

Twenty-five years' experience of modified Lecompte procedure for the anomalies of ventriculoarterial connection with ventricular septal defect and pulmonary stenosis

Hong-Gook Lim, MD, PhD, Woong-Han Kim, MD, PhD, Jeong Ryul Lee, MD, PhD, and Yong Jin Kim, MD, PhD

Objectives: To overcome the drawbacks of the Rastelli operation, the modified Lecompte procedure creates a connection from the left ventricle to the aorta with greater freedom from residual obstruction due to the resection of the outlet septum and avoids the implantation of an extracardiac valved conduit. We evaluated the effectiveness of this technique with analysis of our 25-year long-term results.

Methods: We reviewed the records of 50 patients who underwent the modified Lecompte procedure during the past 25 years. The median age at operation was 1.95 years (range, 0.30-12.48 years). The diagnoses involved anomalies of the ventriculoarterial connection with ventricular septal defect and pulmonary outflow tract obstruction, such as transposition of the great arteries, double outlet right ventricle, and double outlet left ventricle.

Results: There were 2 early deaths (4.0%). During a mean follow-up of 14.2 ± 7.9 years (range, 0.2-25 years), there were 4 late deaths. Actuarial survival was $87.3\% \pm 4.9\%$ at 25 years. The freedom from arrhythmia and reoperation at 25 years was $87.7\% \pm 6.2\%$ and $25.2\% \pm 9.4\%$, respectively. The freedom from reoperation for left ventricular outflow tract obstruction and right ventricular outflow tract obstruction at 25 years was $88.5\% \pm 5.4\%$ and $49.6\% \pm 9.0\%$, respectively. At last follow-up, 43 survivors (97.7%) are in New York Heart Association class I.

Conclusions: The modified Lecompte procedure has excellent long-term results for treating anomalies of ventriculoarterial connection with ventricular septal defect and pulmonary outflow tract obstruction. Early repair is possible with low mortality and morbidity in terms of arrhythmia, reoperation for right or left ventricular outflow tract obstruction, and functional class. (*J Thorac Cardiovasc Surg* 2014;148:825-31)

Anomalies of ventriculoarterial connection with ventricular septal defect (VSD) and pulmonary outflow tract obstruction, such as transposition of the great arteries (TGA), double-outlet right ventricle (RV), double outlet left ventricle (LV), and Taussig-Bing anomaly, have a wide variety of spectrums, and several operative techniques have been performed according to diverse anatomic characteristics without standard operative selection guidelines.¹⁻⁴ The Rastelli operation had been the gold standard for many years,¹ but long-term results have been disappointing.⁵⁻⁷ As an alternative to the Rastelli operation, aortic root

translocation with right ventricular outflow tract (RVOT) reconstruction has been performed, resulting in a more normal anatomic repair. This approach potentially causes several morbidities, such as coronary distortion, aortic regurgitation (AR), arrhythmia, and recurrent RVOT obstruction, and still lacks long-term experience.⁸⁻¹¹

To overcome the drawbacks of the Rastelli operation, the Lecompte procedure creates a connection from the LV to the aorta with greater freedom from residual obstruction because of the resection of the outlet septum, which avoids the spiral shape of the LV-to-aorta tunnel. The pulmonary trunk is directly reimplemented on the RV instead of the implantation of an extracardiac valved conduit.² We modified the Lecompte procedure without applying the Lecompte maneuver. We reported early and midterm results for the modified Lecompte procedure,¹²⁻¹⁴ which was also compared with the Rastelli procedure,¹⁴ but its long-term results have not been reported.¹⁵

We started to apply the modified Lecompte procedure in 1988. In this study, we aimed to focus primarily on the long-term results of the modified Lecompte procedure for optimal operative selection, which are not clear, unlike with the Rastelli operation.

From the Department of Thoracic and Cardiovascular Surgery, Seoul National University Hospital, Seoul, Republic of Korea.

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Address for reprints: Yong Jin Kim, MD, PhD, Department of Thoracic and Cardiovascular Surgery, Seoul National University Hospital, 101 Daehak-ro, Jongno-gu, Seoul 110-744, Republic of Korea (E-mail: kyj@plaza.snu.ac.kr).

