

Acute abdominal pain induced by renal leiomyoma in a young patient: a case report

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Abstract

Renal leiomyoma is a rare benign mesenchymal tumor of the kidney that predominantly originates from the renal capsule or pelvis. However, because of its nonspecific clinical and imaging features, renal leiomyoma remains poorly characterized and may even lead to radical or partial nephrectomy on the basis of preoperative suspicion of renal carcinoma. We herein present a case involving a 12-year-old boy with acute abdominal pain who was diagnosed with renal leiomyoma based on both clinical imaging and histopathological examination. One year after radical nephrectomy, the patient recovered to good condition. This case demonstrates that the comprehensive application of imaging and histology are essential for early clinical diagnosis and effective treatment of renal leiomyoma.

Keywords

Kidney, leiomyoma, capsule, case report, clinical imaging, histopathology, nephrectomy

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Introduction

Leiomyomas are rare benign mesenchymal tumors originating from smooth muscle cells and were first described by Virchow in 1854.¹ The reported prevalence of leiomyomas based on autopsy findings ranges from 4.0% to 5.5%.² Leiomyomas are most common in the uterus; they are exceptionally rare in the kidney, and fewer than 100 renal leiomyomas have been reported in the

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literature.³ The diagnosis of leiomyomas remains challenging.

Renal leiomyomas are well-circumscribed tumors without infiltration into surrounding tissue or invasion and metastasis of distant organs.⁴ Although they were included in the 2004 World Health Organization (WHO) classification of renal tumors, they are not commonly considered during the clinical evaluation of renal masses because of their rare occurrence in this location. The most common symptoms of renal leiomyomas in clinically evident cases are an abdominal mass (57%), abdominal pain (53%), or both (33%). However, only 20% of patients with renal leiomyomas present with gross hematuria.⁵ Thus, it is difficult to clinically distinguish renal leiomyomas from other types of cancer, including renal leiomyosarcoma and malignant renal cell carcinoma (RCC).

In a review of case reports and rare case series of renal leiomyoma, Patil et al.⁶ assessed 24 cases initially diagnosed as renal leiomyoma in 10 institutions from different Western areas. All renal leiomyomas were solitary and occurred in women (mean age, 63 years; range, 44–74 years). The tumor size ranged from 0.6 to 7.0 cm (mean, 2.9 cm). To the best of our knowledge, there are no previously published reports of acute abdominal pain induced by a giant renal leiomyoma in a younger patient.

Case report

The reporting of this study conforms to the CARE guidelines.⁷ A 12-year-old boy was admitted to the hospital because of a half-day history of acute left upper abdominal pain in December 2017. At the time of consultation, he had not experienced abdominal distention, nausea, vomiting, chills, fever, frequent urination, urgent urination, painful urination, gross hematuria, chest tightness, or shortness of breath in the

previous several days. B-mode ultrasonography of the abdomen at a local hospital had shown an approximately 81- × 79-mm mass with low and weak echo, clear edges, less uniform internal echo, and an unclear boundary with the left kidney (Figure 1(a)).

The patient presented to our institution with no significant relief of his abdominal pain. Physical examination showed no protuberance in the area of either the right or left kidney; however, palpation revealed a left renal subcostal mass of about 7.0 × 7.9 cm². No percussion pain was noted in either kidney area, and no vascular murmur was heard during auscultation. The rest of the physical examination, including examination of the respiratory system, was unremarkable.

Abdominal computed tomography demonstrated a round tumor in the left kidney, measuring approximately 7.0 × 8.5 × 8.7 cm³ (Figure 1(b)). The tumor showed contrast uptake, and the enhancement range in the venous phase and delayed phase was enlarged while the degree of enhancement was reduced. Areas of irregular and flaky enhancement were present in the tumor, and the boundary between the tumor and renal parenchyma was unclear (Figure 1(c)–(f)).

Preoperative examinations (blood tests, electrocardiography, and chest radiography) showed no abnormalities. The patient's laboratory results revealed a leukocyte count of $6.82 \times 10^9/L$, erythrocyte count of $4.30 \times 10^9/L$, hemoglobin concentration of 127 g/L, and platelet count of $378 \times 10^9/L$. Biochemical examinations showed a prealbumin concentration of 18.9 mg/dL (reference range, 20–40 mg/dL) and creatinine concentration of 80.9 μmol/L (reference range, 27–65 μmol/L). Urinalysis showed occult blood (+) (reference, negative).

