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## Perineal Endometriosis without Perineal Trauma: A Case Report

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#### Abstract

Vulvar endometriosis can occur after surgery or trauma and cause dyspareunia. Endometriosis of the perineum and vulva is extremely rare without trauma. We report here a case of spontaneously developing perineal endometriosis successfully treated with local excision. A 23-year-old woman was admitted complaining of a vulvar lump. It had gradually increased in size. Gynecological examination showed a subcutaneous mass in the left perineum. The patient was treated by local excision of the vulvar mass, and complete excision was achieved. The pathological diagnosis of the excised tissue was endometriosis. The patient is well without evidence of disease 6 months following the local excision. Spontaneous perineal and vulvar endometriosis is extremely rare. Any vulvar lesion, regardless of its appearance and location, can be related to endometriosis. Surgical resection is recommended to relieve the symptoms and provide histological proof.

Keywords: Endometriosis; Trauma; Perineum

#### Introduction

Endometriosis, a common, benign, estrogen-dependent tumor-like disease affecting 3-10% of women of reproductive age, is characterized by the ectopic growth of endometrial tissue outside the uterine cavity, primarily in the peritoneum, ovaries, and rectovaginal septum [1].

Endometriosis of the perineum and vulva has been reported in the literature, with the most common site being episiotomy scars [2]. However, spontaneous vulvar endometriosis, not associated with episiotomies, is extremely rare. In this case report, we present a rare case of spontaneously developing perineal endometriosis successfully treated with local excision.

### **Case Report**

A 23-year-old female with a perineal mass was examined in october 2014. Her first episode of menarche was at the age of 13. One year later, her menstruation was normal, occurring for 3-5 d/28 d without dysmenorrhea. She denied having experienced either perineal trauma or sexual intercourse. She found a painless papule by accident located between the left labium majus and labium minus. No medical attention was given at the time. More papules appeared from the same site and grew gradually. Gynecologic examination showed a firm mass with dozens of dome shaped papules measuring approximately  $60 \times 30$  mm between the left labium majus and labium minus (Figures 1 and 2).

It was firm and painless. The color of the mass was the same as the skin surrounding it. The uterus and adnexa were found to be normal during ultrasound examination. The mass was completely removed under general anesthesia (Figure 3). Chocolate colored old blood was accumulated inside the resected mass. Pathological examination of the tissue revealed endometriosis. The patient was discharged 3 days later. Postoperative follow-up was scheduled every 3-6 months to monitor any recurrence of the lesion, which was not reported by the patient.



Figure 1: Gynecologic examination of dome shaped papules.

#### Discussion

Endometriosis is a common disease, but the etiology and pathogenesis of endometriosis are still unclear. Many theories have been proposed to explain this condition as the endometrium implantation theory, the coelomic metaplasia theory, the lymphatic and vascular metastasis theories, the mechanical transplant theory of remnant endometrium in spontaneous or operative delivery, the embryonic rests theory, a recent hypothesis based on the relationship of local immune factors [3].



**Figure 2:** Gynecologic examination of dome shaped papules measuring approximately  $60 \times 30$  mm between the left labium majus and labium minus.



Figure 3: Papule removed under general anesthesia.

Perineal trauma, especially perineal tearing or episiotomy during vaginal delivery, can result in perineal endometriosis. The etiology of

this type of endometriosis is that endometrial tissue in the perineal area during parturition or instrumentation causes perineal endometriosis, which is caused by medical profession.

The position of the lesions can be explained by mechanical transplantation of endometrial cells to open episiotomy scars, which supports the transport theory of this extrapelvic endometriosis [4]. It is likely that by the direct implantation of endometrial cells during vaginal delivery, viable endometrial cells are implanted into the episiotomy wound and subsequent cell growth occurs at the healing phase of the wound [2].

This patient had no history of surgical manipulation or trauma to the perineal area. Endometrium may have been disseminating by lymphatic system [5].

Endometrial tissue can pass through these lymphatic ducts and result in perineal endometriosis. In other words, perineal endometriosis without perineal trauma is believed to be associated with multiple factors, especially benign lymphatic metastasis. However, other factors, such as immunological, genetic and familial factors, could be involved in the pathogenesis of this disease.

Pollack et al [5] reported one of the first perineal endometriosis case without perineal trauma in 1990. Three typical characteristics of perineal endometriosis caused by perineal trauma include:

- past perineal tearing or episiotomy during vaginal delivery;
- a tender nodule or mass at the perineal lesion;
- progressive and cyclic perineal pain. If these three criteria were confirmed, the diagnostic accuracy rate was 100% [6].

Our patient experienced neither progressive nor cyclic perineal pain. Furthermore, the disease progressed at a relatively slow rate; the patient did not seek treatment until the diameter of the mass was 6 cm. An accurate diagnosis was not given before the operation.

Another perineal endometriosis without perineal trauma identified by Pollack et al. [5] presented with a tender and progressive mass accompanying cyclic pain, but accurate diagnosis had not been given. It appears that the diagnosis of perineal endometriosis without trauma is difficult and depends on symptoms and gynecologic examination. There are a variety of cases of primary cutaneous endometriosis without previous abdominal surgery at different sites such as umbilicus, vulva, perineum, groin, and extremities reported by Ideyi et al. [7] All of these patients, including the present case, were treated by local excision.

Because of its convenience and non-invasive character, the sonography was useful for discovering the depth of invasion and the precise size of the perineal mass. Although fine needle aspiration and sonography was not used in this case, we believe that a combination of fine needle aspiration cytology with ultrasound would be helpful for diagnosing perineal endometriosis without perineal trauma.

Treatment of endometriosis includes surgical intervention and hormonal suppression. Surgical excision of perineal endometriosis with perineal trauma should be the first choice, while hormonal suppression should be a secondary treatment. If the lesion has been excised entirely, hormonal suppression is not necessary. Usually the lesion is small and limited in location, and can be completely excised, appears that perineal endometriosis without perineal trauma is a curable disorder.

Histological examination is necessary for the diagnosis of endometriosis and critical to exclude malignancies. Surgical excision

of the lesion is the primary treatment. Narrow excision and incomplete excision may result in high recurrences. Adequate, wide excision of endometriotic tissue seems to be the best chance of cure with satisfactory functional results and should be recommended [8].

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