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Case Report

Carotid glomus neurosurgical and neurophysiological monitoring. Report of two cases

Monitorización neuroquirúrgica y neurofisiológica del glomus carotídeo. Reporte de dos casos

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ABSTRACT

Glomus-type tumors are rare neoplasms, representing 0.012% of all tumors worldwide with a median presentation between the fourth and fifth the decade of life. Most of the cases (90%) have sporadic presentation, with only 10% related to family history. Their location varies, with the minority at the head and neck. They present as a slow-growing mass at the neck accompanied or not by pain and symptoms due to compression of adjacent structures such as dysphagia, dysphonia or even dyspnea. In the present work, 2 cases of Shamblin III carotid glomus are reported, both treated surgically. Two different approaches were performed in each patient due to the characteristics and invasion of each one. Both procedures were monitored neurophysiologically. Currently, the patients are stable without the presence of neoplasia with mild to moderate neurological sequelae. As it is a rare neoplasm, there are no standardized guidelines for diagnostic and therapeutic approach, however surgical resection must be considered as the first line treatment, as in the reported cases.

Keywords: *neurophysiological monitoring; neurosurgery; paraganglioma; glomus tumor; carotid arteries; neck.*

RESUMEN

Los tumores tipo glomus son neoplasias poco frecuentes, que representan el 0.012% de todos los tumores a nivel mundial, con una presentación media entre la cuarta y quinta década de la vida. El 90% de los casos tienen presentación esporádica, y solo el 10% se relaciona con antecedentes familiares. Su localización es variable, siendo minoritaria en cabeza y cuello. Se manifiestan como una masa de crecimiento lento en el cuello, acompañada o no de dolor y de síntomas derivados de la compresión de estructuras adyacentes, como disfagia, disfonía o incluso disnea. En el presente trabajo se reportan 2 casos de glomus carotídeo Shamblin III, ambos tratados quirúrgicamente. Se realizaron dos abordajes diferentes en cada paciente, de acuerdo con las características e invasión de cada uno. Ambos procedimientos fueron monitorizados neurofisiológicamente. Actualmente, los pacientes se encuentran estables, sin presencia de neoplasia, con secuelas neurológicas leves a moderadas. Al tratarse de una neoplasia rara, no existen guías estandarizadas para el abordaje diagnóstico y terapéutico; sin embargo, la resección quirúrgica debe considerarse como tratamiento de primera línea, tal como en los casos reportados.

Palabras clave: *monitoreo neurofisiológico; neurocirugía; paraganglioma; tumor glomus; arterias carótidas; cuello.*

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1 INTRODUCTION

Paragangliomas or glomus, correspond to a group of tumors that arise from embryonic cells of the neural crest with a prevalence of 0.012%[1]. Only a few are originated at the head and neck[2,3], distribution varies according to sex, with the carotid glomus being more common in men and the vagal glomus in women[4]. The age of presentation is usually between the fourth and sixth decades of life, affecting predominantly women[5].

Almost 90% of the cases are sporadic, with only 10% associated with family history. Carotid glomus represents close to 65% of neck originated tumors, principally at the carotid bifurcation, with a malignity rate up to 10%[1,5].

Carotid glomus approach is not standardized, however, in cases where surgical resection is viable, it should be considered the treatment of choice[1,5]. Among treatment alternatives, where surgical resection is not a viable option, embolization and radiotherapy are commonly used[6].

Two cases of carotid glomus surgically managed with neurophysiological monitoring under CARE guidelines are reported.

This work is a case report, therefore approval from an Ethics Committee was not required. Only signed informed consent from the patients was obtained for academic use of their clinical information.

2 CASE PRESENTATION

Case # 1

43 years old female from Mexico City with personal history of car crash 20 years ago and family history of diabetes, hypertension and Alzheimer`s Disease. Refers slow growth cervical mass since November 2022, for which an angiotomography was requested. Previously evaluated by oncologists arrived for a second opinion. On physical examination a solid mass on anterior neck from left sternocleidomastoid to left axillary angle was found. The angiotomography showed a glomic-type lesion of the left carotid Shambling III, which extends from the carotid bifurcation to the base of the skull. A contrasted computed tomography (CT) was requested, demonstrating the presence of 3 cm x 3 cm carotid glomus that invades the base of the skull. No genetic alteration was intentionally looked for. Due to the obtained data surgical resection was scheduled.

