

# Metanephric Adenoma in a Solitary Kidney: A Case Report and Review of the Literature

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## Abstract

Metanephric Adenoma is a rarely observed benign-character renal tumor. Most of the time insufficiencies in preoperative evaluations lead to unnecessary surgical intervention which makes these metanephric adenomas important from the point of view of clinicians. Histopathologically, differential diagnosis of this tumor generally includes papillary cell renal carcinoma and Wilm's Tumor. Radiologically there are a limited number of studies related to metanephric adenoma. Though metanephric adenoma is a benign mass, patients follow-up should be carefully undertaken. We present a 67-year old female patient with metanephric adenoma treated with robot-assisted laparoscopic partial nephrectomy in a solitary kidney.

## Introduction

Metanephric Adenoma (MA) is a benign-character kidney tumor rooted in residual embryonic metanephric blastema. This very rare tumor comprises 0.2% of renal epithelial-sourced kidney tumors [1]. This tumor was first described by Brisigotti et al. in 1992 and was accepted as a primary benign renal tumor according to Heidelberg classification in 1997 [2-3].

Most of the time insufficiencies in preoperative evaluations lead to unnecessary surgical intervention which makes these MAs important from the point of view of clinicians. To the best of our knowledge there are nearly 200 cases reported in the literature. In this study we present a 67-year old female patient with MA in a solitary kidney.

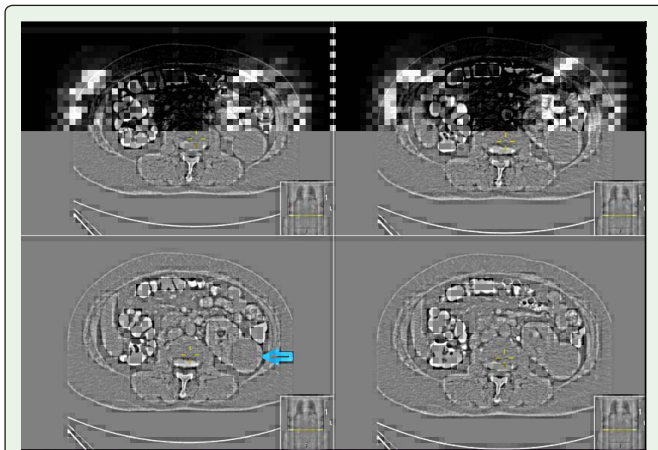
## Case Report

A 67-year old female patient with solitary kidney applied to our clinic with complaint of flank pain for nearly 6 months. Anamnesis found previous history of kidney donation and diagnoses of type-2 diabetes mellitus, hypertension and hyperthyroidism. Physical examination found negative cost vertebral angle tenderness and no palpable mass. Vital signs were stable and routine laboratory tests were hemoglobin: 13g/dl, creatinine: 1.01mg/dl, calcium: 9.1mg/dl, urine analysis: 18p/hpf erythrocytes with no other abnormality identified. Urinary system ultrasonography observed a 4.7x5.1 cm hyperechoic mass lesion in the lower-middle pole of the left kidney. Dynamic renal computed tomography taken after this reported hypodense semi-solid lesion in the central section of the left kidney protruding from the cortex, with maximum diameter 5.5cm; on postcontrast series there was a thick rim in the periphery with heterogeneous contrast in the center (Figure 1). The Renal (radius, exophytic vs endophytic properties, nearness of tumor to the collecting system or sinus, anterior vs posterior, location relative to polar lines) nephrometry score was 7, Chest X-ray was normal.

When the available clinical findings were evaluated, a nephron-sparing surgical procedure was planned for the patient. Robot-assisted standard partial nephrectomy with warm ischemia duration 24 minutes, total surgery duration 90 minutes and total blood loss 50ml was completed without complications (Figure 2). The tumor was cut out with a 1cm margin of normal renal parenchyma surrounding it. After the resection of the tumor and surrounding parenchyma, a renorrhaphy was required to close the collecting system defect, oversaw bleeding vessels and closure of the renal parenchymal residual defect. Mobilized on the 1st day postoperative, the patient was discharged in full health on the postoperative 3rd day with hemoglobin:11.7g/dl, creatinine: 0.97mg/dl. The creatinine value is 1.09mg/dl in 6 months follow-up, while the GFR calculated by the MDRD method was 53 ml/min/1.73m<sup>2</sup>.

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**Figure 1:** Hypodense semi-solid lesion in the central section of the left kidney protruding from the cortex, on postcontrast series there was a thick rim in the periphery (blue arrow) with heterogeneous contrast in the center.



**Figure 2:** Intraoperative view (The tumor protruding from the cortex).



**Figure 3:** A) Macroscopic appearance B) Haematoxylin and Eosin (H&E)-stained section of metanephric adenoma C) Immunohistochemical staining with diffuse positive WT<sub>1</sub>.

