

## Percutaneous retrograde left external carotid artery coil embolization for management of hemorrhage from a persistent proatlantal intersegmental artery type 2

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### ABSTRACT

Persistent fetal communications between the carotid and vertebrobasilar systems are rare and most often discovered incidentally. We present the case of a patient with oropharyngeal cancer status post chemotherapy, radiation therapy, and surgical resection who developed acute oropharyngeal hemorrhage on postoperative day 36, originating from branches of the ligated external carotid artery stump by retrograde flow through a proatlantal intersegmental artery type 2. This hemorrhage was successfully controlled with coil embolization through percutaneous access of the external carotid artery without recurrence at 1-year follow-up. (*J Vasc Surg Cases and Innovative Techniques* 2020;6:250-3.)

**Keywords:** Carotid artery; Coil embolization; Oropharyngeal cancer; Hemorrhage

Grouped together, oral and pharyngeal cancers are the sixth most common cancers in the world, with an annual incidence of roughly 275,000 for oral cancers and 130,300 for pharyngeal cancers, excluding the nasopharynx.<sup>1</sup> Squamous cell carcinoma is estimated to be present in >90% of oral neoplasms. Even with advances in medicine, surgery, and radiotherapy, the mortality remains high at roughly 50% survival at 5 years.<sup>2</sup> Treatment is dependent on stage, with some combination of surgery, radiotherapy, or chemotherapy and radical neck dissection required in those with cervical lymph node involvement.<sup>2</sup> We present the case of a patient who failed to respond to initial chemoradiation for stage 2 human papillomavirus-related squamous cell carcinoma of the left tonsil and underwent subsequent operative resection only to present 36 days later with acute oropharyngeal hemorrhage. The patient's consent was obtained for publication of this report.



**Fig 1.** Selective angiogram of left common carotid artery (CCA) demonstrating occlusion of distal CCA with no contrast material filling of the internal carotid artery (ICA) or external carotid artery (ECA).

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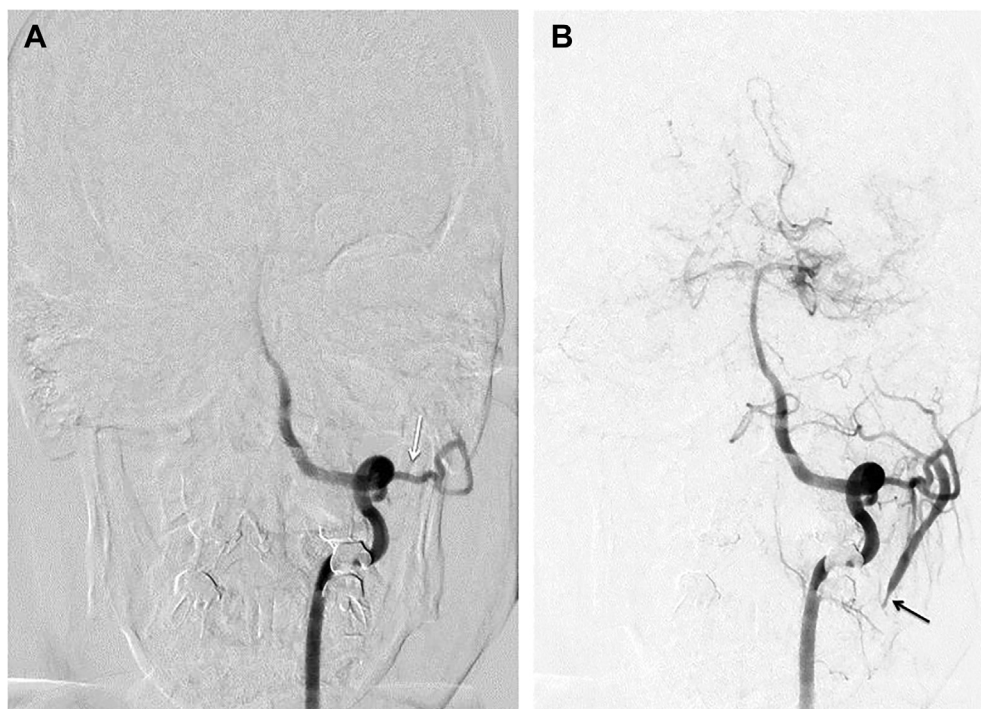
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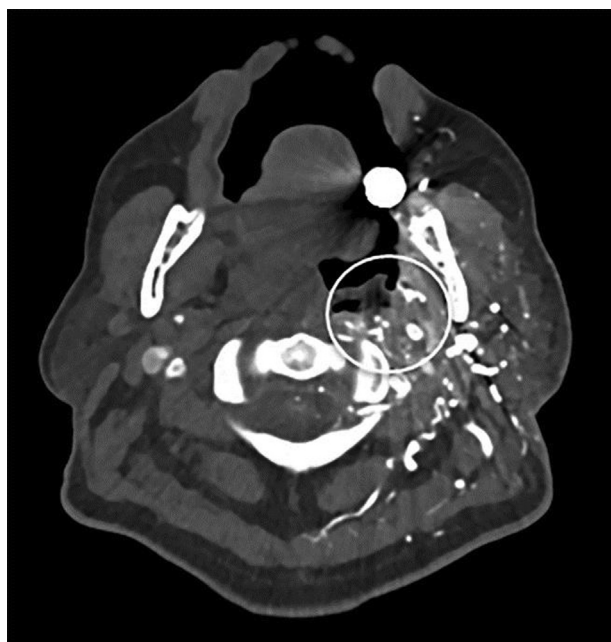
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### CASE REPORT

