CASE REPORT

UROLOGICAL ONCOLOGY

Urinary incontinence as the first clinical symptom of urinary bladder leiomyosarcoma

Łukasz Mazurczyk, Michał Czarnogórski, Aleksandra Czernicka, Paweł Lipowski, Adam Ostrowski, Kajetan Juszczak, Jan Adamowicz, Tomasz Drewa

Department and Chair of Urology and Andrology, Ludwik Rydygier Collegium Medicum, Nicolaus Copernicus University, Bydgoszcz, Poland

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Corresponding author
Michał Czarnogórski
Department and Chair of Urology and Andrology,
Ludwik Rydygier Collegium Medicum,
Nicolaus Copernicus University,
Bydgoszcz, Poland
mcczarnogorski@gmail.com

Urinary bladder leiomyosarcoma is an extremely rare malignancy of the urogenital system.
We would like to present the case of a 59-year-old Caucasian male with a gigantic bladder leiomyosarcoma. The patient was subdued to the surgical excision of the urinary bladder – laparoscopic radical cystectomy with extended pelvic lymphadenectomy, with urinary diversion by bilateral ureterocutaneostomy. The excision was complete both macroscopically and microscopically. No additional adjuvant therapy was administered. In the 6-month follow-up, the patient remained in radiological remission.
The surgical excision with extended pelvic lymphadenectomy seems to be sufficient in the treatment of urinary bladder leiomyosarcoma.

Key Words: leiomyosarcoma ‹› urinary incontinence ‹› minimally invasive surgery

CASE REPORT

We present the case of a 59-year-old Caucasian male without any significant comorbidities, except benign prostate hyperplasia, who was admitted to the Department of Urology.
The patient experienced gradually worsening bladder outlet obstruction symptoms over 6 months. Last month, he experienced urinary incontinence, requiring the utilization of absorbent pads. Retrospectively, he also experienced abdominal fullness, regardless of food intake. He ascribed the symptoms to benign prostatic hyperplasia (BPH) and did seek help in the general practitioner's office.
Basic laboratory tests revealed microhematuria. Based on this sole finding, the patient was referred for an abdominal ultrasound, which showed a pathological mass in the urinary bladder, which at the time did not lead to obstructive uropathy. Based on the mentioned findings above, he was re-ferred for transurethral resection of bladder tumor (TURBT) performed in another hospital a month before admission to the Department of Urology. Due to the tumor size, complete resection was impossi-ble. Multiple biopsies were collected. The histopathological picture indicated leiomyosarcoma – low grade (LG). Differential diagnosis with myofibroblastic inflammatory tumor was performed. The image was ambiguous and required clinical and radiological correlation. Results of immunochistochemical tests are shown in Table 1.
A contrast-enhanced CT scan of the abdomen and pelvis performed a month before admission revealed a urinary bladder with heterogeneous masses,

probably filled with thrombi. In the lower part, the pathological mass of approx. 65 × 58 mm – approximate measurement due to poor secretion. The tumor appeared to grow beyond the walls, infiltrating the seminal vesicles and, in some cases, the prostate gland, including the ostium of the right ureter, with secondary dilatation of the pelvicalyceal system of the right kidney. There was no visible lymphadenopathy, except for a single left internal iliac lymph node up to 9 mm in the short axis (Figure 1). In the contrast-enhanced CT scan, there were no distant metastases.

If staged according to the abovementioned CT – the tumor would be classified as T2N0M0 (AJCC 8th).

After the diagnosis, the patient sought help in the tertiary referral center with experience with bladder tumor treatment with the use of minimally invasive methods and hence was referred to our Department. At admission, the patient had already developed bilateral stage I hydronephrosis with slightly elevated creatinine concentration (1.22 mg/dl) and macroscopic hematuria.

Abdominal ultrasound at admission revealed an extensive, heterogeneous, hyperechoic mass in the lower abdomen measuring 10 × 11 × 20 cm with visible marginal flows in the color Doppler option. However, vascularization was scarce in the central parts of the tumor. Also, in the center, there were homogenous, avascular areas with blurred borders, most probably necrosis-related areas.

After admission to the Department of Urology and reassessment in ultrasound and laboratory tests, the patient was qualified for bilateral percutaneous nephrostomies to prevent acute kidney injury due to obstructive uropathy. A urinary catheter was also placed. A multidisciplinary team consisting of urologists, radiologists, and oncologists qualified the patient for a radical surgical treatment – cystectomy.

