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Case Report

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ABSTRACT

Carotid paragangliomas (CP) are rare tumors, representing 0.6% of the head and neck tumors. These tumors have their origin in the carotid body located in the adventitia of the vascular wall of the carotid bifurcation. Among their principal characteristics are hypervascularity, primarily dependent on branches of the external carotid artery, the proximity and possible involvement of the cranial nerves IX, X, XI, XII, and extension to the base of the skull. Complete surgical resection is the first line of management; however, this procedure can be a surgical challenge due to the potential risk of bleeding, intraoperative neurovascular injuries, and prolonged surgical time. Tumor embolization, carotid stenting, and tumor embolization with carotid stenting have been developed as alternative presurgical endovascular techniques that decrease tumor vascularity and/or provide structural vascular support, reducing bleeding and facilitating tumor dissection. Two cases of carotid tumors of the same classification, Shamblin II, are presented, one treated by preoperative embolization and the other managed with a carotid stent; the indications, advantages, and possible complications of each one are discussed. Two cases of Shamblin II carotid tumors are presented, one treated preoperatively with a carotid stent and the other with preoperative embolization. A literature review was carried out, with a search in PubMed that includes case reports, case series, review articles, meta-analyses on CP, presurgical tumor embolization, presurgical carotid stent placement, and surgical treatment of carotid body tumor. Hypervascularity and adhesion to the carotid wall are the leading causes of difficulties in surgical resection of CP. Optimal tumor embolization and/or preoperative carotid stent placement reduce intraoperative bleeding and provide vascular structural support, reducing intraoperative and postoperative complications.

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Abbreviations: CP, Carotid paragangliomas.

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Case 1

A 69-year-old female with a history of systemic arterial hypertension and rheumatoid arthritis came due to a painless mass in the left lateral region of the neck with progressive growth during the last 6 months. Physical examination revealed the presence of a mass in the left mandibular angle, fixed to deep planes, without other associated lesions; Doppler ultrasound showed a lesion in the carotid bifurcation, with defined borders, homogeneous, hypoechoic, and hypervascular on color Doppler, which displaced and partially covered the carotid vessels (Figs. 1A and B); CT angiography of supra-aortic vessels showed a mass of 5 \times 4.8 \times 4.5mm located in the carotid bifurcation, with avid enhancement after applying the contrast medium, which surrounded the external carotid and internal carotid arteries by more than 180° and did not infiltrate visceral neighbors structures (Fig. 2). This lesion was classified as carotid glomus Shamblin II.

In the clinical assessment between head-neck surgery and interventional radiology service, no absolute contraindication was found for the endovascular procedure. It was decided to perform presurgical management with carotid

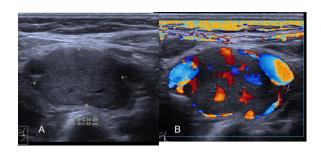


Fig. 1 – Case 1. (A) B-mode ultrasound, (B) color Doppler. Mass with defined borders, homogeneous and hypervascular on color Doppler, located in the left carotid bifurcation that displaces and partially surrounds the carotid vessels. Archive of the General Hospital of Mexico.

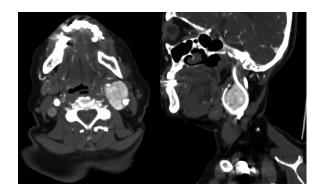


Fig. 2 – Case 1. Tomography angiography of supra-aortic vessels, mass in the left carotid bifurcation, with avid contrast enhancement, classified as Shamblin II. Archive of the General Hospital of Mexico.



Fig. 3 – Case 1. Arteriography. (A) Vascular sufficiency test of circle of Willis shows patency of the anterior communicating artery. Archive of the General Hospital of Mexico.

stent placement. The procedure was performed with vascular access through the right femoral artery. Arteriography of the aortic arch showed the emergence of the carotids through a common trunk, type II variant and the test of vascular sufficiency of the circle of Willis with compression of the left carotid artery showed adequate collateral circulation through the anterior communicating artery (Fig. 3), selective arteriography showed a tumor in the topography of left carotid paraganglioma, with vascularity mainly dependent on the external carotid artery (Fig. 4A). Subsequently, a 7×70 mm endovascular polytetrafluoroethylene prosthesis was placed from the common carotid artery to the internal carotid artery, achieving approximately 95% tumor devascularization (Figs. 4B and C). The procedure was completed without complications and its duration was 1 hour. Patient in current management with acetylsalicylic acid and clopidogrel.

