# Inflammatory Myofibroblastic Tumor of Urinary Bladder

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To the Editor: Inflammatory myofibroblastic tumor of the urinary bladder (IMTUB) is rare clinically. To the best of our knowledge, few Asian cases with IMTUB have been reported currently. IMTUB often mimics soft-tissue sarcoma both clinically and radiologically, which is thereby often mistaken for sarcoma. Typically, surgical resection is its major treatment, but the role of adjuvant or palliative radiochemotherapy remains unclear so far. Herein, this case reported a Chinese female with IMTUB.

A 39-year-old female was admitted to the Yantai Affiliated Hospital of Binzhou Medical University due to massive visible hematuria for one month. She presented with a 1-year history of recurrent urinary tract infection, with no other notable past medical histories. Physical examination on admission revealed no suprapubic pain or palpable abdominal mass. Urinalysis suggested an increased number of homogeneous red blood cells. Moreover, routine blood examination indicated infection and anemia, with a leukocyte count of 12.1 × $10^9$ cells/L and a hemoglobin level of 92 g/L. No abnormality was discovered in other laboratory tests, and malignant cells were not detected in urinary cytology. Ultrasound examination suggested a hypoechoic solid mass in the urinary bladder, in which blood flow signals could be observed. Besides, computed tomography (CT) scanning revealed a visible mass at the top of the urinary bladder, with the greatest cross-sectional area of about 4.5 cm × 4.2 cm, which was well defined with irregular soft-tissue density [Figure 1a]. At the same time, no enlarged lymph nodes were seen in the abdominal cavity or retroperitoneum. In addition, cystoscopy showed a poorly defined mass on the anterior wall of the bladder. Afterward, the patient was treated with radical transurethral resection (TUR). Further histopathological analysis demonstrated the abnormal proliferation of spindle cells with no significant nuclear atypia, together with pleomorphism, partial necrosis, and a low rate of mitosis accompanied by numerous inflammatory cell infiltration (including lymphocytes, eosinophils, and macrophages): SMA+, Desmin−, S-100−, CD34−, CK20+, and CK7+ [Figure 1b–1h]. All these histological findings were compatible with a very rare entity, namely, IMTUB. The symptoms of the patient were not markedly improved after surgery. Subsequently, partial cystectomy was performed to thoroughly resect the tumor. The patient had well recovered at the 1-year follow-up, without hematuria, frequency of urination, and urgent urination. Besides, cystoscopy revealed no evidence of tumor recurrence or distant metastasis.

Inflammatory myofibroblastic tumor (IMT), first described in lung, is the most frequently reported entity. Typically, urinary bladder in genitourinary tract is the most common anatomical site of the origin of this lesion. Besides, other locations in the genitourinary tract are kidney, urethra, prostate, ureter, and rete testis. Most of these tumors are benign, but they are also associated with a certain malignant potential. Tumor invasion and metastasis may occur, mostly at the lateral wall and top wall of the bladder, but rarely in the triangle of the bladder.[1] In our case, the tumor was located at the top of the bladder.

The etiology and pathogenesis of IMT remain unclear at present, which may be related to the following aspects. First, chronic infection has been regarded as an important factor in the pathogenesis of IMTUB. Some microbiological infections, such as mycobacteria, viral hepatitis B, *Corynebacterium*, Epstein-Barr virus, and human papillomavirus, may also play a potential role in the formation of IMT.[2,3] It may also be associated with the history of bladder trauma, application of steroid drugs, and placement of polypropylene mesh (TVT) surgery.[4] Third, the occurrence of IMTUB may be correlated with the overexpression of cyclin D1, mouse double microbody 2, interleukin-6, human herpesvirus 8, and p53. Meanwhile, the rearrangements involving the anaplastic lymphoma kinase (*ALK*) gene on chromosome 2p23 may also be implicated in IMT occurrence. Finally, trauma, vascular causes, and autoimmune disorders have also been proposed to play a certain role.[3] In this case, recurrent urinary tract infection may represent an inciting event of IMT. However, the bladder tumor can cause obstruction of the lower urinary tract, followed by secondary infection, which shows symptoms of urinary irritation. Therefore, the pathogenesis of IMT required to be further studied.

