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## Unilateral lower limb gangrene in neonates, diagnostics and treatment challenges: A case series from a referral hospital

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**Abstract:** Lower limb gangrene is rare in neonates. A cause is frequently not identified, and management is usually directed at identifiable underlying cause, watchful waiting and ultimately amputation.

We present a series of two ill neonates with unilateral lower limb gangrene who both died within 24 hours of admission. This case series highlight the challenges of late presentation and resource constraint to the timely diagnosis, investigation management, timely referral and outcome. It also contributes to the limited literature on this topic.

**Key words:** Unilateral lower limb Gangrene Neonate, Sepsis

**Résumé:** La gangrène des membres inférieurs est rare chez les nouveau-nés. Une cause n'est souvent pas identifiable et la prise en charge est généralement dirigée vers une cause sous-jacente identifiable, une attente vigilante et finalement une amputation.

Nous présentons une série de deux nouveau-nés malades atteints de gangrène unilatérale des membres inférieurs qui sont tous deux décédés dans les 24 heures suivant leur admission. Cette série de cas met en évidence les défis de la présentation tardive, des contraintes de ressources pour les investigations, la référence précoce et l'issue. Il contribue également à la littérature limitée sur ce sujet.

**Mots clés:** Membre inférieur unilatéral, gangrène, nouveau-né, septicémie.

### Introduction

Lower limb gangrene is rare in neonates. A cause is frequently not identified, and management is usually directed at identifiable underlying cause, watchful waiting, dressing, debridement and ultimately auto-amputation or surgical-amputation. Various predisposing factors implicated include prematurity, dehydration, sepsis, umbilical artery cannulation, hypercoagulable state, pregnancy-induced hypertension, and maternal diabetes.<sup>1-5</sup>

We present the cases of two very ill neonates who presented late with unilateral lower limb gangrene and died within 24 hours. Our objective is to highlight the challenges of late presentation and resource constraint to the timely diagnosis, investigation and management of the two cases in our practice.

### Case reports

#### *Case 1: Bluish discoloration of right foot for two days*

A 7-day old male baby, weighing 3 kg, born at term to a 22-year-old primigravida mother was referred to our unit. He developed bluish discoloration of the right foot on 7<sup>th</sup> day of life. It was of a gradual onset, and

started on the sole of the foot, progressively ascending to involve the right ankle. It was associated with blister formation, and was non-tender on touch. There was no history of trauma or intravenous access prior to onset.

There was no history of diabetes in the mother during pregnancy. There was no history of umbilical catheterization or intravenous access on the affected limb. Mother had foul-smelling yellowish vaginal discharge and fever two weeks before delivery. Birth was uneventful, spontaneous vertex delivery, and baby was discharged home within 24 hours. He developed fever on the second day of life, which was associated with convulsions on the 3<sup>rd</sup> day of life, followed by limb discoloration on 7<sup>th</sup> day of life.

On examination, right foot and toes showed bluish-purple discoloration with blisters (Figure 1). Dorsalis pedis, popliteal and femoral pulses were not palpable. Investigations are shown in Table 1. He was started on intravenous antibiotics (Meropenem and Metronidazole) and care of the blisters. He succumbed on the first day of admission.

**Case 2: Bluish discoloration of right toes for one day**

A 5-day old male baby, term weighing 2.7 kg, born to a 32-year-old mother. Baby was referred to us with bluish discoloration of right toes, which started on the fourth day of life, progressing gradually from the toes to mid foot. No history of trauma or tourniquet related. There were also hyperpigmented purplish macules on his back, and a small one measuring around 1cm x 1cm on the right side of forehead.

Pregnancy and birth history were uneventful, and baby was discharged after six hours post-delivery. He developed fever on the 2<sup>nd</sup> day of life, which was associated with one episode of convulsion, followed by limb discoloration on the fourth day of life. Right toes and distal half of the foot showed bluish-purple discoloration (Figure 2). Right popliteal and femoral pulses were present, but dorsalis pedis was absent. Investigations are mentioned on Table 1, showing elevated Prothrombin time and Partial thromboplastin time. Toxoplasma IGM, Rubella IGM and VDRL were all negative. Baby could not be taken to radiology for Doppler USS in time. He was initiated on intravenous antibiotics (Tazobactam/Piperacillin and Metronidazole) and appropriate wound care. Baby succumbed within eight hours of admission.

Investigations	Case 1	Case 2
Haemoglobin (g/dl)	15	13
Total leucocyte count (x 10 <sup>9</sup> /L)	22	26
Absolute Neutrophil count (x 10 <sup>9</sup> /L)	16.19	12.13
Platelet count (x 10 <sup>9</sup> /L)	51.74	49.65
C-Reactive Protein (mg/L)	12.5	34.3
Blood culture	Sterile	Coagulase -ve Staphylococcus
CSF analysis	Sterile	Sterile
Urine culture	Sterile	-
Prothrombin time (seconds)	-	16.3
Partial thromboplastin time (seconds)	-	40.2
Doppler USS	No intraluminal thrombi, normal calibre and flow velocities in femoral artery and vein, reduced flow velocity and spectrum wave form in popliteal vein, and no signal in the tibial and dorsalis pedis artery	-

**Fig 1:** Right foot and toes showed bluish-purple discoloration with blisters



**Fig 2:** Right toes and distal half of the foot showing bluish-purple discoloration



**Discussion**

Lower limb gangrene is a rare entity in neonates. Aetiologies may include congenital bands, maternal diabetes, birth trauma, uteroarterial thrombosis, prematurity,<sup>6</sup> sepsis,<sup>7</sup> intravenous hyperosmolar infusions, umbilical artery catheterization<sup>8</sup> and hypercoagulable state. The possible cause of lower limb gangrene in the index cases is neonatal sepsis due to the presenting symptoms and investigations. The mother in Case 1 had foul smelling discharge and fever two weeks before delivery, which could have increased the risk of infection in the baby. Both cases presented with fever and at least one episode of convulsion. In Case 2, a hypercoagulable state was noted, with focus of infection not easily pinpointed.

Aetiology in majority of neonates presenting with lower limb gangrene is not ascertained.<sup>2,9</sup> Evidence of other risk factors such as diabetes and polycythaemia were not identified. SARSCoV2 could not be done because of the

sample collection process, and because both babies succumbed within 24 hours before arrangements were made. Singh et al reported an unusual case of unilateral lower limb gangrene in a 12-day neonate, who had a negative sepsis screen, normal doppler study, normal magnetic resonance angiography and absence of hypercoagulable state, making the diagnosis of idiopathic unilateral lower limb gangrene.<sup>10</sup> Challenge to ascertain aetiology was also seen by Subramanu et al in their report which lacked definitive aetiology of two neonates who presented with peripheral limb gangrene, who were thoroughly investigated.<sup>1</sup>

Management of lower limb gangrene in the neonate is conservative with systemic antibiotics and dressing with local antibiotic to control secondary infection, followed surgical intervention if severe ischaemic changes are present. If thrombosis present, heparin is used to prevent progression. Progression to spontaneous slough or

autoamputation has been seen most often.<sup>11</sup> Premature amputation should be avoided until definitive demarcation line between healthy and gangrenous area develops.<sup>1</sup>

The rapid mortality seen in our cases could be late presentation to a tertiary hospital due to late referral, which limits timely investigation to ascertain a possible cause after good history and physical examination, hence appropriate management. Case 2 stayed in our unit for only eight hours, before laboratory results to guide the management were obtained. Evaluation with doppler USS in our unit requires movement to the radiology department, which is sometimes impossible when a baby is unstable or early death, as it was seen in Case 2.

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