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Bilateral carotid body tumor resection in a female patient☆

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ABSTRACT

INTRODUCTION: Carotid body tumors also called carotid paragangliomas are rare neuroendocrine neoplasms derived from neural crest cells, approximately 3% of all paragangliomas occur in the head and neck area (Xiao and She, 2015); although they represent 65% of the head and neck paragangliomas (Georgiadis et al., 2008).

PRESENTATION OF CASE: We present the therapeutic management of a 65-year-old woman with bilateral carotid body tumors. The patient presented to medical clinic for unrelated signs and symptoms of weight loss, dyspepsia, and epigastric pain. Physical examination showed bilateral non-tender neck masses for which imaging studies were ordered resulting in the diagnosis of bilateral carotid tumor. Surgical resection was staged with one week of distance between each tumor resection.

DISCUSSION: Carotid Body Tumors can arise from the paraganglia located within the adventitia of the medial aspect of the carotid bifurcation.

Resection is the only curative treatment. Carotid body tumors resection represents a special challenge due to potential neurovascular complications.

CONCLUSIONS: Surgical resection of carotid body tumors represents a special challenge to the surgeon because of the complex anatomical location of the tumor, including close relationship with the cranial nerves, involvement of the carotid vessels and large vascularization of the tumor. With the advance of diagnosis and improvement in surgical techniques as well as the understanding of biological behavior of tumors, surgical treatment has become a safer alternative for treating these tumors.

1. Background

Carotid body tumors are neuroendocrine neoplasms, derived embryonically from the neural crest cells of the autonomic nervous system, forming a group of neoplasms called paragangliomas. Parasympathetic paragangliomas are rare, with a prevalence of 1–2 per 100,000 population representing only 0.012% of all body tumors [1]. Approximately 3% of all paragangliomas occur in the head and neck area, the most anatomically prevalent sites found within the head and neck region are the carotid body, jugular body, the vagus nerve and along the glossopharyngeal nerve and its tympanic branch [1].

Carotid body tumors represent approximately 65% of head and neck paragangliomas [2]; about 5% of carotid body tumors are bilateral [3].

There are 3 different types of carotid body tumors described in the literature, Sporadic, Familial and Hyperplastic.

The most common type is the sporadic form, representing approximately 85% of carotid body tumors. The mean age of onset is reported to be 45 years.

The familial type can occur in 10–50% of the cases; in the familial group the age of onset is typically younger, being in the second to fourth decade, bilateral carotid body tumors are also related to the familial type, being found in 30–40% of the cases compared with 3–4% of the sporadic type [4].

The hyperplastic type is related in patients with chronic hypoxia, which includes, patients who have chronic obstructive pulmonary disease or cyanotic heart disease. Patients living at a high altitude (>5000 feet above sea level) are also related to the hyperplastic type [5].

Surgery is the only curative treatment for this type of tumors, being a special challenge to the surgeon due to the highly complex anatomic area of work, with vital structures being at risk of injury.

2. Case report

A 65-year-old woman presented to the clinic with history of weight loss, epigastric pain, and some degree of dyspepsia. The patient denied any history of hoarseness, dysphagia or voice changes. Past medical history and family history were unremarkable. On physical examination, the only pertinent finding was

☆ This work has been reported in line with the SCARE criteria (Agha et al., 2016 [20]).

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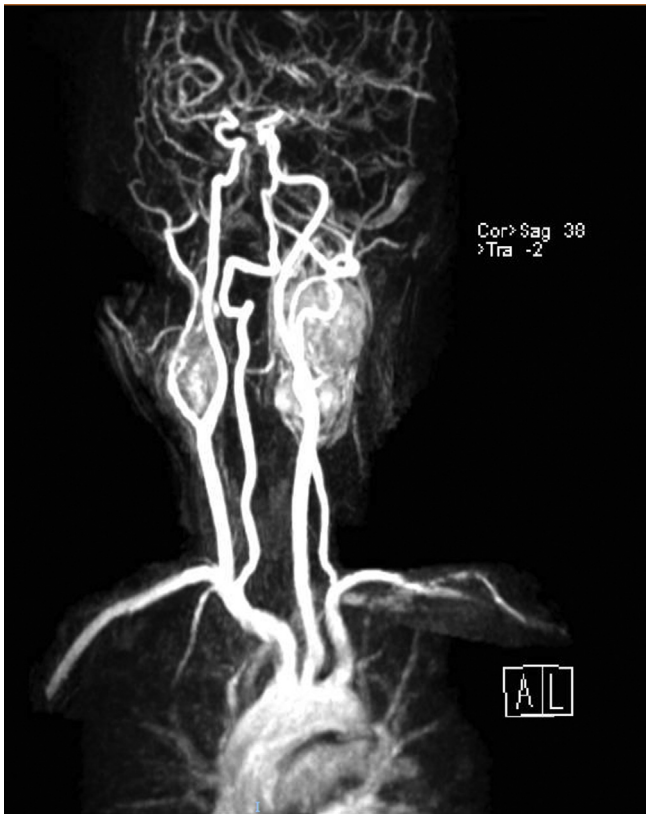

Fig. 1. Magnetic resonance angiography showing bilateral carotid body tumors at the level of the carotid bifurcation.

