Mycotic internal carotid artery pseudoaneurysm secondary to Mycobacterium tuberculosis

Dennis H. Lui, MBChB,a,b Shreena Patel, MBBS,c Ruhaid Khurram, MBBS,c Michael Joffe, MBChB, MMed,c Jason Constantinou, MBBS, MD,d and Daryl Baker, MBChB, PhD,e London, UK

ABSTRACT

Mycobacterium tuberculosis is a rare causative agent for mycotic aneurysms of the extracranial carotid arteries. We describe a case of acute mycotic pseudoaneurysm and abscess in the right proximal internal carotid artery in close proximity to the carotid bifurcation, and subsequent management with antibiotic therapy, surgical debridement and resection with an end-to-end anastomosis. (J Vasc Surg Cases Innov Tech 2022;8:251-5.)

Keywords: Aneurysm; Myotic; Extracranial carotid artery aneurysm; Lymphadenitis; Carotidynia

Mycotic extracranial carotid artery aneurysms are rare lesions and Mycobacterium tuberculosis is a highly unusual causative organism.1-3 Principles of treatment revolve around targeted antibiotic therapy followed by surgical excision and repair, or endovascular exclusion in a hostile neck. We present a case of an internal carotid artery (ICA) pseudoaneurysm secondary to miliary tuberculosis with an associated caseating tubercular lymphadenitis and large neck abscess. The patient gave their consent for their clinical history and images to be published in this report.

CASE

A 50-year-old African Caribbean man was admitted with symptoms of a large swelling and carotidynia in the right neck. The patient described a history of trauma to the neck 6 months prior, secondary to mild penetrating injury at the site from a thorn bush.

He subsequently developed back and hip pains and decreased mobility, prompting admission to another hospital and a biopsy of the lumbar spine. He was discharged until re-presenting with right neck symptoms. No neurological symptoms were present and other past medical history, social and travel histories were unremarkable. On examination, the patient was thin (body mass index of 18.4), had a persistent tachycardia with hypertension (172/87 mm Hg), and was not jaundiced or cyanosed. A large, painful, indurated and pulsatile mass was present in the right neck, in cervical levels II and III. His white cell count was elevated at 11,500/µL and C-reactive protein was elevated at 88 mg/L.

Computed tomography (CT) angiography of the aortic arch, cervical vessels, and head revealed a 10 × 9 mm bilobed pseudoaneurysm arising from the origin of the right ICA, and an overlying 4 × 3 cm collection (Fig 1). These findings were confirmed with duplex ultrasound and contrast-enhanced magnetic resonance angiography, which identified features of an extensive abscess originating from the right carotid space and extending into the adjacent soft tissues (Fig 2). Fluorodeoxyglucose positron emission tomography imaging illustrated intense uptake from the pseudoaneurysm, lungs, and right sacroiliac joint. Because these findings were highly suspicious for tuberculosis infection, results from the previous spinal biopsies were requested, which confirmed chronic granulomatous inflammation. Polymerase chain reaction tests were positive for M tuberculosis. He received a formal diagnosis of multifocal tuberculosis with cervical and mediastinal lymphadenopathy, L3/L4 facet joint and sacroiliac joint involvement, and paraspinal collection.

The patient was started on targeted antimicrobial treatment (rifampicin, isoniazid, ethambutol, and pyrazinamide, with pyridoxine), and counselled for emergent open repair and surgical debridement of right cervical tuberculous abscess. An autologous long saphenous vein conduit for bypass was not an option owing to the poor quality and size of the patient’s leg veins. The case was performed under general anesthesia with electroencephalographic monitoring and routine invasive hemodynamic monitoring. Significant airway edema was noted upon placement of the endotracheal tube. An oblique incision anterior to the sternocleidomastoid muscle was made, and the right common carotid artery,
internal jugular vein, and external carotid artery were dissected and controlled. The pseudoaneurysm could not be dissected free from surrounding structures owing to extensive inflammation. A large abscess containing 200 mL of pus was drained, and the pseudoaneurysm was opened, with good back-bleeding encountered. The distal ICA was then controlled, allowing resection of the damaged proximal ICA. With the common carotid and internal carotid sufficiently mobilized, the external carotid artery was ligated and divided, and a primary end-to-end anastomosis was performed between the common carotid artery and ICA (Fig 3). Careful washout of the wound was performed and a drain was left in situ. Tissue and pus from the wound were positive by polymerase chain reaction for *M tuberculosis*. Subsequent culture and sensitivities were performed, which showed full sensitivity to antimicrobial treatment.

