

Suburethral Endometriosis as Clinical Finding of Extensive Disease

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ABSTRACT

Objective: Endometriosis spreading to the vagina is rare, present in only 0.02% of women with symptomatic endometriosis. Suburethral lesion site is exceptional. In an extensive literature review only 4 cases of suburethral endometriosis were identified. Our objective is to present a case of primary vaginal suburethral endometriosis in a 31-year old patient who underwent laparoscopic evaluation and to perform a literature review on this topic.

Methods and Procedures: Case report presentation based on information extracted from patient database. A review of literature with a Medline search using key words urethral endometriosis, suburethral endometriosis, or urethral diverticulum was undertaken.

Results: This case report describes a case of a 31-year old female patient referred for severe pelvic pain, worsening during menstruation. On physical examination a 2 cm suburethral endometriotic lesion was found as the initial presentation. Her examination was also significant for enlarged, tender uterus and adnexa. Based on examination and imaging, adenomyosis and endometriosis were

suspected. Surgical evaluation revealed extensive endometriosis with lymph node involvement at laparoscopic exploration. The review of literature revealed only 4 cases where suburethral endometriosis was previously identified.

Conclusion: Primary vaginal suburethral endometriosis, although rare, could be an indication of extensive endometriosis. This case highlights the importance of careful clinical examination, surgical excision, and laparoscopic evaluation when identifying suburethral vaginal endometriotic lesions.

Key Words: Endometriosis, Laparoscopy, Suburethral, Surgical management.

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INTRODUCTION

Endometriosis is defined as the presence of endometrial glands and stroma in ectopic locations. Affecting from 6% to 10% of reproductive aged women, endometriosis may result in dysmenorrhea, dyspareunia, chronic pelvic pain, and/or subfertility.¹ Endometriosis has also been described in virtually every location that can be reached by hematogenous, lymphatic, or direct dissemination.² With increasing anatomical distance from the uterus, endometriotic lesions have the tendency to lose hormonal receptors, and appear to be asynchronous when compared to endometrium.² Ectopic endometrium produces symptoms that are cyclic only in 50% of patients, and many distant lesions do not respond to hormonal stimulation.² Urinary tract endometriosis (UTE) may be considered a rare condition affecting 1% to 2.5% of women with symptomatic endometriosis.²⁻⁵ Only 4 cases of suburethral endometriosis have been reported. We report a case of suburethral endometriosis as the initial presentation in a patient with no prior surgical history.

CASE REPORT

A 31-year-old African American woman, G5P1, with a history of normal vaginal delivery eight years prior to her visit, was referred for severe pelvic pain, worse during menstruation. The patient had been evaluated by a reproductive endocrinologist six months prior with pelvic magnetic resonance imaging



Figure 1. Suburethral lesion, approximately 2 Centimeters In diameter as seen during initial consultation.

for myoma that showed a large area of diffuse adenomyosis involving the mid and upper uterine body and fundal region. She was seen in the Emergency Department one week prior to consultation, and pelvic sonogram revealed heterogeneity of the uterus without focal mass. Neither examiner reported evidence of a urethral lesion. She was otherwise healthy with no prior surgery or significant medical history.

On physical examination during her initial consultation at our center, an approximately 2 cm in diameter bluish cystic-appearing lesion was noted on the urethra and was tender (**Figure 1**). There was no evidence or history of trauma, and clinically this appeared to be endometriosis. Her examination was also significant for enlarged tender uterus and adnexa. Based on the examination and imaging, adenomyosis and endometriosis was suspected.

The patient desired to preserve her reproductive system and elected to have surgery. She underwent video-assisted hysterectomy and laparoscopy with multipuncture technique which has been described before.⁶ Intraoperative hysteroscopic findings on day 21 of her menstrual cycle included external fundal compression of intramural mass without cavity distortion and multiple areas of bluish glandular pattern suggestive of adenomyosis. (**Figure 2**).

Videolaparoscopic findings revealed abnormal uterine shape and contour. The uterus had posterior and fundal wall asymmetry, which upon exploration contained no capsule and was compatible with severe muscular hypertrophy and adenomyosis confirmed by frozen section. (**Figure 3**)

Both fallopian tubes appeared to be normal and patent with injection of indigo carmine. The ovaries had cortical endometriosis and filmy adhesions. There were scattered endometriotic lesions along the pelvic sidewalls and anterior and posterior cul-de-sac, which had slight retroperitoneal fibroadipose tissue penetration. (**Figure 4**) There was no evidence of deep infiltration to the retroperitoneal structures.

Treatment of endometriosis was performed with pelvic sidewall and rectovaginal dissection with vaporization and excision of endometriotic lesions and restoration of the anatomy. There was one enlarged presacral lymph node that was excised and was positive for endometriosis on frozen section (**Figures 5 and 6**). A wedge cytoreductive resection of fundal adenomyotic lesion was performed without cavity penetration. The uterus was reconstructed in multiple layers with absorbable polyglactin 910 sutures.

