



## Case Report

# New coronary aneurysm formation and malapposition after zotarolimus-eluting stent implantation in Kawasaki disease

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## ABSTRACT

Coronary artery involvement is the most important complication of Kawasaki disease. Coronary artery bypass surgery has been performed for ischemic heart disease caused by Kawasaki disease, however, long-term coronary graft patency is not satisfactory. Therefore, percutaneous coronary intervention (PCI) has its role in Kawasaki disease-related coronary artery disease. The incidence of new aneurysm is lower following stent implantation than balloon dilatation alone, even if a higher balloon pressure is applied. However, there are few reports about the efficacy of drug-eluting stent implantation for Kawasaki disease with coronary artery disease. Here, we describe a case of new coronary aneurysm formation and malapposition after zotarolimus-eluting stent implantation in Kawasaki disease.

**<Learning objective:** New aneurysm formation after balloon angioplasty for coronary artery lesions in Kawasaki disease is a relatively well-known phenomenon, however there have been no reports about the influence of drug-eluting stents for coronary artery disease with Kawasaki disease. This report is useful when we consider strategies of revascularization for coronary artery disease with Kawasaki disease. >

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## Case report

A 40-year-old male was admitted to our hospital with continuous precordial pain. His coronary risk factors included smoking and dyslipidemia. Electrocardiography showed acute anteroseptal myocardial infarction, and urgent coronary angiography was performed. The culprit lesion was observed in the mid portion of the left anterior descending artery (LAD), which was treated by bare-metal stent (BMS) implantation. Coronary angiography at the time of the intervention and 320-detector computed tomography coronary angiography (CTCA) revealed chronic total occlusion (CTO) of the right coronary artery (RCA) and a coronary aneurysm with calcification considered to be associated with Kawasaki disease in the proximal portion of the RCA (Fig. 1a and b). Adenosine stress myocardial perfusion cardiac magnetic resonance imaging showed ischemia in the inferior wall. For this lesion, coronary angioplasty was performed. Two overlapping zotarolimus-eluting stents (ZES, proximal, 3.5 mm × 24 mm; distal, 2.5 mm × 24 mm) were implanted from the ostium to the mid portion of the RCA using

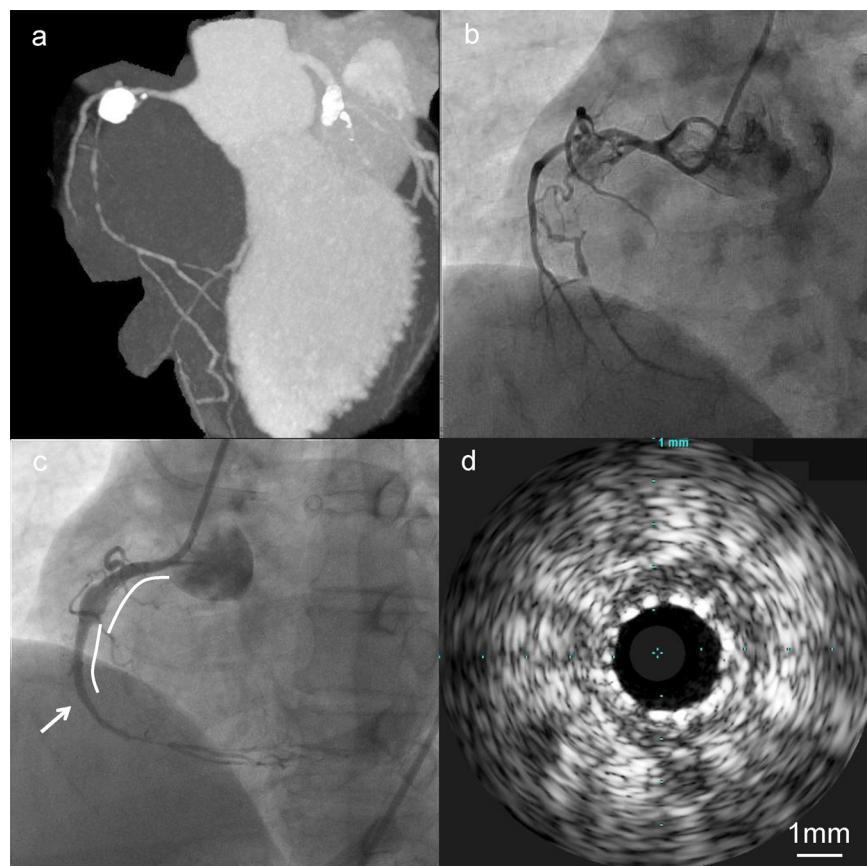
the reverse controlled antegrade and retrograde subintimal tracking technique (Fig. 1c). Intravascular ultrasonography (IVUS) after stenting showed good stent expansion and apposition (Fig. 1d). It was not necessary to use a rotational ablation. CTCA performed 6 months after coronary angioplasty revealed positive remodeling of the coronary artery around the distal ZES (Fig. 2a, b and d). Coronary angiography showed no restenosis in the BMS implanted in the LAD, but restenosis in the ZES implanted in the RCA and new coronary aneurysm formation around the distal ZES (Fig. 2c), which was consistent with CTCA findings. IVUS revealed good stent expansion with a little neointimal proliferation, coronary aneurysm formation, and late acquired stent malapposition (Fig. 2e). Optical coherence tomography (OCT) performed at the same time revealed similar findings (Fig. 2f). After coronary angiography, the restenosis site was treated by balloon dilatation. For the coronary aneurysm, to prevent stent thrombosis, oral administration of cilostazol was added to aspirin and clopidogrel that had been orally administered, and the course was carefully observed.

