Case series – Urethra diaries: Not every bulge is pelvic organ prolapse

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CASE REPORT 1

KEY MESSAGES

- A vaginal bulge is not always pelvic organ prolapse.
- A periurethral mass may be a prolapsed bladder lesion or urethral leiomyoma.
- Examine the mass to its base to identify its origin: this may require imaging or cystoscopy

A 38-year-old woman presented to the emergency department with sudden onset voiding dysfunction, vaginal pain and a new protruding vaginal mass following a sneezing episode. Her presenting creatinine was normal at 79 and GFR was 82. She has a past medical history of mild stress urinary incontinence, non-specified anxiety, and depression disorders on escitalopram daily, and has had two uncomplicated vaginal deliveries. Urogynecology was consulted for an irreducible vaginal prolapse as multiple attempts to reduce the bulge were unsuccessful. On examination, a purple mass was noted at the 6-o'clock position beneath the urethral meatus (Figure 1A). This appeared to be most in keeping with a thrombosed lesion. The patient was in significant pain.

Diagnostic process

Two urogynecology staff examined the patient in the clinic. A Foley catheter was inserted, and urine sent for culture. Local anesthetic jelly was applied, and with patient consent the mass was drained of clots and serosanguinous fluid via a small scalpel incision. Monsel's solution was applied for hemostasis. The patient experienced immediate relief. The fluid was sent for culture and cytology. She was started empirically on Amoxicillin-Clavulanic Acid 875/125 mg as she

reported symptoms of lower urinary tract symptoms prior to cyst prolapse. A kidney and bladder US was ordered that day.

She was booked for an examination under anesthesia, possible cyst excision and cystoscopy the following week. Urology was informed of the case and was made available for intraoperative consultation.

The fluid culture grew pan-sensitive E. Coli. Urine cytology was negative for high-grade urothelial carcinoma.

On the day of her excision, the cyst was no longer visible at the urethral meatus. A 17-French, 30-degree cystoscope was introduced. The bladder was visualized and appeared normal on cystoscopy. The urethra was normal. The right ureter and ureteric jet were visualized. The left ureteric orifice was obscured by a large round mass (Figures 1B -1C).

Treatment and management

Urology was consulted intraoperatively for suspicion of ureterocele. CT urogram was recommended and a large left-sided ureterocele measuring 35 x 26 mm was noted. There was concern for extravasation of contrast between the posterior bladder and ureterocele and the vaginal vault on the portal venous phase of the study, not clearly demonstrated on the urographic phase. The kidneys, proximal and mid portions of bilateral ureters appeared normal. There was no calculi or obstruction. There was no evidence of an intraperitoneal or extraperitoneal urinoma. The patient denied any leaking per vagina. The patient was able to void post-operatively and was sent home without a catheter. Her creatinine was normal.
It was concluded that the ureterocele had prolapsed through the urethra following an abrupt sneeze causing hemorrhage and distention requiring urgent decompression in the clinic the week prior.

Outcome

One month later the patient underwent a cystoscopy with urology. A large left ureterocele was noted to be wide open with left distal ureter dilatation. It did not require surgical resection. There were no concerns for vesico-vaginal fistula. The patient had a complete resolution of symptoms. She will have outpatient follow-up in 6 months with urology.

Conclusions

Ureteroceles are described as a cystic dilation of a distal ureter at the level of the bladder (1). It is a common congenital anomaly in children, and at times associated with a duplex renal collecting system (1,3). They are rare in both adolescents and adults (1). It commonly affects the left ureter in female patients, its incidence estimated to be between 1/5000- 1/2000 (1). A prolapsed ureterocele occurs in 5-10% of cases and has been reported more commonly in caucasian women (1).

There are two classifications of ureteroceles. They are either intravesical or extravesical. An intravesical ureterocele's ureteral meatus is above the neck of the bladder in the intravesicular form and originates from the bladder neck in the extravesicular form. The latter

can be associated with vesicoureteral reflux (1). Our case is more consistent with an intravesical ureterocele.

