

Case Report

Intratumoral pseudoaneurysm in a carotid body tumor

Valeria Ortega¹, Julio Calderon¹, Fernando Ahumada², Alejandro José Quiroz Alfaro³, Orlando Diaz²

¹Department of Radiology, Baylor College of Medicine, ²Department of Interventional Neuroradiology, Houston Methodist Hospital, Houston, Texas,

³Department of Internal Medicine, North Mississippi Medical Center, Tupelo, Mississippi, United States.

E-mail: *Valeria Ortega - vortega730@unab.edu.co; Julio Calderon - julio.calderon@bcm.edu; Fernando Ahumada - fjahumada7@gmail.com; Alejandro José Quiroz Alfaro - quirozq@hotmail.com; Orlando Diaz - odiaz@houstonmethodist.org



*Corresponding author:

Valeria Ortega,
Department of Radiology,
Baylor College of Medicine,
Houston, Texas, United States.

vortega730@unab.edu.co

Received: 15 August 2024

Accepted: 05 November 2024

Published: 29 November 2024

DOI

10.25259/SNI_693_2024

Quick Response Code:



ABSTRACT

Background: The occurrence of secondary vascular pathology in paragangliomas is extremely rare, particularly in carotid body tumors (CBTs).

Case Description: A 73-year-old female presented with computed tomography angiography revealing a right CBT with a large intratumoral contrast collection. Digital subtraction angiography confirmed a CBT with an internal carotid artery (ICA) intratumoral pseudoaneurysm. Interventional neuroradiology performed coil embolization of the pseudoaneurysm, and a covered stent was placed in the ICA. Surgical intervention was deferred, and the patient was closely monitored with Doppler ultrasound.

Conclusion: Diagnostic imaging is crucial in the management of CBT, offering detailed assessment of tumor morphology and their anatomical relationships, particularly in identifying coexisting vascular anomalies. Recognition of vascular pathologies, such as intratumoral pseudoaneurysms, is critical, as failure to do so could lead to life-threatening complications, including severe bleeding during surgical resection. In cases where surgical resection is high risk due to tumor classification or patient comorbidities, palliative endovascular treatment presents a viable alternative. This approach not only reduces tumor vascularity and provides symptomatic relief but also minimizes procedural risks. While surgical resection remains the gold standard for complete tumor removal, embolization serves as an option in complex or inoperable cases and may also be used to reduce intraoperative risks when surgery is feasible.

Keywords: Angiography, Carotid body tumor, Embolization, Intratumoral pseudoaneurysm, Paraganglioma

INTRODUCTION

The carotid body functions as an arterial chemoreceptor and accounts for the largest paraganglia of the head and neck. Carotid body tumor (CBT) is a neuroendocrine neoplasm, mostly nonfunctional. It can cause symptoms by exerting pressure and invading the nearby tissue, thereby affecting cranial nerves IX, X, or XII and may also involve the sympathetic chain. The definitive treatment is surgical resection; however, the current treatment includes conservative treatment with active surveillance or radiotherapy to be considered in cases where morbidity is high.^[4] The proximity to the carotid arteries, the anatomic relationship with cranial nerves, and possible intratumoral abnormalities warrant an accurate characterization of these masses with diagnostic imaging before definitive treatment.^[1] The diagnosis of CBT involves Doppler ultrasound, magnetic resonance angiography, computed tomography angiography (CTA), and angiography. We present a unique case of CBT with a secondary vascular pathology.

This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, transform, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

©2024 Published by Scientific Scholar on behalf of Surgical Neurology International

CASE REPORT

A 73-year-old female presented with a 10-year history of a progressively enlarging right neck mass associated with sensations of pressure and occasional shortness of breath. Her past medical history is significant for diabetes, hyperlipidemia, and a right thyroid lobectomy for papillary thyroid carcinoma. Due to her prior history of thyroid cancer, a biopsy of the neck mass was performed. The biopsy was negative for metastatic thyroid malignancy but positive for paraganglioma. The patient did not experience any complications related to catecholamine secretion following the biopsy. CTA showed an enhancing, hypervascular, and lobulated soft-tissue density mass consistent with a CBT [Figure 1a]. In addition, a large intratumoral contrast collection was present, suggesting an intratumoral pseudoaneurysm [Figure 1b].

