Case series – Urethra diaries

Not every bulge is pelvic organ prolapse

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CASE REPORT 1

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 glomerular filtration rate (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 lower urinary tract symptoms prior to cyst prolapse. A kidney and bladder ultrasound (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. Computed tomography (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 postoperatively 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 followup in six months with urology.

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

Discussion

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.[1,2] 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. 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.[3] Management of ureterocele is also variable. Some surgeons opt for a reduction via incision or ureteric reimplantation.[3]

Manual reduction can also be performed in office or in the operating room with a rigid scope.[3] If post-reduction imaging does not show any obstruction, surveillance with periodic US of the kidneys is recommended.[3] Definitive management can include ureteroneocystostomy, nephroureterectomy, or transurethral unroofing of the ureterocele.[3] Our patient's symptoms and prolapsed ureterocele were reduced and treated with a simple incision of the prolapsed portion of tissue.

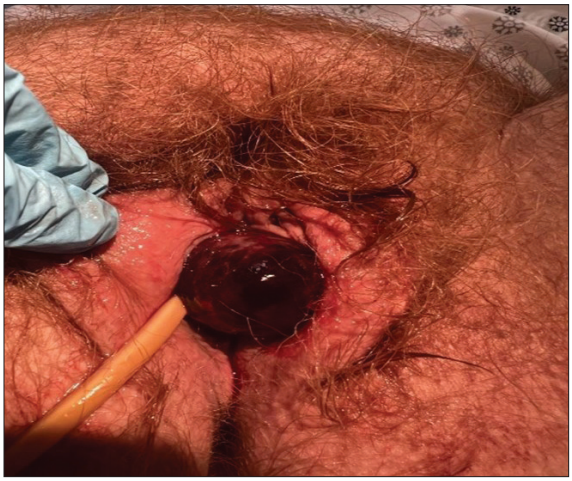

Figure 1A. Prolapsed ureterocele at the time of initial presentation, prior to incision and drainage.

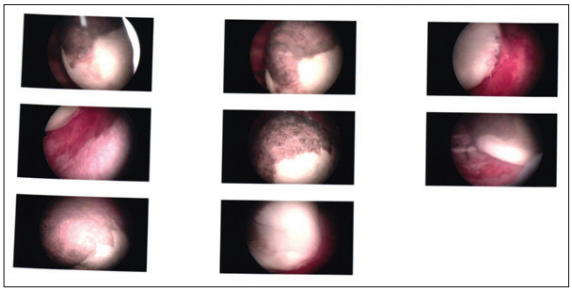

Figure 1B. Serial hysteroscopic images of left ureterocele.

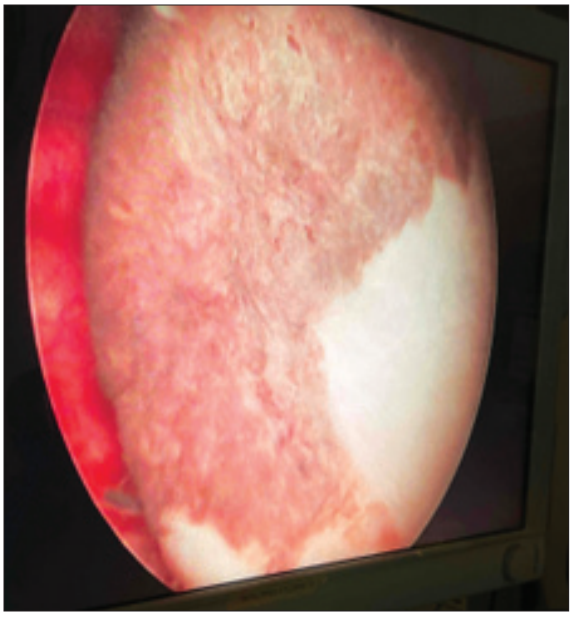

Figure 1C. Hysteroscopic image of the patient's left ureterocele.

Conclusions

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.

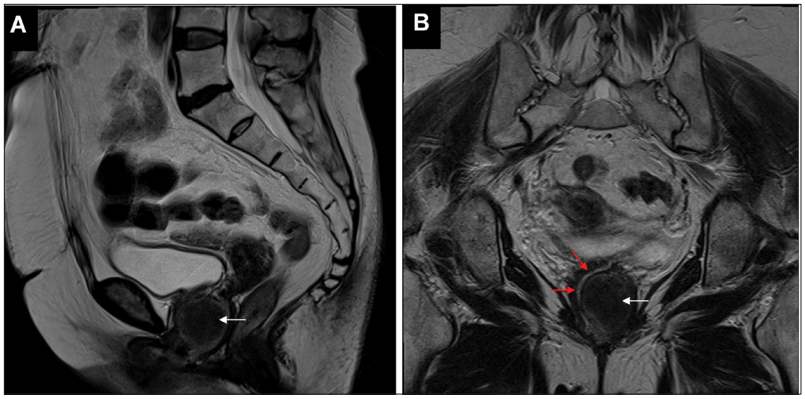

Figure 2. Select T2-weighted sagittal (A) and coronal (B) pelvic magnetic resonance images demonstrating a large sub urethral fibroid (white arrows) measuring 2.7 cm x 3.5 cm x 3.0 cm. Red arrows indicate lateral displacement of the urethra secondary to fibroid mass effect.

