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Total Anomalous Pulmonary Venous Drainage: Overall Results, Comparison of The Sutureless Versus the Conventional Approach, And Predictors of Adverse Events. A 17 Years Single Centre Experience

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Abstract **Original Research Article**

Introduction: Total Anomalous Pulmonary Venous Drainage (TAPVD) constitutes approximately 1.5% of all congenital heart defects. Surgical correction has traditionally been performed using the conventional technique, however, the sutureless approach has emerged as a recent alternative. This study aims to compare the surgical outcomes of the two techniques and to identify the variables influencing them. ***Methodology:*** This was a retrospective study over a 17-year period from 2007 to 2024. There were 55 patients of TAPVD-supracardiac (n=32), intracardiac (n=18), infracardiac (n=3) and mixed (n=2), ***Results:*** Median age of surgical intervention was 43 days (IQR 30,120), median weight of 3.40 kg (IQR 3.00,4.30). Prematurity was present in 3 patients (5.5%), 11(19.6%) had pre-operative pulmonary vein obstruction, 2(3.6%) had Atrial septal defect (ASD) obstruction, 23 (41.1%) had pre-operative pulmonary hypertension. N=12(21.4%) required pre-operative intubation, 4(7.1%) requiring inhaled nitric oxide, n=7 (12.5%) had hemodynamic instability/cardiopulmonary resuscitation (CPR). Surgical correction (supracardiac and infracardiac) was by conventional repair in 20 patients and sutureless in 15 patients. When comparing sutureless and conventional TAPVD repair techniques, no statistically significant differences were observed in rates of pulmonary hypertension crisis, hospital stay duration, non-invasive ventilation (NIV) duration, postoperative infections, or chest re-exploration rates. Similarly, the incidence of pulmonary venous obstruction (PVO) and early mortality was comparable between the two groups. The predictors of early mortality were seen in patients with pre-operative pulmonary hypertension (OR 5.63, p=0.048), pre-operative intubation (OR 9.29, p=0.008) and pre-operative CPR (OR 36.67, p=<0.001). Incidence of post-operative LCOS was strongly associated with pre-operative pulmonary hypertension (OR 3.90, p=0.02) while predictors of post-operative ECMO predominant in patients with pre-operative PVO (OR 24.57, p=0.007), associated pulmonary abnormality (OR 4.57, p=0.031) and pre-operative intubation (OR 3.88, p=0.05) ***Conclusion:*** Early postoperative outcomes were found to be closely associated with the patients' preoperative clinical status. Overall, in our cohort, no difference was seen between the conventional and sutureless technique of TAPVD repair. These findings may be influenced by the study's limited sample size, potentially reducing its statistical power.

Keywords: Aneurysmal bone cyst, Telangiectatic osteosarcoma, Pathological femoral fracture, Differential diagnosis.

INTRODUCTION

Total Anomalous Pulmonary Venous Drainage (TAPVD) is a rare congenital anomaly, accounting for approximately 1.5% of all congenital heart defects, in which the pulmonary veins fail to connect to the left atrium and instead drain into the systemic venous circulation. This anatomical defect necessitates surgical correction early in life, as untreated TAPVD is fatal due to progressive hypoxemia and right heart failure caused by pulmonary venous obstruction (PVO) and pulmonary hypertension [1,2].

Historically, the conventional surgical repair involves directly anastomosing the pulmonary venous confluence to the left atrium (Figure 1). While effective in re-establishing pulmonary venous return, this technique carries the risk of suture-line stenosis, particularly in neonates and in patients with small or hypoplastic veins. Postoperative PVO is a serious complication that can arise from this technique, with reported incidence ranging between 8% and 21% and a strong association with increased morbidity and mortality [3,4].

