

Carotid Body Tumours: Outcomes of a Multidisciplinary Approach

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ABSTRACT

Background: Carotid paragangliomas, or carotid body tumours (CBT), often present significant surgical challenges. The objective of this study was to examine the impact of a multidisciplinary surgical approach on carotid body tumour resection and clinical outcomes.

Methods: A retrospective review of all patients who underwent carotid body resection within the Calgary Health Region from January 1, 1994, to December 31, 2019 was conducted. Patient demographics, operation parameters, co-morbidity, post-operative complications and the usage of pre-operative embolization were compared using appropriate statistical methods. A p-value of < 0.05 was considered statistically significant.

Results: Of the 49 patients included in this study, 28 underwent resection by a single specialty and 21 underwent resection by a multidisciplinary team. The multidisciplinary team operated on significantly larger ($p < 0.001$) and more complex CBTs ($p = 0.006$). When analyzed as a group, operating time ($p < 0.001$) and length of hospital stay ($p = 0.019$) were significantly longer for patients operated on by the multidisciplinary team, reflecting the greater complexity of the CBTs in the multidisciplinary cohort. However, a sub-group analysis of high Shamblin grade tumours by surgical team showed no differences. Pre-operative embolization was performed in 41% of cases. There were no post-operative deaths.

Conclusions: Surgical management of CBTs can be challenging. However, we were unable to find any improvement in OR time, blood loss, length of hospital stays or complication rate in the multidisciplinary group. Pre-operative embolization did not significantly diminish intra-operative blood loss.

Keywords: carotid body tumours, surgical management, multidisciplinary approach

INTRODUCTION

Paragangliomas of the head and neck are rare, with an incidence of 1-2/100 000 people. Of all head and neck paragangliomas, 65% are carotid body tumours. Since the first excision of a carotid body tumour in 1888, surgical excision has been the mainstay of management. However, the significant morbidity and mortality associated with carotid body tumour surgery, including stroke, cranial nerve deficits and death, has resulted in an evolution in management strategies to include pre-operative embolization, vascular shunting and radiation therapy [1-3].

The literature on carotid body tumour management is mostly limited to case reports and series describing the outcomes of single surgeons or single specialty programs. Several large, multicentre reviews have been conducted [4-15], though no Canadian centre has reported on carotid body tumour management in nearly 10 years [16-19]. Core to our management of this pathology in our centre is a combination of a relatively high volume of cases thought to be due to geographical factors (high elevation), and a multidisciplinary surgical team of a vascular surgeon and an otolaryngologist – head and neck surgeon. We are in a unique position to investigate the utility of a multidisciplinary surgical approach to carotid body tumour management. The objective of our study is to review and analyze the experience and outcomes of the management of carotid body tumours at a single centre. The data collected from this study will improve our understanding of the quality, safety and effectiveness of the treatment approach currently used.

METHODS

From January 1994 to present, both a single discipline and multidisciplinary approach to carotid body tumor resection has been used at the Calgary Health Region (now Alberta Health Services). The single discipline consisted of either an otolaryngologist - head and neck surgeon or a vascular surgeon; the multidisciplinary group consisted of both an otolaryngologist and a vascular surgeon. The medical records of patients who underwent carotid body resection between January 1, 1994 and December 31, 2019 were identified through the Calgary Health Region discharge database and the surgeons' clinical database. Patients were identified with ICD codes 9 and 10 with operating codes 1JE87LA excision partial, carotid artery using open approach without tissue, IJ80LA repair, carotid artery using open approach without tissue, 1JD89 excision total, carotid body and carotid sinus using open approach, 39.8 operations on carotid body, carotid sinus and other vascular bodies. The hospital record as well as outpatient clinic charts were reviewed. The study was approved by the University of Calgary Conjoint Health Research Ethics Board.

Patients in the aforementioned databases, who underwent carotid body tumour excision between January 1, 1994 and December 31, 2019 and who were at least 16 years of age were included. Patients with other paragangliomas of the head and neck were excluded. Demographic data obtained included sex, age, pre-operative imaging and embolization, estimated operative blood loss, length of stay, operative time, and postoperative complications.

The decision to embolize tumours before surgery was at the discretion of the surgeon and was based primarily upon the size and extent of the lesion. Tumours were classified according to the criteria established by Shamblin [20]. Preoperative comorbidities, based on the patient's pre-operative assessment were classified according to the Adult Comorbidity Evaluation 27 (ACE-27) score and the Charlson score. Post-operative complications were scored using the Clavien scale [21]. Statistical analysis included Student t-tests and Mann-Whitney U tests which were used to compare continuous and categorical outcomes (Stata, version 11. Stata Corp. College Station, Tx, USA).

