

Renal immature teratoma in a male adult

A case report and literature review

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Abstract

Rationale: Teratomas commonly arise in the gonads, including ovary and testis. The kidney is one of the most rare regions of primary teratoma. To date, about 19 cases of renal teratoma have been reported, and only 3 articles have reported renal immature teratoma; however, all of them occur in infant or children. In the present study, we reported a renal immature teratoma in a male adult.

Patient concerns: The present patient was a middle-aged man with aching pain in the left waist, and contrast-enhanced CT showed a lump in the left kidney with mild-to-moderate enhancement, and a low density small necrotic area was seen in the center.

Diagnosis, interventions, and outcomes: The patient underwent radical nephrectomy. Based on postsurgical histopathology, the final diagnosis of this case was renal immature teratoma. Postoperative chemotherapy was carried out, and the patient has been followed-up for 18 months without tumor recurrence.

Lessons: Adult renal immature teratoma is rare, and the diagnosis is mainly based on the pathological findings.

Abbreviations: CT = computed tomography, NCCN = national comprehensive cancer network.

Keywords: case report, kidney, renal, teratoma

1. Introduction

Teratomas are a type of neoplasms originating from totipotential embryonic cells, and can be differentiated into one or more germinal layer, including ectoderm, endoderm, and mesoderm.^[1] Teratomas commonly arise in the gonads, including ovary and testis. Extragonadal teratoma mostly arise in sacrococcygeal, mediastinal, and sacral regions, and rarely arise in retroperitoneal, alimentary, cervical, and intracranial regions.^[2] The kidney is one of the most rare region of primary teratoma. Teratoma can be divided into benign teratoma and malignant teratoma. Malignant teratoma includes immature teratoma and malignant transformation of benign teratoma.^[3] Immature teratoma is composed of embryonic tissue, mainly neural tissue. To date, about 19 cases of teratoma of kidney have been reported.^[4–7] However, primary malignant teratoma of the kidney is extremely

rare. In the present study, we reported a renal immature teratoma in adult. To the best of our knowledge, our patient is the first known case with renal immature teratoma in adult.

2. Case report

A 47-year-old male patient presented with aching pain in the left waist of about 1-month duration, without frequent micturition and hematuria. There was no significant decline in weight in short term. Physical examination showed positive left renal percussion pain, but no obvious lumps in the left waist. Contrast-enhanced computed tomography (CT) of the whole abdomen and pelvic cavity showed that the size, position, and shape of the kidneys were normal on both sides. It revealed an iso-density occupying shadow in the left kidney measuring about 68 × 72 mm. After enhancement, it had mild-to-moderate enhancement, and small necrotic area with low density was seen in the center (shown in Fig. 1). Enlarged lymph nodes were seen in the retroperitoneum, behind the diaphragm and around the left kidney, renal artery and vein, and the left renal vein was partial filling-defect. Tumor mass biopsy was conducted after hospitalization, and the pathological report considered low differentiated carcinoma. IHC findings are as follows: Inhibin (–), Ki-67 (+ < 3%), P53 (+), RCC (–), S-100 (–), Vim (+), Pax-8 (+), CK7 (–), E-cadherin (–), EMA (+), P63 (–), and CK5/6 (–). Subsequently, he underwent radical nephrectomy. Intraoperative findings revealed 8 × 7.5 × 3.5 cm solid mass in the left kidney. Enlarged lymph nodes were found near the abdominal aorta and the inferior vena, and the renal pedicle vessels were also surrounded by enlarged lymph nodes. Perirenal fascia, fatty sac, kidney, upper ureteral segment, lymph nodes around the renal pedicle, and the adrenal gland were resected. Then the dissection of lymph nodes around the abdominal aorta and the inferior vena were conducted carefully. The surgical excision specimen is shown in Figure 2.

The tumor size was about 8 × 7.5 × 3.5 cm, and it invaded the renal portal, perirenal adipose tissue, and adrenal gland. The

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XLZ, GX, and JLL contribute equally to this study.

