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"No-Touch" Saphenous Vein Grafting and Coronary Aneurysm Ligation in an Adult Patient with Suspected Prior Kawasaki Disease

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Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Female, 68-year-old
Final Diagnosis: Acute coronary syndrome • Kawasaki disease
Symptoms: Chest pain
Clinical Procedure: —
Specialty: Cardiac Surgery

Objective: Rare disease

Background: Coronary artery aneurysms in patients with Kawasaki disease may develop acute myocardial infarction. It is challenging to achieve complete revascularization solely through percutaneous coronary intervention in these patients. Therefore, coronary artery bypass grafting is often necessary.

Case Report: We present a case of a 68-year-old woman who developed multiple acute myocardial infarctions due to giant aneurysms formed in the right coronary artery (RCA) and the left circumflex artery (LCx). We diagnosed the cause of the aneurysms as Kawasaki disease based on the coronary angiogram, laboratory results, and family history. After the primary balloon angioplasty, we conducted coronary artery bypass grafting, which involved grafting 2 vessels to the LCx and 1 vessel to the RCA. The internal thoracic arteries, which are the standard graft vessels, were occluded, most likely due to Kawasaki disease vasculitis. Instead, we used saphenous vein grafts harvested using the "no-touch" technique, which preserves the perivascular adipose tissue, to improve the long-term patency. In addition, we ligated the LCx aneurysm to prevent occlusion of the grafts and rupture of the aneurysm. Four years after the uneventful discharge, the patient is in good health and coronary computed tomography angiography revealed good patency of all grafts.

Conclusions: This report highlights a successful combination of "no-touch" saphenous vein grafting and coronary aneurysm ligation in an adult patient with Kawasaki disease. These techniques may be especially useful for this vasculitic illness which is often associated with occlusion of internal thoracic arteries.

Keywords: Mucocutaneous Lymph Node Syndrome • Coronary Aneurysm • Coronary Artery Bypass • Vasculitis • Cardiac Surgical Procedures • Acute Coronary Syndrome

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Introduction

Kawasaki disease (KD), first reported in 1967 [1,2], is a systemic vasculitis that predominantly affects small children. Patients are treated with intravenous immunoglobulin (IVIG) to prevent the development of coronary artery aneurysms and acute myocardial infarction [3,4]. The number of cases with coronary aneurysm(s) has steadily decreased after the advent of IVIG treatment [5,6]. However, adult patients who developed KD before 1967 may be diagnosed with this illness based on their coronary angiography (CAG) findings at the onset of myocardial infarction [7-9]. In these patients, coronary artery morphology is often characterized by giant aneurysm(s) with stenosis at both the inlet and outlet, leading to massive thrombus formation [6]. Percutaneous coronary intervention (PCI) alone does not always achieve complete revascularization. Vascular prognosis, which is inversely represented as the rate of repeated percutaneous intervention, is better after coronary artery bypass grafting (CABG) than after PCI alone [10,11]. However, an appropriate method of CABG has not been established for KD, unlike for atherosclerotic lesions. Herein, we describe the case of an adult patient with KD for whom a combination of a novel CABG graft and coronary aneurysm ligation was applied.

Case Report

A 68-year-old woman presented to our emergency department with chest oppression and nausea that had persisted for 2 hours. She had no coronary risk factors but had a history of repeated myocardial infarctions. On arrival, her general condition and vital signs were stable (blood pressure: 120/80 mmHg; heart rate: 64/min; respiratory rate: 16 breaths/min; oxygen saturation: 98% in room air). Electrocardiography revealed ST-segment depression in leads V1-V5 and abnormal Q-waves in leads III and aVF. Transthoracic echocardiography identified severe hypokinesia of the posterior and inferior walls, with a left ventricular ejection fraction of 45%. Laboratory test results were normal, except for B-type natriuretic peptide levels of 28.4 pg/mL (reference value: <18 pg/mL) and troponin I levels of 33.2 pg/mL (<26 pg/mL).

