Repair of persistent truncus arteriosus without a conduit: sleeve resection of the pulmonary trunk from the aorta and direct right ventricle-pulmonary artery anastomosis

Shintaro Nemoto a,*, Hideki Ozawa a, Tomoyasu Sasaki a, Takahiro Katsumata a, Kanta Kishi b, Kenichi Okumura b, Yasuhiko Mori b

a Department of Cardiovascular Surgery, Osaka Medical College Hospital, 2-7 Daigaku-machi, Takatsuki, Osaka 569-8686, Japan
b Department of Pediatrics, Osaka Medical College Hospital, 2-7 Daigaku-machi, Takatsuki, Osaka 569-8686, Japan

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Abstract

Objective: Establishing a new continuity between the right ventricle and the pulmonary artery is the mainstay of repair for persistent truncus arteriosus. We used the Tran Viet—Neveux technique without a Lecomte maneuver to construct the connection without a conduit. Here, we retrospectively review the mid-term surgical results to examine the effectiveness of this approach.

Methods: A cylindrical segment incorporating both pulmonary artery branches was sleeve-resected from the truncal artery. The cylindrical segment was cut in the middle and two truncal arterial flaps were combined to form the posterior floor of the new pulmonary arterial trunk. The edge of the floor was attached directly to the superior margin of an oblique incision made in the left-anterior wall of the right ventricle. A polytetrafluoroethylene monocusp was attached to the lower half margin of the right ventricular incision. A large glutaraldehyde-treated pericardial patch was used to form the anterior hood of the new right ventricular outflow tract. Both great arteries were located in a normal spiral configuration.

Results: Ten babies (range: 3 days to 9 months of age) underwent this procedure. The Collett—Edwards classification of persistent truncus arteriosus was type I in five cases and type II in five others. There was one hospital death due to severe respiratory distress. During follow-up (36—60 months, median 54 months), only one re-operation was required to enlarge a left branch pulmonary artery stenosis. Follow-up echocardiography showed pulmonary regurgitation (mild two, moderate seven, and severe one) and mild flow acceleration in the left pulmonary artery branch and right ventricle—pulmonary artery connection in one case.

Conclusion: This simple modification for surgical correction of persistent truncus arteriosus may be an effective alternative that overcomes conduit-related problems.

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Keywords: Persistent truncus arteriosus; Repair without conduit; Right ventricle—pulmonary artery direct anastomosis; Surgical results; Pulmonary hypertension

1. Introduction

Establishing a new continuity between the right ventricle (RV) and the pulmonary artery (PA) is the mainstay of repair for persistent truncus arteriosus. Repair using an RV—PA conduit is currently the preferred choice in most centers. Although conduit repair is costly and limited by the available size, it has several advantages in surgical repair of persistent truncus arteriosus, including enabling a quick and simple surgery and guaranteed pulmonary valve competency. However, regardless of which conduit is used, conduit replacement is not guaranteed in follow-up after surgery because of tissue degeneration and outgrowth of conduits causing stenosis or regurgitation.

RV—PA reconstruction without use of a conduit may overcome the problems in surgical repair of persistent truncus arteriosus and other congenital cardiac anomalies with an RV—PA discontinuity. Several approaches to RV—PA reconstruction without a conduit have been reported. Reconstruction enables somatic growth, but has poor rates of in-hospital mortality in the beginning [1—3] and freedom from re-operation or re-intervention due to stenosis and/or regurgitation at the RV—PA connection, the rate of which varies widely from low [4] to high [2,5,6]. To search for a better approach to RV—PA reconstruction without use of a conduit, we revived the Tran Viet—Neveux technique reported in the early 1980s [7] by excluding a Lecompte maneuver [8]. Here, we present the details of our surgical modification and a retrospective review of the short- and mid-term outcomes.
2. Patients and methods