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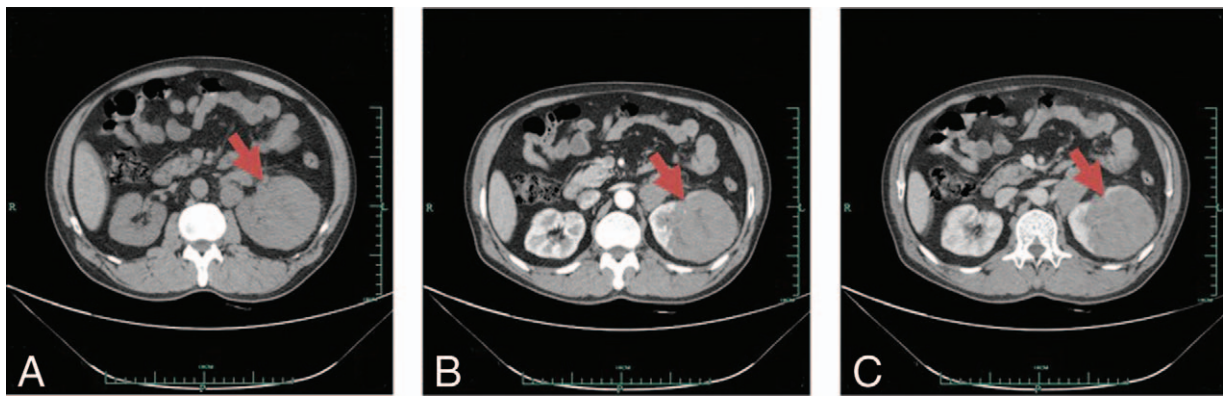


Figure 1. Preoperative contrast-enhanced CT revealed a iso-density occupying shadow in the left kidney measuring about 68×72 mm. After enhancement, it had mild-to-moderate enhancement. (A) Plain scavenging period; (B) enhancement period; (C) substantive period. CT=computed tomography.

tumor thrombus was seen in the pulse tube, and 11 of 12 lymph nodes were positive. Therefore, the pathological stage was T4N1M0. IHC findings are as follows: WT1 (-), CD10 (+), CK7 (-), E-Cadherin (+), EMA (+), Inhibin (-), Ki-67 (+80%), Melan-A (-), P53 (+), RCC (-), S-100 (-), Vim (+), TFE3 (-), CA-IX (+), P504s (-), Pax-8 (+), P63 (-), p40 (-), CK20 (-), and CKpan (+). The tumor tissue mainly contains malignant epithelioid components, with a small amount of brain tissue (shown in Figs. 3 and 4).

The final diagnosis of this case was renal immature teratoma. Postoperative chemotherapy was carried out, and the chemotherapy regimen was BEP (Bleomycin 30 units per week, Etoposide 100 mg/m^2 daily, day 1–5; Cisplatin 20 mg/m^2 daily, day 1–5; repeat every 21 days and 3 cycles). The present case has been followed up for 18 months without tumor recurrence.

The patient provided informed consent for the publication of his clinical data. Medical Ethical Committee approval of the report was waived by Shaoxing People's Hospital (Shaoxing Hospital of Zhejiang University).

3. Discussion

Teratoma and other germ cell tumors rarely occur in the kidneys, and renal immature teratoma is more rare. So far, only 3 articles have reported renal immature teratoma, and all of them occur in

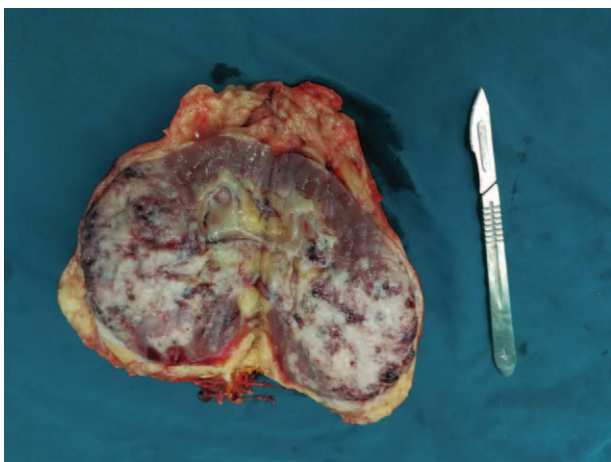


Figure 2. The surgical excision specimen.

