

Inguinal endometriosis in a patient without a previous history of gynecologic surgery

Da Hee Kim¹, Min Jung Kim¹, Mi-La Kim¹, Jong Taek Park¹, Ji Hyun Lee²Departments of ¹Obstetrics and Gynecology, ²General Surgery, CHA Gangnam Medical Center, CHA University, Seoul, Korea

Endometriosis, defined as growth of endometrial stroma and glands outside the uterine cavity, is a chronic and recurrent disease that affects patients' quality of life. Ectopic endometrial tissue can proliferate at any location in the body, but the pelvic organs and peritoneum are the most frequent implantation sites. Among extrapelvic endometriosis, inguinal endometriosis is a very rare gynecologic condition usually associated with previous pelvic surgery. Endometriosis should be preoperatively distinguished from other inguinal masses using computed tomography, magnetic resonance imaging, or ultrasonography. Here, we report a case of right inguinal endometriosis in a patient with no previous history of gynecologic surgery; in addition, we have provided a brief review of relevant literature.

Keywords: Endometriosis; Inguinal; Round ligament; Ultrasonography

Introduction

Endometriosis is defined as growth of endometrial tissue outside the uterine cavity. This condition has a prevalence of 10% to 20% in women of reproductive age, with occurrence mainly in the pelvic organs, particularly the ovaries, pouch of Douglas, and pelvic peritoneum [1]. Additionally, endometriosis can also occur in extrapelvic organs such as the gastrointestinal and urinary tracts, and the thoracic cavity, umbilicus, abdominal wall incisions, central nervous system, bone, episiotomy scars, and abdominal wall, and rarely, in the inguinal canal [2,3]. Candiani et al. [4] reviewed 958 patients who had endometriosis and reported that only 0.6% of patients had inguinal endometriosis. Due to its rarity, inguinal endometriosis is often misdiagnosed or overlooked. Its most common symptoms are palpable inguinal mass, often accompanied by change of mass size and inguinal pain related to menstrual cycle [2,4]. Most cases of inguinal endometriosis are associated with previous surgery in the uterine cavity [2,5]. In Korea, only 6 patients in 4 case reports have been reported to have inguinal endometriosis [6-9]. We report an additional case of right inguinal endometriosis in a patient with no previous history of gynecologic surgery; this patient was treated by a general surgeon using a wide excision.

Case report

A 40-year-old nulliparous woman presented with a painful mass in her right inguinal area. The mass had grown for 2 years and had occasionally been tender, especially during menstrual cycles. The patient had no specific medical or surgical history. She experienced regular menstruation at 30-day intervals and experienced menorrhagia. Her menstruation period lasted 7 days.

Physical examination revealed a non-erythematous swollen and tender mass measuring approximately 2×2 cm located lateral and superior to the pubic tubercle. The size

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Corresponding author: Mi-La Kim

Department of Obstetrics and Gynecology, CHA Gangnam Medical Center, CHA University, 566 Nonhyeon-ro, Gangnam-gu, Seoul 135-913, Korea