The patient developed her first myocardial infarction in the right coronary artery (RCA), complicated by coronary artery ectasia, at the age of 54 years. Since then, she experienced myocardial infarctions of the RCA or the left circumflex artery (LCx) a total of 8 times and had undergone PCI each time. The morphology of coronary arteries had gradually worsened from ectasia to giant aneurysms, with progressive stenosis at both the inlet and outlet of the aneurysms (**Figure 1A, 1B**). She repeatedly developed myocardial infarctions, presumably because turbulence in the aneurysm formed thromboses, especially in the presence of dehydration.

Cardiologists, cardiac surgeons, and pediatricians discussed the possible etiology of her aneurysms. The CAG and contrast-enhanced CT did not identify an arterial fistula that could explain the aneurysms [12-14]. She did not have severe comorbidities that could have caused atherosclerotic coronary aneurysms [15,16]. In addition, congenital coronary aneurysm was unlikely because of the morphology of the aneurysms in our patient [17]. Her aneurysms were bead-like and located in the

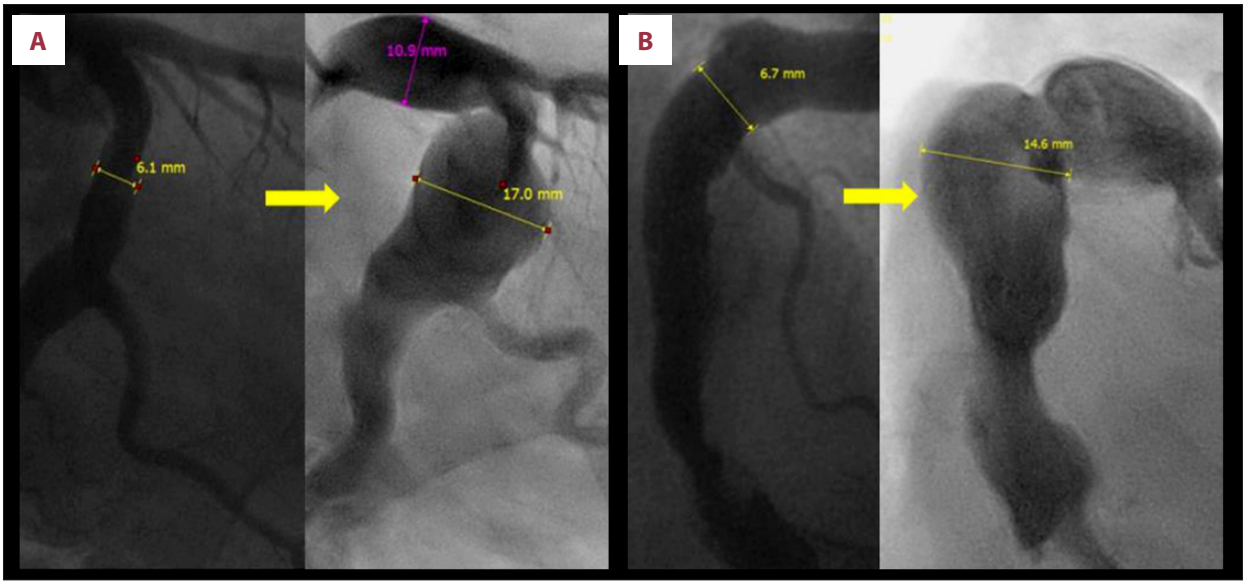

Figure 1. Coronary angiography images showing gradual changes over time of left coronary artery (**A**) and right coronary artery (**B**). The left side of each image shows the coronary artery ectasia at the age of 54 years. The right side presents the giant aneurysms more than 10 years later.

