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CASE REPORT

Gastric carcinoma originating from the heterotopic submucosal gastric gland treated by laparoscopy and endoscopy cooperative surgery

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Abstract

Gastric carcinoma is derived from epithelial cells in the gastric mucosa. We reported an extremely rare case of submucosal gastric carcinoma originating from the heterotopic submucosal gastric gland (HSG) that was safely diagnosed by laparoscopy and endoscopy cooperative surgery (LECS). A 66-year-old man underwent gastrointestinal endoscopy, which detected a submucosal tumor (SMT) of 1.5 cm in diameter on the lesser-anterior wall of the upper gastric body. The tumor could not be diagnosed histologically, even by endoscopic ultrasound-guided fine-needle aspiration biopsy. Local resection by LECS was performed to confirm a diagnosis. Pathologically, the tumor was an intra-submucosal well differentiated adenocarcinoma invading 5000 μm into

the submucosal layer. The resected tumor had negative lateral and vertical margins. Based on the Japanese treatment guidelines, additional laparoscopic proximal gastrectomy was curatively performed. LECS is a less invasive and safer approach for the diagnosis of SMT, even in submucosal gastric carcinoma originating from the HSG.

Key words: Heterotopic submucosal gland; Laparoscopy and endoscopy cooperative surgery; Gastric carcinoma; Gastric submucosal tumor; Less invasive treatment

Core tip: This report describes the rare case of a submucosal gastric carcinoma originating from the heterotopic submucosal gastric gland (HSG) that was safely diagnosed by laparoscopy and endoscopy cooperative surgery (LECS). LECS is a less invasive and safer approach for the diagnosis of submucosal tumor, even in submucosal gastric carcinoma originating from the HSG.

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INTRODUCTION

Gastric carcinoma is commonly derived from epithelial cells in the gastric mucosa and is very rarely initially diagnosed as a submucosal tumor (SMT). We herein presented a case of submucosal gastric carcinoma originating from the heterotopic submucosal gastric gland (HSG) that was safely diagnosed by laparoscopy and endoscopy cooperative surgery (LECS) and treated by subsequent laparoscopic gastrectomy with D1+ lymphadenectomy. We reviewed the clinical features of this rare tumor and selected successful decision-making using the LECS technique.

CASE REPORT

Patient

The patient was a 66-year-old man who underwent upper endoscopy in a medical checkup, which showed a SMT on the upper gastric body. The patient was referred to the hospital for diagnosis and treatment. Endoscopic re-examination detected a SMT of 15 mm in diameter on the anterior wall of the upper gastric body. The tumor did not have a depression or ulceration (Figure 1A). The results of endoscopic biopsy from the gastric mucosa on the tumor were chronic gastritis with no evidence of malignancy. Barium gastrography showed a smooth elevated lesion of 2 cm in diameter on the anterior wall of the upper gastric body near the esophago-gastric junction (Figure 1B). Computed tomography revealed a 15-mm low density area with calcification in the anterior wall of the upper gastric body and no lymph node or distant metastasis (Figure 1C). Endoscopic ultrasound (EUS) showed an 11.2 mm × 13.5 mm SMT that was derived from the third layer of the gastric wall as a heterogeneous lesion with a mixture of a high echoic lesion, low echoic lesion, and calcification (Figure 1D). The tumor could not be diagnosed histologically, even by EUS-guided fine-needle aspiration biopsy at multiple sites. LECS for gastric local resection was selected as decision-making for a pathological diagnosis and safe removal.

LECS for the SMT

Observations in the abdominal cavity by laparoscopy confirmed no distant or nodal metastasis. The SMT was endoscopically detected on the anterior wall of the lesser curvature of the upper gastric body, but not by laparoscopy. To avoid bleeding, the peripheral branches of the left gastric artery near the tumor were coagulated using a laparoscopic ultrasonically activated device. Endoscopic submucosal resection around the tumor was performed using the endoscopic submucosal dissection technique and seromuscular dissection was performed around the tumor along the line of submucosal resection. The incisional line in the stomach was closed using a laparoscopic stapling device. The resected tumor had negative lateral and vertical margins with normal mucosa (Figure 2A). A pathological examination confirmed that the tumor was a SMT that invaded 5000 μm into the submucosal layer, measured 20 mm × 11 mm × 6 mm, and was a well differentiated adenocarcinoma (Figure 2B). Dilated gastric glands were detected in the submucosal layer (Figure 2C). There was no lymphovascular invasion. Immunohistochemical staining revealed the positive expression of MUC5AC and MUC6, indicating differentiation into the pyloric glands (Figure 2D).