2.1. Surgical procedure

All operations were performed under general anesthesia with modified neuroleptanalgesia (NLA; intravenous fentanyl, and midazolam). After median sternotomy, a large piece of pericardium was procured and treated with glutaraldehyde for 5 min. The aorta and pulmonary artery were fully mobilized by dissected down to the all neck vessels and both hila of the pulmonary artery, respectively. During cardiopulmonary bypass, the flow rate was maintained at 150 ml min\(^{-1}\) kg\(^{-1}\) with mild-to-moderate hypothermia. Deep hypothermic arrest was not used. The heart was arrested using intermittent cold-blood cardioplegia. After cross-clamping the aorta, the truncal artery was transected at the level of the upper margin of the PA origin. Another transverse incision was made on the anterior wall of the proximal truncal artery below the transaction line and slightly above the sinotubular junction. The incision was extended posterior just below the pulmonary orifice. To avoid posterior displacement of the newly constructed ascending aorta, the posterior width of the sleeve should be matched to the diameter of the PA opening, and the anterior width was smaller than the diameter most of the time. The cylindrical segment was cut in the middle and two flaps of truncal artery were joined to form the posterior floor of the new pulmonary trunk (Fig. 2(a) and (b)). An oblique incision was made parallel to the left anterior descending coronary artery on the anterior wall of the right ventricle. The ascending aorta is reconstructed by an end-to-end anastomosis, with wider bites taken on the proximal stump to make up for the discrepancy in the size of two ends of the aorta. Unlike in the original technique, in which transfer of the new pulmonary trunk anterior to the aorta is achieved by a Lecompte maneuver\cite{7,8}, the new pulmonary trunk is located in a normal spiral relationship to the aorta in our modification. We expect this normal configuration of the great arteries to prevent sternal compression to the newly constructed pulmonary trunk and excess stretch to both the pulmonary branches which are dislocated far more anterior by a Lecompte maneuver, compared with the transcinated PA after the arterial switch operation for the complete transposition of the great arteries.

The ventricular septal defect was closed with a 0.4-mm polytetrafluoroethylene patch through the right ventricular incision in a continuous suturing fashion. The proximal end of the new pulmonary trunk was directly matched to the superior margin of the right ventricular incision by pledged mattress sutures (Fig. 3). After releasing the cross-clamp, a large piece of 0.1-mm polytetrafluoroethylene membrane was attached to the two-thirds margin of the right ventricular incision as a new pulmonary monocusp. A large glutaraldehyde-treated autologous pericardial patch was used to form the anterior roof of the new right ventricular outflow tract under guidance of a Hegar dilator (12 mm in diameter) placed longitudinally on the posterior floor of the new pulmonary trunk. After completion of the procedure, a pressure-monitoring catheter was directly inserted into the new PA via the free wall of the right ventricular outflow. Modified ultrafiltration was used after discontinuation of cardiopulmonary bypass.

2.2. Postoperative management

Following admission to the intensive care unit, all patients were routinely sedated using a combination of midazolam, morphine and dexmedetomidine, and an optimal sedation level was maintained. Patients who required delayed chest
consent from parents or guardians, sildenafil was administered via a nasogastric tube or orally at a starting dose of 0.5 mg kg\(^{-1}\) every 4–6 h up to a maximum dose of 2 mg kg\(^{-1}\) with careful hemodynamic monitoring. After successful weaning from a ventilator, sildenafil was gradually reduced and discontinued over the next 5–7 days if possible [9].

2.3. Characteristics of the patients

Surgery without a conduit for persistent truncus arteriosus was performed in 10 consecutive cases (five males, five females) between 2005 and 2007. The age of the patients at the time of the operation was neonate, 1–3, 4–6, and >6 months in three, three, three, and one of the cases, respectively. Early surgical repair in neonatal period or within 3 months after birth is our current policy for the persistent truncus arteriosus. However, several patients were referred to us late from the other territorial hospitals because they overcame life-threatening disease, such as respiratory distress, pneumonia, sepsis, and intracranial lesion. The Collett—Edwards classification of persistent truncus arteriosus was type I in five cases and type II in five others. Truncal valve anomalies (with regurgitation in parenthesis) were found in the bicuspid valve in two (one) cases and in the quadricuspid valve in one (one) case. Truncal valve regurgitation of severity greater than mild was present in six cases. Truncal valve plasty was performed in two cases with regurgitation of severity greater than moderate: commissural plication plus De Vega type annular reduction with an absorbable suture in one case, and resection of the accessory cusp and sinus of Valsalva of the quadricuspid valve in the other case.

Concomitant cardiac lesions included a single coronary artery in two cases, a main pulmonary artery stenosis in one, and an abnormal neck branch in one. The sternum was left open immediately after surgery in five cases and delayed chest closure was successfully performed in all these cases.

3. Results

3.1. In-hospital outcome

Severe pulmonary hypertension occurred in the acute postoperative period in eight cases and was treated with targeted pulmonary vasodilators: inhaled nitric oxide alone in one case and inhaled nitric oxide plus sildenafil in seven. One patient developed severe respiratory distress and sustained pulmonary hypertension-related symptoms despite receiving inhaled nitric oxide and sildenafil. Bosentan, a dual endothelin-receptor blocker, was added at a dose of 1.5 mg kg\(^{-1}\) day\(^{-1}\), and the pulmonary artery pressure was controlled well at less than 50% of systemic pressure. The symptoms were relieved and inhaled nitric oxide and sildenafil were discontinued without rebound pulmonary hypertension. Sildenafil treatment was completed in all cases during the hospital stay.

