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**Kidney inflammatory myofibroblastic tumor masquerading as metastatic malignancy: A case report and literature review**

Zhang GH *et al.* Kidney IMT masquerading as metastatic malignancy

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**Abstract**

***BACKGROUND***

Inflammatory myofibroblastic tumor (IMT) is a rare mesenchymal tumor that is characterized by spindle cells differentiated from muscle fibroblasts and infiltration of various types of inflammatory cells. IMT can occur at any age and at any anatomic site. The most common location of IMT is the bladder in the genitourinary tract. Only scarce cases of kidney IMT have been reported in the literature.

***CASE SUMMARY***

A 77-year-old woman, with a history of bilateral renal calculus for 15 years, was admitted to the Department of Urology of our hospital complaining of recurrent painless gross hematuria for one month. The treatment with cephalosporin was ineffective. Computed tomography imaging showed a mixed density and slightly heterogeneously enhanced lesion in the middle pole of the left kidney and ipsilateral adrenal enlargement. The patient underwent surgical treatment by retroperitoneoscopic left radical nephrectomy plus adrenalectomy. A large number of typical spindle cells surrounded by plasma cells and lymphocytes were observed microscopically. Immunohistochemical analyses indicated that these spindle cells were positive for vimentin, cytokeratin (CK), Ki-67, CK7, CD34, and CD31 and were focally positive for CD10 and anaplastic lymphoma kinase (ALK-1). Thus, a diagnosis of IMT was made definitively. The patient recovered well after operation, and no recurrence or metastasis was noted during the 22-mo follow-up.

***CONCLUSION***

Since kidney IMT is very rare and lacks characteristic clinical manifestation, it is easily misdiagnosed as a malignant tumor before operation. Surgery remains the best choice for diagnosis and treatment, and such cases must be followed carefully because of the uncertain biological behavior of this tumor. This report suggests that renal calculus may be one of the causes of IMT, but further investigation is necessary to prove it.

**Key words:** Inflammatory myofibroblastic tumor; Kidney; Diagnosis; Renal calculus; Case report

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**Core tip:** Herein, we present an elderly woman with kidney inflammatory myofibroblastic tumor (IMT) characterized by painless gross hematuria and misdiagnosed as a malignant tumor with ipsilateral adrenal metastases. Kidney IMT is a rare tumor entity with unknown etiology, diverse clinical symptoms, and imaging manifestations. Renal calculus was suspected to be one of the causes or drivers of IMT because of the constant destruction of the kidney mucosa. The diagnosis of IMT is considered a great challenge before operation. However, the awareness of its existence and deep understanding of its clinical characteristics are essential to avoid misdiagnosis in the differential diagnosis of a renal mass and to provide better management of such cases. Despite the uncertain biological behavior of this type of tumor, most patients get a favorable prognosis after surgery.

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**INTRODUCTION**

Inflammatory myofibroblastic tumor (IMT) is a tumor subset belonging to the inflammatory pseudotumor family, which is a generic term for spindle cell proliferation in an uncertain anatomic location with diversified infiltrative inflammatory components. Although recent studies have suggested that IMT is a type of borderline neoplasm, it is still uncertain whetherthis type of tumor originates from an inflammatory or a cancerous process[1]. The most common anatomic location of IMT is the lungs, but there have been many reports of extrapulmonary IMT in recent years[1-9]. In the urinary system, the most common site of IMT is the bladder[10]; its occurrence in the kidney is extremely rare. To the best of our knowledge, only 48 cases of kidney IMT have been reported in the English literature between 1972 and 2019. Herein, we report an additional case of kidney IMT with a history of renal calculus and review the relevant literature to summarize the characteristics to increase our understanding of this disease and provide such patients with better management.

**CASE PRESENTATION**

***Chief complaints***

A 77-year-old woman who presented with recurrent painless gross hematuria was admitted to the Department of Urology of The Second Affiliated Hospital of Shenzhen University (Shenzhen, China).

***History of present illness***

The patient’s symptom started one month ago without other discomfort. Shevisited a local hospital for treatment two weeks ago. Ultrasound (US) examination revealed bilateral renal calculus, and routine urinalysis was positive for leukocytes. At that time, the physician suspected that the symptom was caused by calculous pyelonephritis, but treatment with cephalosporin was ineffective.

