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Large gastrointestinal stromal tumor and advanced adenocarcinoma in the rectum coexistent with an incidental prostate carcinoma: A case report

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ABSTRACT

INTRODUCTION: Gastrointestinal stromal tumors (GISTs) are the leading mesenchymal neoplasia in the gastrointestinal tract, but GIST arising from the rectum is rare. When a secondary neoplasia coexists in the vicinity of a rectal GIST, more aggressive treatment strategies may be needed to cure the diseases.
PRESENTATION OF CASE: We herein describe a 76-year-old man with a large gastrointestinal stromal tumor along with an advanced adenocarcinoma in the rectum that coexisted with prostate carcinoma. Preoperative examination revealed an advanced adenocarcinoma of the upper rectum and a large pelvic mass suggestive of a GIST or a neuroendocrine tumor arising from the anterior wall of the lower rectum. To eradicate the tumor, total pelvic exenteration with ureterocutaneous fistula was carried out after obtaining written informed consent. Immuhistochemical studies revealed the concurrence of an advanced rectal cancer (T3, N1, M0) and a malignant GIST (c-kit-positive, CD34-positive, vimentin-positive, and CAM5.2-negative), and an incidental prostatic acinar adenocarcinoma. The patient was given adjuvant chemotherapy with imatinib and remains disease-free as of 12 months after surgery.
DISCUSSION: A PubMed search for the case of coexistence of GIST with two other malignancies revealed only four cases, making this very rare condition.
CONCLUSION: Radical surgery with perioperative adjuvant chemotherapy using tyrosine kinase inhibitors is the choice for treatment of large GISTs with a malignant potential. Our report suggests that aggressive surgical approach would be feasible, when a secondary tumor is present near the GIST.

1. Introduction

The most common location of gastrointestinal stromal tumor (GIST) is the stomach (60–70%) followed by the small intestine (20–25%), as compared to only approximately 5% in the rectum.[1,2]

The coexistence of GIST with other epithelial cancers of different histological types has been reported, where the second tumor can develop synchronously or metachronously.[3] Of interest are those cases in which one or more tumors were located within the same organ. When a secondary neoplasia coexists in the vicinity of a GIST, more aggressive treatment strategies would be needed to cure the diseases. We report a patient who underwent a total pelvic exenteration for a rectal GIST concurrent with an advanced rectal cancer and an incidental prostate cancer.

2. Case report

A 76-year-old man suffered from constipation for 6 months. At the age of 26 years, he had undergone an appendectomy. The family history of the patient was unremarkable. He visited a local hospital where digital examination revealed a tumor with a hard, elastic and smooth surface in the anterior wall of the rectum at about 4 cm above the dentate line. Magnetic resonance imaging (MRI) showed a mass with a smooth margin, 7 cm × 5 cm in size mainly occupying the anterior wall of the lower rectum (Fig. 1). These findings suggested a GIST or rectal carcinoid originating from the rectal wall. The biopsy was avoided for the risk of intra-abdominal seeding or tumor rupture. Then he was referred to our hospital for further examination and treatment. Laboratory examination was unremarkable. Colonoscopy revealed an irregular tumor in the rectosigmoid colon approximately 15 cm from the anal verge,

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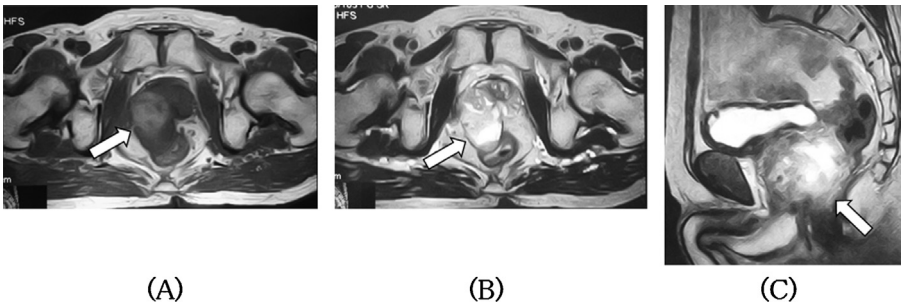

Fig. 1. Magnetic resonance imaging. (A) Transverse T1-weighted image showing a homogeneous mass with intermediate signal intensity (arrow). (B) Transverse T2-weighted image showing a heterogeneous mass with high signal intensity (arrow). (C) Sagittal T2-weighted image could not show clear delineation between the tumor and the prostate (arrow).

aside from the pelvic tumor, and biopsy of the tumor demonstrated moderately differentiated adenocarcinoma. However, no visible mucosal abnormality relevant to the pelvic tumor was found. Contrast-enhanced computed tomography (CT) showed an irregular circumferential mural thickening involving the rectosigmoid colon with no enlarged lymph nodes and a solitary irregular and low-density mass in the lower rectum extending from the anterior rectal wall into the prostate. No distant metastasis including the liver was found.

