


# Surgery for total anomalous pulmonary venous connection: primary sutureless repair vs. conventional repair

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**Abstract** Despite recent advances in surgical technique and perioperative care, the surgical correction of total anomalous pulmonary venous connection (TAPVC) remains a challenge. The major complication and the main cause of reoperation in TAPVC surgery are the occurrence of pulmonary venous obstruction (PVO). In the 1990s, sutureless repair was introduced as a technique to relieve PVO after TAPVC repair. Following the favorable outcomes for postoperative PVO, the indications for sutureless repair as a primary operation have been expanded to include infants who have preoperative PVO or those at risk of developing PVO after the repair of TAPVC. However, the indications of “prophylactic” primary sutureless repair still remain controversial. Recent studies have shown that normal-risk patients have excellent early and long-term outcomes and a low incidence of reoperation for postoperative PVO. Most patients who survived beyond 2 years after TAPVC surgery were in NYHA class I and offered good outcomes. Although favorable early and mid-term outcomes of primary sutureless repair are reported, the long-term outcomes of this technique are still unclear. The influence of non-contractile pericardial tissue interposed between the PV vessel wall and LA myocardium on

the atrial function is also unclear in patients who undergo sutureless repair. Another disadvantage of primary sutureless repair is potential bleeding from the gap between the confluence and pericardium into the posterior mediastinum or pleural cavity. Thus, it might be best for primary sutureless repair to be indicated for high-risk infants, such as those with TAPVC associated with single-ventricular physiology, mixed-type TAPVC, or small PV confluence.

**Keywords** Pulmonary venous obstruction · Total anomalous pulmonary venous connection · Sutureless repair

## Introduction

Total anomalous pulmonary venous connection (TAPVC) is a cardiac malformation in which there is no direct connection between any pulmonary vein (PV) and the left atrium (LA); rather, all of the pulmonary veins connect to the right atrium or one of its tributaries [1]. Despite recent advances in surgical technique and perioperative care, surgical correction of TAPVC remains a challenge [2, 3]. The major complication and the main cause of reoperation in TAPVC surgery are the occurrence of pulmonary venous obstruction (PVO). Patients with PVO present more with respiratory issues, hypoxia, and even low cardiac output, as well as pulmonary artery hypertension. Both obstructed TAPVC and postoperative PVO are associated with a significant risk of recurrent PVO or death, requiring reoperation for stenosis [4–6].

In the 1990s, sutureless pericardial repair was introduced as a technique to relieve PVO after TAPVC repair [7, 8]. The sutureless repair for pulmonary veins namely, the creation of a neo-LA by anastomosing the LA wall to

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the posterior pericardium rather than to the PV tissue itself was initially used for recurrent PVO. This technique was associated with a favorable survival and freedom from reintervention after the treatment of postoperative PVO [4, 9]. Following these favorable experiences with postoperative PVO, the indications for sutureless repair of the PVs as a primary operation have been expanded to include infants who have preoperative PVO or those at risk of developing PVO after the repair of TAPVC [10–14]. Although the efficacy of the management strategy for postoperative PVO based on the use of a sutureless technique is well recognized, the efficacy of the primary sutureless technique in the prevention of postoperative PVO is not well established. The applicability and indications of this technique are also not well established.

In this review article, we assess the early and late results of conventional and sutureless repair of TAPVC. We also describe our experiences in the management of several types of TAPVC to evaluate the adequate indications of primary sutureless repair.

