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CASE REPORT

A young adult who had undergone coronary artery bypass grafting and abdominal aortic replacement with prosthetic vessel later after incomplete Kawasaki disease

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KEYWORDS
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Summary A 33-year-old male with a history of undiagnosed fever at the age of 14 years underwent coronary artery bypass grafting (CABG) and abdominal aortic replacement with a prosthetic vessel. Syncope and chest pain on exertion at the age of 19 years led to the diagnosis of complete occlusion of three major coronary branches and emergency CABG was performed. Fourteen years later, a pulsating abdominal mass was an incidental finding during an outpatient clinic visit and an abdominal aortic aneurysm was confirmed by computed tomography. Based on the recorded symptoms and examination findings, the past history of unexplained fever was suspected to be due to incomplete Kawasaki disease. Co-existing systemic arterial lesions should be sought in patients with multi-vessel coronary disease due to Kawasaki disease, although their prevalence is low.

Introduction

Kawasaki disease (KD) is an acute febrile illness of unknown origin affecting children, especially those under 5 years' old [1]. KD is diagnosed based on the presence of five or more of the six major symptoms, or, in Japan, four symptoms with a coronary aneurysm. Undoubtedly, there are cases of incomplete or atypical KD that fail to meet the usual criteria and in which other diseases have been ruled out. Furthermore, because KD is a vasculitis of small- and medium-sized systemic vessels, the other symptoms found in KD are very varied, making incomplete KD difficult to diagnose, especially in infants less than 6 months and older children over 6 years [2]. We experienced a young man who had undergone coronary artery bypass grafting (CABG) and abdominal aortic replacement due to systemic vasculitis of incomplete KD.

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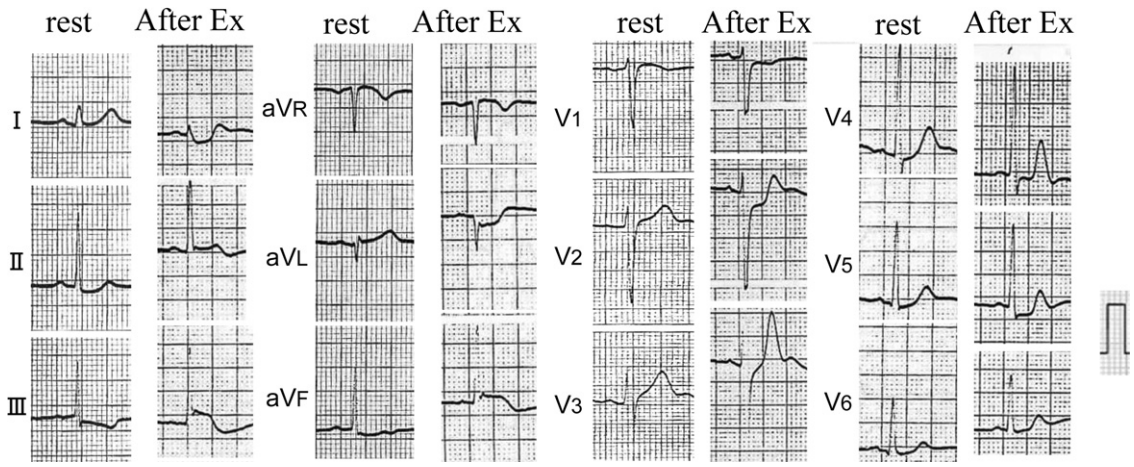

Figure 1 Twelve-lead electrocardiogram before and immediately after Master double testing at the time of chest pain at 19 years' old. The left column shows at rest. Abnormal ST-T segments in leads II, III, and aV_F were detected. The right column shows immediately after exercise. ST-T segments at I, aV_L, V_1—V_5 were depressed, and at III and aV_F were elevated.

