

## A unique presentation of a rare renal cancer: Appearance of bone metaplasia in tubulocystic renal cell carcinoma

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### ABSTRACT

Tubulocystic renal cell carcinoma is a rare cancer that was not defined as a distinct entity until the early 2000s. Due to the recency of its classification, it remains poorly understood and leaves much room for future research. This report looks at a unique case of this rare subtype of renal cancer. This specific case is unique due to both the demographics of the affected patient and the finding of bone metaplasia within the tumor. We believe this is the first ever reported incidence of this phenomenon.

### Introduction

Tubulocystic renal cell carcinoma (TRCC) is a rare form of cancer that remained undefined until the early 2000s. It was previously referred to as low-grade collecting duct carcinoma. As of 2004, TRCC was not a recognized subtype of renal cell carcinoma (RCC) under World Health Organization (WHO) parameters. In 2009, a case series described the unique subtype of RCC.<sup>1</sup> The report analyzed 31 different cases of TRCC and helped define its common presentation. Although the data set was small, it successfully identified some significant trends; the disease has a strong predilection towards men, has a mean age of onset of 54 years, and ranges in size from 0.7 to 17 cm. Further, there has never been a reported case of TRCC with bone metaplasia present. This report describes a significantly unique case of this very rare disease.

### Case presentation

The patient is a 90-year-old female, who presented with a large abdominal mass noted on self-palpation. The patient has an extensive past medical history notable for pure hyperglyceridemia, essential hypertension, cardiac arrhythmia, and multiple UTIs. Her medications at initial presentation included dicyclomine, escitalopram, eszopiclone, and metoprolol succinate. She reported no history of smoking.

After initial presentation, a CT urogram was performed which showed a Bosniak IV renal cyst in the mid anterior left kidney that contained thick enhancing solid components concerning for renal

neoplasm (Fig. 1).<sup>2</sup> The mass was noted to be 16 × 16 cm, with no lymphadenopathy or other masses detected. The patient's right kidney appeared healthy. The patient was informed of the finding and counseled on possible therapy. Due to the size and aggressive appearance of the cyst, a Hand Assisted Laparoscopic Left Radical Nephrectomy was recommended.<sup>3</sup>

During the surgery, in order to visualize renal hilar structures, roughly 400 cc of viscous brown material was drained from the cyst. The aspirate had a heterogenous appearance of blood coagulum and protein. Following this, the affected kidney was removed. There were no affected lymph nodes or local metastases noted during the operation. The patient tolerated the procedure well and was discharged from the hospital on postop day 3 with a prescription for amlodipine, diclofenac, and docusate.

Pathology analyzed the removed left kidney. Macroscopically, the kidney weighed 875 g and measured 18 × 15 × 9 cm. The pathologist identified a large, well-circumscribed multicystic mass that extended laterally from the superior, middle, and a portion of the lower kidney. The cysts were of variable size with no solid areas between. The main cyst measured 17 × 13 × 12 cm. The cyst was bisected revealing extensive coagulative necrosis. The fluid that was drawn from the cyst intraoperatively was analyzed, showing macrophages, lymphocytes, and blood without the presence of malignancy. The internal surface of the cyst had an irregular, spongy surface with a friable lining including areas that resembled tumor mixed with blood clots. The largest segment that resembled tumor measured 4 × 3 × 2 cm. The cyst was compressing the

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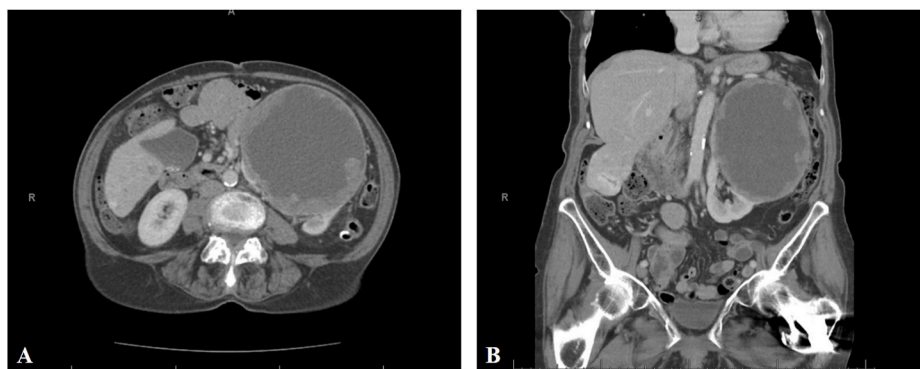
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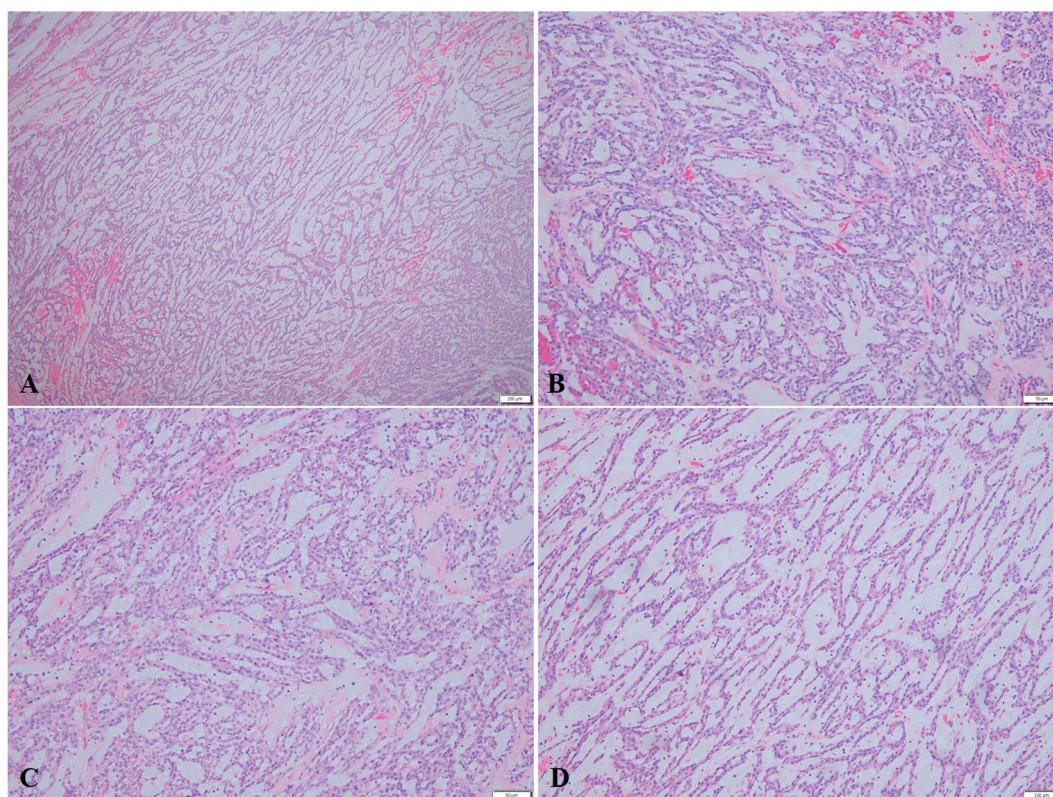
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**Fig. 1.** These images taken during the nephrogenic phase of the CT urogram show the large cystic mass. **Fig. 1B** highlights the extent to which the cystic mass effaces the left kidney.



