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Multiple mesenteric desmoid tumors after gastrectomy for gastric cancer: A case report and literature review

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ABSTRACT

INTRODUCTION: Many patients with desmoids have an antecedent trauma, particularly surgical intervention for familial adenomatous polyposis. However, cases of mesenteric desmoid after gastrectomy are extremely rare. We present a case of multiple mesenteric desmoids after total gastrectomy for gastric cancer.

PRESENTATION OF CASE: A 70-year-old man had undergone a total gastrectomy for early stage gastric cancer. He had no other relevant medical history or family history. A year after gastrectomy, a computed tomography showed three mesenteric masses, and we performed careful observation. The 2-year post-operative examination indicated slowly growing masses. There were no other lesions except for the three masses. We decided to perform diagnostic surgery to evaluate the tumors. There were three mesenteric masses (1.5 cm, 4 cm and 1.5 cm in diameter). We performed partial small intestinal resections for each mass. Histological examination showed that the tumors were desmoids. A year after surgery, he was doing well with no evidence of recurrence of the desmoids or the gastric cancer.

DISCUSSION: Diagnosing intra-abdominal desmoid tumors is often difficult. Especially if a patient has a history of malignancy, it may be extremely difficult to differentiate multiple mesenteric desmoids from a cancer recurrence. In the current case, surgical resection was a useful treatment option as diagnostic therapy.

CONCLUSIONS: Multiple mesenteric desmoids could emerge after gastrectomy for gastric cancer, and surgical resection of the tumors is a useful option as a diagnostic therapy.

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1. Introduction

Desmoids are rare tumors, and the estimated incidence is reported to be 2–4 per million population per year [1]. Although desmoids lack the potential for distant metastases, desmoid tumors are locally aggressive and can destroy vital organs, especially intra-abdominal desmoids that often result in fatal outcomes [2]. Most desmoid tumors are asymptomatic unless the destruction of organs has occurred. It is known that about 30 percent of patients with desmoids have an antecedent trauma, particularly a surgical inter-

vention for familial adenomatous polyposis (FAP) [3–5]. However, cases of mesenteric desmoid after gastrectomy for gastric cancer are extremely rare, and there have been few reports in the literature [6–9]. We present a case of multiple mesenteric desmoids after total gastrectomy for gastric cancer with a review of the relevant literature in line with the SCARE criteria [10].

2. Presentation of case

A 70-year-old Asian man had undergone total gastrectomy for a gastric cancer, and a Roux-en-Y esophagojejunostomy was performed. Histological examination of the tumor showed moderately differentiated tubular adenocarcinoma, and the tumor was classified as pT1N0M0, stage IA according to the Union for International Cancer Control classification, 7th edition. He had an uneventful postoperative course. He had no other medical history or family history. One year post-gastrectomy, he was asymptomatic, but the CT showed three mesenteric masses (0.5 cm, 2.5 cm and 1 cm in diameter). Based on mutual agreement with the patient that he required monitoring, we performed careful observation without surgical

Abbreviations: FAP, familial adenomatous polyposis; CT, computed tomography; CEA, carcinoembryonic antigen; CA 19-9, carbohydrate antigen 19-9; PET, positron emission tomography; FDG, fluorodeoxyglucose; SUVmax, maximum standard uptake value.

