Carotid Body Tumors: Surgical Management and Review of Patients Over 10 Years

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Abstract

Background: Carotid body tumours (CBTs) are rare but highly vascular neoplasms originating in the paraganglionic cells of the carotid bifurcation. Exact etiology of these tumors is not known. Male and female distributions are equal except at high altitude where females appear to predominate. Diagnosis of a chemodectoma usually begins with a color flow duplex scan. Magnetic resonance angiography are also useful, especially to evaluate bilateral disease. Conventional Arteriography with CT Angiography are valuable, especially in larger tumours, and are regarded as the best tools for diagnosis. The treatment of choice for carotid body tumours is surgical removal. Shamblin’s classification system is used to categorize carotid body tumours based on their size and the difficulty of surgical resection.

Results: It was observed that majority of the patients in this study were females numbering 35(79.45%) whereas there were 9 (20.43%) males. commonest age group involved was of patients 50 to 59 years of age. Surgery was done in all 44 (100%) patients with complete resection in 41 (93.07%) and incomplete in 3 (6.81%) patients. 10 (23%) were Shamblin grade 1, 29 (66%) grade 2 and 5 (11%) grade 3. ECA repair was done in 4 (9.08%) of patients, ICA repair in 2 (4.54%), ECA ligation in 2 (4.54%) and vascular graft was used in 1 (2.27%) of patients. 7 (15.89%) had transient cranial nerve palsy most commonly involving hypoglossal nerve. 3 (6.81%) had permanent cranial nerve palsy . In 6 (13.62%) patients there was local wound infection .There was post op stroke in 2 (4.54%) of patients. No operative mortality was seen.43 (97.61%) patients were proved to be paraganglionomas on HPE while 1 (2.27%) patients had inconclusive biopsy on Histopathological examination.

Conclusion: Although rare, Carotid body tumor is still a pathology that we encounter in our experience and it should be kept in mind as a differential diagnosis for painless lateral neck masses.

Key Words: Carotid body tumor, common carotid artery, three-dimensional computed tomography

Introduction

The carotid body (carotid glomus or glomus caroticum) is a small cluster of chemoreceptors and supporting cells located near the bifurcation of the carotid artery. The carotid body is made up of two types of cells called glomus cells; glomus Type I/chief cells are derived from neural crest and glomus Type II/sustentacular cells.[1] The carotid body contains the most vascular tissue in the human body. Carotid body tumors (CBTs) are rare but highly vascular neoplasms originating in the paraganglionic cells of the carotid bifurcation. Described first by Haller in 1743, they are derived from the epithelioid cells of neuroectodermal origin.[2] Incidence is between 0.06 and 3.33 per 100,000 patients. Clinically, CBT typically presents as a nontender, rubbery, pulsatile mass. Classically, the mass can be displaced laterally but not vertically due to carotid artery adherence, which is known as a positive Fontaine sign. Diagnosis is commonly confirmed by duplex ultrasound, computerized tomography (CT), magnetic resonance imaging (MRI), and rarely conventional angiography.[3] Although technically challenging, surgery remains the only definitive treatment. They are generally benign, usually unilateral; and there is no report of spontaneous regression.[4] CBTs are the most common head and neck paragangliomas. The usual histologic criteria for malignancy, i.e., nuclear atypia and nuclear to cytoplasmic ratio, do not apply; and the likely clinical behavior cannot be predicted from the routinely stained tissues. The true proof of malignancy is the presence of lymph node or distant metastases, which may not become evident even years after the original resection.[4,5] CBTs are graded as: Staging; (Shamblin et al)[6,7]

Class I CBT – localized with minimal vascular attachment

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Class II CBT – partially surrounds carotids
Class III CBT – encase carotids. Surgical resection is difficult and may require temporary interruption of cerebral circulation.

Diagnosis of a CBT or chemodectoma usually begins with a color flow duplex scan, MRI, and magnetic resonance angiography (MRA) are also useful, especially to evaluate bilateral disease, conventional arteriography with CT angiography (CTA) are valuable, especially in larger tumors and are regarded as the best tools for diagnosis.[6] The role of FNAC is still not clear because of high vascularity of tumor, but some institutes use it for initial diagnosis.

