

Iatrogenic Parasitic Myoma with Two Recurrence Times after Subsequent Myomectomy: A Rare and Complicated Case Report

Pei-Chen Li¹, Ming-Hsing Lee², Yu-Chi Wei¹, Yung-Hsiang Hsu², Mun-Kun Hong^{1,3*}

¹Minimally Invasive Gynecology Surgery Center, Department of Obstetrics and Gynecology, Hualien Tzu-Chi Hospital, Buddhist Tzu Chi Medical Foundation, ²Department of Pathology, Hualien Tzu-Chi Hospital, Buddhist Tzu Chi Medical Foundation, Tzu Chi University, ³Institute of Medical Sciences, Tzu Chi University, Hualien, Taiwan

Abstract

Increasing number of parasitic myoma (PM) cases due to specimen morcellation during minimally invasive surgery have been reported. The patient was a 46-year-old woman receiving laparoscopic subtotal hysterectomy due to fibroids. She was diagnosed as having PM and had two recurrences after subsequent myomectomies. To prevent recurrence, specimen-contained morcellation was performed during the myomectomies and postoperative ulipristal acetate was given, but with no effects. The interval between each recurrence decreased. Progressive lower abdominal pain and prominent vessels on the myoma were the two distinct clinical characteristics that differentiated PM from general myoma. This case study highlights the importance of specimen containment before morcellation in minimally invasive surgery and implies that the pathogenesis of PM recurrence is unknown.

Keywords: Parasitic myoma, power morcellator, specimen removal

INTRODUCTION

Uterine myomas are the most prevalent benign pelvic tumors in women of reproductive age. Myomas are also the most common indication for hysterectomy, accounting for 30%–50% of hysterectomies in the United States. With the advances in minimally invasive surgery, laparoscopic myomectomy has been extensively utilized for treating fibroids. During laparoscopic surgery, power morcellation devices have been used to remove the specimen through small incisions. However, dissemination of benign or malignant tissue is an increasing concern, and sequelae have been reported.

Iatrogenic parasitic myoma (PM) was considered to originate from fragments of myoma tissue being left in the peritoneal

cavity. The incidence of iatrogenic PM was reported to be 0.12%–0.9%;^[1] however, it has been more frequently reported in the recent decades, probably because of the extensive use of power morcellation during minimally invasive procedures. Herein, we describe a rare case of iatrogenic PM that occurred after laparoscopic subtotal hysterectomy. This case was complicated, with two recurrence times after subsequent parasitic myomectomy.

CASE REPORT

A 46-year-old nulliparous woman visited our hospital due to progressive lower abdominal pain. Sonography

Address for correspondence: Dr. Mun-Kun Hong, Minimally Invasive Gynecology Surgery Center, Department of Obstetrics and Gynecology, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation., No. 707, Chung-Yang Road, Sec. 3, Hualien, Taiwan. E-mail: jeff06038@gmail.com

Article History:

Submitted: 5 September 2019

Revised: 13 November 2019

Accepted: 4 February 2020

Published: 1 August 2020

Access this article online

Quick Response Code:



Website:
www.e-gmit.com

DOI:
10.4103/GMIT.GMIT_88_19

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How to cite this article: Li PC, Lee MH, Wei YC, Hsu YH, Hong MK. Iatrogenic parasitic myoma with two recurrence times after subsequent myomectomy: A rare and complicated case report. *Gynecol Minim Invasive Ther* 2020;9:154-8.

revealed uterine myomas of size 4.4 cm × 3.5 cm and 5.5 cm × 5.3 cm at the anterior and posterior walls, respectively. She underwent laparoscopic supracervical hysterectomy in May 2013 [Table 1]. Laparoscopic observation revealed two globular protruding uterine myomas with prominent vessels on the serosa and a bilateral adnexa that appeared normal [Figure 1a]. After cervical transaction, the uterine body together with myomas was removed using a power morcellator through the 10-mm port. The pathology report revealed leiomyoma with focal hyaline degeneration.