In contrast, the sutureless technique (Figure 1) avoids direct suturing of the pulmonary veins by instead anchoring the left atrial wall to the pericardium surrounding the venous confluence, thereby reducing the likelihood of mechanical distortion and subsequent stenosis [5,6]. Several meta-analyses have supported the benefits of the sutureless method, demonstrating lower rates of postoperative PVO and reintervention [7,8]. Moreover, the technique may offer advantages in early mortality, especially in patients presenting with preoperative PVO, mixed-type anatomy, or severe pulmonary hypertension [9].

However, the sutureless approach is not without drawbacks. It can lead to peripheral PVO which, while less severe, may increase the risk of pericardial bleeding or technical complexity in cases with distorted mediastinal anatomy [10]. Furthermore, the long-term implications of using non-contractile pericardial tissue in atrial reconstruction remain unclear, including its impact on left atrial compliance and pulmonary venous hemodynamics.

Given these evolving surgical strategies, there is an ongoing need to define the optimal repair technique based on patient risk profiles and anatomical subtypes. Thus, this study reviews a 17-year single-center experience comparing clinical outcomes between the conventional and sutureless techniques in TAPVD repair. It focuses particularly on rates of PVO, reintervention, perioperative complications, and predictors of early mortality, aiming to identify which surgical method confers better outcomes in specific clinical scenarios.

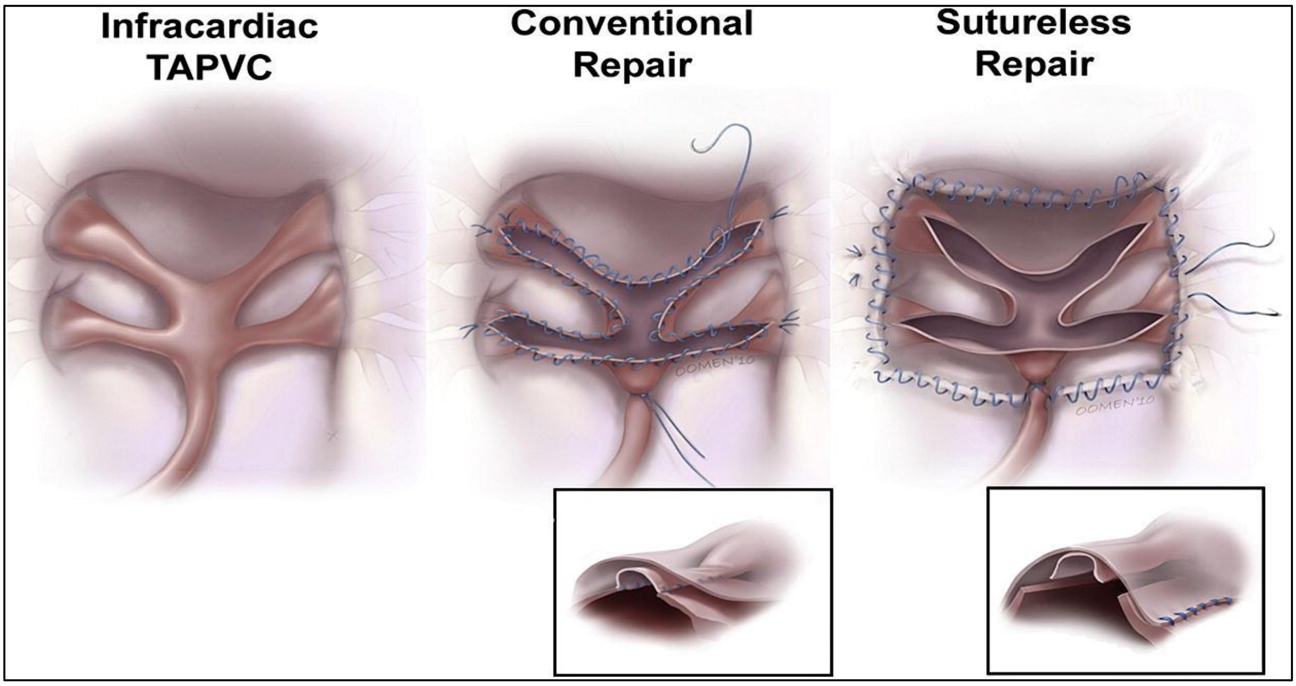

Figure 1: medical illustration comparing infracardiac TAPVD anatomy, conventional repair, and sutureless repair

