Case report

Radical resection of an endometrioid carcinoma arising from endometriosis in the round ligament within the right canal of Nuck: a case report and literature review

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1. Introduction

Since Sampson (1925) first reported the malignant transformation of ovarian chocolate cysts, there have been many reports on malignant transformation of endometriosis. Most malignant tumors associated with endometriosis occur in the pelvic cavity; however, 2.5–6.0% of these tumors were observed at extrapelvic sites (Irvin et al., 1998). The inguinal region was reported to be associated in 1.8% of cases of malignant transformation of endometriosis (Irvin et al., 1998). Among these cases, only 4 were associated with the canal of Nuck.

The canal of Nuck is an embryological remnant in females, which is analogous to a patent process vaginalis in males. The entire fold is normally obliterated within the first year of life in female infants. Occasionally, the canal of Nuck remains patent and permits the seeding of endometriotic tissue in inguinal soft tissues (Irvin et al., 1998; Ito et al., 2010; Mesko et al., 1988; Sun et al., 1979).

Here, we present a case of an endometrioid carcinoma arising from endometriosis in the round ligament within the canal of Nuck and coexisting with a hydrocele of the canal of Nuck. The patient showed favorable prognosis with radical resection of the tumor and adjuvant chemotherapy.

2. Case report

A 40-year-old Japanese woman (gravida 2, para 2) was aware of a gradually enlarging nodule on the right side of her pubis for 3 years. She visited a hospital because of pain and bleeding at the nodule, and she was referred to Kumamoto University Hospital for suspicion of malignancy.

She had a regular 28-day menstrual cycle with no history of hormonal therapy and had no particular medical history. With regard to family history, her paternal uncle had gastric cancer and her paternal aunt had lung cancer.

At the initial examination, a 6-cm mass with an irregular surface was observed on the right side of the pubis. No abnormal findings were identified in the pelvic cavity on bimanual examination. The result of Pap smear was negative for intraepithelial lesion or malignancy, and the endometrial cytology was negative for malignancy.

Biopsy of the inguinal mass indicated an adenocarcinoma composed of columnar cells. The tumor cells were immunohistochemically positive for cytokeratin 7, estrogen receptor, and progesterone receptor, and negative for cytokeratin 20 and gross cystic disease fluid protein 15. These findings suggested that the tumor might have originated in tissues associated with the female genital tract. Pelvic magnetic resonance imaging (MRI) identified a 6-cm solid mass with a 3-cm cystic component in the right inguinal region. The MRI scan revealed that the tumor invaded into the right pectineal, rectus abdominis, and oblique abdominal muscles, and had continuity with the round ligament in the right inguinal canal (Fig. 1A–D). No remarkable abnormalities were
found in the uterus and the bilateral ovaries. Positron emission tomography/computed tomography indicated metastasis to the right inguinal lymph node, but no distant parenchymal metastasis was observed. The serum CA 125 level was 226.8 U/ml and CA 19–9 level was 67.7 U/ml, whereas the CEA level was within the normal range. Based on these findings, the tumor was suspected to be an adenocarcinoma arising from the right round ligament, possibly associated with endometriosis.

*En bloc* resection of the inguinal tumor and the invaded surrounding tissues was performed through partial radical vulvectomy, clitoridectomy, and resection of the right pectineal muscles, rectus abdominis muscles, oblique abdominal muscles, inguinal ligament, and round ligament. Additionally, the bilateral inguinal lymph nodes were resected *en bloc* together with exploratory laparotomy. Laparotomy revealed no macroscopic abnormal findings in the uterus, bilateral fallopian tubes, or ovaries. In addition, the lesions of endometriosis were not identified in the pelvic peritoneum. Intraoperative pathological assessment revealed metastasis to the left deep inguinal lymph nodes; therefore, bilateral pelvic lymph node dissection was performed. Finally, reconstruction was performed with a rectus abdominis myocutaneous flap for the right inguinal area and a sartorius muscle flap for the left inguinal area.

The resected specimen showed a 6-cm yellow solid mass where the right round ligament ended. A 3-cm cyst containing white fluid was located at the lateral distal site of the end of the right round ligament. Microscopic examination revealed the inguinal tumor composed of well-differentiated endometrioid carcinoma, associated with endometriosis on the round ligament within the right canal of Nuck (Fig. 2B, C). The cyst wall was covered by atypical columnar cells and has a bundle of collagen fibers (D: arrowhead). B, C, D: Hematoxylin-eosin staining, scale bars, 200 μm.
Table 1

