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Association between cystitis glandularis and bladder neck leiomyoma: A case report and literature review

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Abstract

This article reported the diagnosis and treatment of cystitis glandularis (CG) co-occurring with bladder neck leiomyomas. We retrospectively analyzed the clinical data of a single case of CG with bladder neck leiomyoma. A 31-year-old Chinese woman was given a diagnosis of CG and bladder neck leiomyoma. The mass and surrounding bladder mucosal lesions were entirely excised via transurethral resection, leaving a clean margin of healthy tissue. Histopathological analyses confirmed the diagnosis of CG and bladder neck leiomyoma. The patient remained asymptomatic throughout the follow-up period, with no indication of recurrence. Cystitis glandularis co-occurring with bladder neck leiomyoma requires careful examination, and surgery remains the best treatment option for these diseases.

Keywords: Cystitis glandularis; Bladder neck leiomyoma; Diagnosis; Treatment

1. Introduction

Leiomyomas, which originate from the smooth muscle, are benign tumors that predominantly affect the uterus in women of reproductive age. [1,2] Its occurrence in the bladder is exceedingly rare, accounting for only 0.43% of all bladder neoplasms. [3] Bladder leiomyomas typically present with distinct clinical symptoms such as urinary tract infections, masses, painful sexual intercourse, urinary retention, and irritating lower urinary tract symptoms. [4]

In contrast, cystitis glandularis (CG)—a proliferative and metaplastic disorder affecting the bladder mucosa—has an incidence of 0.1%–1.9% in the general population. ^[5] Cystoscopic examination often reveals irregular nodular lesions that may mimic tumors. Notably, CG is linked to the development and progression of bladder cancer, ^[6] and its incidence has shown an upward trend in recent years. ^[7] The clinical manifestations of CG include frequent urination, urgency, pain, difficulty in urinating, blood in the urine, discomfort in the lower abdomen and pelvic cavity, and mucus discharge in some patients. ^[8]

A 31-year-old woman presented with a 6-month history of urinary symptoms, including dysuria, hesitancy, intermittency, and persistent bladder irritation. Before visiting our facility, she had undergone 2 transurethral resections for CG at another hospital. Pathological examination after those procedures confirmed the diagnosis of CG. After each surgery, the patient experienced temporary relief from her urinary symptoms; however, the symptoms would soon

Although cases of bladder neck leiomyomas and CG have been documented separately, their co-occurrence has never been

reported before. In this report, we present the unique case of a

31-year-old Chinese woman with a diagnosis of both CG and blad-

der neck leiomyomas. This report can contribute to the existing

medical literature and enhance our understanding of these rare con-

ditions, thereby guiding future clinical practice and research efforts.

2. Case presentation

recur and cause significant discomfort.

Physical examination revealed no remarkable findings. However, during the uroflowmetry test, the patient exhibited a plateau pattern with a peak urinary flow rate of 17.6 mL/s and an average flow rate of 6 mL/s, indicating some degree of urinary obstruction. Pyuria on her urinalysis results and *Escherichia coli* on the bacterial culture of her urine suggest the presence of urinary tract infection. Her serum creatinine was 76 µmol/L, and her hemoglobin was 125 g/L.

Urinary ultrasonography showed residual urine in the bladder, measuring 93 mL after micturition, providing further evidence of urinary dysfunction. In addition, a T2-weighted magnetic resonance imaging scan was conducted, revealing a smooth-edged nodule measuring $25 \times 20 \times 27$ mm at the bladder neck. The nodule appeared slightly enlarged and showed clear and uniform enhancement on the enhanced scan. The mucosal layer of the bladder wall in the trigone

Caixia Zhang and Longlong Fan have contributed equally to this work and share the first authorship.

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appeared slightly thickened and uneven in thickness. An enhanced scan also showed mild enhancement in this area (Fig. 1).

Cystoscopic examination revealed a solitary mass with surrounding bladder mucosal lesions, measuring 30×25 mm, attached to the bladder neck (Fig. 2). The bladder wall surrounding the mass showed follicular changes in mucosal particles.

Given these findings, we decided to completely resect the mass with the surrounding bladder mucosal lesions via a transurethral approach.

