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Case Report

Multi-vessel giant coronary artery aneurysms: An unusual cause of chest pain ☆,☆☆

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ABSTRACT

We describe an unusual case of multi-vessel giant coronary artery aneurysms complicated by acute coronary syndrome despite escalation of therapy. A 65-year-old man with hypertension and hypercholesterolemia presented to clinic with atypical chest pain over 4 months. Outpatient computed tomography coronary angiography (CTCA) demonstrated giant coronary aneurysms involving all 3 major coronary arteries. Outpatient coronary angiogram findings were in concordance with the CTCA with no definite obstructive coronary disease. Myocardial perfusion imaging was normal. He was commenced on dual antiplatelet therapy (DAPT). At 6 months, he presented with chest pain and non-ST-elevation myocardial infarction. Repeat coronary angiogram demonstrated occluded first septal LAD branch which previously had aneurysmal dilatation. DAPT was changed to long-term oral anticoagulation. He remains well at 18 months. This case highlights the importance of multi-modality imaging in the diagnosis and workup of coronary artery aneurysms and challenges in management; an individualized approach is required.

Introduction

Coronary artery aneurysms are an uncommon finding in adulthood and are often incidentally diagnosed. This case report highlights the importance of multi-modality imaging in the diagnosis and workup of coronary artery aneurysms, particularly as functional coronary assessment alone may miss the diagnosis. This report also highlights the challenges faced in the management of coronary artery aneurysms

Abbreviations: CTCA, computed tomography coronary angiography; DAPT, dual antiplatelet therapy; LAD, left anterior descending artery; RCA, right coronary artery.

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and highlights the lack of current consensus on optimal management.

Case presentation

A 65-year-old man with hypertension and hypercholesterolemia presented with 4 months of atypical chest pain. Medications included atorvastatin and amlodipine. There was no significant family history or known childhood illnesses. A 12-lead electrocardiogram was normal. Computed tomography coronary angiography (CTCA) demonstrated giant coronary aneurysms predominantly involving the proximal segments of all 3 coronary arteries (Fig. 1A), particularly the left anterior descending artery (LAD) (16 × 13 mm) (Fig. 1B) and right coronary artery (RCA) (Figs. 1C and D). The RCA was most affected (maximum diameter 17 mm) with a large burden of mural thrombus reducing luminal diameter to 3 mm without obstructive stenosis (Figs. 1C and D). Coronary artery calcium score was 18 Agatston units (32nd percentile for age and sex). Laboratory investigations revealed normal complete blood count and mildly impaired renal function with estimated glomerular filtration rate 52 ml/min/1.73m^2 (N >90 ml/min/1.73m^2). Markers of inflammation and autoimmune antibody testing were all normal. Due to ongoing chest pain, elective coronary angiography was performed (Fig. 2). Findings were concordant with the CTCA, demonstrating severe aneurysmal disease and no obstructive coronary disease. Subsequent rest-stress myocardial perfusion imaging (Tc-99m sestamibi), performed due to ongoing pain, was normal at high workload. CT brain/abdomen/pelvis revealed no extra-cardiac aneurysms. After consensus discussion, he commenced dual antiplatelet therapy (DAPT) (aspirin 100 mg daily and clopidogrel 75 mg daily), and his symptoms, possibly non-cardiac in nature, improved.

However, at 6 months, he developed different chest pain and presented with a non-ST-elevation myocardial infarction. ECG at the time did not demonstrate any ischemic changes while high sensitivity troponin T peaked at 980 ng/L (N < 26 ng/L). Coronary angiography demonstrated an occluded first septal branch of the LAD, which previously had aneurysmal dilatation (Fig. 3). A decision was made to change DAPT to long-term oral anticoagulation and he was commenced on apixaban 5 mg bd. He was discharged home on day 3 post admission. Echocardiography 2 months post-admission was normal. He remains well at 18 months. Subsequently, a CT chest performed 8 years earlier for an unrelated problem became available, showing the coronary aneurysms (not appreciated at the time) to be similar.

Discussion

Coronary artery aneurysms are an uncommon finding in adulthood, often incidentally diagnosed. We describe an unusual case of multi-vessel giant coronary artery aneurysms, likely due to previously undiagnosed childhood Kawasaki disease, complicated by acute coronary syndrome (ACS), despite escalation of medical therapy.

Etiology and clinical presentation of coronary artery aneurysms

Coronary artery aneurysm is defined as a localized dilatation of a coronary artery of ≥1.5 times the adjacent normal segment, while coronary artery ectasia defines similar but more diffuse lesions [1]. Coronary artery aneurysms are reported in up to 5% of patients undergoing coronary angiography [2]. The clinical presentation of coronary artery aneurysms is highly variable, ranging from asymptomatic and incidentally diagnosed to an acute coronary syndrome. Clinical symptoms may develop from concomitant obstructive atherosclerotic disease, local thrombosis of an aneurysm resulting in distal embolization and myocardial infarction, extrinsic compression of surrounding structures, and very rarely aneurysm rupture.