| Case | Author, year | Age | Site | Symptoms | Treatment | Histology | Follow-up | Histology of the disease | Treatment |
|------|--------------|-----|------|----------|-----------|-----------|-----------|-------------------------|-----------|
| 1    | Sun et al. (1997) | 64  | Right inguinal | Enlargement of a mass | Excision | Primary low grade endometrioid carcinoma | Alive with no evidence of the disease at 1 year | 10/1/2014 | Endometrioid carcinoma, right inguinal lymphadenectomy, RT + ILND, right salpingo-oophorectomy |
| 2    | Mesko et al. (1988) | 57  | Right inguinal | Enlargement of a mass | Excision, exploratory laparotomy, resection and ILND | Endometrioid adenocarcinoma | Alive with no evidence of disease 20 months after the third surgery | 10/1/2014 | Endometrioid adenocarcinoma, left omentectomy, left salpingo-oophorectomy, right salpingo-oophorectomy, right salpingectomy |
| 3    | Irvin et al. | 40  | Right inguinal | Enlargement of a mass | Excision, exploratory laparotomy, re-excision and ILND after 3 months, RT | Endometrial sarcoma | Alive with no evidence of disease 6 years after the first surgery | 10/1/2014 | Endometrioid carcinoma, right inguinal lymphadenectomy, RT + ILND, inguinal lymph node dissection |
| 4    | Ko et al. | 50  | Right inguinal | Enlargement of a mass | Excision | Endometrioid adenocarcinoma | Alive with no evidence of disease 20 months after the third surgery | 10/1/2014 | Endometrioid carcinoma, left omentectomy, left salpingo-oophorectomy, right salpingo-oophorectomy |
| 5    | Our case, 2017 | 40  | Right inguinal | Painful mass | Excision, ILND, PLND, reconstruction of inguinal region with the rectus abdominis myocutaneous flap, OMT | Ovarian cancer was also found 3 months after the second surgery, alive with no evidence of disease | 10/1/2014 | Endometrioid carcinoma, left omentectomy, left salpingo-oophorectomy, right salpingo-oophorectomy, right salpingectomy |

IA FIGO, well differentiated (Grade1) endometrioid carcinoma; LS, left salpingectomy; RSO, right salpingo-oophorectomy; OMT, omentectomy.
Extragonadal endometriosis was good with a 5-year survival rate of 100% (Irvin et al., 1998). However, with disseminated intraperitoneal disease, the prognosis was very poor, with a 5-year overall survival rate of 12% (Irvin et al., 1998). Among the 4 previous cases of malignant transformation of endometriosis within the canal of Nuck, all underwent excision of the tumor and no case received adjuvant chemotherapy. Although they showed no invasion to tissues around the canal of Nuck, 2 of the 4 cases (Table 1, case 1, 2) showed recurrence in the inguinal region at 3 months and 3 years after the first surgery, respectively. Additionally, 2 of the 4 cases (Table 1, case 2, 3) showed lung metastasis at 21 months and 2 years after the first surgery, respectively (Irvin et al., 1998; Ito et al., 2010; Mesko et al., 1988; Sun et al., 1979).

In the present case, metachronous occurrence of endometriosis-associated malignancy was confirmed pathologically. When endometriosis-associated malignancy is suspected, clinicians should consider the possibility that persisting endometriosis can contribute to the development of additional cancer in the future. On the basis of these considerations, clinicians need to provide precise information of the patient and determine an appropriate treatment strategy.

Here, we reported a rare case of an advanced endometrioid carcinoma arising from endometriosis in the round ligament within the canal of Nuck, along with a hydrocele in the canal of Nuck. Radical resection and adjuvant chemotherapy were effective.

Ethical consent

Consent was obtained from the patient for publication of this case report.

Conflict of interest statement

The authors have no conflicts of interest to declare.

References

Anderson, C.C., et al., 1995. Hydrocele of the canal of Nusk: ultrasound appearance. Am. Surg. 61, 959–961.
Bergamini, A., et al., 2014. Endometriosis-associated tumor at the inguinal site: report of a case diagnosed during pregnancy and literature review. J. Obstet. Gynecol. 40, 1132–1136.
Candiani, G.B., et al., 1991. Inguinal endometriosis: pathogenetic and clinical implications. Obstet. Gynecol. 78, 191–194.
Clausen, I., Nielsen, K.T., 1987. Endometriosis in the groin. Int. J. Gynaecol. Obstet. 25, 469–471.
Goldman, M.B., Cramer, D.W., 1990. The epidemiology of endometriosis. Prog. Clin. Biol. Res. 323, 15–31.
Huang, C.S., et al., 2003. The presentation of asymptomatic palpable movable mass in female inguinal gernia. Eur. J. Pediatr. 162, 493–495.
Irvin, W., et al., 1998. Endometrial stromal sarcoma of the vulva arising in extraovarian endometriosis: a case report and literature review. Gynecol. Oncol. 71, 313–316.
Ito, M., et al., 2010. A case of primary endometrioid adenocarcinoma arising from the hydrocele of the canal of Nuck. J. Jpn. Surg. 71, 2145–2149.
Kobayashi, H., et al., 2007. Risk of developing ovarian cancer among women with ovarian endometrioma: a cohort study in Shizuoka, Japan. Int. J. Gynecol. Cancer 17, 43–43.
Matter, M., et al., 2003. Cystadenocarcinoma of the abdominal wall following caesarean section: case report and review of the literature. Gynecol. Oncol. 91, 438–443.
Mesko, J.D., et al., 1988. Clear cell (“Mesonephroid”) adenocarcinoma of the vulva arising in endometriosis: a case report. Gynecol. Oncol. 29, 385–391.
Sampson, J.A., 1925. Endometrial carcinoma of the ovary, arising in endometrial tissue in that organ. Arch. Surg. 10, 1–72.
Scott, R.B., 1953. Malignant change in endometriosis. Obstet. Gynecol. 2, 283–289.
Sun, C.J., et al., 1979. Primary low grade adenocarcinoma occurring in the inguinal region. Cancer 44, 340–345.
Sun, Z.J., et al., 2010. A rare extrapelvic endometriosis: inguinal endometriosis. J. Reprod. Med. 55, 62–66.