***History of past illness***

The patient had a medical history of renal calculus for 15 years.

***Personal and family history***

The patient’s personal history and family history were unremarkable.

***Physical examination***

The results of the physical examination were nothing unusual.

***Laboratory examinations***

Routine blood results suggested mild anemia and infection, with a hemoglobin level of 10.0 g/dl and a leukocyte count of 12.2 × 109 cells/L. Urinalysis showed a large number of red blood cells. Other laboratory results were within the normal limits, and no malignant cells were found in three urine cytopathologic analyses.

***Imaging examinations***

Abdominal ultrasound revealed only bilateral renal calculus, but no space-occupying lesions were observed. Computed tomography (CT) imaging showed a slightly heterogeneously enhancing mass in the middle pole of the left kidney and ipsilateral adrenal enlargement, which was suspected to be a metastatic lesion (Figure 1).

**PREOPERATIVE DIAGNOSIS**

Considering the clinical symptoms and imaging findings, a malignancy, particularly renal cancer, was highly suspected.

**TREATMENT**

A retroperitoneoscopic left radical nephrectomy plus adrenalectomy was performed.

**HYPHOLOGY**

A grayish-white, well-defined, and stiff mass measuring 4 cm × 3 cm × 3 cm was discovered in the middle pole of the removed kidney specimens. Microscopically, typical spindle cells were widely distributed, with various types of infiltrating inflammatory cells, capillary hyperplasia, and local necrosis observed (Figure 2).

**IMMUNOCHEMISTRY ANALYSES**

Immunohistochemical analyses revealed that these spindle cells were diffusely positive for vimentin, cytokeratin (CK), Ki-67, CK7, CD34, and CD31 and were focally positive for CD10 and anaplastic lymphoma kinase (ALK-1) (Figure 2).

**FINAL DIAGNOSIS**

This patient was diagnosed with IMT finally. The surgical margins were negative, and the adrenal gland was unremarkable.

**OUTCOME AND FOLLOW-UP**

The patient recovered well, and no complications occurred. No other treatment was performed after surgery. After 22 mo of follow-up, she was still alive, without symptoms or imaging evidence of recurrence or metastasis.

**DISCUSSION**

IMT, a type of rare mesenchymal tumor with intermediate biological potential, is mainly composed of spindle cells and inflammatory cell infiltration. It was first reported in 1937. The early literature suggested that the spindle cell proliferation was mainly a reactive hyperplasia that occurred after inflammation, and thus, IMT was thought to be an inflammatory pseudotumor. Later, as the research progressed, spindle cells were found to be the main component of these lesions; thus, it was classified as a tumor, typically a borderline tumor. However, the potential for local invasion, recurrence, and metastasis of IMT cannot be ignored[3,4]. Therefore, some scholars suggested that IMT should be redefined as a low-grade malignant tumor[3,4]. In recent years, in addition to the lungs, IMT has also been reported in extrapulmonary tissues, including the head and neck, ventricles, liver, retroperitoneum, pelvis, and spinal canal[1-9]. In the genitourinary system, IMT was found to occur in the bladder, ureter, urachal, renal pelvis, and kidney[11-15] . Although most commonly found in the bladder[10], IMT in the kidney is extremely rare. IMT is found more frequently in children than in adults and has no gender predilection, but the extrapulmonary forms occur more commonly in adult women[1].

According to our investigation, few scholars have systematically analyzed the clinical features of kidney IMT. To further study the characteristics of this type of IMT, we reviewed case reports published in MEDLINE and PubMed databases between 1972 and 2019, including 48 cases occurring in the renal parenchyma (37 cases) and renal pelvis (11 cases) (Table 1). Although the patients ranged in age from 3 to 75 years[6,7], those aged over 40 years accounted for 73% (35/48) of cases. Only five patients were younger than 14 years old. Fifty-four percent of these cases occurred in males. Fifty percent of kidney IMT occurred in the left kidney, while the other 50% occurred in the right kidney. Notably, kidney IMT was more inclined to the upper pole than the lower pole (ratio, 13:7). The clinical manifestations included low back or abdominal pain (54%), gross or microscopic hematuria (29%), incidental findings (23%), fever (19%), and weight loss (17%). The tumor sizes reported in the literature varied from 1.5 cm to 13.5 cm (median, 4.85 cm) based on imaging or pathological specimens. The majority of patients opted for radical surgery, while three patients were treated by partial resection, two treated with corticosteroids, and three with antibiotics. All patients achieved satisfactory outcomes, and no clinical or radiographic evidence of recurrence or metastasis was noted during 3 to 66 mo of observation.