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Abbreviations and Acronyms

AR	= aortic regurgitation
LV	= left ventricle
LVOT	= left ventricular outflow tract
NYHA	= New York Heart Association
PR	= pulmonary regurgitation
REV	= réparation à l'étage ventriculaire
RV	= right ventricle
RVOT	= right ventricular outflow tract
TGA	= transposition of the great arteries
VSD	= ventricular septal defect

METHODS**Study Population**

We reviewed the records of 50 patients who underwent the modified Lecompte procedure during the past 25 years. This study was approved by the institutional review board of Seoul National University Hospital (1401-022-547). We started to perform the modified Lecompte procedure in 1988, and 50 patients underwent complete repair with the Lecompte modification of the réparation à l'étage ventriculaire (REV) procedure. The male-to-female ratio was 27:23, and the median age at operation was 1.95 years (range, 0.30-12.48 years). Twenty-six patients (52%) were aged less than 2 years, and 10 patients (20%) were aged less than 1 year. The median weight at operation was 10.1 kg (range, 5.7-35 kg).

Preoperative diagnosis included TGA with VSD and pulmonary stenosis (or pulmonary atresia) in 33 patients (66%), double outlet RV with VSD and pulmonary stenosis (or pulmonary atresia) in 15 patients (30%), TGA with endocardial cushion defect and pulmonary stenosis in 1 patient (2%), and double outlet LV with VSD and pulmonary stenosis in 1 patient (2%). Associated anomalies included atrial septal defect ($n = 31$), patent ductus arteriosus ($n = 30$), juxtaposed atrial appendage ($n = 12$), bilateral superior vena cava ($n = 11$), systemic venous anomaly ($n = 4$), coronary artery anomaly ($n = 4$), dextrocardia ($n = 3$), multiple VSD ($n = 2$), complete endocardial cushion defect ($n = 1$), and criss-cross heart ($n = 1$). In 11 patients, the great arteries were side by side, whereas the other patients had the usual anteroposterior relationship. Four patients had abnormal insertion of the tricuspid tension apparatus on the outlet septum.

Previous Operation

A total of 26 patients underwent 1 Blalock-Taussig shunt, and 2 patients underwent 2 shunts. Two patients underwent balloon atrial septostomy. One patient underwent atrial septectomy after shunt, and 1 patient underwent atrial septectomy and pulmonary valvotomy after balloon atrial septostomy. The mean time interval from the first palliation to complete repair was 1.98 ± 1.06 years.

Operative Technique and Operative Data

We previously described the technical aspects of the modified Lecompte procedure in detail for different anatomic variations of the anomalies of ventriculoarterial connection with VSD and left ventricular outflow tract (LVOT) obstruction.^{12,13} The VSD was baffled from the LV to the aorta after the conal septum was resected, and VSD extension also was undertaken, which enlarges even a small restrictive VSD and constructs a wider and straighter LVOT. In cases involving the abnormal insertion of the tricuspid tensor apparatus, we transferred them onto the right side of the patch after division. The branches of pulmonary artery were dissected and mobilized sufficiently beyond the pericardial reflection for tension-free anastomosis between the RV and the pulmonary artery. The Lecompte maneuver¹⁶ was not applied, and the pulmonary bifurcation

was located orthotopically posterior to the ascending aorta, because the anterior ascending aorta in the anomalies of ventriculoarterial connection with the VSD and pulmonary outflow tract obstruction is usually large, and we also wanted to simplify the procedure and reduce the risk of bleeding by avoiding the transection of aorta and reanastomosis. Right translocation of the main pulmonary trunk was performed in 9 patients, which depended on the initial position of the great arteries. The RVOT was reconstructed with an anterior patch after a monocusp valve was inserted along the margin of the ventriculotomy. Materials for the monocusp valve included autologous or bovine pericardium, homograft monocusp valve, and polytetrafluoroethylene (Gore-Tex; WL Gore & Associates, Inc, Flagstaff, Ariz) membrane. Additional operative procedures were performed in 10 patients: branch pulmonary artery angioplasty in 6 patients, left bidirectional cavopulmonary shunt in 1 patient, tricuspid annuloplasty in 1 patient, aortic valvuloplasty in 1 patient, mitral valvuloplasty in 1 patient, and atrioventricular valve replacement in 1 patient. The cardiopulmonary bypass time was 189.6 ± 75.4 minutes, and the aortic crossclamp time was 93.3 ± 36.3 minutes. In 6 patients, circulatory arrest under deep hypothermia was used.

