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SURGICAL REPAIR OF ISOLATED PARTIAL ANOMALOUS PULMONARY VENOUS CONNECTION WITHOUT ATRIAL SEPTAL DEFECT: A TWO-PATIENT CASE SERIES USING DIFFERENT RECONSTRUCTIVE STRATEGIES

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ABSTRACT

Background: Partial anomalous pulmonary venous connection (PAPVC) without an atrial septal defect (ASD) is uncommon and may remain unrecognized until adulthood. Surgical repair can be challenging when the anomalous pulmonary veins drain high into the superior vena cava (SVC) or when pulmonary venous length is insufficient for direct, tension-free reimplantation.

Case presentation: We report two adults with isolated right-sided PAPVC and an intact atrial septum. A 50-year-old woman presented with exertional dyspnoea, intermittent chest pain and ankle oedema. Imaging demonstrated two anomalous right pulmonary venous channels draining into the SVC. Because direct reimplantation was not feasible, the pulmonary venous confluence was redirected to the left atrium using a 6-mm Dacron interposition graft. A 33-year-old man presented with left-sided chest pain and right atrial and right ventricular dilatation. Two right upper pulmonary venous branches draining near the SVC-right atrial junction were disconnected, fashioned into a common channel and redirected to the left atrium. Both patients had uneventful postoperative recovery. At early follow-up, no pulmonary venous obstruction or residual shunt was identified; Case 1 had documented six-month follow-up.

Conclusion: Isolated PAPVC without ASD requires precise preoperative anatomical definition and an individualized operative strategy. Direct reimplantation is preferable when a wide, tension-free pathway can be achieved, whereas an interposition conduit may be considered in selected anatomies where native venous length is inadequate.

Keywords: partial anomalous pulmonary venous connection; intact atrial septum; superior vena cava; pulmonary vein reimplantation; interposition graft; congenital heart disease

INTRODUCTION

Partial anomalous pulmonary venous connection (PAPVC) is a congenital anomaly in which one or more pulmonary veins drain into the systemic venous circulation rather than the left atrium. PAPVC has been reported in approximately 0.4%-0.7% of autopsy series, and right-sided anomalous drainage into the SVC or right atrium is a common anatomical pattern [1-3]. PAPVC is frequently associated with an ASD; consequently, isolated PAPVC with an intact atrial septum is distinctly uncommon [2,4,6].

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The physiological effect depends on the magnitude of left-to-right shunting. Patients with a small shunt may remain asymptomatic, whereas a significant shunt can produce right-sided volume overload, right ventricular dilatation, pulmonary hypertension, reduced exercise tolerance and atrial arrhythmias [5,11]. Cross-sectional imaging is particularly important in isolated PAPVC because the exact number, course and drainage level of anomalous veins determine the operative strategy [7,8].

We present two adult patients with isolated PAPVC without ASD who underwent surgical correction using two anatomy-driven reconstructive strategies: interposition-graft redirection and pulmonary venous reimplantation.

CASE SERIES AND PREOPERATIVE EVALUATION

This report describes two patients with isolated PAPVC without an associated ASD who underwent surgical correction at our institution. Both patients were evaluated with transthoracic echocardiography and transoesophageal echocardiography to assess cardiac chamber size, ventricular function, pulmonary pressure and the presence of intracardiac shunting. Contrast-enhanced CT coronary angiography and/or cardiac MRI was used to delineate pulmonary venous anatomy and its relationship to the SVC and right atrium. Cardiac catheterization was performed for coronary and haemodynamic assessment. The operative strategy was individualized according to pulmonary venous anatomy and the feasibility of creating a wide, tension-free connection to the left atrium.

CASE 1

A 50-year-old woman presented with exertional dyspnoea, intermittent chest pain and bilateral ankle oedema. Transthoracic echocardiography showed a left ventricular ejection fraction of 55%, no regional wall-motion abnormality, mild mitral regurgitation, an estimated pulmonary artery systolic pressure of 45 mmHg, and right atrial and right ventricular dimensions at the upper limit of normal with preserved right ventricular function. Transoesophageal echocardiography demonstrated no ASD or patent foramen ovale.

Cardiac catheterization demonstrated PAPVC with normal coronary arteries. CT coronary angiography showed right-sided supracardiac PAPVC, with two anomalous pulmonary venous channels draining into the posterior aspect of the SVC: one at the level of the azygos confluence and the second approximately 1 cm caudal to it. The anomalous veins and SVC were well opacified without an obvious filling defect.