Interestingly, aneurysms most often affect the proximal segment of vessels, particularly the right coronary artery, and rarely all 3 vessels [1]. Coronary artery aneurysms are most commonly due to ischemic heart disease but may be a manifestation of autoimmune or inflammatory processes such as childhood Kawasaki disease, part of a systemic multi-organ illness (eg, systemic lupus erythematosus or polyarteritis nodosa), post-infectious, as a result of repeated dynamic wall stress changes such as from cocaine use, iatrogenic from coronary artery manipulation, congenital or idiopathic in nature [1]. Coronary artery aneurysms from atherosclerosis and vasculitic processes usually affect more than one artery, whereas congenital or iatrogenic usually affect a single artery. In this

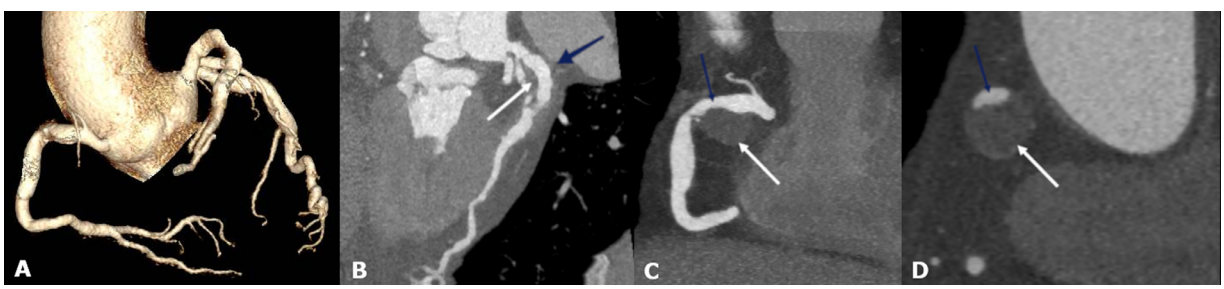

Fig. 1 – CTCA of all 3 major coronary arteries in 3D reconstruction (A) and multiplanar reconstruction images of the LAD (B) and RCA (C, D) demonstrating giant aneurysms proximally in each vessel (blue arrow) and large burden of mural thrombus (white arrow).

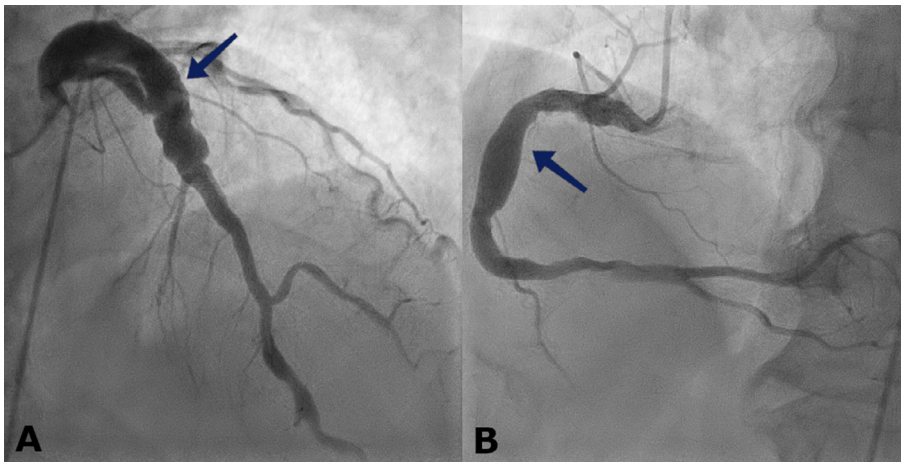

Fig. 2 – Coronary angiogram of the LAD (A) (AP, Cranial 40) and RCA (B) (LAO 34, Cranial 15) also demonstrating giant aneurysms (arrows), concordant with the CTCA.

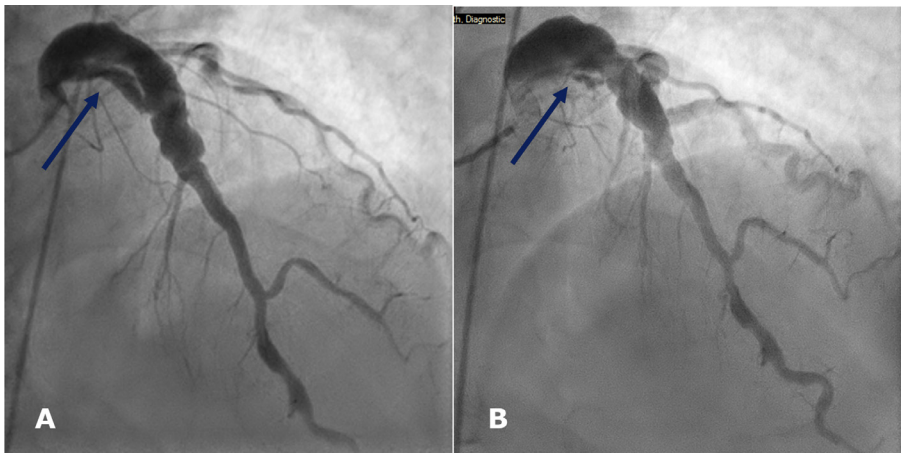

Fig. 3 – Repeat coronary angiogram demonstrating first septal branch of the LAD (arrow) following the myocardial infarct at 6 months. The images demonstrated are pre- (at the time of the first presentation) (A) (AP, Cranial 37) and post-occlusive event (second presentation) (B) (RAO 7, Cranial 35).

