Gastric glomus tumor with a preoperative diagnosis by endoscopic ultrasonography-guided fine needle aspiration: a case report

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Abstract

A gastric glomus tumor (GGT) is a rare gastric submucosal tumor that can become malignant. A preoperative diagnosis would allow for a more informed decision regarding the treatment strategy. We present the case of an asymptomatic man with a GGT that was diagnosed during a preoperative examination. Upper gastrointestinal endoscopy was performed in a 64-year-old man and revealed a submucosal tumor at the lesser curvature of the antrum of the stomach. Endoscopic ultrasonography showed a 12-mm-sized hypoechoic tumor in the second and third layers of the stomach wall. A histologic diagnosis of GGT was made using endoscopic ultrasonography-guided fine needle aspiration (EUS-FNA). Abdominal contrast-enhanced computed tomography was performed, but the identification of the tumor was difficult owing to poor enhancement. The gradual growth of the tumor made it necessary to perform an operation. Laparoscopy and endoscopy cooperative surgery was performed without any complications. The tumor cells were immunohistochemically positive for alpha-smooth muscle actin, h-caldesmon, and collagen type IV but were negative for desmin, discovered on GIST-1, S-100 protein, cluster of differentiation 34, epithelial membrane antigen, and cytokeratin AE1/AE3. The final diagnosis was identical to the preoperative diagnosis made using EUS-FNA. EUS-FNA is a useful method for the preoperative diagnosis of small submucosal tumors, including GGTs.

Keywords Gastric glomus tumor · Submucosal tumor · Endoscopic ultrasonography-guided fine needle aspiration · Laparoscopy and endoscopy cooperative surgery

Introduction

A glomus tumor is a rare mesenchymal tumor that usually occurs in peripheral soft tissue but in rare instances, it can occur in the stomach as a submucosal tumor (SMT). Statistics show that only 1% of glomus tumors are classified as gastric SMTs [1]. Gastrointestinal bleeding accompanied by hematemesis and epigastric pain is the most common

initial symptom of a gastric glomus tumor (GGT) [2]. Symptoms are likely to occur in cases of GGT when the tumors are large. However, similar symptoms can occur with other SMTs. Small SMTs are mostly asymptomatic and are usually identified incidentally or during a regular medical examination. In addition, due to the rarity of SMTs, preoperative diagnosis is difficult. We report an asymptomatic patient with a GGT that was diagnosed preoperatively by endoscopic ultrasonography-guided fine needle aspiration (EUS-FNA), and we review the clinical features of GGTs.

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Case report

A gastrointestinal endoscopic examination was performed in a 64-year-old man, and a gastric SMT was identified. The diagnosis of GGT was made using EUS-FNA, and the patient received follow-up. In the tissue, atypical cells with round-to-oval-shaped nuclei and eosinophilic cytoplasm

compose solid aggregates surrounding capillary vessels. Mitotic figures are not evident. Immunohistochemical staining showed that the tumor cells were positive for alpha-smooth muscle actin, but were negative for c-kit, desmin, S-100 protein, clusters of differentiation 34, and cytokeratin AE1/AE3. Therefore, the gastric tumor was diagnosed as glomus tumor.

The patient's tumor had been growing slowly for 3 years and, therefore, surgery was performed. Prior to the endoscopic examination, the patient had diabetes and hypothyroidism. No abnormal findings were detected in the blood tests. Upper gastrointestinal endoscopy revealed a 10-mm-sized SMT with no dellen at the lesser curvature of the antrum of the stomach (Fig. 1a). Endoscopic ultrasonography showed a 12-mm-sized hypoechoic tumor in the second and third layers of the stomach wall (Fig. 1b). Although the endoscopic examination three years ago revealed that the SMT was 10 mm (Fig. 2a), the EUS 3 years ago showed a tumor size of 8 mm (Fig. 2b). We judged that the tumor was increasing, although the degree of tumor growth was slight. Identification of the tumor on computed tomography (CT) was difficult because of the small tumor size and poor image enhancement. No swollen lymph nodes were detected. Because the tumor was intraluminal and located at the lesser curvature of the pylorus of the stomach, laparoscopy and endoscopy cooperative surgery (LECS) was performed.

