

Coronary artery bypass grafting, endarterectomy, and aneurysmorrhaphy for Kawasaki disease in an infant

Bir yenidoğanda Kawasaki hastalığı nedeniyle yapılan koroner arter baypas greftleme, endarterektomi ve anevrizmorafi

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ABSTRACT

In this article, we report an infant case of delayed diagnosis of Kawasaki disease. Despite intravenous immunoglobulin, acetylsalicylic acid, immunosuppressive and anticoagulant therapy agents, ischemic alterations on electrocardiography and a persistent giant aneurysm in coronary arteries and with a thrombus in the left anterior descending artery on repeated echocardiographies were detected. Computed tomography angiography and conventional angiography confirmed the diagnosis. Coronary artery bypass grafting, endarterectomy, and aneurysmorrhaphy were successfully performed at one year of age. The case still remains uneventful during outpatient clinic visits after discharge.

Keywords: Coronary artery bypass grafting; infant; Kawasaki disease.

Kawasaki disease (KD) may lead to coronary artery disease in 25% of children, if left untreated. Early treatment with acetylsalicylic acid (ASA) and high-dose intravenous immunoglobulin (IVIG) decreases the incidence of coronary artery damage; however, despite appropriate treatment, aneurysms continue to develop in approximately 3 to 5% of children.^[1,2]

In some patients may develop obstructive coronary disease, myocardial infarction, and sudden death.^[1] Coronary artery bypass grafting (CABG) is the preferred treatment procedure for patients with severe coronary stenosis secondary to KD.^[2,3] Herein, we report a case of KD who was diagnosed in the

ÖZ

Bu yazıda geç Kawasaki hastalığı tanısı konulmuş bir yenidoğan olgusu sunuldu. İntravenöz immünglobulin, asetilsalisilik asit, immünsüpresif ve diğer antikoagülan tedavi ajanları kullanılmasına rağmen, elektrokardiyografide iskemik değişiklikler ve ekokardiyografik kontrollerde koroner arterlerde devam eden dev bir anevrizma ve sol ön inen arterde trombus saptandı. Bilgisayarlı tomografi anjiyografi ve konvansiyonel anjiyografi ile tanı doğrulandı. Bir yaşında iken, başarılı koroner arter baypas greftleme, endarterektomi ve anevrizmorafi işlemi uygulandı. Olgu, taburculuk sonrası poliklinik kontrollerinde sorunsuz izlenmektedir.

Anahtar sözcükler: Koroner arter baypas greftleme; yenidoğan; Kawasaki hastalığı.

late stage with a giant thrombotic coronary aneurysm treated by aneurysmorrhaphy and CABG.

CASE REPORT

A 10-month-old female patient was evaluated in a clinic due to fever for five days. Antibiotic and antipyretic treatment were initiated. After six days along with persistent fever and due to eruptions on her body, she was referred to an another clinic. Maculopapular erythema, hyperemia of oropharynx, red strawberry tongue, hyperemia on lips, erythema and edema on back of the both hands, perineal hyperemia were detected. Pericardial effusion (5 mm in diameter),



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fusiform dilatations on left main coronary artery (LMCA), and right coronary artery (RCA) were detected on echocardiography and IVIG (2 g/kg) and ASA treatment (80 mg/kg) were initiated. At 24 hours following the treatment, symptoms and fever slightly decreased. Echocardiography revealed multiple saccular aneurysms of largest measured around 9 mm on LMCA and fusiform dilation measured 5.2 mm on RCA. As a result, warfarin treatment was added to the ASA and dipyridamol treatment.

Twenty days after the initial symptoms, echocardiography revealed an 8x9 mm giant thrombus inside the giant aneurysm on the LMCA. Then, heparin (30 IU/kg/hour) and tissue plasminogen activator (tPA) therapy were started.

Fifty days after the initial symptoms, ST-T segment alterations were detected on electrocardiography (ECG). Troponin I level was titrated up to 0.065 mg/L (normal levels: 0.01-0.023 mg/L). The patient was referred to our clinic after the detection of a giant aneurysm on the left anterior descending (LAD) artery. The patient on enoxaparin, propranolol, ASA, tPA, and clopidogrel treatment was hospitalized in the intensive care unit.

Electrocardiography revealed ST segment depression on anterolateral derivations. The echocardiographic study revealed an aneurysm of 11.9 mm in diameter and a thrombus around 11x12 mm inside the aneurysm on LAD artery. The global segmental wall movements were normal (fractional shortening 32%, ejection fraction 65%) without any mitral regurgitation finding (Figure 1).

The giant aneurysm was confirmed by conventional coronary angiography (Figure 2) and surgery was planned.

