

Minimally invasive surgery for inflammatory myofibroblastic tumor of the urinary bladder

Three case reports

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Abstract

Rationale: Inflammatory myofibroblastic tumors of the urinary bladder (IMTUB) is exceptionally rare. Currently, no standardized treatment has been established for IMTUBs.

Patient concerns: Herein we report three cases presenting with hematuria and anemia. A 25-year-old man experienced painless gross hematuria for 2 days and the hemoglobin level continuously dropped to 88 g/L; a 72-year-old man complaining of gross hematuria for seven days; and a 33-year-old woman presenting with gross hematuria, urgency, and frequency for the duration of 20 days, with a hemoglobin level of 61 g/L.

Diagnosis: Ultrasonography, contrast-enhanced computed tomography (CT) scan and magnetic resonance image (MRI) indicated masses of different sizes on the walls of the urinary bladders. Diagnostic transurethral resection of bladder tumor (TURBT) was performed which revealed the diagnosis of IMTUB.

Interventions: In our cases, we removed the tumors completely with a minimally invasive approach. The first patient received TURBT only. The other patients underwent further laparoscopic and robot-assisted laparoscopic partial cystectomy respectively for the incomplete resection of tumor by diagnostic TURBT.

Outcomes: Histology of the resected specimen had proliferation of spindle cells with inflammation consistent with IMTUB. Immunohistochemical staining revealed that the tumor cells were positive for anaplastic lymphoma kinase (ALK), Vimentin and Ki-67 (20%–40%), negative for smooth muscle actin (SMA), S-100 and desmin confirming the diagnosis of IMTUB. Follow-up cystoscopy and CT or MRI (mean follow-up period: two years) did not detect any local recurrence or distant metastasis.

Lessons: Bladder-sparing treatment by TURBT or partial cystectomy remains the main mode of treatment for IMTUB. Laparoscopic and robot-assisted laparoscopic approach is safe and may yield satisfactory oncological and functional results. Regular follow-up protocol is necessary after operation.

Abbreviations: ALK = anaplastic lymphoma kinase, CT = computed tomography, IMT = inflammatory myofibroblastic tumors, IMTUB = inflammatory myofibroblastic tumor of the urinary bladder, MRI = magnetic resonance image, SMA = smooth muscle actin, TURBT = transurethral resection of bladder tumor.

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1. Introduction

Inflammatory myofibroblastic tumor (IMT) is proliferative lesions arising from submucosal stroma, of low or indeterminate malignant potential with the bladder being the most common site involved in the genitourinary tract.^[1] However, IMT of the urinary bladder (IMTUB) is rare and comprises less than 1% of all bladder tumors,^[2] which was firstly introduced by Roth in 1980.^[3] The average age of presentation is 28 years with hematuria and severe anemia being the most common manifestations.^[4] The treatment option is highly individualized and no standardized treatment protocol has been established for IMTUBs.^[5] Potential treatment options for the masses mainly include transurethral resection of bladder tumor (TURBT), partial cystectomy, and/or radical cystectomy. Herein we reported three cases of IMTUB, who were treated with minimally invasive surgery of TURBT, laparoscopic and robot-assisted laparoscopic partial cystectomy respectively at our institution.

2. Case report

2.1. Case 1

A 25-year-old man presented to our institution with painless gross hematuria for two days. He had no prior urologic disease history and no other medical problems. Complete blood count revealed the hemoglobin level of 104 g/L. Ultrasonography revealed a 46 mm × 25 mm hyperechoic mass in the bladder. Contrast-enhanced computed tomography (CT) scan confirmed a mass of 26 mm × 18 mm in size on the left bladder wall. The hemoglobin level continued dropping to 88 g/L before surgery.

After receiving the bladder irrigation and transfusion, emergency surgery of TURBT was performed because of persistent hematuria and continued dropping hemoglobin level. Cystoscopy revealed a broad-based mammillary tumor located on the left rear wall of urinary bladder. Postoperative histological examination of the resected specimen suggested the diagnosis of IMTUB (Fig. 1). Immunohistochemically, the tumor cells were positive for anaplastic lymphoma kinase (ALK), Vimentin, but negative for smooth muscle actin (SMA), S-100, P63, desmin, GATA3, CD34, myogenin, and β-catenin. No recurrence or progression was observed during the 2-year follow-up time.

