Case Report

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Urgent Rare Surgical Complication Assessment of Intestinal Gastrointestinal Stromal Tumors

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Gastrointestinal stromal tumors (GISTs) are rare mesenchymal subepithelial tumors originating from abnormal proliferation of interstitial cells of Cajal, with worldwide incidence of about 1–2 per 100,000. Herein, we report an unusual case of a 55-year-old man who presented a severe digestive hemorrhage as a rare post-surgical complication after intestinal GIST surgical removal. The patient was admitted to the Emergency Department of our center affected by abdominal epigastric pain. Different imaging techniques were performed leading to the final diagnosis of a GIST and surgical intervention planning. Immediately after intervention the patient developed a severe intestinal hemorrhage. Multidetector computed tomography (MDCT) confirmed the ongoing bleeding and the patient underwent a new intervention to control the hemorrhage. The aim of the paper is to show the different imaging techniques used to assess GIST. MDCT represents the gold standard for diagnosis and in the emergency setting is used to identify post-surgical complications.

Keywords: *Gastrointestinal Stromal Tumors (GIST); Imaging Techniques; Computed Tomography (CT); Magnetic Resonance Imaging (MRI); Surgical Favorable Outcome*

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INTRODUCTION

Gastrointestinal stromal tumors (GISTs) are rare mesenchymal subepithelial tumors originating from abnormal proliferation of interstitial cells of Cajal [1–3]. GISTs can develop anywhere in the gastrointestinal (GI) tract; the most frequent localizations are within the stomach (40–70%) and small intestine (15–44%) while less frequently they may be located in the rectum (5%) or esophagus (<1%) [4,5]. The symptoms most commonly associated with GISTs are anemia, abdominal pain, dyspepsia, nausea or vomiting, constipation, or diarrhea. Nevertheless, 25% of GISTs are discovered incidentally in asymptomatic patients. In some cases, the symptoms may be more severe, such as digestive bleeding, perforation, or bowel obstruction, requiring emergency surgical treatment [6–10].

Treatment is mainly surgical. Radiology plays a key role not only in diagnosis, but also in monitoring the effects of treatment, in case of complications, and during follow-up [11,12]. Computed tomography (CT) with contrast agent administration is currently the imaging technique of choice, although other methods such as positron emission tomography (PET) with florodeoxyglucose (FDG) and magnetic resonance imaging (MRI) can also be used [13].

Herein, we report an unusual case of a 55-year-old man who presented a severe digestive hemorrhage as a rare post-surgical complication after intestinal GIST surgical removal. The aim of the paper is to show the different imaging techniques used to assess GIST. Multidetector computed tomography (MDCT) represents the gold standard for diagnosis and in the emergency setting is used to identify post-surgical complications.

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CASE REPORT

A 55-year-old man presented to the Emergency Department of our center (Ospedale del Mare, Naples, Italy) with severe abdominal epigastric pain for 24 hours. On physical examination, clinical parameters were stable, and the abdomen was painful in the mesogastric and left hypochondriac regions. Laboratory examinations and complete blood count (CBC) revealed neutrophilia 87.3% (40–75%), lymphopenia 6.7% (19–48%), increased D-dimers 1,001 µg/L (10–500 µg/L), and a mild increase in C-reactive protein (CPR) 0.6 mg/L (0–0.5 mg/L). The values in the brackets represent the reference values of our laboratory.

The patient underwent an abdominal X-ray as a first-level imaging technique, which showed overdistention of the intestinal loops in the umbilical and left lumbar regions with associated pathological hydro-aerial levels (Figure 1). In order to investigate the X-ray findings, a contrast-enhanced abdominal CT scan was performed, which showed distention of the proximal jejunal loops with hydro-aerial levels, a finding indicative of bowel sub-occlusion. MDCT showed a 15 mm solid lesion, located in a jejunal loop downstream of the occlusion, characterized by intense enhancement in the arterial phase and wash-out in the subsequent phases, indicative of a GIST (Figure 2). In addition, two