Case 2

A 66-year-old male with a history of arterial hypertension, diabetes mellitus, dyslipidemia, and overweight, BMI 28 kg/m², with an incidental finding, during carotid Doppler, performed due to a recent cerebrovascular event, of a hypervascular carotid bifurcation lesion on Doppler application color; CT angiography of supra-aortic vessels showed a lesion in the right carotid bifurcation, which was displaced laterally and anteriorly and also partially covered the external and internal carotid arteries, which was classified as Shamblin II (Fig. 5). Evaluated by vascular surgery, which found no contraindication for a surgical procedure prior to presurgical endovascular management with embolization. The procedure was performed through femoral vascular access; subsequently, selective cannulation of the glomus carotid nutrient vessels dependent on branches of the occipital artery and direct neoves-

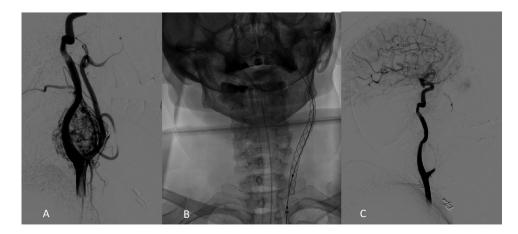


Fig. 4 – Case 1. Arteriography. (A) Tumor in the topography of left carotid paraganglioma, with vascularity mainly dependent on the external carotid artery. (B) Carotid stent 7 x 70 mm, from the common carotid to the external carotid, excluding 95% of the circulation of the external carotid artery. (C) Archive of the General Hospital of Mexico.

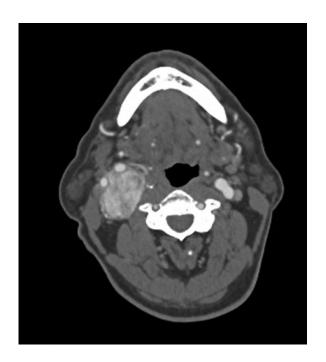


Fig. 5 – Case 2. Angiotomography of supra-aortic vessels, mass in the right carotid bifurcation with avid contrast enhancement, classified as Shamblin II. Archive of ISSSTE Regional Hospital Lic. Adolfo López Mateos.

sels of the external carotid artery (Figs. 6A and B) was performed with a PX SLIM microcatheter, through which the tumor was embolized with coil PC 400 14 \times 50, histoacryl, lipidol and squid with a decrease in tumor vascularity of approximately 80% (Fig. 6C). The embolization was completed without complications and its duration was one hour and a half. At 24 hours postembolization, complete surgical resection of the tumor was performed with intraoperative bleeding of 150cc, without intra- or postoperative complications or postoperative neurological deficit, discharging the patient home the day after surgery.

Discussion

Introduction

Carotid paragangliomas (PG) were first described in 1942 by Albrecht Von Haller, who described the presence of a "ganglion" in the carotid bifurcation [1]. They originate in the carotid body, an anatomical structure located in the adventitia of the carotid bifurcation, composed of paraganglionic cells and support cells, its function is chemoreceptors. Carotid body is mainly irrigated by the ascending pharyngeal artery and innervated by the glossopharyngeal nerve [2].

Epidemiology

PG are infrequent, slow-growing neuroendocrine tumors, generally non-functioning, represent 0.6% of head and neck tumors; they mainly affect women between 45 and 60 years of age in a male-female ratio of 8:1 [2]; 90%-95% are unilateral, and 5%-10% are bilateral [3]. Bilateral tumors have been described in patients with multiple endocrine neoplasia I and II, neurofibromatosis type I, Von Hippel Lindau disease, in familial forms of mutation of the gene of the succinate dehydrogenase (SDH) family SDHA, SDHB, and SDHD and people living at high altitudes [4,5]. Previously they were known as benign tumors, however, in 2017, given their uncertain malignant potential and that between 4% and 16% can become malignant and present regional and distant metastases, the WHO (World Health Organization) classifies it as a malignant tumor and classifies it with an ICD-O code (International Classification of Diseases for Oncology) 8692/3 [6].

