



## Case Report

## “A hybrid approach for GISTs near the esophagogastric junction, a case report”

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## ABSTRACT

**Introduction and importance:** Gastrointestinal stromal tumors are the most frequent mesenchymal tumors of the gastrointestinal tract. Complete resection of GISTs is the only chance of cure for patients. When these tumors are located near the esophagogastric junction, the surgical risk can cause deformity or stenosis in the gastric inlet, leading to higher complications and diminishing their quality of life. In such cases, a more sophisticated and tailored approach should be used.

**Case presentation:** We present the case of a 42-year-old female; she presented to our office with a 3-year history of nausea, vomiting and abdominal distension. Two GISTs were located near the EGJ, and with a combined approach we achieved complete resection. On follow-ups, the patient is doing well.

**Clinical Discussion, Conclusion:** When diagnosis is confirmed, surgical resection must be the first choice for GISTs as complete surgical excision is the only permanent cure. The rise of endoscopic surgery has become a valuable tool and a critical element in surgery. Hybrid techniques that combine laparoscopic and endoscopic approaches can improve the patient's outcomes and provide better results.

## 1. Introduction

The safety and oncologic outcome of laparoscopic gastric GIST resection is well established; negative margins are the preferred surgical choice to improve patients' outcomes and survival [1]. The treatment of GIST tumors at the GE junction is troublesome as it may need proximal gastrectomy esophagectomy [2]. When these events occur, the transoperative decision will directly impact the patient's quality of life [3]. Therefore, laparoscopic and endoscopies techniques are needed to correctly address these lesions, especially when their locations are inconvenient [4,5]. We present the case of a 42-year-old patient, two GISTs tumors were found near the GE junction in the lesser gastric

curvature. A hybrid procedure with endoscopy and laparoscopy achieved complete tumor resection. On follow-ups, the patient is doing well.

Our work has been reported in line with the SCARE 2020 criteria [13].

## 1.1. Case presentation

The patient is a 42-year old, otherwise healthy female. She presented to our office with a 3-year history of nausea, vomiting, abdominal distension, early satiety, and mild but recurrent postprandial abdominal pain. She had no history of fever, dysphagia, weight loss, bleeding, or any other symptoms. Clinical examination was unremarkable and

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complementary exams were requested. Complete blood count and chemistry panel were normal; nonetheless, the upper endoscopy revealed two small ( $1 \times 1 \times 0.5$  cm &  $0.5 \times 0.5 \times 0.3$  cm) gastric lesions covered with normal mucosa. The lesions were found 2 cm away from the esophagogastric junction in the lesser curvature.

To further investigate the submucosal tumor, endoscopic ultrasound (EUS) was performed, revealing two small hypoechoic lesions with clear margins. The low-echo lesions existed in the submucosal layer. The color Doppler image showed a pulsating vascular signal extending into the center of the low-echo lesion from the periphery (Fig. 1A and B). EUS fine-needle aspiration was performed for pathological evaluation of the submucosal tumor and was completed safely without any complications. Immunostaining was performed, revealing positivity for CD34 and CD117, and were negative for S100 and anti- $\alpha$ -Sm-1. Gastric GIST was diagnosed. A contrast-enhanced abdominal computed tomography (CT) was normal and didn't reveal any masses or lymph nodes; thus, surgery was decided.

Due to the tumor's location, several surgical strategies were proposed to achieve complete resection; wedge, subtotal, and partial gastrectomy were considered. Nonetheless, after recognizing the stomach's lesser curvature and its posterior wall during laparoscopy, the tumor was not recognized. Intraoperative endoscopy was completed showing the small tumors near the esophagogastric junction. Therefore, a hybrid trans-gastric resection of the gastric GIST with the surgeon and endoscopist's help was decided (Fig. 2A and B).

The stomach was fixed to the anterior abdominal wall using an absorbable suture (Vicryl, Johnson & Johnson Medical), and a 10mm gastrostomy was completed using an ultrasound device. A 10mm trocar was placed through the gastrostomy, and the inner gastric surface was seen; another 10mm trocar was placed under direct endoscopic visualization. The submucosal tumors were located near the esophagogastric junction and were covered by normal mucosa. A 3-0 Vicryl stitch was placed on the biggest tumor to provide traction, and using two consecutive 2.5mm endo staplers (30mm), the first tumor was completely resected. For the second tumor, another 3-0 Vicryl stitch was placed to provide traction and was removed using an ultrasound energy device (Fig. 3). After this, the trocars were removed from the stomach, and the gastrostomies were closed using a 3-0 sterile synthetic absorbable monofilament. (PDS, Johnson & Johnson Medical) The rest of the procedure was completed without complications (Fig. 4A and B & Supplementary Video). Pathology reported a total tumor excision; the tumors were composed of spindle cells with eosinophilic, fibrillary cytoplasm with focal para nuclear vacuolization and low mitosis. Gastric GIST was the final diagnosis.