Oomen MWN, *et al*., Total anomalous pulmonary venous connection: anatomy and surgical repair. Ann Thorac Surg. 2010;89(5):1638. DOI: 10.1016/j.athoracsur.2009.11.007 [11]

Subjects and Methods:

This is a retrospective study which collected and recorded clinical information on patients who underwent TAPVD repair from the year 2007 to 2024. The overall sample size collected in this study was 55. Baseline characteristics were summarized as mean with standard deviations for continuous variables and as count with percentages for categorical variables. Categorical variables were analyzed using the Chi Square test whereas t-test was carried out to assess the distribution of continuous variables. Risk factors against mortality were analyzed using univariate and multivariate analysis. Correlation was used for analysis between continuous data. All analyses were done using IBM SPSS Statistics 27.0 (Statistical Package of Social Science).

RESULTS

Table 1: Demographics

N= 55	Value
Age (n)	
2 weeks and below	7
2 weeks to 1 month	14
1 month to 3 months	18
3 months to 5 months	7
5 months to 1 year	6
Above 1 year	3
Median age (weeks)	43 (30,120)
Gender (n)	
Male	28 (51.0%)
Female	27 (49.0%)
Race (n)	
Malay	39 (71.0%)
Chinese	5 (9.1%)
Indian	11 (20.0%)
Others	0 (0%)
Median Weight (kg)	3.40 (IQR 3.00,4.30)
Prematurity (n)	
Yes	3 (5.5%)
No	52 (94.5%)
Gestational age (n)	
<36 weeks	1 (1.8%)
36 weeks to 36 weeks 6 days	2 (3.6%)
37 weeks to 37 weeks 6 days	15 (27.3%)
38 weeks to 38 weeks 5 days	31 (56.4%)
39 weeks and above	6 (10.9%)
Syndromic (n)	
Yes	4 (7.3%)
No	51 (91.1%)

Table 2: Pre-operative data

N=55	Value
Anatomical type (n)	
Supracardiac	32 (57.2%)
Intracardiac	18 (32.2%)
Infracardiac	3 (5.4%)
Mixed	2 (3.6%)
Pre-operative pulmonary vein obstruction (n)	11 (19.6%)
Restrictive Atrial septal defect (ASD) (n)	2 (3.6%)
Pulmonary hypertension (n)	23 (41.1%)
Single ventricle (n)	1 (1.8%)
Isomerism (n)	1 (1.8%)
Associated cardiac anomalies	9 (16.1%)
Associated pulmonary anomalies	5 (8.9%)
Mean Left ventricular ejection fraction (LVEF) (%)	69.81±8.42
Pre-operative intubation	12 (21.4%)
Pre-operative non-invasive ventilation	4 (7.1%)
Pre-operative nitric oxide	4 (7.1%)
Pre-operative Extracorporeal membrane oxygenation (ECMO)	0 (0%)
Pre-operative Cardiopulmonary resuscitation (CPR)/Haemodynamically instability	7 (12.5%)
Pre-operative surgery/catheterization	4 (7.1%)

Table 3: Intra-operative data

N=55	Value
Mean Cardiopulmonary bypass time (minutes)	136.64±41.99
Mean Cross-clamp time (minutes)	84.24±28.47
Mean Circulatory arrest time (minutes)	0 mins
Redo sternotomy (n)	0
Type of repair (For supracardiac and infracardiac TAPVD only) (n) Conventional Sutureless	n=35 20 (57.14%) 15 (42.86%)
Patent foramen ovale (PFO) left open (n)	31 (55.4%)
Delayed chest closure	32 (57.1%)
Wean off Cardiopulmonary bypass (CPB) with ECMO	1 (1.8%)

