Superselective carotid body tumor embolization with platinum-based coils

Kirthi S. Bellamkonda, MSc,[a] Julia Fayanne Chen, MD,[a] Britt Tonnessen, MD,[a] Rahmatullah Rahmati, MD, MPH,[b] *and* Naiem Nassiri, MD,[a] *New Haven, Conn*

ABSTRACT

Resection of Shamblin II and III carotid body tumors can be challenging owing to the potential for significant blood loss. Prophylactic use of liquid embolic agents poses a risk of inflammatory reactions and nontarget embolization. On the other hand, coil embolization has traditionally been limited to cases involving external carotid artery sacrifice. Herein we demonstrate that superselective targeting of tumor-feeding vessels using platinum based, fully detachable packing coils is effective at sustained devascularization of Shamblin II and III carotid body tumors without subsequent inflammation, allowing for a longer interval between embolization and tumor resection, and potentially reducing blood loss without need for ligation or reconstruction of the internal or external carotid artery. (J Vasc Surg Cases and Innovative Techniques 2021;7:1-5.)

Keywords: Carotid body tumor; Coil embolization; Paraganglioma; Platinum coils; Chemodectoma

Carotid body tumors (CBT) are rare neuroendocrine neoplasms that typically arise at the carotid bifurcation, resulting in a characteristic splaying of the internal and external carotid arteries on duplex ultrasound imaging and angiography. CBTs, although rarely malignant or functional, are hypervascular and can be locally invasive, at times encompassing nerves and infiltrating the walls of adjacent arterial walls.[1] Surgical resection is the definitive therapy for these hypervascular tumors. As such, preoperative embolization has been suggested to minimize intraoperative blood loss at the time of excision, particularly for higher grade, more extensive tumors.[2] Commonly used embolotherapeutic techniques in this particular clinical area have been associated with nontarget embolization leading to stroke, difficulty in catheterization of tumor-feeding vessels, and inflammatory reactions causing neovascularization.[3,4] Most agents currently used require surgical intervention within 24 to 48 hours after embolization to minimize revascularization of the tumor and the postembolization fibroinflammatory changes that can further complicate surgical excision. We, herein, demonstrate that prophylactic superselective CBT embolization with mechanically detachable, platinum-based coils adequately addresses safety concerns of embolization with preservation of the external carotid artery (ECA) main trunk; provides durable tumor devascularization foregoing the need for immediate postembolization excision, eliminates postembolization fibroinflammatory reactions, and may minimize intraoperative blood loss.

Consent from the patients was obtained to publish this case.

CASE REPORTS

Patient 1. A 41-year-old male patient presented for evaluation of a neck mass at the carotid bifurcation found on imaging. The patient noted years of episodic neck pain and several syncopal episodes of escalating frequency, associated with the Valsalva maneuver. The patient denied dysphagia and voice changes. Physical examination was without sensorimotor deficits. Urine catecholamines and metanephrines were grossly normal. On contrast-enhanced magnetic resonance imaging, a right-sided ovoid enhancing mass measuring 1.8 $\times$ 2.3 $\times$ 2.9 cm displayed arterial enhancement and was consistent with a Shamblin II CBT. Duplex ultrasound examination demonstrated the classic goblet splaying of the carotid bifurcation consistent with a CBT at the carotid bifurcation without evidence of flow-limiting lesions in the carotid branches.

Pre-resection embolization was performed. Transfemoral, selective right common carotid angiography outlined the contour of a hypervascular tumor within the carotid bifurcation. We identified multiple arterial feeders arising from the ECA, namely, the ascending pharyngeal, facial, lingual, and several other unnamed feeders arising from the proximal ECA trunk itself. Following systemic heparinization, a coaxial system was developed through the existing 5F Berenstein catheter placed within the proximal ECA under angiographic roadmap guidance. This consisted of a Headway Duo 1.6F-tip microcatheter (MicroVention Inc, Aliso Viejo, Calif) over an 0.014" Asahi wire used to superslectively access and catheterize each of the aforementioned

From the Division of Vascular & Endovascular Surgery,[a] and Division of Otolaryngology,[a] Yale School of Medicine.

Author conflict of interest: N.N. is a consultant for and on the speakers' bureau of Penumbra, Inc.

Correspondence: Naiem Nassiri, MD, Division of Vascular & Endovascular Surgery, Yale School of Medicine, 333 Cedar St, New Haven, CT 06510 (e-mail: naiem.nassiri@yale.edu).

