A Case of a 3-Month-Old Infant with Incomplete Kawasaki Disease After Rotavirus Gastroenteritis

Rotavirüs Gastroenteriti Sonrası İnkomplet Kawasaki Hastalığı Gelişen 3 Aylık Bir İnfant Olgu

Banu Gülcan Öksüz1, Mahir Iğde2
1Samsun Education and Research Hospital, General Pediatrics Clinic, Samsun, Turkey
2Samsun Education and Research Hospital, Pediatric Allergy and Immunology Clinic, Samsun, Turkey

Abstract

Kawasaki disease is diagnosed on the basis of special findings. Patients who do not have all of the diagnostic criteria are considered as incomplete cases. Infants have more coronary complications because of the delays in diagnosis. We report the case of a 3-month-old with incomplete Kawasaki disease after rotavirus gastroenteritis. The Journal of Pediatric Research 2014;1(1):36-8

Key Words: Kawasaki disease, incomplete, coronary artery, infant, rotavirus

Introduction

Kawasaki disease (KD) is an acute febrile disease of unknown etiology. It is a systemic vasculitis characterized by a systemic vascular inflammation involving the small and medium sized arteries especially including the coronary arteries.

It was first described as mucocutaneous lymph node disease or infantile periarteritis nodosa in Japan in 1967 (1). KD is seen more often in boys (1.5/1) and occurs between the ages of 6 months-5 years. It is the most common cause of acquired cardiac disease in children in developed countries. Although the reason is not exactly known, bacterial toxins, viruses, autoimmunity are accused and it is also thought to be a genetic predisposition (2).

There is no single criterion for the diagnosis so, prolonged fever more than 5 days should be considered as pathological and KD should be suspected.

There is risk of sudden death and chronic disease due to the affected coronary arteries. For this reason early
and correct diagnosis is very important. Administration of intravenous immunoglobulin within 10 days of fever prevents the complications.

The diagnosis may be delayed in the cases at <1 year and >9 years of age because of incomplete forms. We report the case of a 3-month-old female infant with the incomplete KD after rotavirus gastroenteritis.

**Case Report**

A 3-month-old female infant was brought to our hospital with high fever, vomiting and diarrhea that continued for 6 days although oral antibiotic therapy was given. She had a history of non purulent conjunctivitis 2 days before the reference to the hospital. Personal and family history was unremarkable.

Fever was 38.5 °C at physical examination. She was weak and restless. There was a slight injected pharynx, significant cracks and hyperemia on lips. Dermatitis was present at genital area. Other system aspects were normal.

There was a mild leukocytosis (Wight blood cells (WBC): 21660/mm$^3$), anemia (hemoglobin (Hb): 10.7 gr/dl), dominance of neutrophils 69% in peripheral blood smear, increase in erythrocyte sedimentation rate (ESR: 82 mm/hour), mild thrombocytosis (674000/µL) in laboratory findings. C-reactive protein (CRP) value was 36 mg/dl. She had sterile pyuria (17 leukocytes in every area). Chest x-ray was normal. Ultrasounds were normal for subdural effusion and intra abdominal pathologies. Due to the tenderness of bilateral lower extremities, hip joint ultrasound was done to rule out the diagnosis of septic arthritis and found normal.

Ampicillin treatment was initiated and netilmicin was added to the treatment for the suspicion of urinary tract infection.

There was no reproduction in blood, urine and stool cultures. Liver and renal functions were normal. No lumber puncture was done because her family did not give permission. For this reason treatment was changed to vancomycin and ceftriaxone therapy for meningitis. Salmonella and brucellosis serology were negative for differential diagnosis of fever. As the diarrhea continued, series of stool examinations were done on day 3 and 5 and rotavirus antigen screening in stool examination was found positive. She did not have a history of rotavirus vaccination.

Fever continued on day 11. KD was suspected because of the long lasting fever, skin and mucosal findings and conjunctivitis. Echocardiogram revealed dilatation of the left coronary artery (3.2 mm). Diameter of right coronary artery was normal. According to the echocardiogram the diagnosis was confirmed. A single dose of 2 g/kg/day immunoglobulin (IVIG) was administered intravenously in a 12-hour infusion on day 11. Also acetyl salicylic acid was started (80 mg/kg in 4 doses) at the same time. Fever disappeared at the 3rd hour of the IVIG therapy. Mucosal lesions of the lip disappeared 2 days after IVIG. Gastroenteritis improved. A clinical improvement was observed. No periungual desquamation appeared on her hands during follow up.

