ESC European Society of Cardiology

European Heart Journal - Case Reports (2024) **8**, ytae575
https://doi.org/10.1093/ehjcr/ytae575

CASE REPORT
Cardiovascular imaging

Suspected contained rupture of a coronary artery aneurysm followed by a re-rupture: a case report

Ryota Miyamoto [1]*, Norihito Kageyama[1], Ryo Suzuki[1], Tatsuya Kokawa [2], and Hiroyuki Fujinaga[1]

[1]Department of Cardiology, Tokushima Prefectural Miyoshi Hospital, 815-2 Shima, Ikeda-cho, Miyoshi, Tokushima 778-8503, Japan; and [2]Department of Cardiology, Tokushima Red Cross Hospital, 103 Irinokuchi, Komatsushima-cho, Komatsushima, Tokushima 773-8502, Japan

Received 11 March 2024; revised 27 May 2024; accepted 18 October 2024; online publish-ahead-of-print 24 October 2024

Background Most coronary artery aneurysms (CAAs) are clinically asymptomatic and are only detected incidentally during cardiac imaging. However, CAAs can cause fatal complications such as cardiac tamponade following a rupture. Reports of contained ruptures of CAAs are limited.

Case summary An 89-year-old man had a severe, acute-onset chest pain 2 days prior. The patient was referred to our hospital because of residual dyspnoea and a fever. The blood tests exhibited high C-reactive protein and D-dimer levels; moreover, contrast-enhanced computed tomography (CT) and transthoracic echocardiography revealed a diffuse mild pericardial effusion, which had not been detected six months prior. The fever and pericardial effusion persisted, and the main cause was not clearly understood. On Day 19, the patient had a sudden loss of consciousness after chest pain. Contrast-enhanced CT showed an increase in the pericardial effusion, and coronary angiography showed a CAA in the proximal right coronary artery with extravasations that probably sealed. A re-rupture following a contained rupture of the CAA was considered, and it was suspected that the prolonged fever and pericardial effusion were related to the contained rupture associated with the first episode of chest pain.

Discussion Our case showed that a contained rupture of a CAA can cause a fatal event by a re-rupture and may present with a prolonged fever and pericardial effusion. A contained rupture of a CAA should be considered as a differential diagnosis in patients with a prolonged fever and pericardial effusion of unknown origin after an acute onset of chest pain.

Keywords Coronary artery aneurysm • Contained rupture • Fever of unknown origin • Pericardial effusion • Case report

ESC curriculum 2.2 Echocardiography • 2.4 Cardiac computed tomography • 3.1 Coronary artery disease • 3.4 Coronary angiography • 6.6 Pericardial disease