One neonate required extracorporeal lung support on day 3 for severe respiratory distress. This baby was successfully weaned from support after 6 days and was discharged from hospital uneventfully. Re-exploration was required to release cardiac tamponade in one case on day 5. Diaphragm plication was performed in two cases for difficulty with ventilator weaning.

There was one hospital death, in a case of a 2-month-old boy with DiGeorge syndrome, who was mechanically ventilated preoperatively because of severe heart failure with pulmonary hypertension and respiratory failure due to emphysematous lung. His Collett—Edwards classification of persistent truncus arteriosus was type II with abnormal neck branches and a bicuspid truncal valve. Cardiac arrest occurred during skin incision, but the patient was resuscitated immediately. After the surgical correction, the baby
was uneventfully weaned from cardiopulmonary bypass. Delayed chest closure was performed on day 3. However, severe respiratory distress and bilateral pneumonia rapidly progressed and resulted in severe sepsis, leading to death on day 14. The other nine patients were successfully discharged from hospital. Cardiac ultrasound at hospital discharge showed pulmonary regurgitation that was trivial in two cases, mild in seven cases, and moderate in one case. Flow acceleration of 3.0 m s⁻¹ was detected at the origin of the left branch pulmonary artery in one case.

3.2. Mid-term outcome

No deaths occurred during a follow-up period of 36—60 months (median: 54 months) after repair. Two re-operations were performed: one for closure of residual ventricular septal defect with renewal of the polytetrafluoroethylene monocusp 1 year after repair, and the other for patch augmentation using a glutaraldehyde-treated autologous pericardium for left branch PA stenosis at the origin with renewal of the polytetrafluoroethylene monocusp 5 months after repair. Echocardiography showed pulmonary regurgitation that was mild in two cases, moderate in seven cases, and severe in one case at the last follow-up at our outpatient clinic.

Flow acceleration of 2.7 m s⁻¹ was detected at the RV—PA connection and at the origin of the left branch PA in one case 14 months after repair. Cardiac catheterization and RV angiography were performed (Fig. 4). The pressure gradients across the RV—PA connection and the left PA origin were both 15 mmHg. The RV/LV pressure ratio was 0.6 and the RV end-diastolic volume was 149% of normal. Pulmonary regurgitation was moderate and the Nakata index was 388. Catheter intervention was not indicated. The child had no symptoms and has been followed up prescribing oral medication.

The severity of the truncal valve regurgitation at the last follow-up echocardiogram was mild in eight cases, trivial in one case, and faint in one case. The contour of the ascending aorta was normal in all cases (Fig. 5).

4. Discussion

There are three major findings in this study. First, our modification of RV reconstruction without an RV—PA conduit was relatively simple and reproducible in 10 consecutive patients, with provision of an ideal diameter for the new pulmonary trunk and minimization of tension on the bifurcation and the branch PAs. Second, although there is a concern regarding future functional deterioration of the polytetrafluoroethylene-membrane monocusp valve, the monocusp provides sufficient valve competence to overcome life-threatening severe pulmonary hypertension in the early postoperative period. Third, catheter intervention, or re-operation for stenosis, or outgrowth of the newly reconstructed RV—PA route was not required, at least in mid-term follow-up; this is a frequent problem with the RV—PA conduit, regardless of the type of conduit that is used [1,5,10].

As homografts and other prefabricated xenografts are not commercially available in Japan, a handmade valved equine pericardial roll is the preferred RV—PA conduit in surgical repair of persistent truncus arteriosus [11,12]. However, it also has failed to give satisfactory mid- and long-term surgical outcomes as reported in the series using the allograft [1,5,10]. Therefore, the search for an ideal conduit has started, and grafts based on tissue engineering have been developed and applied clinically in a few hospitals [13]. However, these grafts are still not commercially available and have never been used in neonates or early infants. On the other hand, recent reports
encouraged us to explore this challenge because of the fact that valved homograft recipients were more likely to require re-operation than patients who had undergone RV—PA direct connection in direct comparison studies [5,14]. The current topic of the surgical treatment of persistent truncus arteriosus in most centers has stepped forward to morbidity reduction from life-saving surgery. With this background, it is a step in the right direction to establish a better RV—PA continuity without using a conduit. Moreover, two condition must be met; first, the ideal, direct RV—PA connection must have the same advantages of using a conduit (simple, of adequate size, and producing a competent valve); and, second, the ideal RV—RA connection must avert the disadvantages of using a conduit (thus, with improved growth potential and less chance of a re-operation).

