CASE REPORT

Bladder Endometriosis Presenting as Obstructive Uropathy in a Post-hysterectomy Patient

Antolyn E. Exconde Jr., MD; Kathleen R. Gonzales, MD, FPUA and Robert Leeh E. Pedragosa, MD

Urology Service, Department of Surgery, AFP Medical Center

Urinary tract endometriosis is present in approximately 1% of women. Bladder endometriosis (BE) is defined as presence of endometrial glands and stroma in the detrusor muscle. Close collaboration of the urologists and gynecologists in dealing with urinary tract endometriosis is necessary in order to establish both the correct diagnostic procedure and the most adequate management. Presented here is the case of a 42-year old female, G3P3 (3003), who presented with intermittent episodes of right-sided flank pain four months post-hysterectomy. She was initially diagnosed to have a bladder tumor and underwent transurethral resection of bladder tumor. Histopathological examination of the resected specimen revealed endometriosis.

Keywords: Bladder endometriosis, obstructive uropathy

Introduction

Urinary tract endometriosis is present in approximately 1% of women with endometriosis, but its prevalence increases to 19–53% among patients who have the deep infiltrating endometriosis (DIE) type of endometriosis. Its peak incidence is between 30 and 45 years of age.[1-3] Involvement is thought to occur highest in the urinary bladder (85%), ureter (9%), kidney (4%) and urethra (2%).[4] Bladder endometriosis (BE) is defined as presence of endometrial glands and stroma in the detrusor muscle. The bladder base and the dome are the most frequently affected sites.[5] Bladder endometriosis (BE) can be classified as 'primary' or 'secondary': 1) primary BE is a spontaneously occurring disease, diagnosed in 11% of overall patients diagnosed with DIE[6], while 2) secondary BE is defined as an iatrogenic lesion, occurring after pelvic surgery, such as cesarean delivery or hysterectomy.[6] Up to 50% of patients with BE have past history of pelvic surgery .[7] Accompanying symptoms of BE includes frequency, dysuria, hematuria and less frequently, bladder pain and urgency. These symptoms may worsen during menstruation, or may have a non-cyclical presentation.[1] Close collaboration of the urologists and gynecologists in dealing with urinary tract endometriosis is necessary in order to establish both the correct diagnostic procedure and the most adequate management.

The Case

This is a case of a 42-year old female, G3P3 (3003), with regular menstrual cycle, a known case of abnormal uterine bleeding secondary to adenomyosis and endometrial polyp. She presented

with intermittent episodes of right-sided flank pain, described as colicky, non-radiating, 4/10 severity scale, four months post-hysterectomy. There were no associated cyclical gross hematuria, dysuria, dysmenorrhea, pelvic pain, fever and chills, vaginal discharge, lower urinary tract symptoms, such as frequency, urgency nor intermittency. Her obstetrician requested for a urinalysis which revealed pyuria, absence of microhematuria. An ultrasound of the kidneys, ureters and bladder (KUB) showed moderate dilatation of right uretero-pelvo-calyceal system (Figure 1A) and unremarkable left kidney and urinary bladder. The patient was started on oral quinolone and was then referred to a Urologist. An unenhanced abdominal computed tomography (CT) scan was requested which revealed severe dilatation of the right pelvocalyces and ureter down to the pelvic region where an abrupt narrowing at the 3rd sacral level was noted. There was fullness in the cervical region and rectovesical space and an apparent density along the posterior aspect of the urinary bladder (Figure 1C). This was confirmed using transvaginal (TV) ultrasound (Figure 1B).

The patient was then advised to have a serum creatinine determination and to undergo a CT Urography. Serum creatinine was 0.72 mg/dL. The CT urogram revealed a 3.31cm x 2.27cm x 3.06cm polypoid enhancing urinary bladder mass in the lower, posterior area (Figure 1D). There were no pelvic lymphadenopathies. The severe dilatation of the calyceal system seen on ultrasound was again observed extending to the level of the distal ureter (Figure 1E,F). At that time, the primary consideration was a bladder tumor with possible external compression of the right distal ureter resulting in secondary upper tract obstruction. A urine cytologic examination revealed absence of atypical and malignant cells. The patient was then advised to undergo cystoscopy, trans-urethral resection of bladder tumor (TURBT), with bimanual pelvic examination. Intraoperatively, a palpable, movable intravesical mass, measuring approximately 3 cm was confirmed through bimanual pelvic and vaginal examination under spinal anesthesia. Cystoscopy showed multiple nodular, wide-based, finger-like projections, with areas of hyperpigmentation, located

