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Case Series
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Two Cases of Polypoid Uterine Sarcoma Successfully Operated Laparoscopically without Morcellation: Feasibility of Diagnostic Minimally Invasive Hysterectomy

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Abstract

Uterine sarcoma is rare malignant tumor and the prognosis is poor among the aggressive type. It is sometimes difficult to make differential diagnosis correctly between uterine benign tumor such as uterine fibroma or adenomyosis preoperatively. So, diagnostic aim operation is feasible for suspicious cases. Minimal invasive surgery such as laparoscopic operation or robotic surgery is a good indication in such case as they have merit of smaller incision, short time in hospital, early recovery. But if uterine size is large enough that morcellation is necessary for transvaginal specimen extraction, the spread of sarcoma cells is warranted.

We experienced two cases of uterine polypoid sarcoma that laparoscopic hysterectomy was accomplished without morcellation. One case was uterine adenosarcoma with heterologous elements and sarcomatous overgrowth. Another case was low grade ESS. In both cases, recurrence in the abdominal cavity was not occurred and diagnostic hysterectomy was safely accomplished.

Introduction

Uterine fibroma is a high frequency illness for reproductive aged women and the incidence is reported about 25% of the women population [1]. Comparatively uterine sarcoma is quite low frequency; the incidence is reported about 3% of all malignant uterine tumors [2]. Among uterine fibroid patients, uterine sarcoma incidence was reported about 0.2% to 0.3% [3]. Recent minimal invasive surgery has a merit of lower pain, early recovery, short time bed stay, low infection rate, the benign uterine fibroid or adenomyosis cases, minimal invasion surgery is a good indication and total laparoscopic hysterectomy has become widely prevailed [4,5]. The operation has a process of transfer resected uterus outside the body usually through the vaginal sump. In case that the uterus size is too large, the morcellation is necessary. But in suspicious uterine sarcoma case, morcellation might spread malignant cells in the abdominal cavity and poor prognosis by early recurrence in the peritoneal cavity is warranted [6-9]. In February 2020, FDA recommends that contained morcellation in women when laparoscopic power morcellation is appropriate [10]. To avoid spread of sarcoma cells, contained morcellation is necessary [11].

For the aim of diagnosis hysterectomy, without morcellation operation is more desirable so, it is necessary to judge that of the suspicious uterine sarcoma cases, laparoscopic hysterectomy could be accomplished without morcellation or not preoperatively. In this report, we experienced two case of uterine polypoid sarcoma that the laparoscopic operation was performed and discuss about the problems and indication of total laparoscopic hysterectomy without morcellation in the suspicious uterine sarcoma cases.

Case Series

Case 1

A 47 years old woman, gravitas 1 para 1, presented in our hospital with suspicious of uterine endometrial cancer. Annual abdominal echo by internal medical doctor has previously pointed out uterine swelling. She had been bleeding since menstruation two months before her visit to our hospital, so she was referred to a previous gynecological doctor. The previous doctor suspected the malignant lesion and done Pap smear test, and pelvic MRI without enhancement, because she has suffered from kidney failure and dialysis was done three times in a week. Although cervical Pap smear was NILM and endometrial Pap smear negative, MRI showed the suspicious of uterine endometrial carcinoma (Figure 1). So, the doctor did diagnostic curettage of total uterine endometrium, but

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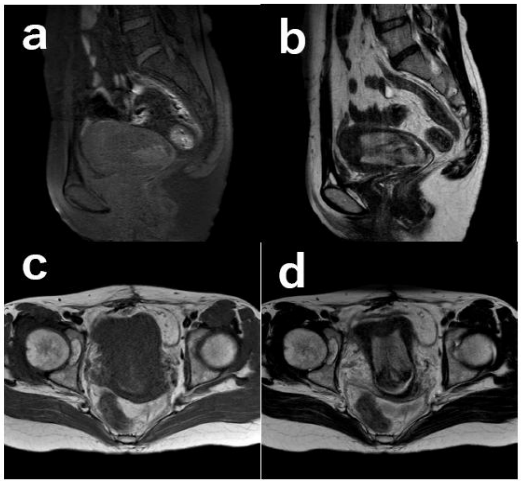

Figure 1: T1 and T2 intensity sagital and transverse MRI images of the patient are shown. As shown in a and c, the high intensity area can be seen, showing that bleed storage was suspected.