The Shamblin classification, which categorizes CBTs based on their relationship to the carotid vessels, provides valuable insights into surgical risk and complexity.^[11] This mass was classified as Shamblin Class III, indicating extensive encasement of the carotid arteries – in this case, the internal carotid artery (ICA) – which makes surgical resection more challenging and increases the risk of complications, such as cranial nerve injury, stroke, and bleeding.^[12] Due to the tumor's size and its Shamblin classification, surgical resection was considered high risk, and palliative endovascular treatment was considered as an alternative.

During intraprocedural angiography, injection of the right common carotid artery showed splaying of the right external carotid artery (ECA) and ICA secondary to the CBT [Figures 2a and b]. Injection of the right ECA demonstrated a hypervascular tumoral blush consistent with a paraganglioma,

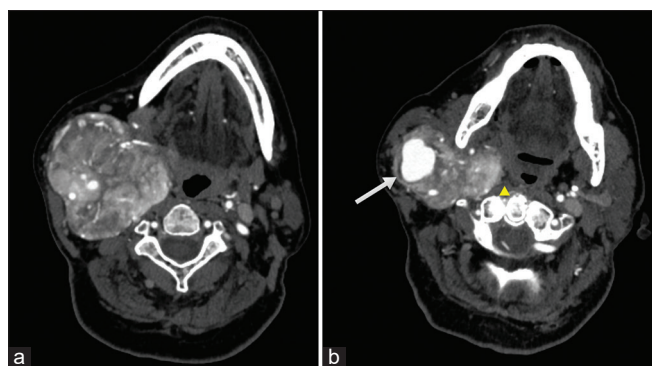


Figure 1: Axial computed tomography angiography of the neck (a,b) demonstrates a large, enhancing, hypervascular, lobulated mass originating in the carotid space, encasing the common carotid and internal carotid arteries, and extending into the masticator and parotid spaces. This mass causes obliteration of the right parapharyngeal fat pad (arrowhead, b). A large contrast-filled collection measuring up to 3.5 cm is noted within the tumoral mass (arrow, b), consistent with a carotid body tumor with an intratumoral pseudoaneurysm.

primarily supplied by the musculospinal branch of an enlarged ascending pharyngeal artery [Figures 2c and d].

Right ICA injection revealed an abnormal jet flow originating from the proximal cervical ICA, filling a 3.5 cm contrast collection within the tumoral matrix, consistent with a pseudoaneurysm contained by surrounding tumor tissue [Figure 3a].

In the late arterial phase, multiple blood vessels were seen originating from this pseudoaneurysm, indicating an arterial component [Figure 3b]. These vessels did not connect to any venous structures, ruling out a fistulous component and confirming the arterial nature of the flow. Overall, these findings are consistent with a large intratumoral pseudoaneurysm of the ICA [Figure 3c].

The patient underwent successful embolization of the pseudoaneurysm using a PX Slim Microcatheter (Penumbra Inc.), multiple PC400 Coils (Penumbra Inc.), and Onyx 34 (Medtronic PLC), with a balloon-assisted technique employing a 5 × 30 mm HyperGlide balloon microcatheter [Figure 3d]. This technique was used to prevent the coils and Onyx from escaping the aneurysm and to reduce the risk of cerebral emboli. Finally, a 5 mm × 50 mm Viabahn-covered stent (W.L. Gore and Associates, Newark, DE, USA) was placed in the ICA [Figure 3e].