Table 4: Post-operative data

N=55	Value
Median Post-op Invasive Ventilation duration (days)	6 (IQR 3,11)
Mean Post-op Non-Invasive Ventilation (NIV) Duration (days)	7.84 ± 21.33
Median Intensive Care Unit (ICU) stay (days)	9 (IQR 6,15)
Median Length of hospital stay (days)	19 (IQR 11,30)
Median Duration chest open (days)	1.00 (IQR 0,2)
Infection/Surgical Site Infection (SSI) (n)	17 (30.4%)
Chest reopens (n)	4 (7.1%)
Arrythmia (n)	14 (25.0%)
Pleural/Pericardial Effusion (n)	11 (19.6%)
Chylothorax (n)	3 (5.4%)
Diaphramatic paralysis	7 (12.5%)
Vocal cord palsy	2 (3.6%)
Low Cardiac Output Syndrome (LCOS)	19 (33.9%)
Post-op Extracorporeal membrane oxygenation (ECMO)	5 (8.9%)
Pulmonary hypertension crisis	37 (66.1%)

Table 5: Mortality and follow up

N=55	Value
Early mortality (n)	8 (14.3%)
Mean time to death (days)	10.88 ±6.31
Discharge to tertiary/referring hospital with Non-invasive ventilation (NIV)/Edotracheal intubation (ETT)	6 (10.7)

Table 6: Post-operative pulmonary vein obstruction

N=55	Value
Post op Pulmonary vein obstruction (PVO) (n)	4 (7.1%)
Reintervention for PVO (Transcath/surgical) (n) Surgical Transcathether	4 (7.1%) 4 0
Surgical/transcathether intervention for asc/dec vein (n)	1 (1.8%)
Surgical/transcathether intervention for Atrial septal defect (ASD) Closure/widening (n)	1 (1.8%)

Table 7: Comparision between conventional and sutureless technique for supracardiac and infracardiac TAPVD

Variable	Sutureless (n = 15)	Conventional (n = 20)	p-value
Mean Cardiopulmonary-bypass time (min)	138.9 ± 21.4	142.9 ± 47.1	0.761
Mean Aortic cross-clamp time (min)	95.9 ± 18.8	83.7 ± 26.5	0.138
Mean Post-op invasive ventilation (days)	8.5 ± 8.8	11.1 ± 12.8	0.519
Mean Post-op non-invasive ventilation (days)	7.1 ± 12.5	11.6 ± 33.7	0.628
Mean ICU stay (days)	11.3 ± 10.2	14.0 ± 13.5	0.524
Mean Hospital stay (days)	26.5 ± 20.8	30.6 ± 38.8	0.719
Infection / SSI (n)	6 (40 %)	8 (40 %)	0.467
Chest re-open (n)	1 (6.7 %)	3 (15 %)	0.619
Arrhythmia (n)	2 (13.3 %)	6 (30 %)	0.419

Variable	Sutureless (n = 15)	Conventional (n = 20)	p-value
Pleural / Pericardial effusion (n)	4 (26.7 %)	5 (25 %)	N.S
Chylothorax (n)	1 (6.7 %)	2 (10 %)	N.S
Diaphragmatic paralysis (n)	1 (6.7 %)	2 (10 %)	N.S
Vocal-cord palsy (n)	0	0	N.S
Low cardiac output syndrome (LCOS) (n)	4 (26.7 %)	8 (40 %)	0.411
Post-op ECMO (n)	2 (13.3 %)	3 (15 %)	N.S
Pulmonary-hypertension crisis (n)	9 (60 %)	14 (70 %)	0.537
Early mortality(n)	3 (20 %)	4 (20 %)	N.S
Post-op pulmonary-vein obstruction (n)	1 (6.7 %)	1 (5 %)	N.S
Re-intervention for PVO(n)	1 (6.7 %)	1 (5 %)	N.S
Ascending/Descending vein intervention (n)	0	0	N.S
ASD closure / widening (n)	0	0	N.S

