

## CASE REPORT

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

O Ait Sahel\*, I Ghfir, H Guerrouj, Y Benameur, N Benraiss

Department of Nuclear Medicine, IBN-SINA Hospital Center,  
University Med V-Rabat, Morocco

### 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

### \*Correspondence

Omar Ait Sahel  
Nuclear Medicine Department  
IBN-SINA Hospital Center  
University Med V-Rabat  
Rabat, Morocco  
Tel: 0021 26284178  
Email: omaraitsahel85@gmail.com

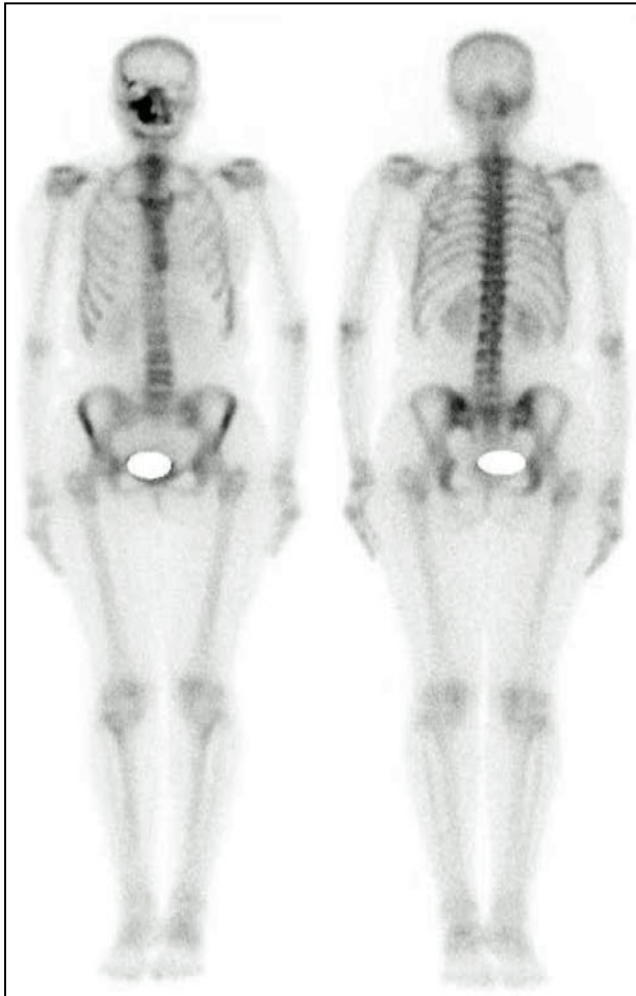
### Introduction

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 <sup>99m</sup>Tc-HMDP bone scan.

### Case report

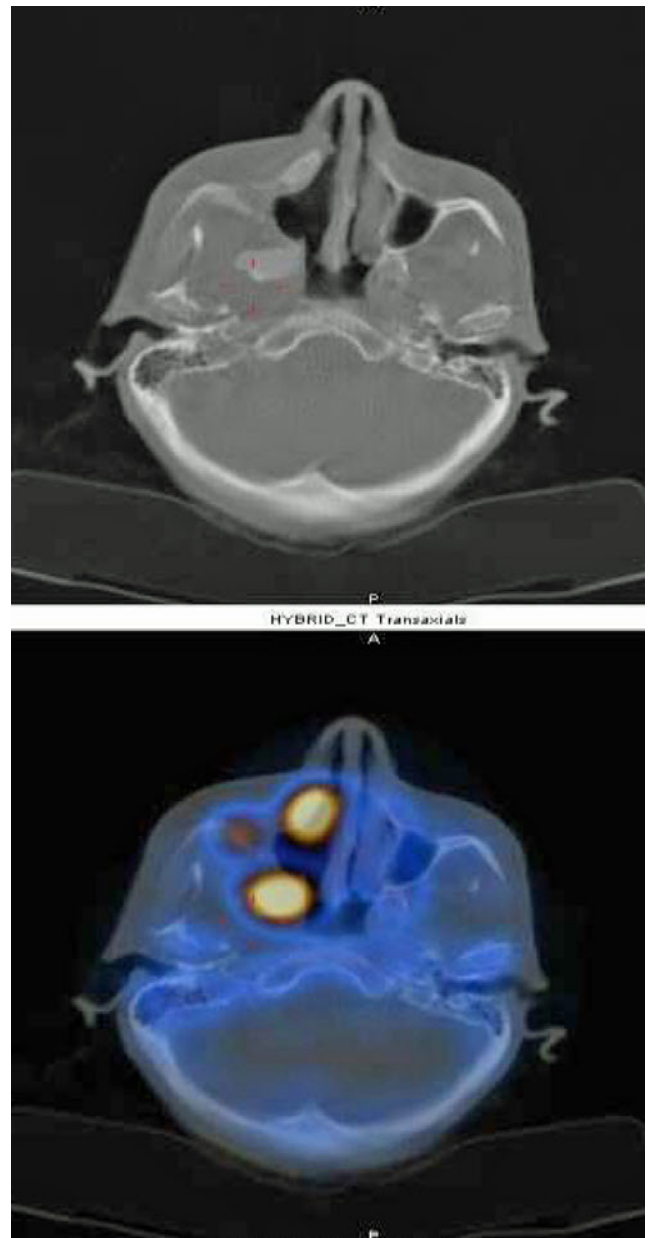
A 45-year-old woman developed ACC of the right maxillary sinus, which was treated by surgery and radiotherapy. The patient presented 9 months later with limb weakness, which had progressed moderately over time. Neurological examination revealed paraparesis with preserved tendon reflexes and sensation.

