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Multiple Coronary Aneurysms: A Case Report

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ABSTRACT

Aneurysmal disease of the coronary arteries is a topic specific to pediatric ages in cardiology practice, with secondary causes in most cases completely related to viral infection by an unknown causative agent, in genetically predisposed people, so encountering an adult patient with these pathological characteristics resulted in a cause of academic discussion.

A coronary aneurysm can be defined as a dilation of an arterial segment greater than 1.5 times the diameter of the adjacent normal-sized arteries, taking into account that the diameter of the coronary arteries ranges from 1.5 to 6 mm for the right coronary arteries (RCA) and left coronary arteries, respectively. The left coronary artery (LC) is 60% larger than the RCA. They can be the result of coronary atherosclerosis, congenital, secondary to connective tissue diseases, or infections.

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Introduction

In 2017, a clear distinction was established between the two phenotypes through angiographic studies in selected patients. Coronary aneurysm was defined as focal dilation >1.5 times the adjacent diameter of the same artery with a normal diameter, and coronary ectasia was defined as more diffuse dilation involving the same vascular bed [1].

Differential diagnoses for coronary aneurysms include those caused by atherosclerotic disease, congenital aneurysms, and cocaine-induced aneurysms [2]. Involvement can be diffuse, affecting the entire course of the artery, or localized, affecting only a segment [3].

In patients who develop aneurysms, neointimal proliferation produces a pseudo-normalization of the lumen, associated with thrombi, calcification and stenosis, especially in aneurysms larger than 6 to 8 mm in diameter. As a consequence of these phenomena, ischemia occurs, patients may have angina, acute coronary syndrome (ACS), serious arrhythmias and/or death. In a young adult presenting with ACS, KD should be considered as a cause, especially if the coronary artery study reveals an aneurysm or aneurysms, which may be calcified, even more so if there are no risk factors [3].

The clinical case of a young male patient is presented. The only relevant history is hospitalization due to acute myocarditis in December 2022. He was symptomatic until the date of admission to our center due to atypical precordial pain that had been developing for months. His complementary examinations revealed multiple coronary aneurysms with a calcified thrombus inside one of them in the DA, with proximal occlusion of said vessel. CRM was decided upon with subsequent favorable recovery and he was discharged.

Clinical Case

A 19-year-old male patient with no known cardiovascular or childhood-juvenile history, symptomatic with atypical precordial pain of 2 months' duration, with stabbing characteristics, located in the precordial region, variable functional class, without irradiation or neurovegetative cortex accompanied by dyspnea, consulted at a hospital due to experiencing angina of greater intensity, the admission electrocardiogram (ECG) showed repolarization disorders with negative T waves in the precordial leads without dynamic changes in subsequent records, positive myocardial damage enzymes were dosed, CPK 212U/L, CK-MB 18.4, Troponin -TNT 181pg/ml, negative acute phase reactants, the picture was assumed to be probable acute myocarditis due to the patient's age and absence of risk factors. A Doppler echocardiogram was performed with the following positive findings: slightly impaired left ventricular systolic function (LVSF) with ejection fraction (EF) 49%, mild global hypokinesia, minimal mitral regurgitation, hyperrefringent pericardium at the posterior level with minimal pericardial effusion. To evaluate myocardial tissue, a cardiac nuclear resonance (C-MRI) with gadolinium was requested, which highlighted "increased signal intensity in the T2 sequences at the lower and inferolateral basal level, compatible with myocardial edema, fibrosis of non-ischemic origin, findings compatible with acute myocarditis". Symptomatic treatment was performed with improvement of the clinical picture and the patient was discharged from the hospital with a request for a new cardiac resonance in 4 months.

The patient presented to the emergency department of the same hospital 4 months later due to presenting 48 hours prior to the consultation with precordial pain similar to those mentioned above, which resolved with analgesics. An ergometry test was requested, which was performed on a careful outpatient basis, where "no significant electrocardiographic alterations suggesting ischemia were evident."

Due to the persistence of anginal symptoms, he repeatedly consulted the emergency service 5 days later, with no changes to the ECG (Figure. No. 1) from his first admission, negative cardiac enzymes, treatment with analgesics was started with a good response, interconsultation with the rheumatology service was suggested and a new follow-up MRI-C showed "absence of late gadolinium enhancement, improvement compared to the previous study", assuming the acute condition was resolved.

