

A case of recurrent smooth muscle tumor of uncertain malignant potential with abdominal dissemination after morcellation

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Summary

Objective: Smooth muscle tumor of uncertain malignant potential (STUMP) shares some characteristics with sarcoma but does not meet the full diagnostic criteria. Morcellation is a technique used to extract surgical specimens through a small incision, thereby spreading unsuspected malignant cells. We report a case of STUMP that recurred postoperatively in the abdominal cavity. **Case Report:** Surgery was performed on a patient diagnosed with uterine leiomyoma, with the tumor morcellated transvaginally. Twelve months later, several abdominal tumors appeared. Pathological specimens were reevaluated, and STUMP was diagnosed. Many tumors in the abdominal cavity of the patient were removed surgically. The postoperative pathologic diagnosis was leiomyosarcoma. **Conclusion:** This case of unsuspected STUMP recurrence after transvaginal scalpel morcellation illustrates the risk of spillage of malignant cells. Surgeons and surgical pathologists must stay alert to the possibility of occult malignancy, and morcellated tissues should be carefully examined.

Key words: Leiomyoma; STUMP; Transvaginal scalpel morcellation; Laparoscopy; Total hysterectomy.

Introduction

Smooth muscle tumor of uncertain malignant potential (STUMP), a variant of smooth muscle tumor, is a rare neoplasia with unclear clinical behavior. Morcellation, by which solid masses are removed through a small abdominal incision or the vagina, has dramatically increased the number of minimally invasive surgeries performed. When a morcellated tumor is malignant, the risk of dissemination of malignant cells within the abdominal cavity increases significantly. Worsened outcomes are reported among patients who have undergone morcellation [1, 2]. The authors describe a case of recurrent STUMP in a patient who underwent laparoscopic surgery and transvaginal scalpel morcellation for presumed uterine leiomyoma. Recurrence was derived from peritoneal and retroperitoneal dissemination. The patient provided consent for her case details to be reported under the conditions of anonymity.

Case Report

A 50-year-old woman (secundipara) previously treated for a degenerating uterine leiomyoma was diagnosed following identification on T2-weighted MRI of a 10-cm mass with mixed signal intensity located under the uterine serosa. The patient's medical history was unremarkable, and her cervical and endometrial cells as well as lactate dehydrogenase were normal, removing suspicions of malignancy. Total laparoscopic hysterectomy, including enucleation, was performed, with the surgical specimen removed via transvaginal scalpel morcellation. Morcellation was not performed in a bag. The postoperative pathological diagnosis was also leiomyoma. Approximately 12 months later, the patient complained of a sensation of ab-

dominal fullness. Computed tomography revealed bilateral ovarian swelling and several solid tumors in the peritoneal and retroperitoneal spaces. She was admitted to the hospital with suspected metastatic ovarian tumor, malignant lymphoma, or gastrointestinal stromal tumor. Upon pelvic examination, a pelvic mass with poor mobility was palpated. The complete blood count and blood serum components were normal. Tumor markers (CEA, CA19-9, CA125, SCC) were normal, but lactate dehydrogenase was slightly elevated at 257 IU/L. Neither upper gastrointestinal endoscopy nor colonoscopy revealed any abnormalities. The authors reevaluated the pathological specimens from the first surgery and diagnosed the mass as a STUMP (Figure 1a). Macroscopically, a few atypical cells were mixed with normal cells, and 3-4 mitotic figures/high-power field (hpf) were observed. The authors suspected recurrence of a smooth muscle tumor and planned a surgical resection. During open surgery, they found numerous tumors in the peritoneal and retroperitoneal spaces of the patient's abdominal cavity and removed all tumors. One tumor in the retroperitoneal space involved the left ureter, so they excised the tumor and a segment of the ureter near its point of entry into the bladder and performed a ureteral anastomosis. The postoperative pathological diagnosis was leiomyosarcoma (Figure 1b). Adjuvant chemotherapy comprising six cycles of docetaxel and gemcitabine was administered. There is no further evidence of disease at seven months after surgery.

