DRESS Syndrome with Peripheral Neuropathy Due to Reactivation of Cytomegalovirus in a Child

Sir,

A 30-month-old toddler with developmental delay due to congenital cytomegalovirus (CMV) infection who was on phenytoin, phenobarbitone, and carbamazepine for epilepsy, was admitted with fever, generalized maculopapular rash, respiratory distress of 10 days with lymphadenopathy, hepatosplenomegaly, and an episode of generalized seizure (initial investigations) [Table 1]. His anticonvulsants were changed to levitracetam for suspected drug hypersensitivity, confirmed by skin biopsy [Figure 1]. His fever and rashes disappeared following intravenous immunoglobulin and oral steroid therapy. One week later, rash, fever, and respiratory distress reappeared with icterus and new onset opacities on chest radiograph. Based on repeat investigations [Table 1], reactivation of CMV with drug hypersensitivity syndrome was diagnosed. During the illness course he developed features of peripheral neuropathy which improved with risperidone and amitriptyline. Fever, lymphadenopathy, skin rash, eosinophilia, hepatic and pulmonary involvement, negative antinuclear antibody (ANA), negative hepatitis serology, and sterile blood cultures indicates a diagnosis of DRESS (Drug Reaction (or Rash) with Eosinophilia and Systemic Symptoms) syndrome based on scoring proposed by Kardaun *et al.*[1]

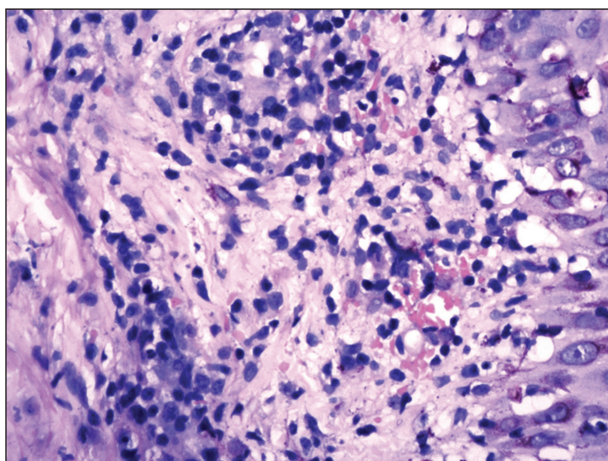

Figure 1: Skin biopsy showing parakeratitis with few dyskeratotic keratinocytes, spongiosis with prominent basal layer vacuolization, increased pigmentation reaching upto epidermis as well as incontinence into dermis, and extravasation of red blood cells. Dermis showed dense perivascular infiltrate compromising of lymphocytes consistent with drug hypersensitivity syndrome

ACKNOWLEDGEMENT

We thank Dr. Laxmisha Chandrasekhar, Associate professor of DVL, Dr. Sarthak Das and Dr. Srinivasa Raghavan, Senior residents in Pediatrics, JIPMER for involvement in management of the case.

Tanigasalam Vasanthan, Ganesan Rajaguru, Chandrasekaran Venkatesh, Parameswaran Narayanan, Reena Gulati, Pampa Ch Toi[1]

Departments of Pediatrics, and [1]Pathology, Jawaharlal Institute of Postgraduate Medical Education and Research, Puducherry, India

Address for correspondence:
Dr. Chandrasekaran Venkatesh,
E-mail: cvenkatesh@hotmail.com

Table 1: Investigations

Initial investigations	
Haemoglobin	8.2 g/dl
Total Leukocyte Count	14,000/mm^3 (lymphocyte predominant-64%)
Platelet count	330,000/mm^3
Erythrocyte Sedimentation Rate	10 mm/h
Serum bilirubin	0.6 mg/dl
Aspartate Transaminase	374 IU
Alanine Transaminase	140 IU
Serum Alkaline Phosphatase	512 IU
Chest radiograph	Suggestive of pneumonitis
Echocardiogram	Normal
Repeat investigations	
Total Leukocyte Count	9,900/mm^3 (eosinophils 8%)
Platelet count	630,000/mm^3
Prothrombin Time (International Normalized Ratio)	1.9 (corrected with vitamin K)
Blood, Cerebrospinal fluid and urine culture	Sterile
Widal	Titers not suggestive of enteric fever
Leptospira serology	Negative
Dengue serology	Negative
Paul–Bunnel test	Not suggestive of Infectious mononucleosis
Weil–Felix test	Titers not suggestive of scrub typhus
Hepatitis A, B, and C serology	Negative
Anti Nuclear Antibody	Negative
Real time polymerase chain reaction for CMV	Positive
Nerve conduction study	Reduced sensory nerve action potential in bilateral median and peroneal nerve
Nerve biopsy	Normal

REFERENCE

1. Kardaun SH, Sidorof A, Valeyrie-Allanore L, Halevy S, Davidovici BB, Mockenhaupt M, *et al*. Variability in the clinical pattern of cutaneous side-effects of drugs with systemic symptoms: Does a DRESS syndrome really exist? Br J Dermatol 2007;156:609-11.