2.2. Case 2

A 72-year-old man complained of gross hematuria for seven days was admitted to our department. Routine blood tests showed that the hemoglobin level was 110 g/L. Ultrasonography revealed a

heterogeneous mass in the bladder and CT scan confirmed a solid-cystic mass of 48 mm × 56 mm in size. We performed diagnostic TURBT, however, failed to remove the tumor completely due to the bottom of tumor located at the bladder diverticulum. The histology of the resected specimen revealed proliferation of spindle cells with inflammation which was consistent with IMTUB (Fig. 2). Immunohistochemical staining revealed the tumor cells were positive for ALK, Vimentin, and negative for SMA, desmin, S-100, CD34, and CK. The magnetic resonance image (MRI) 1 week after the surgery reported a mass in the posterior wall of bladder (Fig. 3A). The patient then underwent partial cystectomy by laparoscopic approach and the tumor resected was 5.0 cm in diameter (Fig. 3B). No local recurrence was observed during the 2-year follow-up.

2.3. Case 3

A 33-year-old woman presented with gross hematuria, urgency, and frequency for the duration of 20 days. No prior urologic disease or other medical problems were reported. Routine blood tests on admission showed that the hemoglobin level was 61 g/L. Ultrasonography revealed a 45 mm × 40 mm heterogeneous mass and contrast-enhanced CT scan revealed a 48 mm × 45 mm solid-cystic mass in the bladder. Diagnostic TURBT was then performed with the histology showed a proliferation of spindle cells with inflammation indicating the diagnosis of IMTUB. Immunohistochemical staining revealed the tumor cells were positive for ALK, SMA, desmin, Vimentin, but negative for S-100, CK, and CD34. However, the tumor could not be removed completely due to the location at the top of bladder. The patient underwent robot-assisted laparoscopic partial cystectomy thereafter and the surgery went out uneventfully. No local recurrence has been detected for the 2-year follow-up.

3. Discussion

IMTUB is a rare tumor of indeterminate malignant potential and comprises less than 1% of all bladder tumors.^[2] The common manifestations of IMTUB include anemia and gross hematuria, accompanied by pain sometimes during urination. Teoh et al^[6] reported that high possibility of IMTUB should be suspected for young patients presenting with gross hematuria and severe anemia. Our three cases were presented with severe hematuria, with blood clots in the bladder and reduced hemoglobin, which was in consistent with Teoh's findings. Radiological examinations usually cannot differentiate IMTUB from bladder cancer, therefore, it is difficult to make the final diagnosis before surgery.

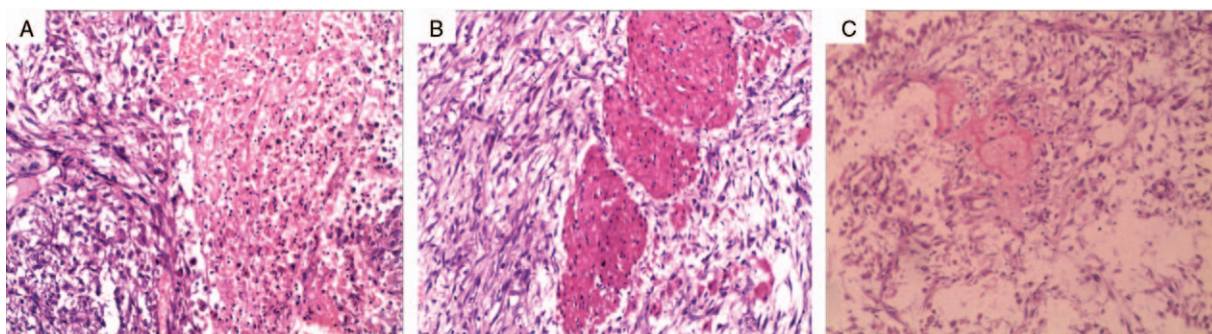


Figure 1. Histological examination of the biopsied specimen in patient 1 (hematoxylin-eosin staining, original magnification ×100) shows: (A) interstaggered spindle cells with atypia and necrosis; (B) the spindle cells involving on smooth muscle and (C) interstitial myxoid degeneration with scattered inflammatory cell infiltration.

