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Leiomyomatosis peritonealis disseminata mimicking peritoneal carcinomatosis 13 years after laparoscopic uterine myomectomy: A case report

Shih-Feng Huang^a, Chen-Yueh Wen^a, Cheng-I. Liao^b, Jung-Chia Lin^c, Cheng-Chung Tsai^{d,*}

^a Department of Surgery, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan, ROC

^b Department of Obstetrics and Gynecology, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan, ROC

^c Department of Pathology and Laboratory Medicine, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan, ROC

^d Division of General Surgery, Department of Surgery, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan, ROC

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ABSTRACT

INTRODUCTION: Leiomyomatosis peritonealis disseminata (LPD) is a rare clinical condition that can be challenging to diagnose because its clinical features mimic other conditions. We present a case of LPD mimicking peritoneal carcinomatosis 13 years after laparoscopic uterine myomectomy using a power morcellator. The aim of this paper is to report a rare case which surgeons can learn from and to provide more clinical information for further studies to investigate LPD.

PRESENTATION OF CASE: A 49-year-old woman was referred to us because sonography revealed abnormal abdominal and pelvic nodules. Thirteen years previously, she had undergone laparoscopic uterine myomectomy using a power morcellator. An exploratory laparotomy revealed nodules on the peritoneum, greater omentum, intestinal mesentery, and terminal ileum. We surgically removed all visible nodules and performed bilateral salpingo-oophorectomy. LPD was confirmed based on the morphology and immunohistochemistry results.

DISCUSSION: Diagnosing LPD preoperatively may be difficult because its clinical manifestations resemble peritoneal carcinomatosis or metastatic lesions. Abdominal pain due to diffuse tumor growth is a common manifestation. LPD degenerating into malignancy is rare, but possible. The probable etiological factors, clinical manifestations, and treatment options which may aid when dealing with LPD have been described in this report.

CONCLUSION: LPD should be considered in women, particularly those with a history of gynecologic surgery presenting with disseminated intraabdominal or pelvic tumors.

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1. Introduction

Leiomyomatosis peritonealis disseminata (LPD) is a rare clinical condition that was first described in 1952 by Wilson and Peale [1]. Till date, there have been less than 200 cases reported in the literature, and studies regarding prevalence and incidence are lacking. The correct diagnosis of LPD is crucial because of the possibility of degeneration into malignancy; however, diagnosis can be challenging because LPD's clinical features mimic peritoneal carcinomatosis and metastatic leiomyosarcomas [2]. In this paper, we present a case of LPD mimicking peritoneal carcinomatosis 13 years after a laparoscopic uterine myomectomy performed using a power mor-

cellator. The aim of this paper is to report a case with a rare diagnosis for surgeons to learn about and to provide more clinical information for further studies to investigate LPD.

2. Presentation of case

A 49-year-old Han woman was referred to our institution from a local hospital because of multiple abnormal abdominal and pelvic nodules noted on sonography. She denied any family history of leiomyomas, leiomyosarcomas of the uterus, or other gynecological diseases. She was married and had an obstetric history of G3P2A1. She was of medium income and there was no clinically significant psychosocial history. She had undergone laparoscopic uterine myomectomy performed using a power morcellator 13 years previously for the treatment of a large uterine myoma, and she underwent laparoscopic total hysterectomy 2 years later due to recurrence of the myoma. She did not have a previous history

* Corresponding author at: No. 386, Ta-Chung 1st RD, Kaohsiung, Taiwan, ROC.

E-mail addresses: odafeng7@vghks.gov.tw (S.-F. Huang), cctsay@vghks.gov.tw (C.-C. Tsai).