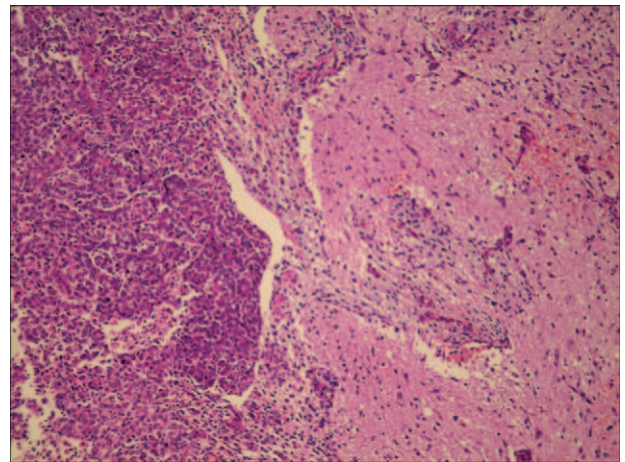


Figure 3. The tumor tissue mainly contains malignant epithelioid components (on the left side), with a small amount of brain tissue (on the right side). Original magnification $\times 100$ (H&E).

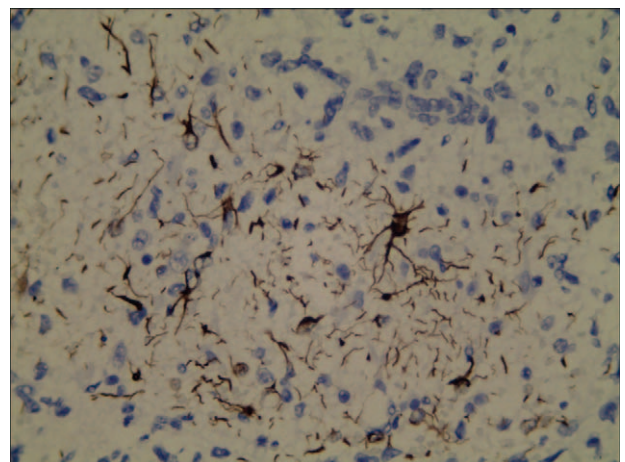


Figure 4. Teratomatous components of brain tissue. Original magnification $\times 400$ [glial fibrillary acidic protein (GFAP) staining].

Table 1**Clinical characteristics and pathologic features of renal immature teratoma.**

First author	Journal, year	Age	Sex	Country	Clinical presentation	Components of teratoma	Lymph node involvement and distant metastasis	Treatment after surgery
Idrissi-Serhrouchni K	<i>Diagnostic Pathology</i> , 2013	6-month-old	Female	Morocco	Abdominal distension and pain	Keratinizing stratified squamous epithelium with skin adnexae, cartilage, mucinous columnar epithelium, bone, melanin containing cells and neuroglial cells with occasional foci of immature neuroectodermal tissue	No involvement of the lymph nodes	No further treatment was given
Evans K	<i>Pediatr Blood Cancer</i> , 2010	6-month-old	Male	UK	Abdominal mass and hypertension	A variety of tissue derived from all the 3 germ cell layers.	The lymph nodes in the hilum were free of tumor as were the cut margins of the renal vein and artery.	No further treatment was given
Liu YC	<i>The Journal of Urology</i> , 2000	2-year 10-month-old	Female	Taiwan	Poor appetite and poor activity 1 week in duration	Yolk sac tumor and immature teratoma	Multiple lymph node involvement in the hepatic hilum and peripancreatic head area.	Administer 4 more chemotherapy sessions

infant or children.^[4,8,9] The clinical characteristics and pathologic features of these renal immature teratoma were shown in Table 1. To the best of our knowledge, our patient is the first known case with renal immature teratoma in adult.

