



# An asymptomatic anterior vaginal wall endometrioma, a rare manifestation of endometriosis: A case report

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## ARTICLE INFO

### Article history:

Received 10 April 2020

Received in revised form 21 April 2020

Accepted 22 April 2020

Available online xxx

### Keywords:

Endometriosis

Endometrioma

Vagina

## ABSTRACT

A 23-year-old patient with an asymptomatic anterior vaginal wall cyst was referred to gynecology for evaluation and treatment. Preoperative assessment with physical examination and magnetic resonance (MR) imaging of the pelvis was most consistent with Gartner's cyst. Following resection of the cyst wall, histologic evaluation demonstrated endometrial glands, hemosiderin-laden macrophages and inflammation, consistent with vaginal wall endometrioma. This case highlights challenges in the diagnosis of endometriosis in the vagina and in other rare locations, possible mechanisms of development, and proposed treatments.

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## 1. Introduction

Normal endometrium consists of endometrial glands and stroma that undergo a series of changes throughout the menstrual cycle. Endometriosis occurs when endometrial-like tissue is located outside of the uterus [1,2]. Endometriosis affects approximately 10% of women of reproductive age; since definitive diagnosis requires surgical visualization, the true prevalence is unknown [1]. The condition frequently causes pelvic pain and infertility. Symptoms can also include dysmenorrhea, dyspareunia, dysuria, dyschezia and fatigue, affecting patient well-being and productivity [1].

Several theories describe the pathogenesis of endometriosis. Sampson first used the word “endometriosis” in 1927 to describe endometrium-like tissue discovered in the myometrium, rectovaginal septum and hemorrhagic-appearing “chocolate” ovarian cysts [3,4]. He cited “menstrual dissemination...of endometrial tissue...transferred to situations favorable to its existence” as the source [3]. The prevailing notion of its origin is Sampson's hypothesis, often termed “retrograde menstruation,” but this does not explain all clinical observations.

Theories that endometriosis arises from mesothelial cell metaplasia and from genetic and epigenetic defects transmitted at birth have emerged more recently, and can explain endometriosis located outside of the pelvis. As Koninckx *et al.* described, hematologic spread and lymphatic spread of endometrial tissue have additionally been implicated as possible causes [4].

Typical locations of endometriosis deposits are within the pelvis (ovaries, pouch of Douglas, broad ligaments, round and uterosacral ligaments, uterus, fallopian tubes and bowel), but implants can occur widely throughout the body [1]. Rarely does a clinician encounter endometriosis and endometriomas in the vagina. This case highlights an uncommon chief complaint and presentation of an endometrioma, and discusses possible mechanisms of endometriosis development in this unique setting.

## 2. Case Presentation

### 2.1. Patient Details

A 23-year-old woman presented with a vaginal cyst identified three months earlier at another institution. She reported inability to “get a good seal” with a menstrual cup and an occasional pinching sensation during vaginal intercourse. She denied any pain (including cyclic pain) or bleeding from the site.

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She experienced menarche at age 13 followed by regular monthly menses. She denied having dysmenorrhea, pelvic pain or sexually transmitted infections (STIs). She was gravida 0 and used NuvaRing for contraception. She had tested positive for human papilloma virus (HPV) on a previous pap test, but this was followed expectantly without intervention, given her young age.

## 2.2. Initial Assessment

Physical exam at preoperative visit revealed a nontender cyst measuring 2 centimeters (cm) arising from the right anterior vaginal wall. Given concerns about the cyst's proximity to the urethra, she underwent magnetic resonance (MR) imaging of the pelvis prior to procedure to rule out urethral involvement.

MR of the pelvis revealed a  $2.0 \times 1.2 \times 1.5$  cm ovoid circumscribed non-enhancing lesion in the mid-upper vagina, with intrinsically intense T1 hyper-intensity (Fig. 1A) and minimal/slight T2 hyper-intensity (not simple fluid T2 signal) (Fig. 1B) and without any enhancement on post-contrast sequences (Fig. 1C), consistent with a debris-filled benign cyst. The radiologist's initial interpretation offered differential diagnoses for a benign thin-walled debris-filled cyst in this location, including Bartholin's cyst, Gartner's cyst, or, less likely, urethral diverticulum. Based on location and morphology, a Gartner's cyst was considered highest on the differential. A Bartholin's cyst was considered less likely, as Bartholin's cysts are usually seen in the lower vagina. A urethral diverticulum was also considered less likely because the lesion did not appear intimately adjacent to the urethra and did not have the typical "U"-shaped configuration of a urethral diverticulum.