Follow-up Evaluation

After operation, Doppler echocardiography was regularly checked. Cardiopulmonary exercise test also was performed. According to the clinical status and echocardiographic findings, special cardiac evaluations were followed, such as repeat cardiac catheterization, computed tomography, magnetic resonance imaging, and myocardial single-photon emission computed tomography.

Statistical Analysis

Statistical analyses were performed using SPSS version 21.0 (SPSS, Inc, Chicago, Ill). All descriptive data were expressed as mean values \pm standard deviation or median with ranges, as appropriate. Estimated survival and freedom from events, including reoperation and arrhythmia, were determined by the Kaplan-Meier method. Subgroups were compared using the log-rank test. Variables were evaluated using by the likelihood ratio test in the Cox proportional hazards regression model. Hazard ratios with 95% confidence intervals were constructed for the significant predictors.

RESULTS**Survival**

There were 2 early deaths (4.0%). Five of 48 early survivors could not be traced for follow-up. During a mean follow-up of 14.2 ± 7.9 years (range, 0.2-25 years), there were 4 late deaths. Actuarial survival was $89.9\% \pm 4.3\%$ at 5 years, $89.9\% \pm 4.3\%$ at 10 years, $87.3\% \pm 4.9\%$ at 15 years, $87.3\% \pm 4.9\%$ at 20 years, and $87.3\% \pm 4.9\%$ at 25 years (Figure 1). The cause of death included low cardiac output syndrome in 3 patients, infection in 2 patients, and unknown cause in 1 patient (Table 1). Aortic cross-clamp time was statistically significant risk factor for mortality in univariate analysis ($P = .047$), but there was no significant risk factor for mortality in multivariate analysis.

Arrhythmia

The freedom from any arrhythmia was $95.7\% \pm 3.0\%$ at 5 years, $95.7\% \pm 3.0\%$ at 10 years, $92.6\% \pm 4.2\%$ at 15 years, $87.7\% \pm 6.2\%$ at 20 years, and $87.7\% \pm 6.2\%$ at 25 years (Figure 2). The freedom from tachyarrhythmia

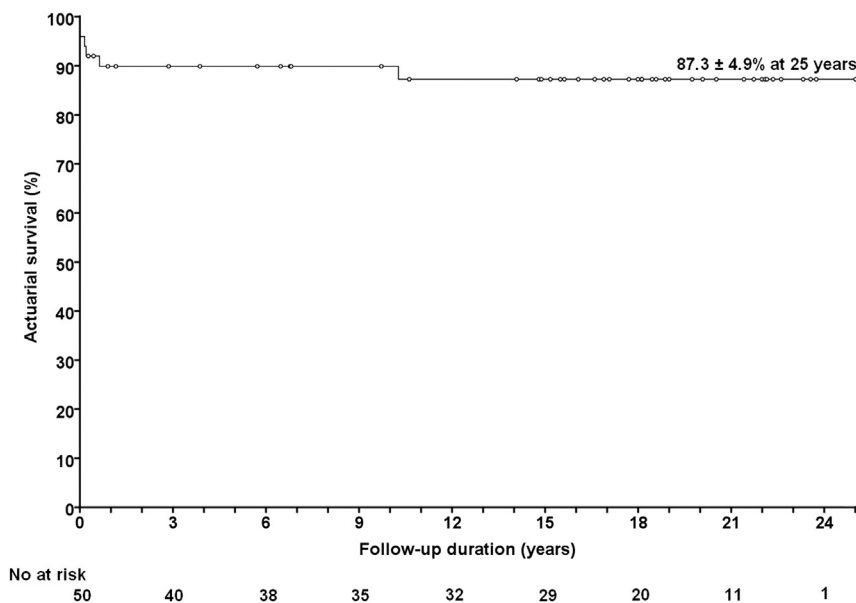


FIGURE 1. Kaplan–Meier curves for actuarial survival.