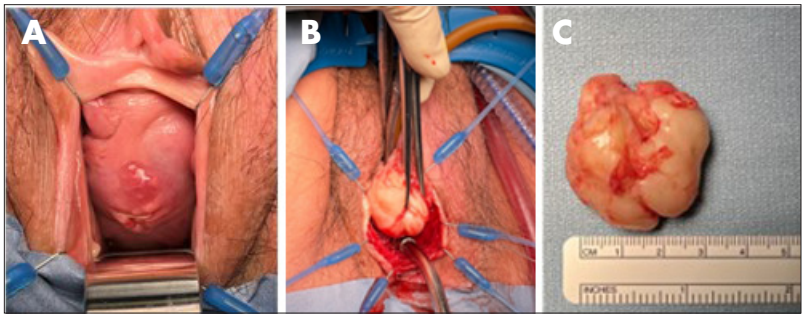

Figure 3. (A) Vaginal bulge with evidence of anterior incision from prior excisional biopsy. (B) Vaginal mass in situ mid-dissection. (C) Operative specimen (leiomyoma) removed in its entirety measuring ~3 cm.

Diagnostic process

Before presenting to the urogynecology clinic, the patient completed a pelvic US, ordered by her general surgeon, whom she was seeing for hemorrhoid management. Completed August 2022, the pelvic US 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 suburethral 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 (POP).

A pelvic magnetic resonance 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 non-specific 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 suburethral 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 suburethral 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 six-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.

Discussion

POP is a common, but distressing condition that affects up to an estimated 75% of women worldwide.[4] POP includes the herniation of anterior, posterior, and/or apical segments of the vagina, with or without descent of neighboring 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 tumors 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 reported 274 patients diagnosed with ectopic leiomyoma within published literature before July 2015.[9] 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.[10-12] When located throughout the external genitourinary tract, patients report symptoms including, but not exclusive to, dyspareunia, urinary incontinence, and dysuria.[10-12] Furthermore, some patients may present asymptomatically or complain of a palpable bulge at the introitus.

The symptomatology of ectopic leiomyomas is nonspecific, 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, thus, a bulge seen at the vaginal introitus could be interpreted as POP.[13] 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 POP.

Conclusions

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 US demonstrated findings worrisome for cervical carcinoma. Fortunately, the etiology of the patient's symptoms was benign, and a diagnosis of suburethral 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 POP, and less common presentations should be considered in the differential diagnosis. An Appendix of differential diagnoses is available online.

COMPETING INTERESTS: The authors do not report any competing personal or financial interests related to this work.

This paper has been peer reviewed.

REFERENCES

1. Delgado Pacheco M, Arias Delgado JA, Imán Izquierdo FJ, et al. Strangulated prolapsed ureterocele in an adult female. *Urol Case Rep* 2023;47:102374. https://doi.org/10.1016/j.eucr.2023.102374
2. Yuri P, Utama ETP. A complete duplicated collecting system with giant ureterocele in adult: Case report. *Int J Surg Case Rep* 2021;79:49-52. https://doi.org/10.1016/j.ijscr.2020.12.083
3. Balzano F, Jellison F. Management of single system prolapsed ureterocele: A case report and review of literature. *MOJ Clin Med Case Rep* 2019;9:57-8. https://doi.org/10.15406/mojcr.2019.09.00304
4. Wilkins MF, Wu JM. Epidemiology of pelvic organ prolapse. *Curr Obstet Gynecol Rep* 2016;5:119-23. https://doi.org/10.1007/s13669-016-0149-z
5. Rogers R, Fashokun T. An overview of the epidemiology, risk factors, clinical manifestations, and management of pelvic organ prolapse in women. UpToDate [Internet]. 2014;1-18. Available from: http://www.uptodate.com/contents/an-overview-of-the-epidemiology-risk-factors-clinical-manifestations-and-management-of-pelvic-organ-prolapse-in-women?source=search_result&search=vaginal+prolapse&selectedTitle=2~27
6. Drayer SM, Catherino WH. Prevalence, morbidity, and current medical management of uterine leiomyomas. *Int J Gynecol Obstet* 2015;131:117-22. https://doi.org/10.1016/j.ijgo.2015.04.051
7. Vilos GA, Allaire C, Laberge PY, et al. The management of uterine leiomyomas. *J Obstet Gynaecol Canada* 2015;37:157-78. https://doi.org/10.1016/S1701-2163(15)30338-8
8. Wise LA, Laughlin-Tomasso SK. Epidemiology of Uterine Fibroids. *Clin Obstet Gynecol* 2016;59:2-24. https://doi.org/10.1097/GRF.0000000000000164
9. Lete I, González J, Ugarte L, et al. Parasitic leiomyomas: A systematic review. *Eur J Obstet Gynecol Reprod Biol* 2016;203:250-9. https://doi.org/10.1016/j.ejogrb.2016.05.025
10. Bolukbasi A, Kandemir B, Bolukbasi S. Urethral leiomyoma. Case report. *Hacettepe Med J* 1984;17:42-5.
11. Özel B, Ballard C. Urethral and paraurethral leiomyomas in the female patient. *Int Urogynecol J* 2006;17:93-5. https://doi.org/10.1007/s00192-005-1316-3
12. Lee MC, Lee S Der, Kuo HT, et al. Obstructive Leiomyoma of the Female Urethra: Report of a Case. *J Urol* 1995;153:420-1. https://doi.org/10.1097/00005392-199502000-00041
13. Weintraub AY, Glinter H, Marcus-Braun N. Narrative review of the epidemiology, diagnosis and pathophysiology of pelvic organ prolapse. *Int Braz J Urol* 2020;46:5-14. https://doi.org/10.1590/s1677-5538.ibju.2018.0581

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