

A Retrospective Study on Carotid Body Tumor, Safe Surgical Treatment without Embolization: A Report of 46 Cases

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Abstract

Background: Carotid body tumors are the most prevalent type of paragangliomas in head and neck region, which present as painless mass in carotid triangle with slowing growth. A palpable mass in the neck is the most common symptom of these tumors and other less common symptoms including hoarseness and dysphagia. The current standards of care for carotid body tumor patients are surgical resection. These tumors don't response to radiation therapy and chemotherapy.

Aim: In the present case series we evaluated surgical treatment without embolization as a safe treatment modality for carotid body tumours.

Methods: In this study, 46 patients with a total of 50 carotid body tumors were investigated in order to recording their demographic data, clinical presentation, diagnostic modality and treatment strategy.

Results: The results demonstrated that almost 78% of patients were female and patients were 21 to 78 years of age, with the average of 49.1. Bilateral carotid body tumors were observed in 8.7% of patients who had familial history.

Conclusion: Carotid body tumors can be removed surgically with minimal morbidity and mortality risks. Understanding the vascular anatomy of this region is critical for the surgeon. However, there is a major surgical challenge due to the nature of the tumor and its vital neighbourhood.

Conflicts of Interest: The Authors declare no conflicts of interest.

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Introduction

Carotid body tumor is a rare head and neck neoplasm (less than 0.5% of all head and neck tumors) that was described in 1743 for the first time by Von Haller (1, 2). Although carotid body tumors are not very prevalent, they are the most common type of the head and neck paragangliomas (65%). Carotid body tumors originate from neural crest cells and often emerge as a painless mass in the carotid triangle with slowing growth. They mostly occur in the fourth and fifth decades of life with female predilection (3, 4). Multiple tumors are observed in about 10% of all cases and the overall incidence of the malignancy is 6–12.5% (5). Surgical resection is the

treatment of choice in management of carotid body tumors. These tumors are resistant to radiotherapy and chemotherapy. Untreated tumors lead to death in about 8% of the cases in the natural course of the disease due to the compression of the adjacent nervous and vascular structures (6, 7). Besides, understanding the vascular anatomy of this region is critical for a surgeon. However, there is a major surgical challenge due to the nature of tumors and its vital area around it.

Methods

In this case study, 46 patients with carotid body tumors referred to Ear, Nose and Throat

Department of Taleghani Hospital in Tehran, Iran over 3 years (2013–2015) were recruited and evaluated retrospectively. Demographic data, clinical presentation, diagnostic modality and treatment strategy were recorded. Preoperative work-up included history, physical examination, ultrasound of the neck, computed tomography (CT) or magnetic resonance imaging (MRI). All carotid body tumors (CBTs) were classified using CT scan or MRI findings based on the Shamblin classification.

In 1971, carotid body tumors were classified into three groups by Shamblin et al. Group I (small tumors that do not involve carotid vessels), Group II (tumors that partially surround carotid vessels), and Group III (tumors encasing carotid vessels) (8).

In all surgically treated cases, tumor resection was carried out under general anaesthesia through a longitudinal cervical incision. Intra-operatively, the carotid sheath was identified. The common carotid artery was dissected and the tumors were detached meticulously. Adjacent neural structures such as the hypoglossal nerve and the vagus nerve were dissected and preserved.

Treatment modality, surgical complications, tumors recurrence and disease survival were recorded.

SPSS statistical software, version 22 was used for statistical analyses. Statistical significance level considered as P-value equal or less than 0.05. The Ethics Committee of Shahid Beheshti University of Medical Sciences approved the study with ethic code: IR.SBMU.MSP.REC.1400.297. Informed consent was obtained from the patients and the rights of the patients were protected

Results

In our retrospective study, 46 patients were investigated for 50 carotid body tumors. There were 36 tumors in females (78.30%) and 10 in males (21.70%). The patients' age ranged between 21 and 78 years old (mean: 49.1 ± 16.1) (Table 1). Four patients (8.7%) had bilateral carotid body tumor. In the other 42 (91.3%) patients with unilateral tumors, 25 (59.5%) tumors were located in the right side and 17 (40.5%) in the left side. The positive family history of CBT was reported in six patients (13%) (Table 1).

Table 1: Demographic data of patients

Variable		Number or mean values
Number of patients		46
Number of tumors resected		49
Age		21_78 (mean: 49.1 ± 16.1)
Male gender		10(21.73%)
Bilateral tumors		4 (8.69%)
Multifocal tumors		1(2.17%)
Family history		6(13.04%)
Presentation	Painless mass	40(80%)
	Painful mass	10(20%)
	Recent growth of neck mass	36(72%)
	Dysphagia	2(4.34%)
	Dysphonia	3(6.52%)
	Flushing/palpitations	-
Imaging	Ultrasound duplex scan	30(65.21%)
	CT	25(54.34%)
	MRI	36(78.26%)
	MRA	2(4.34%)

The Shamblin classification was used for tumors grading.

The average tumors size was 29×29 mm. In 36 patients (72%) the size of tumors increased during 6 months before the surgery.

The most common symptom was neck mass (100%), which was tender in palpation in 20% of the patients. The other less common symptoms included hoarseness (6.52%) and dysphagia (4.34%) (Table1).

