JACC: CASE REPORTS VOL. 29, 2024

IMAGING VIGNETTE

CLINICAL VIGNETTE

Atrial Tachycardia in the Coronary Sinus Communicating With Circumflex Artery Aneurysm

Mitsuaki Itoh, MD, PhD, Akira Shimane, MD, Tomofumi Takaya, MD, PhD, Hiroya Kawai, MD, PhD

ABSTRACT

Coronary artery fistula is a rare anomaly, and localized re-entrant atrial tachycardia (AT) in the coronary sinus (CS) has rarely been reported. We report a case in a patient with a left circumflex artery aneurysm associated with the CS fistula who underwent radiofrequency catheter ablation for localized re-entrant AT, which originated from the CS.
(J Am Coll Cardiol Case Rep 2024;29:102333) © 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/).

A 36-year-old man with a left circumflex (LCx) artery aneurysm associated with a coronary sinus (CS) fistula had recurrent episodes of drug-refractory atrial tachycardia (AT) and underwent radiofrequency catheter ablation for AT. He had an AT episode for the first time 3 years earlier, and his blood pressure and heart rate were 135/77 mm Hg and 136 beats/min, respectively, during AT. Transthoracic echocardiography showed normal left ventricular function, no pulmonary hypertension, and an LCx artery aneurysm. Computed tomography revealed a giant aneurysmal LCx artery communicating with the CS (**Figures 1A and 1B**, Video 1), no other cardiac anomaly, and no thrombus. The CS and great cardiac vein (GCV) were compressed by the LCx artery aneurysm, and the distal portion of the CS was enlarged because of the LCx artery aneurysm fistula (**Figure 1C**). Right-sided heart catheterization demonstrated that the pulmonary-to-systemic flow ratio was 1.25, and the patient had no symptoms of heart failure during follow-up. Oral anticoagulation therapy was administered before catheter ablation for AT. His blood pressure and heart rate during sinus rhythm were 129/73 mm Hg and 73 beats/min, respectively, without signs and symptoms of heart failure on admission.

Clinical AT with a tachycardia cycle length of 260 ms was easily induced. Electroanatomical mapping of the left atrium (LA), the right atrium, and the CS was performed using the Pentaray and CARTO mapping system (Biosense Webster). Moreover, an AT activation map of the LA and right atrium demonstrated a centrifugal activation pattern with the earliest activation site in the inferior LA and the CS ostium, suggesting that the AT originated from the CS (**Figure 1D**). Meanwhile, the LA and right atrium had no low-voltage area (LVA). During CS mapping, the LVA was observed in the distal enlarged portion of CS (**Figure 1E**). Double potentials, comprising the first fragmented and second dull potentials, were recorded in the distal portion of the CS (**Figure 1F**). We hypothesized that the first low-amplitude and fragmented component was the potential of the CS musculature, and the second dull component was far-field LA potential because the distal portion of the CS was separated from the LA by the LCx artery aneurysm. After manually annotating the first potential in the

From the Division of Cardiovascular Medicine, Hyogo Prefectural Harima-Himeji General Medical Center, Himeji, 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.

Manuscript received December 12, 2023; revised manuscript received February 12, 2024, accepted March 12, 2024.

ISSN 2666-0849 https://doi.org/10.1016/j.jaccas.2024.102333

ABBREVIATIONS AND ACRONYMS

AT = atrial tachycardia
CS = coronary sinus
GCV = great cardiac vein
LA = left atrium
LCx = left circumflex
LVA = low-voltage area

distal portion of the CS, mapping of the CS and GCV revealed localized re-entrant AT in the CS (Video 2). AT was immediately terminated after a single radiofrequency application targeting this fragmented potential (gray arrow site, radiofrequency power 20 W, target ablation index 250). After radiofrequency applications only at this site, atrial burst pacing failed to induce atrial tachyarrhythmia.

Coronary artery fistula is a rare anomaly that leads to myocardial ischemia, shunting, and volume overload.[1,2] To our knowledge, 38 patients with an LCx artery aneurysm communicating with the CS have been previously reported. The LCx artery aneurysm with CS fistula resulted in enlargement of the CS and fibrosis of the CS musculature. Localized re-entrant AT was defined as an activation circuit with a diameter of <2 cm, demonstrating continuous fragmented electrograms.[3] The first fragmented potential for maintenance of localized re-entrant AT resulted from compression of the CS from the LCx artery aneurysm. The volume overload and compression of the CS by the LCx artery aneurysm led to the occurrence of localized re-entrant AT in the distal portion of the CS. To our knowledge, this is the first reported case of localized re-entrant AT in the CS communicating with an LCx artery aneurysm.

ACKNOWLEDGMENTS The authors are grateful for the support of the entire staff of the electrophysiological laboratories at Hyogo Prefectural Harima-Himeji General Medical Center.

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 Mitsuaki Itoh, Division of Cardiovascular Medicine, Hyogo Prefectural Harima-Himeji General Medical Center, 3-264, Kamiya-cho, Himeji, Hyogo 670-8560, Japan. E-mail: light0philosophy@yahoo.co.jp. @Liphi6.

REFERENCES

1. Challoumas D, Pericleous A, Dimitrakaki IA, Danelatos C, Dimitrakakis G. Coronary arteriovenous fistulae: a review. *Int J Angiol*. 2014;23:1-10.