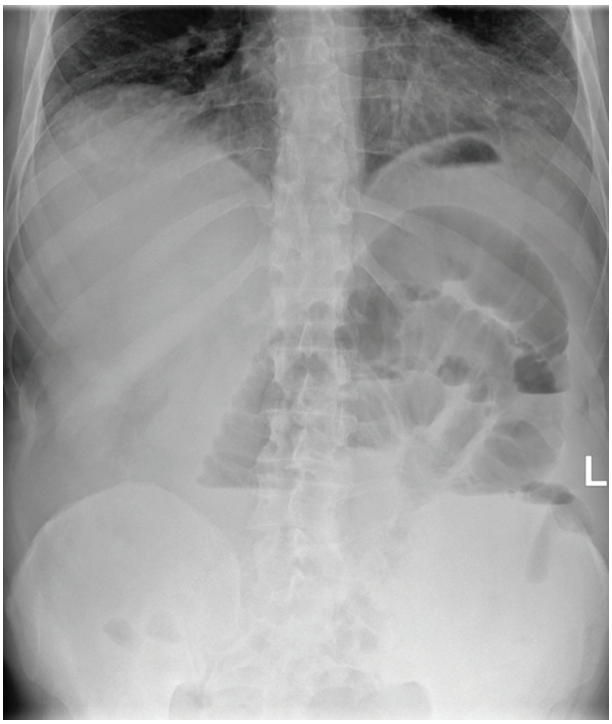

Figure 1 Abdominal X-ray at admission: overdistention of intestinal loops in the umbilical and left lumbar regions with associated pathological hydro-aerial levels.

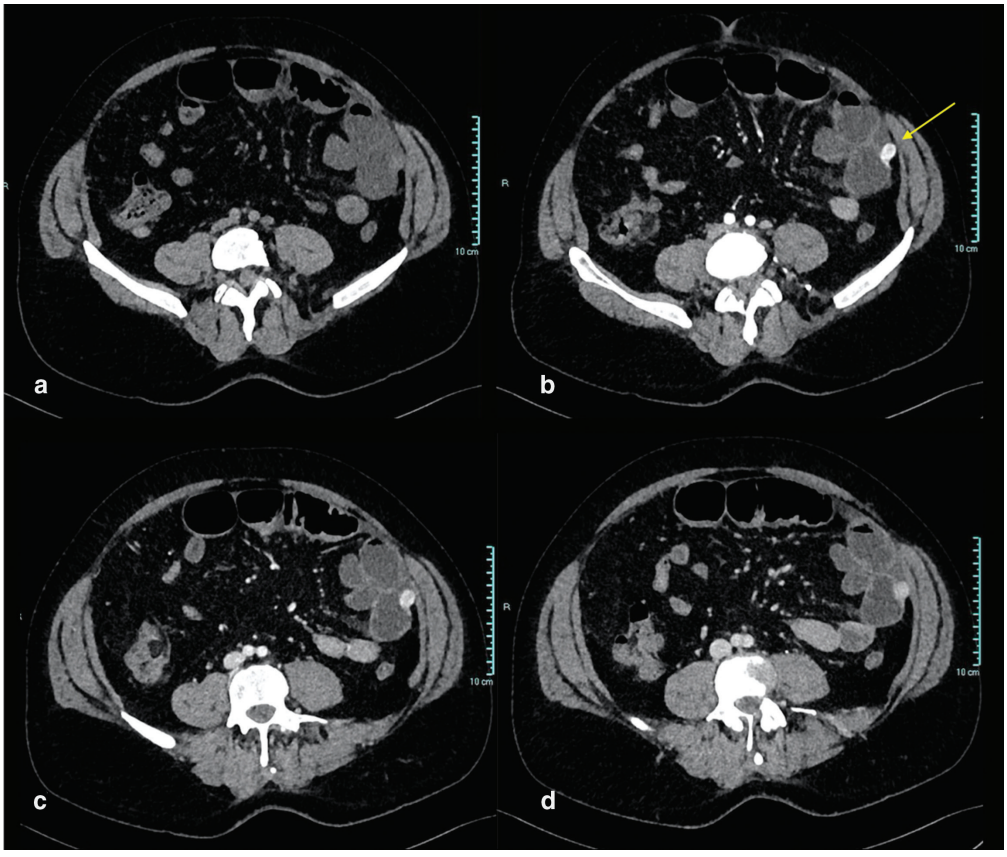

Figure 2 Contrast-enhanced abdominal CT scans, in the axial plane, for non-contrast (**a**), arterial (**b**), venous (**c**), and delayed (**d**) phases. The CT scans showed a 15 mm solid lesion in the wall of a jejunal loop (arrow) with exophytic growth, intense enhancement in the arterial phase (**b**), and wash-out in the subsequent phases (**c**,**d**), indicative of GIST.

analogous smaller solid lesions were evident in other jejunal loops (Figure 3). MRI was then performed confirming a GIST tumor (Figure 4). The MRI was performed with a 1.5 Tesla device, using both an oral biphasic contrast agent, polyethylene glycol (PEG), with a low signal in T1-weighted sequences and a high signal in T2-weighted sequences, and the intravenous contrast agent Gadolinium. Magnetic resonance (MR) enterography confirmed the presence of three jejunal GIST lesions (Figure 5).