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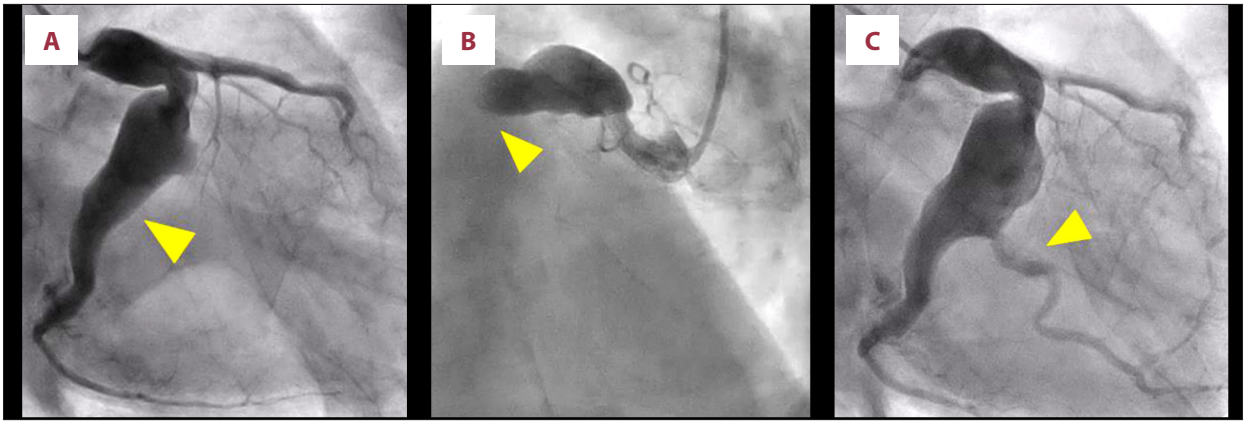
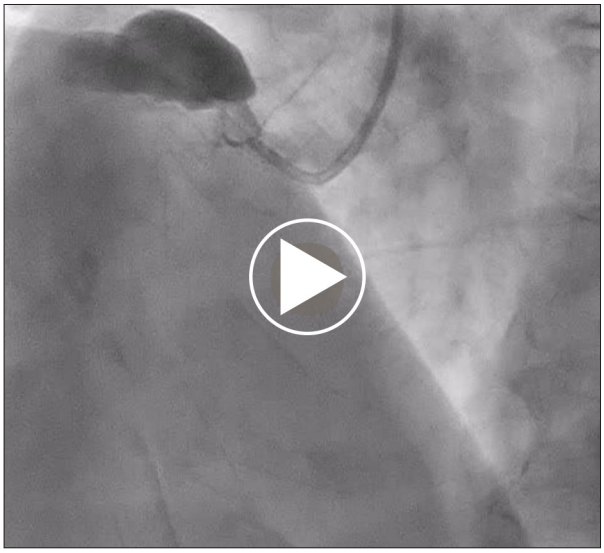


Figure 2. Emergency coronary angiography images show simultaneous total occlusions in the posterolateral branch of the left circumflex artery (**A**) and proximal right coronary artery (**B**) (triangular arrows). (**C**) A post-balloon angioplasty image shows thrombolysis in myocardial infarction grade II in the posterolateral branch of the left circumflex artery (triangular arrow).

Video 1. Total occlusion of the left circumflex artery revealed by the emergency coronary angiography.



Video 2. Total occlusion of the right coronary artery.

central part of coronary arteries, which is the typical morphology of KD aneurysms. She developed a month-long febrile episode with an unknown origin at the age of 2 years. Her son developed KD as well. C-reactive protein (0.2 mg/L) and autoantibodies–anti-nuclear, -Sm, -U1RNP, and Sc170 antibodies–were all negative. Collectively, therefore, the diagnosis of KD was entertained, although she was not diagnosed as such in her childhood. Emergency CAG revealed simultaneous total occlusion of the posterolateral branch of the LCx (#14PL) and the proximal portion of the RCA (**Figure 2A, 2B and Videos 1, 2**). The collateral blood channels extended from the left coronary artery to the distal portion of the RCA (**Video 3**). Therefore, we performed PCI for the #14PL lesion without collateral channels. An intra-aortic balloon pump (IABP) was inserted through the femoral artery before PCI to improve coronary flow. The contrast findings suggested thrombus formation in the #14PL, and monteplase was administered through a pulse-infusion-thrombolysis (PIT) catheter. Subsequently, the thrombus disappeared, and antegrade flow was confirmed in the lesion. A drug-eluting stent had previously been deployed at the origin of #14PL, and in-stent restenosis (ISR) was visible after the PIT procedure. Angioplasty was performed for ISR using a compliant balloon. Finally, thrombolysis in myocardial infarction grade II flow was confirmed in the #14PL (**Figure 2C, Video 4**). We also performed PCI for proximal RCA lesions. The collateral blood channels to the distal portion of the RCA suggested the possibility of chronic total occlusion of the RCA. The lesion had a hard consistency and guidewires could not be advanced beyond the occlusion. Coronary flow did not improve after the administration of monteplase via the PIT catheter from the right ventricular branch. However, following PCI of the #14PL, her chest oppression and electrocardiography

