



# Conventional repair of total anomalous venous drainage without primary sutureless technique: surgical tips to prevent pulmonary vein obstruction

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## Abstract

**Objectives** Although primary sutureless technique for total anomalous pulmonary venous drainage has been introduced to reduce postoperative pulmonary vein obstruction (PVO), controversy still exists about superiority of the procedure between the conventional repair and primary sutureless technique at the initial repair. In our unit, the conventional repair has been consistently used based on four important surgical policies: (1) mark incision lines between 2 chambers to gain anatomically natural alignment, (2) place precise stitches by “intima-to-intima” using monofilament suture, (3) adequate orifice size should be guaranteed in greater than expected mitral valve size, (4) do not hesitate to undertake a redo additional anastomosis by a different approach when an echocardiography shows the velocity more than 1.5 m/s. This study aims to evaluate mid-term outcome of the conventional repair for total anomalous pulmonary venous drainage.

**Methods** Between 2004 and 2016, consecutive 15 patients who underwent the conventional repair without the primary sutureless technique were included in this study. Survival, Freedom from reoperation, and PVO were retrospectively reviewed.

**Results** Mean follow-up period was  $4.6 \pm 3.7$  years. Except for one patient who died of uncontrollable pleural effusion, all other patients survived with 5-year survival rate of 93.3%. For the 14 survivors, there was no PVO, nor reoperation.

**Conclusions** Following these policies, the mid-term outcome of the conventional total anomalous pulmonary venous drainage repair was excellent without the primary sutureless technique showing no obstruction. The conventional repair can be safely applied at the initial operation when the morphological condition allows for it.

**Keywords** Total anomalous pulmonary venous drainage · Congenital heart disease

## Introduction

Pulmonary vein obstruction (PVO) following total anomalous venous drainage (TAPVD) repair has been one of the issues for years. To avoid PVO, various techniques and tips have been introduced. Sutureless technique initially

introduced to reoperation for PVO, and good long-term results were reported [1–6]. Currently, a number of institutions are applying the sutureless technique at the initial TAPVD repair, the so-called “primary sutureless techniques”. Toronto group [6] advocated the superiority of primary sutureless technique to the conventional repair. However, it is true that the conventional TAPVD repair is not necessarily completely abandoned; there are a number of merits of the conventional TAPVD repair. We believe that some reasons for the worse outcomes of the conventional TAPVD repair would be attributed to the technical problems; therefore, we headed important surgical policy for the conventional TAPVD repair. This study aims to evaluate mid-term outcome of the conventional repair for TAPVD.

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## Patients and methods

Between September 2004 and February 2016, consecutive 15 patients who underwent the conventional TAPVD repair without the primary sutureless technique were included in this study. Survival, Freedom from reoperation and PVO was retrospectively reviewed.

Median age at the surgery was 19 days (range, 1–452), and the median body weight was 3.1 kg (range, 2.1–6.5). Type of the TAPVD included 6 supracardiac, 3 cardiac, 2 infracardiac, 4 mixed (supracardiac + cardiac in 1, supracardiac + infracardiac in 1, cardiac in 1, supracardiac in 1) (Table 1). One patient was diagnosed with polysplenia, also had an interruption of inferior vena cava.

## Statistics

Patient characteristics were summarized using mean, standard deviation, median, minimum, and maximum for continuous variables, and counts and percentages for categorical variables. Kaplan–Meier was used to show survival, freedom from PVO and reoperation. All analyses were performed in the XLSTAT software (Version 19.03, Addinsoft SARL, Paris, France).

## Surgical technique

For all the patients, a preoperative trans-thoracic and intraoperative trans-esophageal echocardiography was performed. If the anatomy was complex, a CT scan was performed whenever possible to assess the relation between the pulmonary venous chamber and other surrounding tissues. Through a mid-line sternotomy, a cardiopulmonary bypass was established between the ascending aorta and bicaval cannulations. In several cases, the left superior vena cava was ligated and divided if necessary. The temperature was

cooled down to moderate hypothermia between 28 and 32 °C.

