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【 CASE REPORT 】

Sudden Cardiac Arrest Due to Spontaneous Coronary Artery Rupture - A Case Report with a Diagnostic Challenge -

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Abstract:
Spontaneous coronary artery rupture (SCAR) is a rare, life-threatening disease, and the diagnosis is often challenging. We herein report a 70-year-old man who suffered sudden cardiac arrest due to SCAR with pericardial fluid. At first, emergent coronary angiography (CAG) failed to detect abnormalities. The emergent operation revealed that the presence of pericardial fluid was caused by bleeding that had spontaneously occurred at the left circumflex artery (LCx). A careful retrospective CAG review showed slight contrast spillage from the distal LCx. SCAR should be suspected in patients with unknown etiology of pericardial effusion, and careful inspection of CAG is necessary.

Key words: spontaneous coronary artery rupture, pericardial fluid, coronary angiography

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Introduction

Spontaneous coronary artery rupture (SCAR) is a life-threatening disorder (1). However, since it is a rare entity, its diagnosis is often missed or delayed (2). We herein report a case of SCAR that was difficult to diagnose even with coronary angiography (CAG) and discuss its characteristics and management.

Case Report

A 70-year-old man on dialysis due to diabetic nephropathy was referred to our hospital complaining of dyspnea and abdominal pain. He had no family history of hereditary disease. Ten days before his visit, he had undergone CAG. It showed chronic total occlusion of the right coronary artery (RCA) and significant stenosis of the left anterior descending artery (LAD), and coronary artery bypass grafting was scheduled. On arrival, he seemed uncomfortable and agitated. Vital signs revealed a blood pressure of 144/79 mmHg, pulse rate of 85 beats per minute, and oxygen saturation of 96% with a non-rebreather mask. An electrocardiogram (ECG) demonstrated ST-segment depression in anterior leads, and transthoracic echocardiography (TTE) showed left ventricular hypertrophy with a reduced systolic function. Furthermore, moderate pericardial effusion was observed without signs of cardiac tamponade. We noted no intimal flaps in the ascending aorta by echocardiography. Based on his medical history and examination findings, we assumed that the pericardial effusion was due to uremia.

While we considered performing further investigations, the patient suddenly went into pulseless electrical activity cardiac rhythm. Immediate cardiopulmonary resuscitation (CPR) was initiated, but the return of spontaneous circulation was not achieved. Therefore, veno-arterial extracorporeal membrane oxygenation (VA-ECMO) was inserted. Repeat TTE subsequently revealed increased pericardial effusion (Fig. 1), and the patient seemed to have had cardiac tamponade.

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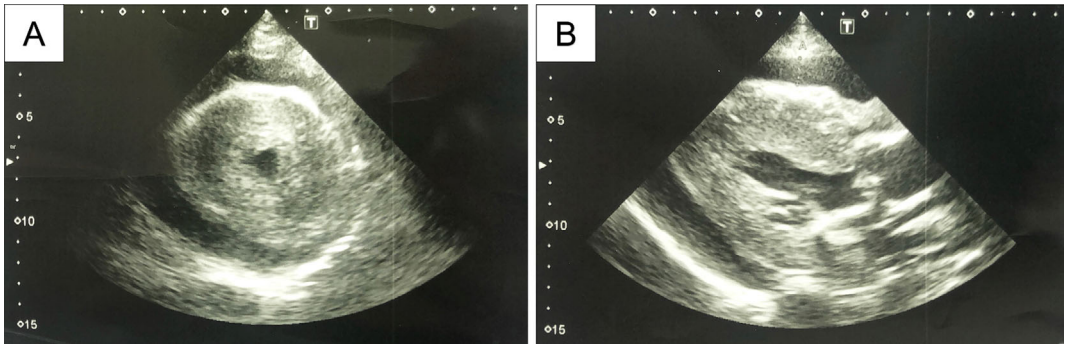

Figure 1. **Transthoracic echocardiography after veno-arterial extracorporeal membrane oxygenation shows a large amount of pericardial fluid. (A) Left chamber short-axis view and (B) long-axis view.**

