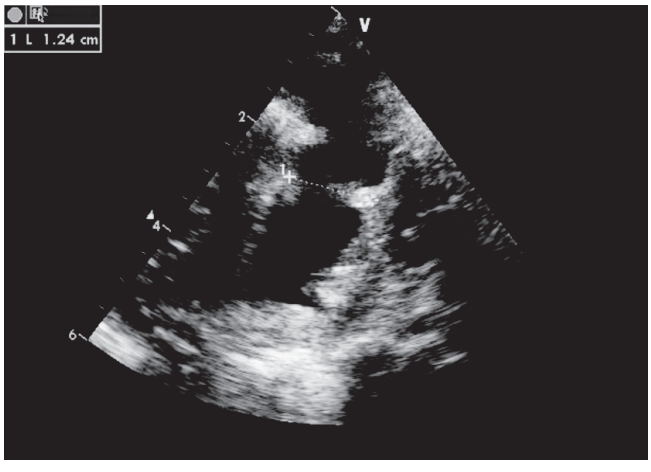



**Figure 7** Early postoperative echocardiogram showing no conduit stenosis

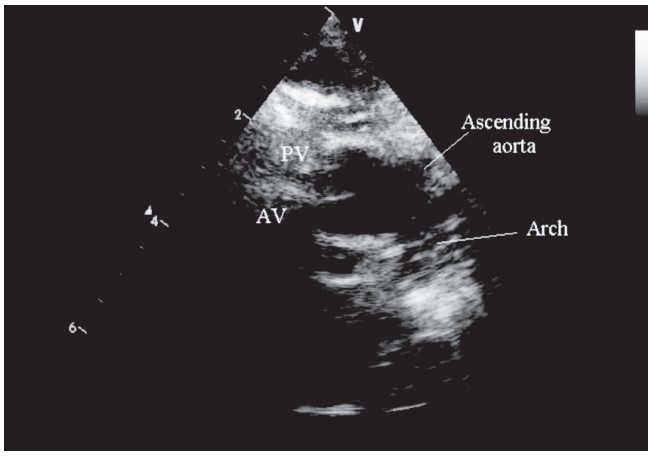



**Figure 8** Early postoperative echocardiogram showing good size of neo-ascending aorta and arch

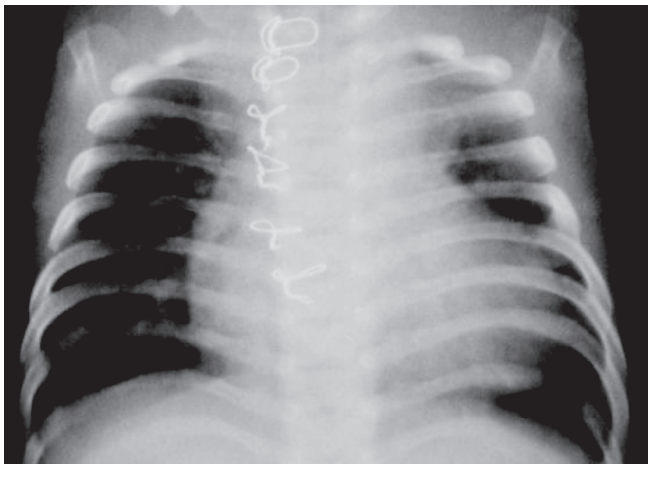


**Figure 9** Early result - Chest X-ray

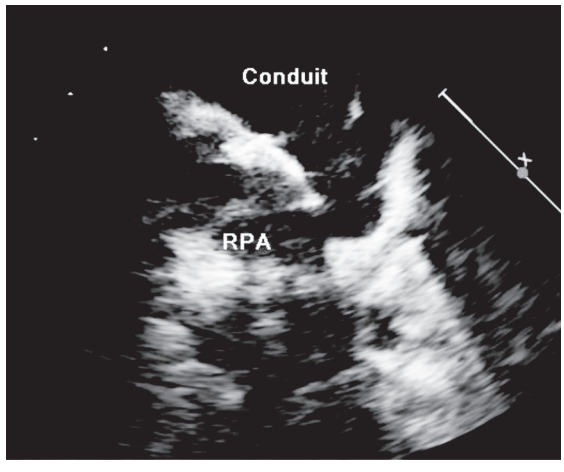



**Figure 10** Mid-term follow up echocardiogram showing stenosis of conduit-RPA junction

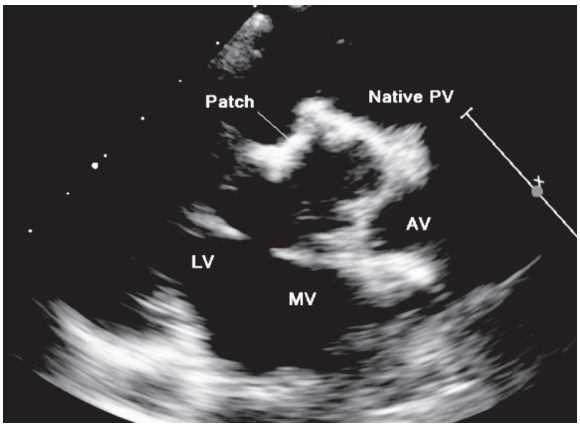



**Figure 11** Mid-term follow up echocardiogram showing no LVOTO

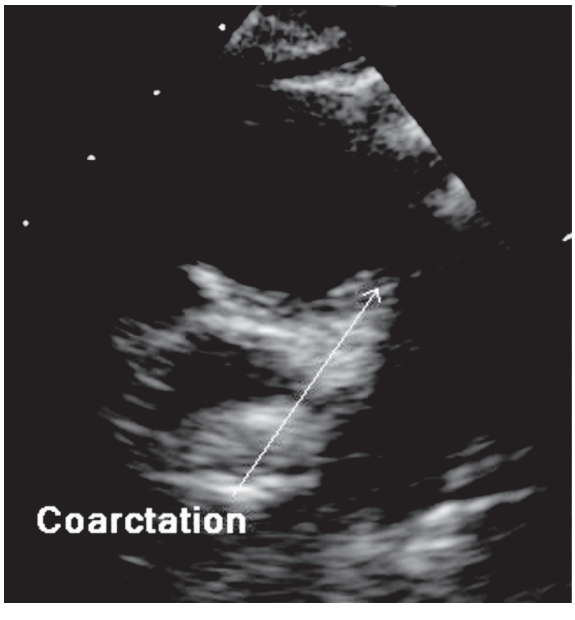



**Figure 12** Mid-term follow up echocardiogram showing new coarctation of aorta

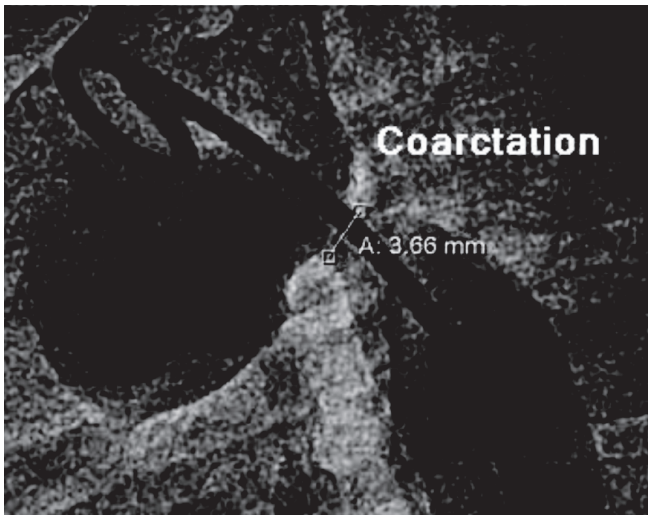



**Figure 13** Angiogram showing recurrent coarctation of aorta

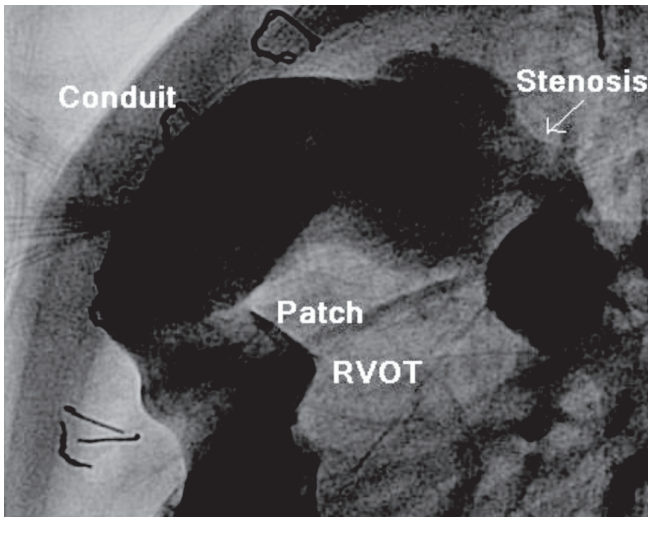



**Figure 14** Angiogram showing new stenosis of conduit-RPA and conduit-LPA junction

stenosis (supravalvular PS) and a new coarctation of aorta. The RVOT-conduit junction was normal. The stenotic part was at conduit-right pulmonary artery (RPA) junction with a gradient of 40 mmHg (Figure 10). Her native pulmonary valve showed mild regurgitation and the conduit valve showed moderate regurgitation similar to the early postoperative echo findings. The native pulmonary valve and VSD seemed to have grown proportionately to the body size (Figure 11). A new coarctation was found at the patch-descending aorta junction with a gradient of 60 mmHg (Figure 12).

At 3 years and 11 months old, she was admitted for interventional cardiac catheterization. An angiogram showed recurrent coarctation of the aorta at the patch-descending aorta junction (Figure 13) and stenosis at the conduit-RPA and conduit-left pulmonary artery (LPA) junction (Figure 14). Balloon angioplasties were carried out and good results were obtained. The gradient across the conduit-RPA junction decreased from 43 mmHg to 28 mmHg. The gradient across the conduit-LPA junction decreased from 42 mmHg to 29 mmHg and the gradient across arch-descending aorta junction decreased from 39 mmHg to 22 mmHg. No complication was found.

At 4 and half years old, she was still in good health with NYHA class 1. Her body weight was 14 kg and height was 104 cm. She will be scheduled to change the conduit to an adult size and repair any residual defects when the body weight reaches 15-20 kg.

## Discussion

HLHS includes a wide spectrum of cardiac malformations with varying degrees of underdevelopment of the structures of the left heart and aorta. At one end of the spectrum is complete atresia of aortic and mitral valves and a diminutive LV incapable of supporting the systemic circulation. At the other end of the spectrum is a milder form of aortic stenosis and mitral stenosis with a reasonable sized LV[2]. In the past, when cardiac lesions did not have all the characteristics of typical HLHS but had the same clinical presentation and treatment, some surgeons and cardiologists called it a HLHS variant[1]. Some describe HLHS variants as HLHS that have hypoplastic LV but may be of sufficient size that a biventricular repair could be possible[3]. For a typical HLHS, when ventricular septum is intact, the LV is markedly hypoplastic or absent. But in approximately 3% of patients with aortic atresia, there is an associated unrestrictive VSD and almost always there is normal LV and mitral valve development[4].