The median age of patients was 43 weeks with an almost equal gender distribution (51% male and 49% female). 7.3% of patients were syndromic (Table 1). Majority of the patients were of the supracardiac type of 57.2%, followed by intracardiac 32.2%, with infracardiac and mixed types being less common.19.6% had pre-operaive PVO, 41.1% had pulmonary hypertension while 21.4% requiring intubation prior to surgery. A small group of patients had some sort of cardiac anomalies of 16.1% while 8.9% had pulmonary conditions (Table 2)

The mean cardiopulmonary bypass time was 137 minutes with an average cross-clamp time of 84 minutes. Only one patient required intra-operative ECMO support. (Table 3). Postoperatively, patients required a median of 6 days of invasive ventilation, and the median ICU stay was 9 days, with an overall hospital stay averaging 19 days. 30.4% developed infections including surgical site infections (SSI), 25% experienced arrhythmias, effusion affected 19.6% of patients, chylothorax in 5.4%, and 33.9% experienced low cardiac output syndrome (LCOS). Pulmonary hypertension was observed in 66.1% of patients, while 12.5% had diaphragmatic paralysis and 3.6% suffered vocal cord palsy. (Table 4)

The early mortality rate was 14.3%, with patients who did not survive typically passing away within 10.9 days of surgery. A proportion of patients (10.7%) were discharged with continued respiratory support such as non-invasive ventilation (NIV) or endotracheal tube (ETT) to the respective referring hospital for further care. (Table 5) Reintervention was necessary in 7.1% of patients due to post-operative PVO, all managed surgically rather than via catheter-based interventions (Table 6)

The conventional or sutureless technique was performed on two groups of patients which are the supracardiac and infracardiac type. 57.4% of these patients (supracardiac and infracardiac) had conventional repair done while 42.8% had the sutureless repair. When comparing the two primary repair techniques, there was no statistically significant difference in mean CPB times between the two techniques (139.93 mins vs 142.95 mins). The ICU stay, hospital stay and post-operative non-invasive ventilatory duration were also not statistically significant. Patients that underwent sutureless TAPVD repair experienced lower incidence of low cardiac output syndrome (26.7% vs 40%), post-operative ECMO (13.3% vs 15%) and pulmonary hypertension crisis (60% vs 70%) albeit no statistical significance was demonstrated. (Table 7)

DISCUSSION

A total of 55 patients were analysed in this study. The median patient age was 43 weeks. Another similar study by Yong et al yielded a median age of 18 days (1 day to 1 year) which highlighted the need for early TAPVD treatment [12]. As in our series, we believe the delay in operation was mainly due to the delayed diagnosis. Majority of patients were probably asymptomatic at the initial stage. However, once they started becoming symptomatic, that is when the majority of the patient's diagnosis were established and thus referred to a tertiary centre for surgical interventions. The male-to-female ratio (51% male, 49% female) is similar; however, male predominance has been noted in some studies, but data remain inconclusive across broader demographics [13]. The study had 71% Malay patients, reflecting the local demographic profile of patients in Malaysia where the majority of the population are Malays. This cohort's TAPVD types supracardiac (57.2%), intracardiac (32.2%), infracardiac (5.4%), and mixed (3.6%) match existing data, where supracardiac TAPVD is most prevalent type such as that by Al Radi et al [14].

