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A rare case of perineal endometriosis in an episiotomy scar

Rzadki przypadek endometriozy umiejscowionej w bliźnie po nacięciu krocza

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Abstract

Introduction: Episiotomy scar endometriosis is an extremely rare entity and often causes diagnostic uncertainty. **Case report:** We report a case of perineal swelling and cyclical pain following obstetric delivery with episiotomy. Magnetic resonance imaging revealed possible episiotomy scar endometriosis confined to the perineum. Wide surgical excision was performed and the histopathological report confirmed the diagnosis. No recurrence was noted after the surgery. **Conclusion:** Episiotomy scar endometriosis should be considered whenever a woman with previous episiotomy presents with cyclical pain or a nodule in the perineum. Magnetic resonance imaging can assist with the diagnosis and wide excision remains the best treatment option for this condition.

Keywords: episiotomy, perineal endometriosis, scar endometriosis

Streszczenie

Wstęp: Endometrioza umiejscowiona w bliźnie po nacięciu krocza (episiotomii) jest niezwykle rzadką jednostką chorobową i częstą przyczyną wątpliwości diagnostycznych. **Opis przypadku:** W pracy przedstawiono przypadek obrzęku w okolicy krocza z towarzyszącym cyklicznym bólem u pacjentki po porodzie siłami natury, podczas którego wykonano nacięcie krocza. Na podstawie badania metodą rezonansu magnetycznego wysunięto podejrzenie ogniska endometriozy umiejscowionej w bliźnie po episiotomii i ograniczonej do okolicy krocza. Wykonano szerokie wycięcie chirurgiczne zmiany. Wynik badania histopatologicznego potwierdził rozpoznanie. Po przeprowadzonym zabiegu nie odnotowano nawrotu choroby. **Wnioski:** Rozpoznanie endometriozy w bliźnie po episiotomii należy brać pod uwagę w każdym przypadku pacjentki z nacięciem krocza w wywiadzie, u której występują cykliczne dolegliwości bólowe lub guzowata zmiana w obrębie krocza. Pomocne w diagnostyce może okazać się badanie metodą rezonansu magnetycznego, natomiast najlepszą opcją leczenia pozostaje szerokie wycięcie chirurgiczne zmiany.

Słowa kluczowe: endometrioza, nacięcie krocza (episiotomia), endometrioza krocza, endometrioza umiejscowiona w bliźnie

INTRODUCTION

Endometriosis is defined as the presence of functional endometrial tissue and stroma outside the uterine cavity. It is one of the most common gynecological disorders, seen in 10–25% of women in reproductive age group[1]. The etiology and pathogenesis of endometriosis remains controversial. Several theories about the pathogenesis of endometriosis have generally been attributed to direct implantation, lymphatic dissemination, coelomic metaplasia, or hematogenous spread. Other factors, such as immunological, familial and genetic factors, may also be involved in the pathogenesis of this disease.

Endometriosis is commonly observed in the pelvic organs, especially the ovaries, fallopian tubes, pelvic peritoneum, uterine ligaments and pouch of Douglas. Extra-pelvic localization of endometrial tissue is rather uncommon, accounting for approximately 12% of all cases[2]. The most common extra-pelvic type is surgical scar endometriosis[3], which is more frequently seen in scars after cesarean sections, and rarely in episiotomy and other obstetrical, gynecologic and non-gynecologic surgeries. Episiotomy scar endometriosis is extremely rare, occurring in only 0.00007% of births[4]. As episiotomy is frequently performed at the time of vaginal delivery, one should know about this rare entity, its etiopathogenesis, various diagnostic measures and management options. Herein, we report a case of episiotomy scar endometriosis successfully treated with wide surgical excision with excellent results.