The patient's postoperative course was uneventful. Liquid diet was started on the first postoperative day and was followed by a full diet. She

was discharged on her third postoperative day without complications. On follow-ups, three months after surgery, the patient is doing well.

## 2. Discussion and conclusion

Gastrointestinal stromal tumors (GIST) are the most common malignant subepithelial lesions of the gastrointestinal (GI) tract. These tumors can appear anywhere along the GI tract [1,2]. Nonetheless, they are found more frequently on the stomach (60%), jejunum and ileum (30%), and duodenum (4–5%). GIST's annual incidence is estimated at only 10–20 per million, and regrettably they hold malignancy potential [1]. These rare tumors originate from the interstitial cells of Cajal from the myenteric plexus in the muscular layer of the GI tract [2]. These cells regulate gut motility and act as peacemakers of the GI tract; they are KIT, and CD117 positive; consequently, these stains are diagnostic for GIST in 75–80% of the patients [2,3]. Mutations on the KIT gene and the platelet-derived growth factor receptor alpha can lead to an unregulated proliferation of the Cajal cells and tumor development [1]. Approximately 10%–30% of GISTs have a malignant clinical course; the risk of malignancy can be classified according to their size and mitotic count [2].

GISTs usually present with non-specific gastrointestinal symptoms, including bleeding, abdominal pain, distention, and discomfort [2]. As our patient experienced. Still, up to 30% of the patients can be completely asymptomatic and are diagnosed incidentally during other gastrointestinal surgery or autopsy [1,4]. These tumors are typically diagnosed during upper endoscopy and appear as a smooth bulge covered with normal mucosa. To fully differentiate these tumors from other malignancies, additional testing is necessary [2,3]. EUS is a crucial test to diagnose subepithelial tumors. It can provide information on which gastrointestinal wall layer it originates, the nature of the lesion, and its actual size [1]. Fine needle aspiration can provide samples to give a conclusive immunohistochemical diagnosis safely [2,3]. In our patient, the tumor was identified during an endoscopy, EUS and fine-needle aspiration aided in reaching the final diagnosis.

When diagnosis is confirmed, surgical resection must be the first choice for GISTs without metastasis [2]. Although GISTs of less than 2 cm have a low frequency to metastasize (not 0%), the medical team should consider close follow up or early aggressive treatment [1,4]. Complete surgical excision is the only permanent cure. R0 resection of all gross and microscopic disease has been associated with improved local recurrence rate and overall survival [1]. GISTs can be, in some cases, friable and must be handled with care to prevent intraoperative rupture or damage its capsule, which can be equivalent to a positive surgical margin [2]. Wedge or segmental resection and less invasive procedures such as laparoscopic, minimally invasive endoscopic