The causes of IMT remain unclear. According to the literature, chronic inflammation, surgery, and trauma are considered predisposing factors for IMT. Some scholars believe that it is related to *Epstein-Barr virus* infection[8], while others have found an association between IMT and hepatitis B virusinfection[9]. Specific pathogen infections, such as *Mycobacterium tuberculosis* infection and *Eikenella corrodens* infection, are also reputed to be a cause[16,17]. The pathogenesis of IMT is also a controversial topic. Most scholars consider that chromosomal aberrations, especially kinase fusions or rearrangements, are the genetic basis of IMT tumorigenesis. The most common gene involved in IMT is the *ALK* gene, which is located on 2p23 and has been implicated in 50% of patients. Fusion partners, including the tropomyosin (*TPM3/4*), clathrin heavy chain, and ran-binding protein 2 (*RANBP2*) genes, have been found in patients with ALK-positive IMT, while the reactive oxygen species (*ROS-1*) and platelet-derived growth factor (*PDGFRβ*) genes were found to be involved in ALK-negative IMT[18,19]. ALK is more commonly expressed in children and is associated with tumor aggressiveness and high recurrence rates[20]. Fibroblast proliferation is proposed to be the intermediate process of IMT formation, but the specific relationship remains to be elucidated[21]. In our case, kinase fusions of *ALK* gene might be a main mechanism. More importantly, based on the view that chronic inflammation is believed to be the cause, we proposed a new point of view that renal calculus is one of the causes of IMT. Chronic injury and inflammation of the kidney mucosa, promoting the process of tumorigenesis, were considered to be the intermediate stages of tumor formation. But more case studies are needed to support this opinion.

Kidney IMT lacks specific clinical symptoms. This disease starts insidiously and develops slowly, mainly occurring in middle-aged and elderly individuals, without a sex predilection. Kidney IMT is frequently circumscribed and confined to a single organ. Pain and hematuria are the most common symptoms, while some patients showed simple low back pain or abdominal pain, some showed simple gross hematuria, and others presented with both types of symptoms. A small number of cases were accompanied by systematic and biological symptoms, such as weight loss, intermittent fever, anemia, naupathia, urinary tract symptoms, weakness, night sweats, and thrombocytosis, which gradually resolved after treatment. In our case, the patient presented with painless gross hematuria and had a history of kidney calculi for 15 years, which was misdiagnosed as urinary calculi or urothelial neoplasms based on the initial examinations.

The imaging manifestations of kidney IMT are also nonspecific and inconclusive. Hypoechoic and intratumoral vascular distribution are shown on US imaging, which is difficult to differentiate from malignant tumors[22]. CT and magnetic resonance imaging (MRI) are more accurate for the diagnosis of kidney lesions. By CT imaging, most kidney IMTs frequently show ill-defined, hypodense, hypovascular, slightly homogeneously enhanced lesions or enhanced thick-wall cystic lesions[23,24]. An area of calcification can be seen in the tumor entities[25]. Kidney IMT often exhibits variable signal intensity on MRI T1-weighted imaging (T1WI) and low signal intensity on T2WI[26]. Although fluorodeoxyglucose (FDG) uptake in IMT vary, positron emission tomography/CT (PET/CT) may possess considerable advantages for detecting primary tumors as well as recurrence and distant metastasis[27]. In our case, the lesion exhibited mixed density, clear boundaries, and uneven enhancement on CT imaging, which is difficult to differentiate from renal malignant tumors. Unfortunately, MRI or PET/CT was not performed, as we did not suspect IMT. Thus, effectively identifying this rare disease by imaging alone is challenging.