## Discussion

The patient had a past history highly suggestive of Kawasaki disease, such as persistent fever, rash, and cervical lymphadenopathy

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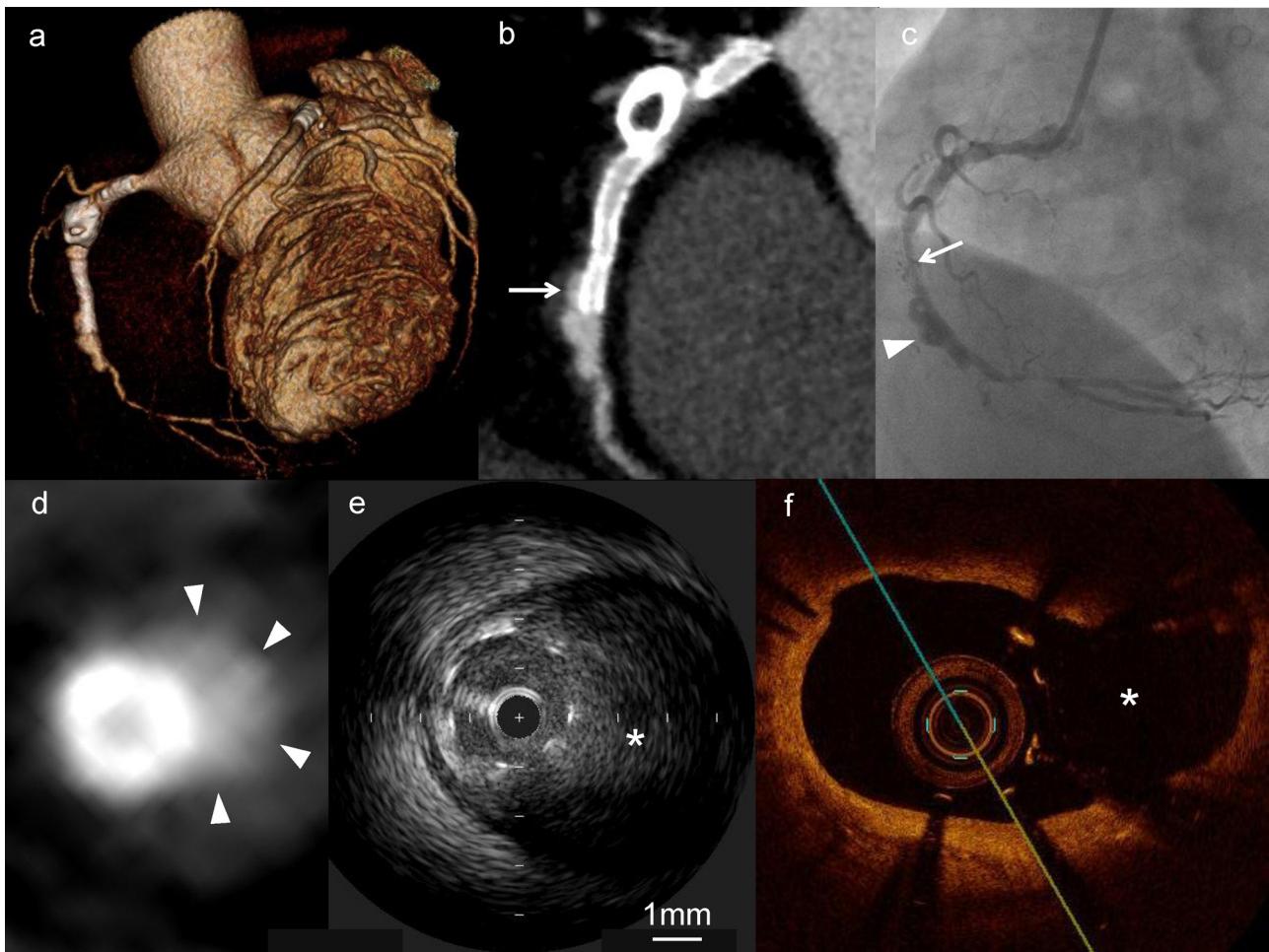