## Surgical tips for conventional repair

The surgical tips for the TAPVD repair contain as follows: First, it is important to place marking lines between the two facing chambers, pulmonary venous chamber and left atrium to gain anatomically natural alignment. This can prevent stresses on the suture lines and torture between the chambers compressing the orifice of the pulmonary veins. Second, it is also important to place fine stitches by “intima-to-intima” using monofilament suture to avoid the indentation to the inside of the suture line. Third, try to gain an adequate orifice size that is greater than the expected mitral valve size from the body size, which would ensure the wide-open area from the pulmonary veins to the mitral valve. Forth, do not hesitate to undertake an additional anastomosis by a different approach, which is usually a right lateral approach, when the trans-esophageal echocardiography shows the velocity more than 1.5 m/s. The final examination after the cardiovascular bypass is very important.

## Case: infracardiac type

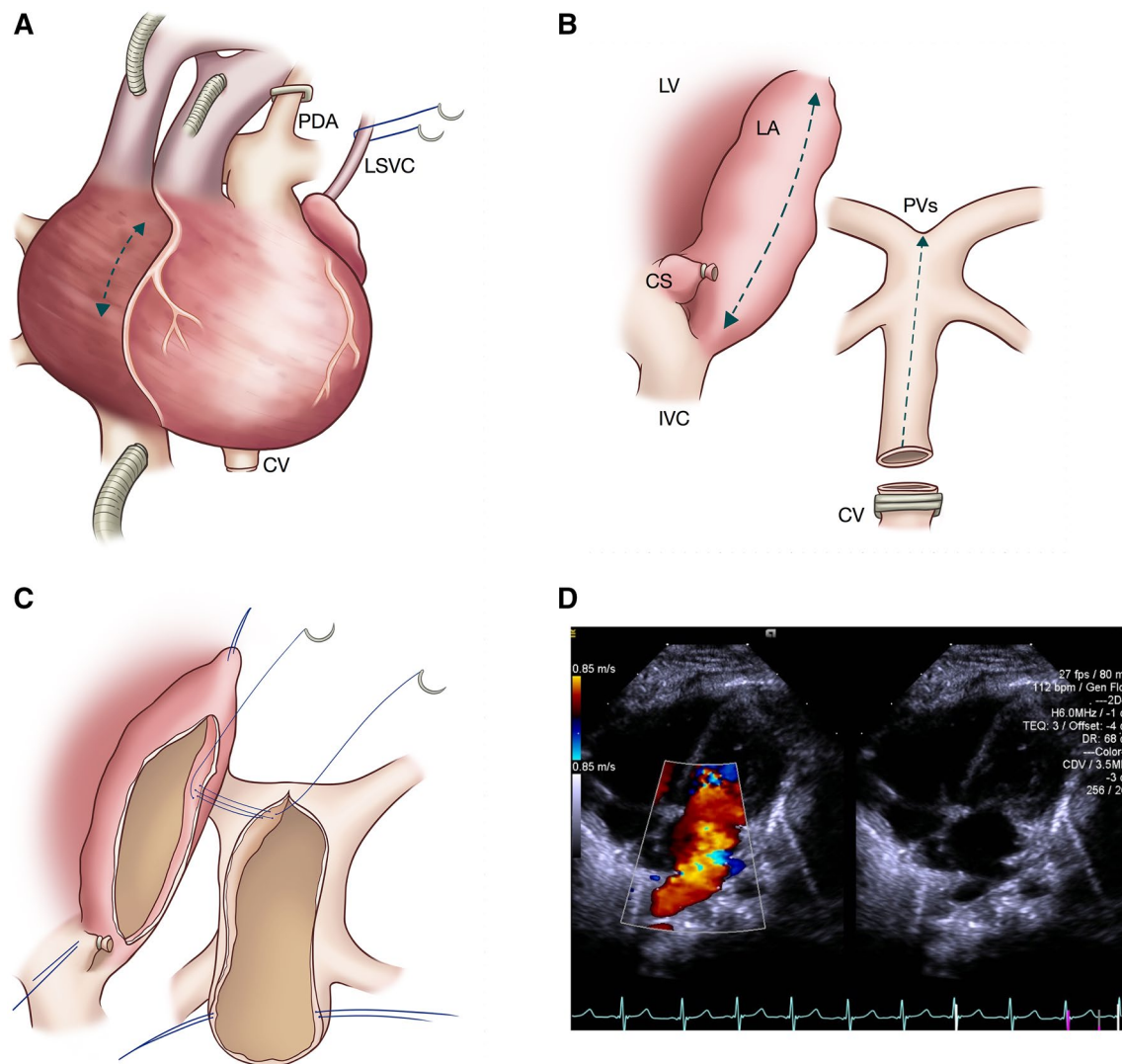
A 9-day-old male weighing 2.5 kg underwent a TAPVD repair. The patient was desaturated preoperatively with an oxygen saturation of 80%. At the surgery, there was the dilated inferior vena cava, and the heart was enlarged (Fig. 1a–c). After the establishment of cardiopulmonary bypass, the connector vein draining toward the diaphragm was ligated and divided at the height of diaphragm. The small left superior vena cava was ligated and divided at the insertion to the coronary sinus. The heart was flipped, and incision lines were drawn between the pulmonary vein chamber and left atrium. Because of enough length of the connector vessel, a sufficient orifice was ensured, which was greater than the mitral valve annulus. A precise intima-to-intima running suture was placed to prevent indentation to the inside of the suture line. The postoperative echocardiography showed the pressure gradient of 0.9 m/s at the anastomosis (Fig. 1d). No PVO was observed during the follow-up.

