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CASE REPORT

Bladder Leiomyoma Presenting with Recurrent Cystitis: A Case Report

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Background: Non-epithelial bladder neoplasms are rare, and there are only approximately 250 cases of bladder leiomyoma reported in the English literature. We present a case of bladder leiomyoma in a patient with recurrent acute cystitis.

Case Presentation: A 53-year-old woman presented to a local clinic with frequent urination and dysuria and was diagnosed with acute cystitis. Her symptoms temporarily improved after taking oral antibiotics, but then flared up repeatedly. Contrast-enhanced computed tomography showed a bladder neck leiomyoma and multiple uterine fibroids. Her body mass index was 27.0 kg/m^2. The patient successfully underwent transurethral resection of the tumor. The histopathological diagnosis was bladder leiomyoma.

Conclusion: There have been few cases of bladder leiomyoma coexisting with urinary tract infection and uterine fibroids. Our findings suggest that female hormones, which might increase body weight and cause fatty liver, are associated with the growth of bladder leiomyomas. If a patient has recurrent urinary tract infection, a bladder leiomyoma should be included in the differential diagnoses.

Keywords: bladder, leiomyoma, female hormone, uterine fibroid, body mass index

Background

Most primary bladder neoplasms are epithelial in origin. The most common bladder neoplasm is urothelial carcinoma, while non-epithelial bladder neoplasms are rare.[1–3] Leiomyomas are found in all anatomical structures and are most common in the uterus but are rarely found in the bladder.[3] Bladder leiomyoma is a benign smooth muscle neoplasm that arises from connective tissue and is the most frequent type of benign neoplasm.[1–3] Bladder leiomyoma accounts for only about 0.3% of all bladder neoplasms, with approximately 250 cases reported in the English literature worldwide.[1,2] Bladder leiomyoma is detected more frequently in women than in men.[3] Anatomically, bladder leiomyomas are classified as endovesical, intramural, or extravesical, with endovesical being the most common.[1] The symptoms of bladder leiomyoma vary by anatomic location and size. The most common chief complaints of bladder leiomyomas are obstructive urinary symptoms.[3]

In this report, we describe a case of bladder leiomyoma in a 53-year-old Japanese woman and review the relevant literature.

Case Presentation

A 53-year-old woman presented to a local clinic with the symptoms of frequent urination and dysuria. She was diagnosed with acute cystitis and prescribed antibiotics. Her symptoms temporarily improved after taking the oral antibiotics but then flared up repeatedly. Therefore, an abdominal ultrasound was performed in the clinic and showed a smooth-surfaced hypoechoic neoplasm of 26×27 × 29 mm in the urinary bladder (Figure 1). She had a small amount of residual urine, but no urinary retention. She was then referred to the urology department in our hospital.

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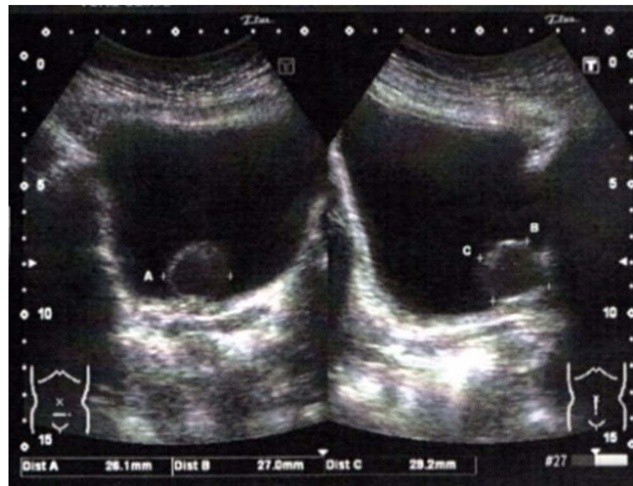

Figure 1 Abdominal ultrasound shows a neoplasm in the urinary bladder.

On presentation, the patient was 161 cm tall, weighed 70 kg, and had a body mass index (BMI) of 27.0 kg/m^2. There was no remarkable medical history or family history. There were no abnormalities on physical examination. A urine analysis was normal (red blood cells: 1–4/high-power field; white blood cells: 1–4/high-power field; and no protein and no glucose). Urine cytology was class I. Contrast-enhanced computed tomography (CT) showed a bladder neck neoplasm with the same contrast effect as the bladder wall, multiple uterine fibroids (leiomyomas), and a fatty liver (Figure 2a–e).