### Sutureless pericardial repair

Sutureless technique was initially used for recurrent PVO in selected patients. Lacour-Gayet et al. [7] introduced a technique that was based on total resection of PV stenosis scar tissue, and the creation of an opening in the LA wall that was left open. They utilized the acquired adhesions between the posterior LA wall and the pericardium for their procedure. Najm et al. [8] reported a similar technique, leaving in place the pathological stenosis tissue; the obstructive PV lesions were incised longitudinally. The main principle of the sutureless technique is to avoid trauma to the pulmonary venous wall and the endothelium, thereby reducing the risk of subsequent intimal hypertrophy. The sutureline is placed on the pericardium, away from the PV ostium. This allows for complex resection of stenotic PVs extending into the pulmonary hilum without the need to create a correspondingly complex anastomosis. Freed from this anastomotic obligation, the restoration of continuity between the PVs and the LA using an atrio-pericardial anastomosis is actually less technically demanding than a direct anastomosis and, importantly, minimizes the geometric distortion of the PVs. The prevention of geometric distortion of the suture line reduces local turbulence which may cause anastomotic stenosis [4, 11, 14, 15].

The conventional management strategies for postoperative PVO have been associated with poor outcomes, with reports describing mortality rate of >50% [16, 17]. Recent reports have supported the use of the sutureless technique to treat postoperative PVO [7–10, 16, 18, 19]. Hickey et al. [16] reported their experiences with 60 patients with

postoperative PVO who underwent 73 redo operations (33 conventional operation vs. 40 sutureless repair). Freedom from reoperation or death in patients who underwent sutureless repair was markedly higher than those in patients who underwent the conventional repair. Ricci et al. [18] reported their experiences with 20 patients who suffered from postoperative PVO. Seventeen patients underwent the conventional repair, and three underwent sutureless repair. Five patients died and six suffered from PVO after the conventional repair, whereas no death and no PVO occurred in three patients who underwent sutureless repair. Yun et al. [10] reported their experiences of 17 patients with postoperative PVO who underwent 24 procedures (7 sutureless repair and 17 conventional repair). They mentioned that the sutureless technique was associated with decreased risk of reoperation or death when used as the initial procedure for postoperative PVO. Lacour-Gayet et al. [7] described that relief of PVO with sutureless technique was successfully applied in 71% of patients, whereas the relief of PVO with classic procedures failed in 67% of patients.

### Conventional repair of TAPVC as an initial procedure

Since the first attempt to correct TAPVC was reported in 1951 [20], improved surgical techniques and hospital care have led to significantly better outcomes of TAPVC surgery. However, surgical correction remains a challenge. A review of the literature suggests that neonates, patients with mixed-type TAPVC, the presence of PVO, very small LA, and patients with single-ventricular physiology are associated with a high mortality rate [5, 6, 17, 21–24]. Seale et al. [22] reported the clinical outcomes of 422 live-births with TAPVC (cases with univentricular physiology were excluded). A total of 406 of these 422 patients underwent the conventional repair of TAPVC, 58 (14.3%) of whom died. Postoperative PVO occurred in 71 of 406 (17.5%) patients undergoing the conventional repair. The estimated 3-year survival for all surgically treated patients was 85.2%. They concluded that mixed connection or hypoplastic pulmonary veins were major risk factors for the development of postoperative PVO. Yong et al. [17] reported the surgical outcomes of 112 neonates with isolated TAPVC. Eleven (10.7%) patients died in the hospital. There were 6 late deaths at a median time of 8.0 months after TAPVC repair. A postmortem histological examination revealed the presence of thickened pulmonary vessel walls and/or pulmonary lymphangiectasia in their lungs. Thirteen patients underwent a total of 16 reoperations for PVO. The authors mentioned that the risk factors for hospital mortality were urgency of surgery and cardiopulmonary bypass time >65 min, while those for late mortality were low-operative

weight and postoperative episode of pulmonary hypertensive crisis. Kelle et al. [24] reported the results of surgical repair of 100 patients who underwent operation at a single institution. They included 83 biventricular and 17 single-ventricle physiology. An anastomosis was created between the orifice in the posterior aspect of the left (or common) atrium and the common PV. There were 12 operative deaths [4 in the biventricular group (5%) and 8 in the single-ventricle group (47%),  $p < 0.01$ ] and 9 late deaths [6 in the biventricular group (7%) and 3 in the single-ventricle group (18%),  $p < 0.05$ ]. Nineteen patients required reoperation for PVO [14 in the biventricular group (17%) and 5 in the single-ventricle group (29%),  $p < 0.05$ ]. The authors mentioned that patients with biventricular anatomy have excellent outcomes and a low incidence of reoperation for anastomotic or PV stenosis. They also mentioned that patients with functional single-ventricle anatomy have a significantly increased operative and late mortality rates and are at increased risk for reoperation from anastomotic stricture and PV stenosis.

