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Surgical Management of Cervical Paragangliomas: A Series of Four Cases

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Abstract

Cervical paragangliomas remain a pathological entity that is both fascinating and difficult to manage. These rare neuroendocrine neoplasms, which originate in non-chromaffin paraganglia (mainly the carotid body and vagus nerve), often present as a simple lateral cervical swelling. However, behind this apparent benignity lies formidable anatomical complexity, particularly their close relationship with the major vascular and nerve axes of the neck. The Shamblin classification guides our approach to carotid forms. The standard curative treatment remains radical surgical excision, which must be planned with extreme care to preserve functional integrity. Before any intervention, it is essential to rule out any multifocal form or the presence of an associated pheochromocytoma.

This study presents experience from the ENT department at Mohammed VI University Hospital, Oujda, focusing on the management of four cases of cervical paragangliomas. Our series was predominantly female (three of the four patients were women) and had an average age of 58 years. It consisted of three paragangliomas of the carotid body and one vagal paraganglioma. The postoperative results highlighted the effectiveness of subadventitial dissection for carotid forms, as well as the persistent challenge posed by vagal paraganglioma surgery in terms of function.

Categories: Otolaryngology, Pathology, Anatomy
Keywords: carotid body tumor, dysphonia, paraganglioma, shamblin classification, vagal

Introduction

Paragangliomas are rare neuroendocrine tumors that arise from paraganglionic tissue. In the head and neck region, they are mainly located at the carotid bifurcation, along the vagus nerve, and in the jugulotympanic region. Most of these tumors are slow-growing and non-secretory, meaning they do not secrete excessive amounts of catecholamines. They usually present as a painless and progressively enlarging cervical mass. However, depending on their location and size, neurological symptoms such as dysphonia or dysphagia may occur [1].

The management of cervical paragangliomas is challenging because of their rich vascularization and their close relationship with major cervical vessels and cranial nerves. Carotid body tumors are particularly related to the internal and external carotid arteries. The Shamblin classification categorizes these tumors into three groups according to their relationship with the carotid vessels: group I includes tumors with minimal vascular involvement, group II includes tumors partially surrounding the carotid arteries, and group III includes tumors extensively encasing these vessels. This classification helps assess surgical complexity and vascular risk [2].

Imaging plays an important role in diagnosis and preoperative assessment. CT and MRI with contrast can determine the location and extent of the tumor and its relationship with surrounding vascular and nervous structures. MRI and angiographic sequences are particularly useful in highly vascular lesions and in planning surgical treatment [1,3].

A genetic predisposition can be found particularly in patients with bilateral, multifocal, recurrent, or familial tumors. Hereditary paragangliomas are associated frequently with mutations involving the genes of succinate dehydrogenase. Evaluation for multifocal disease and associated paragangliomas is therefore an important part of patient assessment [1,3].

Treatment depends on the characteristics of the tumor and the patient. Surgery remains an important treatment option, especially for symptomatic and resectable tumors, but the potential neurological and vascular morbidity must always be considered. This issue is particularly important for vagal paragangliomas because of their close relationship with the vagus nerve. Radiotherapy and surveillance may also be considered in such cases [1,3].

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In this study, we report our experience with four cases of cervical paragangliomas managed in the ENT department of Mohammed VI University Hospital, Oujda. The series includes three carotid body paragangliomas and one vagal paraganglioma. We describe their clinical presentation, surgical management, and postoperative outcomes, highlighting the distinct challenges related to vascular involvement, nerve preservation, and postoperative neurological recovery.

Case Presentation

Case 1

A 76-year-old woman with a history of hypertension, ischemic stroke with residual right-sided deficit, and previous total thyroidectomy was admitted for a left lateral cervical mass that had increased in size over the past two years. It became painful and more pulsatile in the last three months before admission. Clinical examination revealed a firm, painful left cervical mass measuring approximately 5 cm, without inflammatory signs; preoperative flexible nasolaryngoscopy showed normal bilateral vocal fold mobility.

