Case Report

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RENAL METASTASIS FROM SUBMANDIBULAR GLAND ADENOCARCINOMA FIRST REPORTED PATIENT IN LITERATURE HISTORY

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ABSTRACT

This paper presents a patient of primary submandibular gland adenocarcinoma later presenting with renal metastasis. Renal cancers are known for their predisposition to unusual metastasis to other organs. On the contrary, renal metastasis from salivary gland tumours is extremely rare with no adenocarcinoma reported from the submandibular gland to date. Hence the significance of adequate history taking, immunohistochemical identification of pathology and multidisciplinary approach in the management of such rare clinical presentation is discussed here. The metastatic disease should always be a differential when evaluating cancer patients regardless of the interval since previous disease or rarity of occurrence.

Keywords: renal metastasis, submandibular gland tumour, adenocarcinoma, salivary gland cancer, rare metastasis

Renal metastasis from salivary gland tumours is extremely rare with no adenocarcinoma reported from the submandibular gland to date. On the contrary, renal cancers are known for their predisposition to unusual metastasis. We present a case of primary submandibular gland adenocarcinoma that resulted in renal metastasis.

CASE REPORT

A 59-year-old man presented to urology with a self-resolved painless visible hematuria with clots. He was a carpenter by profession, non-smoker, and had no family history of urological malignancies. His only medical history was excision of a 19-mm painless right submandibular gland lesion two-years previous for presumed pleomorphic adenoma on the basis of fine-needle aspiration. Histology reported incomplete excision of poorly differentiated adenocarcinoma with neuroendocrine activity and marked perineural spread. Therefore,

wide local excision and ipsilateral neck dissection were performed with several positive margins and 2 out of 6 lymph nodes showing metastasis. No distant metastasis noted. Postoperative adjuvant radiotherapy was given.

Following standard hematuria investigations, a new non-palpable 56-mm left-renal mass infiltrating the renal pelvis and lower pole pelvicalyceal system was reported on computed tomography (CT) urogram (Figure 1 and 2) which was biopsied. CT findings were similar to renal cell carcinoma with increased enhancement in the contrast phase. Interestingly histology revealed a poorly differentiated carcinoma with morphological and immunohistochemical similarity to the previous submandibular gland cancer. Histology slides were also sent to another tertiary centre for specialist review and validation. After extensive immunohistochemical examination, the final consensus from urology and ENT multidisciplinary team meetings was the diagnosis of metastatic adenocarcinoma

FIG. 1 Surveillance computed tomography showing bilateral normal kidneys in 2015.

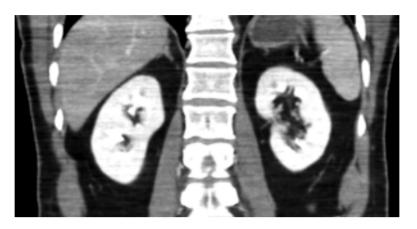
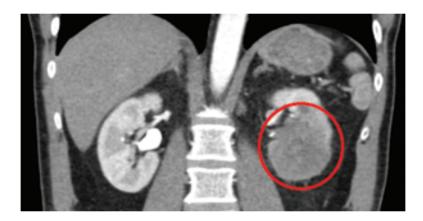


FIG. 2 Computed tomography urogram showing new left-renal lesion in 2016.



of submandibular gland with secondary involvement of the kidney.

Restaging CT scan detected multiple new small irregular speculate nodules in the right lung highly suggestive of metastasis. Due to this evidence of further non-renal metastasis palliative chemotherapy (epirubicin, cisplatin and capecitabine) was commenced. The patient had no further hematuria episodes. Since he is asymptomatic in follow up in relation to renal metastases, no further treatment required.

PATHOLOGY AND DISCUSSION

Literature review suggests renal cancer metastases to salivary glands are extremely rare and histologically either oncocytic or clear cell variants.^{2–5} The reverse metastases are more improbable with 2 reported case of adenoid cystic carcinoma of parotid gland resulting in late renal metastasis.^{1,6} Although renal metastasis from submandibular gland appears not to have been reported, renal metastasis from other salivary gland has, although still very rare.^{7,8} These were treated surgically with very limited benefits of radiotherapy, chemotherapy, and hormonal therapy.

