

## Case Report

# Adenoid cystic carcinoma of maxillary sinus metastatic to the kidney: a case report and review of the literature

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**Abstract:** Adenoid cystic carcinoma (ACC) is a rare malignant tumor of minor salivary gland, accounting for less than 2% of all the head and neck malignancies. It is characterized as slow growth, long clinical course, local recurrences and distant metastases. Once ACC of primary site presents recurrence and distant metastasis, the prognosis is generally poor. Renal metastasis related to this cancer is relatively uncommon. Herein, we presented a 37-year-old man with maxillary sinus ACC metastasis to the right kidney eight years after surgical resection of the primary tumor. A successful right radical nephrectomy was performed followed by local radiotherapy and antineoplastic therapy. Unfortunately, the patient died of respiratory failure within eight months after the operation. To our knowledge, it is the first report of metastatic renal tumor from maxillary sinus ACC.

**Keywords:** Adenoid cystic carcinoma, maxillary sinus, kidney, metastasis

## Introduction

Adenoid cystic carcinoma (ACC) of the sinonasal tract is an aggressive malignancy with a poor five-year survival rate [1], which is the most common salivary gland-type carcinoma of the maxillary sinus [2]. Distant metastasis is one of the characteristics of this cancer, it occurs commonly in the site of lungs. However, metastasis to the kidney is rare. We present a case of metastatic renal involvement from ACC of maxillary sinus.

## Case presentation

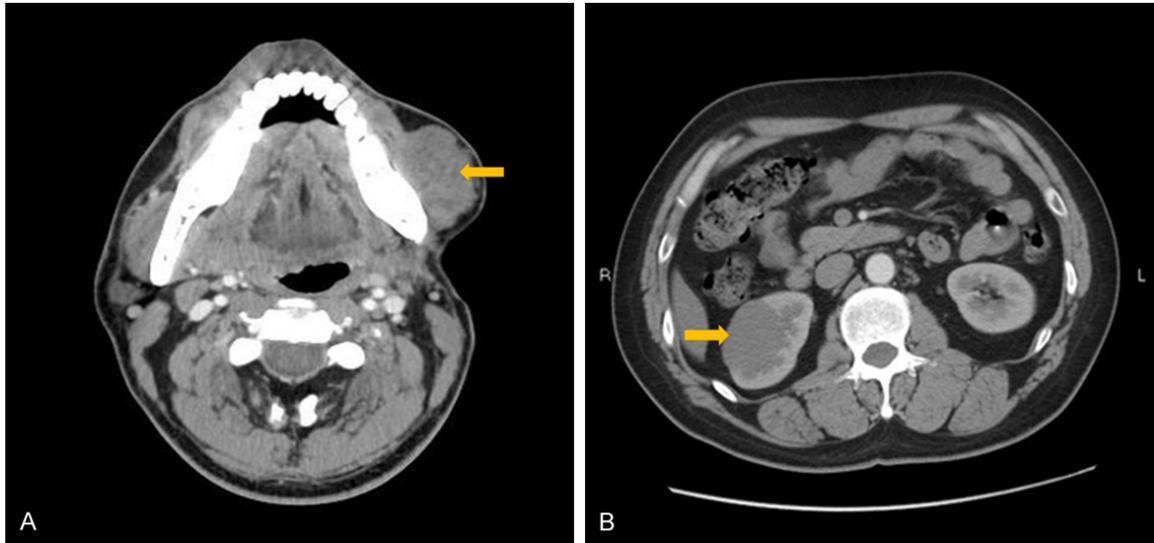
A 37-year-old man presented with left facial swelling of one year duration. The patient gave a history of primary maxillary sinus ACC eight years ago, for which he received an operation, chemotherapy and radiotherapy. A computed tomography (CT) scan suggested a soft-tissue mass measured 3.6×2.3 cm in size in the left mandible (**Figure 1A**). Then, he underwent an extended resection of the tumor, which was finally identified as recurrent ACC by histologic examination.

