Clinics of Oncology

Case Report ISSN: 2640-1037 | Volume 6

# Glomus Tumor of the Stomach. Diagnostic Approach and Surgical Management

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Received: 20 Nov 2022
Accepted: 01 Dec 2022
Published: 10 Dec 2022
J Short Name: COO



**Citation:**

Renzulli P. Glomus Tumor of the Stomach. Diagnostic Approach and Surgical Management.
Clin Onco. 2022; 6(16): 1-4

**Keywords:**

Glomangioma; Tumour; Schwannoma

## 1. Summary

Here, we report on a male patient in his mid-fifties with a known ectasia of the ascending aorta. Computed tomography led to the incidental finding of a 3 cm large submucosal tumor located on the anterior wall of the gastric body, close to the greater curvature. Magnetic resonance imaging and endoscopy confirmed the finding. An endoscopic ultrasound-guided biopsy, however, was not performed. The patient underwent laparoscopic gastric wedge resection under the tentative diagnosis of a gastrointestinal stromal tumor. Further analysis showed a glomangioma, which is a histological variant of a gastric glomus tumour. The patient made a swift and full recovery. Gastric glomus tumors are extremely rare mesenchymal neoplasms. They present as gastric submucosal tumors. About 6% of gastric glomus tumours are reported to be malignant. Surgical resection, without lymphadenectomy, but with adequate free margins, and preferably minimally-invasive, remains the treatment of choice.

## 2. Background

Glomus tumors are mesenchymal neoplasms composed of modified smooth muscle cells originating from perivascular glomus bodies, which are responsible for thermoregulation through arteriovenous shunting of blood [1, 2]. In general, they are found in the fingers and toes of patients, where glomus bodies are abundant [1, 2]. Occasionally, glomus tumors can be found along the gastrointestinal tract. In such cases, it appears that the stomach is disproportionally affected [1, 2, 3, 4]. Gastric glomus tumours (GGTs) are extremely rare with less than 200 cases reported in the literature [2]. The present case report provides the opportunity to discuss the differential diagnoses, the diagnostic approach and the surgical management of gastric submucosal tumours (SMTs) [1, 5, 6].

## 3. Case Presentation

A male patient in his mid-fifties presented to the emergency department with acute onset chest pain. Previous medical history was remarkable for an ectasia of the ascending aorta (tubular portion: 43 mm, sinus portion: 46 mm), which had been stable in size for the last few years in follow-up controls. In addition, the patient had previously undergone a total thyroidectomy for nodular goiter and was under levothyroxine replacement therapy. Due to hypertension, furthermore, the patient was also prescribed an angiotensin II receptor antagonist (valsartan). The patient was also known to have a penicillin allergy causing an anaphylactic reaction as well as a nicotine habit of 20 pack-years. The patient had never undergone previous thoracic or abdominal surgery. Physical examination was largely normal: 183 cm, 84 kg, BMI 25 kg/m2, blood pressure 156/101 mm Hg, heart rate 77 bpm, O2 saturation 96%, temperature 36.9°C.

## 4. Investigations

Heart enzymes and electrocardiography were within normal range, thereby excluding the possibility of an acute coronary syndrome.

In addition, an emergent contrast-enhanced computed tomography (CT) was able to further exclude an aortic dissection or pulmonary embolism. The patient showed a rapid spontaneous recovery and the chest pain was eventually considered to be of musculoskeletal origin.

Upon closer examination, the CT scan revealed an incidental rounded tumor measuring 20 x 30 x 30 mm. The lesion showed inhomogeneous contrast enhancement and was located close to the greater curvature, protruding from the anterior wall of the gastric body. In retrospect, the tumor could be seen in a previous CT scan dating back 2 years. The tumor, which previously escaped detection, appeared stable in size. The patient underwent a gastroscopy showing a 2 cm large indentation of the anterior gastric wall. The overlying gastric mucosa showed no further pathologies and appeared completely normal (Figure 1A). Endoscopic ultrasound (EUS) showed a normal gastric wall with an adherent tumor approximately 2 cm in size (Figure 1B). An endoscopic biopsy was not performed. An abdominal Magnetic Resonance Imaging (MRI) further confirmed the clearly demarcated lesion on the body of the stomach measuring 31 mm (Figure 2). Additionally, two liver haemangiomas in segments 2 and 4a were diagnosed.

