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Drenaje venoso anómalo parcial bilateral en paciente adulto: Una rara entidad

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Bilateral Partial Anomalous Venous Drainage in an Adult Patient: a Rare Entity

Partial anomalous pulmonary venous connection (PAPVC) is a rare, often underdiagnosed entity, in which one or more pulmonary veins are connected to the systemic venous return.

We report the case of a 27-year-old female patient with no cardiovascular risk factors and a history of asthma in her childhood.

The patient was clinically symptomatic for progressive dyspnea FCII-III; physical examination revealed wide splitting of R2, ejection systolic murmur +++/++++ at the base, with symmetrical peripheral pulses. Additional tests included an electrocardiogram (ECG) showing signs of right ventricular (RV) overload, and a chest X-ray showing moderate cardiomegaly and enlarged right atrium. Doppler echocardiography revealed enlarged right chambers and extremely enlarged right ventricle with no evidence of intracardiac shunts, indirect signs of moderate systolic pulmonary artery hypertension, and good biventricular function. The suprasternal axis view showed an image consistent with ascending venous flow draining into the innominate vein.

In order to rule out causes for RV overload, an angiotomography was performed that revealed anomalous connection of the right superior pulmonary vein to the superior vena cava, and of the left superior pulmonary vein to the innominate vein (Figure 1 A and B). The upper and mid-right lobe drained into the right superior vein, and the upper lobe and lingula drained into the left superior pulmonary vein. The findings were also confirmed by digital angiography (Figure 1 C and D).

With a diagnosis of bilateral partial pulmonary venous return anomaly, corrective surgery was performed. The anomalies described above were evident during surgery (Figure 2 A and B). This was performed under extracorporeal circulation, which consisted of right superior pulmonary vein tunneling with autologous pericardial patch, creating an atrial septal defect (ASD) to the left atrium (LA) (Figure 2 C); the drainage in the innominate vein was also closed on the left side, and anastomosis of the left superior pulmonary vein to the left atrial appendage was performed (Figure 2 D).

After an uneventful postoperative course, the patient was discharged on the 5th day, with favorable outcome and early return to normal activities. Follow-up computed tomography scan showed good pulmonary drainage reconstruction.

Partial anomalous pulmonary venous connection

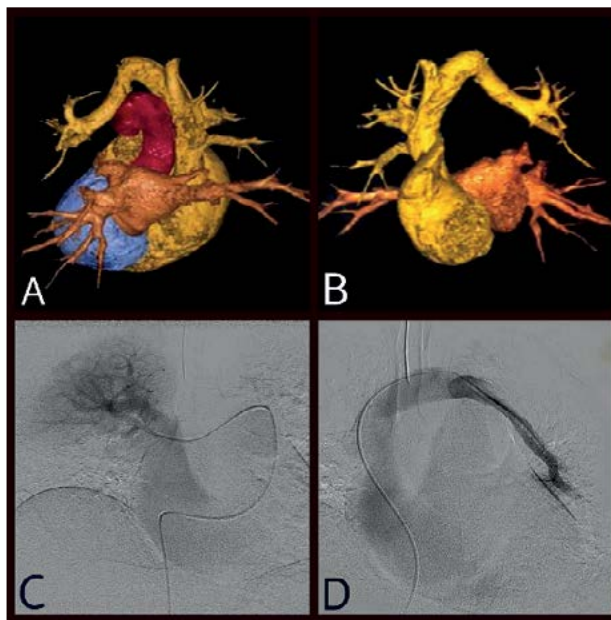


Fig. 1. A and B. Angiotomography reconstruction of cardiac chambers. Digital angiography of right superior pulmonary venous return (C) and left superior and lingula venous return (D).

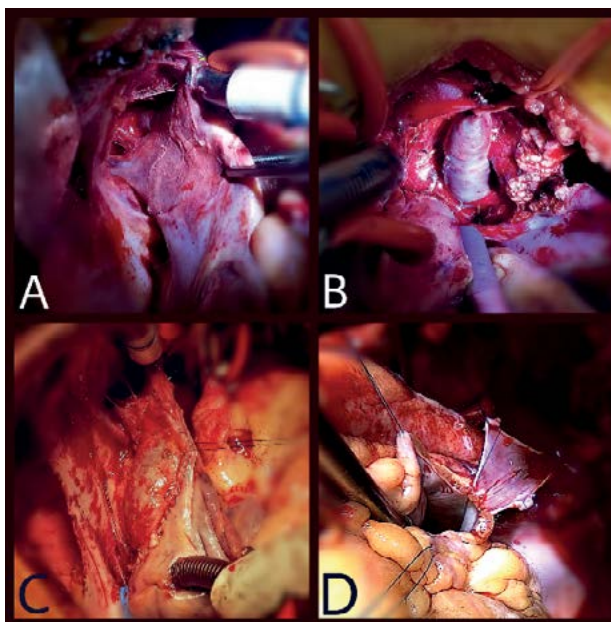


Fig. 2. Anomalous drainage of the right superior (A) and left superior (B) pulmonary veins. Right superior pulmonary vein tunneling with pericardial patch (C) and anastomosis of left superior pulmonary vein to left atrial appendage (D).

