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Case Report

Metanephric Adenoma Associated with Unusual Urinary Candidiasis: A Challenging Morbidity from Postoperative Anastomotic Leak

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ABSTRACT

Metanephric adenoma (MA) of the kidney is a rare benign neoplasm, which is mostly incidental-discovered during imaging studies for other clinical problems. However, this tumor may overlap in morphology with the papillary renal cell carcinoma and there are descriptions of metastatic disease. To date, fewer than 200 cases of MA have been reported worldwide and usually have a good prognosis. In the current report, a case of MA in a middle-aged lady is presented, which developed postoperative morbidity resulting from prolonged perinephric leakage secondary to urinary and perinephric fungus infection as a part of systemic candidiasis. The clinical, morphological and immunohistochemical features are presented together with a review of the current literature.

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Introduction

Metanephric adenoma (MA) is an extremely rare benign neoplasm of the kidney, representing 0.2% of adult renal epithelial tumors [1]. Most cases are seen in the fifth and sixth decades, predominantly in females by a 2:1 ratio [2]. It may have some overlapping features with the solid variant of papillary renal cell carcinoma and is often difficult to distinguish from malignant neoplasms. Immunohistochemistry seems to be sufficient in the diagnosis of MA in the majority of cases and it usually has a good prognosis. Li *et al.* demonstrated multiple variants of MA, suggesting a wider classification spectrum than originally described, which advocate timely surgical treatment in all patients [3]. Partial nephrectomy is a highly recommended treatment option, despite some cases presented with metastatic disease [3].

Invasive fungus infection of the kidney is unusual and is difficult to treat. The guidelines of the Infectious Diseases Society of America (IDSA) recommended empirical antifungal therapy for patients with recent abdominal surgery anastomotic leaks [4]. This report describes a case of

a MA associated with urinary candidiasis, resulting in unpredicted morbidity after successful partial nephrectomy.

Case Presentation

A 42-year-old non-smoker lady was referred to our tertiary care center after incidentally being discovered to have a right renal mass. She has recurrent aching pain localized to the right loin. The patient has no hematuria, bothersome voiding symptoms, or any constitutional symptoms. She was medically free with no past surgical history apart from cesarean sections with a current breast mass. There was no family history of malignancy or relevant medical conditions. Physical examination was unremarkable, and all preoperative laboratory workup was within the average range. Serum creatinine was 0.48 mg/dL, haemoglobin (Hb) level was 12.8 g/dL, and WBCs was $4.68 \times 10^9/L$. Contrast-enhanced CT of the abdomen and pelvis showed a $3.5 \times 4 \times 4$ cm, well-defined exophytic and lobulated soft tissue mass arising from the lateral aspect of the mid pole of the right kidney, which was inseparable from the adjacent liver. There was no evidence of tumor thrombus, lymphadenopathy or radiological signs of distant metastasis (Figure 1).

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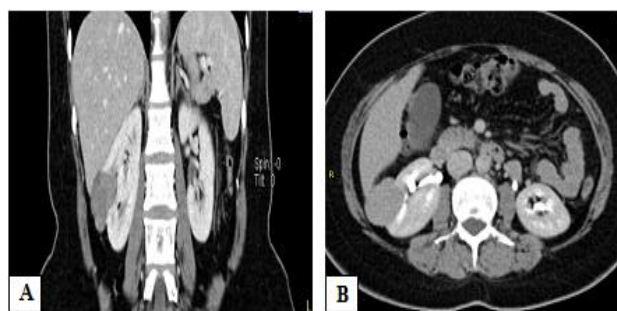


Figure 1: A) Coronal and B) axial views demonstrate a well-defined exophytic lobulated soft tissue mass lesion arising from the lateral aspect of the mid pole of the right kidney. It measures 3.5×4×4 cm in its maximum anteroposterior, transverse and craniocaudal dimensions, and it is inseparable from the adjacent liver.

