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CASE REPORT

CLINICAL CASE

Unmasking of a Giant Coronary Aneurysm by Chronic Total Coronary Occlusion Percutaneous Coronary Intervention Techniques

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ABSTRACT

A young female patient presenting with a non-ST-segment elevation myocardial infarction underwent invasive coronary angiography, revealing a total occlusion of the right coronary artery. During percutaneous coronary intervention with dual catheter access, a retrograde tip injection and peculiar retrograde wiring unmasked a giant coronary aneurysm, which noninvasive imaging confirmed. (J Am Coll Cardiol Case Rep 2024;29:102359) © 2024 The Authors. 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 48-year-old female patient was referred to the Amsterdam University Medical Center after she had been diagnosed with a non-ST-segment elevation myocardial infarction. On admission, the patient had a blood pressure of 127/75 mm Hg, and a resting heart

LEARNING OBJECTIVES

- To provide insight in the treatment options for symptomatic patients diagnosed with a GCA, which is a rare clinical entity.
- To appreciate the potential benefits of preprocedural noninvasive imaging to clarify complex or unclear anatomy.

rate of 68 beats/min. Physical examination revealed no abnormalities. Electrocardiography showed a sinus rhythm with new negative T waves and subtle STsegment depression in the inferolateral leads; no Q waves were present.

PAST MEDICAL HISTORY

The patient had experienced anginal symptoms on exertion before the myocardial infarction. Medical history entailed transient ischemic attack, and a high cardiovascular risk profile (familial hypercholesterolemia, active smoking, and obesity).

DIFFERENTIAL DIAGNOSIS

Based on the electrocardiogram, an acute occlusion of the right coronary artery (RCA) was suspected.

Manuscript received February 6, 2024; revised manuscript received March 25, 2024, accepted March 27, 2024.

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ABBREVIATIONS AND ACRONYMS

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CCTA = coronary computed tomography angiography

CTO = chronic total coronary occlusion

GCA = giant coronary aneurysm

OMT = optimal medical therapy

PCI = percutaneous coronary intervention

RCA = right coronary artery

TTE = transthoracic echocardiography

INVESTIGATIONS

Laboratory analysis demonstrated an elevated high-sensitive cardiac troponin T (273 ng/L). Quick-look transthoracic echocardiography (TTE) was performed, describing a normal left ventricular function with hypokinesia of the infero-basal wall. The TTE report described all other cardiac structures as normal. The patient was scheduled for invasive coronary angiography (ICA), which revealed a total occlusion with thrombus formation of the proximal RCA, and retrograde filling by septal collaterals

originating from the left anterior descending artery (**Figure 1**). Notably, several small coronary artery aneurysms of the left anterior descending artery and first diagonal branch were observed. A short antegrade wire escalation was attempted due to the high thrombus load (suggestive of a subacute lesion), but failure of wire progression suggested the presence of a longer existing total occlusion, possibly a chronic total coronary occlusion (CTO).

MANAGEMENT

The local Heart Team advised optimal medical therapy (OMT) and referred the patient to the department of internal medicine for screening of vasculitic tissue disease. However, because of refractory symptoms after 2 months OMT, the patient was referred for elective percutaneous coronary intervention (PCI) by dedicated CTO operators (S.P. and A.N.). This decision was in concurrence with current clinical guidelines, which state that cardiac symptom alleviation in the presence of OMT is the primary indicator for revascularization of a CTO.¹ The reason for a CTO PCI setup was because we suspected an older occlusion, which requires visualization by selective dual guiding catheter injection. The Japanese CTO score was 4, based on an ambiguous cap, calcification, bending (>45°), and occlusion length (\geq 20 mm). After a brief antegrade attempt with a polymeric tapered wire, the operators switched to a retrograde approach. Subsequent retrograde wire escalation showed an unusual course of the RCA (Video 1). A retrograde tip injection was performed to visualize the distal cap and course of the vessel (Video 2). This revealed the outline of a large, calcified, nonopaque mass, suggestive of a giant coronary aneurysm (GCA) or pseudo-aneurysm (Figure 1). The patient was scheduled for additional

