



Successful Non-Operative Management of Idiopathic Acute Limb Ischemia in a Pediatric Patient

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ABSTRACT

Introduction: Acute limb ischemia (ALI) is a rare occurrence in children, predominantly associated with trauma or iatrogenic causes. Acute limb ischemia can result in the loss of limbs and lifelong complications. The ideal treatment is not yet established. **Case:** A 10-month-old boy was referred with darkening of the extremities, accompanied by fever and seizures. The patient had a continuous fever for the past four days and experienced four generalized seizures, 1-2 minutes each. During treatment at the previous hospital, the nose and fingers of all extremities became dark blue since one day before referral. Upon arrival, examination revealed bluish-purple extremities, a harder texture, and cold acral areas. Pulses in the extremities were weak. Complete laboratory tests showed D-dimer >20,000 ng/mL, CRP 21.6 mg/dL. Head MRI was within normal limits. Doppler ultrasound of the extremities did not reveal thrombosis in the leg veins, and echocardiography also did not show the presence of thrombus. The patient was treated with antibiotics and anticoagulants for 10 days. During hospitalization, the patient showed clinical and laboratory improvement. **Discussion:** Acute limb ischemia in pediatric patients is uncommon, typically associated with trauma or iatrogenic factors. In this pediatric case, the etiology of acute limb ischemia (ALI) remained unclear, as no embolic source or thrombotic occlusion was detected on Doppler ultrasound or echocardiography. This highlights the limitation of the initial diagnostic work-up, since advanced imaging such as CT angiography would be required to further delineate potential underlying vascular abnormalities or occult causes that might not be evident on standard modalities.

Keywords: Acute limb ischemia, case report, non-operative treatment, pediatric.

ABSTRAK

Pendahuluan: Iskemia tungkai akut jarang terjadi pada anak-anak, terutama terkait trauma atau iatrogenik. Iskemia tungkai akut dapat mengakibatkan kehilangan anggota tubuh dan komplikasi seumur hidup. Pengobatan yang ideal belum ditetapkan. **Kasus:** Laki-laki 10 bulan dirujuk dengan keluhan perubahan warna pada ekstremitas, disertai demam dan kejang. Pasien mengalami demam terus-menerus selama empat hari terakhir dan mengalami empat kali kejang umum, masing-masing selama 1-2 menit. Selama di rumah sakit sebelumnya, hidung dan jari-jari semua ekstremitas menjadi biru tua sejak satu hari sebelum dirujuk. Saat tiba, ekstremitas berwarna biru-keunguan, tekstur lebih keras, dan area akral dingin. Denyut nadi ekstremitas lemah. Hasil tes laboratorium lengkap menunjukkan D-dimer >20.000 ng/mL, CRP 21,6 mg/dL. MRI kepala dalam batas normal. Ultrasonografi Doppler ekstremitas tidak mengungkapkan adanya trombosis vena tungkai, ekokardiografi juga tidak menunjukkan adanya trombus. Pasien dirawat dengan antibiotik dan antikoagulan selama 10 hari. Selama rawat inap, pasien menunjukkan perbaikan klinis dan laboratorium. **Diskusi:** Iskemia tungkai akut pada pasien pediatrik jarang terjadi, biasanya terkait dengan trauma atau faktor iatrogenik. Pada kasus pediatrik ini, penyebab iskemia tungkai akut (ALI) belum dapat diidentifikasi dengan jelas, karena tidak ditemukan sumber emboli maupun oklusi trombotik pada pemeriksaan USG Doppler maupun ekokardiografi. Hal ini menunjukkan keterbatasan pemeriksaan awal, sehingga dibutuhkan pencitraan lanjutan seperti CT angiografi untuk menyingkap kemungkinan adanya kelainan vaskular atau penyebab tersembunyi yang tidak tampak pada modalitas standar. **Charolina Margaretha, Rachmanto Heryawan, Nusarintowati, Yehezkiel Nathanael.** Keberhasilan Penatalaksanaan Iskemia Tungkai Akut Idiopatik pada Pasien Pediatrik.

Kata Kunci: Iskemia tungkai akut, laporan kasus, pengobatan non-operatif, pediatrik.



