

Critical arm ischemia—a rare presentation of giant cell arteritis

Ali Hage, MD, MPH,^a and Carman Iannicello, MD, FRCSC,^b *London and Windsor, Ontario, Canada*

ABSTRACT

Giant cell arteritis can rarely present as severe ischemia of the upper limbs. The initial management includes high-dose oral glucocorticoids. However, when patients do not respond to medical therapy, surgical revascularization might be required to reinstitute limb perfusion. We present the case of a 68-year-old woman who had presented with critical arm ischemia that necessitated carotid–brachial artery bypass after initial oral steroid therapy had failed. We have delineated our surgical approach and technical considerations to potentially help increase the long-term patency of the bypass. (*J Vasc Surg Cases and Innovative Techniques* 2021;7:332-4.)

Keywords: Brachial artery; Bypass; Carotid artery; Giant cell arteritis; Ischemia

Giant cell arteritis (GCA) is an autoimmune vasculitis of unknown etiology that affects medium- and large-size arteries. The disease affects women two to three times more frequently than men, and its frequency increases with age, especially after age 50, with an incidence of 15 to 25 cases per 100,000 individuals aged >50 years.¹

Most cases of GCA involve the extracranial branches of the external carotid artery and result in a low-grade fever, malaise, headache, and jaw claudication. In 15% to 20% of patients with GCA, the vasculitis can involve the ophthalmic artery and posterior ciliary arteries, resulting in painless monocular and, more rarely, binocular transient or permanent vision loss.²

Approximately 10% to 15% of patients with GCA will present with upper extremity ischemia caused by stenosis or occlusion of the subclavian and axillary arteries. This can result in a pulseless arm, claudication, rest pain, ulcers, and gangrene of the digits.³ Upper extremity bypass surgery is infrequently performed for GCA, because most patients will improve with the institution of high-dose steroid treatment. Thus, only a few surgical cases have been reported.^{4,5} Angioplasty of the upper extremity vessels affected by GCA has resulted in frequent dissection and a high restenosis rate.⁶

Our patient had presented with severe rest pain and ulcers to the left hand, was not amenable to angioplasty,

and was referred to a rheumatologist, who had prescribed high-dose steroids. However, minimal improvement had occurred after 3 months of medical treatment and, thus, bypass surgery was performed.

CASE REPORT

A 68-year-old woman with a history of dyslipidemia, pituitary adenoma resection, hysterectomy, and remote smoking had presented to the emergency department with a 6-week history of pain, poikilothermia, nonhealing hand ulcerations, and pale discoloration of her left arm that was exacerbated with lifting of the arm. Physical examination revealed absent subclavian, brachial, radial, and ulnar arterial pulses in the left arm. Motor function was intact in both arms; however, her sensation to light touch was reduced in the left hand.

The initial investigation included arterial ultrasound evaluation of her left upper arm, which revealed occlusion of the distal left axillary artery and proximal brachial artery, with only monophasic flow present within the distal left radial and ulnar arteries. Computed tomography angiography (CTA) of the chest revealed smooth, concentric, circumferential wall thickening of the proximal common carotid, subclavian, and axillary arteries bilaterally that was concerning for medium-size vessel vasculitis (Fig. A and B). Angiography revealed no focal stenosis of the renal arteries but did show severe, smooth stenosis of the right axillary artery with near occlusion, with collateral vessels reconstituting the brachial artery distally, multifocal severe stenosis of the left subclavian artery, and occlusion of the left axillary artery with collateral vessels from the subclavian artery reconstituting the brachial artery (Fig. C).

Based on these findings, the patient was referred to rheumatology, which diagnosed severe GCA and initiated a high-dose regimen of oral steroids. GCA was diagnosed on the basis of the patient's symptoms, elevated inflammatory markers (erythrocyte sedimentation rate and C-reactive protein), and CTA findings. Her erythrocyte sedimentation rate was 93 mm/hour, and her C-reactive protein was 32.9 mg/L. However, after 3 months of treatment, the patient had not experienced any significant improvements in her symptoms, and she had continued to have nonhealing ulcers in her left hand. Therefore, given the

From the Division of Vascular Surgery, Department of Surgery, Western University^a; and the Windsor Regional Hospital, Windsor.^b

Author conflict of interest: none.

Correspondence: Carman Iannicello, MD, FRCSC, Division of Vascular Surgery, Windsor Regional Hospital, 1030 Ouellette Ave, Windsor, ON N9A 1E1, Canada (e-mail: cmiannicello@teksavvy.com).

