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A rare case of bilateral renal metastases arising from minor salivary gland adenoid cystic carcinoma

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A B S T R A C T

A 67-year-old female patient was diagnosed with bilateral renal metastases from adenoid cystic carcinoma (AdCC) of salivary gland origin five years after the primary diagnosis of minor salivary gland AdCC. Bilateral renal core needle biopsies were performed to distinguish primary renal cell carcinoma (RCC) from metastases and to guide treatment strategy. Few similar cases have been reported; none had bilateral metastases at the time of discovery or biopsy-verified AdCC metastases prior to the treatment decision. RCC was a tentative diagnosis and renal metastases of AdCC have previously been mistaken for RCC.

1. Introduction

The worldwide crude incidence rate of salivary gland cancers is 0.69/100,000 inhabitants per year including >20 histological subtypes with adenoid cystic carcinoma (AdCC) as one of them.[1] Salivary gland AdCC has a tendency for recurrence, and distant metastases are not uncommon, but renal metastases are rare.[1–4] Recurrent and/or metastatic salivary gland AdCC has an overall survival rate of 40% at 10 years.[1] We report a case of bilateral renal metastases from salivary gland AdCC.

2. Case presentation

A 67-year-old woman was diagnosed with bilateral renal metastases of salivary gland AdCC. The patient was first diagnosed with minor salivary gland AdCC at the base of the tongue (T3N0M0, no perineural growth but focal vascular invasion) and received external beam radiotherapy five years before the present findings.

Recently, a PET-CT scan showed increased metabolism in the primary site as well as bilateral renal masses and small lung nodules. A triphasic renal CT scan visualized contrast-enhancing homogeneous renal masses [Fig. 1]. The patient underwent microscopically non-radical surgical tumor resection at the primary site with confirmation of local recurrence [Fig. 2]. The primary tentative diagnosis was a local recurrence of AdCC in combination with bilateral renal cell carcinoma (RCC) with lung metastases. Histopathology following percutaneous renal core needle biopsies (CNB) showed AdCC in both kidneys [Fig. 3A–B], similar to the primary tumor [Fig. 2]. Of note, the patient had a surgically treated local recurrence a year earlier without free resection margins in a single histopathological section.

Following a multidisciplinary team discussion and shared decision-making, no additional treatment was offered due to incurability and focus on the patient's quality of life. At the latest follow-up, the patient was in good general condition and only bothered by radiation sequelae from the primary treatment.

3. Discussion

The present case demonstrates the rare incident of bilateral renal metastases of salivary gland AdCC. Just under a fifth of the patients with salivary gland AdCC develops distant metastases, predominantly involving the lungs, with a smaller fraction involving bone, liver, and

Abbreviations: AdCC, adenoid cystic carcinoma; RCC, renal cell carcinoma.

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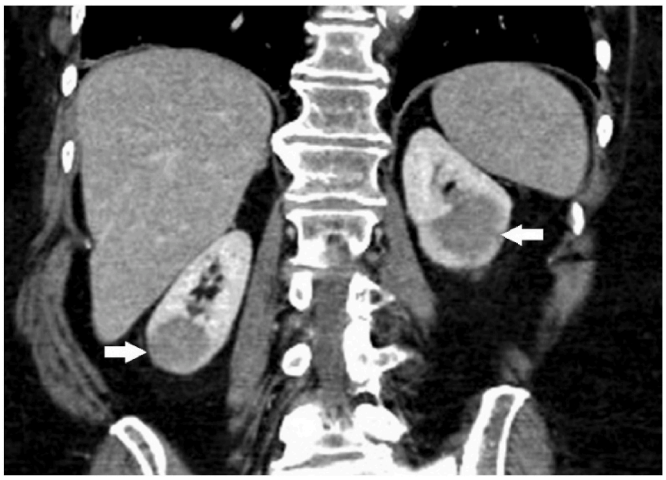

Fig. 1. Renal CT scan
Representative coronal image from renal CT scan in intravenous phase demonstrating bilateral expansive and low vascularized cortical/parapelvic renal masses (white arrows) with a maximal diameter of 4 cm.