Kidney IMT needs to be differentiated from malignant tumors, such as renal pelvic carcinoma, renal cell carcinoma (RCC), sarcomatoid renal cell carcinoma, cystic renal carcinoma, inflammatory fibrosarcoma, low-grade neurogenic tumor, malignant fibrous histiocytoma, and dendritic cell tumors and benign tumors, such as angiomyolipoma, xanthogranuloma pyelonephritis, and plasma cell granuloma. It is most important to distinguish renal IMT from RCC and sarcomatoid renal cell carcinoma. Cystic renal IMT should be carefully differentiated from cystic renal cancer[28]. In our case, the lesion was assumed to be a renal malignant tumor that had most likely metastasized to the ipsilateral adrenal gland before surgery. In actuality, it is often misdiagnosed as a malignancy, which accounts for at least 67% (33/49) according to our calculations.

The final diagnosis usually depends on histopathological and immunohistochemical features. The most common histomorphology is characterized by the differentiation of myofibroblast-derived spindle cells and abundant infiltrative chronic inflammatory components, such as eosinophils and lymphocytes. Mucinous, vascular, and inflammatory areas and dense plate-like collagen are also important histologic features[29]. In addition, osteoblastic cells and heteromorphic bone composition are observed in certain cases[30]. Immunohistochemical analyses confirmed that vimentin, muscle-specific actin, smooth muscle actin (SMA), and CK were positive in IMT[29]. Furthermore, Li *et al*[31] described that the positive rates of vimentin, SMA, desmin, ALK, and CK in 30 patients with extrapulmonary IMT were 100%, 70%, 27%, 27%, and 13%, respectively.

In clinical practice, surgical resection of the lesion is recommended as the first-line treatment for IMT. For patients with only one kidney, bilateral masses, or kidney insufficiency, a biopsy and intraoperative rapid pathological examination were strongly advocated by some authors because of the advantages in excluding malignant tumors and avoiding unnecessary removal[14]. But these approaches are still controversial, as the correct diagnosis is usually made only after nephrectomy[15]. Fortunately, with the widespread application of partial nephrectomy, the function of kidneys are saved gradually.

In addition to surgery, targeted therapy and chemotherapy are also important adjuvant therapies. Based on the importance of the ALK pathway in the progression of IMT, ALK-targeted inhibitors, such as crizotinib, which are mostly used in the treatment of patients with metastatic or unresectable ALK-positive IMT, are effective and even offer surgical opportunities for these patients[32]. However, tumor cells may acquire resistance during the treatment process, which may limit their efficacy[33]. Anti-inflammatory therapy, such as nonsteroidal anti-inflammatory drugs (NSAIDs), is reported to be effective in ALK-negative patients for whom ALK inhibition is not an option[34]. NSAIDs may shrink the tumor and possibly cure the disease, and thus could be used as a diagnostic treatment[34]. If antibiotics cure IMTs, an inflammatory origin may be suggested[35]. Corticosteroids are also a great treatment option, especially for young patients and bilateral renal-infiltrating IMT[36].

Radiotherapy is mainly used for the treatment of IMT in the head and neck; the use of radiotherapy for kidney IMT has not been reported. Studies have shown that high-dose fractional radiotherapy is effective and safe for nasopharynx and skull IMT[37]. Therefore, radiation therapy may be an option in cases where anti-inflammatory drugs, steroids, and surgery are ineffective in controlling kidney IMT. The patient in our case underwent surgery, no other follow-up treatments were performed, and the patient recovered well. Surprisingly, antibiotics were not effective in this case.

The outcome of kidney IMT is quite favorable. Recurrence or metastasis has not yet been discovered from the cases we reviewed, which is consistent with Kapusta's studywith the longest follow-up period being 17 years[29]. However, IMT has been simultaneously detected in multiple tissues, but it was not clear whether they were primary tumors or whether one had metastasized from the other[7]. Worryingly, local recurrence and distant metastasis have been reported in IMT outside of the kidney. The risk factors for recurrence mainly include positive surgical margins, nuclear atypia, ganglion-like cells, DNA aneuploidy, and the abnormal expression of p53[38]. However, there is no consensus on the reliable predictors of the biological behavior of IMT. Although IMT is not a malignant tumor, it is reported to be associated with RCC, which might affect the management and prognosis of patients[10]. Therefore, close follow-up must be advocated in such cases.