After a month, the patient was admitted to the tertiary care center and, after necessary laboratory tests, qualified for surgery. According to the center's experience, the laparoscopic approach was utilized. The patient was placed on the operative table in a supine manner in Trendelenburg position, and the tumor was visible and palpable through the abdominal wall (Figure 2). Due to the tumor size, the higher placement of trocars was necessary in comparison to standard cystectomy. The 11 mm trocar was used for the 30° angular camera, and three 5 mm trocars for the surgical tools. A pneumoperitoneum of 15 mmHg was sufficient for proper visualization of the operative field. The surgery proceeded in the standard manner. The bilateral extended pelvic lymphadenectomy was performed. Due to the risk of bowel involvement, we decided to perform non-continent urinary diversion – ureterocutaneostomies. The procedure was finished with a laparoscopic technique alone. We did not find any macroscopic infiltration of the surrounding tissues.

Table 1. *Results of immunochistochemical tests*

Parameter	Value
Ki67	3%
Atypia	Slight
SMA	+
EMA	–
PAX8	+
Caldesmon	+
CD34	–
S100	+/–
CK	–
Desmin	+
ALK1	–

ALK1 – anaplastic lymphoma kinase 1; CK – cytokeratin; EMA – epithelial membrane antigen; SMA – smooth muscle actin

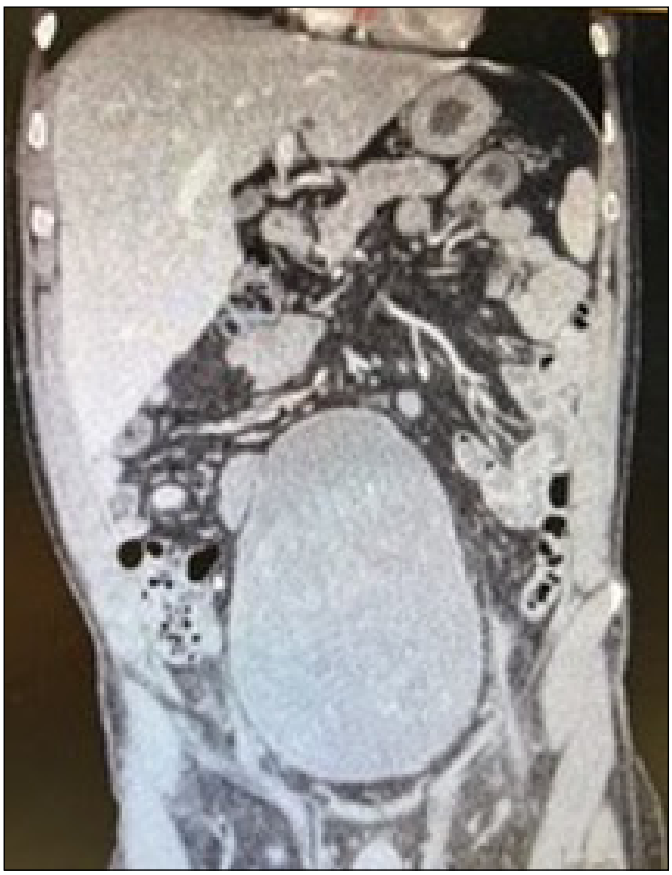

Figure 1. *CT scan of bladder tumor – coronal plane.*

Total blood loss was 300 ml. Due to the bladder's size, we performed Pfannenstiel's incision of approximately 20 cm for the removal of the bladder. The organ was entirely removed (Figures 3, 4 and subdued to histopathological assessment.

Table 2. *Results of immunochistochemical tests*

Parameter	Value
Ki67	5%
Desmin	–
SMA	+
S100	+
EMA	+
Myogenin	–
Calponin	+
ALK1	–
SOX10	–
TFE3	–
HMB45	–
MelanA	–
Mucicarmine	–

ALK1 – anaplastic lymphoma kinase 1; EMA – epithelial membrane antigen; HMB45 – human melanin black 45; SMA – smooth muscle actin