An infratentorial craniotomy was performed with lateral FAR access with tumor exposure, resection, and cranial drilling for tumor release. Cervical glomus was resected; anastomosis and repair of the extracranial carotid artery was performed with graft placement for bridging. The entire procedure was made under neurophysiological monitoring of the facial nerve and brain stem, which reported manipulation of the recurrent laryngeal nerve without persistence. Tumor resection product was sent to anatomopathological study which reported a paraganglioma of 3.5 x 3.0 x 2.0 cm without lymphovascular invasion and immunophenotype with positive GATA3, diffuse SDHB and positive S100 (Figure 1).

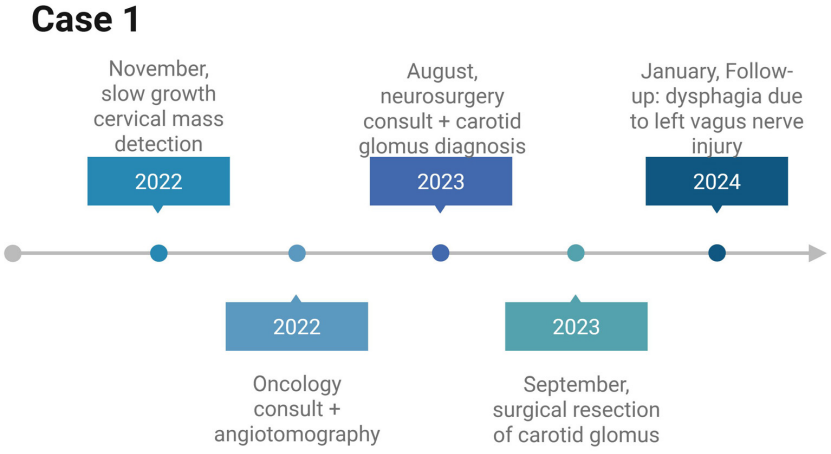

Figure 1. Timeline of case report #1.

Figure 2. Angiotomography with right carotid glomus, Shamblin III.

In the mediate postoperative, patient presented dysphagia due to injury of the left vagus nerve secondary to tumor dependence. At 1 year follow up, patient refers decrease of the previously mentioned dysphagia with no other symptomatology.

Case # 2

72 years old female with personal history of multiple lumbar surgical procedures and 2 years evolution right shoulder pain without sensitivity or strength alterations. In March 2024, neck pain reappeared accompanied by radiculopathy with irradiation to the right arm. On physical examination a right anterior neck mass was found, which comprises from sternocleidomastoid to maxillary angle with Bruit and heartbeat compatible with carotid glomus. A magnetic resonance imaging (MRI) was requested which reveals a tumor compatible with carotid glomus dependent on the right carotid bifurcation, classifying it as Shamblin III. Angiotomography of supra-aortic vessels was requested, which confirmed carotid glomus (Figure 2). No genetic alteration was looked for. Due to the obtained data surgical resection was scheduled.

A cervical approach was performed on anterior carotid space. Jugular vein, common, internal and external carotid were identified, and total resection of the tumor was performed (Figure 3), with preservation of adjacent vascular and neurological structures.

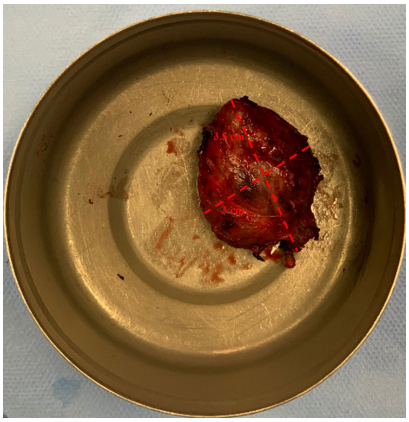

Figure 3. Carotid glomus resection product.

