

CASE REPORT



Multiple and giant coronary artery aneurysm – case report and a review of the literature

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Abstract

We report an unusual case of a 70-year-old female patient admitted in the Department of Cardiology, Emergency County Hospital, Oradea, Romania, for unstable angina and heart failure. Coronary angiography and computed tomography revealed multiple coronary artery aneurysms, some of them giant, associated with significant coronary artery disease. Coronary artery aneurysm is a rare disease, but the frequency of diagnosis increased with the development of coronary angiography. However, very rarely coronary artery aneurysm is very large and can be termed “giant aneurysm”, and a small number of reports have been described in literature. The management and the prognosis of the disease is still controversial.

Keywords: coronary artery, aneurysm, giant, atherosclerosis.

Introduction

Coronary artery aneurysm is defined as a localized dilatation of a coronary artery of saccular or fusiform shape that is larger than the diameter of the normal vessel adjacent segment by 1.5–2 times [1, 2]. A true arterial aneurysm can be fusiform, with the longitudinal diameter larger than the transversal diameter or saccular, with the transverse diameter larger than the longitudinal diameter. Fusiform aneurysms are much more common than saccular aneurysms.

Coronary artery ectasia is a diffuse dilatation that includes 50% or more of the length of a coronary artery [3].

Sometimes, a coronary artery aneurysm dilates very much and can be classified as giant, but the dimensions of such aneurysms are variable in literature. The *Committee of the American Heart Association* has defined giant aneurysm when the diameter is more than 8 mm [4, 5], but other authors consider aneurysm to be giant when are greater than 20 mm [6].

The tunica media of the coronary aneurysms is usually thinned and can be destroyed markedly until is unidentifiable between intima and adventitia. The hyalinized connective tissue replaces the normal smooth muscle cells and elastic fibers from the middle layer of the coronary artery [7]. In pseudoaneurysms or false aneurysms, there is a rupture of the arterial wall contained by the adventitia or a blood clot.

Coronary artery aneurysm is a rare abnormality. It

was first reported in 1761, on post-mortem examination, by Morgagni and by Bourgon in 1812 [8, 9]. The largest autopsy series, by Daoud *et al.* [10], reported a 1.4% incidence of coronary artery aneurysm in 694 patients. Munkner *et al.* [11] presented the first case diagnosed alive with coronary angiography in 1958. The frequency of diagnosis of coronary artery aneurysm increased with the development of coronary angiography. In *Coronary Artery Surgery Study (CASS) Registry*, the angiographic incidence of coronary aneurysm was 4.9% in a group of 20 087 patients [12], which exceeded the incidence reported in many other angiographic studies (0.37–2.53%) [13]. The reported prevalence of giant coronary artery aneurysm is very low (0.02%) [14].

Aim

The aim of this study was to present a rare case of multiple coronary artery aneurysms distributed thorough the coronary system, some of them large enough to be called giant, discovered at coronary angiography.

Case presentation

A 70-year-old woman was admitted in the Department of Cardiology, Emergency County Hospital, Oradea, Romania, in March 2010, for refractory unstable angina despite maximal medical treatment and exertional dyspnea. She had a history of previous old anterior myocardial infarction, which was managed with medical treatment in

a territorial hospital (without thrombolysis) and hyperlipidemia. On physical examination, we found pulmonary stasis at the base of the lungs, peripheral edema, her blood pressure was 110/80 mmHg, and heart rate was 90 beats/min. Electrocardiogram showed old anterior myocardial infarction with QS waves and 2 mm persistent ST segment elevation in V1–V5, negative T waves V1–V6, first diagonal branch–augmented vector left (D1–aVL). Cardiac enzymes [creatin kinase MB isoenzyme (CK–MB) and highly sensitive troponin] were in normal range. Her blood cholesterol level was 246 mg/dL, with a low-density lipoprotein (LDL)-cholesterol of 136 mg/dL. The other blood tests were in normal range, including inflammatory markers. Transthoracic echocardiography revealed left ventricular apical aneurysm, with moderately depreciated left ventricular ejection fraction of 30%, and mild pericardial effusion.

Coronary angiography showed a coronary aneurysm of the proximal segment of left anterior descending artery, with transversal diameter of 4 mm and longitudinal diameter of 5 mm. Left anterior descending artery was completely occluded after the emergency of the D1 artery, with retrograde flow from the right coronary artery. The first diagonal artery was completely occluded after a second saccular aneurysm with transversal diameter of 5 mm and longitudinal diameter of 3 mm (Figure 1).

Left circumflex artery presents in the proximal part, before the emergency of an important obtuse branch, a giant aneurysmal dilatation with transversal diameter of 16 mm and longitudinal diameter of 35 mm (Figure 1).