Learning points

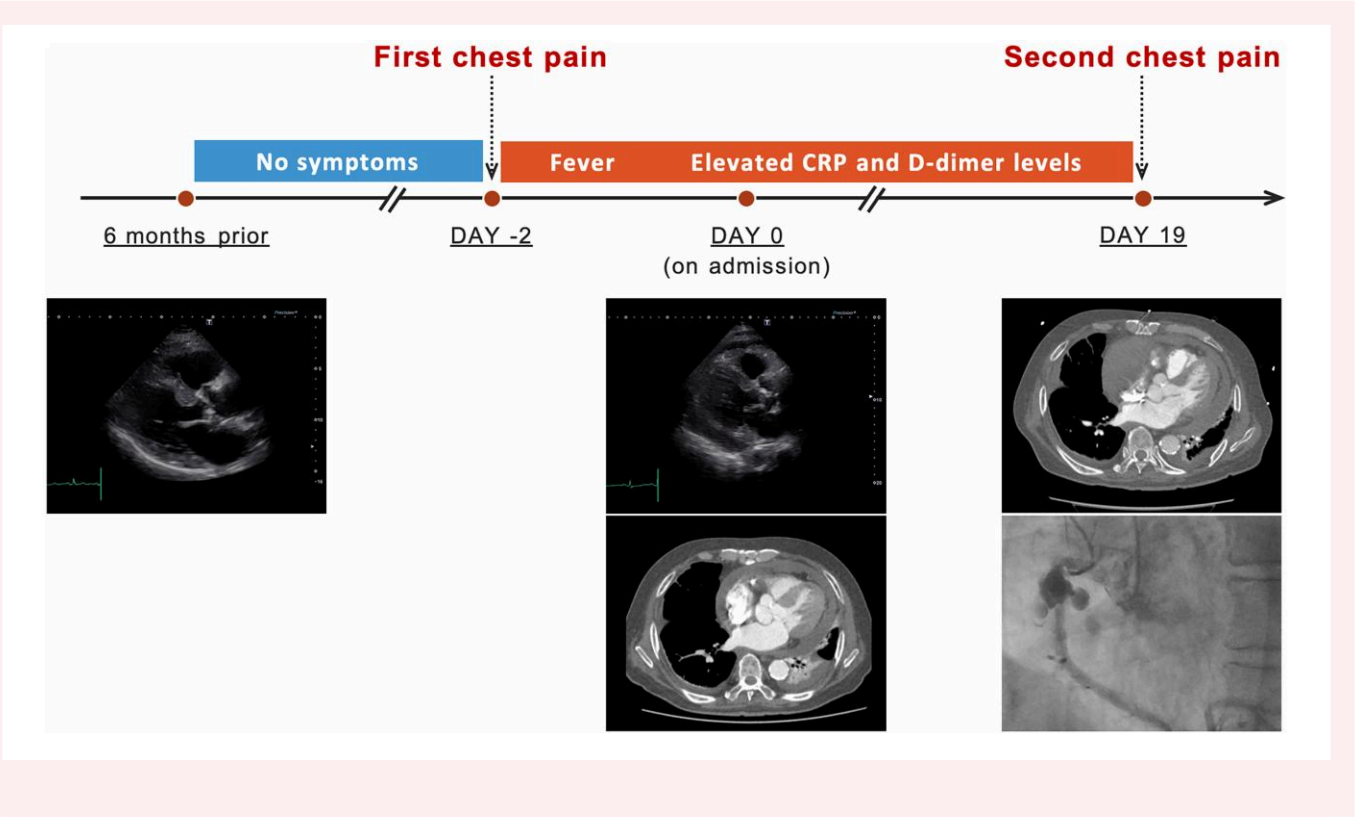
- Coronary artery aneurysms (CAAs) can cause fatal cardiac events including acute myocardial infarction and acute cardiac tamponade.
- A contained rupture of a CAA can cause fatal events by a re-rupture and may present with a prolonged fever and pericardial effusion.
- A contained rupture of a CAA should be considered as a differential diagnosis in patients with a prolonged fever and pericardial effusion of unknown origin after an acute onset of chest pain.

* Corresponding author. Tel: +81 883 72 1131, Fax: +81 883 72 6910, Email: ballballryota8@gmail.com
Handling Editor: Milenko Zoran Cankovic
Peer-reviewers: Giuseppe Musumeci; Jan Henzel; Kashan Ali
Compliance Editor: Franca Morselli

Introduction

Most coronary artery aneurysms (CAAs) are clinically asymptomatic and are only detected incidentally during cardiac imaging. However, CAAs can cause fatal complications such as acute myocardial infarction or cardiac tamponade.[1,2] We herein report a patient with a suspected contained rupture of a CAA, which finally re-ruptured, and who presented with a prolonged fever and pericardial effusion.

Summary figure



Case presentation

An 89-year-old man with a history of hypertension, dyslipidaemia, and moderate aortic stenosis with regurgitation had an extremely severe, acute onset of chest pain radiating to the left shoulder, which lasted for about 2 h, 2 days prior. Although the symptoms improved, the patient was referred to our hospital because of residual dyspnoea and a fever. On the initial assessment, the patient was stable haemodynamically, and the pulse rate was 96 beats/min, blood pressure 128/70 mmHg, respiratory rate 20 breaths/min, oxygen saturation 95% on 1 L of oxygen, and body temperature 37.2°C. A physical examination revealed no abnormalities other than a systolic murmur in the aortic area and slight pretibial oedema. The electrocardiogram (ECG) exhibited sinus rhythm, but no significant ST-T segment abnormalities. The blood tests exhibited elevated white blood cell (10 800/μL, normal value; 3500–9000/μL), C-reactive protein (26.6 mg/dL, <0.4 mg/dL), D-dimer (10.4 μg/mL, <0.9 μg/mL), and brain natriuretic peptide (393.5 pg/mL, <18.4 pg/mL) levels, but the cardiac enzymes including troponin were not elevated. Chest radiography revealed a significant enlargement of the cardiac silhouette as compared to six months prior (*Figure 1A* and *B*). Transthoracic echocardiography revealed a diffuse mild pericardial effusion without a collapse of the right heart, which was not detected six months prior (*Figure 1C* and *D*). Non-ECG-gated contrast-enhanced computed tomography (CT) with a 320-row detector revealed a diffuse mild pericardial effusion, slight bilateral pleural effusion, and severe calcifications of the coronary arteries. The mean CT value near the right heart, 26.5 Hounsfield unit (HU), was slightly higher than that near the left heart, 18.8 HU (*Figure 1E–G*; Supplementary material online, *Video 1*).