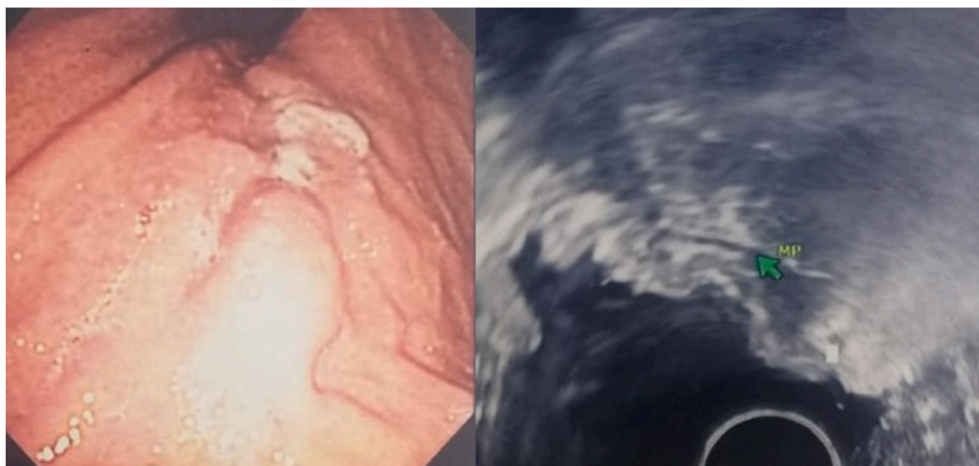
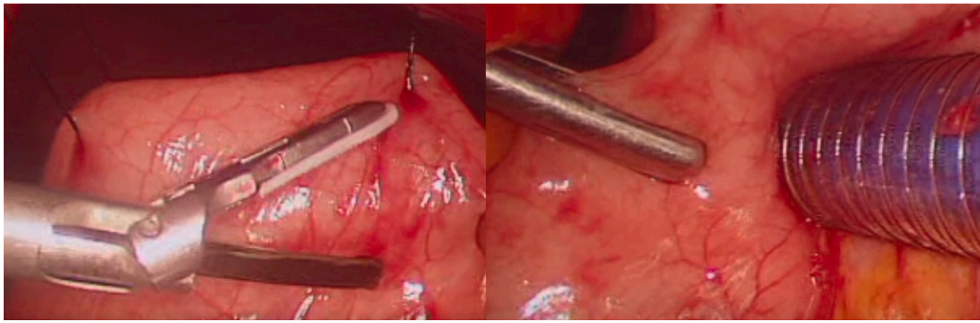
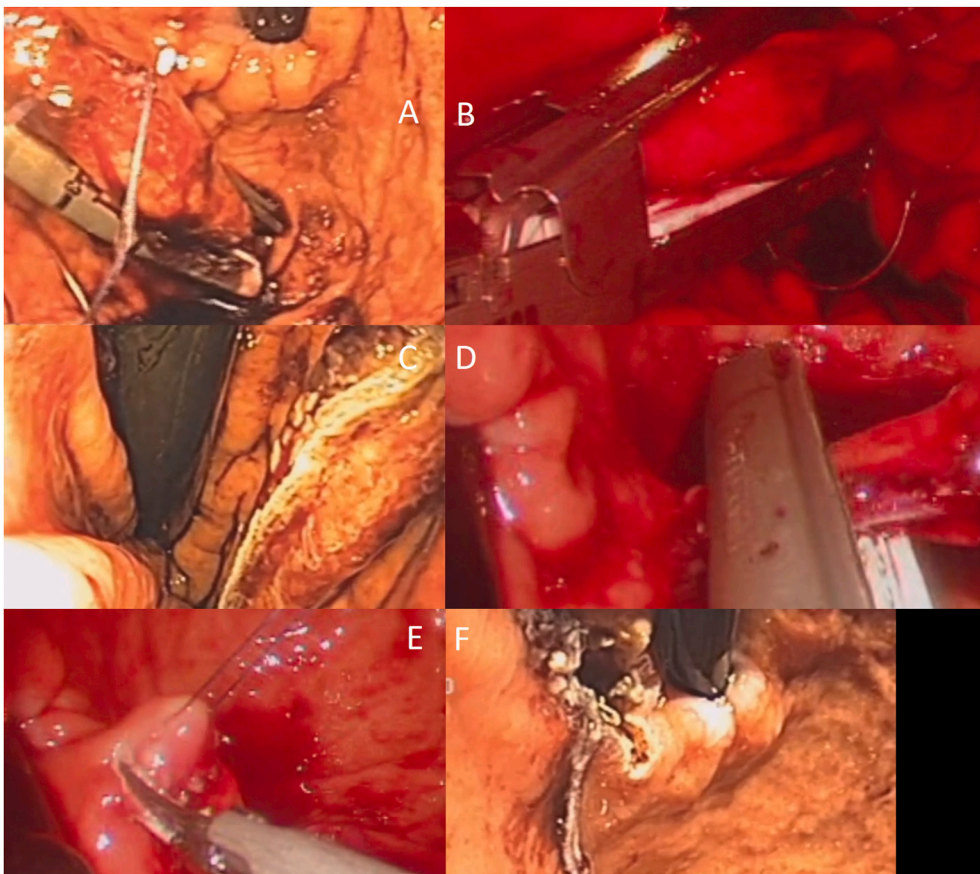


Fig. 1. A: GIST tumor near the esophagogastric junction. B: Endoscopic ultrasound of the GIST tumor.



**Fig. 2.** A: Laparoscopy, the stomach is fixed to the abdominal wall and a gastrostomy is being made using an ultrasound energy device. B: A trocar is placed through the gastrostomy.



