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Percutaneous Transluminal Coronary Angioplasty in a Child with Kawasaki Disease

A successful attempt at percutaneous transluminanl coronary angioplasty (PTCA) to relieve stenosis of the mid-portion of the left anterior descending artery was achieved in a 6-year 9-month old boy who had multiple coronary aneurysms and stenosis due to Kawasaki disease. Despite the progression of coronary stenosis he had been well except for the perfusion defect of the anterior wall of myocardium on ^{99m}Tc-MIBI SPECT with dipyridamole infusion until PTCA was carried out after 4-year 4-months of the onset of illness. The area of stenosis was 70% before PTCA and 20% after PTCA. No restenosis at the site of PTCA was observed on follow-up angiography at 26 months after PTCA. This successful attempt may indicate that this procedure should be considered early in subclinical stenosis to prevent ischemic cardiac damage.

Key Words: *Mucocutaneous lymph node syndrome; Angioplasty, transluminal, percutaneous coronary*

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INTRODUCTION

Kawasaki disease (KD) is an acute vasculitis syndrome with high fever, mucosal inflammation, skin rash, and cervical lymphadenopathy recognized exclusively in children younger than 4 years of age (1). This disease is usually self-limited, but may involve coronary arteries in 15 % to 25% of the patients (2, 3). Coronary arteritis associated with coronary aneurysm and thrombotic occlusions may lead to ischemic heart disease or sudden death (3, 4). It remains a leading cause of acquired heart disease in children. Percutaneous transluminal coronary angioplasty (PTCA) has been rarely performed in KD as a treatment for severe stenosis of coronaries, because of limited applications and unconfirmed effectiveness of this procedure. In Korea, as far as we know, PTCA has not yet been performed in KD.

We report a case of successful PTCA to relieve stenosis of the coronary arteries in a patient with KD.

CASE REPORT

A 6-year 9-month-old boy was admitted to Seoul National University Children's Hospital for elective PTCA to relieve stenosis of the coronary arteries. At 2-year 5-months of age, he presented persistent fever up to 39 ℃ for 10 days accompanied with macular skin rash, conjunctival injection, cervical lymphadenopathy, strawberry tongue and red fissured lip and was admitted to a local clinic. Under the diagnosis of KD, intravenous gamma globulin (2 g/kg over 10 hours) was infused and aspirin (50 mg/kg/day) was prescribed. On the 21st day of illness, aneurysms of both coronary arteries and pericardial effusion were detected on initial two-dimensional echocardiography at the local clinic. He was referred to our hospital and was followed up with the prescription of aspirin (5 mg/kg/day) and dipyridamole (3 mg/kg/day). On the 35th day of KD, coronary angiography revealed diffuse dilatation of long segment in left main, left circumflex, left anterior descending arteries and right coronary artery (maximum diameter about 6 mm) (Fig. 1). There were no abnormal findings on electrocardiography. Repeated cardiac angiography performed 13 months later revealed multiple saccular aneurysms and stenoses [5.76 mm in the left main coronary artery, 3.04 mm in the proximal portion of the left anterior descending (LAD) artery, 2.43 mm in the mid-portion of the LAD, 5.76 mm and 2.4 mm in the proximal portion of the left circumflex, 2.8 mm in the proximal portion of the right main coronary artery, and 0.8 mm in the mid-portion of the right main coronary artery]. He did not show any

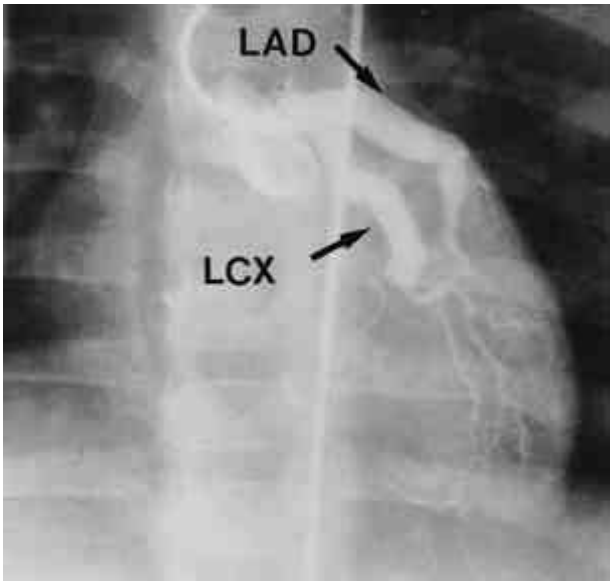

Fig. 1. On the 35th day of KD, coronary angiography reveals diffuse dilatation of long segment in left circumflex (LCX), left anterior descending (LAD) arteries (maximum diameter about 6 mm).

