A Case of Endometriosis in the Inguinal Region

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Abstract
A rare case of endometriosis in the inguinal region is reported.
A 33-year-old unmarried, nulliparous woman presented with a chief complaint of a mass and pain in the left inguinal region. A 10-mm mass with tenderness was noted in the left inguinal region. The patient reported worsening conditions during menstruation. MRI demonstrated a 10-mm mass in the left inguinal region showing low signal intensity on T1-weighted image and high signal intensity on T2-weighted image. Surgery was performed under a diagnosis of inguinal mass with endometriosis. During surgery, a 10-mm mass was found near the external inguinal ring along the round ligament of the uterus. On pathological examination, the mass demonstrated the presence of endometrial tissue and was diagnosed as external endometriosis.

Key words: endometriosis, inguinal region

Introduction
Endometriosis is a benign proliferative lesion in which ectopic endometrium or endometrium-like tissue is observed, and is roughly divided, according to the site of development, into internal endometriosis occurring in the uterus and external endometriosis occurring outside the uterus. Cases of endometriosis in the inguinal region are uncommon, accounting only for 0.8% of all cases of external endometriosis1). In this report, we describe a case of endometriosis in the inguinal region with a review of the literature.

Case
The patient was an unmarried, nulliparous woman aged 33 years. She had noted swelling and pain in the left inguinal region for 2 years, with repeated improvement and worsening of the symptoms, and presented to our hospital in suspicion of inguinal hernia.

A hard mass with a diameter of 10-mm was found in the left inguinal region. The mass was slightly movable, but could not be reduced by compression, which caused tenderness. Enlargement of the mass and increase in pain were observed during menstruation. No abnormalities were found in complete blood cell counting or gen-
eral serum chemistry.

Pelvic MRI reveals a 10-mm mass with low signal intensity on a T1-weighted image and high signal intensity on a T2-weighted image was detected in the left inguinal region (Fig. 1a, b). During observation, pain continued to recur periodically and showed no improvement. Surgery was then performed with a diagnosis of inguinal mass with endometriosis and performed according to the standard procedure for repairing inguinal hernia. After the external oblique aponeurosis was incised and the inguinal canal was opened, a 10-mm cystic mass was found near the external inguinal ring along the round ligament of the uterus. The mass was removed and surgery was completed. Cutting the removed mass revealed accumulation of wine-colored fluid with blood components inside the mass.

Pathological findings: Histological examination of resected specimen showed columnar epithelia with spindle-shaped nuclei were observed. The columnar epithelia were surrounded by aggregations of round cells that appeared like endometrial stromal cells (Fig. 2a). Evidence of hemorrhage was observed in the connective tissue, suggesting the presence of intracystic hemorrhage. On immunohistochemistry, positive staining for estrogen receptor was observed in the nuclei of the columnar epithelia and some of the surrounding stromal cells. The stromal cells were also positive for CD10 in their cytoplasm (Fig. 2b, c). These findings led to a diagnosis of endometrial cyst arising from endometriosis.

The patient was discharged from hospital 2 days after surgery. On examination 2 months postoperatively, no mass was found, but left inguinal pain coinciding with the menstrual period was observed. The presence of residual endometrial tissue was suspected. Since the pain was not as intense as it was before surgery and controllable with analgesics, the patient has been simply placed under observation.

Discussion

Endometriosis is a benign proliferative lesion in which ectopic endometrium or endometrium-like tissue is observed, and is roughly divided into internal endometriosis occurring in the uterus and external endometriosis occurring outside the uterus. It may cause dysmenorrhea, coital pain and infertility, and has been reported to affect 8-15% of all fertile women. External endometriosis commonly occurs in the following areas, in descending order of incidence, ovary (38%), pelvic peritoneum/Douglas’ pouch (21%), rectum/colon (14%), uterosacral ligament (8.1%), and fallopian tube (7.5%), while it rarely occurs in the inguinal region (0.8%). The most common cause of endometriosis is believed to be regurgitation of endometrial tissue through the fallopian tube during menstruation and subsequent dissemination into the peritoneal cavity. Other suggested mechanisms include hematogenous dissemination of endometrial tissue, alteration of Mullerian duct-derived fetal cells and metaplasia of mesothelium cell.