The treatment of choice for many CBTs is surgical removal.[6] There are major risks involved with resecting these tumors because of their close location to the carotid vessels and cranial nerves; these risks increase as the tumor increases in size. Tumors smaller than 5 cm in diameter are associated with a complication rate of 15%; however, tumors with a diameter $\geq$ 5 cm have a 67% complication rate.[6] Thus, tumor size is an important predictor of possible complications.[6] There are some reports on the use of angiographic embolization to reduce the vascularity of the tumor, thus decreasing blood loss during surgical resections.[6]

Materials and Methods

This retrospective hospital-based study was conducted in the Department of Cardiovascular and Thoracic surgery, SKIMS Srinagar, from January 2003 to November 2013. All the cases of CBTs admitted in the Department of Cardiovascular and Thoracic surgery were taken up for the study. The patients were subjected to routine and specialized workup. A detailed history of each patient including age, sex, residence, presenting complaints with emphasis on swelling neck, pain, dysphagia, dysphonia, hypertension, Horner’s syndrome, and smoking was taken as per proforma. A thorough physical examination was done with special emphasis on local examination, cranial nerve assessment. Baseline investigations including complete blood count, kidney function tests, liver function tests, electrocardiogram, coagulogram, and chest X-ray were performed. Ultrasonography USG neck with Doppler was done for initial assessment. CTA/MRA of the neck was done in these patients as per the requirement. After all baseline and specific investigations, patients were taken for surgery with an informed consent. Thereafter, the data from all the patients who were involved in the study were analyzed for the variables which were to be studied. The analysis was undertaken and the results were tabulated and analyzed for statistical significance. Data collected as such were described in mean and percentage form. Intragroup comparison was made by simple t-test. SPSS (IBM Corp. Released 2013. IBM SPSS Statistics for Windows, Version 22.0. Armonk, NY: IBM Corp.) and Graphpad statistical software (http://www.graphpad.com/quickcalc; accessed April 2016) were used for statistical analysis of the data. $P < 0.005$ was considered to be significant.

Results and Observations

A total of 44 patients of CBT who were admitted and evaluated in the Department were included in the study. It was observed that majority of the patients in this study were females numbering 35 (79.45%), whereas there were nine (20.43%) males. All patients were adults, i.e., more than 20 years of age. It was observed that the most common age group involved was of patients 50–59 years of age; youngest patient being 22 years old and oldest patient was 60 years of age, with a mean age of 44.18 ± 10.24 years. Median age of CBT patients in this study was 46.5 years. A large number of the patients in this study were from rural area 36 (83.99%) in number, whereas only eight patients were from the urban area, i.e., 18.16% [Table 1]. Year-wise trends did not show any specific pattern. A total number of cases registered annually varied from 2 in 2003 to 9 each in 2012. Similarly, no particular pattern was seen in annual trends involving gender distribution of patients and the rural–urban distribution [Table 2]. All of the patients enrolled in this study were fully conscious at the time of admission to the hospital. All 44 patients (100%) in study presented with a complaint of neck swelling [Figure 1]. Mean duration of swelling was 23.16 ± 16.34 with a minimum of 3 months to as long as 96 months. Median duration being
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23 months. Twenty (45.45%) had the right-sided swelling and 24 (54.54%) had left-sided swelling. Forty-one (93.07%) had unilateral and three (6.81%) patients had bilateral neck masses. Eight (18.16%) patients had a history suggestive of smoking. Five (11.35%) had a history of hypertension and (9.08%) had a history of neck swelling in the family. Pain was associated feature in nine (20.43%) of patients. Mean duration of pain being 4.222 ± 1.787 months with a minimum duration of 2 months and maximum of 8 months. Median duration being 4 months dysphagia was found in three (6.81%) patients. No patient had a history suggestive of Horner's syndrome [Tables 3a-c]. USG was done in all four (100%) patients with CTA being 32 (72.64%, Figure 2), MRA in eight (18.16%), and both CTA and MRA in four (9.08%) of patients. Surgery was done in all 44 (100%) patients with a complete resection done in 41 (93.07%) and incomplete in three (6.81%) patients. Mean operating time was 3.552 ± 1.038 h with minimum of 2 h and maximum of 6 h and median time being 3 h; 28 (63.56%) patients were operated within time of 2–4 h, six (13.62%) in ≤ 2 h, nine (20.43%) in 4–6 h, and one (2.27%) patient in ≥ 6 h [Table 4]. Ten (23%) were Shamblin Grade 1, 29 (66%) had Shamblin Grade 2, and five (11%) had Shamblin Grade 3 [Table 5]. ECA repair was done in six (13.62%) patients, ICA repair in three (6.81%), ECA ligation in two (4.54%), and vascular graft was used in one (2.27%) patients. An average size of the mass was 2.886 ± 0.8481 cm by 2.477 ± 0.7149 cm, with a minimum of 1.5 cm by 1.0 cm and maximum of 5 by 4 cm with a median size of 3.00 by 2.5 cm [Figures 3 and 4]. Mean blood loss in our study was 213.41 ± 84.412 ml with a minimum of about 100 ml and a maximum of 500 ml and median of 200 ml. In 23 (52.21%) patients, blood loss was in between 101 and 200 ml, 13 (29.51%) 201 to 300 ml, 5 (11.35%) ≤ 100 ml, 2 (4.54%) 301–400 ml, and 1 (2.27%) had blood loss of 401–500 ml. Seven (15.89%) had transient cranial nerve palsy most commonly involving hypoglossal nerve which improved over a period of days to weeks. Three (6.81%) had permanent cranial nerve palsy. In six (13.62%) patients, there was local wound infection which was managed conservatively with dressings and antibiotics. There was a postoperative stroke in two (4.54%) patients. No operative mortality was seen. Mean duration of hospital stay was 7.045 ± 2.524 days with a minimum of 4 days and maximum of 15 days. Median duration of hospital stay was 6 days. Thirty-three (74.91%) patients had a hospital stay of 5–8 days, six (13.62%) had a hospital stay of 9–12 days, three (6.81%) had 3–4 days, and two (4.54%) had ≥ 12 days. Forty-three (97.61%) patients were proved to be paragangliomas on HPE while one (2.27%) patients had an inconclusive biopsy on histopathological examination. Three patients have died while on follow-up because of other ailments. Rest of patients are on regular follow-up and doing fine.