cardiovascular symptoms. Electrocardiogram did not demonstrate any abnormal findings. Twenty six months from the onset, coronary angiography demonstrated the progression of multiple stenosis of the coronary arteries (1.8 mm in the proximal portion of the LAD, 0.84 mm in the mid-portion of the LAD, 5.0 mm and 1.67 mm in the proximal portion of the left circumflex, and 0.84 mm in the mid-portion of the right main coronary artery). ^{99m}Tc-MIBI SPECT with dipyridamole infusion was carried out to evaluate the myocardial perfusion. It showed perfusion defect on the anterior wall of myocardium. Exercise stress test with Bruce protocol, chest roentgenogram, and ECG revealed no abnormal findings. Although he had been well without ischemic symptoms, PTCA was considered to improve myocardial perfusion in order to prevent ischemic event.

On admission, his blood pressure was 100/60 mmHg. His heart rhythm was regular and heart rate was 100/min. His weight was 22.3 kg (25~50 percentile) and his height was 121 cm (50~75 percentile). He looked well and showed normal physical findings on examination. Laboratory studies showed no abnormal findings. Although stenosis of the mid-portion of the right coronary artery (RCA) was more severe than that of the LAD, collateral arteries already had been sufficient to supply blood to the distal portion of the RCA. Accordingly PTCA was carried out uneventfully using 2.5 mm balloon catheter in the stenotic lesion of the mid-portion of the LAD at the 4-year 4-month from the onset of illness. The area of stenosis was 70% before PTCA and 20% after PTCA (Fig. 2). Slight improvement of myocardial perfusion was observed in follow-up ^{99m}Tc-MIBI SPECT with dipyridamole infusion. No restenosis at the site of PTCA was observed recently on follow-up angiography at 26 months after PTCA. He has remained well without any cardiac symptoms so far. He is now taking a follow course of aspirin.

