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A gastrointestinal stromal tumor of the jejunum presenting with an intratumoral abscess: A case report and a literature review

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ABSTRACT

INTRODUCTION: Gastrointestinal stromal tumors (GISTs) are the most common mesenchymal tumors of the gastrointestinal tract. The small intestine is the second-most frequent location where GISTs occur after the stomach. Attention should be paid to small intestinal GISTs because they infrequently present with acute abdomen, which necessitates emergency surgery. This report describes a patient with a small intestinal GIST developing a giant intratumoral abscess, in whom emergency surgery was performed.

PRESENTATION OF CASE: A 56-year-old woman presented with worsening abdominal pain. Computed tomography scan showed an approximately 9.5 cm × 9 cm tumor bearing a thick and hypervascularized wall with an internal air-fluid level. Emergency laparotomy revealed the tumor originated from the jejunum, and partial resection of the jejunum was performed. A large amount of pus was contained inside the tumor. Immunohistochemically, the tumor was diagnosed as a high risk GIST of the Cjejunum, and imatinib mesylate was initiated.

DISCUSSION AND CONCLUSION: When an intratumoral abscess in the abdomen is confirmed, GISTs should be listed as differential diagnosis. Complete surgical resetcion with careful handling and adjuvant chemotherapy with imatinib mesylate are considered to be important for this state.

1. Introduction

Gastrointestinal stromal tumors (GISTs) are the most common mesenchymal tumor of the gastrointestinal tract. They can occur throughout the gastrointestinal tract, and the small intestine is the second-most frequent location after the stomach. Small intestinal GISTs are often asymptomatic until they grow large, and their clinical presentation is highly variable. Attention should be paid to small intestinal GISTs because they infrequently manifest abdominal emergencies such as bowel obstruction, tumor rupture, perforation, and intratumoral abscess formation. This report describes a patient with a small intestinal GIST that developed a giant intratumoral abscess together with a review of published cases. This work has been reported in line with the SCARE criteria [1].

2. Presentation of case

A 56-year-old otherwise healthy woman walked into the emergency room of our hospital with worsening abdominal pain for 4 days. Her body temperature was 38.1°; blood pressure, 92/54 mmHg; and heart rate, 107 beats/min. Physical examination revealed tenderness and muscular defense around the lower abdomen. Blood tests showed elevation in white blood cell count (11,160/μl) and C-reactive protein level (5.4 mg/dl). Levels of CEA and CA19-9 were within normal ranges. Contrast-enhanced computed tomography (CT) scan showed an irregularly shaped tumor in the lower abdomen measuring approximately 9.5 cm × 9 cm × 5.5 cm (Fig. 1a, b). The tumor wall was thick and enhanced with intravenous contrast material, suggesting hypervascularity. An air-fluid level was contained in the tumor. Free air and ascites were not observed. Because her general condition was not bad, and the CT scan findings did not suggest peritonitis, intravenous administration of antibiotics was chosen as initial treatment. In addition, preoperative examinations including upper and lower endoscopies and magnetic resonance imaging were planned. However, because her clinical symptoms and laboratory data did not respond to the treatment, and because her status was accompanied with high fever after admission, an emergency surgical procedure was performed on the 6th day after her admission.

Diagnostic laparoscopy was initially performed. Slightly turbid ascites were observed and collected for the culture test. The procedure also showed that the tumor was movable and was expected to be resectable. Therefore, open exploration was performed through

Abbreviations: GIST, gastrointestinal stromal tumors; CEA, carcinoembryonic antigen; CA19-9, cancer antigen 19-9.