RESULTS

49 patients were eligible for inclusion during the 25-year study period. Patient demographic data are shown in [Table 1](#). Overall, 57.1% of the cohort was female with a mean age of 51.5 years. Patients operated on by the multidisciplinary team were significantly younger than those operated on by a single specialty (45 years vs. 56

years, respectively $p = 0.014$). The mean BMI for the single specialty group and the multidisciplinary group were 27.0 and 28.9, respectively ($p = 0.359$). In the single specialty group, 46.4% of patients presented with Shamblin I lesions and 53.6% presented with Shamblin II or III lesions. The multidisciplinary approach comprised 9.5% of patients presenting with Shamblin I lesions and 90.5% presenting with Shamblin II or III lesions.

Table 1: Patient Demographics.

	Single Specialty (n=28)	Multidisciplinary Team (n=21)	p-value
Gender			
Male	6	9	0.098
Female	22	12	
Age at Procedure			
Mean (years)	56	45.4	0.028
[95% CI]	49.41 - 62.53	38.54 - 52.26	
BMI			
Mean	27	28.9	0.35
[95% CI]	24.94 - 29.08	24.67 - 33.10	
Shamblin			
Grade I	13	2	0.006
Grade II / III	15	19	
ACE 27			
Grade 0	12	11	0.824
Grade 1	12	6	
Grade 2	3	3	
Grade 3	1	1	

All 49 resections were technically successful, with no perioperative deaths recorded. The mean operative time for the single disciplinary group was shorter than the multidisciplinary group (120.5 minutes' vs 197.0 minutes, $p < 0.001$). Estimated blood loss was 173.8mL (range: minimal - 400mL) in the single specialty group and 279.8mL (range: minimal - 2000mL) in the multidisciplinary group ($p = 0.263$). The length of hospital stay was shorter for the single disciplinary group (3.4 days vs. 5.0 days, $p = 0.037$) for the multidisciplinary group. Of the single surgeon operations, only 7.1% required resection of the internal carotid artery (ICA). In the multidisciplinary group, 28.6% required ICA resection; of these, 50% were reconstructed with a vein graft, 17% with prosthetic, and 33% with primary closure. The tumours resected by a multidisciplinary team were significantly larger than those resected by a single specialty (3.90cm vs. 2.35cm, $p < 0.001$).

Embolization was performed on 40.8% of all tumour resections; 28.6% of the single surgeon tumours were embolized compared to 51.7% in the multidisciplinary group ($p = 0.077$). Blood loss between embolized and non-embolized patients was not significantly different (219mL vs. 224mL, $p = 0.960$). Indeed, in a subgroup analysis of Shamblin 2 tumours, there was no difference in blood loss (350mL vs 217mL, $p = 0.513$) or length of hospital stay (3.78 days' vs 5.83 days, $p = 0.150$) when comparing embolized versus non-embolized tumors.

The overall neurological or vascular complication rate was 34.7%. The neurological complications observed in the single specialty cohort included intentional sacrifice of the superior laryngeal nerve, marginal mandibular nerve injury and vocal cord paralysis. The neurological complications observed in the multidisciplinary cohort included vocal paralysis/paresis, marginal mandibular nerve praxia, and spinal accessory nerve injury. Cranial

nerve injury was the most common complication of surgery with an overall incidence of 32.7%. In the single disciplinary group, cranial nerve complications occurred in 21.4% of patients. These complications were mostly seen in Shamblin I and II tumours. In the multidisciplinary group, there was a cranial nerve complication rate of 47.6%, though these mostly occurred in Shamblin II and III tumours. Interestingly, there was no difference in rates of neurological complications comparing low (Shamblin I) versus high grade (Shamblin II/III) tumours ($p = 0.116$). Additionally, higher mean blood loss was not associated with a higher incidence of neurologic complications (170 mL vs 306 mL, $p = 0.159$). There were no neurologic complications associated with the embolization procedure.