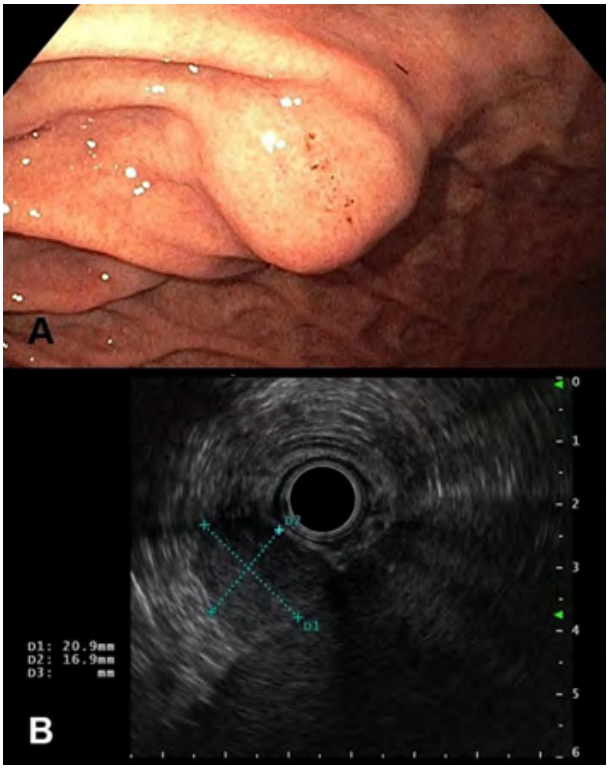


**Figure 1:** Preoperative gastroscopy. A: Bulging of the gastric wall into the gastric lumen with intact mucosal covering. B: Endoscopic ultrasound (EUS).

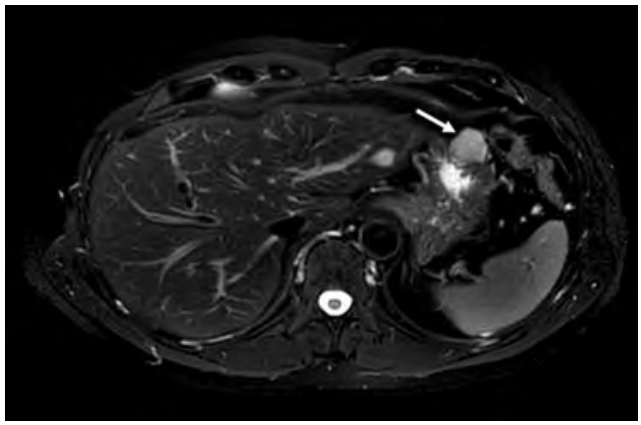

**Figure 2:** Magnetic resonance imaging (T2 weighted MRI). Tumour of the anterior gastric wall (white arrowhead).

## 5. Differential Diagnosis

Based on the imaging described above, a provisional diagnosis of a gastric SMT was made. The most common differential diagnosis in this case was a gastrointestinal stromal tumour (GIST) of the stomach. Further potential diagnoses included, in order of frequency: heterotopic pancreas, leiomyoma, schwannoma, glomus tumor, lipoma, granular cell tumor and neurofibroma [6]. An endoscopic ultrasound-guided biopsy was not performed due to fears of procedure-related complications (bleeding, tumor cell seeding). In addition, given the anatomically favorable location of the tumor, a laparoscopic gastric wedge resection was considered technically straightforward, therefore obviating the need for a preoperative histological assessment. The case was discussed in an interdisciplinary tumor board and surgical resection was recommended.

