



## CASE REPORT

# Plain old balloon angioplasty for severe coronary artery stenosis caused by Kawasaki disease in a young child with a new aneurysm in the remote stage: a case report

Makoto Watanabe<sup>1\*</sup>, Ryuji Fukazawa<sup>1</sup>, Ryosuke Matsui<sup>1</sup>, Kanae Shimada<sup>1</sup>, Yoshiaki Hashimoto<sup>1</sup>, Koji Hashimoto<sup>1</sup>, Masanori Abe<sup>1</sup>, Mitsuhiro Kamisago<sup>1</sup>

1113-8603, 1-1-5 Sendagi, Bunkyo-ku, Tokyo, Department of Pediatrics, Nippon Medical School Hospital



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## ABSTRACT

Our patient was a 12-year-old girl who was diagnosed with Kawasaki disease at 6 months of age, which was refractory to treatment. One month after its onset, bilateral coronary artery aneurysms were confirmed via echocardiography. A stenotic lesion was then observed in the proximal portion of the aneurysm, located in the left anterior descending branch. At 2 years of age, the patient underwent plain old balloon angioplasty to treat the 99% stenosis in the left anterior descending branch caused by coronary artery lesions resulting from Kawasaki disease. This improved her stenosis to 25%. Thereafter, she was treated with aspirin and candesartan and experienced no notable symptoms or cardiovascular events. However, cardiac catheterization at 12 years of age revealed a new giant aneurysm at the plain old balloon angioplasty site. Plain old balloon angioplasty represents an effective treatment method for treating myocardial ischemia in coronary artery lesions after Kawasaki disease and can be safely performed in children. However, identification of new aneurysms after plain old balloon angioplasty is an important concern; hence, low-pressure dilation is recommended in this patient group. Despite the low-pressure dilatation performed in this case, a new aneurysm was found almost 10 years after the plain old balloon angioplasty was performed. Most new aneurysms form within 1 year after plain old balloon angioplasty; however, because new aneurysms can form even after 10 years, careful long-term follow-up is warranted.

## Introduction

Kawasaki disease often occurs in infants around the age of one year, and myocardial ischemia caused by complications such as coronary artery stenosis represents an important concern that can significantly threaten a child's overall prognosis. In general, coronary artery bypass grafting (CABG) and percutaneous coronary intervention (PCI) are the main treatment options for stenotic lesions after Kawasaki disease, but there are no clear indication criteria for their selection. Especially when infants present with myocardial ischemia, treatment options are often limited by considerations related to size and safety. Muta et al. compared CABG and PCI for stenotic lesions after Kawasaki disease and stated that CABG is superior in terms of treatment reintervention in patients younger than 12 years with myocardial ischemia<sup>1</sup>. In 1991, PCI for coronary artery lesions (CALs) after Kawasaki disease was first reported<sup>2</sup>. In general, PCI is performed on stenotic lesions in the remote stage of CALs after Kawasaki disease, but Kawasaki disease is characterized by strong calcified lesions in the remote stage, and repeated PCI was relatively often required<sup>3,4</sup>. Yokoi noted that 8 of 33 patients who underwent Plain old balloon angioplasty (POBA) after Percutaneous transluminal coronary rotational atherectomy (PTCRA) for distant calcified lesions underwent PCI again due to restenosis, but there was no subsequent restenosis and the effect was safe and maintained for a long period of time<sup>5</sup>. However, there is report that PTCRA is difficult for children under 5 years of age due to their physique<sup>6</sup>.

On the other hand, POBA can be safely performed in children, and is considered an effective treatment for myocardial ischemia because calcification at CAL sites is considered minimal within several years following the onset of Kawasaki disease<sup>7</sup>. However, the appearance of new aneurysms within one year following POBA to treat CALs associated with Kawasaki disease has been reported<sup>8-10</sup>, and should be carefully monitored as a potentially serious clinical concern.

