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CASE REPORT

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A case of laparoscopic resection of leiomyosarcoma arising in the mesentery of descending colon: a case report and review of the literature

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Abstract

Leiomyosarcoma of mesenteric origin is rare and may be managed by laparoscopic surgery as a less invasive procedure, on the condition that the tumor can be resected with a safe margin.

KEYWORDS

laparoscopic surgery, leiomyosarcoma, mesenteric tumor, smooth muscle neoplasm

1 | BACKGROUND

Leiomyosarcoma (LMS) of mesenteric origin is extremely rare. We presented the case of a 58-year-old woman, who had 5-cm LMS of mesentery of descending colon, successfully resected by laparoscopic surgery. We showed that laparoscopic resection of localized LMS in the mesentery of the descending colon was safe and feasible.

Leiomyosarcoma (LMS) is a rare malignant tumor that originates from smooth muscle cells.¹ It was reported to occur in any smooth muscle cell-containing organ, such as the uterus: stomach: intestine: retroperitoneum: and blood vessels such as the portal vein and inferior vena cava.²⁻⁷ On the other hand, a mesenteric origin of LMS is extremely rare, with a small number of reported cases.⁸ In this case report, we presented a patient with LMS that originated from the mesentery of the descending colon, which was successfully resected by laparoscopic surgery. Here, we report our case with some literature review.

2 | CASE PRESENTATION

A 58-year-old woman presented with intermittent pain in the lower abdomen for one week. She had no other symptoms at

that time. Computed tomography (CT) was performed and showed a 5-cm mass in the left retroperitoneum and caudal to the left kidney. The patient was referred to our hospital for further treatment. She was taking fluvoxamine maleate, sulpiride, and brotizolam for depression and insomnia. On admission, her height was 151 cm and her weight was 48 kg. Her blood pressure was 147/101 mm Hg, heart rate was 63 beats/min, and body temperature was 36.5°C. The abdomen was soft and flat, with a palpable fist-sized elastic hard mass in the left lumber/middle region. Neither tenderness nor skin changes were observed. Laboratory study results showed elevated gamma-glutamyl transferase 60.4 IU/L (9-32 IU/L), and other serum chemistry results were within normal limits. Complete blood count and serum tumor markers, such as carbohydrate antigen 19-9 and carcinoembryonic antigen, were within normal limits. Esophagogastroduodenoscopy, colonoscopy, and enteroscopy were normal and did not show any epithelial or extramural lesion.

Contrast-enhanced abdominal CT revealed a $36 \times 38 \times 44$ -mm elliptical shaped, heterogeneously enhancing mass with smooth surface (Figure 1). A part of the mass was poorly enhanced, suggesting the presence of a necrotic area or cystic degenerative changes inside the lesion. The mass seemed to be located in the left retroperitoneum or in the mesentery of the small intestine, but the definite location

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arrow: tumor

FIGURE 1 Computed tomography and magnetic resonance imaging findings. A $36 \times 38 \times 44$ mm elliptical shaped heterogeneously enhanced mass with smooth surface was found. A poorly enhanced region was observed, suggesting necrotic area or cystic degenerative change (A: axial image, B: coronal image). The tumor showed the same intensity with the adjacent muscle (psoas muscle) on T1 weighted image, and higher intensity than the adjacent muscle on T2 weighted image (C: axial, D: coronal image, on T1-weighted image)



FIGURE 2 PET findings. PET showed enhanced uptake on the same lesion (A: whole-body and B: axial image)

could not be determined on preoperative imaging studies alone. The small and large bowels were located away from the mass, suggesting that these were unlikely origins of the tumor. Neither dissemination nor ascites were found.

On magnetic resonance imaging (MRI), the intensity of the mass, relative to the adjacent psoas muscle, was identical on the T1-weighted image and higher on the T2-weighted image (Figure 2). There was no area of hypointensity on out-of-phase image, indicating the absence of fat tissue in the lesion. The diffusion-weighted image demonstrated high intensity. Fluorodeoxyglucose positron emission tomography showed enhanced uptake of the lesion (maximum standardized uptake value [SUVmax] 6.03) (Figure 2). All imaging modalities revealed no metastatic lesion. The differential diagnoses included desmoid tumor, leiomyoma/ LMS, gastrointestinal stromal tumor (GIST), and malignant lymphoma.

We chose a laparoscopic approach, because there seemed to be no findings of invasion to the surrounding organs, and we thought that resection with a safe margin could be achieved. Intraoperatively, the tumor was easily dissected by mobilizing the mesentery from the medial to the lateral side. The left ureter was preserved without injury, but the inferior mesenteric vein, left colonic artery, and left ovarian vessels were clipped and cut because of their involvement with the tumor. The descending colon was preserved, because the marginal vessels were >2 cm lateral to the tumor edge. The tumor was removed by cutting the mesentery with laparoscopic coagulating shears from the proximal area, then distally and laterally (Figure 3). The tumor was placed in an extraction bag and removed from the umbilical wound.

The resected specimen was a tumor that measured $7 \times 4.5 \times 3$ cm and was located in the mesentery of the descending colon. The cut surface appeared whitish with necrotic area (Figure 4). Histopathological findings revealed proliferation of spindle cells with fascicle-like formation. Immunohistochemistry was positive for desmin, α -smooth muscle actin (SMA), and h-caldesmon; these indicated a smooth muscle tumor. The negative immunohistochemistry for c-kit and CD34 excluded GIST (Figure 4). There were cellular pleomorphism and atypia: 12 mitotic figures per 10 high-power fields: 20% Ki-67: and coagulation necrosis (Figure 4). Metastasis from uterine cancer was unlikely, based on estrogen receptor (ER) negativity.

