CASE REPORT

Primary leiomyoma of the bladder radiologically mimicking a retroperitoneal tumor - a case report

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Abstract: The case presented is of a 47-year-old patient with an extravesical pedunculated bladder leiomyoma, which was difficult to distinguish from a retroperitoneal tumor. Preoperatively, it was suspected to be a retroperitoneal tumor and a laparotomy with tumor resection was performed. Intraoperatively, the bladder and tumor were connected by a cord-like tissue. A retrospective review of preoperative images revealed that cord-like tissue, identified intraoperatively, was also present. Bladder leiomyomas can grow as extravesical pedunculated tumors. Therefore, when the continuity between the bladder and tumor is only a cord-like object, the finding of continuity is useful to diagnose with bladder leiomyoma. J. Med. Invest. 70: 513-515, August, 2023

Keywords: Primary leiomyoma, bladder tumor, retroperitoneal tumor

CASE RRESENTATION

A 47-year-old man (height, 177 cm; weight, 82 kg; body mass index, $26.2~{\rm kg/m^2}$) presented with hypertension, tachycardia, and hyperglycemia. An abdominal computerized tomography (CT) for investigating the cause of hypertension revealed an 87 mm solid mass located in the anterior part of the bladder. The patient's blood pressure was $232/129~{\rm mmHg}$, heart rate was 115 bpm and the initial hemoglobin A1c was 9.6%. Levels of serum catecholamines, dehydroepiandrosterone and tumor markers, such as carcinoembryonic antigen, α -fetoprotein, neuron-specific enolase and urinary metanephrine fractions were normal. Urinalysis showed no hematuria, and urine cytology was negative. (123) I-metaiodobenzylguanidine scintigraphy showed no uptake. Therefore, the diagnosis was essential hypertension, type 2 diabetes, and non-functioning tumor.

The tumor was a circumscribed round mass, heterogeneously enhanced in the late phase on contrast-enhanced CT (CECT). There was a poorly enhanced area on the left side of the tumor (Figure 1A). Continuity between the right lateral wall of the bladder and the tumor was suspected, but not definitively determined (Figure 1A). Magnetic resonance imaging (MRI) T1-weighted images (T1WI) showed that the right side of the tumor had low-intensity areas like muscle tissue and the left side of the tumor had a heterogeneous moderate-intensity area (Figure 1B). On MRI T2-weighted images (T2WI), the tumor was clearly demarcated from the surrounding tissue, and the right side of the tumor had remarkably low-intensity areas comparable to muscle tissue (Figure 1C). The left side of the tumor contained mixed low- and high-intensity areas (Figure 1D). T2WI also showed suspected continuity, albeit slightly indistinct, between the tumor and the right lateral bladder wall (Figure 1C).

Since a primary retroperitoneal sarcoma could not be ruled out, a laparotomy with tumor resection using a lower abdominal

Received for publication May 16, 2023; accepted July 31, 2023.

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midline incision was performed. The tumor was well demarcated from the surrounding tissue and could be bluntly dissected. Post dissection, a firm cord-like tissue from the right lateral wall of the bladder to the tumor was observed. Intraoperatively, the tumor was suggested to be benign based on demarcated margins with a clear capsule, no tumor invasion into the surrounding tissues, and well-defined morphology. The bladder was intact, except for the cord-like tissue contiguous with the tumor. A partial cystectomy of approximately 10 mm square was performed with resection of the cord-like tissue and tumor. The operative time was 71 min, and 10 ml blood was lost. The specimen weighed

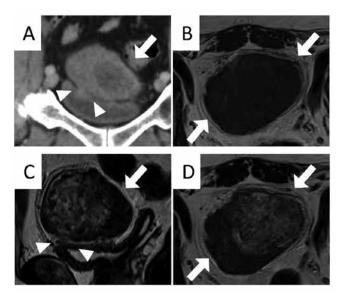


Figure 1. Findings of the tumor on computerized tomography (CT) and magnetic resonance imaging (MRI). White arrows indicate tumors. White arrowheads show continuity between the tumor and the bladder.

A. coronal late phase of contrast-enhanced CT

B. axial T1 weighted image of MRI

C. sagittal T2 weighted image of MRI

D. axial T2 weighted image of MRI

112 g. The cord-like object was contiguous to the tumor (Figure 2A, B). The cut surface was light brown, and some areas were nodular and yellow in color (Figure 2B).

