Lower extremity aneurysmal degeneration of great saphenous venous allograft bypass in an adolescent boy

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ABSTRACT

Chronic limb-threatening ischemia (CLTI) refers to a complex condition that manifests as chronic ischemic rest pain or ischemic skin lesions present for >2 weeks.1-3 The pediatric population represents a minute portion of patients with CLTI, with sparse evidence available to guide treatment. The few case reports documenting open repair in the pediatric population have shown a preference for an autogenous conduit compared with a cadaveric or prosthetic conduit.4,5 However, vein grafts are not without complications, because they can develop graft stenosis, thrombosis, and aneurysmal degeneration and necessitate reintervention.6

In the present report, we have described the case of an adolescent boy with lower extremity ischemia and popliteal artery occlusion due to type III popliteal artery entrapment necessitating above-the-knee popliteal artery to the posterior tibial artery bypass with an autogenous great saphenous vein (GSV) graft. Seven years later, the patient presented with acute limb ischemia with bypass occlusion secondary to aneurysmal degeneration, which necessitated thrombolyis before vein bypass revision. The patient provided written informed consent for the report of his case details and imaging studies.

CASE REPORT

A 10-year-old boy had presented with increasingly prominent claudication symptoms during participation in athletics and a nonhealing ulcer at the plantar aspect of the left foot. The left ankle brachial index was 0.57, and magnetic resonance angiography demonstrated occlusion of a medially displaced popliteal artery with reconstitution of the tibial vessels via a large geniculate branch (Figs 1 and 2). These findings were confirmed by arteriography showing a long segment occlusion of the popliteal artery with reconstitution at the tibioperoneal trunk and posterior tibial artery serving as the dominant run-off vessel.

The patient was electively scheduled for above-the-knee popliteal artery to posterior tibial artery bypass with non-reversed ipsilateral GSV harvest. Intraoperatively, a thick myofascial band consistent with type III popliteal artery entrapment was identified and divided. The left GSV, measuring 4 mm and tapering to 3 mm, was harvested from the saphenofemoral junction to the knee and prepared with valvotomy before tunneling the graft laterally to a prominent medial head of the gastrocnemius. The postoperative ankle brachial index was 1.3 and duplex ultrasound demonstrated a patent bypass. Four years of annual follow-up visits with noninvasive vascular surveillance demonstrated graft patency without thrombus and gradual dilation of the bypass from 8 mm at 1 year to 15 mm at 4 years. During this period, the patient reported inconsistent adherence to a daily aspirin (ASA) regimen.

The patient was lost to follow-up until age 17 when he presented to the emergency department with acute onset of left lower extremity rest pain and absent pedal doppler signals. Computed tomography angiography showed an occlusive thrombus within the bypass at an area of aneurysmal dilatation measuring 20 mm in diameter (Fig 3). Catheter directed thrombolysis was initiated with thrombus resolution after 4 days and...
the patient was discharged with a prescription for ASA and rivaroxaban. Two months later, the patient returned for elective revision of the bypass with contralateral non-reversed right GSV graft prepared with valvotomy. A few centimeters of the proximal and distal portions of the aneurysmal bypass, including the anastomoses, were resected. The new vein bypass was sewn to the same proximal and distal arteriotomy sites, tunneling within the same anatomic tract (Fig 4). Pathologic examination of the excised aneurysmal bypass tissue demonstrated intimal hyperplasia with myxomatous degeneration. At the age of 22 years, the patient continues to endorse resolution of claudication symptoms and has continued a physically rigorous lifestyle as a collegiate athlete with adherence to daily ASA and annual surveillance with stable noninvasive vascular studies.