Postoperatively, the patient reported a significant improvement in pain, with no gross motor or sensory neurological deficit. The cranial nerves were intact save for contralateral deviation in his tongue. A postoperative CT angiogram confirmed patency of the anastomosis and no intracranial changes and aspirin 75 mg once daily was commenced. He was discharged home 4 days after surgery and continued antimicrobial therapy. Pyrazinamide was withdrawn owing to episodes of gout, and moxifloxacin was commenced as a replacement. At the 3-month follow-up, the patient described symptoms of sharp pain around the right face each time he started his meals, which persisted despite consultations with the specialist pain clinic and speech and language therapy. A presumptive diagnosis of first-bite syndrome was made, and the patient was treated with ultrasound-guided botulinum toxin injection around the parotid gland, with moderate relief to his symptoms. The patient had ongoing hyperalgesia of the skin around his scar at the 6-month follow-up. CT angiograms were performed at 2, 6, and 12 months postoperatively, and a magnetic resonance angiogram was performed at the 6-month postoperative follow-up, showing patent carotid anastomosis and no recurrence of pseudoaneurysm. The patient continues to have ongoing surveillance and antimicrobial therapy.

**DISCUSSION**

Extracranial carotid aneurysms account for less than 4% of all peripheral arterial aneurysms. They are most commonly caused by trauma, previous surgery, or atherosclerosis. The first carotid aneurysm was surgically treated by Sir Astley Cooper in 1805, and surgical management remains the mainstay of treatment. More recently, endovascular treatments have also been used to treat extracranial carotid aneurysms, with good short-term results.

Mycotic carotid artery aneurysms, both intracranial and extracranial, are highly unusual lesions and have been associated with bacterial endocarditis, intravenous drug abuse, dental infection, ingested foreign bodies, and bacterial sinusitis. The usual causative organisms of mycotic aneurysms include *Staphylococcus aureus*, *Escherichia coli*, and *Salmonella* species, although *Streptococcus* and *Klebsiella* species have also been implicated. Although historically more prevalent, *M tuberculosis* has been a rare causative pathogen in the formation of mycotic carotid aneurysms for the last century, with only four previous cases reported in literature. Mycobacterial mycotic aneurysms in the carotid artery have also been described after bacillus Calmette-Guerin therapy for bladder cancer.
The pathogenesis of mycotic aneurysms involving *M. tuberculosis* begins with either an (1) external invasion of the bacilli through tunica adventitia and media via seeding from vasa vasorum, adjacent lymph nodes, or adjacent abscess, or rarely (2) direct hematogenous seeding of tuberculous bacilli to the tunica intima (especially around atherosclerotic plaques). Subsequent necrotizing granulomatous inflammation degrades the artery wall integrity, leading to aneurysm or pseudoaneurysm formation. In this case, we believe that the patient with latent tuberculosis developed a reactive lymphadenitis, secondary to penetrating injury to the neck, which subsequently evolved into an abscess and eroded the right ICA.

Mycotic carotid aneurysms are associated with high morbidity and mortality, with potential complications.
including rupture and fatal hemorrhage, fistulation, airway compromise, stroke, cranial nerve palsies, and intracerebral abscesses by hematogenous metastases.\textsuperscript{12,10,16,18,19,21} In patients with an active suppurative infection in the neck, airway edema and compromise may be encountered; therefore, the careful induction of anesthesia and airway management are mandatory. The limited reports of mycotic carotid artery pseudoaneurysms in the literature uniformly support the combined strategy of antibiotic therapy and prompt surgical management.\textsuperscript{1,2,9,14,15} After excision of the damaged artery, different techniques to restore antegrade flow have been described in the literature, including primary end-to-end anastomosis; in situ bypass with autologous deep vein, artery, or synthetic conduit; or repair with patch for very localized lesions. Ligation of the damaged carotid artery is also an option, especially in the emergency setting, but has been associated with poor outcomes.\textsuperscript{1} Although synthetic grafts impregnated with rifampicin could theoretically be used, the implantation of such a graft into an infected surgical field could lead to graft infection and potential anastomotic blow-out. More recently, hybrid and staged management for mycotic carotid aneurysms have been described, where a covered endovascular stent is temporarily deployed to exclude the pseudoaneurysm, followed by an open surgical debridement.\textsuperscript{13,15} In our case, endovascular management was considered carefully, particularly as a potential temporizing measure while antibiotics were initiated. However, we ultimately proceeded with open surgical management and control of sepsis, owing to concerns regarding bacterial embolization and the risk of cerebral abscesses or stent infection. Careful postoperative surveillance is highly important to identify any potential recurrence.

**CONCLUSIONS**

Mycotic carotid aneurysms are highly morbid conditions that require prompt medical and surgical management. Although \textit{M tuberculosis} is a rarely reported causative organism, clinicians should be aware of this condition in patients with any history or suspicion of tuberculosis and a pulsatile neck mass.

