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The Roles of the Microsurgeon and Anesthesiologist in the Treatment of Neonatal Upper Extremity Arterial Thrombosis: A Case Report

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Abstract

We present a rare case of a newborn with spontaneous, non-iatrogenic arterial thromboembolism in the right brachial artery and accompanying ischemic changes to the right upper extremity, who was successfully treated with microsurgical intervention and ultrasound-guided infraclavicular brachial plexus block with a continuous infusion of ropivacaine for 48 hours. This case report highlights the emerging role of both the microsurgeon and anesthesiologist in management of spontaneous neonatal arterial thromboembolism.

Keywords

neonate; thromboembolism; microsurgery; nerve block

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INTRODUCTION

Arterial thromboembolism (TE) in a newborn is a rare event. While over 90% of reported cases are iatrogenic, secondary to arterial catheterization,^{1,2} spontaneous, non-iatrogenic, idiopathic TE in the newborn is exceedingly uncommon and infrequently described in the literature. The few previous reports of TE in infants suggest avoidance of surgical intervention and advocate thrombolytic and anticoagulation therapy. We present a case of a newborn with non-iatrogenic, spontaneous arterial TE in the right brachial artery and accompanying ischemic changes to the right upper extremity (RUE), who was successfully treated with microsurgical intervention to remove the thrombi and an ultrasound-guided infraclavicular brachial plexus block with a continuous infusion of ropivacaine to treat vasospasm. At 15-month follow-up, the infant's RUE was completely healed with excellent perfusion, limited wrist extension, but excellent overall, function.

Written consent was obtained from the patient's family for the publication of this case report.

CASE REPORT

A male neonate was born vaginally without complications to a 24 year-old, G3P0 mother at 39–6/7 weeks. Maternal and antenatal histories were unremarkable. Apgar scores were 5 and 9 at one and five minutes, respectively. At delivery, however, the neonate's RUE was pale and cyanotic, with non-palpable pulses distal to the elbow. Given the concern for thrombus, the infant was transferred to the Neonatal Intensive Care Unit (NICU) at our institution for further evaluation. On examination upon arrival at our institution, he had no palpable or dopplerable right brachial, radial, or ulnar pulses. The RUE was ischemic, mottled, and pale, without capillary refill, and cool to touch (Figure 1). His fingers were flexed, with lack of spontaneous movement below the elbow. Ultrasound revealed a completely obstructing intra-arterial thrombus in the distal axillary artery; the radial and ulnar arteries could not be visualized. A transcranial ultrasound was performed and revealed no evidence of hemorrhage or infarct. Additionally, an abdominal aortic ultrasound was performed and revealed no evidence of thrombus. Vascular Surgery was consulted, and anticoagulation with alteplase and heparin was initiated, with a target PTT of 80 seconds. Plastic Surgery was consulted 7.5 hours after birth and the infant was taken emergently to the operating room (OR) for RUE revascularization. Heparin was initiated prior to surgery with an initial bolus and continuous infusion of 28 units/kg throughout the surgery.

Intraoperatively, there was an abrupt color change in the arm and in the brachial artery at the transition point of the dopplerable signal. An arteriotomy was performed at this location and revealed a yellow irregular, solid material, measuring 16 mm in length, emanating from the lumen (Figure 2). After thrombectomy and arteriotomy repair, perfusion improved with a slow wave of pink progressing distally to the hand. While performing fasciotomies, the hand again became pale and mottled leading us to perform two additional arteriotomies that revealed platelet thrombus proximal to the brachial artery bifurcation and another from within the midportion of the brachial artery. At the completion of the case, there was a strong dopplerable signal at the radial artery at the wrist and palmar arch. The patient

remained on continuous, therapeutic, systemic heparin at a dose of 28 units/kg and was monitored carefully in the NICU. PTT was monitored closely, every 4 hours, with a target value of 80 seconds.

On day of life (DOL) 2, approximately 30 hours after the revascularization procedure, the RUE became cool, edematous, tense, and mottled. There was a palpable hematoma at the right lateral chest near the axilla. The infant was urgently taken back to the OR, and a subcutaneous hematoma was evacuated, but the color and perfusion of the hand continued to fluctuate throughout the case. Intraoperative angiogram revealed patency of the brachial, ulnar, and radial arteries but with evidence of vasospasm. Papaverine and 4% lidocaine were applied topically to the arteries. At the end of the procedure, perfusion varied despite continued anticoagulation, likely related to vasospasm.

