Unexplained Digital Gangrene In A Preterm With Neonatal Hyperviscosity Syndrome

J Okeniyi, O Adegbehingbe, I Dedeke, O Olorunnisola, T Ogunlesi, L Oginni

Citation

Abstract
We report a preterm baby of a hypertensive mother who had borderline polycythaemia but developed unexplained gangrene within 4 hours of delivery with auto-amputation of his left four medial fingers. Clinicians faced with cases of unexplained digital gangrene should consider neonatal hyperviscosity syndrome as a possible explanation.

INTRODUCTION
Polycythaemia (venous haematocrit greater than 65 percent), often associated with neonatal hyperviscosity syndrome, occurs in 0.4 – 12% of neonates especially those small for gestational age (SGA). This follows increased foetal erythropoiesis secondary to hypoxia caused by placental insufficiency in such conditions as preeclampsia, eclampsia, primary renovascular disease, abruptio placenta, maternal diabetes, smoking, cyanotic congenital heart disease and in postdate pregnancy.

Symptoms are due to hypervolaemia, abnormal blood flow kinetics due to microthrombi, decreased tissue oxygenation and metabolic derangements such as hypoglycaemia and hypocalcaemia. Most times, the presenting features are neurological. Thus, we find it imperative to report this case in whom rapid multiple digital gangrene was manifested which otherwise remained unexplained but for hyperviscosity.

CASE REPORT
On the 11th June, 2005 J.B, a low birth weight (1.5 kg), preterm (36 weeks gestation) baby was admitted into the neonatal Unit of the Wesley Guild Hospital, Ilesa, Nigeria with moderate asphyxia (APGAR scores; 4 , 5) immediately following an emergency Caesarean section indicated by preeclampsia and diminished foetal movement. His 32-year-old Para 5 + 2 , mother, a non-diabetic hypertensive with a preceding second trimester spontaneous abortion and a previous fresh stillbirth was unbooked. He was SGA, plethoric, jittery and floppy with a weak cry but adysmorphic, acyanosed and afebrile (rectal temperature was 36.5 ±C). The respiratory and cardiovascular findings were essentially normal. The liver was palpable to 3 cm below the costal margin. His limbs were normal with no amniotic bands. The only abnormalities observed at the age of 1 hour were mild polycythaemia (haematocrit 66%), profound hypoglycaemia (0.2 mmol/ L) and metabolic acidosis (bicarbonate 16 mmol/ L). Initial management included 25% then, 10% Dextrose in water infusion, intravenous Cefuroxime, vitamin K, intranasal oxygen and incubator care.

However, by age 3 hours, when his activity and cry had improved, the left 4 medial fingers had bluish discolouration and were rapidly darkening. A diagnosis of hyperviscosity syndrome was made and partial exchange blood transfusion with 30mL of normal saline was done and the haematocrit reduced to 58%. Nonetheless, by age of 24 hours, the skin overlying the fingers sloughed off and active movements were lost. X ray of the hand showed no bony abnormality. By 28 hours the little finger was completely gangrenous (figure 1). Intravenous Metronidazole was added and he had Gamgee dressing of the hand. By 48 hours, all four medial fingers were completely gangrenous although the thumb remained normal. Clotting file was normal and the blood culture yielded no growth.

On the 8th day of life, he developed fever. Blood culture yielded Klebsiella species sensitive to Ciprofloxacin only and that was given for 14 days. Repeat blood cultures yielded no growth. His hospitalisation was subsequently uneventful. He was discharged home at age 34 days weighing 2.0 kg. Figure 2 shows the baby’s affected limb at
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discharge.

Figure 1
Figure 1: The baby with digital gangrene at age 28 hours.

Figure 2
Figure 2: The baby’s affected limb at hospital discharge.

DISCUSSION
The index baby had polycythemia with moderate birth asphyxia then developed rapid digital gangrene. However, his hematocrit being not so high, gives grounds for doubt about the underlying cause of the gangrene. Nonetheless, being low birth weight, preterm and SGA, plausibly aggravated the impact of thromboembolism. The anatomic distribution of the lesions is indicative of the event occurring along the ulnar artery. Polycythemia and hyperviscosity are common neonatal problems though their symptomatology are non-pathognomonic. 1,2,3,4,5,6 Most affected babies are term or near-term, unlike our baby, a preterm. Diagnosis is largely based on hematocrit values and symptoms, 1,2,3,4,5,6 which can range from subtle to severe, and not on measures of viscosity. 1,2,3,4,5,6 Yet, hematocrits are not routinely drawn in newborn. 1,3,4,5

Most neonatologists advocate dilution transfusions when the peripheral venous hematocrit is above 70% in the absence of symptoms. 3,4,5,6 However, the severe and early digital gangrene in this case with the borderline polycythemia being the only identifiable factor questions the logic in this principle. The poor maternal obstetric history and yet lack of no antenatal care worsened the baby’s prognosis. 1,5

Anticipation and prevention improves prognosis. 1,5 Kanitkar and Gupta have advocated screening high risk babies, such as small for date, asphyxiated and infants of diabetics for polycythemia at between 6 - 12 hours of age for early diagnosis and management to prevent complications. 5

Could this child have had some undetected inherited coagulopathy? This remote possibility exists but ours is a community that lacks the wherewithal for exoteric investigation. In the face of such rapid progression, salvation of gangrenous limbs may also be impossible even in technologically advanced communities where such investigative techniques abound. We are of the opinion that unexplained digital gangrene might be due to borderline neonatal polycythemia. Thus, we believe that the benefits derivable from early and routine screen for polycythemia among high-risk neonates and prompt saline dilution transfusions desire further investigation.

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CORRESPONDENCE TO
Dr. John Akintunde Oladotun Okeniyi. Department of Paediatrics and Child Health, Obafemi Awolowo University, Ile-Ife, Nigeria. E-mail: akinyemiokes2@yahoo.com Tel: +234-803-4014280 Fax: +234-803-4014280

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Author Information

John A. O. Okeniyi
Lecturer, Department of Paediatrics and Child Health, Obafemi Awolowo University (OAU)

Olayinka Adegbhingbe
Lecturer, Department of Orthopaedics and Traumatology, Obafemi Awolowo University (OAU)

Iyabode O. F. Dedeke
Senior Registrar, Department of Paediatrics, Wesley Guild Hospital

Olumide Olorunnisola
Senior Registrar, Department of Orthopaedics and Traumatology, Wesley Guild Hospital

Tinuade A. Ogunlesi
Senior Registrar, Department of Paediatrics, Wesley Guild Hospital

Lawrence M. Oginni
Associate Professor, Department of Orthopaedics and Traumatology, Obafemi Awolowo University (OAU)