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

https://doi.org/10.1016/j.jvscit.2020.10.007

Fig 1. Patient 1's pre-embolization (*right*) and postembolization (*left*) angiogram of the carotid bifurcation, demonstrating a carotid body tumor (CBT).

feeding arterial targets. Through this system, a total of 20 Ruby SMART WAVE and EXTRA SOFT coils (Penumbra Inc, Alameda, Calif) ranging from 1 to 2 mm in width and 1 to 4 cm in length, were positioned and deployed under direct fluoroscopic visualization into the feeding arteries. There was excellent deposition with no evidence of nontarget embolization. Completion cervicocerebral angiogram demonstrated marked tumor devascularization, patent ECA, and no cerebrovascular filling defects (Fig 1).

We performed surgical resection of the CBT 13 days later. Intraoperatively, the tumor was not excessively adherent to the surrounding neurovascular structures and soft tissues. There was no apparent neovascularization or other fibroinflammatory findings. The tumor had also shrunk to 2.5 cm in its largest dimension. In the process of tumor extirpation, as feeder vessels were encounters and ligated, embolization coils were found in the vessel lumen, confirming accuracy of superselective coil deposition. Blood loss was minimal (<50 mL) and no carotid artery ligation or reconstruction was required. There were no perioperative complications. The patient was discharged home on postoperative day 1, recovering uneventfully with no sensorimotor deficits.

Patient 2. A 62-year-old female patient presented for evaluation of odynophagia and dysphagia. She endorsed hot flashes and palpitations. History was notable for hypertension, a cerebral aneurysm for which she underwent clipping twice, and partial hysterectomy. Physical examination was notable for left neck mass, soft and tender to palpation. Urine catecholamines and metanephrines were normal. A computed tomography scan demonstrated a 4.5 × 3.6 × 3.2 cm, heterogeneously enhancing mass splaying the carotid bifurcation, consistent with a Shamblin III CBT.

Pre-resection embolization was performed. Transfemoral, selective right common carotid angiography outlined the contour of a hypervascular tumor within the carotid bifurcation. Catheterization of targets was accomplished as described in case 1. Ruby POD, packing, and SOFT coils (Penumbra Inc) ranging from 2 to 3 mm in width and 1 to 30 cm were deployed under

Fig 2. Patient 2's pre-embolization (*right*) and postembolization (*left*) angiogram of carotid bifurcation, demonstrating a carotid body tumor (CBT).

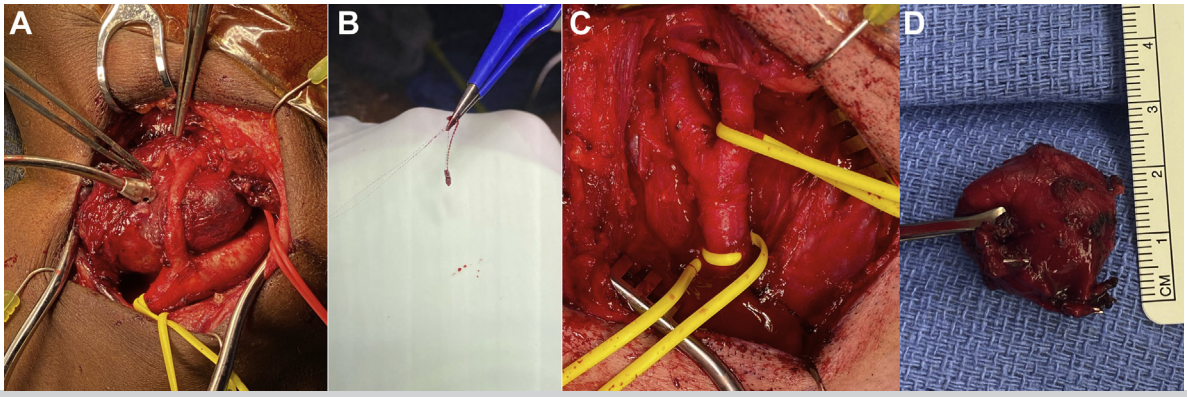

Fig 3. Open resection of carotid body tumor (CBT) in patient 2. **A,** Carotid exposure demonstrating Shamblin III CBT. **B,** Embolization coil, removed from ligated tumor-feeding blood vessel. **C,** Intact carotid bifurcation after tumor resection. **D,** Resected tumor.

fluoroscopic visualization into feeding arteries. There was excellent deposition with no evidence of nontarget embolization. Completion cervicocerebral angiogram demonstrated marked tumor devascularization, without cerebrovascular filling defects (Fig 2). We then performed surgical resection of CBT 6 days later (Fig 3, *A*). There was no apparent neovascularization or other fibroinflammatory findings. When feeder vessels were ligated, embolization coils were found in the vessel lumen (Fig 3, *B*). There was minimal blood loss (<50 mL) and no carotid ligation or reconstruction was needed (Fig 3,*C* and *D*). There were no perioperative complications. The patient was discharged on postoperative day 2 having recovered uneventfully with no sensorimotor deficits.

