International Journal of Surgery Case Reports 131 (2025) 111357

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

A case of gastric glomus tumor with metachronous liver metastasis after laparoscopic partial gastrectomy

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ARTICLE INFO

Keywords:
Gastric glomus tumor
Laparoscopic partial gastrectomy
Liver metastasis

ABSTRACT

Introduction and importance: Gastric glomus tumor (GGT) is a rare submucosal mesenchymal tumor that is typically benign. However, GGT recurrence or metastasis has been reported.
Case presentation: A 55-year-old man was referred to our hospital for gastric submucosal tumor (SMT) examination. Esophagogastroduodenoscopy revealed a 15 mm SMT in the anterior wall of the gastric antrum. Endoscopic ultrasonography demonstrated a hypoechoic solid mass that invaded the proper muscle layer. In contrast-enhanced abdominal computed tomography (CT), the anterior wall of the gastric antrum was thickened. Our provisional diagnosis was gastric leiomyoma. As the tumor grew rapidly, we performed laparoscopic partial gastrectomy. Histopathology revealed solid proliferation of tumor cells with oval-shaped nuclei. Immunohistochemically, the tumor cells were positive for alpha-smooth muscle actin and vimentin but negative for c-kit, CD34, desmin, and S-100. The MIB-1 labeling index was approximately 60 %. We then diagnosed the patient with GGT. After 2 years and 6 months, abdominal CT revealed metastatic lesions over 40 mm in diameter, with ring enhancement seen in segment 8 of the liver and another liver metastatic lesions 15 mm observed in segment 5/6. After being diagnosed with liver metastases of the GGT, the patient continued to receive chemotherapy for 26 months and was in good general condition.
Clinical discussion: Laparoscopic partial gastrectomy was performed for a rare GGT, revealing a case of asynchronous liver metastasis.
Conclusion: We managed a case of asynchronous liver metastasis of GGT.

1. Background

Glomus tumor is a mesenchymal neoplasm arising from the glomus body, which consists of modified smooth muscle cells and exhibits arteriovenous temperature–regulating anastomosis. It occurs mainly in the extremities and peripheral soft tissues and rarely in the gastrointestinal tract [1]. Talijeva et al. first reported gastric glomus tumor (GGT) in 1928 [2]. However, GGT still has no specific clinical, radiological, or endoscopic features for its diagnosis. Currently, GGT is confirmed only by histopathology and immunohistochemistry. The presence of nuclear atypia and mitotic activity indicates malignancy. Most GGTs are benign, with only few reported cases of liver metastasis. Herein, we report a case of GGT that was found to progress to liver metastasis after undergoing laparoscopic partial gastrectomy.

2. Methods

This work has been reported in line with the SCARE criteria [3].

3. Case presentation

A 55-year-old male who was initially diagnosed with gastric submucosal tumor (SMT) following a medical check-up was referred to our hospital for examination. His past medical history included *Helicobacter pylori* gastritis. Laboratory examinations, including tumor marker evaluations, disclosed no abnormalities. Esophagogastroduodenoscopy

Abbreviations: GGT, Gastric glomus tumor; SMT, Submucosal tumor; EGD, Esophagogastroduodenoscopy; EUS, Endoscopic ultrasonography; CT, Computed tomography; GIST, Gastrointestinal stromal tumor; FNA, Fine-needle aspiration.

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https://doi.org/10.1016/j.ijscr.2025.111357
Received 22 March 2025; Accepted 21 April 2025
Available online 23 April 2025

