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ABSTRACT

A 64-year-old asymptomatic man had an incidental finding of a giant left circumflex artery (LCX) aneurysm, with the distal LCX draining into a confluence receiving terminal portions of all coronary arteries and communicating with the left ventricle through a transmural fistulous tract. We believe that this is the first case reported with such a complex LCX abnormality. (Level of Difficulty: Beginner.) (J Am Coll Cardiol Case Rep 2020;–:––) Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

HISTORY OF PRESENTATION

A 64-year-old asymptomatic man underwent a computed tomography (CT) scan of the abdomen and pelvis for evaluation of hematuria. The scan revealed an incidental finding of left circumflex artery (LCX) aneurysm. His physical examination was unremarkable for any acute or chronic cardiovascular findings. Upon admission, his vital signs were: blood pressure, 136/89 mm Hg; pulse, 74 beats/minute and regular; respiratory rate, 18 breaths/minute; temperature, 98.1°F; and pulse oximetry, 97% on room air.

PAST MEDICAL HISTORY

The patient had a history of well-controlled hypertension and no other modifiable or nonmodifiable risk factors or clinical markers of coronary artery disease.

DIFFERENTIAL DIAGNOSIS

Because of the incidental CT findings in this asymptomatic patient, the possibility of congenital versus acquired coronary aneurysm was raised. Given that the patient had not undergone any coronary interventions, the possibility of past iatrogenic causes or coronary manipulation was excluded.

INVESTIGATIONS

Subsequent coronary CT angiography (CTA) showed a giant (3.7-cm), calcified proximal LCX aneurysm with a small thromboatheroma (Figure 1). An additional smaller, noncalcified distal LCX aneurysm was present. Furthermore, the distal LCX drained into a confluent structure receiving terminal por-

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tions of the remainder of the coronary arteries (Figures 2 and 3). This confluence communicated with the left ventricle (LV) through a moderate-caliber transmural fistulous tract (Figures 3 and 4). The left anterior descending artery was markedly enlarged and tortuous proximally, and it appeared to drain into the arterial confluence distally. The right coronary artery was dominant, with a normal caliber, and it terminated distally into the confluence. Increased trabeculation was present in the LV at the anterolateral segment. A transthoracic echocardiogram (Figure 5) showed normal left ventricular and right ventricular systolic function without any wall motion abnormalities or valvular heart disease. Cardiac magnetic resonance revealed normal resting myocardial perfusion, normal biventricular function, a normal pulmonary-to-systemic flow ratio, and no evidence of delayed gadolinium enhancement (Figures 6A and 6B). An exercise single-photon emission computed tomography myocardial perfusion imaging study demonstrated no inducible ischemia, with normal left ventricular ejection fraction. We could not identify any additional congenital abnormalities or signs of infection or vasculitis in the past medical history or during the index evaluation.

**LEARNING OBJECTIVES**

- LCX aneurysm is an extremely rare clinical condition. Careful evaluation of the coronary anatomy is needed to identify any additional coronary anomalies in these patients. Our case represents a unique anatomy, with a giant LCX aneurysm and the distal LCX draining into a confluence receiving terminal portions of all coronary arteries and communicating with the LV through a transmural fistulous tract.
- Complex coronary anomalies require in-depth evaluation, including multimodality imaging to assess the patients for presence of myocardial ischemia and significant left-to-right shunt. Cardiac CTA is a very helpful diagnostic tool in establishing definitive anatomy in these cases.
- The treatment should be individualized on the basis of the patients’ symptoms, objective prediction of risk (presence of ischemia, progression of the lesion), and the presence of associated coronary and cardiac anomalies.
- With a careful work-up and follow-up, this coronary anomaly may have a benign short-term clinical outcome.

**MANAGEMENT**

The patient was managed conservatively using dual antiplatelet therapy (aspirin and clopidogrel) and a statin because coronary artery thrombosis and progressive stenosis within the aneurysm may cause myocardial ischemia, which increases the risk of myocardial infarction and sudden cardiac death in these patients. The patient took warfarin for a month, but it was stopped because of hematuria and hematospermia. He also underwent yearly coronary CTA for surveillance without significant change in the size of the aneurysm, and he has remained without cardiac complications to date for a total of 3 years. At present, nonradiation modalities such as cardiac magnetic resonance are not beneficial in this case because of the need in a 3-dimensional imaging modality with excellent all-axis spatial resolution (isometric voxels), for depiction of such a complex coronary anatomy.

**DISCUSSION**

The incidence of coronary artery aneurysms is 0.02% to 0.04%, and these aneurysms are usually seen in the right coronary artery. To our knowledge, there has been no case reported of a patient with an LCX aneurysm.
These 3D VR images of the inferior and lateral cardiac surface show a giant LCX coronary artery aneurysm, an LV transmural fistula, and confluence receiving terminal portions of all the coronary arteries. Multiplanar reconstruction (MPR) images show a giant left circumflex coronary artery aneurysm and confluence receiving terminal portions of all the coronary arteries. RA = right atrium; RV = right ventricle; other abbreviations as in Figure 1.

These 3D VR images of the apical and lateral cardiac surface show the LAD, LCX, LV transmural fistula, and confluence receiving terminal portions of all the coronary arteries. MPR images show an LV transmural fistula and confluence receiving terminal portions of all the coronary arteries. Abbreviations as in Figures 1 and 2.
These 3D VR and MPR images show a giant LCX coronary artery aneurysm, an LV transmural fistula, and the LCX. Abbreviations as in Figure 1.

This apical 4-chamber echocardiogram shows normal-size cardiac chambers, normal valves, and a calcified LCX aneurysm. Abbreviations as in Figures 1 and 2.
aneurysm communicating with a distal coronary arterial confluence, and this confluence further connected to the LV through a fistulous track. LCX aneurysms with fistulous communication have rarely been reported; among the reported aneurysms, most communicated with the coronary sinus (1). Only 1 case report described LCX aneurysm with a fistulous communication to the LV myocardium through several small vessels (2).

Coronary artery aneurysms may represent a potentially life-threatening condition with important complications including thrombosis or rupture. Of note, our patient was asymptomatic. The available treatment options for coronary artery aneurysm with fistula are surgical or endovascular interventions (1). However, treatment depends on the complexity of the anomalous anatomy. Furthermore, the follow-up duration and treatment with surgical or endovascular interventions in asymptomatic patients are unknown. A systematic approach with possible multicenter registries, development of a specific diagnostic approach, and prognostic estimates in patients with this complex anatomy, requires further research.

FOLLOW-UP

The patient underwent yearly coronary CTA for surveillance without significant change in the size of the LCX aneurysm, and he has remained without cardiac complications to date for a total of 3 years.

CONCLUSIONS

LCX aneurysm is an extremely rare clinical condition. Careful assessment of the coronary anatomy is needed to identify any additional coronary anomalies in these patients. Evaluation of such a unique anomaly as giant LCX aneurysm combined with distal LCX draining into a confluence receiving terminal portions of all coronary arteries and communicating with left ventricle through a transmural fistulous tract requires implementation of advanced 3-dimensional imaging methods.

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