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Giant leiomyosarcoma of the rectum with lymph node metastasis: A case report and review of the literature

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ABSTRACT

INTRODUCTION: Leiomyosarcoma of the gastrointestinal tract is very rare, with a reported frequency of less than 0.1% of all malignancies of the colorectum. It is important to diagnose leiomyosarcoma definitively by immunohistochemical profiling of smooth muscle actin, desmin, and CD34. True leiomyosarcoma of the colorectum diagnosed by immunohistochemical profiling is extremely rare that only 13 reports have been published in reviews of resected gastrointestinal mesenchymal tumors after 1998. In addition, lymph node involvement is rare in patients with leiomyosarcoma. Herein we report an aggressive case of LMS in a rectosigmoid lesion with lymph node metastasis.

CASE PRESENTATION: A 76-year-old woman visited our hospital complaining of intermittent anal bleeding that had lasted 5 months. Image studies aiming at examining the cause of her anal bleeding revealed a tumor located between the right ovary, uterus, and the rectosigmoid. Histopathology of biopsied materials from the colonoscopy suggested a malignant tumor of mesenchymal origin. Surgical resection was performed with curative intent. The tumor was diagnosed as leiomyosarcoma by pathological examination. Moreover, one of the 31 regional lymph nodes retrieved was metastasized by leiomyosarcoma. Eight months later, follow-up CT scans revealed multiple recurrent lesions in the liver and peritoneum. Despite systematic chemotherapy, she deceased 12 months after the surgery.

CONCLUSION: It is crucial to diagnose leiomyosarcoma precisely based on immunohistochemistry, and thereby distinguish it from GIST. Although lymph node metastasis is rare, lymphadenectomy appears to be important for high-risk LMSs to perform R0 resection. Further investigation on leiomyosarcoma cases so far is required to establish standard treatment strategies.

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1. Introduction

Leiomyosarcoma (LMS) of the rectum was first described by Scott in 1923 [1]. In the former classification of smooth muscle tumor in the gastrointestinal tract, LMS was not distinguished from gastrointestinal stromal tumor (GIST). Since the discovery of the oncogenic role of KIT in 1998 [2], GISTs have been differentiated from other mesenchymal tumors such as leiomyomas, leiomyoblastomas, and LMSs [3]. Although 5% of tumors with

the clinicopathologic features of GIST lack KIT expression, it is important to diagnose LMS definitively by immunohistochemical profiling of smooth muscle actin (SMA), desmin, and CD34 [4].

True LMS of the colorectum diagnosed by immunohistochemical profiling is extremely rare, with a reported frequency of less than 0.1% of all malignancies of the colorectum [5]; only 13 reports with immunohistochemical definition which was performed properly have been published in reviews of resected gastrointestinal mesenchymal tumors after 1998 [6–10]. Furthermore, unlike adenocarcinomas, nodal involvement in LMS is unusual [11]. Herein we report an aggressive case of LMS in a rectosigmoid lesion with lymph node metastasis.

Abbreviation: LMS, Leiomyosarcoma; GIST, gastrointestinal stromal tumor; SMA, smooth muscle actin; HPF, high-power field.

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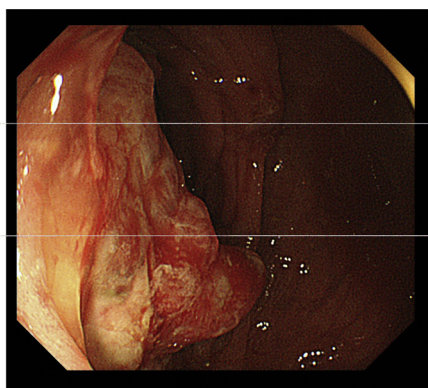


Fig. 1. The colonoscopy showed an elevated tumor with central ulceration in the rectosigmoid.

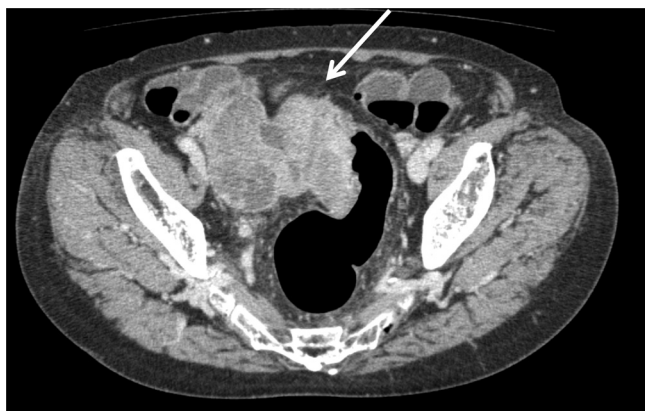


Fig. 2. An axial view of the abdominal CT scan. A giant tumor was found in the space surrounded by the rectosigmoid segment, uterus, and right ovary.