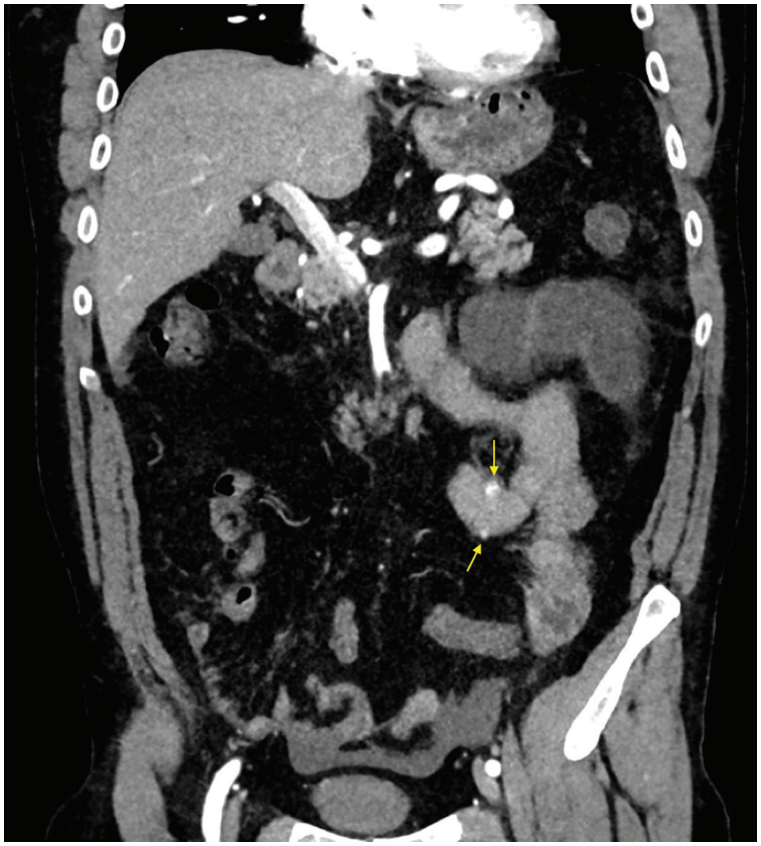

Figure 3 Contrast-enhanced abdominal CT scan, in the coronal plane, with arterial phase. The CT scan showed two lesions in the wall of a jejunal loop (arrows) with exophytic growth and intense enhancement, indicative of GIST.

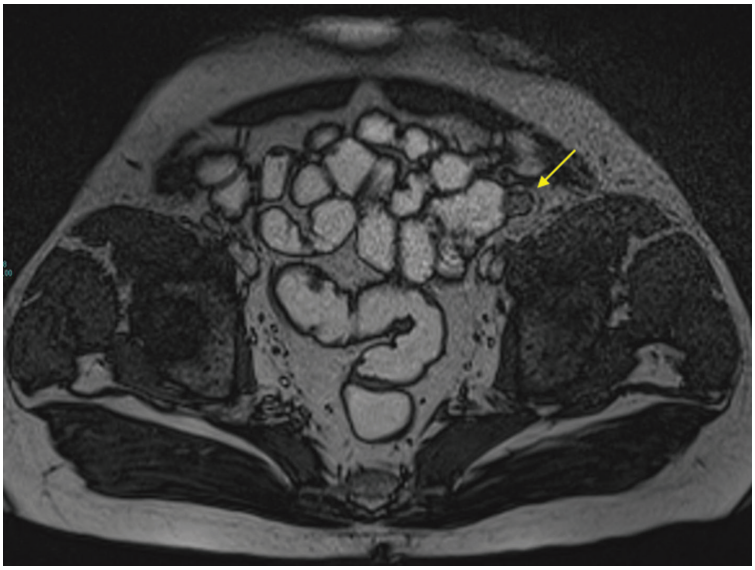

Figure 4 MR enterography, with oral biphasic contrast agent polyethylene glycol (PEG), in the axial plane. MR enterography showed a hypointense lesion in the wall of a jejunal loop (arrow) with exophytic growth, indicative of GIST.

