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Case report

Transanal approach after preoperative imatinib treatment of a rectal gastrointestinal stromal tumors with external anal sphincter invasion: A case report

Makoto Hisanaga [a,*], Takashi Nosanaka [b], Hiroshi Maruta [a], Keizaburou Maruyama [a], Hidetoshi Fukuoka [a], Hiroyuki Yamaguchi [a]

[a] Department of Surgery, Japan Community Health Care Organization Isahaya General Hospital, Nagasaki, Japan
[b] Division of Surgical Oncology, Department of Surgery, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Japan

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ABSTRACT

Introduction and importance: Rectal gastrointestinal stromal tumors (GISTs) are rare, and preserving anorectal function can be challenging. We report the case of a patient with rectal GIST with external anal sphincter invasion, treated via the laparoscopic and transanal approaches.

Case presentation: A 61-year-old man with locally advanced GIST in the right anterolateral wall of the lower rectum was examined. Lower endoscopy revealed a 50-mm submucosal tumor located 4 cm from the anal verge. On immunohistochemistry, the biopsy specimen tested positive for CD34 and C-KIT, and the patient was diagnosed with GIST. Abdominal magnetic resonance imaging (MRI) revealed external anal sphincter infiltration. Because of the large tumor size and proximity to the anal verge, preserving the anus was challenging, and colorectal resection was avoided. Instead, neoadjuvant therapy with imatinib was administered to facilitate local resection of the tumor. Post-treatment MRI showed a reduction in tumor size (30 × 20 × 30 mm), and surgery was performed. We identified an appropriate resection line for diplomatic sphincter resection of the infiltrated area by laparoscopy alone. Thus, we performed a hybrid surgery using the laparoscopic and transanal approaches. The patient had an unremarkable postoperative course and was discharged on postoperative day 23.

Clinical discussion: No study has reported cases of rectal GIST with external anal sphincter invasion wherein anal function was preserved. Here, imatinib was administered preoperatively, and hybrid surgery was performed using the transanal and laparoscopic approaches.

Conclusion: Preoperative treatment and surgery preserved anorectal function in a patient with a massive rectal GIST.

1. Introduction

A rectal gastrointestinal stromal tumor (GIST) is a rare disease, accounting for approximately 5% of all GISTs [1]. Complete resection with a surgically safe resection margin is considered the primary treatment option for GISTs without distant metastases. For locally advanced rectal GISTs, the performance of curative surgery, such as laparoscopic rectal resection or extensive surgery, is required. In locally advanced GISTs with a strong tendency to invade toward the anus, it is challenging to preserve the anus and secure the resection margin.

We report the case of a patient with an anterior rectal wall GIST with external anal meatus invasion, which was treated with imatinib for tumor size reduction, followed by laparoscopic intersphincter rectal resection via a transanal approach and hand-sewn rectoanal anastomosis. This case report was written in accordance with the 2020 SCARE criteria [2].

2. Presentation of case

A 61-year-old man presented with a rectal tumor on undergoing

Abbreviations: GIST, gastrointestinal stromal tumor; CT, computed tomography; MRI, magnetic resonance imaging.

* Corresponding author at: Department of Surgery, Japan Community Health Care Organization Isahaya General Hospital, 24-1 Eishouhigashi-machi, Isahaya, Nagasaki 854-8501, Japan.
E-mail address: hisanaga-makoto@isahaya.jcho.go.jp (M. Hisanaga).