Case report

In 1993, a 19-year-old male first felt chest pain lasting about 1 h after running at midnight. The next day, he experienced an oppressive sensation in his chest on effort. Two days after the first pain, he developed dyspnea and chest pain while going uphill, fainted and had incontinence. Because of chest pain on effort for several days, he visited the local hospital. During Master double-step testing, he felt chest pain, his ST-T segments at I, aV_L, and V_1—V_5 were depressed, and were elevated in III and aV_F in a 12-lead electrocardiogram (Fig. 1). Five minutes after taking isosorbide mononitrate, ST-T change gradually resolved. Unstable angina was suspected, and he was referred to our hospital.

His height and weight were 174 cm and 60 kg, respectively. Blood pressure was 120/70 mmHg. Creatinine kinase, white blood cell, and liver transaminase were within normal limits. Total cholesterol, triglyceride, and blood glucose were 137, 94, and 87 mg/dl, respectively. He had no apparent atherosclerotic disease risk factors. Coronary angiography (CAG) revealed a calcified giant aneurysm in the proximal segment of the right and left coronary arteries and total occlusion of segments 1, 6, and 13 (Fig. 2). Left ventricular ejection fraction (LVEF) was 57%, and regional asynergy of left ventricle was not detected. The ST-T segments in leads I, aVL, V5, and V6 became elevated in his electrocardiogram during the examination, and an intra-aortic balloon pump was inserted. Subsequently emergency CABG was performed. The left internal thoracic artery was anastomosed to the diagonal branch, and the right internal thoracic artery to the left anterior descending artery, and a saphenous vein graft was sequentially anastomosed to two posterolateral branches and the posterodescending branch of the right coronary artery. All grafts were patent in angiograms 24 days after operation. Postoperative exercise electrocardiographic studies showed no ischemic ST-T change. Holter electrocardiogram showed no arrhythmias. After 1 year post-operation he made no follow-up visits to our hospital.

At the age of 32 years, he visited another hospital because of abdominal pain. He was diagnosed with gastroenteritis, and a pulsating abdominal mass was detected at that time. An abdominal aortic aneurysm was suspected by abdominal ultrasound, and an aneurysm extending from distal to the bifurcation of renal artery to the bifurcation of common iliac artery was confirmed by computed tomography. Luminal thrombi were present. The diameter of the aneurysm was 54 mm and its length 95 mm (Fig. 3A and B). Preoperative abdominal aortic angiography revealed spindle dilatation from 21 mm distal to the bifurcation of the renal artery to the bifurcation of common iliac artery, and the maximum diameter of aortic lumen was 30 mm because of apparent luminal thrombi. Preoperative CAG revealed that all grafts were patent, and the LVEF was 50%. Because the aneurysm increased in size after 6 months, abdominal aortic replacement with a prosthetic vessel was performed at the age of 33 years (Fig. 3C). The pathological findings of the abdominal aortic wall showed fibrotic, hyaline, and partial calcification changes due to arteriosclerosis.

He had had an episode of undiagnosed fever at the age of 14 years, his fever exceeded 39° for more than 14 days, and was associated with cough, diarrhea, bilateral conjunctival injection, petechial rash, and left facial palsy. He had been admitted to the department of internal medicine in the local hospital on the 19th illness day. Presence or absence of redness of the lips, strawberry tongue, edema of the hands and feet, eruption, or cervical lymphadenopathy was not recorded. It was written in the nursing record that his lips were rough. White blood cell, platelets, fibrinogen, erythrocyte sedimentation rate at 2 h, and C-reactive protein were 10,800, 211,000 mm^{-3}, 575 mg/dl, 92 mm, and 15.4 mg/dl, respectively. Total protein, albumin, glutamic oxaloacetic transaminase, glutamate pyruvate transaminase, and total cholesterol were 5.4, 2.8 g/dl, 79, 155 U/l, and 56 mg/dl, respectively. Antinuclear antibodies were negative. Chest X-ray revealed a left pleural effusion. His fever persisted despite administration of ceftazidime and minocycline hydrochloride. Steroids had been started on the 26th day and intravenous immunoglobulin was administered on the 33rd day, when his temperature decreased to 37.5°. Steroids were tapered from the 46th day, and he was dis-