Discussion

STUMP has some characteristics of sarcoma but does not meet the full diagnostic criteria. Because STUMP is

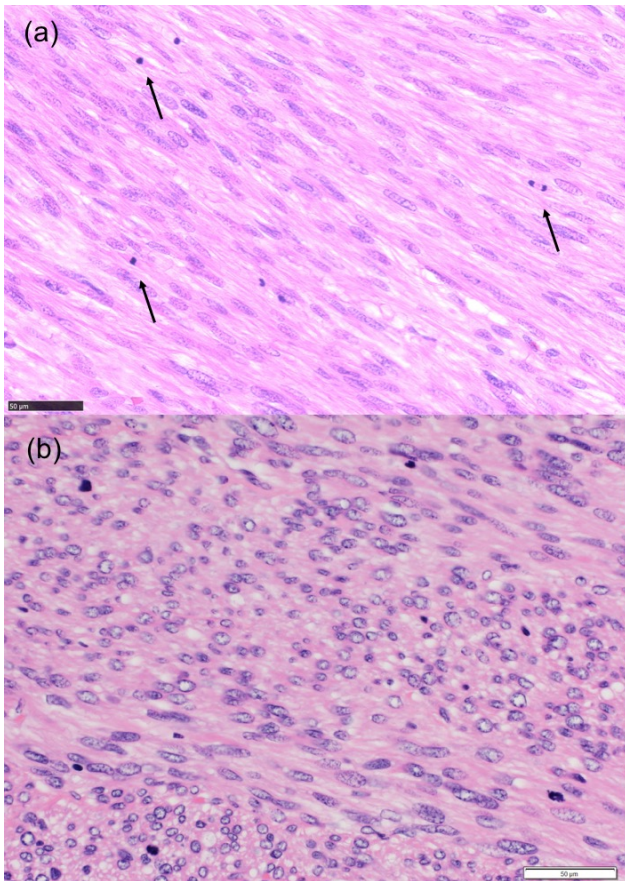


Figure 1. — (a) High-power microscopic view of a tissue section prepared for postoperative histopathologic examination and reexamined at the hospital. Proliferation of spindle-shaped cells is evident. A few atypical cells and 3–4 mitotic figures are observed. (b) High-power microscopic views of a tissue section from the present surgery prepared for postoperative histopathologic examination at the hospital. Proliferation of atypical bundled and overlapping spindle-shaped cells are also observed.

rare, its clinical course is unclear. As a diagnostic criterion, Bell et al. reported the absence of necrosis and < 10 mitotic figures/10 hpf [3]. The reported risk of recurrence is estimated to be 17% [4]. An increasing number of gynecologic laparoscopic surgeries have prompted the need to extract one or more large surgical specimens (uterus or leiomyoma, for example) through a small incision, but extraction is not without risks. The reported risk of unknowingly removing a leiomyosarcoma during surgery for a presumed fibroid is 0.22% [5]. Morcellation of uterine sarcoma appears to be associated with worsened outcomes. A meta-analysis comparing intra-abdominal morcellation versus no morcellation showed significantly higher rates of recurrence (61% vs. 39%) and mortality (48% vs. 29%) with morcellation [3]. Because of the potential for peritoneally seeding malignant cells, the US Food and Drug Administration (FDA) issued an advisory in 2014 against using laparoscopic electric morcellators [6]. Some of these patients had undergone a hysterectomy, and once the uterus is morcellated, pathologic examination of the endometrium, en-

docervix, and ectocervix is much more difficult than that of an intact uterus after open surgery [7]. Recurrence following transvaginal scalpel morcellation in the present patient, which involved a ureter, was evidence that intraperitoneal and retroperitoneal dissemination of the STUMP had occurred. Following reevaluation of the original surgical specimen, the pathological diagnosis was STUMP, but the recurrence was diagnosed as leiomyosarcoma. Postsurgical diagnosis was based on pathologic examination of only two tissue fragments from the morcellated surgical specimen. However, the present authors assume that a high malignant potential would also be noted in the non-examined fragments. FDA recommendations are causing laparoscopic procedures to decrease in popularity. However, they remain advantageous because of the small incisions and relatively quick postoperative recovery. There are increasing reports of “in-bag morcellation”, which is performed to avoid tumor dissemination [8, 9]. Although its safety and efficacy require further study, this method may be beneficial. This case of unsuspected STUMP recurrence after transvaginal scalpel morcellation illustrates the risk of spillage and spread of malignant cells, which were disseminated widely in the peritoneal and retroperitoneal spaces. Laparoscopic surgeons and surgical pathologists must remain alert to the possibility of occult malignancy, and morcellated specimens should be examined carefully.

Acknowledgments

We would like to express my gratitude to all those who helped me during the writing of this manuscript.

Conflict of interest

The authors declare no conflict of interest.

Submitted: February 04, 2019

Accepted: April 29, 2019

Published: August 15, 2020

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