Case Report

A large urinary bladder sarcoma treated with transurethral resection: a case report

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Abstract

The objective of this study is to report a case of a large urinary bladder sarcoma treated with transurethral resection and to evaluate the feasibility and efficacy of endoscopic surgery for urinary bladder sarcoma. We present a case report of a 64-year-old woman who underwent transurethral resection of a large urinary bladder sarcoma. Pathologic findings and immunohistochemistry were used to confirm the diagnosis of undifferentiated sarcoma. The patient was followed up for 48 months for local recurrence and distant metastasis. Nearly complete transurethral resection was performed, yielding 500 g of tissue. The patient underwent repeated transurethral resection because she declined radical surgery. The patient has survived for 48 months without local recurrence or metastasis. Transurethral resection could be an alternative option for urinary bladder sarcoma, particularly in cases where radical resection may not be feasible or desired by the patient. Careful selection of appropriate patients and long-term follow-up are crucial. Further studies are needed to evaluate the efficacy and safety of endoscopic surgery for urinary bladder sarcoma.

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Introduction

Urinary bladder sarcoma is a rare malignant tumor that arises from the mesenchymal tissue of the urinary bladder and represents 0.4% of all histological subtypes of primary invasive urinary bladder tumors.[1] Leiomyosarcoma and rhabdomyosarcoma are the two most prevalent subtypes, accounting for 50.0% and 20.0% of urinary bladder sarcoma, respectively. Additional subtypes include osteogenic sarcoma, angiosarcoma, myxoid liposarcoma, malignant fibrous histiocytoma, and carcinosarcoma.[2]

The standard treatment for urinary bladder sarcoma has not been established due to its rarity and the lack of randomized controlled trials. However, radical resection with negative margins is the cornerstone of treatment due to the aggressive nature of the disease.[3,4] There have, however, been no reports of endoscopic surgery as the only treatment for a large tumor. We thus present the case of a large undifferentiated urinary bladder sarcoma treated with transurethral resection, including the disease-free survival outcome.

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Case Report

A 64-year-old woman presented with painless gross hematuria and had experienced frequent urination for four months. She also felt a lump in her lower abdomen. Ultrasound from the referral hospital revealed a large pelvic mass in the differential diagnosis of an ovarian tumor. The gynecologist performed an exploratory laparotomy; however, no gynecological abnormalities were found intraoperatively except for a substantial anterior pelvic mass. The patient was, therefore, referred to our center for further evaluation.

Upon physical examination, a previous Pfannenstiel incision was observed, and a large firm pelvic mass was palpated two-thirds of the distance between the umbilicus and the pubic symphysis. Ultrasound revealed a 10-cm homogeneous hypoechoic mass in the pelvic cavity. Subsequent magnetic resonance imaging demonstrated a heterogeneous, hypo-intensity, intravesical mass measuring 9.4 x 8.9 x 7.7 cm on T1- and T2-weighted images, without definitive evidence of invasion into the perivesical tissue, although invasion into the detrusor muscle cannot be excluded. Hydronephrosis was not observed (Fig. 1). Based on these findings, the tumor was suggested to be at clinical stage T2, with no regional lymph node involvement or distant metastasis (N0M0). The cystoscopic findings showed a large pedunculated intravesical mass with a stalk located at the posterior wall of the urinary bladder and smooth overlying mucosa (Fig. 2). Both ureteral orifices were seen. The differential diagnoses from the cystoscopic findings included urothelial carcinoma or non-urothelial tumor (i.e., urinary bladder sarcoma).

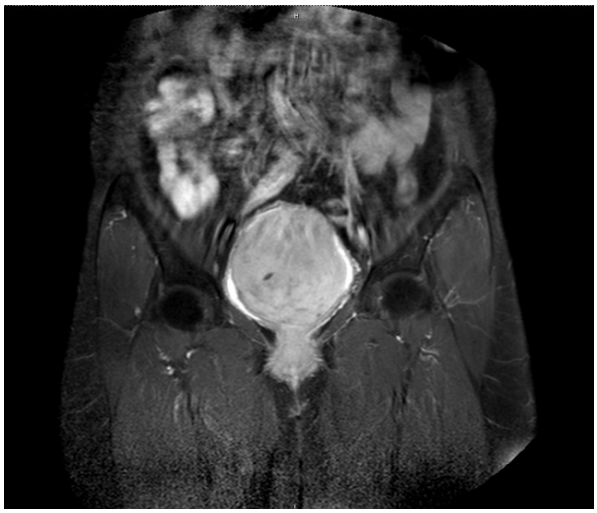

Figure 1. Coronal T2-weighted magnetic resonance imaging demonstrated a heterogeneous hypo-intensity intravesical mass, size 9.4x8.9x7.7 cm.

