

An Unusual Presentation of Sub Urethral Endometriosis: A Case Report

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ARTICLE INFO	ABSTRACT
Article type: Case report	Background & aim: Endometriosis is defined as the presence of endometrial tissue outside the uterus. The most frequent sites of implantation are pelvic viscera and the peritoneum. Extra pelvic endometriosis is a rare phenomenon. The aim of this study was to introduce a rare case with unusual presentation of sub urethral endometriosis.
Article History: Received: 07-May-2023 Accepted: 30-Dec-2023	Case report: A 26-year-old woman, gravida 2, parity 2 with eight months history of vaginal bulging and dyspareunia presented to the pelvic floor clinic of a training hospital in Mashhad, Iran. Physical examination revealed a tender cystic mass measuring 2×2cm at the anterior vaginal wall just below the urethral opening. Vaginal ultrasonography revealed a cystic lesion with size of 25×15mm in the anterior vaginal wall. Surgical incision performed and brownish sticky fluid was drained from the cyst. Cystoscopic assessment confirmed an intact urethra and bladder mucosa. Histopathological evaluation was compatible with endometriosis. The patient was symptom free at sixth month postoperative follow up visit.
Key words: Endometriosis Diverticulum Mass Dyspareunia Tenderness	Conclusion: Endometriosis should be considered in patients with painful vaginal lesion. Complete surgical excision is an acceptable management in these cases.

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Introduction

Endometriosis is defined as the presence of endometrial tissue outside the uterus. The most frequent sites of implantation are pelvic viscera and the peritoneum including ovary and ovarian fossa, uterosacral ligaments, pouch of Douglas (1).

Extra pelvic endometriosis is a rare event. Colorectal and urinary tract involvement are the most common sites of extra pelvic endometriosis. Less common sites include abdominal wall, omentum, perineum, urinary system (ureters, bladder and urethra), cervix and vaginal area which accounted for 0.02% of

cases (2). Most cases of extra pelvic endometriosis are presented to specialties other than gynecologists. The diagnosis of both gastrointestinal and urinary tract endometriosis is often delayed due to the atypical and nonspecific symptoms (3). A definitive diagnosis of endometriosis relies on histological assessment following tissue biopsy or excision (4). The surgical diagnosis of endometriosis has been the gold standard. This study introduces a rare case with unusual presentation of sub urethral endometriosis.

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Case presentation

A 26-year-old woman gravida 2, parity 2 with eight months history of vaginal bulging and dyspareunia presented to the pelvic floor clinic of a training hospital of Mashhad University of Medical Sciences, Mashhad, Iran. She had no concomitant urinary or defecatory symptoms. The menstrual cycles were regular without any history of dysmenorrhea or peri menstrual pelvic pain. She did not use any contraceptive method. She had two normal vaginal deliveries without any past medical history. Physical examination revealed normal vital sign, a tender cystic mass measuring 2×2cm arising from the anterior vaginal wall just below the urethral opening. The mass was very tender, but there was not any urethral discharge after the mass compression (Figure 1).

Figure 1. An anterior vaginal wall mass presented as a sub urethral bulging

Urine analysis and complete blood count (CBC) were normal. Vaginal ultrasonography revealed a cystic lesion with 25×15mm in size located in the anterior vaginal wall. The surgical excision was planned due to severe dyspareunia and tenderness.

In the operation room, after general anesthesia, urethrocytoscopy was performed to evaluate any diverticular opening or another abnormality, but urethra and bladder were completely normal. A small 1 cm vertical incision was made over the cyst wall. At the time of cyst wall dissection from the vaginal epithelium, the cyst ruptured and efflux of

chocolate fluid happened. The entire cyst wall was resected as much as possible and sent for pathological assessment. Cystoscopic evaluation was again performed which confirmed an intact urethra and bladder mucosa. Copious irrigation was done and the anterior vaginal epithelium was closed by continuous 2-0 vicryl suture.

Histological evaluation revealed endometrial glands with hemosiderin macrophages, which was consistent with endometriosis (Figure 2).