The authors acknowledge the assistance of Dr Pencharz, Dr Lucinda Cruddas, Dr Farah Jabeen, Dr Ruwan Weerakkody, Dr Ahmed Helal, and Dr Shahinda Elanwar in the preparation of this article.

**REFERENCES**

1. Jebara VA, Acar C, Dervanian P, Chachques JC, Bischoff N, Uva MS, et al. Mycotic aneurysms of the carotid arteries—case report and review of the literature. J Vasc Surg 1991;14:215-9.
2. Kasangana K, Shih M, Saunders P, Rhee R. Common carotid artery pseudoaneurysm secondary to Mycobacterium tuberculosis treated with resection and reconstruction with saphenous vein graft. J Vasc Surg Cases Innov Tech 2017;3:192-5.
3. Pirvu A, Bouchet C, Garibotti FM, Haupert S, Sessa C. Mycotic aneurysm of the internal carotid artery. Ann Vasc Surg 2013;27:826-30.
4. Li Z, Chang C, Yao C, Guo L, Liu Y, Wang M, et al. Endovascular stenting of extracranial carotid artery aneurysm: a systematic review. Eur J Vasc Endovasc Surg 2011;42:19-26.
5. Attigah N, Kulkens S, Zausig N, Hansmann J, Ringleb P, Hakimi M, et al. Surgical therapy of extracranial carotid artery aneurysms: long-term results over a 24-year period. Eur J Vasc Endovasc Surg 2009;37:127-33.
6. Fankhauser GT, Stone WM, Fowl RJ, O’Donnell ME, Bower TC, Meyer FB, et al. Surgical and medical management of extracranial carotid artery aneurysms. J Vasc Surg 2015;61:389-93.
7. El-Sabrout R, Cooley DA. Extracranial carotid artery aneurysms. Texas Heart Institute experience. J Vasc Surg 2000;31:702-12.
8. Shipley AM, Winslow N, Walker WW. Aneurysm in the cervical portion of the internal carotid artery: an analytical study of the cases...
9. Kenyon O, Tanna R, Sharma V, Kullar P. Mycotic pseudoaneurysm of the common carotid artery: an unusual neck lump. BMJ Case Rep 2020;13:e239821.

10. Budhiraja S, Sagar P, Kumar R, Sharma SC. Mycotic pseudoaneurysm of internal carotid artery induced by skull base osteomyelitis. Otol Neurotol 2019;40:e816-9.

11. Benedetto F, Barillà D, Pipitò N, Deroso G, Cutrupi A, Barillà C. Mycotic pseudoaneurysm of internal carotid artery secondary to Lemierre’s syndrome: how to do it. Ann Vasc Surg 2017;44:423.e13-7.

12. Mazzaccaro D, Stegher S, Occhiuto MT, Malacrida G, Righini P, Tealdi DG, et al. Hybrid endovascular and surgical approach for mycotic pseudoaneurysms of the extracranial internal carotid artery. SAGE Open Med Case Rep 2014;2. 2050313X14558081.

13. Grossi RJ, Onofrey D, Tvetenstrand C, Blumenthal J. Mycotic carotid aneurysm. J Vasc Surg 1987;6:81-3.

14. Wales L, Kruger AJ, Jenkins JS, Mitchell K, Boyne NS, Walker PJ. Mycotic carotid pseudoaneurysm: staged endovascular and surgical repair. Eur J Vasc Endovasc Surg 2010;39:23-5.

15. Eriksen PRG, Hvilsom CB, Hømøe P. Infected “mycotic” aneurysm of the common carotid artery—a differential diagnosis to tumor of the neck. Front Surg 2018;5:75.

17. Patel S, Sharma AK, Meena D, Garg PK, Tiwari S, Chosh TS. Extracranial carotid artery pseudoaneurysm due to Mycobacterium Tuberculosis Asian Cardiovasc Thorac Ann 2020;28:279-81.

18. Geldmacher H, Taube C, Markert U, Kirsten DK. Nearly fatal complications of cervical lymphadenitis following BCG immunotherapy for superficial bladder cancer. Res 2001;68:420-1.

19. Coscas R, Arlet JB, Belhomme D, Fabiani JN, Pouchot J. Multiple mycotic aneurysms due to Mycobacterium bovis after intravesical bacillus Calmette-Guérin therapy. J Vasc Surg 2009;50:1185-90.

20. Long R, Guzman R, Greenberg H, Safneck J, Hershfield E. Tuberculous mycotic aneurysm of the aorta: review of published medical and surgical experience. Chest 1999;315:322-31.

21. Imamura J, Watanabe Y. Multiple brain abscesses associated with a mycotic aneurysm of the left common carotid artery. Case Report J Neurosurg 1986;64:325-7.