The patient is a 64-year-old man with T2N2b (stage 2) human papillomavirus-related squamous cell carcinoma of the left tonsil who, after failed chemoradiation, underwent salvage surgical resection involving transoral partial left pharyngectomy and glossectomy along with left-sided neck dissection (levels 2-4), necessitating ligation of the external carotid artery (ECA) origin. His recovery was uneventful before discharge, but he returned on postoperative day 36 with initially small- and subsequently large-volume



**Fig 2.** Selective left vertebral angiogram demonstrating persistent proatlantal intersegmental artery type 2 (**A**: arrow), which terminates as an irregular stump of the external carotid artery (ECA; **B**).



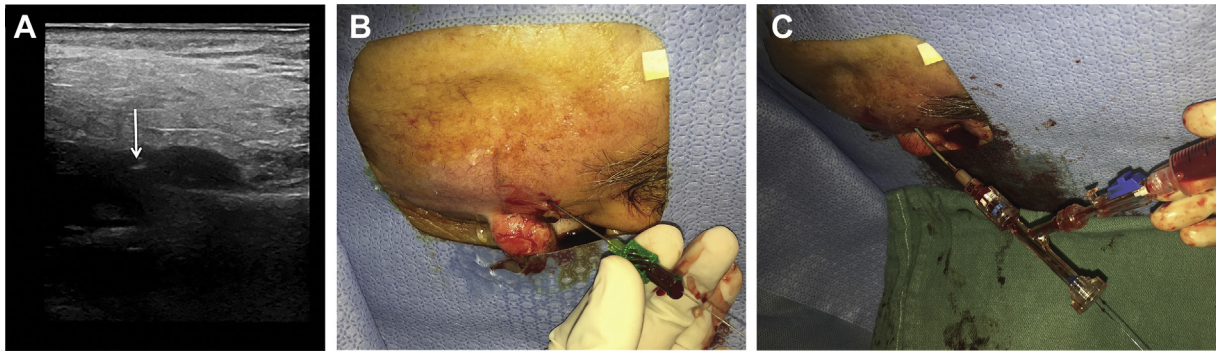
**Fig 3.** Intraoperative computed tomography angiography image demonstrating extravasation of contrast material at the site of surgical ligation of the external carotid artery (ECA).

oropharyngeal bleeding. Because of sudden onset of large-volume hemorrhage, digital pressure was applied transorally, followed by emergent awake tracheostomy for airway protection. Direct laryngoscopy revealed necrotic left stylohyoid