## 6. Treatment and Outcome

The patient underwent laparoscopic wedge resection of the stomach for the tentative diagnosis of a GIST (Figure 3A). The postoperative course remained uneventful and the patient was discharged 2 days later. A follow-up visit after 3 months showed that the patient was in excellent health. Macroscopic examination showed an exophytic, circumscribed tumor of the external gastric wall measuring 25 x 20 x 21 mm (Figure 3B). Histologically, the tumor consisted of polygonal cells embedded in a collagenous stroma surrounding numerous dilated vessels (Figure 4A - 4C). Immunohistochemistry showed a strong expression of smooth muscle actin (SMA) (Figure 4D), whereas CD45a, desmin, DOG1, CD117, and pancytokeratin were negative. The morphology, in conjunction with the immune profile, was consistent with a glomangioma, a histological variant of GGT with a prominent vascular component.

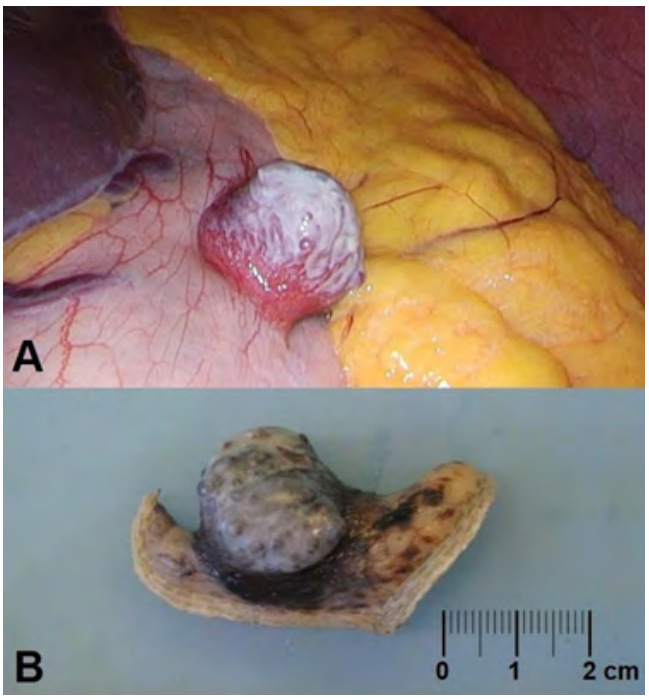


**Figure 3:** Glomangioma of the stomach. A: Intraoperative view during laparoscopy. B: Resected specimen (formalin-fixated).

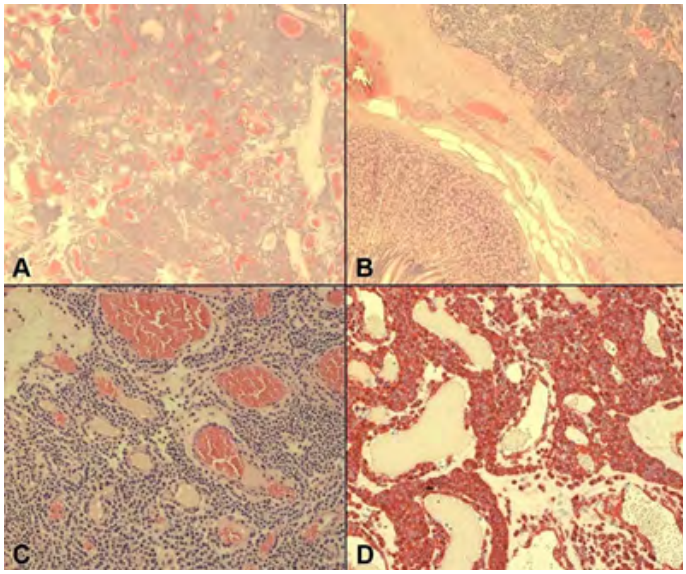


**Figure 4:** Histology of the resection specimen showing the typical high vascularity of the glomangioma (A), its location in the gastric wall (B; right upper corner: glomangioma, left lower corner: gastric mucosa), the bland appearance of the tumor cells at a higher magnification (C) and the strong expression of the tumor cells for smooth muscle actin (SMA). Haematoxylin-eosin and actin staining, overall magnification A: 8x; B: 10x; C and D: 40x