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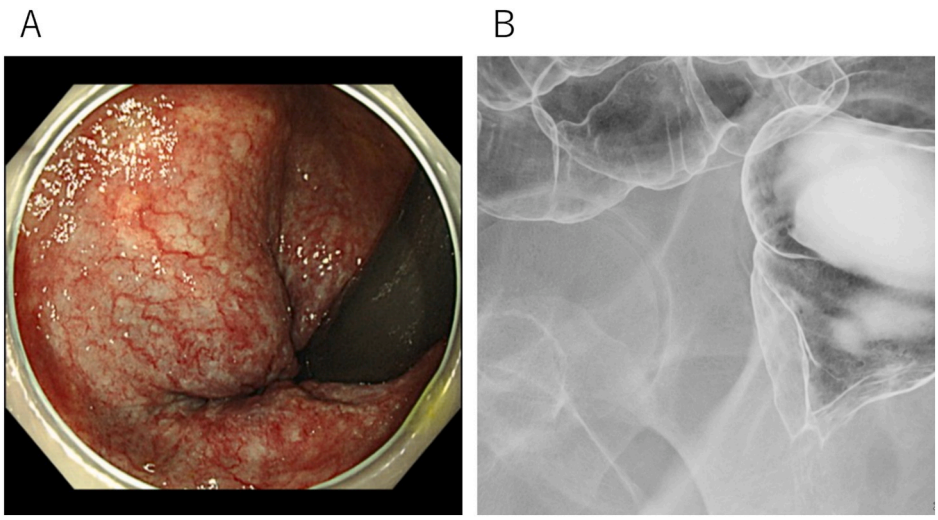

Fig. 1. Lower gastrointestinal endoscopy (A) and contrast enema (B) showing a smooth-surfaced mass approximately 4 cm from the anal verge.

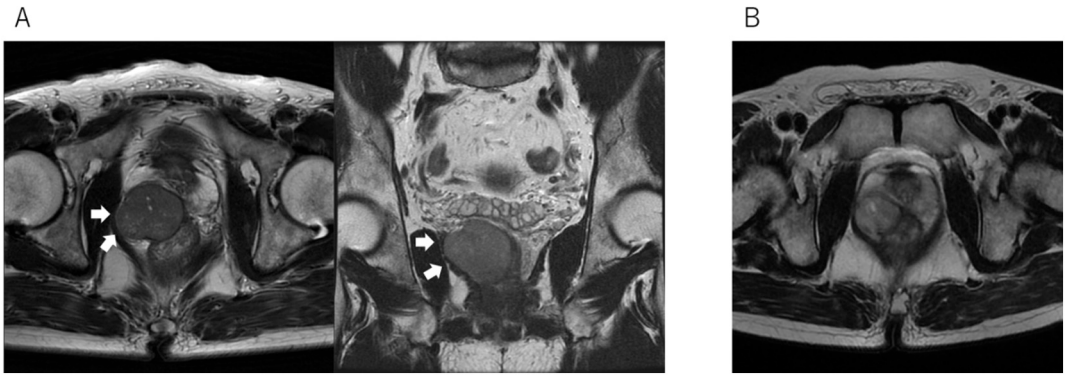

Fig. 2. Tl-weighted magnetic resonance imaging before and after treatment with imatinib.
(A) Before imatinib administration: the tumor (50 mm in diameter) is apparent in the right wall, and involvement of the external anal sphincter is observed (arrows).
(B) After imatinib administration: a reduction in the tumor size (30 mm in diameter) is observed.

computed tomography (CT). Digital rectal examination revealed a smooth-surfaced mass approximately 4 cm from the anal verge on the right rectal wall. Colonoscopy findings revealed a well-defined submucosal tumor approximately 4 cm from the anal verge on the right anterior aspect of the anal canal (Fig. 1A–B). CT findings of the chest, abdomen, and pelvis showed no distant metastases to the lungs or liver. Magnetic resonance imaging (MRI) revealed a tumor, approximately 50 mm dorsal to the prostate, invading the external anal sphincter (Fig. 2A). Transrectal needle biopsy was performed; on histological examination, the biopsy specimen tested positive for CD34- and -KIT. He reported no history of smoking or alcohol consumption. No genetic and syndromic abnormalities, drug allergies, previous surgical history were reported. Abdominoperineal resection was considered because of the proximity of the tumor to the anal verge, suspected external anal sphincter invasion, and large tumor size, which impaired the visual field. Digital rectal examination did not reveal any decrease in muscle tonus; urinary function was normal, and the patient strongly desired sphincter preservation. Therefore, neoadjuvant therapy (imatinib mesylate 400 mg/