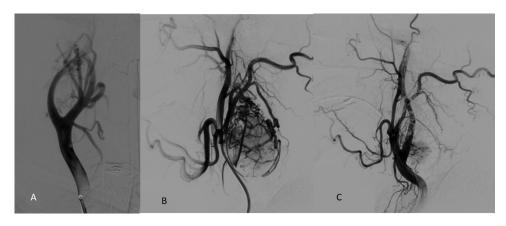


Fig. 6 – Case 2. (A) Arteriography, tumor in the topography of right carotid paraganglioma, with vascularity dependent on direct external carotid neovessels and the occipital artery. (B. C) Arteriography, postembolization control plus Coil placement, 80% decrease in vascularity. Archive of ISSSTE Regional Hospital Lic. Adolfo López Mateos.

Etiology

They originate in the carotid body, an anatomical structure located in the adventitia of the carotid bifurcation that measures $6 \times 4 \times 2$ mm, composed of 2 types of cells: type I paraganglionic cells of the extra-adrenal neuroendocrine system derived from neural crest cells and type II support cells, derived from the mesoderm of the third branchial arch [7], its function is chemoreceptor, regulates oxygen concentration and blood pH, parasympathetic and is generally not secretory [2].

Clinical presentation

PCs are usually asymptomatic tumors, being an incidental finding. However, they can present as a painless, slow-growing mass, associated with dysphonia, stridor, lingual paresis, dysphagia, Horner's syndrome, and cerebrovascular events; less than 1% are functioning tumors with the secretion of catecholamines, and their presentation can be as uncontrolled arterial hypertension, palpitations, headache, pallor/flushing, diaphoresis, weight loss and hyperglycemia [8].

Imaging and diagnostic findings

Imaging studies determine the location, extension, multifocality, metastasis, and tumor vascularity. In Doppler ultrasound, it is observed as a homogeneous lesion in the carotid bifurcation, with hypervascular behavior when applying color Doppler. In computed CT and magnetic resonance angiography, it appears as a homogeneous lesion in the carotid bifurcation, which partially displaces and/or surrounds the carotid vessels and may infiltrate adjacent neural and visceral structures, even presenting intracranial extension; it presents avid enhancement with the contrast medium, the lyre sign is pathognomonic; in T2 magnetic resonance, a classic but not pathognomonic is the salt and pepper sign, due to the voids of a vascular signal. Nuclear medicine studies with Indium-111 octreotide allow the identification of multicentric or metastatic tumors and postoperative residual tumor.

Arteriography is essential as a diagnostic complement and presurgical endovascular treatment: digital subtraction, its multiple angles of vision, and the possibility of 3D reconstructions allow the origin of the nutrient branches of the PC to be determined [9]. These arteries are usually multiple, and the vast majority have their origin either directly from branches of the ipsilateral external carotid artery (superior thyroid, ascending pharyngeal, facial, and occipital arteries), the vertebral artery, or the external carotid artery contralateral [10–12].

Treatment

Complete surgical resection is the choice management due to the risk of regional and distant metastases [13],[14]. However, this management can be a surgical challenge due to the anatomical location of the tumor, its hypervascularity, close relationships or compromise of the cranial nerves IX, X, XI, XII, and their possible extension to the base of the skull with the subsequent risk of intraoperative bleeding, neurovascular injury, cerebrovascular event, the requirement of vascular reconstruction and extensive surgical time. Shamblin's classification, published in 1971 [15], is based on the relationship of the tumor with the carotid vessels. Type I tumors are small tumors that do not surround the carotid vessels and can be easily separated from the adjacent arterial wall; type II tumors are larger lesions that partially surround the carotid vessels, and type III tumors are those that surround the entirely carotid vessels and adhere intimately to the vascular wall and may be associated with stenosis and visceral and intracranial infiltration. This classification determines operative morbidity and mortality based on the characteristics and size of the PG, with a positive predictive value for surgical difficulties and postoperative complications [16]. Radiotherapy is the alternative management in cases of unresectable, residual and malignant tumors, and for patients who are not candidates for surgery due to their underlying conditions.