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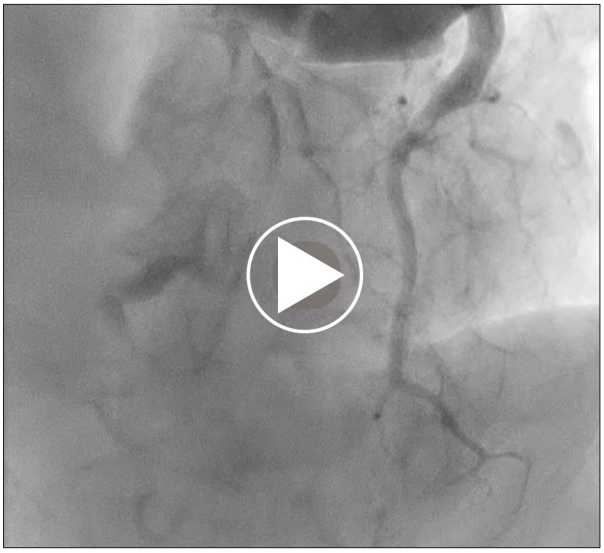
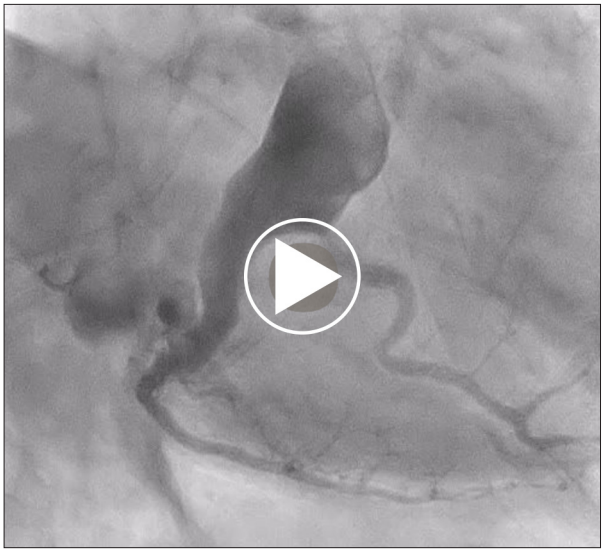


Video 3. Collateral blood channels extending from the left coronary artery to the distal portion of the right coronary artery.



Video 4. Thrombolysis in the myocardial infarction grade II in the posterolateral branch of the left circumflex artery after percutaneous coronary intervention.