The cystourethroscopic examination revealed no evidence of mucosal involvement of the urethra and no evidence of bladder endometriosis. The suburethral lesion was sharply excised and revealed chocolate like material and a fibrotic cyst wall to the paraurethral tissue but no urethral muscularis invasion (**Figure 7**). Transurethral Foley catheter was kept in situ to facilitate identification of the anatomy and sharp dissection and excision of the lesion. Care was taken to excise surrounding healthy margins. The excision site was approximated with 4-0 interrupted polyglycolic suture. Repeat cystourethroscopic examination confirmed intact urethra and normal findings. A proctosigmoidoscopic examination with laparoscopic monitoring confirmed no evidence of bowel disease, restoration of the anatomy and site of dissection to be involved with rectovaginal and pararectal fibroadipose tissue.

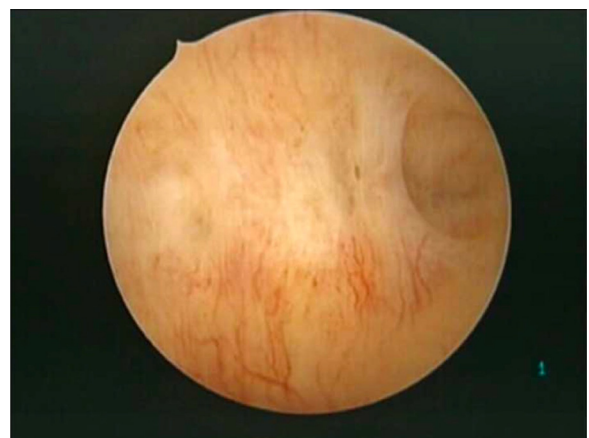


Figure 2. Videohysteroscopic view of bluish discoloration within the myometrial wall.



Figure 3. Videolaparoscopic view of uterine fundus with area of muscular hypertrophy and adenomyosis.

Postoperative bladder drainage was continued with the Foley catheter for 24 hours. The patient was observed overnight and upon discontinuation of the Foley she had spontaneous voiding and was discharged with oral pain medication. At both two and six-week postoperative visits she had progressive recovery, and the site of suburethral excision had healed. (**Figure 8**) The postoperative course was uneventful.

DISCUSSION

The etiology of endometriosis is unknown, although there are several theories aimed at explaining the pathogenesis of the disease. These theories include endometrial and nonendometrial origin. Sampson's theory, proposed in 1920, is based on retrograde menstruation through the fallopian tubes into the peritoneal cavity.¹ This is a widely accepted theory, supported by anatomic distribution of endometriotic lesions. Also proposed in the 1920s is the

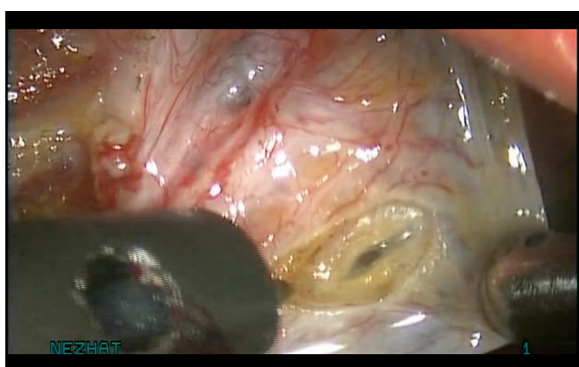


Figure 4. Scattered endometriotic lesions along rectovaginal septum and pelvic side wall.

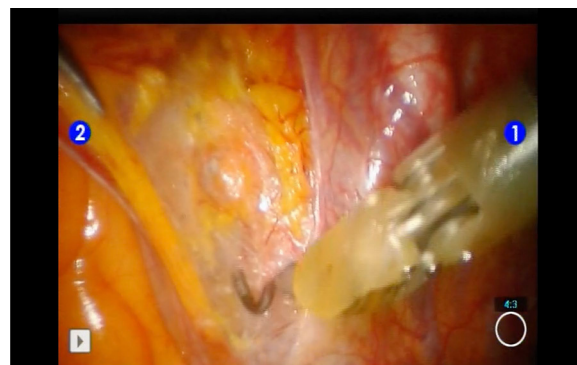


Figure 5. View of presacral space dissection and enlarged lymph node.

theory of benign metastasis through lymphatic or hemogenous spread.¹ Support for lymphatic spread includes presence of endometriosis within 6.5% of women at the time of lymphadenectomy and 6.7% of women at autopsy.¹ Nonendometrial theory includes coelomic metaplasia, in which normal peritoneal tissue transforms via metaplastic transition to ectopic endometrial tissue.¹

The embryonic müllerian rests theory, in which müllerian cells maintain the capacity to develop into endometriotic lesions when influenced by estrogen, is supported by the presence of endometriosis in Rokitansky-Kuster-Huster-Hauser syndrome patients and men with prostate cancer who were exposed to high dose estrogen and were subsequently diagnosed with endometriosis.¹

Endometriotic lesions are usually found within the pelvis; however, extrapelvic sites can be the result of any of these three theories. A recent systematic review reports rates of concomitant or associated pelvic endometriosis ranging from 18% to up to



Figure 6. Microscopic view of lymph node with endometriotic glands within.