Approximately 2 years later, in 2015, she again presented to the emergency department due to progressive lower abdominal pain. Computed tomography (CT) revealed multiple heterogeneous enhancing pelvic tumors ranging from 2 to 10 cm [Figure 1b]. Diagnostic laparoscopy revealed four solid tumors on the dependent part of the abdomen [Table 1], which were attached to the sigmoid colon, right side of the bladder [Figure 1c, PM1 and PM2], peritoneum of the right common iliac artery [Figure 1d, PM3], and *cul-de-sac*. Sigmoid colon injury had occurred during parasitic myomectomy, which was repaired by a general surgeon. The four solid tumors were removed using manual morcellation with the specimen contained in a surgical tissue bag [LapSac, Cook Medical, USA; Figure S1]. The

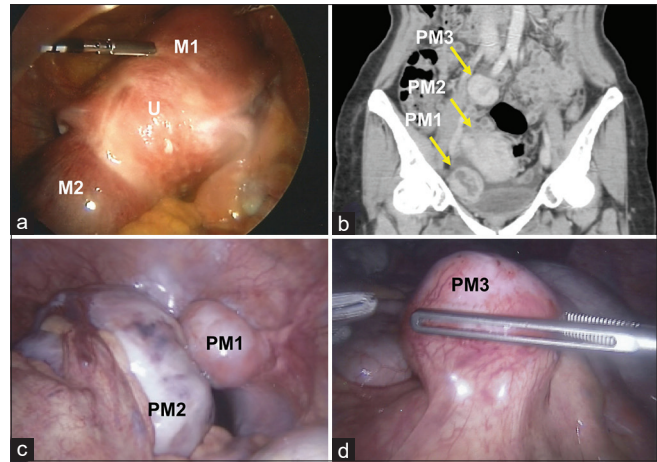


Figure 1: (a) During the initial laparoscopic subtotal hysterectomy, an enlarged uterine (U) with two globular and protruding myomas (M1, M2) were found at the anterior and posterior walls. (b) Computed tomography shows multiple heterogeneous enhanced solid tumors in the pelvic abdomen, with size ranging from 2.5 to 10 cm, located near the right side of the bladder (parasitic myoma 1), on the sigmoid colon (parasitic myoma 2), and the right common iliac artery (parasitic myomas 3) (yellow arrows). (c) Repeat laparoscopic surgery revealed parasitic myoma. The 5.0 cm × 4.5 cm mass (parasitic myoma 1) arises from the right bladder just adjacent to the right ureter and the 10.0 cm × 9.5 cm (parasitic myoma 2) mass arises from the pelvis and is densely adhered to the sigmoid colon. (d) Another 5.0 × 4.0 cm mass (parasitic myoma 3) is present on the peritoneum at the right common iliac artery

Table 1: Summary of the events and characteristics of recurrences of the case

	Event (date)			
	Subtotal hysterectomy (May 03, 2013)	Diagnosis of parasitic myomas (April 22, 2015)	First recurrence (February 24, 2016)	Second recurrence (June 15, 2016)
Clinical presentation	Progressive lower abdominal pain	Progressive lower abdominal pain, especially pain at the right lower quadrant area and radiated to the right flank; poor appetite	Progressive intermittent lower abdominal pain	Left lower abdominal pain
Interval (days)	0	719	308	104
Treatment	Laparoscopic supracervical hysterectomy + bilateral salpingectomy	Laparoscopic parasitic myomectomy + AD	Robotic single-site parasitic myomectomy + AD + UA 5 mg QD till recurrence	Laparoscopy conversion to laparotomic parasitic myomectomy + BO + AD
Myoma/parasitic myoma: Number, appearance, location, size	Two large globular protruding subserosal uterine myomas Posterior wall, 5.5 cm × 5.3 cm Anterior wall, 4.4 cm × 3.5 cm	Four parasitic myomas Sigmoid colon, 10.0 cm × 9.5 cm, densely adhered to the sigmoid colon Right side of bladder, 5.0 cm × 4.5 cm, densely adhered, surrounding the right lower third of the ureter Peritoneum: the right common iliac artery, 5.0 cm × 4.0 cm <i>Cul-de-sac</i> , 2.5 cm × 1.5 cm	Three parasitic myomas in the pelvis Left adnexal anterior near the residual cervix, approximately 4.5 cm × 3.0 cm Right pelvis near the external iliac artery, approximately 3.5 cm × 2.0 cm Peritoneum at the right external iliac artery, approximately 8.0 cm × 5.0 cm	One parasitic myoma was found at the junction of the sigmoid colon and mesentery, 7.5 cm × 5.0 cm
Specimen removal method	Power morcellator	Manual morcellation with specimen contained in a tissue bag	Manual morcellation with specimen contained in a tissue bag	Laparotomy, complete resection
Complication	No	Sigmoid bowel injury	No	Sigmoid colon serosal injury

