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Coronary artery aneurysm. Literature review and cases report in Venezuela

Aneurismas de las arterias coronarias. Revisión de literatura e informe de casos en Venezuela

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SUMMARY

Coronary artery aneurysm (CAA) is an uncommon condition and is defined as dilatation of the coronary artery exceeding 50 % of the reference vessel diameter or more than 1.5 times the diameter of normal adjacent segments or the diameter of the patient's largest coronary artery. The reported incidence of coronary aneurysms ranges from 0.3 % to 5 %, with a predilection to men more than to women, and proximal than to distal segments of the coronary bed. In adults, the most common predisposing condition for CAA is atherosclerotic coronary artery disease. In children, CAAs are typically multiple and associated with Kawasaki disease. The reported incidences in the literature for congenital CAA are wide (1%–30%), so the true incidence remains uncertain. Systemic vasculitis and connective tissue disorders, infectious and iatrogenic causes are also implicated. This is the first report series from a single group of 4 patients (2 females and 2 males) with giant CAA successfully intervened in Venezuela from 1985 to 2012. The mean age of patients was 46.75 years (21-71 years). Surgical therapy is still the most definitive solution to CAA noting that every approach must be carefully tailored to each case.

Keywords: *Coronary artery, aneurysm.*

RESUMEN

El aneurisma de arterias coronarias (AAC) es una condición infrecuente que es definida como la dilatación de la arteria coronaria que excede el 50 % del diámetro del vaso de referencia o más de 1.5 veces el diámetro de segmentos normales adyacentes o del diámetro de la arteria coronaria más grande del paciente. La incidencia reportada de los aneurismas coronarios abarca desde el 0,3 % al 5 %, con mayor predilección por los hombres que por las mujeres y, más a segmentos proximales que a distales del lecho coronario. En adultos, la condición predisponente más común para AAC es la enfermedad ateroesclerótica de las arterias coronarias. En niños, los AACs son típicamente múltiples y asociados a la enfermedad de Kawasaki. La incidencia reportada en la literatura para los AAC congénitos es amplia (1 %-30 %), así que la verdadera incidencia permanece imprecisa. También están implicados la vasculitis sistémica y los desórdenes del tejido conectivo, así como las causas infecciosas y iatrogénicas. Esta es la primera serie reportada de un solo grupo quirúrgico de 4 pacientes (2 femeninos y 2 masculinos) con AAC gigantes exitosamente intervenidos en Venezuela desde 1985 al 2012. La edad promedio de los pacientes fue de 46,75 años (21-71 años). El tratamiento quirúrgico sigue siendo la solución más definitiva para los AAC

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acotando que cada abordaje debe ser cuidadosamente diseñado para cada caso.

Palabras clave: *Arterias coronarias, aneurismas*

INTRODUCTION

Coronary artery aneurysm (CAA) is an uncommon condition and is defined as dilatation of the coronary artery exceeding 50 % of the reference vessel diameter (1) or more than 1.5 times the diameter of normal adjacent segments or the diameter of the patient's largest coronary artery (2). CAAs were first described by Morgagni in 1761 (3). Years after, the first published description of a coronary artery aneurysm (CAA) appeared in 1812, written by French Physician Dr. Charles Bougon. His patient was a male officer who after returning home from military service suffered 4 years of nocturnal chest pain unresponsive to application of sternal leeches that died at 40 years of age after an abrupt escalation of anginal symptoms; and at autopsy, Bougon found hemopericardium and a ruptured right coronary artery (RCA) aneurysm (4).

They are divided into coronary artery ectasia and aneurysms. Coronary artery ectasias (CAEs) are diffuse and involve 50 % or more of the length of the artery, while coronary artery aneurysms (CAAs) are more focal, involving <50 % of the total length of the vessel (2). CAAs are termed giant if their diameter transcends the reference vessel diameter by greater than four times (1) that is exceeding 20 mm in adults and, 8 mm or a coronary artery Z score ≥10 in children (5,6). The biggest CAA described had a maximum diameter of 180 mm (7).