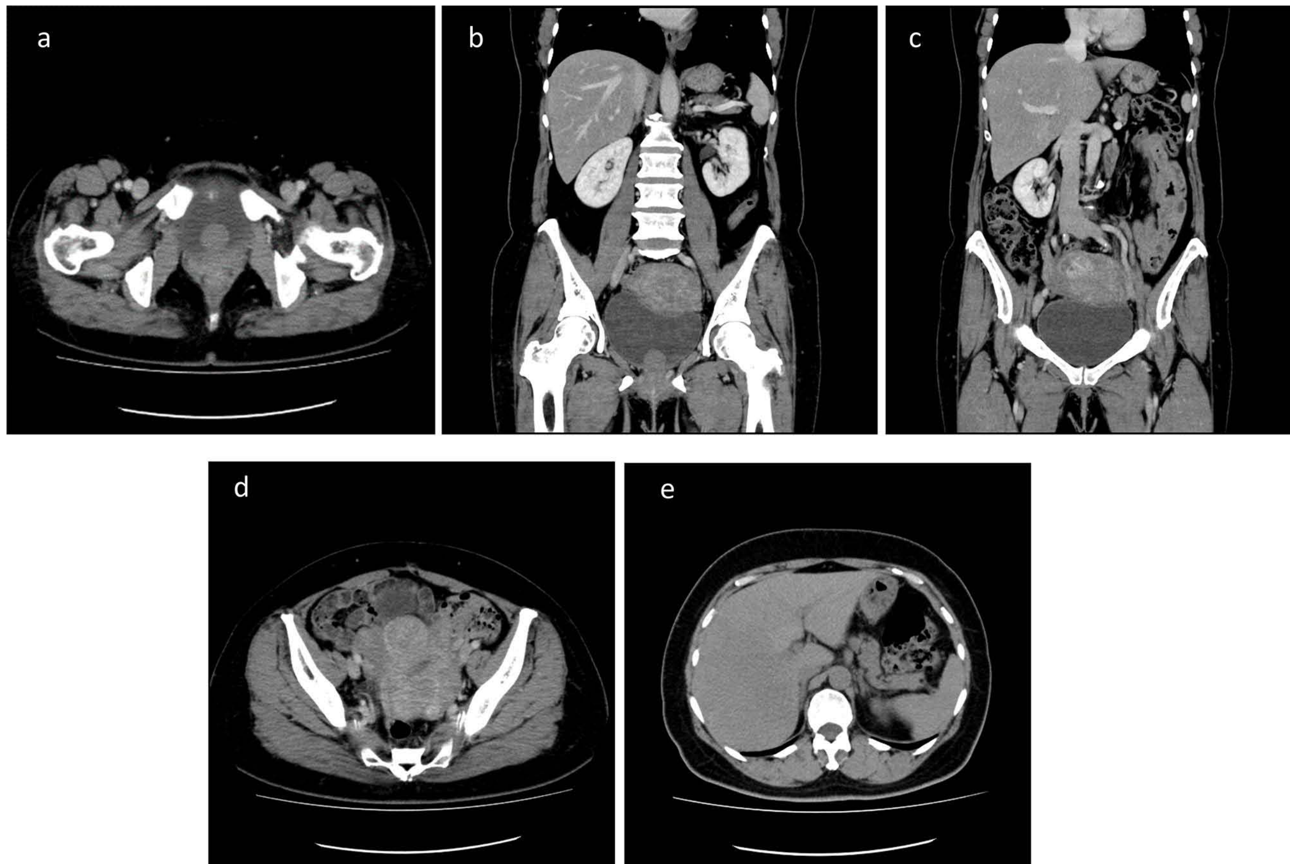

Figure 2 CT images. Contrast-enhanced CT shows axial (**a**) and coronal (**b**) views of a smooth-surfaced mass in the urinary bladder. Contrast-enhanced CT shows axial (**c**) and coronal (**d**) views of multiple uterine fibroids. (**e**) CT axial view of a fatty liver.

