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Port-site implantation of parasitic leiomyoma after laparoscopic myomectomy and its histopathology

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Abstract

Although parasitic leiomyoma could be spontaneous or iatrogenic in origin, port-site implantation of parasitic leiomyoma is an iatrogenic benign sequela of laparoscopic surgery. A 30-year-old, primigravida Japanese woman was referred after unresponsiveness to preoperative gonadotropin-releasing hormone for intramural fibroids. Magnetic resonance imaging showed multiple intramural fibroids and left ovarian endometrioma with no malignant features. Laparoscopic myomectomy with power morcellation and ovarian cystectomy were performed, followed by treatment with a combined oral contraceptive. Seven years after the primary surgery, she underwent abdominal myomectomy for a port-site, and peritoneal recurrence of the leiomyoma and intramural leiomyomas was detected. Microscopic examination revealed that resected specimens from the port-site demonstrated leiomyoma with lesser cell density and more prominent hyalinization than those from the uterus. Therefore, clinicians should counsel patients regarding the risks and benefits of laparoscopy with morcellation versus laparotomy. Further development of techniques for uterine tissues extraction is warranted.

Keywords

Leiomyoma, laparoscopy, uterine myomectomy, morcellation, parasitic leiomyoma

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Introduction

Parasitic leiomyoma (PL), defined as ectopic implantation of the uterine myoma, is supplied by new blood vessels originating from other organs and the peritoneum. It can be spontaneous or iatrogenic in origin.[1] Spontaneous PLs are triggered by the detachment of pedunculated sub-serosal leiomyoma, whereas iatrogenic PLs are non-malignant sequelae of tissue dissemination by power morcellation following laparoscopic surgery.[2,3] Lu et al.[4] observed approximately 8000 cases of laparoscopically removed uterine leiomyoma and estimated the prevalence of spontaneous and iatrogenic PLs at 0.21% and 0.07%, respectively. The safety statement issued by the US Food and Drug Administration (FDA) in 2014 recommended avoidance of laparoscopic power morcellation for hysterectomies or removal of uterine fibroids owing to the risk of occult uterine sarcomas spreading beyond the uterus.[5] Similarly, this alert should be considered important for non-malignant uterine tissue extraction during laparoscopic surgery.

Port-site implantation is defined as tumor recurrence developing within the scar tissue of one or more trocar sites or incision wound in the abdominal wall after endoscopy for malignant diseases.[6] It is neither metastasis nor spread of the primary disease, but the development of residual tumor that was not resected during the previous surgery, suggesting an iatrogenic origin. Because of the rarity and unelucidated pathophysiology of this disease, its management is challenging.

This study reports the case of a 30-year-old Japanese woman with port-site implantation of PL after laparoscopic

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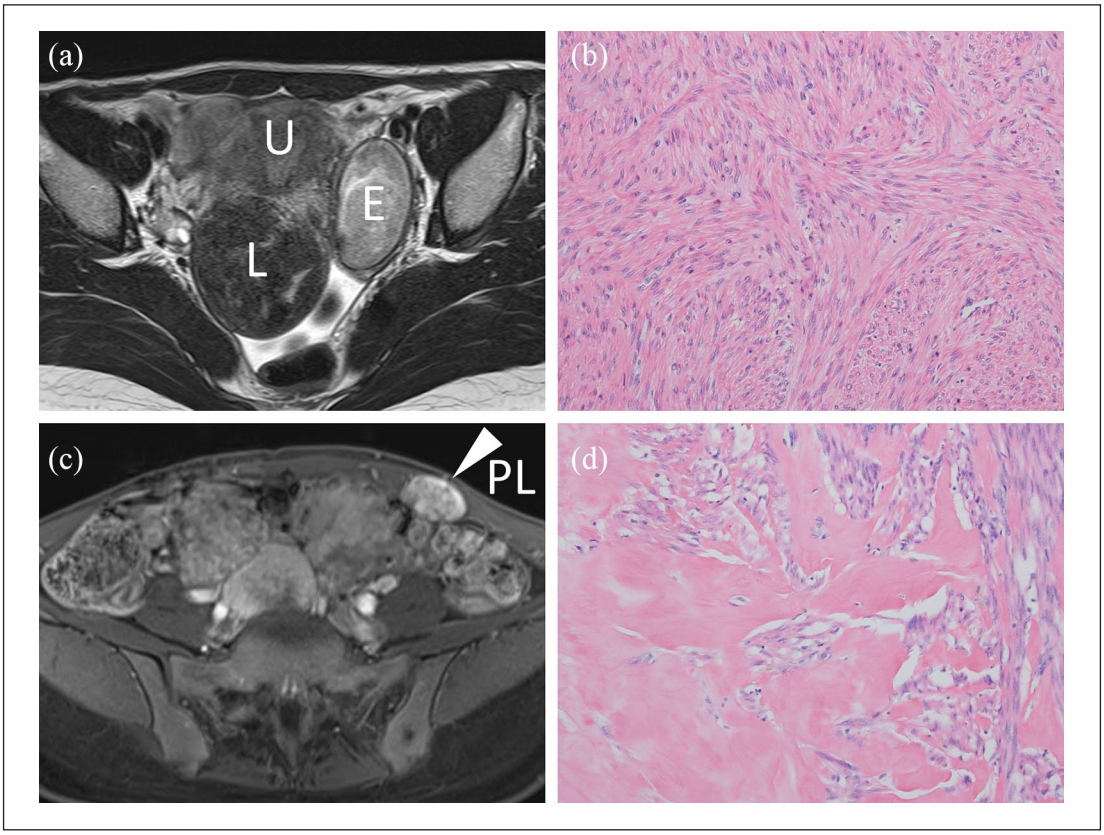