In our case, the salivary gland tumour showed nests, tubules, and trabeculae of monotonous rounded cells with perineural invasion. Ki67 index was less than 30%. A renal biopsy also showed infiltrative tumour composed of nests and tubules of similar monotonous

cells with low mitotic activity. Kidney tumour cells are negative with CD10, p63, CK20, PSA, CD99, TTF1, GATA3, S100, calponin, caldesmon, synaptophysin, and chromogranin; focally positive for CK5/6, SMA, EMA, and diffusely and strongly positive with CK7, CK19, Vimentin, CD56 and WT1. The submandibular tumour was negative for p63, CK20, PSA, TTF1, GATA3, calponin, caldesmon, synaptophysin, and chromogranin and positive with CK5/6, SMA, EMA, CK7, CK19, 34betaE12, Vimentin, CD56 and WT1. Hence, morphology and immunohistochemistry support renal metastasis from submandibular gland cancer. Results were summarized in Table 1 and Figure 3. The rarity of this occurrence resulted in extensive immunohistochemical examinations and specialist reviews to confirm the final diagnosis.

CONCLUSION

This case report emphasizes the rare renal tumour, the significance of histological identification of primary cancer and metastasis as they form the basis of therapeutic decisions. The metastatic disease should always be a differential when evaluating cancer patients regardless of the interval since previous disease or rarity of occurrence. These tumours are difficult to be differentiated from standard renal cell carcinoma (RCC) purely by standard triple phase contrast CT. The learning points from this case report are careful history taking and the role of renal biopsy for these types of patients and careful liaison with histopathologists to differentiate it from RCC.

TABLE 1 Immunohistochemical Results of the Primary and Metastatic Lesions

Ihc	Results	
	Renal	Submandibular Gland
CD10	-	
P63	-	-
CK20	-	-
PSA	-	-
CD99	-	
TTF1	-	-
GATA3	-	-
S100	-	
CALPONIN	-	-
CALDESMON	-	-
SYNAPTOPHYSIN	-	-
CHROMOGRANIN	-	-
CK5/6	+	+
CK7	+	+
CK19	+	+
CD56	+	+
WT1	+	+
SMA	+	+
EMA	+	+
VIMENTIN	+	+
34betaE12		+

IHC RENAL SUBMANDIBULAR GLAND

CK5/6

WT1

H&E

FIG. 3 Comparative histological slides.

REFERENCES

- 1. Manoharan M, Gomez P, Reyes M, Soloway M. Metastatic adenoid cystic carcinoma to the kidney in a young woman. Urology 2006;68:1343e11–2.
- 2. Bedrosian SA, Goldman RL, and Dekelboum AM. Renal carcinoma presenting as a primary submandibular gland tumour. Oral Surg 1984;58:699–701.
- 3. Sist TC Jr, Marchetta FC, Milley PC: Renal cell carcinoma presenting as primary parotid gland tumour. Oral Surg 1982;53:499–502.
- Smits JG, Slootweg PJ. Renal cell carcinoma with metastasis to the submandibular and parotid glands. J Maxillofac Surg 1984;12;235–36.
- 5. Majewska H, Skalova A, Radecka K et al. Renal clear cell carcinoma metastasis to salivary gland A series

- of 9 cases: Clinico-pathological study. Pol J Pathol 2016;67(1):39–45.
- Brown JA and Swanson SK. Bilateral synchronous renal metastases in a patient 13 years status post resection of adenoid cystic carcinoma of the salivary gland. Urology 1998;51(2):1998.
- Nouraei SA, Ferguson MS, Clarke PM, et al. Metastasizing pleomorphic salivary adenoma. Arch Otolaryngol Head Neck Surg 2006;132:788–93.
- 8. Koyama M, Terauchi T, Koizumi M, et al. Metastasizing pleomorphic adenoma in the multiple organs: A case report on FDG-PET/CT imaging. Medicine (Baltimore) 2018 Jun;97(23):e11077.