



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Large leiomyomatosis peritonealis disseminata after laparoscopic myomectomy: A case report with literature review

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ARTICLE INFO

Article history:

Received 25 November 2020

Received in revised form

30 November 2020

Accepted 1 December 2020

Available online 3 December 2020

Keywords:

Leiomyomatosis peritonealis disseminata

Morcellation

Parasitic myoma

Case report

ABSTRACT

INTRODUCTION: Leiomyomatosis peritonealis disseminata (LPD) is a rare disease in which multiple leiomyomas are formed intraperitoneally. Several LPD cases were associated with laparoscopic myomectomy using power morcellators; however, LPD with a large tumor size remains extremely rare. We present a case of large LPD occurring after laparoscopic surgery.

PRESENTATION OF CASE: A 26-year-old woman, gravida 0, underwent laparoscopic myomectomy with power morcellation in our institution. After 5 years, follow-up examination revealed pelvic tumors. Although we recommended resection, she refused and only wanted to be followed up. After 9 years from the first surgery, the tumors became symptomatic and were increasing in number (>10 nodules) and size (>15 cm). Needle biopsy detected leiomyoma. Computed tomography angiography showed that omental and mesenteric arteries were feeding the tumors. We performed laparotomy, and all the 19 tumors emerging from the omentum and mesenterium and weighing 7647 g in total were removed without injuring other organs. The maximum diameter of the largest tumor was 34 cm. The pathological diagnosis was nonmalignant LPD with leiomyoma.

DISCUSSION: Among all reported cases, our case had the largest LPD size. The tumors reached such a huge size because of two possible reasons: (1) they gradually grew asymptotically over a long period from the time of diagnosis, and (2) they were fed by particularly large vessels, including the omental and mesenteric arteries.

CONCLUSION: A large LPD is not always symptomatic. After a laparoscopic myomectomy, especially with power morcellation, long-term follow-up is necessary to detect LPD.

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1. Introduction

Leiomyomatosis peritonealis disseminata (LPD) is a rare disease characterized by the formation of multiple intraperitoneal leiomyomas [1]. LPD was first reported by Willson and Peale in 1952 [2], and currently, approximately 200 cases have been reported [3]. However, LPD still has no standard treatment. Its possible causes include hormonal, subperitoneal mesenchymal stem cells, metaplasia, genetic, and iatrogenic [1]. In particular, iatrogenic LPD is mainly caused by intracorporeal power morcellation during previous laparoscopic surgery [4]. Furthermore, parasitic myoma (PM) is widely used to describe a leiomyoma of extrauterine nourishing. Recently, several cases of LPD or PM have been associated with

laparoscopic myomectomy using power morcellators [5]. To our best knowledge, the case reported by Kumar et al. [6] is the largest LPD associated with laparoscopic surgery, and no cases of LPD larger than 30 cm have been reported to have undergone resection. Here, we present a case of large LPD appearing after laparoscopic surgery. This study conforms to the SCARE guidelines [7].

2. Presentation of case

A 26-year-old woman, gravida 0, underwent laparoscopic myomectomy for menorrhagia and pressure symptoms. Because this operation was performed before the release of the 2014 US Food and Drug Administration (FDA) statement [8], a 13 cm intramural fibroid was removed by power morcellation without in-bag containment system. The patient had no history of drug use. Her family history was unremarkable. After 5 years, follow-up examination revealed pelvic tumors. Magnetic resonance imaging (MRI) detected PMs measuring 9 cm in maximum diameter with no indication of malignancy. Hence, surgical resection was recommended. However, with slight discomfort or pain, the patient refused and wished to be followed up instead. During the follow-up period,

Abbreviations: CT, computed tomography; FDA, Food and Drug Administration; LPD, leiomyomatosis peritonealis disseminata; MRI, magnetic resonance imaging; PM, parasitic myoma; US, ultrasound.

