Case Report

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Disseminated peritoneal leiomyomatosis following laparoscopic myomectomy: a case report

Jianchun Xiao*, Ruopeng Zhang[1,2,*], Yue Teng[1,2] and Beizhan Liu[1]

Abstract

Disseminated peritoneal leiomyomatosis (DPL) is a rare benign disease characterized by tumors derived from smooth muscles throughout the abdominal cavity. DPL has been reported following laparoscopic myomectomy or hysterectomy; however, few reports have described DPL after abdominal myomectomy or hysterectomy. In the present case, two intra-abdominal masses were incidentally discovered on imaging during a routine medical checkup of a 45-year-old woman who had undergone laparoscopic uterine myomectomy 10 years previously. The masses were later found to be near the ascending colon and between the liver and right kidney. A malignancy or stromal tumor was suspected, and the patient underwent exploratory surgery. Histopathologic examination revealed that the masses were leiomyomas. Considering the patient's history of gynecologic surgery, we believe that this was a rare case of DPL following laparoscopic uterine myomectomy.

Keywords

Disseminated peritoneal leiomyomatosis, uterine myomectomy, preoperative diagnosis, benign tumors, smooth muscle, laparoscopy

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Introduction

Disseminated peritoneal leiomyomatosis (DPL) is a rare benign disease characterized by tumors derived from smooth muscles occurring in the abdominal cavity. This condition is thought to be associated with an elevated estrogen level, such as that caused by pregnancy, estrogen replacement

[1]Peking Union Medical College Hospital, Beijing, China
[2]Peking Union Medical College, Beijing, China

*These authors contributed equally to this work.

Corresponding author:
Beizhan Liu, Beijing 100730, China.
Email: pumc2000@163.com

therapy, oral contraceptives, and ovarian endocrine tumors.[1] DPL frequently occurs in women of reproductive age, and its growth patterns can resemble those of malignant tumors with active growth. Thus, DPL is easily misdiagnosed prior to surgery. A definitive diagnosis depends on the operative, histopathologic, and immunohistochemical findings. We herein describe a 45-year-old woman with DPL following laparoscopic uterine myomectomy. This case report provides us with a new perspective on the identification of abdominal swelling and reminds us that it is important to pay attention to the patient's history, especially the surgical history.

Case report

A 45-year-old woman who had undergone laparoscopic uterine myomectomy 10 years previously was found to have abdominal masses on imaging during a routine medical checkup. Pathological examination at that time confirmed a diagnosis of uterine fibroids accompanied by red changes and slight bleeding. She had no history of hormonal therapy or contraception. During the medical checkup 10 years after the myomectomy, a computed tomography (CT) scan revealed a 4.3- $\times$ 5.1-cm mass in the ascending colon that was suspected to be a malignant or stromal tumor. A second 2.5- $\times$ 3.3-cm mass between the liver and right kidney was assumed to be metastatic spread (Figure 1). Both of the masses were intraperitoneal. However, enteroscopy revealed no abnormalities. The patient underwent a positron emission tomography/CT scan to rule out the possibility of a gastrointestinal stromal tumor (GIST). The maximum standardized uptake value (SUVmax) of the two masses were 2.6 and 1.6 Bq/mL, respectively. The SUVmax of GISTs usually ranges from 2.0 to 2.5 Bq/mL, and a malignant tumor is highly likely when the SUVmax is $>$2.5 Bq/mL.

Considering the rapid growth rate and uncertainty of the malignant potential of the masses, the patient underwent

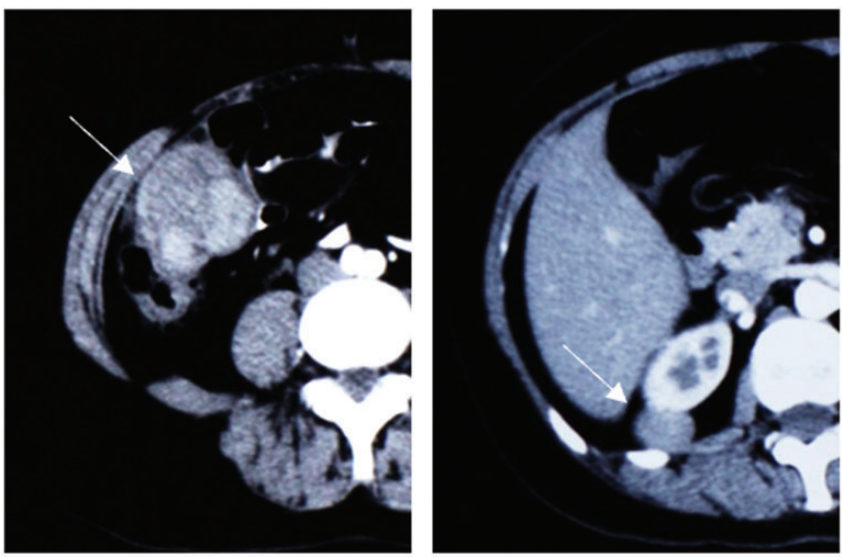

Figure 1. Computed tomography scan revealing the two masses. The left figure shows the extramural colonic mass, and the right figure shows the mass between the liver and right kidney.

