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, Keizaburo 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

ARTICLE INFO

Keywords:
Gastrointestinal stromal tumors
Rectum
Transanal approach
Imatinib
Neoadjuvant chemotherapy

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 definitive 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 undergoing...
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/
reported 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.
Acknowledgments

We would like to thank Editage (www.editage.com) for English language editing.

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

References

[1] M. Miettinen, J. Lasota, Gastrointestinal stromal tumors (GISTs): definition, occurrence, pathology, differential diagnosis and molecular genetics, Pol. J. Pathol. 54 (2003) 3–24.

[2] A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, et al., The SCARE 2020 guideline: updating consensus surgical Case REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230, https://doi.org/10.1016/j.ijsu.2020.10.034.

[3] S. Hirota, K. Isozaki, Y. Moriyama, K. Hashimoto, T. Nishida, S. Ishiguro, et al., Gain-of-function mutations of c-kit in human gastrointestinal stromal tumors, Science 279 (1998) 577–580, https://doi.org/10.1126/science.279.5350.577.

[4] Z.X. Jiang, S.J. Zhang, W.J. Peng, B.H. Yu, Rectal gastrointestinal stromal tumors: imaging features with clinical and pathological correlation, World J. Gastroenterol. 19 (2013) 3108–3116, https://doi.org/10.3748/wjg.v19.i20.3108.

[5] ESMO/European Sarcoma Network Working Group, Gastrointestinal stromal tumours: ESMO clinical practice guidelines for diagnosis, treatment and follow-up, Ann. Oncol. 25 (2014) 21–26, https://doi.org/10.1093/annonc/mdw255.

[6] M. Miettinen, J. Lasota, Gastrointestinal stromal tumors - definition, clinical, histological, immunohistochemical, and molecular genetic features and differential diagnosis, Virchows Arch. 438 (2001) 1–12, https://doi.org/10.1007/s004280000538.

[7] G.D. Demetri, M. Von Mehren, C.R. Antonescu, R.P. DeMatteo, K.N. Ganjoo, R. G. Mak, et al., NCCN task force report: update on the management of patients with gastrointestinal stromal tumors, J. Natl. Compr. Cancer Netw. 8 (Suppl. 2) (2010) S1–41, https://doi.org/10.6004/jnccn.2010.0116, quiz 542.

[8] M. Fiore, C.P. Raut, A. Gronchi, Are we allowed to limit surgical aggressiveness with small rectal gastrointestinal stromal tumors? Ann. Surg. Oncol. 24 (2017) 1153–1156, https://doi.org/10.1245/s10434-016-5719-5.

[9] Y. Fujiimoto, T. Akiyoshi, T. Konishi, S. Nagayama, Y. Fukunaga, M. Ueno, Laparoscopic sphincter-preserving surgery (intersphincteric resection) after neoadjuvant imatinib treatment for gastrointestinal stromal tumor (GIST) of the rectum, Int. J. Colorect. Dis. 29 (2014) 111–116, https://doi.org/10.1007/s00384-013-1769-7.

[10] N. Wachter, M.A. Worms, D.P. Dos Santos, H. Lang, T. Huber, W. Kneist, Transanal minimally invasive surgery (TAMIS) approach for large juxta-anal gastrointestinal stromal tumour, J Minim Access Surg 12 (2016) 289–291, https://doi.org/10.1016/j.jmas.2020.10.034.

[11] M. Kaneko, S. Emoto, K. Murono, H. Sonoda, M. Hiyoshi, K. Sasaki, et al., Neoadjuvant imatinib therapy in rectal gastrointestinal stromal tumors, Surg. Today 49 (2019) 460–466, https://doi.org/10.1007/s00595-018-1737-5.

[12] J. Verweij, P.G. Casali, J. Zalcberg, A. LeCesne, P. Reichardt, J.Y. Blay, et al., Progression-free survival in gastrointestinal stromal tumours with high-dose imatinib: randomised trial, Lancet 364 (2004) 1127–1134, https://doi.org/10.1016/S0140-6736(04)17098-0.

[13] S. Machlenkin, I. Pinkh, H. Tulchinsky, Y. Ziv, J. Sayfan, D. Dwek, et al., The effect of neoadjuvant imatinib therapy on outcome and survival after rectal gastrointestinal stromal tumour, Color. Dis. 13 (2011) 1110–1115, https://doi.org/10.1111/j.1463-1318.2010.02442.x.

[14] K. Kagakari, S. Matoba, S. Toda, Y. Maeda, M. Ueno, H. Kuroyama, Hybrid resection of massive rectal gastrointestinal stromal tumor using laparoscopic and transanal approaches, Asian J Endosc Surg 14 (2021) 102–105, https://doi.org/10.1111/aes.12824.

[15] C. Kosmidis, K. Sapalidis, A. Tsakalidis, S. Atmatzidis, N. Michalopoulos, G. Koinis, et al., Management of low rectal gastrointestinal stromal tumor with neoadjuvant therapy and transanal excision: a rare case report and review of the literature, Int J Gen Med 12 (2019) 121–124, https://doi.org/10.2147/IJGM. SI93988.