Due to the poor appearance and concerns for vasospasm, the Anesthesia team was consulted to perform a RUE peripheral nerve block to improve perfusion by vasodilation. On DOL4, we performed an ultrasound-guided infraclavicular block on the patient in the supine position after withholding heparin for 2 hours. The patient was on continuous sedation and mechanical ventilation during the procedure. After proper sterilization of the block area and proper surgical draping, the brachial plexus at the infraclavicular level was located using the 12Mhz ultrasound probe. The probe was placed inferiorly and perpendicular to the clavicle to identify the pectoralis major and minor muscles, with the axillary artery and vein seen transversely. The brachial plexus cords, located surrounding the artery laterally, medially, and posteriorly, were identified. We then used the ultrasound probe to guide a 20G Touhy needle toward the posterior cord and delivered 1ml of 0.2% ropivacaine (0.45mg/kg) while observing the spread of the anesthetic solution, which separates the nerve from the artery. The needle insertion point was just inferior to the clavicle, and the needle was aimed toward the posterior aspect of the axillary artery. After the initial bolus dose, a 24G catheter was threaded through the Touhy needle, and a continuous infusion of 0.5ml/hour of 0.2% ropivacaine was administered for 48 hours. After catheter placement, heparin was restarted one hour later. Dopplerable pulses were detected hours after nerve block placement and infusion. The appearance of the RUE continued to improve after initiation of ropivacaine, with consistent palpable pulses. Because the patient achieved the vasodilatory effects of the nerve block on DOL6, or 48 hours after the catheter was placed, the decision was made to remove the catheter at this time. In order to remove the catheter, heparin was held 2 hours prior and restarted 2 hours following removal.

Upon discharge on DOL29, the patient was sent home on therapeutic enoxaparin dosing. During his hospital course, a battery of tests and imaging revealed no congenital heart disease, hypercoagulable state, or other significant medical problems. The patient displayed finger movement for the first time at his 2-month follow-up visit. He had no episodes of vascular compromise, and his enoxaparin was discontinued after an unremarkable complete, hematologic work-up at 3 months of age. Over the course of the following 4–5 months, the patient developed significant scar contractures, resulting in the inability to fully extend the wrist and elbow. To improve his function, at 8 months of age he underwent forearm flexor tenolysis and RUE scar contracture release with excision and transposition (Z-plasty closure) (Figure 2B and 2C). A complete list of the patient's surgical procedures is listed in Table 1.

At 15 months of life, the patient has maintained excellent perfusion. Although he has restrictions in wrist extension, he overall has excellent RUE range of motion and function (Figure 3).

DISCUSSION

Peripheral arterial TE in a newborn is rare and requires prompt intervention to salvage the limb and avoid necrosis and subsequent amputation. Knowledge of the coagulation system profile of the newborn can aid in determining the proper course of treatment. Neonatal blood is distinct from that of adults. In the newborn, procoagulants, such as vitamin K dependent factors and contact factors, are <70% of adult values, whereas Factor V, VII, VIII, and von Willebrand's factor are >70% of adult values.³ Additionally, coagulation inhibitors such as antithrombin III, protein C, and protein S are low in the newborn.³⁻⁵ These values largely contribute to hemostasis in the newborn, but slight variations in the concentrations of these factors can predispose the newborn to thrombus or hemorrhage.³

The unique profile of a newborn's blood contributes to the decision to intervene with non-surgical approaches, using thrombolytic and anticoagulation therapy, and less commonly with surgical intervention. The majority of the literature recommends against surgical intervention due to the infant's young age, small blood vessel size, and potentially unknown diagnoses related to the thrombosis.^{2,6} On the other hand, caution must be taken in the use of heparin or enoxaparin in a newborn, largely due to the blood profile. At the Hospital for Sick Children in Toronto, 177 pediatric patients received enoxaparin prophylactically or for treatment purposes. Of these children, 4% suffered major bleeding and 17% suffered minor bleeding.⁷ Although enoxaparin and heparin are determined "efficacious" and "safe" in pediatric patients, these findings warrant attention, and physicians must take caution with the use of anticoagulation and thrombolytic therapy in a newborn. Surgical intervention is conventionally taken at the point of amputation. However, in the event of severe ischemic changes or the failure of anticoagulation/thrombolytic therapy, surgery is indicated.^{6,8} Therefore, there is a fine balance between surgical intervention and medical therapy, based on small blood vessel site and the chance for re-occlusion of the vessels. Although anticoagulation is the mainstay of treatment, significant consideration needs to be taken for the side effects of bleeding and the need for close monitoring of anticoagulation. In our case surgical intervention was pursued given the prolonged ischemic time and reports describing early surgical intervention for successful limb salvage and restoration of function.⁹

We present a case in which the patient's management was not complete without the combination of surgical intervention, anticoagulation therapy, and a brachial plexus nerve block. During the procedure on DOL2, the fluctuating appearance of the arm and the intraoperative findings were consistent with arterial spasm. Usage of local anesthetics and papaverine intraoperatively improved the vasospasm, but the fluctuating perfusion continued postoperatively. The administration of a continuous ropivacaine brachial plexus nerve block was essential for this child's limb salvage as it provided vasodilatory effects. We leveraged the sympathectomy-like effects of the nerve block, which not only led to venous dilatation, but also prevented arterial spasm, both of which improved blood flow in our patient significantly. Additionally, the availability of ultrasound enables precise location of nerve

and vascular structures to deliver local anesthetic solutions by direct visualization of needle and the local anesthetic spread.