AD: Adhesiolysis, BO: Bilateral oophorectomy, UA: Ulipristal acetate, QD: Once a day

whole peritoneal cavity was carefully explored and washed with 3000 mL of normal saline; no residual specimens or morcellation remnant of PM was noted. The pathology report indicated the solid masses to be leiomyomas with variable cellularity and focal degenerative change. No increased mitosis was noted. The specimen from the sigmoid colon was adhered to the adventitial adipose tissue of the colon, and no evidence of mural wall invasion was observed.

Ten months later, in February 2016, the patient developed progressive lower abdominal pain again. Transvaginal ultrasonography revealed a pelvic mass of approximately 4 cm in diameter, which was suspected to be a recurrent PM [Figure S2a]. The patient then received a third surgery of robotic single-site parasitic myomectomy. On exploration, three PMs were found on the peritoneum of the left adnexal anterior near the residual cervix, the right pelvis near the external iliac artery, and the right external iliac artery [Figure S2b]. In the lithotomy position, all PM were excised and contained in the same surgical tissue bag before manual morcellation. The whole peritoneal cavity was carefully explored, and no complication and no residual tumor or remnant of PM was noted. Histopathology confirmed the masses as leiomyomas composed of fascicles of the spindle to epithelioid smooth muscles. After the third surgery, the patient was prescribed a selective progesterone receptor (PR) modulator – ulipristal acetate (UA), 5 mg daily, for preventing recurrence. She exhibited good health status and remained asymptomatic until 3½ months, after which she presented with lower abdominal pain again [Table 1]. A solid pelvic mass was detected on transvaginal ultrasonography [Figure S2c]. The fourth surgery was performed in June 2016, and a 7.5 cm × 5.0 cm recurrent PM with prominent vessels

on the tumor surface firmly attached at the junction of the mesentery and sigmoid colon was identified [Figure S2d]. Due to multiple peritoneal dense adhesions, laparoscopic surgery was converted to laparotomy, and tumor excision with bilateral oophorectomy was performed. Sigmoid colon serosal injury occurred during parasitic myomectomy. We carefully explored the whole abdominal cavity through palpation and found no residual tumor. Peritoneal cavity irrigation was performed with 3000 mL of normal saline. The pathology revealed leiomyoma with interlacing myometrial bundles composed of bland smooth muscle cells. The patient was followed up for approximately 3 years after the last surgery, and she is currently in a stable condition with no evidence of recurrence.

DISCUSSION

Several mechanisms of PM have been postulated. Willson and Peale reported the concept of peritoneal metaplasia under the stimulation of hormones, which explains myomas in unexpected fields of the abdomen.^[2] Another study suggests that pedunculated subserosal myomas revascularize from adjacent organs, such as the bowel, peritoneum, omentum, and mesentery.^[3]

In the present case, the PM occurred 2 years after subtotal hysterectomy with a power morcellator, and all of them were located on the dependent part of the pelvis, such as the colon, mesentery, and *cul-de-sac*. Notably, despite the optimal resection of PM, in-bag morcellation of the PM,^[4] and novel treatment with a PR modulator, two recurrences were

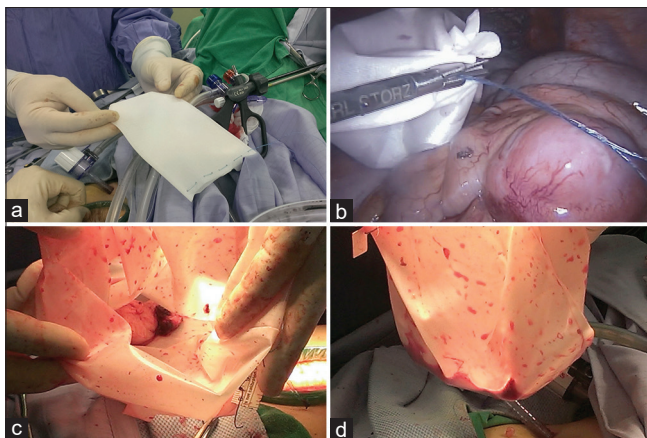


Figure S1: (a) Tissue pouch was folded and inserted into the abdominal cavity. (b) The specimen was contained in the pouch, and the opening of the pouch was closed by pulling up the drawstring. (c and d) Under containment, the parasitic myoma was cut in strips, and all the tissue fragments and fluids from the manual morcellation were contained in the pouch

