

Transverse Tibial Cortex Transport in the Treatment of Kawasaki Disease

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Abstract: Kawasaki disease is a systemic primary vasculitis of medium-sized vessels, predominantly affecting children, whose diagnosis and management in adulthood remain challenging, particularly in the presence of rare peripheral vascular manifestations. Severe ischemic limb lesions in adult patients with sequelae of the disease have limited therapeutic options, with amputation often being indicated. This study reports the first case described in the literature of the use of Transverse Tibial Cortex Transport (TTT) as a limb-salvage strategy in an adult patient with Kawasaki disease and critical lower limb ischemia. A 38-year-old female patient, with a history of spontaneous resolution of the disease in childhood, evolved with severe chronic ischemia, a trophic lesion with bone exposure, absence of a distal runoff bed, and failure of conventional clinical therapies. Given the impossibility of revascularization and a prior indication for amputation, TTT was performed. A significant clinical improvement in perfusion was observed, evidenced by increased ankle pressure, reduced capillary refill time, progressive wound healing, and recovery of functional ambulation. Follow-up computed tomography angiography demonstrated neovascularization and the development of collateral circulation previously absent. TTT proved to be a viable and effective alternative for limb preservation in this pioneering case of Kawasaki disease, suggesting potential application in other primary vasculitides associated with critical ischemia. Prospective studies are needed to better define its indications, pathophysiological mechanisms, and long-term outcomes.

Keywords: Systemic Vasculitis; Kawasaki Disease; Surgical Amputation; Ilizarov Technique.

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1. Introduction

Kawasaki disease (KD) is defined as a primary vasculitis (PV) of medium-sized vessels, with a systemic pattern and idiopathic etiology [1–3]. Despite the wide range of signs and symptoms involved in KD, the diagnosis is based on clinical criteria, requiring the presence of persistent fever for more than four days, associated with at least four of the five classic features: bilateral nonpurulent conjunctivitis; oral mucosal changes (such as oropharyngeal hyperemia, fissured lips, and strawberry tongue); polymorphous rash; extremity changes (erythema and edema of the hands and feet, with periungual desquamation in the subacute phase); and cervical lymphadenopathy, usually unilateral, with a lymph node ≥ 1.5 cm [1, 4, 5].

Once the diagnosis of KD is established, the risk of resistance to intravenous human immunoglobulin (IVIG) is estimated using one of several available scoring systems. Unfortunately, in clinical practice, these scores have limitations, and many patients ultimately require more aggressive therapies that are not initially recommended, given that KD is the most prevalent primary vasculitis in children [4, 6]. Clinical resistance to IVIG is defined as persistent fever lasting more than 36 hours after completion of two weeks of therapy. In KD, there is no tumoral, traumatic, or drug-related etiology, as observed in some secondary vasculopathies [7–10].

Among the multiple factors involved in diagnostic and therapeutic failure, coronary lesions (CL) and peripheral vascular lesions (PVL) stand out, as they are not included among the classic diagnostic criteria. In addition, although coronary aneurysmal lesions are not considered diagnostic criteria, some algorithms allow the use of glucocorticoids in these children [1, 11]. PVL are rare and may be identified in patients who were not adequately treated in childhood and subsequently developed sequelae of KD. In such cases, during adulthood, clinical therapies analogous to those used for other primary vasculitides are recommended [1, 12]. Surgical approaches are exceptional, both in pediatric KD and in adulthood. Conventional revascularization procedures are not standardized in these patients, which has encouraged the search for alternative techniques, such as Transverse Tibial Cortex Transport (TTT) [13, 14].

TTT has already been used in some secondary vasculopathies, such as thromboangiitis obliterans related to smoking and vasculopathies associated with diabetes mellitus; however, there are still no reports in the literature describing the use of this technique in KD [13, 15]. Some authors suggest that TTT promotes the formation of a new bone matrix through osteotomy associated with traction by a metallic external fixator. The formation of this bone matrix is thought to be related to angiogenesis and neovascularization, resulting in improved perfusion of the affected limb [13, 14].

Here, we report the first case described in the literature of the use of Transverse Tibial Cortex Transport as a limb-salvage strategy in an adult patient with Kawasaki disease and critical lower limb ischemia, in the setting of impossibility of conventional revascularization.

