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Case Report

Vaginal cuff endometriosis after laparoscopic-assisted vaginal hysterectomy: a case report and literature review

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Abstract: Background: Endometriosis of the vaginal cuff after hysterectomy is extremely rare. Here we present a case of endometriosis at the vaginal cuff after laparoscopic-assisted vaginal hysterectomy (LAVH). Case: A 41-year-old woman was admitted complaining of sudden massive vaginal bleeding. The patient had a history of hysterectomy 3 years prior due to symptomatic multiple leiomyomas and adenomyosis. A lesion was found at her vaginal cuff, which infiltrated the serous membrane of the rectum and spontaneously ruptured her vagina. However, she had no previous history of endometriosis. The patient was treated with local lesion excision via laparotomy. The pathological diagnosis of the excised tissue was endometriosis. A gonadotropin-releasing hormone (GnRH) agonist was administered three times postoperatively. The patient recovered well without evidence of disease 4 months after the excision. Conclusion: Any lesion that evolves in response to the menstrual cycle should be considered as endometriosis.

Keywords: Endometriosis, hysterectomy, laparoscopy, bleeding, gonadotropin-releasing hormone

Introduction

Postoperative vaginal cuff complications of laparoscopic-assisted vaginal hysterectomy (LAVH) included hematoma, granuloma, keloid, incisional hernia, and or vascular formation at the vault [1]. Although rare, there have been few reports of vaginal vault endometriosis in patients presenting with irregular or cyclic menstrual bleeding several months or years after hysterectomy. Endometriosis is defined as the presence of endometrial tissue that is similar to the endometrium, but presents outside the uterus. Management of endometriosis may include surgical excision and/or medical suppression [2]. Here, we present a rare case of endometriosis at the vaginal cuff, which infiltrated the serous membrane of the rectum and spontaneously ruptured the vagina in a post-hysterectomy patient with no known history of endometriosis.

Case report

A 41-year-old woman was admitted complaining of sudden massive vaginal bleeding. The patient underwent hysterectomy 3 years prior due to symptomatic multiple leiomyomas and adenomyosis. The previous surgery was conducted as a LAVH. Surgical findings showed an enlarged uterus, approximately the size of a uterus at 13 weeks of gestational age. Both adnexa were grossly normal in appearance, and the peritoneum was clear, with no signs of endometriosis. The vaginal vault was sutured vaginally using 1-0 vicryl sutures. No complications were observed during the postoperative period and the patient was discharged as scheduled. The histology of the uterus was confirmed as adenomyosis and leiomyoma with a secretory phase endometrium.

Two months after the operation, the patient presented to our clinic complaining of spotting

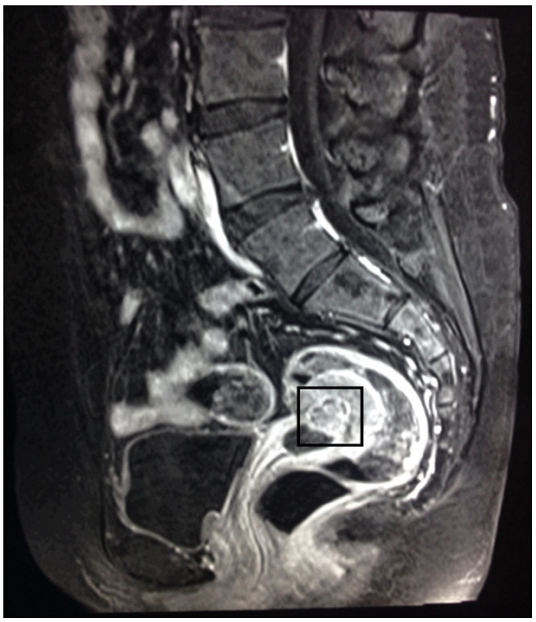

Figure 1. Magnetic resonance imaging (MRI) scan showing an endometriosis mass.