The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

2468-4287

© 2021 The Authors. Published by Elsevier Inc. on behalf of Society for Vascular Surgery. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

<https://doi.org/10.1016/j.jvscit.2021.03.001>

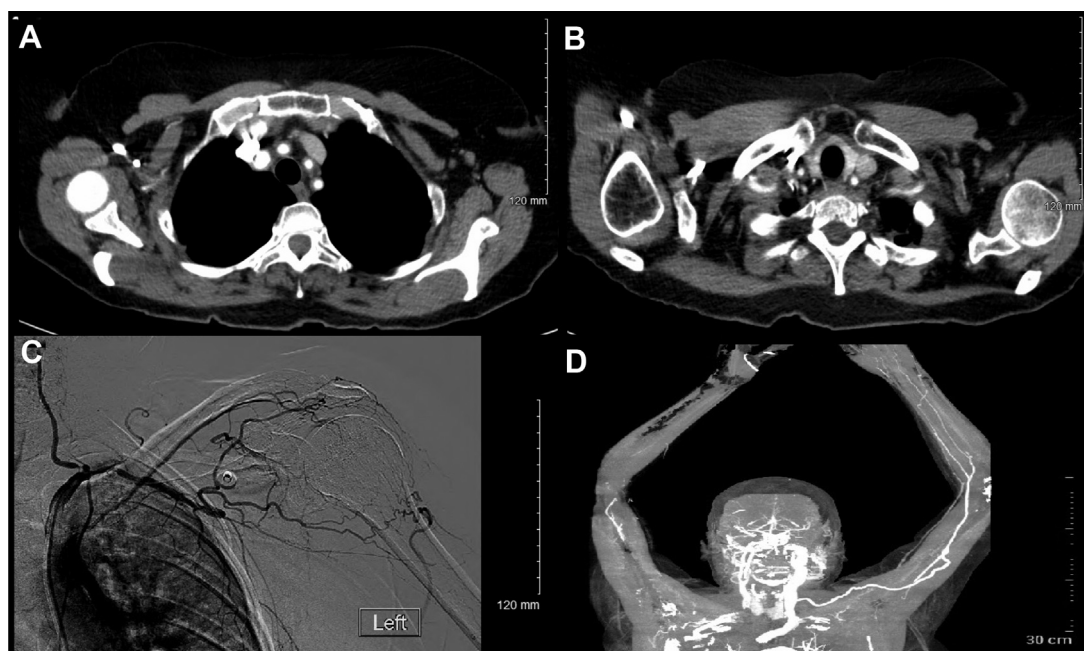


Fig. A. Preoperative computed tomography (CT) angiogram of the chest revealing smooth concentric, circumferential wall thickening of the proximal left common carotid artery. **B.** Preoperative CT angiogram of the chest revealing occlusion of the left axillary artery. **C.** Preoperative angiogram showing multifocal stenosis of the left subclavian artery and occlusion of the left axillary artery, with collateral vessels from the subclavian artery reconstituting the brachial artery. **D.** Postoperative CT angiogram of the chest revealing excellent patency of the ex situ saphenous vein bypass between the left common carotid artery and left brachial artery.

patient's symptoms and that her left upper arm arterial occlusions were not amenable to angioplasty nor stenting, the decision was made to proceed with surgical revascularization in the form of a left common carotid artery to left brachial artery, ex situ, saphenous vein bypass.

Access to the left common carotid artery (ie, the inflow) was obtained between the sternal and clavicular head of the sternocleidomastoid muscle using a 4-cm supraclavicular incision. The left common carotid artery was anastomosed in an end-to-side fashion to a previously harvested long saphenous vein. The mid-brachial artery (ie, the outflow) was isolated by making a transverse incision in the mid-upper arm just below the belly of the biceps brachii muscle. Tunnels were made from the carotid incision down to the left deltopectoral groove, and another tunnel was made from the deltopectoral groove down to the level of the mid left upper arm through which the reversed long saphenous vein was placed and distally anastomosed to the brachial artery in an end-to-side fashion. The mid left common carotid artery and mid left brachial artery anastomotic points were free of "visual" disease. The patient tolerated the procedure well and experienced no postoperative complications. The patient developed a pink, warm hand and had a palpable left radial pulse. Oral prednisone was reinstated, and she was discharged home on postoperative day 6 with instructions to take 81 mg of aspirin daily. The predischarge CTA revealed excellent patency of the bypass (Fig. D). The patient was seen in follow-up at 6 months and continued to do well, with a palpable left radial pulse, intact motor and sensory function, complete

healing of the ulceration, and resolution of the ischemia. However, she has continued to experience the constitutional systemic symptoms of her disease. The patient provided written informed consent for the report of her case.