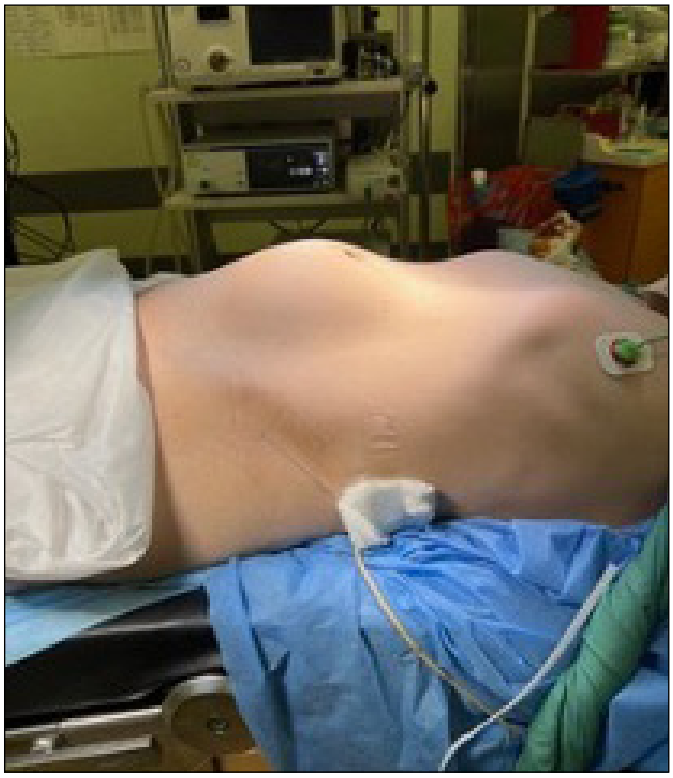

Figure 2. *Patient on the operative table. The tumor's impression on the soft tissues allows for easy identification.*

The sarcoma invaded the entire thickness of the muscularis propria (pT1). The urothelial epithelium over the tumor was normal, without any features of dysplasia. Perivesical fat tissue without cancer infiltration. Minimum tumor distance from the serosal surface – 0.1 cm. No features of angio- or neuroinvasion were visible. Number of mitoses per 10HPF: 4. Necrosis present – less than 50% of cells. Surgical margins were free from tumor infiltration. Immunohistochemistry (Table 2). Surgical specimen was 20 × 10 × 20 cm, including, among others, urinary bladder – 20 × 16 × 5 cm and tumor 15 × 17 × 8 cm, growing on the stem of 5 cm.

The tumor had a smooth surface with ecchymoses and areas of necrosis, comprising 15% of the tumor volume. It was limited to the bladder wall, infiltrating the tunica muscularis with focal infiltration of serosa. The surgical margin on the urethra was 3.5 cm.

The strict follow-up regimen was scheduled. The patient was to be tested every 3 months during the first year after the treatment. At present, the patient remains in follow-up for 6 months. At the 3-month follow-up visit, the patient had no deviations in physical examination and no signs of disease recurrence in contrast-enhanced chest, abdominal, and pelvic CT. Six months after surgery, the patient remains free of the disease's recurrence, as assessed by contrast-enhanced computed tomography. In the 6 months post-surgery, the patient remains content with the treatment and declares a satisfied quality of life.

DISCUSSION

Leiomyosarcoma (LMS) is a malignant tumor belonging to soft tissue sarcomas originating from smooth muscle tissue. It is the most common subtype among malignant mesenchymal tumors, accounting for approximately 10–20% of newly diagnosed soft tissue sarcomas [1].

In the immunohistochemical panel verifying the diagnosis, the LMS sample should express smooth muscle actin (SMA), desmin, and h-caldesmon with negative markers CD117, CD34, and DOG1, which distinguish LMS from GIST [2, 3].

LMS can develop within any smooth muscle tissue in proximity to blood vessels. In 35% of cases, this type of neoplasm is located in the retroperitoneal space and the intra-abdominal area; 30% of the locations are the uterus, followed by the limbs and trunk [4].

Leiomyosarcoma of the urinary bladder is very rare and accounts for approximately 0.1% of bladder malignancies, and is associated with an unfavorable prognosis [5].

In the early stages of the disease, the 5-year overall survival may reach 50% [6]. However, that data is extrapolated from uterine Leiomyosarcoma, a more common disease than bladder leiomyosarcoma. Considering the prognosis and treatment, we have only data from case reports and case series, which contain heterogeneous presentations of the diseases and often differ in immunohistochemical markers. Hence, the exact prognosis is hard to estimate [7].

However, all the available case reports indicate the best therapeutic option remains complete surgical excision with extended pelvic lymphadenectomy [8, 9].

Due to limited data, bladder LMS's 5-year overall survival (OS) rate is difficult to estimate. In the case of uterine LMS, the OS is over 50% when the diagnosis is made at an early stage. However, in FIGO stages III and IV with generated resistance to treatment, the survival rate decreases and ranges between 35% and 29% [6].