During the hospitalization, the patient was observed without antibiotics because of no evidence of a bacterial infection, and repeated blood, urine, and sputum cultures were negative. However, a fever exceeding 38°C and high C-reactive protein and D-dimer levels persisted. On Day 4, the possibility of viral pericarditis was considered, and aspirin 1200 mg/day and colchicine 0.5 mg/day were introduced. The fever over 38°C disappeared and the C-reactive protein level decreased gradually, but a fever over 37°C and a high C-reactive protein level remained (*Figure 2*). On Day 9, edoxaban 30 mg/day was introduced because paroxysmal atrial fibrillation was repeatedly detected. The pericardial effusion did not increase during the hospitalization, and a pericardial paracentesis was considered difficult. The main cause of the symptoms and abnormal findings was not clearly understood.

On Day 19, the patient had a sudden loss of consciousness after complaining of severe chest pain and regained consciousness within 10 s. The ECG exhibited ST-elevation in the inferior leads, but the change recovered within 15 min (*Figure 3A* and *B*). Although the possibility of acute aortic dissection was considered at that time,

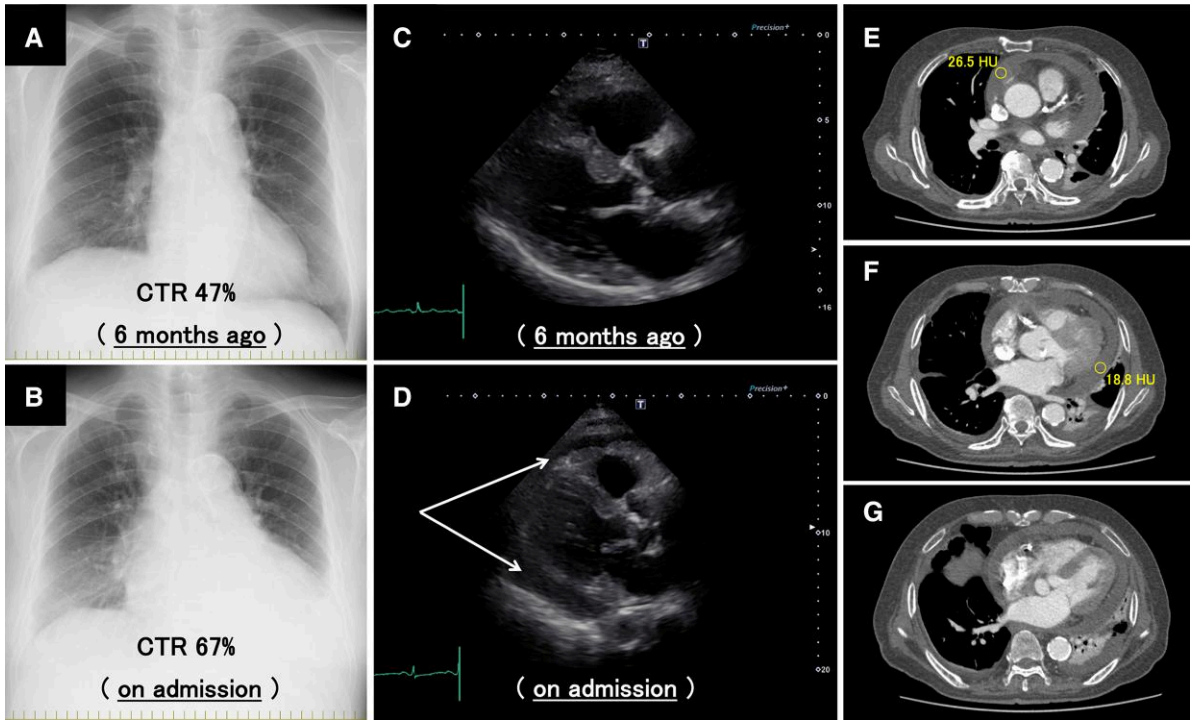


Figure 1 Although chest radiography six months prior did not show any enlargement of the cardiac silhouette [cardio-thoracic ratio (CTR) of 47%] (*A*), that performed on admission revealed a significant enlargement (CTR of 67%) (*B*). Transthoracic echocardiography six months prior did not show any abnormal findings of a pericardial effusion (*C*); however, a diffuse mild pericardial effusion without a collapse of the right heart was observed on admission (arrows) (*D*). Non-electrocardiogram-gated contrast-enhanced computed tomography (CT) with a 320-row detector revealed a diffuse mild pericardial effusion, slight bilateral pleural effusion, and severe calcifications of the coronary arteries (*E*–*G*). The mean CT value near the right heart was 26.5 Hounsfield unit (HU), which was slightly higher than that near the left heart, 18.8 HU (circle) (*E* and *F*).

transthoracic echocardiography and non-ECG-gated contrast-enhanced CT showed an increase in the pericardial effusion, especially on the surface of the right heart, and there was contrast extravasation from the proximal right coronary artery (RCA) detected on the CT. The mean CT value near the right heart, 55.5 HU, was significantly higher than that near the left heart, 23.8 HU (*Figure 3C–G*; Supplementary material online, *Video 2*). Emergency coronary angiography showed an irregular margin of the CAA in the proximal RCA, pulsatile contrast extravasations that probably sealed from three sites of the CAA, and a Thrombolysis in Myocardial Infarction (TIMI) grade 