laparotomy and mass resection. During the surgery, we found that the larger mass was located outside of the colon lumen, originated from the peritoneum, and adhered to the surface of the ascending colon (Figure 2); this was in contrast to the CT findings, which indicated that the tumor was located in the lumen of the ascending colon. The smaller mass was located in the retroperitoneal space. The excised specimens are shown in Figure 3. The final histopathological examination confirmed a diagnosis of leiomyoma (Figure 4) (CD34 and CD117 negativity, which are inconsistent with the characteristics of GISTs, and smooth muscle actin and desmin positivity, which are consistent with smooth muscle tumors).

Discussion

The etiology of DPL is unclear. DPL was first reported in 1952 by Willson and Peale,[2] and only about 100 cases have since been reported. Most researchers believe that DPL is related to increases in the estrogen level in the body.[3] In recent years, however, accumulating evidence suggests that iatrogenic factors are important causes of DPL. Few reports have described DPL after myomectomy or hysterectomy; however, DPL has been reported following myomectomy or hysterectomy using laparoscopic surgery.[4] Residual myoma fragments produced during surgery may implant into normal tissue and continue to grow by forming new blood vessels between normal tissues.[5,6] Patients with DPL have normal

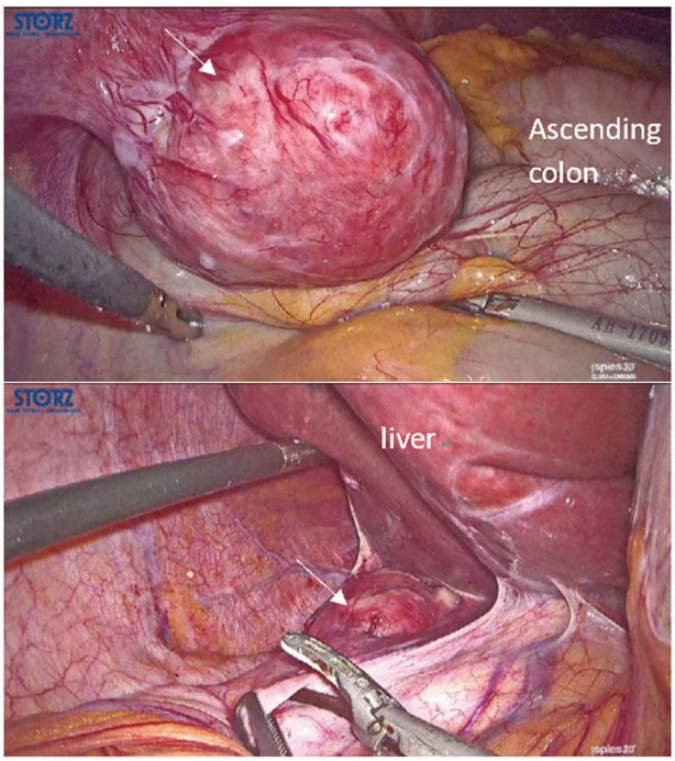

Figure 2. Intraoperative findings. The larger mass was located outside of the colon lumen, adhering to the surface of the ascending colon. The smaller mass was located in the retroperitoneal space.

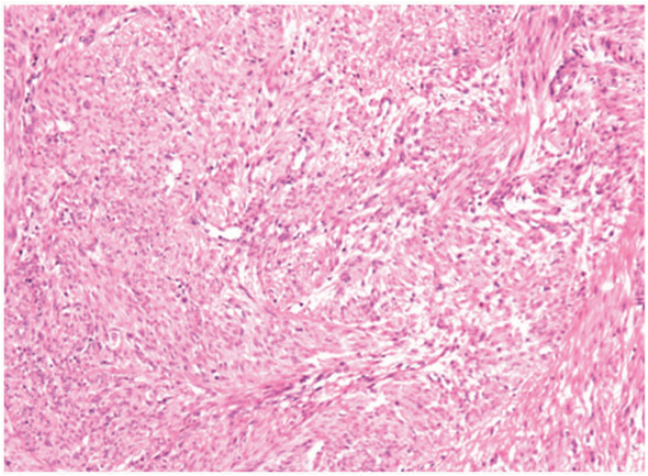

Figure 3. The histological appearance of both masses was compatible with the characteristics of a leiomyoma (hematoxylin and eosin staining). Immunohistochemistry revealed negativity for CD34, CD117, DOG-1, S-100, and succinate dehydrogenase subunit B; positivity for smooth muscle actin and desmin; and a Ki-67 index of 5%.