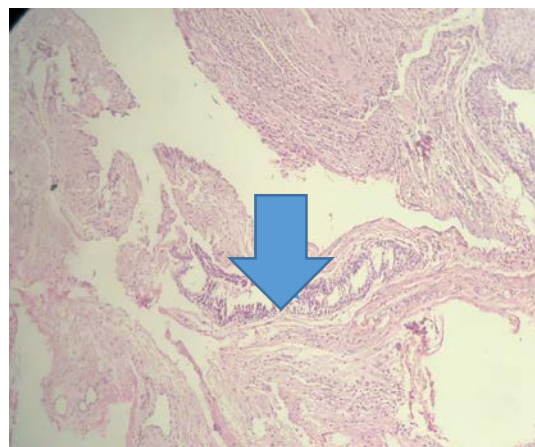


Figure 2 (a). Sections of duct wall with endometrial glands, hematoxylin eosin staining × 100 (thick blue arrow)

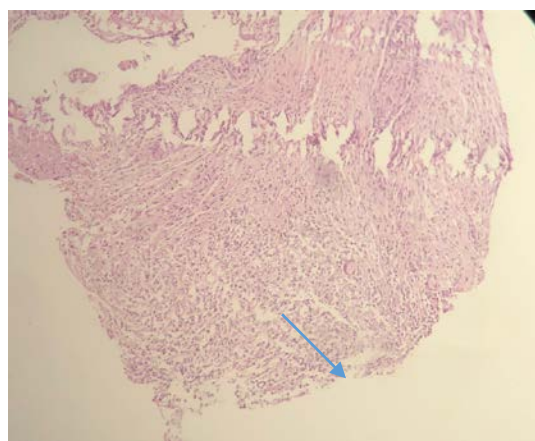


Figure 2 (b). Focal points of hemosiderin macrophages in the duct wall, hematoxylin eosin staining × 100 (thin blue arrow)

The follow-up visits at 2 weeks and then 6 weeks after the surgery showed complete

wound healing without any urinary or bothersome symptoms.

Discussion

Extrapelvic endometriosis is infrequent and can appear in unusual sites. It is often difficult to diagnose and more challenging to treat (5). Few cases of endometriosis in the vagina have been described, actually endometriosis limited only to the vagina in the absence of acute or chronic endometriosis symptoms is a rare entity. It may present as the isolated vaginal endometriotic lesions which commonly located in the upper third of the vagina and the posterior fornix (6), but in this case the lesion was in the lower third of the anterior vaginal wall.

Differential diagnosis of this rare suburethral location is suburethral diverticulum or gartner duct cyst (6).

Similar to the patient described here, another report revealed a vaginal endometrioma in a 43-year-old patient, but she had no other symptoms of endometriosis, which was in contrast with this case which has severe dyspareunia (7). Clinical impression preoperatively was most consistent with urethral diverticulum, whereas US imaging suggested Gartner's duct cyst (7), which again was as similar as the current case.

The most severe form of endometriosis and deep infiltrating endometriosis (DIE), can manifest as the lateral parametrial endometriosis and can involve the rectovaginal septum, vagina, rectum and ureter. This type can lead to severe pain which may require aggressive surgery (8). The finding of study by Foti et al. (2018) was in contrast with the current case because she had no other symptoms of DIE.

The proposed etiology in the case of vaginal endometriosis is its development in Mullerian remnants that persisted in vaginal wall (9). Another theory is that trauma from vaginal delivery may contribute to its pathogenesis. The case of episiotomy scar endometrioma supported the "iatrogenic implantation theory", but in our case, the mass was located in the distal part of anterior vaginal wall which was away from the episiotomy scar (10). In a patient with endometrioma, excision of the entire cyst wall is important for both diagnosis and treatment. This is recommended in all cases with persistent or symptomatic vaginal lesions

in order to prevent recurrence and malignant transformation, which is rare but possible event (10).

Based on experiences with the current patient and another case of anterior vaginal wall mass in the literature, the treatment of vaginal endometriomas with definitive surgical management is an acceptable approach both for accurate treatment and definite diagnosis (6). Locally drainage is another option but may cause more recurrence (6). The diagnosis of endometriosis is surprising when it is found outside the peritoneal cavity and is challenging without histology confirmation (10). As these mass may present with atypical symptoms of endometriosis, diagnosis is based on surgical excision and histopathological examination especially in older age that the vaginal mass may be the only symptom of adenocarcinoma. This affirms the importance of histological and immunocytochemical exams in vulvar and vaginal neoplasms for accurate diagnosis. Postoperative follow-up is essential to rule out any recurrence.

Conclusion

Typical locations of endometriosis are within the pelvis, but rarely a clinician may encounter with endometriosis and endometriomas in the vagina. This case highlights an uncommon presentation of an endometrioma. It is notable that treatment of choice in the case of vaginal endometriomas is surgical excision. Postoperative follow-up is also essential to rule out recurrence.

Declarations

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Conflicts of interest

The authors declared no conflicts of interest.

Ethical considerations

Informed consent was obtained from the patient to publish the information and related images.

Code of ethics

None.

Use of Artificial Intelligence (AI)

None.

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Authors' contribution

LP contributed to conceptualization, data collection and supervising the report preparation. AHJ performed pathological examination and interpretation. AV, MKh, and SA helped in data collection and writing the manuscript. All authors read and approved the final version of the article.

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