Doppler ultrasonography showed a well-defined, hypoechoic mass measuring 37×29 mm adjacent to the left common carotid artery. CT angiography demonstrated a 33×30 mm left carotid mass with intense contrast enhancement. MRI confirmed a 30×29 mm tumor at the carotid bifurcation with intense enhancement and bifurcation widening, producing the characteristic "lyre sign," consistent with a carotid body paraganglioma (Figure *1*). This finding favored a carotid body paraganglioma over other cervical masses, such as schwannomas or lymphadenopathy. Preoperative biochemical assessment, including catecholamine testing, showed no evidence of catecholamine hypersecretion. Additional investigations revealed no other paraganglioma localization. Genetic testing was not performed.

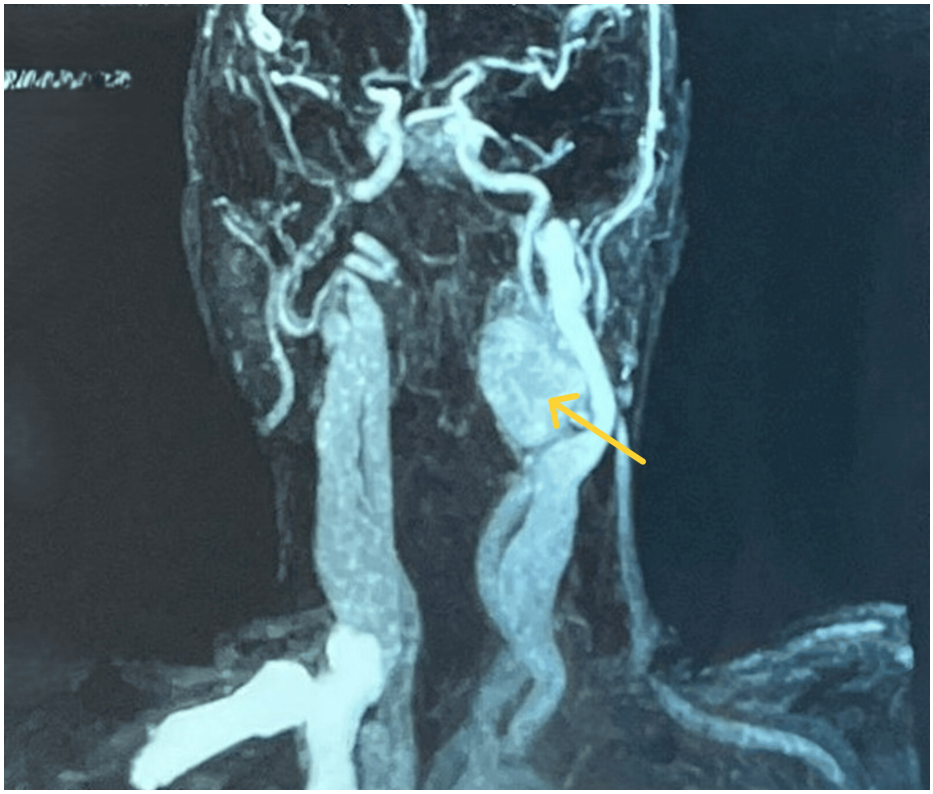


FIGURE 1: Angio-MRI showing mass located at the left carotid bifurcation, consistent with a carotid body paraganglioma (yellow arrow).

The patient underwent complete surgical excision by subadventitial dissection without vascular sacrifice (Figure *2*). No postoperative vascular or neurological complications or sequelae were found. Histopathological examination of the 3×2.5×1.8 cm specimen confirmed a carotid body paraganglioma without aggressive features. Clinical follow-up over 18 months was uneventful.