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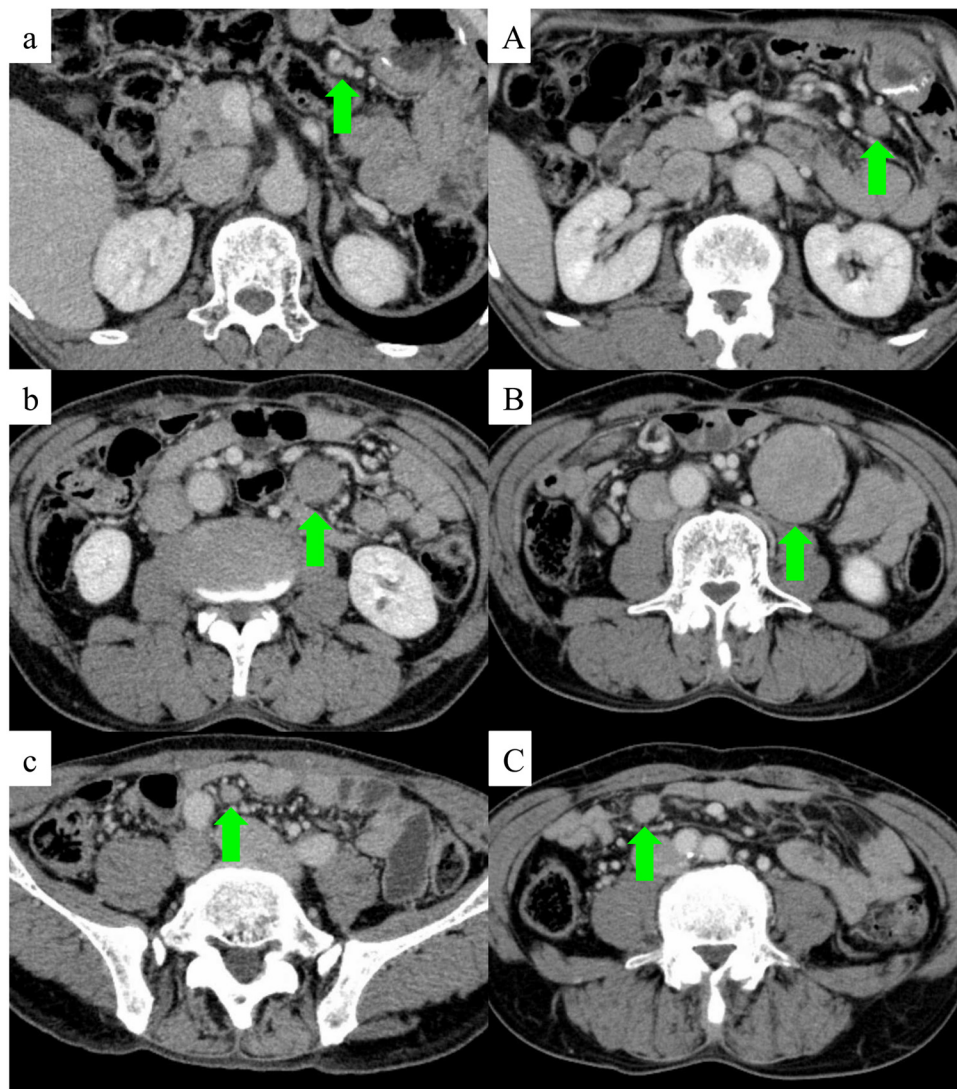


Fig. 1. CT showing 3 enhanced, round-shaped masses mimicking gastric cancer lymph nodes or peritoneal dissemination recurrence that increased in size but not in number. Small characters indicate the CT a year after gastrectomy, and large characters indicate the CT two years after gastrectomy. a: 0.5 cm, b: 2.5 cm, c: 1.0 cm in diameter. A: 1.5 cm, B: 4 cm, C: 1.5 cm in diameter.

resection or chemotherapy. Then, the 2-year postoperative CT indicated slowly growing masses (1.5 cm, 4 cm and 1.5 cm in diameter, as shown in Fig. 1). The tumor marker levels of carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA 19-9) had been within normal limits. Positron emission tomography (PET) revealed fluorodeoxyglucose (FDG) uptake of two masses, and the maximum standard uptake value (SUVmax) of the largest mass was 4.1 (Fig. 2). There were no swollen lymph nodes, and there was neither ascites nor other distant metastases. We firstly suspected relapse of the gastric cancer; however, the recurrence was uncertain, and the possibility of another tumor was suspected because the gastric cancer had been diagnosed at a very early stage, the growth speed of the masses was leisurely and the number of masses was not increased. Therefore, we decided to perform a diagnostic operation to determine the tumor type.