The reported incidence of coronary aneurysms ranges from 0.3 % to 5 %, with predilection to men more than to women, and to proximal than to distal segments of the coronary bed (8). This wide range is likely due to the various reporting methods, with higher incidences found in studies including both CAE and CAA together versus those reporting CAA alone (9).

The right coronary artery is usually the most affected artery (40 %) followed by the left anterior descending (32 %), and the left main being the least affected artery (3.5 %) (10).

Aneurysms are pathologically classified into three groups: atherosclerotic, inflammatory, and noninflammatory. The last category is associated with congenital, inherited, and connective tissue disorders (11).

In adults, the most common predisposing condition for CAA is atherosclerotic coronary artery disease. In children, CAAs are typically multiple and associated with the systemic inflammation of Kawasaki disease. The range of reported incidences in the literature for congenital CAA is wide (1 %–30 %), and therefore, the true incidence remains uncertain. Immune-mediated factors and genetic susceptibilities are implicated in several diseases associated with CAAs, including systemic vasculitis and connective tissue disorders (11). A concomitant thoracic or abdominal aortic aneurysm may be found, which usually is clinically silent (12). Infections and iatrogenic events such as cardiac catheterization are also well-recognized but unusual causes and are actually pseudoaneurysms. Both forms result from a defect in the media (traumatic or inflammatory, respectively) that results in extravasation of blood, along with a fibrous adventitial reaction that walls off the hemorrhage (13-15). Infectious ("mycotic") aneurysms are associated with endocarditis and other conditions predisposing to bacteremia. Histopathologically, infectious aneurysms show acute inflammation and fibrin deposition, along with organisms, findings similar to those in infectious endocarditis (12).

In pathologic autopsy series, CAAs are highly represented by ACAAs, which account for 73 %–90 % of CAAs (16,17). Between one-half and two-thirds of hearts with CAAs have multiple aneurysms. Almost all patients have severe atherosclerosis, with more than 75 % luminal area narrowing of at least one epicardial artery. The CAAs themselves show atherosclerotic change with superimposed thrombosis. Medial thinning is a characteristic and is implicated in the pathogenesis of CAAs (16,17). The fact that atherosclerosis is far more common than aneurysms has led to the theory that a second superimposed inflammatory process exists that results in aneurysms secondary to atherosclerosis (17). Atherosclerotic change can also be superimposed on inflammatory aneurysms, for example, those caused by

Takayasu arteritis (18).

CAAs are frequently reported following brachytherapy due to the late effect of adaptive remodeling, and drug-eluting stent implantation due to the impaired intimal healing effects of the antiproliferative agents (19-21). It has to noted that there have been fewer published reports of CAA formation following second and third generation stent implantation (22,23). Whether this represents a reporting bias versus a true improvement with newer stents remains unknown and requires further investigation. There has been, nevertheless, a concerning number of reports of CAA following PCI with biodegradable stents (24-27).

The symptoms and complications of CCAs may be due to mass effect on adjacent cardiac structures, vasospasm, thrombosis, and/or embolism. Fistula formation occurs when vascular inflammation associated with the CAA causes erosion into adjacent cardiac structures (11). The presence of concomitant obstructive atherosclerotic disease can result in both effort angina or acute coronary syndrome (9) as well as other critical consequences besides myocardial ischemia or infarction, which includes arterial rupture and hemopericardium (11).