In 1981, Norwood and colleagues[5] reported successful operations for HLHS. In 1986, Bailey LL and associates[6] reported a successful orthotopic cardiac transplantation as a management strategy for neonates with HLHS. This was a strategy that could easily be applied to neonates with critical aortic stenosis and other left-sided cardiac abnormalities[7]. As outcomes with surgery remained marginal, and as the morphological variability in the left-sided cardiac

structures of patients with critical LVOTO became more evident, the idea of abandoning a biventricular repair in favor of a single-ventricle palliation became less attractive. In 1987, Yasui and coworkers[8] reported a successful correction of critical aortic stenosis, interrupted aortic arch and VSD in 2 neonates. They used another operative approach that achieved a biventricular repair by a Damus-Kaye-Stansel anastomosis between the ascending aorta and main pulmonary artery with a Rastelli-type intracardiac tunnel and an extracardiac right ventricle to pulmonary artery (RV-PA) conduit. Their approach could be used only if a VSD was present. In 1995, Calhoon JH and Bolton JW[9] reported successful anterior aortoventriculoplasty with the pulmonary autograft or Ross/Konno operation for critical aortic stenosis and complex LVOTO with adequate ventricular function and size, in two neonates.

The degree of LVOTO, the size of the aortic valve, and co-existing malformation such as a VSD typically determines whether neonates with critical LVOTO can be managed with univentricular palliation, biventricular repair or cardiac transplantation. Univentricular repair includes Norwood and other palliative repairs followed by a single-ventricle pathway. Biventricular repairs predominantly include Yasui operation, Ross/Konno operation, and rarely allograft aortic valve replacement[10]. Biventricular repairs can be done in one stage definitive repair or be done in two stages repair after initial palliative operation.

Initial palliation for the staged biventricular repair involves a Norwood aortic arch reconstruction as with a primary repair. Pulmonary blood flow is provided by a modified Blalock-Taussig shunt or a RV-PA shunt (Sano shunt). The eventual biventricular repair in the staged patients channels the flow through the VSD to the pulmonary valve with or without enlargement of the VSD, creation of RV-PA continuity with a valved conduit, and takedown of the shunt[11]. The disadvantages of this staged operation is the existence of a parallel circulation with its resultant volume overload, and the assumption of risks associated with the shunted single ventricle circulation that results in inter-stage mortality[2].

Ross operation was originally described in young adults as an alterative to bioprosthetic, mechanical or allograft aortic valve replacement. A pulmonary autograft is harvested and translocated to the aortic position, and the RVOT is then reconstructed using a valved conduit. Ross operations have since been described in young children, neonates[9] and even in a premature baby[12] with LVOTO. In this scenario, an annular enlargement operation is frequently necessary. The most common approach is to undertake an anterior aortoventriculoplasty (Ross-Konno operation, Figure 15).

Yasui operation was originally described as a strategy to repair neonates with aortic atresia and a VSD[8] in patient subset whose LV function and size is