Thus, the patient underwent two surgical interventions with a wedge resection-type excision, the first of the largest parietal exophytic lesion located in the proximal jejunum, and the second of the remaining two smaller parietal nodules also located in the proximal jejunum upstream of the previous one. Finally, segmental resection of the affected loops of the jejunum was performed with subsequent mechanical latero-lateral anastomosis.

Within a few hours of surgery, the patient began to show signs of hypotension, appearing pale and sweaty, and laboratory tests showed a sharp drop in hemoglobin of more than 5 g/dL, from 15.6 g/dL (pre-intervention values) to 10.5 g/dL (post-intervention values), with initial signs of hypovolemic shock. Therefore, the patient underwent a repeated contrast-enhanced abdominal CT scan in the emergency setting. The CT scan showed a large blush of contrast agent in the arterial phase, increasing in the later phases in the lumen of a jejunal loop in the left lumbar region, a finding indicative of severe active bleeding and associated with profuse blood in the lumen

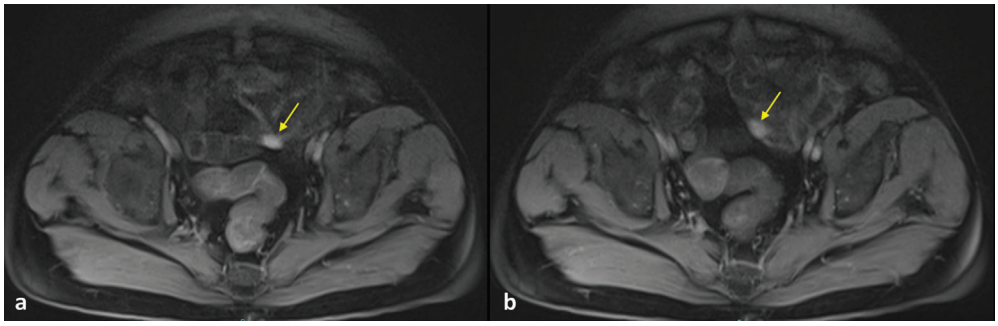

Figure 5 MR enterography, with oral biphasic contrast agent polyethylene glycol (PEG) and Gadolinium by intravenous administration, in the axial plane, with arterial (**a**) and venous (**b**) phases. MRI confirmed the presence of the jejunal GIST lesions with exophytic growth and intense enhancement in the arterial phase (**a**) and wash-out in the venous phase (**b**).

