Case Report Periosteal distraction for foot preservation contributes to favorable clinical effects and prognosis in a foot ulcer patient with critical limb ischemia after multiple operations

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Received March 9, 2023; Accepted April 24, 2023; Epub May 15, 2023; Published May 30, 2023

Abstract: This paper presents the challenges faced by a patient undergoing limb-sparing treatment due to chronic limb-threatening ischemia (CLTI) complicated with long-standing nonhealing foot ulcer and intense pain. However, after multiple vascular surgeries, the foot wound continued to worsen, which could lead to transfemoral amputation and even death. We report a case of an aged male patient admitted after complaining of "pain and ulceration in his left foot for ten months". The patient was diagnosed with arteriosclerosis obliterans of the lower limbs with critical limb ischemia, which improved little after the drug therapy. This patient had undergone three endovascular procedures with a medical history of myocardial infarction and stenting. The main artery could not be directly connected to the foot by open or endovascular surgery due to severe vascular occlusion below the knee. In addition, foot ulcers made it impossible to walk, which induced angina pectoris. After coordination and discussion, we determined to perform a 2-week lateral tibial periosteum distraction (LTPD). The procedure significantly improved the foot wound and relieved the pain. After the two-week personalized wound management, the wound healed, and the pain disappeared. Consequently, the patient was able to walk independently, without recurrence during the 3-month follow-up period. Periosteal distraction has rarely been reported in previous literature and is mainly used to treat patients with diabetic foot, rather than those who have undergone repeated percutaneous transluminal angioplasty (PTA) for CLTI combined with foot ulcers. As most CLTI patients are suffering from cardiac, cerebral, and renal diseases, their blood vessels are difficult to open, with high re-occlusion and recurrence rates and low limb salvage rate. Therefore, we present our case herein so that the CLTI patients whose inferior genicular arteries cannot be opened due to severe infrapopliteal arterial occlusion complicated with nonhealing foot ulcers or intractable pain can be treated with LTPD, thus providing them with the "last kilometer" bloodstream towards the foot.

Keywords: Chronic limb-threatening ischemia, lateral tibial periosteum distraction, percutaneous transluminal angioplasty, peripheral artery diseases

Introduction

Lower limb ischemia is the most common form of peripheral artery diseases (PAD) seen in clinical practice, manifested as intermittent claudication, pain, ulcers, gangrene, or other lower limb ischemic symptoms due to arterial stenosis or occlusion and insufficient blood perfusion of the lower extremities [1].

Chronic limb-threatening ischemia (CLTI) is the severest stage of ischaemia during lower limb PAD progression, when the bloodstream is inhibited by the arterial occlusion in lower extremities. It seriously affects the quality of life of patients and can even lead to amputation or death in some cases, with reduced walking ability, resting pain for weeks, ulcers, and gangrene as the major clinical manifestations [2].

PAD has a global prevalence of 3%-10% [3], of which 10%-20% can develop CLTI [4-6]. The annual incidence of CLTI is estimated to be 220-3500 per 1 million people in countries such as the United States and the United Kingdom, with the incidence of 1% in adults.

CLTI generally occurs in the severest stage of PAD and has a high mortality rate (exceeding 50% within 5 years) [7]. Despite its global prevalence, PAD is still poorly recognized by healthcare workers and patients. This lack of awareness has led to under-diagnosis and undertreatment of PAD [8]. In the past, open surgical bypass was a common treatment for PAD, which was associated with excellent limb salvage rate and clinical durability, but clinical outcomes vary widely due to a number of factors [9].

Herein, we report a case of CLTI with complete occlusion of the inferior genicular artery and reviewed case reports of this disease, based on which we ultimately decided to focus on CLTI patients with nonhealing foot ulcers or intractable pain. By studying this case, we found that periosteal distraction had a positive effect on limb salvage in patients who could not undergo open vasculature due to severe infrapopliteal arterial occlusion but with popliteal arterial patency. Given the extreme rarity of similar cases, it is necessary to document and report this case in order to provide references for the subsequent treatment of similar patients.