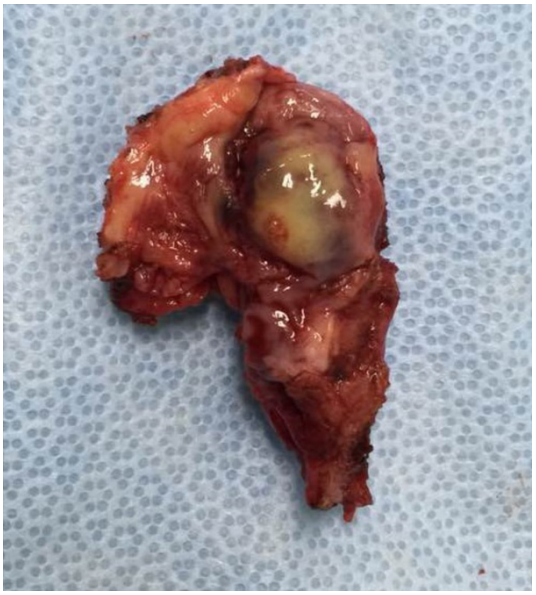

Figure 2. The excised specimen.

that correlated with her menstrual cycle. The symptom disappeared when 25 mg q.d. mifepristone was administered. The dosage of the mifepristone was gradually reduced to 6.25 mg q.d., 2 days before the sudden vaginal bleeding.

Vaginal speculum examination showed no signs of active bleeding. However, a dark wine-colored mass ~3 cm in diameter was observed at the left vaginal vault site. The mass had a hole of ~1 cm in diameter on its surface. The depth of the hole was 2 cm when probed by a cotton swab, and rupture of the vaginal cuff along with endometriosis was suspected. Pelvic examination revealed a hard mass of ~3 cm in diameter on the left vaginal vault site. The mass infiltrated backwards to the rectal wall, and the nodule was palpable on digital rectal examination. However, the mucosa membrane of the rectum was smooth.

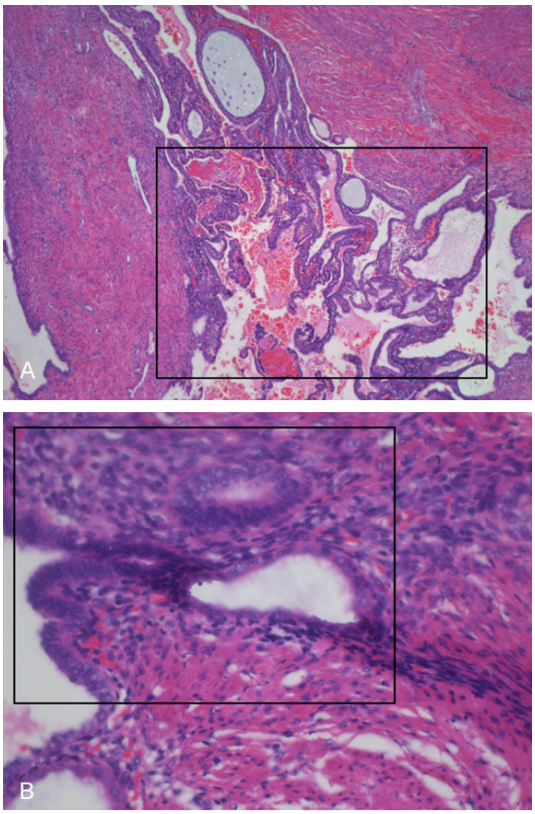

Figure 3. Histological findings of the excised specimen. The tissue consisted of typical endometriotic glands and stroma with dense fibrous structure. Dilated glands are occasionally observed (HE staining, originally magnification ×10 and ×40).

Transvaginal ultrasound (TVUS) revealed an irregular low echo of approximately 33 × 37 × 34 mm at the vaginal vault, showing internal vascularity on duplex color Doppler ultrasound. Pelvic magnetic resonance imaging (MRI) confirmed the diagnosis as vaginal cuff endometriosis with spicule sign at the point of protrusion through the rectum serous membrane (**Figure 1**). The serum cancer antigen 125 level was 25 U/mL.