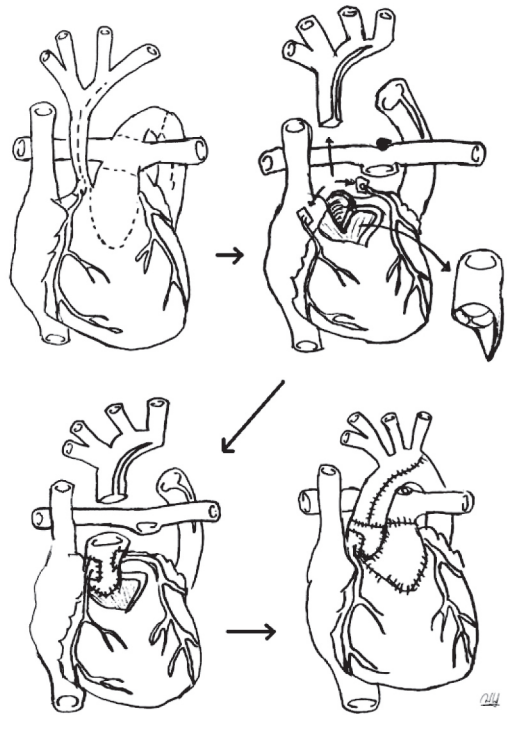


**Figure 15** The Ross/Konno operation[9]

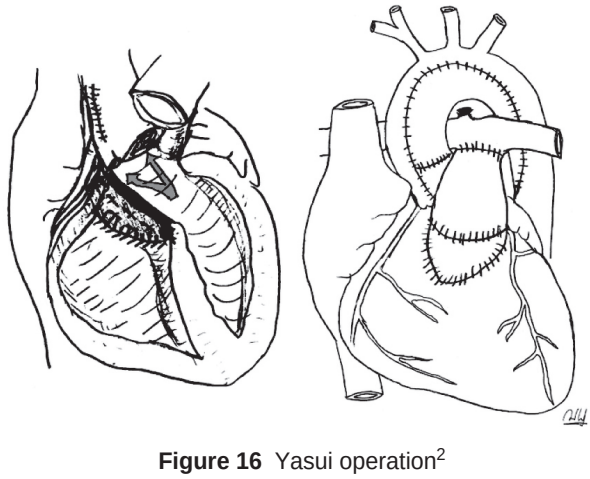


**Figure 16** Yasui operation[2]

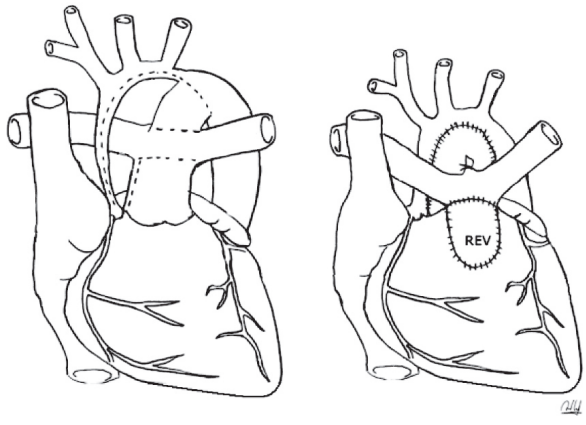



**Figure 17** Modified Yasui operation[13]

often adequate to support a systemic circulation. The repair consisted of tunneling the VSD to the pulmonary valve, reconstructing the aortic arch with a Norwood-type homograft patch, repair of the interrupted aortic arch if present and the creation of RV-PA continuity with a valved conduit (the Rastelli operation) (see Figure 16).

A modification of Yasui operation was reported by Black MD and colleagues[13]. They reported two cases of modified Yasui operation by using Réparation à l'Etage Ventriculaire (REV-procedure) and Lecompte maneuver to avoid a prosthetic or homograft conduit (Figure 17). The first patient was in good condition for 13 months after operation. The other patient required reoperation for recurrent right ventricular outflow tract obstruction (RVOTO) at 10 months after the initial operation and was still doing well at 19 months. Uemura H and colleagues[14] also reported 3 neonatal cases of modified Yasui operation with excellent results without progressive lesions or reoperation up to 60 months.

The Yasui operation seems to have better result than the Norwood operation but the difference is difficult to interpret due to the differences in the patient groups. Carrillo SA and colleagues[15] compared the results of the Yasui and Norwood operations at Lucile Packard Children's Hospital between 2004 and 2013. Eighteen patients underwent the Yasui operation, of whom 15 had a one-stage operation and 3 had a two-stage operation. During this time frame, 113 patients underwent a Norwood operation. The one stage Yasui operation had a significantly lower operative mortality compared with the Norwood operation (6.7% versus 16%). Early and midterm survival was also higher in the Yasui group (85% versus 62% at 1 year, 85% versus 60% at 3 years, and 85% versus 58% at 5 years). They concluded that Yasui operation should be pursued when the anatomy is amenable. Moorthy PS and colleagues[16] reviewed 16 patients who underwent a primary Yasui operation at Birmingham Children's Hospital between 1990 and 2005, and compared with the outcomes of the Norwood procedure over the same period. Yasui operation has an early mortality of 19% (3 patients) with no deaths in the last 12 years (13 patients). Actuarial survival at one and five years was 60% and 53%, respectively. This was comparable to the early mortality of 29% ($p < 0.01$) and actuarial survival of 58% and 50% in the Norwood group. Their conclusion supported the strategy to aim for a primary biventricular repair wherever possible.