Strangulation and necrosis of tissue, as in our case, is a complication of a prolapsed ureterocele. Urethra Diaries: A Case of Ureterocele Prolapse; November 2023
Manual reduction can be attempted, and if this is not helpful, surgical management can be considered (1). Ureteroceles can present differently. Common symptoms include pelvic pain, urinary retention or incontinence, bleeding vaginal mass and flank pain (2). Management of ureterocele is also variable. Some surgeons opt for a reduction via incision or ureteric reimplantation (2).

Manual reduction can also be performed in office or in the operating room with a rigid scope (2). If post-reduction imaging does not show any obstruction, surveillance with periodic US of the kidneys is recommended (2). Definitive management can include ureteroneocystostomy, nephroureterectomy or transurethral unroofing of the ureterocele (2). Our patient's symptoms and prolapsed ureterocele was reduced and treated with a simple incision of the prolapsed portion of tissue.

In conclusion, prolapsed ureteroceles should be considered in the differential diagnosis in those women presenting with a vulvar or vaginal discomfort and concomitant urinary tract obstruction. Although this presentation is uncommon, early management is crucial to prevent hydroureter, hydronephrosis and urinary retention.

CASE REPORT 2

A 73-year-old woman presented to her family doctor in November 2020 with a three-to-four-month history of urinary incontinence, and a several-year history of a palpable bulge at the vaginal introitus. She denied any postmenopausal bleeding. Her past medical history included type-2 diabetes mellitus, hypertension, gastroesophageal reflux disease, and hypothyroidism, all well-controlled with medication. She had one prior pregnancy delivered via Caesarean section. She was menopausal at 48. She was referred to the Urogynecology clinic with a reducible stage 2 anterior compartment prolapse. Due to the evolving COVID-19 pandemic, she was seen in the clinic roughly two years later.

Diagnostic process

Before presenting to the Urogynecology clinic, the patient completed a pelvic ultrasound, ordered by her General Surgeon whom she was seeing for hemorrhoid management. Completed August 2022, the pelvic ultrasound reported a 2.9 x 2.2 x 3.3 cm mass contiguous with the cervix in the anterior vaginal wall concerning for "cervical fibroid or carcinoma". Her referral was expedited, and she was seen in the Urogynecology clinic. Upon examination, a solid, non-fluctuant sub-urethral mass was palpable beneath the vaginal epithelium, separate from the cervix. Moderate to severe vaginal atrophy was apparent throughout. There was no pelvic organ prolapse.

A pelvic MR was completed and described a low T2 signal mass localized to the posterior urethral wall measuring 2.7 x 3.5 x 3.0 cm (Figures 2, 3). The appearance was nonspecific but suggestive of leiomyoma, however, histology was recommended for definitive diagnosis. The remaining surrounding structures were unremarkable.

Given the concern for malignancy raised on ultrasonography, the patient consented to an examination under anesthesia, cystoscopy, hysteroscopy, and biopsy. The bladder and urethra appeared unremarkable, and random biopsies were taken. During cystoscopy, special attention was taken to examine the vesicourethral junction and urethra. Mass effect was noted at the 5 o'clock position at the vesicourethral junction. There was no evidence of communication to the vagina nor disturbance to urethral sphincter coaptation. On hysteroscopy, the endometrium appeared atrophic. Endometrial biopsy was obtained. Lastly, a wedge biopsy of the mass was obtained through a small anterior vaginal incision. All specimens were sent to pathology.

Both the bladder and urethral biopsies were benign. The endometrial biopsy demonstrated atrophic endometrium negative for hyperplasia or malignancy and the sub-urethral biopsy of the mass was consistent with leiomyoma.

Treatment and management

The patient opted for full excision of the mass due to her bothersome symptoms. She was aware of the risk of possible future urethrovaginal fistula and urinary incontinence if the urethral sphincter were to be disrupted. She underwent a myomectomy with an anterior vaginal approach. The mass was shelled out and removed *in toto*. Cystourethroscopy was performed showing no evidence of injury to the bladder or urethra. The sub-urethral space was then repaired in layers and the vaginal epithelium was approximated at the midline. The patient failed our institution's standard trial of void protocol and opted to be discharged with an indwelling Foley catheter for bladder rest. The foley was removed the following week with a successful trial of void.

