Case Report

A case of gastrointestinal stromal tumor of the rectum resected by the transvaginal approach

Yoshiko Kusumoto, Kuniyasu Soda, Yukinobu Shuto, Fumio Konishi

Abstract

A71-year-old woman was admitted to our hospital with a chief complaint of bloody stool. Digital examination revealed a hard mass with a smooth surface protruding from the anterior wall of the rectum. Colonoscopy demonstrated a submucosal tumor with a mucosal ulceration located about 3cm from the anal verge. Biopsy specimens were examined histopathologically and immunohistochemically. The tumor was diagnosed as gastrointestinal stromal tumor (GIST). The patient underwent tumor resection by the transvaginal approach, and an ileostomy on the right side of the abdomen was constructed. The tumor, which measured 55mm×45mm×35mm, was solid and white with a slightly grayish color and soft consistency. Microscopic examination revealed fascicular proliferation of spindle-shaped tumor cells. In immunohistochemical studies, tumor cells were diffusely positive for CD 34 and c-kit protein, and were negative for S-100 protein, α-smooth muscle actin and desmin. The tumor was diagnosed as GIST of the rectum. To our knowledge, 41 cases of rectal GIST have been reported in Japan, but only in one of these was resected by the transvaginal approach. We describe a case of rectal GIST resected by transvaginal approach, with reference to GIST in the literature.

(Key words : gastrointestinal stromal tumor, rectum, transvaginal approach)

Introduction

Gastrointestinal stromal tumors (GIST) are neoplasms that arise in the non-epithelial component of the gastrointestinal tract. Recently, a more widespread understanding of the histological origin of the tumor has increased the number of reported cases. Although, there are many reports of cases of gastric or small intestinal GIST, only 40 cases of rectal GIST could be found in the literature in Japan, and in only one of these was rectal GIST resected by the transvaginal approach. We describe a case of rectal GIST resected by transvaginal approach, with reference to GIST in the literature.

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Case report

A71-year-old woman was admitted to our hospital with a chief complaint of bloody stool. Digital examination revealed a hard mass with a smooth surface protruding from the anterior wall of the rectum. Laboratory tests, including serum levels of carcinoembryonic antigen and CA 19-9, were all within normal limits. Barium enema study showed an elevated lesion, 4cm in size, in the rectum (Fig. 1). Colonoscopy demonstrated a submucosal tumor with a mucosal ulceration located about 3cm from the anal verge (Fig. 2). Endoscopic ultrasonography revealed a homogeneous hypoechoic mass (Fig. 3). Contrast-enhanced CT scan showed a high density mass with 4.0cm in diameter on the anterior rectal wall (Fig. 4). Histopathological examination of the biopsy specimens showed spindle shaped tumor cells and immunohistochemical testing was negative for S-100 protein, α-smooth muscle actin and desmin, and positive for CD 34 and c-kit gene product. The tumor was diagnosed as gastrointestinal stromal tumor (GIST) of the rectum. The patient underwent tumor resection by the transvaginal approach. A midline vertical incision was initiated at the center of the the posterior wall of the vagina, and a transverse incision was made at the mucocutaneous border. The posterior vaginal wall was separated from the rectovaginal septum, until a sufficiently wide area was exposed for tumor resection. A part of external anal sphincter muscle was divided at the anterior midline of the anus, because the tumor was extending under the external anal sphincter. The proximal edge of the tumor could be palpated through the anus, and traction sutures were placed around the tumor on the rectal wall. The full thickness of the rectum wall was cut by the electrocautery, with a tumor free margin of 1cm (Fig. 5-A). After resection of the tumor, the anterior rectal wall was closed by interrupted sutures of 4-0 PDS-II on an atraumatic needle. Subsequently, the posterior vaginal wall and the external anal sphincter muscle were