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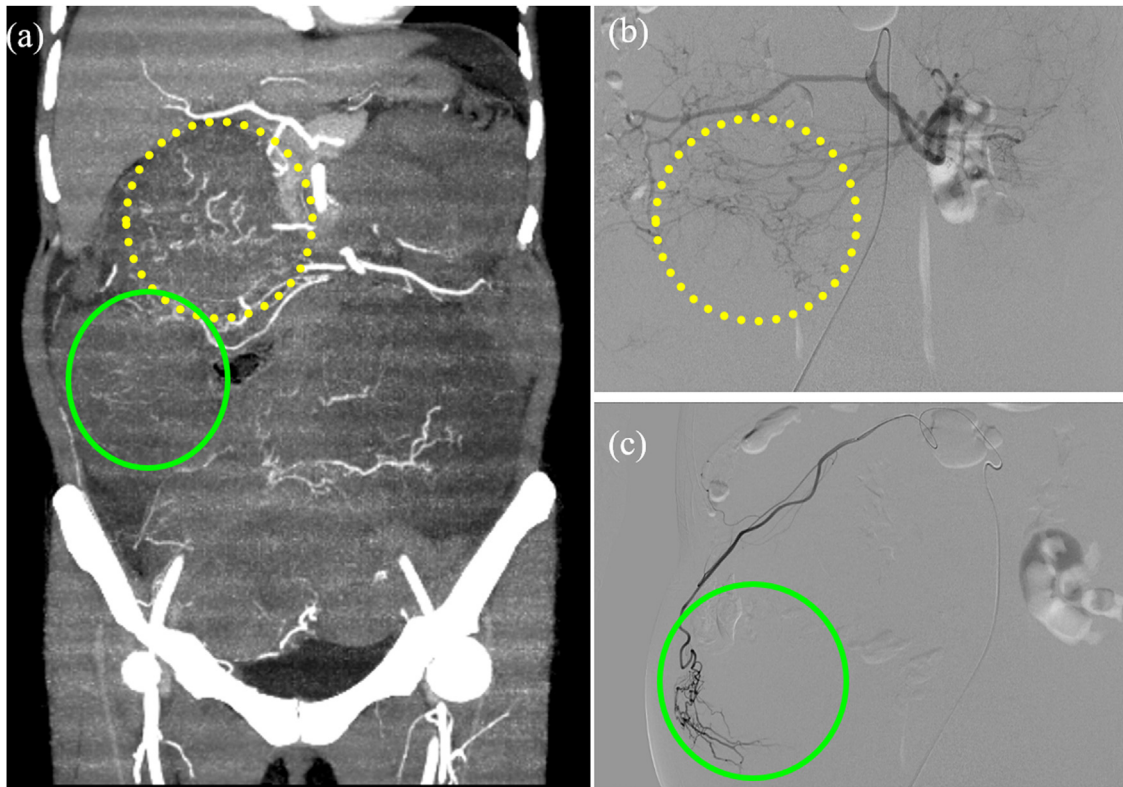


Fig. 1. Radiologic images.

(a) Coronal view from computed tomography angiography demonstrates multiple solid masses fed by a branch of gastroduodenal artery (dotted circle) and the ileal branch of the superior mesenteric artery (solid circle).

(b) Celiac angiography detected a branch of gastroduodenal artery (dotted circle).

(c) Superior mesenteric artery angiography showed the ileal branch (solid circle).

she was monitored closely by MRI and computed tomography (CT). Nine years after the first surgery, the tumors became symptomatic, with a gradual increase in number (>10 nodules) and size (>15 cm in diameter), and CT angiography detected tumors fed by omental and mesenteric arteries (Fig. 1). Considering the possibility of malignant tumors, we performed an ultrasound (US)-guided needle biopsy and revealed the presence of leiomyoma. Hence, the patient was diagnosed with LPD. Her serum cancer antigen 125 level was elevated (553 U/mL), and she manifested pelvic pressure and abdominal distension, thereby consented to undergo surgery. We performed laparotomy and found multiple tumors being fed by vessels from the mesentery or retroperitoneum. All the 19 tumors arising from the omentum and mesentery and weighing 7647 g in total (including four tumors weighing 3052 g, 2118 g, 1120 g, and 786 g, respectively) were removed without injuring other organs because of non-invasive tumor development (Fig. 2). The maximum diameter of the largest tumor was 34 cm (Fig. 3). The surgery lasted for 462 min. A total of 9036 mL of blood (mostly ascites) was lost; thus, two units of red blood cells were transfused. All the procedures were performed by well-trained gynecologists. She fully recovered with no postoperative complications. The pathological diagnosis was nonmalignant LPD with leiomyoma (Fig. 3). No malignant cells were observed in ascites cytology. She was observed without postsurgical hormonal therapy, and 34 months after the second surgery, the disease did not recur.

