Kawasaki disease (KD) is a leading cause of childhood acquired heart disease in Asian population and a small proportion of adult coronary artery stenosis are secondary to childhood KD.[1-3] Giant aneurysms larger than 8 mm seen in few patients may always not regress fully, some may eventually calcify, thrombose, or become stenotic and may get diagnosed for the first time in adult life.[4,5] On long-term follow-up, many patients with aneurysms are asymptomatic in spite of coronary occlusions and few suffer silent myocardial infarctions.[6-8] Tomisaku Kawasaki’s initial description of mucocutaneous lymph node syndrome in childhood was in 1967 and coronary arteritis was recognized later.[9]

Our patient, aged 58 years, is one of the oldest survivor with giant aneurysms on both coronary arteries. His heart failure was contributed by left to right shunt from an uncorrected large atrial septal defect (ASD). He had obesity, hypertension, diabetes mellitus, and features of right heart failure for few years duration, but had no angina. His childhood febrile illnesses were never given much significance and investigated in the past. His large ASD was diagnosed for the first time at 58 years of age. He presented with isolated systolic hypertension, bipedal edema, and features of large pre-tricuspid left to right shunt. There was cardiomegaly and increased pulmonary vascularity on chest X-ray and exercise electrocardiogram showed no ischemia.

The transesophageal echocardiogram showed a 27-mm secundum ASD with left to right shunt and right sided chamber enlargement. There was a giant 6 cm right coronary artery aneurysm in the right atrioventricular groove with a layered thrombus. The left ventricular global and regional systolic function was good. Multi-slice computed tomographic scan [Figure 1, Video 1] and selective coronary angiogram [Figure 2,
Singhi, et al.: Giant coronary artery aneurysms in a 58-year-old

Video 2] showed a giant aneurysm in proximal right coronary artery with layered thrombus, calcification, and sluggish TIMI II flows. Proximal left anterior descending coronary artery was also aneurysmal, dilated to 10 mm, and showed calcifications. There were calcifications, luminal irregularities, and dilatation of the left main coronary artery and the left circumflex artery also [Figure 3, Video 3]. The giant coronary aneurysms with calcification were strongly suggestive of a sequel of old KD. Even though intravascular ultrasound or optical coherence tomographic imaging might have added more information, we did not perform them due to technical difficulties in both these imaging techniques in giant aneurysms of 6 cm and fear of distal embolization of the layered thrombus in the aneurysm walls. The atrial, ventricular end diastolic, and pulmonary artery pressures were mildly elevated. In the absence of significant stenotic coronary artery disease and preserved left ventricular systolic function, the heart failure was caused by left to right shunt through the ASD compounded by the diastolic dysfunction caused by concentric LVH secondary to hypertension.

The ASD was closed with 30 mm Amplatzer atrial septal occluder (St Jude Medical, MN) under transesophageal echocardiographic guidance. The left ventricular systolic and end diastolic pressures of 175 and 20 mm Hg, respectively did not increase after the device closure. One year after the procedure, he had good effort tolerance, no pedal edema, and started active farming and physical activities. His medications included warfarin, low dose aspirin along with statins and losartan. After 1.5 years of transcatheter ASD device closure the patient had sudden cardiac death in home.

Clinical findings may be subtle in atypical and incomplete KD and the coronary lesions get detected later in life. Giant aneurysms lead to silent coronary stenosis and occlusions in about 48% patients and myocardial infarction in about 23% of patients.[3] Many asymptomatic patients may get incidentally detected on angiogram.[5,6] Even though there were sluggish coronary flows due to aneurysmal dilatation in our patient, there was no definite luminal occlusion to warrant revascularization. His sudden cardiac death after 1.5 years could be due to rupture of aneurysm or acute coronary syndrome, and could not be diagnosed precisely due to lack of autopsy. In a detailed review of reports, our 58-year-old patient was the oldest known patient with angiographic features of giant Kawasaki coronary aneurysms.

REFERENCES

1. Burns JC, Kushner HI, Bastian JF, Shike H, Shimizu C, Matsubara T, et al. Kawasaki disease: A brief history. Pediatrics 2000;106:E27.
2. Kato H, Inoue O, Kawasaki T, Fujiwara H, Watanabe T, Toshima H. Adult coronary artery disease probably due to childhood Kawasaki disease. Lancet 1992;340:1127-9.
3. Kato H, Ichinose E, Yoshioka F, Takechi T, Matsunaga S, Suzuki K, et al. Fate of coronary aneurysm in Kawasaki disease: Serial coronary angiography and long-term follow-up study. Am J Cardiol 1982;49:1758-66.
4. Committee on Rheumatic fever, Endocarditis, and Kawasaki Disease of the American Heart Association’s Council on Cardiovascular Disease in the Young. Diagnostic guidelines for Kawasaki disease. Am J Dis Child 1990;144:1218-9.
5. Rozo JC, Jefferies JL, Eidem BW, Cook PJ. Kawasaki disease: A case report and review of the literature. Tex Heart Inst J 2004;31:160-4.

6. Inoue O, Akagi T, Kato H. Fate of giant coronary artery aneurysms in Kawasaki disease: Long-term follow-up study [abstract]. Circulation 1989;80(Suppl II):I262.

7. Canino-Rodríguez A, Cox RA. Giant coronary aneurysms in a young adult patient with Kawasaki disease. P R Health Sci J 2008;27:382-6.

8. Sakai Y, Takayanagi K, Inoue T, Yamaguchi H, Hayashi T, Morooka S, et al. Coronary artery aneurysms and congestive heart failure--possible long-term course of Kawasaki disease in an adult--a case report. Angiology 1988;39:625-30.

9. Kawasaki T. Acute febrile mucocutaneous syndrome with lymphoid involvement with specific desquamation of the fingers and toes in children. Arerugi 1967;16:178-222.

How to cite this article: Singhi AK, Pavithran S, Sivakumar K. Giant coronary artery aneurysms in a 58-year-old. Ann Pediatr Card 2014;7:152-4.

Source of Support: Nil. Conflict of Interest: None declared.