Tel: +82-2-3468-3677 Fax: +82-2-558-1112

E-mail: mila76@naver.com

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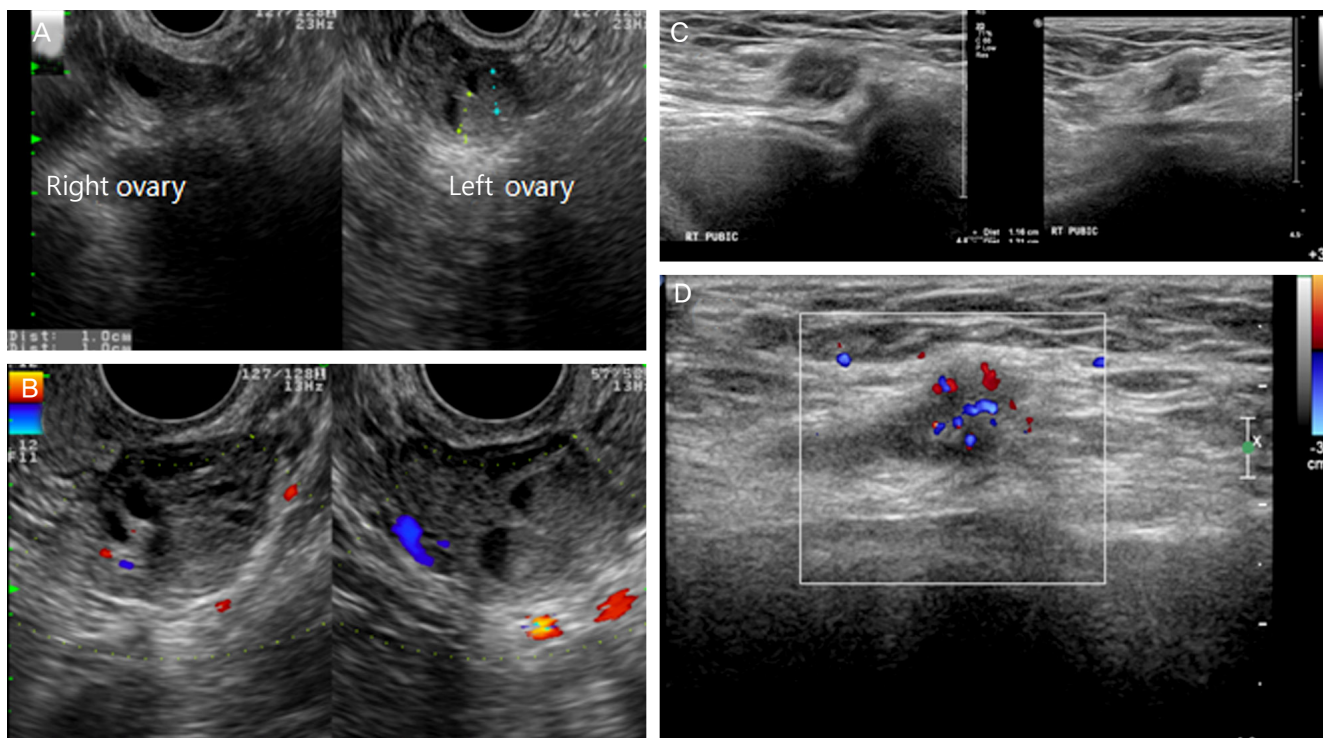


Fig. 1. Transvaginal ultrasonographic findings of the adnexa and inguinal mass. (A) Right ovary was grossly normal. Left ovary showed two low-echoic cysts. (B) Doppler transvaginal ultrasonographic findings of the left ovarian cyst. The longitudinal and transverse view showed two low-echoic cysts of 1.0 cm size. (C) Inguinal ultrasonographic findings of the inguinal mass. The longitudinal and transverse views show an ill-defined hypoechoic lesion (size, 1.1×1.2 cm). (D) On Doppler ultrasonographic findings showed increased vascularity of the mass in the right pubic area.

and position of the mass remained unchanged, even when the patient coughed or changed positions. The patient also reported intermittent vaginal spotting, for which she had undergone simultaneous transvaginal and inguinal ultrasonography.

The transvaginal ultrasonography revealed two left ovarian cysts measuring 1.0 cm that seemed to be endometrioma or hemorrhagic corpus luteal cysts (Fig. 1A, B). Inguinal ultrasonography showed an ill-defined hypoechoic lesion (size, 1.1×1.2 cm) in the right pubic area, with increased vascularity (Fig. 1C, D). She underwent repeated inguinal ultrasonography during her menstrual period. She felt tenderness and swelling of the lesion, but there was no change of mass size on inguinal ultrasonography. After being informed about the presence of hemangioma or arteriovenous malformation, the patient opted for surgical excision of the inguinal mass.

An oblique skin incision was performed along the line running from the pubic tubercle to the anterior superior iliac spine, at the superior margin of the inguinal ligament.

After identification of the femoral vessels, the hard mass was widely excised to include a part of the round ligament. Chocolate-colored fluid was drained from the hard cystic mass during the procedure, and the mass was connected with the intraperitoneal round ligament at the inguinal canal. The inguinal canal was opened and the extraperitoneal portion of the round ligament was resected by gentle traction and then cutting at the level of the internal inguinal ring. The proximal stump was fixed to the surrounding fibrous structure of the inguinal canal. Histological examination showed a cuff of endometrial stroma surrounding the endometrial glands with hemorrhaging from smooth muscle and fibrous tissues (Fig. 2). After 2 months of the surgery, the patient's pain completely resolved and she was fully recovered without complications. Considering the possibility of recurrence of the inguinal endometriosis and progression of the ovarian endometrioma, we recommended her to use the oral contraceptives and planned regular follow-up.