CPB and cross-clamp time averaged 136.64 and 84.24 minutes respectively in this group of patients. Median CPB time for TAPVD repair typically ranges between 60 to 120 minutes while longer CPB times are often associated with complex cases such as infracardiac and mixed TAPVD due to technical challenges [12,15]. Preoperative pulmonary venous obstruction (PVO) significantly impacts CPB time, as additional procedures to relieve obstruction and ensure adequate pulmonary venous flow are necessary [13]. Less complex cases, such as the supracardiac and intracardiac TAPVD, generally have shorter CPB times of around 70–90 minutes due to their less challenging anatomical features. In contrast, infracardiac and mixed TAPVD require more extensive dissection and complex anastomoses, leading to longer CPB durations, often exceeding 100 minutes [16].

In our series, 66.1% of patients developed post-operative pulmonary hypertension crisis. Across literature, pulmonary hypertension is the most common and dreaded post-operative problem occurring in 40-66% of cases [15]. Due to high pulmonary vascular resistance, patients subsequently develop acute right heart failure and systemic hypoperfusion. Nitric oxide, cautious mechanical ventilation, and extracorporeal membrane oxygenation (ECMO) for refractory cases enhance outcomes in high-risk patients [17]. 33.9% of patients developed low cardiac output syndrome (LCOS), which is similar to numbers from other series [18]. We believe the reason behind patients developing LCOS are due to myocardial dysfunction, prolonged cardiopulmonary bypass periods, and inflammatory reactions. The mainstay of treatment for these groups of patients are mainly inotropic support, fluid management and extracorporeal membrane oxygenation (ECMO) whenever necessary.

Arrhythmias occurred in 25% of patients where atrial flutter, junctional ectopic tachycardia, and heart block often develop due to surgery itself, inflammation, or electrolyte imbalances. Overall incidence of sepsis is 30.4%. Infections following TAPVD repair are relatively rare but may occur due to factors such as prolonged surgical time, poor nutritional status, or underlying comorbidities. We believe the higher rates of sepsis (30.4%) is likeley due to the fact that almost half of our patients' chest were closed in a delayed manner. Incidence of pre-operative pulmonary hypertension was 41.1% and 21.4% of pre-operative intubation, we decided to keep the chest open post-operatively to manage the pulmonary hypertension and right ventricular dysfunction. Therefore, once these issues have been resolved, the patient will undergo delayed chest closure and thus, increasing the rate of sepsis. Additionally, due to prolonged hospital stay, these patients are prone to an array of hospital acquired infections. Superficial and deep wound infections have been reported, with rates varying based on institutions and surgical techniques. To minimize surgical site infections, stringent aseptic methods, early antibiotic administration, and attentive wound care are essential.

Postoperative pulmonary venous obstruction (PVO), one of the most feared postoperative consequences, occurred in 7.1% of our patients, which is lower than literature's range 8%-21% of patients undergoing TAPVD repair [19]. Most cases (83%) are usually diagnosed within the first 6 months after surgery, highlighting the importance of early and regular postoperative monitoring [4]. Preoperative PVO is a strong predictor of post-operative PVO, with odds ratios as high as 5.27 [20]. Anatomical subtypes such as mixed-type and infracardiac TAPVD are more prone to developing PVO postoperatively [9]. Factors such as younger age, lower weight, later surgery and prolonged cardiopulmonary bypass (CPB) time further increases the likelihood of PVO [10]. The presence of postoperative PVO significantly reduces survival rates, with a 10-year survival rate of 57.9% in patients with PVO compared to 90.4% in those without [21]. Interventions for PVO include surgical reinterventions and catheter-based therapies.

7.4% of patients that developed PVO post operatively were managed by surgical intervention in our centre. None of the patients underwent cathether based intervention. In general, PVO reinterventions include balloon angioplasty, stenting, and surgery. Despite being less invasive, balloon angioplasty often causes restenosis [22]. Stenting relieves recurrent or severe PVO immediately, however in-stent restenosis can occur, especially in younger groups of patients. For multifocal or severe restenosis, surgical revision, sometimes incorporating reanastomosis or sutureless conversion, is the gold standard [9].