## Results

In 2 patients of supracardiac type, after the first cardiopulmonary bypass, the trans-esophageal echocardiography showed the flow acceleration greater than 1.5 m/s, the bypass was re-started, an anastomosis was added from the right lateral approach (Fig. 2a). After the revision, the echocardiography showed reduced pressure gradient at the anastomosis (Fig. 2b). These two patients who required the

**Table 1** Diagnosis

| Diagnosis (type)            | Number of patients |
|-----------------------------|--------------------|
| Supracardiac                | 6                  |
| Cardiac                     | 3                  |
| Infracardiac                | 2                  |
| Mixed                       | 4                  |
| Supracardiac + cardiac      | 1                  |
| Supracardiac + infracardiac | 1                  |
| Cardiac                     | 1                  |
| Supracardiac                | 1                  |
| Polysplenia                 | 1                  |

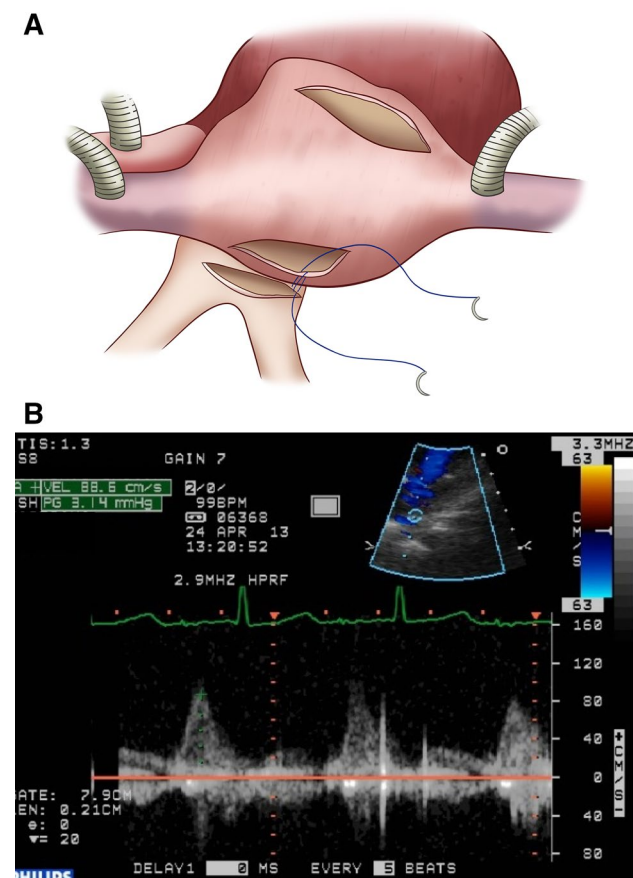


**Fig. 1** **a** Surgical anatomy overview of infracardiac type TAPVD. CV connector vessel, LSVC left superior vena cava (draining into the coronary sinus), PDA patent ductus arteriosus (clipped). **b** The left ventricle was flipped. CS coronary sinus, IVC inferior vena cava, LV left ventricle. Dotted lines indicate incision lines. **c** Anastomosis