### Primary sutureless repair of TAPVC for high-risk patients

Primary sutureless repair was first described in 2005 [10]. Yun et al. [10] extended the original indication of sutureless pericardial repair to patients without the previous cardiac operations and patients at risk for PVO because of the presence of small PVs. They performed the sutureless technique as a primary procedure in 25 patients with PVO ( $n = 18$ ) or at increased risk of PVO ( $n = 7$ ). They showed that the operative mortality was not increased compared with that of patients who underwent sutureless repair as a reoperation, despite the absence of retrocardiac adhesions. Since then, the indications of “prophylactic” primary sutureless repair have been expanded to include infants at risk of developing PVO after the repair of TAPVC [3, 11–15, 25, 26]. Honjo et al. [14] reported their experience of 22 patients with mixed-type TAPVC. They compared the survival and reintervention rates between the conventional group ( $n = 14$ ) and the sutureless group ( $n = 8$ ). There were five early deaths in the conventional group and no deaths in the sutureless group. Three of fourteen patients (14%)

in the conventional group had PVO requiring reoperation, which was associated with two early deaths, whereas no patient required reintervention in the sutureless group. The authors concluded that the primary sutureless repair for the patients with mixed-type TAPVC appeared to be safe and effective. Matsuhisa et al. [12] applied the primary sutureless technique for a patient with cardiac TAPVC whose right PV was compressed by the posterior wall of the right atrium and atrial septum. They avoided right PVO by performing an anterior translocation of the atrial septum and primary sutureless repair of TAPVC. Kan et al. [13], and Buitrago et al. [25] described some modifications of the primary sutureless technique for patients with infracardiac TAPVC. The repair of TAPVC in patients with single-ventricle physiology, especially in right atrial isomerism, is known to be associated with a significant risk of mortality and recurrent PVO [11, 24, 27]. Yoshimura et al. [11] performed primary sutureless repair for three neonates with right isomerism heart. These three neonates had preoperative PVO and required urgent operation. PVO was successfully released in all three patients. They concluded that the sutureless technique might be useful not only for postoperative PVO but also for non-operated TAPVC in neonates with right isomerism heart. Azakie et al. [15] reported their experience of primary sutureless repair for 18 infants, including 9 with single-ventricle physiology. Only one early death occurred in a patient with right isomerism who succumbed to refractory sepsis. There was one late death after a general surgical procedure. No PVO occurred in these patients. They concluded that the sutureless repair technique could be selectively applied as the primary treatment option in infants with TAPVC at high risk for developing postoperative PVO.

### Our experiences

Between January 2005 and December 2016, 36 patients underwent TAPVC repair at our institution. The patient characteristics are summarized in Table 1. The median age at the initial repair of TAPVC was 7.0 days (range 0–228 days). The median body weight was 3.1 kg (range 1.5–6.3 kg). Single-ventricle physiology was noted in 9

**Table 1** Patient characteristics

	IA	IB	IIA	IIB	III	Total
TAPVC with biventricular anatomy	12	2	3	3	7	27
TAPVC with right isomerism	1	4		1	3	9
Age at the initial repair	0–228 (median 7.0) days					
Body weight at initial repair	1.5–6.3 (median 3.1) kg					

TAPVC total anomalous pulmonary venous connection





PVO. Although PVO was successfully released by sutureless repair in these patients, one died of obstruction of the superior vena cava and innominate vein.