Eighty-four days after LECS, additional laparoscopic proximal gastrectomy with D1+ lymphadenectomy was performed based on the Japanese Gastric Cancer Treatment Guidelines[1]. A pathological examination confirmed no residual tumor cells or lymph node metastasis. The postoperative course was uneventful and the patient is alive without recurrence 1 year after surgery.

DISCUSSION

HSG shows that cystic dilated gastric glands exist in the gastric submucosal layer and has been recognized as a benign condition occurring as a result of repeated mucosal damage[2,3]. HSG was previously described

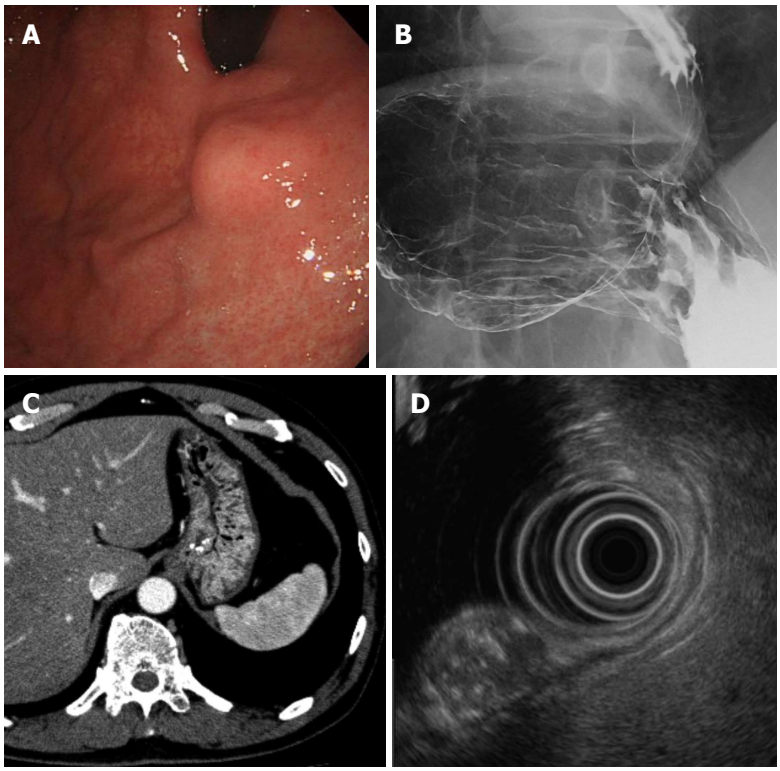

Figure 1 Results of pre-operative examinations. A: Endoscopic findings showing a submucosal lesion of 15 mm in diameter on the anterior wall of the upper gastric body near the esophago-gastric junction. The surface was covered with normal gastric mucosa; B: Barium gastrography showed a smooth elevated lesion of 2 cm in diameter on the anterior wall of the upper gastric body near the esophago-gastric junction; C: Computed tomography revealed a 15-mm submucosal low density area with calcification in the anterior wall of the upper gastric body. No lymph node or distant metastasis was detected; D: Endoscopic ultrasound showed an 11.2 mm × 13.5 mm submucosal tumor derived from the third layer of the gastric wall as a heterogeneous lesion with a mixture of a high echoic lesion, low echoic lesion, and calcification.

