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Carotid body tumors: what have we learned in 5 years?

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Abstract

Objective: Objectives: The present study aimed to retrospectively evaluate patients who were operated for carotid body tumors (CBT) in our clinic.

Methods: The present study involved 19 patients who underwent surgical CBT removal in the Otorhinolaryngology- Head and Neck Surgery Department of Karadeniz Technical University Medical Faculty between 2013 and 2018. Sociodemographic characteristics, clinical presentation and findings of physical examination, Shamblin classifications based on preoperative radiological images, surgical procedure, histopathological diagnosis and complications were recorded from hospital records.

Results: Seven of the patients were male and 12 were female. The mean age of the patients was 48.8 years and the most frequent symptom was a painless mass in the neck. According to the Shamblin classification, seven of our patients had Shamblin type I, nine patients had Shamblin type II and three patients had Shamblin type III tumors. Twelve patients had preoperative embolization of the tumor and seven patients were operated without embolization. Three patients had intraoperative carotid artery injury-related complications, whereas one patient had weakness in tongue movements due to hypoglossal nerve paresis.

Conclusion: The main treatment for CBT is surgery, however, preoperative embolization is still controversial. Successful management of patients with CBT can be achieved via a multidisciplinary approach with the participation of head and neck surgery, cardiovascular surgery and radiology departments.

Keywords: Surgery, carotid body tumor, paraganglioma.

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Introduction

Carotid body tumor (CBT) is a rare neuroendocrine neoplasm originating from extra-adrenal chromaffin cells with a reported incidence of 1 in 30,000.[1] CBT accounts for about 60% of the paragangliomas located in the head-neck region and is sporadic in almost 90% of cases.[2] Hereditary cases are inherited in an autosomal-dominant fashion with incomplete penetrance.[3] CBT is also called glomus caroticum, chemodectoma and carotid body paraganglioma. CBTs occur at the carotid bifurcation within the adventia and usually present as an asymptomatic, slow-growing and highly vascular mass in the lateral neck.[4] The carotid body is composed of chemoreceptor cells that monitor arterial blood oxygen and carbon dioxide levels and are stimulated by hypoxia, hypercapnia and acidosis. Therefore, CBT is more frequent in chronic hypoxic conditions including chronic obstructive pulmonary disease and living at high altitudes.[5,6] Head and neck paragangliomas can secrete catecholamines in about 3 to 4% of cases.[7]

The blood supply of the CBT is provided by the glomic artery mainly from the external carotid artery, carotid bifurcation and the internal carotid artery.[8] Early surgical removal of the tumor is recommended due to the tendency for invasion or compression of adjacent vascular and neural tissues in enlarged CBTs and the malignant potential of the tumor presenting in about 10% of cases.[4] The Shamblin classification system describes the anatomical relation of the carotid arteries with the tumor and is suggested to be a predictor of intra-operative technical difficulty.[9] Preoperative embolization is usually performed due to the high vascularity of CBTs although its benefit remains controversial.[2] In the present study, we aimed to retrospectively evaluate patients who had surgical removal of the CBT in our clinic.

Materials and Methods

Approval for the study protocol was obtained from the Ethics Committee of the Karadeniz Technical University (2020-133) and written informed consent was obtained from the participants. The present study involved 19 patients who underwent surgical CBT removal in the Otorhinolaryngology- Head and Neck Surgery Department of Karadeniz Technical University Medical Faculty between 2013 and 2018. Sociodemographic characteristics, clinical presentation, findings of physical examination, Shamblin classification of the tumor, surgical procedure, histopathological diagnosis and complications were recorded from the hospital records. Statistical analysis was performed using Statistical Package for the Social Sciences (v. 15; SPSS Inc.; Chicago; IL; USA).

