

Giant aneurysm of circumflex coronary artery in asymptomatic patient

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ABSTRACT

We report a case of a 74-year-old woman who presented to the hospital for fever and uncontrolled hypertension. We found, incidentally, a giant aneurysm of the circumflex coronary artery measuring 6.4×5.5 cm. We show suggestive computed tomographic scan images, multi-slice reconstructions and a review of the epidemiology, diagnosis and treatment of this condition.

Introduction

Coronary artery aneurysm (CAA) is a rare condition, characterized by a dilatation of 1.5 times the diameter of an adjacent normal coronary vessel. Finding of a giant CAA is a very uncommon event. We present a case of incidental finding of an asymptomatic giant aneurysm affecting the circumflex coronary artery; then we discuss epidemiology, etiology and clinical approach, referring to published literature data.

Case Report

A 74-year-old woman presented at hospital with fever and uncontrolled hypertension. Over the past

years, she had suffered from chronic obstructive pulmonary disease (COPD) and hypertension that had not been further investigated. While blood specimens showed no significant findings (mild inflammation), chest radiograms revealed the presence of a pulmonary mass and a transthoracic echocardiography revealed an anechogenic interatrial lesion, that measured 56 mm, with apparent internal flow (Figure 1). Chest computed tomography (CT) with radiopaque contrast showed a solid lesion in the superior left pulmonary lobe, while the cardiac angiographic multi-slice CT showed a markedly ectatic and winding circumflex artery (maximum diameter: 8 mm), with a saccular aneurysm placed between the medium and the distal third of the vessel, just after a marginal branch emergence. The aneurysm measured 6.4×5.5 cm and presented no endoluminal thrombosis neither parietal calcifications. The distal branch of circumflex artery overflowed into the extremely dilated and winding

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Key words: Atherosclerosis; coronary disease; aneurysm; echocardiography; CT scan.

Contributions: AB was involved with review of the literature, diagnostic process and drafting of the manuscript; OP, CDF, GF, GB and GPV were involved with radiological and ultrasound analysis and drafting; GM, PF and CP were involved in the diagnostic process and drafting.

Conflict of interest: the authors declare no conflict of interest.

Received for publication: 23 December 2015.

Accepted for publication: 29 January 2016.

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Licensee PAGEPress, Italy
Italian Journal of Medicine 2016; 10:234-237
doi:10.4081/ijm.2016.683

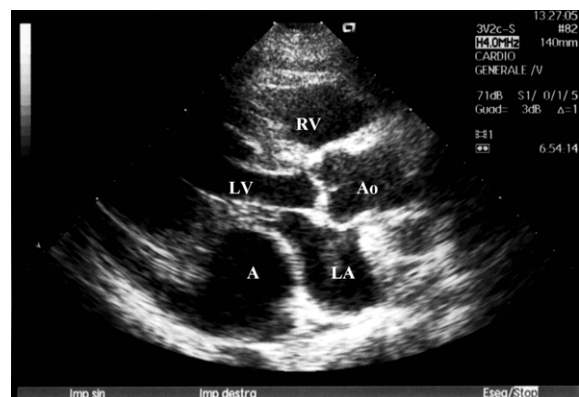


Figure 1. Cross-sectional echocardiography: parasternal long-axis view. RV, right ventricle; LV, left ventricle; Ao, aorta; A, aneurysmatic lesion; LA, left atrium.

coronary sinus, which was displaced to the right posterior and lateral side by the aneurysm (Figure 2A and B). In order to obtain the best resolution of coronary arteries, three-dimensional image reconstructions were performed with maximum intensity projection and volume rendering technique (Figure 2C and D). Invasive coronary angiography was then performed, showing no coronary stenosis but a wide-length aneurysmatic circumflex artery, with a large aneurysm placed next to atrioventricular junction (Figure 3). Diagnostic evaluation for the pul-

monary mass (transthoracic biopsy with CT guide and total-body positron-emission tomography) revealed the presence of non-small cell lung carcinoma, with left adrenal gland metastasis. Clinical history, blood specimens and imaging evaluation led us to a differential diagnosis between a more likely congenital or atherosclerotic etiology and a very unlikely adult-onset Kawasaki disease. The patient, informed about her clinical condition, refused any treatment both for neoplasm and aneurysm. She has died a few months later from neoplastic cachexia.

