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Endometriosis in an Episiotomy Scar: Case Report

Epizyotomi Skarında Endometriozis: Olgu Sunumu

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ABSTRACT

Endometriosis is defined as the presence of endometrial stroma and glands outside the uterine cavity. It is a chronic disease found most commonly in women of reproductive age. In this study, we present a rare case of endometriosis which presented as a mass in the episiotomy scar tissues. A 35-year-old female patient was referred to our clinic with a complaint of painful swelling in the perineal region since 3 months. During physical examination, we palpated a painful mass and performed ultrasonography (USG) and magnetic resonance imaging (MRI) which revealed a fistula tract. The mass was surgically resected and the histopathologic examination of the mass confirmed perineal endometrioma in an episiotomy scar.

Key Words: Endometriosis; episiotomy; perineal pain

ÖZET

Endometrial stroma ve glandların uterus kavitesi dışında yerleşmesine endometriozis denir. Bu çalışmamızda oldukça nadir görülen perineal endometriozisli bir olgunun özellikleri sunulmuştur. Perineal bölgede üç aydır ağrı şikayeti olan 35 yaşında kadın hasta hastanemize başvurdu. Yapılan fizik muayenesinde ele gelen sertlik nedeni ile olguya ultrason (USG) ve manyetik rezonans görüntüleme (MRG) uygulandı. MRG`de fistül traktı ile uyumlu olan lezyonun eksizyon materyalinde endometriozis ile uyumlu histopatolojik görünüm izlendi. Endometriozisin perineal bölgede nadir görülmesi ve klinikte perineal fistül ve maligniteleri taklit etmesi nedeni ile ayırıcı tanıda akılda tutulması gereken bir durumdur.

Anahtar Kelimeler: Endometriozis; epizyotomi; perineal ağrı

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Endometriosis is defined as the presence of functional endometrial tissue outside the uterine cavity.[1] It is the second most prevalent benign gynecologic disease in women of reproductive age, with an incidence of 10% to 25%.[2,3] Despite the presence of endometrial tissue in ectopic locations being a common finding, perineal endometriosis is a very rare condition. Schickele (1923) is thought to be the first

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to report a case of perineal endometriosis.[4] Perineal scar endometriosis, which is characterized by the presence of ectopic endometrial stroma and glands in the perineum, is a rare condition that affects 0.3% to 1% of patients.[5] Few cases of perineal endometriosis have been reported. It generally develops secondary to obstetric or surgical trauma and is most commonly observed in the episiotomy scar after normal vaginal deliveries. In nearly half of the women with perineal endometriosis, the lesions erode into the anal sphincter. Its clinical diagnosis is difficult and is generally confused with abscess, hematoma, suture granuloma, desmoid tumor, sarcoma, and metastatic malignancy. Thus, this could lead to unnecessary investigations and treatments. The pathogenesis of endometriosis still remains controversial. The theories include 1) implantation theory, 2) the coelomic metaplasia, and 3) transplantation of exfoliated endometrium by lymphatic, vascular, transtubal regurgitation and iatrogenic routes that may disseminate endometriosis, 4) altered immunological recognition of endometrial tissues. The clinical manifestations may range from an asymptomatic mass to a mass increasing in size and becoming acutely painful during menstruation periods. On USG imaging, the aspect is non-specific, appearing as hypoechoic or heterogenous nodules (depending on their solid and/or liquid component) or sometimes as hyperechoic nodules with blurred and irregular margins that infiltrate the surrounding tissue; these may differ in shape and size, depending on the amount of blood/fibrosis, the time of the cycle, and/or the medical treatment in progress. On MRI the endometriosis lesions on episiotomy scars appear as a hypodense, fibrous thickening on T2-weighted sequences. The appearance is more pronounced in cases of a stellar or retractile form of infiltration. The treatment of choice is total surgical excision. We should remember that perineal endometriosis is a rare condition and it may mimic perineal fistulas and malignancy.

CASE REPORT

Herein, we present a case of a 35-year-old woman who was referred to us for the development of a tender perineal mass at the episiotomy site associated with perineal pain and pruritus. Digital examination revealed a hard mass measuring 2 cm × 2 cm in the right anterior perineal region, adjacent to an episiotomy scar. The pain intensified before and immediately after the onset of menstrual bleeding. Her symptoms started 3 months ago after a vaginal delivery was performed with mediolateral episiotomy. Prior to this, she had a vaginal delivery with episiotomy seven years ago. Perineal examination showed an episiotomy scar in the right mediolateral position. Diagnostic tools used included USG and MRI (Image 1).

