CASE REPORT

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Carotid Body Tumor Excision in a Limited Resource Setting

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ABSTRACT

The management of carotid body tumors has been well described with the assumption that significant diagnostic and surgical resources are readily at hand. For most patients with these tumors, however, surgical resources remain limited. In this case study, we describe the successful surgical management of a carotid body tumor at Rwanda's University Teaching Hospital despite the lack of certain resources.

Keywords: Carotid Body Tumors; Paragangliomas; Chemoreceptors; Catecholamines; limited resource settings

INTRODUCTION

Carotid body tumors (CBTs) are rare, chemoreceptor tumors arising from the paraganglion cells of the carotid body [1]. Surgical excision is the treatment of choice, however due to their vascular nature, proximity to critical neurovascular structures, and potential morbidity, CBT resection proves to be technically challenging [2]. In limited resource settings, these challenges are multiplied. We report the successful resection of a carotid body tumor at Rwanda's Central University Teaching Hospital in Kigali.

CASE REPORT

A 77-year-old healthy Rwandan woman presented to the national referral teaching hospital with a 2-year history of a progressively enlarging, painless neck mass. Upon examination, a 3cm, firm, pulsatile mass was found. The mass was mobile in the lateral direction but had limited mobility in a cranial-caudal direction (Fontaine Sign). A contrast CT (Figure 1) revealed an enhancing mass at the carotid bifurcation splaying the internal and external carotid arteries (Lyre Sign). Diagnosis was made as a Shamblin II carotid body tumor. Digital subtraction angiography or MRA were not available, nor were urinary VMAs, serum catecholamines, preoperative embolization, or balloon test occlusion. External beam radiation has been shown to arrest CBT growth and is a treatment option for the non-surgical candidate [3], however no radiation facilities existed in the country at the time.

Additionally, there is no vascular surgeon in the country and the patient had no means to travel, therefore after thoroughly detailing the risks of surgery, the patient opted to proceed with surgical excision.

The surgery was technically challenging. A generous transverse incision with a descending limb (Y incision) was made and skin flaps were elevated in the subcutaneous plane. The anterior border of the sternocleidomastoid muscle was identified and retracted laterally. The internal carotid artery was identified in level 3 of the neck and skeletonized in an inferior to superior direction. A makeshift irrigating bipolar (Figure 2) was constructed to limit potential injury to the carotid during dissection. A standard bipolar was used, and then a syringe with saline for irrigation was used concurrently to decrease heat conduction to nearby critical nerves and vessels during dissection over the carotid artery. The vagus nerve and internal jugular veins were isolated and protected during their dissection off the carotid (Figure 3). The external carotid artery was ligated to improve visualization for dissection of the tumor off the internal carotid, which warranted greater care to avoid potential neurovascular incident. Pediatric urinary catheters were used instead of vessel loops. The tumor was dissected successfully off of the pulsating carotid bifurcation without injury to surrounding structures or rupture of the carotid artery itself. The patient recovered without any cranial neuropathies or neurologic sequelae and has been followed for 1 year after surgery without evidence of recurrence.

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Figure 2: Makeshift irrigating bipolar



Figure 3: Surgical dissection



Figure 4: Histology



DISCUSSION

The carotid body is a nest of chemoreceptors that helps the body adapt respirations to changes in oxygen levels. CBTs are rare neoplasms arising from the neural crest ectoderm within the adventitia of the carotid artery.

Histologically they are characterized by a Zellballen appearance: polygonal or spindle cells arranged in nests, surrounded by fibrovascular stroma (Figure 4). While the majority are benign, CBTs can be malignant and metastasize [2]. Eighty five percent of CBTs are sporadic, however, 10% are of a familial form, in which case the incidence of bilateral tumors is 30%[4]. While the cause of CBTs remains unknown, they are associated with chronic hypoxia (e.g. high elevation living or COPD) [5]. The Shamblin classification system divides CBTs into three types: Shamblin I tumors which are localized, Shamblin II tumors which are partially wrapped around the carotid, and Shamblin III tumors which are completely wrapped around the carotid [6]. Our case represents a Shamblin II CBT.

Since surgery remains out of reach for the greater part of the world's population [7], it stands to reason most patients presenting with CBTs do so in limited resource settings. While the literature is replete with management of CBTs in high resource settings, there are few reports detailing their management in limited resource settings [8,9].

Preoperative workup and management differed from what is considered the gold standard when managing patients with CBTs. Diagnosis was limited to contrast CT, which in conjunction with physical exam proved to be adequate. More comprehensive preoperative workup, including urinary vanillylmandelic acid (VMA) and serum catecholamines was not available, which may identify the 5% of active lesions [10]. Preoperative embolization for CBTs has been shown to decrease blood loss and facilitate tumor removal [2], yet this too remained unavailable. Despite this the tumor was resected with minimal blood loss (50cc).

Special attention was given to patient consent, as the patient was made aware of the risks associated with this procedure. The patient was detailed on the risk of intracranial bleeding, stroke, and death, along with risks of non-operative management. Due to the patients age, radiation was discussed, but not feasible at the time.

A Shamblin II CBT treatment, though technically demanding, was managed with minimal diagnostic equipment and instrumentation. CBTs are surgical challenges that while not without risk, are possible to manage in limited resource settings. To the authors knowledge, this is the first carotid body tumor successfully excised at Rwanda's University Teaching Hospital.

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