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Case Report

Scar Endometriosis in a Patient With Bladder Exstrophy

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Endometriosis is an ectopic occurrence of tissue morphologically and functionally resembling endometrial tissue in regions outside the uterine cavity. Although scar endometriosis after surgery has been shown to be most common among all the extrapelvic forms of endometriosis, endometriosis after bladder exstrophy surgery has not been reported, and here we present the first known case. A 26-year-old woman with a history of bladder exstrophy was aware of a painful induration at the operative scar located in the left lower abdominal wall, and presented at our hospital. Although the symptoms resolved, recurring exacerbation was observed after 9 months. Abdominal magnetic resonance imaging showed a heterogeneous mass 16 mm in diameter in the left abdominal wall with high signal intensity on T1W1 and T2W1 images. She underwent excisional biopsy of the lesion under general anesthesia. Histopathology confirmed the diagnosis of endometriosis. Eighteen months after surgery, she was well and free from recurrence.

Key words: Bladder exstrophy – Endometriosis – Scar endometriosis

Endometriosis is an ectopic occurrence of tissue morphologically and functionally resembling endometrial tissue that is present in regions other than the uterus.[1] Although endometriosis occurs most frequently in the intrapelvic organs, many cases of extrapelvic endometriosis throughout the body have been reported. Among them, endometriosis in a surgical scar is rare, and its clinical diagnosis can be confused with other lump lesions, such as abscesses, hematomas, and suture granulomas.[2,3] The most frequent antecedent surgical procedure among patients with scar endometriosis is a Cesarean section.[2] We were unable to find a previous report of endometriosis associated with ectopia vesica in the language we searched.

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Fig. 1 Pelvic CT reveals a cystic subcutaneous mass (white arrow).

Case Report

A 26-year-old woman (gravida 3, para 0), with a history of surgery related to bladder exstrophy in her neonatal and infantile periods, was aware of a painful induration at the operative scar located in her left groin and reported to our hospital. A pelvic computed tomography (CT) scan revealed a subcutaneous cystic lesion without calcification located coincident with the induration (Fig. 1). Suspecting a suture-related abscess, ultrasound-guided fine needle aspiration of the content fluid was performed. The aspirate comprised old blood, and cytology of the fluid was negative for malignancy. Although the symptoms were resolved completely by aspiration, exacerbation recurred 9 months later involving repeated spontaneous remissions and relapses. Interviews revealed that each flare-up coincided with the patient's menstruation. Magnetic resonance imaging (MRI) showed a heterogeneous mass 16 mm in diameter with high signal intensity on T1W1 and T2W1 images (Fig. 2A and 2B).

Based on these findings, the patient underwent excisional biopsy of the mass under general anesthesia, on suspicion of endometriosis. During the operation, the lesion could be approached easily through an incision at the previous scar in the left groin region. Although adhesions to the surrounding tissue were partially severe, the lesion did not have a border with the uterine round ligament. There was no evidence of coexisting inguinal or femoral hernias.

Histopathologically, the removed tissue included fibrous connective tissues and several hollow glands lined with columnar epithelial layers; the glands were surrounded by stromal cells (Fig. 3A). Immunohistochemical analysis of the glands revealed positive immunostaining for progesterone and estrogen receptors (Fig. 3B and 3C). A diagnosis of endometriosis was made. Bladder tissue and mesothelial cells were not found in the excised specimen.

The patient's postoperative course was uneventful, and she was discharged the following day. She achieved spontaneous pregnancy and delivered vaginally 23 months after surgery.

Discussion

Endometriosis is defined as the presence of ectopic endometrial tissue that can respond to ovarian hormonal stimulation. It is a common gynecologic condition that affects up to 22% of all women, about 20% to 30% of patients presenting with subfertility and up to 45% of women with pelvic pain.[4] Endometriosis has been described in almost all body cavities and organs, including the central nervous system, lung, small and large bowel, gallbladder, kidney, extremities, perineum, and abdominal wall. Among these sites, the presence of ectopic endometrial tissue within the abdominal

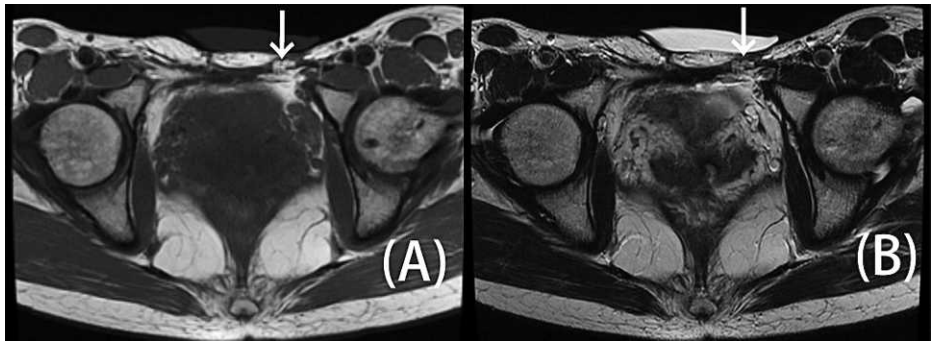

Fig. 2 Abdominal MRI shows a heterogeneous mass in the left abdominal wall with high signal intensity on (A) T1W1 and (B) T2W1 images (white arrow).