**Fig. 2.** **Fig. 2A** shows a histology cross section from the mass at 4X magnification. **Fig. 2B–D** displays the unique histologic characteristics and architecture of TRCC at 10X magnification.

renal pelvis and sinus, distorting the normal renal anatomy. It was noted that the cyst was encapsulated and did not invade the renal sinus or vein. Upon further analysis, the tumor was identified as a WHO/ISUP Grade 2 TRCC via pathologic examination that showed tubular and cystic architecture, fibrotic septa, and septa lined with a single layer of cuboidal cells that stain strongly eosinophilic (**Fig. 2**).<sup>4</sup> The presence of focal metaplastic bone formation and oncocytic features were noted (**Fig. 3**). No sarcomatoid transformation was identified.<sup>5</sup> Extensive hemorrhage, granulation tissue, and giant cells were also noted. The diagnosis of TRCC was made based on macroscopic and microscopic features by two separate pathologists.

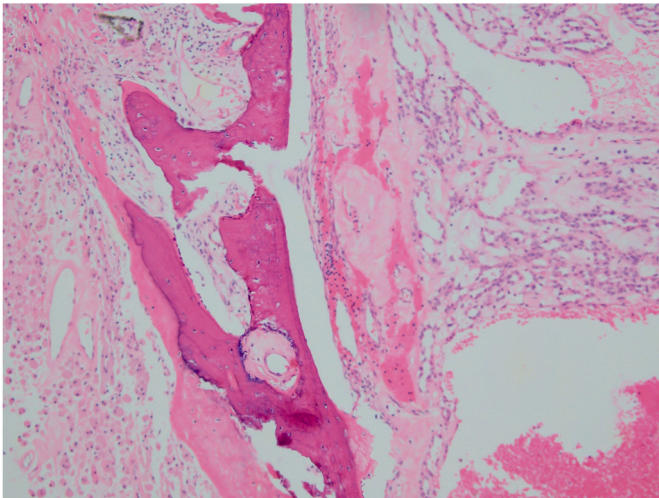
Follow-up CT scans 3 months post-op showed no lymphadenopathy and an intact vascular stump. Her remaining kidney appeared healthy based on imaging and a serum creatinine of 0.9 mg/dL. The patient was informed of these results. She reported no adverse events since discharge

and was pleased with the outcome.

## Discussion

This is a unique case of a remarkably rare cancer. Not only was it found in a patient outside of the classic demographic, but we believe it is the first ever reported occurrence of bone metaplasia in a TRCC. While bone metaplasia has been reported in other subtypes of RCC, this is potentially the first ever reported case of the phenomenon in TRCC. The presence of bone metaplasia was noted by the pathologist in her report, and images of the slides confirm its presence. After extensive research, we were unable to identify another reported case such as this. While the bone metaplasia is the most unique finding in this case, it is far from the only one.

Our patient was significantly older than most patients who present



**Fig. 3.** This figure clearly displays the presence of bone metaplasia seen during pathological examination of the primary cystic mass.

with a TRCC. As mentioned before, the average age of patients with this pathology was 54. In a sample size of 31 cases, all patients were between the ages of 34–74. Our patient was 16 years older than the oldest patient from Amin's influential case series. In that same study, it was reported that men were more susceptible to this disease than women at a ratio of 7:1. Finally, this particular mass is one of the largest TRCC reported. It is approximately the same size as the largest mass from the previously mentioned case series. Given the vague clinical picture of this case, other diagnoses were considered, but we are confident in the diagnosis of

TRCC when considering the macroscopic and microscopic findings.

### Conclusion

This case study describes an incredibly unique presentation of an extremely rare cancer. We believe that this is the first described case of bone metaplasia within a tubulocystic renal cell carcinoma.

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No.

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