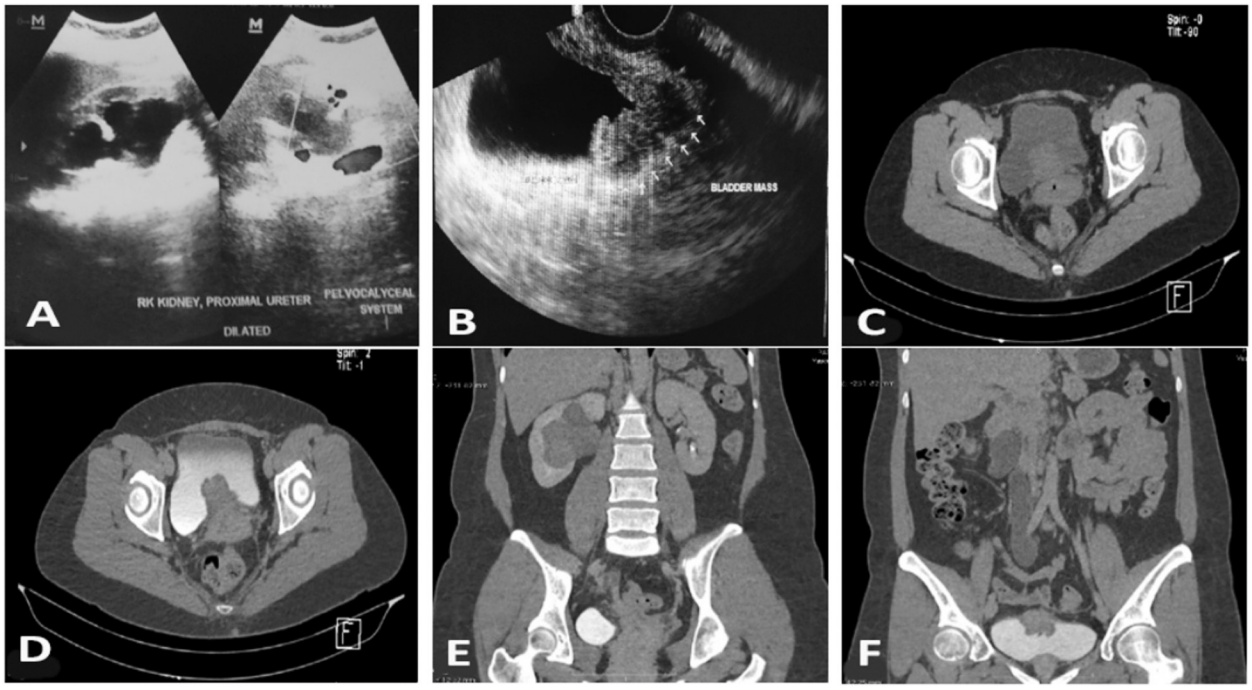

Figure 1. Pre-operative imaging results. A) KUB ultrasound showing moderate dilatation of right uretero-pelvo-calyceal system; B) Transvaginal ultrasound showing a hypoechoic mass with irregular borders at the posterolateral bladder wall; C) Unenhanced abdominal CT scan showing a density along the posterior aspect of the urinary bladder in axial view; D) CT urography revealed a 3.31cm x 2.27cm x 3.06cm polypoid enhancing lower posterior urinary bladder mass and (E, F) severe dilatation of the right collecting system down to the level of distal ureter

on the inter-ureteric ridge and extending to the right postero-lateral bladder wall and trigone (Figure 2). The right ureteral orifice was slit-like, and was adjacent laterally from the bladder mass. The left orifice was unremarkable. The patient underwent TURBT with complete tumor resection. The tissue specimen weighed 2.86 grams and had an aggregate diameter of 2.9 centimeters. A nephrostomy tube was inserted into the right middle calyx and had an initial output of 450 mL of clear urine. Daily nephrostomy output was 200 mL per day. No growth of bacteria on urine gram stain with culture studies was noted. Histologic section showed fibromuscular tissues with varisized and occasional cystically dilated lands lined by cuboidal to columnar epithelium, with round nuclei and ample eosinophilic cytoplasm, and with scattered lymphoplasmacytic infiltrates. This was consistent with endometriosis. The patient was referred back to gynecology service for co-management and a GnRH agonist injection was started, with a 3-month interval. Re-evaluation every after 3 months showed regression in size of the endometrial mass on transvaginal ultrasonography (Figure 3C). Decreasing nephrostomy output, cupping of the calyces, and spontaneous passage of contrast material on the collecting system down to the bladder, complete emptying of dye on delayed nephrostogram films prompted removal of the nephrostomy tube (Figure 3A, B). The patient is doing regular follow-up and regular surveillance ultrasound.