## 2.3. Treatment

The patient proceeded to the operating room for examination under anesthesia and biopsy. The fluctuant mass was palpated at the midline vagina, inferior to the urethra. Given concern for urethral involvement, a cystoscopy was performed with no urethral lesions or openings seen. Incision of the vaginal cyst revealed efflux of "chocolate fluid," consistent with endometrioma. The cyst wall was excised; the cyst cavity was then copiously irrigated and reapproximated using Vicryl suture. Cystoscopy at the end of the procedure showed no lesions in or trauma to the urethra.

On histology, the vaginal cyst biopsy revealed endometrial glands (Fig. 2A), diffuse hemosiderin-laden macrophages and fibrosis (Fig. 2B), consistent with endometriosis. There was no evidence of dysplasia or malignancy.

## 2.4. Outcome

She recovered well and resumed use of the NuvaRing a few days prior to her postoperative visit. Physical examination one month postoperatively revealed that the anterior vagina was healing well at the site of excision. The sutures were dissolving and there were no palpable defects or tenderness.

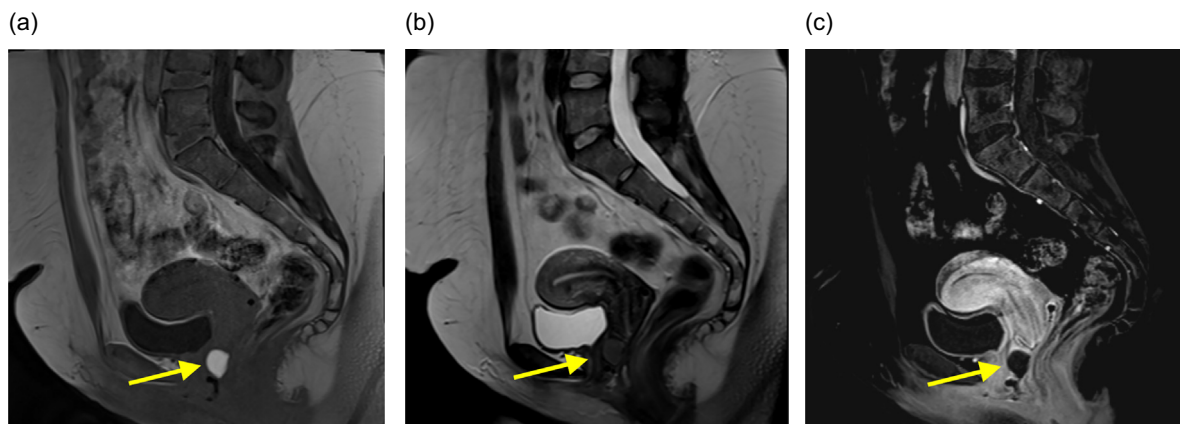
## 3. Discussion

This case highlights endometrioma as an unexpected etiology of a vaginal cyst. The diagnosis of endometriosis is surprising when it is found outside the walls of the peritoneal cavity and is challenging without histology confirmation [2]. Endometriosis can "masquerade" as several other conditions, often leading to missed or delayed diagnosis [6].