<sup>99m</sup>Tc-hydroxyl-methylene diphosphonate (<sup>99m</sup>Tc-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).



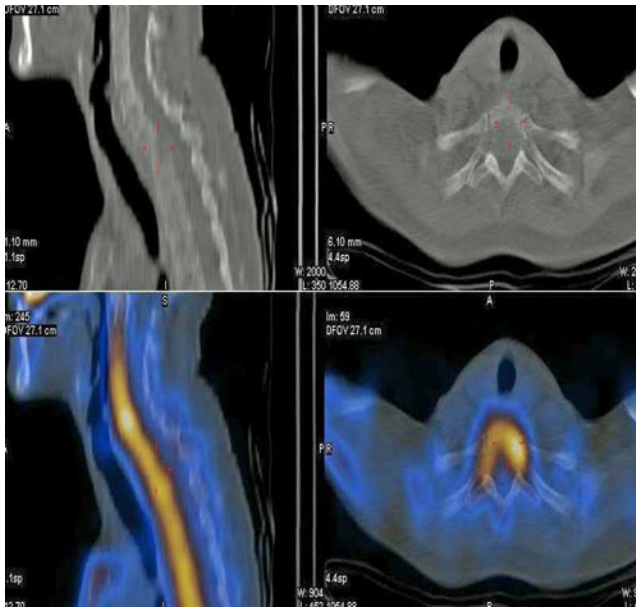
**Figure 1** Planar whole-body scan with localized image, obtained 3 hours after administration of 740 MBq of <sup>99m</sup>Tc-hydroxyl-methylene diphosphonate (<sup>99m</sup>Tc-HMDP), showing increased uptake in the right maxillary sinus, the right orbital roof, with heterogeneous uptake in the region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

SPECT-CT confirmed increased uptake in the right wing of sphenoid bone with cheekbone lysis (Figure 2) together with increased uptake

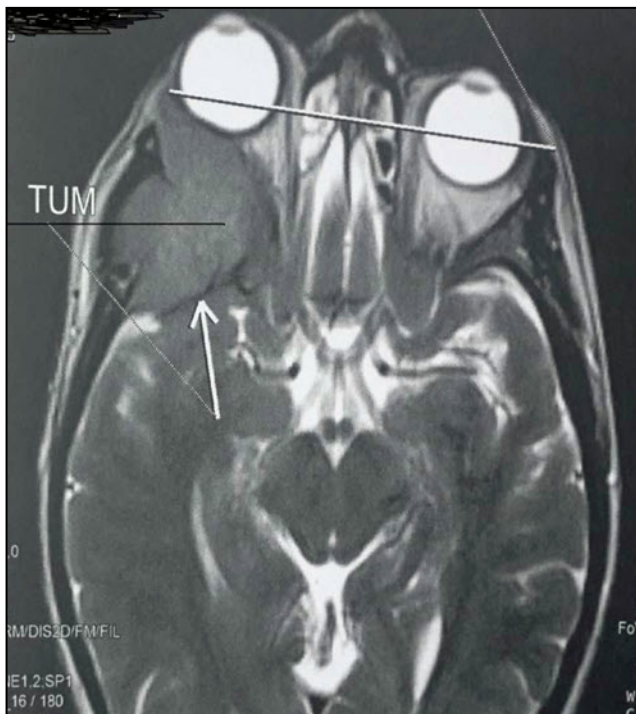


**Figure 2** Figure 2. Axial SPECT-CT scan showing increased uptake of the maxillary sinus extending to involve the right wing of sphenoid bone with cheekbone lysis

