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Case report

Neonatal femoral artery thrombosis at the time of birth: a case report

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Abstract

Neonatal thrombosis is a relatively common disease/condition and most often associated with predisposing genetic factors, underlying medical disorders and acquired trigger factors, such as iatrogenic interventions. Perinatal femoral artery thrombosis with no underlying risk factors has not been presented in the international literature. There has been no evidence of perinatal artery thrombosis with no underlying risk factors in the international literature.

In this case presentation, a neonate was born via normal vaginal delivery at 39^{+4} weeks of gestation, in an otherwise uncomplicated pregnancy without risk factors, with marked discoloration of right lower abdomen and right lower limb, initially pale and eventually evolving to cyanotic. At the time of birth, such clinical presentation was a challenge to clinicians regarding diagnosis. The absence of right-sided palpable femoral pulses raised suspicion and led to a diagnostic approach primarily including ultrasonography.

A right femoral blood clot blocking blood flow and resulting in the pale right lower limb was revealed via Doppler ultrasound. Screening test for thrombophilia was negative and neonatal arterial patency was achieved after administration of Low Molecular Weight Heparin (LMWH). After a

6-week course of treatment, vascular latency was fully restored and confirmed by ultrasound on day 10 post-partum. Clinicians should be aware of this unusual event that can be detrimental and endanger limb survival if immediate further action is not taken. Neonatal thrombosis can occur even at the time of birth; thus, clinicians should be alert in cases of neonates born with pale extremities. Low-risk cases could still pose a great threat to the survival of an extremity if appropriate action is not taken.

Keywords

Pallor, birth complication, benign, neonate, femoral artery thrombosis, low weight molecular heparin.

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Introduction

In a normal vaginal delivery of an uncomplicated birth with no known risk factors, a possible detrimental complication is quite uncommon. Neonatal thrombosis, affecting the femoral artery, in the absence of any known risk factors or sonographic findings of any intrauterine developmental restriction, is considered almost impossible to manifest [1, 2]. In this rare case report, clinicians initially noticed a pale skin color affecting the lower half of the body (right lower limb, and right side of the abdomen). At first sight, in the absence of risk factors, they even suggested a non-typical harlequin color change, a benign physiological transient skin change, completely harmless, which should also be properly identified to avoid initiation of unnecessary interventions and/or treatment. A few minutes later, the pale skin color evolved to bluish, affecting the neonate's limb and lower abdomen. The absence of palpable femoral pulse at the same side led to further evaluation of the vascular patency, and an unlikely femoral artery thrombosis was diagnosed.

Case report

A female neonate was delivered at term (39^{+4} gestation age) during an uncomplicated spontaneous labour, with a birth weight of 3,120 g. The mother received no medication antenatally, and maternal history revealed gestational diabetes mellitus controlled with diet. The neonate had an APGAR score of 9-10 with no further pathological findings or deformities from the rest of the clinical examination. Symmetrical development of all extremities and normal motor function were noted. At the time of birth, the neonate presented with pale right lower limb that turned into bluish color within a few minutes. The discoloration was also affecting the right side of the lower abdomen, leaving intact the rest of the neonatal skin (**Fig. 1**). Due to the disparateness of this color manifestation and the absence of femoral pulse at the right lower limb, the neonate was transferred to the Neonatal Unit for further investigation. There was absence of the right femoral pulse; the limb was slightly colder compared to the other, painless with no obvious restriction in movements. In less than an hour post-partum, the diagnosis of neonatal femoral artery thrombosis was confirmed via sonographic evaluation. The initial Doppler ultrasound revealed a hyperechoic clot in the common femoral artery (CFA), since the lumen is normally anechoic. The exact dimensions of the clot were 4 mm x 2 mm (length x width) with a diameter of the CFA of 2 mm. The clot was located precisely just before the CFA division into the superficial and the deep femoral artery (**Fig. 2**). No blood flow to the peripheral arteries was noted at the time of the initial assessment. Moreover, cranial ultrasound was normal. Blood samples for thrombophilia were taken, and administration of Low Molecular Weight Heparin (LMWH) (Enoxaparin 1.7 mg/kg BD) [3, 4] was initiated, aiming for an anti-factor Xa activity of 0.5 to 1 IU/mL.

Within 24 hours, partial clot resolution was noticed and the Doppler ultrasound at that time revealed monophonic flow in the superficial femoral artery. Interestingly, thrombophilia screening test, assessing free protein S antigen,

protein C, antithrombin III, LA1-2, LupusR 1, factor VII-IX-XI-XII-XIII, vW Ag high-med, vW activation medium and α2-antiplasmin, was negative. Molecular testing showed normal MTHFR PCR, V-Leiden PCR and FII-prothrombin. The echocardiogram was also normal. Further Doppler ultrasound check on day 10 post-partum showed complete clot resolution. Triphasic flow arterial Doppler in the CFA and the popliteal artery was present (**Fig. 3**).