In this report, we describe a case in which a young child underwent POBA to treat severe coronary artery stenosis caused by Kawasaki disease. Although her myocardial ischemia improved and no additional treatment was required, a new aneurysm was found 10 years after the procedure was performed.

## Clinical History

Our patient was a girl aged 12 years and 0 months. She developed Kawasaki disease at 6 months of age and was treated with IVIG (2 g/kg) and Aspirin (30 mg/kg/day), but subsequently relapsed twice. One month after the initial onset of the disease, bilateral coronary artery aneurysms (#1, 5.5 mm; #6, 5.8 mm) were identified via echocardiography. At the age of 1 year and 7 months (i.e., 13 months following the initial disease onset), cardiac catheterization revealed a small aneurysm in the right coronary artery (RCA) and a moderate aneurysm and stenotic lesion proximal to it in the left anterior descending branch (LAD). As a result, the patient was referred to our hospital for further treatment, at the age of 1 year and 9 months.

## Plain Old Balloon Angioplasty

Cardiac catheterization performed in our department when the patient was 2 years and 0 months of age (i.e., 1 year and 6 months following disease onset) revealed an aneurysm in the LAD that had regressed to 3 mm, with 99% stenosis proximal to it and numerous collateral vessels between the RCA and anterior descending branch region (Figure 1). Based on the patient's severe stenosis and the presence of collateral vessels, a diagnosis of myocardial ischemia in the peripheral region of the LAD was made, and POBA was judged to be indicated.

Plain old balloon angioplasty for severe coronary artery stenosis caused by Kawasaki disease in a young child with a new aneurysm in the remote stage: a case report

Figure 1. CAG before POBA

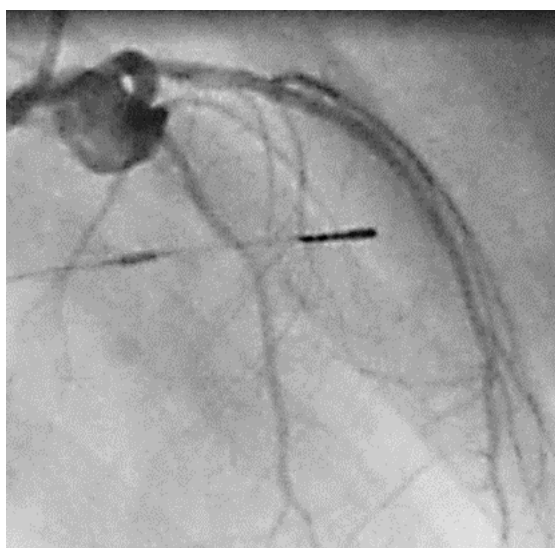


A small 3-mm aneurysm with a proximal 99% stenosis was noted at LAD segment 6. CAG: coronary angiography; POBA: plain old balloon angioplasty; LAD: left anterior descending branch.

Nitroglycerin (0.2  $\mu$ ) was administered the day before cardiac catheterization to prevent worsening of the patient's coronary artery stenosis during the POBA procedure. A 6 Fr sheath was placed in the right femoral artery and a 5 Fr JL 1.5 cm guiding catheter was used. Intraoperatively, a pacing wire was placed in the right ventricle and the balloon size was selected based on the coronary artery diameter before and after the aneurysm. IKAZUCHI Rev (Kaneka Medix Corporation, Osaka, Japan) (1.5 mm) was used as the balloon catheter, and two dilatations of the stenosis (6 atm for 30 s) were performed. The size

was later changed to IKAZUCHI Rev (2.0 mm), and two more dilatations (6 atm for 30 s) were performed. Postoperative results showed that the stenosis improved by 25% after dilatation (Figure 2). Both procedures were completed without any complications. After the examination, nitroglycerin 0.2  $\mu$  was administered until the following morning, then terminated after it was confirmed that there were no issues.