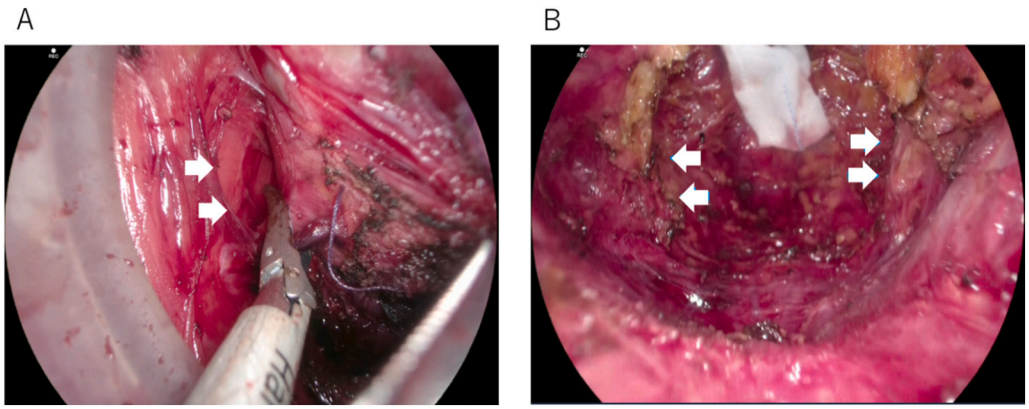

Fig. 3. Transanal view.
(A) Mucosectomy was performed through the anal canal. In areas where invasion was suspected, the external anal sphincter was partially attached to the resection side.
(B) Post-resection photograph: The right and left third and fourth pelvic nerves (arrows) are preserved.

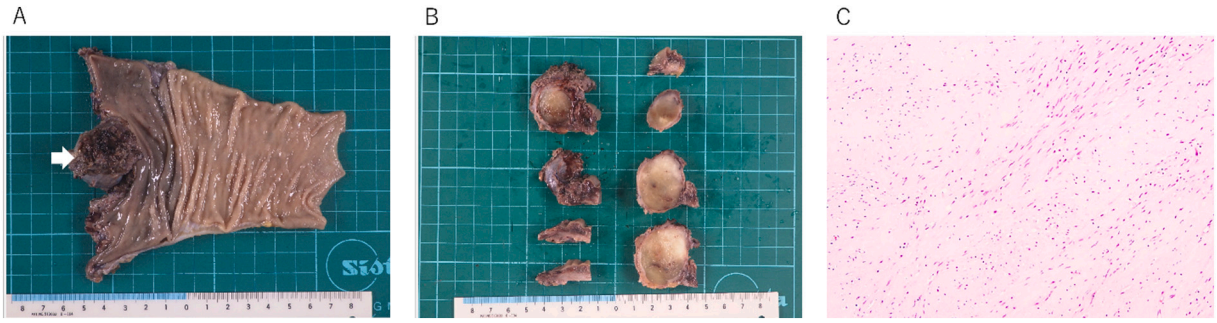

Fig. 4. Macroscopic and microscopic findings.
(A) The specimen measured 35 × 35 × 30 mm with a capsule. The arrow indicates the tumor. (B) The cut surfaces are white and sold. Pathologic examination confirming a negative resection margin with a capsule. (C) Histopathological examination showing spindle shaped tumor cells arranged in diverging bundles (hematoxylin and eosin staining, ×200).

