



Bladder Endometriosis that Invisible to Repeated Cystoscopies: A Case Report

Ida Bagus Putra Adnyana^{1*}, I Made Purnama Adimerta²

- ¹. Department of Obstetrics and Gynecology, Faculty of Medicine, Universitas Udayana/Sanglah General Hospital, Indonesia.
- ². Department of Obstetrics and Gynecology, Bali Royal Hospital (BROS), Bali, Indonesia.

*Corresponding Author: Ida Bagus Putra Adnyana

Abstract

Background: Bladder endometriosis (BE) is a form of Deep Infiltrating Endometriosis (DIE) which affects approximately 1% of women with endometriosis. BE commonly presents with lower urinary tract symptoms such as frequency, dysuria, hematuria, and, less frequently, bladder pain and urgency. The diagnosis and treatment of this condition could be difficult and requires multidisciplinary care. Due to overall rare cases, BE might pose a challenge, both to diagnose and treat. This article aims to share an experience in order to increase awareness and insight about BE for practising gynaecologist and urologist. **Case:** A Thirty-one years old woman, nulligravida, complained of painful, bloody stained urine, which occurs during menstruation since married for the last 18 months. Previously patient had a regular menstruation cycle with moderate pain and normal menstrual volume and no history nor any specific symptoms of disease. Gynaecological examination showed no remarkable finding. Ultrasound scan (US) by gynaecologist showed a mass protruded from the posterior wall of bladder. Cystoscopic finding by a urologist showed an adenomatous reddish nodular mass, and TUR was conducted. Pathological examination showed a cystitic glandulare. After three months temporary improvement, the symptoms reoccur. The patient underwent US and CT scan, again showed a mass, and another two rounds of TUR were conducted, to remove all residual mass. After three surgeries with no significant improvement, a team of gynaecologist and urologist were set to conduct a laparoscopic partial cystectomy with cystoscopy guidance to achieve complete excision of the lesion. The modified technique, which results in symptoms-free recovery, will be described in detail. **Conclusion:** BE could be challenging both in diagnostic and treatment. Cystoscopy and TUR are not recommended to diagnose and manage the lesions. A multidisciplinary approach is needed, and concomitant Laparoscopy with cystoscopy for partial cystectomy might give a satisfying outcome.

Keywords: *Bladder Endometriosis, Deep Infiltrating Endometriosis, Cystoscopy, Laparoscopic partial cystectomy, Lower urinary tract symptoms.*

Introduction

Bladder endometriosis (BE) is a form of Deep Infiltrating Endometriosis (DIE). It affects approximately 1% of women with endometriosis [1]. Bladder Endometriosis is defined as the presence of glandular cells and endometrial stroma in the detrusor muscle. It divided as primary BE when it occurs spontaneously, or as secondary BE when it is related to an iatrogenic lesion occurring after pelvic surgery. The common symptoms of BE are lower urinary tract symptoms such as frequency, dysuria, hematuria, and, less frequently, bladder pain and urgency.

Additional imaging modalities such as ultrasound, MRI, and cystoscopy were needed to support the diagnosis and plan the treatment [1,2]. The pathogenesis of BE proposed by several authors include embryonal theory which proposed that BE might originate from the Mullerian remnants [2]. Migratory or metastatic theory postulates that products of menstruation are regurgitated into the pelvis through the fallopian tubes in a retrograde fashion and then implanted on the bladder wall. An alternative mechanism of pathogenesis which

mainly supported by anatomical-pathological studies, The Transplantation theory, postulates that BE should be defined as bladder adenomyosis. BE thought to arise as an extension of adenomyotic lesions and transported via lymphomatous or hematogenous embolic. Iatrogenic theory, in another view, proposed different pathogenesis in which BE ensues after a cesarean section. In this case, the vesical nodule does not represent a generalised disease, but it is usually isolated and may be caused by intraoperative dissemination of endometrial cells or by a suboptimal surgical technique for closure of the low transverse uterine incision [2]. The diagnosis and treatment of this condition could be difficult and requires multidisciplinary care. Transurethral resection (TUR) can be done to excise the affected tissue, but unfortunately had a high recurrence rate [1,4].

Partial cystectomies, especially by laparoscopic means, is the most effective treatment for BE. This surgical procedure is excisional and consists of the removal of the entire bladder wall affected by endometriosis [1,5]. Due to overall rare cases, BE might pose a challenge to practising gynaecologist or urologist, both to correctly diagnose and delivering the right treatment. This article aims to share an experience in order to give an insight how a case of bladder endometriosis might present in the everyday clinical practice.

