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Case Report

Metastatic carotid body tumor: a rare condition

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ABSTRACT

Carotid body tumors (CBTs), or carotid glomus tumors, are rare, slow-growing benign neoplasms arising from the carotid body chemoreceptor. Metastatic disease is uncommon and poorly understood. We report the case of a 53-year-old male with a Shamblin type IIIB CBT, treated with a hybrid approach involving stent placement followed by en bloc resection. He remained disease-free for 116 months before developing regional recurrence along with pulmonary, bone, and brain metastases, which were managed with palliative radiotherapy. The patient died three years after the onset of the first metastases. This case highlights that CBT metastasis may arise more than a decade after initial treatment, underscoring the importance of long-term follow-up and multidisciplinary management.

Keywords: Metastatic paraganglioma, Gigantic paraganglioma, Carotid body tumors

INTRODUCTION

Carotid body tumors (CBTs), also known as carotid glomus tumors, account for nearly half of all paragangliomas. They are rare, slow-growing benign neoplasms that arise from the carotid body chemoreceptor located at the bifurcation of the common carotid artery. Approximately 2,825 cases have been reported in the literature, with notable differences in the male-to-female ratio.[1-3]

Metastatic disease is rare and poorly understood, as CBTs are histologically benign with apparently low metastatic potential.[2] It is also important to distinguish metastatic disease from multicentric disease. Diagnostic criteria should require prior exclusion of additional lesions at other sites through appropriate testing; however, this is not routinely performed. Consequently, some cases described as metastatic in the literature may have reflected multicentric disease from the outset.

The objective of this report is to describe the clinical course of a patient without prior metastatic disease who subsequently developed metastases during follow-up.

CASE REPORT

A 53-year-old male with a five-year history of progressive cervical swelling presented with a pulsatile mass in the left carotid triangle, showing a positive Fontaine's sign, suggestive of a CBT. Computed tomography revealed a Shamblin type IIIB CBT (Figure 1). A two-stage approach was used. Initially, two 5×50 mm covered stents were placed endovascularly in the left internal carotid artery, covering a 63 mm segment, with adequate perfusion confirmed by follow-up cerebral angiography. This was followed by en bloc surgical resection, including the common, internal, and external carotid arteries, as separation of the tumor up to the skull base was not feasible. The left vagus nerve was infiltrated along its entire cervical course. Estimated blood loss was 3,500 ml, and clamping time was 25 minutes. Histopathological

analysis confirmed a carotid paraganglioma measuring 14×11×5 cm, positive for synaptophysin and CD34, and negative for actin and S-100.

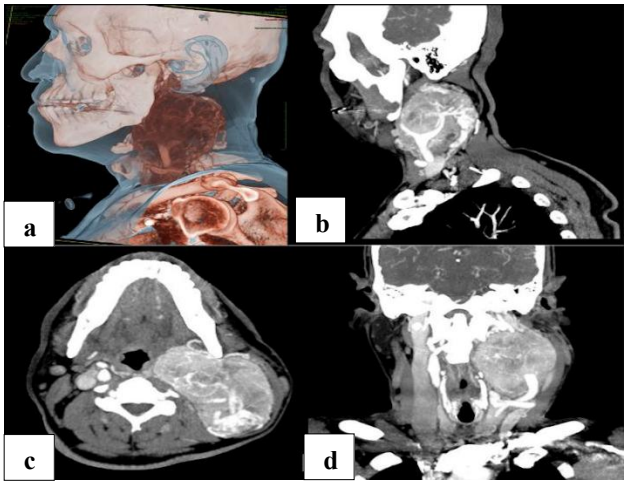

Figure 1: (a) 3D reconstruction of CT angiography showing a large left carotid space mass with intense vascularization, (b) sagittal contrast-enhanced CT demonstrating a highly vascularized mass in the left carotid space, encasing the carotid artery, (c) axial CT revealing the relationship of the tumor to the oropharyngeal airway and major vascular structures, and (d) coronal CT showing the extension of the lesion and displacement of the airway and cervical vessels.

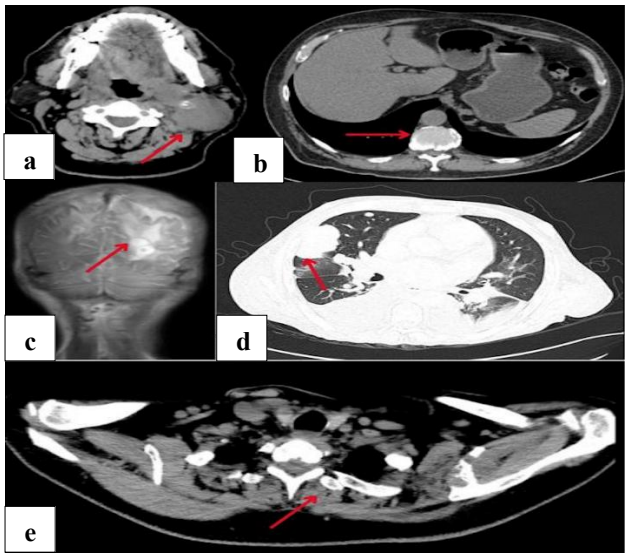

Figure 2: (a) Axial contrast-enhanced CT of the neck showing a recurrent heterogeneous mass in the left carotid space, (b) axial abdominal CT demonstrating an osteolytic lesion in the thoracic vertebral body consistent with bone metastasis, (c) axial chest CT in lung window revealing multiple bilateral pulmonary nodules compatible with metastases, (d) coronal contrast-enhanced brain MRI showing a nodular lesion with peripheral enhancement, and (e) axial contrast-enhanced CT showing a scapular metastasis.