day) was administered for 5 months. Post-treatment MRI showed a reduction in the tumor size (30 × 20 × 30 mm) (Fig. 2B), and the patient underwent surgery. It was challenging to identify an appropriate resection line on the anorectal side of the tumor by laparoscopy alone. Thus, we planned to perform a hybrid operation using the laparoscopic and transanal approaches. After introducing a transanal access platform, mucosal resection was initiated above the dentate line. On the right side of the tumor, the external anal sphincter was partially attached to the resection side. Meanwhile, the mouth side of the tumor was dissected. Next, the tumor was removed continuously by laparotomy, with the dissected anorectal side (Fig. 3A–B). Then, hand-sewn rectoanal anastomosis was performed. Finally, a temporary ileal colostomy was performed. No tumor was found in the resection margins (Fig. 4A–B). Pathological examination was performed (hematoxylin and eosin staining) and showed proliferation of spindle-sharped cells arranged in a fascicle pattern, and the mitotic rate was moderate (less than five mitoses/50 high-power fields) (Fig. 4C). The patient was treated without postoperative complications until postoperative day 23, when he was able to manage his stoma. Ileostomy closure was performed at 6 months after the initial surgery. At present, 12 months have passed since the surgery, and no recurrence has been observed.

3. Discussion

GIST is the most common mesenchymal mass occurring in the gastrointestinal tract. Rectal GISTs are rare and account for 5% of GISTs. GISTs are characterized by mutations in genes involved in the expression of C-KIT (CD117), a receptor tyrosine kinase [3]. CD34 is also frequently expressed but is less specific than CD117 [4]. The main prognostic factors for malignancy are the mitotic rate, tumor size, and tumor origin [5,6]. Surgical resection is the principle treatment for GISTs [7], but larger tumors are more difficult to resect completely and have a higher risk of local recurrence because of intraoperative capsule damage. Treating rectal GISTs is particularly difficult because of the anatomical constraints of the pelvis, which hinder manipulation and preserve anal function [8,9]. In particular, there have been no reports on GISTs wherein anal function was preserved because of suspected external anal sphincter invasion, as in this case.

Imatinib mesylate, a tyrosine kinase inhibitor, has recently been used as neoadjuvant therapy and is indicated for unresectable primary tumors or when major surgery is required because of the tumor location [5]. It reportedly promotes organ- and function-preserving surgeries, especially in cases of rectal GISTs [10]. A review of nine retrospective case series assessed 118 patients with rectal GISTs who were treated with neoadjuvant imatinib; among them, five (4.2%), 78 (66.1%), 35 (29.7%), and one (0.8%) patients showed a complete response, partial response, stable disease, and progressive disease, respectively, according to the Response Evaluation Criteria in Solid Tumors [11].

Neoadjuvant therapy (imatinib 400 mg orally [12,13]) was also administered. After 5 months, the effect of neoadjuvant therapy was evaluated. The tumor size reduction was observed, and therefore, R0 resection while preserving anal function was deemed feasible. The transanal approach has reportedly been useful for rectal GISTs, wherein securing the surgical margin is difficult [14,15]. In our case, the tumor volume was large, and invasion of the external anal sphincter was observed. The transanal and laparoscopic approaches were used to identify the resection line. Therefore, we successfully performed R0 resection without nerve damage, which may have caused sexual dysfunction or urinary dysfunction.

4. Conclusion

We reported a case of rectal GIST wherein anorectal function was preserved by hybrid surgery using preoperative imatinib administration and a transanal approach.

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.

Provenance and peer review

Not commissioned, externally peer reviewed.

Ethical approval

No ethical approval was required.

Guarantor

Hidetoshi Fukuoka.

CRediT authorship contribution statement

Makoto Hisanaga: investigation, writing–original draft, data curation, surgical therapy for this patient; **Takashi Nonaka:** investigation, data curation, writing–review & editing; **Hidetoshi Fukuoka:** investigation, data curation, writing–review & editing, and funding acquisition.

Declaration of competing interest

The authors report no declarations of interest.

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