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Giant aneurysms of pan-proximal coronary arteries including left main presenting as acute coronary syndrome in 23-year-old male: an unusual sequelae of Kawasaki disease

Olbrzymi tętniak obejmujący kilka tętnic wieńcowych, w tym pień tętnicy wieńcowej lewej, powodujący objawy ostrego zespołu wieńcowego u 23-letniego chorego: nietypowe powikłanie choroby Kawasaki

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Abstract

The reported incidence of coronary artery aneurysms (CAA) on angiography varies between 0.3 to 5.3%. Right coronary artery is the most commonly affected followed by left circumflex or left anterior descending artery. Three-vessel or left main involvement is exceedingly rare. Atherosclerosis accounts for the vast majority of CAAs in adults, whereas Kawasaki disease is responsible for most cases in children. We report a rare case of Kawasaki disease associated with giant aneurysms (> 8 mm) in coronary circulation, including left main coronary artery in 23-year-old male having maturity onset diabetes (MODY), who presented with acute coronary syndrome.

Key words: coronary artery aneurysm, Kawasaki disease, giant aneurysm

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Introduction

Kawasaki disease (KD) is an acute vasculitis of unknown aetiology, that predominantly affects children under five years of age. Structural damage to the coronary arteries after the acute, self-limited illness is detected by echocardiography in 25% of untreated patients [1]. Damage to the coronary arteries as a sequela of Kawasaki disease was first recognised by Kato et al. in 1975 [1]. Angiographic and echocardiographic studies have documented coronary artery aneurysms in 25% of untreated patients [2, 3]. Studies of the natural history of these lesions suggest that they may either remodel as a result of myointimal proliferation, or may persist and develop stenosis of the vessel lumen at the outlet of the aneurysm [3].

Case report

A 23-year-old male having mature onset diabetes (MODY) presented with retrosternal chest pain and diaphoresis of three hours duration. He had past history of fever with rash and desquamative lesions of hands along with polyarthralgia in childhood. He also had sensorineural hearing loss (SNHL), as a chronic sequela of Kawasaki disease. His physical examination and biochemistry were all unremarkable. Electrocardiogram showed ST depression

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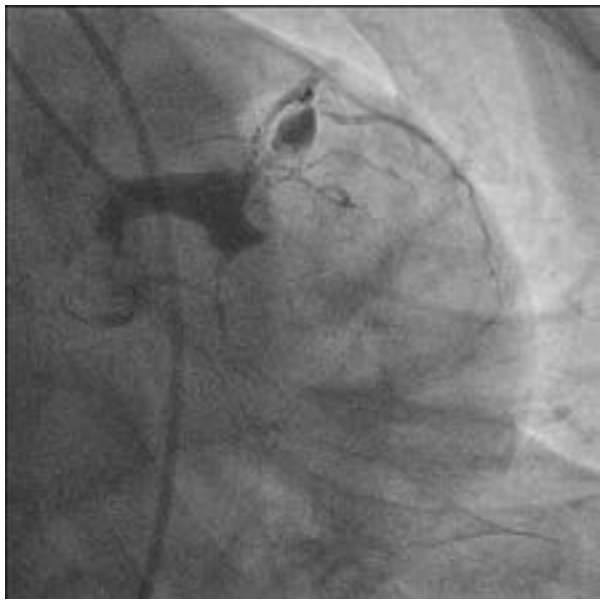

Figure 1. Antero-posterior caudal view showing giant saccular aneurysms involving left main and proximal left circumflex coronary artery

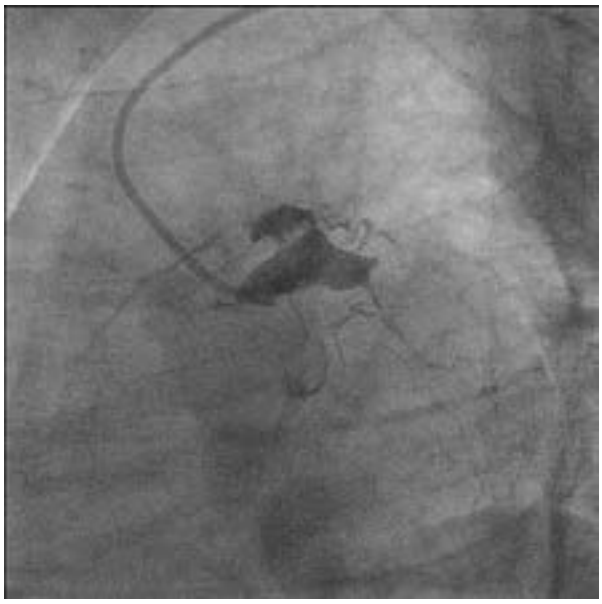

Figure 2. Left anterior oblique-caudal view showing giant saccular aneurysms involving left main, proximal left circumflex coronary artery and fusiform aneurysm of proximal left anterior descending artery

with T wave inversion in precordial leads. Troponin I and T were raised. Echocardiography revealed regional wall motion abnormality in LAD territory with mild LV dysfunction (ejection fraction [EF] 40%). He was preloaded with prasugrel — 60 mg, acetylsalicylic acid — 325 mg and atorvastatin — 80 mg. He was taken to catheterisation laboratory for early coronary intervention in lieu of non ST elevation myocardial infarction after proper consent. 6F JR3.5 and JL3.5 Proflo™ diagnostic catheters (Medtronic, USA) were used for catheterisation after administering 2,500 U of heparin. Coronary angiogram revealed giant aneurysm involving left main coronary artery (11.2 mm), proximal left circumflex artery (10.1 mm) (Figure 1) and proximal left anterior descending artery (8.5 mm) (Figure 2). Giant aneurysm in the proximal and middle segments of right coronary artery (9.8 mm) was also seen (Figure 3). Based on angiographic findings, we decided to continue with medical management. The patient was discharged in stable condition on fifth day and advised surgical revascularisation.

