Cardiovascular Topics

Preference for double-patch technique in adult patients with partial anomalous pulmonary venous return

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Abstract

Introduction: Partial anomalous pulmonary venous return (PAPVR) can be treated with either the single- or double-patch technique. In this study, we evaluated the results of both techniques in adult patients with PAPVR.

Methods: We reviewed the records of 29 patients treated for PAPVR in two groups, according to which surgical technique was used.

Results: Double- and single-patch repair techniques were performed in 12 (group 1) and 17 patients (group 2), respectively. In group 2, three patients with a distance of more than 10 mm between the ostium of the anomalous pulmonary vein and the cava–atrial junction needed surgical re-intervention because of superior vena cava stenosis. There was no sinus node dysfunction and no mortality in either group.

Conclusion: In adult patients with PAPVR, the double-patch repair technique revealed better results than the single-patch technique, especially in patients with the ostium of the distal anomalous pulmonary vein more than 10 mm from the cava–atrial junction.

Keywords: atrial septal defect, partial anomalous pulmonary venous connection, superior vena cava syndrome

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Partial anomalous pulmonary venous return (PAPVR) is diagnosed when one or more pulmonary veins drain into the right atrium or systemic circulation.[1] PAPVR is seen in 10% of patients with superior sinus venosus atrial septal defect (ASD) and cases without an ASD are seen rarely.[2]

PAPVR, which is a congenital abnormality, can also be diagnosed in adults.[3] Anomalous pulmonary venous return can be diagnosed by echocardiography, computerised tomography (CT) or magnetic resonance imaging (MRI).[4]

There are different surgical techniques for the correction of PAPVR. Double- or single-patch techniques are the most preferred procedures. Whatever surgical procedures are used, the aim of the surgery is to redirect the anomalous pulmonary veins into the left atrium.[2] In this study, we aimed to evaluate the indications and restrictions of the surgical procedures used for surgical treatment of PAPVR.

Methods

We reviewed patients who were operated on due to PAPVR from May 2014 to November 2020. We included patients older than 18 years with PAPVR. Demographic and clinical information, including echocardiographic reports and surgical procedure, were gained from the medical records. The study protocol was reviewed and approved by the institutional review board (approval no. 2021 /245).

All patients were evaluated pre-operatively via echocardiography. In all patients, intra-cardiac shunt was from left to right and mean pulmonary arterial pressure was below 30 mmHg. Pulmonary angiography was used in five patients and CT was used in 13 patients for the diagnosis. The patients were divided into two groups according to the surgical procedures used. Patients treated with the double-patch technique were in group 1 and those treated with the single-patch procedure were in group 2.

Echocardiography was performed after weaning the patients from cardiopulmonary bypass (CPB) on postoperative day one and before discharge. A mean gradient higher than 3 mmHg between the proximal and distal part of the superior vena cava or in the ostia of the pulmonary veins was considered significant. All patients were monitored in the intensive care unit (ICU). Daily electrocardiography was done to evaluate rhythm disturbances during the hospitalisation period. Surgical and clinical outcomes were compared between the groups.

All operations were performed with a median sternotomy and standard CPB procedure by the same surgeon. One patient in group 1 and two patients in group 2 had a persistent left superior vena cava, but in all cases, the right superior vena cava was dominant. The persistent left superior vena cava that drained into the left atrium was cannulated and drainage was provided with a

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Y-shaped venous line. A pericardial patch was prepared and fixed with glutaraldehyde to avoid calcification and shortening. Both vena cavae were cannulated selectively. After a right atriotomy, the inter-atrial connection, pulmonary venous return, tricuspid leaflets, coronary sinus and conduction route were examined thoroughly.

In group 2, the right atriotomy incision did not extend through to the cava–atrial junction. Only an ASD patch was used to redirect anomalous pulmonary venous drainage to the left atrium. The PAPVR was redirected to the left atrium during repair of the ASD with a pericardial patch. Care was taken to drain all pulmonary veins into the left atrium and not to damage the conduction system.

In group 1, the right atriotomy incision was extended towards the distal end of the abnormally located pulmonary vein and lateral to the superior vena cava, paying attention to the sinus node. The ASD was repaired with a pericardial patch, redirecting anomalous pulmonary venous return to the left atrium. Then the superior vena cava was dilated with a second pericardial patch that repaired the right atriotomy (Fig. 1).

Statistical analysis

Statistical analyses were performed with the IBM Statistical Package for the Social Sciences 24.0 (SPSS 24.0, SPSS Inc, Chicago, IL) software. Categorical variables are reported as frequency and percentage; continuous variables are reported appropriately as mean ± standard deviation (mean ± SD). Groups were compared with the *t*-test and Wilcoxon tests.