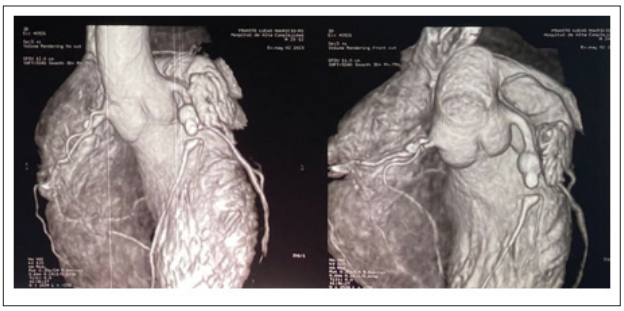

Figure 1

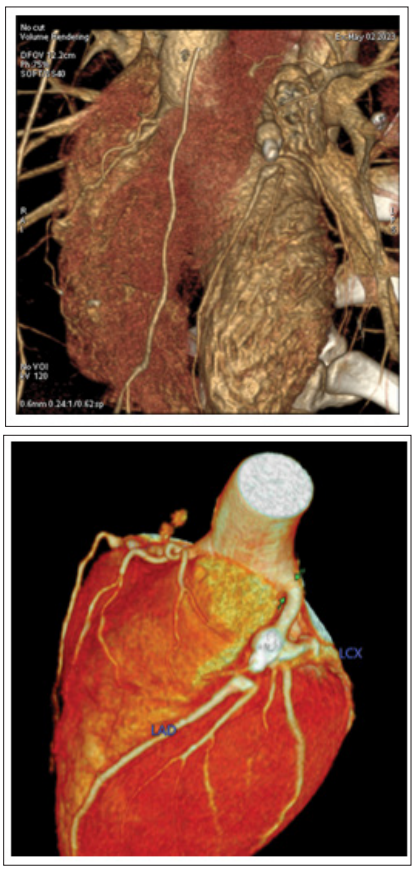

Figure 2:

a) Coronary CT Angiography/Calcium SCORE: Proximal LAD Aneurysm with Calcified Thrombus Inside.

b) 3D Reconstruction

c) 3D MRI-C Reconstruction. LAD: Anterior Descending Artery. LCX: Circumflex Artery

Since the patient persisted with symptoms due to angina, a CT angiography with SCORE for coronary calcium was requested (Fig. 2), which was performed in May. The patient was found to have an "aneurysm of the left anterior descending artery with a calcified thrombus inside". Coronary angiography was then chosen, which is why he was referred to our center, the "Presidente Juan Domingo Perón" High Complexity Hospital (HAC), where he was admitted as an outpatient in July. The coronary angiography (CCG) report revealed a proximal occlusion of the DA artery, as well as multiple coronary aneurysms of the left main coronary artery, the left anterior descending artery, the diagonal artery, the circumflex artery, and the right coronary artery (Figure. 3 and 4). It was decided to admit him. Upon admission, a non-pathological ECG was found, with symmetrical and positive T waves in the precordial leads. A consultation was made with the rheumatology service, which requested an ANCA, ANCA-C/P, Anti-DNA, FAN, RF, anti-CCP antibody, ENA profile, complement factors C3, C5, C7 and C8, all negative or within normal parameters, which ruled out Systemic Lupus Erythematosus (SLE), autoimmune vasculitis such as granulomatosis with polyangiitis (Wegener's disease), microscopic polyangiitis (MPA), rheumatoid arthritis, and scleroderma. HLA-B51 dosage is pending to rule out Behçet's disease.

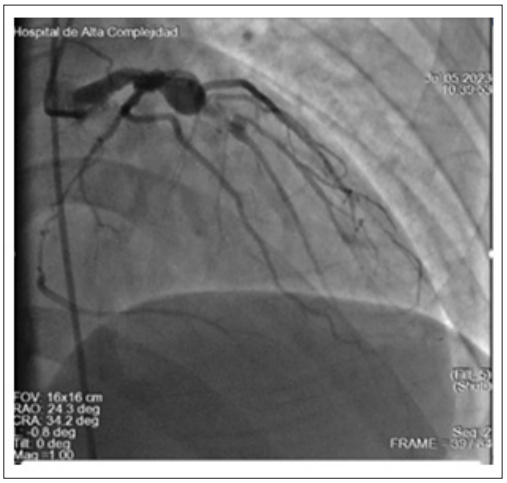

Figure 3: CCG. Right Anterior Oblique Cranial view. Saccular Image in the Distal Segment of the Left Main Coronary Artery (LMCA) at the Bifurcation of the LAD with a Later ventricular branch. A Critical Sub Occlusive Image with Microchannels is Observed in the Proximal Segment of the LAD