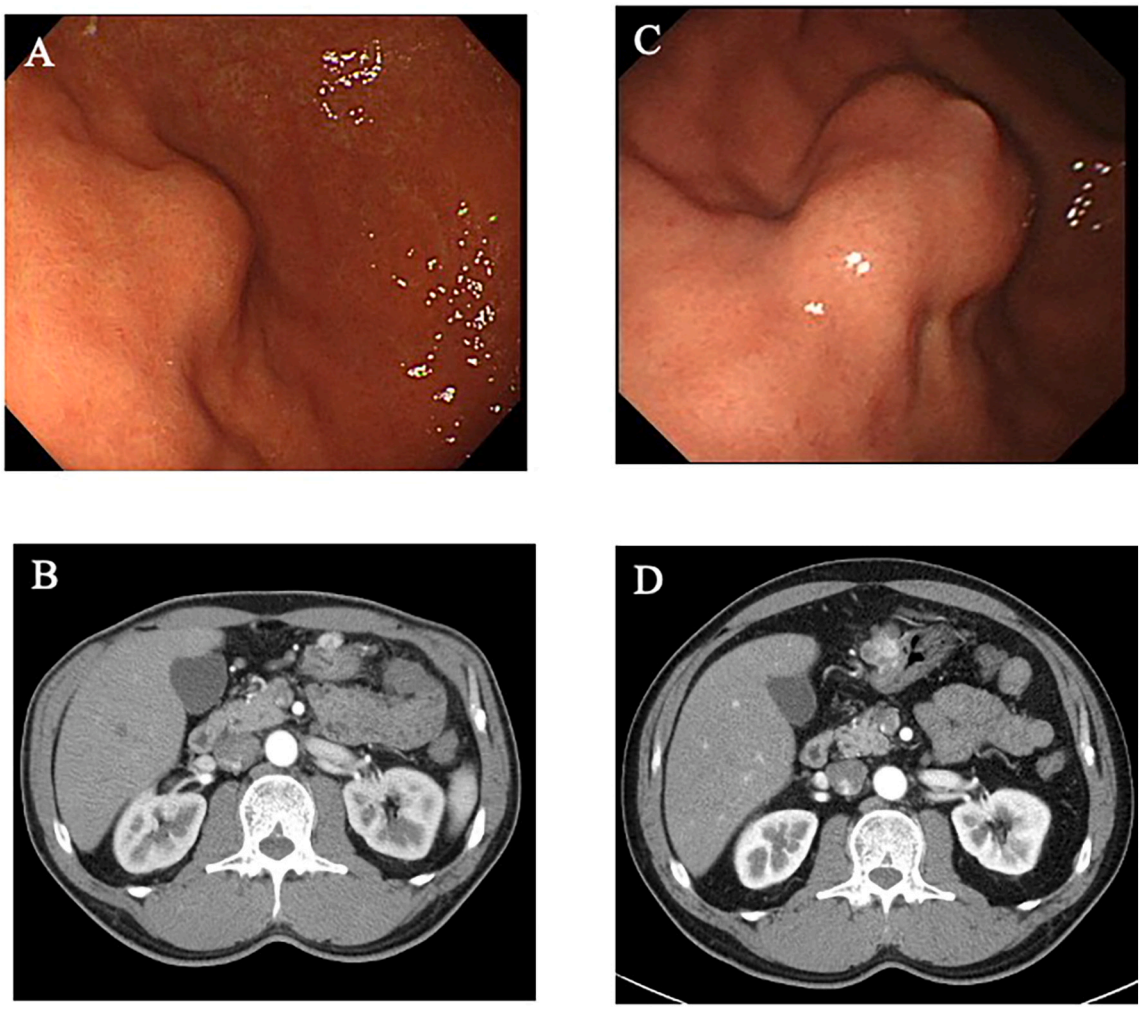

Fig. 1. (A) Esophagogastroduodenoscopy (EGD) revealed a 15 mm submucosal tumor in the anterior wall of the gastric antrum. (B) Contrast-enhanced abdominal CT detected thickening of the anterior wall of the gastric antrum. (C) Seven months after, follow-up EGD revealed an increase in lesion size from 15 mm to 30 mm. (D) Contrast-enhanced abdominal CT detected a 30 mm mass arising from the greater curvature of the stomach.