CASE REPORT

A 36-year-old multiparous woman presented with pain and swelling in perineal region for three years. She was initially evaluated by a general surgeon and diagnosed with a perineal abscess. An incision and drainage were performed, and

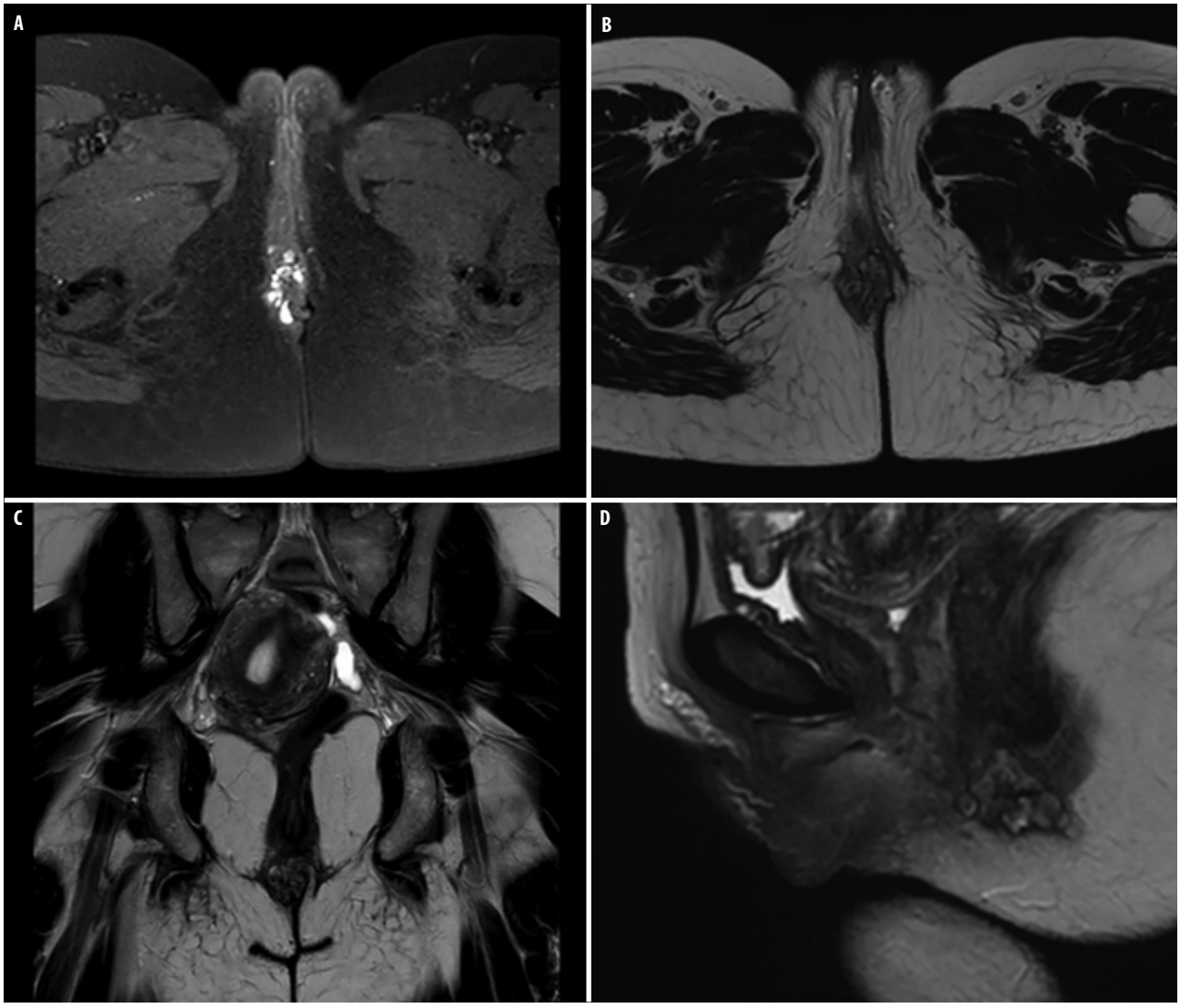

*Fig. 1. Pelvic MRI shows right perineal lesion. (**A**) Axial T1-weighted image, (**B**) axial T2-weighted image, (**C**) coronal T2-weighted image, (**D**) sagittal T2-weighted image*