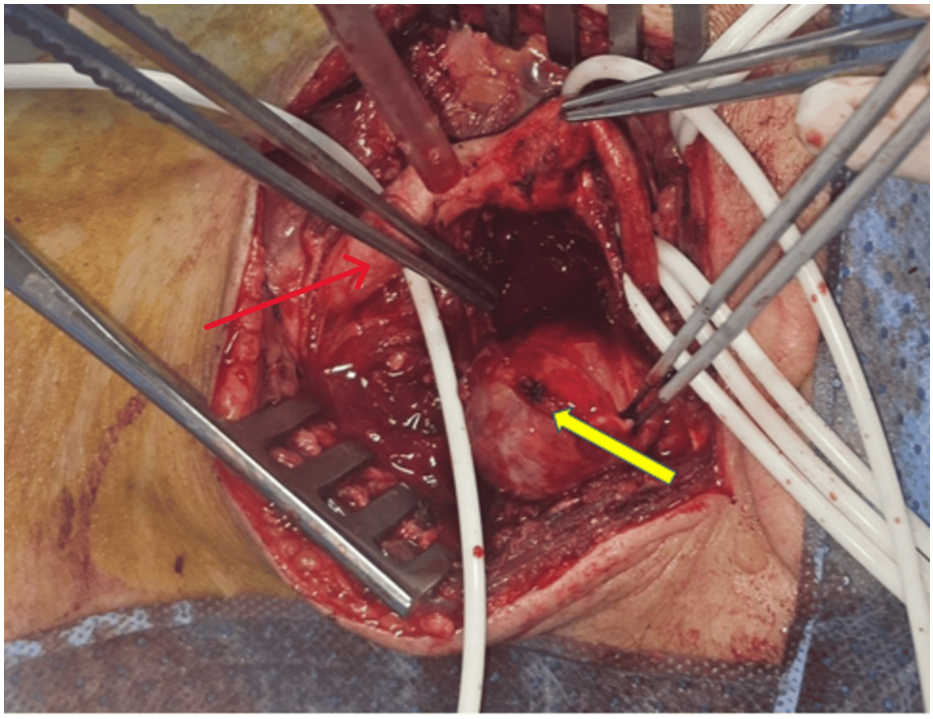


FIGURE 2: Intraoperative view of the left carotid body paraganglioma after dissection.

The yellow arrow indicates the paraganglioma, and the red arrow indicates the common carotid artery.

Case 2

A 54-year-old woman with no significant medical history was admitted for a right lateral cervical mass that had been progressively increasing in size for one year. The mass had recently become painful, without other associated symptoms. Clinical examination revealed a firm, painful right cervical mass measuring approximately 3 cm, mobile relative to the superficial and deep planes, with no inflammatory signs.

CT scan revealed a right cervical mass in close contact with the carotid space; the lesion measured 28×25 mm with a craniocaudal extension of 36 mm and showed marked enhancement after contrast injection (Figures *3A*, *3B*). MRI showed a well-defined right carotid body mass measuring 27×20 mm on axial images and approximately 33 mm on sagittal images; it was heterogeneous and hyperintense on T2-weighted images, with marked enhancement after gadolinium injection (Figure *4*). The mass separated the internal and external carotid arteries and encased the external carotid artery over more than 180°. No other cervical lesions were found. The separation of the internal and external carotid arteries was characteristic of a carotid body paraganglioma and helped distinguish it from other cervical masses, particularly schwannomas and lymphadenopathy. Catecholamine testing and screening for multifocal disease were negative. Genetic testing was not performed.