The average interval between symptom presentation and medical referral was 27.7 months. The diagnostic modalities were CT

(54.34%), doppler sonography (65.21%), MRI (78.26%) and magnetic resonance arteriography (MRA) (4.34%) (Table1). In some patients, more than one modality was used. Forty-five of the cases (97.82%) underwent surgical resection, and only one patient was referred for radiotherapy (Table 2). There was no need for pre-operative angiography, endovascular surgery in any patients. Blood transfusion was done in 12 surgeries (Table 2). Tumors recurrence and malignant transformation were not reported. Only one patient reported transient dysphasia.

Table 2. Management of patients

Variable	Mean values or number
Preoperative embolization	-
Resection alone	49(98%)
Radiotherapy alone	1(2%)
Resection with postoperative radiotherapy	-
Blood transfusion	12(24.48%)
Endovascular surgery	-
Shamblin classification	Type I
	Type II
	Type III
	21(42%)
	24(48%)
	5(10%)

Discussion

Carotid body tumors are attractive neck masses due to their specific location in the carotid bifurcation. Because of their rare nature, most of our knowledge is limited to single-center findings. This study is a retrospective report on a three -year experience of a series of carotid body tumors treated in our center. All patients underwent surgery without embolization preoperatively. Our mortality was zero, with no major postoperative early or late complication.

Interestingly, compared to other centers (9, 10), there were a remarkable number of CBTs in a relatively short time.

Regarding onset age, carotid body tumors usually occurred in 20-60 age category. (11) In our series, the peak age distribution was between 21 and 78 years old.

Contrary to the reports from other centers, our study showed that females are most affected by this disease (78.3%); however, some centers reported their data vice versa (10, 12). This gender preference attributed to the chronic hypoxic stimulation due to menstruation phase in women. Although in the literature, genetic factors are suggested as contributing factors (13).

Carotid body tumors could be bilateral in 5% of the sporadic cases and 33% of the familial cases. Multifocal diseases were reported in 10% to 20% of the sporadic cases and up to 80% of the familial cases. According to the literature, up to 30% of all paragangliomas were associated with germ line mutations in mitochondrial succinate dehydrogenase (SDH) subunit genes—SDHB, SDHC, SDHD and SDHAF2.

Therefore, screening of SDH mutations should be done in all paragangliomas patients to design further treatment strategy (14).

There were four bilateral carotid body tumors cases that two of which showed a familiar pattern. Interestingly, one of the familial cases had multifocal paraganglioma (bilateral carotid body and right-side glomus tympanicum). Because of a high complication rate especially in bilateral cases, treatment in bilateral carotid body tumors is more challenging. All of our bilateral cases were treated surgically. As proposed by other authors, at first, the smaller tumor was approached, because it can be conducted with a minimal risk of the cranial nerve injury and increased probability of preserving neurovascular structures. In the next stage, the larger tumors were removed.

Clinically, the most common presenting symptom of the carotid body tumors is lateral neck mass that was reported in all of our cases. Although some authors reported associated pain, most of them considered carotid body tumor as a painless mass (15).

In this series, the pain was detected in about 20% of the cases. Adjacent cranial nerve impingement may cause dysphagia, odynophagia, hoarseness or other cranial nerve deficits. Occasionally, compression of cervical sympathetic chain causes Horner's syndrome or carotid sinus compression causes syncope (6). We had no clinical symptoms associated with functional tumors. In this series, the interval between neck mass presentation and medical referral was about 27 months. Although carotid body tumor considered as a benign slow-growing mass, early surgical resection suggested, because of malignant potential of these tumors and related neurovascular complication (14). None of our cases showed malignant nature. The embolization of vascular tumors such as carotid body tumors was proposed as an important adjunctive therapy to the surgery;

the effectiveness of preoperative embolization in all cases of carotid body tumors remains controversy. Interestingly, a new investigation found no benefit of embolization preoperatively relating to stroke, blood loss, and cranial nerve injury in the carotid body tumor (16). Although all of our surgically treated cases successfully managed without preoperative embolization, blood transfusion was done in 12 patients. However, it seems that it would be better if blood loss volume documentation was available for further assessment. The management options for the paragangliomas in head and neck region include surgical resection, stereotactic radiotherapy, and fractionated radiotherapy. Surgery is preferred for a small tumor in patients with a good medical condition. Radiotherapy is recommended as the initial treatment for patient in whom the resection of the carotid vessels is anticipated. Also, radiotherapy is proposed as a suitable option for minimal residual tumors after the surgery and as adjuvant therapy in malignant carotid body tumors (6). Only one of these cases was referred for radiotherapy because of the patient preference. Based on a recent review, the rate of post-operative cranial nerve injuries is about 48 %, which is usually transient (31%). Permanent deficit occurred in about 17% of cases. The incidence of arterial injuries estimated to be about up to 27%. Among the neurovascular complications, the vague nerve was the most common cranial nerve and external carotid artery was the common artery affecting by the surgical resection (13). Limitation of our study is the small number of the samples.

Conclusion

In conclusion, carotid body tumor is a rare vascular tumor of the neck that can be treated surgically with minimal morbidity and mortality. However, the distinct rule of preoperative embolization remains a

controversy that it could be considered in a special situation.

Abbreviations

CT: Computed tomography

MRI: Magnetic resonance imaging

CBT: Carotid body tumor

SDH: Succinate dehydrogenase

SDHB: Succinate dehydrogenase [ubiquinone] iron-sulfur subunit, mitochondrial

SDHC: Succinate dehydrogenase complex subunit C

SDHAF2: Succinate dehydrogenase complex assembly factor 2

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Conflicts of Interest

The authors declare no conflicts of interest.

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Ethics

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Patients' consent

Informed consent was obtained from all individual participants included in the study. Ethically, the patients are not identified via the paper and are fully anonymized.

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