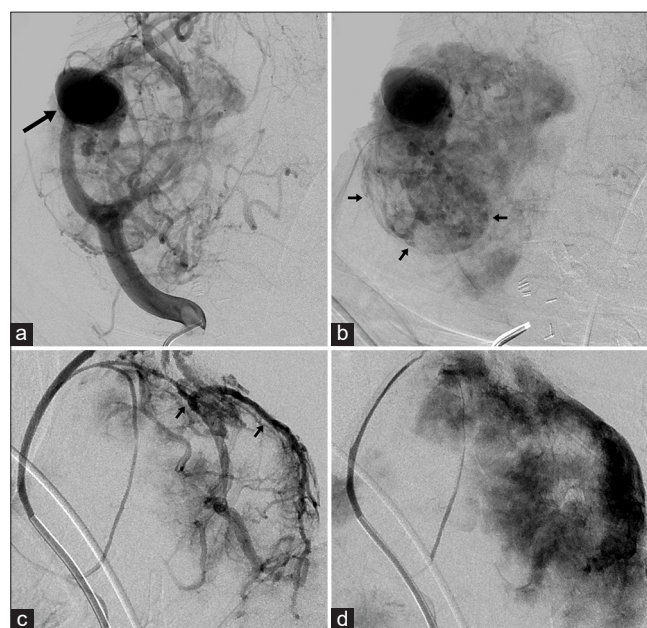


Figure 2: Angiography of the right common carotid artery (a,b) demonstrates splaying of the external and internal carotid arteries. Note the circumscribed contrast collection (arrow, a) originating from the proximal right internal carotid artery, and contrast blush (arrows, b) indicative of a large carotid body tumor with persistent intratumoral contrast collection. Angiography of the right ascending pharyngeal artery (c,d) shows that its musculospinal branch (arrows) is the main arterial supply to this tumor.

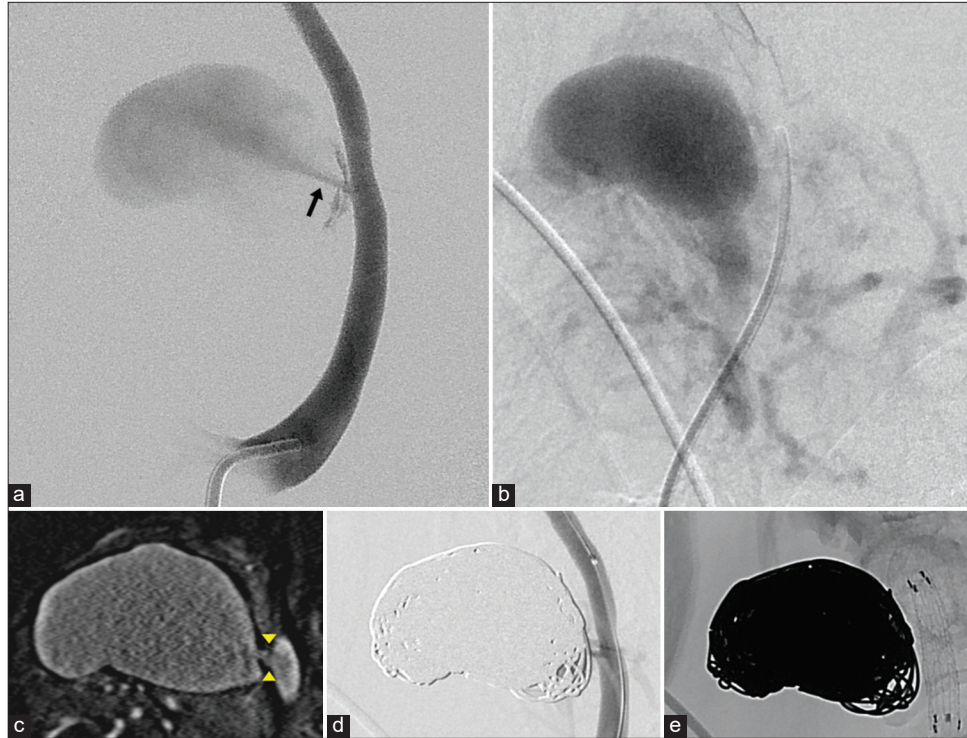


Figure 3: Angiography of the right internal carotid artery (a,b) demonstrates in the early arterial phase (a) a contrast collection that fills the middle portion of the tumor with a jet flow (arrow). During the late arterial phase (b), multiple blood vessels originate from the 3.5 cm contrast collection. Cone Beam CT reconstruction with contrast injection in the right internal carotid artery (c) demonstrates filling of the intratumoral pseudoaneurysm (arrowhead). Post-procedural angiography (d,e) shows near-complete resolution of the pseudoaneurysm and a 5 mm x 50 mm Viabahn covered stent (W.L. Gore and Associates, Newark, DE, USA) deployed in the cervical internal carotid artery.

