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Myocardial Revascularization in an Anterior Descending Coronary Artery of Anomalous Origin Next to the Right Coronary Artery Ostium: Case Report

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1. Summary

The origin of a coronary artery in a sinus other than the usual one is a rare cause of congenital anomaly, estimated at 0.05% to 0.1% for right coronary arteries originating from the left coronary sinus, with an important potential to cause sudden cardiac death. The clinical presentation is generally nonspecific, manifesting as syncope and chest pain on exercise. The diagnosis of this anomaly is difficult, as the individual may be asymptomatic until the lethal event. Once identified, sports should be prohibited and surgical correction, when feasible, restores coronary flow. As for the definitive treatment of anomalous coronary origin, this must be guided by the anomalous artery, its anatomy and implantation, which may include surgery to reconstruct the path, coronary reimplantation, myocardial revascularization and the use of endovascular techniques. Our objective was to report a rare case of anomalous origin of the anterior descending coronary artery next to the ostium of the right coronary artery, associated with episodes of dyspnea, by reviewing the medical records and follow-up of a male patient, aged 72, who was underwent myocardial revascularization surgery with good clinical evolution and is undergoing outpatient follow-up.

2. Introduction

Congenital anomalies of the coronary arteries (CACAs) were first described two millennia ago, by Galen and Vesalius, and are changes in their origin, structure and trajectory [1]. Coronary artery diseases are one of the main causes of morbidity and mortality worldwide. Among these, ACACs, despite being less prevalent, are a potential source of malignant arrhythmias, ischemia and myocardial dysfunction [2]. Coronary artery anomalies result from disorders that occur in the third week of fetal development [3]. The heart originates entirely from the splanchnic mesoderm that forms the floor of the pericardial cavity; This region is known as the cardiogenic area, which gives rise to a pair of endocardial tubes joining together to form the primitive heart tube [4]. The anomalous origin of the coronary artery in the opposite coronary sinus is rare, with an estimated incidence of 0.05% to 0.1% for right coronary arteries originating from the left coronary sinus [5]. Despite their low incidence, they present a high risk of causing sudden death, generally resulting from myocardial ischemia in cumulative outbreaks that, over time, result in scattered fibrosis of the myocardium predisposing to lethal ventricular arrhythmias, by creating an electrically unstable myocardial substrate [6].

ACACs can occur in isolation or be associated with other congenital pathologies, namely transposition of the great vessels, Tetralogy of Fallot and some forms of pulmonary atresia [7,8]. In these cases, symptoms are generally earlier and the diagnosis is made before the patient reaches adulthood. Coronary anomalies are divided into significant or major, those that cause myocardial perfusion disorders, and non-significant or minor, those in which coronary flow is normal. Significant anomalies have a low incidence, corresponding to 0.25% to 0.9% of congenital heart defects [9,10]. Due to this fact, the changes in coronary flow caused by these anomalies and their clinical significance remain unclear [11,12]. It is assumed that the mechanisms involved are three: a) origin at an acute angle and bending or occlusion due to the angulation of the emergence of the coronary arteries; b) coronary spasm due to its torsional movement; c) mechanical compression of the anomalous artery between the pulmonary and aortic artery trunks during effort; the initial portion of the artery may be intramural (within the tunica media of the aorta), which may further aggravate coronary obstruction, especially with aortic expansion during exercise [13,14]. Reports of cases of sudden death related to this anomaly were made by Isner et al. [15] (one case) and Roberts et al. [16] (two cases), suggesting that acute angulation of this artery would lead to interruption of coronary flow and sudden death. The clinical presentation is generally nonspecific, manifesting as syncope and chest pain on exercise. Diagnosis is a challenge, as the individual may be asymptomatic until the lethal event and the physical examination does not reveal changes. Nuclear medicine and magnetic resonance imaging can identify or suspect the existence of these anomalies. Anatomical confirmation is provided by coronary angiography [17]. Once identified, sports should be prohibited and surgical correction, when feasible, restores coronary flow [18]. As for the definitive treatment of anomalous coronary origin, this must be guided by the anomalous artery, its anatomy and implantation. It can be performed through surgery to reconstruct or decompress its path, coronary reimplantation in a suitable coronary sinus, myocardial revascularization and, in some cases, the use of endovascular techniques with stent implantation [19].