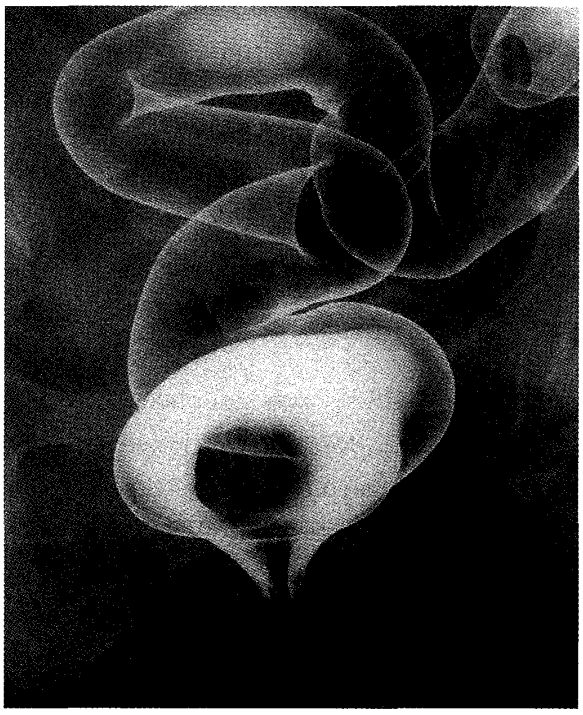

Fig. 1 Barium enema study showed an elevated lesion, 4 cm in size, in the rectum.

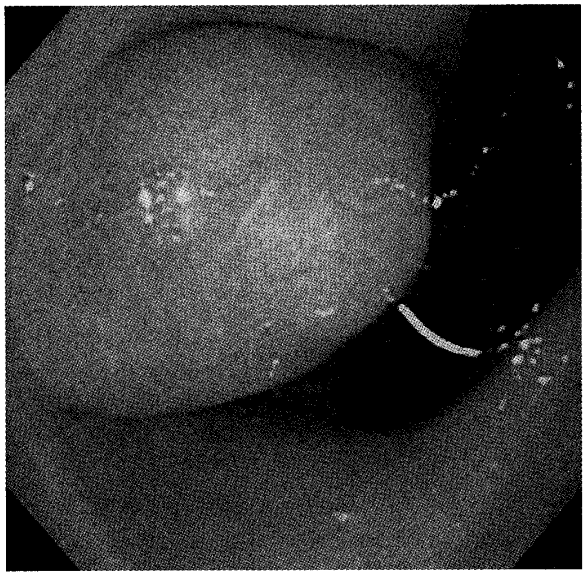

Fig. 2 Colonoscopy demonstrated a submucosal tumor with a mucosal ulceration located about 3 cm from the anal verge.

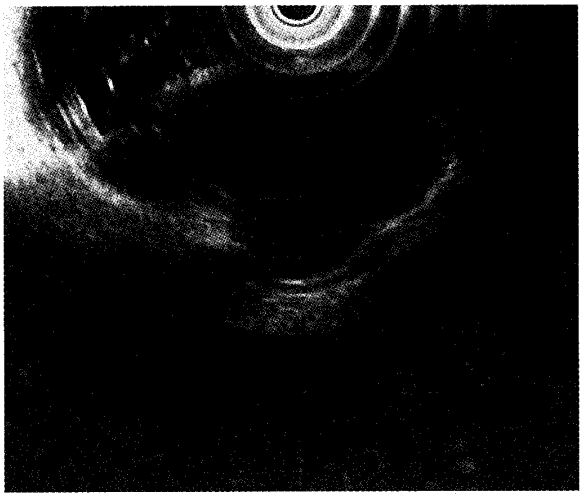

Fig. 3 Endoscopic ultrasonography revealed a homogeneous hypoechoic mass.

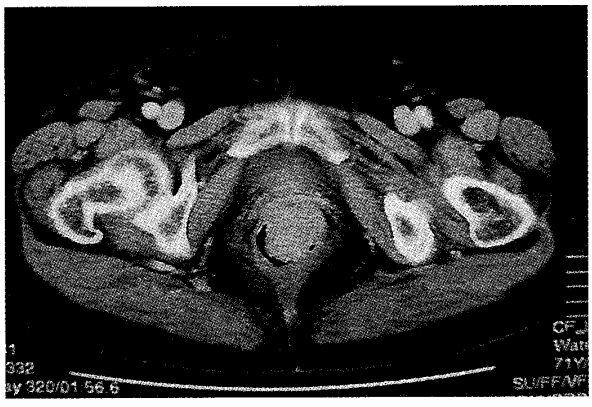

Fig. 4 Contrasted-enhanced CT scan showed a high density mass on the anterior rectal wall.

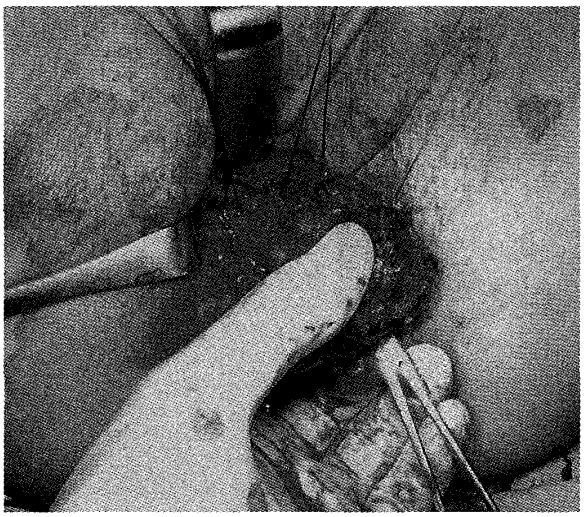

Fig. 5 - A The full thickness of the rectal wall was cut by the electrocautery, with a tumor free margin of 1 cm.

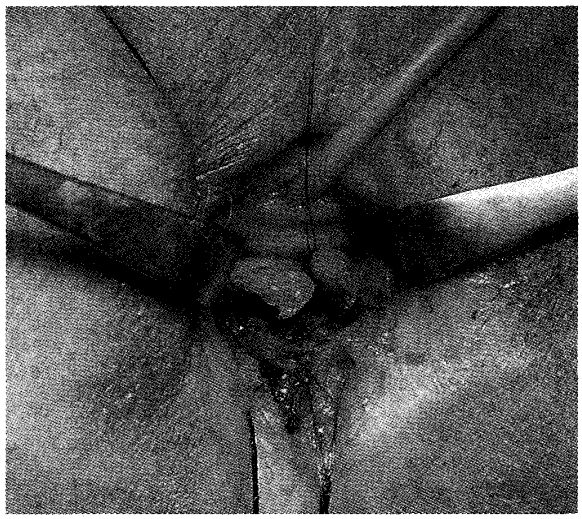

Fig. 5 - B After resection of the tumor, the anterior rectal, posterior vaginal wall and wxternal anal spincter muscle were closed.

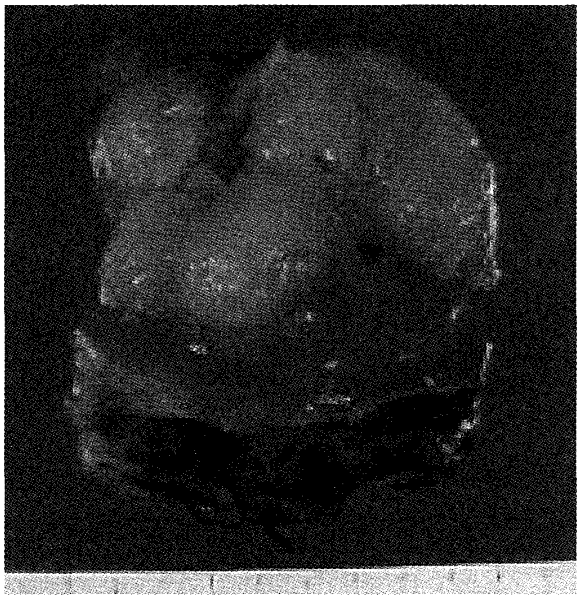

Fig. 6 Histological examination revealed fascicular proliferation of the spindle-shaped tumor cells. In immunohistochemical studies, the tumor cells were diffusely positive for CD 34 and c-kit protein, and were negative for S-100 protein, α-smooth muscle actin and desmin.

closed similarly (Fig. 5-B). Lastly, an ileostomy was constructed on the right side of the abdomen.

The tumor, which measured 55mm×45mm×35mm, involved the external anal sphincter muscle. The mass was solid and white with a slightly grayish color and soft consistency (Fig. 6). Histologic examination revealed fascicular proliferation of spindle-shaped tumor cells. In immunohistochemical studies, the tumor cells were diffusely positive for CD 34 and c-kit protein, and were negative for S-100 protein, α-smooth muscle actin and desmin (Fig. 7). From the above histological and immunohistochemical features, GIST of the rectum was diagnosed.

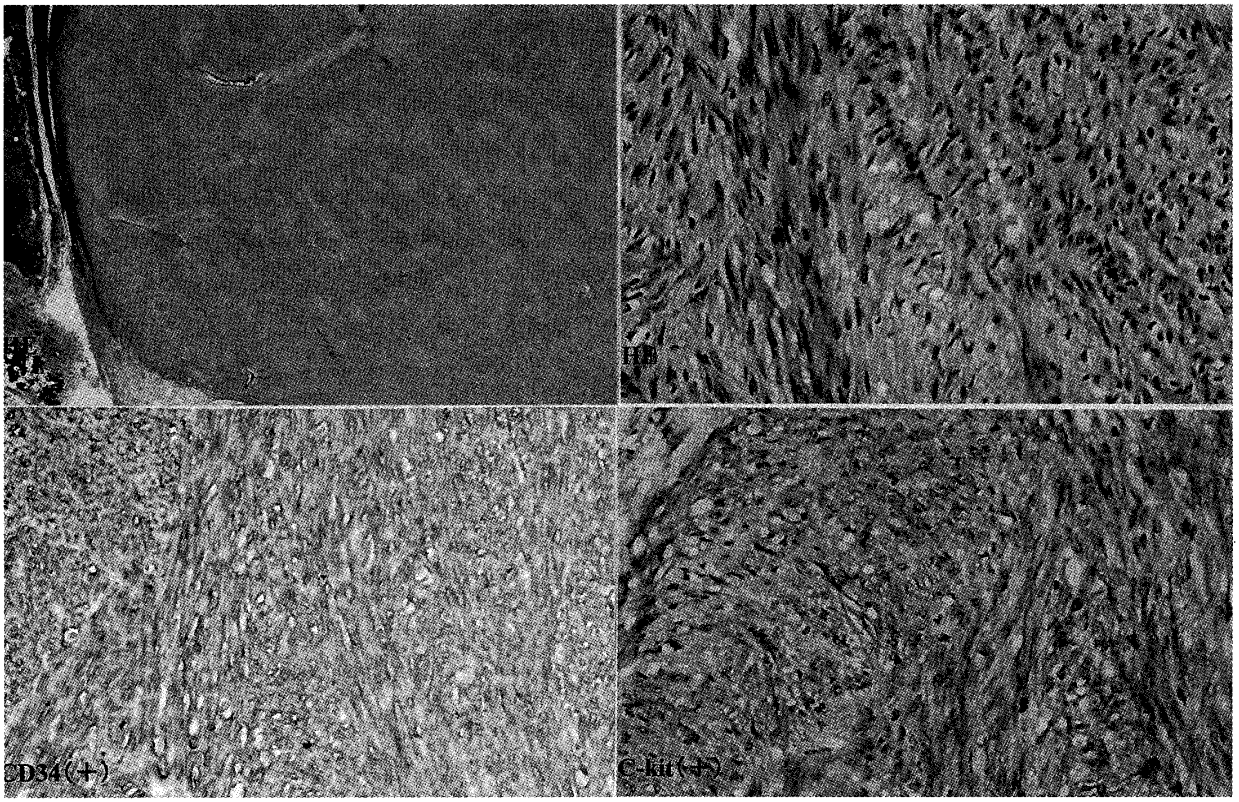

Fig. 7

Seven days after the operation, the patient developed a small bowel obstruction due to a parastomal herniation. A reoperation was performed on that day. A segment of the ileum was resected and the ileostomy was closed. Post-operative course after the reoperation was uneventful and the patient was discharged 34 days after the initial operation.

Discussion

Gastrointestinal stromal tumors (GIST) of the gastrointestinal tract are common in the stomach (39%) and small intestine (32%), but not in the rectum (10%) and colon (5%). Until the diagnostic criteria of GISTs became established, most of such tumors had been reported as leiomyomas or leiomyosarcomas. The sites of metastasis of GIST are mostly in the liver (65%), and lymph node involvement is uncommon (6%)[1].

Surgical resection is the most effective treatment for GIST. Pierie[2] reported the outcome and prognosis of 69 patients with GIST. In 59% of patients, complete gross resection of the tumor was possible. After complete resection, 3- and 5- year survival rates were 54% and 42%, compared with 13% and 9% after incomplete resection. Thirty-nine patients had no metastases at the time of diagnosis and were able to undergo a complete resection. Among these patients, the tumor recurred at a distant site in 35% (18% in the liver only, 9% in the liver and another distant site such as the lung, bone, or skin, 6% intraperitoneal only, and 2% developed lymph node metastases) and locally in 25%.

The operative method in patients with GIST of the rectum should be chosen according to the tumor size and location. In 1992, Vorobyov[3] reported the operative methods of 36 patients with

