Case Report

# Coronary-Cameral Fistula and Aneurysmal Dilatation of the Anterior Descending Artery: A Case Report

***Fístula Coronário-Cavitária e Dilatação Aneurismática da Artéria Descendente Anterior. Relato de Caso***

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## Introduction

Fistulas are abnormal communications between coronary arteries and heart chambers and great vessels[1] such as the pulmonary artery or the coronary sinus. These very rare alterations (accounting for approximately 0.4% of congenital heart diseases)[2] were first described by Krause in 1865.[3] The most common connection occurs between the right coronary artery or the anterior descending artery (ADA) and the right heart chambers, and drainage to the left ventricle is extremely rare.[1,3,4]

In general, fistulas are diagnosed through clinical suspicion or findings from examinations while evaluating heart murmurs or clinical symptoms and suspicious signs in the presence of coronary dilation or abnormal flows in heart chambers on echocardiography and angiotomography. Coronary angiography is the standard method, particularly for diagnostic confirmation and differential diagnosis. Clinical manifestations depend on the amount of flow through the fistula on the chamber or vessel and on the degree of ischemia resulting from abnormal communication, which are usually only observed when the flow through the fistula is high,[1,3,4] compromising the patient's quality of life. Dyspnea on exertion, fatigue, and pain in the precordial region, which are related to myocardial ischemia or heart failure, are the most common signs and symptoms. Heart murmur may occur in the sternal border,[1,5] sometimes similar to the murmur sound produced by a patent arterial channel.[1]

Studies have highlighted the importance of acting on cases of fistulas with high output because maintaining intense continuous flows promotes cardiovascular changes secondary to volume overload and hyperdynamic state, eventually compromising lung function. Coronary communications with the right ventricle (RV) function as heart disease with left–right shunt and can cause biventricular overload by increasing the preload. To avoid these and other complications, early percutaneous approach has been indicated to interrupt the pathological communication as soon as the patient is stable and old enough for the procedure.[4,5] In any case, careful clinical follow-up and periodic reassessments are essential.

## Keywords

Coronary-Cavitary Fistula; Aorta, Thoracic; Aneurysm.

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## Objective

Herein, we report a rare case of coronary-cameral fistula (CCF) in a newborn with high output fistula and large ADA dilation and with a distal aneurysm, adjacent to its outflow at the tip of the RV, in addition to coarctation of the aorta (COA), patent ductus arteriosus (PDA), and interatrial communication (IAC).

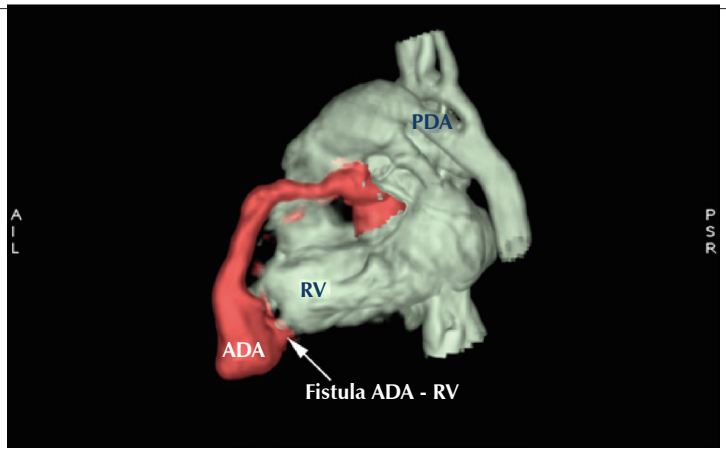
## Case Report

A male patient, born on July 29, 2019, was delivered via cesarean section, at a gestational age of 39 weeks, due to congenital heart disease identified on fetal echocardiography (ECO). Findings in the obstetric examination indicated COA and IAC; nevertheless, the newborn showed good postpartum conditions, weighing 2,820 g and with 9/9 Apgar score. However, respiratory distress occurred after a few hours and, thus, the patient was transferred to the intensive care unit (ICU).

Emergency ECO was performed on his second day of life, identifying a 3.8 mm PDA, 4 mm IAC in the middle third of the interventricular septum, and moderate-to-severe COA (6 mm arc and 2.8 mm isthmus), as well as considerable RV hypertrophy associated with dyskinesia of its apex, which showed a large coronary aneurysm on its surface (Figure 1). Important ADA ectasia was observed throughout its course (Figures 2–4) with evidence of flow from this artery to the tip of the RV and turbulent flow in the RV cavity on Doppler examination (Figures 5 and 6), suggesting a CCF of the ADA in the RV.