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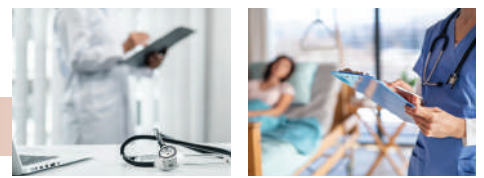
INTRODUCTION

Acute Limb Ischemia (ALI) is a rare occurrence in children, predominantly associated with trauma or iatrogenic causes.¹ Acute limb ischemia can result in the loss of limbs and lifelong complications. The ideal treatment

is not yet definitively known, but one of the therapies is non-invasive therapy.² While acute limb ischemia is well-documented in adult populations, pediatric cases are infrequent,³ leading to limited data and experience in managing this condition in young children.

The rarity of the condition in pediatric patients poses significant diagnostic and therapeutic challenges. Early recognition and timely intervention are crucial to prevent severe outcomes, including limb loss and permanent disability.⁴

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This case report discusses a rare instance of acute limb ischemia in a 10-month-old child presented with systemic symptoms such as fever and seizures, which are uncommon initial presentations for this condition. The patient was successfully managed with non-invasive therapy, specifically anticoagulants, leading to a positive therapeutic outcome. The report highlights the diagnostic challenges, discuss the management strategies employed, and underscore the importance of a multidisciplinary approach in treating acute limb ischemia in pediatric patients.

Etiology

The etiology of acute limb ischemia in infants can be multifactorial, often involving both congenital and acquired factors. Congenital causes may include vascular malformations, thromboembolic events, or genetic predispositions that affect blood clotting mechanisms. Acquired causes are frequently related to iatrogenic factors, such as complications from catheterization, cannulation, or extracorporeal membrane oxygenation (ECMO) procedures. The delicate vascular system of neonates, combined with the high-risk nature of many neonatal interventions, makes this population particularly vulnerable to ALI.^{5,6}

Clinical Presentation

The clinical presentation of Acute Limb Ischemia (ALI) in infants can vary depending on the severity and duration of ischemia. Common signs include pallor, coldness, and decreased pulses in the affected limb. Infants may also exhibit pain, though it can be challenging to assess due to their inability to communicate. Delayed capillary refill time and mottling of the skin are also critical indicators. In severe cases, cyanosis and gangrene may develop, necessitating urgent intervention.⁷

Diagnosis

Acute limb ischemia (ALI) in pediatric patients is a rare but critical condition that requires prompt recognition and management. However, its presentation can be similar to other conditions, making accurate diagnosis challenging. Diagnosis of ALI in infants requires a high index of suspicion, particularly in neonates with known risk factors. Doppler ultrasound is the primary imaging modality used to assess blood flow in the affected limb and identify the location and extent of

the occlusion. In some cases, angiography may be required to provide a detailed map of the vascular anatomy and guide therapeutic decisions. Laboratory tests to evaluate coagulation status and inflammatory markers are also essential in understanding the underlying etiology and guiding treatment.^{8,9}

Differential Diagnosis of Acute Limb Ischemia in Pediatric Patients

1. Compartment Syndrome

Compartment syndrome occurs due to increased pressure within a closed muscle compartment, leading to decreased blood flow and ischemia. It presents with severe pain, pallor, paresthesia, and pulselessness, similar to ALI. However, the presence of tense compartments and pain out of proportion to the injury often helps distinguish it from ALI.¹⁰

2. Vasculitis

Pediatric vasculitis, including conditions like Kawasaki disease and Henoch-Schönlein purpura, can lead to limb ischemia due to inflammation of blood vessels. Symptoms include pain, swelling, and purpura, which can be confused with ALI. Laboratory findings such as elevated inflammatory markers and specific serologies help differentiate vasculitis from ALI.¹¹

3. Deep Vein Thrombosis (DVT)

While DVT primarily involves venous rather than arterial occlusion, it can cause limb swelling, pain, and cyanosis, which might be mistaken for ALI. Duplex ultrasound showing venous thrombus distinguishes DVT from arterial occlusion in ALI.¹¹

4. Embolic Events

Embolic events, such as those originating from congenital heart defects or endocarditis, can cause sudden limb ischemia. This can be mistaken for ALI due to the similar presentation of sudden pain and pulselessness. Echocardiography and imaging studies help identify the embolic source.¹²

5. Trauma

Traumatic injuries, including fractures and dislocations, can lead to vascular compromise mimicking ALI. A detailed history of injury and imaging studies are essential to differentiate between trauma-induced vascular injury and primary ALI.¹²

Management

Management of ALI in infants involves both medical and surgical approaches. Initial treatment focuses on restoring perfusion to the affected limb, often through the administration of anticoagulants such as heparin. In cases where thrombolysis is ineffective or contraindicated, surgical intervention, including thrombectomy or bypass surgery, may be necessary. Postoperative care is crucial and includes monitoring for signs of reperfusion injury, which can lead to further complications such as compartment syndrome. Long-term follow-up is essential to assess limb function and address any residual deficits.¹³

