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Surgical Resection of Painless Carotid Body Tumour without Preoperative Embolization: A Case Report and Review of Literature

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Abstract

Paragangliomas are rare tumors representing a therapeutic challenge. We present a case report of surgical resection of carotid body tumor without preoperative embolization. Our therapeutic attitude is based on controversial benefits of the embolization for those tumors. The major indication for the preoperative embolization is to reduce intraoperative blood loss, but this benefit is not demonstrated. Also, because the relative rarity of this tumor, the confounding factors relative to the surgeon and radiologist experience, no randomized trial can be performed. So, our case report can be useful to participate to increase the number of reported cases, and define the therapeutic approach for this rare tumor.

Keywords: Carotid body tumor; Shamblin; Embolization; Surgical resection

Introduction

Carotid body tumor (CBT) is a rare hypervascular usually benign neoplasm of neural crest origin arising in paraganglial cells of the carotid bifurcation. Usually is found in the region of the head and neck. The clinical presentation can be a cervical mass with lower cranial nerve palsies [1-6].

This tumor is found in any age group but is most common in the 3^{rd} and 4^{th} decades of life with minimal symptoms or signs [2]->1 -4. Those tumors are rare (prevalence of 1– 2 per 100,000), usually benign and only rarely catecholamine-secreting (1–5% of the cases) [5].

Paraganglia is a chemoreceptor and includes the glomus jugular and glomus tympanicum. The tumors can also arise along the nerves such as the vagal or laryngeal nerves [2]. They grow slowly and rarely metastasize [3]. Metachronous mediastinal and abdominal tumors may occur [4].

The vascularization is assured by the branches from external carotid artery or glomic artery arising at the bifurcation of the common carotid artery. There is also the possibility of a supplementary blood supply from an internal carotid artery (ICA) [1].

The surgery is the main treatment option for this tumor [2]. Unfortunately, resection can prove difficult and be fraught with significant complications. Surgical resection of nonfunctional tumors will remove the effect of the chemoreflex on the sympathetic nervous system but there is also sympathetic down-regulation by damage of the baroreflex [5].

Case Report

A 69 years-old male presents to his family doctor for a painless mass in the submandibular region that appears in the last years but who worries the patient and his family. His doctor addressed the patient to our hospitals for a vascular consultation. The patient presents type 1 diabetes, systemic hypertension and tachycardia. The surgical antecedents are total knee prosthesis, Nissen and appendectomy. No allergies were recorded. The clinical exam found the mass in the left submandibular region. The formal suspicion of tumor was posed and the patient was programmed for an angio-CT of the supra-aortic vessels. The angio-CT found a hypervascular mass who measured 30 \times 31 mm placed on the carotid bifurcation which is surrounded (in an

angle of 240°, Shamblin IIIa). A radiological suspicion of glomic tumor/paraganglia was posed.

The patient was programmed for pre-operative exams. Laboratory data showed only an anemia (Hb 11.9 g/dl) and denutrition (total proteins 54 g/l. The chest X-ray was normal and also the spirometry and plethysmography. The electrocardiogram found a left cardiac hypertrophy but no ischemic or sequelae modifications. The echocardiography found a mass of 6×9 mm on the aortic valve (suspicion of fibroelastoma, excluded after trans-esophageal echography) and a hyperkinetic function with hypertrophy of the left ventricular wall. The myocardial scintigraphy found a diminution of the vascular reserve with no consequences, in the inferior and posterior myocardial region.

Surgery

The surgical resection of the tumor with the conservation of the vessels was realized. The commune carotide artery (CCA) was dissected and an important ganglionary group was identified and sent for pathological exam. The internal and external carotid were identified on their distal portions and the small vascular branches around the tumor were ligated. The mass was dissected and removed under clampage (10 minutes) because of the high risk of bleeding. No modifications on the cerebral monitoring were recorded. Blood loss was negligible (about 50 ml). The total duration of operation was 196 minutes (Figures 1-3).

Post-operative course

The patient's Post-operative course was marked by a modification of the voice, which became weaker in intensity and dizzier. The ORL exam found left vocal cord paralysis with no movement during phonation but with a good compensation by the other vocal cord. The patient resumed

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Figure 1: Coronal (a) and axial (b) views of the tumor at the CT-scan.



Figure 2: Intra-operative image before (a) and after (b) tumor resection.



Figure 3: Surgical resection of the tumor.

oral intake on the first Post-operative day. He was discharged from the hospital on the 5th post-operative day.

Pathological results

The histopathologic exam shows lymph nodes hyperplasia without malignancy and paraganglioma with complete resection.

Discussion

The most common clinical presentation and usually the reason for what the patients are presenting to the hospital is the finding of painless mass in the lateral neck region [7]. That was the case of our patient. The surgery was complicated by vocal cord palsy. The recurrent laryngeal nerve was correctly visualized but involved in the mass. So we have not the choice to sacrifice the nerve to avoid any remaining mass tissue [8-15].

