Rare Complications and Atypical findings with Embolization for Carotid Body Tumors: A Review of Three Cases

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Background

- Carotid body tumors (CBTs) account for approximately 60–70% of all head and neck paragangliomas and arise at the bifurcation of the carotid artery.
- CBT resection sometimes necessitates carotid reconstruction and hypervascularity of the tumors presents feared risks of vascular and neurological complications
- Digital Subtraction Angiography (DSA) with embolization and possible balloon test occlusion (BTO) is useful for selected tumors and is associated with uncommon yet substantial potential morbidity and remains somewhat controversial in the literature

Case Presentations

Case 1:43 YO male who presented with a neck mass. Patient had negative genetic testing, and biochemical evaluation was normal. Patient had CT and MRI of the neck, which showed a 3.9 cm mass centered in the left carotid bifurcation. Decision was made to pursue surgical resection with preoperative embolization. Embolization was uneventful. Right radial artery was utilized for access for the procedure. Balloon test occlusion (BTO) of the internal carotid (ICA) was NOT performed and the L ascending pharyngeal artery was successfully embolized. Surgery was successful, with gross total resection with negative pathological margins. Post-operative recovery was complicated by first bite syndrome, which resolved fully, and asymptomatic R radial artery occlusion.

Case 2: 45 YO female who presented with a neck mass. Patient had negative genetic testing, and biochemical evaluation was normal. Patient had CT, MRI, and DOTATE of the neck, which showed a 2.7 cm mass in the L carotid area. Decision was made to pursue surgical resection with embolization. Successful left embolization using Onyx (Medtronic, MN, USA) of the ascending pharyngeal artery utilizing R wrist access. BTO was then performed. After approximately 10 minutes of carotid occlusion, the patient developed subtle asymmetry in the left half of the face indicating failure of BTO. The following day, it was noted the patient had developed a left-sided Horner's syndrome. Decision was made to postpone the CBT resection. Ophthalmology was consulted and recommended no acute intervention. Horner's resolved. She had a gross total resection 4 months later, with no post-operative complications. Embolic material was seen in vasa vasorum if the ICA.

Case 3: 44 YO female with SDHB mutation who had a carotid body tumor with apparent retropharyngeal extension. Successful onyx embolization of the carotid body tumor but failed BTO of Right ICA due to hemiparesis occurred. At surgery, the retropharyngeal mass was thought to be a lymph node. Excision revealed a separate focus of paraganglioma. This did not receive embolic material, but the carotid body tumor did. It was in very close proximity to the sympathetic trunk, but the patient had no Horner's or first bite syndrome postoperatively. The second, separate focus was thought to potentially represent a metastatic lymph node, though no lymphocytes, macrophages, or lymphoid tissue were seen in the specimen, which would be seen in a lymph node. This made the conclusion likely that this was a second, separate paraganglioma.