was 95.2% ± 4.6% at 25 years, and 1 patient developed atrial flutter. The freedom from bradyarrhythmia was 92.6% ± 4.2% at 25 years, and 3 patients developed bradycardia. One patient developed sinoatrial node dysfunction, and 2 patients developed complete atrioventricular block, which required pacemaker placement. The freedom from complete atrioventricular block was 95.7% ± 3.0% at 25 years. There was no significant risk factor for arrhythmia occurrence in multivariate analysis.

Reoperation

Twenty-three patients (46.0%) underwent reoperation after the modified Lecompte procedure. The freedom from any reoperation was 90.5% ± 4.5% at 5 years, 74.0% ± 7.2% at 10 years, 46.3% ± 8.6% at 15 years, 25.2% ± 9.4% at 20 years, and 25.2% ± 9.4% at 25 years (Figure 3). The causes of reoperation are shown in Table 2. Although residual VSD or LVOT obstruction was not indicated for reoperation, these residual lesions were repaired as combined procedure at the same time of RVOT reoperation. The freedom from reoperation for RVOT obstruction was 92.7% ± 4.1% at 5 years, 76.2% ± 7.0% at 10 years,

58.0% ± 8.4% at 15 years, 49.6% ± 9.0% at 20 years, and 49.6% ± 9.0% at 25 years (Figure 4). The cause of reoperation for RVOT obstruction was thickened and calcified immobile monocusp in 13 patients and peripheral pulmonary stenosis in 4 patients. The freedom from reoperation for LVOT obstruction was 97.5% ± 2.5% at 5 years, 94.6% ± 3.7% at 10 years, 88.5% ± 5.4% at 15 years, 88.5% ± 5.4% at 20 years, and 88.5% ± 5.4% at 25 years (Figure 4), which was significantly higher than RVOT obstruction (P = .001). There was no significant risk factor for reoperation in multivariate analysis.

Functional Status

At last follow-up, 43 survivors (97.7%) are in New York Heart Association (NYHA) class I, and 1 survivor (2.3%) is in NYHA class II. Only 6 survivors (13.6%) were taking medications, and 38 survivors (86.4%) do not need any medication. Cardiopulmonary exercise test was performed on 20 patients (45.5%), and maximal oxygen consumption (normal >30 mL/kg/min) was 31.16 ± 5.55 mL/kg/min (range, 19.80-42.40 mL/kg/min), and usually within the normal range except in 2 patients.

Reevaluation for Survivors

Doppler echocardiography was regularly checked in all survivors (n = 44, 100%) to 14.76 ± 7.44 years (range, 0.04-24.02 years) after operation. LV ejection fraction was 63.30% ± 7.90% (range, 48.31%-78.00%), and shortening fraction (normal range, 28%-38%) was 34.63% ± 5.83% (range, 24.24%-45.50%). No patient had AR of more than moderate degree or significant LVOT obstruction. No patient had mitral regurgitation of more than

TABLE 1. Mortality cases

No	Type	Timing	Cause
1	Early	0 d	Low cardiac output syndrome
2	Early	0 d	Low cardiac output syndrome
3	Late	2 mo	Infection
4	Late	3 mo	Infection
5	Late	8 mo	Unknown
6	Late	10 y	Low cardiac output syndrome