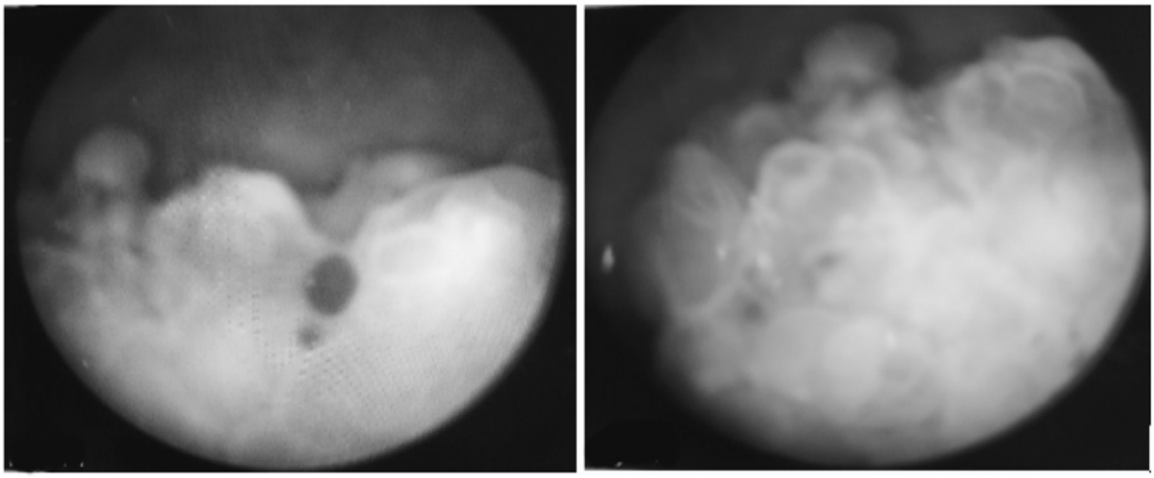

Figure 2. Intra-operative findings. Cystoscopic findings of multiple nodular wide-based finger like projections with areas of hyperpigmentation located on the inter-ureteric ridge extending to the right postero-lateral bladder wall

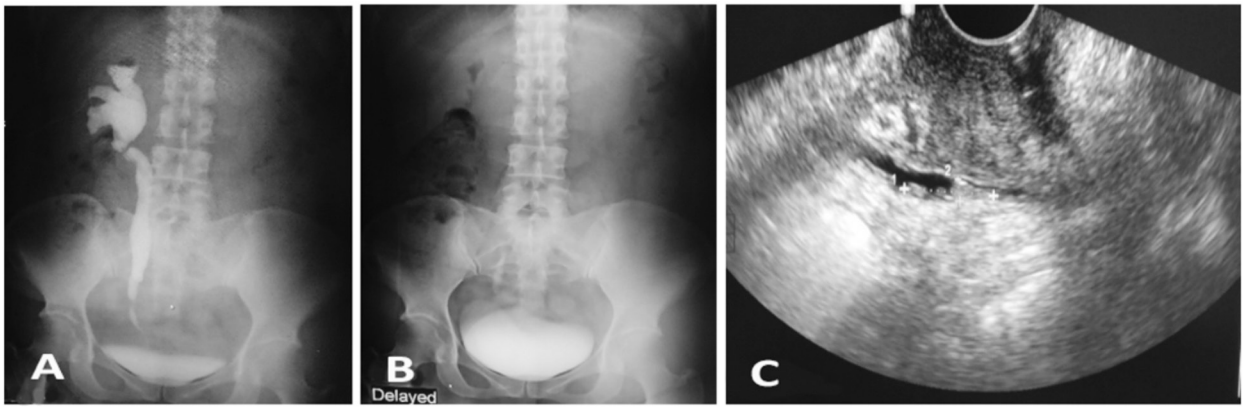

Figure 3. Post-operative antegrade nephrostogram showing A) spontaneous passage of contrast material on the collecting system down to the bladder with, B) complete egress on delayed film and C) repeat transvaginal ultrasound revealing regression in size of bladder endometriosis.