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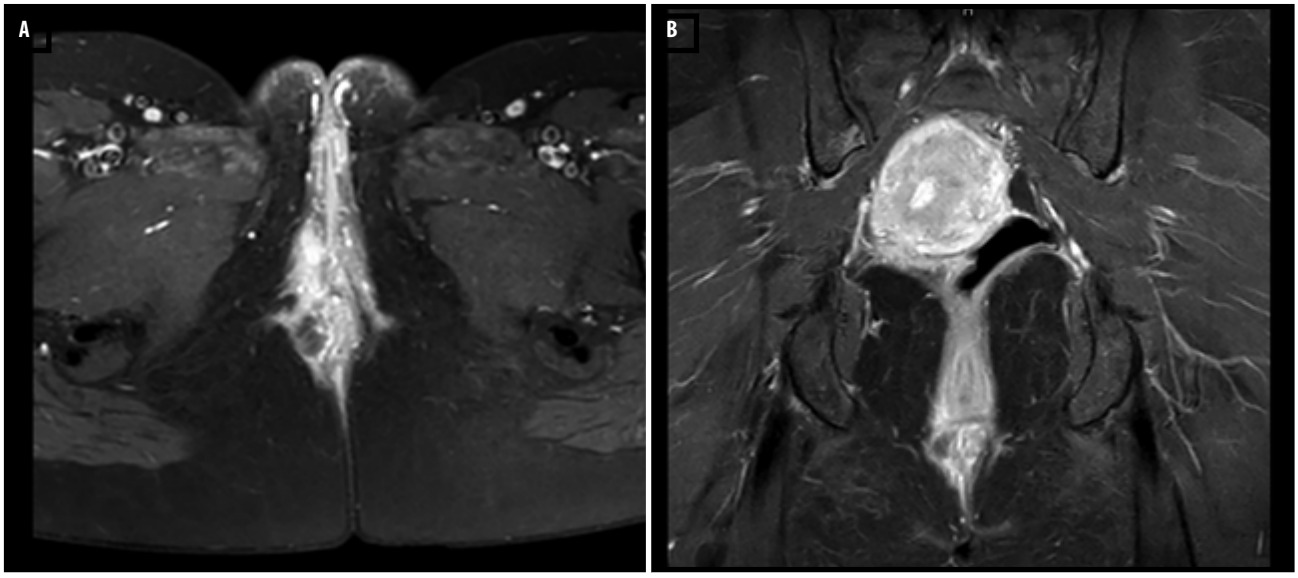

*Fig. 2. Pelvic MRI after injecting IV contrast shows enhancement of the lesion with a linear sinus tract in the right perineal region. (**A**) Axial view, (**B**) coronal view*

blood with dirty material was evacuated. The postoperative period was uncomplicated and the patient experienced no pain. However, five months later a clinical relapse occurred when she completely abandoned oral contraceptives. The pain was progressive and cyclical, correlated with her menstrual cycle and appeared several days before its onset. She had 3 previous vaginal deliveries with a right medio-lateral episiotomy performed each time. Her last parturition was 10 years ago. A year after the last delivery, the patient felt a small swelling at the episiotomy site, and 6 years later,