The carotid body is a highly vascular structure measuring on average 7 mm \times 4 mm \times 2 mm, its location is variable and is usually located in the adventitia of the posteromedial aspect of the carotid bifurcation [16-25]. The carotid body involves a blood volume that is 10 times more than the heart myocardium and 25 times more than brain tissue at the same size due to its rich vascular structure [26-30]. The embryologic origin is from the third branchial arch and contains homeostatic chemoreceptor cells- the sensory nerve endings of the carotid sinus nerve penetrate the clusters to synapse with chemoreceptor cells [31-37]. The carotid body is made up of two types of cells, called glomus cells: glomus type I cells derived from the neural crest, and glomus type II cells and act as supporting cells [38-47]. The glomus or type I cells of the CB are the transducers of hypoxic stimuli, and relay chemosensory information to the brainstem via neurotransmitter release at synaptic contacts with afferent terminals of the carotid sinus nerve.

The anatomical details of the vascularization and the rapports with the main vessels - CCA, ECA, ICA and their branches as well as the presence of the intracranial anastomoses is critical for the safe embolization and for the surgical strategy [22]. Vascular supply is achieved not only by the occipito-pharyngeal trunk but also by the external carotid, ascendant pharyngeal and occipital arteries via one to more than three side branches [8,18,40,47].

Clinical presentation

The most common clinical presentation and usually the reason for what the patients are presenting to the hospital is the finding of painless mass in the lateral neck region [13]. The patient may present symptoms correlated with the cranial nerve involvement. There is a close anatomical relationship with cranial nerves X-XII and the symptomatology can be represented by dysphagia, choking, or hoarseness [8]. Hoarseness is correlated with laryngeal or vagus nerve involvement, dysphagia with glossopharyngeal and hypoglossal nerves involvement. Horner's syndrome with invasion or compression of the cervical sympathetic chain, and syncope, which may be due to CS or ICA compression [47].

Differential diagnosis

Although carotid body tumor is a rare neoplasm, it should always be considered in differential diagnosis of lateral neck masses [9]. The differential diagnosis is very important for planning the surgery, but also for the post-operative treatment. Conventional radiologic methods may result in discrepancies in differential diagnosis and only the pathological exam can clarify the diagnosis [19]. The differential diagnosis of head and neck depends on the location of the lesion. Papillary thyroid cancer has priority because to its location, its higher incidence among women [16]. For the jugulotympanic area the differential diagnosis include middle ear adenoma, meningioma, and schwannoma, among others. In the histologic differentiation the immunohistochemistry plays an important role. The differential diagnosis include also neuroendocrine tumors such as medullary thyroid carcinoma and neuroendocrine carcinoma or the parotid gland tumors as pleomorphic adenoma and Warthin's tumor [8,9], oncocytoma, basal cell adenoma [11]. Hyalinizing trabecular adenoma of the thyroid gland should also be considered [34]. Inflammatory myofibroblastic tumor rarely occurs whereas syncope is infrequently associated with neck mass [17].

Diagnosis

In 1991, Shamblin classified CBTs into 3 categories based on their relationship to the internal and external carotid arteries. Class I tumors are localized between the internal and external carotid arteries and easy to resect; class II tumors are adherent to or partially surrounding the carotid arteries; and class III encases 1 or both carotid arteries [18,21].

The metastatic involvement of neck lymph nodes can be confirmed with the fine needle aspiration biopsy (FNAB) but for the carotid tumors the risks of hemorrhage into the tumor or the damage to the carotid vessel wall have potentially life-threatening implications. The diagnosis can be safely made using the imaging procedures [7].

Familial forms are in relation with the presence of a specific gene multifocal lesions, distant metastases are more likely to occur in familial forms compared with non-familial cases [16].

Color Doppler Sonography and Digital Subtraction Angiography (DSA) play a very important role in confirmation of the clinical diagnosis. DSA instead is regarded as the gold standard for the final diagnosis but it cannot provide information such as intracranial and extracranial blood circulation. The CT-scan and especially 3D-reconstructed image can help to visualize better the relationship of the tumor with the surrounding tissues [33]. The signs of malignancy are suggested by the surrounding of the ECA and ICA, by the infiltration of the tumor up to the base of the skull and by the presence of enlarged and significantly regional lymph nodes and multiple pulmonary and hepatic lesions, signs of metastasis [16].

Treatment options

There is no consensus on the impact of preoperative embolization (POE) on the surgical outcomes of carotid body tumor resections [27].

The use of preoperative embolization has been advocated to aid in surgical resection by decreasing intraoperative blood loss and operative time [!!!]. Other authors have assessed the implications of preoperative embolization on neurovascular complications. So there is no definitive consensus on the impact of preoperative embolization on carotid body paraganglioma management.

In addition, most reports of CBT are small case series, making decisions on optimal treatment strategies difficult.

There are some authors who are not sure about the benefits the embolization and about the advantage in reducing intraoperative blood loss, ease of dissection and in reducing the duration of the operation and more the risk of vascular rupture rate can increase during dissection [26].