3 flow in both coronary arteries (*Figure 4A–D*; Supplementary material online, *Videos 3–6*). Then, a re-rupture following a contained rupture of the CAA was considered. Moreover, it was suspected that the patient's prolonged fever, pericardial effusion, and high C-reactive protein and D-dimer levels were related to the contained rupture of the CAA associated with the first episode of chest pain before the hospitalization. The pericardial effusion was thought mainly to consist of a haematoma, and both haemostasis and removing the haematoma from the pericardial cavity were considered necessary to prevent cardiac tamponade. Thus, the patient was transported to another hospital with cardiovascular surgery support, but haemostasis by percutaneous coronary intervention (PCI) using two 3.5/20 mm PK Papyrus covered stents (Biotronik, Berlin, Germany) was performed because the patient and family refused surgery due to the old age after an explanation by the heart team (*Figure 4E–G*; Supplementary material online, *Video 7*). The haemostasis was successful; however, the haemodynamics did not completely recover and the patient died in the end.

Discussion

The course in this patient provided two important clinical suggestions: (i) a contained rupture of a CAA can cause a fatal event by a re-rupture, and (ii) a contained rupture of a CAA may present with a prolonged fever and pericardial effusion.

Although most patients with CAAs are asymptomatic, CAAs can cause lethal events including acute myocardial infarction and cardiac tamponade following a rupture.[1,2] If a CAA ruptures, blood dissects around the myocardial layers and ruptures internally into the cardiac chambers, or externally into the pericardial cavity.[3] Contained ruptures of atheromatous CAAs and mycotic CAAs have been reported, but reports of contained ruptures of CAAs are limited.[4–6] In this case, the patient had felt no symptoms until the first episode of chest pain; moreover, the transthoracic echocardiography six months prior showed no abnormal findings of a pericardial effusion. However, the pericardial effusion was newly detected after the first episode of chest pain, and it suddenly increased just after the second episode of chest pain. This suggested the possibility that the first episode of chest pain was associated with a rupture of the CAA externally into the pericardial cavity followed by sealing with a haematoma, and the CAA re-ruptured at the time of the second episode of chest pain. Actually, the emergency coronary angiography showed that the CAA had extravasations that probably sealed. The mean CT value near the right heart was significantly higher than that near the left heart on the contrast-enhanced CT just after the second episode of chest pain. That was thought to suggest

