A Case of Atypical Kawasaki Disease with Abducens Nerve Palsy and Sialadenitis

Yılmazer M.M.(1), Meşe T.(1), Arı H.F.(1), Güven B.(2)
Department of Pediatric Cardiology, İzmir Dr.Behçet Uz Children’s Hospital, İzmir, Turkey (1);
Department of Pediatric Cardiology, Tepecik Training and Research Hospital, İzmir, Turkey (2)

Case: A 5-year-old boy presented with 6 days of fever > 39.0. He had bilateral non-purulent conjunctivitis and unilateral cervical lymphadenopathy on the left side. He had normal cardiovascular, neurological and gastrointestinal examination. Two dimensional echocardiography was performed, which showed fusiform dilatation in the left and right coronary arteries. The diameter of the left coronary artery (LCA) was measured to be 5.1 mm (Z-score: +6.22) and the right coronary artery (RCA) was measured to be 6 mm (Z-score: +11.20). The patient diagnosed with incomplete Kawasaki disease according to the clinical, laboratory and echocardiographic data. Intravenous immunoglobulin (IVIG) (2 gr/kg) and high-dose acetylsalicylate (ASA) (80 mg/kg) were given. Fever recovered on the sixth hour of IVIG administration. He was discharged from the hospital with low-dose ASA on the 10th day of IVIG treatment. The patient was re-admitted to our outpatient clinic due to significant submandibular swelling 17 days after the IVIG treatment. He had not fever since he was discharged from the hospital. Cervical USG showed a lymph node which was spherical, 25 x 21 mm in size. In the follow-up swelling continued in the left submandibular region and was characterized as hard by palpation. Biopsy material taken from the submandibular swelling showed that the salivary gland morphology characterized by acini and ductus structures forming the lobules. Eventually eventually the patient was diagnosed with sialadenitis. Meanwhile, 35th day of the illness the patient developed total abducens paralysis in the right eye (Figure 1). Dexamethasone treatment was initiated twice a day at 0.5 mg/kg/dose. Then dexamethasone treatment gradually reduced day 10 and was cut off on day 16. Right abducens palsy did not improved completely but coronary dilatations decreased significantly in the echocardiography. The LCA was measured to be 3.1 mm (Z-score: +1.37), and the RCA was measured to be 2.9 mm (Z-score: +2.49). Three months after the hospital discharge right abducens paralysis and submandibular swelling was regressed and the eye movements completely improved.

Conclusions: It should be kept in mind that Kawasaki disease is a multisystemic vasculitis that can effect different organs such as central nervous system, salivary glands and others.