Twenty-one survivors with biventricular anatomy are doing well. Follow-up echocardiography has shown no PVO and no pulmonary hypertension.

### Patients with single-ventricle physiology

Since November 2007, seven patients including five neonates with right isomerism heart have undergone primary sutureless repair of TAPVC. Before then, two other infants underwent a conventional anastomosis between the PV confluence and common atrium at the age of 1 and 7 months, respectively (Table 3).

There were two hospital mortalities in patients who underwent primary sutureless repair. One patient died of septic shock due to necrotizing enterocolitis 2 days after the operation. Another died of congestive heart failure due to severe systemic atrioventricular valve regurgitation 3 months after the primary sutureless repair. One patient suffered from massive bleeding into the left pleural cavity. Massive bleeding from the incised left inferior PV was repaired under cardiopulmonary bypass support. Among the five patients who survived, three underwent Fontan completion, one underwent bidirectional Glenn procedure, and one is awaiting the Glenn procedure. One patient suffered from left-side branch PVO 8 months after the Fontan operation, and successfully underwent sutureless repair of the PVO.

Both the infants who underwent a conventional anastomosis between the PV confluence and common atrium suffered from right-side branch PVO. Both successfully underwent the release of right-side branch PVO using the sutureless repair technique. One patient showed marked improvement in pulmonary hypertension and underwent Fontan completion following a bilateral bidirectional Glenn procedure. Another patient underwent sutureless repair 5 months after the Fontan operation and recovered from protein-losing enteropathy.

**Table 3** Initial repair of TAPVC with right isomerism heart

IA	Primary sutureless repair: 1
IB	Common PV confluence— common atrium anastomosis: 2 Primary sutureless repair: 2
IIB	Primary sutureless repair: 1
III	Primary sutureless repair: 3

TAPVC total anomalous pulmonary venous connection, PV pulmonary vein, PV pulmonary vein, LA left atrium, LA left atrium

Sutureless repair was performed safely not only for post-operative PVO but also for non-operated neonates with right isomerism heart. All but one case did not suffer from PVO after the sutureless repair.

### Discussion

Although improved surgical techniques and hospital care have led to significantly better outcomes of TAPVC surgery, the risk of progressive PVO continues to be a critical problem. Both obstructed TAPVC and postoperative PVO are associated with a significant risk of recurrent obstruction or death, requiring reoperation for stenosis [6]. The long-term survival is determined primarily by early mortality. Late deaths occur but at a much lower rate than early deaths [1]. Recent literatures have shown that normal-risk patients have excellent early and long-term outcomes and a low incidence of reoperation for postoperative PVO [3, 21, 24, 28]. Most patients who survived beyond 2 years after TAPVC surgery were in NYHA class I and showed good outcomes, at least until early adulthood [17]. In our series, the conventional anastomosis between the PV confluence and LA for the patients with biventricular anatomy had good outcomes. No patients suffered from postoperative PVO.

The indications of “prophylactic” primary sutureless repair are still controversial. Recently, favorable early [11–15] and mid-term [10, 11] outcomes of primary sutureless repair have encouraged us to extend the indications of this technique. However, the long-term outcomes of this technique are still unclear. From the tenth week of human cardiac development, the PVs grow and are incorporated into the expanding dorsal wall of the LA. This process results in a smooth-walled LA body consisting of vessel wall tissue on the inner side covered by myocardium on the outside. In hearts with TAPVC, the common PV forms no open connection between the splanchnic plexus and the LA, resulting in the persistence of the cardinal veins draining the pulmonary venous blood to the systemic venous circulation [29]. In the conventional repair of TAPVC, the vessel wall (PV) is directly sutured to the myocardium (LA), whereas non-contractile pericardial tissue is interposed between the PV vessel wall and LA myocardium in the sutureless repair. It is well known that increased atrial compliance facilitates the reservoir function of the atrium and markedly improves the cardiac performance [1].

