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Case Report

Primary Gastro Hepatic Recess Gastrinoma: A Rare Extra Pancreatic And Extra Intestinal Location Of Gastrinoma.

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Abstract

Introduction : Gastrinomas are rare neuroendocrine tumors that are typically located within the gastrinoma/Zollinger-Ellison Syndrome (ZES) triangle. However, rare cases have been reported in locations such as the body of the stomach, jejunum, peripancreatic lymph nodes, splenic hilum, omentum, liver, gallbladder, common bile duct, and ovary. The average reported size of a gastrinoma is generally less than 4 cm. Gastrinomas in the gastrohepatic recess are extremely rare, with no cases reported to date. We present a case of a gastrinoma originating from an ectopic site with an unusually large size.

Case Report : A 23-year-old male trainee athlete presented with reflux symptoms, vomiting, loose stools, and altered bowel habits for the past 18 years. He had been regularly taking proton pump inhibitors (PPIs) for over 10 years. He had also undergone exploratory laparotomy 7 years ago for a perforated gastric ulcer, though the operative record was unavailable. Initial abdominal ultrasound revealed an epigastric mass, which was further investigated with CT and MRI scans. These imaging studies showed a multilobulated, heterogeneously enhancing mass with central necrosis and multiple calcific foci in the upper abdomen, located in the gastrohepatic recess. The scans also identified multiple variable-sized simple hepatic cysts. An ultrasound-guided core biopsy of the lesion confirmed a Grade I neuroendocrine tumor. Based on this, the patient's serum gastrin levels were found to be >1800 pg/ml (more than 10 times the normal value). The case was discussed in a multidisciplinary team (MDT) meeting, and surgery was recommended. The patient underwent surgical excision of the gastrohepatic recess mass, measuring 10 x 6 cm. No distant metastasis was observed during the surgery. The final histopathology confirmed a Grade I neuroendocrine tumor. Post-surgery, the patient's serum gastrin levels dropped to 189 pg/ml immediately and 79 pg/ml five days after surgery.

Conclusion : In this case, the gastrinoma was located in an extremely rare site, and the mass was unusually large. The mainstay treatment of any well localized/non metastatic gastrinoma is surgical resection like in our case, irrespective of size and rare location.

Keywords : *GaStrinoma, ZES, Surgical resection, Gastrin, Gastrohepatic recess, ectopic location.*

INTRODUCTION

Rare neuroendocrine tumors known as gastrinomas develop from the duodenal mucosa and the pancreas, specifically from non-beta islet cells [1]. Peptic ulcers and an overabundance of stomach acid are caused by these tumors' overproduction of the hormone gastrin. Zollinger-Ellison syndrome (ZES), a hypergastrinemic disease, is characterized by numerous recurrent and frequently resistant to treatment gastrointestinal (GI) tract ulcers. Recurrent peptic ulcers, increased stomach acid production, and a non-beta islet cell tumor in the pancreas are the three classic ZES triads.

The "gastrinoma triangle," which is made up of the intersection of the common and cystic bile ducts, the second and third parts of the duodenum, and the junction of the pancreatic neck and body, is where primary gastrinomas are most frequently discovered [2]. Ectopic gastrinomas are those that are found outside of this triangle. A primary gastrinoma in an ectopic position, such as the stomach body, jejunum, peripancreatic lymph nodes, splenic hilum, omentum, liver, gallbladder, or ovary, affects about 5.6% of patients [3]. There are currently no documented occurrences of a primary gastrinoma in the gastrohepatic recess, making it extremely uncommon. A gastrinoma usually has a size of less than 4 cm.