2. Case presentation

A 76-year-old woman visited our hospital complaining of intermittent anal bleeding that had lasted 5 months. An elevated tumor with central ulceration was found 10 cm from the anal verge by colonoscopy (Fig. 1), and biopsy specimens revealed a spindle cell tumor. The MIB-1 labeling index was high (80%), suggesting a high-grade sarcoma. Abdominal CT scan revealed a 10 cm mass in the rectosigmoid that invaded the uterus (Fig. 2). No distant metastases were observed. Laboratory tests revealed low-grade anemia (serum hemoglobin 12.8 g/dl), but no other abnormalities were noted; serum carcinoembryonic antigen and carbohydrate antigen 19-9 levels were also normal (Table 1).

Surgical resection with curative intent required rectal anterior resection with appendectomy, total hysterectomy, and bilateral salpingo-oophorectomy due to extensive inflammation involving the appendix, uterus, right ovary, and right fallopian tube. The excised mass was 110 × 80 × 60 mm in size. Routine hematoxylin and eosin staining showed proliferation of spindle-shaped tumor cells with enlarged nuclei and eosinophilic cytoplasm (Fig. 3). Immunohistochemically, the tumor was positive for desmin and SMA, whereas c-kit, DOG1, CD34, and S-100 protein were not expressed (Fig. 4). The tumor was of high cellularity, with up to 10 mitoses per high-power field (HPF) and variable areas of necrosis. Direct invasion was histologically proven only in the right ovary, but not in the uterus and right fallopian tube. All surgical margins were negative, and one of the 31 lymph nodes retrieved was metastasized by LMS.

Her postoperative course was uneventful. However, 8 months after the surgery, follow-up CT scanning revealed peritoneal

Table 1
Result of Laboratory Tests.

Variable	Unit		Range
WBC	×1000/μl	10.1	3.5–9.80
Hb	g/dl	12.7	11.0–15.0
Hct		40.4%	38–42%
Plt	×10,000/μl	47.8	15.0–45.0
TP	g/dL	6.1	6.5–8.0
Alb	g/dL	3.3	3.7–5.2
ChE	U/L	279	180–420
AST(GOT)	U/L	20	10–40
ALT(GPT)	U/L	17	5–40
γ-GTP	U/L	58	5–24
T-Bil	mg/dL	0.6	0.2–1.0
D-Bil	mg/dL	0.1	0.1–0.4
Ca	mg/dL	8.7	4.5–5.1
IP	mg/dL	3.0	2.3–4.0
BUN	mg/dL	12.9	8–20
Cre	mg/dL	0.59	0.5–0.9
Na	mEq/L	139	135–146
K	mEq/L	4.3	3.4–4.8
Cl	mEq/L	106	98–108
Amy	U/L	47	50–160
CK	U/L	116	42–138
CRP	mg/dL	0.83	<0.30
CEA	ng/ml	2.5	<5.0
CA19-9	U/ml	19	<7.0
PT		100%	81.0–131.6%
PT-INR		0.93	1.0
APTT	sec	27.6	30–40

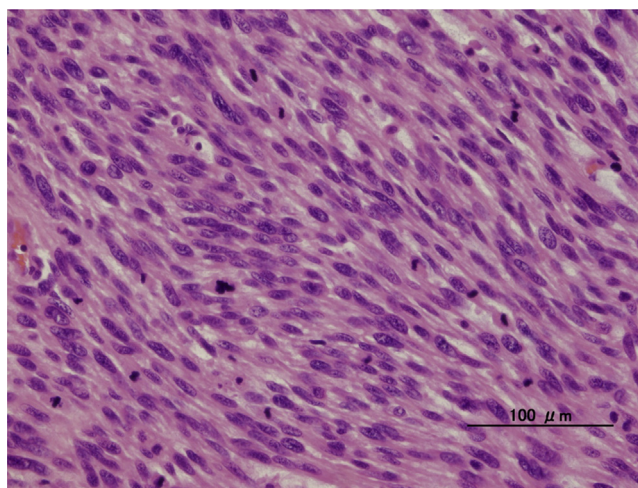


Fig. 3. Hematoxylin-eosin staining of the tumor revealed spindle-shaped cells (original magnification: 40×).

and liver metastases. Palliative chemotherapy using gemcitabine (800 mg/m²) could not control disease progression, and she died 12 months after the surgery.

3. Discussion

Since radiological differentiation between LMS and other mesenchymal tumors is difficult, the final diagnosis needs to be confirmed by postoperative pathological examination. The most definitive feature of LMS is immunohistochemical positivity for desmin and SMA, and negativity for DOG 1, CD34, S-100 protein, and c-kit. True LMS resected completely and with tumor free margins is extremely rare, with only 13 reported cases [6–10]. The clinicopathological profiles of these cases and our patient are reviewed in Table 2. The tumors ranged 20–106 mm in size, with 5–100 mitoses per 50 HPFs in the previous cases. To the best of

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