In 1982, Lecompte et al. reported a new surgical technique without use of a conduit for repair of persistent truncus arteriosus (in four patients) and other related anomalies that required a new RV—PA tract [8]. This method introduced the concept of direct anastomosis of the RV and PA, in which a portion of the pulmonary trunk was detached from the truncus, passed anterior to the aorta (known as the Lecompte or French maneuver), and anastomosed directly to the upper margin of the right ventricular incision. However, the short- and long-term results were poor. This technique was further developed by Tran Viet and Neveux by detaching the pulmonary trunk with a cylindrical rim of the truncal wall [7]. This enables lengthening and widening of the native posterior wall of the pulmonary trunk and lessens the tension between the RV incision and the pulmonary bifurcation and branches. The mid- and long-term results of the Tran Viet—Neveux technique have not been reported, but the technique attracted a great deal of interest for construction of a new pulmonary trunk.

In 1990, Barbero-Marcial et al. reported a new RV—PA construction technique without a conduit for persistent truncus arteriosus [3]. In this technique, the pulmonary trunk was not detached and was pulled down to the RV incision for direct anastomosis, which caused a tendency for the posterior wall of the new pulmonary trunk to be short and narrow. This led to distortion of the bifurcation, as shown in a representative RV angiogram in the report of the long-term results [6]. In fact, a relatively high incidence (23.3%) of pulmonary stenosis was found in the long-term results, although with excellent freedom from re-operation of 89% at 11.4 years after repair [6]. This problem was overcome by interposing the left atrial appendage between the PA incision and the RV incision. Therefore, the Barbero-Marcial technique has become the preferred choice of many surgeons for non-conduit repair of persistent truncus arteriosus [1,5,14].

Regardless of the technique chosen for direct RV—PA anastomosis repair, construction of a competent monocusp valve is necessary for maintaining RV function by reduction of pre- and afterload, especially when severe pulmonary hypertension occurs in the acute critical phase after surgery. A large bullet-shaped polytetrafluoroethylene membrane has been our choice as a monocusp valve in various kinds of RV outflow reconstruction, based on data published by Brown et al. [15] In the acute- and short-term periods, the polytetrafluoroethylene monocusp valves were mobile and pulmonary valve regurgitation remained mild in this series. This may have prevented severe postoperative pulmonary hypertension and led to satisfactory in-hospital survival, along with our targeted treatment for pulmonary hypertension [9] by contrast, direct RV—PA connection repair for persistent truncus arteriosus has a high in-hospital mortality rate reported in the early series [1—3,7,8].

In 3—5 years follow-up in our series, pulmonary valve regurgitation gradually worsened, while the monocusp valve slowly lost its mobility and eventually became fixed. As longstanding pulmonary valve regurgitation may lead to RV failure and ventricular arrhythmia, as seen in long-term follow-up of patients with tetralogy of Fallot who underwent trans-annular patch repair [16], insertion of a competent bioprosthetic valve in the pulmonary position may also be necessary and even required in the long-term after RV—PA direct anastomosis repair for persistent truncus arteriosus. In this context, the pulmonary trunk must be wide and long enough to enable simple insertion of a valve of sufficient size to minimize use of other synthetic material or a xenograft at the time of valve insertion. As the new pulmonary trunk is located anterior to the aorta after the Lecompte technique [8] and original Tran Viet—Neveux technique [7], there is no space for valve insertion in the pulmonary valve position between the sternum and the heart. Therefore, the newly constructed pulmonary trunk must be located in the normal configuration of the great arteries. This is achieved in the Barbero-Marcial technique [3,6], but the pulmonary bifurcation is located very close to the RV incision and the width of the trunk becomes relatively narrow. Therefore, major reconstruction of the pulmonary trunk is required at the time of future prosthetic valve insertion after performance of the Barbero-Marcial technique.

To avoid the inherent problems of previous techniques for RV—PA direct anastomosis repair for persistent truncus arteriosus, we have applied the Tran Viet—Neveux technique [7] with less use of synthetic material or a xenograft by excluding a Lecompte maneuver [8]. Our goal was to construct a pulmonary trunk in the normal configuration with growth potential and competency of the pulmonary valve, at least in the early phase after correction, and with less tension on the bifurcation and branch PAs and no sternal compression to the new pulmonary trunk. As the follow-up period is obviously limited, we have to be cautious about the favorable results of this repair until the long-term outcome is shown.