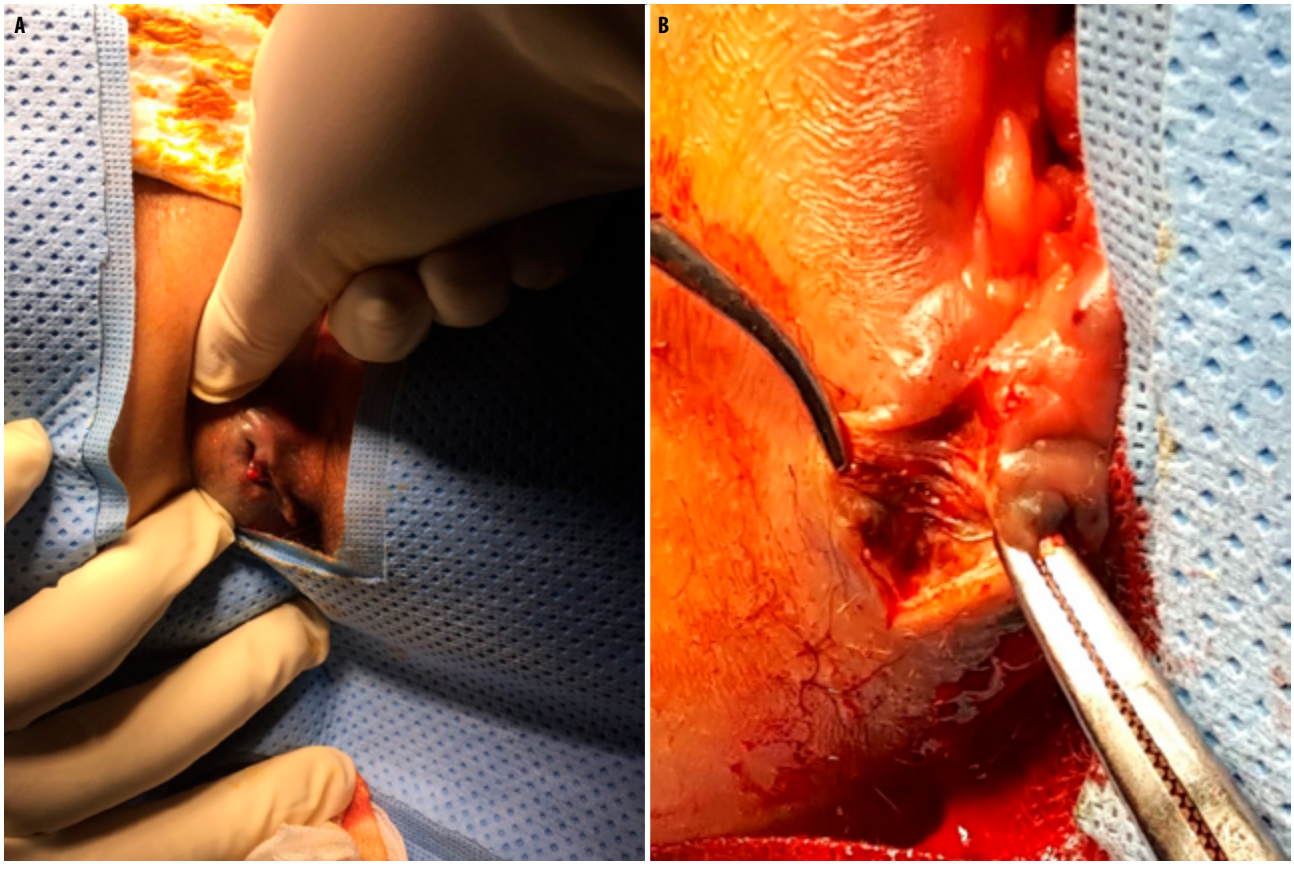

*Fig. 3. Perineal mass with the (**A**) sinus opening discharging blood, (**B**) brownish endometriotic-like deposit*