Discussion

Bladder endometriosis may come with variable symptoms and subtle onset, often mimicking recurrent cystitis. Dysuria has been reported in 21–69% of patients with BE and positive correlation was observed between severity of dsyuria and lesion diameter. Hematuria, a less frequent symptom, has been reported in 0–35% of the cases and is explained by the fact that the bladder lesion rarely infiltrates the mucosal layer. Urinary tract obstruction, a rare symptom, may be observed in <1% of these patients.[8] These are overlapping symptoms occurring in a cyclical manner, with predominance during the pre-menstrual period. A complete and thorough history and physical examination is warranted to rule out malignancy and urinary tract obstruction that should be addressed immediately. Physical examination of bladder pathology includes a detailed bimanual pelvic and vaginal examination, especially if the patient presented with mixed urologic and gynecologic symptoms. Endometriotic infiltration of the pelvis is considered if the bimanual examination reveals a palpable nodule, thickened area, or a palpable cystic expansion with topographic-anatomical correlation to uterosacral ligaments, vagina, rectovaginal space, pouch of Douglas, the rectosigmoid and the posterior wall of the urinary bladder.[4]

Choice of diagnostic modalities used in identifying BE depends upon the clinical presentation of each patient. These would include urine examinations for evaluation of hematuria and urine culture to rule out an infectious etiology. A urine cytologic examination is mandatory in patients with documented bladder tumors. Patients should also be assessed for renal function using creatinine measurements especially if with suspicion of possible urinary tract obstruction.

Imaging modalities are central in determining possible areas of endometrial infiltration. Ultrasound (US) is the first line modality due to its low cost, ready availability, and lack of radiation exposure. Transvaginal (TV) ultrasonography is highly accurate, non-invasive, cost-effective and estimates the distance between ureteral orifices and nodule borders. It is the mainstay for initial diagnostic technique for BE and has a negative predictive value of almost 100%. Sensitivity is under 50% because the detection rate is strongly related to mean lesional diameter.[5] Combination of abdominal, transvaginal and transrectal ultrasound may reveal endometriosis located in uncommon sites. When there is evidence of urinary tract obstruction, like incidental finding of hydronephrosis, further investigation and additional upper urinary tract imaging modalities is warranted. Hydronephrosis is associated with muscular invasion in 70-90% cases, distant metastasis in 55% and is regarded as an important factor in staging of bladder malignancy.[9] Differential diagnoses in patients who had previous pelvic surgery, found to have hydronephrosis, include ureteral stricture secondary to iatrogenic ureteral injury, or in this case, a secondary BE obstructing the ureteral orifice. CT scan and abdominal magnetic resonance (MR) as a complementary examination in complex cases of endometriosis with extensive adhesions is needed as a standard for the diagnosis.[4]

Cystoscopy in BE remains one of the most cost-effective modalities. It is helpful in evaluating the location and configuration of the ureteral orifice or in estimating the distance between the ureteral orifices and nodule borders needed in planning the most appropriate surgical procedure. Scheduling of cystoscopy immediately before or during menstruation is recommended. During menstruation the nodule is larger and more congested, and thus, cystoscopic visualization is clearer.[5] It should be considered that with the exception of transurethral resection (TUR) procedures, biopsy at cystoscopy is frequently not diagnostic for endometriosis[4], owing to its possible intraperitoneal origin. BE lesions often appear as an adenomatous and irregular nodular mass, with different shapes and colors: the blue-red, blue-black, or blue-brown lesions are the more common findings and the urothelium is not usually ulcerated. They might be isolated or multifocal, approximately 1–3 cm in diameter and usually located at the dome or at the base.[10]

In the setting of upper tract obstruction in a bladder tumor with unknown histologic diagnosis, antegrade percutaneous access is generally indicated. This provides drainage, and preserves renal function in long-standing obstruction. Based on the field-cancerization effect and urothelial re-implantation theory, tumor spread may be possible with retrograde placement of ureteral stent.[11] This is avoided by placing an antegrade stent.

Proper tissue diagnosis is necessary prior to initiation of treatment of BE whether it may be surgical or medical. Primarily, medical management for the treatment of BE is mainly hormonal. This may often serve as the initial treatment for younger women and those desiring to preserve fertility.[7,12] Treatment includes 1) GnRH agonists, 2) GnRH antagonists, 3) progestins, and 4) combined oral contraceptives.[4] The ideal candidates for hormonal therapy are bladder lesions with a diameter <5 mm, and post-menopausal women. Hormonal treatment appears to have a high recurrence rate after treatment cessation, and is often considered a palliative modality for the treatment of BE.[7]