Specifically, the most common initial manifestation of IMT is painless gross hematuria, accompanying with other symptoms such as frequency of urination, dysuria, abdominal/pelvic pain, and obstructive symptoms.[2] Radiological imaging is of crucial importance in the preoperative diagnosis and follow-up of patients with IMTUB. Typically, ultrasound, contrast-enhanced

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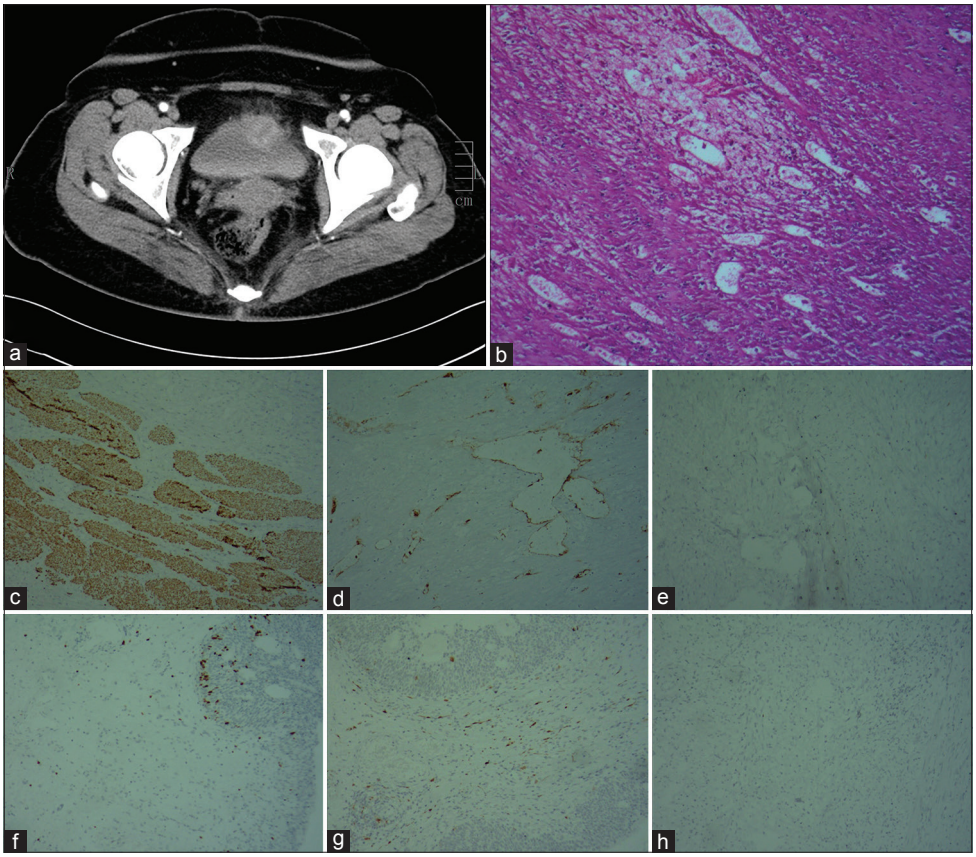


**Figure 1:** A 39-year-old female with inflammatory myofibroblastic tumor of the urinary bladder. (a) Computed tomography showing bladder mass on top wall. (b) Inflammatory myofibroblastic tumor with cytologically bland spindle cells admixed with chronic inflammatory cells and plasma cell infiltrate (HE staining, original magnification ×100). Immunohistochemical test showing negative S-100 stain (c), negative Desmin stain (d), negative CD34 stain (e), positive CK7 stain (f), positive SMA stain (g), and positive CK20 stain (h) (original magnification, ×100).

