We report on a 4-year-old boy admitted to our Institute because of fever (39°C) persisting for 8 days. Physical examination showed bilateral conjunctivitis, strawberry tongue, lip fissures, enlarged cervical lymph nodes, and polymorphous rash, followed by finger and toe desquamation (see Figure 1 in the online-only Data Supplement). Diagnosis of Kawasaki disease was made. According to the Kobayashi risk score, our patient had 8 points, predicting treatment unresponsiveness of 71.4%.1 Over the next 2 weeks, the symptoms of acute phase persisted despite treatment with intravenous immunoglobulin, as well as retreatment with intravenous immunoglobulin and methyl-prednisolone boluses. In week 4 of the disease, he was given infliximab in a single dose of 5 mg/kg. He became afebrile, and his inflammatory parameters normalized after treatment with infliximab.

An initial echocardiography examination performed immediately after the admission showed no specific changes. The second echocardiographic study, performed 2 weeks after the onset of the disease, revealed coronary artery aneurysms involving left anterior descending artery, 8 mm in diameter, and the right coronary artery, 5 mm in diameter (Figure 1A). Antiplatelet therapy (clopidogrel along with aspirin) was administered. Despite the administered therapy, 1 month after the onset, formation of giant aneurysms was diagnosed. The right coronary artery was 24 mm (z score of 62) and left anterior descending artery was 16 mm (z score of 48) in diameter, both filled with intraluminal thrombi (Figures 2). Thrombolytic agent (recombinant tissue plasminogen activator) together with anticoagulant therapy were given, and, 2 weeks later, complete recanalization of coronary arteries was achieved (Figure 1B). Treatment was continued with oral anticoagulants and aspirin to maintain international normalized ratio between 3.0 and 4.0. Nine months later, a cardiac magnetic resonance imaging demonstrated antero-septal subendocardial and cardiac apex transmural scar (Figure 3B–3D), along with hypo-kinesia in the same region vascularized by left anterior descending artery (see Movies I through III in the online-only Data Supplement). Global contractility was preserved, and left ventricle end-diastolic diameter (39 mm) and volume (96.6 mL/m²) were slightly above normal limits (27.4–40.4 mm and 47–92 mL/m², respectively). The patient still had extensive coronary artery aneurysms unchanged in diameter (right coronary artery, 27 mm; left anterior descending artery, 17 mm) but without intraluminal thrombi formation (Figures 3A and 4; see Movie IV in the online-only Data Supplement). After 22 months of follow-up, our patient is symptom free, with spontaneous contrast in the area of dilated right coronary artery (Figure 5; see Movie V in the online-only Data Supplement) but with no echocardiography repolarization abnormalities and additional cardiac function impairment.

Kawasaki disease is an acute vasculitis that occurs mostly in infants and young children.2 To the best of our knowledge, the giant coronary aneurysms detected in our patient are the largest ones ever described in Kawasaki disease.3,4 Risk of thrombosis is very high when giant coronary aneurysms develop promptly. Noninvasive diagnostic imaging techniques, such as echocardiography and magnetic resonance imaging, are very useful in diagnosing subclinical myocardial infarction. Surgical experience with giant coronary aneurysms attributable to Kawasaki disease is still very limited in most of the centers worldwide.

Disclosures

None.

References


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The online-only Data Supplement is available with this article at http://circ.ahajournals.org/lookup/suppl/doi:10.1161/CIRCULATIONAHA.113.002092/-/DC1. Correspondence to Jovan Kosutic, MD, PhD, Cardiology Department, Mother and Child Health Institute of Serbia, School of Medicine, University of Belgrade, R. Dukica St. 6-8, 11070 Belgrade, Serbia. E-mail kosutic@eunet.rs (Circulation. 2013;128:2257-2258.) © 2013 American Heart Association, Inc.

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Figure 1. Parasternal short-axis echocardiographic image. **A**, Initial coronary artery aneurysms. **B**, Enlarged coronary arteries without thrombus. Ao indicates aorta.

Figure 2. Parasternal short-axis echocardiographic image. Giant coronary artery aneurysms almost completely filled with thrombus: **A**, right coronary artery of 24 mm (arrow); **B**, left anterior descending artery of 16 mm (arrow). There were narrow channels through which blood passed into the distal parts of the heart. Ao indicates aorta.

Figure 3. **A**, Cardiac magnetic resonance 3-dimensional reconstruction image of the dilated right coronary artery (arrow). Late gadolinium enhancement magnetic resonance (phase-sensitive inversion recovery) imaging for detecting cardiac apex transmural and anteroseptal subendocardial myocardial scar (**B**, short-axis view; **C**, 4-chamber view; **D**, 2-chamber view).

Figure 4. Magnetic resonance images of giant coronary artery aneurysms. **A**, Right coronary artery is up to 27 mm; **B**, left anterior descending artery (left main coronary artery) is up to 17 mm. The diameter of the aorta (Ao) is 18 mm.

Figure 5. Parasternal short-axis echocardiographic images (**A** and **B**) that revealed spontaneous contrast signal change (arrows) in the area of the right coronary artery aneurysm.
Bilateral Giant Coronary Aneurysms in Kawasaki Disease: How Difficult Can It Be?
Sergej Prijic, Goran Ristic, Srdjan Pasic, Aleksandra Minic, Vladislav Vukomanovic, Oto Adjic, Sanja Ninic and Jovan Kosutic

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SUPPLEMENTAL MATERIAL

1) Supplemental Figure and Figure Legend
Data Supplement Image 1. Red lips with fissures and rash (a), unilateral cervical lymphadenopathy (c), and finger (b) and toe (d) desquamation.
**Movies legend**

Data Supplement Movie 1. Echocardiographic apical four chamber view demonstrates apical hypokinesia (vascularized by left anterior descending coronary artery) with preserved global contractility of the left ventricle (recommended application for viewing movie file is Windows Media Player).

Data Supplement Movie 2. Cardiac apex motion abnormalities evaluated by ECG gated cine magnetic resonance imaging (4-chamber view) (recommended application for viewing movie file is Windows Media Player).

Data Supplement Movie 3. Apical wall abnormalities evaluated by ECG gated cine magnetic resonance imaging (left ventricle 3-chamber view) (recommended application for viewing movie file is Windows Media Player).

Data Supplement Movie 4. Echocardiographic image shows extensive coronary arteries aneurysms without intraluminal thrombi formation (recommended application for viewing movie file is Windows Media Player).

Data Supplement Movie 5. Echocardiographic image demonstrates dilated right coronary artery with spontaneous contrast (recommended application for viewing movie file is Windows Media Player).