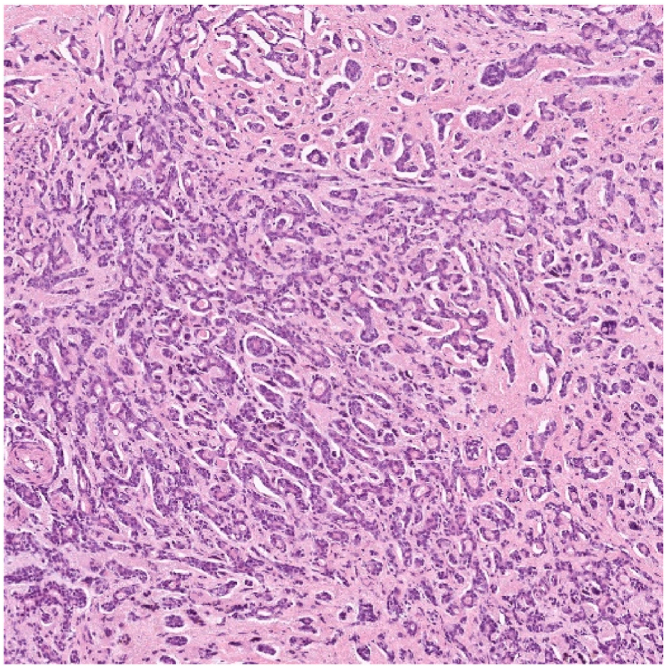

Fig. 2. Histopathological cross-section of tongue tumor
Tumor at the tongue basis demonstrating minor salivary gland tubulo-cribriform adenoid cystic carcinoma. Hematoxylin-eosin-stained.

brain.[1,2] Renal metastases, as in the present case, are rare.[2–4] To our knowledge, only a few cases have been reported, none of which included bilateral renal metastases at the time of discovery or biopsy-verified AdCC metastases prior to treatment decision-making.[3,4]

The most similar case reported was a case of metastatic parotid AdCC to the kidney 13 years after the primary diagnosis.[3] On CT, the renal mass was heterogeneous and lobulated with a diameter of up to 10 cm. One year after the primary nephrectomy the patient had a recurrence in the contralateral kidney.[3] In the present case the bilateral renal metastases were homogeneous, smaller, and appeared closer to the primary diagnosis, and the AdCC originated from a minor salivary gland.

In another case, a renal mass was detected on a CT scan and fine needle aspiration cytology suggested poorly differentiated RCC, which supported the clinicians' tentative diagnosis of RCC.[4] Post-nephrectomy histopathology determined that it was AdCC consistent with the patient's previous history of salivary gland AdCC.[4] RCC with lung metastases was the tentative diagnosis in the present case based on the CT findings and the fact that RCC accounts for approximately 90% of all renal malignancies.[5] The radiographic appearance of RCC can vary from solid and homogeneous to partially cystic and heterogeneous, which seems to be the case for renal metastases of AdCC too.[5]

According to the European Association of Urology (EAU), biopsies of contrast-enhancing solid renal masses are recommended when considering active surveillance or ablative therapy and to select the most suitable surgical and/or medical treatment in the setting of metastatic disease, which was suspected in the present case due to appearance of lung nodules.[5]

EAU recommends CNBs over fine needle biopsies for solid tumors due to higher diagnostic accuracy and at the same time low morbidity related to the procedure.[5] In the present case, with bilateral solid renal masses, a history of extrarenal primary malignancy, and a patient in good general condition, CNBs were performed to obtain histology to distinguish RCC from metastases and to guide treatment in case of metastatic RCC. The AdCC renal metastases were confirmed by histological morphology and immunohistochemical profiles similar to that of the primary tumor in the tongue.