ACACs are a heterogeneous group of rare congenital alterations, whose manifestations are very variable. Although most are benign, some are potentially serious. Due to the hemodynamic repercussions that these anomalies present, their early diagnosis and treatment are essential [20]. The objective of this work was to present the case of a patient through investigation of atypical chest pain, with anomalous origin from the anterior descending artery next to the ostium of the right coronary artery and evolving with a successful surgical approach.

3. Methodology

Through review of physical and electronic records of patients treated in a tertiary hospital, in addition to interviews with them and follow-up of the case.

4. Case Description

This is a male patient, 72 years old, with a pathological history of systemic arterial hypertension, type 2 diabetes mellitus, stage 3b chronic kidney disease with a glomerular filtration rate (GFR) of 39 mL/min/1, 73m² (GFR estimated using the CKD-EPI formula) and dyslipidemia. Active smoker and positive family history of early ischemic cardiovascular disease (brother died of acute coronary syndrome at 35 years of age). He regularly used once a day: fenofibrate 200 mg, rosuvastatin 20 mg, dapagliflozin 10 mg, amlodipine 10 mg, bisoprolol 2.5 mg, hydrochlorothiazide 25 mg, acetylsalicylic acid 100 mg, sitagliptin 25 mg twice a day and metformin 500 mg 3 times a day. In 2019, he underwent total arthroplasty of the left knee due to osteoarthrosis and as a postoperative complication he presented distal segmental occlusion of the left popliteal artery, undergoing arterial thrombectomy with current outpatient follow-up with vascular surgery and orthopedics. During outpatient consultations, he complained of pain in the atypical chest region, as a result of which he was referred to a cardiologist, and an exercise test was requested, which was interrupted due to physical fatigue at 07 minutes and 13 seconds of the 3rd Bruce stage, without reaching the submaximal heart rate recommended for age. A stress echocardiography using dobutamine and atropine was then requested, which was interrupted before reaching the submaximal heart rate due to hypokinesia of the anterolateral walls (middle and basal), with a hyperdynamic response from the other walls, concluding with a pharmacological stress echocardiogram with an ischemic response. in the anterolateral wall. Therefore, in cardiac catheterization carried out during hospitalization for renal preparation, due to a GFR of 39 mL/min/1.73m² (GFR estimated using the CKD-EPI formula), coronary circulation was found with two-vessel obstructive lesion: coronary artery anterior descending with anomalous origin next to the right coronary ostium (Figure 1) with 70% stenosis in the proximal third and circumflex artery reaching the middle third of the left atrioventricular groove with 80% stenosis in the middle third (Figure 2). Other coronaries without significant changes to the method. An angioplasty of the lesion in the anterior descending coronary artery was attempted, however, without success, due to significant tortuosity with an angle of around 25 degrees. For therapeutic planning and to exclude a malignant path, the patient underwent tomography angiography of the coronary arteries, which showed an anterior descending artery of great importance, anomalously emerging from the right coronary sinus in an ostium shared with the right coronary artery, following an intramyocardial path through the interventricular septum. basal, becoming epicardial again in the middle interventricular groove, following it until reaching the apex and absence of the left coronary trunk. In a case discussion with the department of cardiovascular surgery at the Hospital Regional de Presidente Prudente, it was decided to undergo myocardial revascularization with a proposal for intervention in the anterior descending artery and,

secondly, scheduling angioplasty for the circumflex artery. The myocardial revascularization procedure was performed uneventfully, with a graft in the left breast and anterior descending artery. After 5 days of the surgical approach, the patient was discharged from the hospital with a scheduled percutaneous approach to the circumflex coronary artery in the 80% residual lesion in the middle third, in a second phase, on an elective basis. At the moment, the patient is being monitored on an outpatient basis in the cardiology department of the Hospital Regional de Presidente Prudente, progressing satisfactorily, uneventfully and asymptomatic.