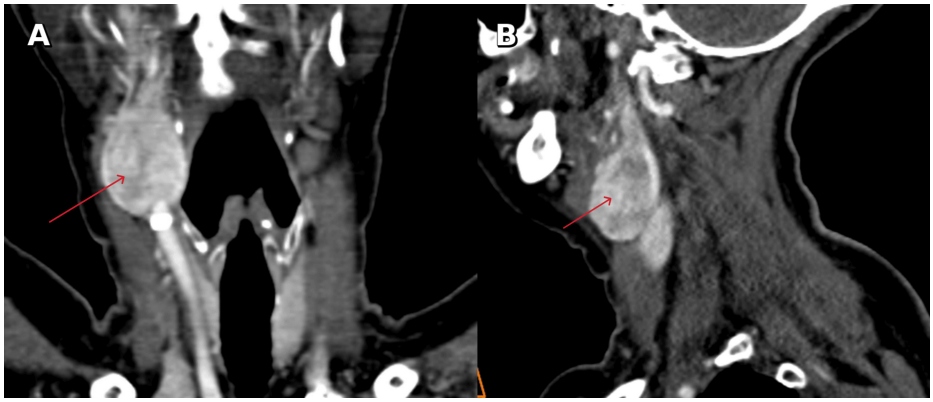


FIGURE 3: Contrast-enhanced cervical CT images in coronal (A) and sagittal (B) planes showing a hyperdense, intensely enhancing right carotid space mass (red arrows), consistent with a carotid body paraganglioma.

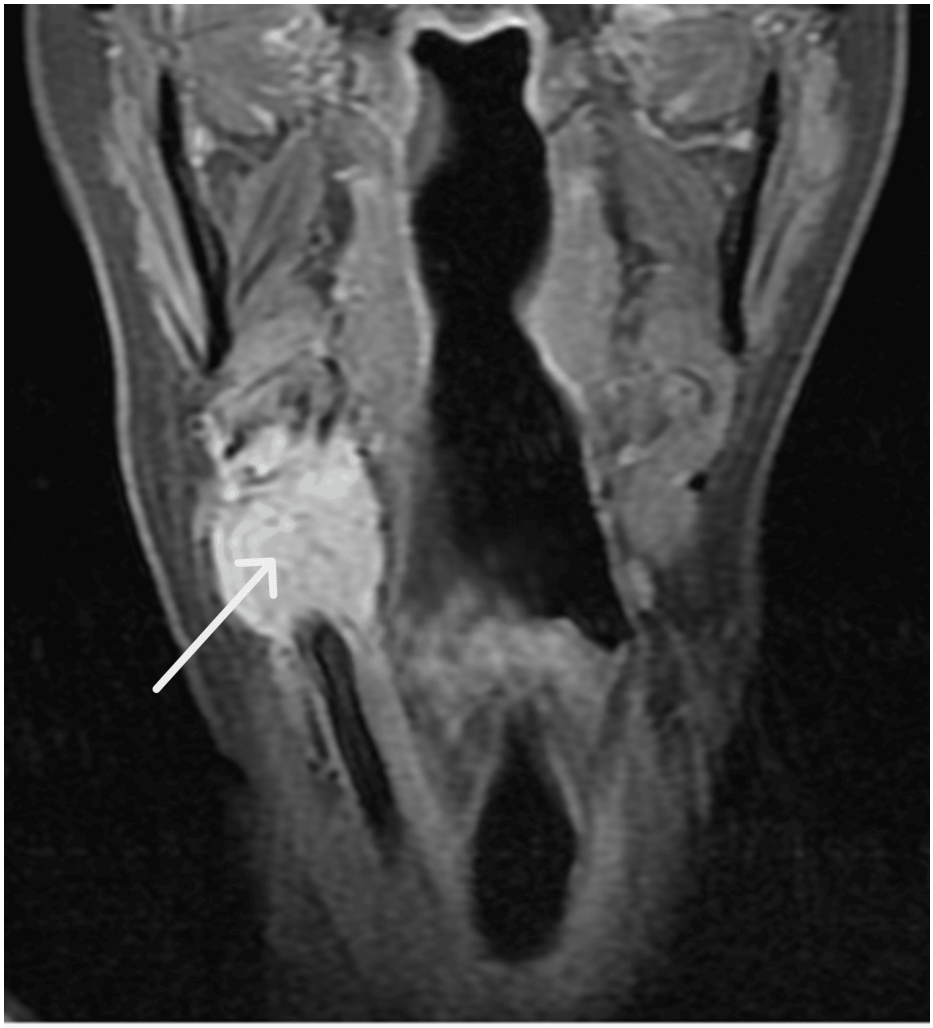


FIGURE 4: Coronal contrast-enhanced T1-weighted MRI showing a right carotid body tumor (white arrow) with marked enhancement after gadolinium injection.