additional anastomosis were referred for a surgery with very late diagnosis suffering from pulmonary hypertension; one 3-month-old girl was 5.7 kg, and another 6-month-old boy weighed 5.8 kg, respectively. Both had supracardiac-type TAPVD draining into the innominate vein. For an additional anastomosis, an incision was added on the right lateral side of the heart, from the top to the bottom of the pulmonary venous chamber about 1.5 cm in length. The right side of the left atrium was incised at the opposing line to the pulmonary vein opening. Stitches were always placed carefully needling into the internal side of the intima, needling out to the intima before the edge of the incision. Care must be taken not to cause torture or stress on the opening of the anastomosis.

between the left atrium and pulmonary vein chamber. Note that precise “intima-to intima” stitches are placed, and adequate orifice size greater than mitral valve size is secured. **d** Postoperative echocardiography in four-chamber view. The adequate orifice was confirmed with low velocity

This principle was applied to the opposed side as well. Both patients have not developed PVO in the long-term follow-up including the added orifice without obstruction. There were two patients with purely mixed-type TAPVD: one 13-day-old boy weighing 2.1 kg, with supracardiac draining into the innominate vein in addition to infracardiac draining; another 52-day-old boy weighing 3.1 kg, with cardiac type draining into the coronary sinus in addition to supracardiac draining to the innominate vein. Both anastomoses were created separately with an adequate opening. The former patient had the isolated right lower pulmonary vein drained close to the inferior vena cava that was detached and anastomosed directly to the left atrium posterior to the interatrial septum. This



**Fig. 2** **a** Additional anastomosis between the right aspect of the left atrium and pulmonary vein. **b** Postoperative echocardiography after the additional anastomosis. Reduced pressure gradient across the anastomosis

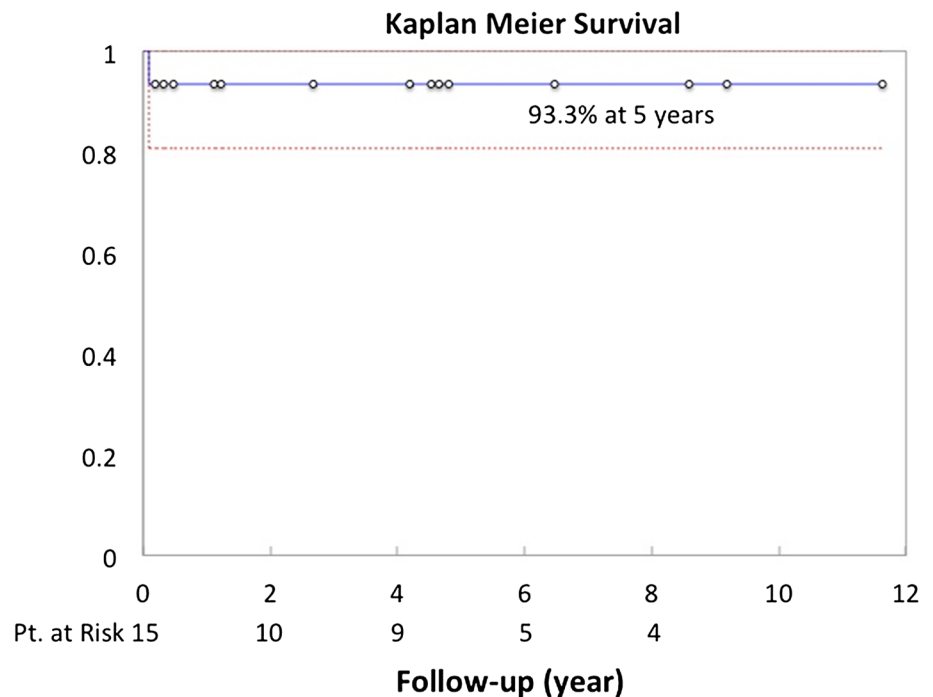
patient developed significant pleural effusion postoperatively and unfortunately died 31 days after the surgery despite mild pulmonary vein stenosis (0.9 m/s). This patient had multiple extracardiac anomalies including tracheal stenosis and preoperative pulmonary hypertension. For the latter patient, the isolated left pulmonary vein was connected to the left atrial appendage that has not shown any vein stenosis. A polysplenia 3.2-kg-girl underwent a cardiac type (coronary sinus drainage) TAPVD repair. A preoperative CT image confirmed polysplenia by bilateral bi-lobed lungs, bilateral superior vena cava, and interruption of the inferior vena cava with the azygos vein connection. Concomitant procedures included 1 ventricular septal defect closure and 1 coarctation of the aorta repair. Mean cardiopulmonary bypass time was  $145 \pm 54$  min while mean cross-clamp time was  $77 \pm 28$  min. Except for one patient who died, follow-up period ranged from 10 months to 12.3 years (mean  $4.6 \pm 3.7$  years); there was no PVO observed, nor reoperation for PVO. The rate of freedom from death or reoperation was 93.3% at 5 years (Fig. 3). Flow acceleration at the surgery was  $0.9 \pm 0.2$  m/s