**Fig. 1.** (a and b) 320-Detector computed tomography coronary angiography and coronary angiography showed chronic total occlusion of the right coronary artery and a coronary aneurysm with calcification. (c) Coronary angiography after zotarolimus-eluting stents implantation (proximal, 3.5 mm × 24 mm; distal, 2.5 mm × 24 mm, white lines). (d) Intravascular ultrasound at site of arrow in (c) indicating stent distal edge showed good expansion and apposition of the stent.

in childhood. In addition to this past history, we predicted that he had suffered from Kawasaki disease because of coronary aneurysm with calcification. Indications of PCI for Kawasaki disease patients have not been established, and the long-term prognosis in Kawasaki disease patients after PCI remains unclear. Balloon angioplasty is only useful for stenotic lesions with mild calcification or the absence of calcification within 6 years of disease onset [1]. However, new aneurysm formation after balloon angioplasty for coronary artery lesions in Kawasaki disease is a relatively well-known phenomenon [2,3]. The development of new coronary aneurysms that can become a risk for thrombosis is a significant clinical concern. The mechanism responsible for this complication is unclear, however, intimal and/or medial dissection due to high pressure balloon inflation could play a role [3]. BMS implantation can reduce the incidence of restenosis and new aneurysm formation after coronary intervention compared with balloon angioplasty alone [4]. In this case, we recommended stent implantation to him, because he had only 1 vessel disease after urgent PCI to LAD lesion, and venous graft to the RCA cannot be expected to have long-term patency. We selected a drug-eluting stent (DES) that can reduce the incidence of restenosis after implantation for CTO lesions compared with BMS [5]. There are few reports about the efficacy of DES implantation for Kawasaki disease with coronary artery disease. A case of coronary aneurysm formation after sirolimus-eluting stent implantation as first generation DES in Kawasaki disease has been reported [6]. ZES used in this case have a biocompatible polymer imitating the red cell membrane, and have excellent long-term safety compared with first generation DES [7]. There have been no reports of new coronary aneurysm formation or malapposition after ZES implantation in Kawasaki disease. The

cause of this complication after stent implantation is still unclear. The mechanism of repair after vascular injury associated with interventional procedures may differ between Kawasaki disease and other atherosclerotic coronary disease, or hypersensitivity reactions to zotarolimus, its coating polymers, or metals may readily occur in Kawasaki disease [6]. In this case, two different vascular reactions occurred in the same stent: stent restenosis and coronary aneurysm formation. It is difficult to describe this mechanism, however, we think that coronary artery specificity of Kawasaki disease and inflammatory reaction after stent implantation and interventional procedures can cause both neointimal proliferation and coronary artery positive remodeling.

In this patient, new coronary aneurysm formation and malapposition after ZES implantation were diagnosed using four imaging techniques (CTCA, coronary angiography, IVUS, and OCT). New coronary aneurysm formation and late stent malapposition after stent implantation are a risk for stent thrombosis [8], and there is a report of stent thrombosis after DES implantation in Kawasaki disease [9]. Therefore, we should consider carefully the use of DES for coronary lesions in Kawasaki disease. Based on this case and previous reports [3,4], we recommend strategies for revascularization for coronary artery disease with Kawasaki disease as follows: (1) if patients have multivessel coronary artery disease or chronic total occlusion of LAD, coronary bypass surgery is recommended; (2) if not, BMS implantation is recommended; (3) for severe calcified coronary lesion, rotational ablation and post balloon dilatation by low pressure are recommended; (4) DES implantation may be one of the choices for restenotic lesions after initial PCI. Additional cases are necessary for the evaluation of revascularization for coronary lesions with Kawasaki disease.



**Fig. 2.** (a and b) Computed tomography coronary angiography 6 months after coronary angioplasty revealed positive remodeling of the coronary artery around the distal zotarolimus-eluting stents (ZES). (c) Follow-up coronary angiography revealed restenosis in the ZES implanted in the right coronary artery (arrow) and marked coronary aneurysm formation around the distal ZES (arrow head). (d) Multi-planar reconstruction cross sectional image at site of arrow in (b) showed the late acquired stent malapposition (arrow heads). (e and f) Intravascular ultrasound and optical coherence tomography at the site of the arrow head in (c) showed marked vessel remodeling and malapposition (\*).

## Conclusion

We report a case of new coronary aneurysm formation and malapposition after ZES implantation in Kawasaki disease. We consider that our report contributes to concepts and future directions of coronary revascularization for Kawasaki disease.

## Conflict of interest

Authors declare no conflict of interest.

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