Hemophagocytic Lymphohistiocytosis Prior to the Diagnosis of Kawasaki Disease

We read the recent case report regarding macrophage activation syndrome secondary to Kawasaki disease [1]. We report a unique case with hemophagocytosis preceding the diagnosis of Kawasaki disease.

A 9-year-old girl was hospitalized on 7th day of illness because of prolonged fever and cervical lymphadenopathy. On admission, leukopenia (total leukocyte count $1.1\times10^9/L$) and thrombocytopenia (platelet count $60\times10^9/L$) were present. Serum aspartate aminotransferase ($128$ U/L), alanine aminotransferase ($99$ U/L), lactate dehydrogenase ($518$ U/L), ferritin ($823$ mg/dL), soluble interleukin-2 receptor ($1,636$ U/mL), interleukin-8 ($371$ pg/mL) and interferon (IFN)-γ ($541$ pg/mL) levels were elevated. Triglyceride and C-reactive protein levels were normal. Coagulation studies revealed increased d-dimer levels. A bone marrow aspiration revealed appreciable numbers of hemophagocytosing macrophages. Serology for cytomegalovirus, Epstein-Barr virus and human parvovirus B19 was negative. On 2nd day of hospitalization, conjunctival injection, lip erythema and erythematous papules appeared. There was no hepatosplenomegaly. Echocardiogram showed dilation of right coronary artery. The patient was diagnosed as having Kawasaki disease complicated by hemophagocytic lymphohistiocytosis (HLH) like illness. Clinical symptoms and laboratory findings improved after the initiations of intravenous immunoglobulin (2 g/kg/dose) and flurbiprofen (4.5 mg/kg/day). Coronary artery lesion regressed at the 13th day of illness.

HLH generally complicates Kawasaki disease with prolonged or relapsing course [2]; it developing before the diagnosis of Kawasaki disease is unusual. The pattern of serum cytokines in the present patient was similar to those in virus-associated HLH [3]. Increased IFN-γ level suggests the exaggerated systemic inflammatory response to viral pathogens [3]. Although no pathogen was identified in this case, an unknown virus might have induced the symptoms.

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REFERENCES

Albendazole-induced Autoimmune Hepatitis

There are no published data on drug-induced autoimmune hepatitis caused by albendazole. We present here a patient with autoimmune hepatitis (AIH) induced by albendazole prescribed for hydatid cyst. A six-year-old girl was referred to our outpatient clinic with the diagnosis of liver hydatid cyst. Her physical examination and routine laboratory analyses were unremarkable. Albendazole treatment (15 mg/kg/day) was given for two weeks to perform puncture, aspiration, injection and re-aspiration (PAIR). But she was lost to follow up and she was admitted with abdominal pain after 2 months. AST, ALT, and GGT were 663, 800, and 92 IU/L, respectively. Laboratory investigations to exclude infectious, autoimmune, and metabolic liver disease were normal. The elevated transaminase levels returned to the normal range after cessation of albendazole. At 9th month, abdominal ultrasound revealed a progressive increase in the size of the cyst. Treatment with PAIR technique was considered. Albendazole treatment (15 mg/kg/day) was initiated again