Complete surgical excision was performed by subadventitial dissection, requiring vascular clamping and

sacrifice of the external carotid artery. No postoperative complications or neurological or vascular sequelae were observed. Histopathological examination confirmed a carotid body paraganglioma without aggressive features. Follow-up at one, three, six, and 20 months showed no abnormalities or complications.

Case 3

A 60-year-old woman with a four-year history of diabetes mellitus and a five-year history of hypertension was admitted for a left lateral cervical mass. The swelling had appeared approximately 18 months earlier and progressively increased in size. It became larger and caused local discomfort six months before consultation. Clinical examination showed a firm, pulsatile left submandibular mass measuring approximately 4 cm, painful on palpation and mobile relative to the deep plane, without inflammatory signs. CT imaging showed a heterogeneous, highly vascularized mass at the left carotid bifurcation, measuring 42×30 mm, with intense enhancement after contrast injection (Figures *5A*, *5B*).

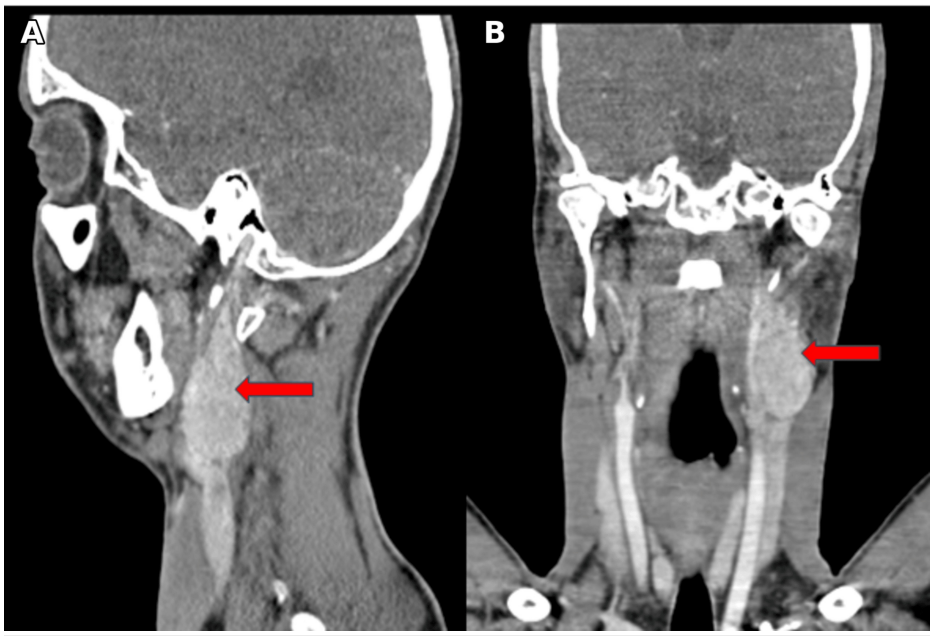


FIGURE 5: Contrast-enhanced cervical computed tomography (CT) showing a left carotid space mass suggestive of a carotid body paraganglioma.

(A) Sagittal and (B) coronal reconstructions demonstrating an enhancing soft-tissue mass in the left carotid region (red arrows).