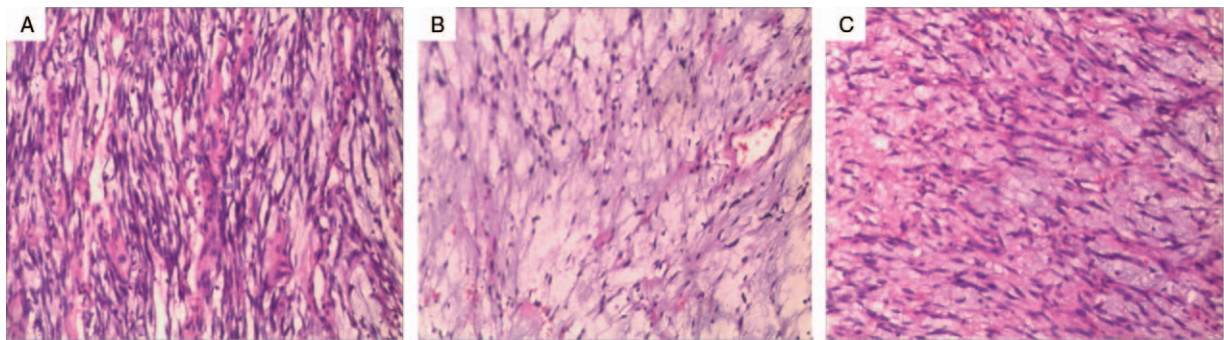


Figure 2. Histological examination of the specimen in patient 2 (hematoxylin-eosin staining, original magnification x100) shows: (A) the spindle cells arranging in bundle with mild atypia; (B) interstitial myxoid degeneration, and (C) lymphocytic infiltration.

