Unusual spinal metastases from an adenoid cystic carcinoma of the maxillary sinus seen on a bone scan: a case report

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Abstract

Adenoid cystic carcinoma (ACC), the second most common cancer occurring in the sinonasal tract, is an aggressive malignancy that presents itself insidiously and is generally advanced when diagnosed. Current treatment modalities include surgery and irradiation. Despite advanced successful therapies, these tumours are notoriously associated with locoregional recurrences. This report presents the original case of a patient with adenoid cystic carcinoma of the maxillary sinus with an unusual clinical course and a thoracic vertebral metastases, with spinal cord compression revealed by a bone scan, occurring only nine months after stopping treatment. The aggressive nature of the tumour and the skull base invasion at the time of diagnosis probably explains the rapid onset of this unusual site of metastases.

Key words: Adenoid cystic carcinoma, bone scan, maxillary sinus tumour, radio biphosphonates, spinal metastases

Introduction

Adenoid cystic carcinoma (ACC) is a rare malignant tumour which represents less than 1% of head and neck malignancies, and 10% of salivary gland tumours [1]. ACC most commonly occurs in the major and minor salivary glands of the aerodigestive tract and skin [3]. The ACC are tumours with slow and insidious growth, often discovered at a late stage and locally evolved. The sinonasal locations of ACC have the worst prognosis. Surgery is often complicated by the importance of the local extension. Recurrence is common and late and can occur many years after the initial treatment [1]. Spinal metastasis of ACC of the maxillary sinus, have only been rarely reported in the literature. We present a case of a patient with maxillary sinus carcinoma with vertebral metastases revealed by $^{99m}$Tc-HMDP bone scan.

Case report

A 45-year-old woman developed ACC of the right maxillary sinus, which was treated by surgery and radiotherapy. The patient presented 9 months later with limb weakness, which had progressed moderately over time. Neurological examination revealed paraparesis with preserved tendon reflexes and sensation.
99mTc-hydroxyl-methylene diphosphonate (99mTc-HMDP), bone scan revealed increased uptake of the right maxillary sinus, the right orbital roof, and also heterogeneous aspect of C7-T1 vertebral bodies (Figure 1).

SPECT-CT confirmed increased uptake in the right wing of sphenoid bone with cheekbone lysis (Figure 2) together with increased uptake in C7 & T1 vertebral bodies (Figure 3). A follow-up MRI of face and neck with and without contrast injection, showed two lesions: one in the right maxillary sinus extending to the pterygopalatine fossa, and the second affecting the right wing of sphenoid bone with cheekbone lysis, and orbital and temporal soft-tissues extension (Figure 4). It also shows the

**Figure 1** Planar whole-body scan with localized image, obtained 3 hours after administration of 740 MBq of 99mTc-hydroxyl-methylene diphosphonate (99mTc-HMDP), showing increased uptake in the right maxillary sinus, the right orbital roof, with heterogeneous uptake in the region of the C7-T1 vertebral bodies.

**Figure 2** Axial SPECT-CT scan showing increased uptake of the maxillary sinus extending to involve the right wing of sphenoid bone with cheekbone lysis.
the presence of a lesion in the T1 vertebral body, with extension to the anterior paraspinal and epidural soft-tissue (Figure 5), causing a compression of the spinal cord at this level, with infiltration of the adjacent vertebral bodies.

An open surgical biopsy of spinal cord lesion was carried out with laminectomies from the 7th cervical to the 2nd thoracic vertebrae. Histopathological examination revealed a metastasis of the known ACC of the maxillary sinus in the vertebral column. Palliative local radiotherapy with chemotherapy was initiated. The patient died two months after completion of chemotherapy.

**Discussion**

Malignancies arising from the sinonasal tract are uncommon. Carcinoma of the maxillary sinus comprises 0.2% to 0.5% of all cancers.
of the head and neck [4]. According to Wang [2], squamous cell carcinomas are the most common form of maxillary sinus tumours, followed by adenoid cystic carcinoma and adenocarcinoma. The choice of optimal therapy for ACC of the head and neck is affected by site, stage, histologic grade, and biologic behavior of the ACC. There are a number of publications that address the efficiency of surgery and radiation therapy in the treatment of ACC of the head and neck [4-6].

ACC of the head and neck, and specifically of the nasal cavity and paranasal sinuses, poses numerous treatment challenges for several reasons: it has a high propensity for local invasion to adjacent structures, making resection more difficult; it is commonly diagnosed late due to its insidious growth; and in 50% of cases it has caused perineural spread at the time of diagnosis [7]. Furthermore, ACC is associated with high rates of distant metastases, which have been noted to occur as late as 10 years after the diagnosis of the primary lesion [8]. Clinicians are all too aware that distant metastases often defeat successful treatment of patients with ACC, despite locoregional control, and are associated with a low long-term survival rate.

Histologically, ACC can be categorized into 3 growth patterns: cribriform, tubular, and solid. In most studies, a solid growth pattern is associated with a worse prognosis, caused by advanced stage and development of distant metastases [9, 10]. A unique feature of ACC is the propensity for perineural invasion, even with early-stage tumours. The tumour is graded according to Szanto et al. [11] as cribriform or tubular (grade I), less than 30% solid (grade II), or greater than 30% solid (grade III). In our report the histopathology revealed a solid subtype. This is probably one factor contributing to the bad outcome.

The reported incidence of spinal epidural compression from head and neck cancer is around 1% [12], which is much lower than that of compression by metastasis from breast (20-26%) or lung cancer (12-13%). Compression of the spinal cord or cauda equina by metastatic disease is almost always extradural [13]. This condition usually results from tumour involvement of the vertebral column affecting either a vertebral body or a neuronal arch, as is the case in our patient. According to literature data, only four patients with carcinoma of the maxillary sinus with spine metastases have been reported: with compression of the cauda equina in two of them. Survival was short (three months) for both [14,17,18].

The clinical course, in our reported case, was atypical in terms of its chronology. The diagnosis of spinal cord metastasis from ACC of the maxillary sinus only nine months after completion of radiotherapy illustrates the highly aggressive nature of this tumour. Decompression and stabilization of the spinal cord can maintain or improve quality of life. The role of decompression and/or fusion in spinal metastases with neurologic deficits is still under debate, although recent studies have confirmed the beneficial role of surgical intervention in selected patients [15, 16]. According to Patchell et al. direct decompressive surgery plus postoperative radiotherapy seems to be superior to treatment with radiotherapy alone for patients with spinal cord compression caused by metastatic cancer [15].

In this reported case, the aggressive nature of the tumour and the presence of intracerebral invasion of the skull base at the time of diagnosis probably explains the rapid onset of this unusual site of metastasis. The presence of perineural invasion on the initial histological examination should have been considered to be a predictive factor of this progression.

**Conclusion**

Spinal metastases of maxillary sinus ACC are uncommon. Decompression and stabilization of the spinal cord can maintain or improve quality of life. The clinical behavior of ACC, and its high propensity for local invasion to adjacent structures makes obligatory a periodic examination throughout life.
References


