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Endoscopic Full-Thickness Resection for a Giant Gastric Glomus Tumor with Extra Luminal Growth: A Case Report

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1. Abstract

1.1. Background

Gastric glomus tumor (GGT) is a rare mesenchymal neoplasm derived from the glomus body. Due to its atypical clinical, endoscopic, and imaging manifestations, the final diagnosis of GGT needs pathological confirmation. Surgical resection is the main therapeutic method for giant GGTs, and some small GGTs have been treated through endoscopic resection in recent years. Here, we report a case of giant GGT originating from the muscularis propria with extraluminal growth that underwent endoscopic full-thickness resection (EFTR).

1.2. Case Summary

A 61-year-old female was admitted to our institution due to a gastric mass observed by esophagogastroduodenoscopy. Contrast-enhanced abdominal CT scans showed that the lesion was located in the gastric antrum area with ill-defined boundaries and heterogeneous enhancement. Endoscopic ultrasonography (EUS) confirmed a 3 cm × 4 cm lesion that was hypoechoic and homogeneous, originating from the muscularis propria and protruding in and out of the lumen. A gastric stromal tumor (GST) was suspected before resection. The patient then underwent EFTR. The postoperative pathological diagnosis was GGT. The endoscopic resection was successful with no complications, and the patient recovered uneventfully. To our knowledge, this is the first case of GGT over 4 cm resected by EFTR in the English literature.

1.3. Conclusion

EFTR is an effective and safe therapy for giant GGTs. It seems to be a promising and less invasive alternative than surgery for gastric submucosal tumors (SMTs).

2. Introduction

Glomus tumors (GTs) are vascular neoplasms arising from neuromuscular artery spheroid cells or glomus bodies involved in the thermoregulation of arteriovenous structures. It is more common in the dermis of the fingers, toes, and peripheral soft tissues of the subcutaneous nailfold area [1, 2]. Other positions include the sublingual area, nerves, nasal cavity, trachea, genitourinary area, gastrointestinal tract, bile duct, and peritoneum [3]. GGT was first reported in 1951 [4]. The clinical symptoms of GGT are upper abdominal pain, bleeding, and vomiting. Some cases of GGT are occasionally found by CT or EUS without symptoms. GGT is extremely rare accounting for approximately 1% of gastric mesenchymal tumors. Only 9 cases of duodenal GT and 20 cases of primary intestinal GT have been described in the reported literature [2, 5].

Most GGTs are benign and cured by surgery. With the development of various endoscopic techniques, GGTs can be treated by endoscopic resections, especially lesions less than 3 cm in size. Here, we

report a 4 cm × 4.5 cm giant GGT that was completely removed by EFTR. No recurrence was observed after 6 months of follow-up in our case.

3. Case Presentation

3.1. Chief Complaints

A 61-year-old female was admitted to our hospital with complaint of a submucosal gastric lesion during health examination.

3.2. History of Present Illness

The patient had no specific symptom.

3.3. History of Past Illness

The patient had a 3-year gallbladder stone in her medical history.

3.4. Personal and Family History

The patient had no special personal and family history.

3.5. Physical Examination upon Admission

Physical examination revealed no palpable mass and tenderness. The patient had no pathological signs.

3.6. Laboratory Examinations

The routine blood test, fecal test, blood biochemistry, immune index and serum tumor markers were within normal limits.

3.7. Imaging Examinations

Contrast-enhanced abdominal CT scans showed that the lesion was situated in the gastric antrum area with ill-defined boundaries and heterogeneous enhancement. The size was about 2.9 cm × 2.7 cm. A few signs of necrosis were observed inside the lesion (Figure 1). The esophagogastroduodenoscopy showed a 3 cm × 4 cm submucosal eminence near the posterior wall of the gastric antrum area with smooth surface. EUS confirmed that the lesion was hypoechoic and homogeneous, originating from muscularis propria and protruding into and out of the lumen (Figure 2). The distal lesion was not clearly visible.

