

ORIGINAL ARTICLE

Surgical treatment of carotid body tumors: a single-center retrospective study

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Summary

Introduction: This study presented our experience in the surgical treatment of carotid body tumors (CBT).

Material and Methods: We retrospectively analyzed patients who underwent carotid surgery between 2009 and 2021. CBT patients were identified, and a control group of patients without CBT was selected using systematic sampling. Demographic and clinical characteristics were compared. Statistical analysis included Student's t-test, Pearson's chi-squared test, and Fisher's exact test.

Results: Among 6728 patients operated for carotid pathology, 16 (0.24%) had CBT. Four CBT patients presented with compressive symptoms, while 12 had medium-sized tumors classified as Shamblin I or II. Subadventitial excision was performed in 12 cases; in four, carotid artery resection and reconstruction were necessary. CBT patients were significantly younger than controls (55.8 ± 17.9 vs. 69.2 ± 7.5 years, $p = 0.009$). The CBT group included fewer males (25% vs. 68.2%, $p = 0.001$) and fewer smokers (18.7% vs. 50%, $p = 0.014$). ASA physical status scores were significantly lower in the CBT group ($p = 0.001$), and hypertension was less prevalent (75% vs. 92.9%, $p = 0.03$).

Conclusion: Subadventitial resection of CBT with deliberate resection of the external carotid artery is a simple and efficient procedure. A resection of the internal carotid artery during CBT surgery requires immediate repair or replacement. Our findings indicate significant differences in demographic and clinical characteristics between patients with CBT and those undergoing carotid surgery for atherosclerotic disease, emphasizing the importance of careful assessment and individualized management of this specific patient group.

Keywords: Cervical paraganglioma, Carotid Body Tumor, Surgical Treatment.

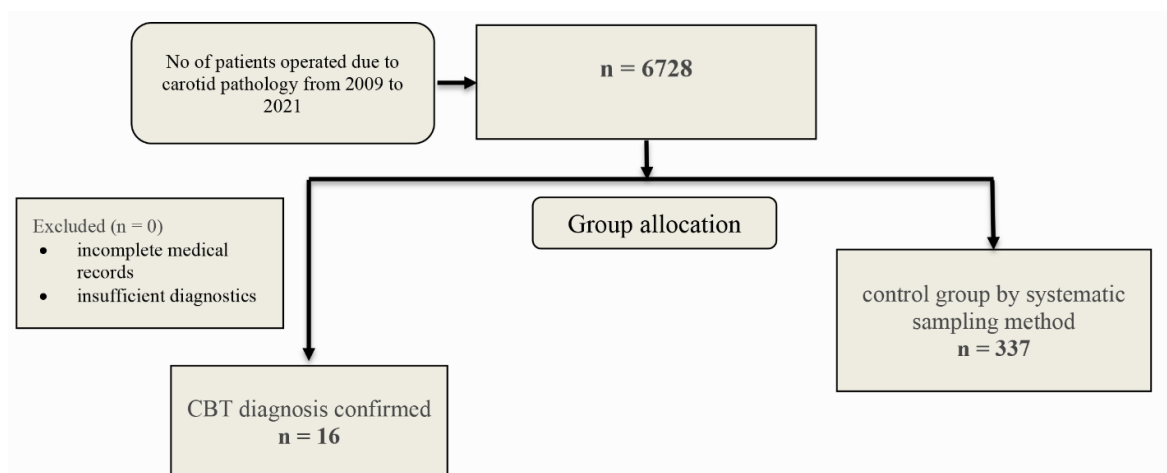


Figure 1. Flow Diagram of the Present Study.

INTRODUCTION

The carotid body is a type of chemoreceptor located at the medial portion of the carotid bifurcation, detecting changes in the level of oxygen, carbon dioxide, and pH in the blood (1). Carotid body tumors (CBT) are the most common paragangliomas of the neck region. Jugular bulb and tympanic paragangliomas are much rarer (2-4). These tumors are mostly benign, and surgical excision is the treatment of choice in the majority of cases. The main aim of this retrospective study was to analyze our experience of CBT's surgical treatment, i.e., to analyze the surgical outcomes of CBTs according to the Shamblin classification and to compare clinical and demographic characteristics of CBT patients against patients with other carotid pathologies.