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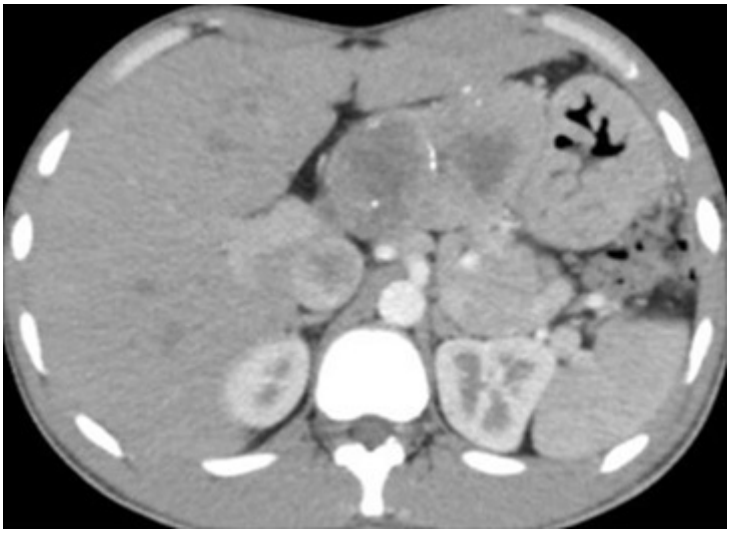
It can be difficult to suspect and diagnose a gastrohepatic recess tumor of extraordinary size as a gastrinoma because of the scant evidence, especially in the preoperative situation.
Histological examination of the removed sample is typically used to confirm the diagnosis after surgery. Computed tomography (CT), somatostatin receptor scintigraphy, and upper gastrointestinal endoscopy are among the diagnostic techniques used to determine whether the primary tumor or potential metastases from other intra-abdominal organs are present [3,4].Based on a combination of symptoms, imaging, histology, and serum gastrin levels, a rare instance of a gastrohepatic recess mass with an unusual size and position was suspected and identified prior to surgery.

CASE REPORT

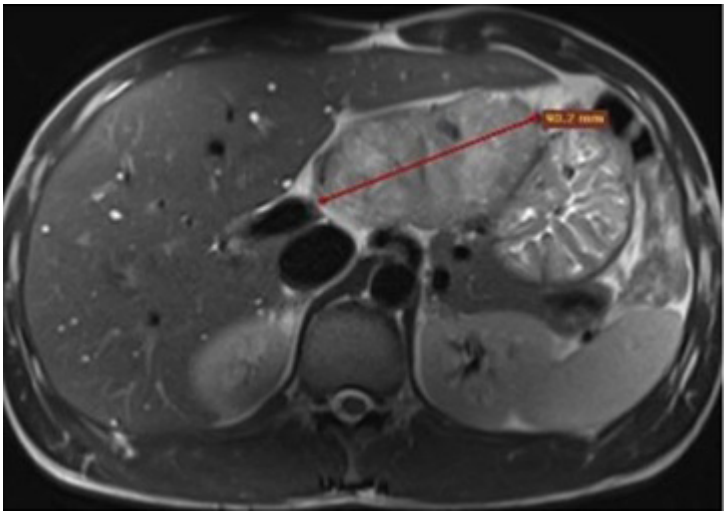
A 23-year-old male trainee athlete presented with reflux symptoms, vomiting, loose stools, and altered bowel habits for the past 18 years. He had been taking proton pump inhibitors (Omeprazole) daily to manage his symptoms. However, he experienced severe vomiting within 24 hours of missing a dose, which led to a hospital visit for intravenous fluid administration. His family history included a maternal aunt diagnosed with esophageal cancer. Ultrasound (US) and computed tomography (CT) scans revealed an epigastric mass, and biopsy and resection were recommended. However, his parents declined the procedure at that time. The patient had undergone exploratory laparotomy seven years earlier for a gastric ulcer perforation and remained hospitalized for one week, though the surgical records were unavailable.
Persistent symptoms led the patient to our clinic, where a well-healed transverse surgical scar was noted on examination in the right upper abdomen. No obvious visceromegaly was observed.
To investigate the cause of his symptoms, a CT scan of the chest, abdomen, and pelvis was performed, revealing a multilobulated, heterogeneously enhancing mass with centrally necrotic areas and multiple calcific foci in the upper abdomen within the gastrohepatic recess. Additionally, multiple bilobar hepatic hypodense lesions were identified, likely benign simple cysts, and mild splenomegaly was noted.

Figure 1. CT scan shows heterogeneously enhancing centrally necrotic mass in the gastrohepatic recess.



