International Cancer Conference Journal (2024) 13:68–73
https://doi.org/10.1007/s13691-023-00639-1

CASE REPORT - PATHOLOGY

Endometrioid adenocarcinoma arising from inguinal endometriosis during follow-up for gastric cancer surgery: a case report and literature review

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Received: 2 October 2023 / Accepted: 30 October 2023 / Published online: 23 November 2023

Abstract

Malignant transformation of inguinal endometriosis is rare. A 56-year-old woman underwent surgery for advanced gastric cancer 5 years ago and received postoperative adjuvant chemotherapy. She had no recurrence since then. However, 5 years after surgery, contrast-enhanced computed tomography (CT) showed a mass in the right inguinal region suspected to be a hydrocele of the canal of Nuck, with a blood test showing a slightly elevated CA19-9 level (63.0 U/mL). Six months later, CT showed an enlarged mass in the right inguinal region and inflammation in the surrounding area. In addition, both inguinal lymph nodes and those in the right iliac artery area were enlarged, suggesting the possibility of malignancy. For diagnostic purposes, a right inguinal mass was excised. Histopathological examination revealed that it was endometrioid adenocarcinoma with ectopic endometriois as the origin. The differential diagnoses for inguinal masses in women include an inguinal hernia, hydrocele of the canal of Nuck, ectopic endometriosis, lymphoma, and metastatic malignancy. The presence of a primary malignancy in the inguinal region is sporadic but must be differentiated. This is the first case of malignant transformation of inguinal endometriosis developed during postoperative follow-up of another cancer.

Keywords Inguinal tumor · Inguinal endometriosis · Adenocarcinoma from endometriosis

Abbreviations

CT	Computed tomography
PET	Positron emission tomography
FDG	Fluorodeoxyglucose
SUV	Standardized uptake value
ER	Estrogen receptor
PgR	Progesterone receptor

Introduction

Endometriosis is common in women of reproductive age, with its lesions generally locating in the ovaries, myometrium, and pelvic peritoneum. However, these lesions can also be detected in the extrapelvic space, including the lungs, intestines, and skin. Inguinal endometriosis has been reported to account for less than 1% of endometriosis cases [1, 2]. Inguinal endometriosis is most commonly diagnosed by an inguinal mass. Preoperative diagnosis is difficult in the absence of symptoms associated with menstruation, and the diagnosis is often made by resection, rarely by finding a malignant tumor. Reports have shown that less than 1% of endometriosis cases develop malignant transformation [3, 4]. Among them, there are even fewer reports of malignant transformation of inguinal endometriosis, and there is no established treatment. In this report, we describe a case of endometrioid adenocarcinoma arising from inguinal endometriosis during postoperative follow-up for gastric cancer.

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Case report

A 56-year-old woman underwent surgery for advanced gastric cancer 5 years ago. The patient was treated with postoperative adjuvant chemotherapy and had no recurrence. Five years after surgery, contrast-enhanced computed tomography (CT) showed a mass in the right inguinal region suspected to be a hydrocele of canal of Nuck (Fig. 1A), with blood test showing a slightly elevated CA19-9 level (63.0 U/mL). No other obvious findings suggested recurrence. Three months later, CT showed no change in size (Fig. 1B), and CA19-9 remained unchanged at 65.4 U/mL. Three months later, CT showed an enlarged right inguinal mass and inflammation in the surrounding area (Fig. 1C), with an elevated CA19-9 level (171.7 U/mL). Furthermore, inguinal lymph nodes and those in the right iliac artery area were enlarged, indicating the possibility of malignancy. She was also aware of a mass, and palpation revealed a hard mass with poor mobility. Positron emission tomography (PET)-CT showed fluorodeoxyglucose (FDG) accumulation in the right inguinal mass (SUVmax 8.3) (Fig. 1D), both enlarged inguinal lymph nodes (Fig. 2A), lymph nodes in the right iliac artery region (SUVmax 2.6–5.0) (Fig. 2B, C), and the subcutaneous vulvar region (SUVmax 5.0) (Fig. 2D). No other obvious accumulations had been noted. For differential diagnosis, we considered gastric cancer metastasis, lymphoma, and other malignant tumors, which prompted us to perform a right inguinal mass excision for diagnostic purposes. Pathological examination of the excised specimen revealed proliferation of the cylindrical atypical glandular epithelium forming papillary and tubular structures within a fibrous stroma. A poorly atypical cuboidal glandular epithelium was observed contiguous to the tumor, while poorly atypical endometriosis with endomembranous stromal cells was observed at the margins (Fig. 3A). Immunostaining of the periductal stroma revealed positivity for CD10 (Fig. 3B), suggesting an endometriosis origin, and PAX8 (Fig. 3C), suggesting a Mullein duct origin. Other immunohistochemistry tests at the same tumor site showed positivity for p53 and partial positivity for ER and PgR. Based on the immunostaining results, a diagnosis of endometroid adenocarcinoma with an ectopic endometriosis as the primary site of origin was established. The patient was then referred to the obstetrics and gynecology department and underwent pelvic lymph node dissection, paraaortic lymph node dissection, inguinal lymph node dissection, and vulvar mass excision for both inguinal lymph nodes, lymph node metastasis in the right iliac artery region, and vulvar mass. There were no other findings in the abdominal cavity indicative of endometriosis. Twenty-four lymph nodes were removed, with pathological examination confirming that all were adenocarcinomas.