One patient suffered a stroke in the multidisciplinary group with permanent arm weakness. At 48 hours postoperatively, 32.1% of patients in the single specialty group had a grade I Clavien score and 3.6% had a grade IIIa secondary to a postoperative hematoma that required return to the operating room for evacuation. No further complications were seen at 1 week or 1 month. 71.4% of patients in the multidisciplinary group had a grade I Clavien score. At 1 week postoperatively, 9.5% continued to have a grade I Clavien score (neurologic deficit due to stroke), and 4.8% had a grade II Clavien score secondary to wound infection. At one month post-operatively, one patient in the multidisciplinary group developed a pneumonia and a further developed a wound infection. There were no mortalities in either group. First bite syndrome developed in 23.8% of patients who underwent multidisciplinary resection, and 7.1% who underwent single specialty resection. Mean follow up was 719 days in the single specialty group (range, 16-2599), and 735 (range 33-2535) for the multidisciplinary group.

As seen in [Table 2](#), in comparing low grade (Shamblin I) versus high grade (Shamblin II/III) tumours, we found no difference during the resection when it came to operative blood loss or length of hospital stay. Higher grade tumours trended towards increased operative time (103 minutes vs. 136 minutes, $p = 0.071$), underscoring the increased complexity of these tumours, although this was only observed in the single disciplinary cohort. Interestingly, although low grade tumors tended to be smaller, the difference in size between low- and high-grade tumours was not statistically significant.

Table 2: Comparison of Low vs. High Grade Shamblin Tumours by Surgical Team.

	Single Specialty (n=28)		p-value	Multidisciplinary Team (n=21)		P-value
	Shamblin I (n=13)	Shamblin II / III (n=15)		Shamblin I (n=2)	Shamblin II / III (n=19)	
Age			0.0476			0.0455
Mean (yrs)	62.7	50.1		25.4	47.5	
[95% CI]	55.18 - 70.24	39.92 - 60.34		22.63 - 28.20	40.63 - 54.38	
Operative Time			0.0712			0.557
Mean (min)	103.1	135.6		159	201.1	
[95% CI]	81.55 - 124.61	106.4 - 167.8		-584.48	154.34 - 247.77	
Estimated Blood Loss			0.4463			0.5445
Mean (mL)	197.2	155.8		100	300.94	
[95% CI]	121.59 - 272.81	70.72 - 240.82		100 - 100	70.57 - 531.31	
Hospital Stay			0.4932			0.3781
Mean (days)	3.67	3.2		3	-25.42	
[95% CI]	2.20 - 5.13	2.60 - 3.80		5.16	3.57 - 6.74	
Tumour Size			0.1158			0.6259
Mean (cm)	2.02	2.63		3.55	3.94	

[95% CI]	1.34 - 2.70	2.16 - 3.11		1.64 - 5.46	3.42 - 4.47	
Preop Embolization			0.221			0.171
No	11	9		2	7	
Yes	2	6		0	12	

A further analysis of high-grade tumours (Table 3) showed no differences in rates of embolization (40.0% vs 63.2%, $p = 0.300$) or estimated blood loss (156 mL vs 301 mL, $p = 0.271$) between the single specialty and multidisciplinary groups. Single specialty operations were again associated with a decreased length of stay (3.20 days' vs 5.16, $p = 0.035$) and decreased operative time (136 min vs 201 min, $p = 0.025$).

Table 3: Subgroup Analysis of High Shamblin Grade Tumours.

	Single Specialty (n=15)	Multidisciplinary Team (n=19)	p-value
Operative Time			0.0251
Mean (min)	135.6	201.1	
[95% CI]	106.41 - 164.79	154.34 - 247.77	
Preop Embolization			0.3
No	9	7	
Yes	6	12	
Blood Loss			0.2712
Mean (min)	155.8	300.9	
[95% CI]	70.72 - 240.82	70.57 - 531.31	
BMI			0.157
Mean	26.7	30.1	
[95% CI]	24.21 - 29.23	25.56 - 34.65	
Hospital Stay			0.0345
Mean (days)	3.2	5.2	
[95% CI]	2.60 - 3.80	3.57 - 6.74	

DISCUSSION

We here present the outcomes of carotid body tumour management in a single, high-volume center in the contemporary era. Surgical resection of these tumours is the standard of care. Due to the complex and rare nature of this pathology, post-operative morbidity remains high, and this is particularly true for high grade disease. In our centre, we selectively apply a multidisciplinary approach to reduce this risk. Our overall neurological or vascular post-operative complication rate was 34.7%. The majority of this was comprised of cranial nerve complications, with an overall incidence of 32.7%. In the single disciplinary group, cranial nerve complications occurred in 21.4% of patients. In the multidisciplinary group, the cranial nerve complication rate was more than double, at 47.6%. The observed rate of postoperative cranial nerve deficiencies was slightly less than that reported by Kasper et al (52%) in their similar multidisciplinary approach [20]. Our multidisciplinary cohort was similar in patient age, complexity of tumour, and rate of embolization. Furthermore, in this study, a single disciplinary team was associated with decreased operative time and decreased length of stay. The present data do not demonstrate a benefit to a multidisciplinary approach.