Considering these findings altogether, the diagnosis of LMS (histological grade 2, and stagellA by the French Federation Nationale des Centers de Lutte Contre le Cancer (FNCLCC) system and TNM classification, respectively) was made.^{9,10}

The postoperative course was uneventful and she was discharged on the 14th postoperative day. To date, the patient remained free from recurrence at two years and three months after surgery without any additional treatment.

3 | **DISCUSSION**

LMS is a frequent type of soft tissue sarcoma, with an incidence of 10%–20% of all newly diagnosed soft tissue sarcomas.¹ LMS of mesenteric origin was believed to originate from mesenteric vessels and had been extremely rare, with limited number of case reports, to date. Its prognosis is very poor with a median survival of 12–14 months.¹¹ LMS can occur in any site of the body where there is smooth muscle tissue, but LMS from a different anatomic origin has been reported to differ in clinical course and prognosis.¹² FIGURE 3 Intraoperative findings and procedure. The tumor was observed in the mesentery of descending colon (A). The tumor was easily dissected by mobilizing the mesentery from medial to lateral approach (B). Left ureter was preserved without injury, however, inferior mesenteric vein, left colonic artery, and left ovarian vessels were cut for their involvement with the tumor. The tumor was removed by cutting mesentery with laparoscopic coagulating shears from proximally and then distally to laterally (C, D)



*: tumor arrow head: dissecting layer (medial to lateral approach) arrow: left ureter (preserved)



FIGURE 4 Histopathological and immunohistochemical findings. The resected specimen is a well-marginated lesion measuring $7 \times 4.5 \times 3$ cm in the mesentery of descending colon with necrotic area (A). The tumor consisted of spindle cells in fascicle-like formation. Coagulative necrosis was also observed (B: H.E. ×20, C: ×100). Immunohistochemical staining was positive for desmin, α -SMA, h-caldesmon, and negative for CD34 and c-kit. Ki-67 was 20% (D-I: ×100)

We searched the PubMed database for reports of cases undergoing laparoscopic resection of LMS arising in the mesentery using with the keywords "leiomyosarcoma," "mesentery," "resection," and "laparoscopy." None of the cases of laparoscopic resection for LMS of mesenteric origin were found.

In the process of human development, the mesentery of intestinal loop rotates and twists around the superior mesenteric artery, whereas that of the descending colon presses against and fuses with the peritoneum of the posterior abdominal wall.¹³ Therefore, determining a mesenteric or retroperitoneal tumor origin is developmentally and pathologically difficult. As shown in our case, surgical dissection of the

fused fascia enabled easy ventral dissection and clarified a mesenteric origin of the tumor. On the other hand, a tumor of retroperitoneal origin would have stayed on the dorsal side of the dissected layer.

A mesenteric tumor may present with palpable abdominal mass, distention, or discomfort, but it rarely causes symptoms, because of its mobility in the peritoneal space or fixation in the retroperitoneum.¹ Therefore, a mesenteric tumor tends to be large when it is found.¹⁴ In this case, the laboratory studies were not diagnostic for LMS, but the imaging study was the key that made us suspect LMS.¹⁵ CT usually shows an enhanced, predominantly solid mass with poorly enhanced irregular cystic regions that represent necrotic

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areas. Moreover, the presence of metastatic lesions in the liver and lungs should be checked. In this case, although the MRI demonstrated the solid components and necrotic areas in the tumor, the imaging modalities were unable to differentiate LMS from other types of tumors that can originate from the gastrointestinal tract, intrapelvic organs, and peritoneum. Therefore, the definitive diagnosis was made by histopathological examination of the surgically resected specimen.

The typical histopathological findings of LMS include cellular pleomorphism or atypia, coagulation necrosis, increased mitotic figures, and sharply marginated fascicles of spindle cells that intersect. Immunohistochemistry was more diagnostic, particularly the positivity for smooth muscle markers, such as α -SMA, desmin, and h-caldesmon. These markers were reported to be positive in more than 70% of cases. Additionally, focal positivity for keratin, epithelial membrane antigen, CD34, and S100 protein may be occasionally seen.¹⁶ CD117 (KIT), which is a diagnostic marker for GIST, should be confirmed to be negative.¹⁷

The only curative treatment for LMS is surgical removal. However, advancement of LMS to a metastatic disease would narrow down the options to chemotherapy and radiation therapy, but no established treatment strategy has been proven.¹⁸ During surgery for localized LMS, complete resection is required, and the resection margin needs to be secured to prevent local recurrence.¹⁹ The other prognostic factors include tumor depth, histological grade, and the presence of metastasis at presentation. Administration of adjuvant chemotherapy has not been shown to be a predictor of local recurrence.²⁰

The treatment strategy for mesenteric LMS is yet to be established. However, early detection and less invasive surgery, mainly laparoscopic surgery, may contribute to the short- and long-term clinical outcomes. Although LMS of mesenteric origin is rare, the treatment strategy needs to be developed by accumulation of cases and evidence.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

RK and YM are the primary investigator and contributed to conceptualization; data collection. RK: drafted the manuscript. All authors have read and approved this manuscript for publication.

CONSENT FOR PUBLICATION

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

DATA AVAILABILITY STATEMENT

The data supporting the conclusion are included in this article.

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