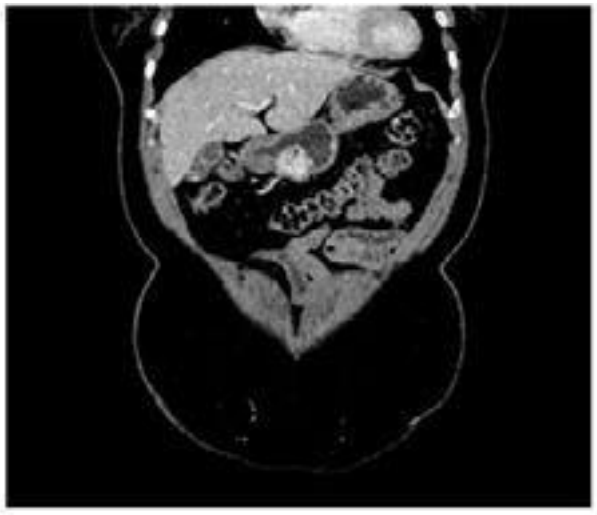

Figure 1: Abdominal computed tomography findings. A 2.9 cm × 2.7 cm lesion was situated in the gastric antrum area with ill-defined boundaries and heterogeneous enhancement.

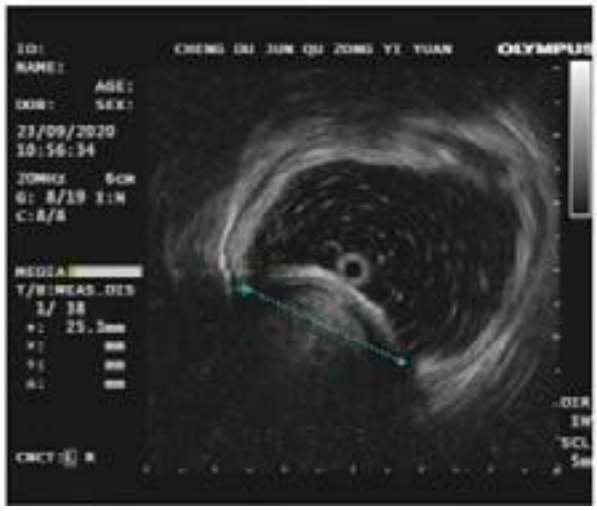

Figure 2: Endoscopic ultrasonography images. A 3 cm × 4 cm submucosal eminence near the posterior wall of the greater curvature of the gastric antrum with smooth surface was observed.

3.8. Histological Examinations

Biopsy histology showed that trabeculae of tumor cells were distributed around blood vessels. Proliferating oval-shaped cells in small nest formation with a high nuclear to cytoplasmic ratio were observed (Figure 3A). The immunohistochemical profile demonstrated SMA (+) (Figure 3B), Syn (+) (Figure 3C), Calponin (+), Vim (+), CD34(-) (Figure 3D), CD45 (-), CD56 (-), CK8/18 (-), CgA (-), ERG (-), S- 100(-), TIF-1 (-), and Ki-67 (+,1-3%).

3.9. Final Diagnosis

Based on the imaging, EUS, and histological findings, the final diagnosis of the present case was GGT.

3.10. Treatment

After comprehensive assessment, the EFTR was performed. The procedures were as follow: first, the lesion area was labeled; second, the indigo rouge saline was injected in the submucosa to create working space; third, the submucosal layer and muscularis propria were gradually incised. The tumor was located outside the muscularis propria and was partially enclosed with the omentum inside the abdominal cavity. A circumferential incision as deep as muscularis propria around the lesion was performed with insulated-tip (IT) knife (T-Type I-Jet, HybridKnife), then full-thickness resection of the tumor including extraluminal growth was completed; finally, the wound and perforation were sutured with nylon rope (Loop30-LD195, LeoMed) and clipped with 9 titanium clips (ROCC-D-26-195-C, Micro-Tech(Nanjing)) (Figure 4). The postoperative specimen size was about 4 cm × 4.5 cm.

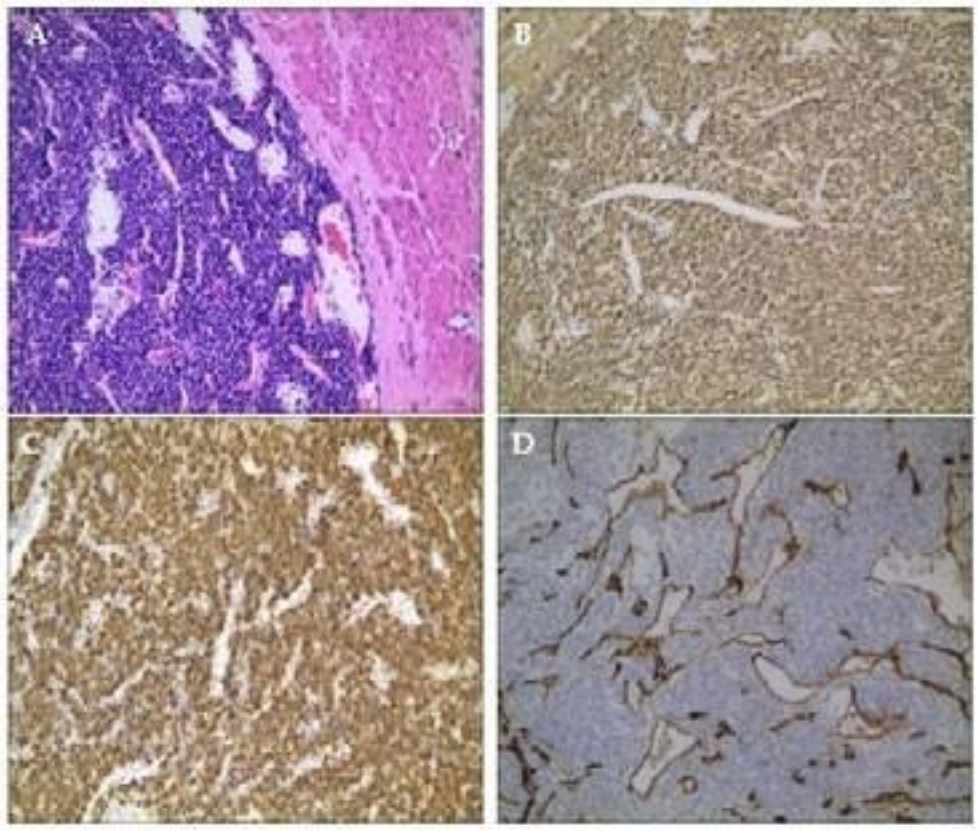

Figure 3: The histological and immunohistochemically profiles of the lesion. A: Trabeculae of tumor cells distributed around the blood vessel (Hematoxylin & Eosin staining ×100); B and C: Tumor cells were positive for smooth muscle actin (B, ×200) and syn (C, ×200); D: Tumor cells were negative for CD34 (×200).