Following the operation, he was advised an 18F-fluorodeoxyglucose positron emission to-

mography (FDG-PET) scan and the results revealed abnormalities in the right kidney and the spine region, but there was no symptom and sign associated with these abnormalities, such as hematuria or back pain. Abdominal CT scan and magnetic resonance imaging (MRI) of thoracolumbar spine were required for further validation. An enhanced CT scan revealed an approximately 4.0 cm×5.0 cm mass with slightly lower-density on the upper pole of the right kidney (**Figure 1B**). The MRI of thoracolumbar spine demonstrated hyperintense on T2 weighted image at the level of T10-T11, L3 with contrast enhancement, and the destruction of some vertebral bodies could be easily found as well. Therefore, right radical nephrectomy was suggested.

Macroscopically, there was a well-circumscribed, firm, gray mass on the upper pole of the right kidney, measuring 5.0 cm×4.0 cm×2.0 cm, and there were no significantly enlarged lymph nodes in the renal hilum. On histologic examination, a large number of cribriform formations of atypical epithelial cells replaced of normal kidney tissues, the cells were round and oval, similar to basal cells, appearing clustered in small groups (**Figure 2A**). Immunohisto-

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**Figure 1.** A plain CT scan of head and neck showing a soft-tissue mass measured 3.6×2.3 cm in size in the left mandible (A). An enhanced abdominal CT scan revealing an approximately 4.0 cm×5.0 cm mass with slightly lower-density on the upper pole of the right kidney (B).

chemically, the neoplastic cells were positive for alpha-smooth muscle actin (SMA), epithelial membrane antigen (EMA), p63 (**Figure 2B-D**), cytokeratins AE1-AE3, cytokeratin 5/6 and AB/PAS. Moreover, there were weak reactions for cytokeratin 8 and Ki-67. Carcinoembryonic antigen (CEA), S100 protein and cytokeratin 10 were negative on the examination. All these pathological findings were consistent with metastatic ACC to the kidney.

Two months after the operation, the patient received local radiation therapy in spine region with a dose of 40 Gy for one time. Meanwhile, we applied the gefitinib for antineoplastic therapy in case of further recurrence and metastasis. Unfortunately, the patient died of respiratory failure within eight months after radical nephrectomy.