Results

There were 19 patients with definitive data. Seven of the cases were male (37%) and 12 were female (63%). The mean age of the patients was 49.6±14.7 years (29-76 years). None of the patients had a family history or association with higher altitudes. All patients had a painless and slowly growing neck mass just anterior to the sternocleidomastoid muscle at the level of the hyoid bone without any findings of increased catecholamine secretion. 13 patients had a CBT on the right side of the neck while six had a CBT on the left side. One patient had a history of surgical CBT removal located at the contralateral side of the present tumor in a different clinic eight years ago. All patients underwent radiological examination including ultrasonography, computed tomography and magnetic resonance imaging with angiography. According to the Shamblin classification, which is based on the involvement of the carotid arteries, seven patients had Shamblin type I, nine patients had Shamblin type II and III patients had Shamblin type 3 tumors (Figure 1). None of the patients had fine-needle aspiration biopsy due to the high vascularity of the tumor. All patients underwent surgical removal of the CBT by the same surgical team. 12 patients were operated after tumor embolization whereas seven patients were operated without embolization (Figure 2). The surgical procedure was

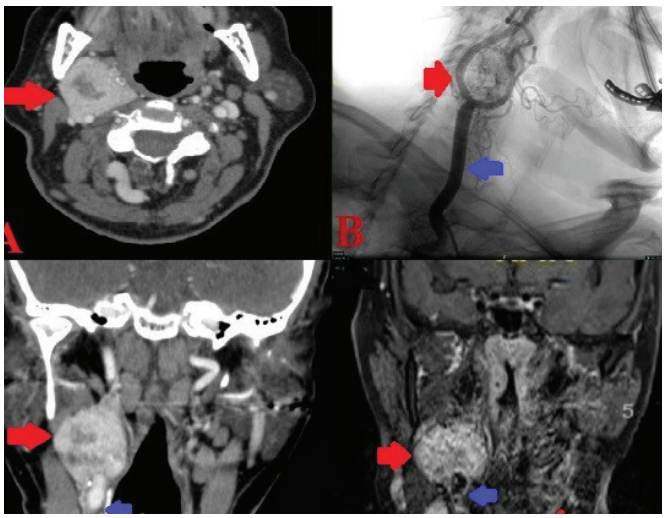

Figure 1. Carotid body tumor at the right carotid bifurcation (red arrow: carotid body tumor; blue arrow: common carotid artery). **A.** Contrast-enhanced axial CT image of the tumor. **B.** Contrast-en-hanced coronal CT of the tumor. **C.** T1-weighted contrast-enhanced MRI of the tumor. **D.** Selective external carotid angiography for tu-mor embolization.

