CASE REPORT

Harlequin Colour Change

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ABSTRACT

Harlequin colour change (HCC) is a rare, benign and transient colour change presenting in normal healthy neonates in initial days of life and resolves spontaneously. It is thought to be due to hypothalamic immaturity, leading to capillary bed dysregulation. Gravity might also play a role, as this is apparent on the dependent side. It is important for physicians to recognize this phenomenon so that they do not order any irrelevant investigations. Here, we present a case of a healthy neonate who had HCC, which was unilateral, well demarcated erythema, with patchy pallor. Baby was vitally stable at that time. This colour change was observed twice and each time resolved within 20 minutes of its appearance and was not observed again to date (after 20 days). Baby was discharged in a stable condition after 48 hours of life.

Key Words: *Harlequin colour change. Newborn. Premature. Infant. Dermatology.*

INTRODUCTION

Harlequin colour change (HCC) is a benign, transient condition apparent in 10% of healthy newborns.[1] This distinctive phenomenon presents as a well-demarcated colour change, with one half of the body displaying erythema and the other half pallor. It can also present in a patchy fashion sparing arms, legs, trunk, face, and/or genitalia. It usually occurs between second and fifth days of age.[2,3] It has also been reported in a 9½-month boy.[4] Episodes last for the duration of 30 seconds to 20 minutes and resolve spontaneously in cephalo-caudal fashion.[5] HCC can recur when the infant is placed on his or her dependent side. Its occurrence varies from 1 to 12 episodes in 24 hours.

The exact mechanism for HCC is unknown; but evidence exists that this colour change is due to functional hypothalamic immaturity, leading to dysregulation of regional capillary vasoconstriction and vasodilation.[2,6] Associated conditions and medications like prostaglandin E or anesthetic agents may possibly accompany and/or enhance HCC phenomenon, either by influencing the peripheral vascular tone or reactivity.[7]

CASE REPORT

A baby was born to a 37-year 7th gravida mother with no comorbid conditions. Antenatal period was uneventful. All the previous pregnancies were uneventful and all children were healthy and alive. Baby was born via elective cesarean section at 38 weeks of gestation with immediate cry and good Apgar score. Birth weight was 3.1 kg and anthropometric measurements at 50th centile. After birth, baby remained vitally stable with unremarkable head to toe systemic examination. At one hour and 30 minutes of birth, the baby developed patchy unilateral discoloration (erythema) of right side of the body. Well demarcated and non-blanching pale patches were observed on right upper limb, trunk and lower limb, not appreciable over face, back and genitals (Figures 1 and 2). Baby was vitally stable during that time with heart rate of 132/min, respiratory rate of 52/min. Baby was maintaining oxygen saturation of 98% pre-ductal and 99% post-ductal. Blood pressures were also normal in all 4 limbs and were coinciding. In cardiovascular examination, there was delayed capillary refill of about 4 seconds; but all the peripheral pulses were palpable with normal rate and volume. Apex beat was in the 5th intercostal space 3 cm medial to mid-clavicular line. Heart sounds were of normal intensity with no added sounds. There was no respiratory distress, and no recessions with bilateral normal vesicular breathing and no added sounds. Abdomen was soft and non-tender with no visceromegaly and gut sounds were audible. Neonatal reflexes were good and complete and CNS examination was also normal.

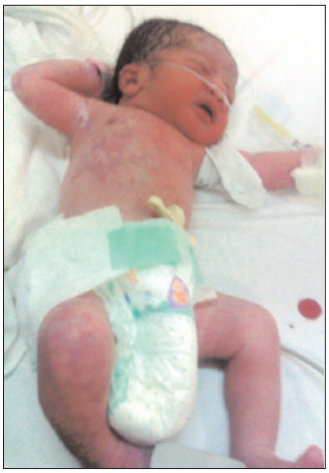

Figure 1: Baby developed erythema.

This phenomenon resolved after 20 minutes of appearance. It was observed again on second day of life as it involved buttocks and lower back. It was also observed that this discolouration was at the dependent side on which the baby was lying. Capillary refill time was 5 seconds on the toes of right lower

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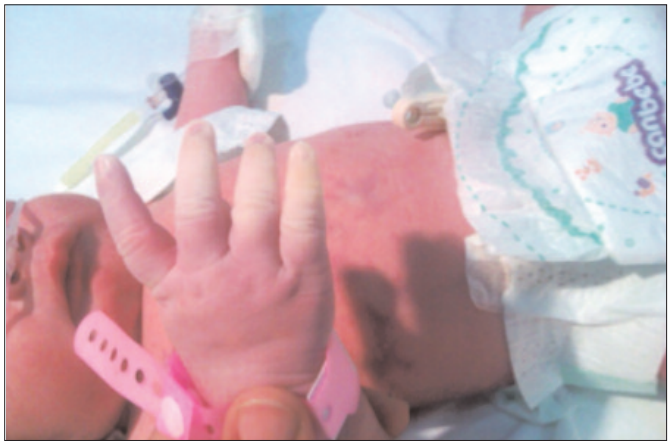

Figure 2: Pale patches on right upper limb, lower limb, and trunk.

limb. The characteristic of auto-resolution was observed as HCC resolved within 5 minutes of appearance

Baby was discharged from hospital in a stable condition after 48 hours. This phenomenon was not observed for next 20 days.

DISCUSSION

The Harlequin colour change (HCC) is described in multiple case reports, mostly in preterm neonates; but HCC has also been reported in term babies, as in our case.[6] HCC is usually observed on the dependent parts of the body. Different patterns have been identified.[8,2] HCC is mostly observed unilaterally with erythema on one side and pallor on the other; but in our case, a different pattern is identified in which the colour change is unilateral as well as patchy at the same time. Discoloration improved within minutes, but it was again apparent with a different pattern involving only lower limbs.[6] HCC is mostly observed from day 2 till day 5 of life;[3,4,6] but in this baby, it was present on first day of life. Multiple case reports are presented with this change to date; but none of them is from our part of the world.

It is important that this phenomenon, with its different patterns of colour change, should be recognized by pediatricians. Awareness is important, as lack of recognition of HCC can be overlooked or underreported. The characteristic of being present for short duration and auto-resolution can also be a cause that it might not be reported.[7]

A condition named Harlequin syndrome HS is a permanent and pathological condition of nerves which is diagnosed by skin conductance responses,[9] so it is important to differentiate HCC from HS, if spontaneous resolution is not observed.

HCC is a benign condition which is present in 10% of normal healthy newborns with no sequelae. It is observed in the initial days of life for a short duration of time and resolves spontaneously without any treatment or maneuvers. The recognition of this condition by pediatricians is important as it will save extra investigations and they will be in a better position to guide and relieve parental anxiety.

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