On intraoperative examination, we found a well-circumscribed, grayish, encapsulated round tumor with soft consistency.

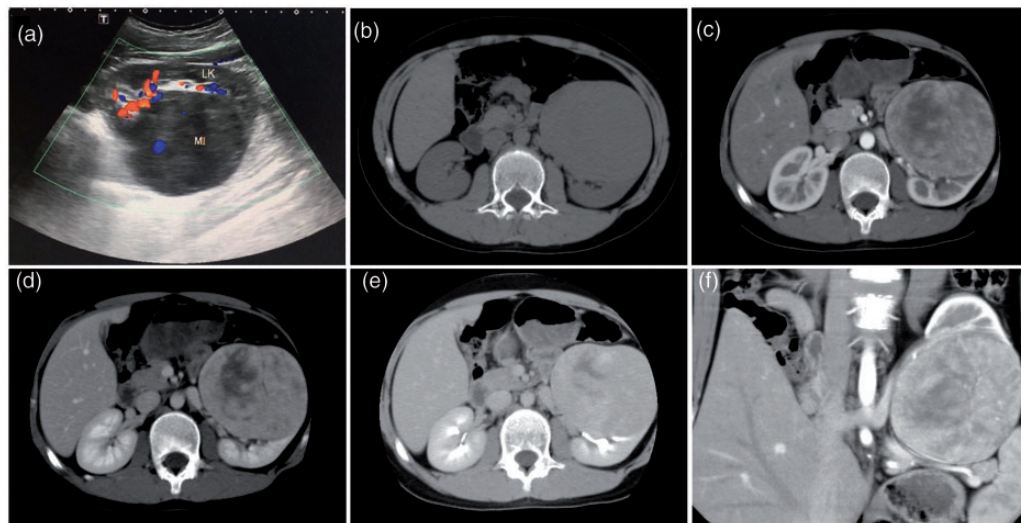


Figure 1. Imaging changes of left renal leiomyoma. (a) Ultrasound image. (b) Plain computed tomography image. Enhanced computed tomography images in (c) portal venous phase, (d) delayed phase, and (e) arterial phase. (f) Coronal enhanced computed tomography image.

The tumor seemed to originate from the renal capsule. Given the size of the lesion and because malignant entities such as cystic RCC or leiomyosarcoma could not be excluded, a surgical approach was deemed necessary. Therefore, radical left nephrectomy was carried out under laparoscopy.

On postoperative examination, the kidney specimen measured approximately $14.0 \times 9.5 \times 8.5 \text{ cm}^3$. The specimen was dissected, and the tumor was gray-white and $8.5 \times 8.5 \times 8.0 \text{ cm}^3$ in volume; it had a clear boundary and soft quality. On microscopic examination, the tumor consisted of well-oriented fascicles of long spindle cells immersed in stromal tissue. No mitosis, necrosis, or atypical cells were found (Figure 2). Immunohistochemical evaluation was positive for vimentin, desmin, and CD34 and negative for pan-cytokeratin, S100, and Ki67 (<1%). The final pathologic diagnosis was renal leiomyoma.

The perioperative course was uneventful, and the patient was discharged on the fifth postoperative day. One year after surgery,

the patient returned for CT re-examination. The results of plain and enhanced CT showed that the patient was disease-free (Figure 3(a)–(d)).

