Case Report

Desmoid Tumor Mimicking Port Site Metastasis after Laparoscopic Surgery for Endometrial Cancer

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Abstract

Desmoid tumors are rare; however, they sometimes form in the abdominal wall after surgery or trauma. We report a case of desmoid tumors in the abdominal wall mimicking port-site metastasis after laparoscopic surgery for endometrial cancer. A 53-year-old woman with familial adenomatous polyposis presented to our hospital with vaginal bleeding and was diagnosed with endometrial cancer. We performed a total laparoscopic hysterectomy and began observation. Two years after surgery, follow-up computed tomography revealed three nodules with a size of approximately 15 mm in the abdominal wall at the trocar sites. Tumorectomy was performed because endometrial cancer recurrence was suspected, but desmoid fibromatosis was finally diagnosed. This is the first report of desmoid tumors at the trocar site after laparoscopic surgery for uterine endometrial cancer. Gynecologists should be aware of this disease because differentiating it from metastatic recurrence is challenging.

Keywords: Desmoid tumor, endometrial cancer, laparoscopy, port site, trocar site

Introduction

Laparoscopic surgery is the standard treatment for early-stage uterine endometrial cancer.[1] During laparoscopy, surgeons carefully remove malignant tumors to avoid the spread of tumor cells at the trocar sites;[2] however, desmoid tumors can also form at the trocar sites because of physical stimulation during surgery.[3-8] Although desmoid tumors are benign,[9] they mimic the local recurrence of malignant tumors. We report a case of desmoid tumors at the trocar sites after laparoscopic surgery for uterine endometrial cancer. Gynecologists should be aware of this disease because differentiating it from metastatic recurrence is challenging.

Case Report

A 53-year-old, gravid 2 and para 2, postmenopausal woman with familial adenomatous polyposis (FAP) presented to our hospital with irregular vaginal bleeding. On admission, transvaginal ultrasonography revealed an endometrial thickness of 22 mm with blood flow. A biochemical marker analysis showed that the tumor markers were not abnormally increased: cancer antigen 125, 17.1 U/mL; carbohydrate antigen 19–9, <2.0 U/mL; and carcinoembryonic antigen 0.9 ng/mL. Contrast-enhanced magnetic resonance imaging revealed an endometrial mass in the uterine cavity with the invasion of less than half of the myometrium. Disseminated nodules and ascites were not observed. Distant metastases were not observed using contrast-enhanced computed tomography. The endometrial tissue biopsy detected endometrial carcinoma G1. Preoperatively, endometrial cancer (Stage IA, FIGO 2008) was diagnosed. Because of the size and localization of the tumor, total laparoscopic

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hysterectomy with bilateral salpingo-oophorectomy and pelvic lymphadenectomy was performed. An umbilical trocar (12 mm), midline lower abdominal trocar (12 mm), right and left lower abdominal trocars (5 mm), and left lateral umbilical trocar (5 mm) were inserted. Bilateral fallopian tubes were ligated at the beginning of surgery. The resected specimen weighed 78.8 g and was vaginally collected in a bag. No bleeding was observed during trocar removal. The total operative time was 4 h and 29 min, and blood loss was 250 mL. The postoperative course was uneventful, and the patient was discharged from our hospital 4 days postoperatively without any complications. The histopathology report revealed that the uterus measured 65 mm × 50 mm × 30 mm and the tumor measured 40 mm × 32 mm × 15 mm. Endometrial cancer and endometrioid carcinoma (G1, Ly 0, v 1, FIGO stage IA, pT1aN0M0, N 0/17) were diagnosed. Because the recurrence risk was low, we monitored the vaginal stump, tumor markers, transvaginal ultrasonography, and computed tomography. Computed tomography performed at 1 year and 2 months postoperatively showed no recurrence. However, three nodules with contrast enhancement in the abdominal wall were detected at 2 years and 2 months postoperatively. The nodule in the right lower abdomen was 16 mm × 13 mm, that in the median lower abdomen was 16 mm × 9 mm, and that in the left lower abdomen was 14 mm × 5 mm [Figure 1a and b]. The three nodules were at the trocar sites. Magnetic resonance imaging also detected three nodules with contrast enhancement, and high signal intensity was detected using diffusion-weighted imaging. There were no other signs of recurrence. Biochemical marker analysis showed that the tumor markers were not abnormally increased: cancer antigen 125, 8.7 U/mL; carbohydrate antigen 19–9, 2.2 U/mL; and carcinoembryonic antigen 1.1 ng/mL. Endometrial cancer recurrence or desmoid tumor was considered, and we contemplated either a tumorectomy or needle biopsy for differentiation. In the likelihood of endometrial cancer recurrence, tumorectomy would be a faster treatment with a lower risk of dissemination. In addition, all tumors were in the abdominal wall and appeared easily and noninvasively resectable. After thorough informed consent, tumorectomy was performed 2 years and 3 months postoperatively. Under transabdominal ultrasonography guidance, a 5-cm skin incision was placed over each tumor, the fascia was then incised, and the mass was removed. The specimen in the right lower abdomen, median lower abdomen, and lower left abdomen weighed 10.0, 2.0, and 4.6 g, respectively. The total operative time was 1 h and 9 min, and blood loss was 36 mL. The postoperative course was uneventful, and the patient was discharged from our hospital 3 days postoperatively without any complications. All three tumors were resected with negative margins. Hematoxylin and eosin staining showed spindle-shaped cell proliferation and abundant collagen fibers [Figure 1c]. An immunohistochemical analysis showed that β-catenin, CD10, and smooth muscle actin were positive, Ki-67 was 5% positive, and estrogen receptor and CD34 were negative [Figure 1d]. Desmoid fibromatosis was diagnosed. There has been no sign of endometrial cancer recurrence since the initial surgery 3 years ago, and no sign of desmoid tumor recurrence since the second surgery 1 year ago. The patient experienced no complications.