MATERIAL AND METHODS

Consecutive patients who underwent surgery due to carotid pathology at the Clinic for Vascular and Endovascular

Surgery of the University Clinic Center of Serbia between 2009 and 2021, were included in the study. Among them, patients with a pathohistologically confirmed diagnosis of CBT were identified. Conversely, the exclusion criteria implemented to maintain the integrity and robustness of the study's findings were: incomplete medical records or insufficient diagnostics (Figure 1). Basic demographic and clinical data, along with clinically relevant characteristics of the disease, as well as data regarding the postoperative course, were obtained from patients' medical records. Additional postoperative data were gathered at regular postoperative check-ups. The decision for surgical treatment was established according to findings of color duplex scan (CDS) and multidetector computed tomography (MDCT) angiography examination (Figure 2). All of those patients were operated on under general endotracheal anesthesia through longitudinal cervicotomy. Shamblin classification of tumors was established based on intraoperative findings (Figure 3). Diagnosis of CBT was confirmed by pathohistological analysis of tumor samples, and the Pheochromocytoma of the Adrenal Gland Scaled Score (PASS) was calculated to predict the

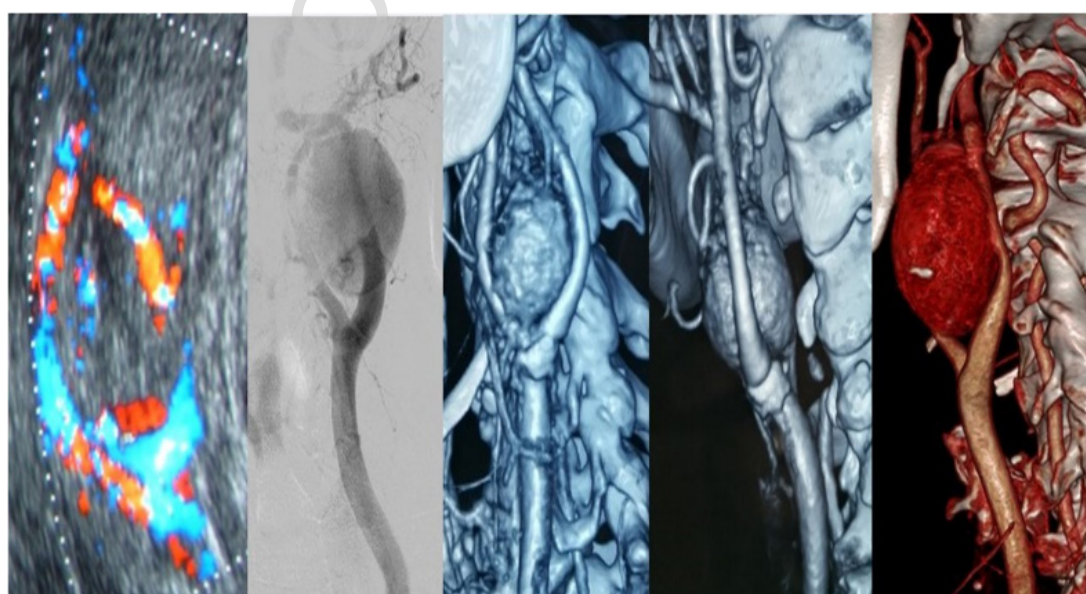


Figure 2. Diagnostic tools for carotid body tumor.