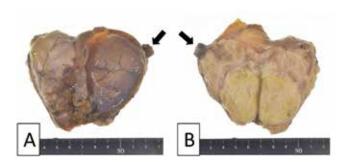


Figure 2. Specimen of the tumor Black arrows show the cord between the tumor and the bladder. A. Outside the tumor B. Inside the tumor

Histopathological examination revealed a well-demarcated (Figure 3A), solid tumor with bundles of spindle-shaped cells and an eosinophilic cytoplasm intersecting in a stereotypical manner. Overall, no nuclear atypia and mitotic figures were observed (Figure 3B). The tumor was positive for alpha-smooth muscle actin (Figure 3C). The left side of the tumor exhibited edematous and hyaline degeneration. Microscopically, histology of the cord-like object was identified as the bladder muscularis propria (Figure 3D). Based on these findings, the tumor was pathologically diagnosed as a primary leiomyoma of the bladder. One year after surgery, no recurrence was observed.

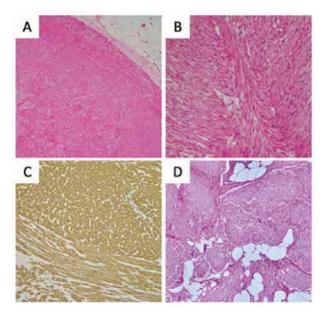


Figure 3. Pathological findings of the tumor

A. Original magnification, ×40; hematoxylin and eosin staining. A well-defined, substantial tumor with well-defined boundaries with the surrounding tissue.

- B. Original magnification, ×100; hematoxylin and eosin staining. Spindle cells with eosinophilic cytoplasm form bundles and intersect three-dimensionally. Overall, nuclear atypia was lacking, and mitotic figures were not observed.
- C. Original magnification, $\times 100$; α -SMA $\times 100$. The tumor was α -SMA positive.
- D. Original magnification, ×100; hematoxylin and eosin staining. The Bladder muscularis propria was contiguous with the tumor.

DISCUSSION

Bladder leiomyoma is a rare disease, accounting for approximately 0.43% of all bladder tumors (1). Primary bladder leiomyomas are classified into intravesical, intramural, and extravesical, with the last accounting for 30% (2). In all three types, bladder leiomyomas are typically continuous with the bladder broadly. This is the first report of an extravesical pedunculated bladder leiomyoma with poor continuity with the bladder that was difficult to diagnose preoperatively.

CT and MRI are generally useful in the diagnosis of bladder leiomyoma; on CT, a round-like, hypodense mass with a circumscribed margin was observed (3, 4). On CECT, tumors show variable enhancement patterns upon contrast enhancement (4). Smooth-margined, low-signal tumors are typical on MRI, characterized by moderate signal on T1W1 and low signals on T2WI (3-5). Furthermore, bladder leiomyomas typically involve extensive lesions in the bladder (3-5). However, in the present case, there was poor continuity between the bladder and the tumor, which is atypical for bladder leiomyoma (Figure 1C). Therefore, performing preoperative imaging was challenging. A retrospective review of the preoperative images revealed that the cord-like object had the same intensity as the bladder muscle layer on MRI T2WI. The cord-like object was pathologically the muscularis propria of the bladder (Figure 3D). Therefore, it is possible that the tumor originated from the outermost layer of the muscularis propria of the bladder and grew outward, resulting in a pedunculated tumor.

Laparotomy was selected based on the possibility that tumor resection with wide margins might be required. Intraoperatively, the tumor was diagnosed as a primary benign bladder tumor and a resection with minimal partial cystectomy was performed. The standard treatment for bladder leiomyomas is transurethral resection or partial cystectomy, depending on the tumor location and size. Recently, several cases of benign bladder tumors treated with laparoscopic or robot-assisted laparoscopic partial cystectomy have been reported (6-8). In the present case, laparoscopic surgery would have been the treatment option if bladder leiomyoma had been preoperatively diagnosed.

In conclusion, preoperative imaging findings of continuity between the bladder and tumor are useful for the diagnosis of bladder leiomyoma.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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