Saphenous vein graft aneurysm formation in a patient with idiopathic multiple aneurysms

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ABSTRACT

True aneurysmal vein graft dilation is rare, and its etiology remains speculative. However, systemic dilation diathesis is regarded as a risk factor. We herein report a case of a rapidly expanding aneurysm in a great saphenous vein graft, resulting in distal malperfusion in a patient who had previously undergone open repair of multiple popliteal artery aneurysms. After an unsuccessful endovascular intervention, the dilated section was eventually replaced by a reversed segment of the contralateral great saphenous vein. Subsequent whole-exome sequencing identified no relevant mutations. This case provides further evidence that aneurysmal disease may be associated with systemic dilation diathesis. (J Vasc Surg Cases and Innovative Techniques 2018;4:197-200.)

Keywords: Vein graft; Systemic dilation diathesis; Etiology

Anastomotic aneurysms are well described in patients with a reversed great saphenous vein (GSV) for peripheral arterial disease.[1] However, true aneurysmal graft dilation of an autologous vein graft is rare, and its etiology is not completely understood. Possible pathogeneses include atherosclerotic degeneration, systemic dilation diathesis, venous graft varicosities, infection, and poststenotic dilations.[2-6] Plaque and cholesterol depositions are common in bypass grafts, suggesting that atherosclerosis may be the main factor in this process. We herein describe a case of a rapidly expanding nonatherosclerotic aneurysm in a GSV graft, resulting in distal malperfusion. The patient's history of multiple aneurysms prompted us to search for potential genetic factors. Although no exact gene mutation was found, this case provides further evidence that aneurysmal disease may be associated with systemic dilation diathesis. Informed consent was obtained from the patient for the publication of the case details and images.

CASE REPORT

A 46-year-old man presented at our institution with a growing pulsatile mass in his right knee. The mass had rapidly expanded during a 2-week period, causing extreme discomfort because of tension of the overlying skin. Computed tomography angiography (CTA) revealed multiple aneurysms in the thoracic aorta, abdominal aorta, bilateral common femoral arteries, and right popliteal artery (Fig 1). The patient's medical history included asymptomatic left popliteal artery occlusion and cerebral infarction. The patient had no hypertension, hyperlipidemia, or any other systemic inflammatory diseases. The multiple popliteal artery aneurysms (PAAs) were successfully excised. A reversed segment of the ipsilateral GSV was implanted for reconstruction through a medial approach. Postoperative recovery was uneventful, and the ankle-brachial index was 0.9 after the operation. The patient was lost to follow-up.

Three years after the first intervention, the patient developed a rapidly expanding pulsatile painless mass in the right popliteal fossa region. Repeated CTA demonstrated a saphenous vein graft aneurysm (Fig 2). Infection was ruled out, as the patient had no fever, and the serum white blood cell count and procalcitonin concentration were within normal ranges. The serum C-reactive protein concentration was 6.10 mg/L (normal range, 0-5 mg/L). Two days after admission, the patient developed rest pain, and the dorsalis pedis artery pulse was impalpable at that time. Emergency angiography through the contralateral common femoral artery showed distal embolization of the graft aneurysm but not of the infrapopliteal runoff (Fig 3). We planned to reconstruct the popliteal artery using a covered stent but were unable to recanalize the occlusion. The patient then underwent resection of the initial vein graft, using a posterior approach to perform the reconstruction with a reversed segment of the contralateral GSV. Resection and repair were exceedingly difficult as the aneurysm was adherent to the adjacent tissues and ruptured during the operative process. Histopathologic examination of a segment of the aneurysmal vein graft revealed intimal thickening without atherosclerotic change and medial thinning. Postoperative recovery was uneventful, without infection or hematoma development. Repeated CTA performed 1 year after the second intervention