Cystoscopy was then performed and revealed a round neoplasm in the bladder neck. The tumor had a smooth surface covered by normal mucosa similar to the bladder mucosa, and was blocking the urethral opening (Figure 3a and b). The patient was diagnosed with bladder leiomyoma and underwent transurethral resection of the bladder tumor with a margin of several millimeters extending to the depth of the muscle layer. Her postoperative course was good, and she was discharged 4 days after surgery. Her symptoms improved after surgery.

Histopathological examination with hematoxylin and eosin staining revealed a proliferation of spindle-shaped cells with fibers and an eosinophilic cytoplasm (Figure 4a). Immunostaining was positive for desmin and the myogenic marker alpha-smooth muscle actin (Figure 4b and c), and negative for S-100 and CD10. Ki-67 was present in only 1%–5% of neoplasm cells, and this was considered negative (Figure 4d). The histopathological diagnosis was bladder leiomyoma.

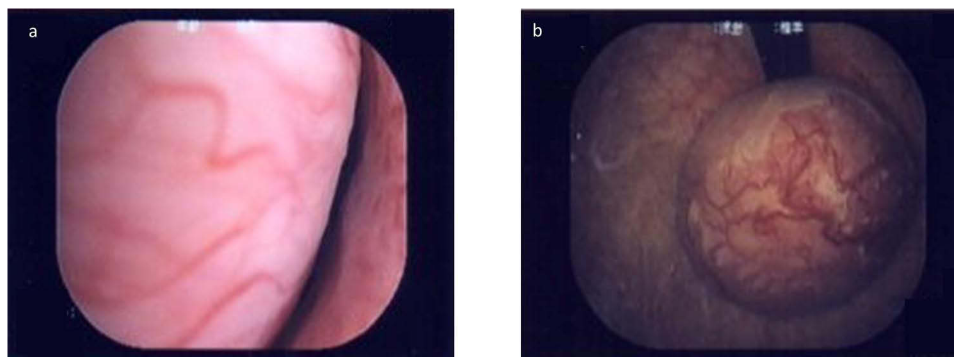

Figure 3 Cystoscopy shows a bladder leiomyoma that protruded from the internal urethral opening at the 9 o'clock position (**a**) and was turned around to observe the internal urethral opening from inside the bladder (**b**).