Immediate postprocedural angiography showed good apposition of the stent into the ICA wall and complete occlusion of the pseudoaneurysm. There were no procedural complications, and the patient had a satisfactory recovery. The patient was started on dual antiplatelet therapy with ticagrelor and aspirin.

A 6-month follow-up carotid Doppler ultrasound showed no flow within the pseudoaneurysm and a reduction in the original tumor size from 8 cm × 7 cm to 4.61 cm × 5.04 cm.

The patient reported complete resolution of the symptoms. Surgical resection or radiotherapy was not pursued at that moment, and surveillance with Doppler ultrasound was indicated.

DISCUSSION

CBTs are highly vascular tumors with blood supply mostly derived from ECA branches, although supply from ICA and vertebral branches can also occur. Angiography provides a greater understanding of the vascular anatomy, allows visualization of underlying vascular lesions such as

aneurysms, pseudoaneurysms, and arterio-venous fistulas (AVF), and helps in the treatment decisions.

In general, paragangliomas are rarely associated with underlying vascular pathologies. Some case reports have described cardiac and intrapericardial paragangliomas with intratumoral aneurysms and AVFs.^[7,14] However, there are no reports of CBTs with intratumoral pseudoaneurysms, but rather, only the presence of associated aneurysms in the ICA and ECA has been described.^[9,10]

Surgical resection remains the definitive treatment for CBTs.^[2] Nonetheless, effective management must be tailored to each patient's specific circumstances, considering factors such as tumor characteristics and patient comorbidities.^[4] Available treatment options include surgical resection, conservative management, and radiotherapy. While surgical resection is often preferred, conservative management, including active surveillance, may be appropriate in select cases, particularly given the typically slow growth of these tumors.^[6] Radiotherapy may also be considered for patients who are not candidates for surgery, although its precise role in simple CBT cases is not fully established and lacks clear indications.^[4,13]

Palliative embolization can be considered a viable alternative to surgical resection for CBTs, particularly in cases of recurrent or unresectable tumors, such as those classified as Shamblyn type III. It is also a suitable option for patients with significant comorbidities or those at high surgical risk.^[8] Embolization has been shown to effectively reduce symptoms and tumor vascularity, providing symptomatic relief when surgery is not feasible.^[8] Some reports highlight its use as a primary treatment for recurrent or unresectable tumors.^[8] However, when surgery is possible, resection remains the standard treatment, as it offers the only chance for complete tumor removal. In many cases, embolization is used as an adjunct to surgery to reduce intraoperative blood loss and facilitate resection.^[3,5]

The use of preoperative embolization is well recognized in the management of CBTs due to their high vascularity. Some studies suggest that preembolization can reduce intraoperative blood loss, shorten operative time, and minimize surgical complications.^[11] In complex cases involving vascular pathologies, such as those with intratumoral pseudoaneurysms, embolization becomes even more critical to reduce the risks associated with surgery.

Balloon test occlusion (BTO) is crucial in preoperative planning for CBTs, assessing collateral cerebral circulation through the circle of Willis. It helps determine if the ICA can be safely sacrificed without ischemia risk by temporarily occluding the artery.^[3] If the BTO is negative, meaning sufficient blood flow exists, the ICA and tumor can be safely removed with no postoperative neurological complications.^[15] BTO is a valuable tool for guiding safe surgical planning when ICA removal is needed.

The absence of standardized guidelines for managing CBTs necessitates a case-by-case assessment. In this case, the patient passed the BTO; however, embolization was prioritized due to the presence of a rare intratumoral pseudoaneurysm and high risk for undergoing surgery. This approach effectively addressed the secondary vascular pathology and provided symptomatic relief, yet surgical resection remains the definitive treatment for CBT when feasible. Consequently, the patient is currently under active surveillance to monitor for any changes in tumor status.

CONCLUSION

In complex vascular paragangliomas, endovascular treatment serves as a critical tool in reducing morbidity and mortality, particularly when surgical resection poses high risks. This case highlights the importance of identifying secondary vascular pathologies, such as intratumoral pseudoaneurysms, during preoperative imaging. Recognizing these vascular anomalies is essential to prevent life-threatening bleeding during surgical intervention. Palliative embolization

provides an alternative, particularly in cases where surgery is not currently the best option. This approach reduces tumor vascularity and minimizes procedural risks.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript, and no images were manipulated using AI.

