Case Report

Mullerianosis of the Urinary Bladder: A Rare Case Report

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Abstract

Endocervicosis, endometriosis, and endosalpingosis are choristomas derived from Müllerian tissue, collectively termed *Müllerianosis*. We report a 40-year-old woman with pelvic pain and dysuria who presented with a 3.2×2.5 cm bladder mass four years post-hysterectomy. Transurethral resection revealed endocervical glands and endometrial stroma deep in the bladder wall. Immunohistochemistry confirmed ER/PR positivity in glandular epithelium and CD10 positivity in stroma, establishing the diagnosis of *Müllerianosis*. Outcome: The patient remained asymptomatic with no recurrence over 8 years of clinical and cystoscopic follow-up. *Müllerianosis* is a rare benign bladder lesion that can mimic malignancy. Accurate histopathological diagnosis with immunohistochemistry is essential to avoid unnecessary radical surgery and enable hormonal therapy when indicated.

Keywords: müllerianosis; *endocervicosis*; *endometriosis*; *endosalpingosis*; case report

Introduction

Müllerianosis refers to the presence of choristomas derived from Müllerian tissue—specifically *endocervicosis*, *endometriosis*, and *endosalpingosis* occurring in extrauterine locations. When affecting the urinary bladder, this rare benign condition can closely resemble malignant tumors both clinically and radiologically, leading to potential diagnostic errors.[1, 2] Definitive diagnosis requires histopathological examination with immunohistochemical demonstration of estrogen and progesterone receptors (ER and PR) and CD10.[1, 2] Recognition of this entity is crucial as it may respond to gonadotropin-releasing hormone (GnRH) agonists, thereby avoiding unnecessary cystectomy, with surgical resection reserved for cases refractory to hormonal treatment.[3]

Case Report

A 40-year-old female patient presented with a 4-month history of pelvic pain and burning micturition (dysuria). She denied hematuria, abdominal pain, nocturia, or incontinence. Her past surgical history was significant for total abdominal

hysterectomy performed 4 years prior for symptomatic uterine fibroids. Physical examination revealed a healed lower abdominal surgical scar with an otherwise unremarkable abdomen.

Transabdominal ultrasound demonstrated a 3.2×2.5 cm soft tissue mass in the posterior bladder wall. Cystoscopy revealed an exophytic lesion on the posterior wall, and transurethral biopsy initially suggested cystitis cystica glandularis. Given diagnostic uncertainty, complete transurethral resection of bladder tumor (TURBT) was performed.

Gross Pathology: The resected specimen consisted of an irregular grey-brown mass measuring 3.6×2.8×1.0 cm. Serial sectioning revealed grey-white tissue extending deep into the muscle layer (Figure 1a).

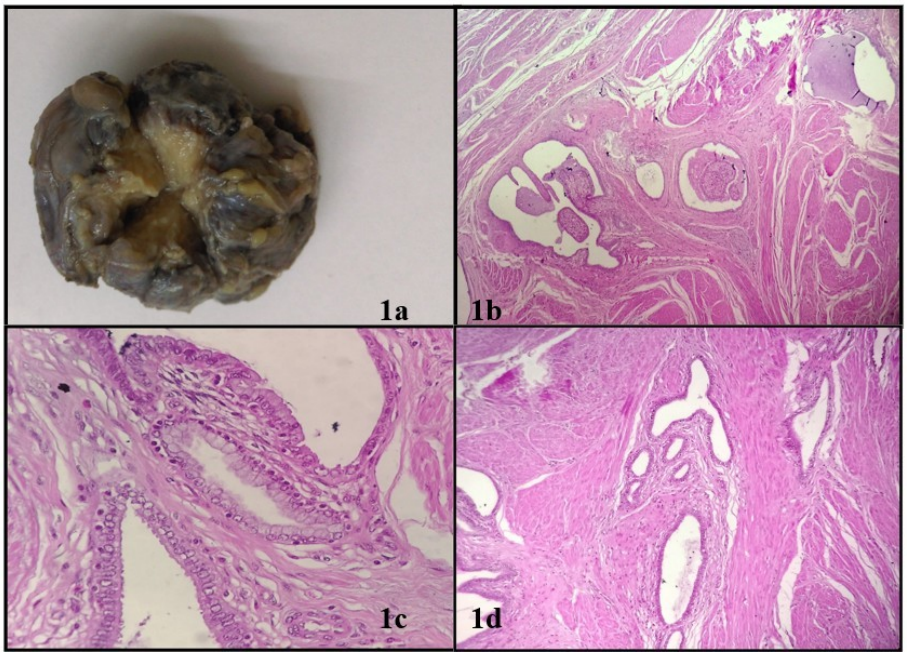

Figure 1: a. Gross examination reveals polypoidal mass-like lesion; cut surface shows grey-white mass in deep bladder wall. b. Benign endocervical glands deep in bladder wall (H&E, 20×). c. Endocervicosis in bladder wall with benign endocervical-type glands in smooth muscle (H&E, 40×). d. Endometrial glands deep in wall of urinary bladder (H&E, 20×).