DISCUSSION

Resection of Shamblin II and III CBTs can be challenging owing to the potential for significant blood loss. This factor can impede dissection, resulting in major vessel or nerve injury and potential for significant morbidity. Tumor vessels may be prophylactically embolized preoperatively, or coils, plugs, and covered stents may be used for branch vessel sacrifice.[2,5-8]

Polyvinylalcohol particles have historically been the predominant embolic agent, with the newer addition of Onyx, n-butyl cyanoacrylate, and coils.[9,10] The advantages of embolization include decreased bleeding and better visualization, as well as tumor shrinkage.[7] However, complications rates from embolization with traditional agents ranges from 2.5% to 3.7%.[11-13] These complications include downstream nontarget embolization and paradoxical emboli.[4,14] Additionally, Onyx and other traditional agents produce intense local inflammation, severely limiting the interval between embolization and resection.[15]

Superselective coil embolization with fully detachable platinum-based coils addresses these risks.[16-18] Platinum coils are inert, offering a longer window between embolization and resection.[19] Polyvinylalcohol and n-butyl cyanoacrylate induce angionecrosis of the tumor wall—inflammation and neovascularity can cause adhesions; resection must be performed in a short window after embolization.[20] Specimens resected 4 days after Onyx embolization demonstrate mononuclear infiltration and intraluminal giant cells.[15] Several authors cite this inflammatory reaction as a limitation on the interval between embolization and resection.[5,11,21] In a meta-analysis of 25 papers, none chose an interval longer than 48 hours.[22] Bercin et al[23] discuss the histologic implications of embolization, postulating that "the hypoxia caused by microparticles may increase the vascular wall stress in the vasa vasorum." They and other investigators postulate that this and increased tumor adherence could result in risk of carotid injury during resection.[24] Despite longer intervals between embolization and resection in our cases, the gross appearance of the tumor was noninflamed and nonadherent, with sustained devascularization. Indeed, compared to average blood loss in other types of CBT embolization (approximately 350 mL), blood loss in both our cases was less than 50 mL and carotid branches were preserved, facilitating a clean dissection and cranial nerve preservation.[2]

In the existing literature, coils have been primarily used in cases involving ECA sacrifice, instead of superselective catherization and embolization of individual feeding branches.[3,25,26] Previous reports of coil embolization have typically described coil placement in larger named arteries such as the ECA itself to occlude the entire vascular region supplying the tumor. Superselective microcatheterization of individual feeding branches has rarely been attempted in coil embolization techniques. In a cohort by Zhang et al,[8] three patients have been reported in whom platinum coils are used as a standalone superselective agent with technical success. Unlike liquid agents, fully detachable coils have greater control and when handled appropriately, can be less likely to be associated with nontarget embolization.[25,27] This greater control can enhance precision of deployment and may mitigate the need for ECA sacrifice at the time of embolization.[3] The precision of our coil placement was underscored by coils discovered in ligated tumor-feeding vessels during open resection.

Platinum-based, fully detachable packing coils are effective at sustained devascularization of Shamblin II/III CBTs without subsequent inflammation, resulting in greatly decreased blood loss and facilitated dissection during resection. The inert metal allows for a longer window between embolization and tumor resection, and the detachable nature allows greater technical precision in superselective targeting of tumor-feeding vessels.