MRI demonstrated a 40×28 mm lesion that was hypointense to isointense on T1-weighted images and hyperintense on T2-weighted images, with marked enhancement after gadolinium injection. The mass was located at the carotid bifurcation, producing the characteristic “lyre sign.” This vascular enhancement and separation of the carotid arteries favored a carotid body paraganglioma over other cervical masses, including lymphadenopathy and schwannomas. Biochemical screening was negative, with no additional paragangliomas identified. Genetic testing was not performed.

The tumor was completely removed by subadventitial dissection without vascular or nerve sacrifice, and no transfusion was required. The postoperative course was uncomplicated. Histopathological examination confirmed a completely resected (R0) carotid body paraganglioma, with no signs of malignancy reported. Follow-up at one, three, six, 12, and 18 months showed no complications or sequelae.

Case 4

A 42-year-old man with a history of asthma was admitted for a right lateral cervical mass. His symptoms had started three years earlier with intermittent cervical pain and a progressively enlarging right cervical swelling. Dysphonia subsequently developed, without compressive or secretory symptoms. Clinical examination revealed a firm, pulsatile, and painful right cervical mass measuring approximately 5 cm, without inflammatory signs.

CT scan showed a homogeneous right jugulocarotid mass with marked contrast enhancement, measuring 25×40 mm and extending over 52 mm (Figures *6A*, *6B*). MRI angiography demonstrated a hypervascular mass

arising from the vagus nerve, measuring 25×39 mm, surrounding the jugulocarotid vessels and extending to the skull base without bone destruction. The location along the vagus nerve and the marked vascular enhancement favored a vagal paraganglioma over other cervical masses, particularly a vagal schwannoma. Catecholamine levels were normal, no additional paragangliomas were detected, and genetic testing was not performed.

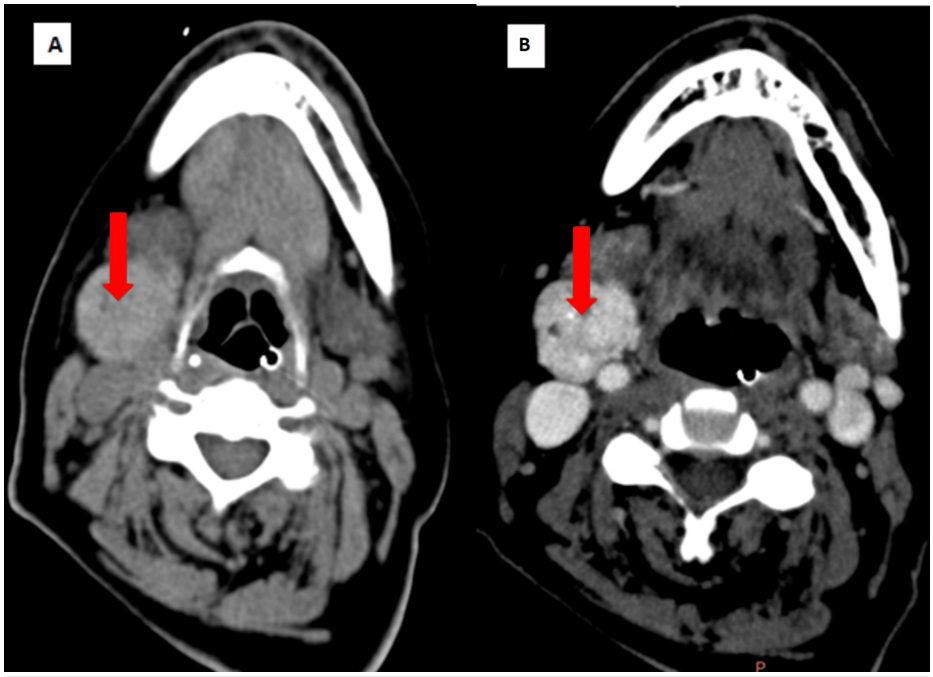


FIGURE 6: Axial cervical CT images before (A) and after (B) contrast injection showing a right-sided vagal paraganglioma (red arrows) displacing the carotid and jugular vessels.