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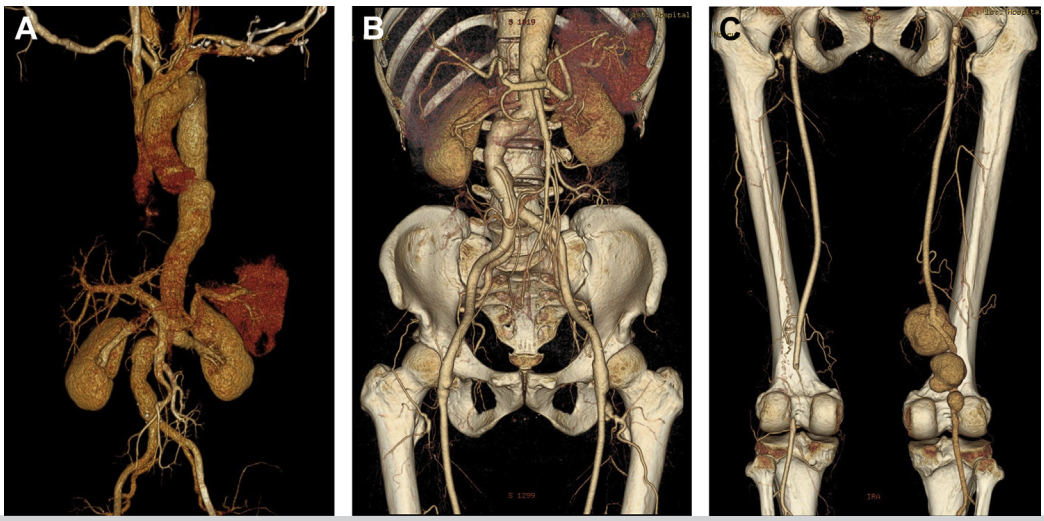

Fig 1. Computed tomography angiography (CTA) revealed formation of multiple aneurysms in thoracic aorta (**A** and **B**; maximum diameter, 45 mm), abdominal aorta (**A** and **B**; maximum diameter, 32 mm), bilateral common femoral artery (**B**; maximum diameter, 15 mm), and right popliteal artery (**C**; maximum diameter, 60 mm).

showed a generally patent graft with mild proximal anastomotic stenosis (Fig 4). However, the left femoral artery was newly occluded, which might have been related to the previous puncture and compression. As the patient was asymptomatic and the ankle-brachial index was 0.7, no further intervention was conducted. The patient's history of multiple aneurysms prompted us to search for potential genetic factors. However, subsequent whole-exome sequencing identified no relevant mutations.

DISCUSSION

Although true infrainguinal vein graft aneurysms are infrequently reported in the literature, a recent study suggested that the incidence may be as high as 8.8%.[7] Dilations in venous bypass grafts may rupture, causing acute ischemia and hemorrhage that require urgent surgical treatment; however, treatment of dilated venous bypass grafts can be challenging because of the presence of extensive scar tissue and a high risk of infection. Despite advancements in endovascular repair for initially untreated PAA, the use of an endoprosthesis to treat a dilation in a venous bypass graft has rarely been reported. In 2009, van Vugt et al[8] described two cases in which covered stents were used. To the best of our knowledge, no similar reports have subsequently been published. In this case, the rapidly emerging distal embolization ultimately resulted in unsuccessful endovascular treatment. Prompt intervention through either an open or endovascular procedure is warranted for rapidly growing or symptomatic defects as well as for those resulting in distal malperfusion.[9]

Aneurysmal disease is associated with several inherited connective tissue disorders, such as Marfan syndrome, Loeys-Dietz syndrome, and some cases of Ehlers-Danlos syndrome.[10] Marfan syndrome is caused by mutation of

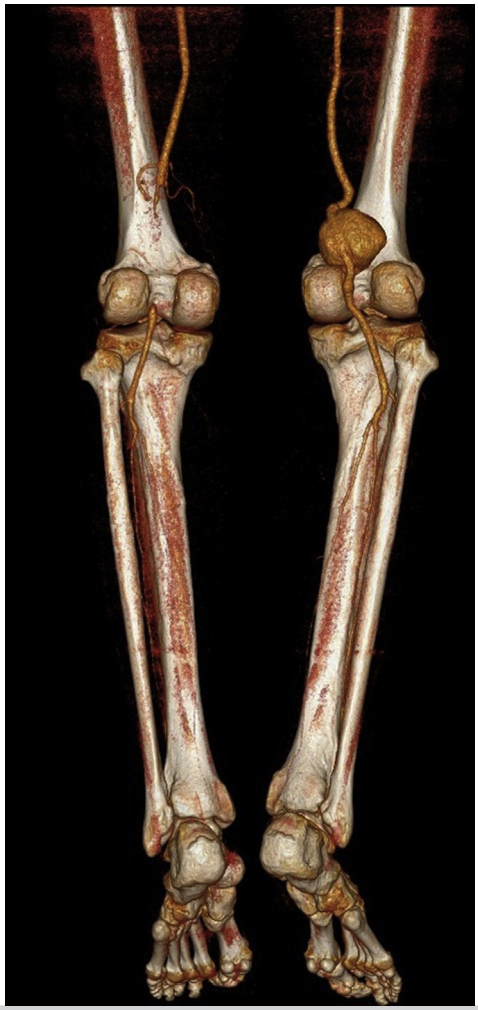

Fig 2. Computed tomography angiography (CTA) showing saphenous vein graft aneurysm formation (maximum diameter, 56 mm).