Table 1 Previous case reports of gastric carcinoma originating from the heterotopic submucosal gastric gland

Total number of reported cases		*n* 17	(%)
Age		64.1 (45-81)	
Sex	Male	11	65
	Female	6	35
Location	Upper	4	24
	Middle	8	47
	Lower	5	29
Size (mm)		20.5 (8-50)	
Ulceration or depression	Present	13	76
	Absent	4	24
Histological type	Well differentiated	16	94
	Unknown	1	6
Depth of invasion	m	1	6
	sm	14	82
	T2 or more	2	12
Diagnosis by biopsy	Present	6	35
	Absent	11	35
EUS-FNA	Present	2	12
	Absent	15	88
Treatment	EMR	1	6
	EMR and surgical resection	3	18
	Surgical resection	12	71
	LECS + surgical resection	1	6

EUS-FNA: Endoscopic ultrasound-guided fine-needle aspiration biopsy; EMR: Endoscopic mucosal resection; LECS: Laparoscopy and endoscopy cooperative surgery. Note: Ref. [2,6-18].

as a para-cancerous lesion found in 4% of resected specimen from the stomachs of patients with gastric carcinoma, and multiple cancers have been detected in 30% of specimens of gastric carcinoma associated with HSG[4]. However, little is known about the carcinogenesis of HSG itself. Kim *et al*[5] described two cases of early gastric carcinoma arising from HSG that were treated by laparoscopic gastric wedge resection. To the best of our knowledge, there have been no other studies in English concerning gastric carcinoma originating from HSG.

Table 1 shows a summary of 17 previously reported cases, including cases in Japan and our case. Gastric carcinoma originating from HSG occurred more frequently in males and in the middle area of the stomach. Regarding histological findings, the well differentiated type was more common. A study has not yet been conducted on lymph node metastasis from gastric carcinoma originating from HSG. This summary showed that more than 65% of patients could not be histologically diagnosed by biopsy and FNA using EUS before resection.

The recent development of endoscopic and laparoscopic techniques has allowed for less invasive diagnoses and treatments. LECS is a novel and excellent approach for local gastric resection, and was developed by Hiki *et al*[19] as an alternative strategy to laparoscopic wedge resection for gastric SMT. The feasibility and safety of this procedure for gastric SMT have been demonstrated