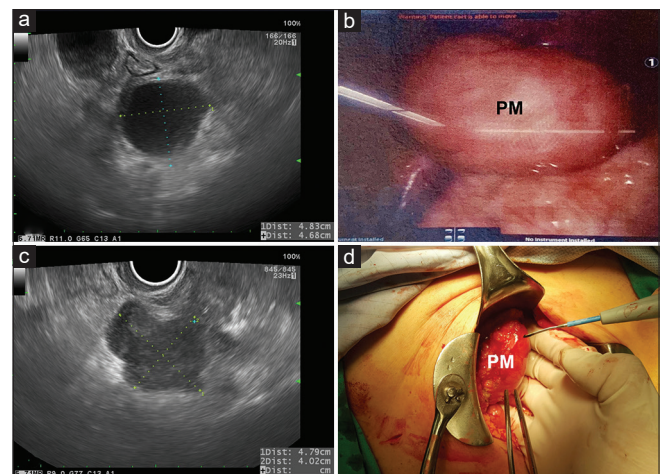


Figure S2: Preoperative transvaginal sonography image shows (a) a 4.83 cm × 4.68 cm hypoechoic pelvic mass before the third operation. (b) Robotic single-site myomectomy shows one of the first recurrence parasitic myomas – a pelvic mass of size 4.79 cm × 4.02 cm – before the fourth operation. (c) A 4.79 cm × 4.02 cm pelvic mass found by transvaginal sonography before the fourth operation. (d) Parasitic myomas at the second recurrence firmly attached to the mesentery of the sigmoid colon detected during laparotomy

