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Case Report

A Very Rare Case of Bilateral Partial Anomalous Pulmonary Venous Connection in an Adult

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Abstract

Partial anomalous pulmonary venous connection is a condition in which some, but not all, of pulmonary veins connect to the right atrium or its tributaries, rather than to the left atrium. This rare condition more frequently occurs unilaterally, in the right or left pulmonary veins. The occurrence of bilateral partial anomalous connections is a very rare anatomic finding. There was noticed that only the right superior pulmonary vein drains into the left atrium, while the right inferior pulmonary vein and all left pulmonary veins drained into a venous collector, localized extra pericardial in the left side , which drained through a large vertical vein to the innominate vein. To our knowledge, there is only one reported case that is similar to our case. We corrected the anomaly successfully and the patient did a good postoperative course.

Keywords: Adult atrial; Congenital heart disease; Partial anomalous pulmonary venous drainage; Septal defect

Case Presentation

A 44-year-old female was hospitalized in our service because of dyspnea and progressive exercise intolerance. She was diagnosed with large Atrial Septal Defect (ASD) type ostium secundum. She was referred to perform surgery after she was completed with all laboratory examinations and coronary angiography. Echocardiography showed a large atrial septal defect with important right ventricle enlargement and severe tricuspid regurgitation. Pulmonary pressure was measured about 60 mmHg. The intervention was realized through standard median sternotomy under cardiopulmonary by -pass. Closure of ASD with pericardial patch and tricuspid valve annuloplasty with Gore-Tex ring of the was done. Meanwhile, the patient was not disconnected from the heart-lung machine we noticed that the color of the blood in the venous cannula was redder than usual. Transesophageal echocardiography showed an abnormal drainage of pulmonary veins. We explored the outside and inside of the heart. There was noticed that only the right superior pulmonary vein drains into the left atrium , while the right inferior pulmonary vein and all left pulmonary veins drain into a venous collector ,localized extra pericardial in the left side , which drained through a large vertical vein to the innominate vein. Figures 1 and 2 show, respectively, anatomy of the case in the operating room and a schematic presentation of this anatomy. The heart rearrested and the atrial septal defect reopened. We ligated the vertical vein and anastomosed the venous collector with the left atrium appendage (Figures 3,4). The atrial septal defect was reclosed with a pericardial patch and we continued as usually.

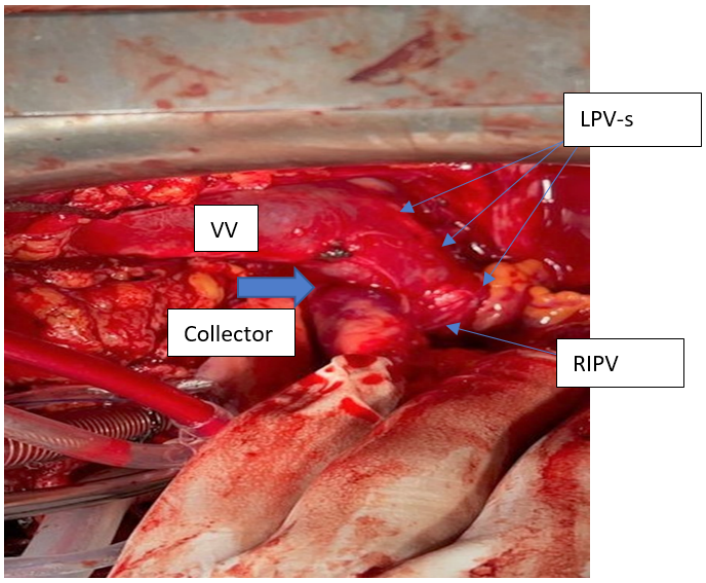

Figure 1: Photo During intervention.Anatomy of anomalous drainage.VV-vertical vein, collector-venous collector, LPV- s left pulmonary veins,RIPV-right inferior pulmonary vein