## 7. Discussion

GGTs are extremely rare mesenchymal tumors. A recent systematic review was only able to identify 187 recorded cases since 2000 [2]. Mean age at presentation appears to be around 52 years of age, with the majority of cases affecting women (61%). Patients became symptomatic mainly due to epigastric pain (33.9%) or upper gastrointestinal bleeding (23%). About a quarter of all patients (23%) remained asymptomatic and the tumor was only discovered incidentally. Most tumors were located within the gastric antrum (75.3%), with a mean tumor size of 2.82 cm. Within the review, a preoperative biopsy, usually endoscopic and ultrasound-guided, was performed in 43 cases. However, even with biopsy, the correct diagnosis could preoperatively only be determined in only 51% of cases (22/43). About two thirds of patients underwent gastric wedge resection (62%). Expression of SMA was found in all patients where immunohistochemistry was documented. Eleven patients (6%) presented with malignant GGTs. Here, the mean size of the lesions was 6.5 cm, versus 2.5 cm in benign GGTs ($p<0.001$). In general, patients with malignant GGTs were significantly older and tumors were preferentially located in the gastric fundus. In patients with malignant GGTs, distant metastases to the liver, kidney, brain and skin were detected; however, no metastases to regional lymph nodes were reported. Therefore, just as for GIST, routine lymphadenectomy may not be indicated [2, 6].

The data garnered from the literature makes clear that both diagnostic approach and the surgical management of GGTs require further elaboration.

To begin, due to the relative paucity of cases, clear diagnostic criteria are as of yet not well defined. GGTs are hypervascular tumors. A contrast-enhanced CT examination will therefore show a rounded lesion within the gastric wall with strong enhancement in the arterial phase. Furthermore, GGTs are gastric SMTs. The overlying mucosa is usually intact. The tumor manifests itself optically as an indentation of the gastric wall. In a minority of cases, however, a mucosal ulceration can be seen [3]. Endoscopic ultrasound (EUS) reveals an iso- or hypoechoic mass, sometimes with intralesional calcifications, arising from the 3rd or 4th layer of the gastric wall [2]. Although the differential diagnosis of SMTs includes a variety of pathologies, GIST is by far the most common [1, 5]. Given the submucosal localization, an endoscopic ultrasound-guided biopsy will provide the correct preoperative diagnosis in only about half of all cases [2]. Therefore, the information gained by a preoperative endoscopic biopsy must be weighed against possible procedure-related complications. Compounding the diagnostic problem, aspects of malignant GGTs need to be addressed. Currently, no definitive histological criteria exist to distinguish benign from malignant GGTs [1]. The occurrence of metastases is the only categorical feature defining malignancy. Mitotic activity and nuclear atypia have thus far been shown to be poor predictors of malignancy. Moreover, vascular invasion is frequently seen in benign as well as malignant GGTs [3, 7]. The metastatic risk of glomus tumors of the extremities seems to be related to depth and a tumor size of more than 2 cm as well as atypical mitotic figures [7]. With regards to GGTs, however, a tumor size of greater than 5 cm, especially if associated with atypical histological features (spindle cell change, atypical mitotic figures, high nuclear atypia, vascular invasion), has been proposed as a more appropriate indicator of

malignant tumor behavior. The presence of such features should result in the inclusion of affected patients in a clinical and radiological follow-up program [1, 2, 3, 4].

The second aspect requiring further evaluation concerns the surgical approach. Most GGTs are located within the gastric antrum (75% - 82%) [3, 6]. Resection with adequate disease-free margins is the mainstay of surgical treatment [2]. A minimally invasive technique is, when possible, preferred. In more than 60% of patients, tumor clearance was achieved through gastric wedge resection [2]. A tumor location in close vicinity to the gastric in- or outlet, however, may necessitate a formal proximal or distal gastric resection [2]. The available data on malignant GGTs remains extremely sparse. However, thus far, no reports on metastases to regional lymph nodes have been published.

Therefore, some authors conclude, that a routine lymphadenectomy is not indicated [2].

In summary, GGTs are extremely rare submucosal mesenchymal tumours of the gastric wall, preferentially located in the antrum. Malignant GGTs are uncommon, mostly arising from lesions larger than 5 cm. Endoscopic ultrasound-guided biopsy will provide a preoperative histological diagnosis in only about half of all cases. Surgical resection, preferably laparoscopic, with adequate free margins, is the treatment of choice. A routine lymphadenectomy is not indicated.

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