The imaging features of metanephric adenoma: a case report and review of literature

Abstract: Metanephric adenoma (MA) is a rare epithelial tumor of the kidney with a characteristic histology. To date, the imaging features of the tumor have not been clearly described. Until now, MA was considered to be benign, but the majority of MA cases underwent nephrectomy. Here, we report a case of MA confirmed by surgical pathology, and we will analyze the ultrasound and computed tomography findings. The radiological features of MA are presented along with a brief review of the pertinent literature to deepen the understanding of MA's imaging features.

Keywords: metanephric adenoma, X-ray computed tomography, histology, ultrasound

Introduction
Metanephric adenoma (MA) is an uncommon tumor of the kidney that can be found in both children and adults, and which shows a characteristic histology. To date, only about 100 cases have been reported, and there are very few detailed radiological reports. MA is not well recognized by either clinicians or radiologists, and the clinical and imaging features of MA have not been well illustrated. Currently, MA is considered to be benign, but the majority of MA cases undergo nephrectomy. Here, we report a case of MA in a 38-year-old male and describe their appearances on computed tomography (CT) and ultrasonography. We also reviewed all of the previous literature involving 17 cases with MA to provide a comprehensive understanding of the imaging features of MA.

Case report
An unspecified lesion in the left kidney was detected in a 38-year-old male by sonogram in a remote hospital during a physical examination 5 years ago. This patient was not treated since he felt no particular discomfort. A noncontrast CT examination revealed that the left kidney lesions became larger at the same hospital 1 month ago, measuring 3.5 cm × 3.5 cm. Three days before admission to Tangdu Hospital, the patient started to have left-flank pain and discomfort without obvious incentive, no fever, chills, nausea, or vomiting. The patient underwent further examination at the Tangdu Hospital.

An ultrasound examination showed a 3.2 cm × 4.5 cm liquid dark area in the lower pole of the left kidney. An irregular 2.4 cm × 2.1 cm, slightly hyperechoic mass was visible in the liquid dark area without acoustical shadowing, which was connected to the left kidney extensively (Figure 1). CDFI (color doppler flow imaging) showed no significant blood flow. Because it was not certain if the nonshadowing hyperechoic mass represented hemorrhage or calcification, CT imaging was performed for further characterization.
Cancer remains one of the most deadly diseases, despite extensive research on its etiology, diagnosis, and treatments. As a rare form of cancer, metanephric neoplasms are an uncommon renal tumor that generally occurs in adults and is considered to have a benign course. It was first proposed and named by Brisigotti et al in 1992. It has recently been identified and classified in the group of metanephric tumors, which also includes metanephric adenofibroma and stromal metanephric tumors. MA often presents as asymptomatic, although symptoms can include abdominal pain, abdominal palpable mass, hematuria, or fever. Among renal lesions, MA has the highest incidence (12%) of polycythemia. Although MA is usually benign, a few cases with atypical histological and distant metastasis have been reported.

Pathologically, MA is often unilateral and solitary, varying in size, is well defined, noninvasive, hard, or pink, and nodular, and it may be associated with hemorrhage, cystic degeneration, and necrosis. Microscopically, MA is composed of very small epithelial cells with small regular nuclei; it has a high nuclei-to-cytoplasm ratio and there are no mitotic figures, which form very small acini in an acellular stroma. Less often, they form tubular, glomeruloid, or polypoid and papillary formations. Most also show evidence of regression in the form of scarring and calcification. In addition, immunohistochemical staining with CD57, WT1, and CK7 may be useful to differentiate MA from papillary renal cell carcinoma (PRCC) when distinction by histological features alone proves difficult.

The imaging features of MA include a lack of specificity, which has been reported in some cases. Ultrasound images of MA have been described as hyperechoic, isoechoic, hypoechoic, or heterogeneous echotexture. Only one case was reported where ultrasound demonstrated a cystic renal mass. Our case showed a slightly hyperechoic mass in the liquid dark area without acoustical shadowing. CDFI shows no significant blood flow.