There was no sign of pelvic endometriosis in USG findings, and serum CA-125 level was normal. Pelvic examination was also normal, and the uterus was mobile. There was no nodularity in the pouch of Douglas or the rectovaginal septum. Under general anesthesia, a complete surgical excision was performed. During excision, we drained the mass

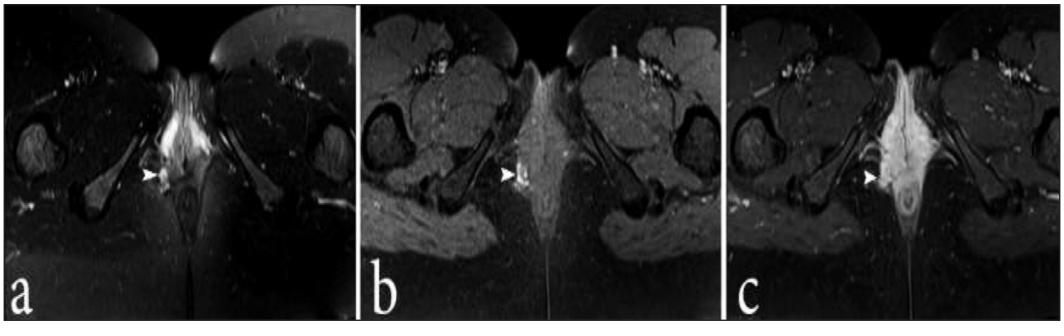

IMAGE 1: Axial T2-weighted (a) and axial fat-saturated T1-weighted (b) MR images reveal a small nodule at the episiotomy site in the right paravaginal area, which was hyperintense in both sequences because of hemorrhagic content (arrowheads). The lesion shows enhancement on post-contrast axial fat-saturated T1-weighted MRI (c) due to the presence of scar tissue (arrowhead).

containing "brownish" fluid. The pathology report confirmed perineal endometriosis.

Intraoperatively, frozen tissue sections showed endometrial-type glands and stroma embedded in a fibroconnective tissue containing mild inflammatory cells (Figure 1). Chronic hemorrhage was evidenced by hemosiderin-laden macrophages scattered among endometrial stromal cells. The intraoperative histopathologic diagnosis was endometriosis externa.

The findings for permanent sections of the formalin-fixed and paraffin-embedded tissue stained with hematoxylin and eosin (H&E) were similar to those of frozen sections. Microscopic examination revealed variable-sized ectopic endometrioid glands within cellular endometrial-type stroma which was surrounded by dense fibroconnective tissue (Figure 2). Pseudoxanthoma cells filled with hemofuscin pigment were also identified in the stroma around glandular structures (Figure 3). CD10 immunohistochemistry showed cytoplasmic positivity in endometrial stromal cells (Figure 4).

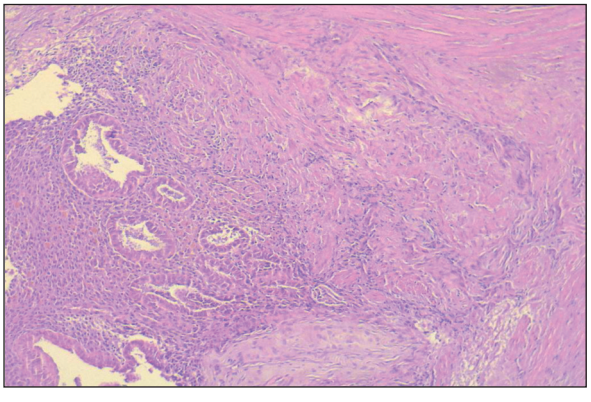

FIGURE 1: X100, H&E, endometrial-type glands and stroma within the fibroconnective tissue.

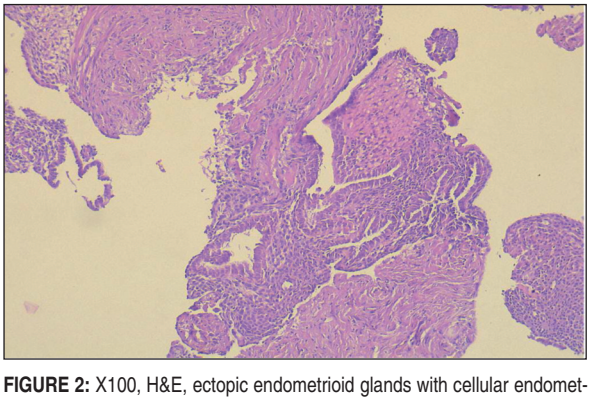

FIGURE 2: X100, H&E, ectopic endometrioid glands with cellular endometrial-type stroma.

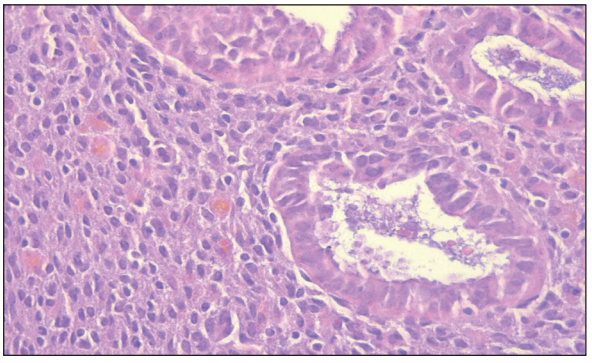

FIGURE 3: X400, H&E, hemofuscin pigment filled macrophages in endometrial-type stroma.