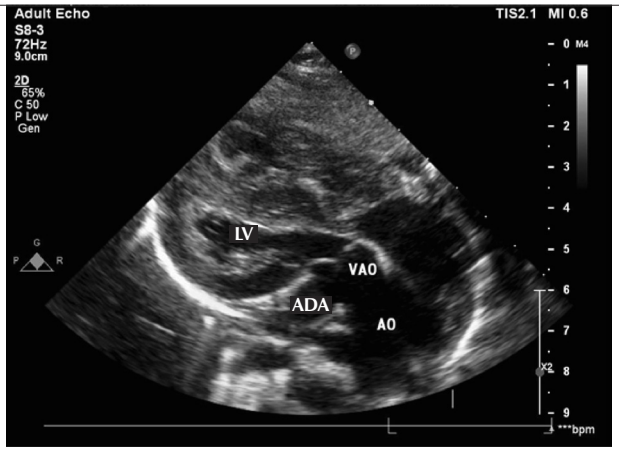
The newborn was under observation in the ICU using prostaglandin to maintain the PCA in order to stabilize his condition, until the surgical intervention was performed on August 6, 2019, when only his COA was corrected. His condition deteriorated hemodynamically during the postoperative period but was subsequently controlled, allowing monitoring in a regular pediatric ward 13 days later. Finally, he was discharged from the hospital, maintaining outpatient follow-ups.

After 4 months, the patient was re-examined, confirming the diagnosis of CCF on angiotomography (Figures 1, 6, and 7) and identifying an abnormal communication between the ADA and RV apex. Images showed ectasia of the ADA 9 mm in diameter in its exit from the left coronary artery trunk (Figures 1, 2, 3, and 7) and expansion when nearing the RV wall, forming a coronary aneurysm adjacent to the mouth in the right ventricular cavity (Figures 1, 4, and 7). Because the patient was oligosymptomatic



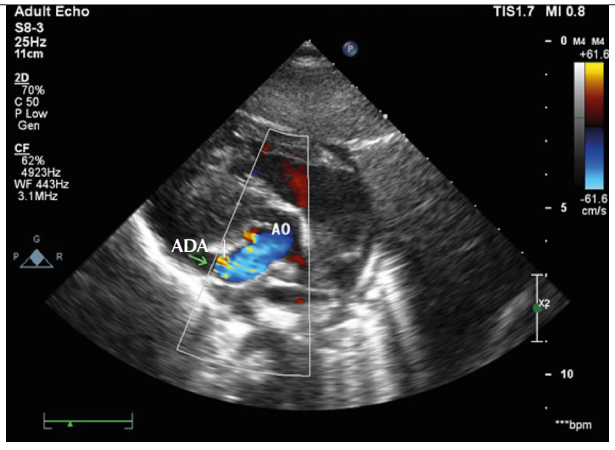

*ADA: anterior descending artery; RV: right ventricle; PDA: patent ductus arteriosus.*

**Figure 1 –** *Three-dimensional heart reconstruction; highlighted in red, the anterior descending artery usually originates from the left main coronary trunk with distal communication to the apical portion of the right ventricle.*



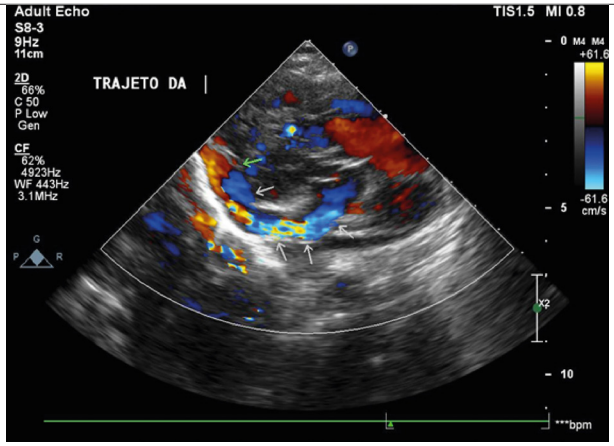

*ADA: anterior descending artery; LV: left ventricle.*

**Figure 2 –** *Subcostal section showing the left ventricular outflow tract and the emergence of the ectatic anterior descending artery.*



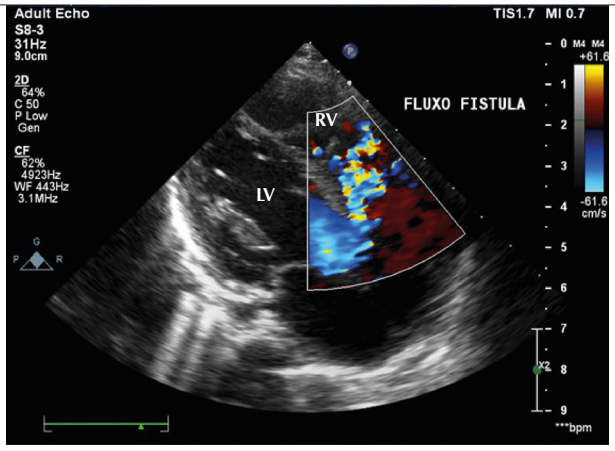

*ADA: anterior descending artery; AO: aorta.*

**Figure 3 –** *Subcostal section showing flow through a color Doppler from the aorta to the dilated anterior descending artery.*



