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Teaching case

Primary synovial sarcoma of the stomach—A case report and review of the literature

Shu Sahara[a], Yoshiro Otsuki[b], Yuki Egawa[b], Shin-ichi Shimizu[b], Yashiro Yoshizawa[a], Yoshisuke Hosoda[a], Kazufumi Suzuki[c], Yoshihiko Sato[a], Hiroshi Kobayashi[b,d,*]

[a] Department of Gastroenterology, Seirei Hamamatsu General Hospital, Hamamatsu, Japan
[b] Department of Pathology, Seirei Hamamatsu General Hospital, Hamamatsu, Japan
[c] Department of Digestive Surgery, Seirei Hamamatsu General Hospital, Hamamatsu, Japan
[d] Department of Pathology, Tachikawa General Hospital, Nagaoka, Japan

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ABSTRACT

Synovial sarcoma (SS) is a mesenchymal spindle cell tumor which displays variable epithelial differentiation. It commonly arises around the major joints or tendon sheaths in young adults, but is not commonly seen in the stomach. We experienced a case of primary gastric SS. The patient is a 22-year-old male, who presented with epigastric pain. Upper endoscopy showed an ulcer of 25 mm in diameter with marginal elevation on the posterior mid-gastric body. Biopsy of the ulcer base showed monotonous proliferation of small spindle-shaped cells on HE-stain. On immunohistochemical staining, these cells were positively stained with vimentin, cytokeratin, epithelial membrane antigen, and CD99, but were negative for KIT, CD34, desmin, and S-100 protein. These findings were compatible with SS of monophasic type. Diagnosis of primary gastric SS was made because there were no other primary lesions, nor metastatic lesions. The wedge resection was performed. Reverse transcriptase polymerase chain reaction (RT-PCR), using the RNA from frozen neoplastic tissue of the resected specimen, detected a fusion gene called SYT-SSX1, specific for SS. Though SS arising in the stomach is rare, it should be considered in the differential diagnosis of KIT-negative gastric spindle cell tumor.

Introduction

Synovial sarcoma (SS) is a mesenchymal spindle cell tumor which displays variable epithelial differentiation, and commonly arises around the major joints or tendon sheaths in young adults. It is named so because its morphology resembles the developing synovial tissue, but its origin is still unknown [8]. Recently, SSs that originate from the digestive tract, including the esophagus, and stomach have been reported, but these are still rare, and the preoperative diagnosis is often difficult to make [1,2,4,5,9,12,17]. However, like SS of soft tissue origin, a specific chromosomal translocation and expression of the fusion gene are also seen in SS of the digestive tract [4,6,12]. In this case, we could suspect primary gastric SS preoperatively by histological features and ancillary staining, and made a definite diagnosis with surgical specimens using molecular pathology.

Case report

The patient was a 22-year-old male who presented at Seirei Hamamatsu General Hospital with intermittent epigastric pain that had continued for one year. Blood testing, including the tumor marker, showed no abnormality. Upper endoscopy showed a well-demarcated ulcer lesion of 25 mm in diameter with marginal elevation on the posterior mid-gastric body (Fig. 1). Endoscopic ultrasound showed a low echoic mass (17 mm diameter, 6 mm thickness) occupying mainly the upper part of submucosal layer (Fig. 2). This lesion was strongly stained with enhanced CT scan, but there was no other site of indicating primary or metastatic lesions (Fig. 3). There was no accumulation of FDG-PET signal on the stomach wall. The differential diagnoses were malignant lymphoma, gastric carcinoma, and mesenchymal tumors occurring in the submucosa.

On endoscopy, 9 biopsy specimens were obtained from the ulcer base. Histologically, 5 specimens showed monotonous proliferation of spindle-shaped cells mainly in the submucosal area (Fig. 4). These cells were relatively small and uniform in size with fusiform or ovoid nuclei and a small or inconspicuous nucleolus. The cytoplasm was scant and the cell borders were indistinct. These findings indicated the possibility of mesenchymal tumors.

* Corresponding author at: Department of Pathology, Seirei Hamamatsu General Hospital, 2-12-12 Sumiyoshi, Naka-ku, Hamamatsu, Japan. Tel.: +81 53 474 2222; fax: +81 53 474 1228.
E-mail address: h-kobayashi@tatikawa.or.jp (H. Kobayashi).

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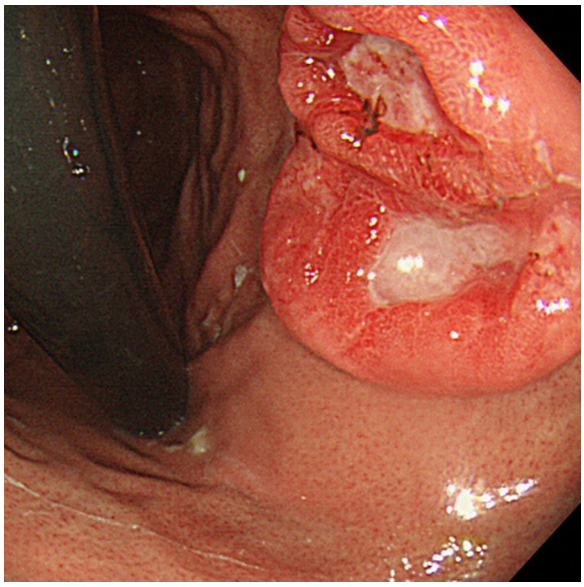

Fig. 1. Upper endoscopy showed a well-demarcated ulcer lesion with marginal elevation on the posterior mid-gastric body.