Case report

A 70-year-old male with a 30-year smoking history was presented to Qingdao Haici Hospital Affiliated to Qingdao University with progressively worsening conditions caused by pain and ulcers in his left foot, combined with coldness and numbness in his lower extremities, for 10 months. He underwent a series of procedures in 2021, including "percutaneous transluminal angioplasty (PTA) + stent implantation of the left femoral, left popliteal, and left anterior tibial arteries" in February, "endovascular atherectomy + balloon dilatation" in June, and "endovascular drug-coated balloon dilatation" in September. During this process, the foot wound became increasingly worse, despite multiple methods employed. This patient was admitted to our hospital on October 28, 2021, presenting with anterolateral ulcer of the left foot, pus exudation and smelliness, destruction of the fifth metatarsophalangeal joint, and haphalgesia (+) pulselessness of the popliteal and infrapopliteal arteries. The ankle brachial index (ABI) was 0.5 and the Visual Analogue Scale (VAS) score was 9 points on admission. Computed tomography angiography (CTA) showed

extensive sclerosis in the thoracic aorta, abdominal aorta, and bilateral iliac arteries. The contrast agent was visible in the tubular stent shadow located in the upper part of the left femoral artery but was not observed in the tubular stent shadow located in the left popliteal artery. The left posterior tibial artery was not clearly visualized, while the left peroneal artery was visualized intermittently. In the left anterior tibial artery, there was also a tubular stent shadow, in which no contrast agent was found (**Figure 1**).

Considering the patient's poor physical condition, intense foot pain, and unfavorable wound progression, we performed lateral tibial periosteum distraction (LTPD) under local anesthesia on November 18, 2021. The specific steps are as follows:

After a 2-centimeter-long longitudinal incision made at the medial tibial flare below the tibial tubercle, the subperiosteal channel was opened by inserting a nerve dissector under the periosteum through a transverse periosteal incision. Then, a special periosteum elevator, with the same length as the pre-inserted steel plate, was placed, and the subperiosteal dissection was performed along the direction of the pre-inserted steel plate, followed by the removal of the periosteum elevator. Using the longitudinal skin incision and transverse periosteal incision as a coordinate axis and the midpoint of the incision as the origin, four quadrants were obtained. In each quadrant, a hole was drilled approximately 1 cm from the incisions along both directions using a 2.0 mm drill bit. Four holes were drilled in total, all of which penetrated the unilateral cortex to decompress the medullary cavity. Then, a thin steel plate (0.8 cm × 8.0 cm) was inserted into subperiosteum via the periosteal incision and was moved backwards after the whole plate was inserted to the distal extremity, thus making it cross the middle of this incision. A flat-head cannulated screw was screwed into the central hole of this plate through the periosteal incision, and a 1.5mm Kirschner wire was drilled into this screw centrally to fix the plate and screw (Figure 2). Finally, a distractor was installed to verify whether it could work properly [10].

After surgery, the patient was able to walk normally, with the pain relieved and the skin temperature and oxygen saturation significantly



Figure 1. Patient condition on admission. A: Exposed state of the fifth metatarsophalangeal joint at admission; B: Angiograph of popliteal arteries (the stent implanted in popliteal and anterior tibial arteries is indicated by an orange arrow); C: Foot angiograph (no straight bloodstream is observed above the ankle joint).

improved. Distraction treatment was performed on the third postoperative day by rotating the screw clockwise to separate the steel plate inserted under the periosteum from the bone cortex for about 0.75 mm/d. Then, X-rays were taken to check whether the periosteum was lifted by the distractor. After two weeks of continuous adjustment, the patient's symptoms improved considerably. At this point, both the Kirschner wire and distractor were removed. Then, the patient's fifth toe was resected, and two weeks later, the foot wound healed. The ABI and VAS scores measured at discharge were 0.9 and 1 point, respectively.