case report, the most likely cause for our patient's coronary artery aneurysms is childhood Kawasaki disease given the distribution of aneurysms predominantly affecting the proximal segment of all 3 major coronary arteries. Less likely is atherosclerotic disease, due to the lack of obvious coronary artery stenoses as well as the similar appearance 8 years prior. While there is no known history of Kawasaki disease in our patient, it is possible and not uncommon for an acute febrile illness in childhood to be overlooked and the diagnosis missed. Although Kawasaki disease is often a self-limiting illness, it can have long-term clinical sequalae with coronary artery aneurysm formation in up to 25% of untreated patients [3].

Prognosis of coronary artery aneurysms

While little is known about the natural history of coronary artery aneurysms, prognostication is closely related to the size of aneurysms both in terms of absolute diameter and body surface area adjusted z-scores. Large or giant aneurysms are those considered with a diameter >8 mm or z-score >10 [4]. The z-score is obtained by dividing the difference between the actual coronary artery diameter measurement and the predicted measurement by the standard deviation as previously described by McCrindle et al, though only normal values for the left main coronary artery, and proximal LAD and RCA currently exist [5]. The patient in our case report has aneurysms in all 3 major coronary arteries and are all classified as giant coronary artery aneurysms as each have an absolute diameter >8 mm and specifically the LAD z-score was 23.8 and RCA z-score was 25.3 (normal population predicted z-score value is 0). In a large study of 1356 children with Kawasaki disease followed for up to 15 years, coronary artery events (thrombosis, coronary artery stenosis >50% on angiography, coronary intervention, myocardial infarction, or death) were significantly more likely to occur in patients with both an aneurysm z-score ≥10 and an absolute dimension ≥8 mm compared to

those with an aneurysm z-score <10 and an absolute dimension <8 mm [4]. While this study was based on a pediatric population, it is reasonable to assume that major adverse cardiovascular events will only increase with age and acquisition of acquired cardiovascular risk factors. Therefore, optimal imaging and sizing of coronary artery aneurysms is important for prognostication of future cardiovascular risk.

Multi-modality imaging of coronary artery aneurysms

Complete assessment of coronary artery aneurysms may require multi-modality imaging. While invasive coronary angiography remains the most utilized imaging modality, optimal image acquisition may be limited by contrast stasis in the aneurysmal coronary segment and delayed antegrade contrast filling, resulting in underestimation of aneurysmal diameter and inaccurate assessment of intraluminal thrombi. CTCA has gained popularity in recent years as a non-invasive, yet sensitive tool in the characterization of aneurysmal features and coronary anatomy, even in the presence of intraluminal thrombi [6]. In our case report, the large burden of mural thrombus with significant reduction in luminal diameter was best appreciated on CTCA. CTCA can also provide 3-dimensional reconstructions of complex anatomical structures. Functional coronary artery assessment may be normal in coronary artery aneurysms, as in our case report, and may miss the diagnosis if utilized alone/upfront. While coronary artery aneurysms without significant coronary artery stenosis have been demonstrated to correspond with preserved myocardial function during stress as well as normal flow reserve values, stress-induced myocardial ischemia from microvascular dysfunction may still occur [7]. In our patient, functional assessment with stress Tc-99m sestamibi was performed after the anatomical diagnosis of coronary artery aneurysm was made to guide potential revascularization of lesions.

Management of coronary artery aneurysms

There is no consensus on the optimal management of coronary artery aneurysms but may include medical therapy, percutaneous, or surgical intervention. Medical management may include the use of DAPT or anticoagulation, although with mixed results seen from predominantly small and retrospective studies [8,9]. Similarly, it is unclear what the optimal duration of medical therapy is. The role of percutaneous coronary intervention of an aneurysmal vessel in acute coronary syndrome to restore flow is debated. There is a high rate of no-reflow or distal embolization due to higher thrombus burden in coronary artery aneurysms as well as a lack of specific stents designed for this purpose. Specifically, in patients with Kawasaki disease, current guidelines recommend restricting percutaneous coronary intervention to those with single vessel or focal multivessel disease [3]. Little is known about the role of surgical intervention in coronary artery aneurysms and may include coronary artery bypass grafting, aneurysm ligation or resection, or placement of an interposition graft [10].

Conclusion

Coronary artery aneurysms are an uncommon finding in adulthood and are often incidentally diagnosed. Multimodality imaging is often required in the diagnosis and the optimal management of coronary artery aneurysms remains debated. Ultimately, an individualized approach based on patient characteristics, clinical presentation, and etiology, location and morphology of the aneurysm/s is required.

Patient consent

Written, informed consent for publication of this case was obtained from the patient.

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