Figure 1. (a) Horizontal view of magnetic resonance imaging (MRI) T2 weighted image demonstrates a 50-mm intramural fibroid (L) and a 60-mm left ovarian endometrioma (E) presumed adhesion to the uterus (U) before primary operation. (b) Microscopic examination of resected specimens at primary surgery shows typical leiomyoma without any specific features (Hematoxylin–eosin stain, 20×). (c) Horizontal view of MRI T2 weighted image 7 years after primary surgery illustrates a 30-mm parasitic leiomyoma (PL, arrowhead) in the left abdominal wall (Hematoxylin–eosin stain, 20×). (d) Microscopic examination of PL in the abdominal wall shows a degenerated leiomyoma with low cell density and prominent hyalinization.

myomectomy (LM) for multiple intramural fibroids and ovarian cystectomy for ovarian endometrioma.

Case

A 30-year-old, primigravid Japanese woman was referred to our department for LM. Six months before, she had visited a primary care doctor owing to menorrhagia and dysmenorrhea and was diagnosed with multiple intramural leiomyomas ranging 4–7 cm in length. A 5.5-cm left ovarian endometrioma was detected on transvaginal ultrasound and magnetic resonance imaging (MRI). Although six cycles of gonadotropin-releasing hormone agonists (GnRHas) were administered, follow-up MRI showed enlarged fibroid and stable endometrioma.

A vaginal ultrasound and MRI at our department demonstrated a 50-mm fibroid in the anterior uterine wall, 72- and 39-mm fibroids in the posterior uterine wall, and a 60-mm left ovarian endometrioma with adhesion to the uterine body (Figure 1(a)). The findings did not indicate a malignant potential, and tumor markers and endometrial cytology screening were both negative. The patient was diagnosed with leiomyoma-related abnormal uterine bleeding (AUB-L), International Federation of Gynecology and Obstetrics (FIGO) type 3 (PALM-COEIN),[7] and ovarian endometrioma. LM and left ovarian cystectomy were performed, and three intramural fibroids were removed by power morcellation without in-bag containment system (this operation was performed before the 2014-FDA statement was issued). Similarly, she was diagnosed with endometriosis stage IV (revised American Society for Reproductive Medicine (r-ASRM) classification).[8] Microscopic examination of the resected specimens showed leiomyoma (with arrangement of spindle cells in intersecting fascicles) with infrequent mitosis and no necrosis (Figure 1(b)) and an endometriotic cyst with no specific findings. She was subsequently administered a combined oral contraceptive (COC) of drospirenone and ethinyl estradiol for 11 months, which was switched to norethisterone and ethinyl estradiol because of frequent spotting.

At a follow-up visit 3 years and 9 months after the primary surgery, she felt a mass beneath the left accessory trocar site scar. Contrast-enhanced MRI demonstrated a 16-mm solid tumor in the left port-site, suggesting PL in the abdominal

wall. Similarly, the MRI demonstrated multiple recurrences of intramural leiomyoma, unlike endometrioma. She was strictly monitored while on COC administration because the findings showed no potential malignancy, such as elevation in lactate dehydrogenase levels. However, MRI 7 years after the primary surgery demonstrated an increase in PL to 30 mm in diameter (Figure 1(c)); furthermore, the lesions showing intramural recurrence equally increased, showing a maximum diameter of 96 mm. Furthermore, the PL protruded from her abdominal body when she changed her position. Abdominal myomectomy was performed to relieve her symptoms and histologically diagnose the PL. Excluding the multiple intramural fibroids and the PL identified inside the abdominal wall in association with the left port-site scar, a new PL attached to the peritoneum of the Douglas' pouch was observed during the operation. All these fibroids (15 intramurals; 1 abdominal wall; 1 Douglas' pouch) were excised and extracted without morcellation. Recurrence of endometriosis was not observed in the abdominal cavity. Surgical time was 230 min, blood loss was 680 mL, and her postoperative course was uneventful. Microscopic examination of the resected specimens of the intra-abdominal PL showed spindle cells arranged in intersecting fascicles accompanied by low cell density and prominent hyalinization (Figure 1(d)). There were 0–3 mitoses per 10 high power field (HPF) without necrosis. Although she was prescribed a levonorgestrel-releasing intrauterine system as adjuvant therapy, she preferred watchful waiting. A 22-mm intramural fibroid recurred 8 months after the second surgery; however, PLs did not recur on the latest follow-up visit 17 months after the second surgery. Due to the retrospective nature of this case report, the need for the approval by institutional review board was waived. Written informed consent was obtained from the patient.