Coronary angiography remains the most commonly used imaging modality to assess ectatic or aneurysmal coronary arteries. However, delayed antegrade contrast filling, segmental backflow, and contrast stasis in the dilated coronary segment often hamper optimal imaging during angiography (28). In these cases, intravascular ultrasound (IVUS) can be extremely helpful; it provides better delineation of vessel wall structures and helps to distinguish between true aneurysm, pseudoaneurysm, and segments with aneurysmal appearance due to plaque rupture or adjacent stenosis. IVUS can also accurately size the CAA and/or any adjacent stenosis and allows proper stent sizing if PCI is planned. Coronary computed tomography is gaining popularity in the assessment of these patients because it allows more accurate evaluation of the aneurysm size and degree of thrombus and calcification than invasive angiography (29). Computed tomography is particularly helpful in patients with giant CAA and those with saphenous vein grafts active (SVGA) because it avoids the pitfalls of luminal angiography and provides a precise assessment of mechanical complications of these aneurysms, but a CAA may even be identified at chest radiography, transthoracic echocardiography, or magnetic resonance (MR) imaging. Depending on the clinical scenario and the imaging findings, a CAA may require medical management or more invasive procedures, including (a) the placement of a percutaneous stent or coil devices or (b) coronary artery bypass surgery (11).

Percutaneous treatment of CAA brings certain practical challenges because there are no covered stents that are specifically designed for their treatment. Several stent graft systems have been used off-label to exclude CAA. Proper sizing of the CAA is key to reducing the risk of stent thrombosis and migration (30) but partially thrombosed CAA can result in underestimation of the true size and true extension of the aneurysm, making an accurate assessment of the stent's landing zone difficult. In long aneurysmal segments, several overlapping stents are often needed to cover the full length of the aneurysm (9,31,32).

It is important to note that covered stents are stiff and are delivered via large coronary guiding catheters or introducer sheaths, this increases the risk of procedural complications (stent migration, dissection, perforation, and so on), especially in tortuous or calcified vessels, with fear of side branch compromise or worst, limited access to them leaving a persistent leak into the aneurysm sac. In such cases where covered stent placement is not possible, the stent-assisted coil embolization technique can be used (9).

The ideal surgical approach must be individually tailored for every patient. Although most of the reviewed references report single cases (33-35), there are several articles reporting case series of various patients successfully intervened. Operative therapy for CAA may include aneurysm ligation, resection, patch closure, or marsupialization with interposition graft (36). The most common surgical practice is, however, to open the CAA, suture its afferent and efferent vessels, and finish with bypass grafting when necessary (37,38).

CASES REPORT

Clinical features of 4 patients (2 females and 2 males) with coronary artery aneurysms are presented in Table 1. The mean age of patients was 46.75 years (21-71 years).

Table 1

Cases of coronary artery aneurysm

Case	Age	Date	Sex	Cause	Origin of CAA	Fistula	Additional procedures			
							1	2	3	4
1	21	8/14/1985	F	Congenital	RCA	RV		CAAPC	AVR	
2	46	5/7/1997	M	Atherosclerotic	RCA + LAD + Cx		CABG x 4			
3	71	5/21/1997	M	Atherosclerotic	RCA		OPCABG x 1			AAA Repair
4	49	10/3/2012	F	Congenital	RCA + LAD + Cx		CABG x 4			

CAA, Coronary artery aneurysm; RCA, right coronary artery; RV, right ventricle; CAAPC, coronary artery aneurysm patch closure; AVR, aortic valve replacement; LAD, left anterior descending coronary artery; Cx, Circumflex; CABG, coronary artery bypass grafting; OPCABG, off-pump coronary artery bypass grafting; AAA, abdominal aortic aneurysm.

In case 1, the patient had a gigantic right coronary artery aneurysm (see Figure 1) fistulized to the right ventricle that produced a theft in the distal circulatory bed, also produced an Aortic insufficiency due to leaflet asymmetry because of a gigantic RCA ostium of 2cm (see Figure 2). In this case, direct closure of the RV fistula was performed with a couple of pledged sutures followed by AVR with a mono disc mechanical prosthesis and RCA ostium closure with a dacron patch (see Figure 3). No CABG was needed distally because of retrograde inflow from LAD.

Figure 1.

Figure 2.

Figure 3.

Drawings by Dr. Mario Duran.

In case 2, the patient had small proximal CAA in RCA and distal to left main bifurcation (LAD and Circumflex) that required proximal and distal ligature (afferent and efferent), followed by a distal confection of CABG comprised of the left internal thoracic artery (LITA) to diagonal and saphenous vein graft (SVG) to LAD, Obtuse marginal (OM) and RCA.