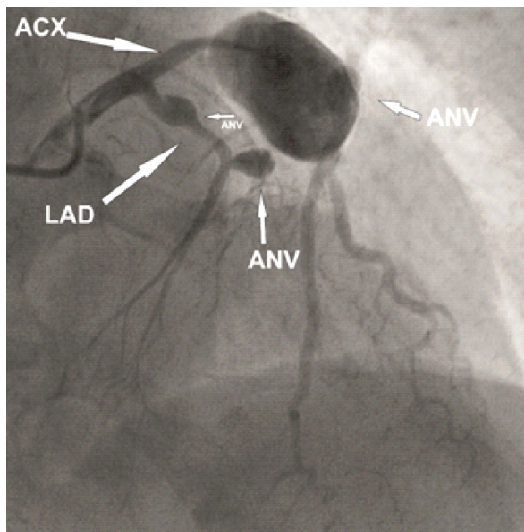


Figure 1 – Coronary angiography. Left anterior cranial view demonstrating a giant aneurysm in the proximal part of the ACX, with transversal diameter of 16 mm and longitudinal diameter of 35 mm. LAD presents a coronary aneurysm in the proximal segment with transversal diameter of 4 mm and longitudinal diameter of 5 mm. LAD is completely occluded after the emergency of the first diagonal branch. The first diagonal artery is completely occluded after a second saccular aneurysm with transversal diameter of 5 mm and longitudinal diameter of 3 mm. ACX: Circumflex artery; ANV: Aneurysm; LAD: Left anterior descending artery.

The right coronary artery presents in the vertical segment an aneurysmal dilatation with transversal diameter of 7.7 mm and longitudinal diameter of 25 mm, followed by

another aneurysmal dilatation with a transversal diameter of 5.3 mm and longitudinal diameter of 7 mm (Figure 2).

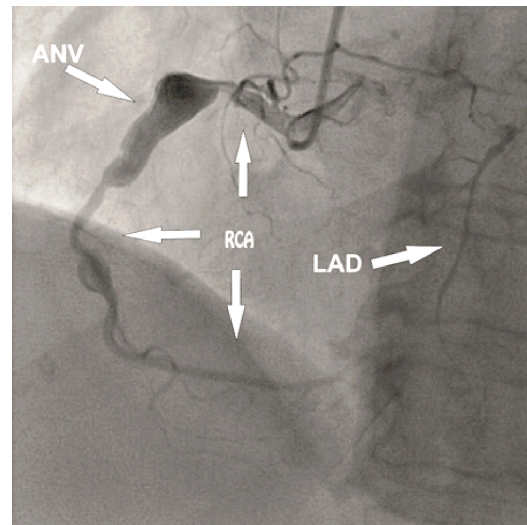


Figure 2 – Coronary angiography. Left anterior oblique view. The RCA presents in the vertical segment an aneurysmal dilatation with transversal diameter of 7.7 mm and longitudinal diameter of 25 mm, followed by another aneurysmal dilatation with a transversal diameter of 5.3 mm and longitudinal diameter of 7 mm. Retrograde flow from RCA to LAD. ANV: Aneurysm; LAD: Left anterior descending artery; RCA: Right coronary artery.

Cardiac computed tomography (CT) enables the further delineation of the topographical anatomy of coronary artery aneurysm. CT angiography provides a non-invasive and accurate evaluation of the localization, the dimensions and the content of thrombus or calcifications of the aneurysm. The size of the left circumflex artery aneurysm was much greater on multislice CT with contrast, the largest transversal diameter of the aneurysm was 44 mm; but the giant aneurysm contains an important amount of thrombus on the right lateral wall of 28 mm (Figures 3–5). Only flow within the lumen can be detected after contrast injection at coronary angiography, so the true dimensions can be undervalued if the aneurysm contains a substantial amount of thrombus as in our patient.



Figure 3 – Multislice computed tomography with intravenous contrast. Axial sections. Saccular giant aneurysm of the circumflex artery with the largest transversal diameter of 44 mm and a filling defect on the right lateral wall of 28 mm (thrombus), and calcification of the aneurysmal wall.



Figure 4 – Multislice computed tomography with intravenous contrast. Coronal multiplanar reconstruction. Giant aneurysm of the circumflex coronary artery with an important amount thrombus.

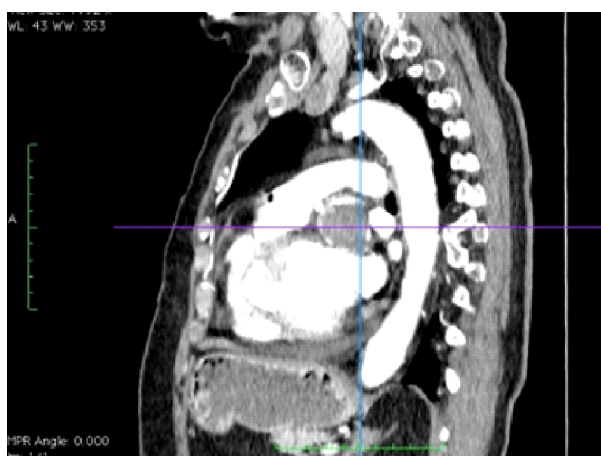


Figure 5 – Multislice computed tomography with contrast. Sagittal multiplanar reconstruction. Giant aneurysm of the left circumflex coronary artery with thrombus.