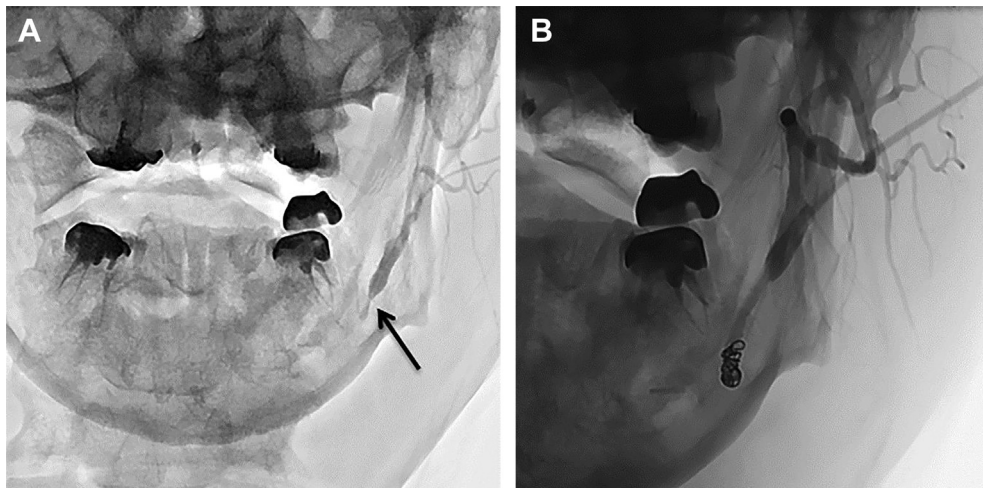
ligament with surrounding soft tissue necrosis and profuse arterial bleeding, which was packed for hemostasis. The vascular surgery service was consulted intraoperatively, and the patient was then transferred directly to a hybrid Miyabi Angio-CT (Siemens Healthcare, Hoffman Estates, Ill) operating room with capabilities of fluoroscopy (zeego C-arm) and computed tomography scan (SOMATOM Edge scanner) for further evaluation and treatment.

Femoral access was obtained under ultrasound guidance, followed by arch angiography and right common carotid artery (CCA) selective catheterization, which revealed brisk cross-filling of the left anterior and middle cerebral arteries. However, it did not reveal a visible bleeding source. The left CCA was selected with angiography, revealing occlusion of the distal CCA and internal carotid artery (ICA), with no contrast material noted in the distal ICA or ECA (Fig 1). A left vertebral selective angiogram demonstrated a variant communication between the left vertebral artery and ECA, which is a rare fetal circulation remnant described as persistent proatlantal intersegmental artery type 2.

The persistent proatlantal intersegmental artery was filling the left ECA trunk in retrograde fashion, which terminated as an irregular stump with vasospasm (Fig 2). Intraoperative computed tomography angiography was then performed, demonstrating an arterial blush at the previous surgical site from proximal ECA branches (Fig 3). Coil embolization of the left CCA was performed to prevent potential recanalization and embolic stroke. After consensus review, ECA trunk embolization was recommended. We considered the option of



**Fig 4.** Intraoperative images demonstrating ultrasound-guided percutaneous access (A) of the left external carotid artery (ECA) with a micropuncture needle (B) with subsequent 5F micropuncture sheath placement and Tuohy-Borst valve with side injection port (C) to allow contrast angiography followed by coil embolization.



**Fig 5. A,** Angiogram obtained through percutaneous access demonstrating irregular termination (*arrow*) of the left external carotid artery (ECA) stump. **B,** Completion angiogram demonstrating successful coil embolization without evidence of extravasation.

retrograde ECA embolization with a microcatheter through the proatlantal branch as more time demanding and higher risk for left vertebral artery complications, especially in the absence of therapeutic anticoagulation and left vertebral dominance. We considered embolization of the ECA trunk to control the bleeding. Therefore, the left ECA was accessed percutaneously under duplex ultrasound guidance through the parotid gland (Fig 4), and through the microcatheter we proceeded with angiography and retrograde coil embolization of the irregular 2-cm ECA trunk, ablating flow to the regional branches and providing prompt control of arterial extravasation (Fig 5). All packings were removed intraoperatively, and hemostasis was confirmed. Given the occluded left CCA-ICA and confirmed brisk ipsilateral cerebral flow through intracranial collaterals, we proceeded with coil embolization of the left CCA to prevent potential future recanalization and embolic stroke. The patient was extubated and awoke neurologically intact in the intensive care unit. He remains neurologically intact without recurrent bleeding at 1-year follow-up.