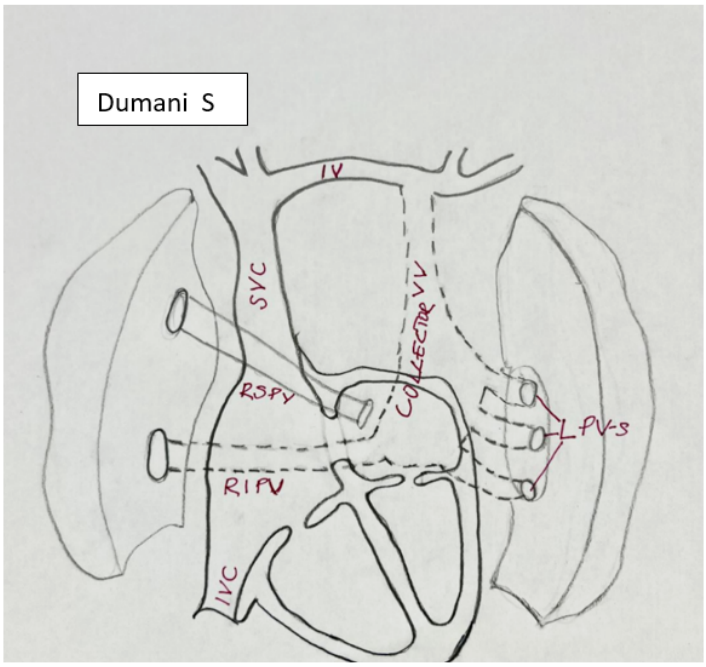

Figure 2: Schematic representation of the pulmonary venous drainage in this case. The anomalous venous drainage; venous collector and vertical vein is in dashed lines

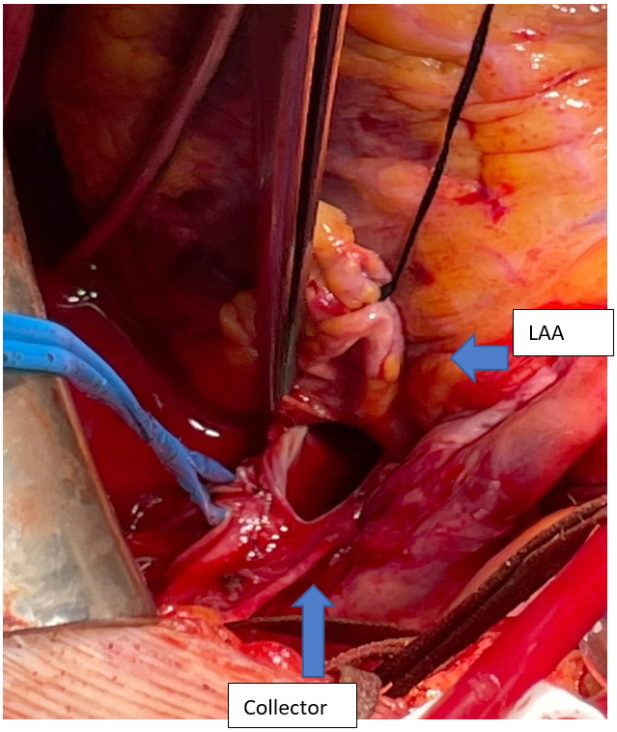

Figure 3: Anastomose between venous collector to left atrial appendage-LAA. The patient did a good postoperative course. We performed Angio Computed Tomography of the heart to check the anatomy after correction.