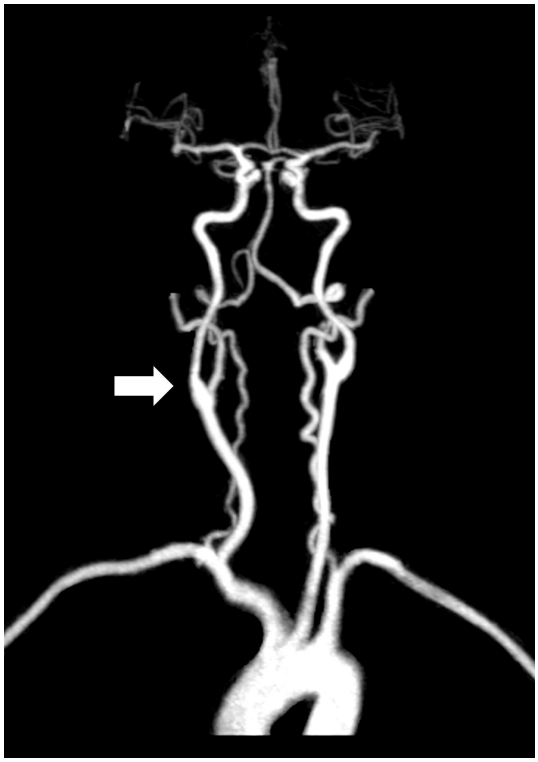

Figure 4. Contrasted computerized tomography demonstrating no occupying process on the vascular pathway.

The entire procedure was made under neurophysiological monitoring, without any reported nerve manipulation. Tumor resection product was sent to anatomopathological analysis that reported a paraganglioma of 3.5 x 3 cm with isolated mild atypia, no immunophenotype study was made. Postsurgical contrasted computerized tomography reported pathway of the common carotid arteries, towards the bifurcation and the course of the jugular vein without any occupying process (Figure 4).

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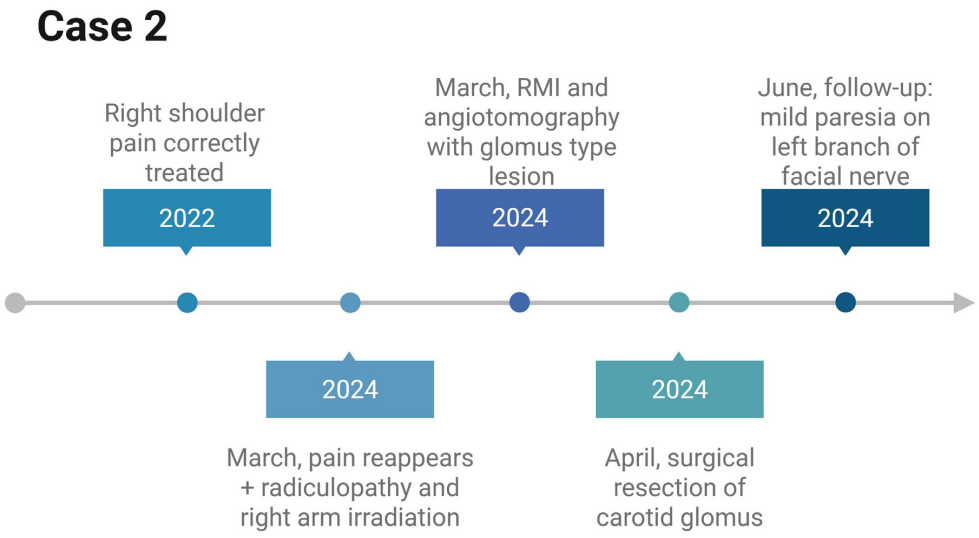

Figure 5. Timeline of case report #2.

In the mediate postoperative, the patient had minimal paresis in the labial branch of the right facial nerve. Long term follow up was lost due to other medical complications secondary to an abdominal hernia (Figure 5).

3 DISCUSSION

Glomus type tumors, also known as paragangliomas, are rare neoplasms originated from the neural crest cells, representing 0.012% of all neoplasms[1], the minority is located on head and neck[2]. Most cases, near 90%, have a sporadic origin, being only the 10% of the cases of genetical and familiar etiology, the last ones commonly are bilateral and/or multiple[2,3]. They usually appear between the fourth and fifth decades of life[2]. According to a study carried out in Mexico in 2014, carotid glomus is more common in women[7] (Table 1), as in the reported cases.

They have benign characteristics and slow growth rate, being initially painless and asymptomatic, with principal prevalence at carotid bifurcation, jugular foramen, middle ear and cervical portion of the vagus nerve[3]. Depending on clinical evolution, patients may present dysphagia, visual disorders, Horner syndrome and cranial neuropathies[2,4]. Additionally, close to 1% of cases present as secreting tumors which gives characteristic symptomatology[4].

