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Hybrid Surgery in Potato Tumor

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Abstract

Objective: To analyze the outcomes of patients with Carotid body tumors of neck in a tertiary care hospital. To compare the effect of preoperative embolization in CBT.

Study design: Retrospective study.

Place and duration of study: Shifa International Hospital, Islamabad. From 1998 to 2019.

Methodology: Vascular data base was searched from 1998 to 2019 for patients who presented with pulsatile, painless neck swelling in anterior triangle of the neck. A total of 998 cases were reviewed out of which 48 cases were included in the study. All had preoperative CT scan done and all underwent surgical excision of the tumor.

Results: A total of 48 cases were included in the study during the period of 1998 to 2019, 17 were male and 31 were female with age range 20 years to 82 years with median age of 45 years. Five cases were referred to us by the radiology department with incidental diagnosis of CBT while performing ultrasound guided FNAC of provisionally diagnosed thyroid swelling or cervical lymph node. All patients presented to the hospital with complaints of painless neck swelling with 27 cases having left sided CBT, 13 with right sided CBT and 8 bilateral CBT reported.

Conclusion: Carotid body tumors do not present with any specific symptoms. Surgical excision is the main stay of treatment. Pre-operative embolization reduces surgical complications.

Keywords: Carotid body tumor; Hypoglossal nerve; Carotid artery; Paraganglioma; Chemodectoma

Introduction

Carotid body Tumors (CBT) also known as chemodectoma or paraganglioma are rare benign neoplasms that arise from neural crest cells with an incidence of 1:30,000 [1]. It is the most common type of paragangliomas of head and neck with highly vascular nature. CBT may be sporadic (60%), familial (10% to 50%) and hyperplastic [2]. Familial CBT are most commonly multifocal and bilateral [3]. CBT present as non-tender, slowly enlarging mass in anterior triangle of neck [4,5]. On examination there may be a firm mass that is mobile only in horizontal plane, known as Fontaine's sign [6]. Majority of these tumors are benign and nonfunctioning in nature but 5% to 30% are functional and malignant [7]. Furthermore, due to highly vascular and infiltrating behavior early diagnosis and management is recommended [8]. CT angiography and MR angiography are used to diagnose CBT [9]. CT scan shows rapid enhancement with contrast and splaying of internal and external carotid arteries (ICA and ECA) known as lyre sign (Figures 2-6). CBT have a salt and pepper appearance on MRI [10]. Surgery is the main line of treatment [11]. CBT is classified according to shamblin classification with type-1 including those tumors that are easily resectable small in size, type-2 tumor partially encase the vessels and type-3 tumors completely surround the vessels which are difficult to resect (Figure 1) [12]. According to a meta-analysis by Jensen et al. [10] cranial nerve damage was 3%, 17% and 39% and complication rate 0%, 1%, 10% according to Shamblin class 1, 2, 3. Shamblin class 3 tumors are associated with increased risk of stroke due to ICA manipulation and ECA ligation [13]. Vaux Robertson et al. [13] found that mean age of patient with CBT was 47.3 yrs with majority of females, 9.6% bilateral and 4.1% malignant cases. 2.3% of these cases were functional. Most common type was shamblin 2 and mean 30 days mortality and stroke 2.29% and 3.53%. Cranial Nerve Injury (CNI) rate was 25.4% with hypoglossal nerve the most commonly injured nerve. 5.24% of neck hematoma was re-explored. This meta-analysis also showed increase CNI and stroke rate in shamblin 3 tumors. Preoperative embolization of tumor had no significant effect on postoperative neck hematoma or drain loss [14]. Pavlos et al. [14] showed lower blood loss during procedure and shorter surgery time in cases with embolization before CBT resection but no difference in stroke, CNI and length of stay [15].