Surgical exploration confirmed that the tumor developed from the vagus nerve. Complete resection required sacrifice of the vagus nerve, while the vascular structures were preserved. Three adjacent lymph nodes were also removed. Postoperatively, the patient developed hoarseness and dysphagia requiring temporary nasogastric feeding. Swallowing function began to improve one week postoperatively and continued to recover progressively over one year with rehabilitation, although complete recovery was not achieved, whereas dysphonia persisted and required speech therapy. Histopathological examination confirmed a vagal paraganglioma, with no signs of malignancy, and the three lymph nodes showed no metastatic involvement. Progressive improvement was observed during follow-up, with recovery of a normal voice at three years. The clinical, radiological, surgical, and postoperative characteristics of the four patients are summarized in Table *1*. The clinical data collection form used to document patient information, medical history, clinical findings, investigations, and treatment is provided in the table in appendix.

Characteristics	Case 1	Case 2	Case 3	Case 4
Age (years)	76	54	60	42
Sex	Female	Female	Female	Male
Tumor location	Left carotid body	Right carotid body	Left carotid body	Right vagal paraganglioma
Shamblin classification	II	III	II	NA
Tumor size on imaging (mm)	37×29	36×28×25 (CT)	42×30 (CT)	52×40×25 (CT)
Operative time (min)	170	194	120	190
Estimated blood loss (mL)	ND	ND	ND	ND
Vascular sacrifice	None	External carotid artery	None	None
Nerve sacrifice	None	None	None	Right vagus nerve
Histopathological results	Carotid body paraganglioma, with no signs of malignancy	Carotid body paraganglioma, with no signs of malignancy	Carotid body paraganglioma, with no signs of malignancy	Vagal paraganglioma, with no signs of malignancy
Postoperative complications	None	None	None	Persistent hoarseness and partially improved dysphagia
Follow-up duration (months)	18	20	18	36
Recurrence	No	No	No	No

TABLE 1: Clinical, radiological, surgical, histopathological, and follow-up characteristics of the four patients.

Discussion

In our series, three patients had carotid body paragangliomas, and one had a vagal paraganglioma. All four tumors were surgically removed, with preservation of the carotid circulation in the three carotid body tumors. These results illustrate that surgery can provide complete tumor removal, but the choice of treatment should take into account tumor location, size, vascular involvement, age, and expected neurological morbidity. Current recommendations emphasize an individualized approach, as observation and radiotherapy are also appropriate options in selected patients, particularly when preservation of neurological function is a major concern [1].

Carotid body paragangliomas and vascular management

In the three patients with carotid body paragangliomas, complete resection was achieved using subadventitial dissection. The carotid circulation was preserved in all three cases, although sacrifice of the external carotid artery was required in one patient because of its close involvement with the tumor. This is consistent with the surgical principle of careful subadventitial dissection, which provides a plane between the tumor and the carotid vessels and may facilitate vascular preservation [4]. The difficulty of resection is closely related to the degree of carotid involvement. The Shamblin classification is useful for describing the relationship between the tumor and the carotid vessels and for anticipating surgical complexity [2]. Higher Shamblin classes are associated with greater vascular and cranial nerve morbidity.

Vagal paraganglioma and neurological morbidity

The vagal paraganglioma in our series illustrates the greater neurological risk associated with this location. Complete tumor removal required sacrifice of the vagus nerve, while the carotid and jugular vessels were preserved. Postoperatively, the patient experienced persistent hoarseness and transient swallowing

difficulties requiring temporary nasogastric feeding. These findings are consistent with previous reports showing that surgical resection of vagal paragangliomas frequently results in loss of vagal nerve function, with dysphonia and dysphagia representing important postoperative complications [5]. Swallowing function began to improve after one week and continued to recover progressively over one year with rehabilitation, although complete recovery was not achieved. Voice quality improved over three years, likely reflecting contralateral vocal fold compensation rather than recovery of vagal nerve function. However, postoperative laryngoscopy was not performed to confirm this mechanism.