The present patient was a middle-aged man with aching pain in the left waist, and contrast-enhanced CT showed 68×72 mm lump in the left kidney with mild-to-moderate enhancement, and a low density small necrotic area was seen in the center. The pathological report of tumor mass biopsy considered low differentiated carcinoma. Therefore, the diagnosis of renal immature teratoma is very difficult before surgery, and the final diagnosis depends on the pathological results. For a tumor to be termed a renal teratoma, Beckwith^[10] suggested that it should meet the following minimal criteria: firstly, the primary tumor should be unequivocally of intrarenal origin, which usually can be established only if the entire lesion is contained within the renal capsule and there are no teratomas in remote sites which might have metastasized to the kidney. Secondly, the tumor should exhibit unequivocal heterotopic organogenesis, with clearly recognizable evidence of attempts to form organs other than kidney. Such organs can be of either somatic or extraembryonic type. The differential diagnosis of renal immature teratoma includes Wilm's tumor, metanephric adenoma, lymphoma, peripheral neuroectodermal tumor, and rhabdomyosarcoma; and rarely metastatic small cell tumors from lung.^[4,11] In the present case, the tumor was unequivocally of intrarenal origin, and the tumor tissue mainly contains malignant epithelioid components, with a small amount of brain tissue. Therefore, characteristics in the present case were consistent with the criteria given by Beckwith, and it can be diagnosed as a renal immature teratoma.

Since the incidence of renal malignant teratoma is very low, and the related literature and follow-up data are limited, there is no unified standard for its treatment. The pathological stage of

the present patient was T4N1M0. According to guidelines of national comprehensive cancer network (NCCN) for the treatment of ovarian teratoma,^[12] postoperative chemotherapy was carried out in the present case. Follow-up data of intrarenal teratomas after surgery are limited, especially for renal immature teratoma in adult. The present case has been followed up for 18 months without tumor recurrence, and we need longer follow-up to provide data for the prognosis of renal immature teratoma.

Author contributions

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References

- Albany C, Einhorn LH. Extragonadal germ cell tumors: clinical presentation and management. *Curr Opin Oncol* 2013;25:261–5.
- Bokemeyer C, Nichols CR, Droz JP, et al. Extragonadal germ cell tumors of the mediastinum and retroperitoneum: results from an international analysis. *J Clin Oncol* 2002;20:1864–73.
- Motzer RJ, Amsterdam A, Prieto V, et al. Teratoma with malignant transformation: diverse malignant histologies arising in men with germ cell tumors. *J Urol* 1998;159:133–8. PubMed PMID: 9400455.
- Idrissi-Serhrouchni K, El-Fatemi H, El madi A, et al. Primary renal teratoma: a rare entity. *Diagn Pathol* 2013;8:107.
- Higgins A, Eisa W, Walton J, et al. Metastatic mucinous adenocarcinoma and carcinoid tumor arising from a mature cystic teratoma of a horseshoe kidney. *Urol Case Rep* 2017;11:39–41.

- [6] Yavuz A, Ceken K, Alimoglu E, et al. Mature cystic renal teratoma. *Iran J Radiol* 2014;11:e11260.
- [7] Jacob S, Ghosh D, Rawat P, et al. Primary intra-renal teratoma associated with renal dysplasia: an unusual entity. *Indian J Surg* 2013;75 (suppl 1):77–9.
- [8] Evans K, Rogers T, Garrett-Cox R. A rare case of intrarenal teratoma in a 6-month-old male. *Pediatr Blood Cancer* 2010;55: 1207–9.
- [9] Liu YC, Wang JS, Chen CJ, et al. Intrarenal mixed germ cell tumor. *J Urol* 2000;164:2020–1. PubMed PMID: 11061908.
- [10] Beckwith JB. Wilms' tumor and other renal tumors of childhood: a selective review from the National Wilms' Tumor Study Pathology Center. *Hum Pathol* 1983;14:481–92. PubMed PMID: 6303938.
- [11] Geethamani V, Kusuma V, Gowda KM, et al. Adult Wilms' tumour: a case report with review of literature. *Diagn Pathol* 2006;1:46doi: 10.1186/1746-1596-1-46. PubMed PMID: 17144931; PubMed Central PMCID: PMC1702367.
- [12] Morgan RJJr, Armstrong DK, Alvarez RD, et al. Ovarian cancer, version 1.2016, NCCN clinical practice guidelines in oncology. *J Natl Compr Canc Netw* 2016;14:1134–63. PubMed PMID: 27587625.