Macroscopic examination revealed a 13 × 11 × 3-mm-sized nodular mass that arose from the submucosa and extended through the muscularis of the stomach. Microscopic examination revealed a proliferation of round-to-oval-shaped cells with round nuclei and eosinophilic cytoplasm that appeared in sheet-like patterns around the vascular channels of the gastric wall. Immunohistochemical staining showed that the tumor cells were positive for alpha-smooth muscle actin, h-caldesmon, and collagen type IV but were negative for desmin, discovered on GIST-1, S-100 protein, clusters of differentiation 34, epithelial membrane antigen, and cytokeratin AE1/AE3 (Fig. 3a–d). Proliferative index evaluated by ki-67 was less than 1% (Supplemental Fig). The final diagnosis of GGT was identical to the preoperative diagnosis made using by EUS-FNA. The patient's postoperative course of care was successful, and the patient was discharged 7 days after surgery. There was no recurrence or complaints from the patient after the operation.

Discussion

Glomus tumors usually occur in the peripheral soft tissues and are most commonly subungual. The first reports of GGTs were documented by De Busscher in [3] and Kay et al. in [4].

Fig. 1 **a** Upper gastrointestinal endoscopy revealed a 10-mm-sized SMT with no dellen at the lesser curvature of the antrum of the stomach at the time of surgery. **b** Endoscopic ultrasonography showed a 12-mm-sized hypoechoic tumor in the second and third layers of the stomach wall at the time of surgery

Glomus tumors in the stomach are extremely rare and account for less than 1% of all gastric soft-tissue tumors. GGTs are more likely to occur in women than in men (female: male = 3:1) and are most commonly found in the antrum of the stomach. Preoperative diagnosis is quite difficult in many cases due to non-specific clinical findings. We report a case of GGT that was diagnosed preoperatively by EUS-FNA. In addition, we performed a PubMed search for cases of GGTs that were surgically removed during the 10-year period from 2010 to 2019 (Table 1 and Supplemental Table).

Fig. 2 **a** Upper gastrointestinal endoscopy revealed a 10-mm-sized SMT with no dellen at the lesser curvature of the antrum of the stomach at the time of initial diagnosis. **b** Endoscopic ultrasonography showed a 12-mm-sized hypoechoic tumor in the second and third layers of the stomach wall at the time of initial diagnosis

A total of 46 cases were identified, including 14 males and 32 females, with a mean patient age of 51.5 years. The majority of tumors were located in the antrum of the stomach, and the reported symptoms varied. EUS-FNA (including written FNA) was performed in thirteen cases, and GGT was diagnosed in eight of these thirteen cases; three of the remaining five cases had a suspected neuroendocrine tumor

(NET) (Table 2). Seven of the eight cases with a preoperative diagnosis of GGT made using EUS-FNA showed a hypoechoic lesion and a median maximum diameter of 24.5 mm (15–30 mm) (Table 3) [5–12].

Our patient also showed a 12 mm hypoechoic lesion, which was the smallest of all the reported cases with a preoperative diagnosis of GGT made using EUS-FNA. A biopsy was performed in five cases, and one of these five cases were diagnosed with glomus tumor. Our PubMed case review revealed that EUS-FNA provides a preoperative diagnosis that is as accurate as a biopsy, and NET is the most difficult to diagnose.

EUS-FNA is an effective tool for the diagnosis of SMT. Akahoshi et al. [13] reported the accuracy and safety of EUS-FNA in the diagnosis of gastric SMTs smaller than 2 cm. They retrospectively evaluated the usefulness of EUS-FNA for gastric subepithelial lesions smaller than 2 cm in ninety patients. They reported that the diagnostic accuracy of EUS-FNA for gastric subepithelial lesions smaller than 2 cm was 73% and, in surgically resected cases, the diagnostic accuracy of EUS-FNA using immunohistochemical analysis was 98%.

Complications from EUS-FNA, including perforation and seeding, can occur; therefore, EUS-FNA on small lesions must be performed carefully. There were no complications associated with EUS-FNA in our patient. Our findings suggest that EUS-FNA is extremely useful for the preoperative diagnosis of small SMTs.

While most GGTs are benign, some can become malignant and the frequency to become malignant is about 1% [14]. Folpe et al. [15] proposed a criteria for malignancy in GGTs. It has defined as (A) deep location, size > 20 mm, (B) atypical mitotic figures, (C) combination of moderate-to-high nuclear grade, and mitotic activity of more than five mitoses per 50 (HPF). It has been argued that the tumor size of 5 cm or more is an indicator of malignancy [16] [17]. In our review, most of the cases were benign GGTs and the size of the tumor was larger than 2 cm. It helps the 5 cm criterion is appropriate.