As in the present case, approximately one-third of the patients with salivary gland AdCC have recurrence. In Denmark, the median time from primary diagnosis to discovery of distant metastases is 2.7 years, a slightly shorter time than in the present case, but recurrence has been reported up to 15 years after primary curative treatment.[2] Furthermore, contrary to the present case with simultaneous local recurrence, almost half of the patients with recurrence exclusively have a distant recurrence, which emphasizes the importance of imaging to discover distant metastases.[2]

Due to the high risk of recurrence and distant metastases, managing salivary gland AdCC is a therapeutic challenge. Radical surgery and disease stage are essential prognostic factors for curing AdCC patients.[2] The former represents a special challenge in managing minor salivary gland AdCC.[1,2] The patient in the present case had primary deep tumor growth in the tongue substance making it functionally unresectable. Systemic treatment and radiotherapy for AdCC have not been shown to improve survival, but radiotherapy improves locoregional control and can be considered for functionally unresectable tumors or in the palliative setting.[1,2] The latter was not an option in the present case due to recurrence in the former high-dose area.[1]

In summary, no curative intended treatment was found for the patient in this case report. Focusing on the patient's quality of life and after shared decision-making, no additional treatment was offered.

4. Conclusion

Renal metastases of salivary gland AdCC are rare and can be mistaken for RCC. In the present case with a history of extrarenal primary malignancy and bilateral renal masses, CNBs were performed to distinguish RCC from metastases and to guide treatment strategy. The histopathological findings were consistent with metastases of salivary gland AdCC in both kidneys.

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Consent

Informed and written consent was obtained from the patient for the publication of this case report.

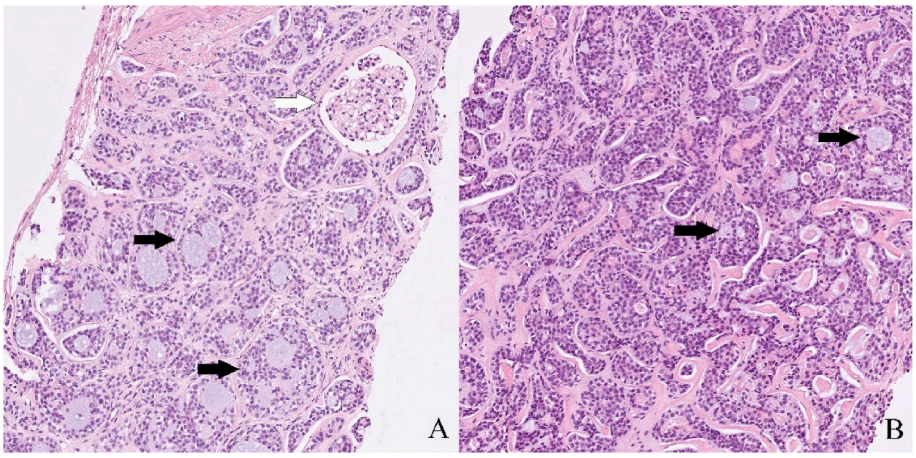

Fig. 3A–B. Histopathological cross-sections of renal masses
Core needle biopsies of the right (A) and left (B) renal masses demonstrating a morphology similar to the primary adenoid cystic carcinoma in the tongue. Note the tubulo-cribriform growth pattern (black arrows) and glomerulus (white arrow). Hematoxylin-eosin-stained.

CRediT authorship contribution statement

Mona Løgtholt Kristensen: Conceptualization, Data curation, Investigation, Methodology, Validation, Writing – original draft, Writing – review & editing, Visualization, Project administration. **Henriette Nelsson:** Writing – review & editing. **Sandra Simony Tornøe Riis:** Writing – review & editing. **Kristine Bjørndal:** Writing – review & editing. **Ole Graumann:** Conceptualization, Writing – review & editing. **Theresa Junker:** Conceptualization, Data curation, Investigation, Methodology, Validation, Writing – review & editing, Visualization, Supervision, Project administration.

Declaration of competing interest

OG: Speaker honoraria, Advisory Board member, and research grant: Boston Scientific. TJ: Research grant: Boston Scientific. Other authors: None.

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