Discussion

We report a case of critical limb ischemia in an elderly male patient. First, he suffered intense pain that was not relieved by various methods, which induced angina pectoris. As the condition exacerbated, the ulcer worsened, which indicates the need for transfemoral amputation and the risk of death. Second, the patient's symptoms did not improve despite multiple visits to different hospitals and repeated PTA procedures. The patient's inferior genicular arteries could not be opened surgically, but he had an intense desire for limb salvage. Third, the critical cardiovascular diseases make him unable to tolerate epidural or general anesthesia.

Hence, after extensive discussions on limb salvage, timely and effective establishment of sufficient foot perfusion is determined as the only solution. Animal experiments on periosteal distraction have been reported in the previous literature, and some reports have focused on the application of this method in the treatment of diabetic foot, which is mainly performed under epidural anesthesia. In our case, local anesthesia was used with regard to the potential risks, and the patient was informed. After surgery, the skin color and temperature of the affected foot

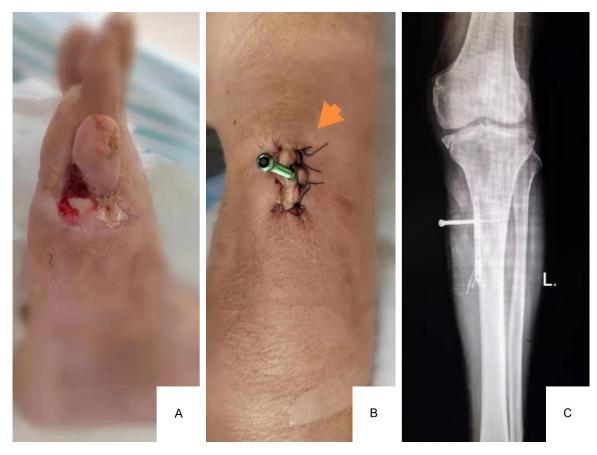


Figure 2. Patient's condition after debridement of lateral forefoot wound. A: The blood supply at the floor of the wound was significantly improved after the wound bed was debrided in the lateral forefoot; B: The surgical incision in the upper part of the tibia, and the exposed distraction screw; C: Locations of the steel plate and screw in X-ray images.

improved significantly, and the pain was also relieved. Thereafter, personalized wound management was performed during periosteal distraction to facilitate foot wound healing.

In addition, the patient's ABI score was tested and a CTA examination was scheduled. CTA results (**Figure 3**) showed a remarkable improvement in symptoms but no significant difference in pre and post-operative ABI and foot circulation. In this report, periosteal distraction greatly improved the symptoms of a patient whose vasculature could not be opened due to severe vascular occlusion below the knee accompanied by nonhealing foot ulcers and intractable pain, while playing an active and effective role in wound healing and pain relief.

However, there are still some shortcomings in this study. First, only one patient was reported in this study. While the report is detailed, the sample size is small and needs to be expanded to improve the accuracy of the findings. Second, patient prognosis was not further analyzed, leading to incomplete results. In future studies, we will improve the study design to address these deficiencies.

Conclusion

At present, the primary treatments for CLTI include medication, endovascular surgery, surgical interventions, and other tentative methods such as gene therapy and stem cell transplantation. Among them, medication only slows down the progression of lower limb arterial occlusion, rather than fundamentally alleviating the vascular stenosis and occlusion caused by arteriosclerosis obliterans. In contrast, revascularization via endovascular surgery or surgical interventions is the preferred treatment for CLTI. However, it is not applicable to all CLTI patients, as many of them cannot tolerate surgery due to advanced age, co-morbidities, or