CT, and magnetic resonance imaging (MRI) can provide valuable information on tumor size and evidence of invasion to the urinary bladder wall. Depending on the particular case, IMTUB can appear as a well-defined and nonspecific mass on ultrasound scanning; alternatively, it may seem to be simple bladder-wall thickening with variable echogenicity described as hypoechoic or hyperechoic. CT examination often shows either a focal or diffuse thickening of bladder wall, with few single tumors; moreover, the lesion may be hypodense and isodense or hyperdense, which is well enhanced after intravenous administration of contrast medium. In MRI, IMTUB is characterized by the low T1 signal intensity and nonhomogeneous high T2 signal intensity. In some cases, IMTUB may show high signal intensity centrally and low signal peripherally on T2 images, reflecting central necrosis. Cystoscopy is particularly important since it may indicate the tumor size and location. Preoperative cystoscopy is the most convenient and helpful method for biopsy diagnosis. Tumor is rounding, cauliflower-like or broad-based in clump, with rough surface, and covered with visible tissue necrosis as well as bleeding under cystoscopy. Besides, biopsy can give a preliminary diagnosis of IMTUB.

Pathological examination maintains the gold standard for diagnosing IMTUB. Histologically, IMTUB is characterized by three features including loose stellate cells in the mucoid background with inflammatory cell infiltration, fascicular spindle cells, and scattered collagenous areas. The tumor is mostly gray-white; in a few cases, there are fish-like changes. Besides, necrosis is rare, and a large number of spindle cells can be seen under microscope, accompanying with a variety of thin inflammatory cells. Moreover, there are more mitotic figures in tumor, but abnormal mitotic figures are rare. Typically, immunohistochemical staining is also significant in that it can confirm the immunophenotype of IMTUB, thus contributing to identifying IMTUB. In our case, results of immunochemical analysis reveal that the spindle cells are positive for ALK (50–60%), vimentin (95–100%), smooth muscle actin (48–100%), cytokeratin (10–89%), desmin (5–80%), and actin (62%), whereas negative for S-100 protein, epithelial membrane antigen, and p53. Recently, ALK protein expression is detected in IMTUB. Notably, IMTUB should be differentially diagnosed with sarcomatoid urothelial carcinoma, leiomyosarcoma, and rhabdomyosarcoma based on their similarities in histological findings. Importantly, findings of cytological atypia and atypical mitotic figures, as well as immunostaining with ALK are useful to differentiate IMTUB from other malignant spindle cell tumors.

At present, surgical resection is the preferred treatment for IMTUB, such as TUR of bladder tumor, partial cystectomy, and radical cystectomy, among which, TUR minimally invasive, and some postoperative patients required a second operation due to recurrence or other causes. Comparatively, patients undergoing partial cystectomy are associated with a lower incidence of re-operation.[5] Therefore, partial resection of bladder is the first choice, followed by TUR of bladder tumor. Generally, a surgical method is selected depending on different individuals. However, postoperative intravesical instillation remains a source of controversy, which requires to be proved in more studies. Some researchers suggested that anti-infection treatment can be adopted to reduce tumor size and

even cure the tumor. Moreover, the application of ALK inhibitors has also been reported in treating IMT.

Most IMTUB is stable or subsides after incomplete resection of bladder; nonetheless, there is still a possibility of recurrence and metastasis of IMTUB. Concretely, the recurrence rate is up to 10–25%,[6] and the prognosis for IMTUB is related to the tumor size, histological expression, and tumor removal. With the application of minimally invasive techniques such as laparoscopy, advanced treatment of chemotherapy or ALK inhibitor therapy is suggested to be carried out first for tumor >5 cm in diameter, followed by laparoscopic partial cystectomy or radical resection after tumor shrinking. Furthermore, pre- and postoperative full anti-infection treatment is of great help to tumor <3 cm in diameter in patients undergoing TUR of tumor or partial cystectomy. However, routine perfusion therapy after operation is not recommended. Besides, interventional embolization has not been reported in IMTUB; nevertheless, it is an alternative scheme for large tumors and elderly patients who cannot tolerate surgery. IMTUB should be followed up regularly after surgery, which can be ascribed to its possibility of recurrence and metastasis. It is suggested that cystoscopy and urinary B ultrasound or lower abdominal CT should be adopted in follow-up examination every 3–6 months within the 1st year, and cystoscopy can be repeated once every 6 months 2 years after surgery. For patients without recurrence, the urinary system can be checked once at least every 6 months.

## Declaration of patient consent

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


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## Conflicts of interest

There are no conflicts of interest.

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