CASE REPORT

Drug-Coated Balloon Treatment for Possible Sequelae of Kawasaki Disease Evaluated by Multi-Modalities

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Summary

The most devastating sequela of Kawasaki disease (KD) is coronary artery complications that may lead to myocardial infarction and cardiac mortality. Percutaneous coronary intervention (PCI) and bypass grafting are recommended for KD patients with inducible myocardial ischemia and amendable coronary anatomy. However, there are few reports about coronary revascularization with drug-eluting balloons among KD patients, especially at an early age. We present a case report of multi-modality guidance of PCI with a drug-coated balloon (DCB) for a young patient with acute coronary syndrome and a history of KD. Post-procedural optical coherence tomography, angiography-derived fractional flow reserve, and 12-month coronary artery magnetic resonance showed favorable outcomes. The present case indicated that DCB therapy with intravascular imaging and physiologic assessment guidance may be an alternative strategy to treat severe coronary artery stenosis in selected patients with KD.

Key words: Drug-eluting balloon, Optical coherence tomography

Kawasaki disease (KD) has been recognized as the most prevalent cause of acquired coronary artery disease that could result in fatal thrombotic events among infants and young children.1-3 Percutaneous coronary intervention (PCI) could be considered to recanalize severely stenotic coronary arteries among KD patients with appropriate coronary anatomy.4 During PCI for KD patients, coronary stent implantation after lesion preparation is now recommended as a standard strategy, although the long-term efficacy and safety remain unclear.4-6 To date, drug-coated balloon (DCB) has emerged as an alternative option to treat de novo coronary artery lesions.7 However, reports of DCB use for KD patients are still limited, especially for younger patients.8,9 Here, we present the case of an 18-year-old female undergoing successful PCI for possible sequelae of KD with multi-modality-guided DCB treatment.

Case Report

An 18-year-old girl was hospitalized with progressive exertional chest pain for 1 month. She was previously diagnosed with Kawasaki disease (KD) when she was 7 years old and was treated with intravenous immunoglobulin and acetylsalicylic acid. The patient was asymptomatic and received an annual echocardiogram follow-up. The echocardiogram showed that the lumen diameter of the proximal right coronary artery (RCA) was 5.0 mm 1 month after KD diagnosis and was persistently less than 5.0 mm during follow-up. Four years before admission, the patient received coronary computed tomographic angiography (CTA), indicating mild-to-moderate stenosis with mild dilation at the proximal RCA. Repeated CTA was performed just before admission, which revealed significant progression of the RCA lesion. An elevated cardiac troponin I level (0.036 ng/mL) and normal inflammatory parameters (C-reactive protein level and erythrocyte sedimentation rate) were detected in a blood test. Echocardiography revealed normal cardiac dimension (left ventricular end-diastolic diameter was 41 mm) and ventricular wall motion (left ventricular ejection fraction was 65%). Invasive coronary angiography showed patent left coronary artery (Figure 1A) and subtotal occlusion at the proximal RCA (Figure 1B) with mild coronary ectasia and suspected thrombus. After thrombectomy (Export Aspiration Catheter, Medtronic, USA), optical coherence tomography (OCT; Dragonfly®, Abbott, USA) revealed a complex coronary lesion with a heavy plaque burden (minimal lumen area 1.1 mm²), layered intima, focal calcification, macrophage accumulation, and microvessel (Figure 1C-H). According to the online OCT quantitative measurement, additional plaque modification was performed with a scoring balloon (NSE 3.0 × 13 mm@14 atm, Goodman, Japan) and a non-compliance balloon (NC Trek 3.5 × 15 mm@16atm, Abbott, USA). Mild residual stenosis (approximately 10%) was found in the following coronary angiogram (Figure 2A, B). Repeated OCT pull-back showed significant lumen enlargement (minimal lumen area 1.1 mm²) from the previous measurement.
Baseline coronary angiography. A: Normal left anterior descending and circumflex coronary artery. B: Subtotal occluded lesion at the proximal right coronary artery with mild coronary dilation and suspected thrombus. C–H: Optical coherence tomography (OCT) after thrombectomy. The distal and proximal reference lumen diameters were 4.0 mm (C) and 3.5 mm (H), respectively; OCT indicated layered intima (D), macrophage accumulation with linear signal attenuation (E), focal calcification (F), and microvessel within plaque (G).

Discussion

KD is considered an acute, self-limited vasculitis that affects medium-sized, extra-parenchymal muscular arteries, mainly the coronary artery. Among KD patients, the major pathological changes in the coronary artery include the formation of a coronary artery aneurysm and subsequent arterial remodeling. A recent autopsy study described the histologic manifestations of the vasculopathy of KD, including necrotizing arteritis, subacute/chronic vasculitis, and luminal myofibroblastic proliferation. Calcification and layered thrombus may occur in advanced lesions with aneurysms. In the present case, although there was no sign of significant coronary aneurysm by transthoracic echocardiography and systemic inflammation by laboratory testing, the typical manifestations of KD were detected by OCT imaging, including macrophage accumulation and layered, proliferated intimal and focal cal-
Figure 2. A–J: Percutaneous coronary intervention procedure and corresponding optical coherence tomography (OCT) imaging. A: Predilation with a non-compliance balloon (3.5 × 12 mm). B: Coronary angiogram after lesion pretreatment showed mild residual stenosis without significant dissection. C–E: Intimal dissection with an enlarged lumen. F: Drug-coated balloon dilation. G: Final CAG showed no significant stenosis with TIMI flow grade 3. H–J: The final OCT image revealed adequate lumen area (minimal lumen area was 8.6 mm²) and intimal dissection without hematoma. K: Post-procedural angiogram-based fractional flow reserve (CaFFR): the CaFFR value was 0.93 after drug-coated balloon dilatation. L: The 12-month follow-up CMRA showed patent right coronary artery without significant stenosis and ectasia.
reported that both intravascular ultrasound and OCT could identify appropriate dissection that facilitates the uptake of anti-proliferation drugs to enter into the vessel wall without acute complications, which could lead to favorable long-term prognosis. In the present case, OCT-guided serial pretreatment and intimal dissection without intramural hematoma may partly explain the excellent outcome at the 1-year follow-up. However, whether it could increase the risk of aneurysms should be monitored in long-term follow-up.

Recently, several observational studies suggested that FFR-guided DCB treatment could be safe and effective for de novo coronary lesions with good anatomic and physiological patency. Meanwhile, increasing data indicated that angiogram-derived FFR could provide accurate measurement consistent with FFR in selected patients. Notably, a recent study indicated that angiography-derived FFR can assist in predicting the angiographic changes and restenosis post DCB therapy; a quantitative flow reserve value less than 0.89 could predict restenosis in the future. In the present case, the optimal post-procedural angiography-derived FFR value could be associated with a beneficial result at the 12-month follow-up. In the future, further investigations are necessary to elucidate whether the combination of intravascular imaging and physiology modality could potentially improve the long-term outcomes for KD patients.

**Conclusions**

The present case suggested that stent-less PCI by DCB dilation under intravascular imaging and physiological guidance could be an alternative revascularization therapy for selected KD patients.

**Disclosures**

**Conflicts of interest:** None.

**References**