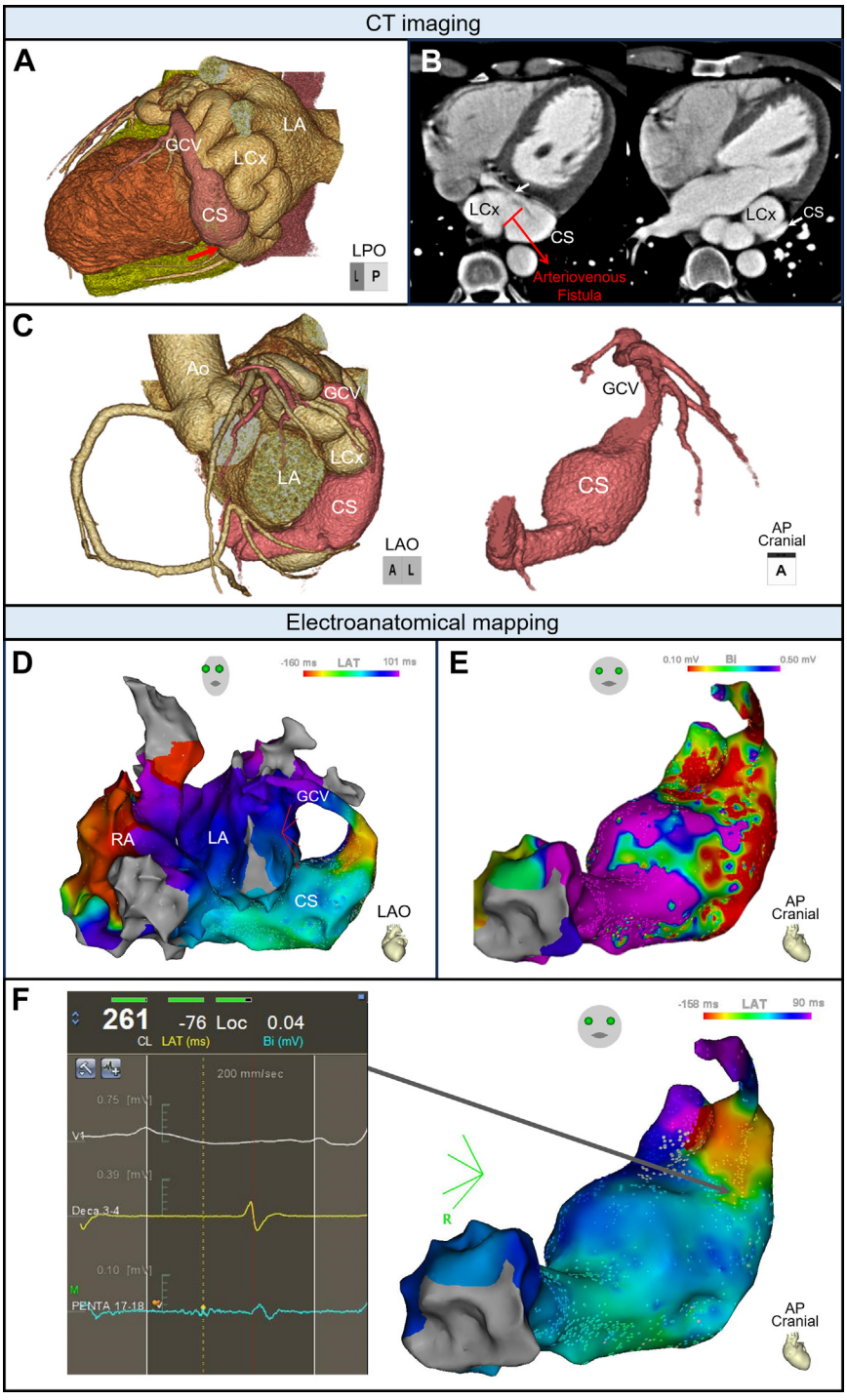
2. Libertini R, Wallbridge D, Jones HR, Gunning M, Satur CM. Giant circumflex artery aneurysm with a coronary sinus fistula. *Ann Thorac Surg*. 2018;106:e223-e225.

3. Sanders P, Hocini M, Jaïs P, et al. Characterization of focal atrial tachycardia using high-density mapping. *J Am Coll Cardiol*. 2005;46:2088-2099.

KEY WORDS atrial tachycardia, circumflex artery aneurysm, coronary sinus fistula

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

FIGURE 1 **CS Communicating With LCx Artery Aneurysm and Atrial Tachycardia Mapping**



(A to C) Computed tomography (CT) imaging reveals a giant left circumflex (LCx) artery aneurysm communicating with the coronary sinus (CS). The red arrow indicates the arteriovenous fistula between the coronary sinus and the left circumflex artery aneurysm. The coronary sinus is compressed by the left circumflex artery aneurysm and enlarged because of this fistula. The white arrow indicates the compressed portion of the coronary sinus. (D) Atrial tachycardia activation map of the left atrium (LA) and right atrium (RA) demonstrates a centrifugal activation pattern with the earliest activation site in the inferior left atrium and coronary sinus ostium, suggesting that the atrial tachycardia originates from the coronary sinus. (E) A low-voltage area was observed in the distal portion of the coronary sinus. (F) A reannotated activation map of coronary sinus demonstrates localized re-entrant atrial tachycardia in the distal portion of the coronary sinus. We hypothesized that the first low-amplitude and fragmented component was the potential of the coronary sinus musculature, and the second dull component was far-field left atrial potential. The gray arrow indicates the site of recording double potentials and radiofrequency application. Supplemental figures for D, E, and F were additional figures of D, E, and F in this article. A = anterior; Ao = aorta; AP = anteroposterior; Bi = bipolar voltage; CL = cycle length; GCV = great cardiac vein; L = lateral; LAO = left anterior oblique; LAT = local activation time; LPO = left posterior oblique; P = posterior.