Bilateral kidney metastases from adenoid cystic carcinoma of lung: a case report and literature review

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Abstract

Adenoid cystic carcinoma (ACC) is a rare malignant tumor, usually arising from salivary glands and rarely found in other locations. ACC is characterized by asymptomatic course, slow growth pattern, perineural invasion and high incidence of late metastasis. Renal localization of metastasis is rare. Only 12 cases of renal metastasis were published and, to our best knowledge, no case of bilateral metastasis to the kidney has ever been reported. We present a case of a 58 years old woman with bilateral renal metastasis from ACC of the right lung after fourteen years from lobectomy and radiotherapy for the primary presentation. The patient underwent bilateral robot-assisted partial nephrectomy in a two-stage approach. Despite its rare incidence and slow growth, ACC metastasis may simulate primary papillary renal carcinoma and occur many years after primary treatment. Therefore, lifelong follow-up, including abdominal imaging, is recommended.

Keywords Adenoid cystic carcinoma · Kidney · Kidney metastases · Partial nephrectomy · Robot assisted-surgery

Introduction

Adenoid cystic carcinoma (ACC) is a rare tumor, accounting for less than 1% of malignant tumor of head-neck district. It is usually found in major and minor salivary glands but can also affect other sites such as breasts or lungs [1]. ACC is a slow growing but extremely aggressive tumor, characterized

by high incidence of neural invasion. Despite its slow biologic growth, ACC is associated with poor outcomes, with a 10-year survival of less than 50%, regardless of the histologic grade [2]. Moreover, ACC have shown high recurrence and metastasis rates [3]. Common metastasis sites are bones, lungs, brain and liver. Kidney is a known, even though rare, dissemination site [4]. To date, only 12 cases of ACC's kidney metastasis have been reported in English literature (Table 1); no cases of histologically verified bilateral kidney metastasis from ACC have been reported. We present a case of a 58 years old woman with bilateral renal metastasis from ACC, fifteen years after treatment of the primary tumor, treated with robot-assisted partial nephrectomy.

Case presentation

A 58 years old woman presented to the Emergency Department at Onze-Lieve-Vrouw Hospital (Aalst, Belgium) with left flank pain. Her past medical history included ACC of the right lung, treated with surgical resection of the upper pulmonary lobe in 1996, and with radiotherapy in 2004 for disease recurrence. At admission, laboratory examinations

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Table 1 Clinicopathological features of 12 cases of metastases to kidney from adenoid cystic carcinoma

References	Age (y)/sex	Primary ACC location	Primary treatment	Time to MTX	MTX location/size (Cm)	Treatment	Outcome
Ladefoged et al. [13]	47/M	Right lung	Pneumonectomy	23 years	Left kidney/9 cm	Left radical nephrectomy	ANED after 12 months
Herzberg et al. [14]	57/F	Breast	Radical mastectomy	12 years	Left kidney/6.5 cm	Left radical nephrectomy	ANED after 20 months
Blöchle et al. [15]	83/F	Left lacrimal gland	Excision + RT	25 years	Left kidney/not reported	Left radical nephrectomy	Unknown
Manoharan et al. [16]	21/F	Left parotid	Parotidectomy	7 years	Left kidney/9 cm	Left radical nephrectomy	ANED after 4 years
Vranic et al. [17]	76/F	Breast	Mastectomy	5 years	Right kidney/9 cm	Right radical nephrectomy	Unknown
Santamaria et al. [18]	71/F	Right palate	Hemimaxillectomy	14 years	Right kidney/2.5 cm	Not reported	Unknown
Kala et al. [19]	35/F	Salivary gland	Excision + CHT + RT	8 years	Right kidney/6 cm	Right radical nephrectomy	Unknown
Goyal et al. [19]	50/M	Right lung	Lobectomy + RT + CHT	7 years	Right kidney/9.3 cm	Right radical nephrectomy	Unknown
Bacalja et al. [26]	26/M	Left salivary gland	Hemimaxillectomy + RT	3 years	Right kidney/6 cm	Right radical nephrectomy	Unknown
Qiu et al. [20]	76/F	Left lacrimal gland	Surgical resection	14 years	Right kidney/7.8 cm	Right radical nephrectomy	Unknown
Liu et al. [12]	65/F	Right parotid gland	Surgical resection	9 years	Left kidney/3.8 cm	Left radical nephrectomy + CHT	ANED after 24 months
Our case	58/F	Right lung	Lobectomy + RT	14 years	Left kidney/7.5 cm Right kidney/1 cm	Bilateral RAPN with two-stage approach	ANED after 12 months

Y years, MTX metastases, M male, F female, ANED alive with no evidence of disease, RT radiotherapy, CHT chemotherapy

showed regular renal function, normal blood cell count and no elevation of the inflammation markers.

Abdominal contrast enhanced computed tomography (CECT) showed the presence of a multilobular hypodense lesion (maximum diameter 66 mm) with irregular contrast enhancement at the lower pole of the left kidney and a second smaller lesion (maximum diameter 9 mm), with similar characteristics, in the upper pole of the right kidney. (Fig. 1) In order to exclude the presence of urothelial carcinoma (UC), urinary cytology was performed, which resulted negative. Both lesions were finally considered as suggestive for papillary renal cell carcinoma (RCC) and, therefore, nephron sparing surgery was planned for the treatment of tumors [5–8].

