Case Report

Rapid recurrence of uterine smooth muscle tumor of uncertain malignant potential

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Abstract

Smooth muscle tumor of uncertain malignant potential (STUMP) is a rare variant uterine smooth muscle tumor with borderline features that reflect neither the benign leiomyoma nor the malignant leiomyosarcoma. The patient, a 30-year-old woman (gravida1, para1), presented with mild hypogastrum pain. As a result of detailed examination, we suspected a right ovarian multilocular cyst. Under the intraoperative view, the multilocular cyst grew under the right retroperitoneum, near the vicinity of the right uterine cardinal ligament. We conducted tumor resection, and microscopic analysis revealed STUMP in the center of the tumor wall. On a follow-up examination 3 months after the surgery, transvaginal ultrasound revealed a solid tumor measuring 3×5×4 cm on the right side of the uterus. We suspected the rapid recurrence of STUMP and conducted hysterectomy and salpingo-oophorectomy. The pathological diagnosis was that STUMP developed from the uterus. At a follow-up 15 months after the first surgery, the patient was alive and well with no evidence of recurrent or metastatic disease.

Key Words:
Smooth muscle tumor of uncertain malignant potential, rapid recurrence, laparoscopy

Introduction

A smooth-muscle tumor of uncertain malignant potential (STUMP) is a rare, variant, uterine smooth-muscle tumor with borderline features that reflects neither the benign leiomyoma nor the malignant leiomyosarcoma. The diagnosis of benign, borderline, or malignant uterine smooth muscle tumors depends on histologic findings such as the number of mitotic figures, nuclear atypia, and the presence of tumor cell necrosis [1]. STUMP is diagnosed if the tumor exhibits any unusual combination of these 3 features. However, it does not satisfy the criteria for leiomyosarcoma [2].

We experienced a rare case of STUMP which had cystic shape and relapsed rapidly after the surgery. We report this case with some literature review.

Case Presentation

A 30-year-old woman (gravida1, para1) presented with mild hypogastrum pain. She had no obvious medical illness and no history of surgery. A transvaginal ultrasound revealed a multilocular cyst measuring 8×8×13 cm on the right side of the uterus. The pelvic Magnetic Resonance Imaging (MRI) showed that there was no solid pattern and no malignant findings, including Diffusion-Weighted (DW) MRI (Figure 1). We diagnosed the right ovarian cyst and planned a laparoscopic assisted ovarian cystectomy. A 10 mm long incision was made in the umbilicus, and a closed laparoscopic method was used for entry into the abdomen. An 11 mm trocar was inserted through umbilicus
and a 10 mm scope was inserted through the trocar. A 30 mm long incision was made in the hypogastrium, a Lap-Protector (Hakko Medical, Nagano, Japan) was inserted into the incision for wound protection, and an EZ Access (Hakko Medical) was mounted on the Lap-Protector. A 5 mm and 12 mm trocars were inserted through the EZ Access port. The multilocular cyst grew under the right retroperitoneum, near the vicinity of the right uterine cardinal ligament.

Although we tried to remove the tumor, while separating the tumor from the tissue around it, the total bleeding volume amounted to 1600 ml. The patient needed to have a blood transfusion following this procedure. We decided to change to the laparotomy and excised the tumor which weighed 250 g containing fluid. The wall of the tumor was firm and rubbery. Microscopic analysis revealed that in the center of the tumor, there were complicating spindle-shaped cells, which were positive in immunohistochemically staining method of the α-smooth muscle actin(αSMA), that dyes a smooth muscle, and partially had moderate atypia (Figure 2). The mitotic rate was low (<10 mitotic figures/10 high-power field) and necrosis was not detected. It was diagnosed as STUMP. We recommended removing the uterus to the patient, but she insisted to preserve it. On the follow-up examination 3 months after the initial diagnosis, a transvaginal ultrasound revealed a solid tumor measuring 3×5×4 cm on the right side of the uterus. The Computerized Tomography (CT) showed that the boundary between the tumor and the uterus was obscure, and it did not spread to other parts of the body (Figure 3). We suspected the rapid recurrence of STUMP. The patient did not wish fertility preservation yet, and there was a possibility that the recurrent tumor was sarcoma. We carefully discussed with her, and we determined to conduct total hysterectomy and bilateral salpingo-oophorectomy. We were apprehensive about the formation of abdominal adhesions as a repeat surgery, and the patient hoped for a less invasive surgery. We selected a laparoscopic surgery with the function of magnifying and reaching the depth of the pelvis. Because the right ureter ran at the edge of a tumor, we inserted bilateral ureteral stents before the surgery.