Surgical Steps:

- (1) Anesthesia and positioning: After successful anesthesia, the patient was placed in the lithotomy position. Routine sterilization and draping of the perineal surgical field were performed.
- (2) Cystoscopy using a resectoscope: The resectoscope sheath and urethra were adequately lubricated with paraffin oil. The resectoscope was then inserted into the bladder, revealing turbid urine. A mass measuring approximately 30 × 25 mm was visible in the bladder neck. The surface of the mucosa was smooth. Mucosal follicular changes on the bladder wall surrounding the mass and trabeculation of the bladder were noted. Evidence of previous surgical scarring was observed on the bladder trigone. Both ureteral orifices were visible with clear urine efflux, and no other significant abnormalities were observed.
- (3) Lesion resection: After connecting the electrosurgical unit, the mass and bladder mucosal lesions were resected, extending to the upper segment of the urethral sphincter and deep into the muscular layer. The resected tissue was collected using an Ellik evacuator and sent for histopathological examination.
- (4) Hemostasis of the surgical site: We inspected any bleeding points in the bladder and applied electrocoagulation for hemostasis. After careful examination confirmed no bleeding or damage to the bi-

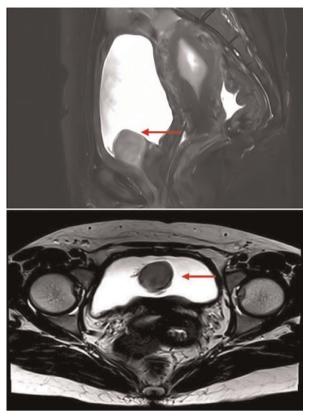


Figure 1. T2 pelvic MRI showing a mass attached to the bladder neck (arrow). MRI = magnetic resonance imaging.

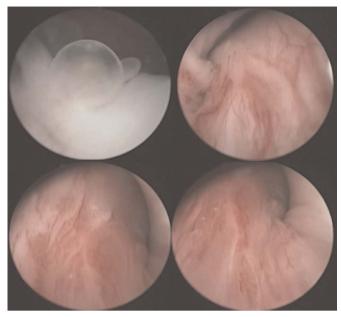


Figure 2. Cystoscopic examination revealing follicular granules of bladder neck mucosa and a single 3.2×2.5 -cm mass attached to the 6-o'clock position on the posterior wall of the bladder neck.

lateral ureteral orifices, the resectoscope was withdrawn. An F22 three-way Foley catheter was inserted, and continuous bladder irrigation with normal saline was initiated. The surgery was concluded, and the patient was transferred to the ward.

The resected tissues were sent to the pathology department for hematoxylin and eosin staining and immunohistochemical staining using the leiomyoma markers, actin and desmin. The patient's recovery is currently being closely monitored to ensure complete resolution of her symptoms.

Pathological examination of the resected mass revealed characteristic findings of leiomyoma and CG (Fig. 3), confirming the diagnosis. This benign smooth muscle tumor is a common type of uterine fibroid; however, its presence in the bladder is less common. The identification of leiomyoma provided valuable insight into the patient's clinical presentation and explained the urinary symptoms she had been experiencing.

Six months after the surgical resection, uroflowmetry performed to assess the patient's urinary function demonstrated a significant improvement in urinary flow rates. Specifically, she achieved a maximum urinary flow rate of 26.9 mL/s and an average flow rate of 20.5 mL/s, indicating a substantial improvement in her urinary tract obstruction.

The patient remained asymptomatic for over 6 months after the surgery, with no recurrence of her previous urinary symptoms. Magnetic resonance imaging at the 6-month follow-up revealed a nearly normal bladder structure (Fig. 4), indicating the successful resolution of the leiomyoma and restoration of bladder function.

The patient's positive outcome highlighted the importance of an accurate diagnosis and prompt surgical intervention in managing leiomyoma-related urinary tract obstruction.

3. Discussion

Buttner first documented a case of leiomyoma in 1894, significantly contributing to medical literature. This tumor, although

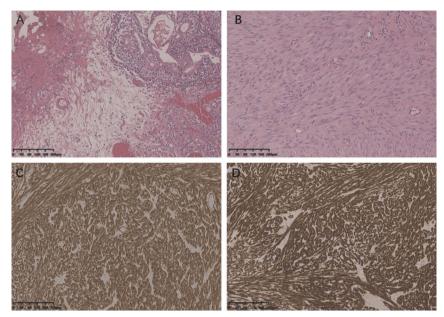


Figure 3. Bladder neck leiomyoma and cystitis glandularis (H-E, original magnification ×100). (A) Cystitis glandularis. (B) Leiomyoma. (C and D) Leiomyoma marked by actin and desmin antibodies, respectively. H-E = hematoxylin and eosin staining.

most commonly observed in women of reproductive age, can affect individuals of any age or sex. [9] Leiomyomas are relatively prevalent in specific parts of the body, particularly in the genitourinary system and gastrointestinal tract, and less frequently occur in the skin; they are even rarer in deep tissues. Overall, the incidence of soft tissue leiomyomas is relatively low.