Microscopic and Immunohistochemical Findings: Histological examination revealed benign endocervical-type glands invading deep into the bladder wall (Figure 1b, 1c, 1d, 2a). While endometrial-type stroma was not definitively identified on routine Hematoxylin and Eosin (H&E) staining, immunohistochemistry was performed to clarify the tissue composition.

Immunohistochemistry Methods: Immunohistochemical staining was performed on 4-μm formalin-fixed paraffin-embedded sections using the following antibodies: estrogen receptor (clone SP1, Ventana Medical Systems, prediluted), progesterone receptor (clone 1E2, Ventana Medical Systems, prediluted), and CD10 (clone 56C6, Leica Biosystems, 1:50 dilution). Appropriate positive and negative controls were run concurrently. The glandular epithelium showed strong nuclear positivity for ER and PR (Figure 2b, 2c), while the surrounding stromal cells demonstrated CD10 positivity (Figure 2d), confirming the presence of endometrial-type stroma.

Final Diagnosis: *Müllerianosis* of the urinary bladder with *endocervicosis* (endocervical-type glands) and *endometriosis* (endometrial-type stroma), fulfilling the diagnostic criterion of at least two Müllerian tissue components.

Follow-up: The patient has been followed for 8 years post-resection without recurrence. Follow-up protocol consisted of clinical review every 6 months for the first 2 years, then annually. Cystoscopic examination was performed at 6 months, 1 year, 2 years, and 5 years post-operatively, with no evidence of recurrence on visualization or biopsy. Ultrasound imaging at 2-year and 5-year intervals confirmed absence of bladder mass. The patient remains asymptomatic at the time of this report.

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

Discussion

The first reported case of *Müllerianosis* affecting the urinary bladder was described by Young and Clement in 1996.[4] Two primary theories explain its etiology: the metaplastic theory proposes hormonal-induced transformation of peritoneal mesothelium, while the implantation theory suggests direct incorporation of Müllerian tissue during pelvic surgery.[4, 5] Our patient's history of hysterectomy 4 years prior supports the implantation theory.

Diagnosis requires high clinical suspicion as the condition mimics malignancy both clinically and radiologically, with

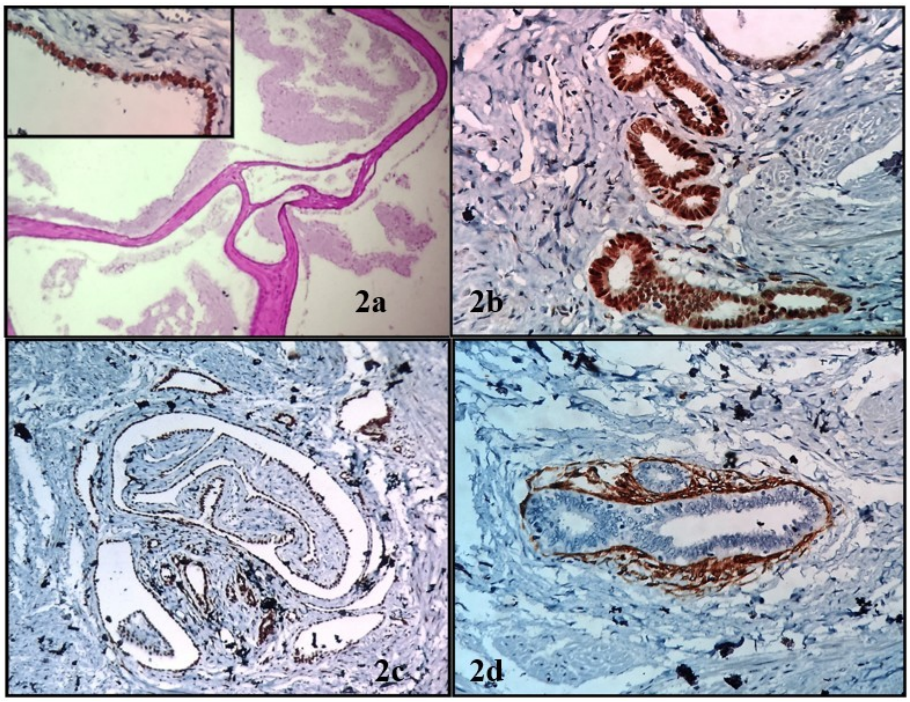

Figure 2: a. Benign cystically dilated glands (H&E, 40×). b. ER positivity in nuclei of benign endometrial glands (IHC, 40×). c. PR positivity in benign glands (IHC, 20×). d. Endometriosis in bladder wall with CD10-positive endometrial stroma (IHC, 40×).