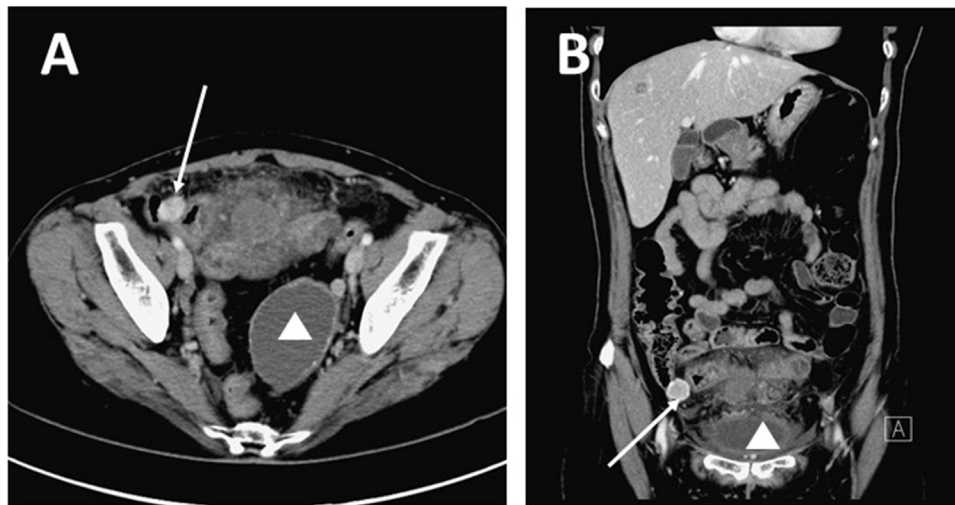


Fig. 1. Preoperative abdominal CT (transverse view) revealed (A) a large left-ovarian cystic lesion (61 × 38 mm) with an irregularly thickened wall (white triangle), a hyperdense terminal ileum nodular lesion (3 × 2.8 cm, white arrow), and multiple mesenteric nodules; (B) coronal view of the abdominal CT.

of prolonged use of contraceptive pills or other medications. The patient had no other significant medical or surgical history. She had started to experience dull intermittent lower abdominal pain approximately 2 months previously and visited a local hospital for this. Abdominal sonography revealed disseminated tumor-like lesions of various diameters in the abdominal and pelvic cavity. Furthermore, a large (6.1 cm) left ovarian cystic mass was noted. Abdominal carcinomatosis of unknown origin was suspected at first, and she was referred to our hospital for further evaluation and management. Abdominal computed tomography (CT) aroused suspicion of seeding tumors in the pelvic fossa and distal ileal mesentery without enlarged lymph nodes. Also noted in the CT study was a poorly enhanced 3.8-cm mass attached to the terminal ileum, a 6-cm cystic lesion with a calcified rim at the left adnexa, and two well-enhanced nodules at the pelvis with approximate diameters of 1.8 cm and 1 cm, respectively, as depicted in Fig. 1. Endoscopic examinations were conducted. Colonoscopy revealed no notable findings, but esophagogastroduodenoscopy revealed a 1.2-cm lesion over the anterior wall of the gastric antrum. However,

the lesion turned out to be a benign tubulovillous adenoma with low-grade dysplasia. Tumor markers carcinoembryonic antigen (CEA), carbohydrate antigen 19-9 and 125 (CA19-9, and CA-125) were all within normal range.

Under the impression of abdominal carcinomatosis of ovarian or unknown origin, an exploratory laparotomy was performed by a gastrointestinal surgeon who had 30 years of experience. The surgery was carried out after keeping the patient on an empty stomach with nothing per mouth for more than 8 h. A 10-cm midline vertical incision was made on the lower abdomen for an exploratory laparotomy. Severe adhesions over the proximal ascending colon were encountered and we performed adhesiolysis first. Multiple nodules of various sizes were noted over the peritoneum, greater omentum, intestinal mesentery, and terminal ileum. We surgically removed all the visible nodules and some of them were sent for intraoperative frozen section biopsy. These were reported to be mesenchymal tumors without mitotic figures or any other evidence of malignancy. A gynecologist with 15-year experience then took over the surgery to perform following procedures. The left

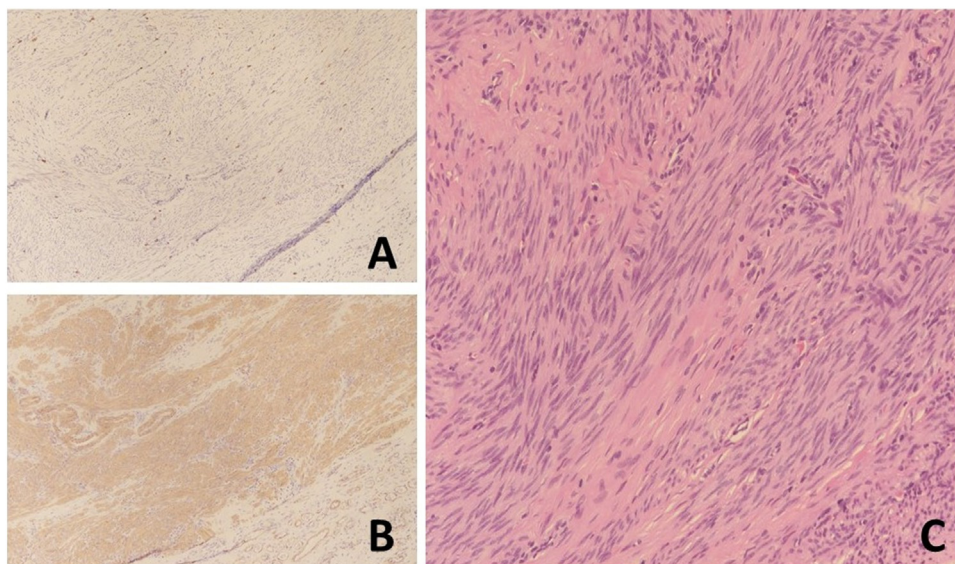


Fig. 2. Under histopathological examination, the mesenteric nodule was found to be composed of spindle cells with (A) negative CD-117 stain under 100× magnification, which could be discriminated from gastrointestinal stromal tumors (GIST), and (B) positive SMA stain under 100× magnification, which was compatible with leiomyoma. (C) The morphology of spindle cells under a high power field (200× magnification) without obvious mitotic activity or necrosis.