Neonates without a VSD are unable to undergo Yasui operation and neonates with aortic atresia are unlikely to be considered candidates for Ross operation[10]. Some patients may have a hypoplastic aortic valve and a small VSD and could be candidates for both Yasui and Ross/Konno operations. There is still no study to compare these two operations in the same groups of patients but Yasui operation seems to be more attractive due to being less extensive and generally has a better outcome. Hickey EJ and colleagues[10] reviewed 1217 neonates presenting with critical LVOTO enrolled in the Congenital Heart Surgeons Society study between 1994 and 2008. The Ross operation (N = 39) was rarely the primary intervention (5/39, 13%). Konno aortoventriculoplasty was performed in the majority of the cases (26/39, 67%). Twenty five patients (64%) were operated to treat iatrogenic aortic regurgitation complications after other previous interventions and four of them were considered emergencies. Infants less than 3 months old had a worse late outcome with a 5-year survival of only 44 ± 10%. Infants more than 3 months had a 5 year survival rate of 76 ± 8%. Yasui operation (N=13) was usually the primary operation (9/13) but occasionally followed Norwood palliation (4/13). None were an emergency. All had a VSD. Survival was better with a 10 years survival rate of 69 ± 13%.

There is still no conclusion about aortic valve size or z-score that is favorable to doing a standard conventional biventricular repair over the Yasui repair. Kanter KR and colleagues[11] suggested that Yasui

operation should be performed on neonates in whom the smallest diameter of the left ventricular outflow tract (LVOT) (either at the valve or the subaortic region) is less than 4.0 mm. For those with a minimum value between 4.0 and 4.5 mm, they would consider either a conventional repair or a Yasui, and for those patients with a value greater than 4.5 mm, they would tend to perform a standard biventricular repair. Ohye RG and colleagues[17] suggested that an aortic valve anulus z-value of -2 or less is considered indicative of a high risk of significant residual LVOTO after standard biventricular repair. Hirata Yand colleagues[18] suggested that preoperative measurement of the difference between the aortic annulus and the patient's weight predicted the need for further reoperation for LVOT. When the aortic annulus is larger than patient's weight + 1.5 mm, excellent survival is expected without further reoperation. If the aortic annulus is between body weight + 1.5 mm and body weight + 1.0 mm, excellent survival is expected although they frequently required reoperation for LVOTO. When the aortic annulus is equal or less than body weight + 1.0 mm, an LVOT bypass operation such as the Yasui operation is recommended.

Although the Yasui operation is now well-established, controversy remains as to whether this should be performed in one stage or in two stages with an initial palliative procedure[16]. Each technique has inherent strengths and weaknesses. An initial Norwood procedure as a part of staged repair carries a shorter ischemic time, no ventriculotomy (if a Sano shunt is not used) and avoids the placement of a RV-PA valved conduit in these small neonates. However, the postoperative course may be more labile because of the shunt-dependent pulmonary blood flow, volume loaded and cyanotic circulation. Nevertheless, the procedure is attractive as it does not preclude from conversion to a biventricular repair at any stage in the future[16]. Ohye RG and colleagues[17] reviewed their experience with primary and staged Yasui operations in 20 infants between 1991 and 1999. Among the 9 patients who underwent staged repair, there were no deaths after the Norwood procedure and 1 death after the biventricular repair. For the 11 patients who underwent primary biventricular repair, there was 1 early death and 2 late deaths from noncardiac causes. Actuarial survival was not significantly different between staged repair (89%) and primary repair (73%). Although their data supported either a primary or staged approach, the authors recommended primary biventricular repair. Kanter KR and colleagues[11] reported 29 cases of Yasui operation at Atlanta, Georgia between 2002 and 2011. Six underwent a primary Yasui repair, 15 were staged with an initial Norwood repair followed by a Yasui completion in 13 (2 await completion). Early mortality was zero with no inter-stage death in the staged patients. One patient died 2 months after staged repair. There was no statistically significance in the survival between the patients with a primary Yasui operation and those with a staged repair. The authors concluded that primary and staged repair have comparable results.

