Case Report

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Title

Congenital ischemic loss of skin of the lower limb in a neonate: A case report

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Abstract

Background: Congenital ischemia of the limbs in a neonate is a rare condition seen in clinical practice.

Case Presentation: We report a case of a 16 hours old full term female neonate born via spontaneous vaginal delivery to a 38 years old woman who had four prenatal visits and one ultrasound scan done during the second trimester of pregnancy which was normal. The diagnosis of congenital ischemia of the left lower limb was made after physical examination. Investigations undertaken were unremarkable.

Discussion: She was managed conservatively until discharge and the outcome were favorable after six months of follow up. Similarly, in the literature, cases of limb ischemia were managed conservatively
Conclusion: Therefore, in the absence of gangrene, cases of limb ischemia should be managed conservatively like in our case.

Key words: congenital ischemia, loss of skin, lower limb, neonate, conservative, skin graft, outcome

Background

Congenital ischemia of the limbs in a neonate is a rare condition seen in clinical practice. Its commonest location is the upper limbs, though in very rare occasions the lower limbs can be affected **(6)**. It could be diagnosed by prenatal ultrasound or postnatally **(5)**. The exact cause is not known but certain etiological factors have been identified, being congenital malformations, infectious, metabolic or obstetric **(7)**. The management could be conservative but if any signs of gangrene are noticed, amputation of the limb is done. We report the case of a patient managed at our pediatric surgery unit and discuss the etiology, diagnosis and management of this rare disease

Case presentation

Prenatal and postnatal history: A 16 hours old female neonate born at 37 weeks 6 days of gestation via spontaneous vaginal delivery was referred to our unit. APGAR scores were 8/10 and 9/10 in 1 and 5 minutes respectively. She was born to a 38-year-old woman who had four prenatal visits and one ultrasound scan done during the second trimester of pregnancy with no anomalies detected. No history of maternal infection nor gestational diabetes was reported. Family history was not significant.

Clinical presentation: The neonate was conscious, not pale, afebrile, anicteric, acyanosed, and had no signs of dehydration. Vital signs were normal. Oxygen saturation and blood sugar were also within the normal range. The patient's birth weight was 3 kilograms. On local examination of the left lower limb, there was an inflammation from the inguinocrural fold to the toes, erythematous and distally covered with necrotic skin **(Figure 1)**. The contra-lateral lower limb was normal. Systemic examination was unremarkable. A diagnosis of congenital left lower limb ischemia was made.

Diagnostic workup: Full blood count (WBC 4 *10^3/mm^3, Hb 15mg/dl, PLT 300 * 10^3/mm^3, C-reactive protein (25 mg/l), Doppler echocardiography, left lower limb doppler ultrasound were all done and came out to be normal.

Management: The neonate was put on analgesics (intravenous paracetamol 15mg/kg four hourly for three weeks, antibiotics (intravenous Ampicillin 75mg/kg/day 8 hourlies for one-month, intravenous Gentamicin 5mg/kg/day for five days) and daily dressings with application of antibiotic cream (fusidic acid) for two months.

Follow up: After two months of admission under the above management, the infant was finally discharged home to be followed up in the clinic. The follow-up was favorable with presence of granulation tissue and secondary epithelialization **(Figure 2)**. She was on physiotherapy with minimal contracture on the foot as seen starting in **(Figure 2)**. However, there was no skin grafting done on the limb.

Discussion: In our case, the diagnosis was based on the physical examination at birth. The investigations were not very contributive. The patient was managed conservatively with intravenous antibiotics, analgesics with daily dressing because there was no gangrene and the outcome was favorable. Similarly, in the literature, cases of limb ischemia were managed conservatively. Intrauterine limb ischemia is considered very rare with a limited number of cases being reported in the literature **(1)**. Lower extremity gangrene is even rarer **(2)**. The incidence of limb ischemia is 1 in 4500 births per year **(3)**. Intrauterine limb ischemia must be differentiated from neonatal limb ischemia, which occurs after birth mostly due to catheterization, sepsis, or coagulopathy.
Etiological factors of limb ischemia include: Congenital malformations (Prematurity, congenital heart disease, amniotic bands, congenital arterial anomaly), infectious (Syphilis, Rubella, chickenpox), metabolic *(*maternal diabetes, thromboembolic disease, protein C deficiency, thrombocytopenia), obstetric (obstetrical trauma, umbilical cord strangulation, presentation abnormality in utero, placental infarction).
On the other hand, intrauterine upper limb ischemia may result due to intrauterine compression by amniotic bands, oligohydramnios, mal-presentation of the fetus with limb prolapse and umbilical cord entanglement, or as a result of thromboembolic events due to embolization from infarcted placental parts or thrombosis due to coagulopathies **(1)**. Antenatal diagnosis can be suggested by ultrasound evaluation which can identify foetal limb developmental abnormalities as a result of vascular disruption and potentially constricting entities such as amniotic bands **(5)**. The diagnosis can also be made by clinical observation at birth or a few hours later as in our case. Color duplex ultrasound is the first imaging test used to confirm the diagnosis **(4),** however, in our case it didn't contribute to the diagnosis. Treatment can be supportive or interventional in selected cases. The salvageability of the limb depends on the length of time between the initial vascular event and presentation. It may sometimes be difficult to evaluate this interval accurately and thus provide the most appropriate treatment. In case of ischemia, you can do conservative treatment with daily dressing and analgesics but wherein gangrene sets in, amputation is often necessary **(3,7)**. In most cases, however, the non-viable limb will be obvious, with fixed skin changes or established necrosis and in such case, amputation is required. Supportive treatment includes adequate intravenous hydration and antibiotics if infection is suspected **(5)**.

Conclusion: Intrauterine or congenital lower limb ischemia is rare. In our case, because there was no prenatal ultrasound in the third trimester, the diagnosis was made after birth and the investigations were not contributive to

the diagnosis. The patient was managed conservatively since there was no gangrene and the outcome was favorable. Therefore, in the absence of gangrene, cases of limb ischemia should be managed conservatively.

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Figure and Legend:

Figure 1. Shows a left lower limb with inflammation from the inguinocrural fold to the toes, erythematous and distally covered with necrotic skin (white arrow).

Figure 2. Presence of granulation tissue and secondary epithelialization (Black arrow).

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