We continuously performed a laparotomy to resect and survey the masses. There were three mesenteric masses near the bowel: a 1.5-cm diameter mass, 20 cm distal from the jejunojejunostomy; a 4-cm diameter mass, 80 cm distant from the jejunojejunostomy, and a 1.5-cm diameter mass, 120 cm distant from the jejunojejunostomy (Fig. 3). We performed partial

small intestinal resections for each mass. Histological examination showed desmoid tumors that were 15 × 12 mm, 40 × 40 mm and 15 × 13 mm (Fig. 4). Immunology testing indicated β -catenin (+). The patient had an uneventful postoperative course. A year after surgery, he was doing well with no evidence of recurrence of the desmoid tumors or the gastric cancer.

3. Discussion

Multiple mesenteric desmoids could emerge after gastrectomy for gastric cancer. Typically, a desmoid tumor is solitary and occurs at the extremities or abdominal wall in premenopausal woman or at the surgical site in patients with FAP [11–13]. It was reported that about 50 percent of all desmoid tumors are intra-abdominal, but cases in patients without FAP accounted for only about 10 percent [1,14]. Although rare, there are some reports of multiple mesenteric desmoids that were difficult to distinguish from the recurrence of malignancy [9,15,16]. We found six cases of intra-abdominal desmoid tumor after gastrectomy for gastric cancer (Table 1) [6–9]. Moreover, two of six cases including our case manifested multiple mesenteric tumors. The diagnostic difficulty would remain unrec-

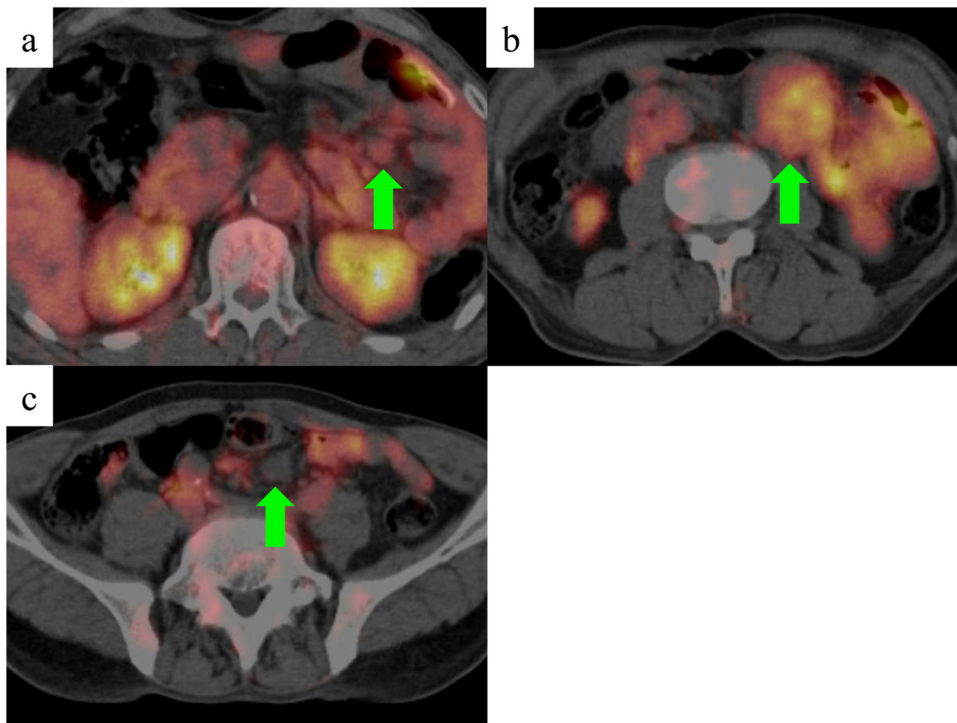


Fig. 2. Positron emission tomography revealing FDG uptake of two masses, although one mass did not take up FDG. a: A small mass with mild uptake of FDG, b: a large mass with uptake of FDG; the maximum standard uptake value of 4.1, c: a small mass without uptake of FDG.