On CT scan, these tumors are consistently well defined, and they do not have distinct attenuation patterns; they are mostly spontaneous and slightly hyperdense in comparison to the normal adjacent renal parenchyma, and they sometimes present as an isodense or hyperdense mass. Calcifications of various sizes, as well as cysts can be seen. Mostly, peripheral areas of the mass had significantly higher attenuation when compared to the central portion, suggestive of central necrosis. However, our case showed lower attenuation in the peripheral areas compared to the central portion, representative of peripheral necrosis and hemorrhage.

Figure 1 Longitudinal sonogram of the left kidney. Notes: Longitudinal sonogram shows a 3.2 cm × 4.5 cm liquid dark area (C) in the lower pole of the left kidney (LK). An irregular 2.4 cm × 2.1 cm, slightly hyperechoic mass (N) is visible in the liquid dark area without acoustical shadowing, which was extensively connected to the left kidney.

An unenhanced CT scan showed a well-defined, high-density mass measuring 3.7 cm × 3.3 cm in the lower pole of the left kidney, with an irregularly shaped lower attenuation area in the mass (Figure 2A). On contrast-enhanced CT scan, the mass appeared to contain necrotic or hemorrhagic material, as demonstrated by the unenhanced peripheral areas in the mass, and this was confirmed by the findings of the ultrasound examination. Progressive enhancement in the central portion was observed, with 46 Hounsfield units (HU) before contrast enhancement, and 64 HU, 81 HU, and 82 HU on the cortex-phase, medullary-phase, and delayed-phase images, respectively (Figure 2B–D). No other remarkable findings were identified in the abdomen and thorax.

Open and radical nephrectomy was performed. The left radical nephrectomy specimen revealed a 3.0 cm × 3.5 cm, well-circumscribed, oval cystic–solid mixed mass bulging out of the cortex of the lower pole of the left kidney, with necrotic or hemorrhagic material. Microscopically, the histological specimen showed homogeneous round or ovoid tumor cells, mainly composed of tiny tubules and papillae, accompanied by very scanty stroma. The tumor cells were negative for immunohistochemical staining with WT1 and CD57. The histological features of the tumor were characteristic for MA (Figure 3). The diagnosis of MA was established.

Discussion
Cancer remains one of the most deadly diseases, despite extensive research on its etiology, diagnosis, and treatments. As a rare form of cancer, metanephric neoplasms represent a spectrum of differentiated lesions that seem most likely to be related to Wilms’ tumor. These neoplasms include a pure stromal lesion, a pure epithelial lesion (MA), and a mixed epithelial and stromal tumor (MEST). MA is an uncommon renal tumor that generally occurs in adults and is considered to have a benign course. It was first proposed and named by Brisigotti et al in 1992. It has recently been identified and classified in the group of metanephric tumors, which also includes metanephric adenofibroma and stromal metanephric tumors. MA often presents as asymptomatic, although symptoms can include abdominal pain, abdominal palpable mass, hematuria, or fever. Among renal lesions, MA has the highest incidence (12%) of polycythemia. Although MA is usually benign, a few cases with atypical histological and distant metastasis have been reported.

Pathologically, MA is often unilateral and solitary, varying in size, is well defined, noninvasive, hard, or pink, and nodular, and it may be associated with hemorrhage, cystic degeneration, and necrosis. Microscopically, MA is composed of very small epithelial cells with small regular nuclei; it has a high nuclei-to-cytoplasm ratio and there are no mitotic figures, which form very small acini in an acellular stroma. Less often, they form tubular, glomeruloid, or polypoid and papillary formations. Most also show evidence of regression in the form of scarring and calcification. In addition, immunohistochemical staining with CD57, WT1, and CK7 may be useful to differentiate MA from papillary renal cell carcinoma (PRCC) when distinction by histological features alone proves difficult.

The imaging features of MA include a lack of specificity, which has been reported in some cases. Ultrasound images of MA have been described as hyperechoic, isoechoic, hypoechoic, or heterogeneous echotexture. Only one case was reported where ultrasound demonstrated a cystic renal mass. Our case showed a slightly hyperechoic mass in the liquid dark area without acoustic shadowing. CDFI shows no significant blood flow.

