Internal carotid artery ligation for excision of a huge Carotid Body Tumor
– Report of a Case –

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Abstract
Paragangliomas (PGLs) are rare neuroendocrine tumors. About 50% of PGLs develop in the head and neck region, of which approximately 50% originate from the carotid body, and classified as Carotid body tumors (CBTs). Proper management and therapeutic options for CBTs have not yet been clearly determined, to date. We hereby report a case of a huge transected CBT treated by ligation of internal carotid artery (ICA), resulting to an unexpected ipsilateral embolic stroke despite an anticoagulation therapy.

Keywords: Carotid body tumors; carotid artery ligation; neck paragangliomas; stroke;

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Introduction

Carotid Body Tumors (CBTs) are non-functional nor-adrenergic tumors located in the head and neck that was derived from cells associated primarily with the parasympathetic autonomic neuroendocrine system. After the first definition of the tumor by Linder in 1953, Shamblin et al. re-established the classification of CBTs in accordance with the size of the tumor and its relationship with carotid artery bifurcation. In this regard, CBTs should not be considered as a usual clinical scenario for vascular surgeons. Most of the reported cases were from single institutions. Here, in this report, we present a rare case of an extremely large CBT that was transected and extracted. In this case, complex anatomical vasculature of the tumor mandated a ligation of an internal carotid artery.

Case Presentation

A 41-years-old male patient was referred to our department with a huge lump in his left neck free from any neurological or other pressing symptoms. The mass was manifested with a slowly-growing, slightly movable swelling. His past medical history revealed no medications or hospitalization, nor his family medical history was negative for any malignancy. Straight laryngoscopy examination demonstrated unaffected movement of the vocal cords. The neurological examination was free from any abnormal finding. On imaging, the Computer Tomography (CT) with contrast enhancement showed a Shamblin’s 3 CBT, splaying the carotid bifurcation, extending from the skull base to the inferior mediastinum with maximum transverse diameter 43.60 mm and sagittal diameter 57.44 mm, encircling the carotid vessels (Fig. 1a-d). Both vertebral arteries and Willis arterial circle was patent (Fig. 1e, f). SDH mutations or other selective test was not carried out, given the lack of prior family history and absence of general symptoms.

The operation was performed under general anesthesia. A left side longitudinal skin incision for carotid endarterectomy was implemented to expose both carotid arteries and the tumor. The tumor was attached to the bifurcation of left carotid artery, outspreaded until the skull base, and a lymph node was stuck right in front of the mass (Fig. 2a). At first, that lymph node was removed (Fig. 2b). The detachment of the mass from the vessel wall was difficult to carry out, resulting in subtraction of the tumor with a large piece of the carotid bifurcation (Fig. 2c). The remaining stump of the internal carotid artery extended enough into the skull base, while the quality of the vessel integrity was poor. Since reconstruction of the carotid vessels seemed to be totally impossible, ligation of the internal carotid artery was the only option.
Although vagus and hypoglossal nerves were protected, a sympathetic nerve segment was resected. Total operating time was 245 minutes and blood loss was 380 mL.

After the operation, the patient developed hoarseness of his voice, and laryngoscopy revealed a swelling of the left side of the vocal cord. Also, a transient Horner’s syndrome few hours after the operation was observed. Histopathologic examination confirmed the preoperative diagnosis of extra adrenal paraganglioma.

At the time of discharge from the hospital, the patient took instructions for treatment with acenocoumarin with an INR target of > 2.5, and 100 mg of acetylsalicylic acid (ASA) was administered daily for protection of post-procedural events.

At the 10th post-operative day, the patient experienced a transient ischemic attack with drop mouth, weakness at right upper and lower extremities. Three days thereafter the patient returned to the hospital with neurological signs indicating embolic infarction of the left middle cerebral artery.

By the advent of continued anticoagulation treatment in combination with optimal supportive treatment, he restored his pre-operative normal neurological status three months after the operation.
Discussion

Head and neck paragangliomas are very rare tumors with an incidence of 1 in 30,000 to 1 in 100,000 people. CBT is the most common form, accounting from 50 to 80% of these tumors. The first successful operation for CBT was conducted by Albert in 1889, while Kohn first used the term paraganglioma in 1903.

Surgical excision is indicated for most of the CBTs. Since surgery is not free from various risks the best results would be achieved by a specialist team. Shamblin’s classification is widely accepted as a useful tool for the evaluation of surgical risks. It is taken for granted that the larger the volume, the greater the risk of postoperative neurological and vascular complications following tumor resection. Since extension of the tumor might alter the vascular structure, arterial reconstruction with an interposition graft or patch angioplasty may be required. After vascular repair, cranial nerve injury was noted in 63% of cases while its incidence was observed in 27% with carotid body resection alone.

Post-operative strokes varied from 0 to 8% in international studies. If the maximum transverse diameter of the tumor is more than 5 cm, the mortality rate is estimated 1%-3%. In diagnosis, the most useful imaging techniques to identify CBPs and planning surgery remain CTA and MRI. Angiography can be used in cases where the diagnosis is unclear and selective embolization of feeding arteries exists as a therapeutic option. Needle biopsy to confirm diagnosis should be avoided because high vascularity of the lesion can cause disastrous complications.

It was the first case in our single center experience that we proceeded into ligation of the internal carotid artery during CBT’s excision. Our decision relied on the fact that the tumor was infiltrating the internal carotid artery and had disrupted its anatomical structure. Moreover, the tumor extended distally to the skull base making any effort for vascular reconstruction technically impossible. Pre-operatively, we had noted the sufficient Willis collateral. Nevertheless, the patient developed an embolic stroke sign in the 10th post-operative day despite preventive anticoagulant therapy.

This paper adds our experience with a Shamblin’s 3 CBT in which tumor extraction alone or resection with artery reconstruction was not possible. Additionally, the source of the embolus despite anticoagulation therapy and its relevance with the internal carotid artery stump could not be fully explained.

Conclusion

Although CBT is a rare pathological entity, it should always be considered at the time of differential diagnosis with palpable cervical mass. Surgical removal of these tumors remains challenging for every surgeon despite the progress in surgical techniques.

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Conflicts of interest

The Authors declare that there is no conflict of interest regarding the publication of this paper.
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