Submandibular approach and use of Ligasure® system in a complex carotid paraganglioma

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Abstract
Carotid paragangliomas are rare tumors of slow growth, progressive, and highly vascularized; surgery is the chosen treatment, and most surgeons perform an approach with a longitudinal cervicotomy.

Objective: We present a case of carotid paraganglioma Shamblin III with skull base extension.

Methods: Submandibular approach and vascular sealing device of the Ligasure® system were used in the surgery.

Results: We achieved complete resection using these adjunctive techniques.

Conclusions: The use of these adjunctive techniques can be useful to manage large carotid body tumors.

Keywords
Carotid body tumor, glomus tumor, surgery

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Introduction
Carotid body tumors (CBTs) are rare neoplasms characterized by slow growth and abundant vascularization.¹,² Surgical resection is the treatment of choice.²,³ The type of tumor based on Shamblin classification partially predicts intraoperative difficulties and morbidity.⁴ Most surgeons perform a longitudinal cervical incision for tumor resection. Submandibular approach is rarely used.³,⁵ Vascular sealing devices of the Ligasure® system are not commonly used in carotid surgery.⁶ We present the use of the Ligasure® system and submandibular approach in a difficult case.

Case
A 53-year-old man with an asymptomatic pulsatile right cervical tumor was referred to our center. His antecedents were paranoid schizophrenia and obesity. Doppler ultrasound showed a highly vascularized tumor that splays the right carotid bifurcation. The distal common carotid artery and both internal and external carotid arteries were encircled by the tumor without causing any hemodynamic flow alteration. Computed tomography angiography (CTA) of supra-aortic trunks confirmed the diagnosis of a carotid paraganglioma Shamblin III with skull base extension (Figure 1). The preoperative study also included urine metanephrines that were negative. Preoperative embolization was performed; it was partial. Not all feeding branches were embolized because some of them were too small and short. Surgery was performed 48 h after embolization. We performed a submandibular transverse cervicotomy approach under electroencephalographic monitoring and general anesthesia. It allowed exposure of the entire tumor and proximal and distal neurovascular control. Osteotomy or subluxation of the mandible was not needed. The tumor was densely adherent to the walls of the vessels. We tried subadventitial dissection, but it was not possible without damaging the carotid arteries.

Therefore, we performed ligation of all branches of the external carotid artery with the Ligasure® Small Jaw Instrument LF1212 system and used it throughout the procedure for most blood vessels (Figure 2). A block resection was performed respecting cranial nerves and a carotid-carotid bypass with reversed saphenous vein. During the clamping and declamping, there were no electroencephalographic changes.

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Complete tumor resection was achieved; intraoperative bleeding was approximately 300–400 mL. After surgery, the patient presented a psychotic break and a multifactorial respiratory insufficiency: right vocal cord paralysis and use of antipsychotic drugs to control schizophrenia, requiring temporary tracheostomy. After 1-year follow-up, the patient was neurologically asymptomatic and was speaking normally. The control computed tomography scan showed the absence of tumor recurrence.

We want to state that the patient provided informed consent for his information and images to be included in this article, and hospital IdiPAZ Clinical Research Ethics Committee provided authorization to submit this case report.

Discussion

CBTs are rare slow-growing tumors that arise from the extra-adrenal paraganglion of the autonomic nervous system. CBTs are mainly nonfunctional, but sometimes may be functional and produce catecholamines, such as norepinephrine and epinephrine, and produce symptoms, such as palpitations, hypertension, and weight loss.

Some studies revealed that matrix metalloproteinase (MMP) enzymes are elevated in patients with CBTs compared to healthy patients. MMPs that regulate cell matrix composition play a role in several conditions, including embryogenesis, wound healing, inflammation, and cancer. MMPs seem to be more elevated on malignant CBTs, but this relation is not clear. Most of these lesions are benign. However, malignant behavior is sometimes encountered, with a rate of malignancy reported to be 6%–12.5%.

There are no histological characteristics to diagnose malignant CBTs. This diagnosis is reserved for tumors with local, regional, and distant metastasis. Therefore, tumor resection is indicated. Surgical resection is the treatment of choice. The type of tumor based on the Shamblin classification partially predicts intraoperative difficulties and morbidity. Surgery can be associated with significant morbidities, especially with large Shamblin III tumors. Complete resection can be very challenging and often requires block resection with interposition graft.

Most surgeons perform a longitudinal cervical incision for tumor resection. Submandibular approach is rarely used. It is commonly used on maxillofacial surgery. It allows exposure of the distal portion of the extra-cranial internal carotid artery, elevation of the mandible, osteotomy, and subluxation of the mandible when needed. Vascular sealing devices of the Ligasure® system are not commonly used in carotid surgery. The device comes in various sizes and indications according to the type of surgery. These devices are designed to reduce blood loss and operative time. Ligasure® system for head and neck surgery is used on thyroid and parathyroid procedures. Carotid surgery is not in the manufacturer specifications. Because of the extension of the tumor to the base of the skull in this case, we considered submandibular approach appropriate. This approach allowed us to completely resect the tumor, and the use of the Ligasure® system helped in the resection, possibly reducing intraoperative blood loss and surgical time. We believed that the use of Ligasure® system can be useful to manage large CBTs.

Declaration of conflicting interests

The authors declare that there is no conflict of interest.

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