Recurrent upper extremity acute limb ischemia secondary to retained axillary polytetrafluoroethylene cuff causing axillary stump syndrome

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ABSTRACT

Acute limb ischemia of the upper extremity is less frequently encountered than in the lower extremity. The etiology is typically cardioembolic. Axillary-femoral stump syndrome is a rare complication associated with an occluded axillary-femoral bypass graft. We present the case of recurrent acute limb ischemia of the upper extremity whose embolic source was a retained cuff of a previously explanted axillary-profunda bypass graft. The patient failed anticoagulation after an initial embolectomy and after a recurrent embolism from the retained cuff, ultimately required cuff exclusion with a covered stent. (J Vasc Surg Cases and Innovative Techniques 2020;6:606-8.)

Keywords: Axillofemoral graft; Graft infection; Axillary stump syndrome

Acute limb ischemia of the upper extremity is less frequently encountered than in the lower extremity, accounting for only 2% to 18% of all episodes of acute limb ischemia.1-3 The etiology of the embolus is typically cardioembolic.1,2 A rare etiology is axillary-femoral stump syndrome (AxFSS) where thrombus embolizes to the upper extremity from an occluded axillary-femoral bypass graft.4-10 We present the case of recurrent acute upper limb ischemia following explantation of an infected axillary-profunda bypass with a retained axillary cuff salvaged with covered stent exclusion of the cuff after repeated distal embolization. The patient agreed in writing to his images and case details being published.

CASE REPORT

A 63-year-old man with a history of diabetes, coronary artery disease, chronic obstructive pulmonary disease, and hypertension presented to an outside facility with right lower extremity disease, chronic obstructive pulmonary disease, and hypertension. Two months following his amputation he re-presented to our institution after a herald bleed with exposed prosthetic and purulent, malodorous femoral drainage. On presentation, he was afebrile with a leukocytosis of 19.11 10^3/μL. He was taken to the operating room for explant of the previous femoral-femoral bypass with sartorius muscle flap for coverage and construction of a right axillary-profunda bypass off of the second portion of the axillary artery with 8-mm polytetrafluoroethylene (PTFE). His postoperative course was complicated by thrombosis of the axillary-profunda bypass on postoperative day 1. A thrombectomy of the axillary-profunda bypass with a jump graft to the above knee popliteal artery with 8-mm PTFE was performed to provide in-line flow to the foot for gangrenous changes. Blood and operating room cultures returned positive for methicillin-sensitive Staphylococcus aureus and he was discharged with a 6-week course of cefazolin and rifampin.

At his follow-up appointment 4 months later, his incisions were completely healed with no signs of infection. He was then lost to follow-up. Four years later he underwent a right above-knee amputation at an outside institution. Two months following his amputation he re-presented to our institution after a herald bleed with exposed prosthetic and purulent, malodorous femoral drainage (Fig 1). He was afebrile with a leukocytosis of 28.53 10^3/μL and negative blood cultures. Physical examination demonstrated a portion of the graft externalized at the right pelvis, superior to the right inguinal regional (Fig 1). Computed tomography angiography showed an occluded bypass with air visualized throughout much of the graft and a rim-enhancing fluid collection with foci of gas and inflammatory stranding surrounding the graft in the proximal right thigh. The bypass grafts were explanted leaving a cuff of well incorporated PTFE over-sewn at the axillary anastomosis. Operating room cultures were positive for Klebsiella pneumoniae and Proteus vulgaris. The patient was discharged with a course of ertapenem.

The patient re-presented 1 month later with significant pain, motor dysfunction, numbness, and cyanosis in his right hand. Computed tomography angiography imaging demonstrated occlusion of the distal brachial artery. There was contrast visualized in the retained axillary-profunda bypass cuff (Fig 2). A thrombectomy through a distal brachial incision restored flow to the right upper extremity. An echocardiogram performed to evaluate for a possible cardioembolic source did not identify any intra-atrial thrombus or any wall motion abnormalities. At
this point, because no cardioembolic source was identified, the cuff was felt to be the likely source of embolism and the plan was for medical management. For this reason, the patient was discharged on rivaroxaban.

He presented 1 month later to an outside institution with recurrent motor and sensory dysfunction of the right upper extremity. An open thrombectomy was again performed through a more proximal brachial artery exposure and a covered stent was placed across the axillary anastomosis to exclude the retained cuff during this procedure. He was maintained on anticoagulation. His hand progressed to necrotizing tissue loss owing to incomplete revascularization of the radial and ulnar arteries during the previous recurrent thrombotic event and ultimately required a transradial amputation. Duplex ultrasound examination performed during this admission demonstrated stent patency.

**DISCUSSION**

AxFSS is a rarely reported phenomenon and, when described, it occurs in the setting of an occluded axillary-femoral bypass.4-10 A blind stump and Y configuration of the bypass have been postulated as mechanisms for thromboembolic events in AxFSS.4-7,9,10 In the few case reports describing the syndrome, management techniques of the axillary stump include both endovascular and open surgical options in combination with anticoagulation4-10 (Table). Kanamitsu et al10 suggested that treating an axillary stump is not always necessary, but once a thromboembolic event occurs, they recommend resection of the graft with arterial reconstruction. In their case, when initially treated with anticoagulation, as in the patient presented here, recurrent thromboembolism was reported.10 No other authors specified the use of anticoagulation in their reports. There is a paucity of data regarding the long-term durability of endovascular management of AxFSS as all cases were initially treated with open surgery.4-8 In the endovascular era, Kallakuri et al9 described the successful exclusion of the cuff as an embolic source via covered stent placement through the arteriotomy used for the embolectomy. Care was taken in stent placement to exclude the stump while ensuring the thoracic outlet was not crossed to avoid compression of the stent between the clavicle and first rib.9

Leaving a cuff of PTFE during the partial explantation of an axillary-femoral bypass is a reasonable option if that segment is well-incorporated without signs of infection.11 In this patient, the graft was occluded, but the Szilagyi grade III infection led to the need for explantation. The infection was localized to the distal anastomotic site with well-incorporated, uninfected graft at the axillary anastomosis, without signs of infection, leading to the decision to leave a proximal cuff. AxFSS in the setting of a retained cuff has not been well-described. Consistent with cases involving graft occlusion, anticoagulation after thromboembolectomy was insufficient to prevent recurrence of thromboembolism in the setting of a PTFE cuff.10 We believe this cuff acted in a similar nature to the blind stump in an occluded axillary-femoral bypass graft. As such, once a thromboembolic event occurs, either endovascular exclusion or complete open surgical excision should be performed to prevent additional recurrences.

**CONCLUSIONS**

AxFSS is a rare complication associated with an occluded axillary-femoral bypass graft. We present the case of recurrent acute upper limb ischemia following explant of an axillary-profunda bypass with retained axillary cuff which initially failed anticoagulation and after recurrent embolism from the retained cuff, ultimately required exclusion of the cuff with a covered stent and the recurrent thrombotic events led to limb loss. AxFSS from a retained cuff should be treated with either endovascular exclusion or open surgical reconstruction.
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