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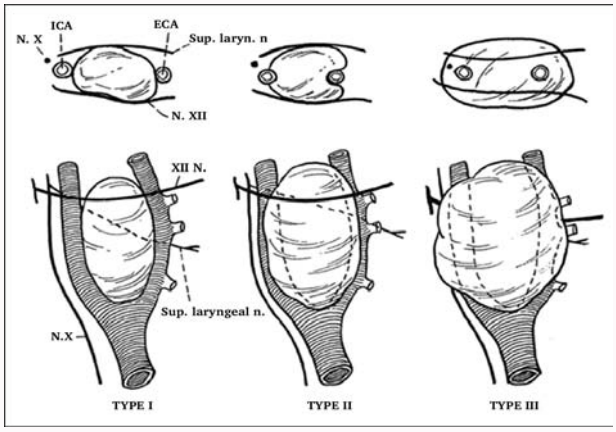


Figure 1: The Shamblin classification of the difficulty of surgical resection. Type I tumours are localised and easily resected. Type II includes tumours adherent or partially surrounding vessels. Type III tumours intimately surround or encase the vessels.

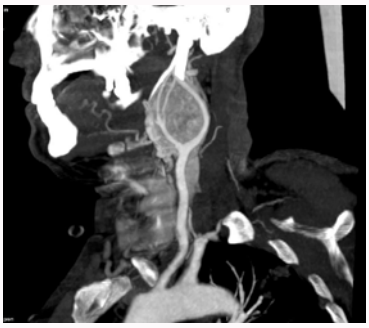


Figure 2:

Figure 3:

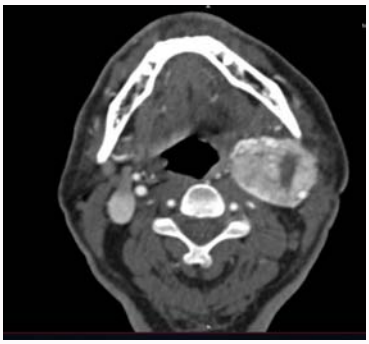
Hinojosa et al. [15] compared type of dissection techniques for surgical treatment of CBT. They concluded that shamblin 2 was the most common type operated on and one group in which Retrocarotid Dissection (RCD) dissection from above was done mean blood loss was 480ml as compared to 690 ml of another group in which Standard Caudocranial (SCC) dissection from below took place. The RCD group had less operative time and shorter hospital stay 172 min *vs.* 260 min and 5 days *vs.* 9 days of the SCC group. Complication rate was found higher in SCC group 34% *vs.* 22% including cranial nerve deficits and cerebrovascular accidents [16]. There is no decent study available in Pakistan sharing the experience of carotid body tumor management. This study entails experience of managing CBTs with surgical management with and without pre-op-embolization in a tertiary care hospital.

Methods

After approval from the hospital ethical and research committee, medical records were retrospectively reviewed, from 1998 to 2019. All patients between ages of 40 to 80 years with chief complain of painless neck swellings were selected. Those patients who had thyroid swelling or enlarge cervical lymph node were excluded from the study. Patients with swelling other than anterior triangle of the neck or vascular swellings like aneurysms and AV malformations and congenital vascular anomalies were also excluded.

A total of 48 cases were selected of the 998 cases reviewed. All had basic serum workup done with CT Carotid angiography confirming carotid body tumor. Twenty-four hour urine Vanillylmandelic Acid (VMA) and plasma metanephrines were done in all patients with history of hypertension. Surgery was planned after explaining risks and benefits of the procedure to patient with informed and written consent taken and referral to anesthetist for further debriefing. Preoperative embolization of the CBT was done only for tumors more than 4 cm in size on CT scan.