There are several limitations in the study. First, the study was not performed as a randomized comparison of our technique with conduit repair or other types of non-conduit right ventricular outflow reconstruction, and, therefore, we are unable to show the surgical superiority of our technique relative to other approaches for persistent truncus arteriosus. However, the study was performed over a short period and in a relatively homogeneous and consecutive patient population, with the procedure performed by a single surgeon and with similar postoperative management, especially for pulmonary hypertension [9]. Therefore, we believe our modification of non-conduit right ventricular outflow reconstruction is sufficiently characterized despite the small retrospective cohort.

Second, the favorable results of our surgical modification may be affected by application of the technique only to less complex cases of persistent truncus arteriosus; that is, Collett—Edwards type I and II. In the study period, no
complex cases of persistent truncus arteriosus of Collett—
Edwards types III and IV were referred to us for surgical
correction. However, conduit repair is suitable for RV—PA
reconstruction in such complex cases because of a lack of
available trunical wall tissue and central PA, as in correction
of pulmonary atresia with ventricular septal defect, espe-
cially in cases with pulmonary trunical atresia and/or major
aortopulmonary collateral arteries. RV—PA direct connection
repair for persistent truncus arteriosus is considered to be
best when anatomically appropriate [5] or in cases of Collett—
Edwards types I and II [6]. Our modified Tran Viet—Neuvek
 technique might also be most appropriate for surgical
 correction of persistent truncus arteriosus in cases with
simple anatomy of the PA.
Within these limitations, we conclude that the relatively
simple technique of RV—PA reconstruction in repair of
persistent truncus arteriosus is an acceptable and effective
alternative to overcome conduit-related problems, at least
until the mid-term after surgery, and has less chance of a
need for re-intervention. Comparison of this technique with
other types of non-conduit repair for persistent truncus
arteriosus and a detailed evaluation of hemodynamics are
required, along with careful long-term follow-up and
accumulation of experience with the technique.

References
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Appendix A. Conference discussion
Dr O. Raisky (Paris, France): I am going to make it short, something which
is sometimes good in surgery.
I think when we are talking about RV—PA connection without a conduit, we
are going in the right direction, given that the ideal connection has not yet
been described. I am talking about something that is easy to do, not time-
consuming, allows ostial and distal pulmonary growth, which avoids early
pulmonary regurgitation and late stenosis.
Here you propose a method that is a modification of a technique that was
proposed by our group 25 years ago, a technique that I can tell you has been
abandoned by the group because of poor results in relation to the pulmonary
artery bifurcation anatomy and growth, probably due to the 90 degree rotation
of the two flaps that are sewn together to reconstruct the back wall of the
neopulmonary artery. Could you explain to us why avoiding the Lecompte
manoeuer has dramatically improved the results?
Second question. You take a segment of the trunical artery, something that
can be very challenging in a few patients due to the proximity of the left
 coronary, known for its frequent anomalous position and acute angle of takeoff
in some patients.
How do you manage that considering that you are doing a direct aortic
anastomosis later, something that we try not to do to avoid coronary and right
pulmonary artery compression.
Dr Nemoto: Let me answer the second question first.
As you said, to take the flap from the trunical artery is very critical, and if
you take too much, the ascending aorta gets shorter and goes back posteriorly,
so you may compress the right pulmonary artery.
Yes, sometimes we had those kind of difficulties you mentioned. To start
just below the pulmonary artery opening and then go a little bit shallower
anteriorly would be a tip.
The important thing is to make the new posterior wall of PA as long as
possible because that way you can lessen the tension and the torsion to the
bifurcation. That is one thing we are trying to judge – how we can take the segment
during the operation.
Regarding the first question, we do not know long-term results after the
 technique, but one thing we are sure of is that no matter what kind of
procedure is performed without a conduit repair, these patients will need a
pulmonary valve insertion in the future. So at that time, it would be better for
the pulmonary trunk to be on the left side of the aorta for a successful
replacement in the future. In that way you can also reduce the torsion to both
PA branches. That might be the answer for the first question.
Dr J. Vazquez-Jimenez (Aachen, Germany): We have used this technique
before. My former chief professor Messmer used to make an anastomosis like
this one. I am curious to know for how long the monocusp valves function,
because the results you have with pulmonary insufficiency are probably
because of tissue retraction or because the monocusp is standing there.
So in your follow-up, for how long does the monocusp function?
Dr Nemoto: That is one of the critical questions. Most of the time in our
series, the mobility of the monocusp starts to reduce and become fixed, let us
say in 2 years.
Dr Vazquez-Jimenez: Two years?
Dr Nemoto: Two years.
Dr Nemoto: That is a long time.
Dr Nemoto: Yes, but strictly speaking, it may be a year and a half. But when
we see the patients 2 years after the surgery, almost all the cusps were fixed.