

Case Report

Idiopathic Upper Limb Ischemia in a Term Infant

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Introduction

Acute Limb Ischemia (ALI) is rare in neonates with overwhelming sequences that could eventually end up with limb loss. ALI may be due to intrauterine causes as in utero thromboembolic or ischemic events or in utero compression. Postnatal limb ischemia is usually a complication after umbilical artery catheter. We present a case of ALI with no apparent underlying cause that was managed conservatively.

Case Presentation

A full term baby boy was born at 41 weeks gestation. He was born by urgent Caesarean section due to fetal bradycardia. His birth weight was 3070 grams and Apgar score was 7 and 9 at 1 and 5 minutes respectively. The mother is 30 years old, primigravida. She had an uneventful pregnancy with unremarkable antenatal scans. There was no history of maternal diabetes mellitus or preeclampsia. Our baby is the product of non consanguineous pregnancy. There was no family history of thromboembolic disorders or ischemic diseases. Gross examination of the umbilical cord and placenta was normal. The patient was delivered at King Salman Armed Forces Hospital. Immediately after birth it was noticed that the baby had pale right upper limb (Figure 1), so he was admitted to our NICU. On examination, the whole upper arm was pale white, cold with delayed capillary refill (5 to 6 sec). Axillary pulse was palpable but brachial, radial

Abstract

Acute Limb Ischemia (ALI) is a rare disease in neonates with overwhelming sequences that could eventually end up with limb loss. ALI may be due to intrauterine causes as thromboembolic events or in utero compression. Postnatal ALI is usually iatrogenic after umbilical artery catheter. We present a case of acute upper limb ischemia with no apparent underlying cause that was managed conservatively.

Keywords: Neonates; Acute limb ischemia; Nitroglycerine patches

and ulnar pulses were not felt. He had symmetrical Moro reflex with full range of spontaneous motion of the right upper arm with intact grasp reflex and pain sensation. Oxygen saturation by pulse oximeter was not detected on the affected limb but was normal on the other limbs. X-ray of the chest and right upper limb showed no fracture or dislocation of the cervical vertebrae. Bedside Duplex ultrasound of the right upper limb done by vascular surgeon showed an evidence of good blood flow in the axillary artery down to brachial artery, with no blood flow in the radial and ulnar arteries. Baby was managed as per our NICU protocol which included; decent hydration, prevention of hypothermia and Nitroglycerin patch 5mg applied over the palmar aspect of the right arm and forearm every 2 hour. After the first two hours, the patient's right arm colour and perfusion down to the elbow greatly improved with good pulsation at the brachial artery (Figure 2). The right forearm and hand colour remained pale, cold with delayed capillary refill. Vascular team recommended starting heparin infusion as prophylaxis against possibility of thrombosis. However, Hematology team did not agree with this recommendation because of the high risk of bleeding beside there was no evidence of thrombosis. After parent counselling we decided to continue the same management rather than to start heparin therapy. Surprisingly progressive improvement of forearm color and perfusion down to the

hand occurred within next few hours. Hand color and perfusion was back to normal within 24 hours (Figure 3). Arterial doppler done at age of 48 hours revealed patent arterial circulation with evidence of good blood flow in the axillary artery down to the radial and ulnar arteries.

Laboratory investigations showed normal Complete Blood Count (CBC) values including platelet and hemoglobin. Normal renal, liver function and the coagulation profile. Echocardiography showed no evidence of congenital heart disease. Protein C and S level were also normal. Follow up of the baby at the age of two weeks and one month showed good perfusion in the right hand with good pulsation down to radial and ulnar arteries with preserved motor function (Figure 4).



Figure 1 :



Figure 2 :



Figure 3 :



Figure 4 :

Discussion

ALI is rare in neonates with an incidence of 0.5 per 10000 live births. It has devastating sequences as limb loss or reduced limb growth [1]. The exact underlying cause is usually unknown. It could be due to thromboembolic and ischemic events [2]. Neonates generally have increased risk for thrombotic and bleeding tendency as the concentrations of clotting and fibrinolytic factors are lower compared to children and adult [3]. Predisposition for neonatal thromboembolic and ischemic events could be aggravated by maternal factors as maternal lupus, diabetes mellitus or neonatal factors as birth asphyxia, sepsis, dehydration, polycythemia or cardiac anomalies associated with poor cardiac function [4]. In-utero mechanical compression, amniotic band or umbilical cord loops could be also additional risk factors. Iatrogenic factors after birth are related to arterial or venous catheter or direct birth trauma [5]. There was no evidence of the above mentioned risk factors in our case.