## DISCUSSION

Aberrant communications between the carotid and vertebrobasilar systems are the result of continued connections that formed during embryonic development to supply circulation to the posterior segment of the brain.<sup>3-6</sup> Four such named connections between the carotid and vertebrobasilar system exist: otic, hypoglossal, trigeminal, and proatlantal intersegmental arteries.<sup>6</sup> These aberrant pathways between the carotid and vertebrobasilar circulations begin forming around the 4-mm embryo stage, with most involuting by the 12-mm embryo stage.<sup>3-5,7</sup> Of these anomalous connections, the most common is the persistent trigeminal artery, which is noted to have an estimated incidence of 1%.<sup>7</sup> The persistent proatlantal intersegmental artery, first described at autopsy in the 19th century, exists as two variants, type 1 and type 2.<sup>5</sup> Type 1 persistent proatlantal intersegmental arteries arise from the ICA before connecting with the vertebrobasilar system, which is in

comparison to the type 2 persistent proatlantal intersegmental artery, which arises from the ECA.<sup>6</sup> In most cases, these anomalies are discovered incidentally. However, persistent proatlantal intersegmental arteries can be of clinical significance if they result in ischemia or, as in this case, become a source of life-threatening hemorrhage.<sup>6</sup>

Although roughly 40 cases of persistent proatlantal intersegmental arteries have been reported in the literature, none have presented with postoperative bleeding through retrograde flow from a proatlantal intersegmental artery of either type 1 or type 2.<sup>7</sup> In this case, hemorrhage was noted from the left proximal ECA branches perfusing retrograde through a proatlantal intersegmental artery type 2 in the setting of surgically ligated ECA origin, eliminating a transfemoral option for antegrade ECA embolization. The alternative option of retrograde ECA embolization with a microcatheter through the left vertebral-proatlantal intersegmental arteries was considered relatively time demanding, increasing the risk of thrombosis in the dominant left vertebral artery because of lack of anticoagulation in the setting of active bleeding. Given these risks, it was decided that a more direct approach to the left ECA for retrograde embolization should be attempted first, reserving the vertebral pathway in the scenario of persistent or recurrent bleeding. This ultrasound-guided approach provided rapid percutaneous access to the left ECA and prompt control of the hemorrhage from the ECA branches without perioperative complications. Whereas the literature does not specifically denote the complication rate of transparotid access to the ECA, there are reviews noting that the overall complication rate for parotid gland biopsies is low, with rare cases of facial nerve branch injury and <1.7% chance of hematoma,<sup>8-10</sup> all of which may be lowered with appropriate experience in use of ultrasound guidance. The patient has remained neurologically intact and free from recurrent oropharyngeal bleeding at 1-year follow-up.

## CONCLUSIONS

Life-threatening oropharyngeal bleeding may be challenging to control in oncologic patients, especially in the presence of aberrant carotid-vertebrobasilar anatomy. Although rare, anatomic variants should be considered and may require a multimodality approach for effective bleeding control in patients with head and neck malignant neoplasms.

## REFERENCES

1. Warnakulasuriya S. Global epidemiology of oral and oropharyngeal cancer. *Oral Oncol* 2009;45:309-16.
2. Markopoulos AK. Current aspects on oral squamous cell carcinoma. *Open Dent J* 2012;6:126-30.
3. Basekim CC, Silit E, Mutlu H, Pekkalı MZ, Ozturk E, Kizilkaya E. Type I proatlantal artery with bilateral absence of the external carotid arteries. *AJNR Am J Neuroradiol* 2004;25:1619-21.
4. Gumus T, Onal B, Ilgit ET. Bilateral persistence of type 1 proatlantal arteries: report of a case and review of the literature. *AJNR Am J Neuroradiol* 2004;25:1622-4.
5. Purkayastha S, Gupta AK, Varma R, Kapilamoorthy TR. Proatlantal intersegmental arteries of external carotid artery origin associated with Galen's vein malformation. *AJNR Am J Neuroradiol* 2005;26:2378-83.
6. Zarghouni M, Marichal D. Persistent bilateral proatlantal type II artery. *Proc Bayl Univ Med Cent* 2013;26:50-1.
7. Luh GY, Dean BL, Tomsick TA, Wallace RC. The persistent fetal carotid-vertebrobasilar anastomoses. *AJR Am J Roentgenol* 1999;172:1427-32.
8. Witt BL, Schmidt RL. Ultrasound-guided core needle biopsy of salivary gland lesions: a systematic review and meta-analysis. *Laryngoscope* 2014;124:695-700.
9. Schmidt RL, Hall BJ, Layfield LJ. A systematic review and meta-analysis of the diagnostic accuracy of ultrasound-guided core needle biopsy for salivary gland lesions. *Am J Clin Pathol* 2011;136:516-26.
10. Haldar S, Sinnott JD, Tekeli KM, Turner SS, Howlett DC. Biopsy of parotid masses: review of current techniques. *World J Radiol* 2016;8:501-5.

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