is a congenital defect observed in 0.4% - 0.7% of autopsies. (1, 2, 4, 6) This rare condition is characterized by one or more pulmonary veins connected to the right chambers, generally involving right pulmonary veins. Anomalous drainage of the left pulmonary veins has been reported in only 10% - 18% of the cases. (2, 6) Bilateral anomalous connection is a rare anatomical finding that represents 0.9% - 1.6% of the cases reported. (2, 3)

Symptoms and severity of the PAPVC clinical condition relate to the magnitude of left-to-right shunting; (5) dyspnea, exercise intolerance, trepidation, and progressive pulmonary hypertension are its common manifestations. (4, 5, 6) It is often associated with other congenital anomalies such as ASD. The venous sinus type ASD is found in 49% to 85% of cases and the ostium secundum type ASD in 10% to 33%. (5)

In the presence of RV overload and in the absence of intracardiac shunt, PAPVC is one of the diagnoses to be ruled out. Sensitivity of transthoracic Doppler echocardiography is low, and its usefulness to evaluate pulmonary venous return is limited among the adult population. (6) Transesophageal echocardiography, on the other hand, allows a correct assessment of the pulmonary venous return, although it has relative usefulness in case of anomalous drainage in left vertical veins. Magnetic resonance angiography and cardiac magnetic resonance imaging are the non-invasive methods of choice in the evaluation of pulmonary venous return (Class I). (6)

The indication for surgery in PAPVC is controversial. The authors agree that, if there is anomalous drainage in more than one vein, there will be significant hemodynamic consequences; therefore, surgical repair is recommended in those cases. (5, 6) Each pulmonary vein provides between 20% to 25% of the venous flow, so more than one vein with anomalous connection would mean more than 50% involvement. Conversely, if less than 50% of the flow is involved, patients tend to be oligosymptomatic until adulthood, when they will become symptomatic coinciding with increased stiffness of the left ventricle and increased left atrial pressure.

Conversely, yearly screening is indicated in asymptomatic patients with no significant hemodynamic involvement.

We can confirm that surgery is indicated in symptomatic patients or in those with >50% of anomalous venous flow involved, in the presence of a QP/QS >1.5/1, in the case of pulmonary arterial hypertension <50% of the systemic hypertension, and pulmonary resistances lower than 1/3 of systemic resistances.

The correct diagnosis and assessment of the connection sites have important implications for surgical decision.

Different surgical techniques have been described so far, including the division of the superior vena cava, tunneling of the pulmonary veins towards the LA using a pericardial patch (1-patch or double-patch tech-

nique), Warden's surgery, consisting of the division of the superior vena cava (SVC) and its anastomosis in the RA, (4, 6) and modifications of these techniques.

In the case of our patient, two combined techniques were performed for anatomical resolution of the anomaly, with positive outcomes.

In the follow-up of surgical patients, decreased volume and LV remodelling was observed from the time of surgery to an average of 2.5 years later, (6) as well as improved functional class, and a drop of 1-2 mmHg in systolic pulmonary arterial pressure.

Postoperative complications include SVC obstruction (5%), pulmonary veins closely associated to the size of the baffle employed and the use of a double patch (12%). The incidence of tachyarrhythmia or sinus node disease has also been associated with the technique used and to the presence of previous arrhythmia, reaching between 12% and 25% depending on the series. (4)

In the immediate postoperative period, our patient had no complications, with improvement of RV volumes and reduction of systolic pulmonary artery pressure.

In conclusion, the case of this patient with bilateral partial anomalous drainage is a considerably particular entity, especially in adult patients. We believe that intensive assessment and therapy pre-planning is a cornerstone for the resolution of highly uncommon cases, in order to reduce morbidity and optimize long-term outcomes.

Conflicts of interest

None declared.

(See authors' conflicts of interest forms on the website/ Supplementary material).

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Simultaneous Multi-Vessel Coronary Thrombosis Resolved with Rescue Angioplasty

Simultaneous multi-vessel coronary thrombosis is a poorly described entity of ST-segment elevation myocardial infarction (STEMI); consequently, clinical presentation, therapeutic approach and prognosis are not clearly known. Published case reports often describe critically ill patients with electrical and/or hemodynamic instability, most of whom were appropriately revascularized through percutaneous coronary angioplasty showing a low in-hospital mortality. We report the case of a patient with simultaneous thrombosis of the left anterior descending coronary artery and right coronary artery that was opportunely treated at our hospital.