Antenatal diagnosis of limb ischemia is quite challenging. Ultrasound evaluation may show fetal limb abnormalities as a result of amniotic band causing vascular disruption of the af-

affected limb. Duplex ultrasound assessment of limb after birth is non invasive diagnostic tool [6]. In our case, bed side Duplex ultrasound of the right upper limb showed an evidence of good blood flow in the axillary artery down to brachial artery, while no blood flow in the radial and ulnar arteries. Follow up duplex revealed patent arterial circulation down to radial artery. The dramatic improvement of the limb perfusion after conservative management including application of nitroglycerine patches raised the suspicion of arterial spasm rather than thrombosis as the underlying cause of ALI in our case.

Proper management of limb ischemia depends on early diagnosis and identification of the underlying etiology. There is no proven standard protocol for the management of ALI. Initial management depends on the severity of limb ischemia; supportive therapy as adequate hydration, initiation of antibiotics if sepsis is suspected and topical nitroglycerine should be started promptly [7]. Nitroglycerine patches as vasodilator has been used to relief vasospasm associated with umbilical and peripheral arterial catheters [8,9]. Milrinone is a Phosphodiesterase-3 (PDE3) inhibitor that has emerged as a novel option for management of limb ischemia. Boyd et reported one case of ALI due to thrombosis of the right subclavian artery that was managed successfully with milrinone infusion [10]. The use of heparin infusion or low molecular weight heparin therapy is limited to significant thrombosis to prevent extension of the clot [10]. Interventional management is reserved for selected cases that do not respond to supportive therapy [2].

Conclusion

Early diagnosis and management of limb ischemia is essential to preserve limb function.

References

1. Lim S, Javorski MJ, Halandras PM, Kuo PC, Aulivola B, Crisostomo P. Epidemiology, treatment, and outcomes of acute limb ischemia in the pediatric population. *J Vasc Surgery*. 2018; 68: 182-8.
2. Aslam M, Guglietti D, Hansen AR. Neonatal arterial thrombosis at birth: case report and literature review. *American Journal of Perinatology*. 2008; 25: 347–52.
3. Veldman A, Nold MF, Michel-Behnke I. Thrombosis in the critically ill neonate: incidence, diagnosis, and management. *Vascular Health and Risk Management*. 2008; 4: 1337–48.
4. Edstromand CS and Christensen RD. Evaluation and treatment of thrombosis in the neonatal intensive care unit. *Clinics in Perinatology*. 2000; 27: 623–41.
5. Zetlitz E, Weiler-Mithoff E, Turner T. Idiopathic neonatal ischemia in the upper limb: the role of the microsurgeon. *Am J Perinatol*. 2008, 25: 513-6.
6. Arshad A, McCarthy MJ: Management of limb ischemia in the neonate and infant. *Eur J Vasc Endovasc Surg*. 2009; 38: 61-5.
7. Downey C, Aliu O, Nemir S, Naik-Mathuria B, Hatef DA, et al. An algorithmic approach to the management of limb ischemia in infants and young children. *Plast Reconstr Surg*. 2013; 131: 573–81.
8. Vasquez P, Burd A, Mehta R, Hiatt M, Hegyi T. Resolution of peripheral artery catheter-induced ischemic injury following prolonged treatment with topical nitroglycerin ointment in a newborn: a case report. *J Perinatol*. 2003; 23: 348e50.
9. Kamar R, van Vonderen JJ, Lopriore E, Te Pas AB. Nitroglycerin for severe ischemic injury after peripheral arterial line in a pre-term infant. *Acta Paediatr*. 2013; 102: e144–143.
10. Boyd S, Shah V, Belik J. A novel role for milrinone in neonatal acute limb ischemia: successful conservative treatment of thrombotic arterial occlusion without thrombolysis. *BMJ Case Rep*. 2019; 12: e232440