Figure 2. CAG immediately following POBA



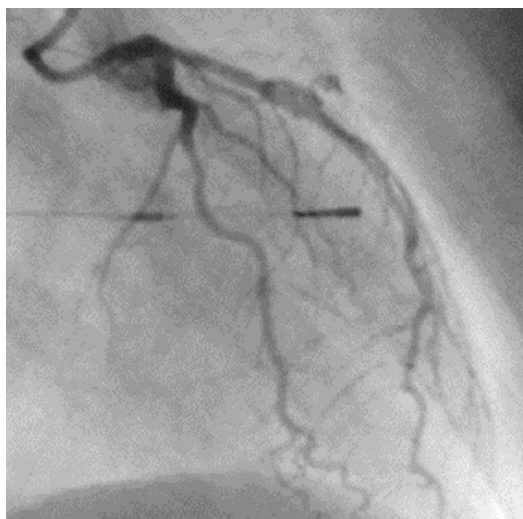
The stenotic lesions improved to 25%. CAG: coronary angiography; POBA: plain old balloon angioplasty.

## Progress After Plain Old Balloon Angioplasty

The patient was prescribed only aspirin and candesartan after the POBA procedure, and no specific symptoms or cardiovascular events were observed.

Cardiac catheterization 6 months after POBA showed no progression of stenosis, with 25% stenosis and disappearance of the collateral vessels (Figure 3). Cardiac catheterization at 5 years and 8 months (i.e., 3 years and 8 months after POBA) showed that the stenosis remained unchanged, at 25%, and that the coronary fractional flow reserve (FFR) of the LAD was 0.87. Therefore, the stenosis was deemed insignificant.

Figure 3. CAG at 6 months following POBA



The stenotic lesion remained at 25%. CAG: coronary angiography; POBA: plain old balloon angioplasty.

Figure 4. CAG at 10 years following POBA



The initial stenosis lesion was at 50% stenosis, but a new aneurysm (measuring 8.5 × 11.6 mm) was identified distal to the original stenosis. CAG: coronary angiography; POBA: plain old balloon angioplasty.

Cardiac catheterization at 12 years and 0 months of age (i.e., 10 years 0 months after POBA) revealed 50% stenosis, but a new aneurysm (measuring 8.5 × 11.6 mm) was identified distal to the stenosis (Figure 4). The FFR at the stenosis was 0.90. An adenosine stress <sup>13</sup>N-ammonia positron emission tomography scan showed that the patient's myocardial flow reserve (MFR) was < 2.0 in the apical anterior and apex regions, but myocardial blood flow quantification (MBF) remained relatively high during adenosine stress (Table 1). Based on these findings, we determined that there was no significant myocardial ischemia. However, as the new aneurysm was a giant one, warfarin was started and the patient's follow-up was increased.

Table 1. MBF and MFR values on a <sup>13</sup>N-Ammonia PET scan

	MBF(stress) ml/min/kg	MBF(rest) ml/min/kg	MFR
LAD	2.3067	1.01	2.284
basal anterior	2.3862	0.9084	2.627
basal anteroseptal	2.4516	0.9426	2.601
mild anterior	2.285	0.9892	2.31
mild anteroseptal	2.3491	1.0382	2.263
apical anterior	1.8769	0.9571	1.96
apical septal	2.5535	1.0629	2.402
apex	2.1036	1.2812	1.642
RCA	2.8275	0.8761	3.227
basal inferoseptal	2.2352	0.7442	3.003
basal inferior	2.3661	0.7544	3.136
mild inferoseptal	3.2864	0.9989	3.0029
mild inferior	3.6685	1.0133	3.62
apical inferior	2.8729	0.9784	2.936
LCX	2.6877	1.0177	2.641
basal inferolateral	2.5117	0.8994	2.793
basal anterolateral	2.5068	1.0101	2.482
mild inferolateral	3.1683	0.9991	3.171
mild anterolateral	2.6381	1.1425	2.309
apical lateral	2.7195	1.11	2.45

MBF: myocardial blood flow quantification; MFR: myocardial flow reserve; LAD: left anterior descending branch; RCA: right coronary artery branch; LCX: left circumflex artery branch.