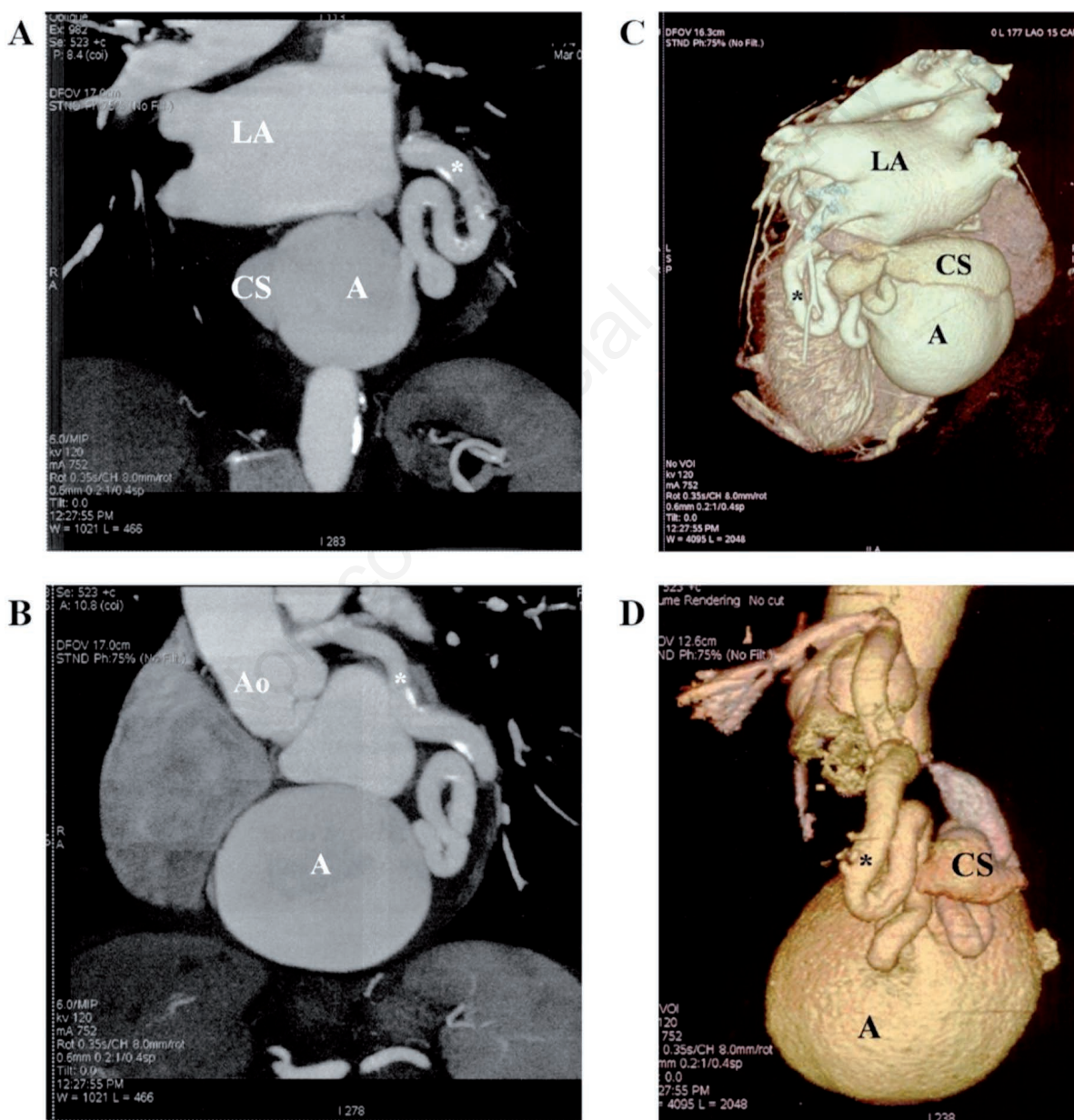


Figure 2. A-B) Angiographic multi-slice computed tomography; C) three-dimensional image reconstruction; D) three-dimensional image reconstruction, without cardiac chambers. *Ectatic circumflex coronary artery; LA, left atrium; CS, coronary sinus; A, aneurysmatic lesion; Ao, aorta.

Discussion

CAA is a relatively uncommon disease, which is characterized by abnormal dilatation of a localized or diffuse segment of coronary vessels of up to one and a half times the diameter of an adjacent normal segment.¹ This finding is usually occasional (generally during cardiac catheterization) and it is reported in 1.1-4.9% of coronary angiography and 1.4% of autopsies, with preference for male gender (2.2% *versus* 1.5% in female gender) and right coronary artery (40-61% *versus* 15-32% in left anterior descending coronary artery, 15-23% in circumflex coronary artery and 0.1% in left main coronary artery).²⁻⁶ Some authors classify aneurysms only on the basis of the shape, making difference between diffuse aneurysmal ectasia (that interests a large part of the coronary vessel) and discrete aneurysm (which is a localized abnormal dilatation with a spherical or saccular shape); other authors classify aneurysms combining their appearance and the number of coronary vessels affected.^{1,6-8} Giant CAA are very rare, with anecdotal incidence of 0.02% to 0.2%. To date, no uniform definition is accepted for *giant* aneurysm, since in different papers they are variously defined as more than 8 to 50 mm in size or when dilatation exceeds the reference vessel diameter by 4 times.^{2-4,9-12} Different etiologies have been suggested for CAA: congenital; atherosclerosis; Kawasaki disease; coronary angioplasty (balloon, laser, atherectomy) and surgery (coronary artery bypass graft); rheumatic; arteritis (polyarteritis nodosa, systemic lupus erythematosus, Takayasu's arteritis,

