Successful bilateral popliteal-plantar bypasses for polyarteritis nodosa induced ischemia

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ABSTRACT

Polyarteritis nodosa (PAN) is a necrotizing medium vessel vasculitis which causes stenosis, thrombosis, and microaneurysms. PAN characteristically affects people in their 40s to 60s with a male predominance and involves visceral vessels, however there are case reports of peripheral vascular involvement. Criteria for diagnosis of PAN include three or more of the following symptoms or signs: weight loss of ≥4 kg since illness began; livedo reticularis; testicular pain or tenderness; myalgias, weakness, or leg tenderness; mononeuropathy or polyneuropathy; diastolic blood pressure <90 mm Hg; elevated blood urea nitrogen >40 mg/dL or creatinine >1.5 mg/dL; hepatitis B virus; arteriogram showing aneurysms or occlusions of visceral arteries; and biopsy of a small- or medium-sized artery containing neutrophils, granulocytes, or mononuclear leukocytes in the artery wall. This case of PAN caused critical limb-threatening ischemia with digital gangrene and contralateral intermittent claudication, and was successfully treated with bilateral revascularization by popliteal-plantar bypass. The patient consented to publication of the details and images included in this report.

CASE REPORT

The patient was a 49-year-old man who had been healthy until he presented to an outside institution with 1 month of malaise, low-grade fever, weight loss, new-onset chest pain, and hypertension. He had quit smoking cigarettes about 15 years before presentation; he had smoked two packs per day for 15 years before cessation. He had no other contributing medical or social history. He had an elevated troponin level and underwent a cardiac catheterization, which demonstrated minimal coronary disease. After an extensive evaluation that revealed elevated inflammatory markers, multifocal scarring of bilateral kidneys with mildly elevated creatinine, right foot short distance claudication, early left first toe gangrene, left foot rest pain, and a testicular infarct, he was ultimately diagnosed with PAN. He was initially treated with prednisone, required the addition of oral cyclophosphamide, and transitioned to intravenous cyclophosphamide owing to intolerance of oral dosing. Over the ensuing 4 months, his constitutional symptoms improved and his inflammatory markers returned to normal. He transitioned to oral azathioprine for maintenance remission therapy. Early in his disease course, he was seen by vascular surgery at an outside institution for his peripheral vascular complaints. His left first toe pressure was unmeasurable. Angiography revealed normal vessels to the proximal calf with occlusion of all three left tibial arteries and reconstitution of the medial plantar artery (Fig 1). As his erythrocyte sedimentation rate and C-reactive protein normalized with treatment, although his constitutional symptoms improved, his left foot rest pain continued to be severe with further progression of dry gangrene. He was referred to the authors’ institution for revascularization. Approximately 6 months after his symptoms began and 2 months after stabilization of his inflammatory markers, he underwent an uneventful left below-knee popliteal to plantar artery bypass using the ipsilateral greater saphenous vein. He was observed overnight for pain management and discharged...
on postoperative day 1. He healed without issue, his rest pain resolved, and he underwent a partial left first toe amputation several months later after his dry gangrene fully demarcated. Over the ensuing 5 years, he was seen regularly for surveillance of his bypass with his most recent left ankle-brachial index normal at 1.1.
At each follow-up visit, he continued to complain of cramping in his right foot when he walked more than 75 feet, with an ankle-brachial index of 0.8 and toe pressure of 72 mm Hg. He maintained a walking program; however, this claudication remained lifestyle limiting. For example, he found it quite frustrating and bothersome that he had to take multiple breaks for the foot pain and cramping to resolve while trying to mow his lawn. During this time, his PAN was controlled with stable renal function and no constitutional symptoms. With his right foot claudication symptoms unimproved despite maximal conservative measures, he consented to a right lower extremity angiogram, which confirmed tibial artery disease in a pattern similar to his previously treated left-sided disease (Fig 2). He subsequently underwent a right below-knee popliteal to plantar artery bypass using cephalic vein, as the right greater saphenous vein was sclerotic. Again, he was observed overnight and recovered uneventfully. Seven months postoperatively, he is able to walk unlimited distance with no claudication with an ankle-brachial index of 1.3 and a normal bypass by duplex imaging (Fig 3).

**DISCUSSION**

This 49-year-old man developed critical limb-threatening ischemia of his left foot with digital gangrene and intermittent claudication of his right foot as manifestations of PAN. Alongside aggressive medical management of his PAN, both lower extremities were successfully treated with popliteal-plantar bypasses. Beyond the less common exposure of the plantar artery (Fig 4), this patient and his treatment must be considered in the context of published reports of peripheral vascular manifestations of PAN (Table).

There are several reports of rapidly progressive ischemic presentations of PAN. Buhl et al reported the case of a 42-year-old patient with acute myeloid leukemia who developed PAN with bilateral lower extremity gangrene with no attempt at revascularization before bilateral lower extremity amputation owing to rapid progression of symptoms despite systemic therapy including high-dose corticosteroids, cyclophosphamide, and plasmapheresis. There are two reports of patients presenting with symmetric digital gangrene of both upper and lower extremities owing to PAN without revascularization attempt. The case reported here similarly had quite symmetric disease of his lower extremities. Héron et al reported a case of a 33-year-old patient with PAN who developed acute limb ischemia with foot parasthesias and rest pain owing to tibial occlusion which was managed with anticoagulation. This patient had slow improvement of her symptoms over the course of a year.
In the case reported here, the patient was lucky to have had early diagnosis of his PAN, allowing aggressive systemic treatment and improvement in his constitutional symptoms and inflammatory markers before he underwent operative revascularization. He did require a partial toe amputation, but the timeline of his diagnosis and initial PAN treatment did not preclude limb salvage, as in the cited cases. Other groups have also reported successful revascularization for PAN induced peripheral ischemia. There are several reports of successful healing of cutaneous ulcers through endovascular interventions and a report of a successful bypass for a patient with an iliofemoral stenosis from PAN causing progressive claudication. The patient reported here similarly has done well in the 5 years since his initial popliteal-plantar bypass. This limited literature suggests that, alongside systemic medical therapy, open or endovascular revascularization is appropriate for PAN induced ischemia.

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