Another disadvantage of primary sutureless repair is potential bleeding from the gap between the confluence and pericardium into the posterior mediastinum or pleural cavity. We experienced intraoperative massive bleeding into the left pleural cavity in one case. Yun et al. [10] reported four cases in which such bleeding occurred. They

described that the longitudinal incision of the PVs into the pulmonary hilum might be associated with the violation of the thin pleura at the junction between the parietal and the visceral pleura. An incision of the PVs should not exceed the pulmonary hilum. It might be best for primary sutureless repair to be performed only in infants at high risk of developing PVO after the repair of TAPVC.

Infants with single-ventricle physiology especially those with heterotaxy syndrome, are a particularly high-risk group [10, 11, 24, 27, 30, 31]. In such patients, the abnormal positioning of the descending aorta and the PVs causes compression of the PVs between the common atrium and the descending aorta or spine [11]. We experienced two infants who suffered from right branch PVO after the conventional anastomosis between the common PV confluence and common atrium. Their branch PVOs were successfully released by sutureless repair. Neonatal repair of TAPVC in patients with right isomerism heart is associated with a significant risk of recurrent PVO or death. The atrial wall of the right isomerism heart is thick, and the pectinate muscles are extensive all around the vestibule. Direct anastomosis between the common atrial wall and the thin-walled PV confluence is unreasonable [11]. We performed primary sutureless repair of TAPVC for five neonates with right isomerism heart. There were no operative deaths, and only one patient suffered from left branch PVO after Fontan completion. Her left branch PVO was successfully released by redo sutureless repair. Primary sutureless repair should be the procedure of choice in patients with single-ventricle physiology, especially those with heterotaxy syndrome.

The surgical outcomes for mixed-type TAPVC are associated with a relatively high mortality rate and an increased risk of postoperative PVO when compared with other types of TAPVC. The morphologic heterogeneity of the PV connections, incomplete ascertainment of the anatomy, the small size of individual PV confluences, and the high incidence of preoperative PVO make surgical treatment very difficult [14, 23]. Honjo et al. [14] mentioned that the major advantage of the primary sutureless repair for mixed-type TAPVC includes wide applicability to almost any configuration of the PV confluences or individual PVs regardless of the size or location. Aggressive resection of the obstructed PV tissue can be achieved and the remote suture line is simple in geometry, which may prevent surgically induced distortion of the PVs. The authors showed the impact of primary sutureless repair on the survival and reintervention.

There are some case reports of patients with infracardiac TAPVC who underwent primary sutureless repair [13, 25]. The common PV confluence is small and quite low, resulting in discrepancy between the PV confluence and LA in some cases with infracardiac TAPVC. We have performed primary sutureless repair for only one patient

with biventricular anatomy. The PV confluence was small (5 mm in diameter) and quite low, resulting in discrepancy between the PV confluence and LA in this patient. We have to incise out to the individual PVs. In addition, the short PV confluence requires the incision to the individual PVs to make enough communication between PV and LA. These cases might be suitable for primary sutureless repair.

There were five cases with very small LA in our experience (Fig. 2). Four of these five patients died in hospital. There was not enough space to make anastomosis between PV confluence and LA. Very small LA did not function as an adequate reservoir during the postoperative period. Marked pulmonary congestion caused low cardiac output syndrome or severe chylothorax in these patients. Primary sutureless repair cannot be performed in such cases, because LA wall is insufficient to cover the incised pulmonary venous confluence.

## Conclusion

The indications of “prophylactic” primary sutureless repair are still controversial. The early and long-term outcomes of conventional repair of TAPVC for normal-risk patients with biventricular anatomy are excellent. Primary sutureless repair may improve the prognosis of high-risk patients, such as TAPVC associated with single-ventricular physiology, mixed-type TAPVC, or small PV confluence.

## Compliance with ethical standards

**Conflict of interest** The authors have declared that no conflict of interest exists.

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