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Renal Metastases from a Nasal Cavity Mixed Squamous Cell and Adenoid Cystic Carcinoma: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Female, 64-year-old
Final Diagnosis: Renal metastases from a nasal cavity mixte • squamous cells and adenoid cystic • carcinoma
Symptoms: Epistaxis
Medication: —
Clinical Procedure: —
Specialty: Oncology

Objective: Rare disease
Background: Adenoid cystic carcinoma (ACC) is a very rare tumor with a high risk of loco-regional recurrence and potential distant metastases. Until now, only a few cases of renal metastases from ACC have been reported in the literature.
Case Report: A 64-year-old, Caucasian, non-smoker female, 8 months after being treated by radio-chemotherapy for a squamous cell nasal cavity tumor, presented two renal lesions associated with lung and vertebral metastases. Histology was consisted with a metastasis from an ACC. The histological revision of the primary nasal tumor confirmed a squamous cells carcinoma with an adenoid cystic component that metastasized to the kidney. Renal lesions appeared hypometabolic at the ¹⁸F-fluorodeoxyglucose (¹⁸F-FDG) PET scan mimicking a primary renal tumor. The patient underwent a systemic, palliative chemotherapy by a weekly carboplatin/paclitaxel/cetuximab regimen that was well tolerated and allowed a lasting tumor control.
Conclusions: The particularity of this case relies on the rarity of renal metastasis from ACC, its difficult diagnosis, and the complexity of its management, as no standard chemotherapy has been validated for metastatic ACC, yet. In our case, a weekly carboplatin/paclitaxel/cetuximab regimen was administered leading to a durable tumor stabilization with an excellent patient's quality of life.

MeSH Keywords: Carcinoma, Adenoid Cystic • Neoplasm Metastasis • Neoplasms, Squamous Cell

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/919781>

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Background

Adenoid cystic carcinoma (ACC) is a very uncommon tumor that shows a characteristic perinervous and perivascular spreading pattern, which can explain its high risk of local recurrence and its potentiality for distant metastases [1–3]. Primary sinonasal tract and nasopharyngeal adenoid cystic carcinoma (STACC) originates from the respiratory epithelium or the underlying mucoserous glands of the nasal cavity or paranasal sinuses [4–6]. Due to the large anatomic sinonasal tract location, STACC is belatedly diagnosed at a locally advanced stage that is more difficult to radically treat [4–6].

Until now, only a few cases of renal metastases from ACC have been reported in the literature [7–14].

In our case, a patient who was treated by concomitant radiotherapy and chemotherapy 8 months before for a primary mixed squamous cell and adenoid cystic carcinoma of the nasal cavity, presented with 2 renal metastases in a context of a lung and vertebral tumor dissemination. A synchronous follicular and papillary thyroid tumor was also diagnosed. Systemic, palliative chemotherapy by weekly carboplatin/paclitaxel/cetuximab regimen was started leading to a durable tumor control with excellent patient quality of life.

Case Report

In January 2018, a 64-year-old, Caucasian, non-smoker female consulted for epistaxis and nasal obstruction. Clinical examination documented a right nasal polypoid lesion. She had no relevant comorbidities. Her history was uneventful. Computed tomography (CT) scan and magnetic resonance imaging (MRI) confirmed the presence of a tumor mass of the right nasal cavity, involving the pterygoid fossa, the sphenoid sinus, and the posterior maxillary sinus wall, without loco-regional lymph nodes and distant metastases. Biological tests were in the normal ranges. Tumor biopsy revealed a squamous cells carcinoma, p16 positive. An external radiotherapy (70 Gy in 35 fractions over 7 weeks) concomitant to a systemic chemotherapy with cisplatin (3 cycles at 100 mg/m² every 3 weeks) was administered from March to April 2018. The post-treatment radiological evaluation (July 2018) found a partial tumor response. The patient's follow-up revealed a stable disease until December 2018 when a whole-body CT scan documented the presence of a lung metastasis in the left lower lobe and 2 tissular lesions of the right kidney (Figure 1A, red arrows). The patient referred to a percutaneous ultrasound-guided core biopsy of a renal lesion. Histology showed a cribriform tumor massif with numerous round punched lights surrounded by a proliferation of myoepithelial cells with a poorly limited amphophilic cytoplasm and monomorphic, homogenous,

basophilic, and round nuclei (Figure 1B). Tumor cells were positive for CD117, heterogeneously (Figure 1C). Basal cells were also positive for P63 (Figure 1D). This histology pattern was consisted with a renal metastasis from an ACC. Based on these data, the histology of the primary nasal tumor was revised and confirmed a squamous cells carcinoma with an adenoid cystic component that accounted for renal metastases. A coupled CT/¹⁸F-fluorodeoxyglucose (¹⁸F-FDG) PET scan was also performed documenting a bi-focal hypermetabolism of the left thyroid lobe and 2 lung and vertebral metastases (Figure 1E, yellow arrow), the two renal metastases showing a hypometabolic pattern (Figure 1E, red arrows). Considering the pluri-metastatic context and the absence of clinical symptoms related to this secondary tumor renal involvement, a surgical approach was not considered. Indeed, in March 2019, a systemic chemotherapy by a weekly carboplatin/paclitaxel/cetuximab regimen was started. The radiological evaluation by CT/¹⁸F-FDG PET scan after 6 and 12 cycles of chemotherapy showed a stable disease (Figure 1F, red arrows). The patient underwent also a percutaneous cytology of the 2 left thyroid hypermetabolic lesions revealing a follicular (oncocyctic) and a papillary tumor, respectively, which, considering the patient's metastatic setting, were not surgically removed and showed a radiological stabilization after 12 cycles of chemotherapy. Presently, the patient is in good clinical conditions and still on chemotherapy treatment.

