A rare cause of upper gastrointestinal bleeding: Primary gastrinoma of the lesser omentum

Une cause rare hémorragie digestive : un gastrinome primitif du petit épiploon

Gastrinomas are rare neuroendocrine tumors (NET) characterised by an abnormal secretion of gastrin resulting in a clinical syndrome known as Zollinger-Ellison syndrome (ZES). Diagnosis is often delayed and can be missed as symptoms lack specificity. More than 90% of primary gastrinomas classically occur in the pancreas and duodenum within the borders of the gastrinoma triangle. However, other ectopic localisations have been reported such as jejunum, splenic hilum, root of the mesentery, liver, gallbladder, lymph nodes and ovary [1]. To the best of our knowledge, only five cases of gastrinomas arising from the lesser omentum have been reported in literature [2-5]. Our case was about a patient with ZES due to primary gastrinoma of the lesser omentum with gastrointestinal bleeding as the revealing manifestation.

A 26-year-old male patient with no past medical history presented with melena and epigastric pain. Hemoglobin measured 5 g/dL. The patient was admitted to the intensive care unit. He was treated with intravenous proton pump inhibitors (PPI) and received a blood transfusion. Once hemodynamic status was stabilized, esophago-gastroduodenoscopy (EGD) was performed. It showed moderate oesophagitis, erosive gastritis, and multiple duodenal ulcers. Colonoscopy was normal. Fasting serum gastrin (FSG) level after tapering PPI doses was moderately elevated at 306 pg/mL (normal < 121 pg/mL) and chromogranin A level was also elevated at 704 ng/mL (normal < 94 ng/mL). Serum calcium, phosphorous, parathyroid hormone, and prolactin levels were all within the normal ranges. Magnetic resonance imaging was performed and showed a solitary 4.1 cm × 3.5 cm well-circumscribed mass in the lesser omentum with no evidence of adjacent organ invasion (figure 1). Whole body somatostatin-receptor scintigraphy showed a focus of markedly increased octreotide uptake in the lesser omentum corresponding to the lesion seen on magnetic resonance imaging with no evidence of tumors at any other sites. Primary gastrinoma of the lesser omentum was suspected. An exploratory laparotomy was performed. At surgery, a 5 × 4 cm lesion close to the lesser curvature of the stomach was identified (figure 2). Thorough bimanual palpation of the pancreas, duodenotomy with exploration of the duodenal mucosa and regional lymph node dissection did not identify evidence of alternative primary sites of the gastrinoma or evidence of metastases. The tumor was totally resected. Pathology revealed a well-differentiated NET grade 1 (mitotic index < 1; Ki67 < 2%) consistent with gastrinoma. The excision margins were clear. Stains were positive for synaptophysin and chromogranin (figure 3). Postoperative course was uneventful. The patient remained symptom-free and showed normal gastrin level after 6 months of follow-up.

Discussion

The common clinical manifestations of gastrinoma include abdominal pain, secretory diarrhea and heartburn [6]. Gastrointestinal

Figure 1
Abdominal magnetic resonance image showing a 4.1 cm × 3.5 cm well-circumscribed mass in the lesser omentum (red arrow). This mass was of low signal intensity on T1-weighted sequence (a) and high central signal intensity on T2-weighted sequence (b) with moderate enhancement after gadolinium injection (c)
**FIGURE 2**
Operative photo showing a well-defined 5 x 4 cm solitary mass in the lesser omentum attached to the lesser curvature of the stomach.

**FIGURE 3**
Microscopic images of the tumor. (a) Hematoxylin and eosin stain (×100). The tumor cells were arranged in an insular-type pattern within a highly vascular stroma (b) Hematoxylin and eosin stain (×400). The tumor had relatively uniform cells having centrally located nuclei and eosinophilic cytoplasm with rare mitoses. Immunohistochemical stains (×400) were positive for chromogranin (c) and synaptophysin (d).
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bleeding secondary to duodenal ulcers is the presenting symptom in 25% of patients. The diagnosis of ZES can be established in patients with an appropriate clinical setting and high FGL in the presence of raised gastric acid production. If FGL is not very high (<10-fold), achlorhydric conditions including atrophic gastritis and pernicious anemia should be excluded and then a secretin stimulation test should be performed [7]. Although most guidelines recommended proton pump inhibitors (PPIs) be stopped for at least a week prior to gastric analysis, abrupt PPI discontinuation can cause complications due to acid hypersecretion [6]. Thus, to make diagnosis of ZES, cautious gradual tapering of PPI before biochemical testing has been suggested. In our patient, FGL was not very high. Therefore, provocative tests should have been performed.

Gastrinomas are, in 75% of the cases, sporadic or occur in patients with multiple endocrine neoplasia (MEN) type 1 in association with pituitary adenoma and parathyroid tumours [8]. Gastrinomas can be detected with a computed tomography scan or magnetic resonance imaging [9]. However, their sensitivity depends on the gastrinoma’s size and location. The octreoscan is widely used to localise gastrinomas, especially to detect metastasis.

Medical therapy for ZES consists primarily of acid suppression therapy with high-dose proton pump inhibitors. Surgical resection represents the main curative treatment. For patients who underwent a complete surgical resection of a non-metastatic gastrinomas, the 15-year survival rate is >80% [10]. However, in patients with metastatic gastrinomas, the 5-year survival rate ranges from 20% to 38% [11]. Even after complete surgical tumor removal, regular follow-up with clinical and imaging surveillance is obligatory to detect disease recurrence.

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**References**


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