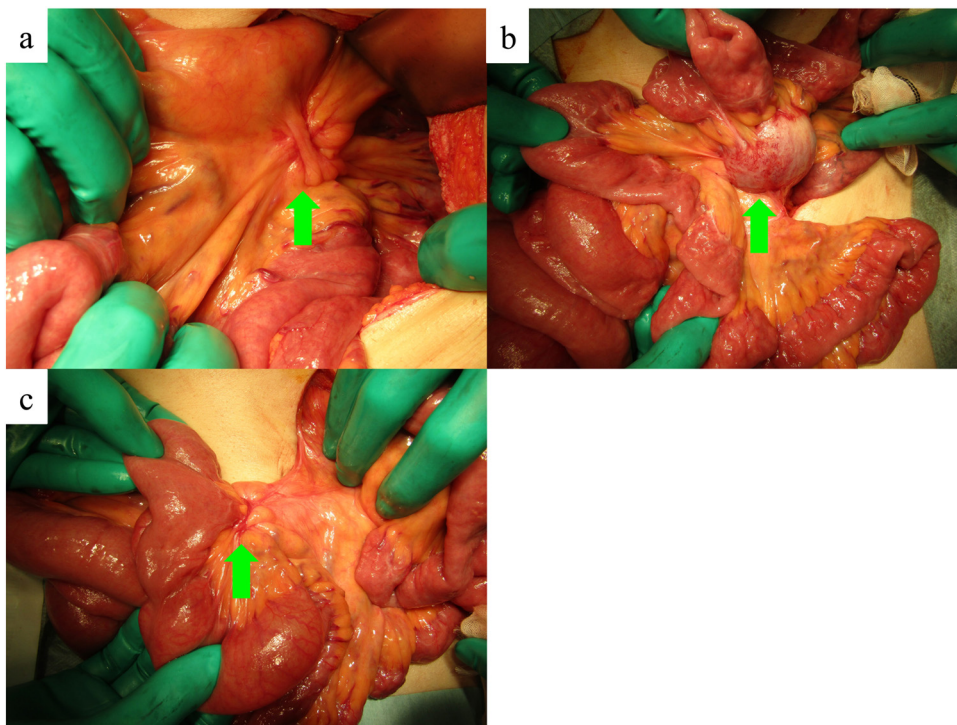


Fig. 3. Intraoperative pictures showing 3 masses of the jejunal mesentery close to the jejunum. a: 3-cm mass mimicking peritoneal dissemination, 20cm from the jejunojejunostomy, b: a 4-cm round mass with a pellicle, 80cm from the jejunojejunostomy, and a 1.5-cm mass mimicking peritoneal dissemination, 120 cm from the jejunojejunostomy.

ognized due to the wrong diagnosis of gastric cancer recurrence, and as matter of fact, in two of six reported cases, chemotherapy had been performed prior to the surgical resection. Therefore, we presumed that there could be more hidden desmoid cases. We

should be aware that desmoid could manifest as multiple mesenteric tumors after gastrectomy for gastric cancer.

Surgical resection of the tumors could be a useful diagnostic and treatment option in this setting. Diagnosing intra-abdominal