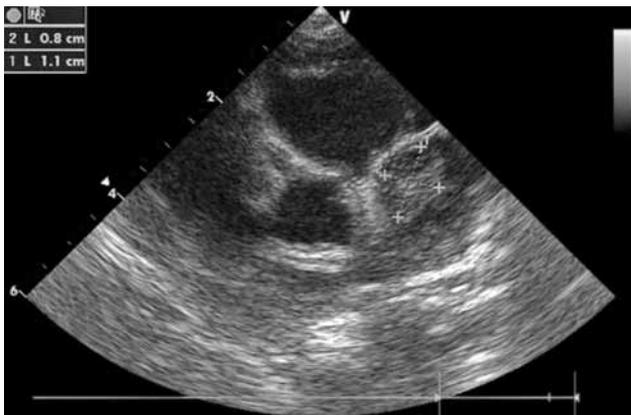


Figure 1. Echocardiographic image demonstrating an aneurysm of 11.9 mm in diameter and a thrombus around 11x12 mm inside the aneurysm of the left anterior descending artery.

The operation was performed under mild hypothermic cardiopulmonary bypass with aortobicaval cannulation. Cold blood antegrade cardioplegia was used and maintained with direct intracoronary and retrograde route during operation. There was a giant aneurysm in the left anterior coronary artery, right after the circumflex artery takes off, a full of organized thrombus. Aneurysm wall showed inflammatory alterations such as atherosclerosis. There were several diagonal and septal branch ostia in the aneurysm sac. The distal LAD ostium was small as the proximal side. Endarterectomy including small branch ostia followed thrombectomy. Then, aneurysm sac was plicated cutting both side walls. Before closing the sac, the optimal place for anastomosis was marked on the LAD artery with a coronary probe. An anastomosis of the left internal thoracic artery (LITA) and LAD artery was performed. Left anterior descending artery perfusion by LITA was confirmed before the cross-clamp was removed (Figure 3).

At four months following surgery, echocardiography showed excellent global ventricular function, showing a good surgical result.

DISCUSSION

The main systemic effect of KD is cardiac involvement. It is strongly associated with long-term mortality and morbidity. In addition to coronary

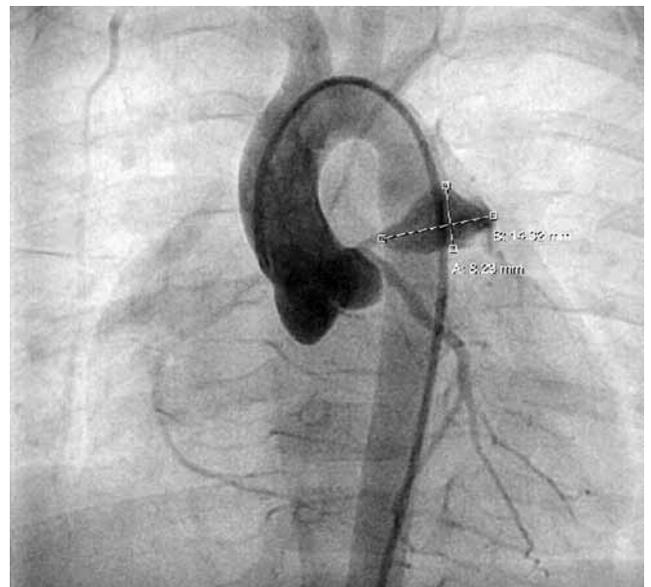


Figure 2. An anteroposterior image of a left coronary artery angiogram demonstrating an aneurysm of the left anterior descending artery and an obstructive thrombus restricting the flow inside. The circumflex artery were normal.

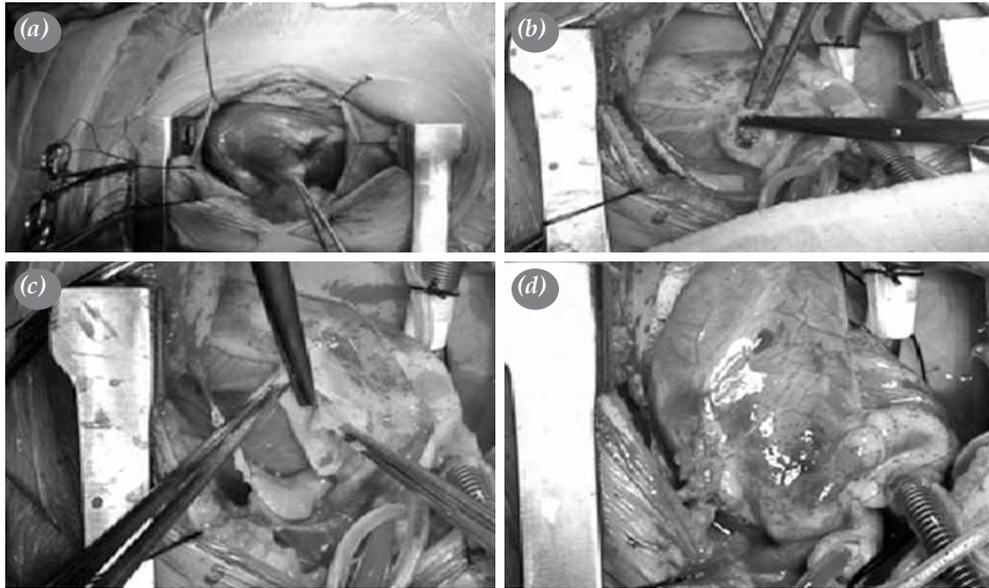


Figure 3. (a) An intraoperative view reveals a giant aneurysm in the left anterior coronary artery right after the circumflex artery takes off (b) An intraoperative view after the opening aneurysm sac. (c) An intraoperative view of endarterectomy of the left anterior descending artery following thrombectomy. (d) An intraoperative view after the aneurysmorrhaphy of aneurysm sac and left internal thoracic artery-left anterior descending artery coronary artery bypass grafting.