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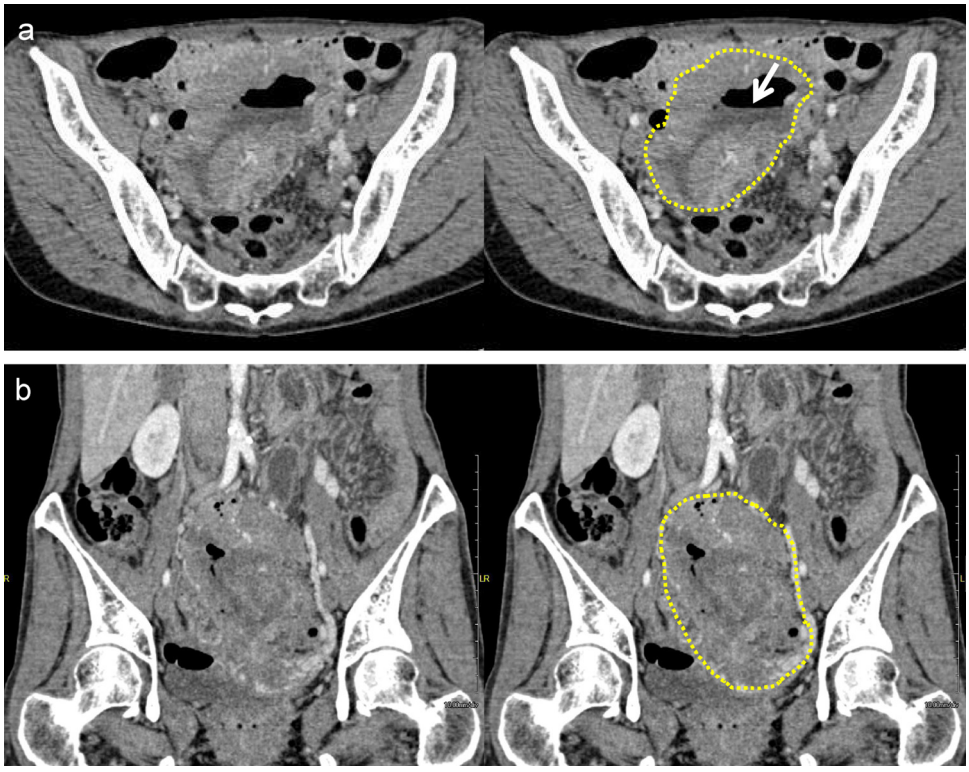

Fig. 1. (a, b) Contrast-enhanced computed tomography scan of the lower abdomen showing a large tumor (dotted line) bearing a thick and enhanced wall with an internal air-fluid level (arrow).

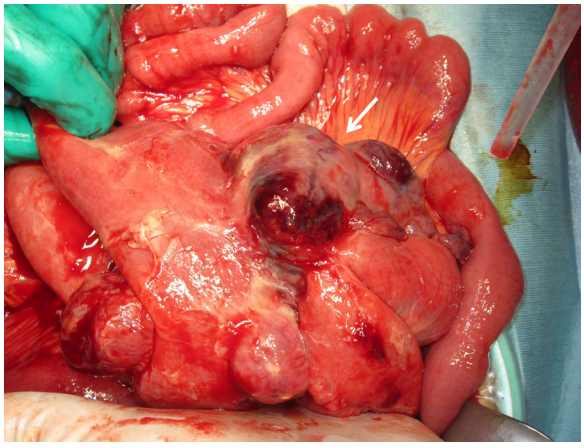

Fig. 2. Intraoperative photograph showing a tumor arising from the jejunal wall (arrow).

a midline incision. The laparotomy revealed that the tumor arose from the jejunum wall, and was located 20 cm from the ligament of Treitz (Fig. 2). Loose adhesion without invasion was observed and the tumor was easily separable with blunt dissection. Macroscopic perforation was not observed to be apparent. Partial resection of the jejunum with the tumor and sampling of some nearby lymph nodes were performed, and a small portion of the tumor was sent for frozen section, which revealed proliferation of spindle-shaped tumor cells. Thus, this patient was diagnosed as having a mesenchymal tumor of the jejunum, and lymphadenectomy was omitted.

At macroscopic observation, the excised tumor, measuring 9 cm in diameter, was irregularly shaped with a soft tumor wall, and had a central cavity containing a large amount of pus (Fig. 3a, b). There was a small fistula between the cavity and the intestinal lumen (Fig. 3c, d). The culture test of the ascites was negative for bacteria.