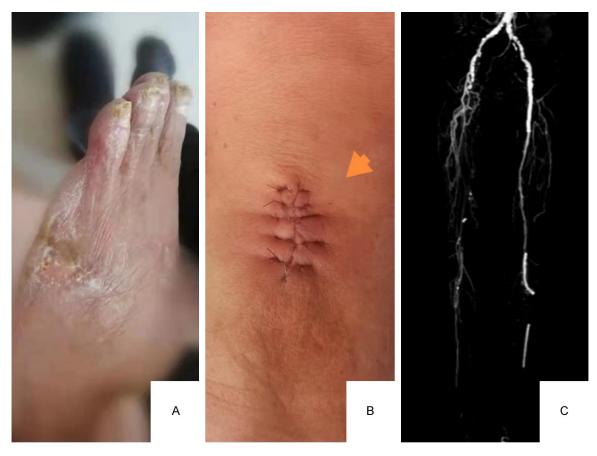


Figure 3. Stump surgical wound healing after removal of the fifth toe. A: The wound healing of the stump after the resection of the fifth toe; B: The incision after the removal of periosteal distractor; C: CTA image of wound healing (the main artery is not visualized below the left knee). Note: CTA, computed tomography angiography.

other factors. Nor is it infeasible for those with unfavorable conditions in the distal outflow tract of lower limb arteries and with restenosis or reocclusion of the treated site. It is reported that 10%-50% of CLTI patients do not undergo revascularization [4, 11, 12], and as a result, they face a high above-ankle amputation rate of 40% and a high mortality rate of 20% within six months [13].

This report presents an effective clinical therapy for intractable pain and complicated wound in an elderly patient with foot ulcers during the difficult limb salvage process. In recent years, tibia transverse transport (TTT) based on llizarov technique [14] has achieved satisfactory clinical results in the treatment of middleadvanced foot and ankle diseases caused by chronic ischemic diseases of lower limbs [15-26]. However, this method requires osteotomy of the midshaft tibia, which expands the operative wound and damages the bone structure, carrying the risk of postoperative fracture. Therefore, professor Zeng Naxin of China adopted special devices in clinical practice and standardized the application of LTPD [10]. Previous animal experiments have verified that the stretch-stress mechanical stimulus can promote the regeneration of capillaries and tissues, and distraction histogenesis can stimulate the formation of "vascular network" [10, 27, 28]. Limited literature has reported the clinical application of LTPD, mostly of which focuses on the treatment of diabetic foot, and no report is concerned with its application in CLTI patients who cannot tolerate revascularization (Table 1). In our case, we found LTPD to be less invasive than TTT, but more simple, economical, and convenient.

However, surprisingly, there was no difference in pre- and post-operative foot angiograph and ABI scores in this case. Therefore, it is necessary to further study whether the accelerated wound healing and pain relief by LTPD are relat-

No.	Number of cases	Time	Combined diseases	Wound description	Type of anesthesia	Age (year)	Follow-up visit
1	1	2019	Type 2 diabetes	Necrosis of the second toe of the right foot	Epidural anes- thesia	71, female	The dry gangrene of the toe was unhealed [29].
2	1	2021	Arteriosclerosis obliterans of lower limbs	Ulcer of the first toe of the right foot	Unspecified	81, female	The wound healed.
3	1	2021	Hypertension, diabetes, coronary heart disease, and cerebral infarction	Necrosis of the fourth toe of the left foot	Epidural anes- thesia	75, female	The wound healed.
4	13	2019~ 2020	5 cases of DF, 1 case of DF combined with ASO, 2 cases of TAO, and 5 cases of ASO	12 cases of wound infection and (or) gangrene, and 1 case of ASO without ulcer	Local anaesthesia or epidural anesthesia	9 males and 4 females at the age of 39~77, with an average age of 66	2 patients underwent amputation, 1 patient still felt painful, and 10 patients had improved wound [10].
5	1	2021	Hypertension, coronary heart disease, heart failure, cerebral infarction, and after the surgical treatment of myo- cardial infarction and PTAS	Ulcer in the anterolat- eral part of the left foot combined with destruc- tion of the metatarso- phalangeal joint	Local anaes- thesia	70, male	The wound healed within 3 weeks, without recur- rence in the following 3 months.

Table 1. Previous case reports about the treatment of lower limb ischemia by periosteal distraction

ed to reduced intrapulpal pressure in the tibia and systemic stem cell mobilization.

Disclosure of conflict of interest

None.

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