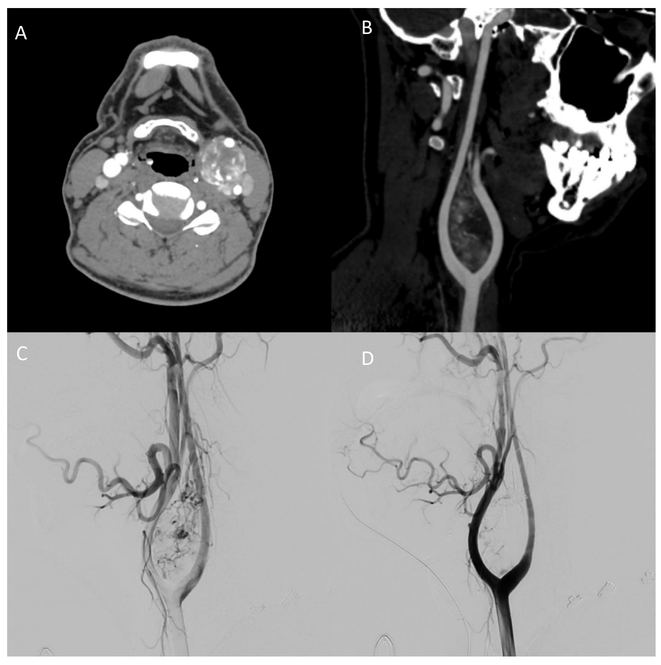
Conclusions

- Angiography and BTO and embolization clearly play a role in carotid body tumor surgery.
- The decision for surgery, and in turn angiography, is complex. Our algorithm at this time is to offer this to most patients but not require it unless we believe the risk of surgery without it is too high and selection criteria relate to size, classification and growth pattern of the tumor on cross-sectional imaging.
- Our policy is to do the angiography the day prior to surgery so we can be certain the procedure is uncomplicated and allows for full reversal of heparinization from the cerebral angiogram and time for embolic material to occlude the selected vessels Had we done the embolization at the same OR session as the surgery, we would not be sure the Horner's was not due to surgery which would seem to validate this policy.
- Reasons for angiography, other than embolization which can reduce bleeding risk, would be to better understand the vascular anatomy, and to do BTO to evaluate risk if vascular repair or reconstruction is required. Patients should be counseled about the risks of angiography including the risk of access in the wrist or groin and patients who undergo wrist access should undergo Allen's test prior.
- Because of a thorough multidisciplinary team approach, which includes otorhinolaryngology, vascular surgery, nephrology, interventional neuroradiology, and pathology, all patients in this group had no additional morbidities and recovered fully. Review of our experience over decades confirmed that these occurrences are rare in our institution prompting more thorough analysis.

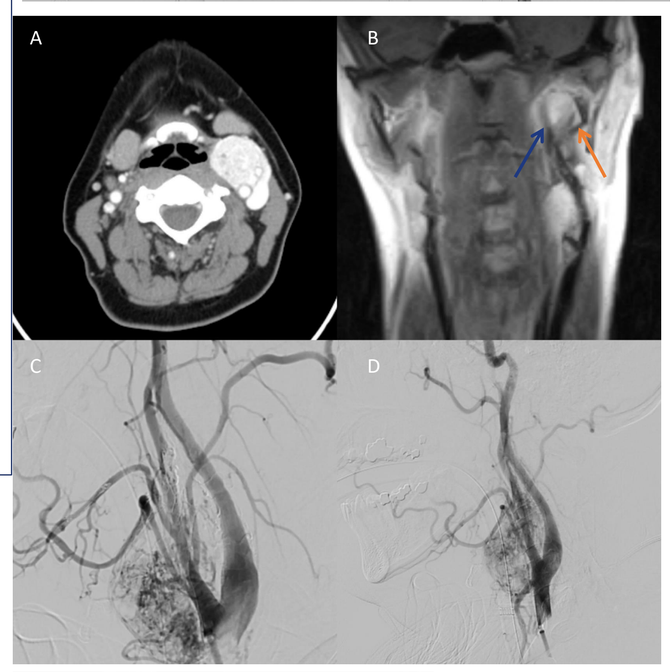
Discussion

These cases illustrate the complexity of decision-making in paraganglioma surgery. While angiography provides important anatomical information, it has known risks. Numerous systemic reviews and meta-analyses have been done and have come to different conclusions about the decision to perform embolization for carotid body tumors.[1-4] In addition, test occlusion adds additional risks but provides important information about risk of stroke in the event there is carotid artery injury during the procedure.[5]