Discussion

ACC, firstly described by Robin et al. in 1853 [1,2], is a very rare tumor, representing 1% of head and neck cancers and 10% of malignant salivary gland tumors [1–3]. ACC usually involves the submandibular gland (15% to 30%) or minor salivary glands (30%), the involvement of the nasal cavity being very uncommon and less frequent than this one of the maxillary sinuses [1–3].

Primary sinonasal tract and nasopharyngeal adenoid cystic carcinoma (STACC) is separated into salivary gland-type and non-salivary gland-type adenocarcinoma and it can originate from the respiratory epithelium or the underlying mucoserous glands [4–6]. STACC represents approximately 10% to 18% of all malignant sinonasal tract neoplasms, and 13% of all head and neck ACCs [4–6].

ACC shows a characteristic perinervous and perivascular spreading tumor pattern that accounts for its high local recurrence risk, adjacent skull base invasion by bone lysis and/or spreading within the skull base foramina, and distant metastases [1–6]. Several histological subtypes have been described, including tubular, cribriform, solid or mixed type, the solid one representing the most malignant subtype [1–6]. Recent studies showed

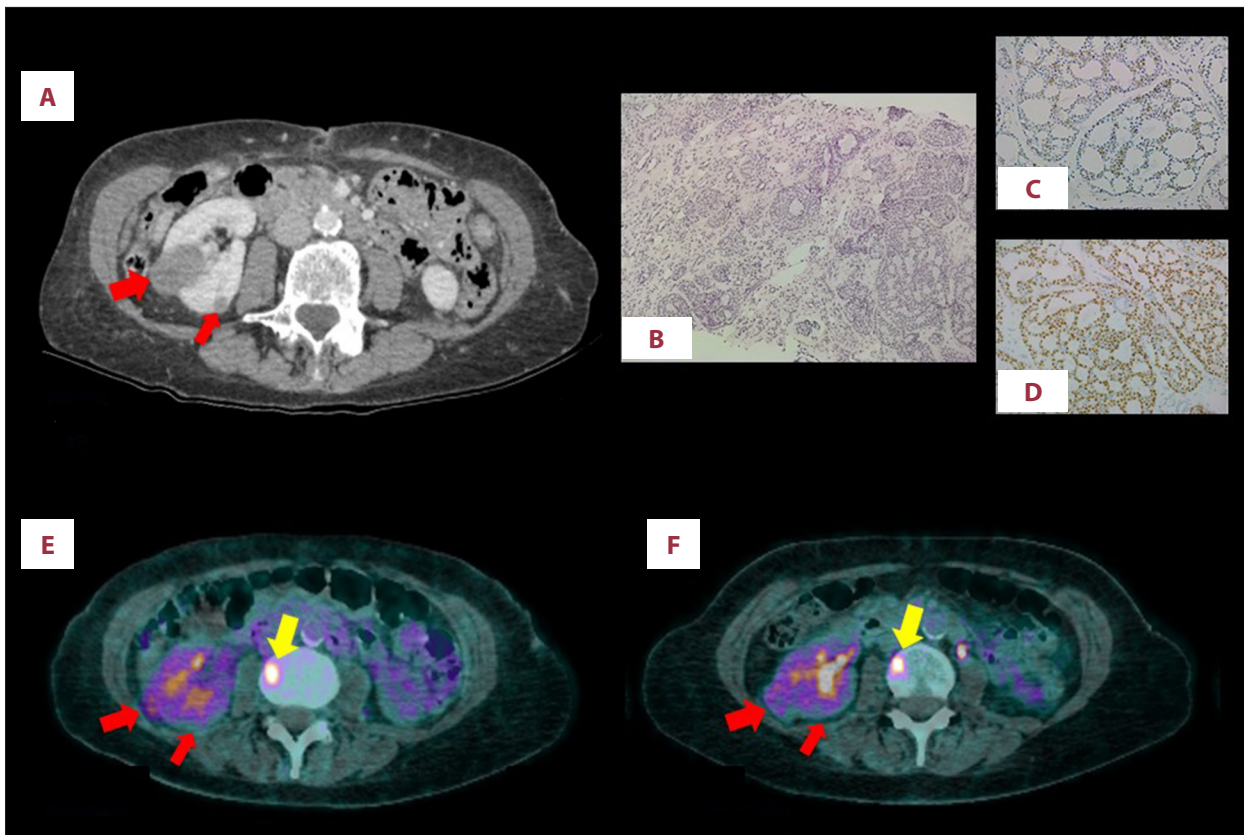


Figure 1. Diagnosis of 2 renal metastases from adenoid cystic carcinoma (ACC). (A) Two hypodense renal tumor lesions, the larger one partially exophytic (abdominal axial section computed tomography scan; red arrows). (B) Cribriform tumor masses with numerous round punched lights surrounded by a proliferation of myoepithelial cells with a poorly limited amphophilic cytoplasm and monomorphic, homogenous, basophilic, and round nuclei (histology; hematoxylin and eosin stain, 200 \times). (C) The inner neoplastic epithelial cells are positive for CD117 (hematoxylin and eosin stain, 200 \times). (D) Basal neoplastic cells show a myoepithelial differentiation and are positive for P63 (hematoxylin and eosin stain, 200 \times). (E) Renal metastases appear hypometabolic at the ^{18}F -FDG-PET-scan (red arrows). A hypermetabolic vertebral metastasis is also found (yellow arrow). (F) ^{18}F -FDG PET-scan, performed 6 months after the beginning of the systemic chemotherapy, shows a stabilization of the vertebral metastasis (yellow arrow) and the 2 hypometabolic renal lesions (red arrows).

that c-Kit, vascular endothelial growth factor (VEGF), and Ki-67 are usually associated with a significantly poor prognosis, while epidermal growth factor receptor (EGFR) overexpressing tumors present a better 3-year survival [15,16].

The main clinical symptoms include unilateral nasal deformation and/or obstruction, pain and recurrent epistaxis. Horner's syndrome and persistent unilateral sero-mucous otitis have been also described [17,18].

Due to the large anatomic sinonasal tract location, STACC can silently permeate the air-filled spaces, reaching a considerable size before clinical symptoms appearing. These aspecific symptoms can mimic a chronic sinusitis or an obstructive lesion and usually precede the diagnosis of about 2 years. This diagnostic delay and the STACC characteristic slow tumor growth

could explain the large size of these neoplasms at the diagnosis. In contrast to salivary gland ACC, pain or other neurological symptoms are not commonly seen in STACC (12.8%) [17,18].