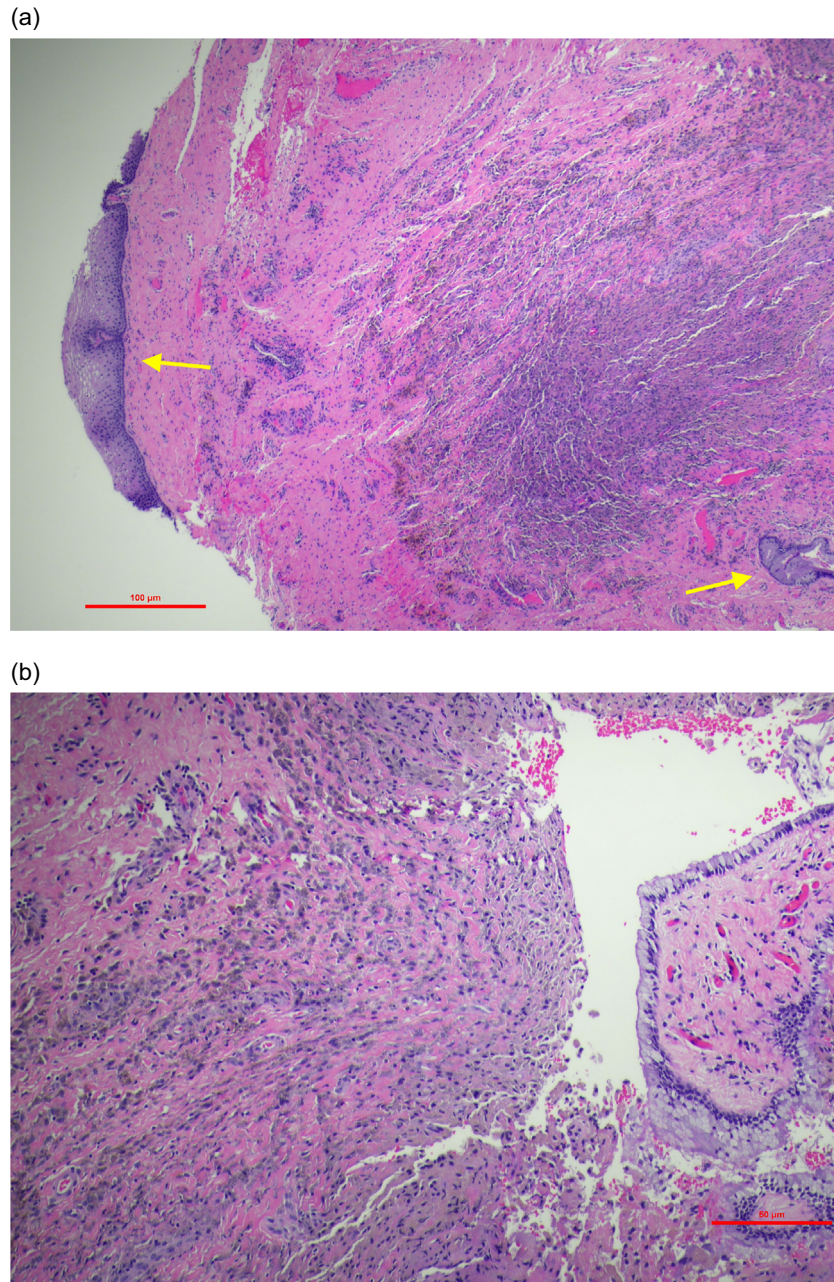
Few cases of endometriosis in the vagina have been described. Review of the literature demonstrated endometriosis in an episiotomy scar with a tender nodule and cyclic pain [7]. Similar to the patient described here, another report revealed a vaginal endometrioma in a 43-year-old patient who had no other symptoms of endometriosis [2]. Clinical impression preoperatively was most consistent with urethral diverticulum, whereas MR and US imaging suggested Gartner's duct cyst [2]. She remained asymptomatic three months postoperatively [2].

Endometriosis can develop in areas of the body as diverse as the pericardium and surgical scars, among others [5,6,8,9]. A case with clinical features and cardiac MR images highly suggestive of pericardial endometriosis was described in a 42-year-old woman with chronic catamenial chest pain and normal physical exam [8]. Another report portrayed a 20 cm abdominal wall mass in a patient's laparotomy scar after myomectomy [6]. One case illuminated cutaneous endometriosis of the eyelid, and an additional example showed endometriosis in a nasal mucosa nodule [5,9].

MR imaging is a useful modality preoperatively to diagnose, to assess local extent and to rule out coexisting endometriosis at other pelvic sites [7]. In this case, the diagnosis was not suspected on preoperative MR imaging. Retrospectively, the MR features are completely concordant with the pathologic diagnosis of an endometrioma. Blood products on MR imaging are usually T1 hyper-intense, and although they can have variable T2 features, blood products always have less T2 signal than simple fluid (i.e. relatively T2 hypo-intense compared to a cyst filled with simple fluid). Endometriomas are unilocular thin-walled cysts, which repetitively bleed into themselves with the menstrual cycle. As a result, these lesions are always filled with hemorrhagic debris, and therefore always demonstrate characteristic T1 hyper-intensity and relative T2 hypo-intensity. The MR findings of this patient's vaginal lesion detailed



**Fig. 1.** Sagittal MR sequences demonstrate an ovoid circumscribed non-enhancing lesion in the upper vagina – with intrinsic intense T1 hyper-intensity typical of a debris-filled cyst. 1A. T1 weighted sequence demonstrates intense hyper-intensity within the lesion. 1B. T2 weighted sequence demonstrates only slight T2 hyper-intensity (nearly iso-intense to the adjacent soft tissues) within the lesion. 1C. T1 weighted post contrast subtraction sequence demonstrates the lack of enhancement within the lesion, confirming it as likely a benign cyst.