**CONCLUSION**

Kidney IMT is an uncommon neoplasm with an unknown etiology and diverse clinical manifestations. Renal calculus is suspected to be one of the causes or drivers. Despite its rarity, awareness of its existence is essential to avoid misdiagnosis in the differential diagnosis of a renal mass. Surgery and adjuvant chemotherapy are the current mainstream treatments. Although most patients obtain satisfactory results, follow-up is still indispensable because of the potential for recurrence and metastasis.

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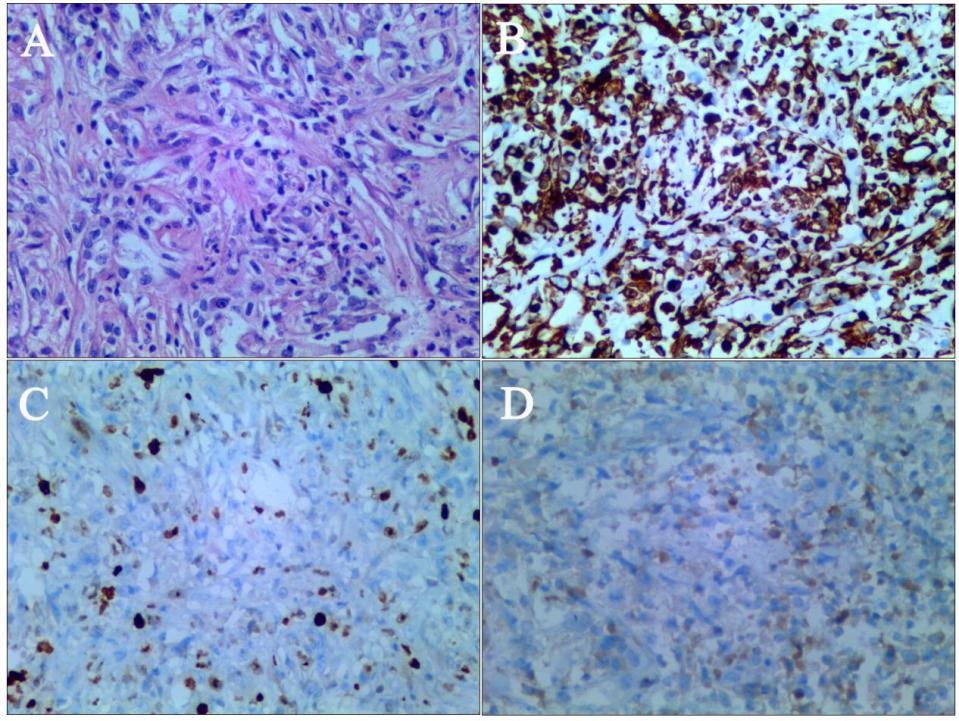
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图一

**Figure 1 Pre-operative computed tomography images.** A: computed tomography (CT) image showing a 4.1 cm × 3.2 cm mass in the middle pole of the left kidney. B: Contrast-enhanced CT image showing slight enhancement.



**Figure 2 Microscopic findings and immunohistochemical analysis results.** A: Photomicrograph showing abundant spindle cells and collagen with infiltrating lymphocytes and plasma cells (hematoxylin-eosin staining; magnification, ×200); B-D: Immunohistochemically, the tumor cells were positive for (B) vimentin, (C) Ki-67, and (D) CK (magnification, ×200).