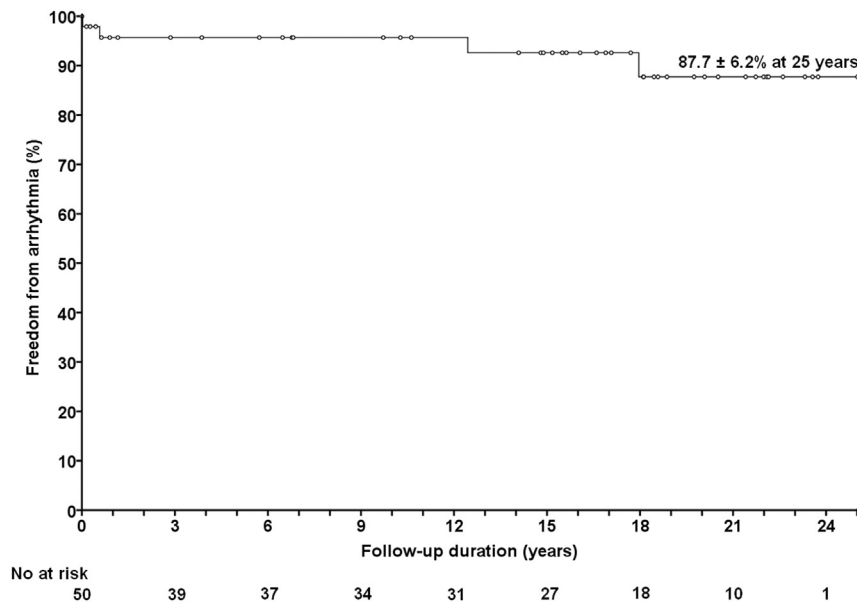


FIGURE 2. Kaplan–Meier curves for freedom from any arrhythmia.

mild degree and tricuspid regurgitation of more than moderate degree. Two patients had significant pulmonary stenosis, and 2 patients had pulmonary regurgitation (PR) of more than moderate degree. Repeat cardiac catheterization was performed in 27 survivors (61.4%) to 10.87 ± 6.43 years (range, 0.62-23.67 years) after operation. LV end-diastolic pressure was 10.16 ± 3.14 mm Hg (range, 4-16 mm Hg), and RV end-diastolic pressure was 9.57 ± 3.33 mm Hg (range, 4-16 mm Hg). Computed tomography was performed in 22 survivors (50%) to 13.81 ± 3.63 years (range, 5.55-19.77 years) after operation and as a routine evaluation before reoperation in 7 cases. Additional abnormal results were not demonstrated. Magnetic resonance imaging was performed in 14 survivors (31.8%) to 15.60 ± 6.94 years (range, 1.48-24.11 years) after operation. LV ejection fraction was $51.21\% \pm 8.08\%$ (range, 37.20%-66.60%). LV end-diastolic volume index was 93.48 ± 22.80 mL/m² (range, 70.70-147.10 mL/m²), and LV end-systolic volume index was 46.56 ± 17.68 mL/m² (range, 25.10-88.40 mL/m²). RV ejection fraction was $47.60\% \pm 7.58\%$ (range, 36.60%-61.30%). RV end-diastolic volume index was 138.53 ± 32.54 mL/m² (range, 101.40-210.60 mL/m²), and RV end-systolic volume index was 74.00 ± 26.00 mL/m² (range, 41.10-133.60 mL/m²). Myocardial single-photon emission computed tomography was performed in 4 survivors (9.1%) to 15.57 ± 3.12 years (range, 13.20-20.16 years) after operation, and within normal range.

DISCUSSION

A quarter of a century ago, we started the Lecompte procedure for the anomalies of ventriculoarterial connection

with VSD and pulmonary stenosis, and have reported its effectiveness and early to midterm results.¹²⁻¹⁴ In this study, we evaluated the long-term results of the Lecompte procedure to help optimal selection among variable surgical options, because there is much debate on the best operation of this anomaly. In the Rastelli operation, small-sized VSD and the presence of tricuspid valve straddling or abnormal attachment of the tricuspid chordae on the conal septum are limiting factors and risk factors for mortality. For RV-pulmonary artery connection, the availability of homografts is limited and the durability of xenografts is disappointing, particularly in small infants. The timing of operation is usually delayed for the use of large conduit with enhanced durability, and multiple reoperation is inevitable for conduit change.^{1,5-7} However, when using the Lecompte procedure, the size of the VSD and abnormal attachment of the tricuspid chordae on the conal septum are not limiting factors. In this study, the Lecompte procedure was successfully applied in the patients who had contraindications or risk factors for the classic Rastelli operation even at the early age of 3 months and with a body weight of 5.7 kg. Because a prosthetic conduit was not used, age and body weight were not limiting factors, and multiple reoperation was avoided, unlike in the Rastelli procedure.¹⁴