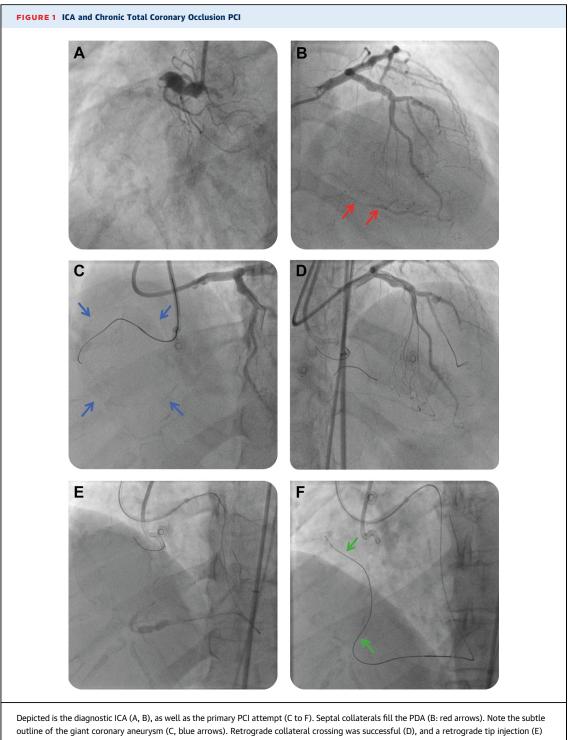
imaging (Figure 2). Coronary computed tomography angiography (CCTA) reported a fully thrombosed saccular aneurysm of the mid-RCA, estimated diameter 50 mm, and a thrombus in the distal RCA over a trajectory of 25 mm. A new TTE was performed, which demonstrated a hypoechogenic mass parallel to the RCA, with visual signs of mild compression of the right atrium.

SURGICAL INTERVENTION

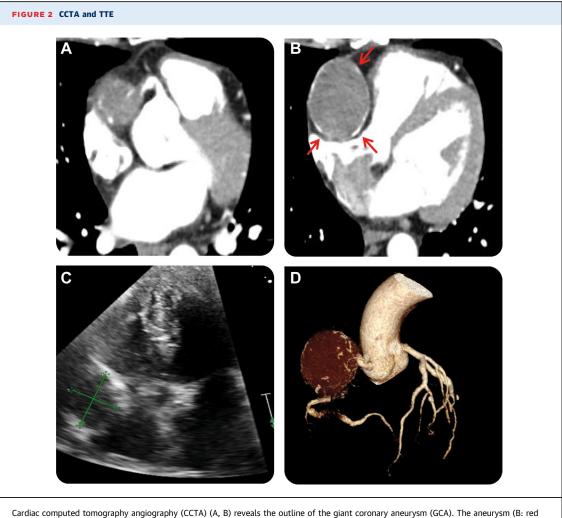
A percutaneous approach was considered, with a covered stent across the GCA to seal the entrance. However, the combination of right atrial compression, the size of the GCA, and risk of distal embolization and no reflow during PCI, led us to opt for surgical intervention as first choice of treatment. The time between CTO PCI attempt and surgical intervention was 54 days. The RCA was revascularized with a free right internal mammary artery anastomosed to the distal RCA before the crux (Figure 3, Video 3). A ligation of the proximal and mid-RCA severed the native coronary vessel path. Complete resection of the GCA was not possible because of its close relation to the surrounding structures. Part of the roof of the aneurysm was resected, and its contents were aspirated (Video 4). Histological analysis confirmed the GCA was an aneurysm with atherosclerotic changes, befitting of the cardiovascular risk profile (Figure 4). There was no clear evidence indicating vasculitic or infectious diseases, and although Kawasaki's disease could not be excluded, the presence of vasculitis or a connective tissue disease was considered unlikely.

DISCUSSION

GCAs (Figure 5) are exceedingly rare, and its management mostly empirical. The largest registry to date on coronary artery aneurysms² describes a prevalence of 0.35%, of which only 5.3% was considered to be a GCA. The presentation of a GCA has been described as predominantly silent, and its discovery incidental during ICA. Alternatively, the patient may present with cardiac symptoms, or acute coronary syndrome.³ Even more rare is the presentation of a GCA as a cardiac tumor⁴ or CTO. The latter has been described by Fanai et al,⁵ who reported a patient presenting with myocardial infarction caused by compression of the coronary arteries due to GCAs arising from the RCA and left anterior descending artery. To the best



outline of the giant coronary aneurysm (C, blue arrows). Retrograde collateral crossing was successful (D), and a retrograde tip injection (E) and peculiar course of the retrograde wire (F, green arrows) suggested wiring into the cavity of a large aneurysm. ICA = invasive coronary angiography; PCI = percutaneous coronary intervention; PDA = posterior descending artery.