Discussion

Mathews (1915) remarked that “this rare tumor presents unusual difficulties to the surgeon and one should encounter it without suspecting the diagnosis, the experience will not be forgotten.”[10] The carotid body was first described by von Haller in 1743.[11] It is highly specialized organ located at the common carotid artery (CCA) bifurcation. Paraganglionic cells constituting this body sense pH, pO₂, and pCO₂ changes in the blood. While the size of the body may vary, the mean size is 5 mm × 3 mm × 1.5 mm. Weight for adults ranges from 1 mg to 47.4 mg, with a mean of 12.1 mg. Its feeding vessels run primarily from the external carotid artery. The function of the carotid body is related to autonomic control of the respiratory and cardiovascular systems, as well as blood temperature. The CBTs are uncommon, accounting for only 0.03% of all neoplasms, and 0.06% of head and neck tumors. Carotid body paraganglioma is more common in women with female: male ratio of 2.7:1. Moreover, that
was noticed in most of the reviewed articles and in our series females outnumbered males with a ratio of 3.88:1, possibly because the place of study is at an altitude of >5000 m from sea level. Luna-Ortiz et al. in 2005 concluded that women significantly predominated (96.9%) with a female: male ratio of 31.2:1. Although CBP has been observed in children, they are predominantly a disease of the middle age with the average age of onset at 45 years. In our study, the most common age group involved was of patients 50–59 years of age, youngest patients being 22-year-old, and oldest patients were 60 years of age, with a mean age of 44.18 ± 10.24 years. Median age of CBT patients in our study was 46.5 years. A large number of the patients in this study were from the rural area 36 (83.99%), whereas only eight patients were from the urban area, i.e., 18.16%. In this study, 44 patients were diagnosed with CBT over a period of 10 years, with no significant pattern detected over the decade. Erdogan et al. had in their study five patients with three of their patients were female and two were male. The ages of patients ranged from 44 to 68 years with a mean of 59.6 years. Koskas et al. described 39 carotid chemodectomas of which 14 men and 22 women, with a mean age 44.4 ± 5 (range: 21–78) years. Nazari et al. in their study had CBT patients which were predominantly females (82%). Age of diagnosis was 18–75 years old.