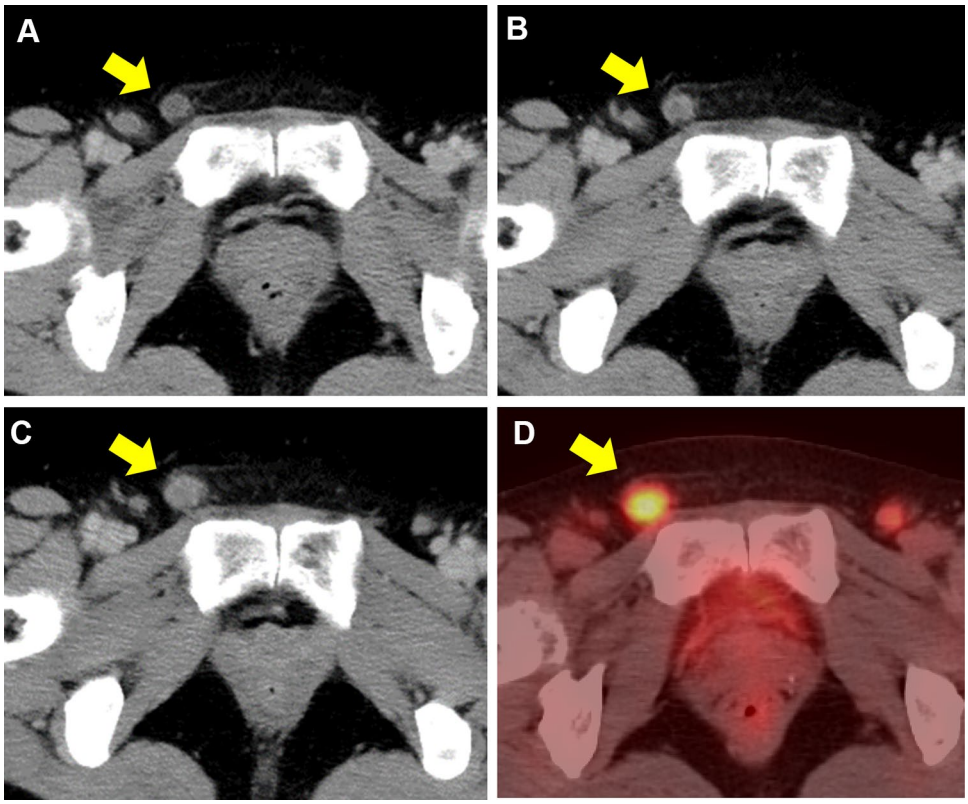

Fig. 1 **a**–**c** Computed tomography showing a right inguinal mass (yellow arrow). **a** Initially noted, **b** 3 months later, **c** 6 months later. **d** Fluorodeoxyglucose-positron emission tomography-computed tomography showing a right inguinal mass (yellow arrow) 6 months later