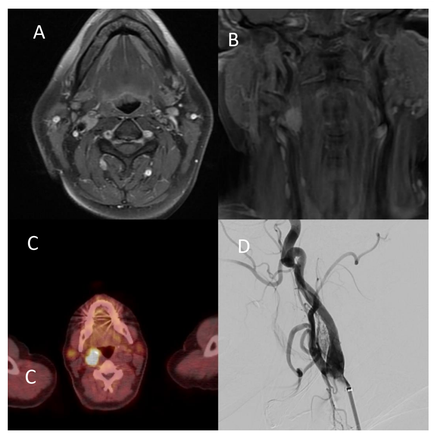
- Our practice has been to offer embolization to most patients who undergo carotid body tumor surgery, and this is the primary reason for angiography. Larger tumors, Shamblin II and III tumors and those that have atypical cross-sectional imaging are ideal candidates for angiography and embolization. [6] In most cases, we then advise patients to consider test occlusion if embolization is performed. Angiographic techniques and embolic material have evolved significantly over the last several decades. Currently, onyx is favored and has some very specific features conferring advantages over other embolic material.
- Test occlusion may predict the risk of complications but has some false negatives[7],so in cases where concerns about carotid artery patency may arise, we also employ EEG and SSEP monitoring intraoperatively.
- Vascular access for angiography has some risks of local complications in the groin or the arm. Traditional femoral access for angiography is associated with some significant morbidity and femoral access restricts mobility significantly.[8]
- More recently, access via the radial artery has become a popular alternative but has a steep learning curve.[5,6] Generally, the right radial artery is preferred for access for interventional neuroradiology. [8-11]
- Radial artery occlusion occurs in 5.5-7.7% of transradial procedures and results from acute arterial thrombosis due to catheter-related endothelial injury, local hypercoagulable state, and decreased blood flow from compressive hemostasis. Understanding this risk, it is very important to perform Allen's test prior to utilizing the radial artery for access of for any other procedure.[11]
- First bite syndrome (FBS) occurs in 7-40% of carotid body tumor cases and is believed to result from sympathetic denervation of the parotid gland, causing severe cramping pain with the first bite of each meal that diminishes with subsequent bites. Most FBS resolves over time. [12-15]
- Horner syndrome can rarely be associated with carotid body tumors on presentation and may occur after treatment.[16] In our case, it developed after test occlusion, but it is unclear if the test occlusion or the embolization was the cause. We chose to observe the patient for recovery and stage the procedure to another time understanding the risk of losing the benefit of the embolization.



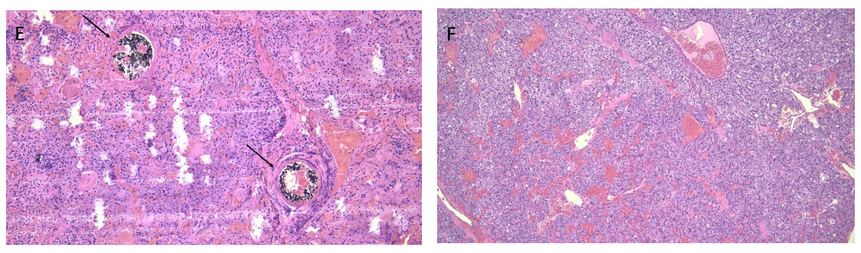
Case 1: Figure 1A) CT axial showing a heterogeneous mass, with regions of prominent enhancement centered around the carotid bifurcation. The mass splays and partially encases internal and external carotid arteries, over 180 degrees
1B) CTA sagittal of neck shows widely patent great vessels, with no carotid stenosis
1C) Pre-embolization DSA of left carotid artery, showing prominent tumor blush signifying frank vascularity
1D) Tumor blush post-embolization of the left ascending pharyngeal artery. Vascularity is significantly reduced



Case 2: Figure 2A) CT axial showing the tumor to be Shamblin II classification carotid body tumor with less than 270 degrees of encasement of the carotid artery and displacement of the submandibular gland and extension medial and deep to the external carotid artery and displacement of the internal carotid artery and internal jugular vein.
2B) Coronal MRI showing the relationship to the external carotid artery (orange arrow) and internal carotid artery (blue arrow)
2C) Pre-embolization DSA of left carotid artery showing vascularity
2D) Post-embolization of the lingual artery and ascending pharyngeal artery.



Case 3: Figure 3A) MRI axial demonstrating a hyperintense lesion centered around the right carotid bifurcation. The second mass cannot be demarcated.
3B) MRI Coronal showing tumor relationship to the external and internal carotid artery.
3C) PET CT Dotatate scan with a hyperdense mass abutting the right carotid space. There is some subtle asymmetries to the shape, which upon a retrospective review, could be representative of the two tumors relation to each other.
3D) Pre-embolization DSA of right carotid artery showing vascularity of a Shamblin I classification tumor



3 EF) H&E of Carotid Body Tumor (Left). Vessels surrounding and within the tumor are filled with embolic material (arrows). Second tumor (right) lacks embolic material. The absence of lymphoid tissue in the background argues against nodal metastasis

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