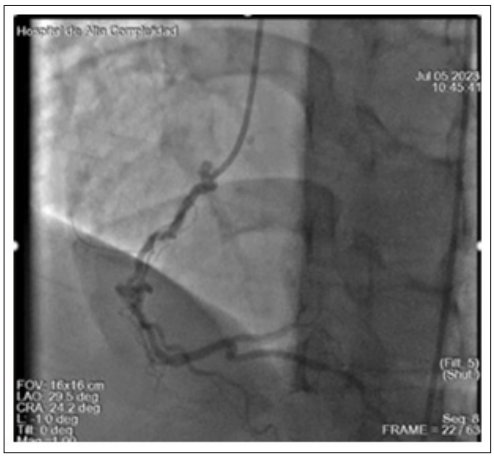

Figure 4: GCC. Left Anterior Oblique Cranial View. Well-Developed, Dominant CD, Irregularity of the Proximal Segment and Half Aneurysmal Sac at the Ostial Level

In consensus with the patient and family, surgical treatment of occlusive coronary lesion of DA was opted for, after completing presurgical studies, including a Doppler echocardiogram with improvement compared to the previous one during the acute phase of myocarditis with preserved left ventricular diameters, wall thickness and systolic function Fey 60%, without regional asynergies, non-dilated cavities, preserved aorta in all its portions, mild mitral regurgitation, mild tricuspid regurgitation that does not allow estimation of PSAP, mild pulmonary regurgitation. A CT angiography of the brain, thorax, neck vessels, abdomen and pelvis was also performed, which did not show aneurysmal dilations in any vascular territory.

He was admitted to the operating room where coronary artery bypass grafting (CABG) was performed with 2 arterial bypasses: Left Internal Mammary Artery-LAD and diagonal branch of LAD plus 2 venous bypasses to the lateroventricular branch of CX and CD with a biopsy of the right mammary artery which at macroscopy presented a bead-like appearance, at microscopy without acute inflammatory signs in the thickness of the arterial wall. The procedure was uncomplicated with subsequent cardiovascular recovery in (ICU) with favorable evolution, asymptomatic from the cardiovascular point of view, medication was titrated among which we highlight antiaggregation with Aspirin 100 mg / day and anticoagulation with Rivaroxaban 20 mg / day.

Development

Coronary artery lesions are a concern because there may be no signs of the disease until a serious event occurs, such as chest pain, myocardial infarction, or even sudden death. A child may have remained asymptomatic throughout childhood, and their parents may be completely unaware that they actually have coronary artery disease. Those who engage in strenuous or athletic activities, as is the case with our patient, may be at risk for sudden death and should modify their exercise routines. This disease is the second most common cause of sudden death among young athletes. Anomalous coronary arteries are also suspected of increasing the risk of premature development of coronary atherosclerosis [1].

Coronary aneurysms in adults are a rare entity. The most commonly associated risk factors are similar to those for coronary artery disease, such as chronic arterial hypertension, smoking, and male sex (male:female ratio 3:1) [2]. Their initial manifestation may be myocardial infarction and sudden death, resulting from rupture or from the presence of intracoronary thrombi [4]. They occur most frequently in the right coronary artery, followed by those of the left anterior descending coronary artery and circumflex coronary artery. Aneurysms of the left main coronary artery are even rarer. The disease most frequently associated with this entity is coronary atherosclerosis with severe stenosis in the adjacent areas [5].

They represent an exaggerated form of expansive vascular remodeling in response to the growth of an atherosclerotic plaque. Several enzymatic factors contribute to the degradation of the extracellular matrix of the media; among these are elevated homocysteine levels, hyperinsulinemia, increased nitric oxide (NO) metabolites, accumulation of lipoproteins in the intima, activation of the renin-angiotensin-aldosterone system (RAAS), and endothelial inflammatory processes. All of the above leads to a disruption of the internal elastic intima that promotes the exit of inflammatory cells into the media, which favors expansive remodeling and ultimately ectasia [4].