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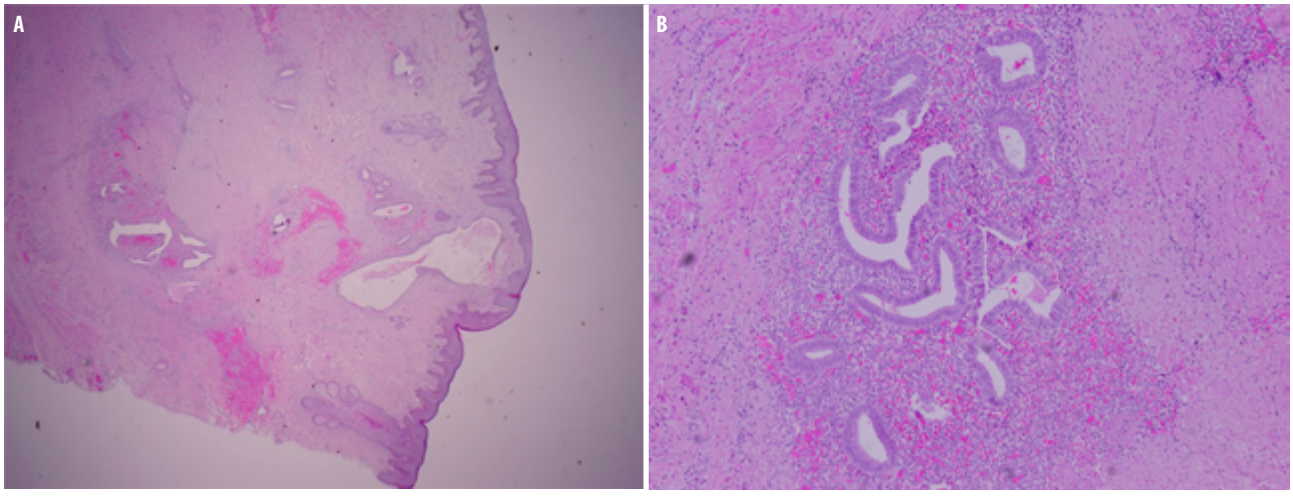

*Fig. 4. Endometrial glandular epithelium and stroma in the perineal region (**A**) in the subcutaneous tissue, (**B**) in the perineal tissue*

when oral contraceptive pills were changed, she observed slight growth of the swelling, which became larger and more painful during menses. The swelling was also reported to have sporadic drainage that ranged from dark red to brown. Her menarche was at the age of 13 years. Her menstrual cycles were regular; the flow was normal but accompanied by pain in the abdomen as well as the perineal region. She described the pain as severe, preventing her from sitting, sexual intercourse and ordinary daily activities. There was no personal or family history of endometriosis.

On examination her vitals were normal and systemic examination revealed no abnormality. On local examination, a sinus opening was seen externally at the site of episiotomy scar, and did not seem to be communicating with the vagina or anal orifice. Episiotomy scar site felt indurated and was tender on touch, hindering us from proper examination. A professional diagnosis of possible episiotomy scar endometriosis with fistula-in-ano was made.

Magnetic resonance imaging (MRI) was requested to assess the deeper extension of the lesion and to exclude involvement of the anal sphincter complex. MRI showed a 1.5 × 1.3 × 1.3 cm right perineal hemorrhagic fluid collection just below the level of the anal orifice in the midst of the scar tissue with blood lined sinus tract extending to the right buttock cutaneous opening and also multiple hemorrhagic foci seen around the perineal body to the right of the midline, probably related to the episiotomy scar (Fig. 1A–D and Fig. 2A, B). There was no definitive evidence of anal canal fistula or anovaginal communication. External and internal sphincter muscles appeared normal. Pelvic organs, such as uterus, both ovaries, rectum and urinary bladder, were unremarkable. The overall picture suggested episiotomy scar endometriosis confined to the perineum without disturbing the anal sphincter complex. The findings and impression were discussed with the patient, and the need for examination under anesthesia with excision of the endometriotic tissue was explained.

