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Case Report

Leiomyomatosis peritonealis dissemianata five years after laparoscopic uterine myomectomy: A case report

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ABSTRACT

Introduction: Leiomyomatosis peritonealis disseminata (LPD) is a rare disease that can be challenging to diagnose. In this report, we present a case of LPD arising 5 years after laparoscopic uterine myomectomy using a power morcellator.

Presentation of case: A 32-year-old woman was admitted to our hospital with complaints of vaginal bleeding and abdominal discomfort. Five years previously, she had undergone laparoscopic uterine myomectomy using a power morcellator. Pelvic ultrasonography and magnetic resonance imaging demonstrated multiple pelvic tumors closely attached to peritoneum with no indication of malignancy. An exploratory laparotomy revealed multiple sites of leiomyomatosis in the peritoneum, especially on the parietal peritoneum at the port site of the previous laparoscopic surgery. We surgically removed all visible tumors and performed a total hysterectomy. Histologic examination confirmed the diagnosis of LPD.

Discussion: The use of a power morcellator without in-bag containment system might have played a role in the pathogenesis of LPD in our case. Ultrasonography, computed tomography, and MRI are among the most effective to distinguish between leiomyomas and other solid tumors in the pelvis, but they are not of great help in the differential diagnosis of malignancies.

Conclusion: The physicians need to combine medical history, clinical findings, imaging techniques and histopathological examination to establish a correct diagnosis of LPD. The application of containment bags in the setting of power morcellation should be considered to reduce the risk of developing LPD. The optimal intervention strategy should be chosen according to the particular features of each patient.

1. Introduction

Uterine leiomyomas are the most common benign pelvic tumors in women, occurring in approximately 25% of reproductive-aged women [1]. Leiomyomatosis peritonealis dissemianata (LPD) is a rare condition characterized by the formation of multiple intraperitoneal leiomyomas [2]. LPD was first described in 1952 by Wilson and Peale [3], and currently, there have been about 200 cases reported in the literature [4]. Several risk factors have been identified for LPD, including female sex hormones, subperitoneal mesenchymal stem cells, metaplasia, genetic, and iatrogenic [5]. It is believed that LPD is associated with an increased steroid hormone status, such as oral contraceptive use, pregnancy, and steroid-secreting tumors [3].

In recent years, certain surgical manipulations, including the use of power morcellation, can significantly increase the risk of dissemination of uterine benign and malignant tumor tissues during laparoscopic surgery [6]. Morcellation is a procedure for dividing and removal of organs and tissues directly from a cavity without excessive tissue dissection. After morcellation, small fragments of the uterus or myoma can be implanted into the abdominal cavity and take a blood supply from neighboring structures. Although parasitic leiomyomas still grow in a benign fashion, they can be dangerous depending on their location, especially if they involve the heart or lungs [7].

LPD has an atypical clinical presentation and can be misdiagnosed with peritoneal carcinomatosis. Basically, diagnosis of LPD is established based on a combination of clinical and radiological findings

Abbreviations: LPD, Leiomyomatosis peritonealis dissemianata; MRI, magnetic resonance imaging; CA-125, cancer antigen 125; CEA, Carcinoma Embryonic Antigen; CA19-9, cancer antigen.

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(ultrasonography, computed tomography, and magnetic resonance imaging). Pathological examination of the surgical specimens helps confirm the final diagnosis. There is no consensus regarding the optimal treatment strategies for LPD; however, hormonal therapy and surgery are still considered as the mainstays of treatment.

In this report, we describe the unique case of a patient who developed disseminated peritoneal leiomyomatosis five years after a laparoscopic myomectomy with power morcellation.

2. Presentation of case

A 32-year-old woman, gravida 2, para 2, was admitted to our hospital with complaints of vaginal bleeding and abdominal discomfort for the past two months. After checking the clinical notes, we found that she underwent laparoscopic uterine myomectomy using a power morcellator 5 years ago for the treatment of 8 × 7cm uterine myoma. The patient had no history of drug use. Her family history was unremarkable.

On the day of admission, a physical examination revealed a mild rebound tenderness over her left adnexa. Her uterus was enlarged to 16 weeks' size. Laboratory evaluation demonstrated a hemoglobin value of 151 g/l, red blood cells of 4.72 × 10 T/l. Her serum cancer antigen 125 level increased (553 U/mL). Pelvic ultrasonography detected an indeterminate left adnexal mass measuring 11 × 9,5cm with normal Doppler. Magnetic resonance imaging (MRI) was then performed to determine the exact location of her adnexal mass. It demonstrated multiple pelvic tumors closely attached to uterus and peritoneum, measuring 13 cm in maximum diameter with no indication of malignancy (Fig. 1). She was diagnosed with leiomyomatosis peritonealis disseminata. The surgical procedure (type of incision, risks, possible complications …) was explained by our consultant and informed consent for total hysterectomy was signed by the patient taking into consideration she had completed her family. Laparotomy with midline incision revealed a uterus enlarged with 2 subserous tumors (8 and 6 cm in diameter). Multiple sites of leiomyomatosis were visualized in the peritoneum over the anterior wall of rectum (15 × 12cm) and the bladder (6 × 5cm). Of note, 3 fibroids (ranging from 1 to 3 cm) were identified on the parietal peritoneum at the port site of the previous laparoscopic surgery. A total hysterectomy was successfully performed and all tumors were completely resected. Hemostasis was achieved with electrocautery and stitches (Vicryl 2-0 or 3-0).

