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Upper Limb Ischemia in Preterm Infant Leading to Autoamputation of Hand – a Case Report

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ABSTRACT

Acute limb ischemia(ALI) in a paediatric patient is a rare condition but may result in limb loss and life long complications. We present here a case of a 34 weeks premature infant, with birth weight 1530 grams, who was having respiratory distress at time of birth. Infant was referred to advanced pediatric center for further management, where he was kept in neonatal ICU. He was put on C-PAP and oxygen support and an infant feeding tube for feeding. On 21st day infant developed ischemia of the right upper limb. Color Doppler flow study was done which revealed thrombosed right radial and ulnar artery and biphasic flow in brachial artery. He was put on low molecular weight heparin and was advised referral to higher center for further management. Patient developed dry gangrene of right hand and wrist resulting in the autoamputation of the hand and wrist after a period of 15 day from development of gangrene. In order to prevent such complications there should well trained staff to detect early ischemic changes in the limb so that timely treatment can be started and the long term chronic complications can be prevented.

Keywords: Premature infant, Acute limb ischemia, Gangrene, Complications.

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INTRODUCTION

Acute limb ischemia (ALI) is a potentially catastrophic event that can lead to death of nerve and then muscle tissue within 4 to 6 hours unless the limb is revascularised.[1] ALI is rare but potentially devastating condition in the paediatric population which could be iatrogenic, post traumatic and is rarely secondary to arterial occlusion disease as in the adult population. [2] Most common consequences of iatrogenic arterial occlusion can be gangrene and limb loss. [3] There are few published guidelines related to management of arterial occlusion in premature sick infant. [4] In case of incomplete ischemia where there is no loss of tissue or less neuromuscular changes, a trial of non operative management can be attempted. [5] Several series of reports have demonstrated successful management of pediatric acute limb ischemia with non-operative measures. [6-7] With the recent advances of medication and endovascular surgery, there has been decrease in morbidity, limb loss (amputation), mortality and hospital stay [7]. In upper limb most common consequences of iatrogenic injury may be gangrene and limb loss. In general there is agreement that early diagnosis and therapeutic intervention helps in reestablishment of limb perfusion.

