Implications of Carotid Sinus Hypersensitivity Following Preoperative Embolization of a Carotid Body Tumor
An Indication for Prophylactic Intraoperative Cardiac Pacing

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Carotid body tumors (CBTs) are rare neoplasms of neural crest origin, also called paragangliomas. Even though most CBTs are benign, these tumors exhibit an infiltrative growth pattern, especially in the region of the carotid bifurcation. While some patients are observed or treated with radiation therapy, most undergo surgical resection. Because of the vascularity of these tumors, preoperative embolization may be performed to decrease intraoperative blood loss. While well tolerated in most, tumor embolization has associated risks and complications.

In this Case Report, we describe a patient with bilateral CBTs who underwent staged surgical resection with preoperative embolization prior to each procedure. Following his second preoperative embolization, mild hemodynamic instability suggestive of carotid sinus hypersensitivity (CSH) occurred. The postembolization cardiovascular instability was a harbinger of the significant CSH that resulted in severe intraoperative cardiovascular complications and the need for temporary cardiac pacing.

Report of a Case

A man in his early 30s was seen in consultation for CBTs. He had noticed a mass on the right side of his neck approximately 6 months before he went to see his primary health care provider (PCP). Other than noticing the mass, the patient was completely asymptomatic and denied any pain, difficulty swallowing or breathing, voice changes, difficulty moving his tongue, tremors, or palpitations. The patient did have a 5-year history of hypertension that had been relatively resistant to medical management. In addition to his family history being significant for hypertension, it was also notable that his paternal grandmother, 2 paternal cousins, and his sister had CBTs.

IMPORTANCE  Carotid body tumors are rare neoplasms of neural crest origin that are both highly vascularized and locally invasive. Treatment options for these tumors often include surgery with preoperative embolization, which can pose major cardiovascular risk to patients. As demonstrated by this case report, hemodynamic instability following preoperative embolization of a carotid body tumor may indicate severe carotid sinus hypersensitivity and the need for temporary cardiac pacing.

OBSERVATIONS  This case report describes a man in his early 30s who presented for staged surgical resection of bilateral carotid body tumors with preoperative embolization. After embolization of the second tumor, the patient displayed transient episodes of bradycardia and hypotension, which resolved with medical management. Surgery commenced, and with minimal manipulation intraoperatively, the patient became asystolic and required resuscitation. Following a negative cardiac workup, a temporary pacemaker was implanted, and surgical resection of the tumor was successfully completed.

CONCLUSIONS AND RELEVANCE  Carotid sinus hypersensitivity is a rare but serious risk of preoperative embolization of carotid body tumors. Postembolization bradycardia or hypotension should be assessed as potential harbingers of carotid sinus hypersensitivity, and the need for temporary intraoperative cardiac pacing should be strongly considered.
Prior to the consultation, an ultrasonographic image had been obtained by his PCP and showed a soft-tissue mass associated with the right carotid artery. Subsequently, a computed tomographic scan of the neck with contrast was obtained and showed a large, intensely enhancing mass measuring 2.6 × 2.6 × 4.6 cm at the level of the right carotid bifurcation. A slightly smaller mass was seen in the left side of the neck in a similar location, measuring 2.0 × 2.5 × 3.9 cm. These findings were consistent with bilateral CBTs.

Given the patient’s history of poorly controlled hypertension at a young age, blood and urine studies were completed to assess for tumor functionality and revealed no signs of catecholamine or dopamine production. Genetic testing was also obtained, confirming a succinate dehydrogenase D abnormality. Treatment options of surgical resection, radiation therapy, and observation were discussed with the patient. Given his young age and relative progressive growth noted over the past months, it was agreed that staged surgical resection of his tumors was the most appropriate therapy. It was decided to operate on the smaller tumor first, with the thought that this side had a lower risk of a permanent cranial nerve injury.

Given the size of the left CBT, preoperative embolization using nondetachable Vortex coils (Stryker) as well as Embospheres (Merit Medical) was completed the day prior to surgical resection (Figure 1). After embolization, the patient experienced some discomfort in the tumor but remained hemodynamically stable and without any complications.