Discussion

Leiomyomas are rare benign mesenchymal tumors that mainly originate from smooth muscle cells. Renal leiomyoma is an uncommon benign mesenchymal neoplasm of the urinary system and is included in the 2016 WHO classification of renal tumors.⁸ Renal leiomyomas can be classified into three groups based on the detection scenario: (1) discovered at autopsy, (2) rare clinically significant symptomatic lesions, and (3) discovered incidentally on imaging examination.⁹ This tumor shows a female predilection (2:1), and patients' mean age is 47 years.¹⁰ The most common symptoms are a palpable flank mass, abdominal pain, and hematuria.^{11,12} To the best of our knowledge, the present case report is the first to describe acute abdominal pain

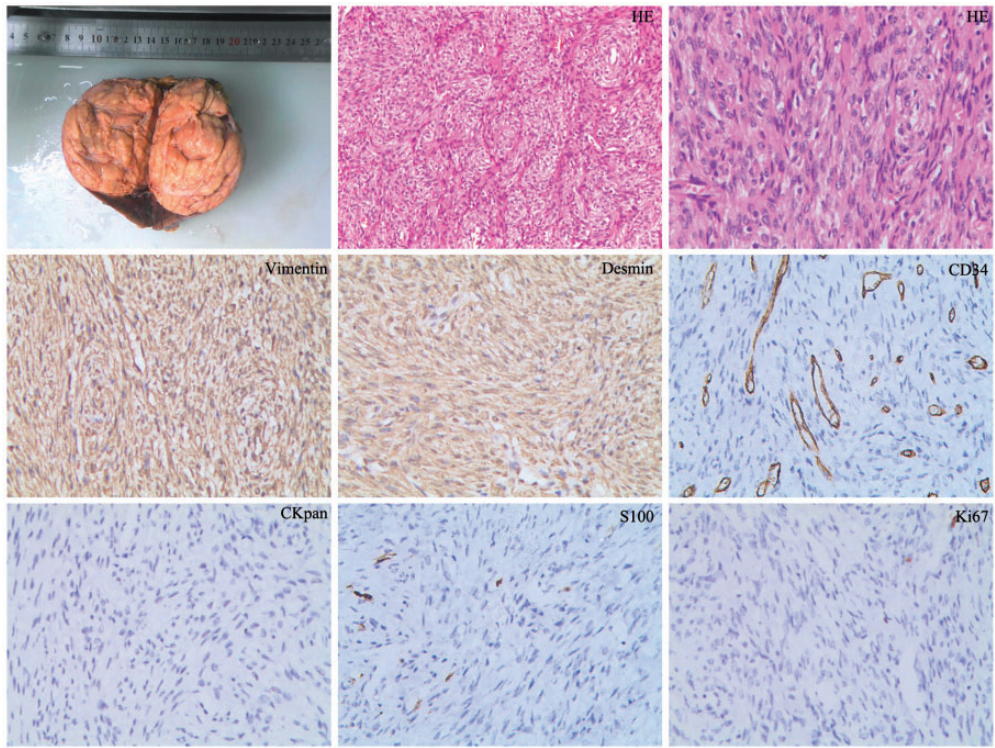


Figure 2. Histopathologic examination and immunohistochemical staining results of renal leiomyoma ($\times 200$). The immunohistochemical findings were as follows: vimentin (+), desmin (+), CD34 (+), CKpan (–), S100 (–), and Ki67 (–).

HE, hematoxylin–eosin staining; CKpan, pan-cytokeratin.

induced by a renal leiomyoma in a pediatric patient (12 years of age).

Because of degenerative phenomena of the tumor, most renal leiomyomas have lost their typical homogeneous properties, making it difficult to differentiate them from benign lesions such as angiomyolipoma (AML) and oncocytoma as well as malignant cancers such as leiomyosarcoma and RCC.¹³ CT and magnetic resonance imaging are widely used to achieve a clinical diagnosis; these imaging techniques show that the lesion has regular margins and no evidence of local invasion. However, regular and well-defined margins and the absence of radiological signs of local invasion are uncommon features of

leiomyosarcomas.¹⁴ RCCs typically appear hyperintense in T2-weighted images, and large RCCs usually show areas of necrosis. Meanwhile, with the absence of a macroscopic fat component and the presence of calcifications and hemorrhagic areas on imaging examination, AML and its less common fat-poor variant can be ruled out with a reasonable degree of certainty.¹⁵ Mixed epithelial and stromal tumor of the kidney often presents as a well-circumscribed, multicystic, and solid mass with delayed enhancement and often occurs in perimenopausal women.¹⁶ However, rare kidney sarcomas such as dedifferentiated liposarcoma or undifferentiated pleomorphic sarcoma also have fat