Many papers acknowledged the importance of Shamblin classification and tumor size but then did not stratify the data for EBL and operative time based on this information. Power et al. reported a correlation of the level of difficulty operative time, blood loss, and nerve injuries to higher Shamblin-class tumors but recognized the subjectivity of this system [!!!!]. Yet, Ozay et al. compared Shamblin I and II with Shamblin III and found increased blood loss, cranial nerve injury, and hospital stay for Shamblin III tumors, but their experience was limited to only 14 patients [18]. Lim et al. also compared Shamblin I and II with Shamblin III tumors in 13 patients and found increased operative time, blood loss, and cranial nerve deficit in patients with Shamblin III tumors.

There are multiple choices for POE that includes re-absorbable material (shredded gelatin sponge) but is preferred non-reabsorbable agents such as Ethylene vinyl alcohol (EVOH) or PHIL[™] (injectable precipitating hydrophobic liquid), acrylic glue, endovascular coils [13]. EVOH may have better tumor penetration because it can be injected slowly for precise delivery into the feeding vessels compared to acrylic glue, which polymerize immediately on contact with blood [31].

If a malignant behaviour of the tumor is recorded then the radiation therapy may also be used [25]. Radiation therapy is used following an incomplete surgical resection or before surgical resection of large lesions. Radiation therapy results in tissue fibrosis that may possibly hamper the surgery and elongate the healing process and also in palliative treatment of patients [24,29,30].

Surgical excision remains the only curative treatment for this type of tumor, and the recurrence is rare [10,12]. The tumor removal is challenging because of location near vascular structures and cranial nerves. The excision should be as conservative as possible in preserving the main vessels and the nerves. The extensive resections must be limited to the well selected patients with loco-regional invasion to minimize the risk of complications [13].

To perform a safe resection a complete preoperative evaluation is required [14]. The size and extension of the tumor should be determined by preoperative imaging for the correct planning of surgical procedure. It should be taken into consideration that the rate of Postoperative cranial nerve damage is high [9]. Early surgical excision has been recommended to reduce the risk of perioperative complications and malignancy [21]. Some authors suggest that ligation of the external carotid artery provides a safe opportunity for operation in cases with larger tumors that are densely adherent to the carotid artery [45]. The ligation of the ICA must be avoided if possible because of the 66% prevalence of stroke [46]. The surgical technique must perform precise anatomic dissection and vascular control before the tumor excision. The dissection is carried out along the arterial subadventitial plane to allow for complete local tumor excision and the preservation of the critical vascular structures [20]. Adjacent lymph node dissection is recommended [18]. The major morbidity is related to the intraoperatively cranial nerve lesion. The risk of cranial nerve palsy as a complication of CBT surgery has been reported to range from 10% to 40% [5,15,41]. The use of clamping of all carotid arteries with placement of shunt and the immediate replacement or repair of damaged vessel, can reduce clearly massive bleeding, cerebrovascular accidents and overall morbidity [44].

Post-operative complications

Post-operative neurovascular complications are the most frequent and can comprise: stroke (after ICA ligation or transient ischemic attacks) and peripheral neurologic deficit. The nerves most often involved are the vagus and hypoglossal followed by cervical sympathetic, glossopharyngeal and superior laryngeal [18]. The other nerves who can be involved are mandibular branch of the facial nerve, the pharyngeal branch and the glossopharyngeal. [43]. Non-neurological Post-operative complications can be generally represented by local (abscess and hematomas), vascular (bypass thrombosis) or general (pulmonary embolism, pneumonia or non-controlled hypertension) [18,44]. Shamblin III had a high risk of Post-operative neurovascular complications. The early detection and prompt surgical attitude will decrease surgical morbidity [42].

Our attitude

We decided to not perform a pre-operatively embolization for these patient for multiple reasons. In the first place, at the pre-operatively Citation: Cimpean S, Belhaj A, Rondelet B (2017) Surgical Resection of Painless Carotid Body Tumour without Preoperative Embolization: A Case Report and Review of Literature. J Clin Case Rep 7: 915. doi: 10.4172/2165-7920.1000915

imaging, we didn't identify clearly the vessel that emerged to the tumor. Also we consider the risk of emboli that were potentially able to migrate in the ICA with the neurological consequence as higher. In addition to surgical complications, complications may arise from the embolization procedure itself, and pre-operatory selective embolization is still controversial. During the operation no visible nerve injuries were observed but in the post-operative period the patient presented a vocal cord paralysis on the left side, the operation side. We encounter some difficulties during the operation because of the developed collateral circulation around the mass and a careful dissection was performed due to the increase risk of bleeding. After the tumor and the vessels were liberated from the surrounding tissue we preferred to excise the mass after the clampage of the ICA, ECA and CCA because we were not sure that there is a cleavage zone between the mass and the vessels and no risks were taken. The vessel conservation is important and the integrity of the vessel was preserved but if the tumor invade the arterial wall, no hesitation must be have for resection and reconstruction.

Conclusion

Carotid paraganglioma is a rare tumor of neck, and no randomized trial, given the relative rarity of these tumors, can be performed. Therefore, future research should focus on standardization of reporting as well as performance of prospective comparative studies with or without preoperative embolization.

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