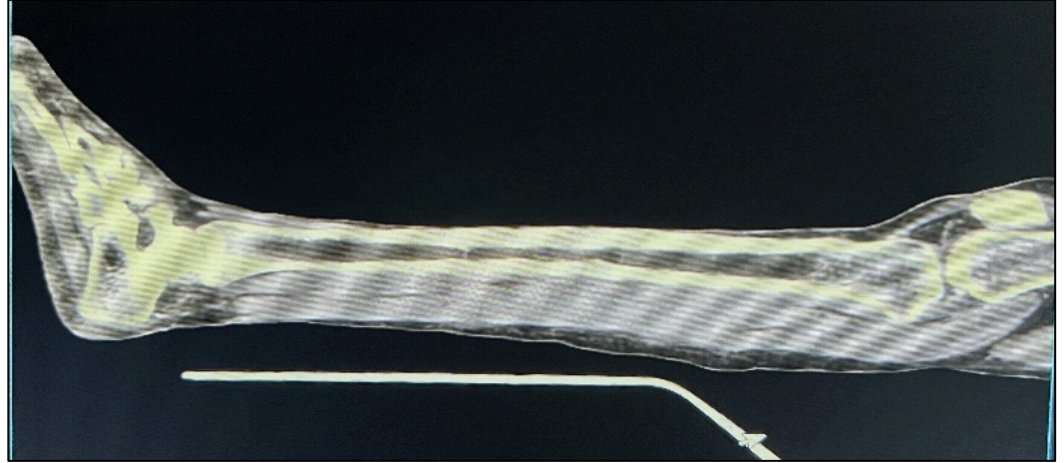
2. Case Report

A 38-year-old female patient was referred to the Santo André Municipal Hospital Center (CHMSA) due to therapeutic failure related to Kawasaki disease (KD). With a history of KD and spontaneous resolution of the disease in childhood, she evolved in adulthood with lower limb ischemia associated with heart failure. Physical examination revealed a distal trophic lesion with bone exposure in the left lower limb, fixed cyanosis of the forefoot, a capillary refill time (CRT) of 5 seconds at the heel, ankle pressure (AP) of 42 mmHg with an ankle-brachial index (ABI) of 0.38, a marked thermal gradient, an 8-cm area of erythema, and a left ventricular ejection fraction of 38% on echocardiography, consistent with New York Heart Association (NYHA) functional class III [16].

After optimization of clinical therapy, improvement in the ejection fraction to 52% was observed; however, there was worsening of the trophic lesion in the left lower limb and intensification of rest pain, refractory to the use of analgesics, broad-spectrum antibiotics associated with prednisone, and antiplatelet therapy. Computed tomography angiography (CTA) and magnetic resonance angiography did not reveal coronary lesions.

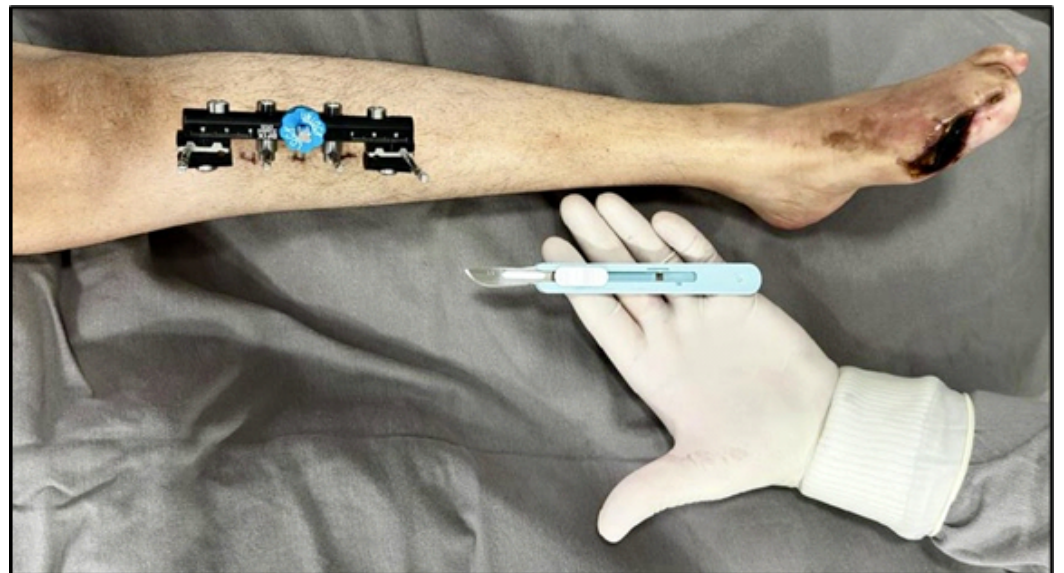
Radiological studies, Doppler ultrasonography, and lower limb CTA demonstrated absence of compensatory collateral circulation consistent with chronic ischemia (Figure 1), associated with complete absence of a distal runoff bed. Aware of previous attempts at surgical debridement in other medical services and given the impossibility of revascularization due to the lack of a distal bed, the decision was made to perform Transverse Tibial Cortex Transport (TTT).