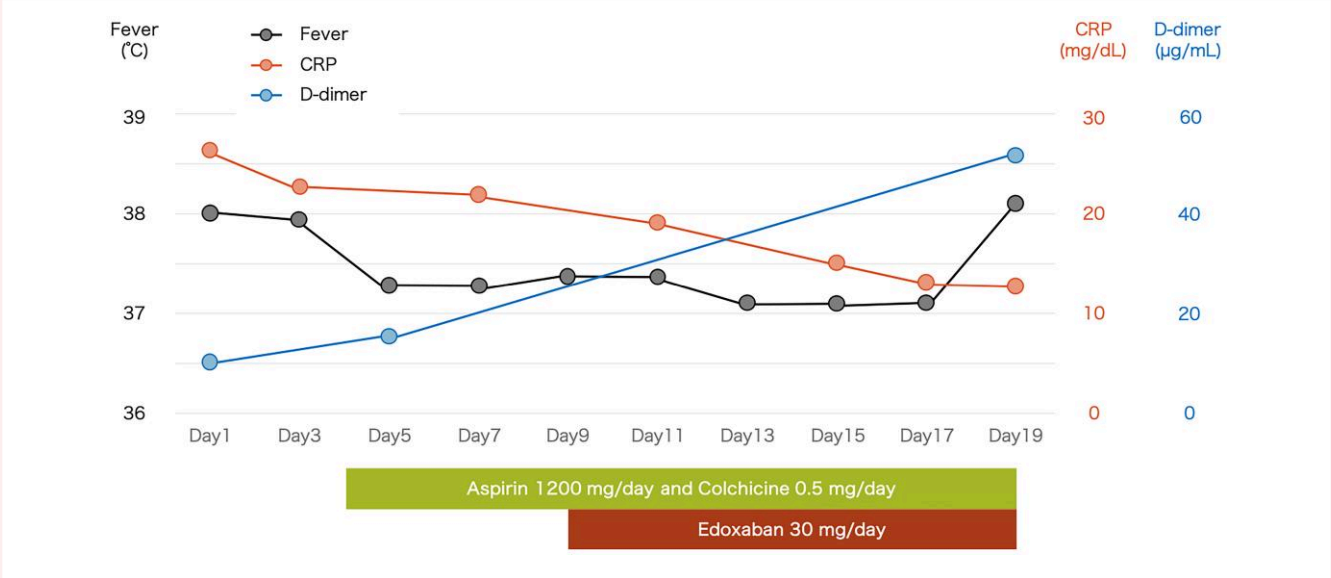


Figure 2 Time course of the fever and C-reactive protein and D-dimer levels in addition to the medications during the hospitalization. CRP, C-reactive protein.