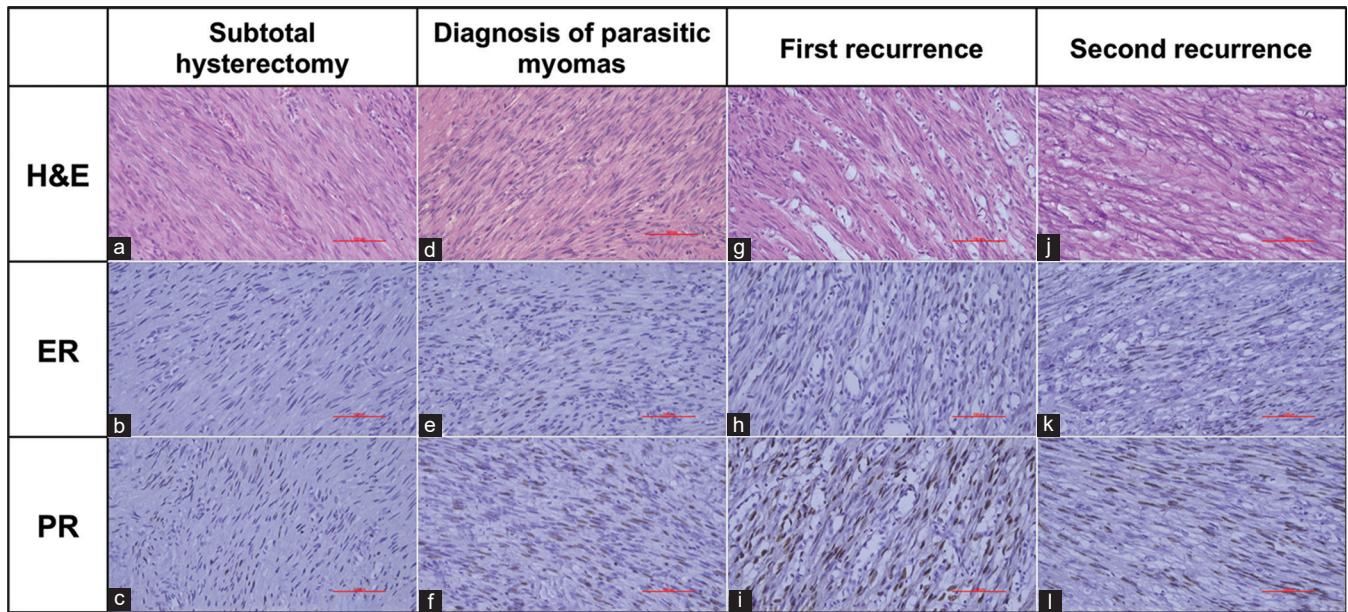


Figure S3: Histopathology findings of the myomas. (a-c) Leiomyoma with focal hyaline degeneration resected during initial laparoscopic subtotal hysterectomy without expression of estrogen receptor and low expression of progesterone receptor. (d-f) Parasitic myoma resected during the second surgery shows variable cellularity and focal degenerative change with a focal expression of estrogen receptor and strong expression of progesterone receptor. (g-i) Parasitic myoma resected at the third surgery showed strong positive estrogen receptor and progesterone receptor. (j-l) Strong expression of estrogen receptor and progesterone receptor of parasitic myoma resected at the fourth surgery

observed. In addition, the interval between each recurrence in this reported case decreased. The recurrence of parasitic leiomyoma is very rare, and only a few cases of recurrent PM after laparoscopic hysterectomy and myomectomy have been reported.^[5,6] Multiple parasitic leiomyoma recurrences have not been reported thus far.

PM present no specific symptoms or typical radiologic features. Thus, the preoperative diagnosis of PM becomes challenging, and the frequency of recurrence is difficult to establish. In one review, 25% of patients were asymptomatic, which depends on the size and location of PM.^[7] In our case, progressive lower abdominal pain and prominent vessels on the myoma were the two distinct clinical characteristics that differed from the general impression of myoma. The ultrasonography of PM is more hypoechoic [Figure 1b] than that generally found in the ultrasonography of myoma. Fibroids usually exhibit degeneration or central necrosis on CT scan when they grow sufficiently large. However, PM appear heterogeneous and contain areas of hypoattenuation on a CT image even in cases of small PM. This feature may be a unique finding on CT for PM and may imply that these PM experienced hypoxia initially and later obtained revascularization and blood supply from the peripheral and adjacent organs.

Laparoscopic excision of PM is the treatment of choice. However, patients should be informed about the possibility of extensive and repeated surgery, as was required in the

present case. Some preventive measures for recurrence have been reported, including containment before morcellation using a tissue pouch, reverse Trendelenburg position after morcellation, and irrigation to reduce seeding of cellular debris. Following such methods during the procedure may reduce the risk of tissue remnants that may cause symptoms in the future. However, in the current case, despite retrieving all specimen fragments by using a surgical tissue pouch during the second and third parasitic myomectomies and washing the abdominal cavity with abundant amounts of normal saline after surgery, PM recurred. This implies that the actual pathogenesis of the recurrence of PM remains unknown.

Immunohistochemical (IHC) staining of the myoma obtained from the initial surgery showed no expression of the estrogen receptor (ER) and low expression of the PR. However, low ER and high PR expression were observed in the nuclei of the PM cells. The PR expression remained strong throughout, whereas the ER expression became stronger in the following two PM recurrences [Figure S3]. Upregulation of ER and PR expression was observed in the PM cells. Growing lines of evidence suggest that ER and PR signaling pathways can crosstalk with growth factors to promote the proliferation of myomas^[8] and neovascularization, supporting the key role of estradiol and progesterone in the pathogenesis of leiomyoma growth.

UA, a selective PR modulator, reversibly blocks the PR in the tissue and exerts antiprogesterone and antiproliferative

effects on fibroid cells. A recent randomized control trial showed that UA effectively eliminates uterine bleeding, reduces fibroid-associated pain, and shrinks fibroid size. Despite positive ER and PR of the leiomyoma tissue in the IHC staining, treatment with UA appeared to be refractory toward the recurrence of PM in this case.

CONCLUSION

In summary, this case emphasizes the importance of specimen containment before morcellation because failure to do so may result in iatrogenic PM throughout the peritoneal cavity and lead to severe morbidity. Despite optimal preventive management, the multiple recurrences, in this case, imply that the pathogenesis of PM recurrence is unknown.

Ethical statement

The study was approved by the Research Ethics Committee of Hualien Tzu Chi Hospital (IRB number CR108-03).

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 understand that name and initials will not be published and due efforts

will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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