Diagnosis requires a clinical examination, CT scan and tumor biopsy for an accurate histological definition. MRI is essential for a precise assessment of tumor extension before planning the appropriate treatment [17,18].

Surgery, including endoscopic approaches, followed by conventional or neutron radiotherapy, represents the standard treatment [17–23]. Recently, interesting results have been reported with the post-surgery photodynamic therapy in recurrent STACCs [21–23]. As far as proton therapy is concerned, it should be reserved for tumors with extensions to the sphenoid bone or the clivus, and chemotherapy for palliative

treatment [17–23]. There is no validated systemic chemotherapy for metastatic tumors [17–23].

Based on autopsy series, the incidence rate of renal metastasis ranges from 3% to 15%, lung, breast, gastric and colon cancer, and melanoma being the most frequent tumors described [24].

Renal metastases from ACC are extremely rare and only a few cases have been reported in the literature until now [7–14].

Renal metastasis is frequently misdiagnosed and can mimic radiologically a primary renal tumor [7–14,24]. It is often asymptomatic and rarely exhibits hematuria as it is usually located in the vascular plexus cortex without any tumor infiltration of the adjacent urothelium [24].

Histology remains necessary for an accurate diagnosis and it should be interpreted according to the patient's medical history.

Management of renal metastasis is not well defined and should be adapted to each individual situation, considering the histology, the time of relapse after the treatment of the primary tumor, the presence of other concomitant metastases, the clinical symptoms and the patient's comorbidities. Surgery ought to be considered only in selected cases. Other loco-regional approaches, such as thermoablation, renal embolization, radiotherapy and stereotactic radiotherapy may be used, particularly in patients with painful lesions [24]. Finally, systemic chemotherapy plays a palliative role with an efficacy that is strictly related to tumor histology.

In our case, the patient, presenting a primary STACC treated by a radio-chemotherapy 8 months before, developed 2 renal

metastases in a context of lung and vertebral tumor dissemination. This metastatic renal involvement appeared hypometabolic at ^{18}F -FDG PET scan mimicking a primary neoplasm of the kidney. Histology was necessary for an accurate diagnosis.

In addition, the patient showed 2 synchronous thyroid tumors, a follicular (oncocyctic) and a papillary cancer, which, considering the patient's metastatic setting, were not surgically removed. They remained stable after 12 cycles of systemic chemotherapy.

Conclusions

The particularity of this case relies on the rarity of the renal metastasis from STACC and on the difficulty of its diagnosis and management.

Effectively, there is not a validate chemotherapy for metastatic ACC. Considering the limited data in the literature and the mixed, adenoid cystic and squamous cells, histology of the primary tumor, we started a systemic chemotherapy with a weekly carboplatin/paclitaxel and cetuximab protocol that led to a tumor control over 6 months with an excellent patient's quality of life.

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Conflicts of interests

None.

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