Table 1. Summary of all cases

Case (reference)	Age (years)	Primary Operation	Time of Endometriosis Complication after Surgery	Treatment
1 [4]	NA	Total vaginal hysterectomy	NA	NA
2 [5]	NA	NA	NA	NA
3 [6]	31	Total abdominal hysterectomy and bilateral salpingo-oophorectomy	5 years after total abdominal hysterectomy, 4 years after bilateral salpingo-oophorectomy	NA
4 [7]	37	Total abdominal hysterectomy	6 months	Laparoscopic excision of the mass and fistula after ureterolysis and bowel dissection
5 [8]	44	Laparoscopic-assisted vaginal hysterectomy	2 years	Laparotomy excision
6 [9]	42	Abdominal hysterectomy and right salpingo-oophorectomy	6 months	Laparotomy left-oophorectomy and fistula repair
7 [10]	42	Total laparoscopic hysterectomy	13 months	Excision
8 [11]	41	Ward Mayo's vaginal hysterectomy	2.5 years	Laparotomy excision
9 [12]	45	Single-port laparoscopic-assisted vaginal hysterectomy	13 months	Emergency exploratory laparoscopic operation

NA, Not Applicable.

An exploratory laparoscopy operation was performed to control vault bleeding and remove the lesion. The vaginal cuff closely adhered to the anterior rectal wall and bladder peritoneum. After dissecting the adhesion, a full thickness excision of the vault lesion was performed (**Figure 2**). The remaining vaginal vault was sutured using 1-0 vicryl sutures. The rectal wall, which was adhered to the vault, showed infiltration of endometriosis. The lesion on the serous membrane of the rectum was removed; however, the mucosa of the rectum remained intact. Upon surgery, other peritoneal structures, including both the ovaries, were grossly normal and showed no signs of endometriosis. Pathology of the excised lesion was confirmed to be consistent with endometriosis (**Figure 3**). Deep external endometriosis of the vaginal cuff and surrounding tissues was the cause of menstruation. All lesions were surgically removed. To maximize the outcome, as well as decrease the chances of recurrence, a gonadotropin-releasing hormone (GnRH) agonist was used three times postoperatively to reduce recurrence. No significant adverse drug reaction was noted, and the patient is satisfied with her current therapy.

Discussion

In cases of sudden vaginal bleeding with a history of prior total hysterectomy, granulation tissue formation must be excluded. Additionally, endometriosis at the vaginal vault should also be taken into consideration [3]. Although rare, there have been few reports of vaginal vault endometriosis in patients presenting with regular or cyclic menstrual bleeding several months or years after hysterectomy [4-12]. PubMed, MEDLINE, and Google Scholar were searched. The literature was limited to English-language case reports. We present a summary of these cases to highlight their clinicopathological profiles (**Table 1**). However, most patients had a history of ovarian endometriosis with adhesion or a fistulous tract formation to the vault or even some endometriosis spots left behind near the vault site. For those who had no prior evidence of endometriosis, the possible pathophysiology is suspected to be endometrial implantation during surgery.

In this case, the risk of development into vault site endometriosis could be due to a uterus showing adenomyosis. Some endometriosis cells might have scattered in the vaginal cuff intraoperatively, indicating that seeding of the endometriosis cells at the vagina might have been the reason for cell adhesion. This hypothesis also explains some other disease such as c-scar endometriosis and episiotomy scar endometriosis [13]. These transplantation cells further grow into endometriosis lesions under

hormone stimulation. However, the seeding and adhesion theory has not yet been proved. Thus, further research is required.

The treatment for endometriosis needs to be individualized depending on factors such as the patient's age, clinical symptoms, and location. Owing to its infiltrating biological behavior and high recurrence rate, surgical excision is the first choice. Although being efficacious, GnRH agonists are associated with a high incidence of hypoestrogenic side effects and substantial decrease in bone mineral density when used for a long period of time in the absence of hormonal 'add-back' therapy [14]. As our patient has both ovaries, a GnRH agonist was used three times postoperatively to reduce recurrence.

In summary, endometriosis after LAVH is relatively uncommon, and determining a diagnosis is challenging, considering both the clinical and pathological aspects. Vaginal vault endometriosis must be considered when delayed bleeding occurs after total hysterectomy. Clinicians should always be aware of endometriosis for any female patients presenting with symptoms in accord with their menstrual cycle.

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Disclosure of conflict of interest

None.

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