Solitary Fibrous Tumor of the Stomach Treated with Laparoscopic and Endoscopic Cooperative Surgery

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A 43-year-old Japanese woman with melena underwent an upper gastrointestinal endoscopy and was preoperatively diagnosed with sarcoma of the stomach. Physical examination revealed no abnormalities. Findings on the upper gastrointestinal endoscopy showed a pedunculated submucosal tumor measuring 17 mm in the antrum. An enhanced computed tomography showed wall thickening in the gastric antrum. The patient underwent a laparoscopic and endoscopic cooperative surgery (LECS) for wedge resection of the stomach. The excised tumor measured $27 \times 20 \times 15$ mm in size. Histopathology showed spindle-shaped cells in the submucosal layer. Immunohistochemistry showed that the tumor was positive for CD34, bcl-2, and MIC-2. The final diagnosis was solitary fibrous tumor (SFT) of the stomach. The postoperative course was uneventful, and no evidence of recurrence was observed at the 8-month follow-up. We report a case of SFT arising from the stomach that was treated with wedge resection by LECS.

Key words: Solitary Fibrous Tumor of the Stomach, Laparoscopic and Endoscopic Cooperative Surgery, Barbed suture

INTRODUCTION

Solitary fibrous tumor (SFT) is a rare neoplasm. In 1931, the first report of an SFT arising from the pleura was presented by Klemperer and Rabin [1]. This tumor has been reported to originate in nonpleural sites, including the peritoneum, pericardium, mediastinum, retroperitoneum, paranasal sinuses, nose, and upper respiratory tract [1].

To the best of our knowledge, this report presents a description of the first laparoscopic surgery of an SFT arising from the stomach. Here we report a rare case of SFT that presented as a gastric submucosal tumor.

CASE REPORT

A 43-year-old Japanese woman with melena underwent an upper gastrointestinal endoscopy. A physical examination revealed no abnormalities, and laboratory data on admission were within the normal range.

An endoscopic examination showed a pedunculated submucosal tumor in the anterior wall of the antrum (Fig. 1), and an endoscopic ultrasonography confirmed the submucosal tumor. An endoscopic biopsy revealed a sarcoma on histopathological findings. Upper gastrointestinal series showed a pedunculated tumor (17 mm in size) of the gastric antrum infiltrating the duodenum (Fig. 2). An enhanced computed tomography (CT) of the abdomen showed slight wall thickening in the antrum (Fig. 3). No distant metastatic lesions or lymph node metastasis was seen in the abdominal cavity. A positron emission tomography–computed tomography revealed slight accumulation in the tumor, and the maximum standardized uptake value was 3.6. A preoperative diagnosis of sarcoma of the stomach was made.

Wedge resection of the stomach by laparoscopic and endoscopic cooperative surgery (LECS) was performed. Under general and epidural anesthesia, the patient was placed in a supine and open-leg position. The abdomen was entered through an umbilical incision (approximately 4 cm) and using a Lap protector with EZ access (Hakko Medical, Japan), a 5-mm camera port and two parallel 5-mm working ports were placed through the EZ access, together with a plus 1 12-mm port on the left side of the abdomen (Endopath Xcel trocar, Ethicon Endo-Surgery, USA). Through an intraoperative endoscopy, surgical margins around the tumor were determined and marked using a needle knife (Fig. 4). After a penetrating incision of the serosa with a needle knife, full-thickness laparoscopic dissection by THUNDERBEAT (Olympus Medical Systems, Japan) was completed. The stomach was hand sewn by V-LOCTM (Covidien, USA) and closed under laparoscopic guidance. The operation lasted for 281 min, and additional blood loss was 40 mL.

The tumor size of the resected specimen measured $27 \times 20 \times 15$ mm in size (Fig. 5). Histopathology revealed spindle-shaped cells in the submucosal layer (Fig. 6A, 6B). Immunohistochemistry showed that the tumor strongly stained positive for CD34 (Fig. 6C), bcl-2 (Fig. 6D), and MIC-2 and negative for SMA, c-KIT, DOG-1, ALK-1, and ALK-EML4. The MIB-1 labeling index was 20%. The final diagnosis was SFT of the stomach.

The postoperative course was uneventful. The patient's melena resolved after removal of the tumor, and no evidence of recurrence was observed at the 8-month follow-up.

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Fig. 1 Upper gastrointestinal endoscopy showed a submucosal tumor in the anterior wall of the prepylorus. The mucosal surface of the tumor showed slightly redness.

Fig. 2 Enhanced contrast radiography revealed a submucosal tumor of the anterior wall of the antrum infiltrating the duodenum.