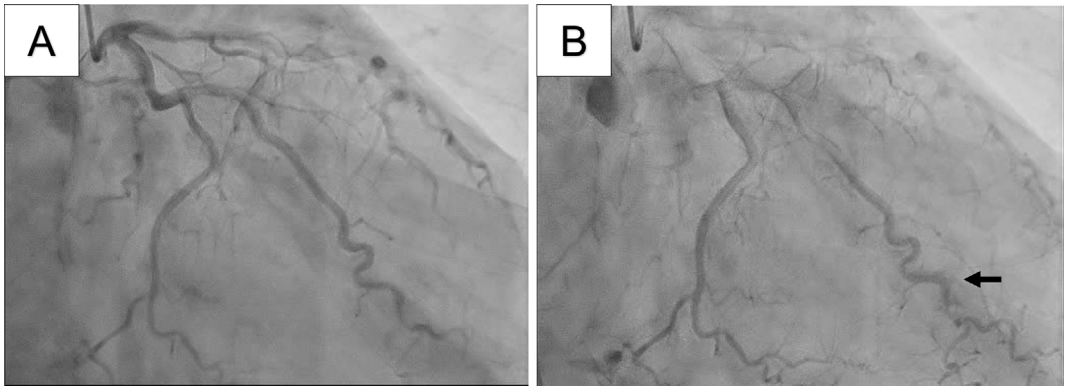

Figure 2. **Left coronary angiography after veno-arterial extracorporeal membrane oxygenation insertion. (A) In the early phase, there were no findings that might have caused pericardial effusion. (B) The late phase detected slight spillage of contrast media from the left circumflex artery (arrow).**

To investigate the cause of pericardial effusion, emergent CAG was performed. At first glance, CAG demonstrated the same findings of RCA occlusion and LAD stenosis as seen 10 days earlier, and there were no findings that might have caused pericardial effusion (Fig. 2A). Pericardial drainage was then performed. However, a large amount of bloody pericardial fluid continued to drain, leaving the patient hemodynamically unstable. Thus, he underwent emergent surgery for the diagnosis and treatment.

Transesophageal echocardiography just before the surgery showed no findings of aortic dissection. Intraoperative inspection revealed bleeding in the distal left circumflex artery (LCx) without coronary dissection or aneurysm (Fig. 3). Hemostasis was achieved by direct suture, and coronary artery bypass grafting to the LAD and the RCA was also performed. In the retrospective review, the late phase of CAG revealed slight spillage of contrast media from the distal LCx corresponding to the bleeding site (Fig. 2B). After surgery, his hemodynamics gradually stabilized, and VA-ECMO was successfully removed six days after the operation. However, the patient did not regain consciousness due to hypoxic encephalopathy caused by cardiopulmonary arrest. He remained in a coma and died 41 days after the operation due to sepsis.

Discussion

Coronary artery rupture is usually associated with various underlying conditions, such as percutaneous coronary intervention procedures, aneurysm, and Ehlers-Danlos syndrome (3, 4). Furthermore, blunt injury to a vessel associated with CPR can cause coronary rupture (5). In the present case, CAG performed 10 days before the onset confirmed no angiographic abnormality at the ruptured site, and no invasive manipulation was done, such as wire insertion. In addition, a moderate level of pericardial effusion was already present before CPR was initiated. Given with his medical history, the sudden increase in the pericardial fluid after the visit, and the intraoperative findings of no anatomical changes such as aneurysm and dissection, we concluded that the rupture occurred spontaneously.

In this patient, given the diffuse atherosclerosis in the entire coronary artery, particularly in the LAD and RCA, atherosclerotic change at the bleeding point not detectable in CAG may have been an underlying cause of SCAR (2). Although SCAR without any known underlying vascular disease is a rare entity, we believe that this disease can be missed and may be underreported due to its sudden onset and fatal outcome in a short period. Therefore, an early and

accurate diagnosis is essential.