Outcome

At her 6-week postoperative visit the patient reported an unremarkable recovery. On examination moderate to significant vaginal atrophy was again noted and the suture line was healing appropriately. She was prescribed twice weekly vaginal estrogen cream for ongoing vaginal atrophy and discharged from the Urogynecology clinic.

Conclusions

Pelvic organ prolapse (POP) is a common, but distressing condition that affects up to an estimated 75% of women worldwide (4). Pelvic organ prolapse includes the herniation of anterior, posterior, and/or apical segments of the vagina, with or without descent of neighbouring pelvic organs (5). Patients often report sensations of pelvic pressure or a noticeable vaginal bulge. Other accompanying symptoms that should be screened for include urinary incontinence or retention, dyspareunia, vaginal bleeding, and defecatory dysfunction.

Leiomyomas are benign tumours of uterine smooth muscle cells and are a common gynecologic finding in those of reproductive age. Approximately 70-80% of individuals are affected with uterine leiomyomas by the age of 50 (6,7). Prevalence declines throughout menopause (8). For those who are postmenopausal, special consideration for malignancy should be made.

Extrauterine leiomyomas, also known as ectopic or parasitic leiomyomas, have been reported (9). Due to their rare nature, current literature consists primarily of case reports. The prevalence of ectopic leiomyomas is therefore unknown. A systematic review by Lete et al. (9) reported 274 patients diagnosed with ectopic leiomyoma within published literature before July 2015. Authors attest that prior myomectomy or hysterectomy, particularly procedures requiring morcellation, were significant risk factors for the development of an ectopic leiomyoma. Interestingly, approximately 60% of patients with ectopic leiomyomas had no prior uterine manipulation, and 11% of those for which information regarding the location of the leiomyoma was provided reported no concomitant uterine leiomyoma (9).

Locations of ectopic leiomyomas vary. Most are located throughout the abdominopelvic cavity; either diffusely, termed diffuse peritoneal leiomyomatosis, or localized, in the greater omentum, anterior abdominal wall, colon, or pelvic wall (9). When located here, abdominal pain, bleeding, or a mass-like sensation within the pelvis are the most reported presenting symptoms (9). However, there are rare cases of leiomyomas located adjacent to the urethra or within the vaginal wall (10,11,12). When located throughout the external genitourinary tract, patients report symptoms including, but not exclusive to, dyspareunia, urinary incontinence, and dysuria (10,11,12). Furthermore, some patients may present asymptomatically or complain of a palpable bulge at the introitus.

The symptomatology of ectopic leiomyomas is non-specific, and the diagnosis of such masses requires a high degree of suspicion particularly when POP occurs much more commonly. POP can affect 50% of women (13). Thus, a bulge seen at the vaginal introitus could be interpreted as POP. Pelvic examination, including palpation for contour, consistency, size, and reducibility of suspected prolapse, along with imaging, can improve the assessment of patient complaints.

Although rare, sarcomas, ectopic leiomyomas, and cervical, vulvar, and vaginal cancers should be included in the differential diagnosis when investigating any patient presenting with a palpable vaginal bulge. Not every vaginal bulge is pelvic organ prolapse.

The patient outlined in this case was seen and referred prior to the pandemic but, unfortunately, the pandemic began to develop shortly thereafter. Ultimately, imaging studies were not complete until roughly two years following her referral to the Urogynecology clinic, and initial pelvic ultrasound demonstrated findings worrisome for cervical carcinoma. Fortunately, the etiology of the patient's symptoms was benign, and a diagnosis of sub-urethral leiomyoma was made. Given the patient's age and post-menopausal status, the outcome could

have been different. This case demonstrates the importance of imaging for risk stratifying and triaging patients, especially at the time of referral.