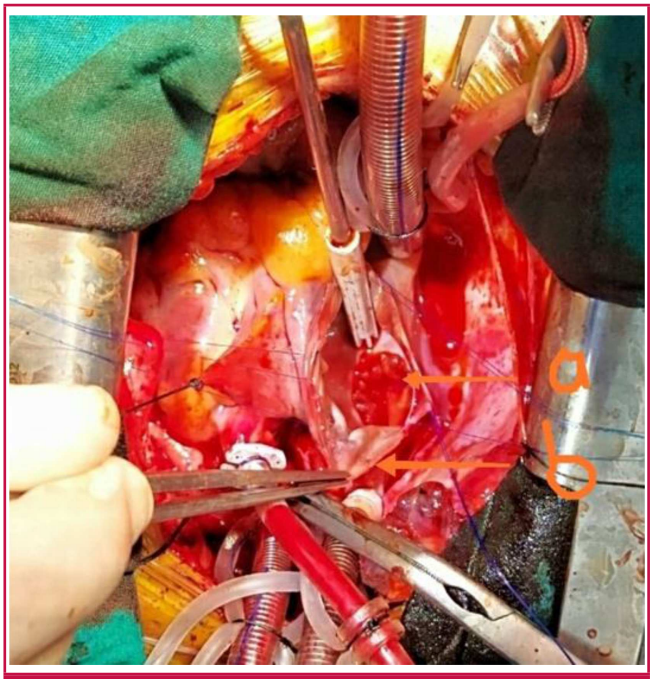

Fig. 1 Double-patch technique. A: first pericardial patch used for the repair of the ASD; B: second pericardial patch used for the repair of the superior vena cava and right atrium.

Results

Twenty-nine patients were enrolled in the study, 12 (41%) in group 1 and 17 (59%) in group 2. There were 13 males (45%) and 16 females (55%). Dyspnoea was the most common symptom pre-operatively. There was no statistically significant difference in terms of demographics and pre-operative variables between the groups ($p < 0.05$) (Table 1).

All patients had an accompanying ASD with the PAPVR. There was no need for additional surgical intervention during the surgery. There was no statistically significant difference between the groups with regard to operative data ($p < 0.05$). In comparison of the groups with regard to postoperative variables, there was no statistically significant difference in intubation period, drainage and length of ICU stay between the two groups ($p < 0.05$). Hospitalisation time was longer in group 2 ($p = 0.017$) (Table 2).

In group 2, three patients necessitated re-intervention due to superior vena cava stenosis. Two were done intra-operatively and one was at postoperative day one. In the evaluation of the possible reasons for stenosis of the superior vena cava, we noticed that the distance between the ostia of the anomalous pulmonary veins and the cava–atrial junction was longer than 10 mm in those three patients.

There was no re-operation caused by haemorrhage and rhythm abnormality. There was no mortality in the postoperative

Table 1. Comparison of pre-operative data

Variables	*Group 1* (n = 12)	*Group 2* (n = 17)	p-*value*
Age (years) mean ± SD	29.1 ± 5.3	27.3 ± 6.2	0.952
Gender (male/female)	5/7	8/9	0.601
EuroSCORE, mean ± SD	2.17 ± 1.12	2.38 ± 1.04	0.345
Hypertension, *n* (%)	3 (25)	5 (29.4)	0.102
Diabetes mellitus, *n* (%)	2 (16.6)	3 (17.6)	0.913
Superior sinus venosus ASD, *n* (%)	10 (83.3)	13 (82.4)	1.000
Mild tricuspid regurgitation, *n* (%)	9 (75)	13 (76.4)	0.815
Mild mitral regurgitation, *n* (%)	2 (16.6)	3 (17.6)	0.913
Ejection fraction, mean ± SD	54 ± 2.7	53 ± 3.2	0.716

ASD: atrial septal defect.

Table 2. Comparison of operative and postoperative data

Variables	*Group 1* (n = 12)	*Group 2* (n = 17)	p-*value*
ACC time (min) mean ± SD	46.3 ± 7.2	49.1 ± 5.6	0.230
CPB time (min) mean ± SD	61.5 ± 11.7	63.2 ± 10.1	0.607
Temperature (°C) mean ± SD	33.5 ± 1.1	33.2 ± 1.3	0.803
Concomitant persistent left vena cava, *n* (%)	1 (8.33)	2 (11.76)	0.092
Inotropic support, *n* (%)	3 (25)	5 (29.4)	0.102
Intubation period (h) mean ± SD	5.4 ± 2.7	6.1 ± 2.3	0.605
Drainage (ml) mean ± SD	570 ± 55	520 ± 80	0.207
ICU stay (days) mean ± SD	2.1 ± 1.1	2.4 ± 0.8	0.095
Hospital stay (days) mean ± SD	6.4 ± 1.3	7.2 ± 1.6	0.017

ACC: aortic cross clamp, CPB: cardiopulmonary bypass, ICU: intensive care unit.

follow-up period. There were no signs of residual ASD or stenosis of the pulmonary vein or superior vena cava during echocardiographic examination in the postoperative first month.