3. Discussion

According to a retrospective study, the incidence of LPD or PM after laparoscopic surgery using morcellation was between 0.12% and 0.95% [9]. In a systematic review, Lete et al. examined



Fig. 2. Intraoperative image.

A large tumor arising from the mesentery of the small intestine was removed by ligating the feeding vessels without intestinal resection.

274 patients with LPD/PM and observed that the mean age of these patients was 40 years; furthermore, 120 (44%) had previous myomectomy or hysterectomy, and 106 (39%) had a history of power morcellation [10]. Though rarely practiced, previous laparoscopic power morcellation has been recognized as a major cause of LPD. In 2014, FDA has issued a safety statement about the use of

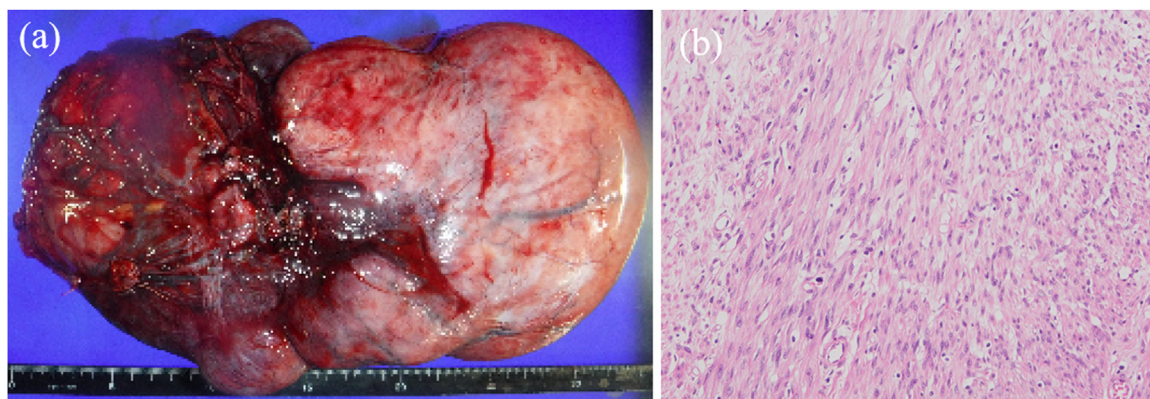


Fig. 3. Histopathological findings of resected specimens. (a) Macroscopic appearance of the largest tumor: the tumor measured 34 × 24 cm, with a smooth surface. (b) Microscopic examination showed a typical leiomyoma without malignancy (Hematoxylin–eosin staining, 200×).

Table 1
Reported cases of large leiomyomatosis peritonealis disseminata or parasitic leiomyoma (≥30 cm in maximum diameter).

Author	Year	Age at surgery for LPD	Maximum tumor diameter (cm)	History of morcellation	Time after previous surgery (months)	Treatment	Tumor location	Outcome
Kumar et al.	2008	24	30	Yes	9	laparotomy	omentum and pelvic cavity	No recurrence for 3 months
Dashraath et al.	2016	31	30	None	–	laparotomy	omentum	N/A
Present case	2020	35	34	Yes	112	laparotomy	omentum and mesenterium	No recurrence for 34 months

LPD, leiomyomatosis peritonealis disseminata; N/A, Not acceptable from the literature.

laparoscopic power morcellation for removing fibromatous uterus or uterine fibroids to avoid spreading occult uterine sarcomas [8]. The same point is valid regarding nonmalignant uterine fibroids because using laparoscopic power morcellation may contribute to LPD development. Therefore, to minimize scattering leiomyoma fragments in the peritoneal cavity, surgeons should take intraoperative precautions including in-bag containment system, minilaparotomy for specimen retrieval instead of morcellation, and irrigation after laparoscopic power morcellation [11,12]. However, the onset of iatrogenic LPD cannot be completely prevented even with in-bag morcellation because minor leakage or tissue dissemination may be inevitable [13,14].