Simultaneous multi-vessel coronary thrombosis is defined as STEMI associated with direct angiographic visualization of two or more thrombi causing partial or complete occlusion of at least two major epicardial coronary arteries. (1) Little is known about its clinical presentation and therapeutic management. Two systematic reviews published in 2009 and 2015 included a population of 47 and 56 patients with unidentifiable etiology of simultaneous multi-vessel coronary thrombosis, which means the population whose myocardial infarction was caused by coronary atherosclerotic plaque rupture but not by other secondary causes. Electrical and hemodynamic instability was the most common clinical presentation, however, there was low percentage of in-hospital mortality. (1, 2)

We describe the case of a 68-year-old man with a history of smoking and type 2 diabetes mellitus, without any other personal or family history of diseases, who presented at the emergency department with one hour episode of chest pain, dyspnea and diaphoresis. On examination, the heart rate was 40 beats per minute (bpm) with mean arterial pressure of 70 mmHg. He was anxious and showed bradycardia with normal breath sounds on auscultation; the rest of the examination was normal. Initial electrocardiogram (ECG) revealed subepicardial lesion of the anterior and inferior wall with right ventricular (RV) involvement, as well as a 3rd degree atrioventricular block (AV) (Figure 1). Treatment with aspirin 300 mg, clopidogrel 300 mg, enoxaparin 30 mg and a weight adjusted single bolus of tenecteplase within the first 15 minutes of the diagnosis of myocardial infarction was administered. However, the patient developed cardiogenic shock and the electrocardiogram did not show any reperfusion criteria after 60 minutes of thrombolysis. At that moment, STEMI risk scores were GRACE 181 points (40% probability of death from admission to 6 months) and TIMI 9 points (35.9% risk of all-cause mortality at 30 days). Therefore, the patient was

transferred to the cardiac catheterization laboratory to undergo a rescue angioplasty supported by norepinephrine and dobutamine. First, a pacemaker electrode was placed in the right ventricle through the femoral vein and was programmed in VVI mode at 70 bpm; coronary angiography showed intraluminal thrombus and 70% stenosis in the proximal segment of the right coronary artery (RCA) with TIMI 2 flow. Simultaneously, the left anterior descending artery (LAD) showed intraluminal thrombus and 80% stenosis in the mid-segment with TIMI 2 flow. Everolimus-eluting stent (Xience-Alpine: 4.0 × 28 mm) was implanted in the RCA and two everolimus-eluting stents (Xience-Alpine: 3.5 × 18 mm and 4.0 × 18 mm) were implanted in the LAD with stent overlap. The final angiography showed both arteries with TIMI 3 flow (Figure 2). Subsequently, the ECG showed normal sinus rhythm and markers of reperfusion with normalization of ST-segment and T wave inversions in the affected leads. The echocardiogram revealed RV dyskinesia with fractional area change of 22% and left ventricular ejection fraction of 46%. The inotropic support and the vasopressor were withdrawn in less than 48 hours. Finally, after cardiac in-hospital rehabilitation, he was discharged 8 days later with medical follow-up in the outpatient cardiology clinic. In the last cardiologic consultation, six months after the event, the patient denies cardiovascular symptoms in his daily activities or during the aerobic exercises, so his condition has a functional class I, according to the New York Heart Association.

Obstructive coronary artery disease in other epicardial arteries unrelated to STEMI is a common finding and has been found approximately in 52.8% of cases. (3) However, simultaneous multi-vessel coronary thrombosis is a rare entity of STEMI, which has been reported in 1.7% to 4.8% of cases. (2) The specific trigger of this condition remains unclear, but a theory suggests that inflammatory pathophysiological processes exert adverse effects throughout the coronary

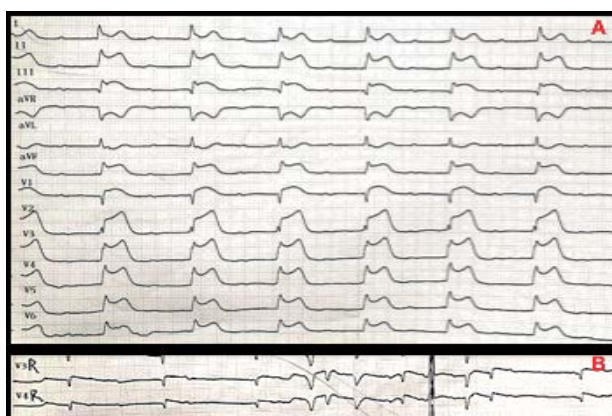


Fig. 1. Initial ECG: **A)** ST-segment elevation in leads DII, DIII, AVF, V1-V6, DI + AV dissociation. **B)** V3R and V4R with ST-segment elevation + Qr + ST.