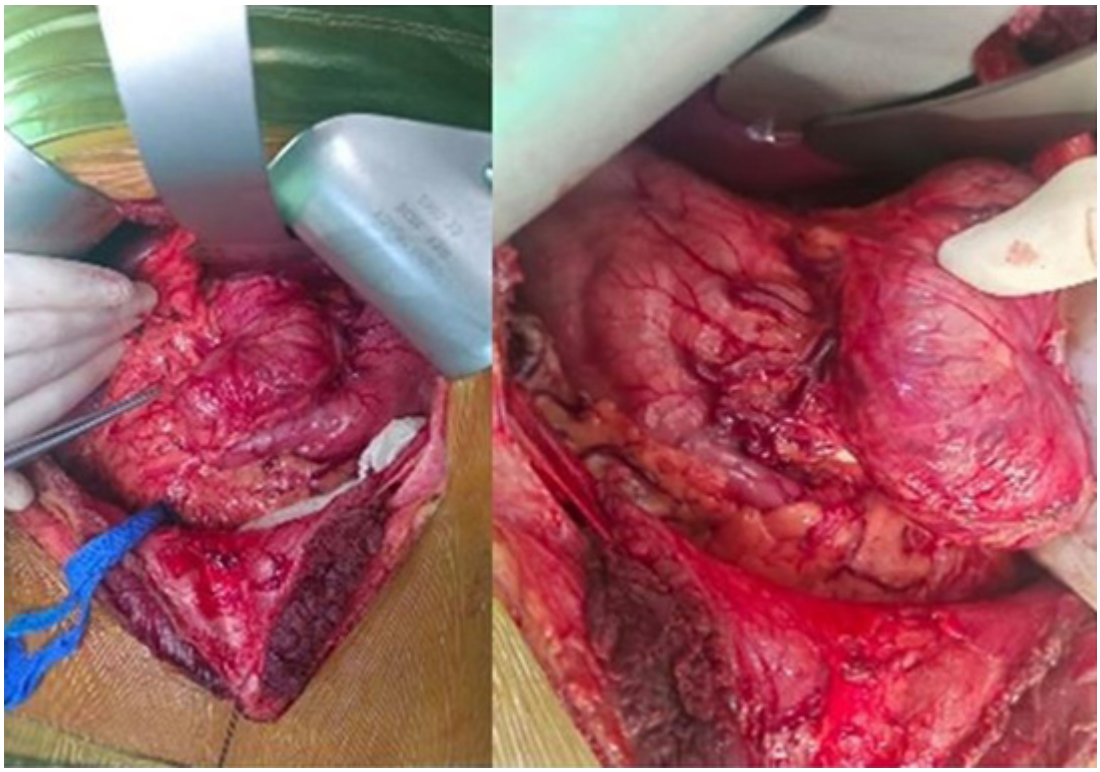
An ultrasound-guided core biopsy of the mass confirmed the diagnosis of a Grade I neuroendocrine tumor (NET). The case was discussed in a multidisciplinary team (MDT) meeting, and the recommendations included performing endoscopic ultrasound (EUS) and magnetic resonance imaging (MRI) of the abdomen to assess the hepatic lesions before proceeding with surgery.

Figure 2. MRI demonstrates loculated abnormal signal intensity lesion in the region of gastrohepatic recess.



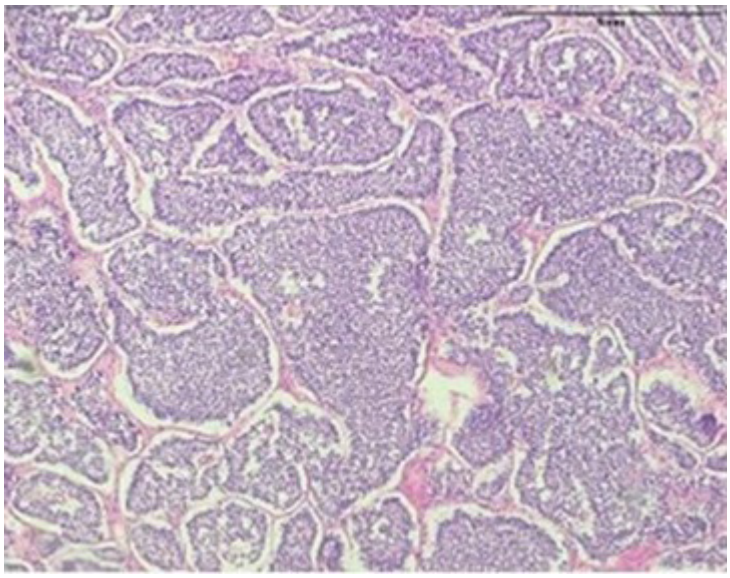
The MRI demonstrated a stable-sized biopsy-proven neuroendocrine tumor, along with multiple benign hepatic cysts and no suspicious hepatic lesions. EUS revealed a large, bilobed mass in the gastrohepatic area, measuring 75 x 52 mm. The mass was primarily solid, with cystic and necrotic areas, and scattered calcifications. While the coeliac artery was unaffected, other vessels could not be clearly identified due to the mass's size.

Figure 3. Intraoperative image showing mass in the Gastrohepatic recess.