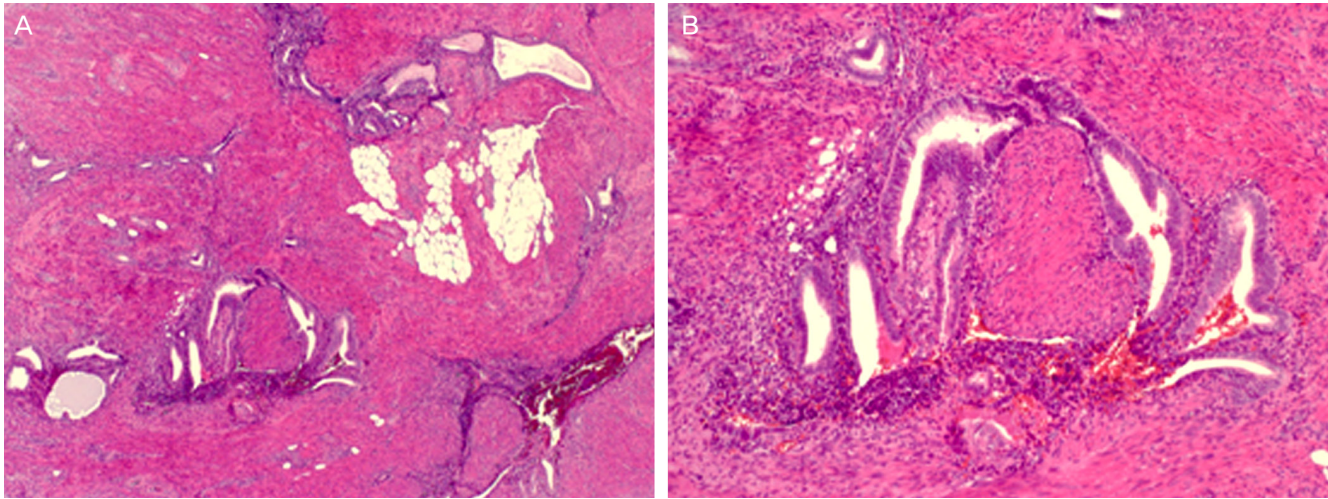


Fig. 2. Microscopic findings of the excised specimen of inguinal endometriosis. The endometrial glands are surrounded by a cuff of endometrial stroma, with hemorrhage involving smooth muscle and fibrous tissue in the round ligament. (A) H&E, $\times 40$; (B) H&E, $\times 200$.

Discussion

Inguinal endometriosis is a rare clinical condition occurring in only 0.6% of women [4]. Patients with inguinal endometriosis complain of inguinal mass and pain, in particular, acute pain during menstrual cycles [2,4].

Inguinal endometriosis is usually concomitant with pelvic endometriosis [4,10], suggesting a similar pathogenesis for both conditions. Clausen and Nielsen [11] suggested several possible theories, including implantation of endometrial tissue by transtubal regurgitation during the menstrual cycle, metastasis via venous or lymphatic channels, congenital hormonal activation of embryonal cells from the Müllerian duct, metaplasia of mesothelial cells, and direct extension from the pelvis along the round ligament. In the case of inguinal endometriosis, the right side is more commonly affected than the left, and is often associated with inguinal hernia, and its occurrence on both sides is rare [4,5,8,12]. The reasons for the right-predominance are unclear. Sun et al. [13] proposed two different theories: 1) the sigmoid colon protects the left inguinal canal and 2) endometrial cells remain on the right side for a longer period due to clockwise flow of intraperitoneal fluid. Our patient exhibited right-sided inguinal endometriosis, the most common presentation.

Differential diagnosis is essential in cases of suspected inguinal mass. Causes of inguinal mass include hernia, lymphadenopathy, neuroma, abscess, hydroceles, hematoma, lymphoma, lipoma, sarcoma, subcutaneous cysts, and cancer

[9]. Computed tomography (CT), magnetic resonance imaging (MRI), and ultrasonography are useful diagnostic tools. The CT appearance of inguinal endometriosis is often not specific [9,14], presenting as either solid, cystic, or complex (i.e., the same density as muscle). MRI is more accurate than CT. Gaeta et al. [14] described two MRI patterns for inguinal endometriosis. Type 1 pattern consist of cystic hyperintense lesions, on both T1- and T2-weighted images. Type 2 lesions have solid components, showing a high signal intensity on T1-weighted images and either hypointensity or moderate hyperintensity on T2-weighted images with some "shading signs" for cystic lesions [14]. The ultrasonographic findings of inguinal endometriosis are variable. Round or oval cystic masses, representing intracystic bleeding associated with menstruation are found in most cases, although homogeneously hypoechoic solid or combined cystic and solid masses have also been described [9]. The differential diagnosis of cystic masses includes inguinal hernia and hydrocele, and solid masses should be differentiated from sarcoma, lymphoma, hematoma, and abscesses. Since no available diagnostic imaging tools are specific for inguinal endometriosis, differential diagnosis based on imaging results must be combined with a careful review of the history of cyclic menstrual pain associated with inguinal mass [9,14]. Fine-needle aspiration cytology allows accurate preoperative diagnosis and exclusion of malignancy and can be useful in selected cases [9,15].

