ADVANCED

JACC: CASE REPORTS © 2021 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/).

IMAGING VIGNETTE

CLINICAL VIGNETTE

Immunoglobulin G4-Related Coronary Artery Aneurysm-Associated Stent Migration



Keisuke Shoji, MD, Noriyuki Wakana, MD, PHD, Kan Zen, MD, PHD, Yusuke Hori, MD, Satoaki Matoba, MD, PHD

ABSTRACT

A 65-year-old woman with immunoglobulin G4-related disease (IgG4-RD), which was managed with a corticosteroid, underwent percutaneous coronary stent implantation in the left anterior descending artery as a result of angina pectoris. After 9 months, coronary angiography revealed stent migration and occlusion caused by progression of a coronary aneurysm potentially associated with IgG4-RD. (Level of Difficulty: Advanced.) (J Am Coll Cardiol Case Rep 2021;3:1895-1897) © 2021 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/).

CASE PRESENTATION

A 65-year-old woman with Mikulicz disease, an immunoglobulin G4-related disease (IgG4-RD), who achieved clinical remission with a corticosteroid (prednisolone, 4 mg/day), had angina pectoris refractory to medications. Coronary computed tomography angiography (CTA) showed severe stenosis in the left anterior descending (LAD) artery, and coronary angiography (CAG) revealed hemodynamically significant stenosis in the LAD artery (resting full-cycle ratio: 0.77). Two sirolimus-eluting stents were implanted, overlapping each other in the LAD artery, under intravascular ultrasound (IVUS) guidance (**Figures 1A and 1E**, Supplemental Figures 1 and 2, Videos 1 and 2). However, the patient presented with recurrent angina 9 months after the index percutaneous coronary intervention (PCI), and CAG revealed stent migration and occlusion resulting from progression of a coronary artery aneurysm (CAA) (**Figures 1B, 1C, 1F, and 1G**, Video 3). Retrospective analysis of IVUS images on the index PCI showed isoechoic perivascular thickening in the LAD artery and left main trunk (Supplemental Figures 2A to 2D, Video 4). Coronary CTA revealed stent migration, a gap between stents, instent occlusion, and abnormal coronary flow through the aneurysm (**Figures 1D and 1H**). Technetium-99m-tetrofosmin single-photon emission computed tomography revealed myocardial ischemia in the anterior and apex segments. There was no significant uptake in the CAA on fluorine-18-fluorodeoxyglucose positron emission tomography combined with computed tomography.

On the basis of the heart team's decision, isolated coronary artery bypass grafting of the LAD artery was performed using a graft from the left internal thoracic artery.

Coronary arteritis is a phenotype of IgG4-RD that leads to stenosis, aneurysm, and diffuse wall thickening (1). Although corticosteroid treatment is often effective for IgG4-related periarteritis, some patients can show

Manuscript received August 5, 2021; revised manuscript received August 27, 2021, accepted September 1, 2021.

From the Department of Cardiovascular Medicine, Kyoto Prefectural University of Medicine, Kyoto, Japan.

The authors attest they are in compliance with human studies committees and animal welfare regulations of the authors' institutions and Food and Drug Administration guidelines, including patient consent where appropriate. For more information, visit the Author Center.

ABBREVIATIONS AND ACRONYMS

CAA = coronary artery aneurysm

CAG = coronary angiography

CTA = computed tomography angiography

IgG4-RD = immunoglobulin G4-related disease

IVUS = intravascular ultrasound

LAD = left anterior descending

PCI = percutaneous coronary intervention

poor outcomes, such as life-threatening complications secondary to myocardial infarction or aneurysmal rupture (1).

This case is a rare presentation of stent migration and occlusion caused by progression of a CAA potentially associated with IgG4-RD. On the basis of pathology data, IgG4-related coronary arteries are characterized by the expansion of adventitia and a lymphoplasmacytic infiltrate with increased fibrosis involving the adventitia, media, and intima (2). Thus, these vascular degenerations exacerbate the fragility of vessels, and mechanical stimulation and activated inflammation caused by stent placement may have promoted a CAA in our case.

These pathologic findings of coronary arteritis may be visualized using several imaging modalities (3). Consistent with previous reports' findings, isoechoic perivascular thickening on IVUS imaging was observed in the present case. This case should serve as a warning when performing PCI on coronary arteries with acute inflammation. We should recognize the risk of progression of an IgG4-related CAA after stent implantation, focus more on imaging findings, and carefully consider PCI for coronary disease associated with IgG4-related arteritis.



(A to C, E to G) Left coronary artery seen on coronary angiography: (A to C) right anterior oblique and cranial views; (E to G) spider view. (D and H) Coronary computed tomography angiography. (A and E) Final angiography on index percutaneous coronary intervention (dotted lines represent implanted sirolimus-eluting stents [SES]). (C and G) Corresponding to B and F, respectively: stent migration (white dotted line in C and G) and occlusion with coronary artery aneurysm (CAA) (pink area in C and G) on follow-up coronary angiography. (D and H) Corresponding to B and F, respectively: stent migration (gray in D and H) and coronary flow snaking through the aneurysm (pink in D and H) on coronary computed tomography.

FUNDING SUPPORT AND AUTHOR DISCLOSURES

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

ADDRESS FOR CORRESPONDENCE: Dr Keisuke Shoji, Department of Cardiovascular Medicine, Kyoto Prefectural University of Medicine, 465 Kajii-cho Kawaramachi-Hirokoji, Kamigyo-ku, Kyoto 602-8566, Japan. E-mail: shjksk@koto.kpu-m.ac.jp. Twitter: @ksk09Cardiology.

REFERENCES

1. Akiyama M, Kaneko Y, Takeuchi T. Characteristics and prognosis of IgG4-related periaortitis/ periarteritis: a systematic literature review. *Autoimmun Rev.* 2019;18:102354.

2. Patel NR, Anzalone ML, Buja LM, Elghetany MT. Sudden cardiac death due to coronary artery involvement by IgG4-related disease: a rare, serious complication of a rare disease. *Arch Pathol Lab Med.* 2014;138:833–836. **3.** Koseki K, Yahagi K, Okuno T, et al. Immunoglobulin G4-related coronary periarteritis with multiple intracoronary images. *J Am Coll Cardiol Intv.* 2019;12:e59-e61.

KEY WORDS immunoglobulin G4-related coronary arteritis, immunoglobulin G4-related disease, stent migration **T** APPENDIX For supplemental videos and figures, please see the online version of this paper.