

Metastatic Smooth Muscle Tumor of Uncertain Malignant Potential after Laparoscopic Presuming Myomectomy

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Abstract

A 38-year-old para-2 female underwent laparoscopic myomectomy with uncontained morcellation. Three years later, she complained of epigastric pain. An intraperitoneal 3 cm mass beneath the umbilicus was showed on computed tomography (CT) scan. With the impression of gastrointestinal stromal tumor, she underwent open laparotomy at the general surgery department. A tumor was excised. Pathological examination showed that the tumor was consistent with a smooth muscle tumor of uncertain malignant potential smooth muscle tumors of uncertain malignant (STUMP). Six years postlaparoscopic myomectomy, during a regular follow-up, three parauterine masses were found on ultrasonography and CT scan. She underwent laparoscopic surgery for hysterectomy, bilateral salpingectomy, and excision of the masses. The masses were again diagnosed as STUMP. This case presents a recurrence of a rare type of smooth muscle tumor after uncontained morcellation. If myomas are to be removed with morcellation, it should only be used appropriately with a compatible containment system, and the risk of occult malignancy should be counseled.

Keywords: Laparoscopic myomectomy, morcellation, morcellator, parasite smooth muscle tumors of uncertain malignant potential, smooth muscle tumors of uncertain malignant potential

INTRODUCTION

Uterine leiomyoma is the most common benign pelvic tumor and is estimated to occur in 70%–80% of reproductive-aged women. For women over 35 years of age, approximately 20%–40% of women have symptomatic myomas requiring intervention.^[1] Treatment modalities include myomectomy for those considering future childbearing capability and hysterectomy for definitive management. Due to the technological advance in laparoscopy, laparoscopic surgery is now used with most patients with uterine myoma. Among different types of uterine myoma, intramural and subserous myomas were associated with a higher successful rate.^[2] Ulipristal acetate has been introduced as preoperative treatment of patients with large fibroids; however, studies

showed that it results in a more technically difficult situation with the potential risk of incomplete resection due to anatomical distortion and unclear cleavage planes.^[3] Although minimally invasive surgery represents the gold standard treatment, laparoscopic myomectomy still requires adequate knowledge of the risk factors for complications, of which the most common is bleeding. The myoma diameter, the number of myomas, and the operative time were found to be significantly associated with blood loss and transfusion rate.^[4] Furthermore, checking all items after the laparoscopic surgery is important to prevent the incidence of operative

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instrument breakage.^[5] Specimen fragmentation during the retrieval process in laparoscopy may leave behind tissue particles that can survive, implant, and grow. As such, the incidence of iatrogenic metastatic leiomyoma has been increasingly reported due to the growing use of laparoscopic surgery.^[6,7] Moreover, smooth muscle tumor of uncertain malignant potential (STUMP) after laparoscopic surgery is extremely rare. This report describes a case of metastatic recurrent STUMP after laparoscopic myomectomy with uncontained morcellation.

CASE REPORT

A 38-year-old para-2 female presented with urinary frequency. Physical examination and preoperative ultrasonography revealed an anterior subserous myoma measuring 8.8 cm. She underwent laparoscopic myomectomy with electric tissue morcellation in 2012. Intraoperatively, the uterus was irregularly enlarged to 12-week pregnancy size, and there was an 8 cm × 8 cm subserous myoma at the anterior uterine wall. The process of laparoscopic myomectomy included making an incision into the overlying serosa using monopolar scissors, enucleation of myoma using sharp and blunt dissection, and repairing the incision with two layers of simple interrupted stitches using 1-0 polyglactin 910. The myoma was removed by uncontained electric morcellation. The entire abdominopelvic cavity was inspected and thoroughly irrigated with a large amount of isotonic solution before the closure of the incision. No postoperative complications were noted, and she was discharged from the hospital. Histopathology confirmed the mass to be a leiomyoma.

Three years after the laparoscopic myomectomy, the patient complained of epigastric pain and decreased appetite and was subsequently directed to the general surgery outpatient department. An abdominal computed tomography (CT) scan showed an intraperitoneal 3 cm mass beneath the umbilicus. With an impression of gastrointestinal stromal tumor (GIST), she underwent open laparotomy. A 3 cm mass originating from the mesentery of the transverse colon was noted, and a tumor excision was done. Her postoperative course was uneventful. Histopathology showed a tumor composed of fascicles of spindle cells; the tumor could be GIST or smooth muscle neoplasm. Further, immunohistochemical examination was processed; the specific markers for GIST, CD117, and DOG1^[8,9] were negative. Desmin, which is a specific marker for smooth muscle tumor was positive. Estrogen receptor was diffusely positive, and Ki-67 was 10% positive. Ki-67 is the proliferative marker, which is generally increased more than 33% in leiomyosarcoma.^[10] In summary, the tumor featured smooth muscle tumor by immunohistochemical examination. For histopathology, according to the WHO Classification 2014,^[11] the tumor

was consistent with a smooth muscle tumor of uncertain malignant potential (STUMP). She had regular follow-ups every 6 months with CT scan until 6-year postlaparoscopic myomectomy. When a parauterine tumor was noted, she was referred to the gynecology department. Ultrasonography revealed three parauterine masses, with the largest mass 4 cm in diameter. The patient was informed that the masses could be recurrent myoma or STUMP as in the previous operation. Surgical intervention was advised, and she underwent laparoscopy in 2018. The operative findings revealed the uterus to be globularly enlarged to 12-week size. There were two extrauterine masses: one 4 cm mass located at the right anterior broad ligament, and another 2 cm mass located at the left pelvic sidewall, lateral to the infundibulopelvic ligament [Figure 1]. Total laparoscopic hysterectomy with bilateral salpingectomy was performed with an excision of mass at the left pelvic sidewall. All specimens were retrieved through the vagina without morcellation. She was discharged from the hospital in an improved condition. Histopathology revealed the uterus contained leiomyoma. The extrauterine parasitic tumors were again diagnosed as STUMP [Figure 2]. The microscopic picture depicted a well-circumscribed nodule composed of interlacing bundles and fascicles of spindle tumoral cells with nuclear atypia.

The patient underwent consistent follow-ups every 6 months. Surveillance using abdominopelvic CT scan. The latest CT scan was performed in May 2019, 10 months postoperatively has not shown the recurrence of any intra-abdominal tumor.

DISCUSSION

Smooth muscle tumor of uncertain malignant potential is a rare entity, and the occurrence of a parasitic STUMP is even rarer. The patient had a history of leiomyoma, a benign pathologic condition, during the first operation. The recurrence of a smooth muscle tumor with a different pathology from the first operation is rare. Moreover, an intraperitoneal parasitic smooth muscle tumor is extremely difficult to occur *de novo*. Metastatic myoma due to peritoneal seeding of myometrial tissues during morcellation has been reported since 1997 due to an increased prevalence of laparoscopic surgery.^[7] However,

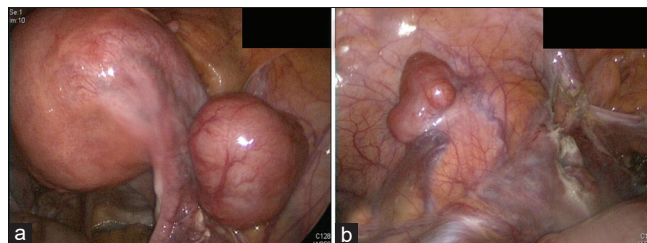


Figure 1: Picture from the third operation performed on July 30, 2018. (a) 4 cm mass at the right anterior broad ligament. (b) 2 cm mass at the left pelvic sidewall, lateral to infundibulopelvic ligament

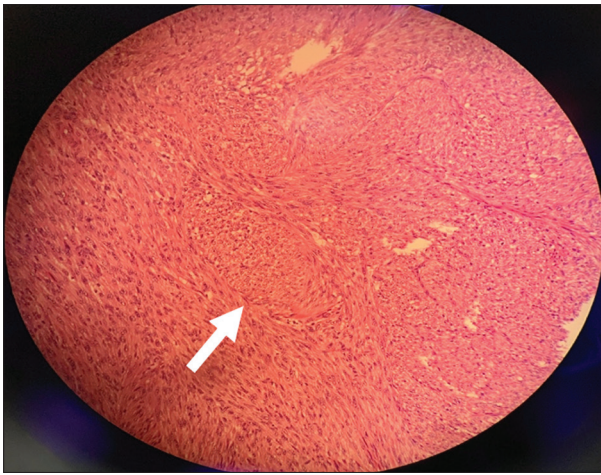


Figure 2: Smooth muscle tumor of uncertain malignant potential (H and E stain, at $\times 40$). Circumscribed nodule composed of interlacing bundles and fascicles of spindle tumoral cells with nuclear atypia characteristic of STUMP

intraperitoneal metastatic STUMP was reported only once in 2016 by Bogani *et al.*^[12] The initial surgery was laparoscopic myomectomy, and the histology confirmed the excised tumor to be a leiomyoma. Six years later, the patient noted a 1 cm nodule at the port site. Nodule excision was performed, and the pathology revealed a smooth muscle tumor. The two specimens from the first and second surgeries were then reexamined side by side, and the reexamination revealed that they were both STUMP. The patient consequently underwent total abdominal hysterectomy with bilateral salpingectomy and a wide excision of the parasitic port site. There was no residual disease.

In the presented case, metastatic smooth muscle tumors of uncertain malignant potential were detected 3 and 6 years after laparoscopic myomectomy for a leiomyoma. Pathological review from the second and third operation presented the same characteristics as stump as in Figure 2. The specimens from the first operation may present stump in some small parts. Generally, if the pathology of the recurrent tumor showed a lesion of greater severity, the first specimen should be reexamined. A side-by-side review of all the specimens is recommended. Since STUMP presents a condition that is between the spectrum of benign and malignant conditions and its clinical behavior differs from that of a simple leiomyoma, it is important to differentiate and confirm the pathology of the tumors. Second, this case affirms the importance of implementing methods for tissue spillage prevention during the specimen retrieval process, especially when using a morcellator. The myoma or any uterine tumors should be placed in a specimen bag to avoid small tissue particles from being left inside the abdominopelvic cavity, which could potentially implant to become a tumor years later. In cases where malignancy is considered or the malignant status of

a tumor has not been determined, the use of a morcellator should be avoided.

The reported recurrence rate for STUMP is 7%–27%.^[13] Recurrence can occur many years after the initial operation, and long-term follow-ups must be implemented. Due to the indolent nature of the tumor and previous history of recurrence in this case, it is recommended to follow-up with the patient every 6 months with abdominopelvic CT imaging.^[14,15]

CONCLUSION

Recurrence of intraperitoneal tumors is a potentially deleterious complication of laparoscopic myomectomy with uncontained morcellation. Extreme caution is called for to prevent specimen spillage during the retrieval process. Furthermore, histopathology reexamination of the initial surgery specimen is recommended if there are findings of the pathology of greater severity.

Ethical statement

This study was approved by Chang Gung Medical Foundation Institutional Review Board, IRB number: 202000804B0. The patient consent was waived by the IRB.

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Conflicts of interest

Dr. Kuan-Gen Huang, an editorial board member at *Gynecology and Minimally Invasive Therapy*, had no role in the peer review process of or decision to publish this article. The other authors declared no conflicts of interest in writing this paper.

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