All patients were prepped from mandible down to sternal joint in supine mild reverse trendelenburg position with head rotated to



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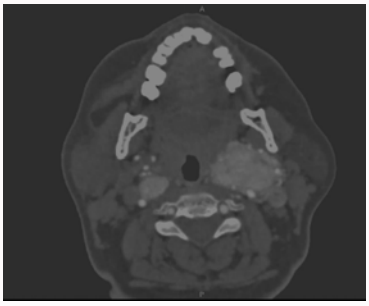

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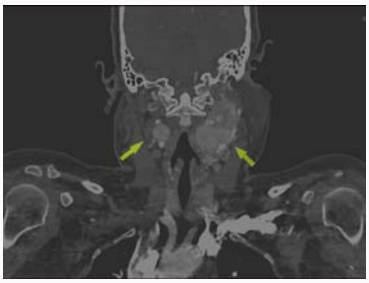

Figure 6:

opposite side and pad under shoulder. Conventional general anesthesia was used with Mean Arterial Pressure (MAP) kept between 80 mmHg to 100 mmHg. Oblique incision along the anterior border of the sternocleidomastoid muscle was used to expose carotid bifurcation dividing fascial vein in the process. Proximal and distal control was achieved early in shamblin 1 and 2 tumors while latter in shamblin 3. Hypoglossal nerve was protected with nerve loops away from surgical field in some cases. Subadventitial plane was used to dissect tumor of the vessel in caudo-cranial fashion. Branches supplying the tumor were ligated early with most commonly the ascending pharyngeal artery originating either from ECA or carotid bifurcation. In some cases of shamblin 3 types ECA was ligated due to complete adherence of tumor to the vessel. All precautions were taken to preserve the nerves, mainly vagus, accessory, hypoglossal, and glossopharyngeal. Careful anatomical dissection was done around ICA to preserve it but in few cases reconstruction was done with interposition graft. At the time of extubation vocal cords were checked by the Anesthetist in all cases. The tumor resected was marked and sent for histopathological examination in all cases. After the surgical procedures all the patients were shifted to ICU for close monitoring for one day and shifted to surgical ward once stable. Oral fluids were allowed early and assisted supervised mobilization was done 4 h to 6 h after surgery. Prophylactic dose of subcutaneous low molecular weight heparin was administered to all patients postoperatively and dual anti platelets resumed once orally allowed. A single preoperative dose of cefazolin 1 g was used in all patients. Drain was usually removed on first post-op day and patient discharged home on second post-op day with one week follow up visit.

All data including demographics, duration of complain, preoperative CT scan reports, operative findings, hospital stay, post-surgical recovery and complications, were collected after reviewing the medical records and entered in SPSS 21. Patients follow up consults were also explored and few patients was called for in case of any query. All the data was analyzed by our expert analyst in SPSS 21.

The shamblin hour's classification of the difficulty of surgical resection. Type I tumors are localized and easily resected. Type II includes tumors adherent or partially surrounding vessels. Type III tumors intimately surround or encase the vessels [12].

Results

A total of 48 cases were included in the study during the period of 1998 to 2019, 17 were male and 31 were female with age range 20 years to 82 years with median age of 45 years (Table 1 and 2). Out of 48 cases 15 cases were referred from district hospitals located in mountainous areas like Muzaffarabad and surrounding northern areas (altitude >2400 feet). Five cases were referred to us by the radiology department with incidental diagnosis of CBT while performing ultrasound guided FNAC of provisionally diagnosed thyroid swelling or cervical lymph node. All patients presented to the hospital with complaints of painless neck swelling with 27 cases having left sided CBT, 13 with right sided CBT and 8 bilateral CBT reported (Table 3). 85.7% of CBT were asymptomatic with 14.2% having symptoms. The proportion of CNI, difficulty swallowing, and headache as chief complains on presentation was 2% while 1% of patients had symptoms of vertigo, dizziness and tinnitus.

Duplex ultrasound showed a highly vascularized solid hypoechoic mass causing separation of ICA and ECA. CTA showing a rapidly enhancing mass causing splaying of ICA and ECA was done in all cases. Majority of the tumor were 4 cm to 6 cm in size (Table 4). All (48) the patients underwent surgical excision the tumor. Twenty patients (41.7%) had preoperative embolization of the tumor to reduce intraoperative bleeding. According to Shamblin classification 35.4% were shamblin I, 52.0% were shamblin II and 12.5% were shamblin III (Table 5).

Table 1: Gender distribution.

Gender Distribution	
Males	Females
17	31

Table 2: Age distribution.