(EGD) revealed a 15 mm SMT in the anterior wall of the gastric antrum (Fig. 1A). Endoscopic ultrasonography (EUS) detected a hypoechoic solid mass reaching the proper muscle layer. In contrast-enhanced abdominal computed tomography (CT), the anterior wall of the gastric antrum thickened (Fig. 1B). Using 18-fluorodeoxyglucose, we found no significant radioactivity in the stomach. Our provisional diagnosis was gastric leiomyoma.

Seven months later, follow-up EGD revealed that the lesion size increased from 15 mm to 30 mm (Fig. 1C). Contrast-enhanced abdominal CT detected a 30 mm mass arising from the greater curvature of the stomach (Fig. 1D). The preoperative diagnosis was gastrointestinal stromal tumor (GIST) or leiomyosarcoma; thus, laparoscopic partial gastrectomy was performed. Macroscopically, the resected specimen appeared to be a 35 × 30 mm solid grayish tumor covered by the gastric mucosa (Fig. 2A). The patient had an uneventful postoperative course, thereby discharged 10 days after surgery.

Microscopic examination revealed solid proliferation of tumor cells with oval-shaped nuclei and scanty cytoplasm around the disorganized vessels in the submucosa and the muscular layer (Fig. 2B). Immunohistochemical staining showed that the tumor cells were positive for alpha-smooth muscle actin and vimentin but negative for c-kit, CD34, desmin, and S-100. The MIB-1 labeling index was approximately 60 % (Fig. 3). Thus, we diagnosed the patient with GGT. After 2 years and 6 months, abdominal CT revealed metastatic lesions over 40 mm in diameter, with ring enhancement seen in segment 8 of the liver and another liver metastatic lesions of 15 mm observed in segment 5/6 (Fig. 4). Given the diagnosis of liver metastases of GGT, he was treated with three cycles of doxorubicin chemotherapy. Unfortunately, the liver metastases persisted to progress; hence, the patient continued to be treated with eribulin chemotherapy for 26 months and was in good general condition with no particular discomfort.

4. Discussion

GGT is extremely rare, accounting for approximately 1 % of the gastric mesenchymal tumors [4]. Talijeva et al. first reported GGT in 1928 [2]. GGT has a male-to-female ratio of 1:3, mainly affecting individuals in the 50s, but most of them are asymptomatic. Its most common diagnostic methods include EUS and CT. EUS reveals well-defined masses frequently located in the third or fourth gastric layer (submucosa and muscularis propria, respectively), whereas CT provides valuable information regarding size, morphology, internal stricture, growth type, and blood supply [5]. However, both cannot establish clear diagnostic criteria. Thus, techniques such as fine-needle aspiration (FNA) or endoscopically guided fine-needle biopsy aids in obtaining a correct, minimally invasive diagnosis [4].

Most patients with GGT are asymptomatic. Asymptomatic cases can be detected accidentally during a gastrointestinal endoscopy [6]. The most common site is the gastric antrum, which often presents with a submucosal tumor, and differential diseases include GIST, leiomyomas,