A nearly complete monopolar transurethral resection (TUR) was performed, yielding a tumor weight of 500 g. The tumor's base measured approximately 2 cm in diameter (Fig. 3). We used 79 l of sterile water for irrigation during the 5-h operation. The hospital course was uneventful, and the patient was discharged on the third postoperative day.

The histopathological findings revealed high-grade tumor cells arranged in sheets demonstrating infiltrative patterns and comprising elongated nuclei, hyperchromatic nuclei, moderate pleomorphism, and a lack of mitotic figures. The tumor invaded the detrusor muscle. Immunohistochemistry revealed GATA3 +, Vimentin +, AE1/AE3 -, Desmin -, SMA -, S100 -, EMA -, CK7 -,

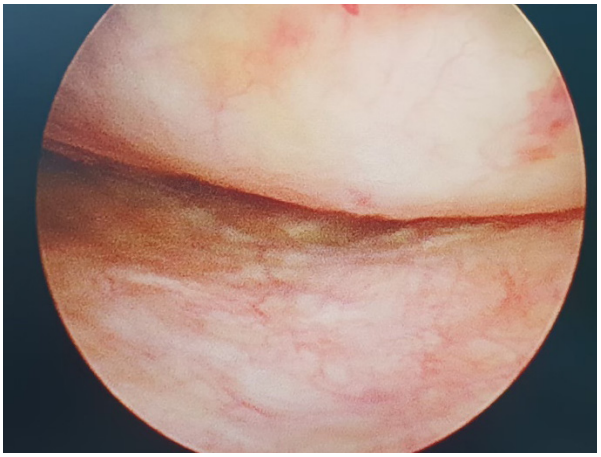

Figure 2. Cystoscopy showed the bladder tumor with normal overlying urothelium.

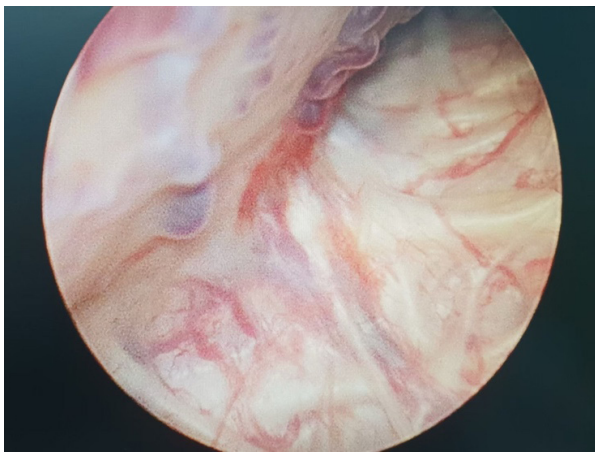

Figure 3. Cystoscopy showed the stalk of tumor.

CK20 -, CD117 -, and CD138 -, suggesting an undifferentiated sarcoma.

We discussed the curative treatment options, including partial cystectomy and anterior pelvic exenteration. After understanding the potential risks and benefits, the patient expressed a desire to preserve her bladder and maintain her quality of life, and she elected to undergo repeated TUR as an alternative management strategy, which aimed to remove as many tumors as possible. After resecting all gross disease, the pathological investigation indicated that the residual tumor had persisted. After discussing adjuvant treatment approaches, the patient declined chemo- and/or radiation therapy. We did, however, schedule her for surveillance cystoscopy every three months, which showed scar formation with negative biopsies (Fig. 4). The annual whole-abdomen computed tomography did not detect any abnormalities. Up to the end of our study, she was alive and had survived for 48 months with no evidence of a local recurrence or metastasis.

The Human Research Ethics Committee of Khon Kaen University reviewed and approved the study per the Helsinki Declaration and the Good Clinical Practice Guidelines (HE631371). Furthermore, the eligible patient signed informed consent before enrollment.

Discussion

We presented the case of urinary bladder sarcoma with a large pelvic mass and hematuria, which had been initially misdiagnosed as an ovarian tumor. Most urinary bladder sarcoma patients generally present with painless gross hematuria, followed by lower urinary symptoms.[3-5] However, these patients sometimes present a large mass at the time of diagnosis. According to a systematic review of 210 cases of urinary bladder leiomyosarcoma, the mean tumor size is 6 cm (range, 0.5-16).[5] Our case report highlights the importance of thorough medical history taking, physical examination, and appropriate investigations when diagnosing a patient with a pelvic mass. In particular, patients with a history of gross hematuria must undergo cystoscopy and cross-sectional imaging to initially rule out urinary tract cancer.