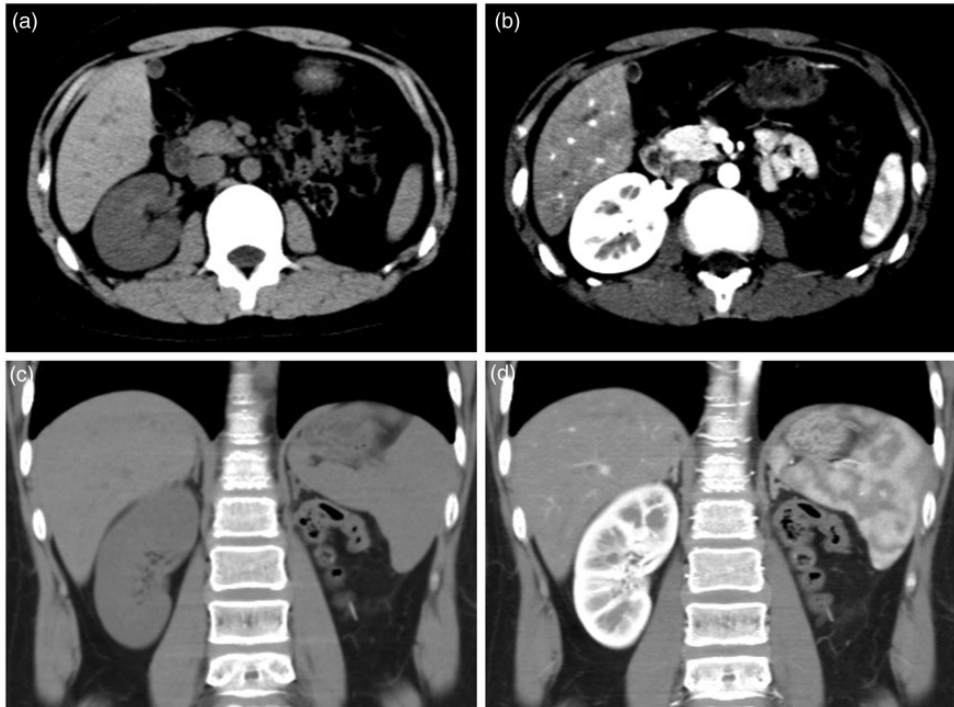


Figure 3. Imaging changes after radical nephrectomy. (a) Plain and (b) enhanced computed tomography images in horizontal section. (c) Plain and (d) enhanced computed tomography images in coronal section.

components, and they must also be included as differential diagnoses. Thus, these radiologic features are not completely sufficient to exclude papillary RCC and other rare kidney sarcomas as differential diagnoses.

Microscopically, malignant spindle cells usually display variable degrees of nuclear pleomorphism, nuclear hyperchromasia, and mitotic activity. Immunohistochemical staining of HMB45 in renal leiomyosarcomas is reportedly negative and can be used to differentiate these tumors from leiomyomas.^{17,18} In one study, cathepsin K reportedly showed positive expression in 8 (67%) of 12 leiomyosarcomas.¹⁹ Desmin is also a useful marker in the distinction between lipid-poor AML and leiomyoma.⁶ Cathepsin K is 100% sensitive in both common and leiomyoma-like AMLs and

shows reactivity in >80% of cells.²⁰ However, the expression of cathepsin K has not been investigated in leiomyomas. Despite the potential role of hormones in the pathogenesis of renal leiomyomas, estrogen receptor and progesterone receptor expression cannot be used in distinguishing renal leiomyoma from AMLs.

In conclusion, because of the nonspecific clinical and imaging features of renal leiomyoma, comprehensive application of imaging and histology is an effective way to achieve a diagnosis. Thus, better identification of this neoplasm is still needed to avoid unnecessary aggressive treatments, particularly in younger patients.

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Ethics statement

This study was reviewed and approved by the Ethics Committee of Tungwah Hospital of Sun Yat-sen University. Written informed consent was obtained from the patient's legal guardian for the publication of any potentially identifiable images or data included in this article.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.


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Author contributions

WNM, HLiJ, and YZZ wrote the case report. JZ and HLoJ reviewed and edited the manuscript. All authors contributed to manuscript revision and approved the final version.

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