Regarding the probable rheumatological etiology, the hypothesis is that coronary aneurysms are a consequence of one of the various vasculitis, between 10 to 20% are secondary to connective tissue diseases, mainly systemic sclerosis, Ehlers-Danlos syndrome, Kawasaki disease and ANCA antibody positive vasculitis, among others [5].

KD is a self-limiting acute systemic vasculitis affecting medium-sized arteries, with a predilection for coronary arteries. It begins with fever, mucocutaneous involvement, and cervical lymphadenopathy. It predominantly affects children under 5 years of age, especially between 6 and 11 months of age, with a certain predominance in males. It is most common in Japan, with an incidence of 217 to 265 cases per 100,000 children under 5 years of age per year. KD in adults is extremely rare, with very few published cases. The classic diagnosis of this disease is based on the presence of fever lasting 5 or more days and 4 or more of the following clinical signs: skin changes on the extremities, polymorphic rash, bilateral non-exudative conjunctivitis, involvement of the lips and oral cavity, and cervical lymphadenopathy in patients with fever. If 4 of the previously described clinical signs are present, this diagnosis can be suspected and will be confirmed if coronary artery disease is detected [5].

Coronary aneurysms have also been linked to the presence of several coronary risk factors, arterial hypertension, dyslipidemia, smoking, diabetes mellitus and family history of ischemic heart disease, which have been associated with atherosclerotic aneurysms that usually affect one or two coronary vessels, with degradation of collagen and elastin fibers with disruption of the internal and external elastic lamina [6]. Potential complications of coronary aneurysms include rupture, thrombosis, embolization, dissection, mechanical obstruction and erosion to surrounding structures [5].

Coronary angiography (CCG) has been the tool of choice for the diagnosis of coronary aneurysmal disease, since it not only evaluates the diameter of the coronary arteries but also their flow, often finding ectatic patterns, with turbulent flows, slow filling of the contrast medium and local deposition of contrast in dilated arterial segments [4].

There is still much controversy regarding antiplatelet therapy and anticoagulation in patients with coronary aneurysmal disease. Multiple studies have shown no benefit in anticoagulating patients with coronary aneurysmal disease; however, a recent study suggests a possible benefit of anticoagulating patients who also have acute coronary syndrome. In the study, patients who received adequate anticoagulation with warfarin (time in therapeutic range > 60%) had a 0% incidence of major cardiovascular events (MACE) compared with 33% of patients who did not receive anticoagulation ($p < 0.03$). Additionally, there is evidence that in patients with Kawasaki disease, anticoagulation helps reduce the rate of thrombotic events, so its use is recommended in this population [7].

Both antiplatelet agents (APD) and anticoagulants (OCA) are statistically significantly effective in preventing recurrences. In coronary artery disease, both antiplatelet and anticoagulant effects are required, since ectasia shows continuous platelet activation due to the existing turbulent flow, but also procoagulant activation due to endothelial damage. This means that (dual) DAPT in combination with OAC is effective for long-term management in patients with coronary ectasia. However, studies with larger

study populations are required to reach an accurate conclusion. The combination of APD and OAC may also be essential when there is already thrombosis in the ectatic segment, because when a residual thrombus is identified within an aneurysm (i.e., a thrombus that remains after attempted aspiration). Some authors advocate triple antithrombotic therapy with replacement of a conventional OAC with a new oral anticoagulant (NOAC): rivaroxaban. It has been proposed that the use of NOACs may have greater benefit than conventional OACs in the management of coronary ectasia [8].

Conclusion

Speaking about a young adult, let alone a 20-year-old, who complains of chest pain that has been developing for months is no easy task. We are faced with a world of possibilities, syndromes that share common signs and symptoms, and a complex patient due to his age and lack of cardiovascular risk factors that so often lead us to suspect coronary artery disease. But a range of causes that we don't consider on a daily basis opens up in cases like this one, which is why we bring it up for discussion. Coronary aneurysmal disease is common in the category of congenital heart conditions, but not in adults. Precordial pain is its most common symptom, and if we have learned anything from our patient, it is that in the daily life of the cardiology department, we find that not all chest pain is synonymous with myocardial infarction; we must always look beyond what is expected.

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