Preoperative embolization

Preoperative embolization was not performed in any of our three patients with carotid body paragangliomas. Direct surgical resection without prior embolization was considered feasible based on preoperative imaging and surgical assessment. All three tumors were completely removed by subadventitial dissection. In one case, sacrifice of the external carotid artery was necessary. The role of embolization before surgery remains debated in the literature. It may be useful in selected highly vascular tumors, but its benefit is not clearly established in all patients [6].

Follow-up and treatment alternatives

Follow-up is important because paragangliomas are slow-growing tumors and recurrence or new lesions may occur [3]. In our series, no recurrence was found during the available follow-up. All four patients underwent surgery, but other approaches can be considered depending on the patient and the tumor. Observation or radiotherapy may be appropriate when the expected morbidity of surgery is high [1,3].

Limitations

Our study has some limitations. First, our experience is based on only four patients, so the results cannot be generalized. The duration of follow-up was also different between patients, and intraoperative blood loss was not documented in the available medical records, limiting the assessment of perioperative bleeding. Despite these limitations, our cases show some of the difficulties encountered in the surgical management of cervical paragangliomas, particularly when the carotid vessels or the vagus nerve are involved.

Conclusions

Cervical paragangliomas remain challenging tumors because of their close relationship with major vascular and neural structures. In our series, complete surgical resection was achieved in all four patients. The common and internal carotid arteries were preserved in the three carotid body paragangliomas, although sacrifice of the external carotid artery was required in one case. The vagal paraganglioma required sacrifice of the vagus nerve, resulting in neurological sequelae, with gradual improvement in swallowing function and voice quality likely reflecting contralateral vocal fold compensation rather than neural recovery. Although limited to four patients, our institutional experience illustrates the importance of careful preoperative assessment and individualized surgical planning, particularly regarding tumor location, vascular involvement, and neurological risk. Genetic screening and long-term imaging follow-up should also be considered because of the potential for hereditary disease, multifocal tumors, and late recurrence.

Appendices

Section	Clinical parameters	Details
Patient identification	Name and origin	
	Age	
	Sex	Male/female
Medical history	Personal history	Hypertension/diabetes/asthma/renal insufficiency
		Hypoxemic conditions
		Heart disease/pulmonary disease/ischemic stroke
		Current medications
		Previous surgical history
	Family history of similar cases	
Reason for presentation	Main symptoms	Neck mass/pain/dizziness or vertigo/headache/dysphonia/tachycardia/sweating/dysphagia/hearing loss
	Duration of symptoms	
Clinical findings	Cervical examination	Location of the mass
		Consistency, side, mobility, dimensions, and presence or absence of inflammatory signs
	ENT and general physical examination	
Investigations	Ultrasound	
	CT scan	
	Angiography	
	MRI	
	Scintigraphy	
	Blood tests	
	Urine tests	
Treatment	Type of excision	Total/partial
	Extent of resection	
	Structures involved in resection	Internal carotid artery/External carotid artery/Jugular vein/vagus nerve/glossopharyngeal nerve/peripheral lymph nodes
	Embolization	Yes/no
Postoperative follow-up	Postoperative sequelae	
	Histopathological results	
	Outcome and follow-up	

TABLE 2: Clinical data collection form for patients with cervical paragangliomas.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Safae Elmelhaoui

Acquisition, analysis, or interpretation of data: Safae Elmelhaoui, M'barek Ben Youssef, Achraf Amine Sbai, Drissia Benfadil, Azzedine Lachkar

Drafting of the manuscript: Safae Elmelhaoui

Critical review of the manuscript for important intellectual content: M'barek Ben Youssef, Achraf Amine Sbai, Drissia Benfadil, Azzedine Lachkar

Supervision: Achraf Amine Sbai, Drissia Benfadil, Azzedine Lachkar

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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