Operative mortality for TAPVD repair varies widely from 2.7% to 35%, depending on factors like the anatomical subtype and the presence of preoperative pulmonary venous obstruction (PVO). This study demonstrated an ealy mortality rate of 14.1% with a mean time to death following surgery of 10.88 days [23]. We believe that the higher mortality rate is likely due to the critical pre-operative condition of the child. Close to 50% of the children had pre-operative pulmonary hypertension with 7.1% requiring nitric oxide and 12.5% had a history of cardiopulmonary resuscitation (CPR). As the median age of diagnosis is 43 weeks in our series, the delay in diagnosis may be a contributing factor leading to this critical pre-operative state. Neonatal surgery is associated with higher mortality (7.9%-13%) compared to infants [12]. PVO is the leading cause of early and late mortality, with a survival rate of 38.6% at 20 years for patients with PVO, compared to 92.2% for those without [19]. Emergency surgery, low body weight (<2.5 kg), and prolonged cardiopulmonary bypass time (>90 minutes) significantly increase mortality risk. After effective repair, 10- and 20-year survival rates are 88% to 90% [12].

In this series, the predictors of early mortality were seen in patients with pre-operative pulmonary hypertension (OR 5.63, p=0.048), pre-operative intubation (OR 9.29, p=0.008) and pre-operative CPR (OR 36.67, p=<0.001). Incidence of post-operative LCOS was strongly associated with pre-operative pulmonary hypertension (OR 3.90, p=0.02) while predictors of need for post-operative ECMO were pre-operative PVO (OR 24.57, p=0.007), associated pulmonary abnormality (OR 4.57, p=0.031) and pre-operative intubation (OR 3.88, p=0.05).

Comparing between sutureless and conventional TAPVD repair in this series, the sutureless technique showed no statistical difference to the conventional group in terms of pulmonary hypertension crisis, hospital stay and NIV duration. Pulmonary venous obstruction (PVO) rates, a major cause of reintervention, were similar in both groups. However, other studies have shown that the sutureless technique has a lower incidence of PVO (3-5% vs 13-17%), reflecting the benefits of a tension-free anastomosis [7]. Conventional repairs are associated with central PVO, while sutureless repairs more commonly result in peripheral PVO, which is generally less severe [20] Wu et al also demonstrated a lower early mortality rate (3-4% vs 5-11%) in the sutureless cohort [8]. While conventional repair remains viable for straightforward cases, its higher complication rates emphasize the need for careful patient selection and management . Yoshimura et al advocates the sutureless techniques for high-risk cases such as pre-operative PVO or mixed/infracardiac types while the conventional technique for low-risk cases [6].

The main drawback in this study is that the retrospective design and single-center setting restrict the study's generalizability. The small sample size reduces statistical power, especially for surgical method subgroup analyses. Larger, multicenter, long-term follow-up studies are needed to improve surgical and postoperative treatment.

Conclusion

In this 17-year study involving 55 patients undergoing total anomalous pulmonary venous drainage (TAPVD) repair, early postoperative outcomes were found to be closely associated with the patients' preoperative clinical status. When comparing sutureless and conventional TAPVD repair techniques, no statistically significant differences were observed in rates of pulmonary hypertension crisis, hospital stay duration, non-invasive ventilation (NIV) duration, postoperative infections, or chest re-exploration. Similarly, the incidence of pulmonary venous obstruction (PVO) and early mortality was comparable between the two groups. These findings may be influenced by the study's limited sample size, potentially reducing its statistical power.

However, preoperative factors such as cardiopulmonary resuscitation, mechanical ventilation, and pulmonary hypertension were identified as strong predictors of adverse outcomes, including the need for extracorporeal membrane oxygenation (ECMO) and the development of low cardiac output syndrome. These results highlight the critical importance of early risk stratification and suggest that while the sutureless technique may not show broad statistical benefits in this cohort, its selective use in high-risk patients could enhance surgical outcomes and reduce postoperative complications.

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