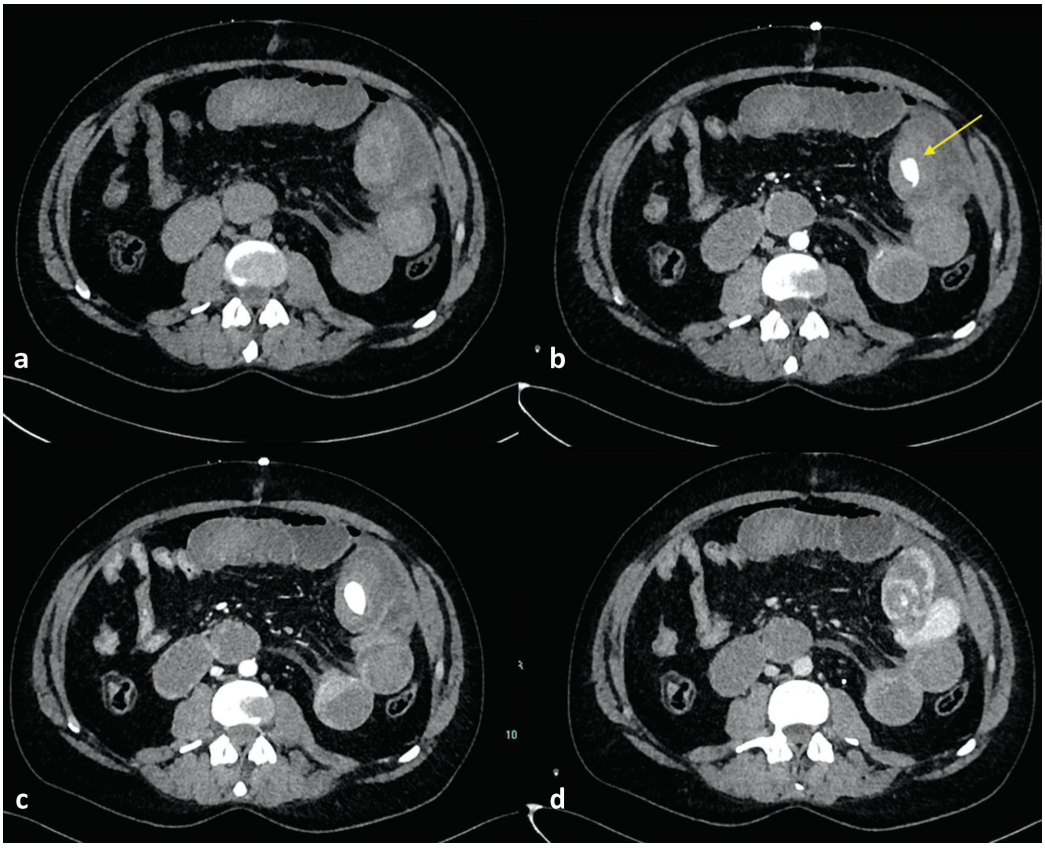

Figure 6 Contrast-enhanced abdominal CT scan, in the axial planes, with non-contrast (**a**), arterial (**b**), venous (**c**), and delayed (**d**) phases. The CT scans showed a large blush of contrast agent in the arterial phase (**b**, arrow) increasing in the later phases (**c**,**d**) in the lumen of a jejunal loop in the left lumbar region, indicative of severe active bleeding and associated with profuse blood in the lumen of multiple jejunal loops downstream (**d**). The CT scan also showed hemoperitoneum adjacent the jejunal loops in the left lumbar region.

of multiple jejunal loops downstream. The CT scan also showed diffuse hemoperitoneum (Figure 6). The patient underwent emergency surgery again with reopening of the recent laparotomy, confirming predominantly localized hemoperitoneum in the resection and perianastomotic sites, and evidence upstream of the jejunal loop of resection/anastomosis of profuse active bleeding. Bleeding control was performed with subsequent resection of approximately 20 cm of the small bowel including the bleeding site and packing of a new downstream latero-lateral anastomosis.

Ethical Approval and Informed Consent

Ethical approval to report these cases was not required. Informed consent was not acquired. All the data referring to the patient were anonymized.

DISCUSSION

GISTs are rare tumors originating from the interstitial cells of Cajal in the muscularis propria, although they are considered the most common mesenchymal tumor of the GI tract (incidence worldwide of 1–2 per 100,000). They arise from abnormal proliferation of interstitial cells of Cajal that are electrical pacemakers and mediators of enteric neurotransmission in the muscularis propria of the GI tract, with 95% of the tumors staining positive for CD117 (c-KIT) and 70% for CD34 [14], in most cases caused by oncogenic activating mutation of the receptor tyrosineproteinkinase [1–3]. Their incidence is very uncommon when compared to GI carcinoma [15] and they usually occur after 40 years of age, most commonly in older patients (median age 60–65 years) with a slight male predilection [9,10]. They can originate throughout the GI tract, most commonly in the stomach (40–70%), followed by the small bowel (15–44%) and rarely the omentum, mesentery, and retroperitoneum [4,5]. Histopathological evaluation is essential for both diagnosis and risk stratification in patients affected by GISTs [13].

The clinical presentations of GISTs vary according to their site, size, and biological behavior: they may present with non-specific GI symptoms (e.g. abdominal pain, nausea, and vomiting) [10], and small tumors are more likely to be asymptomatic, regardless of location, and found incidentally [10]. If they are large tumors they can cause bowel obstruction, intussusception or ulceration, and cause GI hemorrhage [9]: up to 40% of cases manifest with GI bleeding, ranging from chronic anemia to uncommon acute massive GI bleeding [6–8]. Aggressive forms may present with metastases or symptoms relating to local disease [9]. A peculiarity of small intestine GISTs is a higher incidence of bleeding compared with gastric GISTs [16,17]. The tendency of GISTs to bleed is due to the high tumor vascularity, progressive destruction of the mucosa, and tumor invasion of blood vessels, leading to vascular rupture [18].

Multiple imaging modalities can be used to evaluate a tumor's local extent, determine staging, predict risk, conduct surveillance after surgery, and monitor the response to molecularly targeted therapy [13]. Ultrasonography (US) is usually the first-line technique for the investigation of a patient with abdominal pain or palpable abdominal mass. Most commonly, US alone is not diagnostic because GISTs are frequently discovered to be so large that the organ of origin cannot be identified, and they present as heterogeneous masses due to the presence of necrosis, hemorrhage, or cystic changes; liver metastases can also be detected [19,20]. Abdominal X-ray is not diagnostic for the presence of a GIST but may show indirect signs such as overdistention of the loops with hydro-aerial levels as per a sub-occlusive or occlusive state [21]. Contrast-enhanced MDCT is the gold-standard technique for the diagnosis of a GIST, defining its exact location, growth type (exophytic, intraluminal, and mixed/combined/endophytic), size, and features, and whether it has metastasized to other organs [13]. The CT scan is also fundamental to define surgical planning, postsurgical surveillance, and monitoring of therapy response [22]. GISTs are defined as small when the lesion size is <2 cm, and large when the lesion size is >5 cm: small GISTs are usually homogeneous in content, while large GISTs are usually more inhomogeneous due to the presence of hemorrhagic and necrotic areas [13]. In larger lesions, often an afferent arterial vessel is observed due to the phenomena of neoangiogenesis [23,24]. Generally, the growth pattern of GISTs is categorized as exophytic, intraluminal, and mixed/combined/endophytic [13]: exophytic (54%) and mixed growth patterns (39%) are common in the small bowel [25]. On MRI, GISTs typically show low signal intensity on T1-weighted and high signal intensity on T2-weighted sequences, and enhanced signal intensity on post gadolinium images, especially in the arterial phase [26]. MRI provides morphological imaging findings similar to those obtained from a CT scan; additionally, quantitative parameters such as the apparent diffusion coefficient (ADC) and degree of enhancement, and perfusion parameters are helpful in assessing the malignancy and response to treatment [27–31]. FDG PET is more sensitive than morphological imaging in evaluating therapy response [22].

Primary management of small localized primary disease is surgical resection, while large GISTs or locally advanced GISTs may benefit from neoadjuvant Imatinib followed by surgery; for metastatic and recurrent GISTs, Imatinib remains the mainstay of treatment [11,12]. Major postoperative complications include anastomotic leakage, prolonged ileus, infection, delayed perforation, tumor rupture with intraperitoneal seeding, and blood loss [32–35]. Rarely postoperative bleeding can be potentially life-threatening: Ihn et al. report that of 95 patients undergoing surgical resection of a small bowel GIST, 13 patients (13.7%) had postoperative

complications and 1 patient (1.1%) died from postoperative hemorrhage [36]. The incidence of complications has been shown to be lower in laparoscopy than in the open technique [33; 36,37]. Postoperative bleeding is one of the surgical factors associated with a higher risk of recurrence, along with incomplete resection, intraperitoneal rupture, and tumor associated factors such as tumor size, mitotic index, or localization [38–40].

In our case, the diagnosis was made on hospital admission by contrast-enhanced CT, which showed three jejunal lesions indicative of a GIST that was further confirmed by MRI. Our patient subsequently underwent surgery but there was an early complication of bleeding, which was quickly recognized by further abdominal contrast-enhanced CT examination. Thus, the patient underwent a life-saving reoperation.

CONCLUSIONS

To conclude, in this report we have presented an unusual case of a patient affected by jejunal GIST that presented a rare postoperative complication, a potentially life-threatening event that quickly needed prompt CT diagnosis and further surgery.

The aim of this study is to show the use of the different imaging techniques to assess the diagnosis of a GIST and to highlight the role of contrast-enhanced CT, which represents the gold-standard imaging method not only in the diagnosis of a GIST but also in detecting immediate operative complications such as hemorrhage that, although statistically rare, can be life-threatening for patients. In order to promote the increasing use of contrast-enhanced CT in the emergency setting, this case is reported both for the excellent iconography and the favorable outcome of the patient.

Ethics Statement

(1) All the authors mentioned in the manuscript have agreed to authorship, read and approved the manuscript, and given consent for submission and subsequent publication of the manuscript.
(2) The authors declare that they have read and abided by the JEVTM statement of ethical standards including rules of informed consent and ethical committee approval as stated in the article.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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Author Contributions

All the authors made a substantial contribution to the manuscript and participated sufficiently in submission to take public responsibility for its content. Publication was seen and approved by all authors and by the responsible authorities where the work was carried out.

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