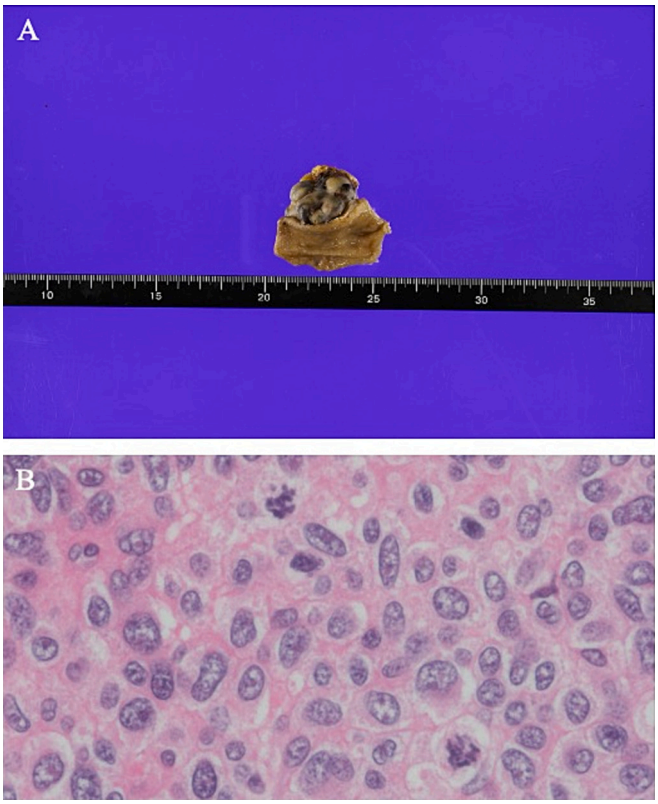

Fig. 2. (A) The resected specimen was a 35 × 30 mm solid grayish tumor covered by the gastric mucosa. (B) Microscopically, the tumor cells with oval-shaped nuclei and scanty cytoplasm proliferated around the disorganized vessels in the submucosa and the muscular layer.

neurogenic tumors, and ectopic pancreas. GGTs are rare and difficult to diagnose preoperatively; however, cases wherein resection was performed after preoperative diagnosis by EUS-FNA have been reported [7–9]. The present case had a rapidly growing submucosal tumor, and preoperative diagnosis using EUS-FNA was considered. Nonetheless, in any case, resection would be necessary; hence, a laparoscopic partial gastrectomy was performed as a diagnosis.

While most GGTs have a good prognosis, metastatic cases have been reported. The mean tumor diameter in patients with metastatic GGT was reported to be 6.7 cm, with 3 cm as the smallest [10]. Malignant lesions are at risk of metastasis according to the following World Health Organization criteria: (1) a tumor diameter of 2 cm or more in gastric development, (2) atypical fission in imaging, and (3) moderate-to-severe nuclear atypia and five or more fission images/50 HPF [11]. In our case, the tumor was approximately 3 cm in diameter and had abundant nuclear fission images. This type of tumor was reported to be a malignant lesion that could cause metastasis; thus, strict follow-up is necessary. Most of the GGT metastases are hematogenous, affecting the liver, lungs, brain, and kidneys, with no reports of lymph node involvement. In fact, few cases of recurrence or metastasis have been reported. Song et al. reported a case of a 3-cm GGT causing simultaneous metastasis to the brain, bone, kidney, and lung, resulting in death after 7 months [12]. However, all of the cases that metastasized to the liver were tumors with a primary lesion of 6.5 cm or larger, and they survived for more than 36 months. In the present case, the patient was diagnosed with liver metastases, and he has survived for 26 months at this time. He may survive in the future depending on the effect of chemotherapy.

In conclusion, we managed a case of asynchronous liver metastasis after performing laparoscopic partial gastrectomy for a rare GGT.

CRediT authorship contribution statement

Yoshitaka Toyomasu drafted the original manuscript. Kenji Nakazato, Yoshinori Shitara, Masatoshi Ishizaki, Hiroshi Saeki, and Ken Shirabe reviewed and critically revised the manuscript. All authors read and approved the final manuscript.

Consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

This study is exempt from ethical approval from the Fujioka General Hospital, Gunma, Japan.

Since this is a case report that does not involve statistical analysis or additional analysis beyond the scope of routine medical practice, ethical review is not required.

Guarantor

Dr. Yoshitaka Toyomasu.