A literature review by Darii et al. [15] showed that most reported cases of iatrogenic PM measured <10 cm. To our best knowledge, only two reported cases of extrauterine LPD tumors measured 30 cm or more in maximum diameter [6,16]. Of these two cases, only one [6] had a history of power morcellation (Table 1). Among those reported previously, our case had the largest LPD size after morcellation. The possible explanations of this enlargement were 1) our patient’s tumors were relatively asymptomatic over a long period from the time of diagnosis compared with other reported cases with large tumors that were mostly symptomatic, and 2) our patient’s tumors were fed by large vessels such as the omental and mesenteric arteries as revealed by CT angiography. A study by Dashraath et al. [16] reported that large LPD tumors are supplemented by engorged vessels from the greater omentum, thereby highlighting the possibility that large LPD may develop along large blood vessels.

Van der Meulen et al. reported that the median interval between diagnosis and surgery using morcellation was 48 months (range: 1–192) [9]. Conversely, our patient was followed up without surgery for 9 years from the first surgery because of an asymptomatic increase in size of the LPD. In Erenel et al.’s [17] study, 47% of patients with PM did not complain of abdominal pain, and 25% were asymptomatic. Symptoms are related to location as well as size. As

presented in our case, a large LPD is not always symptomatic, and symptoms may not be strong until the tumors become considerably enlarged. Therefore, although routine follow-up may not be justified considering the infrequency of LPD, a long-term follow-up can be considered for early detection of LPD after laparoscopic myomectomy with power morcellation. Visualization of the upper abdomen should be included in follow-up visits because large LPDs often occur in the omentum [6,16]. Moreover, for long-term observation, the possibility of malignancy should be considered carefully because LPD can mimic a malignant process or malignant transformation in LPD [18]. In general, the preoperative diagnosis of LPD is extremely difficult because its diagnosis is based on medical history, intraoperative observations, and pathological findings [19]. Nonetheless, as shown in the current case, preoperative pathological evaluation through US-guided biopsy can be considered, depending on the tumor location, especially for large tumors. Kumar et al. also performed an US-guided biopsy for large LPD to rule out malignancy preoperatively [6].

In patients with pelvic masses and a history of morcellation, iatrogenic LPD should be considered as a differential diagnosis, and surgical resection should be considered for symptomatic or suspected malignant LPD. Even in cases with large tumors, LPD is likely to be resected without injuring other organs, resulting in successful outcomes, as in the present case; however, the possibility of combined resection of the surrounding organs should be fully explained.

4. Conclusion

A large LPD is not always symptomatic. After laparoscopic myomectomy, especially with power morcellation, long-term follow-up should be considered to detect LPD and survey tumor growth.

Declaration of Competing Interest

The authors report no declarations of interest.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

Due to the retrospective nature of this case report, the need for the approval by institutional review board was waived in our institution. Written informed consent was obtained from the patient.

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

Yasunori Yoshino: writing the paper, manuscript preparation.

Naoyuki Yoshiki: study concept and design, performing the operation.

Reiko Nakamura: data collection, data analysis and interpretation, performing the operation.

Yuki Iwahara: manuscript preparation.

Tomonori Ishikawa: manuscript preparation.

Naoyuki Miyasaka: study concept and design.

Registration of research studies

Not applicable.

Guarantor

Yasunori Yoshino, Naoyuki Miyasaka.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgements

The authors would like to thank Enago (www.enago.jp) for the English language review.

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