The most critical point for the diagnosis is to be highly suspicious of SCAR. A literature search in PubMed identified 16 SCAR cases (1, 2, 6-17) (Table). The age ranged between 37 to 74 years old, with most in their 50s or 60s. More than 80% (13 of 16) of cases occurred in men. One patient was on hemodialysis. Patients with SCAR usually present with symptoms associated with pericardial effusion with two different clinical scenarios: nonspecific symptoms, such as fatigue and dyspnea, maintaining the patient's hemodynamics; and cardiogenic shock. In both situations, the patient's unstable condition often makes it challenging to examine the bleeding site thoroughly. While the usefulness of computed tomography angiography (CTA) has been reported (9), CAG seems to be the standard modality for diagnosing SCAR, with its high spatial and temporal resolution (2, 7, 13, 15, 16). If needed, transcatheter hemostasis can be performed following diagnostic CAG. However, CAG does not always demonstrate extravasation clearly, as in our case, because the increase in pressure in the pericardial space and the decrease in coronary flow due to cardiac tamponade reduce blood leakage. In addition, heparin use and pressure release by drainage may worsen bleeding, so caution should be exercised. In our case, if CAG had been performed after pericardial drainage, the spillage of blood might have been clearer. In any case, we should evaluate CAG images carefully to avoid missing any contrast spillover, a sign of coronary rupture. If the patient is hemodynamically stable, ECG-gated coronary CTA may be a viable option for detecting coronary rupture and ruling out other causes of pericardial effusion. However, patients in critical conditions who cannot tolerate these examinations need emergent surgery for the diagnosis and treatment (1, 8, 10, 12, 14, 17).

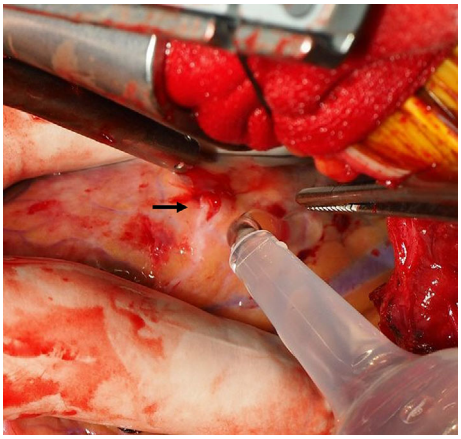

Figure 3. Intraoperative inspection revealed a rupture in the left circumflex artery (arrow).

The treatment strategy of SCAR depends on several factors, including the etiology, the location and severity of bleeding, and the general condition of the patients. If CAG can identify the bleeding source clearly, we can consider transcatheter hemostasis with covered stents (15) or coil embolization (2, 7) according to the patient's status and coronary anatomy. Various strategies have been reported in cases requiring surgery. In addition to the direct suture performed in this case, the usefulness of ligation with bypass grafting (11, 16) and venous patch repair (10, 12, 14) has been reported. In our case, if we had been able to diagnose the rupture by carefully reviewing the CAG findings, transcatheter hemostasis may have been a choice.

In conclusion, the diagnosis of SCAR can be missed or delayed, even if CAG is performed. SCAR should always be suspected in patients with pericardial effusion of unknown etiology. We need to review images with great care when we perform CAG.

The authors state that they have no Conflict of Interest (COI).

Table. Cases of Spontaneous Coronary Artery Rupture without Underlying Condition in the Literature.

References	Age	Sex	HD	Symptoms	Diagnosis	Treatment
(1)	56	M	No	Cardiogenic shock	Intraoperative	Surgery (suture)
(2)	62	F	Yes	Non-specific	CAG	Catheter (coil embolization)
(6)	48	M	No	Non-specific	Intraoperative	Surgery (suture)
(7)	74	M	No	Non-specific	CAG	Catheter (coil embolizatoin, gelatine sponge)
(8)	67	M	No	Cardiogenic shock	Intraoperative	Surgery (suture)
(9)	37	M	No	Non-specific	Intraoperative	Surgery (ligation+suture)
(10)	41	M	No	Non-specific	Intraoperative	Surgery (patch repair)
(11)	65	F	No	Non-specific	CT	Surgery (ligation+CABG)
(12)	43	M	No	Cardiogenic shock	Intraoperative	Surgery (patch repair)
(13)	65	M	No	Non-specific	CAG	Surgery (hematoma evacuation+CABG)
(14)	50	M	No	Non-specific	Intraoperative	Surgery (suture)
	62	M	No	Cardiogenic shock	Intraoperative	Surgery (patch repair)
	69	M	No	Cardiogenic shock	Intraoperative	Surgery (patch repair)
(15)	52	F	No	Cardiogenic shock	CAG	Catheter (covered stent)
(16)	69	M	No	Cardiogenic shock	CAG	Surgery (ligation+CABG)
(17)	58	M	No	Cardiogenic shock	Intraoperative	Surgery (ligation)

CABG: coronary artery bypass grafting, CAG: coronary angiography, CT: computed tomography, HD: hemodialysis

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