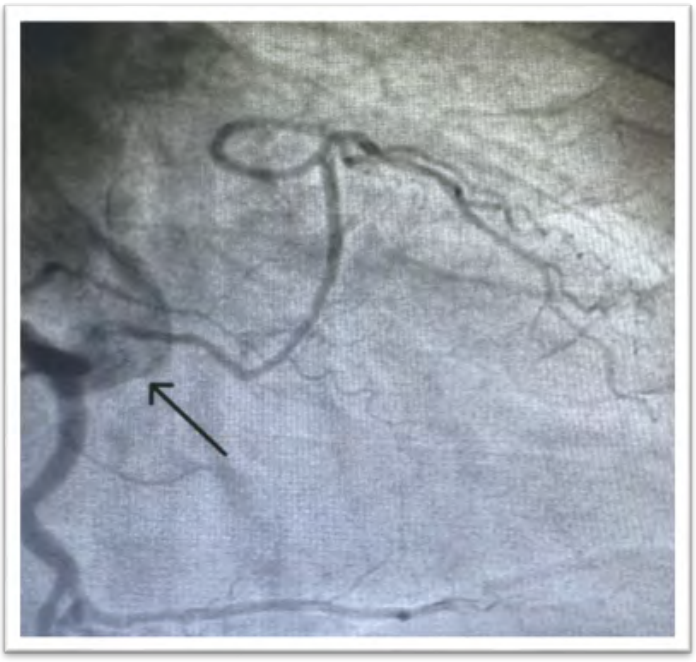

Figure 1: Cardiac catheterization demonstrating anterior descending coronary artery with anomalous origin next to the ostium of the right coronary artery (arrowhead), with 70% stenosis in the proximal third. Source: patient record.

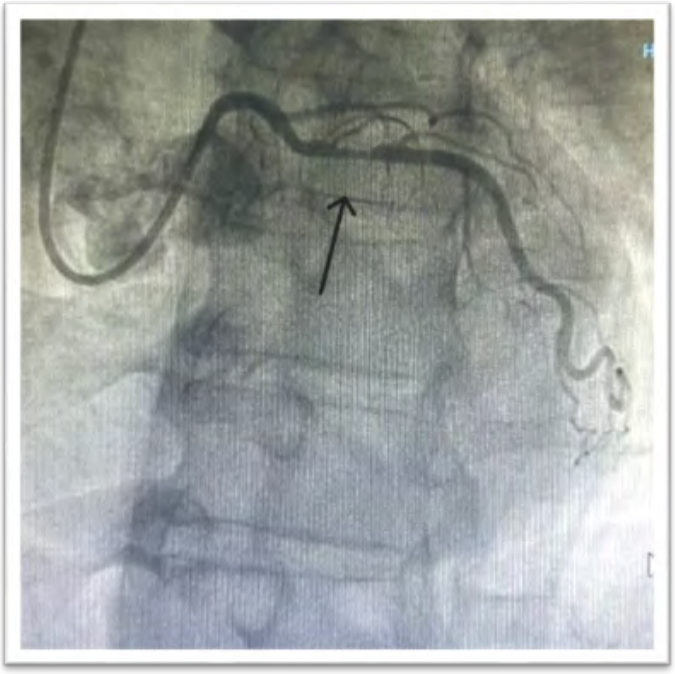

Figure 2: Cardiac catheterization demonstrating circumflex artery reaching the middle third of the left atrioventricular groove with 80% stenosis in the middle third (arrowhead). Source: patient record.