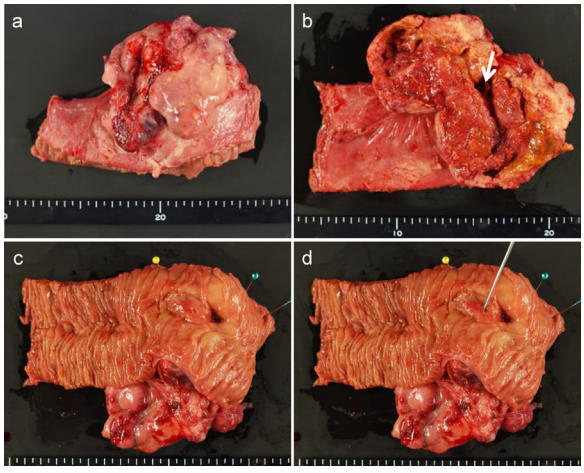

Fig. 3. Macroscopic findings of the tumor. (a) Gross appearance of the tumor. (b) A cavity surrounded by a thick tumor wall (arrow). (c, d) A small fistula between the cavity and intestinal lumen was observed.

Hematoxylin and eosin staining showed proliferation of spindle-shaped tumor cells. Immunohistochemical staining showed that the tumor cells were positive for c-KIT, CD34, SMA, and negative for Desmin, S-100. Mitotic count was 6/50 High Power Fields. Metastasis to the sampled lymph nodes was not observed. Therefore, the final diagnosis was high-risk GIST of the jejunum. Postoperative recovery was uneventful and she was discharged home on the 10th postoperative day. Thereafter, adjuvant imatinib mesylate was administered.

3. Discussion

GISTs are the most common mesenchymal tumors of the gastrointestinal tract. They can occur throughout the gastrointestinal

tract, but mostly in the stomach (60%) and the small intestine (30%). Less frequently, they can occur in the colon, rectum, appendix, esophagus, mesentery, omentum, or retroperitoneum [2]. Because GISTs of the small intestine often show an exophytic growth pattern [3], they tend to be asymptomatic until they grow large. A previous report described the median size of GISTs in the small intestine being as large as 7 cm at the time of diagnosis [4]. The clinical presentation of the GISTs in the small intestine is highly variable. The most frequent symptom is gastrointestinal bleeding, and others include vague abdominal pain, a palpable mass, weight loss, and fever [4]. Infrequently, small intestinal GISTs cause acute abdomen such as bowel obstruction, hemoperitoneum secondary to tumor rupture, and peritonitis secondary to tumor perforation [5]. Cases with intratumoral abscess formation as in our case are also rare.

When an abscess bearing a thick and hypervascularized wall is observed at contrast-enhanced CT scan, as in our case, an intratumoral abscess rather than a simple abscess should be suspected. There is a wide variety of potential diseases such as tumors of digestive organs, tumors of reproductive organs, and malignant lymphomas [6]. In addition, emergent surgery is often necessary because of strong symptoms without sufficient preoperative examinations. Therefore, it could be often difficult to narrow differential diagnosis. However, because GISTs are a relatively common disease and they can cause intratumoral abscesses as mentioned above, GISTs should be listed as a differential diagnosis.

Our literature search with MEDLINE revealed only 13 cases of small intestinal GISTs that presented an intratumoral abscess including the present case (Table 1). The median tumor diameter was 9.7 cm, larger than the ordinary size of the small intestinal GIST described previously [4]. In nine cases, a fistula between the abscess cavity and the intestinal lumen was observed. In six cases, the tumors were perforated and accompanied peritonitis. In seven cases, emergency surgery was performed. In all cases, partial resection of the small intestine with the tumor was performed. In seven cases, imatinib mesylate was initiated as adjuvant therapy.

The mechanism of intratumoral abscess formation is considered to comprise enteric bacteria entering through the fistula infected with necrotic tissue inside the tumor and developing into an intratumoral abscess. In cases with perforation, the process is considered to comprise intratumoral pressure that elevates rapidly along with the abscess formation and finally perforating the tumor wall. Although some reports have described conservative treatments as being effective, immediate surgery is considered to be eligible when symptoms are strong or when conservative treatments are found to be ineffective because this state may easily lead to perforation and peritonitis.