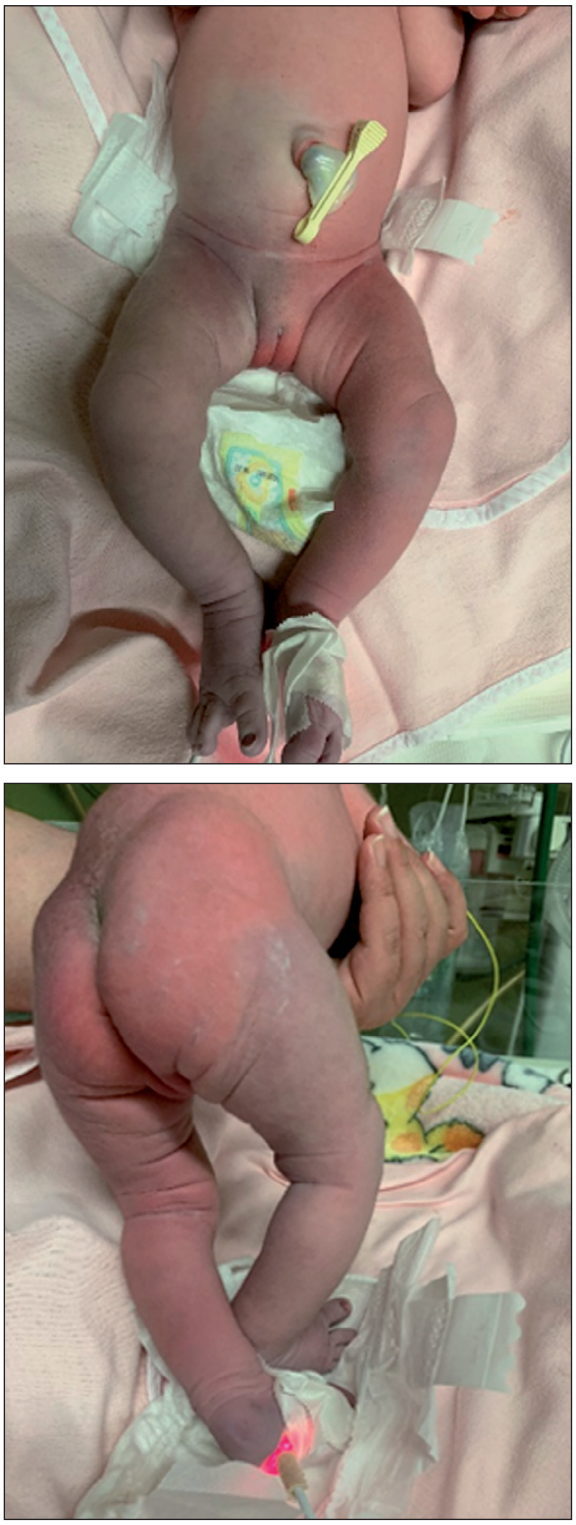

Figure 1. A. Right femoral artery thrombosis affecting the right lower limb and right lower abdomen. **B.** Different view of the neonatal extremity.

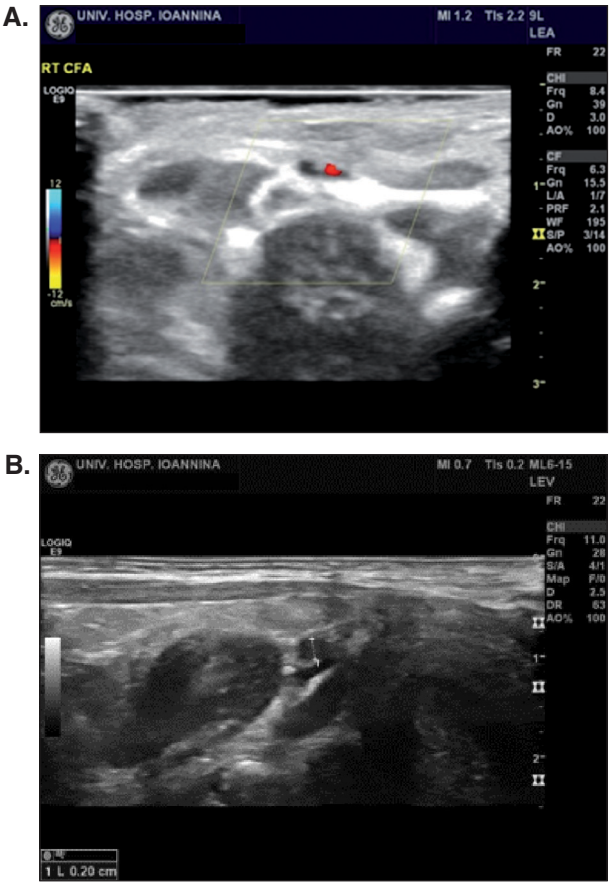

Figure 2. Sonographic images of neonatal femoral artery thrombosis. **A.** No blood flow in the right common femoral artery (CFA). **B.** Clot at the right CFA causing a blockage of the blood flow.