In this series, 20-year overall survival was 87.3%. In comparison with the study by Di Carlo and colleagues,¹⁵ 20-year overall survival was 52% to 95% (52%-59% in the Rastelli procedure, 95% in the Nikaidoh procedure, and 85% in the Lecompte procedure). An early Lecompte operation achieves better long-term results by avoiding the negative effects of long-standing cyanosis, ventricular

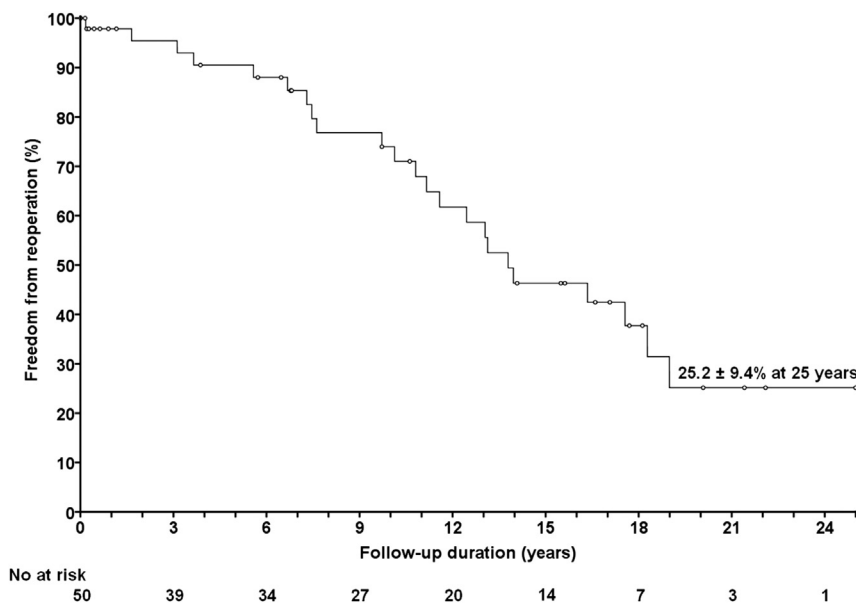


FIGURE 3. Kaplan–Meier curves for freedom from reoperation.

volume overload, and RV hypertension. The LVOT was more straightly constructed than in the Rastelli procedure, and because the Lecompte maneuver was not applied, the RVOT was orthotopically posteriorly positioned. Conversely, in the Rastelli procedure, the LVOT was not straight, and the RVOT was positioned anteriorly to the large aorta, which was already in an anterior position. The risk of sternal compression was increased, and the long-term results of the Rastelli procedure were disappointing because of recurrent RVOT or LVOT obstruction, ventricular dysfunction, arrhythmia, and sudden death.⁵

Conal septum resection and VSD enlargement may carry a potential increased risk for complete atrioventricular block, and its scar tissue play a role as a substrate for late-onset arrhythmia. However, in this series, the freedom from any arrhythmia and complete atrioventricular block was 87.7% ± 6.2% and 95.7% ± 3.0% at 25 years, respectively. Conversely, the prevalence of early and late arrhythmia including sudden death was high after Rastelli

repair.⁵⁻⁷ The cause of arrhythmia may include right or left ventricular hypertension due to RVOT or LVOT obstruction. The right bundle branch block also progresses to bifascicular and complete atrioventricular block.

According to the degree and anatomic characteristics of complex forms of LVOT obstruction, a substantial percentage of patients with dynamic or resectable LVOT obstruction may undergo an arterial switch operation with or without relief of LVOT obstruction.¹⁷⁻²⁰ The patients with moderate or severe mechanical LVOT obstruction and unresectable or not easily resectable LVOT obstruction may undergo a nonarterial switch operation repair (Rastelli, Lecompte, or Nikaidoh procedure). However, the guidelines for selection of the best anatomic LVOT obstruction type according to various surgical options have not been established. After the Rastelli operation, the LV undergoes a geometric change, such as increased wall thickness, decreased cavity dimensions, and decreased VSD size, which subsequently develops LVOT obstruction according to the patient’s growth.²¹ RV hypertension also produces septal hypertrophy and leftward septal displacement, which may risk LVOT obstruction. Although anterior VSD enlargement is usually recommended,⁵⁻⁷ resection of the infundibular septum was reported to diminish recurrent LVOT obstruction risk in the Rastelli procedure.²² In this series, the freedom from reoperation for LVOT obstruction was 88.5% ± 5.4% at 25 years. In our previous study¹⁴ comparing the outcomes of the Lecompte procedure and the Rastelli repair in TGA with VSD and LVOT obstruction, reoperations for LVOT obstruction in the Rastelli group (40%) were more frequently required than in the Lecompte group (4%)