Complete surgical resection is the only potential curative treatment for GISTs [7]. For small intestinal GISTs, partial small bowel resection with the tumor is the standard procedure. Gentle handling is necessary because injury of the pseudocapsule may cause dissemination of the tumor cells and may lead to poor patient prognosis [7]. In cases with intratumoral abscess, dissection with extreme care is considered to be necessary because the pseudocapsule is expected to be fragile with inflammation. Lymphadenectomy is not routinely performed because lymph node metastasis is rare [7,8]. Intraoperative frozen section is useful because lymaphadenectomy can be omitted without hesitation when typical spindle-shaped cells are confirmed.

GISTs are categorized as low, intermediate, or high risk for recurrence risk based on tumor size, mitotic rate, location, and presence of rupture [9–11]. Imatinib mesylate, a tyrosine kinase inhibitor targeting KIT, was reported to improve prognosis after radical resection of high risk GISTs [12,13]. Small intestinal GISTs are reported to have more malignant potential than gastric GISTs [9]. In addition, GISTs with intratumoral abscess formation tend to

Table 1
Summary of small intestinal gastrointestinal stromal tumors with intratumoral abscesses in the available medical literature.

First author	Year	Age	Sex	Tumor size (cm)	Location	Emergency surgery	Macroscopic perforation	Peritonitis	Surgical procedure	Intraoperative frozen section	Macroscopic fistula	Mitotic count	Adjuvant imatinib	Outcome (months)
Karagulle E	2008	70	M	6.6	Jejunum	NR	+	NR	SBR	NR	NR	0/50	–	13 ANED
Ku MC	2010	33	F	6.5	Jejunum	+	+	+	SBR	NR	+	NR	–	NR
Feng F	2010	45	F	10	Jejunum	+	+	+	SBR	NR	+	<5/50	NR	NR
Kitagawa M	2010	66	F	7	Jejunum	–	–	–	SBR	NR	+	<5/50	NR	NR
Andican A	2012	48	M	21	Jejunum	NR	–	–	SBR	+	+	NR	+	12 ANED
Chen HW	2012	22	M	9.1	Jejunum	–	–	–	SBR	NR	NR	low mitotic count	–	2 ANED
Beltran MA	2013	46	M	7.1	Ileum	+	+	+	SBR	NR	NR	15/50	+	NR
Cabral FC	2015	49	F	14	Jejunum	+	+	+	SBR	NR	+	NR	–	NR
Rubini P	2016	51	M	7.5	Ileum	NR	–	–	SBR	NR	+	<5/50	+	72 ANED
Prakash JS	2017	60	F	6	Ileum	+	+	+	SBR	NR	NR	NR	+	NR
Sato K	2017	74	M	14	jejunum	+	+	+	SBR	NR	+	NR	+	22 Dead
Gorelik M	2018	63	F	9	Ileum	NR	–	–	SBR	+	+	<5/50	+	12 ANED
Our case	2018	56	F	9	Ileum	+	–	–	SBR	+	+	6/50	+	4 ANED

Abbreviations: SBR small bowel resection, NR not reported, ANED alive with no evidence of disease.

grow large and to frequently accompany perforation, as mentioned above. Therefore, adjuvant treatment with imatinib mesylate should be considered for the small intestinal GISTs developing into intratumoral abscess. In our case, recurrence risk was diagnosed to be high because the tumor size was large, the mitotic rate was high, and the possibility of perforation could not be denied. Therefore, imatinib mesylate was administered.

4. Conclusion

This report describes a patient with an intratumoral abscess caused by a small intestinal GIST, in whom emergency surgery was necessary. This case can provide an opportunity to review a rare presentation of small intestinal GISTs as an intratumoral abscess, and the management of this state.

Conflicts in interest

All authors have no conflicts of interest.

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Ethical approval

This research was approved by the ethics committee of Iwate Prefectural Iwai Hospital, japan.

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.

Author contribution

Soichi Ito is the main author of this article; Soichi Ito and Kazunori Katsura performed clinical treatment including surgery; Yuma Tsuchitani, Yuro Kim, Souhei Hashimoto, Yuichi Miura, Takuji Uemura, Takayuki Abe, Koichiro Sato, and Hirotaka Kato reviewed the manuscript; all authors have read and approved the final manuscript.

Registration of research studies

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