The incidence of bilateral carotid body lesions is approximately 10%. Most of these lesions are benign however malignant behavior is often encountered. For diagnosis of malignant carotid body paraganglioma, there are no clear histological characteristics that differentiate it from benign lesions. This diagnosis is reserved for the tumors with local, regional, and distant metastasis. The rate of malignancy is reported to be 6–12.5% of all cases. The 7–9% of the cases are hereditary. In our study, twenty (45.45%) had right-sided swelling and 24 (54.54%) had left-sided swelling, 41 (93.07%) had unilateral, and three (6.81%) patients had bilateral neck masses. Nazari et al. in their study had five patients which had bilateral CBT. The family history of CBT was positive in seven patients. Carotid body paraganglioma often present as slow growing, nontender neck masses located just anterior to the sternocleidomastoid muscle.
at the level of the hyoid bone. The tumor is mobile in the lateral plane, but its mobility is limited in the cephalocaudal direction. Occasionally, the tumor mass may transmit the carotid pulse or demonstrate a bruit or thrill. Because of its location in close approximation to carotid vessels and X–XII cranial nerves, tumor enlargement causes progressive symptoms such as dysphagia, odynophagia, hoarseness of voice, or other cranial nerve deficits. The patients may give a history suggestive of symptoms associated with catecholamine production such as fluctuating hypertension, blushing, obstructive sleep apnea, and palpitations. All patients n = 44 (100%) in our study presented with a complaint of neck swelling. Mean duration of swelling was 23.16 ± 16.34 with a minimum of 3 months to as long as 96 months. Median duration being 23 months eight (18.16%) patients had a history suggestive of smoking, five (11.35%) hypertension and four (9.08%) neck swelling in family. Pain was associated feature in nine (20.43%) patients. Mean duration of pain being 4.222 ± 1.787 months with a minimum duration of 2 months and a maximum of 8 months. Median duration being 4 months. Dysphagia was found in three (6.81%) patients. No patient had a history suggestive of Horners Syndrome. Nazari et al. in their study had all of the patients presented with a neck mass, mostly without pain (84%). Other symptoms included vertigo 4%, dysphasia 4%, and tinnitus 2%. There was no patient with cranial nerve involvement at presentation. In the study by Davidovic et al., eight of their cases presented as a large asymptomatic non tender neck mass and two each presented with dysphagia and hoarseness of voice. The size of the tumor has a great importance not only for its clinical manifestations but also for treatment. In 1971, Shamblin introduced a classification system based on tumors size. In our study, ten (23%) were Shamblin Grade 1, 29 (66%) Shamblin Grade 2, and five (11%) were Grade 3. As per Shamblin classification, seven of tumors were Type II and five were Type III in the study by Davidovic et al. Luna-Ortiz et al. in their study had four tumors grouped in Shamblin’s Class I, 24 in Class II, and 35 in Class III. Histologically, carotid body paraganglioma resembles the normal architecture of the carotid body. The tumors are highly vascular and between the many capillaries are clusters of cells called Zellballen. Thirty-five (80%) patients underwent FNAC of swelling and nine (20%) did not have FNAC done. Forty-one (93.07%) patients were proved to be paraganglionomas on HPE, while three (6.81%) patients had inconclusive biopsy on histopathological examination. Masilamani et al. described the fine needle aspiration cytology findings of one such tumor in a 27-year-old man. The aspirate was hemorrhagic with clusters of the round to oval cells showing moderate anisokaryosis. Delicate fibrous strands with spindle cells were observed within these clusters suggesting a diagnosis of CBT. The carotid angiography is the most useful diagnostic test for paragangliomas. The angiography demonstrates tumor blood supply and widening of the carotid bifurcation by a well-defined tumor blush (“lyre sign”), which is classic pathognomonic angiographic finding. MR and contrast CT are more effective noninvasive imagining modalities comparing with duplex ultrasonography, especially for small tumors. Radioimmunodetection of carotid body paraganglioma by In labeled anti-CEA antibody has also been described in literature. The differential diagnosis includes other tumors in this area, carotid artery aneurysms, and elongation. For this reason, using of percutaneous fine needle aspiration for preoperative diagnosis of carotid body paraganglioma can be very dangerous. USG was done in all 44 (100%) patients with CT angiography being 32 (72.64%), MRA in eight (18.16%), and both CTA and MRA in four (9.08%) patients. Resection of carotid body paraganglioma carries inherent risks of injury to the cranial nerves, carotid arteries, as well excessive blood loss. Reigner first attempted the resection of a carotid body paraganglioma in 1880, but the patients did not survive. Maydel was the first to remove a carotid body paraganglioma successfully in 1886, but the patient became aphasic and hemiplegic due to the internal carotid artery (ICA) ligation. In 1903, Scudder performed the first successful removal of carotid body paraganglioma. The surgical excision with careful subadventitial dissection is the treatment