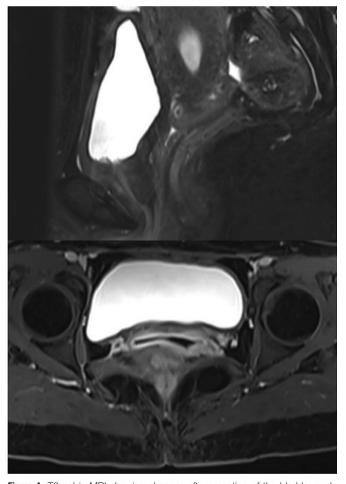
Although bladder leiomyoma is an extremely rare type of tumor, it represents a common histological type within the category of benign bladder tumors.^[10] Bladder leiomyoma originates from smooth muscle tissue, and although its exact etiology remains unknown,^[11] factors such as hormonal imbalances, chromosomal abnormalities, repeated infections in the bladder region, inflammation around blood vessels, and dysplasia may contribute to its development.^[12,13]

In patients with bladder leiomyoma, voiding symptoms are the most common, occurring in 50% of the cases. This was followed by storage symptoms, which occur in 25% of the cases, urinary tract infections in 14%, and hematuria in 11%. [14] Furthermore, the severity of symptoms may be related to the size and location of the tumor; for instance, lesions located closer to the bladder neck may elicit more symptoms. [15] Thus, when assessing and managing patients with bladder leiomyomas, it is necessary to consider the size and location of the tumor to comprehensively understand their symptoms and formulate an appropriate treatment plan.

Management strategies for bladder leiomyoma include endoscopic or ultrasonographic surveillance, transurethral resection, and partial or total cystectomy. These methods are crucial for the diagnosis and treatment of bladder leiomyomas and are applicable to patients with different conditions and symptoms. [16]

Currently, the best treatment option for bladder leiomyomas is local surgical resection, which involves removal of the tumor and surrounding tissue to ensure complete excision and minimize the risk of recurrence. Although other treatment options may be considered in certain cases, surgical resection remains the gold standard for managing this rare but potentially debilitating condition.

Cystitis glandularis, a proliferative disorder that affects the bladder mucosa, is characterized by the transformation of transitional



 $\begin{tabular}{ll} Figure 4. T2 pelvic MRI showing changes after resection of the bladder neck mass. MRI = magnetic resonance imaging. \\ \end{tabular}$

cells into glandular structures.^[17] It typically occurs concurrently with von Brunn nests, and both are regarded as reactive lesions.^[18] The exact etiology and pathogenesis of this condition remain controversial in the medical community.

Cystitis glandularis is commonly classified into 2 distinct subtypes: common and intestinal. [19] Although both subtypes share certain characteristics, they differ in terms of their histological features and potential clinical implications. Notably, an association between CG and bladder adenocarcinomas has been reported. However, a causal relationship between these 2 conditions has not yet been definitively established. [20]

Previous studies have reported contradictory findings. For instance, Morton et al.^[21] observed that telomerase length in intestinal CG was significantly shorter than that in adjacent normal bladder tissue and typical CG. Telomerase deletion is considered an important mechanism in the development of bladder cancer.

Cystitis glandularis can often be managed conservatively; its symptoms can be effectively managed using anti-inflammatory therapy with levofloxacin and cyclooxygenase-2 inhibitors. [19] However, severe cases may necessitate surgical interventions, with electrocautery/excision and bladder irrigation as the main surgical options.

Our team observed that obstructive lower urinary tract diseases might play a significant role in the pathogenesis of CG. [22] We hypothesized that these conditions could lead to recurrent chronic bladder inflammation, which, in turn, could induce CG development. Specifically, we hypothesized that bladder leiomyomas located in the bladder neck may cause urinary obstruction, which can lead to recurrent urinary tract infections and may ultimately be the underlying cause of CG. Although further research is required to confirm this hypothesis, it provides a potential new direction for the prevention and treatment of CG.

4. Conclusions

Cystitis glandularis combined with bladder neck leiomyoma is a rare clinical condition that necessitates more careful examination and appropriate treatment.

Acknowledgments

None

Statement of ethics

In accordance with our institutional regulations, a single-case report that excludes any identifying patient information does not require formal ethical approval or consent from the patient. All procedures involving human participants were conducted in strict adherence to the ethical standards set forth by the institutional and national research committee, as well as the 1964 Helsinki Declaration and its subsequent amendments or comparable ethical norms.

Conflict of interest statement

The authors declare no conflicts of interest.

Author contributions

CZ: Conceived the study and drafted and edited the manuscript; LF: Collected data and drafted and edited the manuscript;

KL, LH: Made substantial contributions to the editing of the manuscript;

LC: Provided invaluable pathological support;

YY: Conceived the study and approved the final version.

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Data availability

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

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