Patients with inguinal endometriosis may have a history of gynecologic or abdominal surgery [2,5,10]. In the absence

of a surgical history, however, inguinal endometriosis may be misdiagnosed as another groin diseases and managed by a general surgeon. In our case, the patient had no history of previous surgery, and ultrasonograph showed an ill-defined hypoechoic lesion with increased vascularity. We made a presumptive diagnosis of hemangioma or arteriovenous malformation and referred the patient to a general surgeon for treatment. If inguinal endometriosis been correctly diagnosed preoperatively, the patient could have undergone laparoscopic surgery rather than wide excision of the inguinal area. It is therefore important to consider inguinal endometriosis in the differential diagnosis in patients with inguinal mass, even if there is no history of gynecologic or abdominal surgery.

The treatment for inguinal endometriosis is radical surgical excision of the lesion and the extraperitoneal round ligament [12]. In patients with concomitant inguinal endometriosis and pelvic endometriosis, which occurs in most cases, either laparoscopy or laparotomy can be employed for the excision. However, no such associated disease is present, neither laparoscopy nor laparotomy is necessary [11,13]. Hormonal therapy is recommended in addition to surgery, as it plays an important role in preventing recurrence of endometriosis [7].

In conclusion, inguinal endometriosis is a very rare disease compared to pelvic endometriosis. Nonetheless, extrapelvic endometriosis should be considered in the differential diagnosis in cases in which women are of reproductive age and present with an inguinal mass, even in those without a history of gynecologic surgery. Pain associated with the menstrual cycle and characteristic ultrasonographic findings also are helpful for diagnosis.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

References

1. Goldman MB, Cramer DW. The epidemiology of endometriosis. *Prog Clin Biol Res* 1990;323:15-31.
2. Seydel AS, Sickel JZ, Warner ED, Sax HC. Extrapelvic endometriosis: diagnosis and treatment. *Am J Surg* 1996;171:239.
3. Cullen TS. Adenomyoma of the round ligament. *Bull Johns Hopkins Hosp* 1896;7:112-4.
4. Candiani GB, Vercellini P, Fedele L, Vendola N, Carinelli S, Scaglione V. Inguinal endometriosis: pathogenetic and clinical implications. *Obstet Gynecol* 1991;78:191-4.
5. Majeski J. Scar endometriosis manifested as a recurrent inguinal hernia. *South Med J* 2001;94:247-9.
6. Whang JD, Park CS, Bae DS, Lee JH, Noh JS, Kim JS, et al. Two cases of endometriosis in the extraperitoneal portion of the uterine round ligament. *Korean J Obstet Gynecol* 1999;42:189-93.
7. Lee SE, Jo DH, Moon SH, Chong HI, Shin SI, Kim HG, et al. A case of inguinal endometriosis in the absence of previous gynecologic surgery. *Korean J Obstet Gynecol* 2008;51:261-4.
8. Lim MC, Choi JY, Lee DO, Yoo JW, Park SY, Seo SS. Inguinal endometriosis connected to intraperitoneal round ligament: complete excision with extraperitoneal wide dissection. *Korean J Obstet Gynecol* 2008;51:1533-8.
9. Yang DM, Kim HC, Ryu JK, Lim JW, Kim GY. Sonographic findings of inguinal endometriosis. *J Ultrasound Med* 2010;29:105-10.
10. Wong WS, Lim CE, Luo X. Inguinal endometriosis: an uncommon differential diagnosis as an inguinal tumour. *ISRN Obstet Gynecol* 2011;2011:272159.
11. Clausen I, Nielsen KT. Endometriosis in the groin. *Int J Gynaecol Obstet* 1987;25:469-71.
12. Fedele L, Bianchi S, Frontino G, Zanconato G, Rubino T. Radical excision of inguinal endometriosis. *Obstet Gynecol* 2007;110(2 Pt 2):530-3.
13. Sun ZJ, Zhu L, Lang JH. A rare extrapelvic endometriosis: inguinal endometriosis. *J Reprod Med* 2010;55:62-6.
14. Gaeta M, Minutoli F, Mileto A, Racchiusa S, Donato R, Bottari A, et al. Nuck canal endometriosis: MR imaging findings and clinical features. *Abdom Imaging* 2010;35:737-41.
15. Catalina-Fernandez I, Lopez-Presa D, Saenz-Santamaria J. Fine needle aspiration cytology in cutaneous and subcutaneous endometriosis. *Acta Cytol* 2007;51:380-4.