CLINICAL REPORT

A Hybrid Approach Towards Successful Resection of a Huge Carotid Body Paraganglioma Using Coil Embolization and Traditional Surgical Techniques

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Abstract Carotid Body Paraganglioma (CBPGL), is a type of neuroendocrine tumor that should be managed promptly due to their malignant potential and locally aggressive nature making resection at a later stage difficult. The objective of this case report is to explore the benefit of coil embolization and describe the surgical techniques employed in successful resection of a huge CBPGL.

Keywords Carotid body tumor · Paraganglioma · Micro coil embolization · Shambli III · Extra adrenal

Introduction

Paragangliomas (PGLs) are a group of neuroendocrine tumors derived from chromaffin cells of the neuroectoderm, paravertebral sympathetic or parasympathetic chain ganglia [1]. Approximately 10% of PGLs are extra-adrenal, 70% of which are found in the Head and Neck region

[1, 2]. The Carotid body is the site of origin in 60% cases of head and neck PGLs and are very vascular tumors. Very few studies have explored the benefits of using microcoils over traditional embolization techniques for this indication. The objective of this case report is to explore the role of coil embolization and describe the surgical techniques employed in safe resection of CBPGL, without the need for external carotid artery (ECA) or internal carotid artery (ICA) ligation, stenting, repair or reconstruction.

Case Report

A 40-year-old Indian woman presented with an asymptomatic, pulsatile, neck swelling, progressively increasing in size for 13 years. Family history was negative. On examination a 6 × 4 cm non-tender pulsatile mass anterior to the left sternocleidomastoid muscle extending from, left angle of the mandible to the level of cricoid cartilage not crossing the midline was palpable. *Fontaine's sign* was positive. Cranial nerve examination revealed an afferent sensory deficit in the gag reflex pathway.

Preoperative workup including a 24 h urine VMA estimation was normal. CT Angiography revealed an intensely enhancing mass of size 8.5 × 5.1 cm in the left carotid sheath at the level of left carotid bifurcation splaying the external and internal carotid arteries (*Lyre's sign*) with multiple dilated feeding arteries from the left ECA and multiple draining veins into the left internal and external jugular veins. The tumor completely encased both ECA and ICA (Fig. 1). A halo around the carotid arteries suggested a free periadventitial plane of dissection. There was no evidence of vascular infiltration, Aneurysms or Arteriovenous malformation. All vessels showed normal luminal diameter without flow-limiting plaques or stenosis. There

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Fig. 1 CT Angiography with 3D reconstruction. **a** anterior view, **b** posterior view, **c** lateral view

was no evidence of a contralateral tumor. As per the surgical classification described by Shamblin et al. [3] the tumor was classified into category III.

Preoperative embolization of the Left ECA was done using micro coils, 36 h prior to surgery. Three fiber coated stainless steel coils of 0.035 inch thickness; 6 mm diameter; 6 cm(1) and 5 cm(2) lengths were used. Post-embolization tumor blush was absent and there were no

neurovascular complications following the procedure (Fig. 2).

Surgery was performed under General Anaesthesia with the patient in supine position, neck extended and rotated to the right with head end elevation to minimize venous bleeding. Under aseptic precautions, 2% Xylocaine with adrenaline was infiltrated along a modified Schobinger's incision line. Subplatysmal flap elevation and dissection

Fig. 2 Digital subtraction Angiography and Coil embolization. **a** tumor blush pre-embolization, **b, c** coil embolization being carried out, **d** post coil embolization with absence of tumor blush

exposed the tumor pseudocapsule which was tightly adherent to the Common Carotid Artery(CCA), ECA and ICA. Meticulous dissection of the lower border of the tumor was done to identify the “*White Line*” which marks the Peri adventitial plane [4]. Periadventitial dissection was carried out in a caudocranial direction from the CCA towards the carotid bifurcation with the help of bipolar cautery under microscopic magnification. Proximal control of the CCA was obtained just above the omohyoid muscle. The distal ECA was dissected free after ligating all the feeders. The tumor seemingly encased the ICA; however, it was successfully taken off the ICA, by retro-carotid dissection in the periadventitial plane in a caudocranial direction with the use of a fine-tipped right-angle instrument between the carotid sheath and the ICA adventitia, obviating the need for arterial resection or replacement. Major cranial nerves including the X and XII nerves were not encountered during dissection. The carotid artery system in the neck was covered using a partial sternocleidomastoid muscle rotation flap.

The patient recovered without any neurovascular complications. Histopathological examination of the tumor showed features typical of CBPGL such as *Zellballen cell nests* and Immunohistochemistry was positive for *Synaptophysin*, *Chromogranin* and *S100* [5] (Fig. 3). Postoperative MRI was negative for residual tumor or thrombosis of the Carotid artery system.

Discussion

Surgical resection is the treatment of choice for CBPGL and it involves a multidisciplinary team to achieve maximum results [6–9]. Excision was first carried out in 1889 by *Sir Albert* [10] and in 1940 *Sir Gordon Taylor* described the Peri adventitial dissection technique [9]. In 1980s devascularisation of the tumor by ligating feeding vessels from the ECA gained popularity and first pre-op embolization was done in 1983.

Previous studies have reported various benefits of embolization such as reduction in tumor size, intra-operative blood loss, operative time, reduced complications and hospital stay [11–13]. The unique advantage of Microcoil embolization as we have experienced, is the ability to deploy the coil selectively, precisely and proximal to the feeding vessel thereby preventing complications of unwarranted distal embolization [13, 14]. Intraoperatively an added advantage is the ability to palpate the coil and it acts as a surgical marker in identifying the feeding vessels [14]. A recent advancement in the endovascular approach to CBPGLs is the use of covered stents for vascular exclusion and studies have shown promising results. Limitations of covered stents are its high cost, unavailability in our country, lifelong antiplatelet or anticoagulation therapy and followup to rule out stent thrombosis/displacement [15–18].

Fig. 3 Histopathological examination and Immunohistochemistry. **a** hematoxylin and Eosin staining showing Zellballen cell nests, **b** chromogranin positivity, **c** S100 positivity of sustentacular cells, **d** synaptophysin positivity

Studies have demonstrated shorter procedure time and hospital stays with retrograde caudo-cranial dissection technique when compared to the cranio-caudal approach [19]. In our case, we used both retrograde caudo cranial and retro carotid dissection in a periadventitial plane. Dissection adhering to the tumor pseudocapsule helps prevent any neurological complication [20]. A relative contraindication to surgery is a distance of less than 2 cm between the upper border of the tumor and the skull base [21, 22].

Shamblin 3 CBPGL as it is in our case, is a major challenge due to carotid encasement, cranial nerve involvement that may lead to neurovascular complications like intra op haemorrhage, stroke and cranial nerve injuries [23]. In our case, the patient recovered without any such complications. Post-embolization tumor size reduction was not appreciated but intraoperative blood loss was minimal amounting to approximately 180 ml and procedure time was 150 min.

In conclusion, a hybrid approach employing endovascular micro coiling of the feeding vessels and meticulous surgical dissection techniques under the microscope can be successfully used for safe resection of advanced CBPGL.

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