in average and for those who undergo the echocardiography routinely with concern, the recent velocity was  $1.4 \pm 0.3$  m/s.

## Discussion

Despite the development of surgical and postoperative management, TAPVD repair is one of the congenital heart anomalies which still holds high mortality rate [7]. PVO is an important cause of late mortality in TAPVD [1, 4, 8–10]. The report from the United Kingdom group [8] showed that of 406 patients undergoing repair of TAPVD, 71 (17.5%) had postoperative PVO. Three-year survival for patients with postoperative PVO was 58.7% (95% confidence intervals, 46.2–69.2%). Lately, to prevent postoperative PVO, the primary sutureless technique has been introduced and proved better operative outcomes [6]. Lo Rito and colleagues described that the sutureless group had a lower incidence of PVO of moderate or greater degree (2.9%) than the standard repair group (11.3%) especially in infracardiac and mixed-type TAPVD. Although the primary sutureless technique provides better outcomes, this technique does not necessarily prevent all postoperative PVO. As Hussain [10] described two patients who underwent sutureless technique at the time of initial re-intervention and both were also noted to have very diminutive pulmonary veins extending distally. We have selected the strategy to perform the conventional repair not using the primary sutureless technique for TAPVD. We believe that there should be several technical keys to improve the conventional repair of TAPVD. As described, four surgical tips may have contributed good surgical outcomes. (1) Drawing lines between the facing chambers confirms the natural alignment of the opening, which may lessen the stress to the suture line and decrease flow disturbance. (2) Meticulous intima-to-intima suture will provide smooth endocardial surface wall without indentation inside of the suture line, which is also important to prevent small vortex flow by dents, causing stimulus to inflammation reaction. (3) Creating the orifice size greater than mitral valve is essential for the blood return from the pulmonary vein to the mitral valve with smooth curvature, and to prevent flow acceleration at the anastomosis site. (4) Revision and adding extra-anastomosis after the assessment of trans-esophageal echocardiography is also important. This is the only chance to revise the anastomosis to provide sufficient outcome. This additional anastomosis may lessen the blood flow in each anastomosis and can prevent intima hyperplastic reaction by turbulent flow. When revision is necessary for elevated pressure gradient across the anastomosis, except for finding a detrimental error on the anastomosis that is easy to fix, placing an additional anastomosis can be an option to solve the problem in a