**Fig. 3.** Hybrid view of the procedure. A: Endoscopy, the tumor is being grasped by the stapler. B: Laparoscopy, the tumor is being grasped by the stapler. C: Endoscopy, resection is being completed with another stapler. D: Laparoscopy, resection is being completed with another stapler. E: Laparoscopy, resection of the small tumor with an ultrasound energy device while being tractioned with the endoscopic grasper. F: Endoscopy, complete resection of the GISTs tumors.

procedures, or hybrid (laparoscopic-endoscopy) techniques have recently shown promising clinical outcomes, especially in challenging locations [3,4].

In our case, the tumor was located near the esophagogastric junction. This place would have required total or proximal gastrectomy if an intraoperative endoscopy or a trans gastric approach were not performed.

For tumors near the EGJ, early resection is more necessary to avoid a total or a proximal gastrectomy if there is cardia involvement [4,5]. These patients have a high possibility of stenosis or deformity in the gastric inlet with the surgical procedure [5,6]. Therefore total laparoscopic proximal gastrectomy has been proposed by some authors [4,5,7]. Still, this is considered too invasive for the resection of GISTs [8,9].

To solve this issue, Tagaya et al., in 2002 reported the technique of intragastric resection; balloon-type ports were inserted into the gastric lumen via the gastric wall to remove the tumor, yet this equipment wasn't always available and created a difficulty for the surgical team [10–12].

This difficulty can be surmounted with endoscopy in a hybrid procedure; this equipment can help locate the tumor, serve as a traction point, and facilitate simultaneous dissection, among others [11,12].

In metastatic or unresectable cases, imatinib (first-line tyrosine kinase inhibitor) has dramatically improved the management of GISTs. It is also used as Adjuvant therapy in patients in which the tumor is bigger than 2–3cm or complete resection could not be achieved [2,3].

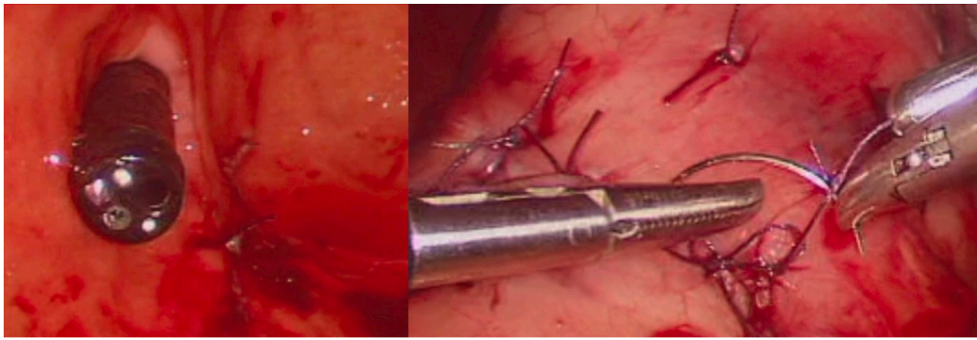


Fig. 4. A: Complete resection with staple line. B: The anterior gastric wall is being sutured.

### 3. Conclusions

Early histologic diagnosis and early surgical resection is currently the most reliable and seems to be the only promising way to improve patients' quality of life and prognosis. When approaching small GISTs or GISTs in troublesome locations, The surgical decision will be the one that will define the future of the patient and, ultimately, his life. Surgeons must have the ability to change strategy and be prepared for these rare and challenging situations.

### 4. Patient perspective

At first, the patient was unsure about her treatment, how long it would last, whether it would hurt, and whether she could be "normal" again, were her main concerns. However, after surgery and seeing the results, her mood improved.

### Declarations

- The submission has not been previously published, nor is it with another journal for consideration
- All authors listed on the paper have reviewed and approved the manuscript in its current form and are aware that it has been submitted.

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No funding was obtained for this study.

### Ethics approval and consent to participate

"This article does not contain any studies with human participants or animals performed by any of the authors."

### Consent for publication

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

### Authors contributions

GM analyzed and interpreted the patient data. WA, CR and AC were a major contributor in writing the manuscript. SE and EP, VB, FG and JJ, revised the manuscript and reviewed all the available data.

All authors read and approved the final manuscript.

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### Availability of data and materials

Not Applicable.

### Declaration of competing interest

The authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest, or non-financial interest in the subject matter or materials discussed in this manuscript. We have no conflict of interest to disclose.

The submission has not been previously published, nor is it with another journal for consideration.

All authors listed on the paper have reviewed and approved the manuscript in its current form and are aware that it has been submitted.