Case Description

A Thirty-one years old woman complained of painful, bloody stained urine, which occurs during menstruation in the last 18 months. The patient has been married for 18 months without any pregnancy. Previously patient had a regular menstruation cycle with moderate pain and normal menstrual volume. There is no history of any fever, urinary bladder stones, weight loss or other symptoms related to malignancy. Due to the complaint, the patient went to a gynaecologist.

The physical examination showing typical vital signs, abdominal inspection showing no distention, bowel sound was normal, no palpable mass, and no tenderness: genital inspection showing no vaginal discharge or bleeding and smooth cervix. The uterus was anteflexed normal-sized and no mass, pain, or nodule palpable during vaginal toucher.

An ultrasound scan was done and revealed a hyperechoic mass measured 2, 6 cm x 2, 6 cm with vascular supply protruded from posterior wall to the bladder lumen. In order to confirm the diagnosis, the patient consulted to a urologist and underwent cystoscopy showing adenomatous reddish nodular mass at the bladder wall measured 3 cm x 3cm, transurethral resection (TUR) was performed. The excised mass was a cystitis glandulare on pathological examination. After the surgery, the patient received a ciproterone acetate (Diane 35®) for three months.

During the treatment, the symptoms were slightly improved, accompanied by the absence of bloody urine. After three months, the patient reexperienced the previous symptoms. Then the patient underwent another ultrasound scan, which shows a mass on the bladder wall measured about 29.8 mm x 16.7 mm.

CT scan was conducted, and the result indicates a solid, isodense mass, with clear border, and irregular shape attached to the upper left portion of the bladder wall measured about 31.9 mm x 27.23 mm x 32.32 mm. The second cystoscopy with transurethral resection was conducted in order to excise all the lesion. The subsequent pathologist report was cystitis glandulare with interstitial metaplasia. Unfortunately, after the second TUR, there was no significant improvement achieved.

The patient underwent a third cystoscopy aimed to remove the residual mass and the third pathologic result, also showing a chronic cystitis glandulare. After three surgeries, the patient requests a second opinion and willing for more advanced management. After reviewing the history of treatment, a team of gynaecologist and urologist were set. The teams then manage to prepare a laparoscopic partial cystectomy with cystoscopy guidance to achieve complete excision of the lesion and conserve the remaining healthy bladder wall.

The surgery starts with a cystoscopy performed by the urologist to evaluate the interior lining of the bladder. The adenomatous reddish nodular lesion was found, and electrocautery was done to mark the lesion's border. DJ stents were applied to characterise the ureter orifice at the bladder dome to avoid injury. Simultaneously,

laparoscopy was prepared. A primary umbilical port, left upper and lower ancillary port, and the right ancillary port was placed, and then the laparoscopic procedure was started. Severe adhesion on the anterior part of uterus with bladder wall was found, both ovaries and fallopian tubes were normal, and no other endometriosis lesions were visible. Adhesiolysis was performed, starting with dissecting the bladder peritoneum and open the right and left paravesical space to separate the uterus from the bladder. The

bladder wall excision guided with the cystoscopic light to illuminate the lesion site. Once the bladder wall was exposed, the full thickness excision was continued using the previous electrocautery mark as an excision margin. The bladder wall then closed with two layers of suture. The abdominal drain was applied for 24 hours for urinary leakage monitoring. The urinary catheter was placed for seven days to avoid bladder distention and provide the appropriate condition for bladder wall healing.