Discussion

Drainage of one or more pulmonary veins into the systemic circulation or the right atrium rather than the left atrium, which is identified as PAPVR, has been reported in 0.4–0.7% of paediatric autopsy series and most were associated with superior sinus venosus ASD. Ho *et al*.[5] declared the rate of PAPVR at 0.1% in the evaluation of 45 538 live cases.

Surgical treatment of PAPVR was first described in 1956.[6] However, there are different surgical approaches for the treatment of PAPVR, with the single- and double-patch repair techniques being the most popular methods. In our department, we perform both single- and double-patch repair techniques, according to the extent of the ASD and ostium of the pulmonary veins. In our retrospective analyses of re-operation rates and causes of the re-operations, we noticed that the distance between the cava–atrial junction and ostium of the anomalous pulmonary vein was a risk factor for caval vein stenosis in the single-patch repair technique. In patients with a distance longer than 10 mm, the double-patch repair technique would be performed.

Transthoracic echocardiography is the first step in the diagnosis of PAPVR. It has advantages in evaluating the atrial septum and pulmonary veins. In some conditions, trans-oesophageal echocardiography may be used. CT and MRI are reliable and useful methods to evaluate pulmonary vascular structures.[7,8] Oxygen saturation assessment with selective pulmonary angiography is the gold-standard method for diagnosis.[9]

We needed pulmonary angiography in five patients and CT in addition to echocardiography in 13 patients for the diagnosis. Most patients with PAPVR have sinus venosus ASD. There are rare PAPVR cases with an intact atrial septum.[10] In our study, 80% of the patients had co-existence of superior sinus venosus ASD with PAPVR.

In patients with anomalous pulmonary veins far from the cava–atrial junction, the surgical approach may be difficult. In our routine practice, we prefer a median sternotomy in sinus venosus ASD cases, since it is easier tunnelling the anomalous pulmonary veins into the left atrium.

Hospital mortality of non-complex congenital cardiac diseases was reported at 2.6–3%. Pulmonary venous stenosis, superior vena cava stenosis and supraventricular arrhythmias are the most important complications in the postoperative period of PAPVR.[11] In our patients, there were three re-operations. There was no mortality, sinus node dysfunction or atrioventricular block. We managed to maintain a normal sinus rhythm with medical therapy for supraventricular arrhythmias in two patients in both groups. The hospitalisation period was longer in group 2 due to re-operation.

There are different surgical techniques for the treatment of PAPVR. The superior vena cava division and re-implantation technique, which is known as the Warden procedure, could avoid pulmonary venous stenosis.[12] However, there are cases where superior vena cava stenosis has been reported after this procedure.[13] In our department we do not perform this technique due to the risk of stenosis of the superior vena cava.

The single-patch technique can be used to redirect the pulmonary veins into the left atrium and dilate the superior vena cava.[14] A separate double-patch technique for closure of the ASD and right atriotomy and correction of the PAPVR may be the easiest procedure to avoid superior vena caval stenosis. In this technique, the atriotomy incision is extended to the lateral side of the superior vena cava to protect the sinus node.

In our department, we prefer the double-patch repair technique in patients with a longer distance between the ostium of the distal anomalous pulmonary vein and the cava–atrial junction. In group 2, three patients needed surgical re-intervention due to superior vena cava stenosis. In our re-operated patients, we measured the distance between the ostium of the distal anomalous pulmonary vein and the cava–atrial junction and found that this distance was more than 10 mm in all of them.

Measurement of this distance by echocardiography or CT may be unavailable pre-operatively. MRI imaging could be helpful. Measurement by the surgeon during the operation is the best way to measure this distance.

This study has limitations. The nature of retrospective studies depends on the availability of information from medical records. We did not have exact indications of the surgical approach for PAPVR repair, however, we prefer the double-patch technique for extension of the defect through to the superior vena. Additional surgical follow up in a larger series is needed to confirm our results.

In our study all patients were older than 18 years and we found that the double-patch technique was more advantageous than the single-patch technique in adult patients whose anomalous pulmonary vein was more than 10 mm from the cava–atrial junction, since it does not cause any pulmonary or systemic venous obstruction. Tissue mobility is greater in children and cardiac dimensions differ in different age groups. Therefore, it is hard to set up a cut-off value for children. Although our study gives an indication for paediatric age groups, studies with a larger series are needed for paediatric age groups.

Conclusion

In the surgical repair of PAPVR, the double-patch repair technique had the advantage of avoiding stenosis of the superior vena cava, especially in patients with a distance between the distal anomalous pulmonary vein ostium and the cava–atrial junction of more than 10 mm. Determination of this distance pre-operatively could be helpful in deciding on the best surgical procedure.

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