In case 3, there was only one CAA emerging from RCA. The aneurysm was proximally and distally ligated and a reverse saphenous vein graft (SVG) bypass of the pump was used to assess the lesion plus infrarenal AAA resection with aorto-aortic dacron bypass.

Case 4 was a female patient who presented with angina and underwent to coronary angiogram finding multiple CAA (see Figure 4) in LM (A), LAD and Diagonal (B) and Cx (C) with significative obstructive coronary artery disease, late filling of a gigantic CAA partially thrombosed in the left territory (D), RCA (E1) and late filling of multiple gigantic CAA (E2) which filled by reflux from LAD. Coronary angiotomography was performed (see Figure 5) with tridimensional reconstruction (see Figure 6).

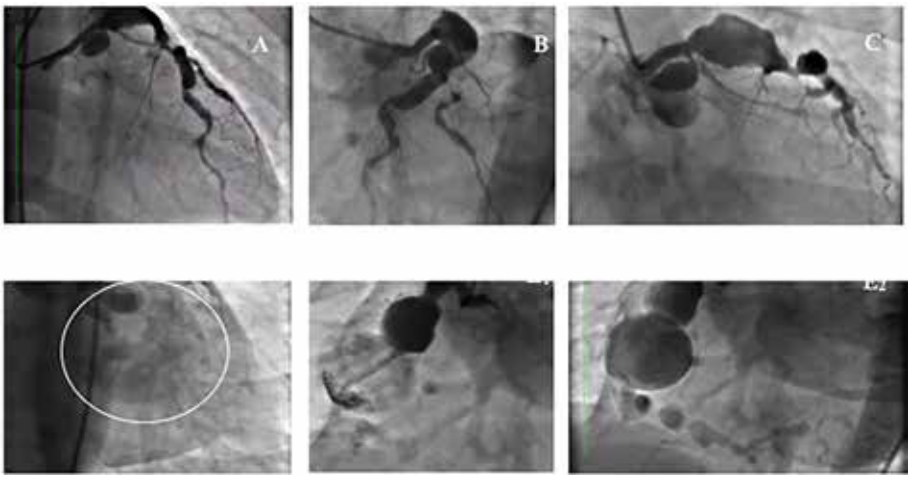

Figure 4. Photography series 1. Diagnostic Angiogram series. Note late filling inside RCA.

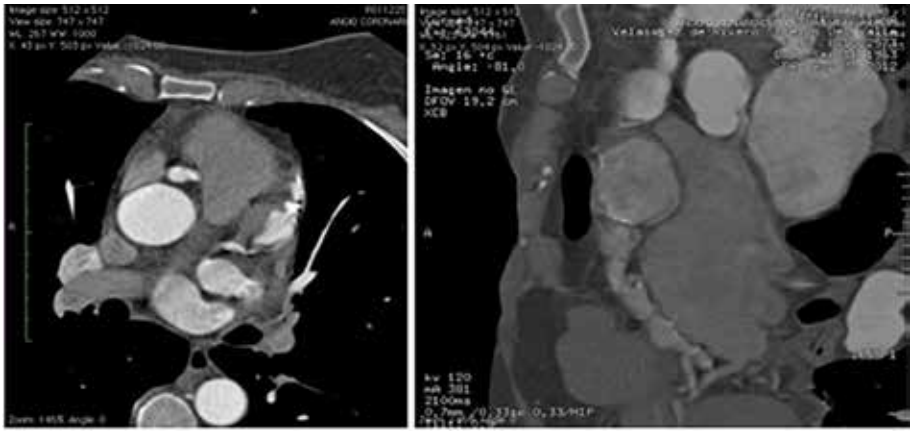

Figure 5. Photography series 2. Diagnostic AngioTomography series.

