UNILATERAL ERYTHEMA IN A PREMATURE NEONATE

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A male neonate was born at 32 weeks of gestation due to a premature rupture of the membranes nine days before delivery. His birth weight was 1830 g (50th percentile), and Apgar scores were 6 at 1 minute and 8 at 5 minutes. He was admitted to the neonatal intensive care unit with respiratory distress and was placed on respiratory support with nasal continuous positive airway pressure, for the first 24 hours of life. On the third day of life, he had progressive respiratory deterioration and hemodynamic instability related to a bloodstream infection, with Klebsiella pneumoniae isolation in blood culture. Antibiotic treatment, volume expansion, and dopamine infusion were necessary to manage the septic shock. On the sixth day of life, he suddenly developed unilateral erythema with simultaneous contralateral pallor with a well-demarcated line separating the left and right sides of the body (Figure 1). During this episode, the physical examination was otherwise unremarkable, with no changes in vital signs. It resolved spontaneously within a few minutes.

What is the diagnosis?
Harlequin color change. It is a benign, idiopathic, and self-limited feature. It is characterized by a clearly demarcated color change, with one half of the body displaying erythema and the other half pallor as reported in this case. This condition has been described in up to 10% of healthy neonates and, although it has been firstly described in association with prematurity and low birth weight, it is more frequent in full-term newborns. This phenomenon’s etiology remains unexplained. The possible physio pathological mechanism described is sympathetic autonomic dysfunction in the control of cutaneous capillary tonus due to hypothalamic functional immaturity. Associated medications and conditions described to date include anesthetic agents, prostaglandin E, and central autonomic disturbance such as meningitis, and congenital, acquired, or iatrogenic brain lesions. Dysfunctional vasoreactivity in our newborn may have occurred due to multiple factors: prematurity; septic shock; hemodynamic disturbance; and vasoactive drug usage. The diagnosis is clinical and no treatment is needed. Awareness of Harlequin color change is important when reassuring worried parents.

Compliance with ethical standards
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References: