Case Report

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Leiomyosarcoma of the Sigmoid Colon Causing Sigmoido-Rectal Intussusception: A Case Report

구불결장-직장 장중첩증을 유발한 구불결장의 평활근육종: 증례 보고

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The most common malignant tumors in the colon are adenocarcinomas, while leiomyosarcoma (LMS) are rare. Here, we report a case of LMS of the sigmoid colon in a 73-year-old man who presented with sigmoido-rectal intussusception, which was discovered by abdominal computed tomography. As LMS of the colon is uncommon and is rarely associated with intussusception, we have described the imaging features in this case report.

Index terms Leiomyosarcoma; Colonic Neoplasm; Intussusception; Multidetector Computed Tomography

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INTRODUCTION

Leiomyosarcoma (LMS) is a malignant tumor originating from smooth muscle cells and occurs mainly in the uterus, stomach, and retroperitoneal space. Of these, LMSs occurring in the large intestine are rare and account for about 0.1% of the malignant tumors that occur in the large intestine; most commonly in the rectum (1). The most common symptom of LMS is abdominal pain. It may be difficult to detect until symptoms develop due to disease progression, such as palpable mass, bleeding, perforation, or intestinal obstruction (1, 2). To date, rare cases of colonic LMS causing intussusception have been reported (2-5). Here, we present a case of LMS of the sigmoid colon causing sigmoido-rectal intussusception, with emphasis on imaging features.

CASE REPORT

A 73-year-old man visited the emergency room due to hematochezia occurring on the same day. There was no specific past and family medical history, and nausea, vomiting, and abdominal distention did not appear to be present. Over the last 10 days, the patient complained of a change in bowel habits. Vital signs were within the normal range, with a blood pressure of 183/67 mm Hg, heart rate of 74 beats per minute, respiratory rate of 13 breaths per minute, and body temperature of 36.3℃. On physical examination, the mass was not palpable and anemic conjunctiva was observed. Laboratory tests showed leukocytosis (11930/mm^3), decreased hemoglobin (6.6 g/dL), increased platelets (407000/mm^3), and increased C-reactive protein (9.69 mg/dL). There were no specific findings in the plain chest and abdominal radiography.

The colonoscopy showed a large lobulated intraluminal mass filling the entire lumen of the sigmoid colon and a thick stalk behind the mass (Fig. 1A). A biopsy was performed and revealed granulation of tissue.

The contrast-enhanced abdominal computed tomography (CT) showed colo-rectal intussusception with an approximately 5 cm sized sigmoid colonic tumor. The tumor showed relatively heterogeneous enhancement in the portal venous phase. A necrotic portion that appears to be an internal low attenuation in the tumor was not clear. It had a lobulated contour and caused sigmoido-rectal intussusception about 6 cm long. Also, there was a stalk-like structure on the backside of the tumor, and it seemed to be connected to the posterior wall of the sigmoid colon (Fig. 1B). No visible mechanical bowel obstruction or ischemia was observed. The authors initially considered colorectal adenocarcinoma, which appears as a large pedunculated colonic polyp that triggers intussusception. There was no evidence of distant metastases or lymphadenopathy.