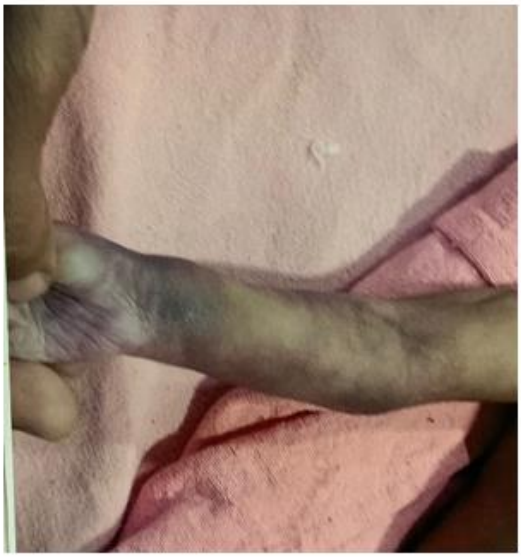

Figure 1-Initial stage of ischemia

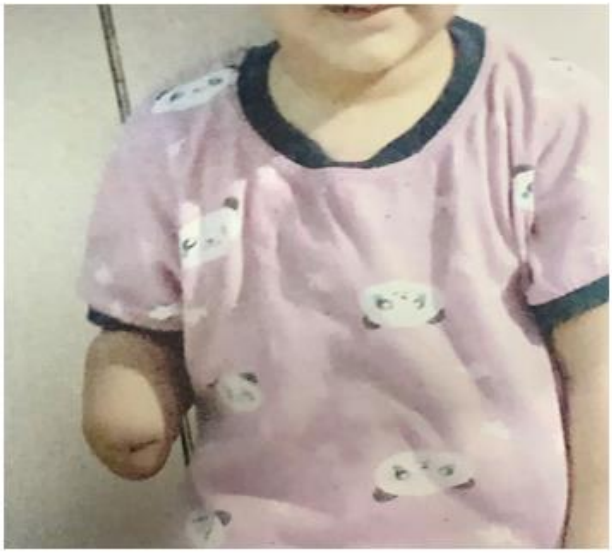

Figure 4-autoamputaion

Figure-2 Demarcated gangrene Right upper limb

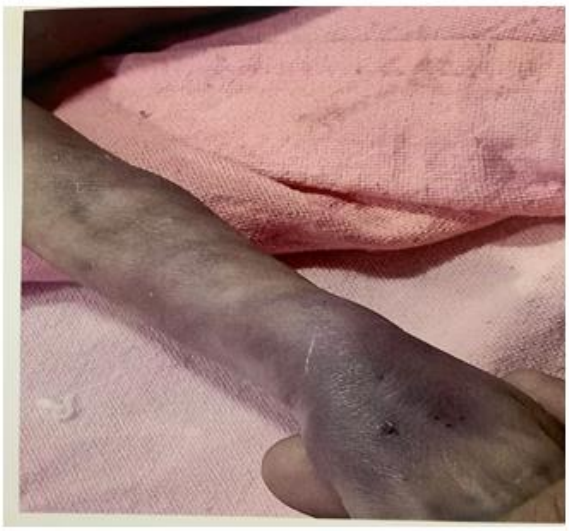

Figure 3- multiple pricks at cannulation site

CASE REPORT

A 34 week premature infant with birth weight 1530 was delivered at a private nursing home and developed respiratory distress. He was shifted to NICU of a private paediatric center within two hours of birth.

On further evalution it was found that he was anemic (Hb-6gm), RR=68/m, HR=140/m, SpO_2=98%. He was lethargic and was immediately put on C-PAP and oxygen support. Ryles tube feeding along with I/V line was started. He was put on inotropes and antibiotics coverage was given. On 5th day he had an episode of epileptic fit and was put on ventilator support as he did not maintain SpO_2.He had increased levels of serum bilirubin=13.5 mg i.e. physiological jaundice for which he was put under phototherapy.

On 21st day child developed blanching of right upper limb as per history from hospital staff followed by development of bluish discoloration of the right forearm and hand (Fig.1)

He was put on Low molecular weight heparin(injection enoxaparin1.5 unit S/c BD) and was referred to Paediatric department of Guru Nanak Dev Hospital Amritsar care center. On examination child was sick and febrile. His upper limb was cold, clammy and there was no response to external stimulation. There were scattered, gangrenous changes and multiple pricks over the dorsum of the hand as in Fig.2 Pricks were also noticed in the right cubital region due to insertion of cannula for I.V line. Radial and ulnar pulse were not palpable. Brachial pulse was feeble. He was advised Color Doppler flow study and his routine investigations were done Hb=10.3 gm, TLC=18640, CRP=17.7, Platelet 10,000, Blood urea=20 mg.

Color Doppler study revealed absence of flow in ulnar and radial artery, biphasic flow in axillary and brachial artery with PSV 20-25 cm/s. He was put on LMW heparin (Injection Enoxaparin 3-unit S/c BD), Inj. Amphotericin-B, Inj. Colistin 25000 I/V. Opinion of plastic surgeon was taken and he was referred to the PGI Chandigarh for further management. In the meantime, child developed dry gangrene of the right hand (Fig.3)

At PGI Chandigarh he was treated by Plastic Surgeon, Vascular Surgeon and Pediatrician. Color Doppler flow was repeated and found that ulnar and brachial artery had low flow but there was no flow in radial artery.

As advised by Pediatric Surgeon, dry dressing of the right gangrenous hand was carried. On 7th day, child developed autoamputation of the right hand just above wrist joint (Fig.4). At PGI Chandigarh child was put on Inj. Enoxaparin 1.5 mg S/c BD and Aspirin 5 mg (one sachet daily) alongwith antibiotics and B-complex and calcium preparation. Child was discharged under satisfactory condition.

DISCUSSION

Acute limb ischemia in pediatrics is rare and may lead to loss of limb as a life long complication.In this case, the child developed right upper limb ischemia and it resulted in autoamputation of right hand and wrist.

The causes of acute upper limb ischemia in pediatric cases may be due to congenital reason; iatrogenic and traumatic injury. Iatrogenic causes are common in infant but traumatic injuries leading to ALI is more common in children and adolescents. [8] In some cases, it may be due to intrauterine ischemia. [9] In situ thrombosis or thromboembolism may arise from inborn error of the coagulation cascade. Oligohydramnios and amniotic band may cause, extrinsic compression of extremity arteries that leads to necrosis or contraction. In most cases unintentional cannulation in the arterial system in upper limb for Intra venous administration of medication has been reported. [10]

The pathophysiological mechanism of peripheral gangrene remains unclear. Many theories have been put forward. All concepts agree with the mechanism of peripheral under perfusion due to endarteritis and thrombosis leading to tissue necrosis. [10,11]