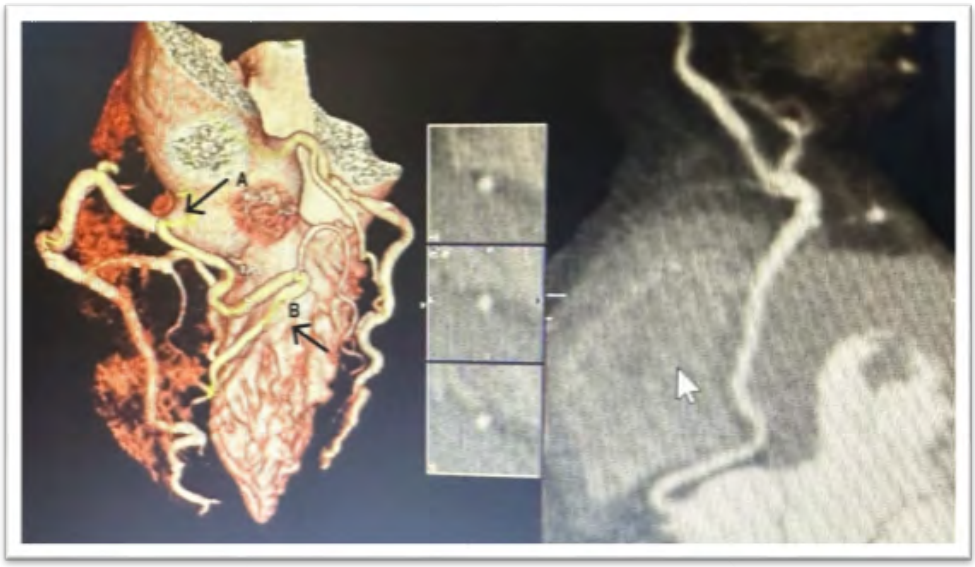

Figure 3: Coronary tomography angiography demonstrating an anterior descending artery of great importance, anomalously emerging from the right coronary sinus (arrowhead A) in an ostium shared with the right coronary artery, following an intramyocardial path (arrowhead B) through the basal interventricular septum, becoming it it becomes epicardial again in the middle interventricular groove, following it until reaching the apex.

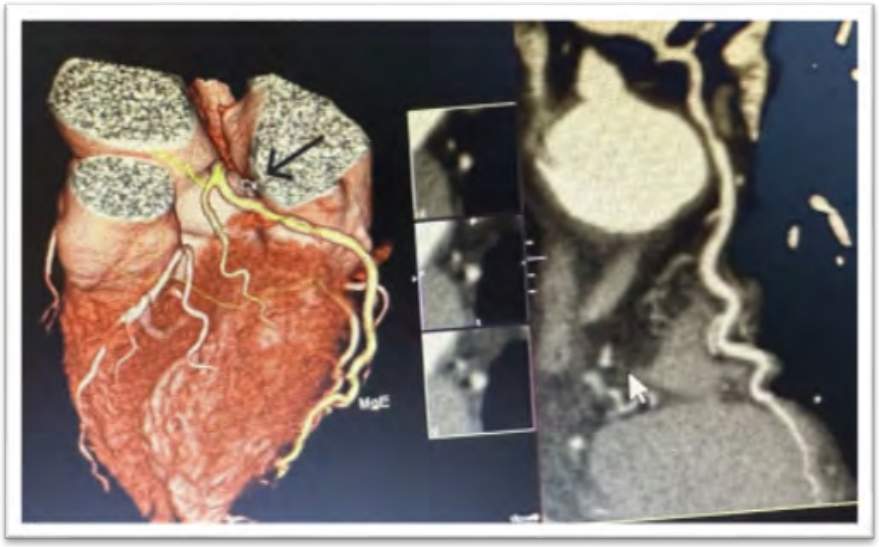

Figure 4: Coronary angiotomography demonstrating circumflex artery (Cx) of moderate importance, which emerges from the left coronary sinus (arrowhead) and reaches the middle third of the left atrioventricular groove, presents a non-calcified plaque after the emergence of the first marginal, determining reduction important luminal.