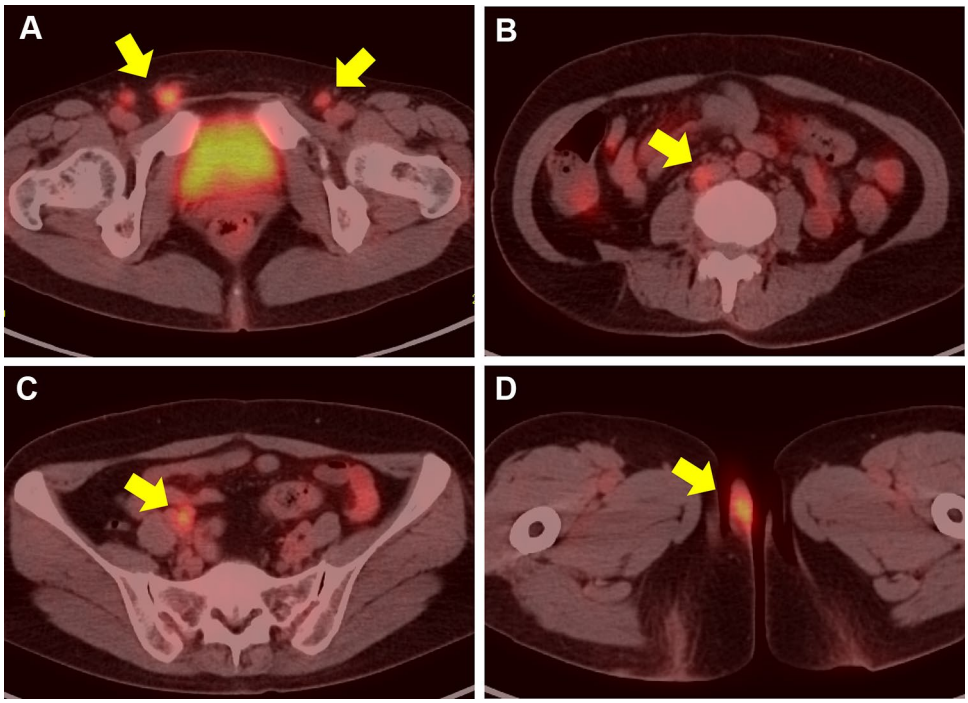

Fig. 2 **a**–**d** Fluorodeoxyglucose-positron emission tomography-computed tomography showing an inguinal and vulvar mass and lymph node metastasis (yellow arrow) 6 months later initially noted