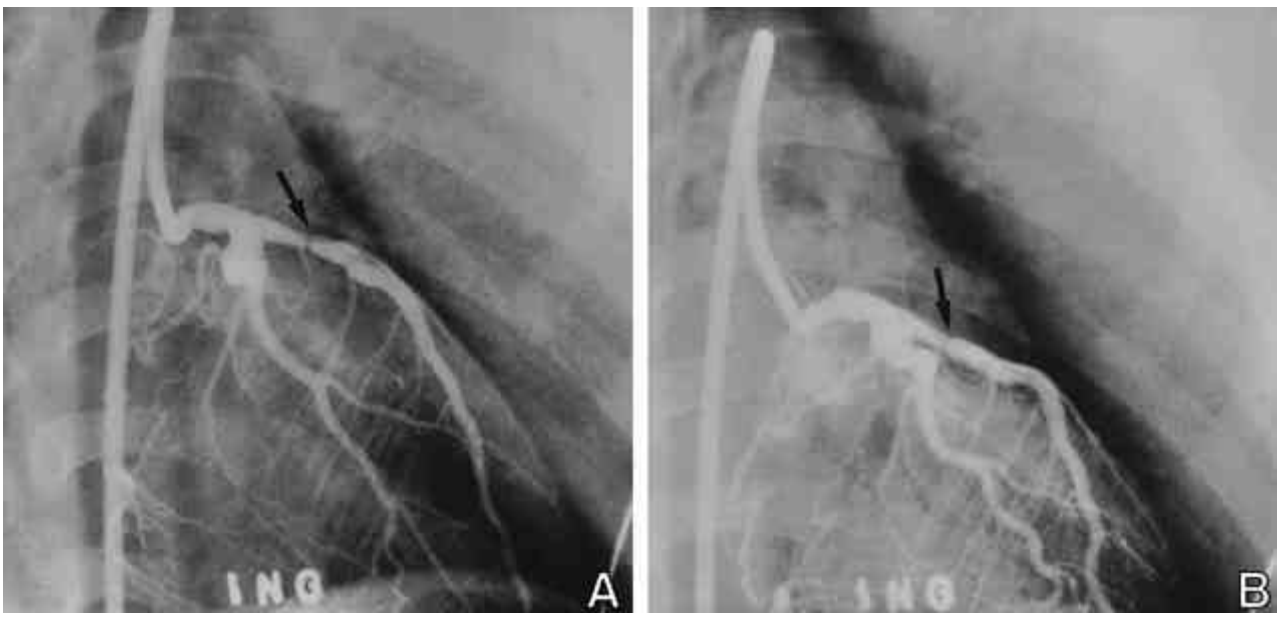

Fig. 2. Coronary angiogram before (A) and after (B) PTCA demonstrates the change of stenosis. The area of stenosis in the midportion of left circumflex artery was 70% (arrow) before and 20% (arrow) after PTCA.

DISCUSSION

The long-term clinical issues in KD are associated with coronary artery lesions that lead to aneurysmal formation, thrombotic occlusion, progression to ischemic heart disease, sudden death, and premature atherosclerosis (2-4). Since the first description by Kawasaki in 1967, the mortality rate has been decreased to 0.8% with the development of the advanced diagnostic methods and treatment (3).

However, coronary artery aneurysm, a serious complication of KD, has remained a leading cause of acquired heart disease in children (5). Kato and his colleagues (2) have reported the result of serial coronary angiography to clarify the fate of the coronary aneurysms. The incidence of coronary aneurysm in acute Kawasaki disease was 15%, 1) 50% of which showed regression and 2) the remainder of which showed persistent coronary aneurysms although reduced in size, 3) complete obstruction or marked stenosis of coronary arteries, or 4) irregularities of coronary arterial wall without stenosis. The regression in coronary aneurysms was recognized within one or two years from onset, but half of the patients with regressed coronary aneurysms continued to demonstrate a dense or bright echo in the coronary artery wall on 2D-echocardiography (3). Ino et al. (6) has observed that no patients revealed clinical symptoms or positive perfusion defect on the thallium scintigraphy despite angiographic findings of significant stenosis. In this case, the patient has remained asymptomatic and electorcardiography and chest roentgenography revealed no abnormal findings despite the progressing coronary stenosis on the scheduled follow-up angiography.

Aortocoronary bypass surgery has been performed as the standard therapy in patients with severe stenosis of coronary arteries due to KD (6). However the long-term patency of vein grafts is still unsatisfactory in small children because of the small caliber of the coronary artery and the limited potential for growth of the graft. Therefore, PTCA should be considered in small children to postpone bypass surgery until patients are of sufficient age and size, so that the bypass grafts are likely to maintain satisfactory long-term patency. During the past decades balloon valvuloplasty and angioplasty have become an accepted treatment for various congenital obstructive heart disease. Although the mechanism of balloon dilatation of stenotic vessels has remained unclear, according to a recent report about the mechanism of balloon angioplasty assessed by intravascular ultrasound (IVUS), three distinctive morphologic types were identified: 1) vessel stretching, 2) superficial or deep intima-media tearing, which was the most common type and 3) arterial wall dissection (7). Although the relation between the morphologic outcome and restenosis has not yet been established, sufficient luminal enlargement could be obtained when PTCA was performed until deep arterial tearing or dissection developed. Only vessel stretching may not persist for a long period. Recently Ino and his colleagues (6) reported that the only predictor of successful PTCA seemed to be the elapsed time from the onset of KD to the performance of PTCA: the attempt within 6 to 8 years of the onset of the disease may lead to successful dilatation, and factors such as age at disease onset, lesion site, sex, clinical symptoms, and presence or absence of a perfusion defect on a ^{201}Thallium scintiscan did not seem to affect outcomes of PTCA.

In this case, despite the progression of coronary stenosis he was well except the perfusion defect on ^{99m}Tc-MIBI SPECT with dipyridamole infusion until PTCA was carried out after 4-year 4-months of the onset of illness. In our opinion, PTCA was a necessary and a feasible procedure to prevent further ischemic heart damage even though there were no apparent clinical symptoms. However, it has not been determined when PTCA is carried out in subclinical stenosis. Our successful attempt may indicate that this procedure should be considered early in subclinical stenosis to prevent ischemic cardiac damage.

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