The patient underwent trans-peritoneal partial nephrectomy with insertion of an intraoperative ureteral stent. Postoperatively, the patient developed low-grade fever with a drop of haemoglobin to 9.2 gm/dl. She reported paravaginal itching with vaginal secretion, whereas the urine culture showed moderate growth of *Candida albicans*, which was appropriately managed with local and systematic antifungals. The patient was discharged home in a good general condition with serum creatinine of 0.6 mg/dl, normal WBCs count, and Hb level of 10.3 gm/dl. The bilateral screening mammogram showed right breast skin calcification with no suspicious malignant findings (BIRADII). Histopathology revealed morphological features of metanephric adenoma of 6.0×5.0×2.5 cm, which was 0.5 cm away from the excision margin and reaching up to the outer painted renal capsule (Figures 2A-2C). Properly controlled immunohistochemical showed the focal patchy positive CK-7 in tumor cells with negative EMA and CD-57 in tumor cells (Figure 2D).

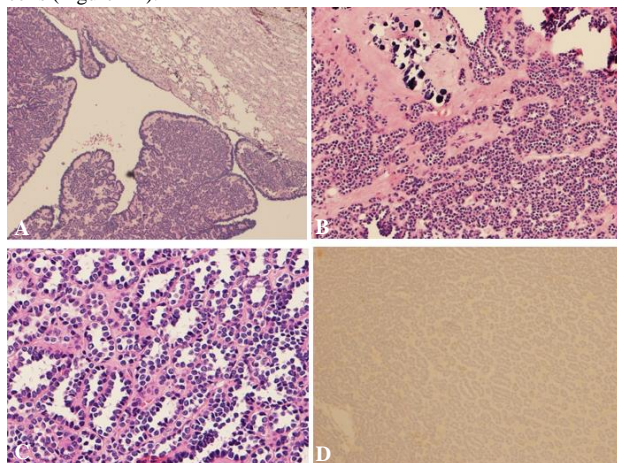


Figure 2: Non encapsulated, multinodular tumor composed of small, uniform, closely packed tubules and papillae lined by small cells with minimal cytoplasm, bland nuclei that may overlap, uniform chromatin and no mitoses was seen. Psammomatous calcification and fibrosis were seen at places. A) Adenoma with adjacent normal kidney 4X, B) metanephric adenoma 10 X, C) metanephric adenoma 20X and D) negative CK 7 and EMA immunohistochemical staining.

Four-week later, the patient came to the ER with fever of 39.6°C and remittent right loin pain, which was aggravated by inspiration and relieved by NSAID. Serum creatinine was 0.76 mg/dl, Hb was 11.6 g/dl,

and WBCs was $10.11 \times 10^9/L$. CT/KUB revealed perinephric localized collection of 4.6×3.7×5.6 cm attached to the surgical bed, with normal enhancement of the right kidney, with no residual tumor local recurrence (Figures 3A & 3B). This finding has not been detected in the postoperative contrast imaging performed on the 5th postoperative day. Percutaneous drainage of the perinephric collection was performed guided by CT with insertion of a pigtail catheter, while the ureteral stent was removed under local anaesthesia. During cystoscopy, the bladder showed heavy pyuria with precipitation of purulent *Candida albicans* biofilm on the ureteral stent, as approved by culture and sensitivity, especially at its renal end (Figure 3C). Postoperative urinary leakage continued for 14 days, where its creatinine contents was <10 mg/dl. The culture of the perinephric fluid showed heavy *Candida albicans*, similar to the high vaginal swab, which was managed with fluconazole orally daily for 7 days.

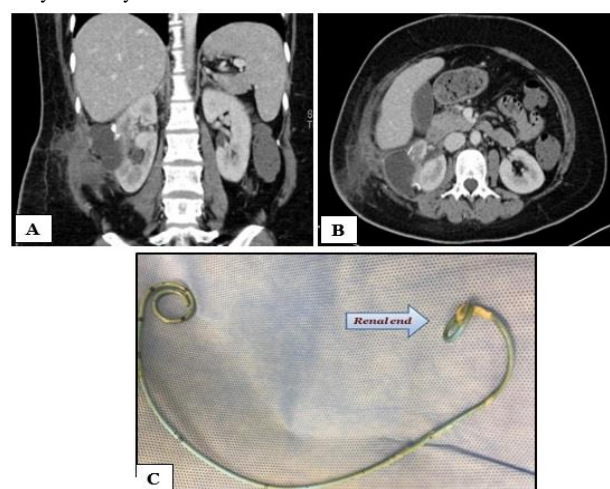


Figure 3: A & B) Coronal and axial imaging demonstrate right perinephric collection measuring about 4.6×3.7×5.6 cm. No obvious local recurrence or residual tumor. The right kidney demonstrates normal enhancement with no obvious focal lesion, with an unremarkable appearance of the left kidney. C) The double-J ureteral stent showed precipitation of purulent biofilm on the renal end of the ureteral stent.