Histologic examination of the surgical specimens confirmed the diagnosis of leiomyomatosis peritonealis disseminata. Histological structure of biopsy material was characterized by the fibrous and muscular bundles with the presence of spindle-shaped cells (Fig. 2). The patient made an uneventful recovery and was discharged from hospital 5 days after surgery. She was followed up without postsurgical hormonal therapy. At a scheduled visit two months later, she was asymptomatic with a hemoglobin value of 130 g/l, red blood cells of 4.5 × 10 T/l.

3. Discussion

LPD is an extremely rare clinical condition. Two main theories of the

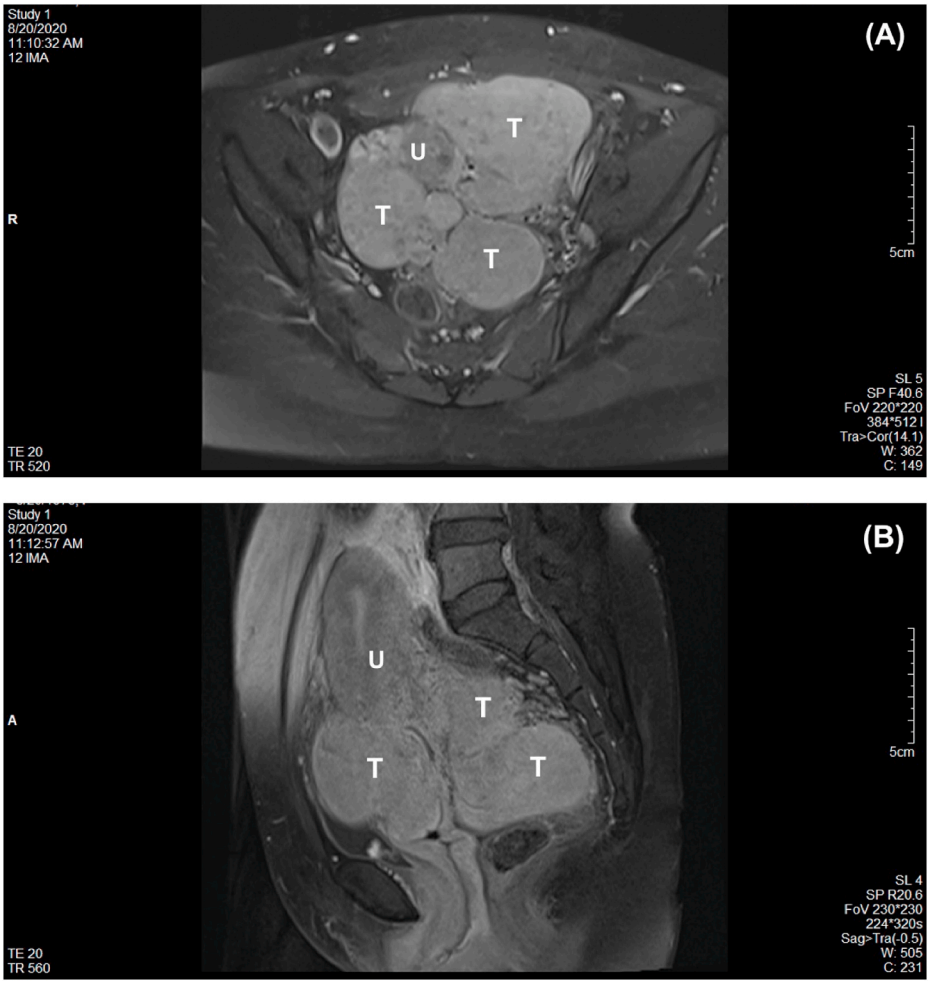

Fig. 1. Preoperative MRI revealed multiple pelvic tumors attached to uterus and peritoneum (T: tumor, U: uterus).
(A) Transverse view
(B) Sagittal view.