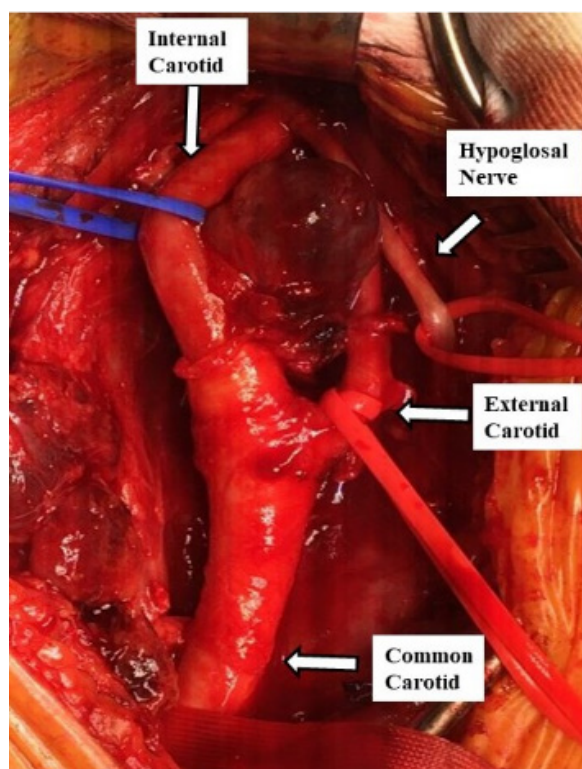


Figure 3. Intraoperative findings of a carotid body tumor.

potential malignant behavior of the tumor. According to the protocol of our Institution, patients were followed up by ultrasound (at one and six months following surgery, and annually thereafter) for five years. A follow-up of five years was completed in 9 patients, and the remaining were followed from 6 months to 2 years. Being that this is a retrospective study involving anonymized patient data, the requirement for ethical approval was waived by the institutional policy of the Clinic for Vascular and Endovascular Surgery, University Clinical Center of Serbia, and the principles outlined in the Declaration of Helsinki.

To compare demographic and clinical characteristics, a control group of patients without CBT who underwent carotid surgery was selected using systematic sampling. Starting from a randomly selected patient among the first 20, every 20th patient was included based on the chronological order of surgery, provided that complete data of interest were available. Relevant clinical and demographic variables were extracted from medical records and compared between the two groups.

Statistical analysis

Descriptive statistics methods were employed to analyze and interpret the collected data. Continuous variables are reported as means $\pm$ standard deviation (SD). Categorical variables are presented as absolute counts (n) and percentages (%). Continuous variables were compared between the groups using Student's t-test. In contrast, Pearson's chi-squared test or Fisher's exact test was used to compare the differences in the frequency of categorical variables. The level of significance was set at <0.05 ,

and statistical analyses were performed using SPSS 22.0 (IBM Corp., Armonk, NY, USA).

Being that this is a retrospective study involving anonymized patient data, the requirement for ethical approval was waived by the institutional policy of the Clinic for Vascular and Endovascular Surgery, University Clinical Center of Serbia, and the principles outlined in the Declaration of Helsinki. Shortly, this study has been conducted in full accordance with national and international ethical guidelines and standards relevant for this type of study. All statistical analyses were performed on anonymized patient data, ensuring that individual identities could not be disclosed or compromised.

RESULTS

A total of 6728 patients were operated on due to carotid pathology from 2009 to 2021. Among them, 16 patients (0.24%) with CBT who underwent surgical treatment during these 12 years were identified. Basic characteristics of CBT patients are presented in Table 1, while Table 2 summarizes Shamblin classification, surgery characteristics, and PASS score. Male/female ratio was 1:3 (N male=4, N female=12). The mean age of CBT patients was 55.8 ± 17.9 years (range 23–82 years). In ten patients (63.5%), the tumor was right-sided, and no patients presented with bilateral tumors. In twelve patients (75%), the tumor presented as an asymptomatic neck mass.

In comparison, in four patients, the tumor caused symptoms of compression on surrounding structures (2 patients with dysphagia, one with odynophagia, and one with stridor). In twelve patients, the tumor was medium-sized and classified as Shamblin I and II. Others infiltrated the carotid arteries and were classified as Shamblin III. Subadventitial tumor excision was performed in 12 patients with Shamblin I and II, while in four patients with Shamblin III, carotid arteries were resected and then reconstructed. In three of them, reconstruction was performed with a prosthetic Dacron graft, and in one with significant internal carotid artery elongation, end-to-end anastomosis was performed. In five patients (31%), enlarged local lymph nodes were found. They were subjected to pathohistological analysis, which showed hyperplastic enlargement. None of our patients had systemic metastasis according to the findings of preoperative MDCT.