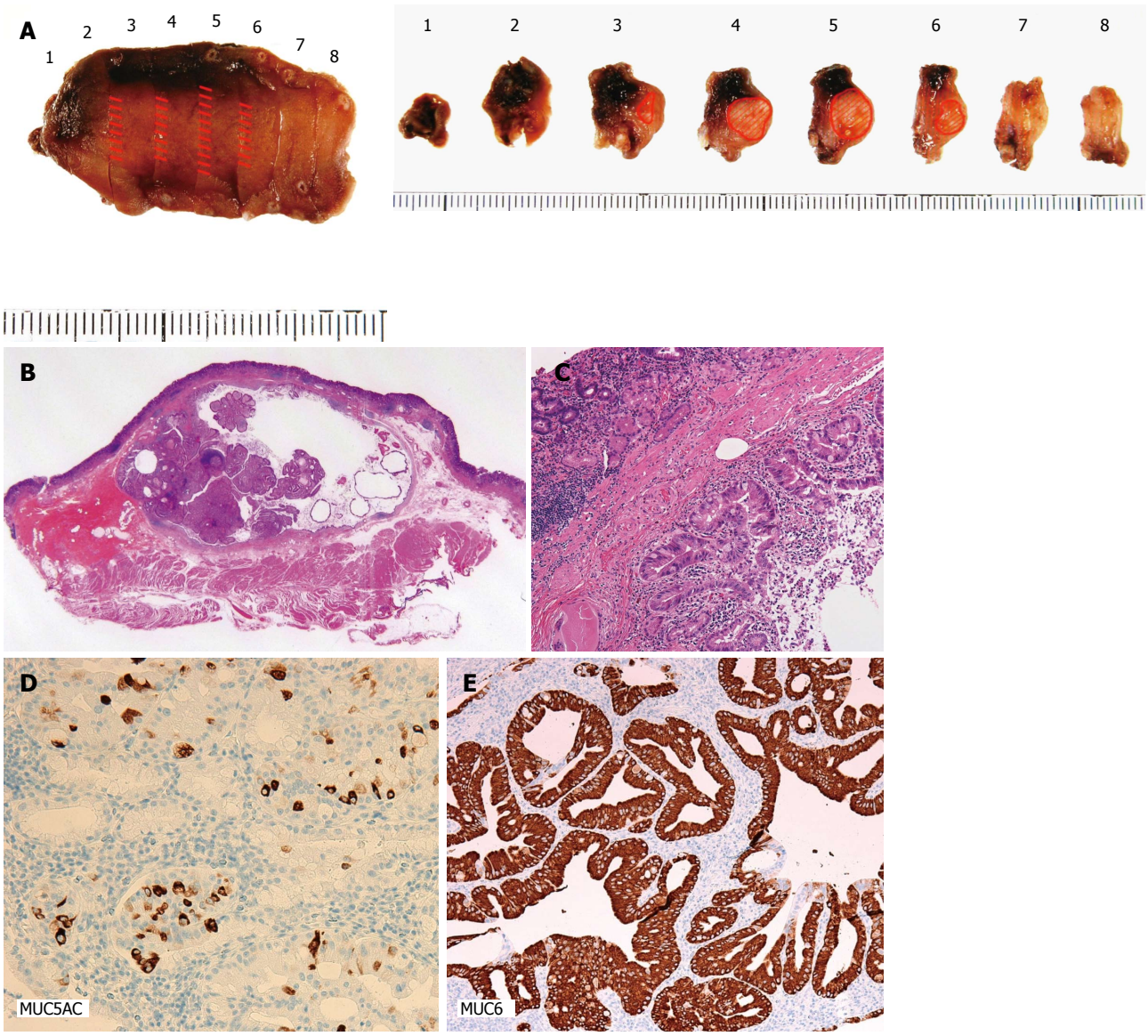

Figure 2 Results of histopathological examinations. A: The resected specimen had negative lateral and vertical margins with normal mucosa; B: A pathological examination confirmed that the tumor was intrasubmucosal (the depth of invasion into the submucosal layer was 5000 μm), measured 20 mm × 11 mm × 6 mm, and was a well differentiated adenocarcinoma; C: Dilated gastric glands were found in the submucosal layer. There was no lymphovascular invasion; D: An immunostaining method showed MUC5AC (+) and MUC6 (+), indicating differentiation into the pyloric glands.

in several studies[20-22]. LECS is now being applied to the treatment of early gastric cancer[23]. The most critical issue associated with its application to gastric cancer is the dissemination of cancer cells into the peritoneal cavity during surgery. Therefore, several methods have been investigated for LECS[24-26]. LECS is a promising approach for the diagnosis of SMT, even in gastric carcinoma originating from HSG.

COMMENTS

Case characteristics

A 66-year-old man who underwent upper endoscopy in a medical checkup, which showed a submucosal tumor (SMT) on the upper gastric body.

Clinical diagnosis

The presented patients had submucosal gastric tumor that could not be diagnosed histologically by endoscopic biopsy.

Differential diagnosis

Gastrointestinal stromal tumor, early gastric tumor, smooth muscle tumor.

Laboratory diagnosis

There were no abnormal findings in laboratory examinations including tumor markers.

Imaging diagnosis

Endoscopic ultrasound and computed tomography showed that the tumor was derived from the third layer of the gastric wall.

Pathological diagnosis

Pathological examination confirmed that the tumor was an intra submucosal tumor that was a well differentiated adenocarcinoma.

Treatment

Laparoscopy and endoscopy cooperative surgery (LECS) for gastric local resection was selected as decision-making for a pathological diagnosis and safe removal.

Term explanation

LECS: Laparoscopy and endoscopy cooperative surgery; HSG: Heterotopic submucosal gastric gland.

Experiences and lessons

Gastric carcinoma originating from the HSG forms a submucosal gastric tumor and is often difficult to diagnose by endoscopic biopsy. If unable to deny malignant disease, resection of the tumor should be considered.

Peer-review

This manuscript described a rare case of submucosal gastric carcinoma originating from the HSG and the authors also described the treatment of the carcinoma by LECS.

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