Thrombocytosis was observed 5 days after IVIG treatment (850.000 K/µl). Dose of acetyl salicylic acid was decreased to antithrombotic dose (5 mg/kg/g) on the 14th day of the treatment. The patient’s acute phase reactants (ESR, CRP) improved at discharge. Her coronary artery dilatation was in boundary value so acetyl salicylic acid was continued. She was discharged and called for outpatient controls. Because of her parents’ movement to another city, her controls were made in another hospital. Her echocardiogram findings were normal in the 3rd month of follow up and she was clinically healthy so acetyl salicylic acid treatment was discontinued.

**Discussion**

Kawasaki disease is an acute febrile disease of unknown etiology. Bacterial toxins, viruses, autoimmunity are accused (3). As in our case, a case of KD was reported after rotavirus gastroenteritis but in this case anasarca edema developed because of microcapillary hyper permeability secondary to massive inflammation (4). And also an association of rotavirus with Kawasaki disease was found about 74% (5). Seasonal-relationship with rotavirus was investigated but no relationship was found because rotavirus had seasonal but KD had year round pattern (6).

In addition to main diagnostic criteria, if fever and signs of coronary artery involvement in echocardiography are present, diagnosis can be made with less than four of the diagnostic criteria. Restlessness (50%), arthritis (15%), diarrhea (25%), vomiting (44%) coughing (28%), common cold (20%), fluid loss (30%), abdominal pain (18%) and other nonspecific symptoms like sterile pyuria, hydrops of gallbladder, myozitis, stomatitis, aseptic meningitis, cranial and peripheral nerve paralysis and hepatosplenomegaly also can be seen. A 10-month-old infant with hemiparesis and a 9-month-old infant with indurations around the BCG scar were reported. These two infants had coronary aneurisma and were evaluated as incomplete KD (7). Patients whose illness does not meet the diagnostic criteria but have fever and coronary artery abnormalities are classified as incomplete KD. Complete and incomplete forms can not be distinguished histologically. Our patient had only two main diagnostic criteria, bilateral conjunctivitis and hyperemia of lips. She had other clinical findings like sterile pyuria and diarrhea and for this reason diagnosis was delayed. But her diarrhea was not a nonspecific symptom, it was due to rotavirus.

Incomplete KD is the case with the absence of full diagnostic criteria and coronary abnormalities develop later. 10% of patients have incomplete courses. In particular <1 year of age and >9 years of age cases are characterized as incomplete cases. Therefore, KD should be considered for <1 year and >9 years of age groups when there is long-term, unexplained high fever and laboratory findings indicate an abnormal inflammation. Because of the incomplete cases, failure in diagnosing the disease at <6 months and >8 years of age was found > (50%) in general pediatricians, 25% in infectious disease subspecialists (8).
The frequency for those <6 months is 3%-11%. The disease is rare in neonatal and infant period. Only 1.6% of patients is under 90 days of age (9). In incomplete cases, coronary involvements are more common because of the delays in diagnosis. In another study coronary abnormality frequency was observed to be similar between complete and incomplete cases (31.3% vs. 37.5%) (10).

A case of a 35-day-old KD has been reported after hepatitis B vaccine (11). Also KD after vaccinations was analyzed according to the reports of the vaccine adverse event reporting system and a low relationship was found between rotavirus vaccination and KD (12).

Especially in <6 months incomplete cases, mucosal involvement, lymphadenopathy, pharyngitis, and conjunctivitis are less common. Prolonged fever and rash occurs most frequently in this age group. Fever lasts longer than the older age group. Restlessness and diarrhea are more common. Sterile pyurias, aseptic meningitis, respiratory symptoms, desquamation of the skin are more frequent. Our patient also had sterile pyuria.

The goal of the treatment is to prevent thrombosis in coronary artery walls to stop inflammation. Therefore a single dose of 2 g/kg IVIG is sufficient. In order to avoid thrombosis acetylsalicylic acid is given at a high dose of 80%-100 mg/kg/day and is decreased to antithrombotic dose at 5 mg/kg/day. Although the effectiveness of IVIG treatment is known to be less after >10th day of the fever, IVIG should be given. Therapy with IVIG <10 days reduced the frequency of coronary complications. Our case benefited from IVIG treatment although the treatment was given on day 11. But coronary aneurysm was due to the delay in diagnosis and her age group.

In conclusion, because of the non-specific symptoms in incomplete KD, delay in the diagnosis determines the prognosis. So physicians should be careful in incomplete cases, especially in infants for early and accurate diagnosis and treatment.

References