The first case of inguinal endometriosis was reported by Cullen in 1896. Our search of the Japa-
na Centra Revuo Medicina database with keywords “endometriosis” and “inguinal” identified 61 reports over 27 years between 1983 and 2009. Although it occurred in the left inguinal region in the present case, a large majority (76.1%) of the Japanese cases of endometriosis occurred in the right inguinal region. The most widely accepted theory for this laterality is as follows. Body fluid circulates in the clockwise direction in the peritoneal cavity and flows through the pelvic floor and the right lower abdominal region toward the head. Endometrial tissue flowing with this fluid may be retained in the right lower abdominal region during this process.

Inguinal endometriosis rarely occurs in girls who have not reached menarche and postmenopausal women. It commonly manifests as a mass showing an increase in size and pain during menstruation. It is thought that the onset of inguinal endometriosis is preceded by pelvic endometriosis, which extends through the round ligament of the uterus into the inguinal canal. Some cases of inguinal endometriosis associated with inguinal hernia have also been reported. In the present case, although no clinical signs of pelvic endometriosis were observed, close examination was deemed necessary considering the possibility of later development of the condition.

For the diagnosis of inguinal endometriosis, imaging modalities, such as ultrasonography, CT, and MRI are useful in determining the presence and location of a mass while clinical findings, such as changes in the size of the mass and the intensity of pain associated with menstruation, are important for qualitative diagnosis. Fine-needle aspiration cytology (FNAC) has been shown to be effective for definitive diagnosis, but is also associated with the risk of causing mechanical transfer of endometrial tissue. We did not perform FNAC in the present case.

Treatment includes hormone therapy and surgical therapy. The commonly used hormone therapies are 1) pseudo-pregnancy therapy consisting of continuous administration of estrogen and syn-

Figure 2 Columnar epithelia with spindle-shaped nuclei were observed in the inner wall of the removed mass. The columnar epithelia were surrounded by aggregations of round cells that appeared like endometrial stromal cells (a) (HE × 200). Positive staining for estrogen receptor was observed in the nuclei of the columnar epithelia and some of the surrounding stromal cells (b). The stromal cells were also positive for CD10 in their cytoplasm (c).
thesized gestagen, 2) pseudo-menopausal therapy using gonadotropin-releasing hormone analogue, 3) gestagen therapy consisting of continuous administration of progestin and 4) danazol therapy that uses danazol that is conductor of male hormon. However, for young women who wish to be pregnant and to have a baby, hormone therapies are not suitable and complete removal of a mass should be selected. In the present case, pain coinciding with menstruation period recurred 2 months postoperatively. Since no rupture of the mass occurred and complete removal of the mass was considered to be achieved during surgery, we speculated that endometrial tissue had been extended to the surrounding tissues. With no means to macroscopically confirm complete removal of the mass, it is difficult to determine the presence or absence of residual endometrial tissue. Removal of a mass including the round ligament of the uterus or surrounding subcutaneous fat may be effective in preventing residual endometrial tissue.

**Conclusion**

We encountered and report here a case of endometriosis in the left inguinal region.

The possibility of inguinal endometriosis should be considered when diagnosing and treating a mass with pain coinciding with the menstruation period.

**References**

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和文抄録

亀径部に発生した稀な子宮内膜症の症例を経験したので報告する。
症例は未婚・未経産婦の33歳の女性。左亀径部の腫瘤と疼痛を主訴に外来受診。左亀径部に直径10mmの圧痛を伴う腫瘤を認め、月経時に症状の増悪を認めた。子宮内膜症を合併した亀径部腫瘤の診断で手術を行った。手術所見では外亀径輪の近傍に直径10mm大の腫瘤を認めた。病理組織学的検討では組織内に子宮内膜組織が混在しており外性子宮内膜症と診断した。

キーワード：亀径部腫瘤、子宮内膜症