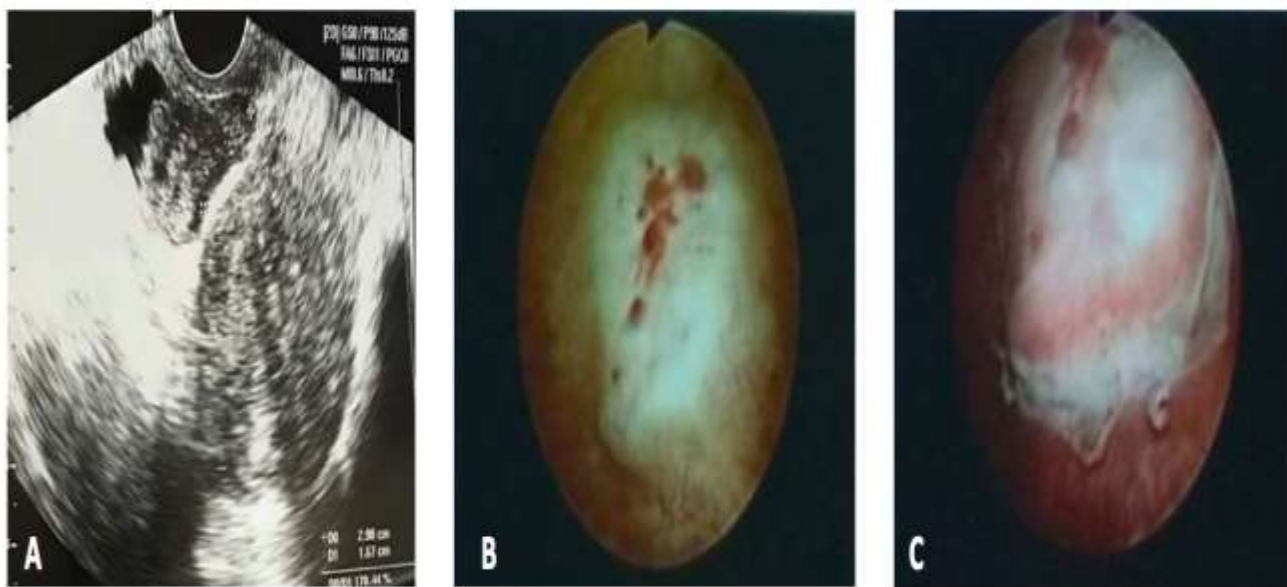


Figure 1: A) Ultrasound imaging of mass on the bladder wall; B) Cystoscopy finding of a mass lesion on the bladder wall; C) Electrocautery marks to delineate the lesion from surrounding healthy tissue

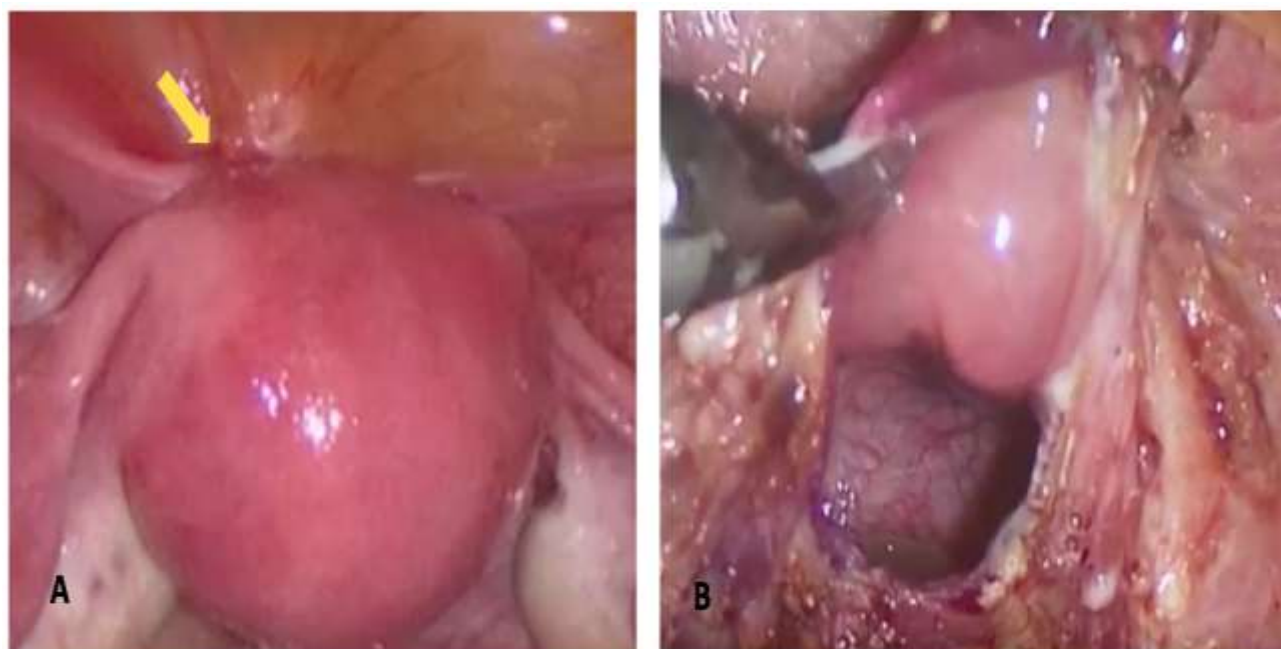


Figure 2: A) Endometriosis lesion on the bladder wall and adhesion with the uterus (yellow arrow); B) Partial cystectomy

The patient discharged the next day after the surgery without any complications, and the patient gains a normal voiding function after the urinary catheter was removed.