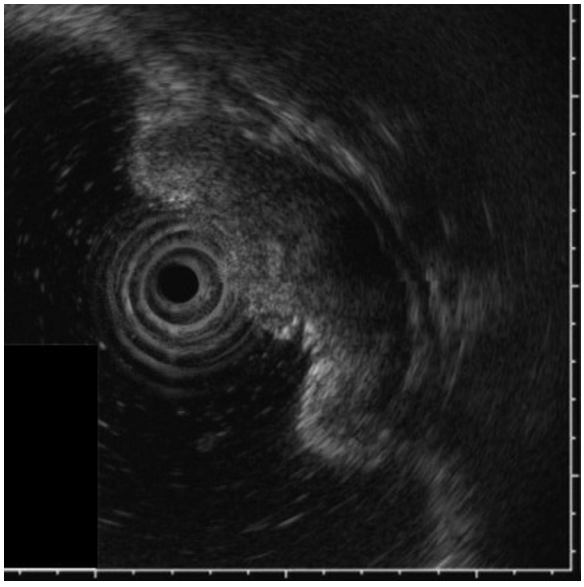

Fig. 2. Endoscopic ultrasound finding shows a low echoic mass occupying the upper part of submucosal layer.

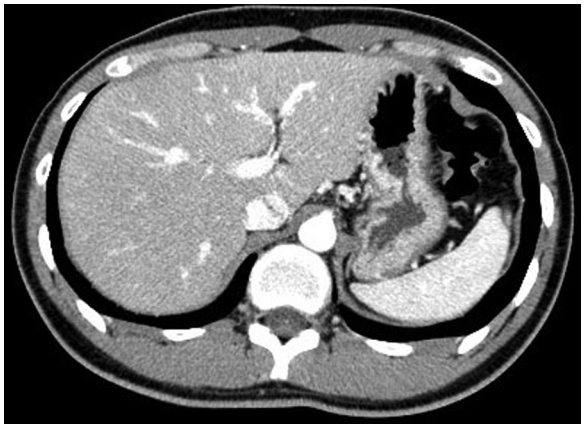

Fig. 3. CT scan finding. Enhanced CT shows the strongly stained lesion on the posterior gastric body.

Immunohistochemical staining was performed using Ventana NX system (Ventana Medical Systems, AZ, USA). These methods are summarized in reference [16]. The antibodies, their sources, their dilution, and the method used for antigen retrieval are listed in Table 1. Immunohistochemically, the tumor was negative for KIT, CD34, desmin, and S-100, while it was diffusely positive for CD99, bcl-2, and focally positive for pan-cytokeratin and epithelial membrane antigen (EMA) (Fig. 5). Rearrangement of the SYT gene (18q11.2) was detected by dual-color break-apart fluorescent in situ hybridization analysis of the SYT locus on an interphase cell nucleus (data not shown). Accordingly, SS was strongly suspected. The wedge resection was performed.

Grossly, the resected specimen exhibited that the tumor formed a luminal side lesion, measuring 25 mm × 25 mm in size. The luminal surface of the lesion exhibited an irregular ulceration, measuring 14 mm × 13 mm in size (Fig. 6a). On cut section, the tumor showed a whitish-gray solid appearance with focal hemorrhage, and mainly involved both the mucosa and the submucosa with focal involvement of the muscularis mucosa (Fig. 6b).

The histology of the resected tumor was almost the same as that of the gastric biopsy. Moreover, it showed that the tumor cells densely proliferated in whorled or fascicular arrangement (Fig. 7). The stromal collagen sometimes intermingled with the spindled tumor cells. The collagen was always thin and wiry. The average mitotic figures were 14 per 10 high power fields. There was no epithelial structure in this tumor. The immunohistochemical results of the resected tumor were almost the same as those of the biopsy specimen.

SYT-SSX1 fusion transcripts were detected using reverse transcription (RT)-PCR method (Fig. 8). Total RNA was extracted from frozen neoplastic tissue of the resected specimen.

Table 1
Panel of antibodies applied in immunohistochemical staining.

Anti-	Clone	Source	Dilution	Retrieval
Pan-cytokeratin	AE1/AE3	DAKO, Glostrup, Denmark	1:100	PK
EMA	E29	DAKO, Glostrup, Denmark	1:2[a]	–
Desmin	D33	DAKO, Glostrup, Denmark	1:200	–
S-100 protein	Polyclonal	DAKO, Glostrup, Denmark	1:2000	–
KIT	Polyclonal	DAKO, Glostrup, Denmark	1:400	Boil
CD34	QBEnd/10	Novocastra Laboratories, Newcastle upon Tyne, UK	1:50	Boil
CD99	12E7	DAKO, Glostrup, Denmark	1:50	–
bcl2	124	Scytec Laboratories, Logan, UT, USA	1:80	Boil

[a] Antibodies were already diluted for ready to use by the company.