Urinary bladder sarcoma has various histopathological features, making a definitive diagnosis difficult; however, immunohistochemistry can help differentiate these subtypes of non-epithelial tumors.[4,6] In our patient, the positive result for the mesenchymal origin marker Vimentin and the negative results for epithelial markers [viz., pan-cytokeratin (AE1/AE3), CK7, CK20, and EMA] gave rise to the differential diagnosis of some type of sarcoma; however, the negative results for myogenic differentiation markers (Desmin and SMA) decreased the likelihood of its being leiomyosarcoma or rhabdomyosarcoma. Thus, the final pathological diagnosis suggested an undifferentiated sarcoma.

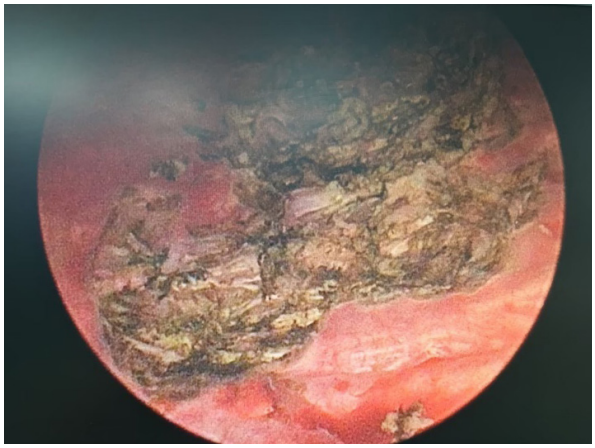

Figure 4. Cystoscopy showed the tumor's base located in the posterior wall of the urinary bladder after transurethral resection.

The Memorial Sloan Kettering Cancer Center (MSKCC) sarcoma staging system has been used to determine the prognosis of urinary bladder sarcoma in a small series. The prognostic factors include grade (low versus high), size of the tumor (≤ 5 cm versus > 5 cm), depth of invasion (superficial versus deep), and the presence of metastasis.[3] Russo et al reported that the 3-year relapse-free survival rate for stage 3 or 4 was only 26.0%. In contrast, Spiess et al reported a series of 19 patients with urinary bladder sarcoma and found that the 5-year disease-specific survival rate was 59.0%, with a median survival rate of 6 years, and the essential factor for recurrence-free survival was surgical margins status, not the stage of disease.[4] Thus, they concluded that complete surgical resection (radical or partial cystectomy) with negative resection margins offers the best treatment outcomes.[3,4]

Only a few studies have reported on the treatment outcomes of transurethral surgery. Russo et al reported that two patients with small leiomyosarcoma (2 cm) underwent complete TUR with

no evidence of local recurrence after seven years of follow-up.3 Rodriguez et al reported 183 cases of leiomyosarcoma of the urinary bladder; those patients who underwent cystectomy appeared to have a higher median disease-specific survival than those undergoing TUR only, albeit the difference was not statistically significant (48 and 24 months, respectively, p = 0.260). However, Rodriguez et al did not mention the size of the tumor.[7] Furthermore, chemotherapy and radiotherapy were proposed as multimodal treatments for local recurrence or distant metastasis, providing a wide range of therapeutic effects.[3-5,8]

We are the first to demonstrate a case of an extremely large urinary bladder sarcoma treated with TUR alone. Although the space for endoscopic resection is quite difficult, it had a narrow base (2 cm in diameter) and was located away from the ureteral orifices. Notwithstanding, we needed to discuss the chances of recurrence and/or metastasis with the patient. In addition, we needed to provide information about the risk of complications (i.e., urinary bladder perforation or TUR syndrome) that can occur due to the prolonged operative time needed for resection of a high-volume tumor.[9] The tumor was completely resectable using TUR without any complications. The patient survived for 48 months after TUR without any recurrence or metastasis; despite having been diagnosed as MSKCC stage 3 (a high-grade tumor > 5 cm). Further research is recommended to understand the nature of tissue invasion in a pedunculated urinary bladder sarcoma, whether it is related to the tumor's size or base diameter.

Conclusion

Urinary bladder sarcoma is a rare tumor, but it should be considered in the differential diagnosis of pelvic mass and hematuria. Although complete surgical resection provides the best curative treatment, TUR could be an alternative option worth considering in terms of its minimal invasiveness and urinary bladder preservation. We suggest that this approach be reserved for narrow-based, pedunculated tumors that can be completely resected, do not involve the ureteral orifice, and where imaging shows no evidence of perivesical invasion. Long-term follow-up should be required to monitor disease progression.

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Conflicts of Interest

The authors declare no conflicts of interest.

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