**Fig. 2.** A. This image shows endometrial glands at the 3 o'clock position. The vaginal mucosa is at the 9 o'clock position. B. The H&E images show a collection of hemosiderin-laden macrophages admixed with glands and inflammation.

in Fig. 1 are classic MR features of an endometrioma. However, the diagnosis of an endometrioma was not considered here, given the very atypical location. Imaging diagnosis of an endometrioma would have been made were it not for the location of this lesion; i.e. such MR features would be diagnostic of an endometrioma if seen in the ovary.

While this patient was asymptomatic and her disease was undeniably mild, other forms of endometriosis in the vagina can cause pain or bleeding. The most severe form of endometriosis, deep infiltrating endometriosis (DIE), can manifest as lateral parametrial endometriosis (LPE) and can involve the rectovaginal septum, vagina, rectum and ureter. DIE and LPE lead to severe pain and require aggressive surgery [10]. For another patient discussed in the literature, endometriosis of the cervix caused massive vaginal hemorrhage and persistent postcoital

bleeding; cervical conization was effective treatment [11]. Many patients with cervical endometriosis were asymptomatic and diagnosed by histopathology incidentally [11]. Endometriosis has also appeared in a patient with cyst of canal of Nuck who presented with swelling and discomfort in the right groin [12]. This affirms the importance of histological and immunocytochemical exams in vulvar and vaginal neoplasms for accurate diagnosis [12].

The proposed etiology of the case of cervical endometriosis is its development in Mullerian rests that persisted in cervical stroma [11]. Another theory is that trauma from cone biopsy, curettage or electrocautery contributed to its pathogenesis [11]. The case of episiotomy scar endometrioma supported the "iatrogenic implantation theory" – namely, that endometriosis developed through mechanical



transposition during surgery [7]. Like in the case of cervical endometriosis, it is possible that this patient's endometrioma developed in persistent Mullerian tissue in the upper one-third of the vagina. It is also imaginable that long-term use of either the NuvaRing and/or menstrual cups contributed to development of an endometrioma in the vagina, especially if the foreign bodies caused local tissue injury and if endometrial cells from menstrual blood were retained in the vaginal wall.

In this patient, and with endometriomas in general, excision of the entire cyst wall is important for both diagnosis and treatment. This should be performed in all cases to prevent recurrence and malignant transformation, which is rare but possible [7,13]. Studies dating back to Dr. Sampson's work have described ovarian cancer arising from endometriosis [13]. Higher risk of malignant transformation exists in endometriosis patients with longstanding histories of endometriosis, diagnoses of endometriosis at an early age, histories of infertility and/or infertility treatment, and ovarian endometriomas [13].

Medical treatment of endometriosis involves hormonal therapy with the goal of decreasing local estrogen production [1]. This can then inhibit tissue proliferation and local inflammation at endometriotic implants [1]. In clinical practice, oral contraceptive pills (both combined or progestin only) are used frequently as they can serve as a treatment for dysmenorrhea and chronic pelvic pain in patients with endometriosis [1]. For future control of endometriosis, we recommend that this patient continues medical management with NuvaRing (a vaginal ring containing ethinyl estradiol and etonogestrel) or change to progestin-only contraception, as daily or depot progestins are also effective [1]. Although one-third of women with endometriosis have infertility [1], we predict that the infertility risk is low for this patient with an isolated vaginal endometrioma in the vagina and with no symptoms of pelvic pain or pelvic adhesive disease.

Based on experiences with this patient and another instance of anterior vaginal wall mass in the literature, we encourage the treatment of vaginal endometriomas with definitive surgical management [2] with or without medical management. Treatment of choice in the case of perineal scar endometriosis was surgical excision with wide margins [7]. Postoperative follow-up is essential to rule out recurrence [7]. This patient was asymptomatic, had no other evidence of pelvic endometriosis, and resumed use of NuvaRing postoperatively. Therefore, we are optimistic about her recovery, fertility potential and future evolution of symptoms.

### Contributors

Elizabeth A Dilday is the primary author and was the primary provider for the patient described, conducted the review of the literature and wrote the manuscript.

Michael S Lewis provided histology slides and descriptions of images.

Kiarash Vahidi provided patient MR images and descriptions of images.

Sanaz Memarzadeh is the anchor author and was the attending physician for the operating room case, provided guidance and edited the manuscript.

### Conflict of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

### Funding

No funding from an external source supported the publication of this case report.

### Patient consent

Obtained.

### Provenance and peer review

This case report was peer reviewed.

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