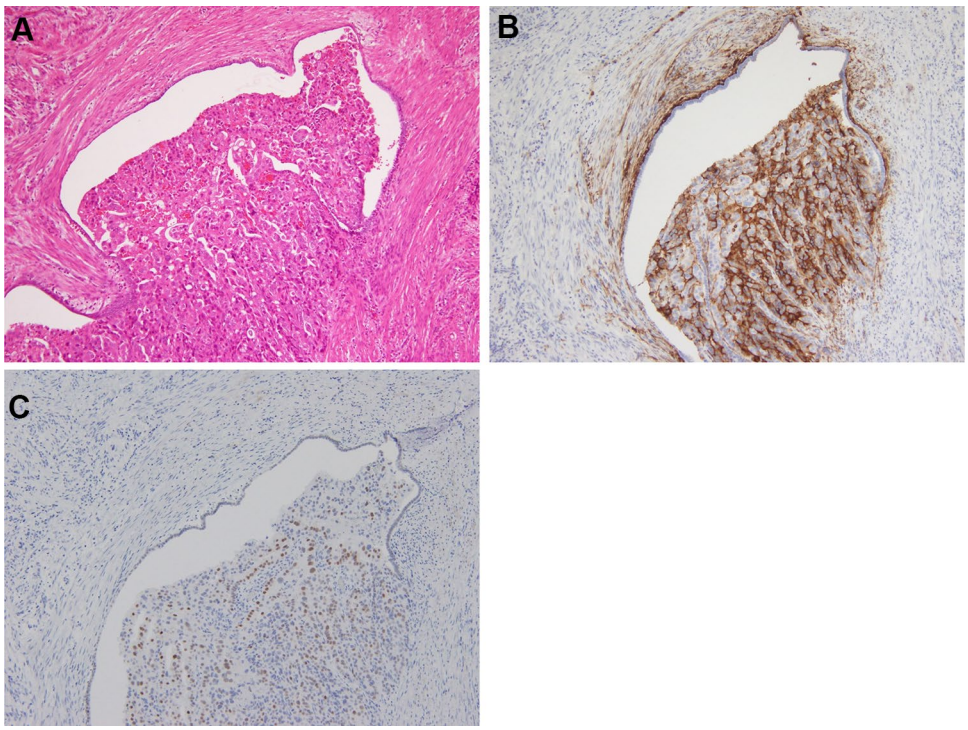

Fig. 3 Microscopic findings of the resected right inguinal tissue. **a** Hematoxylin–eosin staining × 100. **b** CD10 immunostaining × 100, **c** PAX8 immunostaining × 100

Postoperative chemotherapy with paclitaxel/carboplatin was started. However, after treatment initiation, the patient developed pelvic lymph node recurrence 6 months after surgery and underwent lymph node dissection, abdominal total hysterectomy, bilateral salpingo-oophorectomy, and radiotherapy, followed by resumption of chemotherapy.

Discussion

We herein detail our experience with a patient who developed endometrioid adenocarcinoma of endometriosis origin in the inguinal region during her 5-years follow-up

Table 1 Malignant tumors arising from inguinal endometriosis: literature review

Author, year	Age	Site	Symptoms	Treatment	Histology	Follow-up
Mesko et al. 1988	57	Right	Mass	Excision, re-excision [a]LND after 3 months and [b]RT	Clear cell adenocarcinoma	Local recurrence at 3 months, alive with probable pulmonary metastasis at 2 years
Ahn and Scolly et al. 1991	43	Right	Mass	Excision, right groin dissection	Clear cell adenocarcinoma	Right retroperitoneal recurrence after 10 years, treated with surgery and RT [c]DOD at 11 years and 2 months
	68	Left	Mass	Excision	Clear cell adenocarcinoma	[d]NED at 4 years and 8 months
Elemenoglou et al. 1993	52	Right	Mass	Excision, [f]TAH, [g]BSO, and LND, and adjuvant [e]CT	Clear cell adenocarcinoma	DOD 6 months after resection
Irvin et al. 1998	34	Left	Mass	Excision	Endometrioid stromal sarcoma	Lung metastasis at 21 months, treated with surgery. Focal recurrence in lungs at 9 months after the second surgery and treated with surgery. NED 6 years after the first surgery
Klein et al. 1999	67	Right	Mass	Excision and RT	Clear cell adenocarcinoma	Local recurrence at 2 years, treated with surgery. NED at 34 months after diagnosis
Slomowitz et al. 2002	34	Right	Mass	Excision and staging laparoscopy, LND and adjuvant CT	Serous adenocarcinoma	NED at 3 years after surgery
Milam et al. 2006	47	Right	Mass	Excision	Adenosarcoma	NED at 12 months
Bergamini et al. 2014	34	Left	Mass	Excision, staging laparoscopy, LND, and adjuvant CT	Endometrioid adenocarcinoma	NED at 10 months
Motooka et al. 2018	40	Right	Mass	Excision and LND, and adjuvant CT	Endometrioid adenocarcinoma	Endometrial cancer was found 12 months after the first surgery, ovarian cancer was also found 3 months after the second surgery, NED at 20 months after the third surgery
Yoshida et al. 2018	53	Right	Mass	Excision, TAH, BSO, and LND, and adjuvant CT	Clear cell adenocarcinoma	NED at 20 months after surgery
Gakuhara et al. (present case) 2023	56	Right	Mass	Excision and LND, and adjuvant CT	Endometrioid adenocarcinoma	Lymph node reoccurrence at 6 months and treated with LND, TAH, BSO, RT and chemotherapy

[a]LND: lymph node dissection

[b]RT: radiation therapy

[c]DOD: died of disease

[d]NED: no evidence of disease

[e]CT: chemotherapy

[f]TAH: abdominal total hysterectomy

[g]BSO: bilateral salpingo-oophorectomy

after having undergone surgery for gastric cancer. During her follow-up after gastric cancer surgery, an inguinal mass was noted, with CT findings suggesting a hydrocele of the canal of Nuck as a possible diagnosis, which prompted follow-up. FDG-PET-CT was useful in identifying malignant tumors. The differential diagnoses for inguinal masses in women include an inguinal hernia, hydrocele of the canal of Nuck, ectopic endometriosis, lymphoma, and metastatic malignancy.