Serum gastrin levels were found to be >1800 pg/ml. The patient then underwent surgery for excision of the gastrohepatic recess mass via a rooftop incision. Intraoperative findings revealed a 10 x 6 cm, bean-shaped, extra-pancreatic, extra-intestinal mass in the gastrohepatic recess, which was adherent to the stomach and pancreas with flimsy adhesions. Intraoperative ultrasound showed no pancreatic or duodenal lesions, although multiple suspicious liver lesions, mostly cystic, were noted. A representative lesion from segment 4b was excised and sent for histopathology. The surgery lasted 2 hours, with minimal blood loss (30 mL).

Figure 4. Histopathology sections show neoplastic proliferation of cells with organoid and nested arrangement.



The final histopathology confirmed a Grade I neuroendocrine tumor for the gastrohepatic mass, while the liver lesions were benign.

Post-surgery, serum gastrin levels dropped to 189 pg/ml immediately and 79 pg/ml five days after surgery.

Given the unusual size and malignant potential of the gastrinoma, a Gallium-68 DOTA-PET/CT scan was performed, which showed no avidity at the surgical site or elsewhere. On follow-up, the patient was doing well, with resolution of his symptoms, and had resumed his regular activities.

DISCUSSION

The unusual instance of a gastrinoma discovered in an ectopic position and its surgical treatment are highlighted in this article. The incidence of gastrinoma, a rare neuroendocrine tumor (NET), ranges from 0.1 to 15 incidences per million people worldwide [4]. After insulinomas, it is the second most prevalent form of NET. With the pancreas being the most common site of occurrence, followed by the duodenum, these tumors usually cause excessive gastrin secretion. The "gastrinoma triangle," which is surrounded by the pancreatic neck/body medially, the second and third portions of the duodenum inferiorly, and the junction of the cystic and common bile ducts posteriorly, is where the majority of gastrinomas are found during surgery [5].

There are two categories of gastrinomas: those linked to multiple endocrine neoplasia type-1 (MEN-1) and spontaneous gastrinomas (SG), which account for 75% of all occurrences [6]. Although the majority of gastrinomas grow slowly, 50–60% are malignant and have spread by the time they are diagnosed. Usually, the liver and local lymph nodes are where metastasis starts. Excessive stomach acid output is the main cause of symptoms in gastrinoma patients [7]. Abdominal discomfort is the most prevalent symptom, and it is followed by vomiting, diarrhea, heartburn, and gastrointestinal bleeding.

For the detection of gastrinomas, a variety of imaging methods are necessary, including somatostatin receptor imaging (SRI), endoscopic ultrasound, cross-sectional imaging (CT and MRI scans), and ultrasonography. However, histological and immunohistochemical analysis are necessary for the final diagnosis. Positive immunostaining for synaptophysin and chromogranin A (CgA) is one of the primary diagnostic characteristics of NETs. Gastrin immunostaining also aids in differentiating gastrinoma from other pancreatic NETs [8].

Proton pump inhibitors (PPIs) and other oral antisecretory drugs are advised for medical care in order to regulate the hypersecretion of stomach acid observed in individuals with Zollinger-Ellison syndrome (ZES) [9].

For patients with Zollinger-Ellison syndrome (ZES), surgical excision of the localized tumor and metastatic lymph nodes is the recommended course of treatment.(10) Depending on the size and location of the tumor, either a laparotomy or a laparoscopy can be used to execute this surgery. Following surgery, positron emission tomography (PET) should be performed 18–24 months after the procedure, and CT or MRI scans should be performed every 3–6 months [4].

CONCLUSION

Gastrinomas can vary in size and arise in ectopic locales, necessitating careful examination and therapy. As we showed in our instance, surgical excision is the main treatment for any well-localized, non-metastatic gastrinoma, regardless of its size or peculiar location.

ABBREVIATIONS

ZES Zollinger-Ellison Syndrome
PPI proton pump inhibitors
GI gastrointestinal
CT computed tomography
NET neuroendocrine tumor
MDT multidisciplinary team
EUS endoscopic ultrasound
MRI magnetic resonance imaging
MEN multiple endocrine neoplasia
CgA chromogranin A
PET positron emission tomography

AUTHOR'S CONTRIBUTIONS

Dr Sayed Moosa Kazim: Conception of the work AND Design of the work.
Dr Zulqarnain Hyidar: Drafting the work AND Revising the work critically for important intellectual content.
Dr Fatima Tu Zahara: Drafting the work AND Revising the work critically for important intellectual content.
Dr Zain Tayyab: Conception of the work AND Design of the work.
Dr Muhammad Awais: Drafting the work.
Dr Aamir Ali Syed: Final approval of the version to be published AND Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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