DISCUSSION

CLTI is rare in the pediatric population, with previous studies primarily focused on iatrogenic etiology, with minimal data regarding alternative causes such as vasculitis-related stenosis, hypercoagulable state-related thromboses, structural anomalies, and infectious or paradoxical emboli. The present patient had been diagnosed with type III popliteal artery entrapment due to a myofascial band directly compressing the popliteal artery. Popliteal artery entrapment is caused by direct compression of the popliteal artery, and long-term compression can lead to intimal damage and occlusion. A review of the reported data pertaining to acute and chronic pediatric lower extremity ischemia necessitating open repair showed a preference for use of GSV conduit, driven primarily by the anticipated long-term life expectancy of the pediatric population and associated long-term complications. Dalsing et al performed five lower extremity bypasses with an autogenous conduit in patients aged <13 years with three reversed GSV bypasses remaining widely patent at a mean of 35.6 months. Reed et al treated four patients aged <18 years with a reversed GSV graft for popliteal artery injury with normal duplex waveforms and segmental pressures at 10 to 42 months of follow-up without secondary intervention.

The present patient was followed up with noninvasive vascular studies for 4 years without incident or an indication of bypass irregularity. Aneurysmal degeneration of the bypass was only noted when the patient had returned with rest pain due to acute thrombosis of the aneurysmal bypass 7 years after its creation, including 3 years without surveillance. An extensive review of the reported data pertaining to lower extremity bypass in the pediatric population failed to find outcomes data beyond 3 to 4 years. The importance of long-term outcomes data is emphasized by the events described in the present case report. The data lacking regarding the pediatric population can be cautiously extrapolated.
from the adult population. Huang et al\textsuperscript{12} reviewed 358 lower extremity GSV bypasses in adults and reported 5-year primary and secondary patency rates of 85\% and 94\%, respectively. Hoelting et al\textsuperscript{13} reported follow-up primary patency rates that had decreased to 57\% to 65\% during a period of 8 to 10 years, stressing the importance of continued follow-up for patients with autogenous bypass owing to the occurrence of late complications. As shown from these studies, the importance of ongoing surveillance beyond the initial 3 to 4 years cannot be understated, because the early detection of irregularities can vastly improve the overall, long-term assisted patency.\textsuperscript{14}

Aneurysmal degeneration of an autogenous vein bypass is a rare, but known, late complication.\textsuperscript{15} The underlying etiology is speculative. Some have theorized that structural differences between the autogenous vein compared with that of the native artery leads to an accelerated rate of atherosclerotic change and sequelae of ulceration, obliteration of elastic lamina, and fibromuscular intimal thickening. An alternative proposed mechanism is a systemic dilatory process encompassing inflammation and immune responses, biochemical wall stress, and molecular genetics.\textsuperscript{16,17} Various studies have supported both theories, suggesting the process is likely multifactorial.

Patients with an autogenous lower extremity bypass will require lifelong surveillance.\textsuperscript{18} It has been our practice to monitor these patients every 3 months for the first 2 years, every 6 months for 1 year, and then annually. Noninvasive vascular imaging studies provide inexpensive and valuable information regarding the flow

**Fig 3.** A, Completion angiogram of the index bypass procedure using ipsilateral non–reversed great saphenous vein (GSV) from the above-the-knee popliteal artery to posterior tibial artery tunneled in anatomic fashion. B, Computed tomography angiogram obtained during acute onset of rest pain and absence of pedal Doppler signals 7 years after the index procedure showing occlusive thrombus 9 cm distal to the proximal anastomosis within an aneurysmal segment of the bypass measuring 20 mm in diameter. C, Completion angiogram after 4 days of catheter-directed thrombolysis demonstrating patency of the aneurysmal bypass without residual thrombus or recurrence of external compression.
and an interposition graft. They reported that during rysm ligation and bypass or endoaneurysmorrhaphy up 55 popliteal artery aneurysms repaired with aneu-
tural abnormality (ie, graft stenosis, vein graft aneu-
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CONCLUSIONS
Aneurysmal degeneration in autogenous bypasses is a rare, but known, late complication that is of particular concern in the pediatric population owing to the extended, lifelong dependency of the patient on the bypass. The long-
term outcomes will be optimized with ongoing surveil-
ance and the early detection of irregularities, allowing for elective, rather than emergent, intervention.

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