The following day, surgical resection was completed without any complications. The tumor was dissected away from the carotid artery without difficulty and with only moderate manipulation of the cranial nerves. Postoperatively, the patient did have hypernasality and dysphagia but no hemodynamic instability. The cranial nerve deficits did resolve over the following few months with the patient returning to normal function.

Six months following his initial surgery, resection of the right CBT was scheduled. Similar to this first resection, preoperative embolization with nondetachable coils and Embospheres was performed the day prior to surgery (Figure 2). That evening, the patient became light-headed and felt nauseated. On telemetry, it was noted that the patient’s heart rate had dropped from approximately 60 beats per minute (bpm) to approximately 30 bpm. With the bradycardia, the patient was also noted to be hypotensive with systolic blood pressure measurements in the 70s to 80s. The bradycardia and associated hypotension did spontaneously resolve, but he subsequently had similar episodes during the night.
A cardiology unit consultation was obtained. Evaluation included an electrocardiogram, which did not show any signs of cardiac ischemia or primary heart blockage. Medical management was initially implemented with isoproterenol hydrochloride, but the patient continued to have symptoms. Therapy with low-dose dobutamine hydrochloride was then initiated, and this stabilized his vital signs. It was felt that cardiac pacing would not be required. He was closely monitored overnight and did not have any other episodes.

Given the stability overnight following the addition of low-dose dobutamine, it was felt that proceeding with resection was appropriate. The patient was stable throughout induction of anesthesia and intubation. He was then prepared for surgery and draped in standard fashion. After making the neck incision, subplatysmal flap elevation was performed to expose the sternocleidomastoid muscle. As the muscle was being mobilized, the patient became acutely bradycardic with progression to asystole within a minute. Resuscitation was initiated using Advanced Cardiovascular Life Support protocol, including chest compressions and the administration of epinephrine hydrochloride and atropine sulfate. The patient regained a pulse and achieved a measurable blood pressure within a few minutes of becoming asystolic. At that point, it was determined that the procedure should be terminated. The neck incision was promptly closed, and the patient was taken to the intensive care unit (ICU) for future diagnostic evaluation and monitoring.

In the ICU, the patient remained stable, with no signs of any cardiac ischemia or neurologic deficits. The cardiology unit reevaluated the patient and decided to place a temporary pacemaker prior to any additional surgery. The patient had the pacemaker placed without difficulty and remained stable for the next 48 hours. The patient was then taken back to surgery for resection of the CBT. During the resection, the pacemaker was triggered multiple times, and the patient had a paced rhythm during most of the procedure. The resection was completed without complication, and the carotid artery and cranial nerves were all preserved. Following completion of the procedure, the patient did not have any episodes of bradycardia and did not require any pacing to maintain an appropriate heart rate. With this resection, he also did not experience any postoperative cranial nerve deficits. After showing stability for 24 hours postoperatively, the pacer wires were removed without complication. The remainder of the hospitalization was uneventful. Subsequently, he has done remarkably well, with good exercise tolerance and no signs of any cardiovascular issues.

![Figure 2. Angiographic Image of Right Carotid Body Tumor (CBT)](image)
Discussion

The carotid sinus is located immediately above the carotid bifurcation and contains pressure-sensitive receptors important for blood pressure homeostasis. The carotid sinus baroreceptor transmits sensory information via an afferent reflex arc comprising the nerve of Hering, a specialized nerve to the carotid sinus from the glossopharyngeal nerve. This signal is relayed to the nucleus tractus solitarius and the paramedian reticular formation in the brainstem with the efferent reflex arc carried through the vagus and sympathetic nerves; the former nerve increases parasympathetic inhibition of cardiac output by increasing conduction delay at the sinoatrial and atrioventricular nodes, and the latter nerve works to decrease vasmotor tone.4

Carotid sinus hypersensitivity, or carotid sinus syndrome, is an exaggerated response of this reflex arc to stimulation. The symptoms of CSH can range from mild dizziness or syncope to asystole and cardiac arrest. Carotid sinus hypersensitivity is categorized into 3 subtypes: (1) cardioinhibitory, which comprises 70% to 75% of CSH and causes a decrease in heart rate, leading to bradycardia or cardiac arrest; (2) vasodepressor, which makes up 5% to 10% of CSH and has no effect on heart rate but causes hypotension owing to effects on vasmotor tone; and (3) mixed, which comprises 20% to 25% of CSH and is a combination of both bradycardia and hypotension.5 Although the most common type of CSH is cardioinhibitory, tumors of the head and neck have been documented to more commonly exhibit the vasodepressor effect.6,7 In this case, the patient experienced cardiac arrest while in surgery and had clinically significant bradycardia and hypotension following embolization, categorizing his CSH as the mixed subtype.