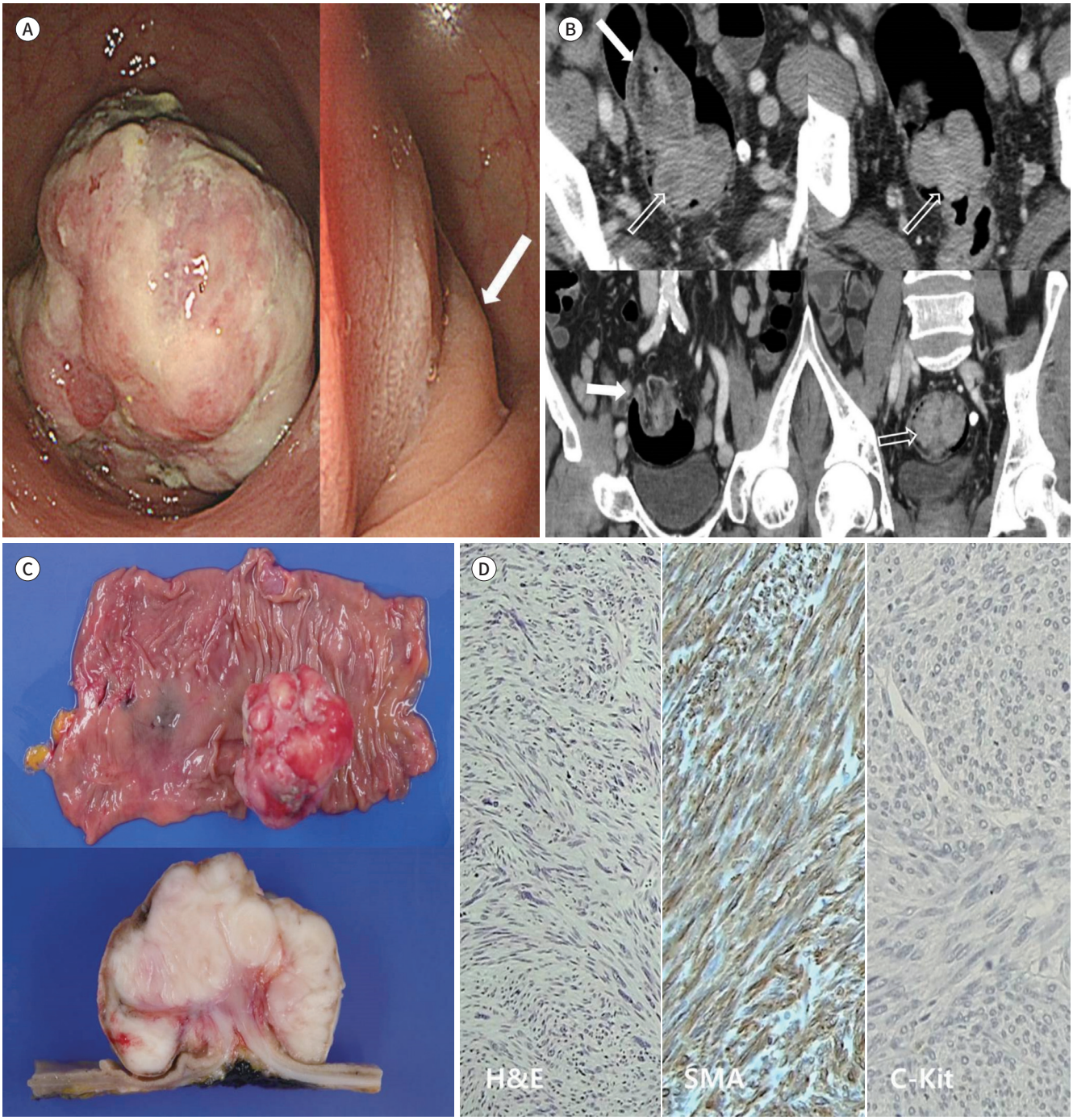
The patient underwent laparoscopic anterior resection. A polypoid tumor located about 30 cm from the anal verge was identified and resected. Macroscopic gross imaging revealed the intraluminally protruding mass of about 5.5 cm × 5.0 cm × 5.0 cm covered with ulcerated mucosa in the lumen (Fig. 1C). Under microscopic examination, the submucosal tumors consisted of interlacing bundles of spindle cells, each of which had cigar-shaped pleomorphic nuclei. Immunohistochemistry testing showed strong positive staining for smooth muscle actin (SMA), and negative staining for c-kit (CD117) and S-100, which was consistent with LMS (Fig. 1D). No regional lymph node metastasis was observed.

No definite local recurrence or metastatic disease was observed on CT after six months, and no gross change of nonspecific small mesenteric and retroperitoneal lymph nodes were noted.

DISCUSSION

LMS, which occurs in the gastrointestinal tract, is a malignant tumor originating mainly from smooth muscle cells, and usually occurs in patients between 50 and 60 years of age. The most common symptom is abdominal pain but it is often not detected until the disease develops and is diagnosed after surgery for the treatment of bleeding, perforation, or intestinal

Fig. 1. A 73-year-old man with leiomyosarcoma of the sigmoid colon causing sigmoido-rectal intussusception.
A. Colonoscopy images show large lobulated intraluminal mass filling the lumen of the sigmoid colon and a thick stalk (arrow) behind the mass.
B. Axial and coronal contrast-enhanced abdominal computed tomography images show the sigmoido-rectal intussusception (white arrows), accompanied by a pedunculated type mass (empty arrows) approximately 5 cm × 4.4 cm × 3.5 cm in the lumen of the distal sigmoid colon.
C. Gross findings show a polypoid tumor covered with ulcerated mucosa. The cut surface has a whitish and vaguely trabeculated appearance.
D. The tumor cells have cigar-shape pleomorphic nuclei (H&E stain, × 100). The tumor cells are strongly positive for SMA but negative for c-kit (× 100).
H&E = hematoxylin and eosin, SMA = smooth muscle actin



obstruction (2). The most common site of incidence is the stomach (50%), followed by the small intestine (30%), colon/rectum (15%), and esophagus (5%) (6). In this case, our patient complained of hematochezia and a change in bowel habits, without abdominal pain or abdominal distention, and the LMS was discovered in the sigmoid colon.

