Case Report

Peripheral Symmetrical Gangrene of the Neonatal Extremities: A Case Report

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Abstract

Background
Limb gangrene in neonates is an extremely rare clinical problem and very few cases have been recorded up to now. The clinical findings were mild involvement of skin to full necrosis and gangrene of the involved regions. Unfortunately, in most cases the etiology cannot be established. However, a variety of etiological causes may account for this condition.

Case Presentation
We present a newborn baby with symmetrical peripheral gangrene of the limbs, both flanks, chin, and scrotum due to leukocytoclastic vasculitis with unknown cause. He was treated by parenteral glucocorticoid, broad-spectrum antibiotics, heparin, hydralazine and fresh frozen plasma (FFP). He died after 2 weeks of intensive treatment. There is history of the same disease in two infants of his paternal aunt, both of them were dead soon after birth.

Conclusion
Peripheral gangrene with symmetrical involvements, due to vasculitis, is a very unique disorder in newborn infant, which no case was reported up to now.

Key Words
Neonates, Leukocytoclastic Vasculitis, gangrene

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Introduction
Peripheral limb ischemia and gangrene, especially symmetrical type, occurring in the first week of life is an uncommon condition. Many etiologic factors are postulated, but the exact causes are in doubt. We report a very unusual case of neonatal symmetrical gangrene, due to leukocytoclastic vasculitis, there is a history of the same disease, which died soon after birth. His parents had no history of any significant disorder. This suggests Mendelian pattern of inheritance of the disorder in this family. Gangrene due to leukocytoclastic vasculitis is an uncommon finding at birth. The most common presentation of leukocytoclastic vasculitis is skin lesion with good response to supportive treatment. In present case, the lesions were started as erythematous macules on upper and lower limbs with unremittingly progress to fulminant necrosis and gangrene of the affected regions.

Case Report
This full-term male infant was delivered vaginally at home to a 28-years old mother. The mother had three pregnancies before. First pregnancy ended with abortion at 4-month age of gestation. In second pregnancy a healthy baby was born and he is three years old now. In third pregnancy normal baby was born, but unfortunately he died soon after birth. The Apgar score and birth weight of this baby were unknown; but weight of day five (the day of admission) was 3000 grams. He had erythematous macules on 3rd, 4th and 5th toes of both feet appeared on 2nd days of life, followed by the same lesions on hands and scrotum (Fig 1). Other regions, including flanks, chin, perioral area and buttock, also were involved, after a week (Fig 2). All lesions changed to necrosis and finally to full thickness gangrene. The baby had a high fever (39.5°C) and poor feeding from day five after birth. Laboratory study revealed hematocrit 27 per cent, WBC count 32600/µl (> 76% PMN), platelet count 28000/µl, INR 3.4, and total bilirubin 13.4mg/dl (direct =0.5), C3 and C4 were in normal range, RF Latex was negative. The baby also suffered from gross hematuria a few days before death, due to acute tubular necrosis. Histopathology of the gangrenous regions (from anterior aspect of the left leg) showed perivascular infiltration by inflammatory cells (including neutrophils, lymphocytes, mastocytes and eosinophils), suggested of leukocytoclastic vasculitis (Fig 3).

Discussion
Gangrene of the newborn is an uncommon condition resulting from decreased perfusion of a part, usually an extremity. Exact cause of newborn gangrene is unknown. Birth asphyxia, Rhesus disease, respiratory distress, severe congenital anomalies (such as congenital heart disease), infection and sepsis, dehydration, maternal diabetes, direct pressure from the maternal pelvis, arterial thrombosis, emboli, vascular involvement (such as leukocytoclastic vasculitis), venipuncture, umbilical catheterization, trauma, contraction bands, polycythemia and coagulopathies are possible causes, reported by others (1, 2, 3, 4, 5, 6, 7, 8, 9, 10, 11, 12). The treatment is general support, allowing the ischemic area to demarcate and slough, along with specific interventions in respect to cause (1, 2, 13, 14, 15, 16, 17, 18).

Conclusion
Peripheral ischemia and gangrene presenting at birth or in neonatal period is a rare clinical problem with a heterogeneous etiology, of which leukocytoclastic vasculitis is a unique etiology. Clinical presentations of leukocytoclastic vasculitis in newborn and childhood ages are very divers [19, 20, 21, 22, 23]. Literature review reveals that upper limb gangrene is more common than lower limbs [2, 24, 25], with no report of four limbs gangrene up to now.
thus it is concluded that this case, could be the first report of peripheral Symmetrical gangrene of newborn neonate due to leukocytoclastic vasculitis.

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Conflict of Interest
None

References


Fig 1: Note to symmetrical involvement of hands, feet and midline of scrotum at the first days of admission.

Fig 2: The baby after two weeks. There was obvious necrosis of perioral region, flanks, midline of abdomen, scrotum, dorsal of hands and both feet.

Fig 3: Histopathology of the gangrenous region showed pattern of leukocytoclastic vasculitis.