**Table 1 Summary of clinical features of reported cases of kidney inflammatory myofibroblastic tumor**

|  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Case** | **ref.** | **Age (yr)/gender** | **Clinical features** | **Location** | **Size (cm)** | **Treatment** | **Follow-up (mo)** | **Recurrence** |
| 1 | [Heerwagen](https://www.ncbi.nlm.nih.gov/pubmed/?term=Heerwagen%20ST%5bAuthor%5d&cauthor=true&cauthor_uid=17963086) *et al*[6] | 75/M | Accident finding, anemia | Rt lower | 10 | Nephrectomy | 10 | No |
| 2 | [Gwynn](https://www.ncbi.nlm.nih.gov/pubmed/?term=Gwynn%20ES%5bAuthor%5d&cauthor=true&cauthor_uid=16230166) *et al*[10] | 46/M | Abdominal discomfort, gastrointestinal symptoms | Rt | 13 | Nephrectomy | 13 | No |
| 3 | [Tarhan](https://www.ncbi.nlm.nih.gov/pubmed/?term=Tarhan%20F%5bAuthor%5d&cauthor=true&cauthor_uid=15368679) *et al*[23] | 10/F | Recurrent high fever, headache | Rt middle | NA | Nephrectomy | 18 | No |
| 4 | Wu *et al*[32] | 43/M | Low back pain, hematuria | Lt renal pelvis | 1.5 | Nephrectomy | 26 | No |
| 5 | Li *et al*[36] | 61/M | Lethargy, night sweats, weight loss | Rt and Lt | NA | Biopsy, corticosteroid | 48 | No |
| 6 | [Liang](https://www.ncbi.nlm.nih.gov/pubmed/?term=Liang%20W%5bAuthor%5d&cauthor=true&cauthor_uid=26181565)[28] | 60/F | Accident finding | Lt upper | 10.1 | Partial nephrectomy | 63 | No |
| 7 | Li *et al*[9] | 48/F | Accident finding | Lt upper | 2.9 | Nephrectomy | 6 | No |
| 8 | [Dogan](https://www.ncbi.nlm.nih.gov/pubmed/?term=Dogan%20MS%5bAuthor%5d&cauthor=true&cauthor_uid=26018807) *et al*[7] | 3/M | Low back pain, recurrent fever | Rt upper | 6 | Nephrectomy | 6 | No |
| 9 | Ho *et al*[14] | 3/F | Abdominal pain, intermittent fever | Lt UPJ | 8 | Conservative surgery | 9 | No |
| 10 | Sun *et al*[21] | 54/F | Accident finding | Rt upper | 1.5 | Nephrectomy | NA | NA |
| 11 | [Nakamura](https://www.ncbi.nlm.nih.gov/pubmed/?term=Nakamura%20Y%5bAuthor%5d&cauthor=true&cauthor_uid=18085406) *et al*[26] | 60/M | Accident finding | Lt middle | 5.5 | Nephrectomy | NA | No |
| 12 | Tazi *et al*[15] | 56/M | Low back pain, weight loss | Rt lower | 6 | Nephrectomy | NA | NA |
| 13 | [Sugimoto](https://www.ncbi.nlm.nih.gov/pubmed/?term=Sugimoto%20K%5bAuthor%5d&cauthor=true&cauthor_uid=15518129) *et al*[35] | 20/F | High fever, right lumbago | Rt renal pelvis | 2.5 | Antibiotics | NA | NA |
| 14 | [Babu](https://www.ncbi.nlm.nih.gov/pubmed/?term=Babu%20P%5bAuthor%5d&cauthor=true&cauthor_uid=26458713) *et al*[24] | 51/M | Low back pain | Lt upper | 8.2 | Nephrectomy | NA | NA |
| 15 | [Ishikawa](https://www.ncbi.nlm.nih.gov/pubmed/?term=Ishikawa%20T%5bAuthor%5d&cauthor=true&cauthor_uid=15147553) *et al*[25] | 38/M | Accident finding | Lt upper | 11 | Nephrectomy | NA | NA |
| 16 | [Khallouk](https://www.ncbi.nlm.nih.gov/pubmed/?term=Khallouk%20A%5bAuthor%5d&cauthor=true&cauthor_uid=21864369) *et al*[39] | 57/M | Low back pain, hematuria | Rt middle | 9 | Nephrectomy | 14 | No |