After surgical treatment, transient damage to the hypoglossal nerve was found in two patients. There were no postoperative complications such as hematoma, stroke, or Horner syndrome. Only one patient had a pathological PASS score higher than 3, which was established as borderline for malignancy. That patient had a PASS score of 8. Also, there was no in-hospital mortality. Postoperative follow-up and control color Doppler sonography were performed in all patients. Control angiography was performed in 7 patients (43.8%). We did not find any tumor recurrence.

Table 1. Basic characteristics of patients with carotid body tumor, symptoms, and diagnosis

Patients' characteristics	Number (Percentage)
Age - mean (years)	55.8 ± 17.9
Sex - male	4 (25%)
Preoperative symptomatology - yes	4 (25%)
Tumor side - left	10 (63.5%)
Diagnosis	
Angiography	7 (43.7%)
Duplex + angiography	9 (56.3%)

Table 2. Shamblin classification, surgery characteristics, and Pheochromocytoma of the Adrenal Gland Scaled Score

	Number (Percentage)
Shamblin classification	
I	3 (18.7%)
II	9 (56.3%)
III	4 (25%)
Tumor-related procedure - excision	16 (100%)
Vascular procedure	
No	12 (75%)
Resection + synthetic graft	3 (18.7%)
Resection + end-to-end anastomosis	1 (6.3%)
Complications	
No	14 (87.5%)
Hoarseness	2 (12.5%)
PASS score	
III	15 (93.7%)
VIII	1 (6.3%)

PASS score -Pheochromocytoma of the Adrenal Gland Scaled Score

Using a systematic sampling method, a control group of 337 patients who underwent carotid surgery without CBT was identified during these 12 years. Patients in the CBT group were significantly younger compared to the control group (the mean age 55.8 ± 17.9 years vs. 69.2 ± 7.5 , $p = 0.009$). Furthermore, the difference in sex distribution was also statistically significant: there were four male patients in the CBT group (25%), compared to 230 males (68.2%) in the control group ($p = 0.001$). When the two groups were compared according to body mass index, no significant differences were noted: 26.3 ± 3.7 kg/m² (control group) vs. 27.5 ± 4.2 kg/m² (CBT group), $p = 0.42$. Similarly, no significant differences were observed when the groups were compared in terms of symptomatology. In the CBT group, four patients (25%) presented with symptoms, while 131 (38.8%) were symptomatic in the control group ($p = 0.261$). Significantly fewer patients were active smokers in CBT than in the control group (18.7% vs. 50%, $p = 0.014$). The American Society of Anesthesiologists (ASA) physical status score was also significantly higher in the control group compared to CBT patients: ASA 3 score was noted in 90.5% in the control group vs. 12.5% in the CBT group ($p = 0.001$). Significantly more patients in the control group had hypertension, compared to the CBT group (92.9% vs. 75%, $p = 0.03$). Still, no significant differences were noted when

those two groups were compared in terms of diabetes mellitus (36.5% vs. 31.3%, $p = 0.794$).

DISCUSSION

Carotid body tumors (CBT) are extremely rare and highly vascularized (5). The arterial supply typically derives from the ascending pharyngeal and occipital branches of the external carotid artery. However, contributions from the internal carotid artery, vertebral artery, and thyrocervical trunk can develop (6). Due to this, surgical treatment of the CBT is associated with significant bleeding, which at the same time, makes it more difficult or even disables tumor removal, increasing the risk of iatrogenic injuries to surrounding structures (7). According to the same reports, preoperative embolization reduces intraoperative blood loss and decreases technical difficulties during surgical removal of carotid body tumors (8-12). To be more precise, a selective preoperative embolization includes the major feeding arteries that originate from the external carotid artery. It requires a very precise technique to prevent potential central neurologic complications (13). The role of preoperative embolization before surgical treatment of CBT is controversial (14-17). We do not use it before CBT surgery.