Macroscopic investigation of the partial nephrectomy specimen observed a dirty-white color tumor of 6x6x5.5 cm with lobule contours, separated from the adjacent tissues of the renal capsule by smooth boundaries. Microscopic investigation observed separation from non-tumoral renal parenchyma by a fibrous capsule and tumoral cellular proliferation formed of tightly packeted small a sinus and tubules. The tumor was uniform, comprised of slightly

eosinophilic-scarce cytoplasm, small rounded oval nuclei, without definitions of nucleoles, thin-chromatine cells. Atypia, necrosis and mitosis were not observed in the tumor. Tumor cells were studied with immunohistochemical methods and stained strongly diffuse positive with CD<sub>57</sub>, diffuse positive WT<sub>1</sub>, negative EMA, negative Alpha-Methylacyl-Coa Racemase (AMACR), negative CD<sub>10</sub>, negative RCC, negative CK<sub>7</sub>, and negative desmin. Accompanied by histopathologic and immunohistochemical findings, it was reported as “metanephric adenoma” (Figure 3).

## Discussion

MA is a rarely observed benign-character renal tumor. MA is identified in females at 2 times the rate for males and affects people of wide age interval (5-83 years). Tumor size varies from 0.3 to 15 cm. While it is identified incidentally in half of cases, patients may present such conditions as flank pain, hematuria and palpable mass. In 12% of patients with MA erythrocytosis is identified [4]. Erythrocytosis was shown to develop via multiple cytokines secreted by tumor cells [5]. Additionally, though rare, there are cases reported with hypercalcemia and chyluria [6-7].

Histopathologically, differential diagnosis of this tumor generally includes Papillary Cell Renal Carcinoma (PCRC) and Wilm’s Tumor (WT). The tumor macroscopically shows solid growth pattern, with a homogenous surface not containing necrosis and hemorrhage. Tumors are generally yellowish-white or gray in color and are surrounded by a capsule containing solid or cystic material [8]. Microscopically the tumor cell nuclei are oval and have a smooth appearance. The tumor does not include mitosis, but has a well-differentiated adenomatous pattern. It is different from WT as it does not contain any blastema [2]. Immunohistochemistry shows diffuse strong staining especially for CD<sub>57</sub>, WT<sub>1</sub> and S<sub>100</sub>, while CK<sub>7</sub> is rare and EMA and AMACR are negatively stained. Negative staining with CD<sub>56</sub> differentiates the tumor from WT, while CK<sub>7</sub> and AMACR are usually not expressed or only focally expressed in MA which allows exclusion of PRCC [9].

After definition of this rare entity, studies at molecular and genetic level are increasing continuously. The most common variations observed with MA are BRAF v600e gene mutation and 2p16 gene changes. A comprehensive study by Calio et al. identified BRAF v600e gene mutation in 41 out of 48 cases with MA [10]. It is thought that deletion of BRAF v600e and 2p gene shows synergic effect for development of MA [11]. Additionally gains in the 7th and 17th chromosome and loss of the Y chromosome were identified to have a role in development of this tumor, with these traits showing similarity to PCRC in terms of genetics [12].

Radiologically there are a limited number of studies related to MA. The majority of these tumors are hyperechoic on ultrasonography, while they may also be observed as hypoechoic or isoechoic [13]. On color Doppler ultrasonography blood flow is not observed, or reduced. On computed tomography, these tumors have regular boundaries, and appear hyperdense compared to normal renal parenchyma. The peripheral section of the mass contains attenuation at a higher rate compared to the central section [14]. MA has higher calcification rates compared to other renal masses and on radiology these findings were stated to be an important clue [15]. Contrary to this, one study assessed 18 MA patients radiologically and identified calcification in only 1 patient. Again the same study divided the 18 patients into

10 with pure MA and 8 with MA with malignant component and emphasized that the most important determinant difference was enhancement pattern [8]. Radiologically it does not appear possible to differentiate these masses as malignant or benign yet.

The benign nature of MA has led to questions on whether a preoperative renal biopsy is necessary or not. When the literature is viewed, there are cases of diagnosis with Fine Needle Aspiration Biopsy (FNAB). Blanco et al. diagnosed MA from FNAB and 20 months follow-up showed that the mass was stable on imaging [16]. Again, the lack of a specific finding for MA on preoperative imaging makes patient choice for biopsy complicated. Additionally, treatment may be misdirected due to problems such as insufficient material from aspiration biopsy and pathological confusion of MA with WT and PRCC. An example of this is given by Barroca et al. who identified a mass on the right kidney with radiologically in an 8-year old male child applying with vomiting complaints. FNAB was identified Wilm's tumor, so the patient was administered chemotherapy. Then radical nephrectomy was performed. Pathological specimen reported as metanephric adenoma [17].

Thompson et al. in a study of 2675 patients showed that as the size of renal tumors increased the risk of malignancy increased [18]. In this study, 90% of tumors from 5 to 6 cm in size were malignant. In our case, due to the large tumor size FNAB was not performed.

Though MA is a benign mass, patients follow-up should be carefully undertaken. Renshaw et al. reported a 7-year old female child with paraortic, hilar and aortic bifurcation lymph node metastasis [19]. Death linked to tumor was also reported for a female patient with mixed MA containing sarcomatoid structure [20].