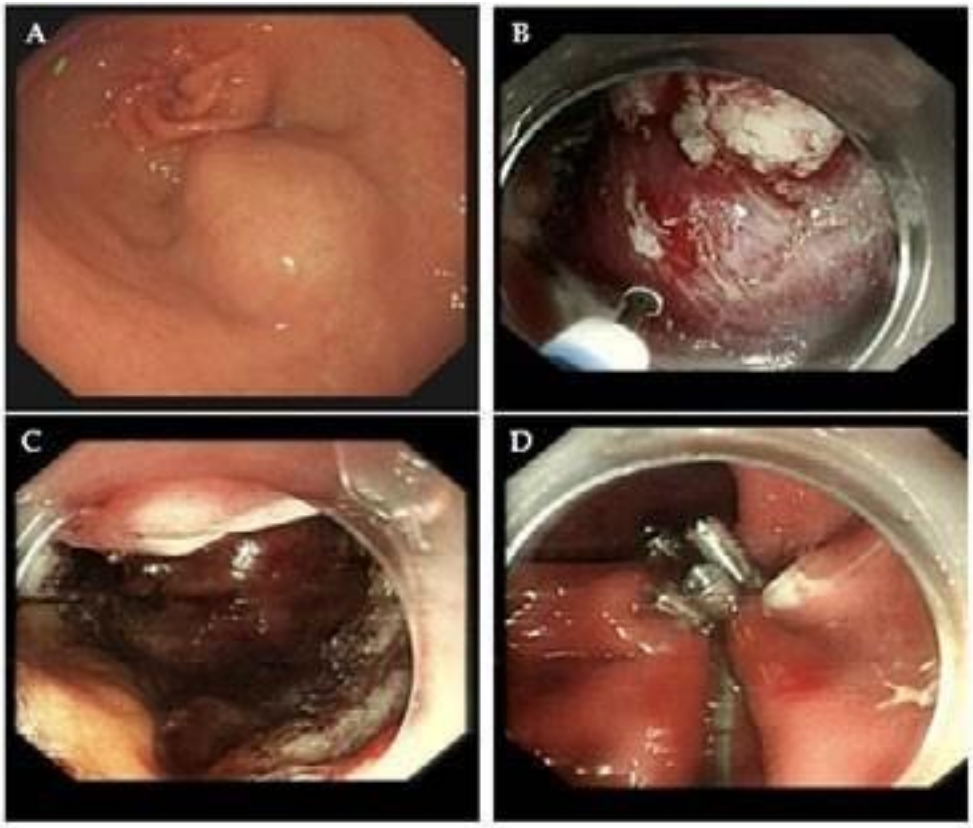

Figure 4: The gastric glomus tumor was respected by endoscopic full-thickness resection. A: The esophagogastroduodenoscopy showed a giant submucosal eminence with smooth surface; B: The lesion was incised with Hook and IT; C: Circumferential incision including extra luminal growth was completed; D: The perforation was closed with the nylon rope and clips.

3.11. Outcome and Follow-Up

There was no intraoperative and postoperative complications. The patient was in good overall and local condition at discharge. No recurrence was observed after 6 months of follow-up.

4. Discussion

GGT more commonly occurs in females than in males (the female to male ratio is 1.6:1). The median age is 45 years (range from 28 to 79 years) [6]. GGTs are more likely located in the gastric antrum and are rarely situated in other parts of the stomach [7-9]. The clinical symptoms are atypical and include abdominal pain and discomfort, loss of appetite, gastrointestinal bleeding, and ulcers with or without nausea and vomiting. Some patients with GGTs have no complaints [10].

GGTs and GSTs exert analogous clinical symptoms and imaging manifestations, making it difficult to differentiate between the two types of tumors [11]. Wang et al. analyzed the features of CT scans between GGTs and GSTs. GGTs are mainly located in the antrum with endophytic growth and heterogeneous enhancement in the arterial phase (AP). The CT attenuation value of GGT in the AP, portal venous phase (PP), and the degree of enhancement (DE), which include DE [AP] (CT attenuation value of the AP minus that of the unenhanced phase) and DE [PP] (CT attenuation value of the PP minus that of the unenhanced phase), are significantly higher than those of GST [12]. Endoscopically, GGT is a submucosal mass with either normal mucosa or ulceration. EUS is a practical imaging technique for diagnosing gastric submucosal tumors. Hu et al. reported that typical EUS properties of GGTs were as follows: the shape of GGT is round or oval on EUS; it is a mild-hyperechoic lesion from the fourth layer [13]. Histopathological examination is necessary for the accurate diagnosis of GGTs. Histologically; most GGTs had solid nests of tumor cells clustered around the wall of blood vessels. The tumor nodules were separated by bundles of smooth muscle and fibrous tissue with hyalinization. Immunohistochemistry (IHC) contributes to the final diagnosis of GGTs when the cell staining of smooth muscle actin (SMA), vimentin, calponin, h-caldesmon, collagen type IV, and laminin are positive, while desmin, chromogranin A, cytokeratin (AE1/AE3b), S-100 protein, creatine kinase, C-KIT (CD-117), CD34, DOG1 protein (K9), p53 protein, and neuron-specific enolase are negative. The proliferation marker Ki-67 was positive in less than 5% of tumor cell nuclei [14-18]. The findings of the present case are consistent with the histological and immunohistochemical features of GGT.