OVERALL CONCUSIONS

Both cases highlight that not every vaginal bulge is pelvic organ prolapse, and less common presentations should be considered in the differential diagnosis. Below is a table of differential diagnoses.

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FIGURES AND TABLES

Figure 1A. Prolapsed ureterocele at the time of initial presentation, prior to incision and drainage.

Figure 1B. Serial hysteroscopic images of left ureterocele.

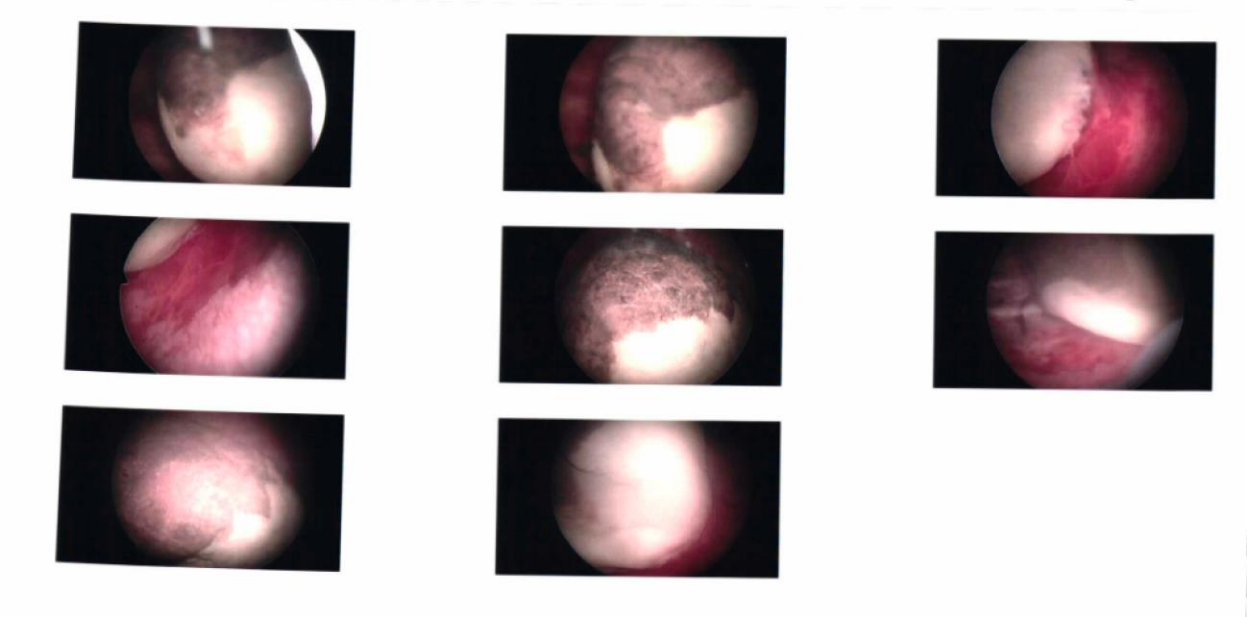

Figure 1C. Hysteroscopic image of the patient's left ureterocele.

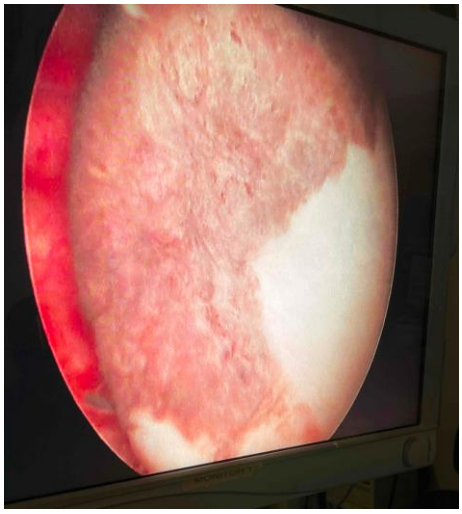

Figure 2. Select T2-weighted sagittal (A) and coronal (B) pelvic magnetic resonance images demonstrating a large sub urethral fibroid (white arrows) measuring 2.7 cm x 3.5 cm x 3.0 cm. Red arrows indicate lateral displacement of the urethra secondary to fibroid mass effect.