of choice for most carotid body paragangliomas (Shamblin I and II). The Shamblin III of carotid body paraganglioma may require resection of the external and/or ICA. If the ICA is encased in tumor or damaged during resection, immediate repair/replacement should be performed. Surgery was done in all 44 (100%) patients with complete resection in 41 (93.07%) and incomplete in three (6.81%) patients all of whom were Shambling Grade 3. Mean operating time was 3.552 ± 1.038 h with a minimum of 2 h and maximum of 6 h and median time being 3 h. Twenty-eight (63.56%) patients were operated within the time of 2–4 h, six (13.62%) in < 2 h, nine (20.43%) in 4–6 h, and one (2.27%) patient in ≥ 6 h. The second problem during tumor excision is bleeding, which sometimes can be massive. In such cases, clamping of all carotid arteries is useful, with the placement of an internal carotid shunt. In our study, mean blood loss was 213.41 ± 84.412 ml with a minimum of about 100 ml and maximum of 500 ml and median of 200 ml. In 23 (52.21%) patients, they had a blood loss in between 101 and 200 ml, 13 (29.51%) 201–300 ml, 5 (11.35%) ≤100 ml, 2 (4.54%) 301–400 ml, and 1 (2.27%) had blood loss of 401–500 ml. David et al. in their study had an average blood loss of 973 ml (range: of 150–3000 ml). Some other articles recommend angiographic embolization preoperatively. The preoperative embolization of a carotid body paraganglioma can be performed by ethanol or polivinyl alcohol. The final result is a complete devascularization. Embolization was not done in any of our patients. Most authors recommend radiotherapy for giant and recurrent carotid body paragangliomas and with malignant carotid body paragangliomas metastatic to the regional lymph nodes.
The modern surgical techniques have reduced the risk of postoperative stroke in carotid body paraganglioma resection to less than 5%. However, the incidence of cranial nerve injury remains strikingly high, ranging from 20% to 40%. In 20% of patients, the neurological deficits are permanent. The patients with ICA reconstruction should undergo duplex scanning periodically to identify graft stenosis. Seven (15.89%) had transient cranial nerve palsy most commonly involving hypoglossal nerve which improved over a period of days to weeks. In our study, ECA repair was done in six (13.62%) patients, ICA repair in three (6.81%), ECA ligation in two (4.54%), and vascular graft was used in one (2.27%) patients. Vascular injuries were repaired on the table with no adverse sequelae and three (6.81%) had permanent cranial nerve palsy. In six (13.62%) patients, there was local wound infection which was managed conservatively with dressings and antibiotics. There was postoperative stroke in two (4.54%) patients. No operative mortality was seen. Mean duration of hospital stay was 7.045 ± 2.524 days with a minimum of 4 days and maximum of 15 days. Median duration of hospital stay was 6 days. Thirty-three (74.91%) patients had a hospital of 5–8 days, six (13.62%) had 9 to 12 days, three (6.81%) had 3–4 days, and two (4.54%) had ≥12 days. David et al. in their study had external carotid artery ligated in seven patients. In one patient, the CCA was injured during dissection and was repaired. In another, the proximal ICA was inadvertently injured needing temporary shunting followed by resection of a segment and reanastomosis to the CCA. There was no in-hospital mortality, and none of the patients developed perioperative cerebrovascular complications. Two patients (10%) developed postoperative cranial nerve deficits (9th, 10th, and 12th) and one of them was discharged on a nasogastric, and one of them was discharged on a nasogastric tube for feeding. None of the patients developed Horner’s syndrome. The average postoperative hospital stay was 4.18 days (range: 3–8 days).[17]

Conclusion

Although rare, CBT is still a pathology that we encounter in our experience, and it should be kept in mind as a differential diagnosis for painless lateral neck masses. Our long-term experience is comparable with other reported case series where surgical intervention conferred a long-term survival advantage despite associated cranial nerve comorbidities. Excision of a CBT is recommended at the time of initial diagnosis in patients in good general health to avoid the difficulty of subsequent excision of an enlarging and highly vascular tumor with possible encaement of the carotid arteries. Preoperative status of patient and Shamblin class of tumor are the important factors for determining the surgical outcome of patients. The outcome improves with expertise of the surgeon. Diagnostic gadgets available are of paramount importance in the diagnosis of CBT. Meticulous subadventitial dissection and excision are the key to achieve complete excision with minimal morbidity. This method should therefore be used for the excision of CBT. Facilities for shunting and arterial repair should always be available. Ideally, preoperative assessment of cross cerebral circulation, meticulous preoperative planning, and multidisciplinary team approach leads to successful outcome with reduced postoperative morbidity. These highly vascular tumors although rare should be operated by experienced vascular surgeon to minimize complications.

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Conflicts of interest

There are no conflicts of interest.

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