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Images

Harlequin phenomenon in a newborn

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A baby was born via urgent cesarean section at 37 + 4 weeks of gestation, with hypoxic-ischemic encephalopathy (HIE). Amplitude-integrated electroencephalography (aEEG) and therapeutic hypothermia were initiated 2 h after birth. An episode of generalized tonic-clonic seizures occurred during the first day of life (DoL) and was treated with phenobarbital. The patient developed a transient blanchable erythematous rash on the right side (from the face to the foot) on the third DoL with a clear demarcation line separating each side of the body (Fig. 1). Simultaneously, aEEG indicated seizure activity; a midazolam bolus was injected leading to the resolution of the seizure and rash within 10 min, and no other events occurred. This cutaneous phenomenon is called Harlequin syndrome (HS),

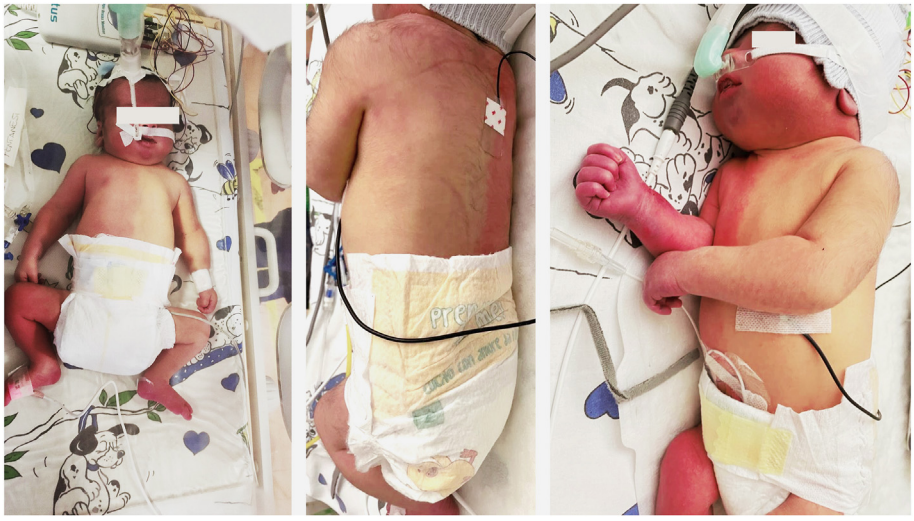

Figure 1 Harlequin phenomenon: transient blanchable erythematous rash on the right side from the face to the foot.

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which is a rare autonomic dysfunction, and extremely rare in cases of HIE. An idiopathic etiology is the more frequent cause but other possible causes are iatrogenic lesions, central nervous system infection, autoimmune disease, prostaglandin E1 or anesthetic infusion, or hypoxia [1—3]. In our case, a brain magnetic resonance image taken on Day 7 revealed multiple microhemorrhages in the falx cerebri and the subdural and subarachnoid spaces. Neurological development at 3, 6, and 12 months was normal. Thus, HS is an epiphenomenon with central nervous system involvement that needs further investigation.

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Declaration of competing interest

Mario Diplomatico, Clara Coppola and Sabino Moschella declare that they have no conflict of interest.

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