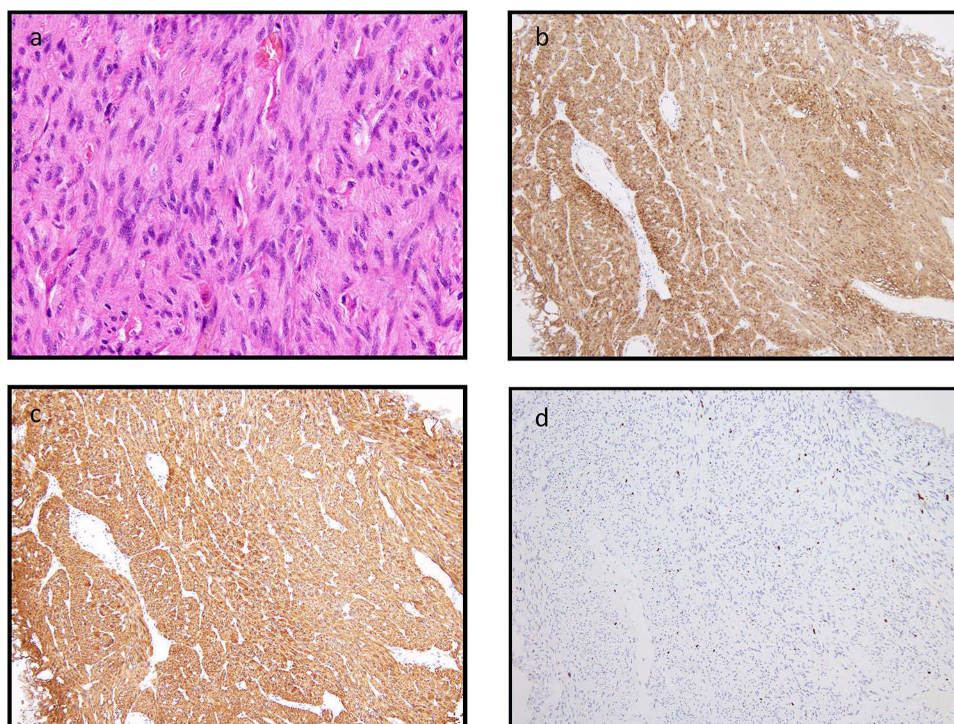

Figure 4 Pathological findings of the neoplasm. (**a**) Histopathological examination shows the proliferation of spindle-shaped cells with fibers and eosinophilic cytoplasm under low magnification (hematoxylin–eosin staining). (**b**) Immunohistochemical staining of desmin is positive under low magnification. (**c**) Immunohistochemical staining of alpha-smooth muscle actin is positive under low magnification. (**d**) Immunohistochemical staining of Ki-67 is negative under low magnification.

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Approximately 1 year after transurethral resection of the bladder tumor, no recurrence was observed. We considered that another follow-up should be performed, but the patient did not visit our hospital again.

Discussion and Conclusions

Most primary urinary bladder neoplasms are epithelial bladder neoplasms. However, bladder leiomyomas are rare epithelial bladder neoplasms, accounting for only 0.3% of all urinary bladder neoplasms.[2] Leiomyomas are benign smooth muscle neoplasms that are composed of connective tissue and are found in all anatomical structures.[1–3] Leiomyomas are most commonly found in the uterus but are rarely found in the bladder.[3]

Only approximately 250 cases of bladder leiomyoma have been reported in the English literature worldwide in the last 90 years.[1] Bladder leiomyoma was first reported in the English literature in 1931,[3] and was first reported in the Japanese literature in 1916.[4] Sugimoto et al reported the 153rd case of bladder leiomyoma in Japan.[4] Taniuchi et al reported the 169th case of bladder leiomyoma in Japan, although they did not mention this condition in their article.[5] To the best of our knowledge, our case is the 170th reported case of bladder leiomyoma in Japan.

The incidence of bladder leiomyoma in women is approximately three times as high as that in men.[6] The median age at diagnosis of bladder leiomyoma is 45.3 years (range: 19–85 years).[6] Bladder leiomyoma tends to develop in women at 30–40 years of age, and in men at 50–60 years of age.[7] The age of predilection for uterine fibroids and bladder leiomyomas is similar, as the most common age of onset of uterine fibroids is 30–40 years of age.[8]

While bladder leiomyoma is a rare disease worldwide, it appears to be more common in Japanese people. However, no study has clarified why bladder leiomyoma is more common in the Japanese population. Risk factors for uterine fibroids include age, race, genetics, reproductive factors, hormonal factors, endocrine factors, a high BMI, lifestyle, and diet.[9] In women, the risk factors for uterine fibroids may overlap with those for bladder leiomyomas because bladder leiomyomas are pathologically similar to uterine fibroids, and these conditions occur at approximately the same age. Men have an older age of predilection for bladder leiomyomas than women and have low blood concentrations of female hormones, suggesting that the cause of bladder leiomyomas might be different in men and women. Further studies are required to clarify the specific mechanism of bladder leiomyomas.[10]

Little is known about the etiology of benign neoplasms. However, Teran and Gambrell suggested the following four theories: dysontogenesis, perivascular inflammation, bladder infection, and hormonal influence.[11] The association between bladder leiomyomas and uterine fibroids and the fact that the age of onset of bladder leiomyomas coincides with that of uterine fibroids suggest the involvement of female hormones as a possible mechanism of pathogenesis. Receptors of estradiol and progesterone have been identified in bladder leiomyomas.[12] Therefore, these ovarian steroid hormones and their receptor formation may be involved in the growth of bladder leiomyomas.

