



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

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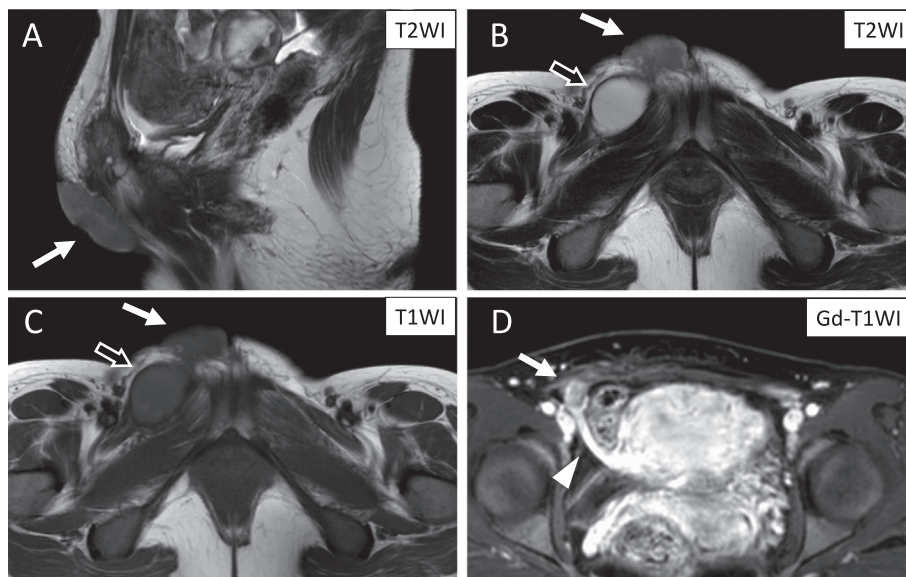


Fig. 1. Contrast-enhanced pelvic magnetic resonance imaging. A 6-cm solid mass (A, B, C, D: white arrow) is seen in the right inguinal region. A cystic component is located at the lateral caudal site of the solid mass (B, C: black arrow), with a thin, regular, smoothly demarcated wall. The solid mass is thought to have continuity with the round ligament (D: arrowhead) in the right inguinal canal. (A: sagittal section, T2-weighted image; B: transverse section, T2-weighted image; C: transverse section, T1-weighted image; D: transverse section, contrast-enhanced T1-weighted image).

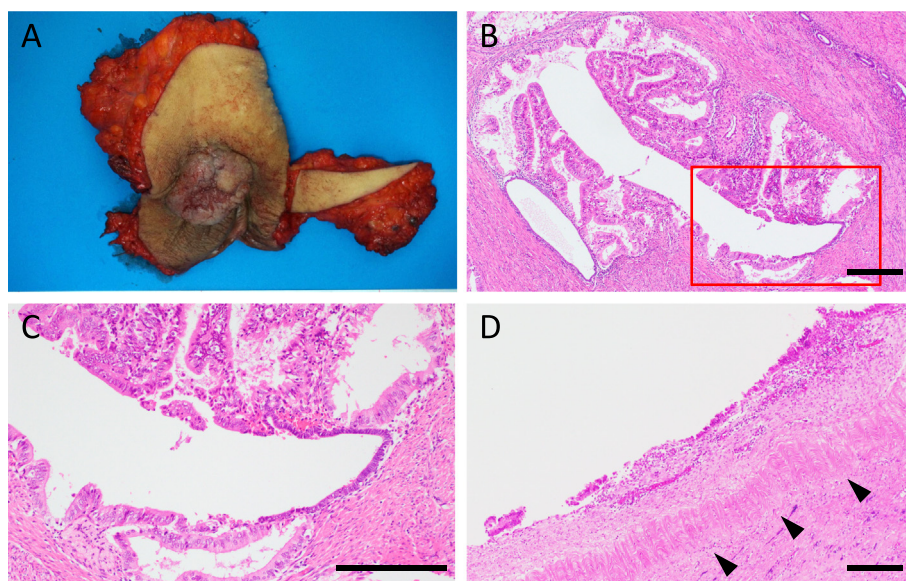


Fig. 2. Macroscopic and microscopic findings of the resected inguinal tissue. The tumor was resected *en bloc* with invaded surrounding tissues (A). Microscopically, a well-differentiated endometrioid carcinoma is observed in continuity with the endometriotic foci in the round ligament within the right canal of Nuck (B, C). The cyst wall is covered with atypical columnar cells and has a bundle of collagen fibers (D: arrowhead). B, C, D: Hematoxylin-eosin staining, scale bars, 200 μm .

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 in the right canal of Nuck (Fig. 2B, C). The cyst wall was covered by atypical columnar cells and did not contain normal epithelium or mesothelium. Well-organized bundles of collagen fibers arranged in a parallel fashion were also observed in the superficial portion of the cyst wall, indicative of its peritoneal-origin. Based on these findings, the tumor was considered to have involved hydrocele of the canal of Nuck (Fig. 2D). Metastatic tumor was identified in the both inguinal and left external iliac lymph nodes. Margins of the surgical specimens were uninvolved by the tumor. Consequently, the diagnosis of endometrioid carcinoma, associated with endometriosis in the right round ligament within the canal of Nuck, was rendered. Adjuvant chemotherapy with paclitaxel (175 mg/m²) and carboplatin (area under the curve, 6) was administered.

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