The procedure was performed under spinal anesthesia. Intraoperative findings revealed a firm mass about 4 × 4 cm in the perineal region, at the site of episiotomy scar. The mass was extending from the posterior fourchette up to 1 cm lateral to the right side of the external anal sphincter. A sinus opening was seen over the mass, discharging old blood (Fig. 3A). Probe examination was attempted and found that the sinus was very superficial, and no fistulous tract seen. Per speculum and vaginal examinations were normal. Anal sphincter and rectal mucosa were found to be uninvolved on per rectal examination. After identification of the limits of the mass, incision was made in the skin overlying it at the level of prior episiotomy scar lateral to the sinus opening. The mass was deep and adherent to the surrounding tissue, separated by sharp dissection. During dissection, brownish endometriotic-like deposit was seen in the mass tissue (Fig. 3B). Wide local excision of the mass with 1 cm margin of surrounding normal tissue was done to prevent recurrence. The mass with the overlying skin and the sinus opening were excised completely and sent for histopathology. Reconstruction of the perineum was done in layers. Histopathological examination revealed endometrial glandular epithelium and stroma confirming the diagnosis of scar endometriosis, and no evidence of atypia or malignancy was seen (Fig. 4A, B). The postoperative recovery was uneventful, and on receiving the histopathological report, the patient was given injection Zoladex (goserelin) 3.6 mg intramuscular to prevent recurrence. At the sixth week's postoperative visit, the wound was primarily healed with no residual pain. Sexual activity was resumed without any discomfort. Eight months later, the patient remains asymptomatic with painless subsequent menstruation and no signs or symptoms of recurrence.

DISCUSSION

Although pelvic intraperitoneal surfaces are the most common sites of endometriotic disease, perineal endometriosis is a relatively uncommon condition, accounting for 0.31% of women with endometriosis treated surgically at Peking