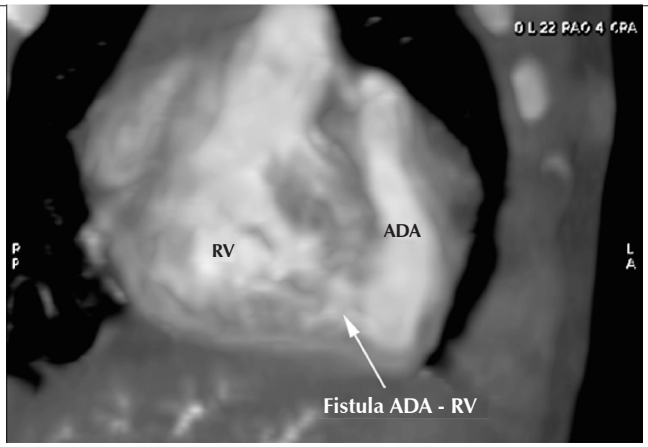

*ADA: anterior descending artery.*

**Figure 4 –** *Modified parasternal section: path of the dilated anterior descending artery.*



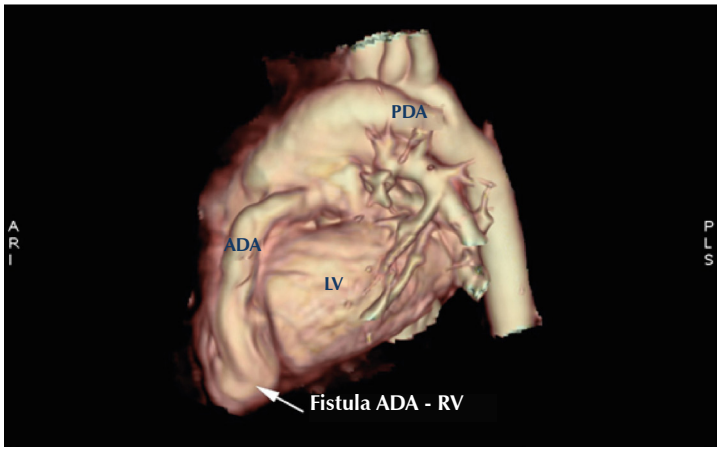

*LV: left ventricle; RV: right ventricle.*

**Figure 5 –** *Apical 4-chamber view showing the turbulent flow of the fistula inside the right ventricle.*




*RV: right ventricle; ADA: anterior descending artery.*

**Figure 6 –** *Multiplanar reconstruction (MPR) highlighting the communication between the anterior descending artery and the right ventricle.*




*ADA: anterior descending artery; PDA: patent ductus arteriosus; RV: right ventricle.*

**Figure 7 –** *Three-dimensional heart reconstruction showing the fistula of the anterior descending artery to the apical portion of the right ventricle, also showing patent ductus arteriosus.*

after the COA correction, he was under observation to ensure a safer future approach and at the right time.

## Discussion

The patient case report highlights the early neonatal diagnosis of a large dilation of the ADA, from its exit in the left coronary artery trunk to its distal portion, where it forms an aneurysm before flowing, as a fistula, into the RV apex, with biventricular overload, signs of high fistulous output in a relatively oligosymptomatic newborn, and a significant association with COA at birth.

High output fistulas can receive up to 50% aortic flow versus approximately 10% normally received by the coronary system. In these fistulas, invasive approaches are considered immediately after the diagnosis to prevent the onset or worsening of symptoms and complications, whereas small fistulas are monitored for years, observing its spontaneous closure.[1,3,4]

High output heart failure, myocardial ischemia, bacterial endocarditis, acute myocardial infarction, atrial fibrillation, and pulmonary arterial hypertension, with reports of coronary steal syndrome, are the most common complications.[2]

## Author contributions

Study conception and design: Oliveira NAM; Data collection: Fusco AS, Oliveira ANM, Cunali VCA, Fernandes FV, Barsam FJG; Data analysis and interpretation: Fusco AS, Oliveira ANM, Fernandes FV; Manuscript writing: Fusco AS, Oliveira ANM; Critical revision of key intellectual content of the manuscript: Fusco AS, Oliveira ANM.

## Conflict of interest

The authors have declared that they have no conflict of interest.

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