| 17 | Taheri *et al*[40] | 15/F | Low back pain, hematuria, weight loss | Lt upper | 13.5 | Nephrectomy and adrenalectomy | 12 | No |
| 18 | Selvan *et al*[41] | 56/M | Accident finding | Lt middle | NA | Nephrectomy | 12 | No |
| 19 | Leroy *et al*[42] | 37/M | Low back pain, hematuria | Rt renal pelvis | NA | Nephrectomy | 12 | No |
| 20 | Navale *et al*[30] | 32/M | Lower abdominal pain, urinary tract symptoms | Rt lower | 4.5 | Nephrectomy | 18 | No |
| 21 | Bektas *et al*[43] | 51/F | Low back pain | Lt upper | 4.5 | Nephrectomy and adrenalectomy | 18 | No |
| 22 | Heerwagen *et al*[6] | 40/F | Accident finding | Rt lower | 7 | Nephrectomy | 24 | No |
| 23 | Pothadiyil *et al*[44] | 50/M | Low back pain | Lt | NA | Nephrectomy | 24 | No |
| 24 | [Vujanić](https://www.ncbi.nlm.nih.gov/pubmed/?term=Vujani%C4%87%20GM%5bAuthor%5d&cauthor=true&cauthor_uid=1409154) *et al*[45] | 8/M | Painless gross hematuria | Rt lower | 6 | Nephrectomy | 36 | No |
| 25 | Epaulard *et al*[46] | 60/M | Fever, weight loss, weakness, night sweats | Rt middle | 3 | Nephrectomy | 3 | No |
| 26 | [Sugimoto](https://www.ncbi.nlm.nih.gov/pubmed/?term=Sugimoto%20T%5bAuthor%5d&cauthor=true&cauthor_uid=18260352) *et al*[47] | 61/M | Abdominal pain | Rt renal pelvis | 3 | Nephroureterectomy | 3 | No |
| 27 | [Ryu](https://www.ncbi.nlm.nih.gov/pubmed/?term=Ryu%20KH%5bAuthor%5d&cauthor=true&cauthor_uid=20119595) *et al*[48] | 61/F | Accident finding, weight loss, gastrointestinal symptoms | Lt lower | 3 | Nephrectomy | 3 | No |
| 28 | [Iida](https://www.ncbi.nlm.nih.gov/pubmed/?term=Iida%20K%5bAuthor%5d&cauthor=true&cauthor_uid=25511942) *et al*[49] | 54/F | Painless gross hematuria | Rt renal pelvis | 4.7 | Nephroureterectomy | 44 | No |
| 29 | Wang *et al*[50] | 74/F | Low back pain, weight loss | Lt upper | 8 | Nephrectomy | 66 | No |
| 30 | Boo *et al*[51] | 9/F | intermittent abdominal pain, weight loss | Lt upper | 5.5 | Nephrectomy and enlarged lymph dissection | 6 | No |
| 31 | Leroy *et al*[42] | 54/F | Low back pain, hematuria | Lt renal pelvis | 1.5 | Nephrectomy | 7 | No |
| 32 | [Petrescu](https://www.ncbi.nlm.nih.gov/pubmed/?term=Petrescu%20A%5bAuthor%5d&cauthor=true&cauthor_uid=18060198) *et al*[52] | 57/M | Hematuria, minimal grade fever, right flank pain | Rt middle | 2 | Nephroureterectomy | NA | NA |
| 33 | [Mukkamala](https://www.ncbi.nlm.nih.gov/pubmed/?term=Mukkamala%20A%5bAuthor%5d&cauthor=true&cauthor_uid=28727376) *et al*[53] | 54/M | Accident finding | Lt middle | 4.7 | Nephrectomy | NA | NA |
| 34 | [Hori](https://www.ncbi.nlm.nih.gov/pubmed/?term=Hori%20J%5bAuthor%5d&cauthor=true&cauthor_uid=19470689) *et al*[54] | 24/F | Accident finding | Lt | 7.5 | Nephrectomy | NA | NA |
| 35 | [Yoshida](https://www.ncbi.nlm.nih.gov/pubmed/?term=Yoshida%20S%5bAuthor%5d&cauthor=true&cauthor_uid=16479987) *et al*[55] | 44/M | Painless gross hematuria | Lt UPJ | NA | Nephroureterectomy | NA | NA |