Due to the rare occurrence of this disease, there are several difficulties in determining its etiology and distinguishing a strictly defined therapeutic protocol [7].

Due to the significant malignancy of LMS, surgical treatment was preferred over systemic therapy. Surgery is the standard of treatment in patients with LMS of a known location. The method of choice is complete, radical surgical excision of the urinary bladder with extended pelvic lymphadenectomy. In the case of R1 or R2 margin invasion, reoperation is recommended in highly experienced tertiary referral centers [8, 9].

Systemic therapy and radiotherapy have been most often described based on data on uterine LMS. Adjuvant systemic therapy is only used in clinical trials. Based on historical data, it can be concluded

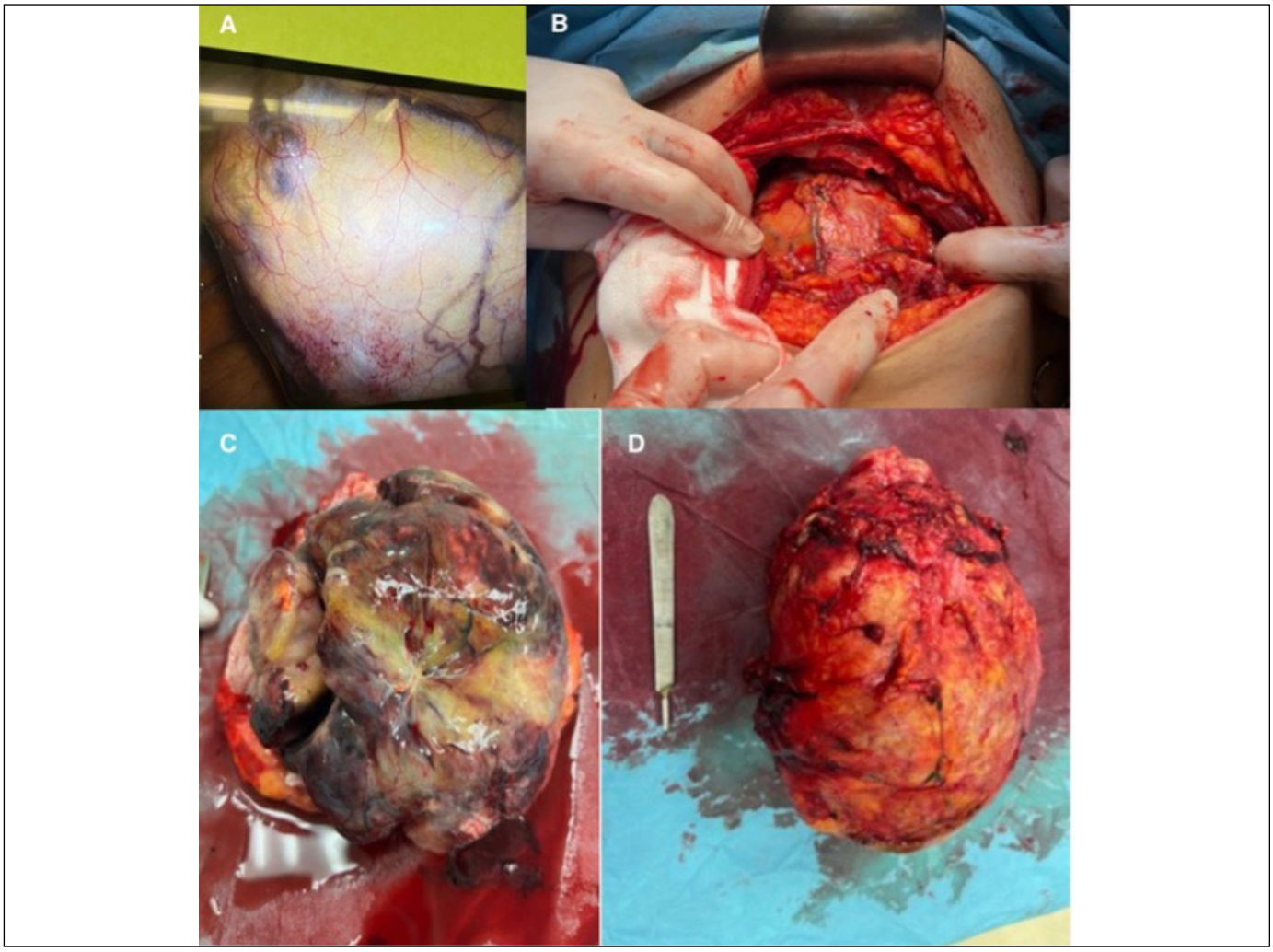

Figure 3. A) *Intraoperative, laparoscopic view of the bladder dome with widened, thrombotic vessels.* **B)** *Intraoperative view of the bladder before organ excision.* **C)** *Bladder tumor after opening the bladder.* **D)** *Bladder size in comparison to the scalpel handle.*