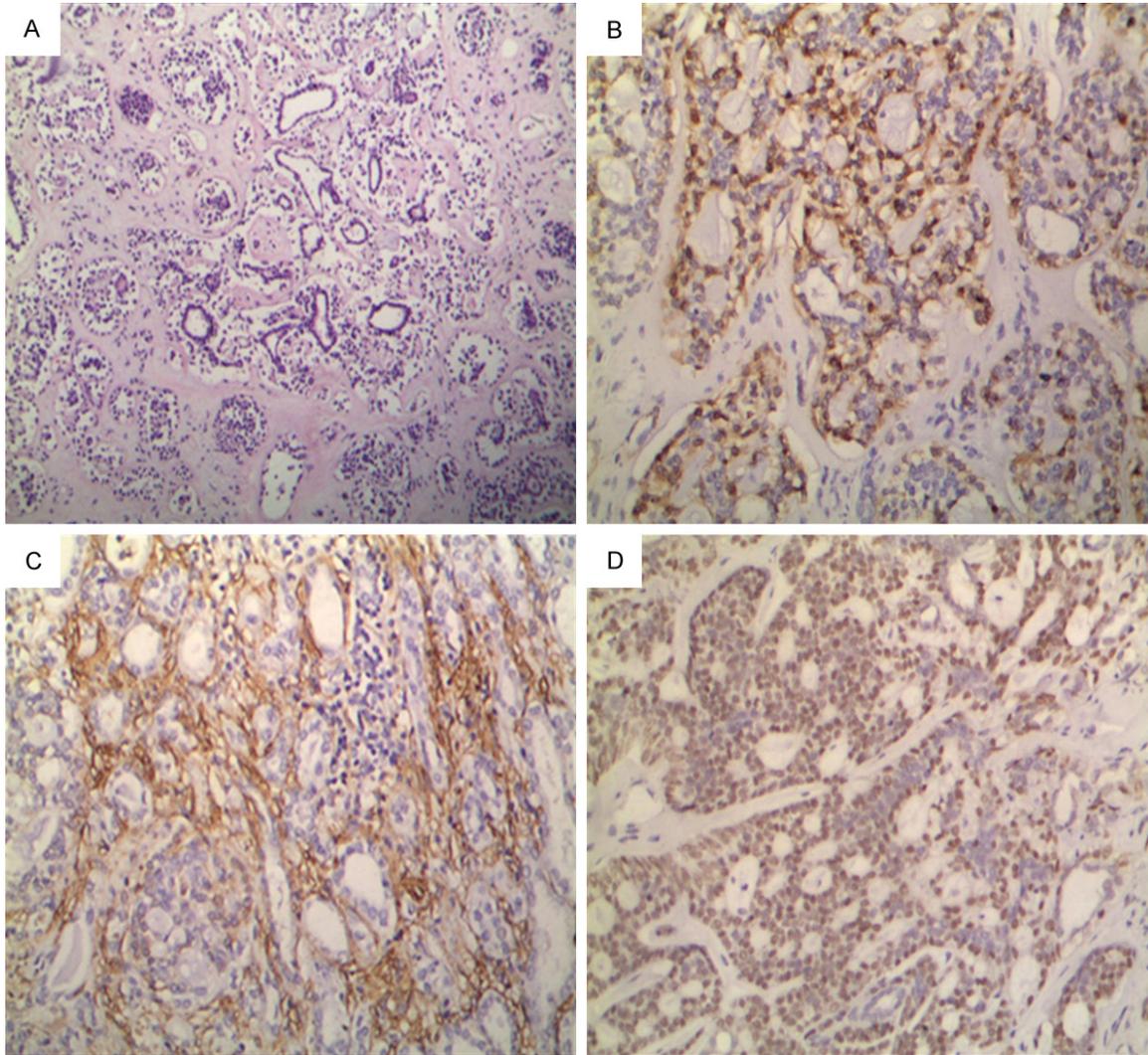
### Discussion

ACC of the head and neck is a salivary gland malignancy. The sinonasal tract is a common site for this malignancy, accounting for 10%-25% of all head and neck ACCs [3]. The maxillary sinus is the most commonly affected primary site, followed by the nasal fossa, ethmoid sinus, and sphenoid sinus [4]. It has distinct characteristics of an indolent but persistent growth and a high rate of perineural spread, local recurrences, and distant metastases [5, 6]. In long-standing cases, distant metastases

were frequently occurred and spread through the bloodstream in 25-55% of cases. Metastases affected the lungs, liver, lymph nodes, long bones, axial skeletons and other uncommon sites [7, 8]. Through reviewing the literature, we summarized a total of 15 cases with renal metastasis from ACC between 1984 and 2014 (**Table 1**) [7, 9-22]. The mean age of these cases was 51.5 years (ranged from 21 to 83 years). Nine patients had at least two metastases and the average interval time between surgery for primary tumor and renal metastasis was 9.8 years, which indicated that the slow growth pattern and high potential of metastasis were associated with this malignancy.

Radical surgery combined with postoperative radiation therapy is regarded as the treatment protocol for primary maxillary sinus ACC [2]. This combination has been proved to be effective in the improvement of overall and disease-specific survival time compared with the patients who received other treatment options [4]. Nevertheless, 58% of the patients develop local recurrences and 38% develop metastases. Radiotherapy is associated with a 96% initial response rate, but 94% of these patients develop recurrence [22]. In the present case, the patient was found local recurrence and distant metastases eight years after surgical resection of the primary tumor combined with chemotherapy and radiotherapy.

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**Figure 2.** Renal metastasis of adenoid cystic carcinoma with cribriform formation of atypical epithelial cells (hematoxylin and eosin stain, magnification, 100 $\times$ ) (A). Immunohistochemical staining showing SMA (B), EMA (C) and p63 (D) positivity in renal tumor (magnification, 200 $\times$ ).