In the present case, the patient had multiple uterine fibroids and a bladder leiomyoma. She also had a high BMI and a fatty liver on CT. These findings suggest a potential hormonal and metabolic basis for neoplasm development. Bladder leiomyomas and uterine fibroids share histological features, and both may express estrogen and progesterone receptors.[12] The presence of excess adipose tissue increases the activity of aromatase, an enzyme that exists in adipose tissue and converts androgens into estrogens.[13] Additionally, fatty liver decreases the production of sex hormone-binding globulin (SHBG), which binds estrogen and limits its biological activity. Therefore, a decreased SHBG concentration results in a higher concentration of free estrogen. These changes may create a hormonal environment conducive to the growth of estrogen-sensitive smooth muscle neoplasms.

Fatty liver and high BMI are also associated with hyperinsulinemia and insulin resistance, which may increase the circulating concentrations of ovarian hormones and promote the proliferation of myometrial smooth muscle cells.[9] While bladder leiomyoma is a rare condition, the overlapping risk factors and hormonal mechanisms between bladder leiomyomas and uterine fibroids may provide clues about the etiological pathogenesis of bladder leiomyoma in women.

Bladder leiomyomas are classified into three pathogenic types: endovesical (63%), intramural (7%), and extravesical (30%).[10] Generally, obstructive urinary symptoms and irritative urinary symptoms tend to appear after the bladder leiomyoma has increased in size and is located endovesically. The main complaints of bladder leiomyomas are obstructive urinary symptoms (49%), irritative urinary symptoms (38%), and hematuria (11%), while approximately 20% of patients are asymptomatic.[10] As more people have health screening performed worldwide, the proportion of

patients with asymptomatic bladder leiomyoma will increase. In the present case, a bladder leiomyoma was discovered following repeated urinary tract infections. Haddad et al reported a case of bladder leiomyoma presenting with a febrile urinary tract infection.[14] Therefore, bladder leiomyomas should also be considered in patients with urinary tract infection.

Pathologically, bladder leiomyomas resemble uterine fibroids and consist of fascicles of smooth muscle cells and an eosinophilic cytoplasm.[10] Smooth muscle cell nuclei are centrally located, oval, or cigar-shaped.[10] These findings suggest the involvement of female hormones in the pathogenesis of bladder leiomyoma.

Ultrasound, magnetic resonance imaging (MRI), CT, and cystoscopy play an important role in the diagnosis of a bladder leiomyoma. Ultrasound is the most sensitive technique for diagnosing bladder leiomyomas and can show a smooth, homogeneous, hypoechoic mass. CT and MRI are used to evaluate the tumor location and its relationship with adjacent structures, but MRI is superior because it can distinguish the boundary and detect the origin of the neoplasm.[15] Cystoscopy can reveal the size and anatomic location of a bladder neoplasm. However, clinicians must be aware that bladder leiomyomas are a type of submucosal tumor that may not be detected by cystoscopy. Bladder leiomyoma may instead be diagnosed incidentally on ultrasound, CT, or MRI, but these imaging studies cannot be used to definitively diagnose bladder leiomyoma. The diagnosis of bladder leiomyoma must be based on pathological examination.

The treatment for bladder leiomyoma is surgical resection. In the past, open surgery was performed in many cases. However, minimally invasive surgeries are increasingly being used. Transurethral resection of bladder tumors is commonly performed, while laparoscopic surgery or robot-assisted surgery is considered for large bladder neoplasms.[10,16,17]

There have been no reports of malignant transformation of bladder leiomyomas. The prognosis of urinary bladder leiomyomas is generally favorable, although there have been a few cases of postoperative recurrence.[4,12,18] Careful long-term follow-up is required after surgery.

In conclusion, bladder leiomyomas are clinically rare worldwide but might be more common in the Japanese population. The findings in our case of bladder leiomyoma suggest that estrogen and/or progesterone are associated with the growth of this neoplasm. Future case reports or studies of bladder leiomyoma should include the insulin and estrogen concentrations. If a patient has recurrent urinary tract infections, bladder leiomyomas should be included in the differential diagnoses.

Abbreviations

BMI, body mass index; CT, computed tomography; MRI, magnetic resonance imaging; SHBG, sex hormone-binding globulin.

Data Sharing Statement

The datasets used during the current case are available from the corresponding author upon reasonable request.

Ethics Approval and Consent to Participate

Institutional approval was not required for the publication of these case details. Written informed consent was obtained from the patient.

Consent for Publication

Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors declare that they have no competing interests in this work.

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