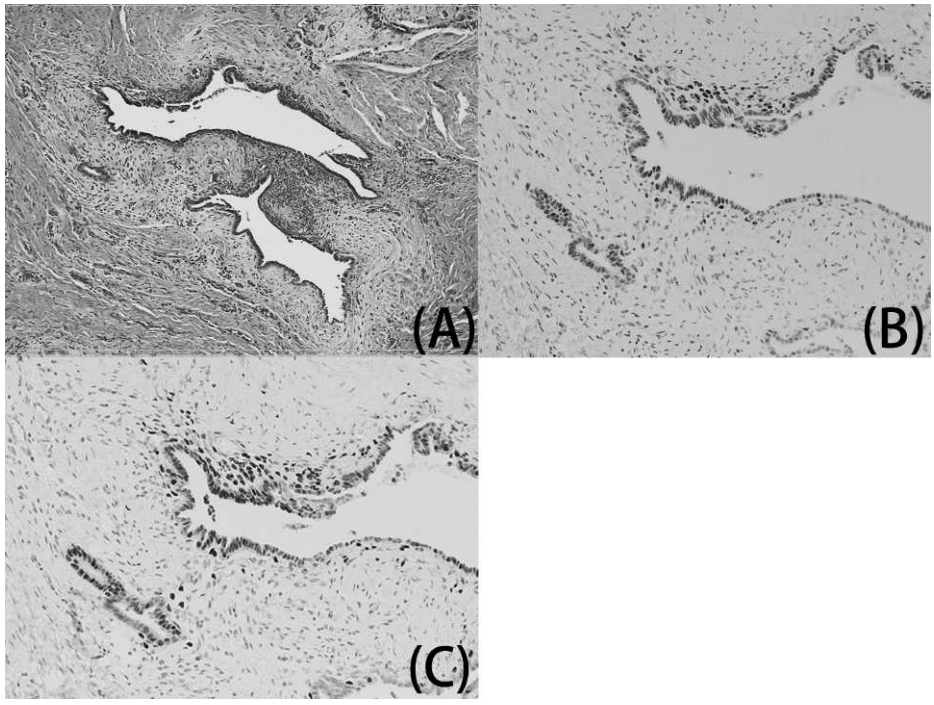

Fig. 3 (A) Hematoxylin and eosin staining of the removed tissue shows small and large cysts with fibrillar connective tissue and endometrial glands and interstitium (×100). Immunohistochemistry revealed that both the endometrial glands and interstitium were immunopositive for (B) estrogen receptors (×200) and (C) progesterone receptors (×200).

wall is uncommon.[5] Of all cases of endometriosis, 1.9% to 2.6% are reported in the abdominal wall. Horton *et al* reviewed 29 articles describing 455 such patients.[2] Endometriosis was most frequently associated with gynecologic operations, particularly following a Cesarian section or hysterectomy in 57% and 11% of cases, respectively.[2] However, 20% of patients had abdominal-wall endometriosis that did not arise in association with an abdominal scar. Endometriosis can be found associated with scars not associated with gynecologic operations, including acute appendicitis, but to our knowledge there have been no reports of this entity subsequent to surgery for bladder exstrophy.

In this case, the endometriosis might have been associated with the bladder exstrophy or with the primary lesion. Regarding the primary inguinal lesion, the reported initial sites are the uterine round ligament, the peritoneum of the inguinal hernia sac, and the peritoneum of a femoral hernia.[1] However, in this case, the lesion was not adherent to the round ligament, and there was no evidence of coexisting inguinal or femoral hernias, so an association with the bladder exstrophy should be considered.

Endometriosis occurring in the bladder is rare, affecting approximately 0.2% to 0.6% of all patients with endometriosis.[6–9] Multiple pathogenic causes for the ectopic occurrence of endometriosis have been proposed. They include (1) an endometrial implantation theory based on the reflux of menstrual blood through the oviducts; (2) a coelomic metaplasia theory involving differentiation in peritoneal mesothelial cells; (3) theories of lymphogenous or hematogenous routes involving lymphatic and vascular metastases; (4) a mechanical transplant theory involving the persistence of remnants of endometrium following a spontaneous or operative delivery; (5) an embryonic remnant theory based on residues of Wolffian or Müllerian ducts; (6) a composite theory based on a combination of the implantation and metastasis theories; and (7) a recent hypothesis based on the involvement of local immune factors. As for bladder endometriosis, its etiology and pathogenesis remain highly controversial, because the affected patients show features ranging from solitary lesions to concomitant nonvesical lesions; moreover, they might or might not have a history of gynecologic surgery. At least 3 different alternative pathogenetic mechanisms have

been hypothesized: (1) development from metaplasia of Müllerian duct remnants; (2) extension of adenomyotic lesions arising in the myometrium and subsequently invading the bladder; or (3) iatrogenic displacement of the decidua after gynecologic surgery, particularly after a Cesarean section.[10–12]

As for the other pathologic conditions including endometrial lesions, fewer than 20 cases of Müllerianosis of the bladder have been reported.[13–15] This rare entity is characterized by the presence of an admixture of at least 2 types of Müllerian tissue leading to endometriosis, endocervicosis, and endosalpingosis in the lamina propria and muscularis propria of the bladder. Müllerianosis can occur without any history of pelvic surgery and the histologic and immunohistochemical features support the idea of a metaplastic origin in embryonic duct remnants.[10]

Although we could not show any conclusive pathologic evidence of an association between bladder endometriosis and the bladder exstrophy in the present case, metaplasia from residual tissue during surgery for the bladder exstrophy or retention of embryonic duct remnants during organogenesis are possible etiologies. Future accumulation of similar cases would be helpful for clarification of this association between bladder exstrophy and endometriosis. It could also help clarify the pathogenesis of bladder endometriosis.

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