Initial Assessment and Stabilization

The management of acute limb ischemia (ALI) in children begins with the immediate assessment of the patient's cardiovascular status, focusing on stabilizing vital signs and ensuring adequate oxygenation. Early identification and intervention are critical to prevent irreversible tissue damage. The initial evaluation includes a detailed history, physical examination, and the use of Doppler ultrasound or angiography to confirm the diagnosis and assess the extent of ischemia.¹⁴

Medical Management

Once the diagnosis is confirmed, anticoagulation therapy is typically initiated to prevent further thrombus formation and to stabilize the existing thrombus. Heparin is the most commonly used anticoagulant, often administered as a continuous intravenous infusion. The dose of heparin is carefully titrated based on regular monitoring of coagulation parameters such as activated partial thromboplastin time (aPTT). In cases where heparin is contraindicated or ineffective, low molecular weight heparin (LMWH) or direct thrombin inhibitors may be considered.¹⁴

Thrombolysis

In selected cases, especially when there is a significant clot burden or when immediate restoration of blood flow is necessary, thrombolytic therapy may be used. Agents such as tissue plasminogen activator (tPA) are administered either systemically or via catheter-directed infusion directly into the thrombus. Thrombolysis is generally more effective when initiated early but carries a



risk of bleeding, particularly in the pediatric population, so careful patient selection and monitoring are essential.¹⁵

Surgical Intervention

When medical management is insufficient, or in cases of extensive ischemia where rapid intervention is required, surgical options are considered. Surgical thrombectomy, where the thrombus is physically removed from the vessel, is one approach. Alternatively, bypass surgery may be necessary in cases where the affected artery is extensively damaged or occluded. The choice between thrombectomy and bypass surgery depends on the location and severity of the ischemia, the underlying cause, and the overall condition of the child.¹⁶

Postoperative and Long-term Care

Postoperative management includes continued anticoagulation, close monitoring for signs of reperfusion injury, and management of any complications such as compartment syndrome. Long-term care involves regular follow-up with vascular specialists, physiotherapy to optimize limb function, and ongoing assessment for any recurrence of ischemia or late complications, such as growth disturbances in the affected limb. Lifelong anticoagulation may be required

in cases with a persistent prothrombotic tendency.¹⁷

Prognosis

Prognosis for infants with ALI depends on the timeliness of intervention and the underlying cause of ischemia. Early diagnosis and prompt treatment are associated with better outcomes, including the preservation of limb function and avoidance of amputation. However, delays in treatment or severe ischemia can result in significant complications, including limb loss and long-term disability. Continuous monitoring and rehabilitation are often required to ensure optimal recovery and developmental outcomes.^{18,19}

CASE

A 10-month-old male presented to the hospital with darkening of the extremities, accompanied by fever and seizures. The patient was referred from a private hospital after four days of continuous fever and multiple seizures. The seizures occurred four times, each lasting between 1–2 minutes and affecting the entire body.

During the initial treatment at the referring hospital, it was noted that the patient's

nose and fingers had turned dark blue a day before the referral. Upon arrival at our facility, the examination revealed bluish-purple extremities, hardened texture of the affected areas, and cold acral regions (**Figure 1**). Pulses in the extremities were weak.

Complete laboratory tests showed: hemoglobin (Hb) 9.3 mg/dL, leukocytes 11,970/ μ L, platelets 7,000/ μ L, D-dimer >20,000 ng/mL, and C-reactive protein (CRP) 21.6 mg/dL. Additional supporting tests include head MRI with returned normal results, Doppler ultrasound of the extremities did not reveal thrombosis in the leg veins and echocardiography which also did not show the presence of thrombus.

Diagnosis was based on clinical presentation, laboratory findings, and imaging results, indicating acute limb ischemia potentially complicated by sepsis and disseminated intravascular coagulation (DIC).

The patient was promptly started on antibiotic therapy (cefotaxim injection 200 mg/every 12 hours) and anticoagulants (Heparin drip 3500 units diluted in 50 mL of 0.9% NaCl at a rate of 2 mL/hour). Anticoagulants were administered to manage the ischemia and prevent further thrombotic events, despite the absence of visible thrombosis in the Doppler ultrasound (**Figure 2**).

Throughout 10-day hospitalization, the patient showed remarkable clinical and laboratory improvement. The fever subsided, and no further seizures were reported. The color and temperature of the extremities gradually returned to normal, and the hard texture of the affected areas softened.