infundibulopelvic (IP) ligament of the ovary was identified and carefully dissected separating it from the left ureter. The IP ligament was then ligated and transected. A large left ovarian cyst was noted, and left salpingo-oophorectomy was completed smoothly. Right salpingo-oophorectomy was also performed smoothly with the steps just described. Altogether the surgery took 190 min. Seven days later, the surgical histopathological results of the excised nodules revealed spindle cells that were positive for smooth muscle actin (SMA), estrogen receptor (ER), progesterone receptor (PR), and beta-catenin (cytoplasmic staining) and negative for human melanoma black 45 (HMB45), S100, DOG-1, and cluster of differentiation 117 (CD117) immunostains, as illustrated in Fig. 2. Neither necrosis nor obvious mitotic activity was observed microscopically. Based on the morphology and immunohistochemistry results, LPD was confirmed.

The left ovarian cyst was eventually confirmed to be a mucinous cystadenoma. Chest CT was performed, revealing no pulmonary or pleural involvement. No further treatment was administered, and the patient was discharged 9 days after surgery. At the first outpatient visit after discharge the patient stated that her abdominal pain had improved greatly and we recommended that she should remain under close observation through abdominal CT or MRI every 6 months.

This work has been reported in line with the SCARE 2020 criteria [3].

3. Discussion

LPD is a rare clinical condition, and the etiology and pathophysiology are not yet well understood. Most reported cases have been related to a previous history of laparoscopic uterine myomectomy. The use of a power morcellator in a laparoscopic myomectomy was thought to be a possible cause of LPD [4]. To potentially reduce the risk of such dissemination, Devassy et al. developed a safe and feasible specimen retraction technique using contained endobags (Morsafe®) during laparoscopic myomectomies and hysterectomies [5]. In our case, the use of a power morcellator might have played a role in the pathogenesis; however, cases have been reported without a history of morcellator use [2].

Preoperative diagnosis of LPD can be challenging because of its nonspecific clinical manifestations and radiological features. Abdominal pain, as in this case, is a common manifestation of LPD in reported cases [2]. Many patients with LPD manifest only a few or no symptoms, and therefore, a considerable number of cases may go undiagnosed.

LPD is often mistaken for gastrointestinal or ovarian tumors [6], or peritoneal carcinomatosis, due to similar radiological features. Given that tumor markers such as CEA, CA19-9 and CA-125 were reported to be normal in many cases of LPD in the literature [7,8], they may help clinicians differentiate between LPD and carcinomatosis. Further studies would be needed to investigate the reliability of the tumor markers used in this scenario.

LPD is histologically benign; nevertheless, it could, though rarely, degenerate into malignancy if left untreated [9]. No standard treatment exists for LPD because cases are rare and evidence is lacking. Hormonal therapy with gonadotropin-releasing hormone (GnRH) agonists is usually the first-line treatment option and surgical excision of the leiomyomatosis lesions was reserved for patients who were symptomatic or refractory to conservative treatment. Surgical procedures include total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, myomectomy, and excision/debulking of the nodules [10]. Both conventional laparotomy and minimally invasive laparoscopy are reasonable, though comparison of these two approaches in this scenario is lacking in the literature. Furthermore, estrogen and progesterone levels may be related to the development of leiomyomatosis. Long-term and

high-dose progesterone administration has been reported to cause mesenchymal stem cells to develop into leiomyomatosis in an animal model [11]; thus, discontinuation of oral contraceptives may be helpful. Another study reported a diminished tumor size after discontinuation of contraceptives or after childbirth [12].

4. Conclusion

In conclusion, LPD is a rare clinical condition that mimics abdominal carcinomatosis, and it is mostly associated with previous laparoscopic uterine myomectomy performed using a power morcellator. LPD should be considered in the differential diagnoses of women presenting with disseminated intraabdominal or pelvic tumors, especially those with a history of gynecologic surgery.

Declaration of Competing Interest

There is no existing conflict of interest.

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Ethical approval

This case report was conducted in accordance with the Declaration of Helsinki and was approved by the Institutional Review Board of Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan, R.O.C.

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.

Author contribution

C.C. Tsai, C.Y. Wen, and C.I. Liao were involved in the management of the case. J.C. Lin helped with the pathological interpretation. The images were collected and the main text was written by S.F. Huang. All authors helped in data collection and preparation of the final article to be submitted. The article has been read and approved by all the authors, the requirements for authorship have been met, and each author believes that the article represents honest work.

Registration of research studies

Not Applicable.

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