Table 1: Differential diagnosis of *Müllerianosis*.

Entity	Location/Morphology	Immunohistochemistry	Key Distinguishing Features
Cystitis Cystica Glandularis	Superficial bladder mucosa	ER/PR negative	No desmoplasia, no atypia
Urachal Remnant	Bladder dome, midline	ER/PR negative	Fragmented tubules, no goblet cells, preserved muscularis propria
Nephrogenic Adenoma	Superficial/lamina propria	ER/PR negative	Small tubules with hobnail cells, no mucin
Bladder Adenocarcinoma	Invasive growth	Racemase positive, ER/PR negative	Desmoplasia, cellular atypia, older age
Urachal Adenocarcinoma	Bladder dome	Variable	Mucinous features, invasive pattern
Secondary Spread from Cervix	Variable	ER/PR may be positive	May mimic adenocarcinoma; requires cervical examination

imaging often revealing bladder wall thickening or mass lesions.[4, 6, 7] The diagnostic hallmark is histological identification of at least two Müllerian-type tissues (endosalpinx, endometrium, or endocervix) within the bladder wall. [4, 8] Our case demonstrated endocervical glands and endometrial stroma confirmed by immunohistochemistry.

Immunohistochemistry is crucial for diagnosis and differentiation from other entities. The ER/PR positivity in glandular epithelium and CD10 positivity in stroma are consistent with endometrial-type tissue.[2, 6, 9] Table 1 outlines key differential diagnoses. Misdiagnosis may lead to unnecessary radical surgery. Hormonal therapy with GnRH agonists may be effective, though surgery remains definitive for persistent symptoms.[3, 9]

Fewer than 40 cases have been documented worldwide (Table 2), with recent reports describing ureteral involvement and recurrence.[6, 8, 9] Our case is unique in providing 8-year follow-up data, demonstrating the benign nature and supporting conservative surgical management with meticulous pathological assessment.

Conclusion

Müllerianosis of the urinary bladder is a rare benign entity that can mimic malignancy. Diagnosis relies on histopathology with immunohistochemistry (ER, PR, CD10) to differentiate it from other lesions. Early accurate diagnosis enables hormonal therapy and avoids radical surgery. TURBT is the preferred treatment. Given its rarity, optimal management strategies require further investigation through larger studies, including evaluation of possible genetic factors.

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Table 2: Documented cases of bladder *Mullerianosis* in the literature.

Author (Year)	Age (years)	Symptoms	Histological Components	Diagnostic Method	Treatment	Reference
Young & Clement (1996)	28, 42	Hematuria, dysuria	*Endometriosis*, *Endocervicosis*, *Endosalpingosis*	Cystoscopy + Biopsy	TURBT	[1]
Donné et al. (1998)	35	Frequency, hematuria	*Endometriosis*, *Endocervicosis*	Cystoscopy + Histopathology	TURBT	[2]
Koren et al. (2006)	36	Hematuria	*Endometriosis*, *Endosalpingosis*	Cystoscopy + Biopsy	Surgical resection	[3]
Branca et al. (2014)	30	Dysuria	*Endometriosis*, *Endosalpingosis*	Histopathology	TURBT	[4]
Vella et al. (2011)	34	Urinary urgency	*Endometriosis*, *Endocervicosis*, *Endosalpingosis*	Cystoscopy + Histology	Excision	[5]
Guan et al. (2012)	29	Microscopic hematuria	*Endometriosis*, *Endocervicosis*	Urine cytology + Biopsy	Excision	[6]
Ogah et al. (2012)	33	Urinary retention	*Endometriosis*, *Endosalpingosis*	Cystoscopy + Imaging	Surgical resection	[7]
Mardi et al. (2024)	32	Hematuria, dysuria	*Endometriosis*, *Endocervicosis*	Histopathology	TURBT	[8]
Wegrzyn et al. (2024)	31	Pelvic pain, dysuria	*Endometriosis*, *Endocervicosis*, *Endosalpingosis*	Cystoscopy + Histopathology	TURBT	[9]
Current case (2025)	40	Dysuria, pelvic pain	*Endocervicosis*, *Endometriosis*	Cystoscopy + Histopathology	TURBT; No recurrence at 8 years	—

Note: The current case was diagnosed in 2016 and is being reported in 2025 with 8-year follow-up data.

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Competing Interests: None

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