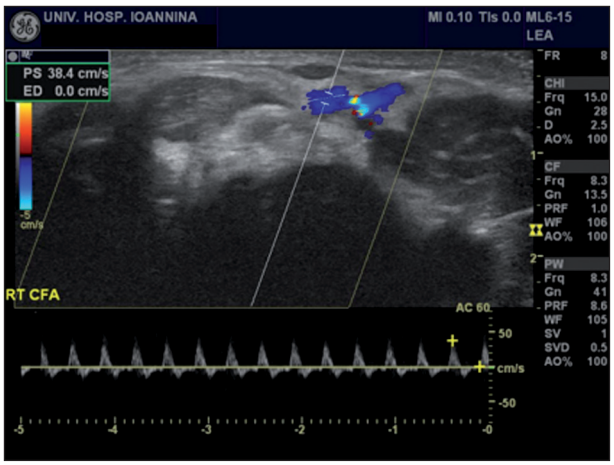

Figure 3. Sonographic confirmation of restored right common femoral artery (CFA) patency.

Discussion

In this case report, clinicians, at first sight, thought that a non-typical harlequin phenomenon could be the case, due to the unusual skin manifestation and the absence of any other risk factor or concern. Harlequin phenomenon is a rare skin color distribution presenting with demarked cutaneous erythema of one half of the neonatal body separated by the pallor of the other half [5]. This condition was first described in 1952 and has been linked with prematurity, low birth weight, maternal general anesthesia, use of prostaglandins E1, meningitis, asphyxia or intracranial injury [6]. Paradoxically, this condition is benign and resolves within 30 seconds to 20 minutes, usually presenting at day 2 to 5 of early neonatal life [7]. The presentation of this phenomenon is quite variable and is considered as a result of a temporal dysfunction of the hypothalamic center, although its cause remains unknown [8]. The diagnosis is strictly clinical and this condition also affects healthy neonates without underlying severe causes [3]. The absence of palpable pulse at the right lower limb easily eliminated this condition from the differential diagnosis.

In our case, the pallor affected the right lower limb and the right side of the lower abdomen but not the whole half of the neonatal body. The absence of femoral pulses on the same side led clinicians to a further sonographic assessment. Doppler ultrasound revealed right CFA thrombosis.

Furthermore, thrombophilia screening revealed no genetic predisposition for this rare condition at the time of birth.

Thrombosis is a disease attributed to many factors and involves genetic predispositions, underlying disorders, and acquired triggers [9, 10]. The most common risk factors for this condition include race/ethnicity, protein C, protein S, and antithrombin deficiencies. The neonatal period poses the highest risk of thrombosis throughout childhood, even without genetic predispositions [11]. Usually, thromboembolism is diagnosed as a complication of sepsis, cancer, congenital heart disease, therapy-related events of drugs, and intravenous catheters. In our cases, none of these risk factors were noted [12].

The neonate received a 6-week course of LMWH, while the arterial patency was fully restored within 10 days, according to the neonatal guidelines regarding the management of neonatal thrombosis [2, 13]. Although there are no specific recommendations regarding the use of LMWH in the treatment of thrombosis in neonates, some studies suggest that in critical cases, such as the one presented, its use might improve neonatal outcome and viability of the limb [14-16]. This is the first case reported in the literature regarding femoral artery thrombosis with no related genetic predisposition or other risk factors at the time of birth [17].

Conclusion

Thrombosis in the neonatal period is well documented in the presence of genetic predisposition, underlying disorder, or acquired trigger. Misdiagnosing this condition is detrimental. Clinicians should be cautious of the unexpected and rare event of arterial thrombosis as a cause of this manifestation. In our case, CFA thrombosis was diagnosed, which could be detrimental for the survival of the limb if it remains untreated, despite being unusual for a neonate with no risk factors or thrombophilia.

Study layout

According to CARE guidelines.

Statement of Ethics

The mother of the neonate provided informed consent for the publication of this case report. All patient data has been de-identified, and the Authors obtained the necessary approval from the Ethical Committee of the University Hospital of Ioannina.

Declaration of interest

All Authors declare no conflict of interest nor funding of any kind for this case report.

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