REFERENCES

1. Butt N, Back WK, Lachkar S, Iwanaga J, Mian A, Blaak C, *et al.* The carotid body and associated tumors: Updated review with clinical/surgical significance. *Br J Neurosurg* 2019;33:500-3.
2. Davila VJ, Chang JM, Stone WM, Fowl RJ, Bower TC, Hinni ML, *et al.* Current surgical management of carotid body tumors. *J Vasc Surg* 2016;64:1703-10.
3. Duffis EJ, Gandhi CD, Prestigiacomo CJ, Abruzzo T, Albuquerque F, Bulsara KR, *et al.* Head, neck, and brain tumor embolization guidelines. *J Neurointerv Surg* 2012;4:251-5.
4. Gonzalez-Urquijo M, Castro-Varela A, Barrios-Ruiz A, Hinojosa-Gonzalez DE, Salas AK, Morales EA, *et al.* Current trends in carotid body tumors: Comprehensive review. *Head Neck* 2022;44:2316-32.
5. Kelblová M, Vaníček J, Gál B, Rottenberg J, Bulik M, Cimřlová P, *et al.* Preoperative percutaneous Onyx embolization of carotid body paragangliomas with balloon test occlusion. *Front Neurol* 2023;14:1132100.
6. Langerman A, Athavale SM, Rangarajan SV, Sinard RJ, Netteville JL. Natural history of cervical paragangliomas: Outcomes of observation of 43 patients. *Arch Otolaryngol-Head Neck Surg* 2012;138:341-5.
7. Lee CC, Chua S, Huang SC, Lee FY, Chung SY. Intrapericardial paraganglioma with intratumoral coronary arterial aneurysm and an arteriovenous fistula. *J Am Soc Echocardiogr* 2009;22:211.
8. Maia M, Vidoedo J, Pinto JA. Recurrent carotid body tumors:

- Embolization as a treatment option. *Rev Port Cir Cardiorac Vasc* 2014;21:129-32.
9. Mutlu V, Ogul H. Magnetic resonance imaging and magnetic resonance angiography findings of external carotid artery aneurysm and coexisting carotid body tumor. *J Craniofac Surg* 2016;27:772-3.
 10. Rohlfing ML, Yang B, Jalisi S. Carotid body tumor with hidden internal carotid artery aneurysm. *Head Neck* 2019;41:E79-81.
 11. Shiga K, Katagiri K, Ikeda A, Saito D, Oikawa SI, Tsuchida K, *et al.* Challenges of surgical resection of carotid body tumors-multiple feeding arteries and preoperative embolization. *Anticancer Res* 2022;42:645-52.
 12. Shamblin WR, ReMine WH, Sheps SG, Harrison EG. Carotid body tumor (chemodectoma): Clinicopathologic analysis of ninety cases. *Am J Surg* 1971;122:732-9.
 13. Valero C, Ganly I, Shah JP. Head and neck paragangliomas: 30-year experience. *Head Neck* 2020;42:2486-95.
 14. Wan J, Li W, Zhou Z, Shang J, Chen D. Primary cardiac paraganglioma with intratumoral coronary artery aneurysm. *Ann Thorac Surg* 2014;97:e75-7.
 15. Zeitoun IM, Borhamy GH, Fata MM, Salloum AM. Sacrificing the internal carotid artery in infiltrating neck tumours: A study of four clinical cases. *Int J Oral Maxillofac Surg* 2017;46:11-5.

How to cite this article: Ortega V, Calderon J, Ahumada F, Quiroz Alfaro AJ, Diaz O. Intratumoral pseudoaneurysm in a carotid body tumor. *Surg Neurol Int.* 2024;15:446. doi: 10.25259/SNI_693_2024

Disclaimer

The views and opinions expressed in this article are those of the authors and do not necessarily reflect the official policy or position of the Journal or its management. The information contained in this article should not be considered to be medical advice; patients should consult their own physicians for advice as to their specific medical needs.