Other results of Yasui operation have been reported from many centers. Nathan M and colleagues[2] reported 17 Yasui operations in neonates at the Children's Hospital, Boston between 1986 and 2002. Median follow-up was 6 years. Three of the 17 (17.6%) died within 30 days. There have been no deaths in this series since 1992. Nine patients (38.9%) required one reoperation, 7 of which were for conduit stenosis, 1 for LVOTO, and 1 for residual VSD with a left ventricle to right atrium shunt. Freedom from death at 10 years was 82% by Kaplan-Meier's estimate.

Gruber PJ and colleagues[19] reported 21 neonates who underwent a Yasui operation at the Children's Hospital of Philadelphia between 1995 and 2005. All patients with aortic stenosis had annular diameters of 3 mm or smaller. There were no operative or in-hospital deaths. No patients required extracorporeal membrane oxygenation support. But more than half of the patient (11/21) had baffle leaks which required reoperation in four of them. The authors could not identify a consistent location of these defects, nor was there a consistent correlation with VSD enlargement. They noted that their patches were completed in a running fashion. They could not exclude the possibility that an interrupted approach may be more secure. On median follow-up of 34.2 months, there was 1 late death and half of the survivors (10/20) required at least 1 reoperation.

Nakano T and colleagues[20] reported 17 patients who underwent a Yasui operation at Fukuoka Children's Hospital, Japan between 1985 and 2014. The minimum diameter of the left ventricular outflow tract (LVOT) and the z-score in patients with aortic stenosis were 3.7 ± 0.4 mm and -9.2 ± 1.2 respectively. Primary repair was

performed in 6 patients, and 11 patients were staged, with bilateral pulmonary artery banding (PAB) in 8 patients, arch repair with PAB in 2 and Norwood operation in 1 patient. The mean age and body weight at the time of the Yasui operation was 4.7 ± 5.3 months and 4.5 ± 1.8 kg, respectively. VSD was enlarged in 6 patients. RV-PA continuity was established with a valved conduit in 14 patients, the Lecompte maneuver in 2 patients and another method in 1 patient. The mean duration of the follow-up was 7.6 ± 9.2 years. There was one early death due to a myocardial infarction and one late death due to a non-cardiac cause. The actuarial survival at 10 years was 87.8%. Six patients underwent reoperation, including five conduit exchanges, two LVOT repairs and two aortic arch repairs. The freedom from reoperation for all causes at 5 and 10 years were 71.3 and 28.5%, respectively.

There is a concern about late recurrent LVOTO from a restrictive VSD when the patients grow. Takabayashi S and colleagues[21] reported a long term outcome of LVOT after a Yasui operation in two neonatal cases. The first patient was followed up for 16 years and the second patient for 10 years. Their results showed no progressive LVOT stenosis, as was indicated by linear growth in each site of DKS, VSD, and the subaortic stenosis. There was no stenosis of the intracardiac baffle over long-term follow-up. No pressure gradient across the reconstructed LVOT and preserved semilunar valve function indicated that their primary biventricular repairs, which added an LVOT route, resulted in excellent outcomes.

## Conclusions

One stage primary Yasui operation in neonates can be performed safely with good results.

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