| 36 | [Bildirici](https://www.ncbi.nlm.nih.gov/pubmed/?term=Bildirici%20K%5bAuthor%5d&cauthor=true&cauthor_uid=15368680) *et al*[56] | 42/F | Low back pain | Rt upper | 7 | Nephrectomy | NA | NA |
| 37 | [Kobayashi *et al*](https://www.ncbi.nlm.nih.gov/pubmed/?term=Kobayashi%20TK%5bAuthor%5d&cauthor=true&cauthor_uid=10834015)[57] | 50/M | Accident finding | Rt middle | 3 | Corticosteroid | NA | NA |
| 38 | Lee *et al*[58] | 48/M | Accident finding | Rt middle | 3.5 | Partial nephrectomy | NA | NA |
| 39 | [Iwaki *et al*[59]](https://www.ncbi.nlm.nih.gov/pubmed/?term=Iwaki%20H%5bAuthor%5d&cauthor=true&cauthor_uid=11517572) | 73/M | General malaise, minimal grade fever | Lt lower | 3.5 | Nephrectomy | NA | NA |
| 40 | [Kim](https://www.ncbi.nlm.nih.gov/pubmed/?term=Kim%20TJ%5bAuthor%5d&cauthor=true&cauthor_uid=11752959) *et al*[60] | 60/M | Painless gross hematuria | Rt renal pelvis | 1.6 | Nephrectomy | NA | NA |
| 41 | [Satoh](https://www.ncbi.nlm.nih.gov/pubmed/?term=Satoh%20E%5bAuthor%5d&cauthor=true&cauthor_uid=10723660) *et al*[61] | 61/M | Painless gross hematuria | Rt | 1.8 | Nephroureterectomy | NA | NA |
| 42 | [Shah](https://www.ncbi.nlm.nih.gov/pubmed/?term=Shah%20VB%5bAuthor%5d&cauthor=true&cauthor_uid=22234141) *et al*[62] | 42/F | Abdominal pain, hematuria | Rt renal pelvis | 6 | Nephrectomy | NA | NA |
| 43 | [Bildirici](https://www.ncbi.nlm.nih.gov/pubmed/?term=Bildirici%20K%5bAuthor%5d&cauthor=true&cauthor_uid=15368680) *et al*[56] | 42/F | Low back pain | Rt upper | 7 | Nephrectomy | 12 | No |
| 44 | [Doga](https://www.ncbi.nlm.nih.gov/pubmed/?term=Dogra%20VS%5bAuthor%5d&cauthor=true&cauthor_uid=20068512)n *et al*[7] | 15/F | Abdominal pain,  recurrent fever | Lt upper | 7 | Biopsy, antibiotics | 8 | No |
| 45 | [Bell](https://www.ncbi.nlm.nih.gov/pubmed/?term=Bell%20ND%5bAuthor%5d&cauthor=true&cauthor_uid=9824189) *et al*[63] | 37/M | Abdominal pain, hematuria, fever | Lt upper | 2.5 | Nephrectomy | 8 | No |
| 46 | [Ma *et al*[64]](https://www.ncbi.nlm.nih.gov/pubmed/?term=Ma%20Y%5bAuthor%5d&cauthor=true&cauthor_uid=18155540) | 42/F | Abdominal pain, nausea, fatigue, weight loss | Rt and Lt | NA | Biopsy, corticosteroid | NA | No |
| 47 | [Tambo](https://www.ncbi.nlm.nih.gov/pubmed/?term=Tambo%20M%5bAuthor%5d&cauthor=true&cauthor_uid=12822456) *et al*[65] | 46/F | Accident finding,  microscopic haematuria | Rt | 5 | Retroperitoneal tumor resection and additional right nephrectomy | 9 | No |
| 48 | [Hattori](https://www.ncbi.nlm.nih.gov/pubmed/?term=Hattori%20S%5bAuthor%5d&cauthor=true&cauthor_uid=19068729) *et al*[66] | 60/F | Low back pain | Lt renal pelvis | 4 | Nephroureterectomy | NA | NA |
| 49 | Present case | 77/F | Painless gross hematuria | Lt middle | 4 | Nephrectomy and adrenalectomy | 22 | No |

M: male; F: female; Rt: right; Lt: left; NA: data not available; UPJ: uretero pelvic junction.