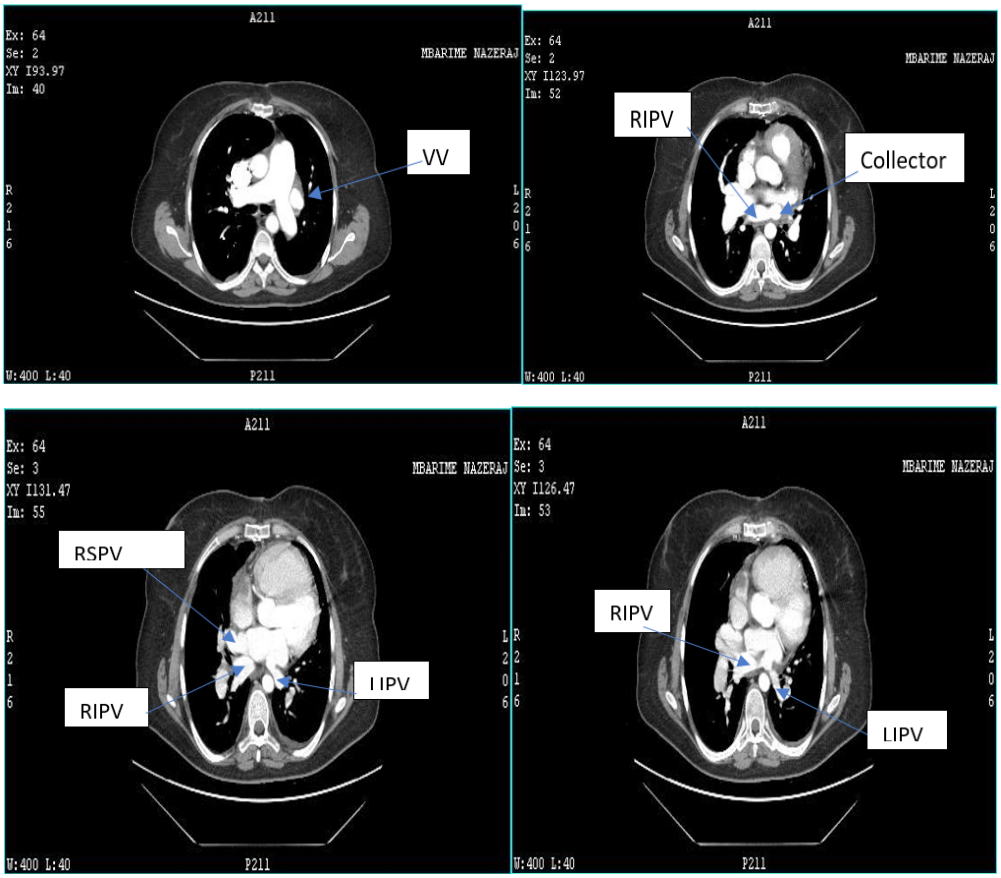

Figure 4: Photo of postoperative computed tomography. VV-vertical vein,RIPV-right inferior pulmonary vein,RSPV-right superior pulmonary vein,LIVP-left inferior pulmonary vein. Post operative echocardiography showed no residual shunt, trivial tricuspid regurgitation and pulmonary pressure about 30 mmHg. The patient is discharged in good health condition.

Discussion

Partial anomalous pulmonary venous connection (PAPVC) is a condition in which some ,but not all, of the pulmonary veins connect to the right atrium or its tributaries ,rather than to the left atrium [1] This rare condition more frequently occurs in the right lung. Anomalous pulmonary veins originating from the left lung have been reported in only 10% to 18% of cases. Bilateral partial anomalous connection is a rare anatomic finding [2-4], accounting for 0.9% to 1.6% of all reported cases. Bilateral anomalous connections are reported in the PAPVC as well, where at least one pulmonary vein from each side drains into a different venous compartment [2] Our case has a very rare anatomy. We found only one case reported in which all the left lung venous drainage and the right medium and inferior pulmonary veins drained into a large and extremely tortuous vertical vein [5]. There was a venous collector in which drained the right inferior and all left pulmonary veins. Right superior pulmonary vein drains normally into the left atrium. Clinical and diagnostic criteria are related to the conditions of left to right shunt. Symptoms may be absent for decades but consist of effort breathlessness and recurrent respiratory infections in infants or signs of chronic heart failure in adults. Diagnostic tools include all imagery studies such as chest radiography ,echocardiography and cardiac catheterization. The last one is not necessary when the diagnosis is sure with noninvasive methods. Presence of ASD or PAPVC with evidence of right ventricle volume overload is an indication for surgery [1] Now day MRI and CT are the methods of choice for demonstration of congenital pulmonary vein anomalies for accurate diagnosis to plan further management [6].

Conclusion

In adults with congenital heart disease catheterization or imaging studies may be reasonable to estimate the cardiac anatomic anomalies or variation and in the operating room we should be prepared for every diagnostic mismatch.

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