Discussion

After LM for multiple intramural fibroids with power morcellation and ovarian cystectomy, the patient subsequently developed port-site implantation of PL and recurrence of intramural fibroids and intraperitoneal PL. Although all lesions were successfully removed by laparotomy, two important clinical issues were learnt in this case. First, PLs could simultaneously develop in the peritoneal cavity and abdominal wall. Second, port-site implantation of PLs showed pathological characteristics different from those of eutopic leiomyomas.

Lete et al. reviewed 274 patients of PL and observed that 154 (56%) had no history of uterine surgery, while 120 (44%) had a previous history of myomectomy or hysterectomy, of which 106 (88%) patients had a history of power morcellation.[1] Power morcellation is considered to contribute to the onset of iatrogenic PLs. PL in the abdominal wall of this case supports this hypothesis because the abdominal wall is anatomically protected by the parietal peritoneum unless disrupted by iatrogenic factors. The efficacy of tissue containment by in-bag morcellation is controversial regarding the non-negligible spread of microscopic cells.[9] In addition, tissue fragments of the leiomyoma may be scattered into the peritoneal cavity during myomectomy or hysterectomy before they are bagged.[10] The efficacy of tissues extraction by scalpel, vaginal, and hybrid mini-laparotomy for PL has not been elucidated in the English literature. Surgeons should perform repeated and copious peritoneal lavage and careful inspection to avoid any tissue remnants.

Since 1997, only four cases of port-site implantation of PL after LM have been reported in the English literature (Table 1). Although the pathological diagnosis was leiomyoma in all cases, there was no description of the morphological difference between eutopic and ectopic lesions except in this case. In this case, the PL showed a leiomyoma with a low cell density and prominent hyalinization. We speculated that these changes were degenerative and derived from secondary growth after implantation caused by an insufficient blood supply from other organs. This finding is in accordance with those reported by Iida et al.[2] However, it is unclear why only a few patients among many who underwent LM or hysterectomy developed PLs.

Intracapsular myomectomy, the removal of fibroids from its pseudocapsule, might be a promising surgical technique not only for uterine myometrial heeling but also for preventing PLs. A pseudocapsule is defined as an anatomic structure surrounding fibroid and separating it from the normal myometrium which has a high angiogenesis potential derived from a growth factor.[14] Iatrogenic damage and spread of pseudocapsule by power morcellation could promote the implantation of minced leiomyoma fragments at an ectopic site. Furthermore, considering the estrogen-dependent nature of PLs, postsurgical hormonal therapy should be considered. Haung et al. explored the effect of four different reagents on morcellation-induced PL in a mouse model and observed that aromatase inhibitors (AIs) significantly decreased the implantation of PLs compared with control; on the contrary, GnRHa and selective estrogen receptor modulators (SERMs) showed no significance difference.[15] Nevertheless, in premenopausal women, 99% of circulating estradiol (E2) is produced in the ovaries. Therefore, E2 suppression by GnRHa through the downregulation of the hypothalamic–pituitary–ovarian axis and AIs through the inhibition of conversion from testosterone to E2 should be theoretically effective.

Conclusion

Port-site implantation of PL is a rare but laparoscopy-specific non-malignant sequela. We observed that PLs are benign leiomyomas with some pathological differences from eutopic leiomyomas. To date, the effect of CO_2 insufflation and specific gene mutation, as *MED12* in eutopic fibroids, on PLs remains unclear. Similarly, the reduction of the risk of benign or malignant sequelae after laparoscopic surgery

Table 1. Reported cases of port-site implantation of parasitic leiomyoma.

Author	Year	Age (years)	History of morcellation	Adjuvant therapy	Time after previous surgery (months)	Treatment	Pathology	Outcome
Ostrzenski et al.[11]	1997	43	None	COC[a]	2	Local removal	Leiomyoma	N/A[b]
Moon et al.[12]	2008	31	Yes	None	36	Local removal	Leiomyoma	N/A
Oindi et al.[13]	2018	44	Yes	None	36	Abdominal hysterectomy	Leiomyoma	No recurrence of PL 6 months after surgery
Present case	2019	36	Yes	COC	45	Abdominal myomectomy and local removal	Leiomyoma with low cell density and prominent hyalinization	No recurrence of PL 17 months after secondary surgery

PL: parasitic leiomyoma.
[a]Combined oral contraceptive.
[b]Not acceptable from the literature.

by in-bag morcellation remains unclear. Therefore, clinicians should counsel patients regarding the risks and benefits of laparoscopy with morcellation versus laparotomy.

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Informed consent

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