TABLE 2. Cause of reoperation

Cause of reoperation	No. of patients
RVOT obstruction	17
PR	7
VSD	5
LVOT obstruction	4
Others	6
Arrhythmia	2
Mitral regurgitation	2
AR	1
Subacute bacterial endocarditis	1
Total	23

AR, Aortic regurgitation; LVOT, left ventricular outflow tract; PR, pulmonary regurgitation; RVOT, right ventricular outflow tract; VSD, ventricular septal defect.

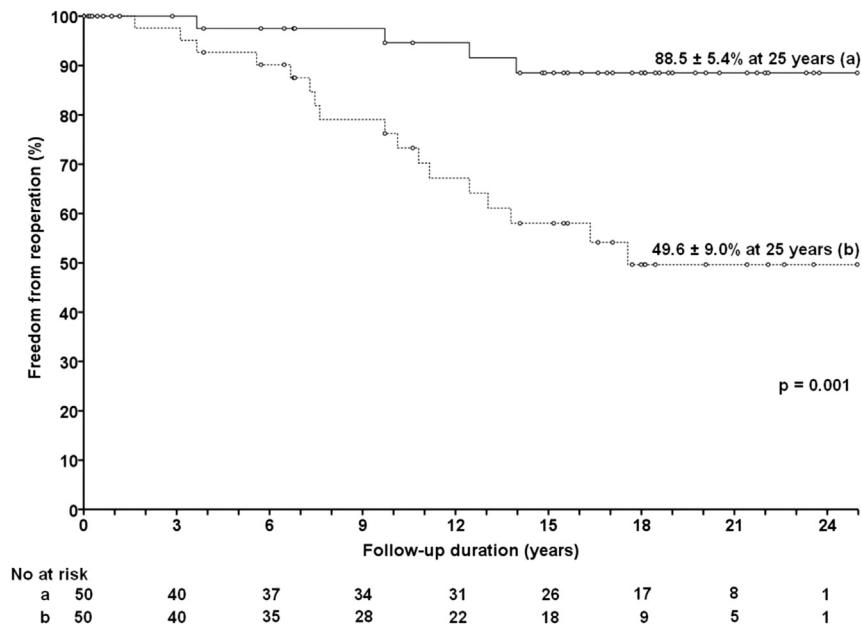


FIGURE 4. Kaplan–Meier curves for freedom from reoperation for LVOT obstruction (a) and RVOT obstruction (b).

($P = .02$), because the radical conal septum resection and use of a shorter and straight patch resulted in a wider left ventricular-aortic tunnel, decreasing the risk of subaortic stenosis in the Lecompte procedure.

Our modified Lecompte procedure avoids the problems of limited durability that are usually inherent in an RV-pulmonary arterial valved conduit and the obstruction of the RVOT, which is positioned orthotopically posteriorly, and not susceptible to sternal compression, unlike in the Rastelli operation. In this series, the freedom from reoperation for RVOT obstruction was 58.0% and 49.6% at 15 and 25 years, respectively. The main cause of recurrent RVOT obstruction was degeneration of the monocusp valve, which needs resolution.¹³ In comparison with the study by Di Carlo and colleagues,¹⁵ 15-year freedom from RVOT reoperation was 20% to 75% (20%-32% in the Rastelli procedure, 64% in the Nikaidoh procedure, and 75% in the Lecompte procedure). In our previous study,¹⁴ reoperations for RVOT obstruction in the Rastelli group (80%) were more frequently required than in the Lecompte group (21%) ($P = .05$), and the risk factors of RVOT obstruction were a younger age at operation, the Rastelli operation, and ductus ligation during the operation. In our previous study,¹⁴ although age in the Lecompte group was lower than in the Rastelli group (23.4 ± 18.2 months vs 39.1 ± 36.1 months, $P = .10$) and body weights in the Lecompte group were lower than in the Rastelli group (10.2 ± 3.0 kg vs 13.8 ± 6.8 kg, $P = .04$), the freedom from reoperation at 5 and 10 years was $40.0\% \pm 15.5\%$ and $26.7\% \pm 15.0\%$, respectively, after the Rastelli operation and $95.7\% \pm 4.3\%$ and $63.5\% \pm 12.6\%$, respectively, after the Lecompte procedure ($P = .02$). In this series, 26 patients (52%) were aged less