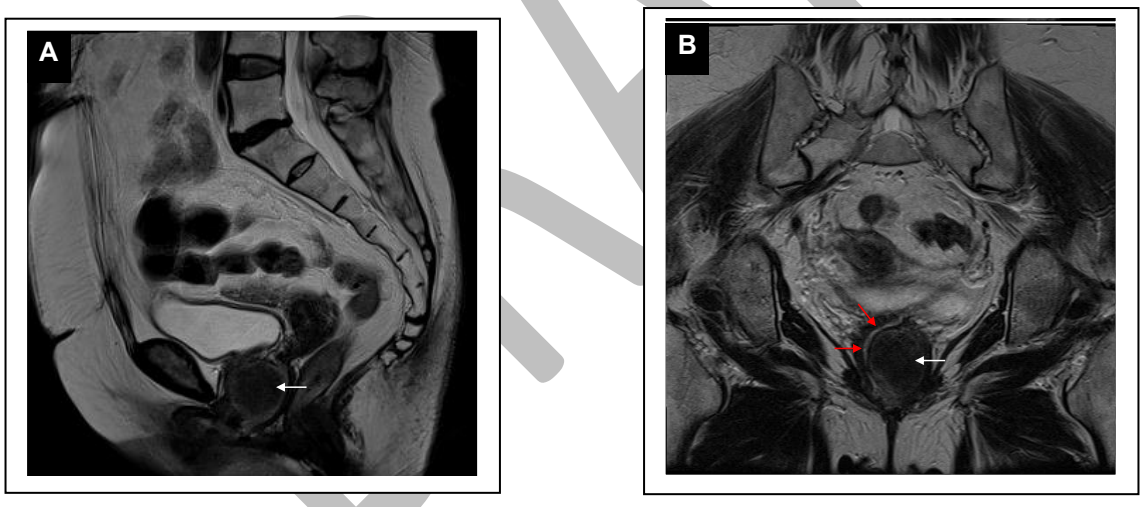

Figure 3. Left: Vaginal bulge with evidence of anterior incision from prior excisional biopsy. Middle: Vaginal mass in situ mid-dissection. Right: Operative specimen (leiomyoma) removed in its entirety measuring ~3cm.

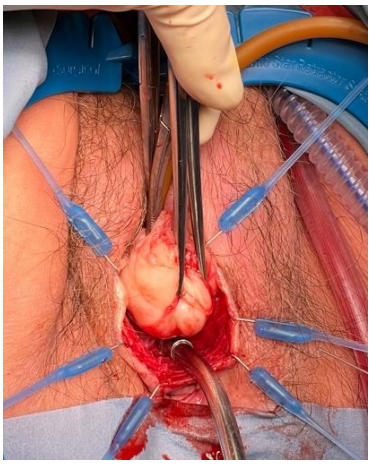

Table 1			
Diagnosis	**Presentation**	**Treatment**	**Incidence**
Benign masses **1) Cystic tumors** **Gartner duct cysts:** Remnants of the mesonephric duct.	These cysts are usually found in the upper lateral vagina. Patients may feel discomfort with intercourse.	If they are symptomatic, they can be excised. Symptomatic cysts can also be biopsied. Vaginal adenosis can be ruled out with Lugol's solution staining. If the cysts are small and asymptomatic, they can be left.	Incidence is 12.5%
Paramesonephric duct cyst: Remnants of the paramesonephros and lined with secretory epithelium. Mullerian in origin. These are lined with secretory cells and often contain mucous.	Can be found anywhere in the vagina. Patient may feel discomfort with intercourse.	If symptomatic, they can be excised. Vaginal adenosis can be ruled out with Lugol's solution staining.	Incidence is 1–5%
Inclusion cysts: These cysts are formed by trapped vaginal mucosa in the submucosal layers. They contain keratin and squamous cells. They are often secondary to vaginal trauma from childbirth, vaginal repairs for tears, due to episiotomies or colporrhaphy.	As above	They can be excised if patient have symptoms.	Incidence is 1/200