Clinically, tumor metastasis to the kidney is not infrequent, but it is still hard to detect because lacking of clear symptom at the early phase. Diverse CT features of renal metastases have been reported, mainly presenting as solid or cystic masses and hemorrhagic or diffuse lesions. However, it remains difficult to determine whether the tumor is a metastatic or primary one [23]. Fine needle aspiration (FNA) of ACC could provide a distinct cytological morphology [7]. The patient in our case did not take the FNA. Once the singular mass on the kidney was confirmed, radical nephrectomy would be subsequently performed. Based on his medical history, imaging examinations and postoperative pathological findings, we made an initial

diagnosis of metastatic ACC to the kidney which was validated by histologic examination. Nevertheless, considering the poor condition of the patient, further surgical resection of spinal mass was not suggested.

ACC always exhibits a poor prognosis. Approximately 80%-90% of the patients die of this cancer within 10-15 years after initial diagnosis [24]. Once distant metastases occur, nearly 33% of the patients will die within two years [23]. In this case, the patient died within eight months after the diagnosis of local recurrence and renal metastasis. Currently, there is still no effective management of metastatic ACC. Perhaps patients could benefit from early

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**Table 1.** Literature review of metastatic renal tumor from adenoid cystic carcinoma

No	Author (Ref.)	Publish time	Sex	Age	Primary site	Interval <sup>a</sup> (years)	Treatment of renal metastasis	Clinical events <sup>b</sup>
1	Ladefoged <i>et al.</i>	1984	M	47	Bronchial	23	Radical nephrectomy.	None.
2	Fujii <i>et al.</i>	1991	M	79	Lung	3	Chemotherapy.	None.
3	Herzberg <i>et al.</i>	1991	F	57	Breast	12	Radical nephrectomy.	Lung metastasis.
4	Blochle <i>et al.</i>	1993	F	83	Lacrimal gland	25	Radical nephrectomy.	Local recurrence; Intracranial metastasis; Lung metastasis.
5	Matthew <i>et al.</i>	1997	F	30	External auditory canal	3	Chemotherapy.	Liver and lung metastases.
6	Brown <i>et al.</i>	1998	F	48	Salivary gland	13	Chemotherapy; Nephrectomy.	None.
7	Awakura <i>et al.</i>	2001	F	40	Parotid gland	5	Radical nephrectomy.	Local recurrence; Contralateral kidney, liver, lung and brain metastases.
8	Manoharan <i>et al.</i>	2006	F	21	Parotid gland	7	Radical nephrectomy.	Lung metastasis.
9	Jimenez-Heffernan <i>et al.</i>	2007	F	55	Parotid gland	6	N.M.	None.
10	Santamaria <i>et al.</i>	2008	F	71	Palate	13	N.M.	Lung metastasis and local recurrence.
11	Kala <i>et al.</i>	2010	F	35	Salivary gland	8	Radical nephrectomy.	None.
12	Goyal <i>et al.</i>	2011	M	50	Lung	7	Radical nephrectomy.	Heart metastasis.
13	Grimm <i>et al.</i>	2012	F	55	Mandible	5	N.M.	Lung, breast and pancreas metastases.
14	Qiu <i>et al.</i>	2014	M	26	Submandibular gland	3	Radical nephrectomy.	None.
15	Bacalja <i>et al.</i>	2014	F	76	Lacrimal gland	14	Radical nephrectomy.	Lung metastasis.

N.M = Not mentioned; M = male; F = female; <sup>a</sup>Interval between surgery for the primary tumor and for the kidney or spine metastasis; <sup>b</sup>Clinical events since primary tumor surgery.

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metastasectomy combined with chemotherapy or other adjuvant therapies, but none has been proved to improve the survival rates [22].

In conclusion, ACC of maxillary sinus is an uncommon malignancy with slowly growing manner and highly aggressive behavior. With the increasing numbers of case reports on its unusual metastatic pattern, as an urologist, more attention should be paid to the patient who present with a renal mass and give a past history of primary ACC. For these patients, it is very important to make the early correct diagnosis and apply proper treatment. Besides, life-long follow-up is still necessary in case of tumor recurrence and additional metastasis.

### Disclosure of conflict of interest

None.

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