Surgical treatment of BE, should be always performed to exclude a malignant lesion, and to define the exact location of the nodule and its relationship to the ureteral orifices. Two techniques have been proposed for surgical treatment of BE: Trans-urethral resection (TUR) and segmental bladder resection. TUR offers fast recovery and is indicated for pre-menopausal women, patients who desire pregnancy, and bladder lesions that are 0.5 – 1 cm deep.[5] However, TUR is associated with incomplete lesion removal, persistence of symptoms, high risk for bladder perforation, and rate of recurrence of 20-30%.[4] TUR, combined with hormonal therapy, yields disappearance of clinical symptoms in most of the patients, and an estimated recurrence of 25–35%.[13] Partial cystectomy is a safe and simple bladder-preserving surgery that can be performed via laparotomy or laparoscopy, with excellent long-term results, adequate symptom relief, and high recurrence prevention.[4,14] Concomitant cystoscopy is useful for defining the margins of the endometriotic lesion to be resected. Pre-operative catheterization of the ureters may be advisable when the distance between the caudal border of the endometriotic lesion and the interureteric ridge is <2cm.[15] In rare cases, a ureteroneocystostomy might be required.[4]

In conclusion, BE is a rare condition that has a wide variety of presentations and symptomatology. Hence, a detailed gynecologic history, which includes asking for previous pelvic surgery should be done. Not all BEs present with common symptoms such as dysuria, hematuria and lower urinary tract symptoms. Upper tract obstruction is rare, and warrants supplementary diagnostic modalities such as transvaginal ultrasound or CT urography. Management of BE warrants a symptom-specific approach to determine the type of management that would address symptom resolution, preservation of quality of life, fertility and prevention of recurrence. It is important to have a close collaboration between the urologist and the gynecologist when planning treatment protocols for BE.

References

1. Gabriel B, Nassif J, Trompoukis P, Barata S, Wattiez A. Prevalence and management of urinary tract endometriosis: a clinical case series. Urology 2011;78:1269–74.
2. Knabben L, Imboden S, Fellmann B, Nirgianakis K, Kuhn A, Mueller MD. Urinary tract endometriosis in patients with deep infiltrating endometriosis: prevalence, symptoms, management and proposal for a new clinical classification. Fertil Steril 2015; 103: 147–52.
3. Chapron C, Fauconnier A, Vieira M, et al. Anatomical distribution of deeply infiltrating endometriosis: surgical implications and proposition for a classification. Hum Reprod 2003; 18: 157–61.
4. Maccagnano C, et al. Diagnosis and treatment of bladder endometriosis: State of the art. Urologia Internationalis 2012; 89: 249–58.
5. Maggiore UL, et al. Bladder endometriosis: A systematic review of pathogenesis, diagnosis, treatment, impact on fertility and risk of malignant transformation. Eur Urol 2017; 71: 790–807.
6. Donnez J, Van Langendonckt A, Casanas- Roux F, et al. Current thinking on the pathogenesis of endometriosis. Gynecol Obstet Invest 2002; 54(suppl 1): 52–62.
7. Fedele L, Piazzola E, Raffaelli R, Bianchi S. Bladder endometriosis: deep infiltrating endometriosis or adenomyosis? Fertil Steril 1998; 69: 972–5.
8. Mehrsai A, et al. A comparison between clinical and pathologic staging in patients with bladder cancer. Iran Urol J 2004; 2: 22-6.
9. Villa G, Mabrouk M, Guerrini M, et al. Relationship between site and size of bladder endometriotic nodules and severity of dysuria. J Minim Invasive Gynecol 2007;14: 628–32.
10. Douglas C, Rotimi O: Extragenital endometriosis – a clinicopathological review of a Glasgow hospital experience with case illustrations. J Obstet Gynaecol 2004; 24: 804–8.
11. Jones TD, Wang M, et al. Molecular evidence supporting field effect in urothelial carcinogenesis. Clin Cancer Res Sept 2005.
12. Westney OL, Amundsen CL, McGuire EJ. Bladder endometriosis: conservative management. J Urol 2000; 163: 1814–7.
13. Sánchez Merino JM, Guillán Maquieira C, García Alonso J. The treatment of bladder endometriosis. Arch Esp Urol 2005; 58: 189–94.

14. Seracchioli R, Mabrouk M, Montanari G, Manuzzi L, Concetti S, Venturoli S. Conservative laparoscopic management of urinary tract endometriosis: surgical outcome and long-term follow-up. Fertil Steril 2010; 94: 856–61.

15. Berlanda N, Vercellini P, Carmignani L, Aimi G, Amicarelli F, Fedele L. Ureteral and vesical endometriosis. Two different clinical entities sharing the same pathogenesis. Obstet Gynecol Surv 2009; 64: 830–42.