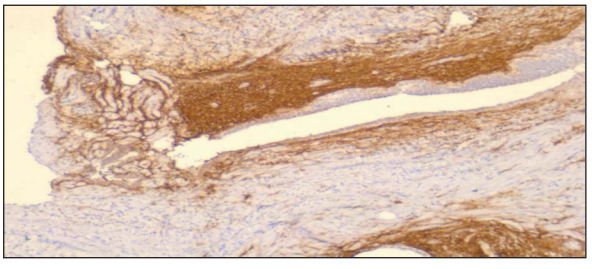

FIGURE 4: X100, CD10 immunopositivity in cytoplasms of endometrioid-type stromal cells.

The recovery was uneventful with excellent functional and esthetic results. No postoperative recurrence was noted on the last follow-up at 2 months, and the woman remains asymptomatic.

DISCUSSION

Endometriosis is the extrauterine occurrence of endometrial glands and stroma, most often involving the ovaries or dependent visceral peritoneal surfaces. The etiology of endometriosis is unclear; however, autologous transplantation of vital endometrial cells to an open episiotomy wound during vaginal delivery (particularly when manual uterine exploration and postpartum curettage are performed) seems to be the pathogenic mechanism of perineal endometriosis.[6] In addition, in our case, there was a history of postpartum curettage because of insufficient placental separation. Perineal lesions often occur during vaginal delivery; however, the incidence of scar endometriosis is rare. The reasons

for rare incidence may include the following: (1) Bacteria existing in the perineal wound which can cause infection or even necrosis of the local tissues. The infection and necrosis are not appropriate conditions for the transplanted endometrial cells to survive in. (2) After delivery, the level of estrogen decreases, which also makes the growth of transplanted endometrial cells difficult. Past history and physical examination are important in detecting scar endometriosis. The most common findings are swelling, pain, and occasional bleeding during the menstrual cycle in the lesion area. The mass is generally hard and is frequently located adjacent to an existing episiotomy scar or a previous site of tearing or injury. For some patients, the skin color over the perineal lesions may be brownish on examination. Some may have cyclic ulceration or bleeding from the perineal mass. Endometriosis should be considered when cyclic perineal pain is present, particularly in episiotomy scars. The onset of symptoms may be years after delivery.[7] Similar signs and symptoms can be caused by hematoma, neuroma, hernia, granuloma, and neoplasia, which should be considered in the differential diagnosis of perineal endometriosis. Menstruation-related pain and swelling in the anamnesis should be considered pathognomonic for scar endometriosis. Three typical characteristics of perineal endometriosis for women of reproductive ages should be considered when taking a history: (1) past history of perineal tearing of episiotomy during vaginal delivery, (2) the presence of a tender nodule or mass at the perineal lesion, and (3) progressive and cyclic perineal pain. In a study implementing these criteria for diagnosis, if these three criteria were met, the predictive value of perineal endometriosis was 100%.[8] Early diagnosis and treatment are important to prevent the progressive involvement of the anal sphincter, thus decreasing the risk of postoperative fecal incontinence.[9] However, a definite diagnosis can be made only by histopathological examination of the lesion. Macrophages loaded with endometrial glands, stroma, and hemosiderin on histological examination are helpful in diagnosis. The possibility of malignant transformation of the lesion should be considered in cases showing recurrence after surgery. This rare but important condition should be considered in all patients and monitored carefully during follow-ups. The primary treatment for scar endometriosis is total excision of the lesion. To prevent recurrence, it is crucial to not leave residual tissue during surgery. Although malignant degeneration of perineal scar endometriosis is rare, cases have been reported in the literature.[10,11] All cases require long-term clinical follow-up because the delay between benign endometriosis and malignant transformation can vary from a few months to over 40 years.

CONCLUSION

A comprehensive history and detailed pelvic examination are essential for the diagnosis of perineal endometriosis. Surgical intervention is the best approach for treatment, and permanent cure is usually achieved after complete excision of the perineal endometriosis. Recurrence due to incomplete removal usually appears within one year. According to the literature and our own experience, wide excision of endometriotic tissue seems to be the best chance of cure with satisfactory functional results and should be recommended. Although scar endometriosis is rare, it should be considered if patients having had a vaginal delivery complain of a painful vulvar lump with swelling during their menstrual cycle. An early diagnosis is important as a delayed diagnosis results in extension of the lesion causing increased damage to the anal sphincter and rectum. The peculiarly of this case was that the patient only presented with extra-genital endometriosis, and no other implants or cysts were found. Further, elevated levels of CA-125 were not found in the beginning of the disease or later. To minimize the chance of perineal endometriosis development, the episiotomy scar should not be contaminated with the blood and debris coming from the uterine cavity. Therefore, gloves should be changed before episiotomy repair is performed.

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