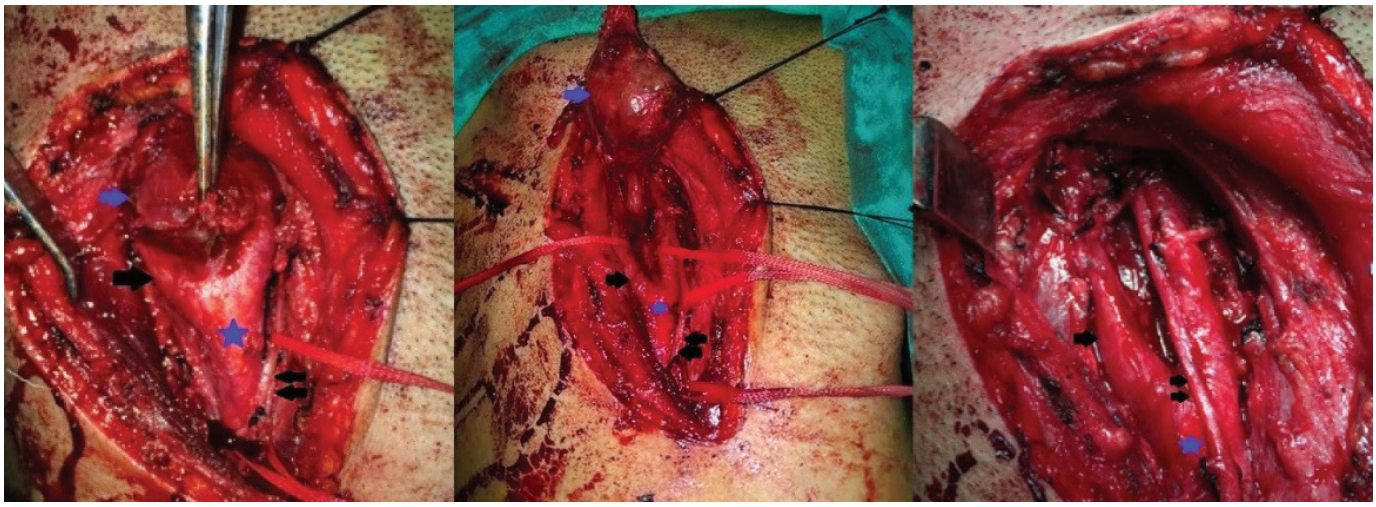

Figure 2. Intra-operative images of right carotid body removal (Blue arrow: carotid body tumor; black arrow: external carotid artery; double black arrow: vagus nerve; blue star: common carotid artery).

initiated with an incision following a skin crease located approximately in the lower aspect of the submandibular region. After exposing the common carotid artery, vagal nerve, hypoglossal nerve, accessory nerve, carotid bifurcation, and external and internal branches of the carotid artery, the CBT was identified (Figure 2). Magnifying loops were used for dissection of the tumor from the carotid artery. In all patients, the internal carotid artery above the tumor was dissected and taped safely before tumor removal, except for one patient who had a malignant infiltration of the tumor to the carotid artery. In this patient, the tumor was excised with the infiltrated part of the carotid artery via an intraluminal shunt and the carotid artery was reconstructed with a reversed saphenous vein graft. Histopathological examination of the tumor revealed a malignant CBT. The patient received postoperative radiotherapy and is disease-free with no sequela in the third year follow-up. Intraoperative carotid injury also occurred in two patients during tumor dissection. The injury site was repaired via primary suturing and one of the patients had permanent hemiplegia. In the postoperative period, one patient had ipsilateral tongue paresis, which was healed after two weeks of steroid regimen. According to the histopathological examinations, 18 patients had a benign CBT whereas one patient had a malignant CBT. The mean follow-up period for the patients was 22 months (range 6-60 months) with no sign of disease.

Discussion

CBTs are usually slow-growing tumors that occur more commonly in women between the ages of 30 and 60.[10] CBTs are usually asymptomatic, but as the tumor grows, dysphagia, odynophagia, dysphonia and compression findings in the 9th and 12th cranial nerves can be seen.[11] Bilateral CBT can occur in 5% of sporadic tumors whereas it can be seen in 33% of familial cases.[12] In the present study, CBT was found to be more common in female patients, in accordance with the literature. The mean age of our series was 48.8 years. 18 patients had a unilateral CBT, while one patient had a history of CBT removal located on the contralateral side of the neck. The most frequent symptom of the patients was a painless mass in the neck and none of the patients had a family history. All patients had benign tumors except for one patient with a malignant CBT.

In 1971, Shamblin et al suggested a surgical classification of CBTs into three groups according to the relationship between the tumor and the carotid artery.[9] Shamblin class I is defined as a small tumor with minimal attachment to the carotid arteries. Shamblin class II refers to larger tumors with moderate attachment to the carotid vessels but the tumor is resectable with the preservation of carotid vessels. In class III, the tumor encases the carotid arteries and the removal of the CBT may necessitate arterial resection with reconstruction. It is well established that a requirement for carotid artery reconstruction is increased with enlargement of the tumor. According to a 30-year experience,

Torrealba et al [13] performed carotid artery reconstructions in 83.3% of Shamblin III tumors. In the study of Sevil et al [14] involving 60 patients who had surgical CBT removal, 29 patients were reported to have vascular reconstruction after tumor resection. In their study, Shamblin class III tumors constituted 62.1% of the patients requiring vascular reconstruction. In the present study, seven patients had Shamblin type I, nine patients had Shamblin type II and three patients had Shamblin type III tumors. One out of three patients with Shamblin class III tumor necessitated carotid artery reconstruction with reversed saphenous vein graft after tumor removal. Histopathological examination of the tumor revealed a malignant CBT.