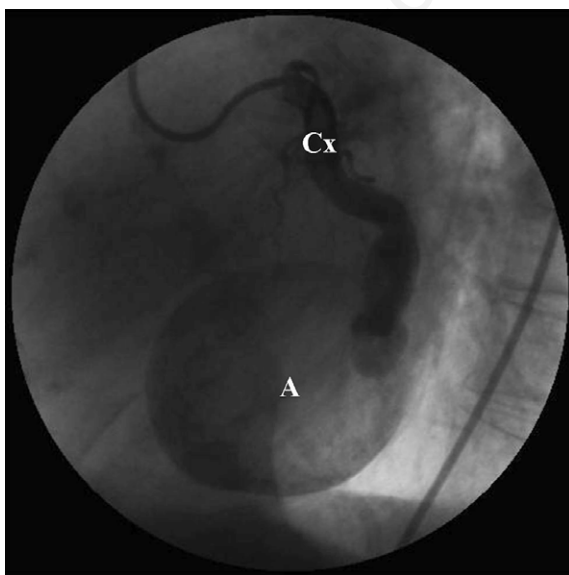


Figure 3. Cardiac angiography: right anterior oblique projection. Cx, ectatic circumflex coronary artery; A, aneurysmatic lesion.

Behçet's disease, syphilis); mycotic coronary emboli; coronary artery dissection or trauma; connective tissue disorders (Marfan's and Ehlers-Danlos syndromes).^{1,2,13,14} In adult population, atherosclerosis accounts for the majority of cases (at least 50%), followed by congenital diseases (20-30%) and Kawasaki disease (10-20%), while the latter is the leading cause for the young and in childhood.^{1,3,14,15} Natural history and prognosis of CAA is largely unclear: some authors report no difference in outcome for patients with or without aneurysm; other authors consider CAA a variant of coronary artery disease with similar reduced overall 5-year survival.^{1,3} Cardiac symptoms, such as chest pain, dyspnea, palpitation, may be present, although sometimes CAA may be totally asymptomatic.² Natural evolution of untreated CAA is increasing in size *via* Laplace's law, at a rate of growth of about 5 mm per year. CAA may be asymptomatic even at a size >80 mm.^{3,11} The primary complications are myocardial ischemia and infarction due to thrombosis and distal embolization, dissection, fistula formation with hemopericardium or tamponade, hemodynamic disturbances due to compression of adjacent structures. Spontaneous rupture is rare, but more likely with the increasing diameter.⁹⁻¹² Diagnosis and characterization of CAA can be made with imaging tools. Although some authors consider echocardiography useful in accurately diagnosing giant CAAs⁵ and non-invasive methods (echocardiography both transthoracic and transesophageal, CT, magnetic resonance) are more readily available and can detect some CAAs, coronary angiography is usually considered the gold standard, since it can provide more accurate information about site, number, size and shape of CAAs and it can have also therapeutic utility.^{10,12} No consensus exists yet on better acute and long term therapeutic approach (medical *versus* surgical therapy), especially in asymptomatic patients: therapeutic algorithms have been proposed only for CAA with acute coronary syndrome or for isolated aneurysm without obstructive coronaropathy.^{3,6} Most cases (particularly in small uncomplicated aneurysms) can be safely managed conservatively with careful follow-up. Indeed, all patients with coronary aneurysm should receive aggressive modification of coronary risk factors. Anticoagulation and antiplatelet therapy (low-dose aspirin together with warfarin, ranging an international normalized ratio between 2.0 and 2.5, or with a therapeutic dose of low-molecular-weight heparin) are effective in giant coronary aneurysms due to Kawasaki's disease, while its benefit is less evident in atherosclerotic etiology, although sometimes proposed in order to prevent thromboembolic complications.^{1,9,15} Some authors recommend surgical approach for all aneurysms >30 mm or 3 to 4 times the normal vessel diameter and for complicated or symptomatic CAAs,

with percutaneous coronary intervention with polytetrafluoroethylene-covered stents, or coronary artery bypass graft. The latter is most frequently performed with resection of coronary artery aneurysm as the treatment of giant aneurysms, due to lack of commercially available stents to cover them effectively.^{2-4,9,10}

Conclusions

CAAs are rare diseases and once we made the diagnosis we faced a clinical challenge, since no validated decisional algorithm still exists. Every patient should then receive a truly *tailored* approach, based on comprehensive clinical evaluation (accurate assessment of global cardiovascular risk, accurate determination of CAA characteristics, accurate evaluation of existing comorbidities and life expectancy) in order to propose the own specific optimal treatment (medical or surgical).

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