The incidence of bilateral CBT is 5% (18-20). The tumor is three times frequent in female patients, and patients are mostly in their 5th decade (21). Five to 9% of patients with CBT have a positive family history. An equal distribution between men and women in patients with familial CBT supports an autosomal mode of genetic transmission (22). We did not have patients with bilateral and familial CBT, while the mean age of treated patients was 55.8 years.

The majority of CBT grows slowly and has benign characteristics (23-30). The malignant potential of these tumors is controversial (31, 32). The reported rates of malignancy range from 2% to 50% (23-30). The metastatic rate of CBT is approximately 5% (23-30). The metastatic spread generally occurs in regional lymph nodes. However, systemic metastases that involve the kidney, thyroid, pancreas, cerebellum, lungs, bone, brachial plexus, abdomen, and breast are also described (23-30). In our study, there were no CBTs with malignant characteristics.

An asymptomatic, rubbery, firm, and non-compressible neck mass located below the angle of the mandible is the most common clinical presentation of the CBT (33-37). The mass is often pulsatile due to its close relationship with the carotid artery. Other nonspecific symptoms may include neck or ear pain, as well as compression abnormalities caused by vagal, hypoglossal, cervical sympathetic, and esophageal involvement (20, 33-37). Approximately 5% of CBT may have neurosecretory activity. Due to this, some patients describe symptoms of hypertension, tachycardia, arrhythmias, headache, and so on (20, 37-

40). CBT's intracranial extension can cause lateralizing and central neurologic symptoms (38). Death due to asphyxia in patients with CTB is described as well (39, 40). The majority of our patients were asymptomatic, but four patients presented with symptoms of surrounding tissue compression. For instance, CBT patients may experience vocal cord paralysis, facial palsy, and ischemic stroke (41).

In all patients with neck mass located below the angle of the mandible, the initial diagnosis should begin with duplex ultrasonography. If CBT is suspected, the diagnosis continues with MDCT angiography (10, 19, 20, 37, 42). Characteristically, a carotid body tumor separates the internal and external carotid arteries, widening the bifurcation (10, 19, 20, 37, 42). In addition, diagnostic procedures should provide information about the concomitant carotid occlusive disease. Since MDCT angiography has been introduced, standard angiography has lost its importance for the diagnosis of CBT. In all our patients, the final diagnosis has been established using MDCT angiography.

Some authors have reported palliation using radiation therapy alone in patients with CBT. However, radiation therapy can be followed by significant local complications, including osteonecrosis of the mandible, carotid radiation arteritis, and laryngeal nerve injury (25, 30, 33).

Most agree that surgery is the method of choice for the treatment of CBT (43). Reigner performed the first resection of the CBT in 1880, but his patient did not survive (44). Six years later, Maydl removed a carotid body tumor, but the patient became aphasic and hemiplegic (45). Finally, in 1903, Scudder performed the first successful removal of a CBT with preservation of the carotid arteries as well as surrounding cranial and cervical nerves (46).

General anesthesia is preferable during CBT surgery, and we applied this anesthesia technique in all our patients. Patient positioning is identical to that for carotid endarterectomy (18, 47, 48). A procedure begins with a longitudinal skin incision along the anterior border of the sternocleidomastoid muscle (47, 48). In the case of large CBT, a modified-T neck incision is recommended (18). Surgical treatment of CBT can be complicated because of its intimate relationship with the carotid arteries and cranial and cervical nerves. However, not all tumors are equally challenging for surgical treatment.