The etiology of CSH is complex owing to the fact that the hypersensitivity can occur anywhere along the reflex arc or at the target organs. Commonly, CSH has been associated with recurrent syncopal episodes in elderly individuals with atherosclerotic plaques in the carotid arteries. Carotid sinus hypersensitivity has also been recently associated with medullary autonomic nuclei degeneration correlating with other disease processes, such as Alzheimer disease, Parkinson disease, and dementia with Lewy bodies.7 While case reports have also linked CSH to internal carotid artery occlusion,6 Takayasu arteritis,9 and radiotherapy for bilateral glomus tumors,9 the most common cause of CSH is a neck mass.3 While intraoperative CSH resulting in cardiac arrest is relatively rare, it has been documented in patients with clinically significant risk factors, such as head and neck tumors, prior radiation, and/or scarring in the area of the carotid sinus, local mechanical stimulation of the carotid sinus by endoscopic instruments, and preoperative embolization of paragangliomas, such as in this patient.3

In this case, the patient did not present with any symptoms of tumor-induced CSH prior to treatment. Furthermore, the patient was a healthy adult without any clinically significant cardiac history, and findings from his postoperative cardiovascular evaluation were negative for other etiologies for intraoperative cardiac arrest. It was only after embolization and resection of the left CBT and then embolization of the right CBT were signs of CSH noted. While surgical manipulation in the area of the carotid sinus can result in cardiac arrest by direct mechanical stimulation of the carotid sinus, in this patient, signs of CSH were noted prior to any surgical manipulation. In addition, embolic agents, such as Onyx (EV3), have been implicated as a cause of bradycardia and asystole during embolization procedures.10 In this patient, Onyx was not used, and no reports of cardiovascular instability have been documented with the use of coils or Embospheres for embolization. It is also unlikely that the embolization products would be directly responsible in this case because the same embolization approach was for both tumors, but CSH was only encountered following the second embolization.

There is considerable complexity of the baroreceptor reflex arc, and much remains unknown regarding the interplay between both carotid sinuses. While the exact etiology of postembolization CSH is unclear, one possible explanation may be embolization-induced tumor edema. Tumor swelling due to ischemia has been documented, and tumor edema, producing a mechanical stimulation to the carotid sinus, could explain the new-onset CSH seen in this patient.11 However, the unique aspect of this case is the difference in outcomes between the treatment of the 2 tumors, even though they were relatively the same size and each was managed using the same protocol. During the embolization and resection of the tumor on the left side of the neck, no hemodynamic instability was encountered. It is possible that the contralateral baroreceptor reflex, which likely was normal functioning, could have affected the physiologic response to any abnormal signals from the left carotid sinus that occurred as a result of the embolization. After resection of the tumor on the left side of the neck, it is likely that the left baroreceptor reflex was lost. Subsequently, when the right-sided tumor was embolized, any hypersensitivity in the right reflex arc would have been unopposed.

While medical management after embolization was able to stabilize the patient, this approach was not successful once the surgical procedure was initiated. In retrospect, if cardiac pacing had been implemented based on the fact that the embolization alone resulted in CSH, it is likely the intraoperative cardiovascular collapse could have been avoided. Prophylactic pacing could have allowed the resection to be completed during the initial procedure and avoided the need for a second operation to resect the right CBT.

Conclusions

This case highlights the possibility of preoperative embolization of a CBT being associated with intraoperative, life-threatening CSH arrest. The significance of this patient’s postembolization hemodynamic instability became obviously apparent the following day during surgery. Physicians should have a heightened awareness of the potential significance of any signs of CSH following preoperative embolization. In patients with any signs of postembolization CSH, a temporary cardiac pacemaker should be seriously considered prior to surgical resection of a CBT to prevent intraoperative hemodynamic instability and ensure a successful outcome.
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