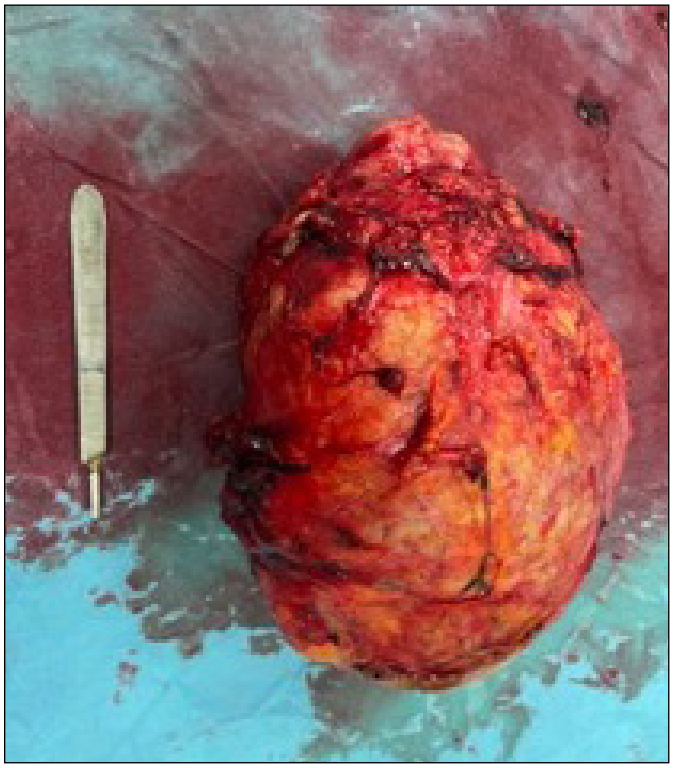

Figure 4. *The removed organ, the urinary bladder, contained the tumor. For comparison, the 13 cm scalpel handle. The size of the bladder was 20 × 16 × 5 cm. The lumen was filled with tumor mass (15 × 17 × 8 cm).*

that the use of combined treatment with docetaxel/gemcitabine followed by doxorubicin increases survival in LMS limited to the uterus alone compared to monotherapy or without chemotherapy. There are reasons to claim that the use of radiochemotherapy also increases 3-year progression-free survival while increasing the toxicity of the therapy [10].

In our case, discrepancies in the tumor size cannot be unequivocally ascribed to rapid tumor growth since the radiologist's CT description clearly states that the measurement is only an approximation due to poor contrast secretion. Moreover, abdominal CT and ultrasound are different imaging modalities.

Adjuvant treatment remains a subject of debate due to the high rate of relapse and progression despite the therapy used and the side effects resulting from the treatment. There are also opinions recommending active surveillance after complete resection without adjuvant therapy due to questionable reports about its effectiveness [11].

Epidemiological data are also conflicting.

Some sources claim that the average age of patients diagnosed with the tumor is approximately 65 years, and there is no clear predominance of the incidence of urinary bladder leiomyosarcoma in relation to gender [12].

Other data show that this cancer occurs with an increased frequency in men, and the median age is approximately 52 years. Probably the most common symptoms reported by patients with this cancer are mainly painless hematuria. Less common symptoms include painful urination, symptoms of constipation, nocturia, increased frequency of urination with abdominal and pelvic pain, etc. [13].

In short-term follow-up, radical cystectomy with extended pelvic lymphadenectomy has proven effective treatment for Leiomyosarcoma of the urinary bladder. Due to conflicting data, systemic therapy, especially without surgical excision, should not be attempted. Strict follow-up is necessary. Minimally invasive techniques, such as laparoscopy and robot-assisted laparoscopic surgery, should be considered when applicable.

CONFLICT OF INTERESTS

The authors declare no conflict of interest.

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ETHICS APPROVAL STATEMENT

The ethical approval was not required.

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