Accidental intra-arterial injection occurs more commonly in the antecubital fossa. [10] where

ulnar, radial and brachial arteries are superficial and easily entered and, in most cases, arterial cannulation involved the radial artery and its branches of forearm and hand. Injuries to superficial ulnar arteries also have been reported in 2 to 3% of patients. [12] In iatrogenic intra-arterial injection, pain is often the initial symptom. [10] Other sign and symptoms are blanching, pulselessness, tingling, numbness, cyanosis, paresis of muscles of involved part, oedema of limb. Necrosis and gangrene are permanent functional deficit that may occur as a complication. [10,13]

In our case, also it is suspected that injury of radial, ulnar artery may have occurred inadvertently that had produced the sign and symptoms as mentioned in the studies.

Color Doppler flow study is the investigation of choice for detection of flow in the arterial and venous system. It helps in diagnosis and even to know the prognosis of the patient. In our case the initial color Doppler flow study revealed absence flow in radial as well ulnar artery and biphasic flow in brachial artery.The thrombosis of radial and ulnar artery of the right forearm and hand that had resulted in the ischemia followed by cool, clammy, gangrenous changes in the limb. Fig 2. But after treatment with LMW heparin, there was biphasic flow in ulnar and brachial artery, except radial artery in which there was complete occlusion.

In children, smaller limbs and relatively underdeveloped limb muscle mass require less oxygen and may result in improved tolerance to hypoxemia. In children, vessels being non diseased and typically unscathed by atherosclerosis, the redirection of blood flow is more facile from the intact blood vessels to the ischemic body part through ischemia -induced vasodilatation mechanisms. Furthermore, angiogenesis of collateral vessels starts early in pediatric patients. Matos et al demonstrated fully developed arterial collaterization within 24 days after femoral artery thrombosis. [9]

Based on pathophysiological features of pediatric limb ischemia. [7] The use of conservative management of pediatric ALI has been supported by many reports/studies e.g. Kayssi et al[8] and Matos et al. [5] Kayssi et al[8] described 151 pediatric patients with ALI. The majority of patients (91%) had iatrogenic. Injury secondary to catheterization. Treatment was started with LMW Heparin. Similar Matos et al[5] studied 12 infant limbs with acute ischemia, 11 patients were due to iatrogenic injury from catheterization. All the patients were treated with low molecular weight heparin. A total of 63% of ALI was successfully managed conservatively.

In our case also the patient was put immediately on LMW Heparin(Injection Enoxaparin 1.5-3.0 S/c BD) along with Aspirin sachets once daily. As a result of this although initially the whole right forearm and hand was affected but due to heparinsation, flow was restored in the ulnar artery, thereby increasing the revascularization of the affected part. Only hand and wrist underwent gangrenous changes and finally autoamputation of the part occurred (Fig.3).

Surgical management of pediatric vasculature is technically challenging. This is due to smaller area of crossection of the vessels in infant/children as compared to the adult population. So whenever injured vessel experiences vasospasm, blood flow distal to damaged vessel will be vastly affected and surgical treatment is even more complicated. Similarly in our case the patient being premature and surgical correction was not possible, so he was managed conservatively.

In term of overall management, the infant population with acute limb ischemia appears to be a higher risk group. This risk increases if there is associated comorbid or congenital diseases which further affect the surgical procedures, making conservative management the most appropriate option.

Further the most important aspect in the management is early diagnosis and treatment. In our case, there was delay in referring the patient to higher Centre and in the meantime, gangrenous changes had already set in. Although patient was put on low molecular

heparin but the patient did not recover and as a result autoamputation of the affected part occurred.Fig.4

Further it is suggested that ancillary staff involved in cannulation or catheterization in pediatric population should have through knowledge of the site and technique to insert cannula. They should have knowledge of the anatomy of vessels and their anomalies in the cubital region and the wrist area. They should be well trained in early detection of ischemic changes resulting due to iatrogenic injuries in the pediatric population to avoid long term chronic complications. [14]

CONCLUSION

Acute upper limb ischemia in pediatric population is a rare entity and is associated with auto amputation and high mortality rates. Timely, diagnosis and treatment helps in avoiding long term complications. Conservative treatment may be considered as initial treatment modality for most pediatric acute limb ischemia. Open surgical revascularization is not very effective in pediatric population and may not help in decreasing the rate of amputation. To avoid iatrogenic injuries in the pediatric population, the paramedical staff should be well trained in cannulation and should be aware with anatomy and anomalies of circulation in the limbs.

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