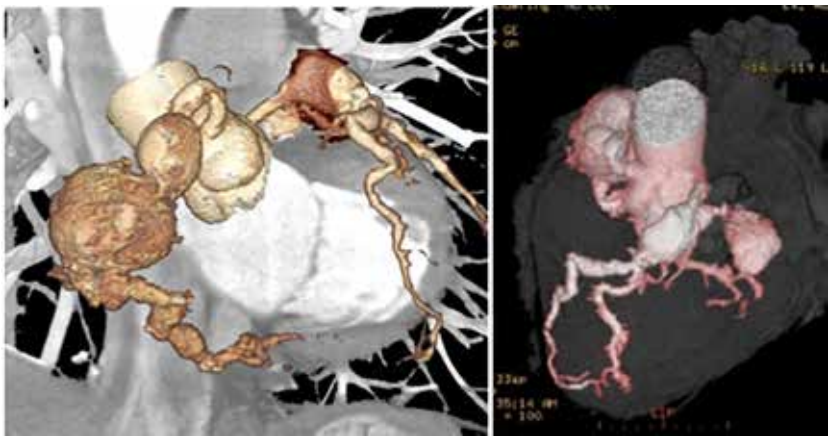

Figure 6. Photography series 3. Diagnostic AngioTomography series with tridimensional reconstruction.

Photographs from surgical findings showed multiple aneurysms in both territories, right and left (see Figure 7), where ligation and CABG were performed on the pump (see Figure 8).

Figure 7. Photograph series 4. Surgical findings. White circles signal coronary aneurysms.

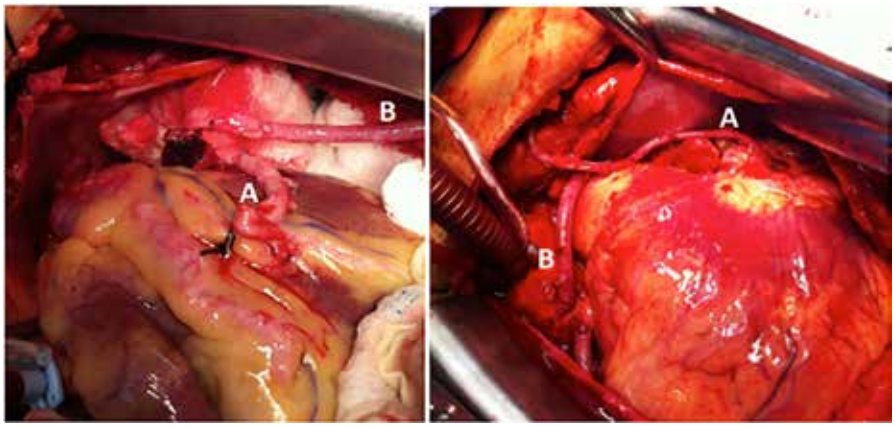

Figure 8. Photograph series 5. CABG with LITA (letter A in white) and SVG (letter B in white).

The postoperative evolution of these four patients was satisfactory and expected for any patient with coronary atherosclerotic or thrombotic submitted to CABG.

DISCUSSION

Coronary artery aneurysm is an uncommon disease, although it has been diagnosed with increasing frequency thanks to the advent of coronary angiography, moreover, giant coronary artery aneurysm is rarely seen. This is the first report series, from a single surgical group, were from 3 469 patients submitted to cardiac surgery for the main author, only 4 patients with giant coronary artery aneurysm were found in Venezuela in the period comprised from 1985 to 2012 which represents an 0.11 % incidence while other authors (38) had a smaller incidence (0.01 %) with a larger number of patients intervened (only 6 patients with CAA of 30 268 cardiac surgical cases)

The most common causes of CAA were seen in this case review, concordant with international literature been congenital and atherosclerosis the main causes.

Surgical therapy is still the most definitive solution to CAA noting that every approach must be carefully tailored to each case, mainly because allows the proximal and distal ligature of the aneurysmal sac, preventing the continued growing and rupture of CAA and distal embolism and consecutive acute coronary syndrome.

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