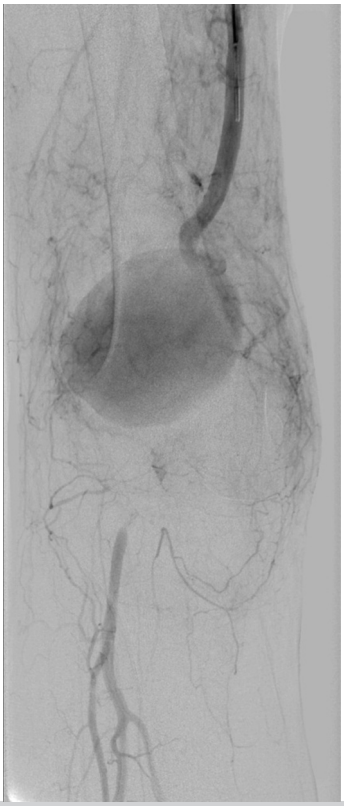

Fig 3. Angiography showing saphenous vein graft aneurysm formation with distal embolization.

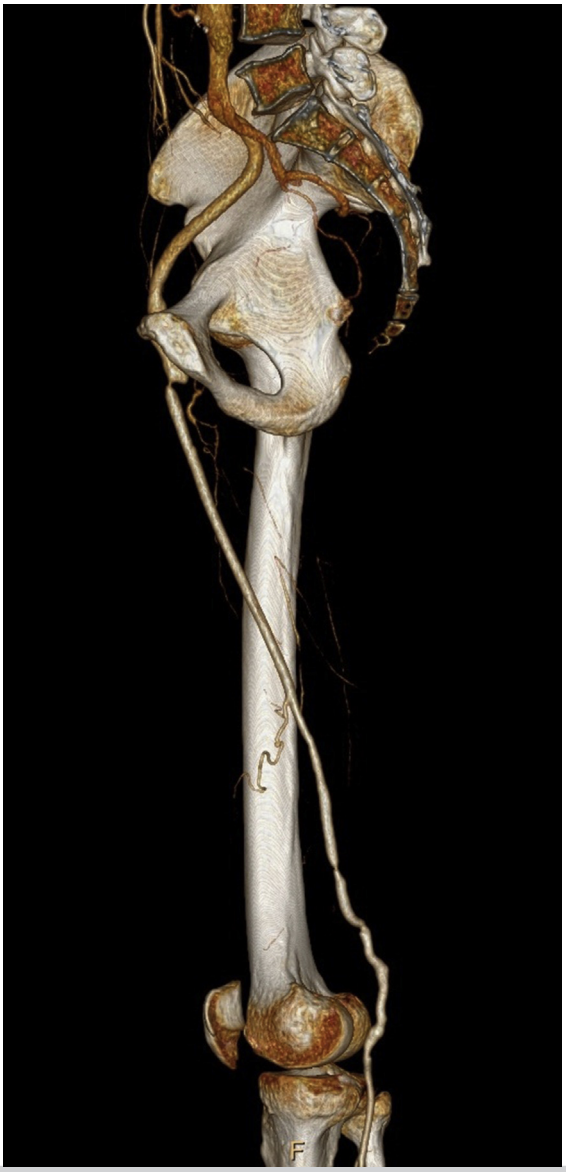

Fig 4. Repeated computed tomography angiography (CTA) showing a generally patent graft with mild proximal anastomotic stenosis. The proximal superficial femoral artery had newly found stenosis.

the *FBN1* gene, and >1500 mutations of this specific gene have been identified so far.[11] However, subsequent whole-exome sequencing of this patient showed no relevant mutations of the *FBN1* gene, the *TGFBR1/2* gene for Loeys-Dietz syndrome, or the *COL3A1* gene for Ehlers-Danlos syndrome.[10] Mutations in some novel genes, including *SMAD3*, *MYH11*, *ACTA2*, and *MYLK*, have recently been reported to be associated with familial thoracic aortic aneurysms and dissections, which may help to define some patients with idiopathic aneurysms.[12]

Enlarged peripheral arteries are noted in patients with abdominal aortic aneurysm (AAA), and the incidence of AAA is increased in patients with PAA.[13,14] This finding suggests a systemic dilation diathesis. Patients with AAA also have an increased incidence of inguinal hernias, diastasis recti, and postoperative incisional hernias, implying defects in collagen and elastin metabolism.[15-17] Aneurysmal degeneration is related to a proportional decrease in the elastin concentration.[18] The abdominal aorta is an elastic artery, whereas the popliteal artery is muscular. It is difficult to explain why the popliteal artery is the second most common site for aneurysm formation (after the aorta) in accordance with the discrimination of arterial types. However, the popliteal artery reportedly manifests some of the same behaviors as a central elastic artery[19]; the stiffness, diameter, and intima-media thickness of the popliteal artery increase with age, whereas the cross-sectional artery wall compliance coefficient and distensibility coefficient decrease with age.[19] These similarities provide further evidence for a systemic dilation diathesis.