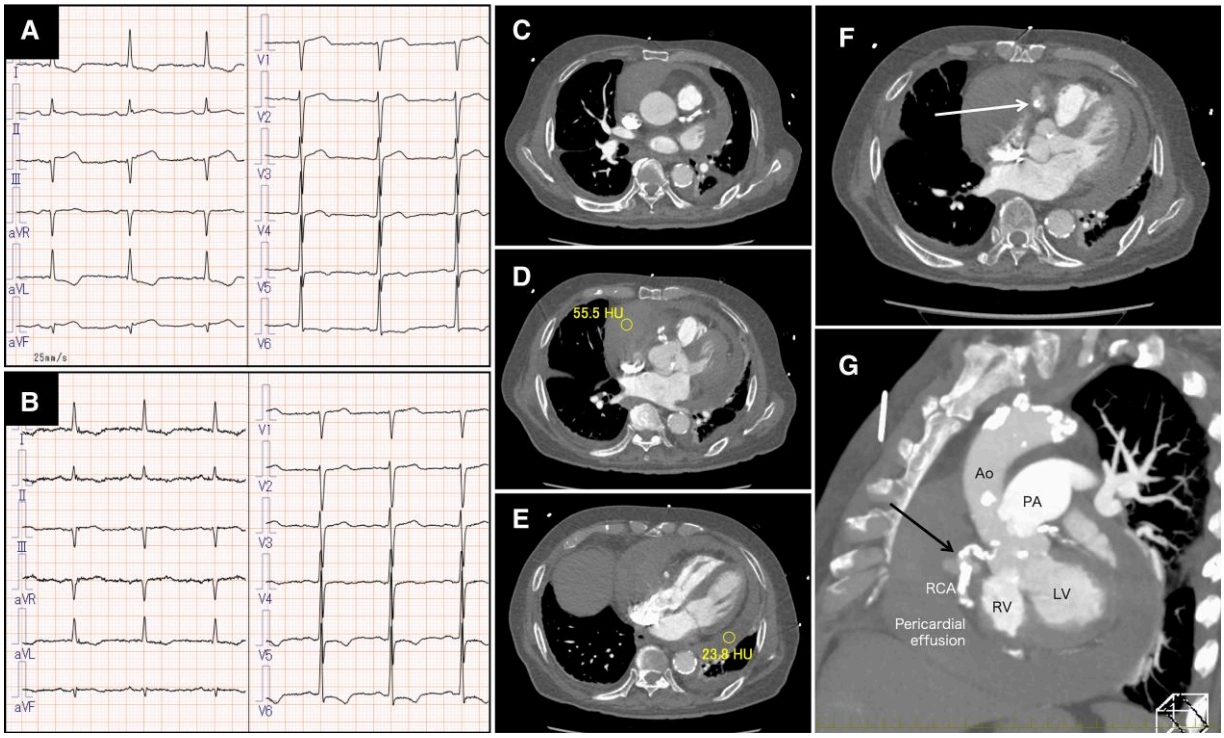


Figure 3 Just after the second episode of chest pain and loss of consciousness, the electrocardiogram (ECG) exhibited ST-elevation in the inferior leads (*A*), but the change recovered within 15 min (*B*). Non-ECG-gated contrast-enhanced computed tomography (CT) showed an increase in the pericardial effusion, especially on the surface of the right atrium and ventricle (*C*–*E*), and the mean CT value near the right heart was 55.5 Hounsfield unit (HU), which was significantly higher than that near the left heart, 23.8 HU (circle) (*D* and *E*). There was contrast extravasation from the proximal right coronary artery in the axial plane (white arrow) (*F*) and in the maximum intensity projection (black arrow) (*G*). Ao, aorta; LV, left ventricle; PA, pulmonary artery; RCA, right coronary artery; RV, right ventricle.

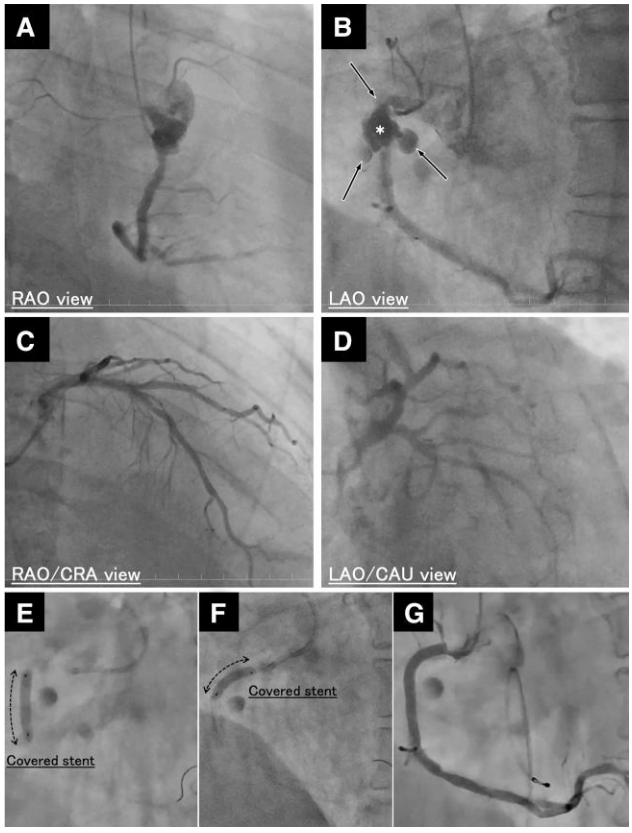


Figure 4 Emergency coronary angiography showed an irregular margin of the coronary artery aneurysm (CAA) in the proximal right coronary artery (RCA) (asterisk), pulsatile contrast extravasations that probably sealed from three sites of the CAA (black arrows) (*A* and *B*), and a Thrombolysis in Myocardial Infarction (TIMI) grade 3 flow in the right and left coronary arteries (*A*–*D*). Haemostasis by percutaneous coronary intervention using two 3.5/20 mm PK Papyrus covered stents was performed (dot arrow) (*E* and *F*). The final angiography showed that the CAA was successfully sealed by the covered stents, and there was a TIMI grade 3 flow in the RCA (*G*). CAU, caudal; CRA, cranial; LAO, left anterior oblique; RAO, right anterior oblique.