The patient underwent left robot-assisted partial nephrectomy (RAPN) 30 days after the diagnosis. Pathological examination showed a solid lesion, measuring 75 × 75 × 60 mm, covered by a thin layer of healthy renal parenchyma. Tumor had cribriform pattern and were composed by basophilic cells with normochromic nuclei (Fig. 2b). Immunohistochemical studies showed diffuse positivity to pancytokeratin and p63, the staining with

synaptophysin was negative and cells showed a diffuse expression of CD117. Of note, the pathological examination of the primary pulmonary lesion, treated in 1996, showed a similar cribriform pattern with basophilic cells and normochromic nuclei; immunohistochemistry showed positivity for PAS positivity and a similar diffuse positivity to pancytokeratin; the primary tumor showed also positivity for vimentin, actin and s100 protein (Fig. 1a). These findings were suggestive for metastatic adenoid cystic carcinoma [9].

After a multidisciplinary meeting with urologist, nephrologist and oncologist, considering the absence of surgical margins, we decided to leave the kidney in place and proceed with the asportation of the contralateral lesion via right robot-assisted partial nephrectomy (Fig. 3). Immunohistochemical studies confirmed the metastatic origin of the lesion. The patient was dismissed regularly 3 days after surgery on both occasions. No intraoperative and postoperative complications were recorded. The patient is currently alive and free from disease at 18 months follow up (Fig. 4).

Fig. 1 Abdominal contrast enhanced CT scan (CECT) demonstrating the left (a) and right (b) renal masses

Fig. 2 Pathological examination of the primary pulmonary lesion (a) and of the left renal metastases (b)

Discussion and literature review

Adenoid cystic carcinoma is a high-grade rare tumor usually affecting major salivary glands. As results of their location, accurate clinical examination and follow up of these tumors is frequently impeded by altered anatomy and post-operative fibrosis. Therefore, recurrence and metastasis spread go clinically unnoticed during early stages. Common metastasis sites are lungs (80%), bones (14%), brain (3%) and skin (1%) [10] and no standardized follow-up protocols have been validated for detection of abdominal metastases [11]. Kidney metastases are anecdotic, with only 12 cases of kidney metastases are reported in English literature (Table 1) [12–20]. Despite the presence in literature of another case described by Saha et al. [21], no

Fig. 3 Intraoperative aspect of the right renal metastasis

Fig. 4 Abdominal contrast enhanced CT scan 12 months after the two surgeries

histological examination were performed and the metastatic origin of the tumor can be only supposed; therefore, we excluded the aforementioned report from our review of literature. Certain diagnosis of metastases by ACC, indeed, require the histological identification of two cell types, including ductal and modified myoepithelial cells. Immunohistochemically, ductal cells are positive for CK7, CEA and CD117, while myoepithelial cells are positive for P63, SMA, CK5/6, SMA, and Vimentin [22].

Clinicopathological features of the 12 cases, including our case, are reported in Table 1. 75% ($n=9$) of patients were female with a median age of 53 years [Interquartile Ranges (IQR) 22–83]. Of note, the reported female:male (F:M) ratio (3:1) appeared to be higher than the one reported for non-metastatic ACC (1.3:1) [23]; furthermore, it appeared to be not in line with the incidence of renal cell carcinoma (F:M ratio 1:2) [24]. Common primary site was lung (25%), followed by breast (17%) and lacrimal glands (17%) and median time from primary treatment to metastasis was 10 years (IQR 7–14). Treatment strategies mainly relied on radical asportation of the affected kidney. Liu et al. [12] reported the use of adjuvant chemotherapy after surgical treatment, consisting of six cycles of combined therapy with docetaxel and cisplatin. Based on the radiological characteristics of both tumors, the scarce incidence of ACC and the long time passed between the primary treatment and the metastases detection, we initially suspected a papillary renal carcinoma, and therefore, the left tumor was treated with robot-assisted partial nephrectomy (RAPN). Considering the exiguous dimensions of the contralateral tumor and the suspected histology [25], after a multidisciplinary consensus meeting, we decided to apply the same minimally invasive treatment to the other kidney. At last follow up, our patient has normal renal function (last serum creatinine

1.06 mg/dL) and no evidence of local or systemic recurrence at 18 months after surgery. To the best of our knowledge, this is the only case of bilateral renal metastases from ACC ever reported and the only case of kidney metastases from ACC treated with minimally invasive surgery.

Conclusion

We presented a case of bilateral renal metastasis from ACC, 14 years after the primary treatment. Kidney metastases should be considered in case of newly diagnosed renal tumors in patients with a history of ACC. Since metastasis may occur many years after primary treatment, lifelong follow-up, including abdominal imaging, is recommended.

Compliance with ethical standards

Conflict of interest All authors have declared that no conflict of interests exists.

Ethical standards All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research committee at which the studies were conducted (IRB approval number 93/2012/U/Oss) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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