The patient recovered satisfactorily and was discharged for follow-up without neurological complications. The patient remained free of metastatic disease for 116 months, until presenting with a painful left supraclavicular mass. CT revealed metastasis in the left scapula and multiple pulmonary nodules (Figure 2), which were confirmed by biopsy. Radiotherapy (35 Gy in 5 fractions) was administered to the neck and scapula, while pulmonary lesions were monitored. One year later, metastatic lesions developed in the thoracic and lumbar spine and were treated with 8 Gy to T4 and T8, and 20 Gy to L3 delivered in 5 fractions. One year later, progression to the brain parenchyma was observed, suggestive of secondary metastatic deposits, and whole-brain radiotherapy (20 Gy in 5 fractions) was administered. Immunotherapy was proposed but not initiated, and the patient died one year later.

DISCUSSION

CBTs are rare neoplasms arising from the carotid chemoreceptor and account for nearly half of all paragangliomas. While they generally exhibit benign histologic behavior, a small subset develops metastatic disease—a phenomenon that remains poorly understood given their low incidence and benign nature.[1] Several tumor-related factors increase morbidity and mortality. The first is a size greater than 5 cm; second, the proximity of the tumor to the skull base, which raises morbidity due to intraoperative bleeding and multiple cranial nerve injuries;[5] and third, direct vascular invasion of the carotid wall.[2] Thus, assessing the patency of the circle of Willis via cerebral angiography is crucial, as it allows greater flexibility if preservation of the internal carotid artery is not feasible. The present case encompassed the risk factors mentioned earlier: a 14×11 cm Shamblin type IIIB tumor with vascular and neural invasion, in close proximity to the skull base, resulting in extreme surgical complexity.[5-8] In anticipation of high morbidity and mortality, the approach was planned as a two-stage hybrid strategy. In the first stage, covered stents were placed in the common and internal carotid arteries, extending up to the skull base to maintain cerebral perfusion in the event of significant arterial injury and to reduce blood flow to the tumor, while excluding the external carotid artery, which provides most of the tumor vascular supply. Although preoperative tumor embolization continues to be reported, we did not perform this technique, as we consider it no longer has a role in contemporary practice.[4]

During the subsequent surgery, despite the placement of stents, tumor bleeding occurred, necessitating en bloc resection of the tumor, along with the common, internal, and external carotid arteries, and the ipsilateral vagus nerve, followed by vascular reconstruction. However, the stent provided a secure site to place a common carotid to internal carotid bypass near the skull base, allowing preservation of short-term functional integrity and facilitating postoperative recovery.

The criteria for malignancy are defined by the presence of metastatic disease, either in regional lymph nodes or distant organs. The diagnosis of metastatic disease demands strict clinical criteria, as even we have previously questioned its presence in cases lacking well-defined histological features, or in which the depth of invasion was not associated with more aggressive tumor behavior. Moreover, metastatic disease must be carefully distinguished from multicentric disease, since the latter may mimic a disseminated pattern. In this regard, it is essential to rule out synchronous primary lesions through comprehensive staging studies; however, in clinical practice this is not usually achieved. In the present case, the course of the disease ultimately allowed for a reliable characterization of its metastatic behavior.[3]

In the present case, the observed disease course (116 months) is consistent with reports describing late recurrences or metastases, highlighting the need for long-term follow-up even in patients without initial residual disease.[9] Although distant metastases are rare, they have been reported in the liver, bone, kidney, lung, breast, pancreas, retroperitoneum, and thyroid.[10] In our patient, progression to bone, lung, and brain metastases demonstrates the capacity of these tumors to manifest aggressive behavior, even after complete initial resection and prolonged follow-up. Metastasis first appeared regionally in the supraclavicular area, where treatment consisted of radiotherapy, and later progressed to multiple bone and cerebral sites, managed with temporary symptomatic control and radiotherapy. Immunotherapy was considered but could not be initiated due to clinical deterioration and the patient's decision. For systemic treatment of distant metastatic CBTs, most of the available evidence is derived from studies on paragangliomas. Due to the rarity of CBTs and their diverse clinicopathological features, the effectiveness of active systemic therapy in metastatic CBTs remains to be determined.[11]

In this context, the unfavorable prognosis was driven by the extent of metastatic disease and progression within the central nervous system. This case highlights that, although uncommon, CBT metastases can occur even more than a decade after primary treatment. Tumor size, invasion necessitating complex resections with vascular reconstruction, and the development of multiple metastases are critical determinants of prognosis. Long-term follow-up and a multidisciplinary approach are essential to optimize patient management.

CONCLUSION

Although distant metastasis of CBTs is uncommon, long-term follow-up is necessary because recurrence can occur even more than a decade after resection of the primary tumor. Factors such as large tumor size, vascular invasion, and proximity to the skull base increase surgical complexity. In the absence of specific criteria to distinguish metastatic from multicentric disease, management must be individualized; therefore, long-term follow-up and multidisciplinary care are essential to optimize prognosis.

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