DISCUSSION

GCA is the most common form of systemic granulomatous vasculitis affecting medium- and large-size arteries in individuals aged ≥ 50 years, with the disease being extremely rare in younger patients.⁷ In a large multicenter, multinational study of 382 patients with GCA, Gribbons et al⁸ reported that patients with GCA were more likely to have bilateral involvement of their axillary and subclavian arteries. The mainstay of treatment includes high-dose oral glucocorticoids, and previous studies have reported a return of peripheral pulsation even in patients who had presented with subclavian artery occlusion.⁹ Surgical revascularization is, therefore, rare, and only a very few cases of revascularization have been reported in which the patients had presented with critical arm ischemia requiring carotid-brachial artery bypass, as was the case for our patient.^{4,5,10} Also, even when significant stenosis of the arteries is discovered, the distal tissue viability will rarely be compromised, even when ischemic symptoms, such as the loss of pulses, are present, because the gradual development of arterial narrowing will have allowed for the establishment of extensive collateral circulation.¹¹

In patients presenting with critical limb ischemia not improving with high-dose steroids, the only two available options include angioplasty or surgical revascularization. When angioplasty is feasible, this option can provide excellent short-term results. However, it has been associated with very high rates of restenosis at mid-term follow-up. Both et al⁶ reported their mid-term follow-up data at 24 months for patients with GCA who had undergone balloon angioplasty of the upper extremity arteries and reported a poor patency rate of only 65%. However, for surgical revascularization, the long-term results are largely unknown, with the longest reported follow-up at 6 months.¹⁰ We also believed that initiating steroids would probably help control the inflammatory process and would subsequently allow us to perform the anastomosis in segments of the arteries that were free of "visual" disease (ie, when the disease was in its quiescent phase),¹¹ which could help increase the long-term patency of the bypass.

REFERENCES

1. Salvarani C, Cantini F, Boiardi L, Hunder GG. Polymyalgia rheumatica and giant-cell arteritis. *N Engl J Med* 2002;347:261-71.
2. Biouesse V, Newman NJ. Ischemic optic neuropathies. *N Engl J Med* 2015;372:2428-36.
3. Berti A, Campochiaro C, Cavalli G, Pepe G, Praderio L, Sabbadini MG, et al. Giant cell arteritis restricted to the limb arteries: an overlooked clinical entity. *Autoimmun Rev* 2015;14:352-7.
4. Almeida-Morais L, Galego S, Marques N, Pack T, Rodrigues H, Abreu R, et al. Acute upper limb ischemia, a rare presentation of giant cell arteritis. *Rev Port Cardiol* 2016;35:237.e1-4.
5. Shibutani S, Obara H, Kan S, Takahashi S, Ogawa K, Kitagawa Y. Atypical giant cell arteritis presenting as lack of a pulse in the upper extremity. *Ann Vasc Dis* 2011;4:241-4.
6. Both M, Aries PM, Müller-Hülsbeck S, Jahnke T, Schäfer PJ, Gross WL, et al. Balloon angioplasty of arteries of the upper extremities in patients with extracranial giant-cell arteritis. *Ann Rheum Dis* 2006;65:1124-30.
7. Crowson CS, Matteson EL. Contemporary prevalence estimates for giant cell arteritis and polymyalgia rheumatica, 2015. *Semin Arthritis Rheum* 2017;47:253-6.
8. Gribbons KB, Ponte C, Carette S, Craven A, Cuthbertson D, Hoffman GS, et al. Patterns of arterial disease in Takayasu's arteritis and giant cell arteritis. *Arthritis Care Res (Hoboken)* 2020;72:1615-24.
9. Swinson DR, Goodwill CJ, Talbot IC. Giant cell arteritis presenting as subclavian artery occlusion: a report of two cases. *Postgrad Med J* 1976;52:525-9.
10. Ratschiller T, Müller H, Pirklbauer M, Silye R, Sulzbacher G, Zierer A. Giant cell arteritis as unusual cause of critical arm ischemia. *J Vasc Surg Cases Innov Tech* 2018;4:248-51.
11. Ponte C, Rodrigues AF, O'Neill L, Luqmani RA. Giant cell arteritis: current treatment and management. *World J Clin Cases* 2015;3:484-94.

Submitted Nov 1, 2020; accepted Mar 4, 2021.