Operative technique: Through a median sternotomy, the anomalous pulmonary veins draining into the SVC were identified and the pulmonary venous confluence was dissected. The right atrium was opened to confirm the absence of an ASD. The pulmonary venous confluence was divided from the high SVC. Because the available pulmonary venous length did not permit a tension-free direct connection, a 6-mm Dacron interposition graft was used. The proximal end was anastomosed to the pulmonary venous confluence and the distal end to the left atrium using 6-0 polypropylene sutures, creating an unobstructed pathway. The SVC and right atrial openings were closed.

Outcome: Postoperative recovery was uneventful. At six-month follow-up, no pulmonary venous stenosis or graft-related complication was identified.

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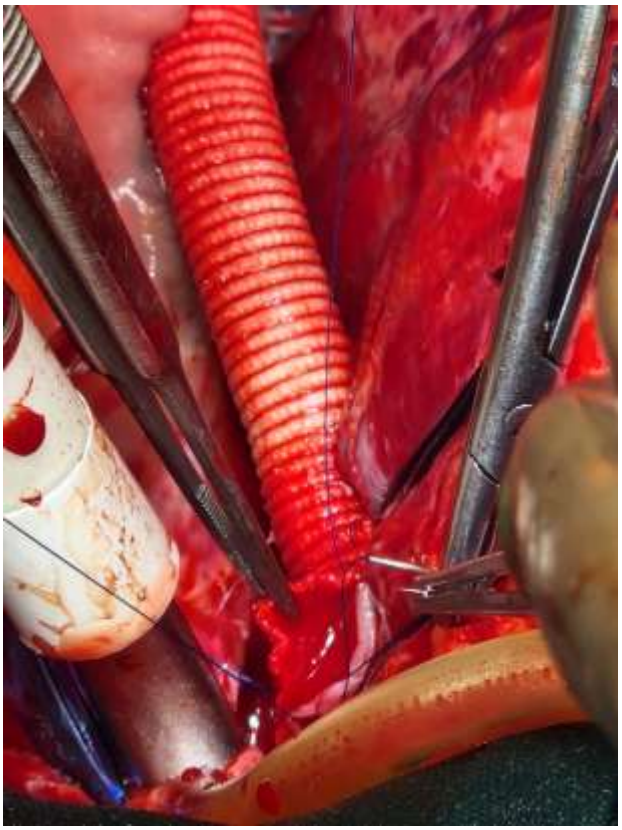

Figure 1. Intraoperative view during surgical correction of isolated PAPVC in Case 1. The anomalous pulmonary venous pathway has been exposed before reconstruction.

CASE 2

A 33-year-old man presented with left-sided chest pain. Transthoracic echocardiography demonstrated right atrial and right ventricular dilatation, a left ventricular ejection fraction of 55% and mild pulmonary hypertension. Transoesophageal echocardiography confirmed right-sided chamber dilatation without an obvious ASD; a bubble-contrast study was negative.

Cardiac catheterization demonstrated PAPVC with normal coronary arteries. CT coronary angiography showed anomalous termination of the right superior pulmonary vein into the SVC, with imaging features of pulmonary hypertension.

Operative technique: Through a median sternotomy, the SVC and pulmonary veins were exposed. Two branches of the right upper pulmonary vein were identified draining into the SVC/right atrial junction. The anomalous branches were isolated and disconnected from the systemic venous side. The two pulmonary venous branches were joined to create a common channel used as part of the reconstruction, and the resulting pathway was reimplanted into the left atrium to establish pulmonary venous drainage. The SVC opening was closed primarily with 6-0 polypropylene sutures.

Outcome: Recovery was uneventful, and no pulmonary venous obstruction was observed on postoperative follow-up. The exact duration of follow-up should be inserted before submission if available.

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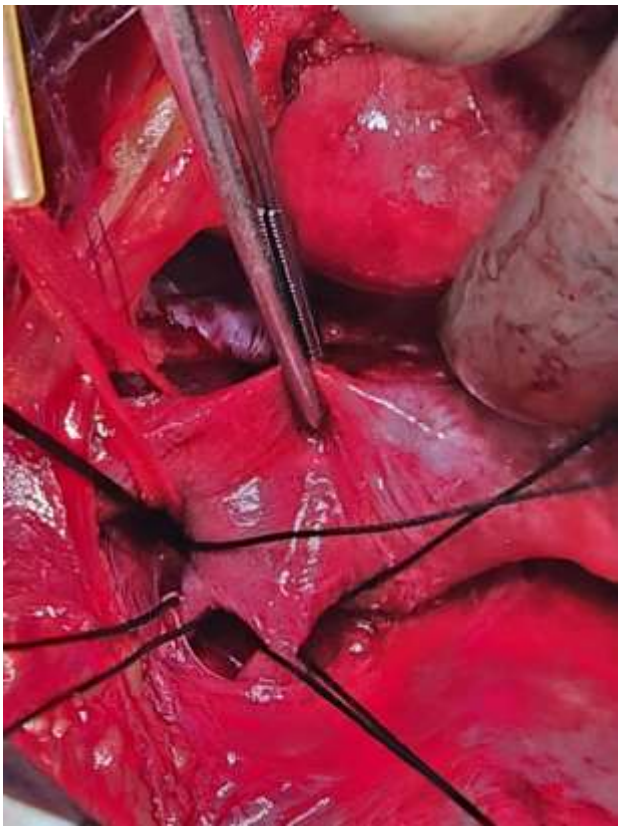

Figure 2. Intraoperative view of the reconstructed pulmonary venous pathway.