Cardiac computed tomography angiography (CCTA) (A, B) reveals the outline of the giant coronary aneurysm (GCA). The aneurysm (B: red arrows) was estimated to have a diameter of 47 mm. Transthoracic echocardiography (TTE) shows a hypoechogenic mass (C, \pm 40 × 40 mm, green cross) with signs of mild compression of the right atrium. The GCA can be fully appreciated in a 3-dimensional reconstruction of the vascular system (D).

of our knowledge, our clinical case is the first to describe the unmasking of a GCA arising from the RCA through CTO PCI techniques. The combination of dual contrast injection and conversion to a retrograde approach led to the clarification of the true course of the native vessel, and finally the suspicion of a GCA. Most GCAs are surgically managed,⁶ although PCI has also been described.⁷ The decision for surgical intervention in our patient was justified on the basis of mechanical complications. It should be noted that the ligation of the RCA vessel hampers any future percutaneous revascularization options. It is unclear whether the total occlusion was already present (as a chronic severe stenosis which predisposed to a GCA), or if an aneurysm of the RCA obstructed the natural course of the vessel due to its considerable growth. The histological analysis noted predominantly atherosclerotic changes, but the presence of aneurysms in the left anterior descending artery could not exclude the hypothesis of a previous and unnoticed Kawasaki's disease. An important learning point from the presented case is the detection of the GCA through CCTA. The most common imaging modality for the assessment of coronary artery aneurysms is ICA, despite its shortcomings in reliably assessing the true size of

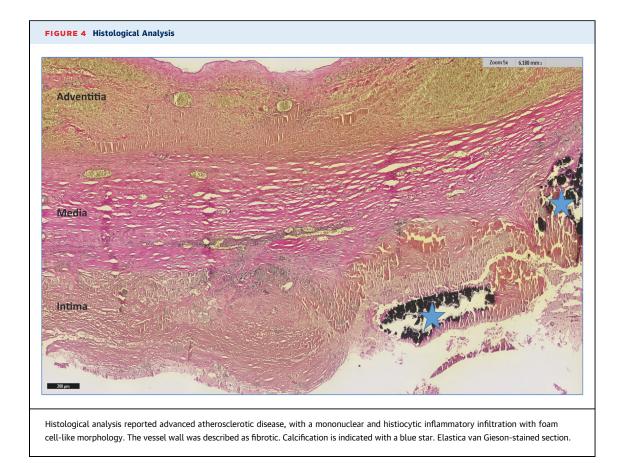
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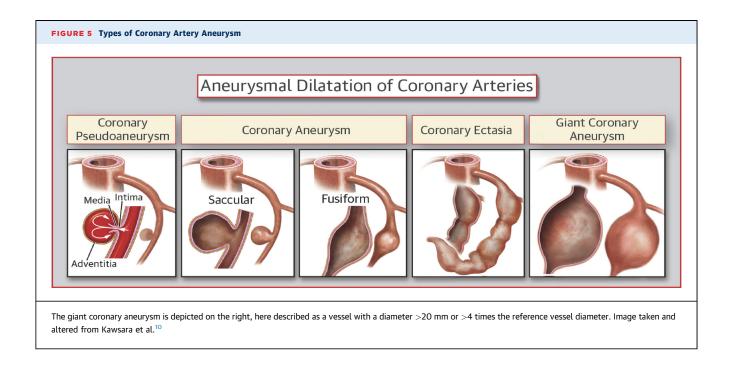
Intraoperative imaging of off-pump coronary artery bypass grafting and partial resection of the giant coronary aneurysm (GCA) (cranial view). After medial sternotomy, the GCA was visualized (A, blue arrows, B). The right coronary artery (RCA) was revascularized with a free right internal mammary artery anastomosed to the distal RCA. Aspiration of the GCA (C, green arrows) produced a dense liquid and thrombi. After partial resection of the roof of the GCA (±30 mm), the aneurysm was sutured with 4-0 Prolene (D, yellow arrows).