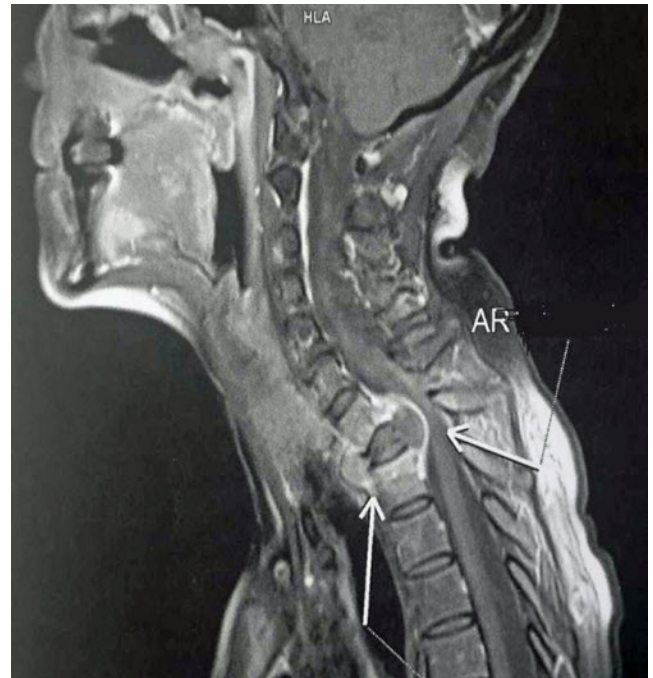
in C7 & T1 vertebral bodies (Figure 3). A follow-up MRI of face and neck with and without contrast injection, showed two lesions: one in the right maxillary sinus extending to the pterygopalatine fossa, and the second affecting the right wing of sphenoid bone with cheekbone lysis, and orbital and temporal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



**Figure 3** Axial and sagittal SPECT-CT scan images showing increased uptake in the 7th cervical and 1st thoracic vertebrae



**Figure 4** Brain MRI scan showing intracerebral invasion of the adnoid cystic carcinoma via the right maxillary nerve



**Figure 5** Sagittal MRI scan showing the lesion in the first thoracic vertebra with posterior involvement and circumferential epidural soft-tissue mass compressing the spinal cord

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

1. Chummun S, McLean NR, Kelly CG. Adenoid cystic carcinoma of the head and neck. *Br J Plast Surg* 2001; 54:476-80.
2. Wang JH, Lee JH, Han JH. Contralateral maxillary sinus lesions in patients with nasal cavity and/or paranasal sinus carcinoma: analysis of computed tomography findings. *Ann Otol Rhinol Laryngol* 2008; 117(12):909-13.
3. Seab JA, Graham JH. Primary cutaneous adenoid cystic carcinoma. *J Am Acad Dermatol* 1987; 17(1):113-8.
4. Le Q-T, Fu KK, Kaplan M. Treatment of maxillary sinus carcinoma. *Cancer* 1999; 86:9.
5. Lupinetti AD, Roberts DB, Williams MD. Sinonasal adenoid cystic carcinoma. *Cancer* 2007; 110:2726-31.
6. Rhee CS, Won TB, Lee CH. Adenoid cystic carcinoma of the sinonasal tract: treatment results. *Laryngoscope* 2006; 116:982-6.
7. Amit M, Binenbaum Y, Sharma K. "Adenoid cystic carcinoma of the nasal cavity and paranasal sinuses: a metaanalysis," *Journal of Neurological Surgery B, Skull Base*, vol. 74, no. 3, pp. 118-125, 2013.
8. Zald PB, Weber SM, Schindler J. Adenoid cystic carcinoma of the subglottic larynx: a case report and review of the literature, *Ear, Nose and Throat Journal*, vol. 89, no. 4, pp. E27-E32, 2010.
9. Matsuba HM, Spector GJ, Thawley SE. Adenoid cystic salivary gland carcinoma: a histologic review of treatment failure patterns. *Cancer* 1986, 57:519-524.
10. Westra WH. The surgical pathology of salivary gland neoplasms. *Otolaryngol Clin North Am* 1999, 39:919-943.
11. Szanto PA, Luna MA, Tortoledo ME. Histologic grading of adenoid cystic carcinoma of the salivary glands. *Cancer* 1984, 54:1062-1069.
12. Ampil F, Nanda A, Aarstad RF, Hoasjoe DK, Chin HK, Hardjasudarma M. Spinal epidural compression in head and neck cancer Report of five cases. *Journal of Cranio-Maxillo-Facial Surgery*(1994) 22;49-52.
13. Stark RJ, Henson RA, Evans SJW. Spinal metastases, a retrospective survey from a general hospital. *Brain* 105 (1982) 189-203.
14. Ampil FL, Lall C, Willis BK. Maxillary sinus cancer and metastatic compression of the cauda equina: report of two cases. *J La State Med Soc* 2001; 153:497-500.
15. Patchell RA, Tibbs PA, Regine WF. Direct decompressive surgical resection in the treatment of spinal cord compression caused by metastatic cancer: a randomised trial. *Lancet* 2005;366:643-8.
16. Ulmar B, Naumann U, Catalkaya S. Prognosis scores of Tokuhashi and Tomita for patients with spinal metastases of renal cancer. *Ann Surg Oncol* 2007; 14:998-1004.
17. Caillot A, Veyssi re A, Ambroise B, B nateau H. Spinal cord metastasis of squamous cell carcinoma of the maxillary sinus. *European Annals of Otorhinolaryngology, Head and Neck diseases* 132 (2015) 97-99.
18. Hopf-Jensen S, Buchalla R, Rubarth O, Peters J, Dunker H, Hensler HM, M ller-H lsbeck S, B rm W. Unusual spinal metastases from an adenoid cystic carcinoma of the maxillary sinus. *Case Reports / Journal of Clinical Neuroscience* 19 (2012) 772-774