TABLE 1. CLINICAL, ANATOMICAL AND OPERATIVE CHARACTERISTICS

Characteristic	Case 1	Case 2
Age/Sex	50 years/Female	33 years/Male
Presentation	Exertional dyspnoea, intermittent chest pain, bilateral ankle oedema	Left-sided chest pain
LVEF	55%	55%
Right heart	RA/RV upper limit of normal; preserved RV function	Dilated RA and RV
Pulmonary pressure	PASP 45 mmHg on echocardiography	Mild pulmonary hypertension
ASD/PFO	Absent on TEE	No ASD; negative bubble study
Anatomy	Two anomalous pulmonary venous channels draining to SVC	Right superior pulmonary venous branches draining to SVC/RA junction
Surgical strategy	6-mm Dacron interposition graft from pulmonary venous confluence to LA	Pulmonary venous branches joined as common channel and redirected/reimplanted to LA
Early outcome	Uneventful	Uneventful
Follow-up	6 months; no pulmonary venous stenosis/graft complication	No pulmonary venous obstruction Was seen

DISCUSSION

Isolated PAPVC with an intact atrial septum is uncommon and may present in adulthood because the absence of an interatrial communication can delay recognition [4,6,11]. Clinical consequences are

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related primarily to the magnitude of anomalous pulmonary blood flow and chronic right-sided volume loading [5,11]. The two patients in this report illustrate that adult presentation may range from symptoms with only borderline right-sided chamber enlargement to clear right atrial and right ventricular dilatation.

Accurate anatomical definition is central to operative planning. Echocardiography establishes the haemodynamic consequences and excludes an ASD, while CT or cardiac MRI defines the number of anomalous veins, their drainage level and the available venous length [7,8]. These anatomical details determine whether direct reimplantation can be achieved or whether an alternative extracardiac reconstruction is required [7,8,13].

Several surgical techniques have been described for PAPVC draining to the SVC, including intracardiac baffling, Warden-type procedures and direct or extracardiac redirection [3,9,10,13]. In patients with an intact atrial septum, an intracardiac baffle may require creation or enlargement of an atrial communication; therefore, an extracardiac approach can be attractive when anatomy permits [6,13]. Regardless of technique, the principal objectives are unobstructed pulmonary venous drainage, avoidance of SVC obstruction, preservation of sinus-node function and creation of a tension-free pathway without kinking [10,13].

The principal feature of this two-patient series is the use of different reconstructive strategies according to anatomy. In Case 1, the anomalous confluence drained high into the SVC and lacked sufficient length for direct reimplantation; an interposition graft therefore provided a tension-free route to the left atrium. In Case 2, the anomalous branches near the SVC-right atrial junction could be mobilized, combined into a common channel and redirected to the left atrium. Both patients had uncomplicated early recovery without documented pulmonary venous obstruction.

The use of a prosthetic conduit in the pulmonary venous circulation warrants careful long-term surveillance because of potential thrombosis, stenosis, intimal hyperplasia or size mismatch [3,13]. The present report is limited by its small sample size and short follow-up, particularly because only six-month follow-up is explicitly documented for Case 1. The patient was discharged on tab Ecospirin and tab warfarin to maintain PT-INR in the therapeutic range of 2 to 3; similarly, Case 2 was also discharged on tab Ecospirin and warfarin. Consequently, these cases demonstrate technical feasibility and favorable early outcomes but cannot establish long-term durability. Serial echocardiography and, when clinically indicated, cross-sectional imaging are important to assess conduit or anastomotic patency [7,8].

CONCLUSION

Isolated PAPVC without ASD is a rare but surgically correctable congenital anomaly [4,6,11]. Multimodality imaging is essential to define pulmonary venous anatomy and select the operative strategy [7,8]. A wide, tension-free direct reimplantation should be used when anatomically feasible; an interposition conduit may be considered in carefully selected patients when pulmonary venous length is inadequate [3,10,13]. Longer follow-up is necessary to establish the durability of prosthetic pulmonary venous reconstruction [3,13].

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DECLARATIONS

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