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Case Report
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Surgical Management of Carotid Paraganglioma

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Abstract

Paragangliomas are rare and usually benign well vascularized neuroendocrine tumors. They develop from diffuse neuroendocrine system cells. They may present an extensive growth and compromise vital neurovascular structures in the neck, such as carotid arteries. The main treatment for carotid body paragangliomas is surgical resection, which can be difficult and challenging in case of adherence to the carotid wall.

We report the case of a 30-year-old woman, who was admitted to our department with a pulsatile and painless neck tumor which had grown slowly. CT scan confirmed the presence of a tumor measuring 30.7 mm × 20.8 mm invading the carotid bifurcation and the internal carotid artery. The neck tumor had been successfully resected, and paraganglioma was diagnosed by histological examination of the surgical specimen. The postoperative course was uneventful.

Keywords: Paraganglioma; Carotid artery; Surgery; Excision

Introduction

Paragangliomas are chromaffin tumors arising from neural cleft cells that can originate from sympathetic or parasympathetic ganglia [1]. The carotid body is the most common site for these tumors' formation [2]. Only 5% of these tumors are bilateral [3].

Their growth is slow, but they have a risk of local compression and of associated malignancy [4]. Carotid involvement is frequent and makes resection of these tumors difficult. So, it is critical for surgeons to identify tumors likely to have arterial involvement and to avoid and manage injuries to these vessels [2].

We report a case of paraganglioma of the carotid body, resected surgically, and diagnosed by histological examination of the surgical specimen.

Case Presentation

A 30-year-old woman with no past medical history was admitted to our department for neck mass. She was symptomatic of a painless mass in the right lateral region of the neck with progressive growth during the last 6 months.

Physical examination revealed a pulsatile and painless mass in the right carotid region of the neck. Duplex ultrasound showed a lesion in the carotid bifurcation, with defined borders, homogeneous and hypervascular on Doppler. She had undergone a supra-aortic trunk Computed Tomography (CT), revealing a tumor measuring 30.7 mm × 20.8 mm in the right carotid space (Figure 1a, 1b).

Complete surgical resection of the tumor was performed (Figure 2 and 3), without intra or postoperative complications or postoperative neurological deficit, discharging the patient home two days after surgery.

Discussion

Paragangliomas are neoplasms accounting for 0.6% of all head and neck tumors [5]. They affect women with an average age between 45 and 60 years, being the female to male ratio of 8:1 [6]. They are related to the parasympathetic nervous system and are found near the arteries and cranial nerves.

According to some studies, 30% of carotid paragangliomas are hereditary [7]. They have a possible involvement of the cranial nerves, and extension to the base of the skull. Tijani et al. [8] reported 12 cases of unilateral paragangliomas during 15 years treated surgically.

They are usually asymptomatic tumors. Their diagnosis is based on discovery of a pulsatile and

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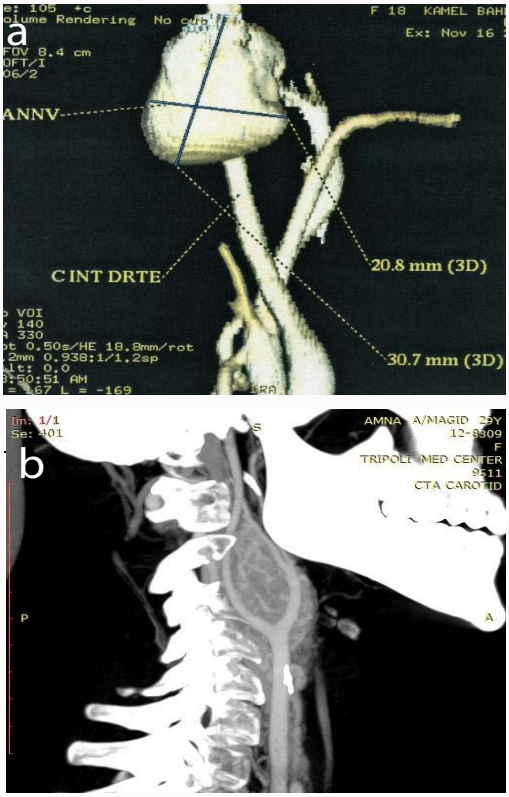

Figure 1a, 1b: CT scan of supra-aortic vessels showing mass in the right carotid bifurcation with contrast enhancement.

painless lateral neck mass at the level of the hyoid bone, and confirmed by imaging findings. Patients may present signs of catecholamines hypersecretion like headaches, palpitations, swelling, flushing, hyperglycemia, fever, nausea, pallor, hypertension, arrhythmias, stroke or even anxiety that can lead to a myocardial infarction [9,10].

Carotid paragangliomas can also be beating, transmitting the carotid pulse and producing a thrill [11]. Duplex ultrasound is the main diagnostic method since it confirms the anatomical relationship of the tumor to the carotid bifurcation and its vascularization. Magnetic resonance imaging allows definitive diagnosis [12]. 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.

Conventional angiography is important if embolization is to be attempted to minimize intraoperative bleeding [13].

Shamblin et al. [14] developed a surgical classification published in 1971, based on the relationship of the tumor with the carotid vessels, and divides the paragangliomas in three different groups based on operative notes [15]. This classification determines operative morbidity and mortality based on the characteristics and size of the paragangliomas, with a positive predictive value for surgical difficulties and postoperative complications.