Endometriosis: Endometrial tissue growing outside the uterus. Endometriosis can be found in the vagina due to erosion/implants along a surgical site.			
Skene duct cyst: These para-urethral glands release mucous which protects and lubricates the urethral opening. These glands are homologous to the prostate.	Endometriosis can appear blue or brown in color. At times, lesions can be palpated in the posterior vaginal wall fornixes or along the uterosacral ligaments. These would cause pain and discomfort with intercourse.	Excision may require laparoscopy. Small lesions visualized in the vagina could be vaporized with laser or excised. Medical management of endometriosis could also help treat these lesions.	Incidence is approximately 10%
Bartholin gland cyst: Glands at the vaginal introitus at the 4 and 8 o'clock positions. They are lined with secretory epithelium and secrete mucous.	Can present as a painful or asymptomatic mass inferolateral to the urethra.	If they are symptomatic, they can be excised.	1/2000–1/7000
*The differential for a midline anterior vaginal mass includes a urethral diverticulum, fibroepithelial polyp, skene duct cyst, cystocele (pelvic organ prolapse) or malignant tumors.	Patients can present with non-infected cysts or abscesses. They can present with vaginal discomfort, fullness, bleeding, or discharge.	These cysts can be treated with incision and drainage, word catheter insertion of marsupialization.	Symptomatic cysts make up about 2% of yearly gynecologic visits.
2) Solid tumors			

Leiomyoma (fibroids): Vaginal, cervical, or urethral fibroids are rare benign tumors which have arisen from a smooth muscle cell. You can also have prolapsed submucosal fibroids through the cervix. These fibroids are considered "parasitic" as they are not uterine.			
Fibroepithelial polyp: An uncommon polyp which can enlarge in pregnancy. They are lined by squamous epithelium and have a fibrovascular stalk.	Patients can present with vaginal bleeding and/or discomfort with intercourse. Urethral fibroids may present with lower urinary tract symptoms.	These can be surgically excised.	Fibroids can be found in up to 70% of women.
Condyloma acuminatum: HPV related tumors which can be found on the cervix, along the vagina, vulva, and perianal areas.			
Urethral caruncle: red and friable lesion at the urethral meatus, often found in post-menopausal women.	Patients may present with vaginal bleeding and discomfort.	These can be excised.	Rare.
3) Cervical polyps			
Prolapsed endometrial polyps: 5% of endometrial polyps are malignant.	Patients may present with bleeding, cosmetic concerns, discomfort, or pruritus.	These can be treated with topical therapies, cryotherapy or excision.	Annual incidence is 1%.

	Women may present with bleeding, pain (as well as dysuria). Patients may present with vaginal bleeding. Patients may present with vaginal bleeding.	Topical estrogen therapy, systemic hormone replacement therapy. They can be excised. Patients often require hysteroscopy. Polyps can be excised at that time.	There is a 1.6% incidence of malignancy. Incidence ranges between 2–5%. Incidence in reproductive aged women with abnormal uterine bleeding is between 20–40%.
Pelvic organ prolapse Cystocele Rectocele Complete procidentia	Women may present with vaginal bulge, discomfort, bleeding. They may present with urinary or fecal incontinence.	Treatment can include pessary insertion, surgical repair.	Incidence can be as high as 50%.
Prolapsed ureteroceles **Urethral prolapse**	Can present with pain (including dysuria), urinary retention, urinary incontinence. As above	May require cystoscopy, surgical reduction. As above	1/5000–1/12 000 Urethral prolapse is uncommon and can present in 1/3000 children.
Malignant tumors of the vulva, vagina, and cervix	Patients may present with vaginal bleeding, vulvar bleeding, pain, discomfort.	Discussion of treatment is not within the scope of this case series.	