Figure 1. Computed tomography angiography performed after cardiologic compensation (LVEF = 52%) and prior to the surgical approach with TTT demonstrated occlusion of the popliteal artery and absence of contrast opacification of the crural arteries (posterior tibial, peroneal, and anterior tibial arteries).



After 29 days, due to infection of the necrotic tissue with the presence of purulent secretion attributed to increased tissue perfusion, surgical debridement was performed (Figures 2 and 3) without removal of the metallic device (external fixator).

Figure 2. TTT in progress, with surgical isolation of the procedure to prevent contamination by necrotic tissue measuring approximately 9.4 cm (a centimeter-scaled scalpel was used as a reference for the extent of necrosis).

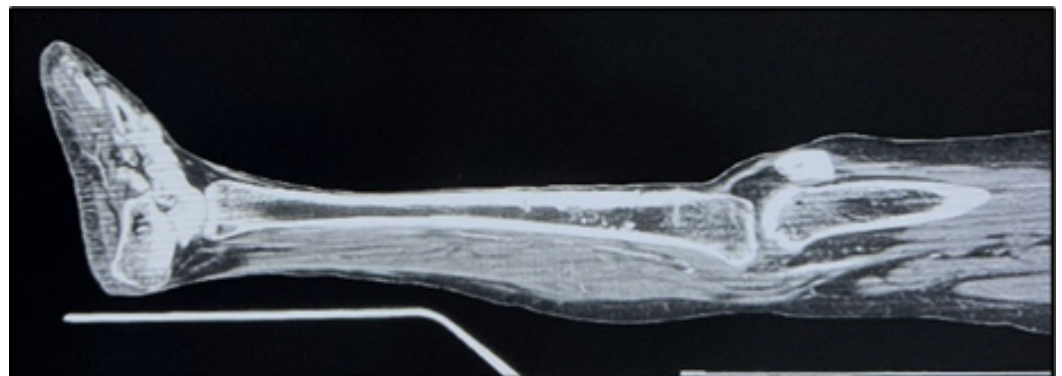


The patient began ambulation with the aid of a walker from the 23rd postoperative day after TTT. The metallic fixator was removed on an outpatient basis 44 days after its placement. On physical examination, ankle pressure (AP) was 68 mmHg, the ankle-brachial index (ABI) was 0.68, and the surgical wound was in the process of healing. Independent ambulation without a walker was achieved six days after fixator removal. Follow-up computed tomography angiography, performed after removal of the metallic device, revealed significant changes in the crural arteries and collateral circulation that had not been present prior to TTT (Figure 4).

Figure 3. Postoperative aspect after surgical resection of devitalized and infected tissue (wet necrosis), demonstrating good perfusion of the plantar fascia and surgical bed, with the presence of bleeding, a finding not observed in previous procedures.



Figure 4. Computed tomography angiography performed 49 days after surgical removal of the TTT demonstrated visualization of the popliteal artery. Absence of complete contrast opacification of the crural arteries (posterior tibial, peroneal, and anterior tibial arteries) was observed.



3. Discussion and Conclusion

Although anatomopathological (AP) examination does not constitute a diagnostic criterion for Kawasaki disease (KD), many rheumatology-specialized medical centers authorize more aggressive therapies, such as the use of CD20-targeted monoclonal antibodies, only when abnormalities are documented on radiologic studies or AP analysis. This approach is justified by the high morbidity associated with these medications used in the treatment of primary vasculitides [1, 17]. In this context, it is noteworthy that the patient did not receive specific aggressive therapies, such as monoclonal antibodies or cyclophosphamide, due to the absence of radiologic and anatomopathological abnormalities. Although it is recognized that findings such as neutrophilic fragmentation are not part of the diagnostic criteria for KD, biological material (digital arteries and veins) collected during

multiple debridements was submitted for histopathological analysis. The potential interference of prednisone with AP results was acknowledged. Corticosteroid and antiplatelet therapy were maintained throughout the entire reported period [1, 4, 8, 18].