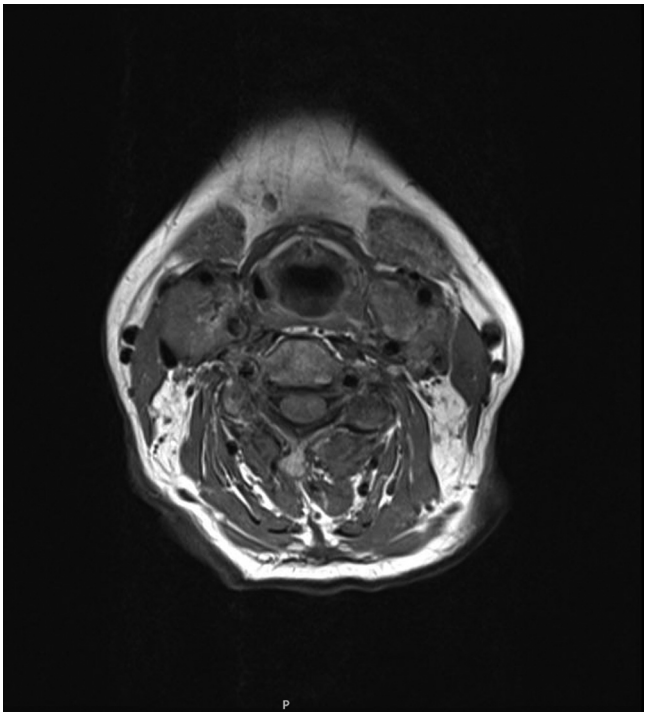

Fig. 2. Axial T-1 weighted MR image showing bilateral carotid body tumors; both with complete encasement of the ICA and ECA; Shamblin type III.

related to the neck, where on the right anterior triangle, a 4.0 × 3 cm non-tender, rubbery mass could be palpated over the common carotid artery. The mass was freely movable horizontally but not vertically (Fontaine sign). The left side of the neck was unremarkable. Neurological and cardiovascular examinations were intact.

Thyroid disease was suspected, for which general laboratory studies were sent, including thyroid function tests and neck ultrasound, the laboratory findings were unremarkable except for an elevation of the C- reactive protein; The ultrasound showed a single nodular cyst of the thyroid with benign characteristics, it also showed bilateral neck masses at the carotid bifurcation.

Further imaging evaluation was ordered, magnetic resonance imaging (MRI) and MR angiography showed the presence of two multilobulated, vascular masses located in both carotid spaces, anterior to the sternocleidomastoid muscle, at the level of the carotid bifurcation in T1 weighted images, with the characteristic heterogenic image of "salt and pepper" appearance (Fig. 1). Both lesions encased the external and internal carotid arteries giving a Shamblin type III classification (Fig. 2). The mass dimensions on the left carotid artery were 5.9 × 3.9 × 3.1 cm (Fig. 3A); on the right carotid artery the mass dimension were 2.3 × 3.0 × 2.6 cm (Fig. 3B). On the MRA sequence, we identify the feeder vessels in both lesions, which apparently depended on the ascending pharyngeal artery. These findings were consistent with bilateral carotid body tumors.

We recommend resecting the largest tumor first based on the reviewed literature. Therefore, the patient underwent surgical resection of the left-side tumor first. A left lateral cervical incision was made parallel to the anterior border of sternocleidomastoid muscle; careful dissection to expose the carotid vessels was critical to preserve all the surrounding structures (Fig. 4A). Proximal and distal control of the common carotid artery, internal carotid artery, and external carotid artery were performed before tumor resection. The inferior pole of the tumor was easily resected from the carotid arteries; we faced a special challenge with the resection of the superior pole of the tumor due to the extension of the tumor into the base of the skull, the Vagus nerve was identified at both the superior and inferior poles of the mass. All feeding vessels were ligated. The tumor was completely removed (Fig. 4B).

One week after the left tumor resection the patient had a second intervention to resect the right carotid body tumor, the same principles of the first intervention applied for this procedure. A right lateral cervical incision was made parallel to the anterior border of sternocleidomastoid muscle; followed careful dissection to expose the carotid vessels, proximal and distal control of the common carotid artery, internal carotid artery, and external carotid artery were performed before tumor resection. The right tumor was resected without any complications. All feeding vessels were ligated. The tumor was completely removed.

Both carotid body tumors were sent to pathology for analysis and final diagnosis (Fig. 5).

The patient presented post-operatively with mild dysphagia, voice changes and tongue deviation; nerve injury was suspected, therefore speech therapy was ordered. After a few sessions of therapy the patient dysphagia, voice changes, and tongue deviation improved.

3. Discussion

The carotid body originates from the embryological third branchial arch and from neuroectodermal derived neural crest lineage. The normal carotid body is ellipsoid, red-brown formation, located in the adventitia or periadventitial tissue at the bifurcation of the common carotid artery [6]. The healthy gland measures 3–5 mm in diameter and weighs less than 15 mg on average. The carotid body is mainly innervated by the Hering nerve, branch of the glossopharyngeal nerve, although many other important nerves pass in close proximity to the carotid body. The carotid body is highly vascular and receives its blood supply from feeder vessels running through the Mayer ligaments, predominantly from