than 2 years at operation, but early correction was not a risk factor for increased reoperation. The Lecompte group¹⁵ reported that the freedom from any reoperation at 25 years was 45%, and the most frequent reason for reoperation was recurrent RVOT obstruction. The hazard function of reoperation for RVOT obstruction after the REV procedure remains constant over time, and lower age at operation stood out as an independent risk factor for reoperations because of RVOT obstruction.¹⁵

The potential for somatic growth of the native pulmonary trunk and pulmonary valve was preserved; moreover, a functioning monocusp valve in the pulmonary outflow tract consistently helps to prevent PR and RV volume loading. The Lecompte group¹⁵ reported that PR has not emerged as a frequent cause for reoperation, and patients seem to tolerate PR well in the long-term follow-up after the REV procedure, compared with tetralogy of Fallot repair. In our series, the cause of reoperation included PR in 7 patients (14.0%). The early surgical intervention for PR was performed to reduce the RV volume and improve the RV function. Our surgical indications for more than moderate PR included decreased RV function on echocardiography, standardized RV end-diastolic volume index greater than 170 mL/m^2 in cardiac magnetic resonance imaging, and reduced exercise capacity.²³

At our last follow-up, 43 survivors (97.7%) were in NYHA class I without medication in 38 (86.4%). Ventricular performance was good according to our data analysis of most recent cardiac evaluations. The resection of the conal septum along with VSD enlargement, which constructs the straight and short LVOT, and produces a limited bulge in the RV cavity, an orthotopically positioned durable

RVOT, and corrective surgery at a young age might favorably affect both LV and RV function.^{7,15} However, after the Rastelli operation, LV function is usually abnormal and residual left ventricular dilation and wall hypertrophy remain.²⁴ RV and LV function were decreased, particularly in older patients undergoing the Rastelli operation, which was caused by the deleterious effects of hypoxia, obstruction, volume overloading, and RV hypertension over a prolonged period. Although patients aged 4 years or more at the time of the Rastelli operation require fewer reoperations for conduit exchange, early Rastelli repair is recommended because patients aged 4 years or more are at risk for a higher long-term mortality.⁷ Contractile dysfunction also was explained by RVOT obstruction by sternal compression, large VSD baffle in the RV, and LVOT obstruction by septal displacement.²⁵

As an alternative surgical option, aortic root translocation has been reported in a small number of patients.⁸⁻¹⁰ However, although the follow-up interval was not long, AR was frequently encountered after aortic root mobilization, and reoperations for conduit replacement were inevitable (more common in valved conduits than valveless reconstruction). Myocardial ischemia occurred because this operation risks the integrity and function of the coronary arteries, and there was still a debate for coronary reimplantation. Atrial and ventricular arrhythmia also occurred, which required maze procedures or implantable defibrillators. In a recent report for modified operative technique,¹¹ the Nikaidoh procedure was more time-consuming and required postoperative extracorporeal membrane oxygenation support and more mechanical ventilation support. Even with a short follow-up interval, the desired result of avoiding PR was not accomplished, and early evidence of AR was noted.

CONCLUSIONS

The modified Lecompte procedure has excellent long-term results for treating anomalies of ventriculoarterial connection with VSD and pulmonary outflow tract obstruction. Early repair is possible with low mortality and morbidity in terms of arrhythmia, reoperation for RVOT or LVOT obstruction, functional class, and ventricular function.

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