Although total surgical removal of the tumor is the well-established treatment option for CBT, the benefit of preoperative embolization is highly controversial in the literature as to whether it improves surgical outcome or not. Proponents of preoperative embolization claim that the embolization reduces the blood supply of the tumor and facilitates surgical removal of the tumor.[15] However, the benefit of preoperative tumor embolization has been questioned in recent studies. In the study of Cobb et al [4] including 547 patients with CBT, preoperative embolization was shown to have no benefit in terms of blood loss, stroke, cranial nerve injury and length of hospital stay. Sevil et al [2] reported total surgical removal of CBTs in 67 patients, composed of 11 Shamblin class I (16.4%), 30 Shamblin class II (44.8%) and 26 Shamblin class III (38.8%) tumors without preoperative embolization. The authors reported that total surgical removal of the CBT can be accomplished without requiring preoperative embolization. Preoperative embolization may cause stroke due to the development of emboli in the internal carotid artery. Also, Bercin et al [16] reported that the risk of carotid artery injury increased during dissection of the CBT in patients who underwent preoperative embolization. The excessive risk of carotid rupture was considered to be likely associated with inflammation, which may increase the fragility of the adventitia of vessels in the embolization patients. The authors demonstrated that the congestion was sufficiently obvious in the adventitial vasa vasorum, which may play a pivotal role in the pathophysiology of carotid rupture in embolization patients.[16] In the present study, there were three patients who experienced carotid artery injury during surgical removal of the tumor. In the patient with a malignant CBT, preoperative embolization was performed, but the carotid artery injury occurred due to the tumor infiltration. In contrast, Bercin et al [16] reported two patients with intraoperative carotid injury that had no preoperative tumor embolization. However, the number of patients in the present study was quite low for drawing a conclusion regarding the association between preoperative embolization and carotid artery injury, the amount of blood loss during surgery and perioperative morbidity.

Surgical management of CBTs may cause serious complications, including cranial nerve injury (CNI), stroke and mortality. In a recent systematic review and meta-analysis including 4418 patients with CBT, the rate of CNI, stroke and mortality was reported as 25.4%, 3.53% and 2.29%, respectively.[17] The prevalence of neck hematoma requiring re-exploration was reported to be 5.24% in this meta-analysis. Pacheco-Ojeda [18] reported postoperative complications in 59 of 214 (27.5%) patients who underwent CBT surgery, which were mostly minor and related to cranial nerve dysfunction. In that study, hypoglossal nerve dysfunction was the most frequent postoperative complication with a rate of 7.9%. In the present study, no mortality occurred, however one patient had temporary hypoglossal nerve dysfunction (5.3%) and one had permanent hemiplegia (5.3%). In our opinion, a careful preoperative evaluation of the patients with adequate radiological examination and meticulous surgical dissection would lower the complication rates of surgical CBT removal.

Conclusion

CBTs are rare tumors that can be treated with surgery. Preoperative embolization is still controversial in these patients. Successful management of patients with CBT can be achieved via a multidisciplinary approach with the participation of head and neck surgery, cardiovascular surgery and radiology departments.

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Ethics Committee Approval: The study protocol was approved by the Ethics Committee of the Karadeniz Technical University (2020-133).

Informed Consent: Informed consent was obtained from all individual participants included in the study.

Author Contributions: Designing the study – B.C., O.B.; Collecting the data – B.C., S.A., O.B., M.I.; Analysing the data – B.C., S.A., O.B., A.U.I., M.I.; Writing the manu-

script – B.C., S.A., O.B., A.U.I.,; Confirming the accuracy of the data and the analyses – B.C., S.A., O.B., A.U.I., M.I.

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