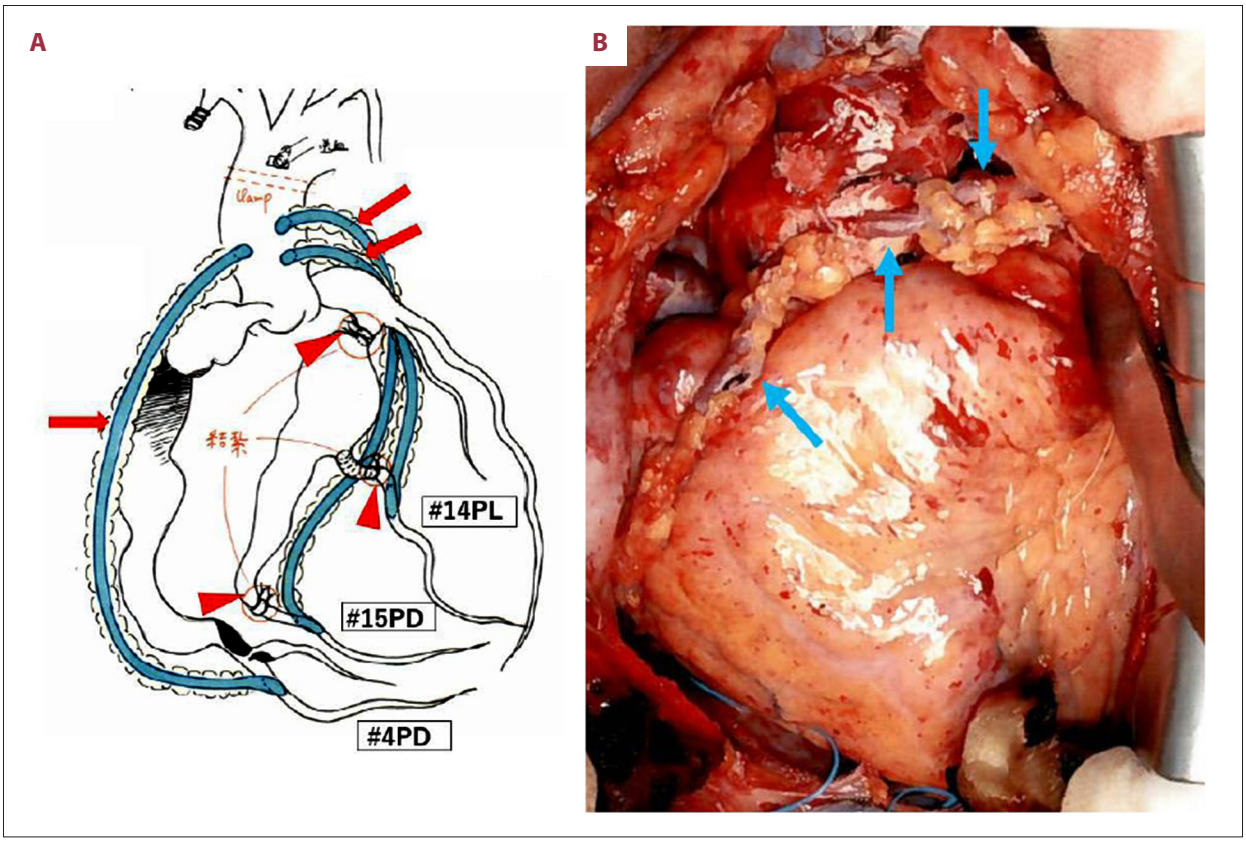


Figure 3. (**A**) An intraoperative illustration shows coronary artery bypass grafting using saphenous vein grafts (arrows) harvested using the "no-touch" technique in combination with left circumflex artery aneurysm ligation (triangular arrows). (**B**) An intraoperative image shows saphenous vein grafts harvested using the "no-touch" technique (arrows).

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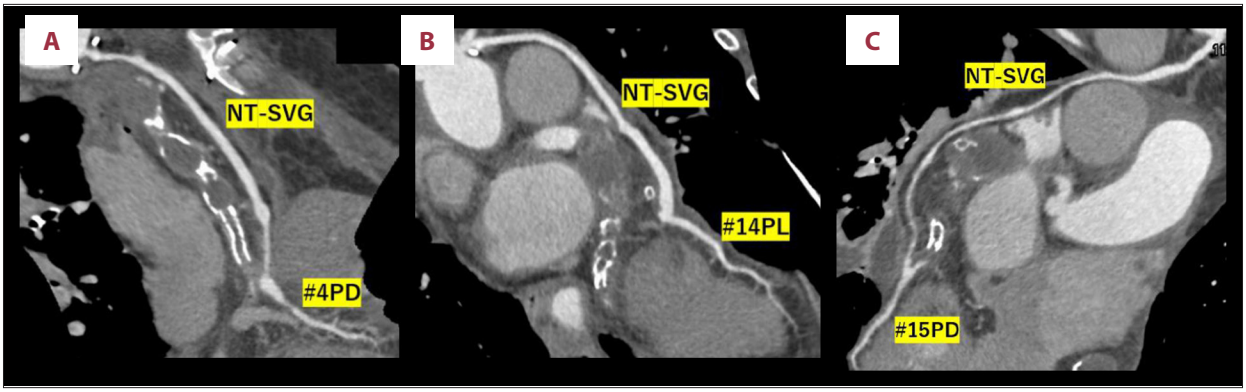