The decision to utilize a brachial nerve plexus block in the newborn requires multiple considerations relative to systemic absorption, metabolism, excretion, and toxicity profiles. In particular, aminoamide local anesthetics are primarily metabolized by CYP450 enzymes in the liver, whereas the aminoesters are predominantly metabolized by pseudocholinesterases in the periphery. The CYP450 system and phase II enzymes are immature in the newborn, and therefore have markedly reduced metabolic capacity.¹⁰ Additionally, neonates have larger relative amounts of extracellular and total body water volumes, and the adipose tissue has an increased proportion of water.¹¹ Therefore, more hydrophilic local anesthetics that gain access to the systemic circulation will distribute into the peripheral tissues and effectively decrease the plasma concentration.¹¹ This effect is less pronounced with more lipophilic local anesthetics.¹¹ Moreover, local anesthetic systemic toxicity is of great concern as local anesthetics have the potential to damage any cell with sodium channels, most notably the neurons, cardiac myocytes, and skeletal myocytes. Toxicity due to systemic absorption of local anesthetics is a serious complication and care must be taken to avoid intravascular injection. The unique anatomic and physiologic profiles of the neonate and the local anesthetic drug profile must be carefully evaluated in the decision to utilize a peripheral nerve block in the neonate.

Our case strongly suggests consideration of a peripheral nerve block to improve tissue perfusion and prevent arterial spasm in complicated surgical cases such as this. Additionally, we reveal that microsurgical intervention can be beneficial and successful in the treatment of arterial TE in infants in select cases.

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Figure 1. Preoperative photographs of the infant's right upper extremity at 7 hours of life. Note the line of demarcation of ischemia beginning at the elbow and extending distally to the flexed fingers.

Figure 2. Intraoperative intra-arterial findings during the first arteriotomy on day of life (DOL) 1. Thrombus emanating from the brachial artery (yellow arrow, *A*) was irregular in shape and measured 16 mm (*B*).

Figure 3.

Photographs of the infant's right upper extremity (RUE) at 5 months (*A* and *B*) and 15 months (*C*) after initial surgery. Note the extremity is well perfused in all images, and that flexion of the right wrist and digits is present (*A*). He developed scar contractures in his RUE (*B*), but these were released with scar excision and Z-plasty closures at 8 months after initial surgery. At 15 months, the scar is well healed with markedly less contracture (*C*).

Table 1

Complete list of patient's operations

Day of Life	POD from initial surgery	Procedure
1	N/A	R brachial artery thrombectomy and embolectomy R carpal tunnel release R forearm fasciotomies R brachial artery arteriotomy in 3 locations Vein patch to R brachial artery Angiogram of RUE
2	1	R hand fasciotomies Evacuation of hematoma from RUE Angiogram with microsurgical repair of access site Placement of Integra on volar and dorsal forearm
4	3	Placement of brachial plexus nerve block catheter
6	5	Removal of brachial plexus nerve block catheter
27	26	Complex closure of RUE wound from axilla to elbow Full-thickness skin graft (FTSG) from right side of abdomen to dorsal and volar open wounds on forearm Primary closure of abdominal donor site
34	33	Dressing change to RUE wounds and R abdominal wall Noted failure of FTSG to the volar and dorsal forearm wounds
40	39	Dressing change to RUE wounds
46	45	Dressing change to RUE wounds and R abdominal wall
60	59	Dressing change to RUE wounds and R abdominal wall Treatment of hypergranulation tissue on R abdominal wall with silver nitrate
243	242	Scar contracture release with scar excision and Z-plasty closures Revision carpal tunnel release of R arm Guyon's canal release of R arm Flexor tenolysis of FDS to index, long, ring, and small finger at the level of the forearm and wrist Flexor tendon tenolysis of FCR and FCU Neurolysis of median nerve

Abbreviations:

DOL- day of life

POD- post-operative day

FTSG- full thickness skin graft

FDS- flexor digitorum superficialis

FCR- flexor carpi radialis

FCU- flexor carpi ulnaris