5. Discussion

Coronary anomaly is an anatomical entity that, in most situations, is associated with a certain congenital heart disease, with no difference between the sexes. With a worldwide incidence of 0.05% to 0.1%, although low, it is a cause of sudden death in young and asymptomatic individuals from a cardiological point of view. Most cases of anomaly of coronary origin produce a reduction in survival, except for the origin of the right coronary artery with the pulmonary artery trunk, a rarer form with a generally benign prognosis [21]. Imaging exams are essential for the diagnosis of ACACs, as it is practically impossible to determine the diagnosis through anamnesis, physical examination and electrocardiogram or even with functional tests. Because of this, it is necessary to find screening methods, ideally non-invasive, as well as defining the target population for this screening. Traditionally, conventional coronary angiography was considered the “gold standard” in the diagnosis of coronary anomalies. However, this is an invasive exam and involves the use of nephrotoxic contrast and ionizing radiation [22]. On the other hand, the evolution of new imaging techniques, namely coronary tomography angiography (CT Angio) and cardiac Magnetic Resonance Imaging (MRI), which allow a three-dimensional assessment of the origin, path of arteries and their relationship with adjacent structures, has highlighted some failures of invasive coronary angiography in the diagnosis [23].

Currently, CT Angio is an important method in the evaluation of coronary arteries, in determining the presence of anomalies in the origin and course of these vessels, and there are several studies that demonstrate its accuracy in this last diagnostic purpose [24-28]. The American Heart Association considers cardiac CT Angio to be an appropriate method for diagnosing ACACs, giving it a score of 9 (maximum classification attributable to a complementary diagnostic method for a given purpose) [29]. The guidelines of the American Heart Association, 2008 Guidelines for the Management of Adults with Congenital Heart Disease, assign a class I indication, level of evidence B, to CT angiography and cardiac MRI in the diagnosis of ACAC.

As for treatment, there are three forms of treatment: 1) observation/drug treatment; 2) angioplasty with stent placement; 3) surgical treatment. Defining the coronary anatomy and relationship with surrounding structures is very important in the preoperative assessment, allowing a correct assessment of the anomaly and planning of surgical repair. In this evaluation, multidetector CT angiography, allowing three-dimensional visualization of the coronary arteries with high spatial resolution, is the method of choice in the study of these patients. According to recommendations from the American College of Cardiology and American Heart Association (ACC/AHA), published in 2008, surgical revascularization is indicated (class I) for: 1) anomaly originating from the left coronary trunk with an interarterial course; 2) anomaly originating from the right coronary artery with an interarterial path associated with evidence of myocardial ischemia; 3) evidence of myocardial ischemia in the territory of the anomalous coronary without another evident causal factor. Furthermore, according to ACC/AHA recommendations, surgical revascularization could be beneficial (class IIa) in: 1) cases of significant stenosis demonstrated by intravascular ultrasound; 2) vascular hypoplasia; 3) coronary compression or signs of coronary stenosis, even without proof of associated ischemia. In recent decades, there has been an increasing evolution in the surgical techniques used to treat coronary artery anomalies. The anomalies that most frequently require surgical intervention include coronary artery fistulas, anomalous origin of the coronary arteries in the pulmonary artery, and anomalous origin of the coronary arteries in the aorta. The choice of surgical intervention for each type of coronary anomaly depends on many variables, namely anatomical, physiological and those associated with the patient. Despite the progression of techniques and improvement in results, there is still controversy regarding the most appropriate treatment for this type of patient.

6. Conclusion

The anomalous origin of the coronary arteries is a rare disease, but potentially lethal if not diagnosed and treated early. Young athletes constitute a risk group for catastrophic consequences. In these patients, surgical therapy brings good results and is the definitive treatment. Identification of the disease in asymptomatic patients remains a challenge, there are several controversies regarding its incidence, classification, screening, heredity and treatment. And new studies are needed to define the ideal treatment for these patients.

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