Figure 4. Coronary computed tomography angiography (CCTA) images of the bypass grafts on postoperative day 8. Curved multi-planar reconstruction of CCTA images show the good patency of saphenous vein grafts, harvested using the "no-touch" technique, in the right coronary artery (**A**) and the left circumflex artery (**B, C**). #4PD: posterior descending branch of the right coronary artery, #14PL: posterolateral branch of the left circumflex artery, #15PD: posterior descending branch of the left circumflex artery.

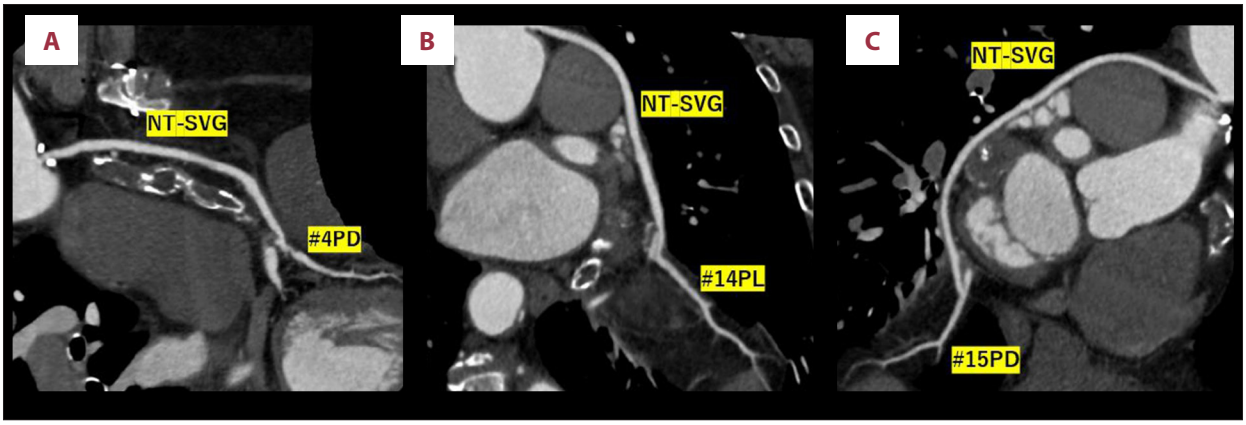

Figure 5. Coronary computed tomography angiography (CCTA) images of the "no-touch" saphenous vein grafts (NT-SVG) at 4 years postoperatively. All the NT-SVGs show good patency in the right coronary artery (**A**) and the left circumflex artery (**B, C**).

findings showed signs of improvement. Therefore, the procedure was completed.

The patient was stable, and the IABP was removed on postoperative day 3. Aspirin, warfarin, and prasugrel were continued. The patient was discharged from the ICU on postoperative day 6 without complications. However, the PCI had not completely revascularized the RCA, while the giant aneurysm in the LCx persisted. In addition, multiple antithrombotic drugs rendered the patient at risk of major bleeding. Therefore, we decided to perform CABG for the posterior descending branch of the RCA (#4PD), as well as for the LCx (#14PL and #15PD) prophylactically. In contrast, the aneurysm in the left main trunk (LMT) and the left anterior descending artery (LAD) never developed thrombosis. Therefore, we did not conduct CABG on these segments because the prophylactic benefit of bypass may not outweigh the risk of failure.

Transthoracic ultrasonography conducted prior to the surgery revealed severe stenosis and occlusion in both internal thoracic arteries (ITAs). Although we tried to harvest ITAs, these arteries were not pulsatile. Therefore, we used saphenous vein grafts (SVGs) instead. SVGs were harvested by the "no-touch" (NT) technique, which preserves the perivascular adipose tissue (PVAT). With this technique, PVAT continues to releases beneficial factors which improve long-term patency, while potential damage to vaso vasum and nerves is minimized [18,19]. In this patient, we harvested the graft by using skip incision to prevent wound complications. Ligation of the LCx aneurysm was also necessary because the competitive flow between the LCx and SVGs might lead to occlusion of the graft. After obtaining written informed consent from the patient, we performed the CABG and ligation of the aneurysm on day 18 (**Figure 3A, 3B**). Postoperative coronary computed tomography angiography (CCTA) showed good patency of all grafts, as well as occlusion of the LCx aneurysm (**Figure 4A-4C**). Prasugrel was discontinued, while aspirin and warfarin were continued.