artery involvement, pericardial effusion, myocarditis, cardiac failure, and dysrhythmia can be also seen.^[1-3]

In the acute phase, echocardiography is the method of choice for cardiovascular evaluation and should be performed as soon as the diagnosis is suspected; however, treatment with IVIG should not be delayed.^[1-4] As aforementioned, coronary artery aneurysms can develop in 25% of the untreated patients and this rate may decrease to 3 to 5% in patients treated in the early stage (i.e. within first 10 days of illness). The main goal of acute treatment is to prevent long-term sequela and probably, most importantly, the inflammation of the wall of the coronary arteries by controlling the acute inflammation. After the basal cardiac examinations, high-dose IVIG (2 g/kg, 10-12 hours infusion) and ASA (80-100 mg/kg/day) therapy should be initiated immediately. In unresponsive patients, there are limited reports on application of intravenous high-dose steroid therapy (30 mg/kg/day, 1-3 days), or in refractory cases, tumor necrosis factor-alpha (TNF- α) (infliximab), methotrexate, plasmapheresis, and cytotoxic agents can be used.^[1-4]

In the present case, IVIG therapy was introduced in the late stage of the disease with an increased risk of coronary complications. Also, pericardial effusion was detected during echocardiography.

Furthermore, surgical revascularization for coronary artery disease secondary to KD is relatively uncommon. Aneurysms tend to develop most frequently in the LMCA, followed by LAD artery and RCA. The left circumflex artery is rarely involved.^[4] A successful CABG with saphenous vein grafting (SVG) for KD was first reported in 1976.^[3] Kitamura et al.^[5] reported the first successful CABG with internal mammary artery (IMA) graft for KD in 1985 (Kitamura operation) and thereafter IMA grafts have been widely used.

The preferred grafts for CABG in these patients are the IMA, since they grow with the somatic growth of children, do not appear to be affected by atherosclerosis, and may have some advantages in terms of endothelial function. Saphenous vein grafts have shown good results in the right coronary circulation with excellent long-term outcome in children with KD.^[5]

There can be rarely ruptures of coronary artery aneurysms in the late stage, as fibrous scars or calcifications appear on aneurysm walls in this period. Some authors suggest partial coronary artery aneurysmectomy combined with CABG for myocardial ischemia of KD; however, most others do not recommend coronary artery aneurysmectomy due to small branches from aneurysms.^[6]

In the present case, the left IMA was used for the LAD. Also, aneurysmorrhaphy (thrombectomy, endarterectomy, and plication simultaneously) was performed. To the best of our knowledge, our case was the youngest patient in whom surgery was performed.

Moreover, we preferred dual supply for LAD artery due to the relative small size of LITA in the early postoperative period. It would be a problem in terms of myocardial perfusion to choose only antegrade flow through the plicated aneurysm sac or only LITA. As the development of a new occlusion with a thrombus or a wall thickening in the sac due to endarterectomy was possible, LITA would be insurance in the late postoperative period. However, LITA would not have been a good blood source for early postoperative period due to its small size. Therefore, the aneurysm sac was used for antegrade blood supply. Although the LITA occlusion is possible due to run-off, the result of this decision would be seen in further catheterization. Beside this, early postoperative period was excellent in our case.

Coronary artery bypass grafting is indicated in symptomatic patients with severe stenosis, as evidenced by coronary angiography in asymptomatic patients with progressive stenosis, as assessed by angiography or in asymptomatic patients with ischemic alterations as demonstrated by noninvasive testing.^[2]

It is even indicated in young children with giant coronary aneurysms >8 mm in diameter with or without a stenotic region, when myocardial ischemia can be detected noninvasively. Coronary artery bypass grafting is the standard therapy for severe coronary artery stenosis.^[7]

Kitamura et al.^[5] reported a survival rate of 95% and an event-free rate of 60% at 25 years of follow-up after CABG surgery in KD. We also suggest that regular follow-up visits should always be maintained, since there is a progressive decline in the event-free rate and endothelial function abnormalities persist many years after the acute phase, even in patients without coronary involvement. Wagner et al.^[8] also reported an in-hospital mortality rate of 1.6% and 5.2% at 4.5 years after surgery in patients with KD.

In conclusion, this case report highlights the importance of early diagnosis and treatment of

Kawasaki disease, bearing in mind its potential late complications. Coronary artery bypass grafting is a safe and effective method for myocardial ischemia even in infants with Kawasaki disease and yields good short-term results. However, further studies are required to obtain long-term results.

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