We generally reserve the use of a multidisciplinary team for complex disease. They were responsible for managing most high-grade tumors, and the average size of tumour resected was 1.55cm larger. A subgroup analysis of

Shamblin II and III tumours revealed no statistical difference between the multidisciplinary and single specialty group for blood loss, but there was a difference in length of stay and operative time favoring the single specialty group. Overall, these findings should be taken in context, given the selectivity bias between the two groups with the multidisciplinary group operating on more complex tumours, as indicated by longer resection times and length of stay. It should be kept in mind that patients in the complex, multidisciplinary group may require complicated vascular (e.g. ICA reconstruction) or otolaryngologic (e.g. mandibular resection) procedures that are typically outside of the scope of practice for the single specialty surgical team. We feel that it is unlikely that this additional team posed additional harm to patients.

Historically, higher grade CBTs have been associated with increased rates of surgical complications¹⁰. This was not demonstrated in our study, where we found equivalence in our rates of blood loss, hospital stay, operative times, and rates of vascular/neurological complications when comparing high versus low-grade tumours resected by single or multidisciplinary teams. The reason for this is unclear, but may reflect the elevated level of combined expertise associated with the multidisciplinary team, particularly when dealing with the highest grade tumors. This may also reflect improvements in surgical technique. This may also indicate that there may be other anatomical features that may prove to be more predictive of perioperative complications.

First bite syndrome is a frequent complication following surgery in the parapharyngeal space [22]. It is characterized by pain in the parotid region following the initiation of mastication and may be the result of loss of sympathetic innervation to the parotid and a denervation supersensitivity of salivary gland myoepithelial cells. First Bite Syndrome was observed in 23.8% of all patients; the multidisciplinary group comprising 71.4% of these patients. Multiple authors have speculated a relationship between first bite syndrome and lesion extent as well as resection grade [23]. Both of these results are similar to those reported in other series [6,24].

A strategy incorporating preoperative embolization as an adjunct to carotid body tumor resection has demonstrated inconsistent results in the literature [25]. Theories as to the discrepant results include difficulty in demonstrating compatibility amongst series regarding tumor grade and extent. Indeed, even in the subgroup analysis of high grade Shamblin tumours, there was no significant difference in blood loss with or without pre-operative embolization.

A multidisciplinary approach has become the standard of care for the treatment of various conditions [26-28]. As surgical teams continue to address increasingly complex pathologies, having the 'second opinion' of a colleague is of major benefit for decision making and problem solving. These opinions may involve defining the objectives of the operating, clarify anatomy, the overall surgical technique, and the conduct of the operation. Each discipline instils a distinct set of knowledge and techniques, allowing for the cross-fertilization of different ideas and innovations. Each discipline can focus on different aspect of the case, offloading the primary surgeon team. Outside of the operating room, each team enables access to different resources, allowing for more comprehensive pre and postoperative care. For these reasons, despite our findings, we do stand by our multidisciplinary approach to CBT management.

There are limits to a working within a multidisciplinary approach. Perhaps most challenging is to find a group of individuals willing to participate in a team approach, and therefore sacrifice a degree of autonomy and control. These individuals need to be available at the same time, be comfortable with complexity, and oftentimes flexible with reimbursement. There should be consensus that the management of these complex cases should, to some degree, be shared from beginning to end. The institution must also have the financial, diagnostic, and clinical resources to support the needs of the team. Dysfunction can arise when there are perceived disparities in power, different treatment principles, poor attitude, or difficulties with scheduling [28].

This study is limited by its small size and retrospective nature. However, prospective and / or randomized studies for a rare condition such as CBT are not feasible.

CONCLUSION

Carotid body tumours are difficult to manage surgically, and complex tumor resections may be benefited by the cumulative expertise of multidisciplinary teams. There is a continuing high rate of cranial nerve injury. We were unable to find any benefit with respect to OR time, blood loss, length of hospital stay, or complication rate with a multidisciplinary approach. However, the tumours resected by a multidisciplinary team were larger, of higher grade, and required more complex reconstruction. Interestingly, tumour grade was not associated with increased blood loss, nerve injury, or length of hospital stay. We did not find a benefit of preoperative embolization in reducing blood loss.

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