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Oncology

Concomitant perinephric Castleman disease and renal cell carcinoma initially suspected to be metastasis: A case report and 24 months follow up

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Introduction

Castleman disease (CD) is an autoimmune, lymphoproliferative disease that shows itself with the enlargement of lymph nodes and varied clinical presentations originally described by Castleman in 1954. The incidence of CD is estimated at 21–25 cases per million person-years[1] and the usual location is the mediastinum, although this pathology can theoretically affect any nodal station, extrathoracic sites have been reported in the neck, axilla, pelvis, and retroperitoneum.

Renal cell carcinoma (RCC) ranks among one of the most prevalent tumors in China, with the estimated 66,800 new cases and 23,400 deaths in 2015. RCC often presents at an advanced stage. Nearly 30% of the patients are diagnosed with metastatic RCC (mRCC) at their first visit to hospital, which results in the poor prognosis.

Enlarged hilar or retroperitoneal lymph nodes of RCC patients detected by CT or MRI commonly harbor the malignant change and metastasis. Whereas, comprehensive consideration should be taken, in case certain special cases would be misdiagnosed.

Case report

A 53-year-old man underwent an abdominal ultrasonography revealed a solid mass with a diameter of 4 cm in the right kidney at the annual health checkup. The results of the physical examination were negative, and no gross hematuria or constitutional symptoms had been observed. Full blood count showed hemoglobin 166 g/L, white blood cells 5400/μL with normal differential counts, and platelets 803,000/μL. Serum protein was 67 g/L, albumin 40 g/L and calcium 2.74 mmol/L. The hepatorenal and coagulation profiles were all normal, the patient was screened negative for human immunodeficiency virus (HIV).

Abdominal contrast computed tomography (CT) revealed a 4.0 cm × 3.8 cm × 3.5cm mass arising from the right renal upper pole (Fig. 1A) and showed a single welled fined homogeneously enhanced mass close to the right renal pedicle, measuring 4.8 cm × 4.0 cm × 3.3cm with no calcification but partial necrotic area inside (Fig. 1B). Due to the possibility of malignancy and metastasis, the patient underwent a Fluorine-18-labeled 2-fluoro-2-deoxy-D-glucose positron emission tomography-computed tomography (^{18}F-FDG PET-CT) scan, which showed metabolic activity and considered the mass close to the right renal pedicle as the metastatic lymph node lesion, but was negative for pulmonary or bone metastasis. Blood circulating tumor cell (CTC) is measured by iFISH technology, however, the result was negative.

Based on the imaging findings, RCC with lymph node metastasis was highly suspected. Consequently, the patient underwent a retro-laparoscopic right radical nephrectomy and perinephric mass dissection. Intraoperative found the perinephric mass (Fig. 2) was close to the right renal pedicle. Besides, there were no significant adhesions to the pelvis or to the renal parenchyma itself, the renal vessels were effectively shaved off the mass and the mass was resected with satisfactory margin.

The post-operation pathologic examination showed the renal tumor was clear cell RCC and Fuhrman nuclear grade 2, the perinephric mass specimen was negative for malignancy but showed abundant small follicles with regressed germinal centers and hyalinization (Fig. 3). There were scattered follicles of lymphocyte depletion with follicular dendritic cells and hyaline deposits and penetration of sclerotic blood vessels, these findings were compatible with hyaline vascular CD. The patient was cured with no evidence of recurrence in 24 months follow-

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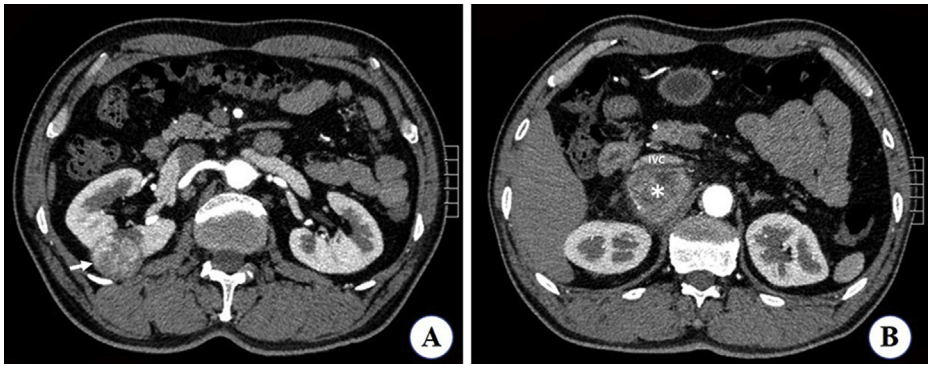

Fig. 1. CT scan (1A)demonstrate a 4.8cm diameter heterogeneously right renal mass (arrow) and (1B) show a single welled fined homogeneously enhanced mass close to the right renal pedicle. IVC: inferior vena cava.