REFERENCES

1. Metheetrairut C, Chotikavanich C, Keskool P, Suphaphongs N. Carotid body tumor: a 25-year experience. Eur Arch Otorhinolaryngol 2016;273:2171-9.
2. Economopoulos KP, Tzani A, Reifsnyder T. Adjunct endovascular interventions in carotid body tumors. J Vasc Surg 2015;61:1081-91.e2.
3. Alqaim M, Puri AS, Vaezi AE, Schanzer A. Carotid body tumor resection utilizing a covered stent graft to enable resection of the tumor en bloc with the internal carotid artery. J Vasc Surg Cases Innov Tech 2019;5:481-4.
4. Gemmete JJ, Pandey AS, Chaudhary N, Potti TA. Paradoxical embolus to the brain from embolization of a carotid body tumor. J Neurointerv Surg 2012;4:e12.
5. Abu-Ghanem S, Yehuda M, Carmel NN, Abergel A, Fliss DM. Impact of preoperative embolization on the outcomes of carotid body tumor surgery: a meta-analysis and review of the literature. Head Neck 2016;38(Suppl 1):E2386-94.
6. Chen KS, Vicenty-Padilla J, Aziz-Sultan MA. Carotid body tumors: pre-operative management and a review of the literature. In: Park MS, Kalani MYS, de Havenon A, McNally JS, editors. Carotid Artery Disease. New York: Springer International Publishing; 2020. p. 247-68.
7. Scanlon JM, Lustgarten JJ, Karr SB, Cahan JI. Successful devascularization of carotid body tumors by covered stent placement in the external carotid artery. J Vasc Surg 2008;48:1322-4.
8. Zhang J, Fan X, Zhen Y, Chen J, Zheng X, Ma B, et al. Impact of preoperative transarterial embolization of carotid body tumor: a single center retrospective cohort experience. Int J Surg 2018;54:48-52.
9. Borges LF, Heros RC, DeBrun G. Carotid body tumors managed with preoperative embolization: report of two cases. J Neurosurg 1983;59:867-70.
10. Kalani MYS, Ducruet AF, Crowley RW, Spetzler RF, McDougall CG, Albuquerque FC. Transfemoral transarterial Onyx embolization of carotid body paragangliomas. Neurosurg 2013;72:9-15.
11. Britt CJ, Mydlarz WK. Should Pre-operative Embolization Be Utilized Routinely for Carotid Body Tumors Prior to Surgical Excision? In: Gooi Z, Agrawal N, editors. Difficult Decisions in Head and Neck Oncologic Surgery. New York: Springer International Publishing; 2019. p. 171-83.
12. Griauzde J, Gemmete JJ, Chaudhary N, Pandey AS, Sullivan SE, McKean EL, et al. A comparison of particulate and Onyx embolization in preoperative devascularization of carotid body tumors. Neuroradiol 2013;55:1113-8.
13. Power AH, Bower TC, Kasperbauer J, Link MJ, Oderich G, Cloft H, et al. Impact of preoperative embolization on outcomes of carotid body tumor resections. J Vasc Surg 2012;56:979-89.

14. Smith TP. Embolization in the external carotid artery. J Vasc Interv Radiol 2006;17:1897-913.
15. Jahan R, Murayama Y, Gobin YP, Duckwiler GR, Vinters HV, Viñuela F. Embolization of arteriovenous malformations with Onyx: clinicopathological experience in 23 patients. Neurosurg 2001;48:984-97.
16. Chiu Y-J, Hsu H-L, Lai S-T, Hsu C-P. Hybrid treatment of a carotid body tumor. Interdiscip Neurosurg 2017;8:75-7.
17. Hans SS. Resection of a malignant carotid body tumor with carotid artery resection, Hans SS. Challenging Arterial Reconstructions. New York: Springer International Publishing; 2020. p. 135-8.
18. Zhang T-h, Jiang W-l, Li Y-l, Li B, Yamakawa T. Perioperative approach in the surgical management of carotid body tumors. Ann Vasc Surg 2012;26:775-82.
19. Ohyama T, Nishide T, Iwata H, Sato H, Toda M, Taki W. Vascular endothelial growth factor immobilized on platinum microcoils for the treatment of intracranial aneurysms: experimental rat model study. Neurol Med Chir 2004;44:279-87.
20. Sheth RA, Sabir S, Krishnamurthy S, Avery RK, Zhang YS, Khademhosseini A, et al. Endovascular embolization by transcatheter delivery of particles: past, present, and future. J Funct Biomater 2017;8:12.
21. Davis FM, Obi A, Osborne N. Carotid body tumors. In: Hans SS, editor. Extracranial Carotid and Vertebral Artery Disease. New York: Springer International Publishing; 2018. p. 253-60.
22. Texakalidis P, Charisis N, Giannopoulos S, Xenos D, Rangel-Castilla L, Tassiopoulos AK, et al. Role of preoperative embolization in carotid body tumor surgery: a systematic review and meta-analysis. World Neurosurg 2019;129:503-13.e2.
23. Bercin S, Muderris T, Sevil E, Gul F, Kılıcarslan A, Kiris M. Efficiency of preoperative embolization of carotid body tumor. Auris Nasus Larynx 2015;42:226-30.
24. Dickinson P, Griffin S, Guy A, McNeill I. Carotid body tumour: 30 years experience. BJS 1986;73:14-6.
25. Ong H, Fan X, Ji T. Radical resection of a Shamblin type III carotid body tumour without cerebro-neurological deficit: improved technique with preoperative embolization and carotid stenting. Int J Oral Maxillofac Surg 2014;43:1427-30.
26. Zhang H, Li F, Zheng Y. An aneurysm-like entity inside a giant carotid body tumor reaching lateral skull base. Translational Surgery 2019;4:32.
27. Janakiram T, Nadakkavukaran J, Sharma SB, Sathyanarayanan J. A hybrid approach towards successful resection of a huge carotid body paraganglioma using coil embolization and traditional surgical techniques. Indian J Otolaryngol Head Neck Surg 2019;71:136-9.

Submitted Aug 8, 2020; accepted Oct 6, 2020.