There are some reports of malignant GGTs, Toti et al. [18] reported a case of GGT that had multiple liver metastases. Surgical resection and intraoperative radiofrequency ablation were performed on the stomach and liver, and no post-surgical recurrence occurred. Zaidi et al. [16] reported a 10-cm-sized malignant GGT; this report also contained a summary of cases of malignant glomus tumors, the majority of which underwent surgical or endoscopic resection. GGTs may become malignant; therefore, it is desirable to make a diagnosis while the tumor is small. If surgery is performed at the stage of a small tumor, it may be advantageous that the surgical invasion will be small; LECS, or partial resection.

Fig. 3 Microscopic examination. **a** Hematoxylin and eosin staining revealed a proliferation of round-to-oval-shaped cells with round nuclei and eosinophilic cytoplasm that appeared in sheet-like patterns

around the vascular channels of the gastric wall. Immunohistochemical staining showed that the tumor cells were positive for **b** alpha-smooth muscle actin, **c** h-caldesmon, and **d** collagen type IV

Furthermore, it is considered that surgical resection will be considered when symptoms occur. There are 33 cases with symptoms in our review, for example, epigastric pain, bleeding, etc.

In our case, the patient was asymptomatic and the tumor size was extremely small, so follow-up was also considered instead of resection as a strategy. In fact, it has been followed-up for about 3 years, and the resection has been performed due to the gradual increase of the tumor size and the wishes of the patient. It is important to diagnose

accurately gastric glomus tumor with EUS-FNA, which enables surgical resection at the appropriate term. In cases of small gastric glomus tumors, such as our case, it may be appropriate to follow up if possible without immediate surgery. However, surgical resection should be considered when malignant findings are suspected or symptomatic, or if the patient wishes to undergo surgery.

In conclusion, our case details the usefulness of EUS-FNA to diagnose small glomus tumors. Gastric submucosal tumors have various pathological findings and are often

Table 1 Characteristics of the 46 cases in Pubmed

Age(y)		Endoscopic ultrasonography	
Median (range)	51.5 (20–72)	Hypoechoic mass	17
		Hyperechoic mass	4
Preoperative size (Maximum, mm)		Others	5
Median (range)	30 (2–97)	NA	21
Location		Preoperative diagnosis	
Antrum	25	EUS-FNA	13
Body	7	Biopsy	5
Angle	3	Others	1
Fundus	2	NA	27
Pylorus	2		
Prepyloric	2	Surgery procedure	
NA	5	Wedge (Partial) resection	22
		Subtotal gastrectomy	4
Symptoms		Distal gastrectomy	3
Asymptomatic	12	LECS	3
Symptomatic	33	Others	11
NA	1	NA	3
Endoscopy		Postoperative size (Maximum, mm)	
SMT	33	Median (range)	26 (12–100)
Polypoid mass	4		
No abnormal findings	1		
Others	3		
NA	6		

SMT submucosal tumor, *EUS-FNA* endoscopic ultrasonography-guided fine needle aspiration, *LECS* laparoscopy and endoscopy cooperative surgery, *NA* no available information

Table 2 Details of the cases which preoperative diagnosis was performed

FNA (<i>n</i> = 13)	
Diagnosed	8
Misdiagnosed (NET)	5 (3)
Biopsy (<i>n</i> = 6)	
Diagnosed	1
Misdiagnosed	4

NET neuroendocrine tumor

difficult to diagnose. However, we recommend performing EUS-FNA even if the tumor is small, as was observed in our case. A GGT that is preoperatively diagnosed can be treated by surgical resection.

Table 3 Details of the cases which preoperative diagnosis was performed with EUS-FNA

Case	Age	Sex	Location	Size (mm)	EUS	Reference
1	47	Female	NA	25	Hypoechoic pattern	5
2	55	Male	Antrum	24	Hypoechoic pattern	6
3	50	Female	Gastric angle, Ant	15	Hypoechoic pattern	7
4	52	Male	Antrum, Gre	30	Hypoechoic pattern	8
5	70	Female	Antrum, Gre	20	Hypoechoic pattern	9
6	45	Female	Antrum, Post	30	Mosaic echo pattern	10
7	60	Male	Antrum, Gre	20	Hypoechoic pattern	11
8	75	Female	Antrum	33	Hypoechoic pattern	12
9	64	Male	Antrum, Less	12	Hypoechoic pattern	Present case

Less lesser curvature, *Gre* greater curvature, *Ant* anterior wall, *Post* posterior wall, *EUS* endoscopic ultrasonography, *NA* no available information

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Compliance with ethical standards

Conflict of interest None.

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Human and animal rights All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all human participants.

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