The histopathology result confirms the presence of endometrial glands and stromal tissue in the detrusor muscle, which was pathognomonic for bladder endometriosis.

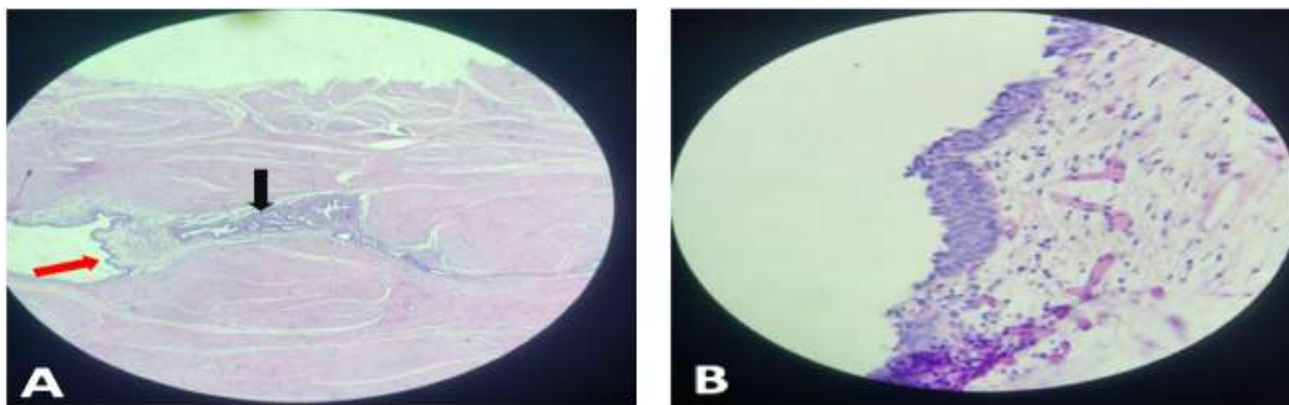


Figure 3: A) Endometrial stroma (black arrow) and gland (red arrow) between the detrusor smooth muscle layers; B) Bladder surface with transitional epithelium without atypia

Discussion

Bladder endometriosis may present with lower urinary tract symptoms and/or infertility. In our case, the patient complained about painful, bloody stained urine and dysuria, and the patient also had infertility issue that may be related to the endometriosis. Hematuria seems to be a less frequent symptom, being described by 20-35% of patients.

Menouria (i.e. hematuria coinciding with menstruation), appeared only in 20-25% of cases when the bladder mucosa is affected. About 40% of patients affected reported these symptoms occurring cyclically, with predominance during the premenstrual period. These symptoms are almost similar to other urological conditions, such as recurrent cystitis, overactive bladder, bladder carcinoma, interstitial cystitis and chronic urethral syndrome [2,3].

In the context about the symptoms of pain, several pathophysiological mechanisms might explain the relation between endometriosis and pelvic pain, which include recurrent cyclic micro-bleeding in the endometriosis lesions responsible for hyper pressure, production of inflammatory mediators by endometriosis lesions, which can stimulate the nerves, the adhesions responsible for fixed position of pelvic structures and compression and/or infiltration of the sub-peritoneal nerve fibers by deep implants [3].

Ultrasound could be taken as the first step in the diagnosis of BE, due to its low cost, ready availability, and lack of radiation exposure. It also can be used to evaluate the location and size of the lesion and estimate the distance between the lesion border and urethral

orifice [1]. As reported by different authors, the specificity and positive and negative predictive values of TVS for BE are about 100%. The sensitivity is 50%-62% because it related to the lesion size (>3cm) [1]. [2,6]. In our case, a transvaginal ultrasound (TVS) result is showing hyperechoic mass sized 2,6 cm x 2,6 cm with vascularisation. Cystoscopy remains one of the most cost-effective procedures to assess the interior lining of the bladder. Due to the intraperitoneal origin of the lesion, cystoscopy findings may be normal. Nevertheless, cystoscopy may demonstrate an intraluminal mass of the posterior bladder wall or dome [2].

However, in our case, three cystoscopies followed by TUR were performed, and the pathological report showed cystitis glandulare instead of BE. Another author also reports several cystoscopies for a patient for 17 years with cystitis on pathologic examination. Another case series reporting that cystoscopy (TUR) alone was not an adequate modality to diagnose and treat the BE [4, 7].