Based on these findings, the patient was judged to have adenocarcinoma of the upper rectum concurrent with a malignant submucosal tumor of the lower rectum. To minimize the risk of tumor spread during the dissection between a large fragile GIST and the prostate in the lower pelvic cavity and to accomplish complete en bloc resection of the two concomitant malignant tumors, total pelvic exenteration (TPE) with ureterocutaneous fistula was selected (Fig. 2). At operation, a 3 cm well-circumscribed nodule was identified in the mesentery of the sigmoid colon, and therefore fine needle aspiration biopsy of the pelvic tumor and incisional biopsy of the mesenteric was performed. However both specimens failed to identify malignancy.

Postoperatively, histopathological examination of the surgical specimen revealed a moderately differentiated rectal adenocarcinoma (T3, N1, M0), rectal GIST with the same pathology as the mesenteric nodule with malignant biological behavior, and an incisional prostatic acinar adenocarcinoma with a combined Gleason score of 6 (3 + 3) that was confined to the left prostate lobes.

Further examination of rectal GIST revealed stromal cell neoplasm with necrotic and hemorrhagic areas and large tumor size (>5 cm), a high index of mitotic count: above 5 mitoses per high-power fields (HPF). Immunohistochemical analysis revealed positive staining for CD117 (c-kit), CD-34 and Vimentin, and negative CAM5.2 (Fig. 3). Based on the histopathological finding, the GIST was diagnosed as a high grade malignancy. Resection margins were free of both the tumors. Although the postsurgical recovery was uneventful, the patient wanted to receive postoperative rehabilitation in the hospital as long as possible and was discharged on postoperation day 46.

Following the histopathological confirmation, treatment with imatinib was initiated on postoperative day 60. The imatinib was introduced at a dose of 400 mg/day in order to minimize the risk of relapse associated with the drug interruption. Postoperatively, patient was followed up with CT at 12 months after the operation; no metastasis has been observed.

3. Discussion

GISTs are the most common type of non-epithelial neoplasia diagnosed in the gastrointestinal tract (0.1–3% of all gastrointestinal tumors) and expresses CD117, a c-kit proto-oncogene, which can be detected immunohistochemically.[4]

Recently, cases of synchronous or metachronous development of GISTs and concomitant another neoplasm with different incidence, etiology, evolution and prognosis have been reported. Abbas et al. reported about 10% of patients with GISTs develop other cancers either synchronously or metachronously.[5] The major types of GIST-associated malignancies reported in the literature include gastrointestinal carcinomas, lymphoma/leukemia, gynecological carcinomas, and carcinomas of the prostate, breast, pancreas, lung, liver or kidney as well as carcinoid of the pancreas or stomach.[5–7] Regarding the simultaneous development of GIST and other cancers, various hypotheses such as gene mutation, expression of metallothioneins, neighboring tissues being influenced by the same carcinogens, have been proposed.[8,9] However, the limited number of such cases cannot confirm the existence of a common factor in tumorigenesis of these histopathologically completely different

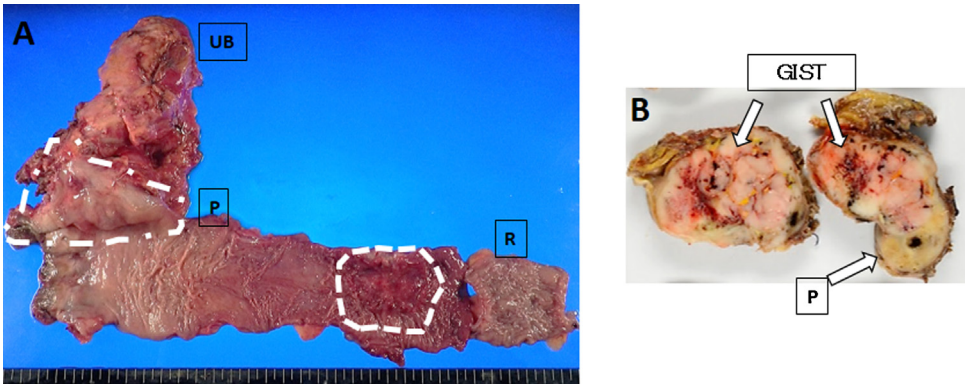

Fig. 2. (A) Resected specimen showing concurrent rectal GIST and adenocarcinoma of rectum. (B) Rectal GIST without prostatic infiltration. UB: urinary bladder; P: prostate; R: rectum.