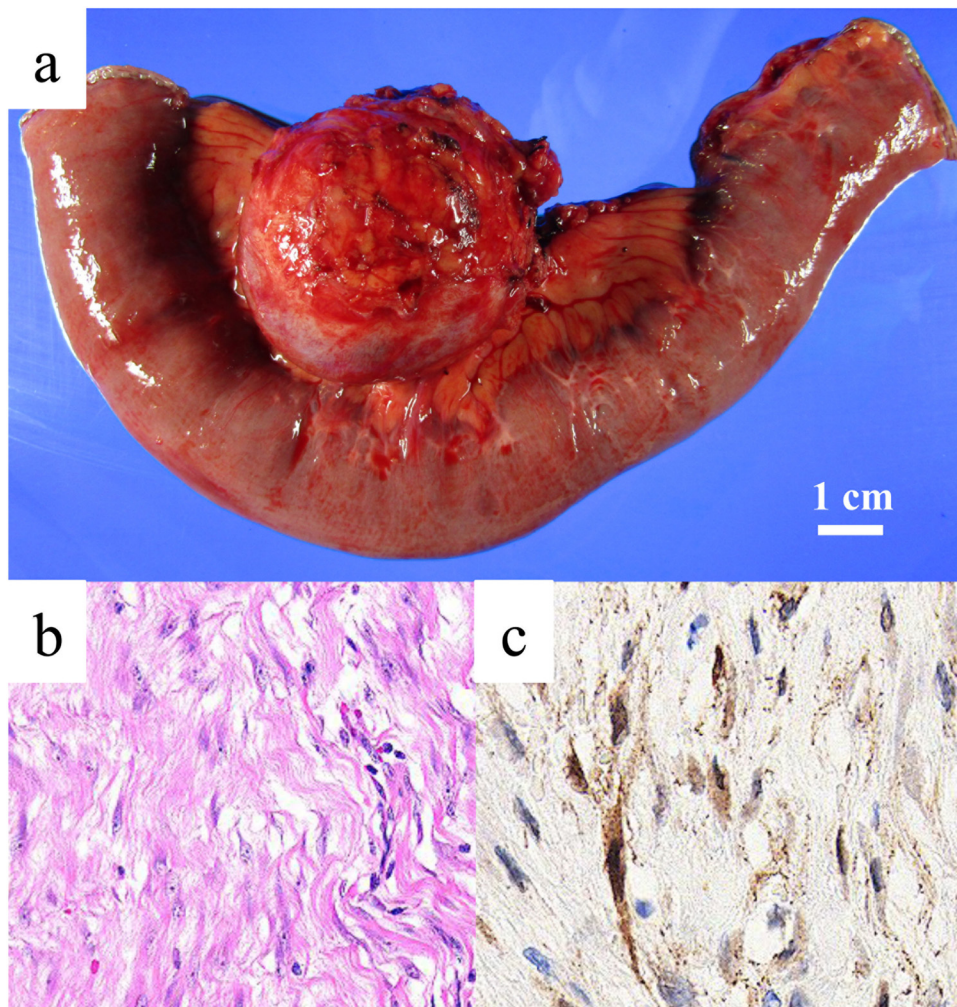


Fig. 4. a: Gross appearance of the largest mesenteric desmoid tumor that was resected with negative surgical margins. b: Histological examination revealing spindle-shaped cells and collagen fibers (hematoxylin and eosin stain). c: Immunostaining showing diffuse β -catenin positive.

desmoids is often difficult. In this case, the desmoids did not have a specific feature on CT, which showed an enhanced round-shaped mass mimicking gastric cancer lymph nodes or peritoneal dissemination recurrence [6–9]. The desmoids had mild uptake of FDG on PET, and the SUVmax did not allow us to make a differential diagnosis between a desmoid and a recurrence of the gastric cancer [8,9]. Tumor marker levels might help us to distinguish a desmoid tumor from the recurrence of gastric cancer. Although CEA is often elevated in a case of gastric cancer recurrence, it is not relevant for desmoid tumors; nevertheless, a case of desmoid tumor with cryptogenic elevation of serum CEA levels was reported [8]. Thus, the diagnosis of desmoid tumors can be established only by histological examination of the biopsy specimen. However, core needle biopsy of an intra-abdominal tumor has a risk of dissemination. Moreover, if a patient has a prior history of malignancy, it may be extremely difficult to differentiate between multiple mesenteric desmoids and disease recurrence. Because desmoid tumors lack metastatic potential, treatment strategies for these tumors are careful observation or local therapies [17–20]. On the other hand, most cancer recurrences would be treated with systemic therapy, but a metastasectomy is hardly indicated, especially in cases with multiple metastatic masses. In terms of the treatment strategy, there is a big difference between these two diagnoses. Therefore, differential diagnosis

is very important, but it remains difficult to distinguish multiple mesenteric desmoid tumors from a cancer recurrence preoperatively. We elucidated that the surgical resection is, if technically possible, a useful option as diagnostic therapy. Further reports should be accumulated about the preoperative differentiation between multiple mesenteric desmoid tumors and disease recurrence.