Regarding that, Shamblin classified CBTs into three groups (18, 25). *Group I* includes relatively small tumors, which are minimally attached to the carotid vessels. Due to this, surgical excision is not difficult (18, 25, 47, 48). *Group II* includes larger tumors with moderate attachments to the carotid arteries. Finally, *group III* includes massive tumors that encase the carotid arteries (18, 25, 47, 48). The first step in surgical treatment of CBT involves proximal and distal bleeding control, which requires isolating the standard, external, and internal carotid arteries with surgical tapes (18, 19, 37). It is recommended to isolate the external and internal carotid arteries above

the upper pole of the tumor. The next step is identifying the hypoglossal and vagal nerves that must be protected during tumor dissection.

In cases with tumors Shamblin types I and II, a dissection proceeds in a subadventitial plane along the carotid arteries (10, 18, 37, 42, 49). Gordon Taylor introduced subadventitial dissection during the removal of the CBT in 1940 (50). The injury of cranial and cervical nerves may be avoided by meticulous dissection around the tumor and by the use of bipolar electrocautery to minimize the conductive heat effect. Temporary clamping of the external carotid artery in patients with Shamblin II reduces bleeding during tumor dissection (10, 18, 37, 42). The Shamblin III of CBT requires resection of the external and/or internal carotid artery (18, 19, 37, 48, 51). In such cases, earlier ligation and division of the external carotid artery reduce bleeding and facilitate further resection of the tumor. Reconstruction of the external carotid artery following tumor removal is not necessary. However, ligation of the internal carotid artery results in a stroke incidence ranging from 23% to 50% and a mortality rate of 14% to 64% (52). Due to this, an excision of the internal carotid artery that is involved with the tumor requires its immediate repair/replacement with end-to-end anastomosis or vascular graft (18, 19, 37, 48, 51). Three of our patients required graft replacement of the carotid artery after tumor excision. Still, in one patient with significant internal carotid artery elongation, reconstruction was done with end-to-end anastomosis. If prolonged clamping and reconstruction of the internal carotid artery is necessary during CBT surgery, the same authors recommend the use of the carotid shunt (19). However, ipsilateral hemispheric hypoperfusion after internal carotid artery clamping during CBT surgery is an infrequent cause of neurological events. Due to this, we do not use carotid shunts during CBT surgery.

Advancements in subadventitial dissection and vascular surgery techniques have significantly decreased the perioperative stroke occurrence after CBT surgery from 30% to below 1%. At the same time, perioperative mortality, which was 50% five decades ago, has decreased to 0% in nowadays studies (2, 18, 19, 30, 37, 42, 51). Unlike central neurologic complications, the incidence of local neurologic complications (iatrogenic injuries of surrounding cranial and cervical nerves) after CBT surgery remained relatively significant (53, 54). In the most recently published papers, the incidence of these injuries is nearly 10% (2, 5, 19, 55-57). Transient hypoglossal nerve injury occurred in one of our patients (16.67%). Our previous reports included 17 cases of CBT treated between 1982 and 2007 (47, 48). Perioperative stroke rate and cranial nerve injuries were 6% without perioperative mortality.

Finally, the present study highlights several important clinical characteristics of patients with CBT that warrant attention. Our results show that CBT patients exhibit distinct profiles compared to individuals undergoing carotid

artery surgery for atherosclerotic disease. Notably, CBT patients are significantly younger and more frequently female. They also have a lower prevalence of smoking and hypertension, and generally present with a better ASA physical status score. These differences likely reflect the non-atherosclerotic, often benign nature of CBT, which is typically of neuroendocrine origin and may be influenced by genetic or familial factors rather than conventional cardiovascular risk factors (58).

In contrast, patients undergoing carotid revascularization for atherosclerotic disease tend to be older and have a higher burden of comorbidities, such as smoking, hypertension, and cardiovascular disease (59). Interestingly, no significant differences were observed between the groups in terms of BMI, presence of symptoms, or the prevalence of diabetes mellitus. This suggests that, while some clinical and metabolic features may overlap, CBT patients constitute a fundamentally different population. Recognizing these distinctions is essential not only for accurate surgical risk stratification but also for optimizing preoperative evaluation and tailoring long-term follow-up strategies.