rectal tumors that were diagnosed as leiomyoma. Endoscopic resection of the tumor was performed in 33.3% of the patients with tumors mostly less than 1cm. Transanal excision was performed in 27.8% of the patients. In one female patient, a tumor measuring 5cm and located in the rectovaginal wall was removed through the vagina. Abdominoperineal resection of the rectum was performed in two patients with large neoplasms exceeding 8cm in diameter and located in the lower ampulla recti, whereas abdominal resection with colo-anal anastomosis was necessitated in five patients with tumors 8-20cm in diameter. In our case, a tumor was located on the anterior rectal wall about 3cm from the anal verge, and the patient was very fat. We predicted that complete unblock resection of the tumor would be extremely difficult through an abdominal approach because operative management in the pelvic cavity was very difficult. Further, the rate of lymph node metastases of GIST was reported to be very low. Therefore, we chose the transvaginal approach for the resection. To our knowledge, only40 cases of rectal GIST were reported in Japan (Table 1). Abdominoperineal resection (APR), transanal resection, low anterior resection (LAR) and total pelvic exenteration were performed in 25, 4, 2 and 3 cases, respectively. Among the Japanese series, there was only one report of rectal GIST that was resected by transvaginal approach.[4)~27)] Wide resections including APR, LAR and total pelvic resection were performed in 30 cases (75%), and local resections were performed in 10 cases (25%). The average tumor size that necessitated wide resection was 9.3 cm and the average tumor size that necessitated local resection was 5.8cm.

Khalifa[28)] reported that the recurrence rate of leiomyosarcoma of the rectum was 67.5% after local resection and 19.5% after abdominoperineal resection. However, the crude survival figures at five and ten years were found to be similar, in both of the surgical procedures. Patients survival, however, was related to the degree of tumor differentiation, and patients with poorly differentiated tumors had poor prognosis. Survival was also better for patients with smaller tumors. Quan and Berg[29)] are of the opinion that when the tumor is small (<2cm in diameter) and of low-grade malignancy, local excision, including a wide area of normal tissue, is sufficient.

With the experience of our present case, we consider that transvaginal approach is recommended as an appropriate method of surgical resection for a GIST situated on the anterior wall of the lower rectum.