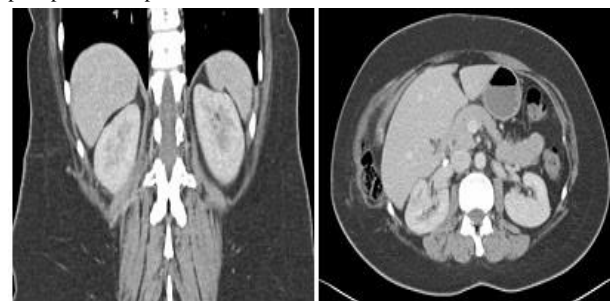


Figure 4: Coronal and axial imaging after complete resolution of the right perinephric leak. No obvious local recurrence or residual tumor. The right kidney demonstrates normal enhancement with no obvious focal lesion, with an unremarkable appearance of the left kidney.

The tube drain was removed 48-hours of complete non-drainage, and a contrast-enhanced CT scan demonstrated normal enhancement of the right kidney and complete resolution of the previously mentioned perinephric collection. Follow-up till 10-month, showed normal kidney function and complete blood count. Last contrast-enhanced CT study revealed no local recurrence or distant metastasis (Figure 4).

Discussion

MA of the kidney was classified by the World Health Organization (WHO) as an epithelial benign tumor, which generally occurs in adults. However, it may include heterogeneous histological subtypes, with the coexistence of malignant components [3]. Nephron-sparing surgery is the treatment option of choice, with an excellent prognosis. In the current indexed case, partial nephrectomy was performed, and the postoperative contrast-enhanced CT showed no leakage. At the early postoperative course, the patient had severe pruritis vulva with whitish vaginal discharge, which was diagnosed by a gynaecologist as a fungal infection necessitating local antifungals. It seems that this was the source of ascending renal infection with *Candida albicans*, as proved later with urine culture and sensitivity. Despite the fact that funguria may be common in hospitalized patients, invasive kidney infection is unusual and is difficult to manage. Risk factors in this patient, including indwelling urethral catheter, intraoperative ureteral stent, and prior antibiotic therapy, may predispose to such infection [5]. Symptoms and signs may include flank pain, costovertebral angle and tenderness, which were the presenting manifestations after discharge.

Complications associated with fungal infections of the kidneys include fungus ball formation, perinephric abscess and surgical wound infection. Prolonged urinary leak from the partial nephrectomy bed seems to result from renal candidiasis; therefore, the ureteral stent was removed once the infection was confirmed, with discontinuation of antibiotics to reduce risk factors for acquisition of candiduria. Drainage of the perinephric collection alone may cure some patients with fungus infection. However, the guidelines of the Infectious Diseases Society of America (IDSA) recommended empirical antifungal therapy for patients with recent abdominal surgery, especially if they have anastomotic leak [4]. Considering the positive culture for *Candida albicans* from urine and perinephric collection, oral fluconazole was started, especially it will target the most likely species and will achieve adequate urinary concentration. After intravenous administration, fluconazole achieves equal serum and peritoneal concentrations [4]. Intravenous amphotericin B deoxycholate is an accepted alternative to fluconazole at a dose of 0.3-0.6 mg/kg daily.

Conclusion

Metanephric adenoma is a rare entity, which is managed by partial nephrectomy with good prognosis. Attention should be focused upon reducing the risk factors for candidiasis in surgical patients, such as removal of catheters and ureteral stents and discontinuation of antibiotics. Systemic antifungal is recommended for all patients with symptomatic candiduria.

Consent

A written consent was obtained from the patient for publication of this case report and accompanying images.

Conflicts of Interest

None.

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