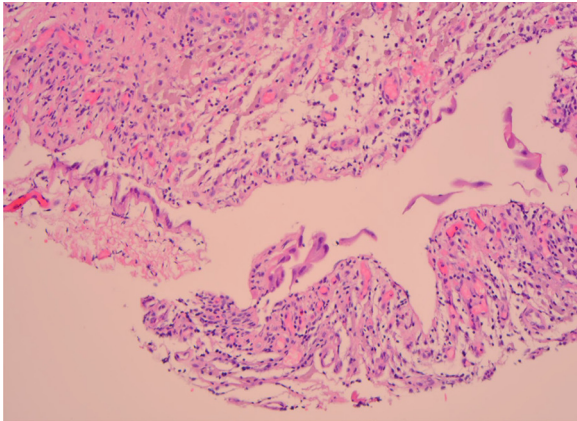


Figure 7. Microscopic view of suburethral lesion with areas of degeneration and endometriotic stroma.

62% in cases of diagnosed extrapelvic endometriosis.⁷ Although an evidence-based approach is lacking, high correlation rates have led several experts to recommend thorough pelvic survey when encountering abdominal extrapelvic endometriosis.⁷

Urinary tract endometriosis can cause loss of renal function in 5%–30% of women with endometriosis making early diagnosis critical.^{2,8} The most common site affected by UTE is the bladder (80% – 84%), followed by the ureter (15%). Left sided ureteral lesions are present more frequently (64%) and the bilateral incidence is 15.2%, kidney (4%), and urethra



Figure 8. Site of lesion at two weeks post-operative visit.

(2%).^{3,9,10} When considering urinary involvement, radiological imaging of the ureter, including intravenous pyelogram (IVP), retrograde pyelogram, or computed tomography IVP may be useful pre-operative tools.² Histologic diagnosis not only confirms endometriosis, it can also identify malignant lesions. In 2002, we reported one instance of malignancy in 15 patients who had bladder resection for endometriosis.³ Vaginal endometriosis accounts for 0.02% of cases,¹¹ and usually present with intrinsic lesions, which can be easily seen on speculum examination and are usually found in the posterior fornix. The presence of vaginal or vulvar endometriotic lesions usually represents a secondary manifestation of pelvic disease.¹²

Suburethral location is exceptional. A review of literature with a Medline search using key words urethral endometriosis, suburethral endometriosis, or urethral diverticulum revealed only 4 cases of suburethral endometriosis previously identified. Cabral Ribeiro et al.⁹ reported a case of 3 – 4 cm size lesion in the anterior vaginal introitus causing voiding dysfunction. The lesion was resected vaginally, and the patient had some urinary incontinence post procedure. Wu et al.¹³ reported a case involving a patient with pain and voiding difficulties, who had an echolucent mass over the suburethral area measuring 3.7 cm in diameter diagnosed with ultrasound. Complete excision was performed with pathologic confirmation of endometriosis. Nelson¹¹ reported a case of 3 × 3 cm suburethral cyst causing discomfort with walking, dyspareunia, and urinary frequency. The lesion was excised and histologic evaluation was consistent with endometriotic cyst. Ewadh, et al.¹⁴ reported a case of a recurrent suburethral 3 × 3 cm cyst causing discomfort on sitting and dyspareunia. Surgical excision was performed and endometriosis was confirmed on pathology report. None of the previously reported cases informed concomitant video laparoscopic pelvic evaluation. The clinical manifestations presented in these cases are similar to diverse entities that present with cystic lesions involving the vagina and urethra. Differential diagnoses of anterior cystic vaginal lesions include Müllerian cyst, Gardner's cyst, Skene's and Bartholin's gland cyst, and urethral diverticulum.¹² These must be considered upon initial examination and a histologic evaluation is required for a definitive diagnosis.

Our case report of suburethral endometriosis is unique due to her thorough surgical exploration and proven extensive pelvic involvement. The goal of conservative operative treatment of endometriosis is to remove all implants, resect

adhesions, relieve pain, and restore involved organs to normal anatomic and physiologic condition. We report the fifth case of suburethral endometriosis in the literature. Additional unique findings of our patient are her widespread endometriosis and adenomyosis. In spite of advancements in imaging techniques, a thorough physical examination remains of paramount importance in evaluation and treatment of patients suspicious for endometriosis. Although further research is required, primary vaginal suburethral endometriosis, though rare, could be an indication of extensive endometriosis. Careful clinical examination and surgical excision is diagnostic and therapeutic.

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