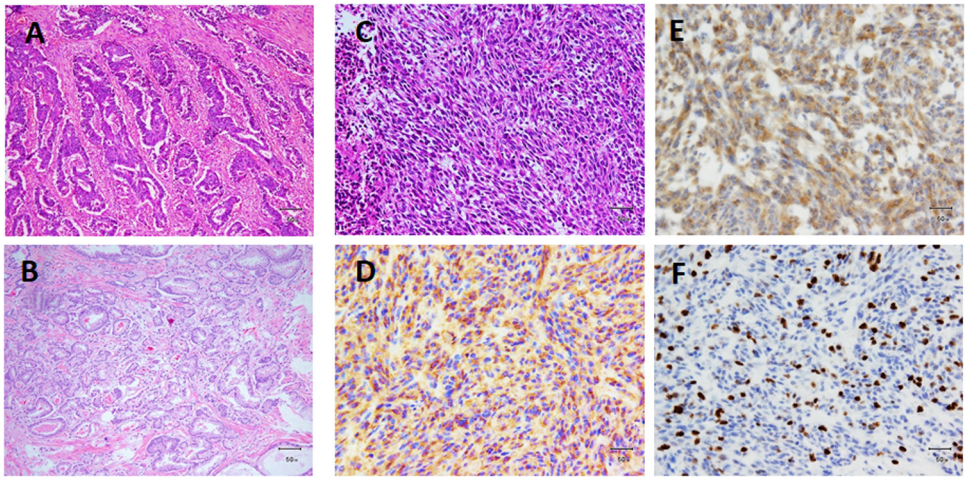

Fig. 3. Microscopic images. (A) Rectal adenocarcinoma (H-E stain, ×20). (B) Prostate adenocarcinoma (H-E stain, ×10). (C) Rectal GIST (H-E stain, ×20). (D and E) Immunohistochemistry indicating strong staining for CD117 (D, ×20), and CD-34 (E, ×40). (F) MIB-1 index was about 15% (×20).

tumors. Simple coincidence could be the most reasonable explanation.

What is unique in our case is that coexistence of GIST of the rectum with two other malignancies. To our knowledge, only four cases of coexistence of GIST with multiple other malignancies have been reported.[5,10] In all four such cases, GISTs coexisted with malignancies of distant organs. The current case seems to be the first report wherein a coexistence of GIST of the rectum, coexisted with rectal adenocarcinoma and prostate carcinoma within neighboring organs and resected together in a single surgical specimen is being reported. The majority of the coexistent GISTs are discovered incidentally during work-up or during therapeutic procedures for GI malignancies. In the current case, coexistence of rectal adenocarcinoma and prostate carcinoma was discovered incidentally, during examination and post laparotomy, respectively.

Neoadjuvant chemotherapy was not selected, because the preoperative diagnosis of GIST was not confirmative and complete resection of the tumors was possible with TPE. And furthermore, to reduce the tumor size and achieve a restorative surgery with increased costs, the benefit and risk of neoadjuvant imatinib therapy for a rectal GIST were discussed.

Adjuvant therapy for GIST was started according to a growing evidence supporting the benefits of tyrosine kinase inhibitors after surgical resection of GISTs with a high malignant potential.

4. Conclusion

A second malignant tumor is not so rare in patients who undergo or had undergone surgery for GISTs, because aging is the important risk factor for GISTs and cancers. The treatment strategy for GISTs could be altered by the biological features of a concurrent tumor.

Conflict of interest

None.

Funding

None.

Ethical approval

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.

Author Contributions

Toshiaki Suzuki contributed to data collections and writing.

Katsuhito Suwa, Ken Hanyu, Tomoyoshi Okamoto, Tetsuji Fujita and Katsuhiko Yanaga contributed to data collections.

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