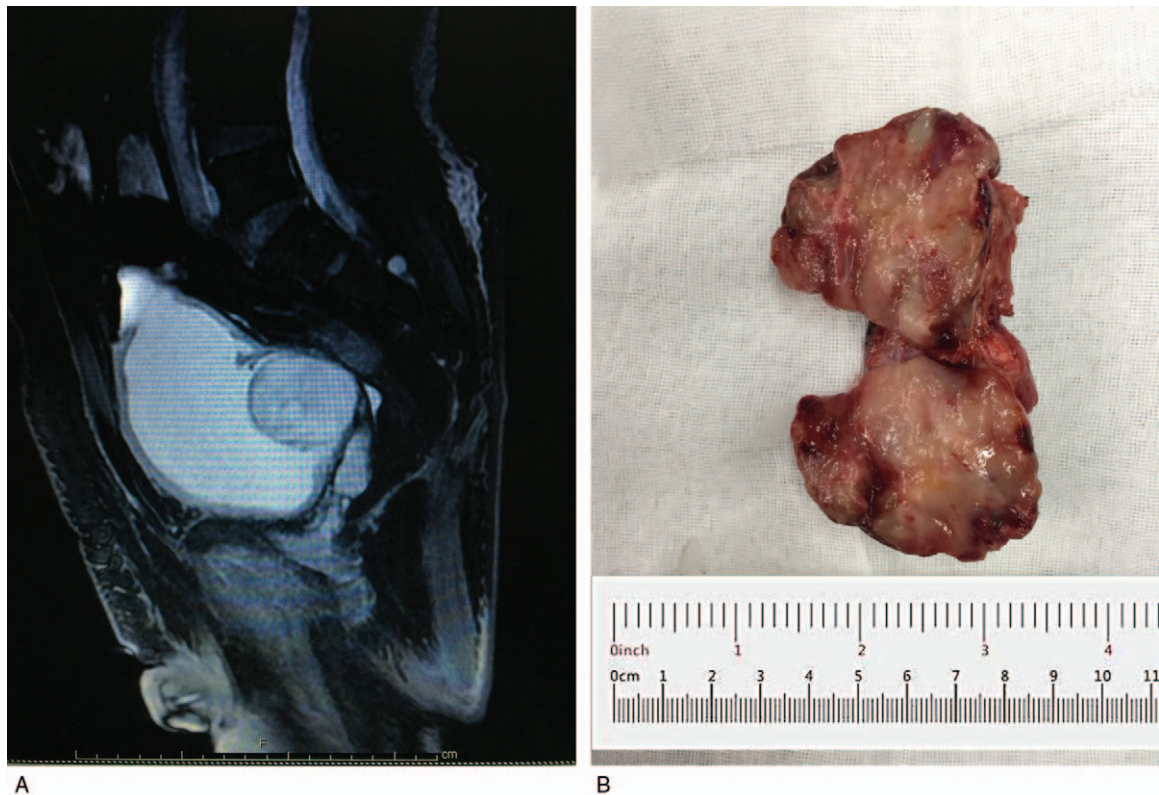


Figure 3. A. Post-operative MRI of patient 2 after TURBT: a mass in the posterior wall of bladder. B. Pathological specimen of patient 2 after laparoscopic partial cystectomy. MRI=magnetic resonance imaging, TURBT=transurethral resection of bladder tumor.

However, Liang et al^[7] found that IMTUBs commonly appeared on the anterior wall of the bladder. Ring enhancement on the contrast-enhanced CT of polypoid nodules may indicate the diagnostic imaging of IMTUB.

Due to the insufficiently specific imaging features, IMTUB is usually not considered as the first diagnosis. Histopathological examination is necessary for the accurate diagnosis. Histologically, IMTUB is divided into 3 types:

1. compact spindle cells accompanied with intermingled inflammatory cells (lymphocytes, eosinophils and plasma cells) resembling fibrous histiocytoma;

2. vascular, myxoid, and inflammatory areas resembling nodular fasciitis; and
3. dense plate-like collagen resembling a desmoid or scar.^[8]

Immunohistochemically, the tumor cells expressing ALK, SMA, and Vim in 65%, 71.9%, and 98.3% of the cases.^[9]

Currently, no standardized treatment protocol has been established for IMTUBs. Potential treatment options for the masses mainly include TURBT, partial cystectomy, and/or radical cystectomy.^[10] Given the benign property of IMTUB, bladder-sparing treatment by TURBT or partial cystectomy would not increase the risk of recurrence and intraperitoneal implantation

which was different from other aggressive urothelial cancer.^[11] And such minimally invasive method remains the main mode of treatment. In a systematic review, Teoh et al^[5] reported that most patients (60.8%) with IMTUB were treated by TURBT. However, for patients who were provided with TURBT initially, further treatment was re-provided including 5.5% with second TURBT, 17.8% of patients with partial cystectomy. Thus, partial cystectomy may be a better choice compared with TURBT especially for patients with tumor invading the muscularis propria.^[12]

Laparoscopic approach is safe in selected patients and may yield satisfactory oncological and functional results.^[13] Recently, Rotenberry et al^[2] reported a patient of IMTUB who was treated by robotic-assisted laparoscopic partial cystectomy with good prognosis. Compared with open partial cystectomy, laparoscopic or robot-assisted laparoscopic approach has several advantages, including cosmetic factors, lower risk of postoperative complications and shorter postoperative hospital stay. Challenges may also exist with the most obvious one aimed at the surgeon who is facing reconstructive procedure laparoscopically. The other 1 of note, from our experience, is that the surgeon should catheterize either 1 or both ureters before surgery to prevent inadvertent injuries of ureter or ureteral orifice while dissecting or suturing the incision. Moreover, laparoscopic and robot-assisted laparoscopic partial cystectomy may add additional operative time and increase cost for patients.

Distant metastasis of IMTUB has hardly been reported. In our three cases, we removed the masses completely with minimally invasive approaches. Follow-up cystoscopy, CT or MRI (mean follow-up period of 2 years) did not detect any local recurrence or distant metastasis. Nevertheless, Kim et al^[10] reported an unusual case of recurrent IMTUB with malignant transformation with multiple metastasis. This patient succumbed to his tumor after 1 and a half years. Accordingly, long-term follow-up is necessary for early identification of recurrence in patients with IMTUB after operation.

4. Conclusion

IMTUB is a rare tumor which is of low or indeterminate malignant potential. The diagnosis of IMTUB is mainly based on histological and immunohistochemical features. Currently, bladder-sparing treatment by TURBT or partial cystectomy remains the main mode of treatment. Laparoscopic and robot-assisted laparoscopic approach is safe and may yield satisfactory oncological and functional results. Regular follow-up protocol including cystoscopy and CT or MRI is necessary after operation.

Author contributions

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