On CT scan, these tumors are consistently well defined, and they do not have distinct attenuation patterns; they are mostly spontaneous and slightly hyperdense in comparison to the normal adjacent renal parenchyma, and they sometimes present as an isodense or hyperdense mass. Calcifications of various sizes, as well as cysts can be seen. Mostly, peripheral areas of the mass had significantly higher attenuation when compared to the central portion, suggestive of central necrosis. However, our case showed lower attenuation in the peripheral areas compared to the central portion, representative of peripheral necrosis and hemorrhage.
The tumor appeared with progressively mild enhancement and a persistent peripheral low-attenuated area following intravenous contrast medium administration.\textsuperscript{14–16}

MA has been described as being hypointense on T1- and T2-weighted magnetic resonance imaging (MRI) scans.\textsuperscript{24} MRI does not further elucidate the image diagnosis of MA. A mass of intermediate signal intensity in a T1-weighted image, or which is sometimes slightly hypointensity on a T2-weighted image, was reported,\textsuperscript{12,22} as was a hypointense mass on both T1- and T2-weighted MRI scans,\textsuperscript{16} with some hemorrhagic foci and no sign of perirenal invasion.\textsuperscript{18,21} MA shows excentric slight contrast enhancement after the administration of intravenous contrast.\textsuperscript{17,22}

The differential diagnosis of MA includes PRCC, Wilms’ tumor, atypical renal angiomyolipoma (AML), and reninoma. PRCC is more often hypovascular and homogeneous on CT than the other subtypes of renal tumors.\textsuperscript{25} Calcifications of various sizes, necrotic and hemorrhagic areas, as well as cysts can be seen in MAs. It is sometimes very difficult to identify PRCC and MA that share the similar characteristics of mild enhancement and homogeneous density. Needle biopsy and a clinical follow-up study may be helpful. Wilms’ tumor is the most frequent solid malignancy of the kidneys that typically occurs in children. Its peak incidence is at 3–4 years of age, and 80% of patients present before 5 years of age.\textsuperscript{23} Most Wilms’ tumors were single, solid, quite large at the time of detection, and poorly enhanced with necrosis and cystic changes; the residual kidney presented with enhancement in a crescent or ring-like pattern, and it is rarely associated with hemorrhage and calcifications. Minimal fat renal AML tumors are typically hyperdense relative to the normal kidney parenchyma and they demonstrate homogenous enhancement with the administration of intravenous contrast on CT.\textsuperscript{26} It is easy to identify MA from Wilms’ tumor and AML according to their characteristic imaging features. Reninoma is a tumor of the renal juxtaglomerular cell apparatus that causes hypertension and hypokalemia via hypersecretion of renin.\textsuperscript{27} It is a single, well-defined, poorly enhanced ovoid mass on

\section*{Figure 2 CT images of the left renal tumor.}

\textbf{Notes:} (A) Unenhanced CT scan shows a well-defined, high-density mass measuring 3.7 cm $\times$ 3.3 cm (long arrow) in the lower pole of the left kidney, with an irregularly shaped lower attenuation area (short arrow) in the mass. (B–D) Enhanced CT scan shows that the tumor appears to have no enhancement in the peripheral portion (white arrow), and progressive enhancement in the central portion (black arrow).

\textbf{Abbreviation:} CT, computed tomography.
CT scan, and its typical clinical manifestations contribute to the final diagnosis.

Conclusion
MA is a rare renal tumor without specific imaging features, and its final diagnosis depends on histopathological examination. Most MAs present as well-defined, ovoid, cystic–solid or solid renal masses with calcifications of various sizes, as well as necrotic and hemorrhagic areas. Needle biopsy, clinical follow-up study, or partial nephrectomy should be planned when a mass that is highly suggestive of MA is encountered. More research is needed to elucidate the pathology, radiologic findings, and biologic behavior of MA.

Disclosure
The authors report no conflicts of interest in this work.

References