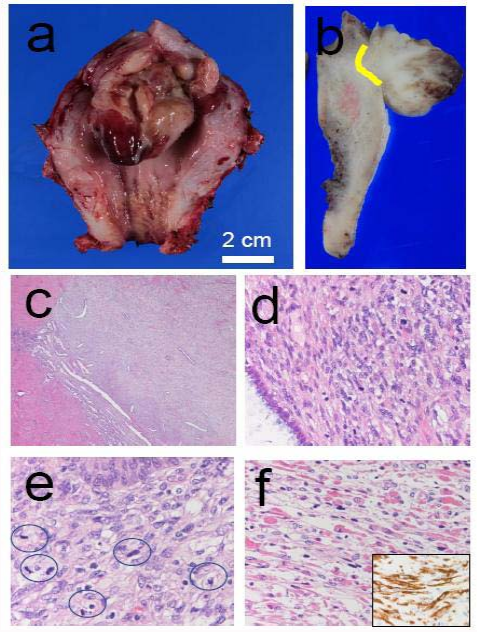

Figure 2: Laparoscopically hysterectomized uterus is shown in a. The polypoid originated from fundus of the uterus, the size of which was 6.8 cm × 4.2 cm × 3.5 cm, was seen as shown in b. At the top of the polyp, hematoma was attached, showing the site of the bleeding. b) Cross section of the uterine specimen after fixation by formalin. The border of the tumor was lined by the yellow. c to f) Show the HE staining of the specimen. The nuclear atypia of cells are poor but the mitosis was seen maximal 22 per HPF as shown in the circle of Figure 1e. The striated muscle differentiation of cancer cells were observed as shown in Figure 1f. Desmin positive stroma cells were observed as shown in the window of Figure 1f.

histology was not malignant and only normal endometrium with necrotic tissue was observed in HE stain of the tissue.

Then the patient was introduced to our hospital. We did curettage of uterine endometrium but the result was only normal endometrium with no malignancy. But judging from uterine abnormal bleeding, abnormal MRI finding, and finding of necrotic tissue from endometrium, we decided total laparoscopic hysterectomy aiming diagnosis. Total laparoscopic hysterectomy without morcellation was done and the patient's uterus was hysterectomized with minimally invasive and she suffered no complication. The macroscopic findings showed a polypoid lesion in the uterine cavity and hematoma was attached at the tip. The formalin fixed HE staining of the resected specimen was poor in cell nuclear atypia, maximal 22 mitotic cell in HPF, vimentin positive, desmin positive, S100 negative, striated muscle differentiation was observed, altogether find the pathological diagnosis as adenosarcoma of the endometrium, with heterologous element and sarcomatous overgrowth (Figure 2) [12]. We performed postoperative radiation to the pelvis because she was suffering from renal failure [13]. After the postoperative treatment, no recurrence was observed in the abdominal cavity, vaginal sump, and vagina. Instead Th10 bone metastasis that located outside the radiation field was found 6 months after the operation and the succeeded treatment was not effective and she died of the disease 7 months after the recurrence. As reported the sarcomatous overgrowth has poor prognosis [14,15].

Case 2

A 50 years old woman, 3 gravitas 2 para 1 spontaneous abortion, was represented with complaint of lower abdominal pain, palpitation, and shortness of breath. At first, she visited to our hospital on suspicion of uterine fibroids and hydrometra. No special notes on medical history were seen. Ultrasonography revealed uterine fibroids, and pelvic MRI with enhancement revealed degenerative submucosal and intramuscular fibroids, and left endometriotic cysts (Figure 3). Cytology of uterine cervical was NILM, uterine endometrium was negative. Since anemia due to menorrhagia was observed, 6 courses of GnRH agonist therapy were performed followed by Dienogest. However, uterine bleeding was continued with anemia, the operation was decided. After 3 courses of GnRH agonist therapy were performed to improve the anemia, total laparoscopic hysterectomy was performed. The uterus was carried out of the body without morcellation. The pathological diagnosis was low-grade endometrial stromal sarcoma as the tumor cell invaded in the myometrium of the uterus and had poor in nuclear atypia, mitosis cell was scarcely found, CD10 immunostaining was positive in tumor cell as shown in Figure 4 [16,17]. Laparoscopic bilateral oophorectomy was added 1 year and 6 months after the initial surgery [18]. When the abdominal cavity was observed, a suspicious lesion of recurrence was found in the vaginal stump, but the histopathological result was an inflammatory

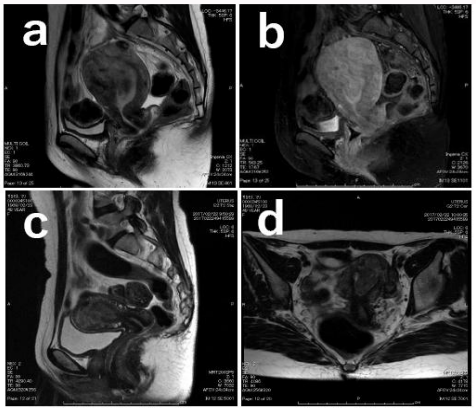

Figure 3: a, b) MRI image of uterus for the first time. The uterine lesion contained myofibroid and adenomyosis but no malignant finding was detected by radiology special doctor. c, d) MRI images of uterus after 6 course of GnRH agonist treatment. The uterus shrunk in the rate of 53%. MRI diagnosis had no change.

Table 1: Character of the two cases.