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The patient was discharged on day 40 without complications. All grafts are patent on CCTA (**Figure 5A-5C**) and the patient is in good health at 4 years postoperatively.

Discussion

We present a rare case in which SVGs were used in CABG for a KD patient. In 97% of cases with CABG for KD, ITAs are used as grafts because of the excellent long-term patency of these arteries: the 20-year patency rates were 87% and 44% for ITAs and SVGs, respectively [17,20]. However, both ITAs were occluded in our patient. This was possibly due to vasculitis because atherosclerotic occlusion rarely occurs in ITAs [21,22]. KD is vasculitis that predominantly affects the intimal/medial layer [23], particularly of medium-sized arteries, including ITAs [24]. Therefore, obstruction of ITAs, as observed in our patient, may not be uncommon in KD patients. This underlines the importance of finding alternative graft vessels for KD.

We considered the radial arteries, the gastroepiploic arteries (GEAs), and SVGs as the possible vessels for grafts. However, the patient's radial arteries (<2 mm) were too small for use as grafts. No consensus exists regarding the long-term outcomes of GEAs and SVGs [14]; however, SVGs are widely utilized for CABG because of their ease of use.

To the best of our knowledge, this is the second report of using no-touch SVGs (NT-SVGs) for KD [25]. However, NT-SVGs have been increasingly used because of the excellent outcome of this technique: the 16-year patency rates of SVGs, NT-SVGs, and the left ITA were 64%, 83%, and 88%, respectively [26]. Conventionally, SVGs are harvested by removing the PVAT and are subsequently expanded with high-pressure saline. In contrast, the NT technique preserves PVAT and avoids the expansion. PVAT releases vasodilators such as nitric oxide and leptin, effectively mitigating vasospasm [27]. Furthermore, without excessive expansion, turbulent flow and low shear stress, which cause intimal hyperplasia, are avoided [28].

We ligated the outlet of the LCx aneurysm at the proximal portions of #14PL and #15PD: otherwise, the competitive flow between the LCx and SVGs might lead to occlusion of the grafts. In addition, we ligated the inlet of the LCx aneurysm to circumvent the possible formation of a thrombus which may extend from the aneurysm to LMT and LAD. Ligation also prevents potentially fatal rupture of the aneurysms [29]. We did not ligate the inlet of RCA because this would have blocked the perfusion to the right ventricular branch, and there was no competition between the graft and the completely occluded main trunk of RCA.

CABG can be complicated by graft hypoperfusion and resulting myocardial damage [30]. However, postoperative myocardial damage was not observed in our patient. The NT-SVGs in our patient remained at sufficient diameters and showed no signs of vasospasms, and the coronary arteries beyond the anastomoses had normal diameters. As a result, a diameter imbalance did not occur between the NT-SVGs and coronary arteries, indicating adequate coronary perfusion.

Conclusions

Surgical treatment for an aneurysm in a patient with KD is challenging in multifold ways. A giant aneurysm characteristic of KD has a potential risk of rupture and therefore should be ligated in some cases. In addition, ITAs, which are the standard graft vessels, may be occluded due to the vasculitis of KD. We ligated the aneurysm and used a new technique – NT-SVGs – for the graft preparation in our adult patient with KD. The outcome has been excellent to date, and the patient will receive long-term monitoring. A future clinical trial is warranted to validate the safety and efficacy of this new practice for KD patients.

Department and Institution Where Work Was Done

Department of Cardiology, Fukuoka Tokushukai Hospital, Fukuoka, Japan.

Ethics Statement

This case report was approved by the Ethics Committee of Fukuoka Tokushukai Hospital (reference number: G-2405). Written informed consent was obtained from the patient.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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