Group one are easily resectable tumors with minimal adherence to vessels. Group two tumors are partially surrounded by vessels and adhered to the adventitia. Finally, group three tumors are adherent and intimate with the carotid bifurcation.

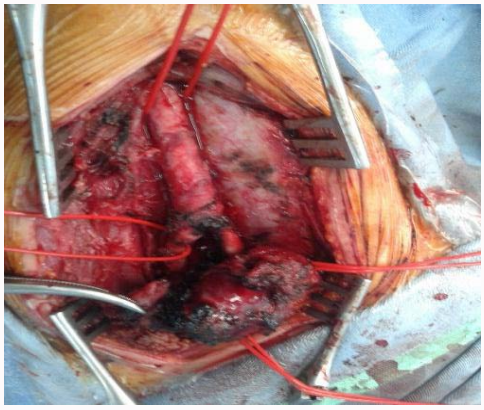

Figure 2: Intraoperative view of the resection of the tumor.

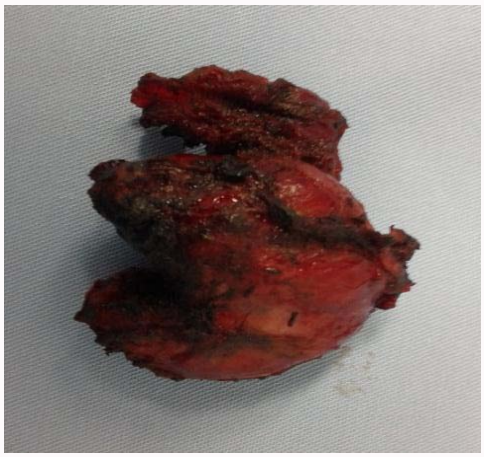

Figure 3: Photography of the tumor after resection.

The main treatment for carotid paragangliomas is surgical resection of the tumor due to the risk of regional and distant metastases [16]. Surgical resection for carotid and vagal paragangliomas is extremely technically challenging due to hypervascularity, their close relationships or compromise of the cranial nerves, and presents a high risk for carotid artery injury [17]. Preoperative planning for vascular reconstruction should always be performed, especially for larger tumors.

Tumors larger than 6 cm, which tend to splay the angle of the carotid bifurcation, are less likely to be separable from the carotid arteries without arterial injury [18]. Bougrine et al. [19] reported a 4 cm paraganglioma in the carotid bifurcation removed surgically in a 35-year-old woman.

The surgeon should be prepared to manage a carotid artery injury. Reconstruction of the internal carotid artery may be required. It can be made by a vein patch, by graft interposition which be anastomosed end-to-side or end-to-end on the distal internal carotid artery. A shunt should be available, the proximal common carotid artery and distal internal carotid artery should be exposed, and one leg should be prepared for saphenous vein harvest. Saphenous vein or prosthetic grafts can be used to reconstruct the internal carotid artery, if necessary.

The external carotid artery may be difficult to dissect free from the tumor, and may limit the exposure of the tumor. So, in many cases

the external carotid artery may need to be ligated during the resection [2].

Borghese et al. [20] reported a case of a 28-years-old woman operated for paraganglioma which infiltrated the internal carotid artery, without plane for dissection between the tumor and the arterial wall. So, they placed a shunt to preserve cerebral flow, ligated the external carotid artery, and excised the tumor. Vascular reconstruction was performed by a 6 mm bypass between the common carotid artery at the bifurcation and the internal carotid artery.

Embolization of the tumors main artery prior to surgery may help reduce bleeding associated with the removal of large tumors, facilitating their resection [21]. Tumor embolization with carotid stenting has been developed as alternative presurgical endovascular techniques that decrease tumor vascularity and facilitate resection.

A meta-analysis of Texakalidis et al. [22] showed that there were no differences in the rates of complications in patients undergoing or not preoperative embolization, even if patients who received preoperative embolization had statistically significant lower intraoperative blood loss and duration of the procedure.

Markiewicz et al. [23] published a case series of 5 patients in whom internal and common carotid artery coated stents were placed preoperatively and subsequently underwent successful sub-adventitial resection of head and neck tumors without the need for revascularization.

In patients with high risk of internal carotid artery injury, and who have tumors precluding extra-cranial carotid artery management, Westin et al. [2] suggest consideration of treatment with radiation or observation without intervention.

Also, in case of incomplete resection, or malignant paragangliomas, or surgery adjuvant radiation therapy may slow tumor growth and improve survival [24].

Lalyaa et al. [25] reported a case of good local control of an unresectable paraganglioma after external radiation of a large carotid paraganglioma involving the temporal bone.

The mortality rate in the absence of an aggressive malignancy is approximately 4% and the graft patency rate approximately 90% to 95% at 6 months, 82% to 95% at 5 years, and 73% at 13 years [26]. The risk of postoperative stroke is now very low.

Postoperative peripheral neurological morbidity, mainly involving the cranial nerves, is between 14 and 49% of early deficit, and between 6 and 23% of persistent deficit, reported in some recent studies [27].

Conclusion

Carotid paragangliomas are rare neoplasms. Surgical resection remains the only curative treatment, with low vascular morbidity. Challenges in their resection include their close relationship to cranial nerves, involvement of the carotid artery, and significant tumor vascularity. So, early resection is necessary to avoid major surgery-related complication and/or the need for carotid artery replacement.

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