	Case 1	Case 2
Age	47	50
Gravita Para	1 gravita 1 para	3 gravita 2 para
Chief complaint	Abnormal bleeding	Anemic symptom
Cx smear	NILM	NILM
EM smear	Class I	Class I
MRI finding	Endometrial hyperplasia	Adenomyosis and myoma
3D uterine size from MRI (depth × width × length)	48 mm × 51 mm × 86 mm	44 mm × 42 mm × 84mm
EM curettage	Normal endometrium with necrotic tissue	Not done
Pathology	Adenosarcoma with heterologous elements and sarcomatous overgrowth of the uterus	Endometrial stromal sarcoma of the uterus, low grade
Recurrent site	Th10, Th1	No recurrence
Outcome	Die of the disease	Without recurrence

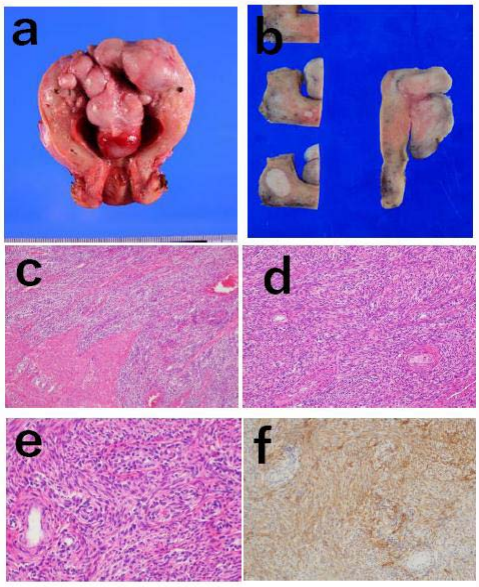

Figure 4: a) Macroscopic finding of the laparoscopically resected uterus that was added vertical incision. The polypoid lesion was observed. b) Cross section of the uterus is shown. The polypoid tumor was originated from the fundus of the uterus. The border of the tumor is shown by yellow line. c-e) HE staining of the tumor. The tumor cell invaded in the myometrium of the uterus and had poor in nuclear atypia. The mitosis cell was scarcely found. CD10 immunostaining was positive in tumor cell as shown in f.

change, not a recurrence. Washed cytology of the abdominal cavity was class I. There were no other suspicious findings of recurrence. Currently she is being managed as an outpatient with no recurrence of the disease 2 years after the first operation.

Discussion

Even though the progress of image radiology, the differential diagnosis of uterine fibroid and malignant uterine diseases has limitation [19,20]. So, in some cases, diagnosis aimed hysterectomy is necessary. Laparoscopic hysterectomy is minimally invasive and superior to open laparotomy in performing diagnostic aim of hysterectomy, because of earlier surgical wound healing, shorter duration at hospital, less painful, and early recovery for social life, and in case of the necessity of postoperative treatment, early adjuvant treatment can start [6]. So, in sarcoma suspicious cases, diagnostic laparoscopic hysterectomy is feasible. However, uterine sarcoma that is often accompanied by uterine swelling often requires uterine morcellation to transfer uterus outside the body. But if do so in case of uterine cancer or sarcoma, malignant cells spread inside the body recurrence will occur and the prognosis of the patient must be poor. To avoid such situation, we must choose the appropriate cases that the TLH without morcellation is possible or not in suspicious uterine sarcoma before operation. But there is no criteria that swelled uterus after resection can be successfully transferred from abdominal cavity to the outside the patient body. We estimated that in both cases, we thought that uterus swelling was small enough to transfer trans-vaginally. In our experience of the two cases, the uterine size is shown in Table 1. In both cases, the resected uterus could be transferred out of the pelvic cavity trans-vaginally without morcellation or splitting, but it was limiting. We think 50 mm to 60 mm in depth and width of the uterine corpus size from MRI finding is the maximal size that the uterus may be limiting size to transfer the resected uterus vaginally without morcellation who had experience of vaginal delivery. Even in such cases, in bag transfer is desirable because strong pulling by forceps sometimes split uterine cervix and corpus.

Clinical study of contained morcellation is not yet reported. So, in the future it is necessary to ascertain the safety of total laparoscopic hysterectomy with contained morcellation even if the malignant uterine lesion existed in the benign lesion of uterine diseases.

In case 2, being smaller size by use of GnRH agonist administration was important. If the doctor did not use GnRH agonist, the uterus corpus size was 64 mm × 44 mm and it must be difficult to transfer the resected uterus through vagina without morcellation. We propose the management of uterine myoma or adenomyosis, that before hysterectomy, any myofibril and adenomyosis cases, if possible, GnRH agonist or GnRH antagonist should be administered to make uterine size smaller and increase the possibility that uterus is laparoscopically resected more successfully and more safely that transferring the resected uterus trans-vaginally outside the body without morcellation.

We experienced to cases of uterine sarcoma that glowed with poly shape and the size of which was modest and was operated laparoscopically and the resected uterus was carried out of the body without morcellation. They tell us the important of down-sizing of the uterus by GnRH agonist or antagonist to accomplish total laparoscopic hysterectomy more safely and if the uterus was not become smaller in spite of using GnRH agonist or antagonist, it should be better to do hysterectomy using contained morcellation or

by laparotomy. In such case, the disease is very suspicious of uterine malignant lesion and better to accomplish hysterectomy with no residue, no spread.

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