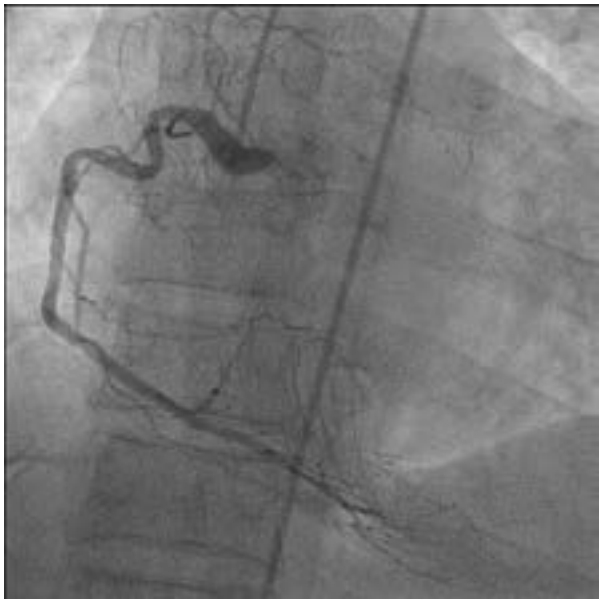

Figure 3. Antero-posterior view showing saccular aneurysm of proximal right coronary artery

Discussion

The coronary artery abnormalities in patients with Kawasaki disease include coronary artery aneurysm with calcification, stenosis, and extensive collateral formation. Spectrum of clinical complications included acute coronary syndrome, arrhythmia, heart failure and sudden death. Patients with aneurysms caused by Kawasaki disease had a strikingly high prevalence of left main coronary artery disease (42%), as compared with patients with atherosclerotic coronary aneurysms reported in the National Heart, Lung, and Blood Institute Coronary Artery Surgery Study (CASS) (4%) [4]. In our case, CAA was the sequela of KD, as the patient was an already known case of it. Secondly, he had no risk factors for coronary artery disease. The unusual feature on angiogram was giant aneurysm of left main trunk along

with pan-proximal affliction of all three arteries. Coronary aneurysms with a diameter > 8.0 mm are categorised as giant coronary aneurysms (GAs). In their study by Kato et al. [5], all patients with small to medium coronary aneurysms showed regression of the coronary aneurysm, but none of the patients with GA showed complete regression. Furthermore, they also demonstrated that the remaining patients with moderate to giant aneurysms showed either persistent coronary aneurysms or the development of stenosis over time. In the world's largest cohort of patients with KD complicated by GA, who have been followed up for the longest time, as shown by Suda et al. [6], a survival rate was 88% up to 30 years. They have also demonstrated, that ongoing remodelling of coronary arteries with GA might continue long after KD, leading to the development of coronary stenosis, even > 21 years after the onset of the disease. They showed an increasing frequency of coronary stenosis in patients with coronary aneurysms up to 17 years after the diagnosis. The potential mechanisms underlying these findings include ongoing remodelling of coronary arteries as a result of ongoing inflammation of coronary arteries, demonstrated by higher levels of serum high-sensitivity C-reactive protein and amyloid A [7] and increased oxidative stress [8].

Another unusual feature of the patients with antecedent Kawasaki disease is ring calcification on the chest radiograph, which was lacking in our patient. Although conclusive documentation of antecedent Kawasaki disease may lack in many patients, as it primarily affects children under five years of age, the patients are usually unable, as adults, to recall details of their childhood illness. Also many cases of Kawasaki disease are misdiagnosed during the acute illness, because the disease shares many features with other childhood illnesses, such as scarlet fever and measles. The possible contribution of antecedent Kawasaki disease to the genesis of cardiovascular disease in adults was investigated by Kato et al. [9] in Japan. Among a survey of 354 hospitals, they identified 130 adult patients with coronary artery aneurysm, where 109 patients might have had antecedent Kawasaki disease, but information regarding childhood illness was not available. Therefore, giant aneurysms, especially involving the left main, parental information about childhood history is very important. Kawasaki disease in childhood can cause permanent coronary artery damage, that may remain clinically silent until adulthood. Coronary artery bypass graft surgery remains the treatment for their obstructive coronary artery disease, as these aneurysms are usually localised to the proximal vessel segments and the distal segments are free of disease.

Conflict of interest(s)

The authors declare no conflict of interest.

Streszczenie

Częstość wykrywania w koronarografii tętniaków tętnicy wieńcowej (CAA) wynosi od 0,3 do 5,3%. Zmiany są najczęściej zlokalizowane w prawej tętnicy wieńcowej, nieco rzadziej w gałęzi okalającej i gałęzi przedniej zstępującej. Tętniaki obejmujące trzy tętnice czy pień lewej tętnicy wieńcowej są niezwykle rzadkie. U dorosłych przyczyną rozwoju CAA w przeważającej większości przypadków pozostaje miażdżyca, natomiast u dzieci najczęściej są one następstwem choroby Kawasaki. Autorzy opisują rzadki przypadek choroby Kawasaki związanej z ogromnym tętniakiem (> 8 mm) w krążeniu wieńcowym, obejmującym pień lewej tętnicy wieńcowej, u 23-letniego chorego na cukrzycę typu MODY (*maturity onset diabetes of the young*), u którego występowały objawy ostrego zespołu wieńcowego.

Słowa kluczowe: tętniak tętnic wieńcowych, choroba Kawasaki, olbrzymi tętniak

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