In medical evaluations prior to admission at CHMSA, amputation of the affected limb was indicated. Given the clinical progression, with a Wifi classification of 2-3-2 and TASC D, this indication was considered appropriate, as no other therapeutic options were available. Unfortunately, the literature lacks therapeutic guidelines for cases in which lower limb revascularization is anatomically impossible [13.1, 14.1]. In such situations, amputation becomes an acceptable alternative to control rest pain or prevent sepsis.

Encouraged using Transverse Tibial Cortex Transport (TTT) in secondary vasculopathies, an attempt was made to preserve the limb in a case of KD [15]. Contrary to what has been described in the literature, improvement in perfusion was observed through clinical parameters such as capillary refill time and ankle pressure shortly after osteotomy. This improvement was evident even before the initiation of traction cycles promoted by the metallic device [15.1]. In accordance with the principles advocated by Ilizarov, it is believed that osteotomy alone may represent a therapeutic option in future situations or in settings where access to TTT is not available.

Due to the unprecedented and pioneering nature of the reported case, it was not possible to establish comparative analyses with the existing literature. Nevertheless, the use of TTT in KD yielded a satisfactory outcome, allowing preservation of a limb previously indicated for amputation, as well as restoration of motor function with recovery of ambulation [16.1]. Despite these results, prospective studies are required to evaluate the use of TTT both in patients with KD and in other primary vasculitides, in which limb amputation often represents the only available therapeutic option.

Prednisone and acetylsalicylic acid were maintained both during the period of clinical deterioration and after TTT. In line with the literature, it is believed that these medications contribute to improvement in KD by reducing the inflammatory process. However, there is no documentation in the literature regarding the effects of concomitant corticosteroid and antiplatelet therapy during or after TTT [10.1].

The classifications currently used for limb ischemia have significant limitations and are subject to criticism when applied to patients with atherosclerotic peripheral arterial obstructive disease. In individuals with primary vasculitides or KD, these classifications are even less applicable [19, 20]. Nevertheless, patients who evolve with worsening perfusion and lack adequate therapeutic options were staged [11.1]. The Wagner classification was not used, even in an adapted form, as it does not adequately differentiate the etiologies of infection and necrosis, even in diabetic patients [19].

In the present report, both the traditional Fontaine classification and the more recent Wifi stratification system were considered as adjuncts to the diagnostic and therapeutic approach [19, 20]. None of these classifications is fully adequate for staging primary vasculitides. The American College of Rheumatology provides no specific guidance on stratifying patients with primary vasculitides who candidates for revascularization are. Due to the active inflammatory process in acute settings, the literature is consensual in prioritizing clinical treatment, reserving surgical approaches for exceptional situations [12.1]. The mere presence of a trophic lesion already classifies the patient as Fontaine IV, while the multiple arterial occlusions observed in Figure 1 characterize a TASC D pattern [19, 20].

This case demonstrates that Transverse Tibial Cortex Transport (TTT) can be a feasible and effective limb-salvage strategy for adult patients with Kawasaki disease and critical lower limb ischemia when conventional revascularization is anatomically impossible and amputation is the only standard therapeutic option. Despite the absence of radiologic and anatomopathological criteria supporting the use of aggressive immunosuppressive therapies, TTT was associated with early and sustained clinical improvement in perfusion, wound healing, and functional recovery of ambulation, culminating in successful limb

preservation. These findings suggest that TTT, and possibly osteotomy alone, may represent a valuable alternative in selected cases of primary vasculitides complicated by critical ischemia, particularly in scenarios with limited therapeutic options. Given the lack of validated staging systems and therapeutic guidelines for ischemia related to vasculitis, this report highlights the need for prospective studies to better define the role, mechanisms, indications, and long-term outcomes of TTT in Kawasaki disease and other primary vasculitides.

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Conflicts of Interest: All other authors declare no conflicts of interest.

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