Fig. 3 Abdominal enhanced Computed Tomography (CT) demonstrated slight wall thickening in the anterior aspect of the antrum (white triangle). No evidence of metastatic lesion.



Fig. 4 The state of intraoperative endoscopy. Intraoperative endoscopy was conducted, and the surgical margin around the tumor was defined using a needle knife. (The left side is a monitor of the endoscopes. The right side is a monitor of the laparoscopes.)



Fig. 5 Gross pathological appearance of resected specimen was a soft lobulated mass, measuring $27 \times 20 \times 15$ mm in size, mass involving submucosa layer.



Fig. 6 Histopathological findings by Hematoxylin and Eosin (H.E.). (A) Histopathological features were consistent with spindle-shaped cells (original magnification, ×200, H-E stain). (B) High power photomicrograph (original magnification, ×400, H-E stain). (C) The tumor cells strongly stained imunoreactivity positive for CD34 (×200). (D) The tumor cells stained positive for Bcl-2 (×200).

DISCUSSION

Here we describe a case of SFT of the stomach successfully treated with wedge resection by laparoscopic surgery.

Since the first report of a pleural SFT in 1931 [1], SFTs arising from other nonpleural sites have been reported in the literature. This tumor has been reported to extrapleural lesions including the peritoneum, pericardium, mediastinum, retroperitoneum, paranasal sinuses, nose, and upper respiratory tract [1]. An SFT arising from the stomach is an extremely rare neoplasm [3, 4].

Accurate diagnosis preoperatively of SFT is difficult, due to non-characteristic clinical symptoms and physical examination findings. In our case, the preoperative diagnosis was a sarcoma by the biopsy specimen with endoscope. There was no report about preoperative diagnosis of SFT arising stomach.

SFTs of the pleura typically showed a well-defined, lobular, solitary nodule or mass. Small SFTs show homogeneous, well-defined, non-invasive, lobular, soft tissue masses [4, 5]. On CT scan findings, this tumor typically appears as well delineated, smooth, lobulated soft tissue masses. It may occasionally include calcification to be scattered [2].

In case of SFT arise at other extrapleural lesion, differential diagnosis from other soft tissue tumors was difficult. Differential diagnosis of an SFT of stomach includes gastrointestinal stromal tumor (GIST), fibromatosis, schwannoma, leiomyoma, leiomyosarcoma, inflammatory myofibroblastic tumor, fibrosarcoma, malignant fibrous histiocytoma, hemangiopericytoma, synovial sarcoma, and malignant mesenchymoma [3, 4]. Differential diagnosis of mesenchymal tumor in the stomach using only CT imaging is difficult. It is useful ideally to make a histopathologic diagnosis before surgery.

Commonly, SFT are diagnosed pathologicaly by the presence of a collagenous matrix with arrays of spindle cells exhibiting diffuse CD34, bcl-2, vimentin positivity, and S100, actin, and keratin negativity on immunohistochemistory [3, 5, 6]. In this case, an SFT was diagnosed on pathological examination by the presence of spindle cells that exhibited diffuse CD34, bcl-2, and MIC-2 positivity and SMA, c-KIT, DOG-1, and desmin negativity on immunohistochemical staining. ALK-1, and ALK-EML4 which became positive in inflammatory myofibroblastic tumor was negative.

Most SFTs are histopathologically benign, but up to 20% of all SFTs may be malignant [7]. Malignant SFTs have high cellularity, high mitotic activity (> 4/10 HPF), pleomorphism, hemorrhage, necrosis [8] fibro-sarcoma-like and synoval sarcoma-like growth patterns and tumor size greater than 10 cm [8].

With regard to treatment of SFT, surgical resection is more effective. Recently, LECS for GIST was developed by Hiki *et al.* and reported in 2008 [9]. LECS is indicated in gastric tumors (e.g., GIST, gastric carcinoid tumor, gastric cancer, etc.) [10]. To the best of our knowledge, there are no reports in the literature of an SFT arising from the stomach treated by laparoscopic surgery, particularly LECS. This is the first report of an SFT arising from the stomach treated by LECS.

The prognosis in SFT is usually good, when a tumor is completely removed surgically. The most reliable prognostic indicator appeared to be the gross appearance of the tumors and resectability. But a recurrence case has been reported 7 years after surgery [4]. Follow-up radiological examination is necessary at least annually to check for recurrence [4]. In the present case, the tumor was pathologically benign and completely resected, with no recurrence 8 months after surgery. However, careful observation is necessary during a long period in this case.

In conclusion, we report a case of SFT arising in the stomach that was treated with wedge resection by LECS, along with a review of the literature.

STATEMENT OF CONFLICT

The authors have no conflicts of interest or financial disclosures.

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