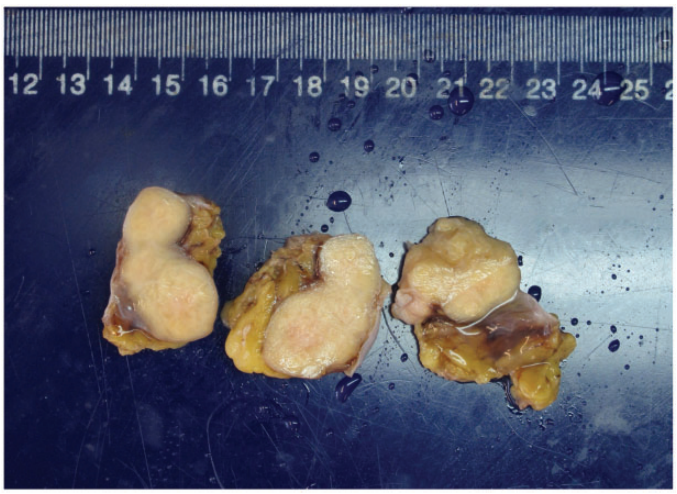

Figure 4. Excised mass specimens. The specimens were soft, yellow masses with a uniform texture.

tumor markers and no specific imaging findings, nor do they have special clinical manifestations or signs. Approximately 50% of patients are asymptomatic, while the remaining patients develop pelvic pain, pelvic cavity discomfort, pelvic and abdominal compression symptoms, and even intestinal obstruction.[7] This lack of specific characteristics results in a very high rate of preoperative misdiagnosis. The definitive diagnosis depends on intraoperative examination findings and pathological results. Several differences exist between DPL and other gastrointestinal tumors such as GISTs. The main pathological manifestation of DPL is dense arrangement of

smooth muscle cells and fibroblasts. Immunohistochemical examination often reveals smooth muscle actin and desmin positivity. GISTs generally consist of spindle cells and epithelial-like cells with CD117 CD34 positivity, and most of the spindle cells have unclear boundaries. Pathological examination can generally determine the tumor type.

In the present case, CT revealed that one of the masses was located in the ascending colon in close proximity to the adjacent intestinal wall, with its outer part close to the peritoneum. Considering its rapid growth rate, it was suspected to be a malignant tumor. We also considered that this mass could be a GIST, which is the most common mesenchymal tumor in the gastrointestinal tract and can occur in any part of the gastrointestinal tract.[8] GISTs also lack specific clinical manifestations and usually present as round masses with a smooth rim on CT images. The smaller mass between the liver and right kidney was assumed to be metastatic spread.

During the intraoperative exploration of the relationship between the larger mass and the peripheral tissue, we found that the mass had not grown on the wall of the intestine but had instead originated from the peritoneum. The intestinal wall was squeezed inward with the tumor growth, explaining why it was closely related to the intestinal wall on the CT image. Histopathological examination revealed that both of the masses were leiomyomas. Considering the patient's history of laparoscopic uterine myomectomy, we believe that these two masses were DPL possibly caused by the spread of tumor cells by uterine myomectomy. In most other cases, patients have been diagnosed with DPL no more than 10 years after uterine myomectomy. In the present case, however, the patient had undergone uterine myomectomy 10 years previously. This case suggests that even many years after surgery, regular medical examinations are required to detect the occurrence of DPL.

Conclusion

DPL is a very rare disease that can develop after myomectomy. DPL should be considered as a differential diagnosis of intra-abdominal masses in patients with a history of myomectomy. Care should be taken during laparoscopic extraction of myomas.

Abbreviations

DPL: disseminated peritoneal leiomyomatosis, GIST: gastrointestinal stromal tumor; CT: computed tomography; SUVmax: maximum standardized uptake value

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Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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Human/animal rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008(5).

Informed consent

Informed written consent was obtained from the patient for publication of this case report and accompanying images.

ORCID iD

Ruopeng Zhang https://orcid.org/0000-0003-0878-2900

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