When the bladder mass or lesion has been documented by imaging techniques and/or cystoscopy, the differential diagnosis should include bladder carcinoma, angiomas, leiomyoma, amyloidosis, malakoplakia, glandular cystitis, nephrogenic adenoma and extravesical processes such as diverticulitis with histologic study necessary in almost all cases [2]. It should be considered that with the exception of transurethral resection (TUR) procedures, biopsy at cystoscopy is frequently not diagnostic for endometriosis.

Maggiore et al. recommended it that cystoscopy should not be routinely performed, except in cases of suspicion due to

malignancy or if the distance between the nodule and the ureteral orifices cannot be determined using TVS [1]. After no improvement of previous three TUR procedures, our patient underwent a laparoscopic partial cystectomy with concomitant cystoscopy that involves both a gynaecologist and urologist.

BE is challenging so that clinical management should include collaboration between gynaecologists and urologists [1]. Surgery is considered to be the gold standard in the treatment of patients with deep infiltrating endometriosis. Partial cystectomy is a bladder-preserving management procedure that involves complete surgical excision of the BE and surrounding bladder wall.

This procedure is relatively safe and straightforward, and the surgical outcome is excellent in terms of symptom improvement and recurrence. The procedure itself can be carried out with laparotomy or laparoscopy approach. Concomitant cystoscopy can be useful to delineate the margins of the endometriosis lesion to be resected [2].

According to the surgeon's preference, catheterisations of the ureters may be advisable, especially when the distance between the caudal border of the endometriosis lesion and the inter-ureteric ridge is <2 cm. The laparoscopic partial cystectomy offers the same results as open surgery with several advantages, including less bleeding, shortened operative time, shortened hospital stay, quicker return to

work, a major reduction in postoperative pain and decreased risk of postoperative morbidity [2]. As reported by other authors, complete excision with bladder conservation is the major goal of the surgery. Partial laparoscopic cystectomy with a modified light-to-light technique is a method that provides adequate identification of the lesion border via cystoscopy with excellent results in preserving healthy unaffected bladder wall [1, 2, 5].

Conclusion

Bladder endometriosis could be challenging both in diagnostic and treatment. Bladder endometriosis should be considered as the pathology behind cyclic hematuria. Cystoscopy and transurethral resection are not recommended routinely to diagnose and manage the lesions.

A multidisciplinary approach is needed to get adequate treatment. Concomitant Laparoscopy with cystoscopy for partial cystectomy might give an excellent outcome.

Funding

The authors are responsible for the funding of study without the involvement of grant, scholarship, sponsorship, or any other resource of funding.

Author Contribution

IBPA is responsible for the study concept, supervisor, and written manuscript. IMPA is responsible for developing the study concept finalize the manuscript.

References

1. Maggiore ULM FS, Candiani M, Somigliana E, Vigano P, Vercellini P (2017) Bladder Endometriosis: A Systematic Review of pathogenesis, Diagnosis, Treatment, Impact on Fertility, and Risk of Malignant Transformation. *Eur. Urol.*, 71(5):790-807.
2. Maccagnano C, Rocchini L, Ghezzi M, Scattoni V, Montorsi F, Rigatti P, Colombo R (2012) Diagnosis and Treatment of Bladder Endometriosis: State of the Art. *Urol. Int.*, 89: 249-58.
3. Chapron C BA, Chopin N, Dousset B, Leconte M, Amsellem-Ouazana D, de Ziegler D, Borghese D (2010) Surgery for bladder endometriosis: long-term results and concomitant management of associated posterior deep lesions. *Human Reproduction*, 25(4):884-9.
4. Le Tohic A CC, Yazbeck C, Koskas M, Madelenat P, Panel P (2009) Bladder endometriosis: diagnosis and treatment. A series of 24 patients. *Gynecologie, Obstetrique & Fertilité*, 37(3):216-21.
5. Stopiglia RM F, Faundes DG, Petta CA (2017) Cystoscopy-assisted laparoscopy for bladder endometriosis: modified light-to-light technique for bladder preservation. *Int. Braz. J. Urol.*, 43(1):87-94.
6. Guerriero S AS, Minguez JA, Jurado M, Mais V, Melis GB, Alcazar JL (2015) Accuracy of transvaginal ultrasound for diagnosis of deep endometriosis in uterosacral ligaments,

rectovaginal septum, vagina and bladder: systematic review and meta-analysis. *Ultrasound Obstet. Gynecol.*, 46:534-45.

7. Kotarski J M-PM, Bobinski M, Kotarski J (2017) The endometriosis of urinary tract. *Archives of Perinatal Medicine*, 23(2):67-9.