Endovascular treatment

Complex CPs often require surgical management combined with preoperative endovascular therapy to obtain the best possible results in local tumor resection while maintaining neurovascular integrity [17]. In 1973, Hekster performed the first presurgical embolization of the carotid glomus using particles from the patient's muscles through a transfemoral approach [1]; since then and with technological advances, this technique has been perfected, and an alternative technique has been developed and/or complementary to embolization: carotid stenting.

Presurgical embolization

Presurgical embolization can be performed by direct percutaneous or endovascular technique using N-butyl cyanoacrylate, ethylene vinyl alcohol, purified porcine skin gelatin, coils, and microcoils. The benefits of this therapy are decreased vascularity and tumor size, less intraoperative bleeding with better visualization of the surgical field, easier tumor dissection, fewer neurovascular lesions, and shorter surgical time [18-20]. Potential complications include leakage of embolizing material into the internal carotid artery and the subsequent risk of cerebrovascular events. In a meta-analysis of 22 studies with a total of 578 patients, Jackson et al. [21] concluded that surgical resection with preoperative embolization reduces the volume of bleeding and surgical time compared to surgery without embolization. Texakalidis et al., in another meta-analysis, which included 35 studies and 1326 patients, compared the efficacy of preoperative embolization versus nonembolization and concluded similar results to those described by Jackson et al. in terms of bleeding reduction and surgical time with preoperative embolization [22]. A standard time between embolization and surgical resection has not been determined; different studies describe a time between 1 and 72 hours between embolization and surgery [23-26]. However, different authors recommend that surgery should be performed on the same day as the embolization [27]. This variability in the time between embolization and surgery, the different embolizing materials used, and patient selection bias is the probable reason that some studies report the non-efficacy of presurgical embolization in the treatment of CP [28-30].

Preoperative stenting

The first case of stent placement in the internal carotid artery was performed in 2000 by Nussbaum et al. [31], they reported a case of bare metal stent placement one month before the planned resection of a malignant neck tumor that compromised the internal corotid artery (ICA); in 2017, Markiewicz et al. [32] published a case series of 5 patients in whom ICA/common carotid artery (CCA) coated stents were placed preoperatively and subsequently underwent successful subadventential resection of head and neck tumors without the need for revascularization.

In preoperative stenting, self-expandable stents, balloonexpandable stents, or coated stents can be used [33]. The stent can be deployed in the external carotid or from the common carotid to the internal carotid. The primary use of this procedure is to provide vascular structural support [34],[35] and, secondly, tumor devascularization, allowing the surgeon a better dissection plane, with tumor resection either subadventential or including or not the vascular wall or external carotid artery [36]. In addition, by allowing continuous anterograde flow in the internal carotid artery, the stent reduces the risk of intraoperative cerebrovascular events, which is especially important in cases where the Matas test demonstrates an incompetent circle of Willis.

The carotid stent is an alternative to cases in which, due to the close relationship between the tumor and the carotid vessels, bypass, prosthetic revascularization, or grafting of the small saphenous is required [37]. Among the described complications of this stent are thrombosis, intra-stent stenosis, stent migration, and vascular dissection [38]. Furthermore, this procedure requires continuous platelet antiaggregation. Surgery is carried out 4 to 6 weeks after stent placement, allowing for the formation of a stable neointima on the stent's luminal surface [39].

About the embolization plus carotid stent, few cases have been described in the literature [36], mainly in malignant tumors of the head and neck; Its benefits are devascularization of the tumor and structural vascular support; its indications are bilateral tumors, tumors that compromise the carotid bifurcation, Shamblin III tumors, tumors with intracranial extension, failure of the balloon occlusion test or Matas test.

Patient consent

The authors confirm that informed consents were obtained from the patients for the publication of this article.

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