Age Distribution			
20 years to 40 years	41 years to 60 years	61 years to 80 years	>80 years
5	36	6	1

Table 3: Site Distribution of CBT patients.

Site of Tumor		
Left	Right	Bilateral
27	13	8

Table 4: Number of patients with different size of CBTs.

Size Distribution			
≤ 4 cm	4 cm to 6 cm	6 cm to 8 cm	>8 cm
10	26	9	3

Table 5: Number of patients with Shambilin Classification.

Shamblin Class/Case Distribution		
Shamblin I	Shamblin II	Shamblin III
17	25	6

Average operative time was 220 min (45 min to 280 min) in the non-embolization group while in the pre-op embolization group was 120 min (30 min to 260 min). Average blood loss was 132 ml (10 ml to 1300 ml). Blood loss was measured by amount of blood in suction bucket at the end of operation including the squeezed surgical sponges into the container. The amount of blood loss for pre-op embolization patients (average 260 ml) was much less than those without pre-op embolization (average 670 ml).

Mean hospital stay was 2.5 days. Patients were followed at 1 week, 4 weeks, and 8 weeks.

Postoperative stroke over 30 days' span was 4.7%. Two patients had a major MCA infarct with major disability. One of the patients had thrombolysis with TPA out of the contrary and recovered completely in 48 h, later carotid stenting was performed. Other patient was managed conservatively who recovered partially as seen over the follow-up visits. There was no mortality.

Both the patients who had stroke were shamblin III CBT. The overall CNI rate was 14.5% with hypoglossal nerve the most commonly injured nerve (71.4%). Out of 5 cases 3 patients had tumor involving hypoglossal nerve completely so it had to be sacrificed for complete excision of the tumor, one was partially transected accidently and repaired, and the other incurred a temporary neuropathy due to retraction. Patients with permanent nerve injury had adapted to tongue deviation in follow-up visits. The last two recovered completely. Horner syndrome complicated

Table 6: Complication rates after resection of CBT.

Complications	Pre-Operative Embolization Group	Non Embolization Group	Number of Cases
Injury to hypoglossal nerve	2	3	5
Horner's syndrome	1	1	2
Injury to Vagus nerve	0	0	0
Injury to recurrent laryngeal nerve	0	1	1
Superior laryngeal nerve	0	0	0
Injury to Glossopharyngeal nerve	0	1	1
Injury to Accessory nerve	0	0	0
Mandibular branch of facial nerve	1	0	1
Injury to ICA with interposition grafting	1	2	3
Ligation of ECA	2	2	4
Stroke	1	1	2
Wound hematoma	2	8	10
Death	0	0	0

Table 7: Type of Carotid Body Tumor.

Type of Carotid Body Tumor				
Asymptomatic CBT	Symptomatic CBT	Catecholamine Secreting CBT	Benign CBT	Malignant CBT
40	8	8	45	3

4.16 % of tumor excisions; the mandibular branch of fascial nerve was affected in 2.0%, the glossopharyngeal and recurrent laryngeal nerve in 2.0%. Injuries to these nerves were temporary and full recovery was reported in follow-up visits. One of the patient with Horner's syndrome required tarsorrhaphy after 6 months for cosmetic reasons. Accessory nerve and vagus nerve didn't incurred any injury at all. External carotid artery was ligated and resected en mass with tumor in 8.33% of patients due to large and adherent tumor. Internal carotid was resected and reconstructed with great saphenous vein interposition graft in 6.25% cases. Wound hematoma was present in 20.8% cases and only one case required evacuation of hematoma due to bleeding from tributary of external jugular vein. The overall 30 days mortality was nil. All the resected tumors were sent for histopathology which reported tumor arranged in zellballen like growth pattern, well developed nested pattern of tumor cells with intervening stromal component of delicate fibrovascular tissue and sustentacular cells at the periphery of cell nests. The tumor cells are round with hyperchromatic nuclei, disposed chromatin and abundant granular eosinophilic cytoplasm. Mitosis is rare. On Immunohistochemistry, Synaptophysin was positive, chromogranin was positive and S-100 was also positive in sustentacular cells. 6.25% of histopathology report confirmed malignant tumor. These cases were referred for radiation.