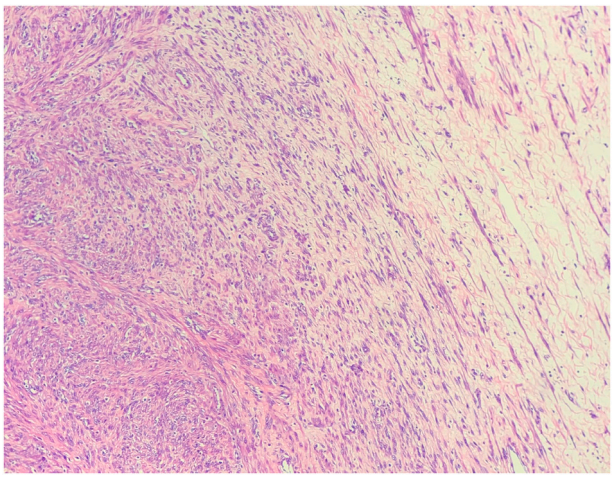

Fig. 2. Histopathological findings of resected specimens: microscopic examination showed the fibrous and muscular tissue with the presence of spindle-shaped cells (Hematoxylin–eosin staining, 100 ×).

etiology and pathophysiology of LPD have been reported: a hormonal theory with metaplasia of mesenchymal stem cells and an iatrogenic origin after laparoscopic surgery [2]. According to the hormonal theory, it is assumed that LPD results from the metaplastic change of mesenchymal stem cells with exposure to high levels of female gonadal steroids [8]. However, recent publications have highlighted a link between laparoscopic uterine myoma morcellation and the development of LPD [9]. Morcellation is a useful procedure for fragmenting and extracting specimens during laparoscopic surgery without the need to perform a laparotomy. However, the main morcellation-related complication is the risk of spreading unsuspected cancerous tissues during hysterectomy or myomectomy. Laparoscopic in-bag morcellation of fibroids or uterus may be helpful in preventing tumor seeding [10,11]. In our case, the use of a power morcellator without in-bag containment system might have played a role in the pathogenesis of LPD.

LPD is associated with non-specific clinical signs and symptoms, such as irregular, heavy uterine bleeding, pain or discomfort, gastrointestinal bleeding and peritonitis [12]. Abdominal pain, as in this case, is a common manifestation of LPD [13]. Some reported cases described the presence of ascites and endometriosis in patients with LPD [14]. However, we were completely unaware of these conditions in our patient.

Clinical history, physical examination and imaging techniques are beneficial to preoperative detection of LPD. In present case, clinical history is especially important. The physician should suspect parasitic leiomyoma when the woman has a history of hysterectomy or myomectomy, particularly if a power morcellator was used. Ultrasonography, computed tomography, and MRI are among the most effective to distinguish between leiomyomas and other solid tumors in the pelvis, but they are not of great help in the differential diagnosis of malignancies. LPD may be confused with peritoneal carcinomatosis [15] or gastrointestinal tumors due to similar imaging characteristics. In these circumstances, tumor markers such as CEA, CA19-9 and CA-125 seem not to be useful for the differential diagnosis, since LPD are sometimes associated with elevated levels of these markers [16,17]. The final diagnosis is normally based on intraoperative observations and histopathological examination.

There is little data on the most appropriate treatment for LPD. Recently, determination of the therapy according to the patient's age, symptoms, child-bearing requirement and past treatments has been proposed [18]. For women with reproductive desire, hormonal therapy with gonadotropin-releasing hormone injection, aromatase inhibitor, or selective progesterone receptor modulator is usually the first-line treatment option. This approach is also preferred in the prevention of postoperative recurrence [19,20]. For women without reproductive desire, a more extensive surgical procedure with total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, myomectomy, and excision/debulking of the nodules may be the best alternative [21]. Surgical treatment is also strongly recommended in cases of high risk of malignant degeneration.

4. Conclusion

Our case report illustrates that LPD is remarkably challenging to diagnose and manage. Firstly, the physicians need to combine medical history, clinical findings, imaging techniques (ultrasound, computed tomography, MRI) and histopathological examination to establish a correct diagnosis. Secondly, the physicians need to be aware of the complications of morcellation during laparoscopic myomectomy or hysterectomy. It is essential to carefully check the abdominal cavity after using a power morcellator to avoid seeding myoma fragments. The application of containment bags in the setting of power morcellation should be considered to reduce the risk of developing LPD. Thirdly, hormonal therapy and surgery are still the primary treatments for LPD. The optimal intervention strategy should be chosen according to the particular features of each patient.

Ethics approval

Since this study used only de-identified patient data, and published data from literature, no approval from our institutional review board (IRB) was required.

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Author contributions

NMT, DHT, NTHA, TDC: Conceptualization, Methodology.
NTHA, NMT: Writing – Original Draft, Visualization.
NMT, DHT, TDC: Supervision, Writing – Review & Editing.
The final manuscript was approved by all authors.

Registration of research studies

1. Name of the registry:
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Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Availability of data and materials

Not applicable.

Declaration of competing interest

The authors declare that they have no competing interests.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102377.

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