Histologically, LMS is very similar to gastrointestinal stromal tumor (GIST) but is negative for CD117 (c-kit) and CD34, and positive for SMA and desmin by immunohistochemistry. It can be distinguished from GIST, which is positive for CD34 and CD117 (6). Complete surgical resection is the most common treatment and is more effective than conventional radiation therapy and chemotherapy, which may leave behind surviving tumor cells. The prognosis is worse than that of other smooth muscle tumors such as GIST because of the high probability of local recurrence. The known prognostic factors include the grade of the mitotic index and the size of the tumor. Tumor location is not strongly associated (7).

CT and magnetic resonance image (MRI) findings are nonspecific in retroperitoneal and peripheral soft tissue LMS. In the case of a large mass, central low attenuation can be observed on a CT scan. These findings are known to be due to bleeding, necrosis, or cystic changes. To date, no systematic reports of the radiological findings of gastrointestinal LMS have been published. According to one case report that reported 10 cases of colonic LMS, the masses can be observed with an internal heterogeneous attenuation and enhancement that shows varying degrees of necrosis and lobulated margins. 3 out of 10 tumors showed exocolic growth in the large bowel, and the remaining 7 cases showed combined growth (endocolic and exocolic growth) (8). In the other study, intraluminally protruding LMS was observed in the sigmoid colon and inducing adult intussusception (2). In our case, the mass showed endocolic growth in the lumen of the colon with a pedunculated appearance. It was heterogeneously enhanced and observed as lobulated margins, but internal necrosis was unclear, which was different from the previous study. This may be due to the small size of the mass, which caused intussusception before necrotic changes occurred. If the mass shows exocolic or complex growth and there is internal necrosis, LMS may be considered, but as in our case, if the mass shows endocolic growth and the necrotic part is not clear, it is difficult to distinguish it from colonic adenocarcinoma before surgery. On MRI, the masses may show various signal intensities on T1-weighted images and T2-weighted images with heterogeneous enhancement when contrast medium is injected (1, 8).

Intussusception in adults is not common and causes about 1–5% of mechanical obstructions. The ileo-cecal (24%) type is the most common type of intussusception, followed by colocolic (19%), ileo-colic (16%), ileo-ileal (16%), jejuno-jejunal (11%), sigmoido-rectal (9%), and appendicocecal-colic (4%) (9). Benign, malignant, or idiopathic disease can cause intussusception. Common causes of colonic intussusception in the adult are benign lipoma, adenomatous polyp, malignant adenocarcinoma, and lymphoma. CT imaging is a useful tool because it can not only discover intussusception but also the cause of the disease.

To date, rare cases of colonic LMS causing intussusception with radiologic findings have been reported. In one case report (2), the LMS was about 3 cm and was in the sigmoid colon in the form of an intraluminally protruding mass, which is similar to our case. On CT, the mass showed homogeneous enhancement, no internal necrosis, and induced sigmoid colon intussusception. In the other case report (3), CT showed the LMS in the sigmoid colon and

nearly total colonic occlusion. Abdominal ultrasonography showed a thickened sigmoid wall with intussusception of the sigmoid colon.

When the causative disease acts as a lead point and triggers intussusception, mechanical obstruction is usually caused (10). However, in our patients, the LMS acted as a lead point, causing sigmoido-rectal intussusception, but mechanical obstruction or proximal bowel dilatation was not apparent.

In conclusion, little is known about the imaging features of LMS in the colon; however, further information on LMS and other rare tumors are warranted to allow for proper diagnosis and treatment by radiologists and clinicians.

Author Contributions

Conceptualization, J.Y.Y., K.Y.; data curation, all authors; investigation, K.Y.; formal analysis, all authors; project administration, J.Y.Y., K.Y.; resources, J.Y.Y., K.Y.; supervision, J.Y.Y., K.Y.; visualization, all authors; writing—original draft, J.Y.Y., K.Y.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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구불결장-직장 장중첩증을 유발한 구불결장의 평활근육종: 증례 보고

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결장에 영향을 미치는 악성 종양은 주로 선암종이며 결장의 평활근육종은 드물다. 저자들은 73세 남자 환자에서 복부 컴퓨터단층촬영상 구불결장-직장 장중첩증과 함께 발견된 구불결장의 평활근육종의 증례를 보고하고자 한다. 결장의 평활근육종은 드물고 장중첩증과 관련되어 보고된 바는 더욱 드물기에, 저자들은 증례 보고와 함께 영상의학적 특징을 기술하였다.

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