A 10 mm long incision was made in the umbilicus again, and a closed laparoscopic method was used for entry into the abdomen. An 11 mm trocar was inserted through the umbilicus and a 10 mm scope was inserted through the trocar. A 10 mm long incision was made and a 12 mm trocar was inserted in the left upper abdomen. A 5 mm long incision was made and a 5 mm trocar was inserted in the bilateral lower abdomen. Under the laparoscopic view, the greater omentum formed wide peritoneum adhesion. The recurrent tumor was found in the right lower part of corpus uteri (Figure 4). We performed laparoscopic hysterectomy and bilateral salpingo-oophorectomy, retrieving the specimen with a collecting bag from the vagina. The total bleeding volume amounted to 5 ml. The tumor was solid and weighed 210 g. Microscopic analysis revealed that the spindle-shaped cells in the tumor were positive by an immunohistochemically staining method of the αSMA and had partially moderate atypia (Figure 5). The mitot-

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**Figure 1.**

MRI (T2 intensity). The multilocular cyst on the right side of the uterus.

**Figure 2.**

Microscopic photograph. The initial tumor diagnosed as STUMP.
ic rate was low (<10 mitotic figures/10 high-power field), and a necrosis was not detected. It was diagnosed as the recurrence of STUMP of the uterus. The progesterone and estrogen receptor expression of the tumor were positive. The patient was young, but STUMP relapsed early postoperatively. It is not clear whether STUMP is hormone sensitive bibliographically [3]. We determined not to conduct hormone replacement therapy as a precaution. Her postoperative process was going well. At the follow-up 15 months after first surgery, she was alive and well with no evidence of recurrent or metastatic disease.

A preoperative diagnosis is generally difficult, and, in many cases, the pathologic diagnosis after the surgery reveals STUMP for the first time [2]. In the event of STUMP diagnosis in uterus conserving surgery, considering the possibility of recurrence, hysterectomy represents the gold standard for those women who have completed their childbearing. Successful pregnancies following fertility-sparing surgery have been reported, however, these patients should be adequately informed of the risk of recurrence. A strict follow-up through clinical and imaging techniques is mandatory [4].

Discussion

The clinical presentation of STUMP resembles that of uterine leiomyoma. Typical clinical features include abnormal vaginal bleeding, symptoms of anemia, rapidly growing pelvic mass, pressure symptoms, and pelvic pain [4]. Bonneau et al. (2014) reported that using ultrasonic inspection, STUMP was usually described as an oval-shaped tumor, lacking acoustic shadowing. MRI enhances the sensitivity of detecting STUMP. For conventional MRI, single tumor, large tumor, poorly defined margins, thickened endometrium, peritoneal implant, intermediate or high signal intensity in T1 and T2 sequences, heterogeneous T1 signal, and heterogeneity of the tumor’s enhancement are significantly associated with STUMP. One of the features is cystic alteration. The odds ratio of cystic alteration is 3.3 (P = 0.03). Furthermore, a high signal at DW imaging seems to be associated with STUMP [5]. In our case, cystic alteration was recognized but high signal intensity on DW image was not indicated.

Among 76 patients with STUMP treated with only tumorectomy, 5 (6.6%) experienced recurrence of the disease. The period from the operation until the recurrence ranged from 6 months to 10 years [2]. It has been suggested that recurring STUMP may represent a low-grade leiomyosarcoma or underdiagnosed leiomyosarcoma [2]. The treatment in the event of recurrence is surgical excision. Progesterone, Gonadotropin-Releasing Hormone (GnRH analogue), or chemotherapy have been proposed as adjuvant therapy, but none, as of yet, are proven to be effective in preventing recurrent disease [4]. Furthermore, there is lack of consensus regarding implementation of follow-up protocols. Ip PP et al. (2010) suggested an intense follow-up program with an evaluation performed every 6 months in the first 5 years followed by annual surveillance for the next 5 years [6]. In our case, since the patient was treated by uterus sparing surgery, we performed a clinical evaluation every month after surgery. The early detection and the early stage treat-
ment of the recurrence were possible as a result of this frequent medical check-ups. A patient who underwent just tumorectomy of STUMP has a possibility of early recurrence. Therefore, we need to follow-up cautiously.

Conclusion
We encountered a case which we suspected an ovarian cyst at first, but later the tumor was revealed to be STUMP. Frequent medical check-ups after the first operation enabled both the early detection and the early stage treatment of the recurrence.

Acknowledgement
None

Declaration of Interest
None

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