This study has several limitations that should be taken into consideration. Most notably, the retrospective design of the present study limits the level of control over data collection, as the analysis relies solely on pre-existing medical records. The relatively small number of patients (due to the low incidence of CBT) restricts the generalizability to broader populations. Therefore, the results should be interpreted with caution. Nevertheless, the study provides critical observational data on the surgical management of rare carotid body tumors and has observational and hypothesis-generating potential for future studies.

CONCLUSIONS

CBTs are rare and mostly benign. Surgery is the method of choice for the treatment of these tumors. Subadventitial resection of CBT with deliberate resection of the external carotid artery is an efficient procedure for the

surgical management of this tumor. A resection of the internal carotid artery during CBT surgery requires immediate repair or replacement. Our findings indicate a marked difference in demographic and clinical characteristics between patients with CBT and those undergoing carotid surgery for atherosclerotic disease, highlighting the need for careful evaluation and individualized management of this unique patient population.

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Ethical approval: Being that this is a retrospective study involving anonymized patient data, the requirement for ethical approval was waived by the institutional policy of the Clinic for Vascular and Endovascular Surgery, University Clinical Center of Serbia, and the principles outlined in the Declaration of Helsinki. Shortly, this study has been conducted in full accordance with national and international ethical guidelines and standards relevant for this type of study. All statistical analyses were performed on anonymized patient data, ensuring that individual identities could not be disclosed or compromised.

Informed consent statement: Informed consent was obtained from all subjects with carotid body tumors involved in the study.

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HIRURŠKO LEČENJE TUMORA KAROTIDNOG TELAŠCA: RETROSPEKTIVNA STUDIJA U OKVIRU JEDNOG CENTRA

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Sažetak

Uvod: Ova retrospektivna studija prikazuje iskustva tercijarnog centra u hirurškom lečenju bolesnika sa tumorom karotidnog telašca (glomus tumorom).

Metode: Analizirani su pacijenti koji su podvrgnuti karotidnoj hirurgiji u periodu 2009–2021. Identifikovani su pacijenti sa glomus tumorom, a kontrolna grupa bez tumora selektovana je sistematskim uzorkovanjem. Upoređene su demografske i kliničke karakteristike među grupama. Za statističku analizu korišćeni su Studentov t-test, Pirsonov hi-kvadrat test i Fišerov test tačne verovatnoće.

Rezultati: Među 6728 operisanih, kod 16 (0,24%) dijagnostikovano je glomus tumor. Kod četiri bolesnika, tumor je izazivao kompresivne simptome, dok je kod 12 klasifikovan kao *Shamblin* I/II. Kod 12 bolesnika urađena je subadventicijska ekscizija tumora, dok je kod četiri bila neophodna resekcija i rekonstrukcija karotidnih

arterija. Pacijenti sa glomus tumorom bili su značajno mlađi od kontrolne grupe ($55,8 \pm 17,9$ godina naspram $69,2 \pm 7,5$ godina, $p = 0,009$), sa manje muškaraca (25% naspram 68,2%, $p = 0,001$) i pušača (18,7% naspram 50%, $p = 0,014$). ASA fizikalni status bio je značajno bolji kod pacijenata sa glomus tumorom ($p = 0,001$), a hipertenzija je bila ređa (75% naspram 92,9%, $p = 0,03$).

Zaključak: Subadventicijska resekcija glomus tumora sa resekcijom spoljašnje karotidne arterije predstavlja efektivni vid hirurškog lečenja ovih bolesnika. Resekcija unutrašnje karotidne arterije tokom ovih operacija zahteva istovremenu rekonstrukciju. Naši rezultati ukazuju na značajne demografske i kliničke razlike između pacijenata sa glomus tumorom i onih koji se podvrgavaju karotidnoj hirurgiji zbog aterosklerotske bolesti, naglašavajući značaj pažljive procene i individualizovanog pristupa u tretmanu ove specifične grupe pacijenata.

Ključne reči: Paragangliom, tumor karotidnog telašca; glomus tumor; hirurško lečenje

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