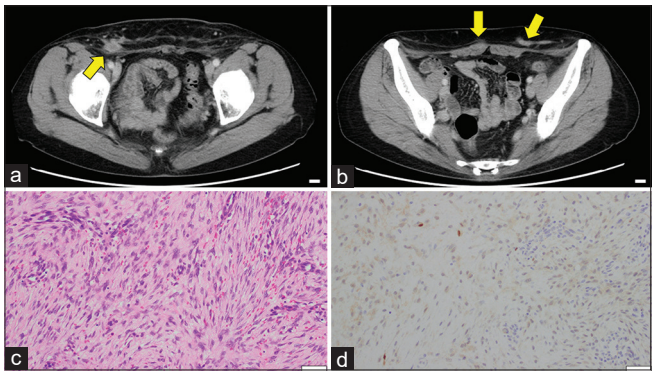

Figure 1: (a and b) Contrast-enhanced computed tomography (axial) shows three nodules at the trocar sites (yellow arrows) (scale bars = 10 mm), (c) Spindle-shaped cell proliferation and abundant collagen fibers (hematoxylin and eosin), (d) β-catenin is positive immunohistochemically (scale bars = 50 μm) are × 200

Discussion

We report a case of desmoid tumors at the trocar insertion sites after laparoscopic surgery for uterine endometrial cancer that mimicked metastatic recurrence of endometrial cancer. The patient had a history of FAP, which is a risk factor for desmoid tumors;[9] however, there were multiple tumors, and it seemed difficult to deny the metastatic recurrence of malignancy and diagnose them as desmoid tumors. Desmoid tumors are not a common disease in the field of gynecology, but it is important to be aware of this disease.

A desmoid tumor is a rare type of benign monoclonal fibroblastic tumor that does not cause metastasis, but it invades aggressively and enlarges locally. Its incidence is approximately five cases in 1 million people per year. It is more prevalent among 10-to 40-year-old, and it is two to three times more common in women than in men. Most desmoid tumors are sporadic, but 10% of desmoid tumors are related to FAP, as in the present case. The APC gene mutation (known as the mutation in FAP patients) and CTNNB1 gene mutations are associated with β-catenin overexpression. In addition, estrogenic involvement has been suggested with this condition. Desmoid tumors can be classified as extra-abdominal or intra-abdominal, but they can occur anywhere and are often solitary; however, sometimes they are multiple. They often occur in association with surgery

Table 1: Characteristics of previous desmoid tumors at the trocar site after laparoscopic surgery

Case	Age	Sex	Primary disease	Primary laparoscopy	Time todesmoid tumors	Number of tumors	Dominant tumor (cm)	Mesh repair
1	35	Female	NA	Tubal ligation	10 years	1	6	+
2	34	Female	FAP	Total colectomy	1 year 8 months	2	40	+
3	24	Male	Testicular cancer	Retroperitoneal lymph node dissection	1 year	1	4	−
4	35	Female	Gallstones	Cholecystectomy	2 years	1	9	−
5	53	Female	Gallstones	Cholecystectomy	8 months	1	5	+
6	34	Female	Gallstones	Cholecystectomy	2 years	1	10	+
7 (our case)	53	Female	Endometrial cancer	Hysterectomy	2 years 2 months	3	1.6	−

FAP: Familial adenomatous polyposis, NA: Not applicable

or trauma[9] and are difficult to distinguish from metastases, especially when they occur at the site of malignant tumor surgery, as in the present case. Fewer cases of desmoid tumors have been reported with laparoscopic surgery than with laparotomy for patients with FAP.[10] Moreover, desmoid tumors in postoperative laparoscopic wounds are extremely rare; only six cases have been reported[3-8] [Table 1]. Owing to the limited number of reported cases, the incidence of desmoid tumors in different laparoscopic surgeries or diseases operated thereon is not clear. Although other cases involved a single occurrence of a desmoid tumor, our case involved multiple tumors. The surgical time for hysterectomy may have been longer and stimulated the abdominal wall more than that in other surgeries, such as cholecystectomy. In addition, the patient, in this case, had a history of FAP. However, there is still no clear explanation for the multiple desmoid tumors observed in this case. Desmoid tumors cannot be confirmed by imaging, such as computed tomography or magnetic resonance imaging; they are usually confirmed pathologically through biopsy.[9] The general management of desmoid tumors depends on the symptoms and localization. Surgical resection has been the main treatment for the management of desmoid tumors; however, recently, it has been proposed that patients without symptoms can be followed up for observation, and surgery is performed only when tumor growth is observed.[9,11] In this case, the standard policy for desmoid tumors was also considered, namely, biopsy and observation. However, although extremely rare, approximately 1% of patients with uterine endometrial cancer develop trocar site metastases after surgery,[1,2] and early treatment is necessary in the case of malignant tumor recurrence. If a biopsy was performed initially, it would have extended the time until tumorectomy. In addition, because the tumors in the present case were not very large and were in the superficial layer, it was possible to perform a less invasive resection without mesh repair. Therefore, we explained both scenarios to the patient, namely, cancerous tumor recurrence and desmoid tumor, and we collectively decided to perform surgical treatment immediately. Regarding future recurrence, desmoid tumor and uterine endometrial carcinoma will be differentiated; however, because this case involved desmoid tumor, careful follow-up is an option. In this case, our policy is to follow-up on a tumor that is only in the abdominal wall and perform a needle biopsy if the tumor is intra-abdominal, as this is not an uncommon site for the recurrence of endometrial cancer. If the tumor in the abdominal wall enlarges under observation, we will also perform a needle biopsy.

Ethical statement

The IRB approval of this study was exempted.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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