Discussion

Carotid Body Tumor (CBT) is a rare benign tumor with incidence of less than 1 in 30000 [17].

Carotid body being situated at carotid bifurcation originates from neural crest cells and helps in keeping homeostasis in response to altering pH, carbon dioxide and oxygen levels by increasing the ventilation. There are mainly three types of carotid body tumors; familial, sporadic and hyperplastic. Sporadic type is the most common form comprising of 85% CBT with reported age of onset at 45 years. Familial CBT are more commonly present in younger age groups in second to fourth decade. Hyperplastic type is present in population living at high altitudes (usually more than 2000) or obstructive pulmonary diseases [18]. CBT are usually unilateral in 95% of sporadic cases and 67% of familial cases. Familial cases are usually caused by germ line mutations in Succinate Dehydrogenase genes [19]. CBTs are asymptomatic and slow growing tumors with mean Doubling Time (TD) estimated by Jansen et al being 7.13 years with a median growth rate of 0.83 mm/year [20]. On clinical examination CBT is vertically fixed with Bruit heard in few cases. 10% present with symptoms of pain, hoarseness, Horner syndrome, dysphagia or shoulder drop. Functional CBT presents with hypertension, diaphoresis, and palpitations.

MRI is considered as the gold standard investigation for the diagnosis of CBT showing the classical salt and pepper appearance on T2 weighted images [21]. mCT scan with contrast is also helpful in demonstrating the splaying of internal and external carotid arteries by a hypervascular tumor. Angiography is helpful in finding the feeder vessels and preoperative embolization of the tumor. Metaiodobenzylguanidine [MIBG] scintigraphy can be done only in functional CBT [22]. Biopsies are usually not recommended due to highly vascular nature of the tumor [23].

CBT is aggressive tumor with invasion of nearby vessels and nerves and has a tendency to turn into malignant lesions in 10% of the cases, thus surgical excision is the main stay of treatment [24].

Due to highly vascular nature of the tumor preoperative embolization is advised and is found to be beneficial by shrinking the tumor and facilitating easy surgical removal [25,26]. While, on the other hand it is not recommended especially in small tumors due to associated risk of cerebrovascular events [27]. Although surgical resection is the recommended treatment it comes with significant comordities especially in shamblin grade 3 tumors [28]. Preoperative angiography and embolization is advised in large tumors shamblin grade 3 or tumors more than 4 cm in size to decrease perioperative bleeding and neural damage. However, a review by Abu-Ghanem et al. [28] and study by Cobb et al found no benefit of preoperative embolization in CBT resection [29].

Radiotherapy is another treatment option in these patients though the tumor is not radiosensitive. This modality is usually reserved for elderly, surgically unfit, or those patient with very large or multifocal tumors in whom surgical intervention might result in increased morbidity. The goal of radiotherapy in these patients is to decrease the progression of tumor. Postsurgical complications are not uncommon, a study by Kim et al. [29] concluded that every 1 cm decrease in the distance to the skull base results in 1.8 times increase in >250 mL of blood loss and 1.5 times increased risk of cranial nerve injury. Management of CBT should be carefully planned accurately weighing the pros and cons of treatment offered.

Our study had certain limitations as it was a single center retrospective study. Two different surgeons were involved at different time intervals. Due to expensive treatment offered in our center the number of cases were limited.

Conclusion

Carotid body tumor is an aggressive but benign tumor. Surgical treatment should be offered to patients after discussing the case

in multidisciplinary meeting including intervention radiologist, neurologist and vascular surgeon. Preoperative embolization should be considered in shamblin II or higher tumors. This hybrid technique reduces per operative bleeding, avoid the need of clamping carotid arteries and facilitate surgical excision eventually reducing the surgical risk and improving surgical outcomes.

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