Although most GTs are benign, malignant GGTs with various organ metastases have been reported [19-23]. Resection is a radical treatment with a favorable prognosis. Previously, surgery was the priority choice of treatment for GGT [8, 9, 24]. Surgical treatment includes subtotal gastrectomy, wedge resection, and excision of the tumor depending on its location and size [7].With the development of endoscopic technologies, endoscopic resection, such as endoscopic submucosal dissection (ESD) and EFTR [25, 26], is increasingly

used in treating gastric submucosal tumors (SMTs). For the past few years, laparoscopy endoscopy cooperative surgery (LECS) has been performed in patients with GGT and SMT [27, 28]. ESD is used to separate the tumor originating in the submucosal layer, while EFTR is applied to resect full-thickness tumors originating in the muscularis propria layer. Since an iatrogenic perforation of the gastric wall was created, clips or purse-string sutures are used to close the gastric wall defect [29]. Furthermore, EFTR has a high rate of microscopic margin-negative resection [30]. Zhou et al. successfully removed 26 gastric SMTs originating from the muscularis propria that were adhesive to the serosa with EFTR. The reported lesion size ranged from 1.2 cm to 4.5 cm, and the pathologic diagnosis included gastrointestinal stromal tumors (GISTs) (16/26), leiomyomas (6/26), GGTs (3/26), and Schwannoma (1/26) [31]. A retrospective study analyzed 11 patients with GGTs, 3 of which were treated by ESD, 1 underwent EFTR [13]. Several reports analyzed clinical features and treatments of GGTs (Table 1). Seventeen patients with GGTs were excised by endoscopic resection. The size ranged from 0.8 cm to 2.7 cm. EFTR was performed in one patient with GGT, which was 2.7 cm in size [13]. We report the first case of GGT over 4 cm resected by EFTR in the English literature. There was no fatal bleeding, residual tumor, or recurrence.

Table 1: The clinical features and treatment of 17 patients with GGT.

Reference	No.	Sex	Age	Symptom	Size (mm)	location	Treatment	Fol low-up (month)	Recurrence
Hu et al[13], 2019	1	M	38	Abdominal pain	19	Antrum	ESD	3	No
	2	F	62	Abdominal discomfort	10	Body	ESD	62	No
	3	F	56	Abdominal discomfort	11	Antrum	ESD	48	No
	4	F	52	Abdominal pain	27	Antrum	EFTR	60	No
Lin et al[32], 2020	5	F	34	Epigastric pain	20	Antrum	Endoscopic resection	97	No
	6	M	54	Epigastric pain	8	Antrum	Endoscopic resection	58	No
	7	M	60	Epigastric pain	27	Antrum	Endoscopic resection	48	No
	8	F	55	None	27	Antrum	Endoscopic resection	13	No
Bai et al[33], 2021	9	F	36	None	10	Antrum	ESD	36	No
	10	F	47	None	12	Antrum	ESD	48	No
	11	M	63	Epigastric pain	15	Antrum	ESD	60	No
	12	M	65	Epigastric pain	18	Antrum	ESD	24	No
	13	M	58	Heartburn	23	Antrum	ESD	48	No
	14	F	54	Heartburn	21	Antrum	ESD	72	No
	15	M	64	Epigastric pain	22	Antrum	ESD	12	No
	16	M	70	None	13	Antrum	ESD	60	No
	17	F	56	Epigastric pain	17	Antrum	ESD	36	No

M: male; F: female; ESD: endoscopic submucosal dissection; EFTR: endoscopic full-thickness resection

5. Conclusion

Endoscopic resection has some advantages, including minimal surgical trauma, quick wound healing, and easy acceptance by patients. EFTR is an effective and safe therapy for giant GGTs. It appears to be a promising and less invasive alternative than surgery for gastric SMTs.

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