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### Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.amsu.2021.01.022>.

### References

- [1] L. Theiss, C.M. Contreras, Gastrointestinal stromal tumors of the stomach and esophagus, *Surg. Clin.* 99 (3) (2019) 543–553, <https://doi.org/10.1016/j.suc.2019.02.012>.
- [2] K. Akahoshi, M. Oya, T. Koga, Y. Shiratsuchi, Current clinical management of gastrointestinal stromal tumor, *World J. Gastroenterol.* 24 (26) (2018) 2806–2817, <https://doi.org/10.3748/wjg.v24.i26.2806>.
- [3] N.R. Sharma, H. Gopakumar, S. Harrison, N. Ehmke, C. Zelt, Gastric gastrointestinal stromal tumors (GIST): a case series and current state of the art in the workup and treatment of this rare disease, *J. Gastrointest. Canc.* 50 (3) (2017) 548–555, <https://doi.org/10.1007/s12029-017-0034-7>.
- [4] X. Xu, K. Chen, W. Zhou, R. Zhang, J. Wang, D. Wu, Y. Mou, Laparoscopic transgastric resection of gastric submucosal tumors located near the esophagogastric junction, *J. Gastrointest. Surg.* 17 (9) (2013) 1570–1575, <https://doi.org/10.1007/s11605-013-2241-2>.
- [5] S. Shimizu, H. Noshiro, E. Nagai, A. Uchiyama, K. Mizumoto, M. Tanaka, Laparoscopic wedge resection of gastric submucosal tumors, *Dig. Surg.* 19 (3) (2002) 169–173, <https://doi.org/10.1159/000064209>.
- [6] S. Kitano, N. Shiraiishi, Minimally invasive surgery for gastric tumors, *Surg. Clin.* 85 (1) (2005) 151–164, <https://doi.org/10.1016/j.suc.2004.09.004>.

- [7] K. Wakamatsu, E. Lo Menzo, S. Szomstein, Y. Seto, S. Chalikhonda, R.J. Rosenthal, Feasibility of laparoscopic resection of gastrointestinal stromal tumor of the stomach, *J. Laparoendosc. Adv. Surg. Tech.* 28 (5) (2018) 569–573, <https://doi.org/10.1089/lap.2017.0564>.
- [8] K.R. Shen, S.D. Cassivi, C. Deschamps, M.S. Allen, F.C. Nichols, W.S. Harmsen, P. C. Pairolero, Surgical treatment of tumors of the proximal stomach with involvement of the distal esophagus: a 26-year experience with Siewert type III tumors, *J. Thorac. Cardiovasc. Surg.* 132 (4) (2006) 755–762, <https://doi.org/10.1016/j.jtcvs.2006.05.064>, e1.
- [9] H. Ismael, Y. Ragoza, J. Caccitolo, S. Cox, Optimal management of GIST tumors located near the gastroesophageal junction: case report and review of the literature, *International Journal of Surgery Case Reports* 25 (2016) 91–96, <https://doi.org/10.1016/j.ijscr.2016.06.006>.
- [10] N. Tagaya, H. Mikami, H. Kogure, K. Kubota, Y. Hosoya, H. Nagai, Laparoscopic intragastric stapled resection of gastric submucosal tumors located near the esophagogastric junction, *Surgical Endoscopy And Other Interventional Techniques* 16 (1) (2001) 177–179, <https://doi.org/10.1007/s004640080158>.
- [11] N. Hiki, Y. Yamamoto, T. Fukunaga, T. Yamaguchi, S. Nunobe, M. Tokunaga, Y. Seto, Laparoscopic and endoscopic cooperative surgery for gastrointestinal stromal tumor dissection, *Surg. Endosc.* 22 (7) (2007) 1729–1735, <https://doi.org/10.1007/s00464-007-9696-8>.
- [12] W. Xiong, J. Zhu, Y. Zheng, L. Luo, Y. He, H. Li, W. Wang, Laparoscopic resection for gastrointestinal stromal tumors in esophagogastric junction (EGJ): how to protect the EGJ, *Surg. Endosc.* 32 (2) (2017) 983–989, <https://doi.org/10.1007/s00464-017-5776-6>.
- [13] for the SCARE Group R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, The SCARE 2020 guideline: updating consensus surgical Case Report (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.

### List of abbreviations

*Gastrointestinal stromal tumors:* (GISTs)  
*Esophagogastric junction:* (EGJ)  
*Endoscopic ultrasound:* (EUS)  
*Computed tomography:* (CT)