The presence of a primary malignancy in the inguinal region is extremely rare but must be differentiated. In fact, previous studies have reported that various malignancies, including gastric cancer, can cause inguinal metastasis and seeding within the inguinal hernia sac [5–7]. In both cases, treatment consisted of a diagnostic and therapeutic resection biopsy, followed by carcinoma-specific treatment. In the present case, given that the patient was undergoing follow-up examination after her surgery for gastric cancer, an excisional biopsy was performed on suspicion of gastric cancer metastasis, but different results were obtained, highlighting the need for pathological examination with other differential diagnosis in mind. This is the first case of malignant transformation of inguinal endometriosis developed during postoperative follow-up of another cancer.

Endometriosis is a disease in which the endometrial gland epithelium with the endometrial stroma grows and proliferates outside the uterine cavity, with reports estimating this disease to occur in approximately 10% to 15% of women of reproductive age [8]. Most endometriosis cases generally involve the pelvic organs, such as the ovaries and peritoneum, whereas extrapelvic lesions, such as those involving the gastrointestinal tract, lungs, and umbilicus, have been reported, albeit very rarely. Accordingly, endometriosis quite rarely develops in the inguinal region, with reports showing a frequency of only 0.3–0.6% [1, 2].

There have been scattered reports of malignant tumors developing from inguinal endometriosis [9–18]. According to a previous cohort study, only around 0.7% of endometriosis cases undergo malignant transformation [19]. However, given that only a few reports exist on malignant transformations of rare-site endometriosis, the actual situation still remains unclear.

The following Sampson and Scott criteria have been used to diagnose malignancies arising from endometriosis [3, 20]: (1) the presence of benign endometriosis lesions within the tumor, (2) the absence of malignant invasion from other tissues, (3) the presence of tissues similar to the endometrial stroma around the glandular epithelium, and (4) a benign to malignant transitional picture. In the current case, the lesion satisfied all of the mentioned criteria for proper diagnosis. To date, only 12 cases, including the current one, have been reported in English, which we summarize in Table 1 [9–18]. The median age was 47 (34–68) years, with the lesions more commonly occurring on the right side (9 and 3 on the right and left sides). This may be because inguinal endometriosis is more common on the right side. The reason inguinal endometriosis is more common on the right side is not clear, but various theories have been offered: (1) the sigmoid colon covers the left round ligament and (2) endometrial cells remain on the right side because of the clockwise movement of intraperitoneal fluid [1]. Clear cell adenocarcinoma was the most common pathological type (five cases), followed by endometrioid adenocarcinoma (three cases) and then serous adenocarcinoma, endometrioid stromal sarcoma, and adenosarcoma each having one case. In recent years, surgical treatment combined with chemotherapy has been the treatment of choice for many patients. In the current case, chemotherapy was imitated after resection. Although recurrence had been observed in half of the cases, some did achieve long-term survival with multidisciplinary treatment after recurrence.

No consensus has yet been established regarding the standard treatment for endometriosis-related cancer occurring the extra-ovarian inguinal region. Resection or radical resection of the inguinal mass (with lymph node dissection in some cases) has been described in the literature [9–18]. Moreover, studies have reported the administration of carboplatin and paclitaxel as chemotherapy, similar to ovarian cancer [11, 14, 16–18].

In the current case, an inguinal mass was noted during postoperative follow-up for gastric cancer. By performing an excisional biopsy with the differentiation of inguinal masses among women in mind, we were able to establish a diagnosis of endometrioid adenocarcinoma of endometriosis origin in the inguinal region.

Author contributions AG performed the surgery; AG drafted the manuscript; SF, TY, YF, CK, NH, JH, TW and YK conceived the study, participated in its design and coordination, and helped draft the manuscript. All authors approved the final manuscript.

Funding None.

Data availability Not applicable.

Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Ethics approval and consent to participate Not applicable.

Consent for publication Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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