## Discussion

Findings suggestive of intimal thickening in the remote stage of coronary artery aneurysms resulting from Kawasaki disease are often found in areas of coronary artery enlargement measuring > 4 mm after the acute stage<sup>11</sup>. However, during intimal thickening caused by vascular remodeling, excessive vascular remodeling can occur—particularly at the entrance and exit points of aneurysms—resulting in stenotic lesions. Such lesions can cause myocardial ischemia, which significantly affects quality of life and general prognoses in pediatric patients.

In local stenotic lesions of single CAL branches resulting from Kawasaki disease, POBA using a cardiac catheter can be safely performed even in young children, and can significantly improve myocardial ischemia. Ino et al. reported POBA as a catheter-based treatment for stenotic lesions in childhood in five cases, and stated that the cases in which POBA was most effective were those who were ≤ 8 years of age within 6 years of the onset of Kawasaki disease, and this was related to the strength of calcification at the site of the lesion<sup>12</sup>.

By contrast, Mitani et al. reported that subluminal or shallow calcification is generally observed at the site of focal stenosis after 6 years<sup>7</sup>. In other words, it was assumed that calcified lesions would gradually appear over time, likely ~6 years after disease onset, when calcification intensifies and the success rate of POBA is lower. In addition, re-stenosis and new aneurysm formation after POBA for CALs resulting from Kawasaki disease have long been noted as significant challenges as well<sup>6,13</sup>. The incidence of re-stenosis after POBA is high, with ~25% of cases resulting in re-stenosis or occlusion.<sup>5</sup> This is thought to be caused by excessive vascular remodeling that causes re-stenosis even if the vessels are sufficiently dilated. The development of new aneurysms also represents an important concern because, in cases of CALs resulting from Kawasaki disease, OCT shows that the internal and external elastic plates and the three-layer structures of the vessels are destroyed by the inflammation of the coronary arteries that typically occurs during the acute phase of Kawasaki disease<sup>14</sup>. Therefore, it can be hypothesized that the high-pressure balloon dilation used in POBA may transfer the dilatation pressure to the outer



membrane side of the coronary artery, thus forming a new coronary aneurysm. Therefore, low-pressure dilatation is recommended for POBA to treat CALs resulting from Kawasaki disease<sup>15</sup>. In our case, we also used a low balloon dilation pressure of 6 atm. We initially observed no obvious morphological changes, except for progression of the patient's mild stenosis; however, 10 years after the POBA, a new aneurysm appeared at the site. Since most reports of re-stenosis and new aneurysms after PCI are generally reported within 1 year<sup>8-10</sup>, the formation of a giant aneurysm 10 years after POBA in our case was considered extremely rare. The reason behind this was thought to be because of the improvement in reflux pressure following the release of the stenosis, in addition to the further destruction of the vascular structure induced by the POBA. Although chronic inflammation has been reported to persist in CALs that result from Kawasaki disease, even in the remote stage<sup>16,17</sup>, the destruction of the three-layered coronary artery wall structure and vascular reconstruction with chronic inflammation can lead to the formation of aneurysmal lesions. At this point in follow-up, it is typically unnecessary to monitor patients for significant ischemia, but careful follow-up is still warranted. If ischemia progresses as a result of further stenosis or dilation of the aneurysm, coronary artery bypass surgery may be indicated.

## Conclusion

In this case, POBA improved the patient's myocardial ischemia in the first few years, and the patient progressed favorably without the need for additional treatment. However, despite the low-pressure dilation procedure used, a new aneurysm was found in the remote stage. POBA is an effective method for ameliorating myocardial ischemia in younger pediatric patients; however, long-term observation is important.

## Conflict of Interest:

The authors have no conflicts of interest to declare.

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## Patient Permission/Consent statement

Permission for publication of this report was obtained from the patient's' guardians.

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