Fig. 3 Kaplan–Meier survival



simple way. Hussain [10] advocated that the mean intraoperative trans-esophageal echo gradient at area of confluence 2.0 mm Hg or greater was associated with the risk of postoperative pulmonary venous stenosis ( $p \leq 0.001$ ). Our criteria, though, peak velocity across the anastomosis is less than 1.5 m/s would be an alternative one since the pulsatile flow is always observed by the contraction of the left atrium. The flow peak velocity at the end of the surgery was  $0.9 \pm 0.2$  m/s in average in our cases. Even for the mixed-type TAPVD, the same principles can be applied: the anastomosis should not be twisted, and an adequate orifice should be confirmed. For the infracardiac type, after the division of the connector vein at the level of the diaphragm, the connector vein can be used to augment the orifice area as Fig. 1b, c. Although a single orifice might be smaller than the expected mitral valve size, keep in mind to create each anastomosis as large as possible. All but one patient did not progress the PVO or necessitated reoperation for PVO. We believe that the above-mentioned surgical strategy have played an important role to prevent the postoperative PVO in any types of TAPVD.

## Conclusions

In conclusion, by applying these above-mentioned surgical tips, the mid-term outcomes after conventional repair of TAPVD were excellent without using the primary sutureless technique to avoid postoperative PVO.

## Compliance with ethical standards

**Conflict of interest** There is no conflict of interest to declare.

## References

1. Yun T-J, Coles JG, Konstantinov IE, Al-Radi OO, Wald RM, Guerra V, et al. Conventional and sutureless techniques for management of the pulmonary veins: evolution of indications from post repair pulmonary vein stenosis to primary pulmonary vein anomalies. *J Thorac Cardiovasc Surg.* 2005;129:167–74.
2. Viola N, Alghamdi AA, Perrin DG, Wilson GJ, Coles JG, Caldaroni CA. Primary pulmonary vein stenosis: the impact of sutureless repair on survival. *J Thorac Cardiovasc Surg.* 2011;142:344–50.
3. Honjo O, Atlin CR, Hamilton BCS, Al-Radi O, Viola N, Coles JG, et al. Primary sutureless repair for infants with mixed total anomalous pulmonary venous drainage. *Ann Thorac Surg.* 2010;90:862–8.
4. Yanagawa B, Alghamdi AA, Dragulescu A, Viola N, Al-Radi OO, Mertens LL, et al. Primary sutureless repair for “simple” total anomalous pulmonary venous connection: midterm results in a single institution. *J Thorac Cardiovasc Surg.* 2011;141:1346–54.
5. Viola N, Caldaroni CA. Surgical repair of post-repair pulmonary vein stenosis using “sutureless” techniques. *Oper Tech Thorac Cardiovasc Surg.* 2011;16:12–21.
6. Rito Lo M, Gazzaz T, Wilder T, Saedi A, Chetan D, Van Arsdell GS, et al. Repair type influences mode of pulmonary vein stenosis in total anomalous pulmonary venous drainage. *Ann Thorac Surg.* 2015;100:654–62.
7. Committee for Scientific Affairs, The Japanese Association for Thoracic Surgery, Masuda M, Okumura M, Doki Y, Endo S, Hirata Y, et al. Thoracic and cardiovascular surgery in Japan during 2014: annual report by The Japanese Association for Thoracic Surgery. *Gen Thorac Cardiovasc Surg* 2016;64:665–97.

8. Seale AN, Uemura H, Webber SA, Partridge J, Roughton M, Ho SY, et al. Total anomalous pulmonary venous connection: outcome of postoperative pulmonary venous obstruction. *J Thorac Cardiovasc Surg.* 2013;145:1255–62.
9. Seale AN, Uemura H, Webber SA, Partridge J, Roughton M, Ho SY, et al. Total anomalous pulmonary venous connection: morphology and outcome from an international population-based study. *Circulation.* 2010;122:2718–26.
10. Husain SA, Maldonado E, Rasch D, Michalek J, Taylor R, Curzon C, et al. Total anomalous pulmonary venous connection: factors associated with mortality and recurrent pulmonary venous obstruction. *Ann Thorac Surg.* 2012;94:825–31 (**discussion 831–2**).