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Union Medical College[5]. Perineal trauma, such as perineal tearing or episiotomy scars, appear to be more commonly affected especially if the episiotomy is associated with a vaginal delivery and subsequent uterine curettage[6]. The etiology of perineal scar endometriosis can be explained by the theory of transplantation; however, the incidence of endometriosis is small compared to the spillage of endometrial cells into surgical incisions that probably occurs quite frequently during obstetrical or gynecologic surgery. This implies that women with endometriosis have additional factors, such as genetic, immunological or biochemical factors that contribute to the survival of endometrial fragments against the body defences, which then attach to surfaces, and consequently invade and modify normal tissue in order to form an endometriotic lesion[7]. Many factors, such as receptor-binding cancer antigen expressed on SiSo cells (RCAS1), metallothionein (MT), and DNA fragmentation factor-45 (DFF45), have been proposed to play an important role in the pathogenesis of endometriosis[8–10]. The ability of endometrial cells to regulate cytotoxic activity (RCAS1 expression) and high protection against DNA damage or apoptosis (MT expression) with associated changes in the immune cells appears to be essential for pathological characteristics of endometriosis[8]. The expression of RCAS1 and MT by the endometrium may support the survival of ectopic endometrial cells in scar endometriosis[9]. DFF45 also appears to play an important role in the apoptotic process, and a decreased level of DFF45 found in endometriotic lesions might be a part of an apoptosis resistance mechanism contributing to the progression of the disease[10]. All these studies indicated that these factors are significantly involved in the pathogenesis of endometriosis; however, it is still unclear if these factors are responsible for endometriosis development, and more research is needed to reach a better understanding of this condition.
Symptoms and/or signs of endometriosis usually appear shortly after ectopic endometrial cell implantation, with some cases having a prolonged latent period of up to 20 years after implantation[4,11,12]. In our patient, symptom onset was ten years after last childbirth with episiotomy. Although the patient first noticed the swelling one year after her last delivery, she presented to the clinic only when she could no longer cope with the pain. Her past oral contraceptive usage with its temporary suppression effect on the symptoms may explain the delay of consultation and treatment. The diagnosis of perineal scar endometriomas can be made by detailed history and thorough pelvic and perineal examination. Usually, a classical triad of cyclic pain, perineal mass and previous episiotomy or tear during vaginal delivery is sufficient to clinch the diagnosis in perineal scar endometriosis. Although this triad was met in our patient, the diagnosis of a perineal abscess was initially suspected by the general surgeon. The rarity of perineal endometriosis and the limited knowledge of the disease cause difficulty in diagnosing this condition, especially among specialists who do not normally treat these cases and may not have included perineal endometriosis in the differential diagnosis of perineal masses.
Various imaging modalities have been used to establish differential diagnosis of this entity. Ultrasonography can reveal the size and character of the masses, depth of invasion and surrounding structures. However, the sonographic features are non-specific, which are usually hypoechoic or heterogeneous nodules, sometimes hyperechoic, with external outlines often fuzzy and irregular, having a variable shape and size depending on the timing of menstrual cycle or current medical treatment[4,13]. Endoanal or endorectal sonography has been recommended particularly for assessing anal sphincter involvement[13]. Computed tomography (CT) scan is rarely advised as it lacks resolution and has radiation hazards[4]. MRI is currently considered the best imaging modality to evaluate the extension of endometriotic lesions and its relation to the nearby structures. Pelvic MRI shows greater sensitivity (90–92%) and specificity (91–98%) for diagnosing endometriomas compared to CT and ultrasound[12]. It is particularly useful for identification of small lesions and differentiation from other integument tumor-like lesions, such as lipoma or an abscess. The use of fine needle aspiration is controversial, although some authors assert that the use of this technique provides a pathological diagnosis before surgery, others suggest that it might increase the risk of a new implant in the puncture site[4]. CA-125 is a glycoprotein antigen expressed in the endometrium. Women with endometriosis often have serum CA-125 greater than 35 IU/mL. However, its levels can also be elevated in normal women at the time of ovulation, menstruation, pregnancy and following peritoneal irritation by infection or surgery[14]. Therefore, serum CA-125 is not an ideal marker for the diagnosis of endometriosis, but could be helpful in monitoring treatment outcomes and recurrence.
Wide excision of the endometriotic tissue with 1 cm margin of the surrounding normal tissue on all sides is the treatment of choice for perineal endometriosis[15]. This procedure, which usually cures the patient, was performed in our case. Therefore, a thorough preoperative evaluation to define the limits of the lesion is important to ensure its complete removal. It has also an advantage of providing a sample for biopsy to confirm the diagnosis and exclude malignancy, though rare[4]. Incomplete excision predisposes the patient to recurrence of the disease as shown in the described case, when the initial diagnosis was considered as perineal abscess and only an incision and drainage was done, and the patient presented again five months later with the same problem. The option of medical treatment alone does not appear to be effective. Although symptomatic relief might be achieved with hormonal suppression, most patients have a recurrence of symptoms after treatment discontinuation. Medical therapy might be a preoperative option in cases with a large endometriomas or coexisting with pelvic endometriosis[5,11], and a postoperative option to prevent recurrence. The drugs most commonly

used are oral contraceptives, danazol, progesterone and gonadotropin releasing hormone agonists. In our patient, a single dose of 3.6 mg Zoladex postoperatively was helpful. There is a question as to whether perineal endometriosis cases are preventable. There are no consistent data in the literature to support any preventive measure. Hypotheses have been suggested, such as washing the episiotomy wound with normal saline before suturing, avoiding manual uterine exploration and postpartum curettage; however, further studies are needed to support these actions.

In conclusion, although perineal scar endometriosis is a rare condition, it should be suspected whenever a woman in reproductive age group with previous history of episiotomy presents with perineal pain or a nodule coinciding with her menstrual cycle. MRI is the preferred imaging modality to reinforce the diagnosis and assess the deeper extension of the lesion. Wide surgical excision remains the best treatment option for perineal endometriosis, and follow-up is essential as recurrence is not uncommon. Luckily, in our patient, a good recovery with a favorable outcome was achieved and no evidence of recurrence was noted in the eight months of follow-up.

Conflict of interest

The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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