Carotid glomus represents 65% of neck paragangliomas, with 6-10% malignancy rate, determined by the extension, adjacent organs invasion and nodular metastases[3,4]. During physical examination involvement of adjacent organs, metastases, stability, Fontaien sign (craniocaudal immobility), heartbeat and nodular growth must be evident. Paraclinical approach should include angiotomography and magnetic resonance imaging (MRI), which allows making differential diagnosis with other cervical injuries. Magnetic resonance T1 sequence shows the classic "salt and pepper" pattern[2].

Surgical resection is the standard and choice treatment, being the only one with a curative objective[8], commonly with a difficult approach due to the high vascularity and cranial nerves presence[9-11,]. Among treatment preoperative alternatives embolization or radiotherapy are commonly used techniques, however, studies have not shown significant advantages[11,12] (Table 1).

Shamblin criteria, proposed in 1970, is used to classify carotid tumors, evaluating the surgical risk based on tumor and carotid artery relation[7,9,12]. The classification confers 3 different grades depending on the size, location and adjacent vessels relationship. Shamblin I correspond to small and well localized tumors that allow an easy split from the vessels[8,13,14]. On the other hand, Shamblin II refers to tumors that partially surround the vessels and adhere to their adventitial layer. Finally, Shamblin III are those tumors completely attached to vessels, which surgical resection commonly requires prosthesis or graft for vascular repair[10,13], as in the currently reported cases.

Table 1. Comparison with previous studies conducted in Mexico.

	(Palos, 2014)	(López, 2005)	(Kemeberly et al, 2022)
Age	55.3 years	41 years	52 years
Gender			
Female	24 (82.7%)	74 (80.43%)	4 (100%)
Male	5 (20.8%)	18 (19.56%)	
Tumor laterality			
Unilateral	27 (87%)	88 (95.65%)	4 (100%)
Bilateral	2 (13%)	4 (4.34%)	0 (0%)
Clinical presentation			
Cranial nerve affection	-	14 (39.13%)	1 (25%)
Hearing loss, nystagmus, fainting			
Adjacent organs compression	-	5 (5.43%)	-
Neck swelling			
	-	30 (32.6%)	1 (25%)
	-	-	3 (75%)
Classification			
Shamblin I	4 (13.7%)	54 (62.8%)	2 (50%)
Shamblin II	15 (51.7%)	25 (29.0%)	1 (25%)
Shamblin III	12 (41.3%)	7 (8.4%)	1 (25%)
Treatment			
Embolization	-	-	0 (0%)
Radiotherapy	-	1 (1.16%)	0 (0%)
Surgical resection	-	82 (95.34%)	4 (100%)

Note: Comparison of glomus-tumor between previous studies conducted in Mexico.

Surgical resection is not exempt of complications, with cranial nerves lesion being the most prevalent (19-49%), specifically hypoglossal and vagus nerves. During surgery patients may present electroencephalographic changes, bradycardia, hypotension and hemorrhage, reason why neurophysiological monitoring was performed during both reported cases.

In this report, 2 carotid glomus were presented, both treated with surgical resection and neurophysiological monitoring during the procedure.

4 CONCLUSIONS

Due to the rarity of the neoplasm, no standardized guidelines exist for diagnostic and therapeutical approach, although, many medical specialists are involved such as oncology, head and neck surgery, angiology and neurosurgery. Whenever there is the possibility, surgical resection should be considered as first line treatment, as in the reported cases, which may or may not be followed by radiotherapy.

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Ethics Committee Approval: The present research corresponds to a **clinical case report**, *a modality that in academic and scientific practice does not require approval from an Ethics Committee, provided that* **informed consent** *is obtained from the patients involved. No experimental interventions or procedures outside of standard medical care were performed; therefore, no additional risks were generated for the patients. Informed consent was duly signed by the participants, authorizing the use of their clinical information for academic and publication purposes. For this reason, submission to an Ethics Committee was not considered necessary, in accordance with international and national regulations which establish that* **clinical case reports** *require only the documentation of informed consent.*

Informed consent: Patients have informed consent of all diagnostic, therapeutic procedures and publication in scientific article done.

Institution: Hospital Angeles Puebla.