Although the differential diagnosis of multiple mesenteric desmoid tumors was difficult, it was evident that the treatment plan should be decided based on the diagnostic imaging. In general, initial careful observation seemed to be an acceptable treatment option for asymptomatic desmoids, whereas surgical resection with negative surgical margins was the feasible treatment when it was technically practicable [17–20]. In cases with an antecedent malignancy, however, a recurrence of that cancer should be excluded, especially for the observation strategy. In our case, we considered the possibility of primary mesenteric tumors for the following reasons: the original gastric cancer was early stage; the three masses were on the mesentery, distant from the previous surgical site without regional lymph node swelling; the masses increased in size but not in number; ascites was never seen; and the tumor marker levels had been within normal limits. Additionally, we considered those three tumors resectable on the

Table 1
Desmoid tumors after gastrectomy for gastric cancer.

Author	Age	Sex	Antecedent gastrectomy	Pathology (adenocarcinoma)	Stage	Duration (months)	Size (mm)	SUVmax of PET	Tumor markers	Sites of mesentery	Initial diagnosis	Treatment	Recurrence
Komatsu et al.	50	M	Total	Well	T3N0M0, stage IIA	12	50	*	*	Jejunum*	Recurrence	Chemotherapy > Operation	No
Tamura et al.	73	F	Total	*	T1N0M0, stage IA	24	63	*	Normal	Jejunal pouch	*	Operation	No
Okamura et al.	57	M	Distal	Poorly	T2N0M0, stage IB	27	40	3.9	Elevated	Transverse colon	Recurrence	Chemotherapy > Operation	No
Kim et al.	50	M	Distal	Poorly	T1N1M0, stage IB	24	50, 60	*	*	Jejunum nearby and 100–cm distal from gastrojejunostomy	Recurrence or Benign tumor	Operation	No
Kim et al.	60	M	Total	Mucinous	T2N2M0, stage IIB	23	60	Mild	*	Ileum 30–cm proximal from ileocecal valve	Recurrence	Operation	No
Our case	70	M	Total	Moderately	T1N0M0, stage IA	12	15, 40, 15	4.1	Normal	Jejunum 20–cm distal and 80–cm distal and 120–cm distal from jejunojejunostomy	Recurrence or Benign tumor	Operation	No

PET: positron emission tomography; SUVmax: maximum standard uptake value. *Details were not described.

diagnostic imaging. Taken together, these factors led us to perform surgical resection as diagnostic therapy.

4. Conclusions

Multiple mesenteric desmoids could emerge after gastrectomy for gastric cancer. If a patient has an antecedent history of malignancy, it may be extremely difficult to differentiate between multiple mesenteric desmoids and disease recurrence. In terms of the treatment strategy, there is a big difference between the two diagnoses. We stressed that the surgical resection was, if technically practicable, a useful treatment option as diagnostic therapy.

Conflicts of interest

All authors declare that they have no conflicts of interest.

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Ethical approval

The institutional ethics committee approved the publication of this case report.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Author contribution

All authors contributed to the concept of this case report. Ryu-sei Yamamoto drafted the manuscript. Yasuji Mokuno revised the manuscript. Hideo Matsubara, Hirokazu Kaneko and Sinsuke Iyomasa supervised the writing of the report. All authors participated in interpreting the results and writing the report; all authors approved the final version of the manuscript.

Registration of research studies

This case report is not mandatory for human study.

Guarantor

Iyomasa Shinsuke.

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