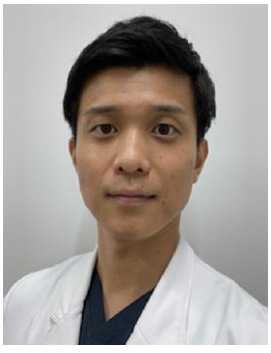
there was a haematoma surrounding the CAA. In the literature, the CT values have been reported to be higher, especially in patients with a haemopericardium.[7]

In this case, the patient had a prolonged fever after the first episode of chest pain, and high C-reactive protein and D-dimer levels persisted during the hospitalization. The main cause could not be clearly explained as there was no evidence of a bacterial infection, collagen disease, or malignancy, which are well-known causes of a fever of unknown origin. The fever and high C-reactive protein and D-dimer levels might have been related to the inflammation induced by the contained rupture of the CAA. Actually, post-cardiac injury syndrome including pericarditis and a high C-reactive protein level caused by some reasons such as a distal wire perforation during PCI has been reported.[8]

The CAA was first diagnosed on the contrast-enhanced CT and coronary angiography just after the second episode of chest pain. The reason why the CAA could not be clearly diagnosed on the first contrast-enhanced CT on admission was not completely understood, but the CAA might have been partially thrombosed. Moreover, severe calcifications of the coronary arteries and non-ECG-gated CT might have also precluded a detailed assessment. A pericardial paracentesis was considered difficult because the mild pericardial effusion did not increase, but it could have been useful for the diagnosis. Although the haemostasis with the covered stents was successful and the final angiography showed a TIMI grade 3 flow in the RCA, the patient's haemodynamics did not completely recover. Cardiac tamponade due to a haematoma in the pericardial cavity was considered to have been involved.

Herein, we reported a patient with a suspected contained rupture of a CAA followed by a re-rupture. A contained rupture of a CAA may be overlooked; thus, it should be considered as a differential diagnosis in patients with a prolonged fever and pericardial effusion of unknown origin after an acute onset of chest pain.

Lead author biography



Ryota Miyamoto, MD, works as a cardiologist at Tokushima Prefectural Miyoshi Hospital in Japan. He graduated from Tokushima University, Faculty of Medicine in 2017. His areas of interest include structural heart disease, cardiogenic shock, and cardiac rehabilitation.

Supplementary material

Supplementary material is available at *European Heart Journal – Case Reports* online.

Acknowledgements

We are grateful to Mr John Martin for his grammatical assistance.

Consent: The authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: None declared.

Funding

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

Data availability

The data underlying this article will be shared upon reasonable request to the corresponding author.

References

1. Kawsara A, Núñez Gil IJ, Alqahtani F, Moreland J, Rihal CS, Alkhouli M. Management of coronary artery aneurysms. *JACC Cardiovasc Interv* 2018;**11**:1211–1223.
2. Hiramori S, Hoshino K, Hioki H, Yahikozawa K, Shinozaki N, Ichinose H, et al. Spontaneous rupture of a giant coronary artery aneurysm causing cardiac tamponade: a case report. *J Caldiol Cases* 2011;**3**:e119–e122.
3. Berrizbeitia LD, Samuels LE. Ruptured right coronary artery aneurysm presenting as a myocardial mass. *Ann Thorac Surg* 2002;**73**:971–973.
4. Ramirez JL, Kratz JR, Wiselthaler GM. Giant right coronary artery aneurysm presenting as cardiac tamponade. *Interact Cardiovasc Thorac Surg* 2018;**27**:787–789.
5. Khoshbin E, Sogliani F, Tang A. Staged hybrid treatment for contained rupture of a large right coronary artery aneurysm. *Innovations* 2014;**9**:69–71.
6. McNamara NJ, Plunkett BT. Management of a mycotic right coronary artery aneurysm with contained rupture. *JACC Case Rep* 2022;**4**:694–698.
7. Çetin MS, Özcan Çetin EH, Özdemir M, Topaloğlu S, Aras D, Temizhan A, et al. Effectiveness of computed tomography attenuation values in characterization of pericardial effusion. *Anatol J Cardiol* 2017;**17**:322–327.
8. Malik J, Zaidi SMJ, Rana AS, Haider A, Tahir S. Post-cardiac injury syndrome: an evidence-based approach to diagnosis and treatment. *Am Heart J Plus Caldiol Res Pract* 2021;**12**:100068.