## Conclusion

MA is a rarely observed renal tumor. As it does not have specific radiological findings, diagnosis of this benign tumor is difficult, as in our case. This case report reveals the necessity of considering FNAB for special situations like solitary kidney even with a large size tumor to prevent unnecessary treatment.

## References

- Amin MB, Tamboli P, Javidan J, Stricker H, de-Peralta VM, Deshpande A, et al. Prognostic impact of histologic subtyping of adult renal epithelial neoplasms: an experience of 405 cases. *Am J Surg Pathol.* 2002; 26: 281-291.
- Brisigotti M, Cozzutto C, Fabbretti G, Sergi C, Callea F. Metanephric adenoma. *Histol Histopathol.* 1992; 7: 689-692.
- Kovacs G, Akhtar M, Beckwith BJ, Bugert P, Cooper CS, Delahunt B, et al. The Heidelberg classification of renal cell tumours. *J Pathol.* 1997; 183: 131-133.
- Davis CJ Jr, Barton JH, Sesterhenn IA, Mostofi FK. Metanephric adenoma. Clinicopathological study of fifty patients. *Am J Surg Pathol.* 1995; 19: 1101-1114.
- Yoshioka K, Miyakawa A, Ohno Y, Namiki K, Horiguchi Y, Murai M. Production of erythropoietin and multiple cytokines by metanephric adenoma results in erythrocytosis. *Pathol Int.* 2007; 57: 529-536.
- Mahoney CP, Cassady C, Weinberger E, Winters WD, Benjamin DR. Humoral hypercalcemia due to an occult renal adenoma. *Pediatr Nephrol.* 1997; 11: 339-342.
- McNeil JC, Corbett ST, Kuruville S, Jones EA. Metanephric adenoma in a five-year-old boy presenting with chyluria: case report and review of literature. *Urology.* 2008; 72: 545-547.
- Li G, Fu F, Song H, Niu Y, Su Y. CT imaging spectrum and the histopathological features of adult metanephric adenoma. *Br J Radiol.* 2015; 88: 1051.
- Mantoan Padilha M, Billis A, Allende D, Zhou M, Magi-Galluzzi C. Metanephric adenoma and solid variant of papillary renal cell carcinoma: common and distinctive features. *Histopathology.* 2013; 62: 941-953.
- Calliò A, Eble JN, Hes O, Martignoni G, Harari SE, Williamson SR et al. Distinct clinicopathological features in metanephric adenoma harboring BRAF mutation. *Oncotarget.* 2016; 8: 54096-54105.
- Dadone B, Ambrosetti D, Carpentier X, Duranton-Tanneur V, Burel-Vandenbos F, Amiel et al. A renal metanephric adenoma showing both a 2p16e24 deletion and BRAF V600E mutation: A synergistic role for a tumor suppressor gene on chromosome 2p and BRAF activation?. *Cancer Genet.* 2013; 206: 347-352.
- Brown JA, Anderl KL, Borell TJ, Qian J, Bostwick DG, Jenkins RB. Simultaneous chromosome 7 and 17 gain and sex chromosome loss provide evidence that renal metanephric adenoma is related to papillary renal cell carcinoma. *J Urol.* 1997; 158: 370-374.
- Fielding JR, Visweswaran A, Silverman SG, Granter SR, Renshaw AA. CT and ultrasound features of metanephric adenoma in adults with pathologic correlation. *J Comput Assist Tomogr.* 1999; 23: 441-444.
- Bastide C, Rambeaud JJ, Bach AM, Russo P. Metanephric adenoma of the kidney: clinical and radiological study of nine cases. *BJU Int.* 2009; 103: 1544-1548.
- Ojea Calvo A, Rodríguez A, Pérez, D Domínguez F, Alonso Rodrigo AE, Rodríguez Iglesias B. Metanephric adenoma. A new embryonal tumor of the kidney. *Actas Urol Esp.* 1998; 22: 150-153.
- Blanco LZ Jr, Schein CO, Patel T, Heagley DE, Cimbalk DJ, Reddy V et al. Fine-needle aspiration of metanephric adenoma of the kidney with clinical, radiographic and histopathologic correlation: A review. *Diagn Cytopathol.* 2013; 41: 742-751.
- Barroca HM, Cirnes L, Lopes JM. Metanephric adenoma: Cytological, histological, and molecular diagnosis of a case. *Diagn Cytopathol.* 2016; 44: 263-266.
- Thompson RH, Kurta JM, Kaag M, Tickoo SK, Kundu S, Katz D, et al. Tumor size is associated with malignant potential in renal cell carcinoma cases. *J Urol.* 2009; 181: 2033-2036.
- Renshaw AA, Freyer DR, Hammers YA. Metastatic metanephric adenoma in a child. *Am J Surg Pathol.* 2000; 24: 570-574.
- Picken MM, Curry JL, Lindgren V, Clark JI, Eble JN. Metanephric adenosarcoma in a young adult: morphologic, immunophenotypic, ultrastructural, and fluorescence in situ hybridization analyses: a case report and review of the literature. *Am J Surg Pathol.* 2001; 25: 1451-1457.