The optimal management in this patient was established to be cardiac surgery, since patient had a giant circumflex artery aneurysm, multiple other coronary artery aneurysms, which were coexisting with significant coronary artery disease, respectively occlusion of the left anterior descending artery and of the D1 artery.

The patient refused surgical treatment and was treated only with medical therapy. She received anticoagulants, antiplatelet agents, angiotensin converting enzyme inhibitors, beta-blockers, statins, and diuretics. She was discharged after nine days with improved medical condition, chest pain diminished, electrocardiogram was stable, and the markers of myocardial necrosis were not elevated.

Long-term evolution was unfavorable, she was admitted repeatedly for angina and she died after one year.

Consent

Written informed consent was obtained from the patient for invasive coronarography examination in accordance with the investigation protocol and also for the publication of this case report. No data that revealed the patient identity were presented in the manuscript.

Discussions

The overall incidence of coronary artery aneurysm ranges from 0.3% to 5.3%, with a mean incidence of 1.65% [15], but giant artery aneurysms are very rare, with a reported prevalence of 0.02% [14]. The incidence of coronary artery aneurysm is higher in men [13], and although coronary artery aneurysm can occur at any age, those with atherosclerotic etiology appear later in life [15]. Most frequently is involved the right coronary artery (40.4%), followed by the left anterior descending artery (32.3%) and by the left circumflex artery (23.4%). Left main artery is rarely involved (3.5%) [6].

Our illustrative case had multiple coronary aneurysms on all three major coronary arteries: right coronary, left anterior descending and circumflex artery with a giant aneurysm on the circumflex artery.

The major characteristics revealed by histological examination consists of lipid deposition of the intima, alongside hyalinization deposits, intramural hemorrhage and an inflammatory reaction which reflects the intensity of the arteriosclerotic process. The chronic overstimulation of the nitric oxide represents an important factor which alters the medial layer of the vessel wall [6]. Other pathological mechanisms, which affect the elasticity of the vessel wall, consists in the presence of different areas of fibrosis, calcification, and the existence of large cholesterol crystals. All these factors contribute to the aneurysm formation. The atherosclerotic disease further affects the wall structure in the context of transmural inflammation [8, 16]. Due to the continuous intraluminal pressure, the stress tolerance is reduced, determining dilation of the vessel wall.

It is supposed to exist a genetic contribution in atherosclerotic coronary artery disease associated coronary artery aneurysm. This was proved in some genomic-wide association studies, which reflected the connection between different variants on chromosome 9p21.3 and coronary artery disease. This specific locus may also play a role in the formation of intracranial and abdominal aortic aneurysm. Apparently, it determines an altered proliferative phenotype, which causes adverse vascular remodeling [17].

Thrombosis is common in giant aneurysm, and an important amount of thrombus was present on the right lateral wall of the giant circumflex artery aneurysm, in our patient. The internal surface of the aneurysmal wall is irregular and predispose to slow blood flow, thrombus formation and subsequent embolization.

Daoud *et al.* observed, in a postmortem study, the presence of thrombus in seven of 10 patients with coronary aneurysms [10]. The clinical presentation of our patient can be associated with the presence of thrombus and possible distal embolization, but we must consider that our patient had also significant coronary artery with occlusion of two coronary arteries. In our patient, the true size of the aneurysm was much greater as revealed on angio-CT, explained by the fact that the giant aneurysm contains an important amount of thrombus.

Coronary angiography is a “luminogram” that does not provide information about the wall of the vessel and can underestimate the true size of the aneurysm or can overlook an aneurysm occluded by a large thrombus or

plaque [15]. The study performed by Murthy *et al.* also observed that true size of the aneurysm with thrombus content can be undervalued [18]. CT angiography is a noninvasive investigation that can accurately assess the aneurysm dimensions and location and provide information about thrombus content and calcification. The extent of thrombus compared to luminal flow is also described with confidence [18].

☒ Conclusions

Giant coronary artery aneurysm is a rare disease with diverse clinical symptoms that can rise problems of differential diagnosis with various disease. The management remains a clinical challenge and should be individualized for each patient considering: clinical presentation, etiology and aneurysm size, location and its expansion in time, comorbidities, the presence and extent of associated atherosclerosis.

Conflict of interests

The authors declare that they have no conflict of interests.

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