the aneurysm and its relation to surrounding structures.³ The evaluation of a coronary artery aneurysm through multimodality imaging, such as cardiac ultrasound and CCTA, are important tools in determining the optimal therapeutic strategy.⁸ In particular, CCTA has several advantages, including the 3-dimensional evaluation of the coronary artery and adjacent structures, accessibility in most centers, and low radiation exposure. In CTO PCI, CCTA is an established tool in pre-procedural planning, as it provides anatomical knowledge that may guide interventional strategy.⁹ In our patient, CCTA could have prevented delay of appropriate treatment. Finally, the operators were proficient in performing complex PCI, and the techniques applied during CTO PCI proved to be useful in recognizing the anomalous vessel course.

FOLLOW-UP

The patient did not experience postoperative complications. Cholesterol levels were managed with a statin and ezetimibe, with a low-density lipoprotein at 2.1 mmol/L. At 2.5 years following surgical intervention, the patient remained free of cardiac symptoms.





CONCLUSIONS

Giant coronary artery aneurysms are a rare entity and may present as a total coronary occlusion. Our case report adds to the limited body of evidence on surgical intervention of GCAs in symptomatic patients. Furthermore, this case highlights the added value of CTO PCI techniques in clarifying complex anatomy and shows the potential benefit of pre-procedural noninvasive imaging using CCTA.

FUNDING SUPPORT AND AUTHOR DISCLOSURES

The authors have reported that they have no relationships relevant to the contents of this paper to disclose.

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REFERENCES

1. Neumann F-J, Sousa-Uva M, Ahlsson A, et al. 2018 ESC/EACTS guidelines on myocardial revascularization. *Eur Heart J*. 2019;40(2):87-165.

2. Núñez-Gil IJ, Nombela-Franco L, Bagur R, et al. Rationale and design of a multicenter, international and collaborative Coronary Artery Aneurysm Registry (CAAR). *Clin Cardiol*. 2017;40(8):580-585.

3. Pham V, Hemptinne Q, Grinda JM, Duboc D, Varenne O, Picard F. Giant coronary aneurysms, from diagnosis to treatment: a literature review. *Arch Cardiovasc Dis.* 2020;113(1):59–69.

4. Dadkhah Tirani H, Aghajanzadeh M, Pourbahador R, Hassanzadeh R, Ebrahimi H. Giant right coronary artery aneurysm mimicking a mediastinal cyst with compression effects: a case report. *Res Cardiovasc Med.* 2016;5(3). e32086e32086. **5.** Fanai V, Malviya A, Mishra A, Lynser D, Ete T. Giant coronary aneurysm causing ostial occlusion of coronary artery by mass effect: a case report. *Cureus*. 2021;13(7):e16280.

6. Ebina T, Ishikawa Y, Uchida K, et al. A case of giant coronary artery aneurysm and literature review. *J Cardiol.* 2009;53(2):293–300.

7. Matta A, Zouari F, Campelo-Parada F, Carrie D. A giant left anterior descending artery (LAD) coronary artery aneurysm treated by covered stent angioplasty: a case report. *Am J Case Rep.* 2020;21:e925820.

8. Jha NK, Ouda HZ, Khan JA, Eising GP, Augustin N. Giant right coronary artery aneurysm–case report and literature review. *J Cardiothorac Surg.* 2009;4:18.

9. Andreini D, Collet C, Leipsic J, et al. Pre-procedural planning of coronary revascularization by cardiac computed tomography: An expert consensus document of the Society of Cardiovascular Computed Tomography. J Cardiovasc Comput Tomogr. 2022;16(6):558-572.

10. Kawsara A, Nunez Gil IJ, Alqahtani F, Moreland J, Rihal CS, Alkhouli M. Management of coronary artery aneurysms. *J Am Coll Cardiol Intv.* 2018;11(13):1211-1223.

KEY WORDS acute coronary syndrome, computed tomography, coronary vessel anomaly, hybrid imaging, percutaneous coronary intervention

APPENDIX For supplemental videos, please see the online version of this paper.