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

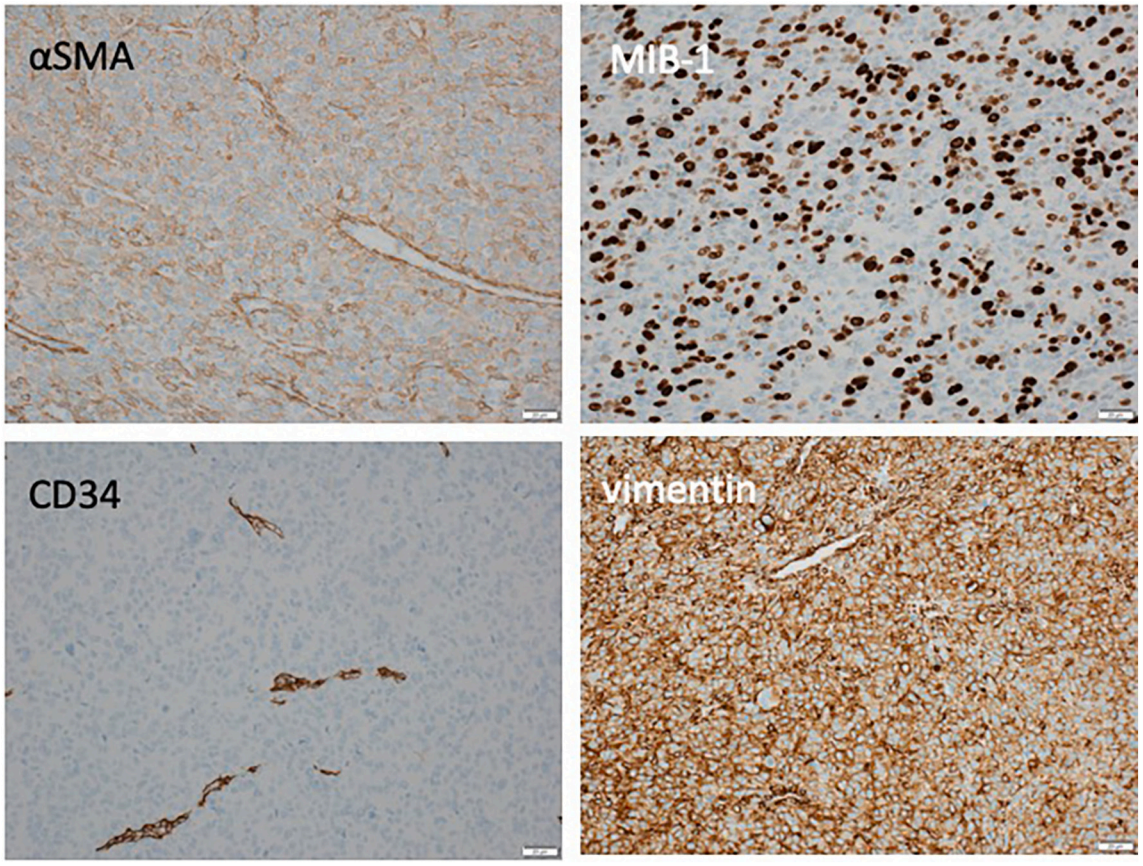

Fig. 3. Immunohistochemical staining showed that the tumor cells were positive for alpha-smooth muscle actin and vimentin but negative for c-kit, CD34, desmin, and S-100. The MIB-1 labeling index was approximately 60 %.

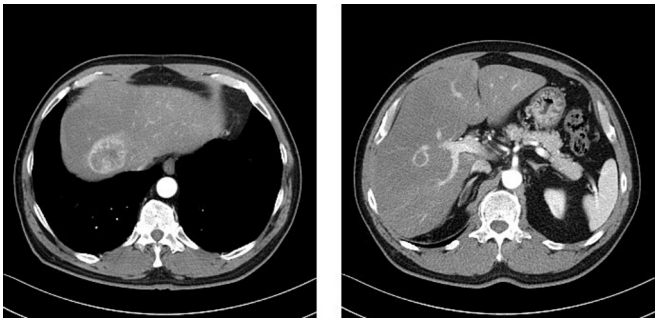

Fig. 4. Abdominal CT revealed metastatic lesions over 40 mm in diameter, with ring enhancement seen in segment 8 of the liver and another liver metastatic lesions of 15 mm observed in segment 5/6.

Declaration of competing interest

The authors have no conflicts of interest to disclose.

Acknowledgement

The authors would like to thank Enago (www.enago.com) for the English language review.

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