True aneurysmal graft dilation is rare, and its etiology remains speculative. Some genetic factors were excluded in this case, which suggests that the pathogenesis may be systemic dilation diathesis. Such patients require lifelong graft surveillance as early diagnosis and intervention can prevent the progression of dilation and minimize potential complications.

REFERENCES

1. Szilagyi DE, Smith RF, Elliott JP, Hageman JH, Dall'Olmo CA. Anastomotic aneurysms after vascular reconstruction: problems of incidence, etiology, and treatment. Surgery 1975;78:800-16.
2. Tao MJ, Aljundi W, Rochenagle G. Aneurysmal degeneration of vein conduit used for vascular reconstruction—case report and literature review. Int J Surg Case Rep 2016;28: 289-92.
3. López MT, Dorgham AS, Rosas FC, de Loma JG. Aneurysmal degeneration of a saphenous vein graft following the repair of a popliteal aneurysm: case report and literature review. Vascular 2012;20:294-8.
4. Nishibe T, Muto A, Kaneko K, Kondo Y, Hoshino R, Kobayashi Y, et al. True aneurysms in a saphenous vein graft placed for repair of a popliteal aneurysm: etiologic considerations. Ann Vasc Surg 2004;18:747.
5. Cassina PC, Hailemariam S, Schmid RA, Hauser M. Infrainguinal aneurysm formation in arterialized autologous saphenous vein grafts. J Vasc Surg 1999;29:756-7.
6. Loftus IM, McCarthy MJ, Lloyd A, Naylor AR, Bell PR, Thompson MM. Prevalence of true vein graft aneurysms: implications for aneurysm pathogenesis. J Vasc Surg 1999;29:403-8.
7. Sharples A, Kay M, Sykes T, Fox A, Houghton A. Vein graft aneurysms following popliteal aneurysm repair are more common than we think. Vascular 2015;23:494-7.
8. van Vugt R, Kruse RR, Fritschy WM, Moll FL. Treatment of dilated venous bypass grafts with an expanded polytetrafluoroethylene-covered nitinol endoprosthesis. Vasc Endovascular Surg 2009;43:190-2.
9. Makela JP, DeBoard ZM, Cisek P. Giant aneurysm of in situ saphenous vein graft. Ann Vasc Surg 2017;40:296.e1-4.
10. Saratzis A, Bown MJ. The genetic basis for aortic aneurysmal disease. Heart 2014;100:916-22.
11. Judge DP, Dietz HC. Marfan's syndrome. Lancet 2005;366: 1965-76.
12. Renard M, Callewaert B, Baetens M, Campens L, MacDermot K, Fryns JP, et al. Novel MYH11 and ACTA2 mutations reveal a role for enhanced TGFβ signaling in FTAAD. Int J Cardiol 2013;165:314-21.
13. Iwamoto T, Kimura A, Nakai T, Kanaya K, Ishimaru S. Implications of carotid arteriomegaly in patients with aortic aneurysm. J Atheroscler Thromb 2004;11:348-53.
14. Diwan A, Sarkar R, Stanley JC, Zelenock GB, Wakefield TW. Incidence of femoral and popliteal artery aneurysms in patients with abdominal aortic aneurysms. J Vasc Surg 2000;31:863-9.
15. Lehnert B, Wadouh F. High coincidence of inguinal hernias and abdominal aortic aneurysms. Ann Vasc Surg 1992;6: 134-7.
16. Mcphail I. Abdominal aortic aneurysm and diastasis recti. Angiology 2008;59:736.
17. Takagi H, Sugimoto M, Kato T, Matsuno Y, Umemoto T. Postoperative incision hernia in patients with abdominal aortic aneurysm and aortoiliac occlusive disease: a systematic review. Eur J Vasc Endovasc Surg 2007;33: 177-81.
18. Wilson WR, Schwalbe EC, Jones JL, Bell PR, Thompson MM. Matrix metalloproteinase 8 (neutrophil collagenase) in the pathogenesis of abdominal aortic aneurysm. Br J Surg 2005;92:828.
19. Debasso R, Astrand H, Bjarnegård N, Rydén AA, Sandgren T, Länne T. The popliteal artery, an unusual muscular artery with wall properties similar to the aorta: implications for susceptibility to aneurysm formation? J Vasc Surg 2004;39:836.

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