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Table 1 Reported cases of the rectal GIST in Japan

case	year	author	size(cm)	operarion method
1	1997	Ito	8	APR
2	1999	Takahashi	10×10× 6	APR
3	1999	Takahashi	6 × 7 × 4	APR
4	1999	Takahashi	8 × 9 × 7	APR
5	1999	Takahashi	7.5×6.5× 5	APR
6	1999	Yokoi	8	APR
7	1999	Oohira	9.0×6.2	total pelvic exenteration
8	1999	Takahashi	6.5×7.5× 5	LAR
9	1999	Ikehata	6	APR, hysterectomy, vaginectomy
10	2000	Matsumoto	7	APR
11	2000	Imazu	5	APR
12	2000	Hirahara	5	APR
13	2000	Yamaguchi	3.5×3.5×7.0	APR
14	2000	Ito	9 × 8	TAE → APR
15	2001	Sawada	4	APR
16	2001	Nozawa	11	APR
17	2001	Kuruma	5	APR
18	2001	Kirizuka	7.5× 6 × 6	APR
19	2001	Shibata	8 × 7 × 6	APR
20	2001	Yamazaki	6 × 6	APR
21	2001	Sato	4.3×2.5	APR
22	2001	Nakasaki	8 × 7	APR, hysterectomy, bilateral salpingoophorectomy resection of the posterior wall of the vagina
23	2001	Ogata	6	transvaginal resection parcial resection of the anterior wall of the rectum
24	2001	Hama	9.8×7.5×5.3	tumor resection, radical prostatectomy
25	2002	Takahashi	8 × 6	APR
26	2002	Ishikawa	3.7	APR
27	2002	Kimura	4.5×4.5	APR
28	2002	Saito	5	APR
29	2002	Shinohara	3 <	super LAR, total prostatectomy
30	2002	Yano	4	transanal resection
31	2002	Oouchi	8	transanal resection
32	2002	Nagamachi	4 × 5	transanal resection
33	2002	Kameyama	3.2×2.2×2.7	transanal resection
34	2002	Katsuno	9.5×8.6×8.0	transmural partial resection
35	2002	Toyoshima	2	transsphincter local resection
36	2002	Kimihira	4 × 5 × 5	total pelvic exenteration
37	2002	Ando	18×18×10	total pelvic exenteration
38	2003	Yamamoto	5	APR
39	2003	Sasaki	6 × 6 × 3	transsacrally parcial resection of the rectum
40	2004	present ca	5	transvaginal resection

APR : abdominoperineal resection
LAR : low anterior resection
TAE : transcatheter anterial embolization

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経膣的切除術を施行した直腸 gastrointestinal stromal tumor の1例

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要　約

症例は71歳女性。下血を主訴に近医を受診し，精査加療目的に当科に入院した。直腸指診で，直腸前壁に表面平滑な硬い腫瘤を触知した。大腸内視鏡検査では，肛門縁より約3cmの位置に中心に潰瘍を伴う粘膜下腫瘍を認めた。生検組織標本の病理組織学的検査および免疫染色にて gastrointestinal stromal tumor（GIST）と診断し，経膣的腫瘍切除術および回腸瘻造設術を施行した。膣後壁に縦切開を加え，更にその下端に横切開を置き，膣壁を直腸膣中隔より剝離した。次いで腫瘍より約1cm離して直腸壁を腫瘍周囲より切離し腫瘍を摘出した。55mm×45mm×35mm大の灰白色，軟な充実性腫瘍で，病理組織学的には紡錘形細胞の増殖を認め，免疫染色にてS-100蛋白，α-smooth muscle actin陰性，CD34，c-kit陽性でGISTと診断した。直腸GISTの本邦報告例は自験例を含め47例であるが，経膣的切除は本症例の他には1例にしか施行されていない。今回我々は，直腸GISTに対する経膣的切除の適応と手技についての考案を加えて本症例を報告する。

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