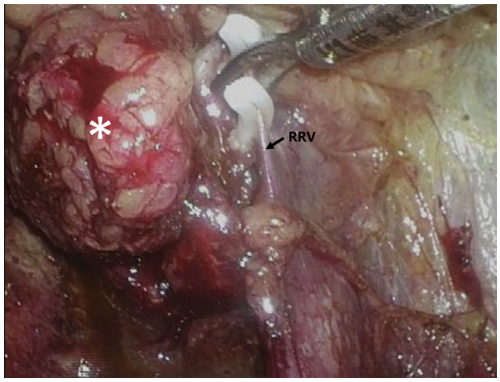

Fig. 2. Intraoperative imaging show the perinephric mass(star) was close to the right renal vein(arrow).

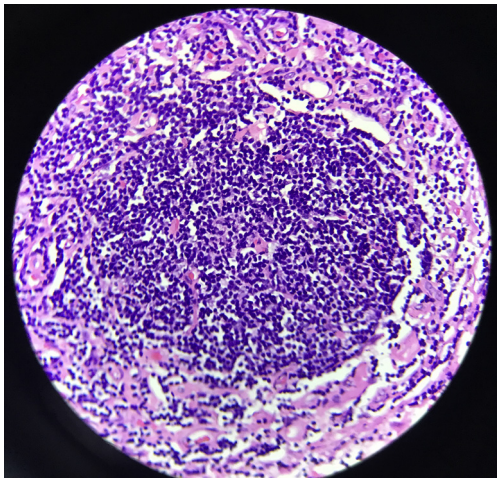

Fig. 3. Histopathologic examination perinephric mass (3) shows distinctive follicles with expanded mantle zones, mainly atrophic germinal centers with hyalinization, and regressed germinal center showing hypervascularity with a "lollypop" appearance.

up.

Discussion

CD is principally distinguished by the presence of localized or generalized lymphadenopathy designated unicentric CD (UCD) and multicentric CD (MCD), respectively. UCD was mainly associated with hyaline-vascular histopathological features, and most patients were asymptomatic. Complete surgical resection is curative for UCD, leading to excellent long-term outcomes and no further treatment is needed. Unfortunately, the outcome for UCD patients was better than those of MCD patients, with 5-year overall survival rates of 93.6%–51.2%.[2]

Concomitant RCC and CD is a comparatively rare occurrence, only three cases have been reported previously.[3–5] First report presented concomitant MCD and renal chromophobe cell carcinoma, the second case was coexistence of UCD and locally advanced papillary RCC, and the last case presented with abdominal mesenteric CD with RCC and stomach leiomyoma.

Imaging findings are nonspecific and may demonstrate lymphadenopathy and organomegaly, the UCD lesion is hard to distinguish from other primary or metastatic tumors. The rarity and heterogeneity of CD have limited thorough understanding of its etiopathology and management. There are no reliable diagnostic methods, and the definitive diagnosis should be based on pathologic examinations.

Even though that the clinical significance and therapeutic implications are not clear yet, the concomitant RCC and UCD could have favorable prognostic significance based on the fact that the UCD described so far have benign behavior and the patient shows no evidence of recurrence in 24 months follow-up.

Hence enlarged perinephric nodal mass on CT coexistence with a renal tumor should not always be considered as a malignant change. Misdiagnosis could cause unnecessary resection of organ. In our case, the optimal treatment strategy for the patient was partial nephrectomy and UCD resection, which may equivalently yield comparative long-term oncological outcomes and better renal function preservation compared to those of RN.

Raising the awareness of CD and other lymphoproliferative diseases may improve the early diagnosis and suitable therapy for CD. The confirmed postoperative diagnosis is important to avoid over diagnosis and unnecessary further treatment, such as molecular targeted therapy or immunotherapy.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initial will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

The case has been first published in the Journal of Clinical Urology (just publish in Chinese) in 2017, which described the 6 months follow-up. Now we would like to share the case and the follow-up result in English.

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Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.eucr.2018.08.019.

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