Follow-up laboratory tests indicated improvement in hemoglobin levels, normalization of leukocyte and platelet counts and a decrease in D-dimer and CRP levels. A few days after hospital discharge, the patient returned for follow-up evaluation. On examination, the previously darkened discoloration of the extremities had markedly diminished and the skin had nearly returned to its normal color, as illustrated in **Figure 3**. The patient was also afebrile, without fatigue or other complaints. According to the caregiver, the child appeared more active, with a noticeable improvement in appetite.



Figure 1. Extremities upon initial admission to the hospital.



Figure 3. At outpatient care.



Figure 2. Clinical improvement after anticoagulant therapy.

Discussion

This case highlights several important aspects of managing acute limb ischemia in pediatric patients. The initial presentation with systemic symptoms such as fever and seizures posed a diagnostic challenge, emphasizing the need for a thorough examination and consideration of ischemic conditions even in the absence of typical risk factors. The viability of the affected limb was confirmed by Doppler examination, which demonstrated preserved perfusion consistent with Rutherford category I. According to current guidelines, such findings justify the use of systemic anticoagulation with heparin as the first-line management, without the immediate need for invasive revascularization procedures. This conservative approach aligns with the principle of minimizing procedural risks in pediatric patients while ensuring adequate restoration of tissue perfusion.¹⁹

In this case, empirical antibiotics were administered due to the presence of persistent fever and systemic inflammatory response at admission. Pediatric patients with acute limb ischemia are particularly vulnerable to secondary infections, including sepsis, which may exacerbate tissue ischemia. Early initiation of antibiotics helped to control potential infectious triggers and reduce systemic inflammation, thereby supporting recovery

and preventing complications. The use of anticoagulants in this case underscores their importance in managing acute limb ischemia. Anticoagulant therapy was initiated because laboratory findings demonstrated markedly elevated D-dimer levels, suggesting ongoing coagulation and thrombus formation, even though no definitive thrombus was detected by Doppler or echocardiography. The use of anticoagulants, such as heparin, is recommended to stabilize the coagulation cascade, prevent extension of microthrombi, and preserve limb perfusion. In this patient, anticoagulation contributed to the resolution of ischemic changes without the need for invasive intervention.²⁰

Serial monitoring of D-dimer and C-reactive protein (CRP) provided valuable information about the dynamic course of the disease. D-dimer is a sensitive marker of fibrin degradation and indicates ongoing thrombotic activity, whereas CRP reflects the degree of systemic inflammation. The initial extreme elevation of D-dimer (>20,000 ng/mL) and CRP (21.6 mg/dL) supported the decision to continue anticoagulation and antibiotic therapy. Progressive normalization of these markers paralleled the patient's clinical improvement and served as a reliable guide for therapy duration and discharge planning.²¹

Acute limb ischemia (ALI) in infants is a rare but serious condition that can lead to significant morbidity and mortality if not promptly diagnosed and treated. In this pediatric case, the etiology of acute limb ischemia (ALI) remained unclear, as no embolic source or thrombotic occlusion was detected on Doppler ultrasound or echocardiography. This highlights the limitation of the initial diagnostic work-up, since advanced imaging such as CT angiography would be required to further delineate potential underlying vascular abnormalities or occult causes that might not be evident on standard modalities.²²

The favorable outcome in this patient can be explained by several factors. First, the absence of underlying comorbidities such as atherosclerosis or diabetes, which are known to worsen the prognosis of acute limb ischemia. Second, the patient was diagnosed early and received prompt anticoagulation therapy, which reduced thrombus progression. Third, serial monitoring of coagulation and inflammatory markers (such as D-dimer and CRP) enabled close therapeutic guidance. These factors are consistent with previous reports, which have shown that pediatric patients with idiopathic ALI may achieve good outcomes with conservative management when treated promptly.²³

CONCLUSION

Acute limb ischemia in infants is a critical condition requiring immediate attention. Advances in neonatal care have improved the prognosis for affected infants, but challenges remain in early diagnosis and management. Ongoing research is needed to better understand the risk factors, optimize treatment protocols, and improve long-term outcomes for these vulnerable patients. Acute limb ischemia, although rare in pediatric patients, should be considered in cases of unexplained extremity discoloration and systemic symptoms. Early diagnosis and prompt management with anticoagulants can lead to favorable outcomes. This case demonstrates the importance of a multidisciplinary approach in the diagnosis and management of pediatric acute limb ischemia, providing valuable insights for clinicians in similar scenarios.



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