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Case Report

Jejunal perforated gastrointestinal stromal tumor as a rare cause of acute abdomen: case report and review of literature

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ABSTRACT

Gastrointestinal stromal tumors (GIST) are the most common mesenchymal tumor of the GI tract. Most cases are asymptomatic, or present as an abdominal mass. Less frequently, they can present as an upper GI bleed or acute abdomen. We hereby present the case of a 47-year-old male who visits the ER department complaining of abdominal pain and melena, with physical exploration findings of acute abdomen. An abdominal computed tomography (CT) scan was performed, where a free intra-abdominal fluid was seen in left upper quadrant and a tumor like mass was detected in small intestine causing bowel obstruction for which he underwent exploratory laparotomy, finding a perforated jejunal GIST. An emergency exploratory laparotomy was performed due to the clinical findings of acute abdomen, upon organ examination, hemoperitoneum was found and intestinal leakage due to a mass in a segment of jejunum 25 cm from Treitz angle, an intestinal resection and anastomosis was performed. GISTs are common tumors that rarely present to the emergency room as an acute abdomen. It is important to the surgeon to bear this in mind and have knowledge of the specifics of management of these type of patients to provide an adequate surgical resection and postoperative risk specific follow up.

Keywords: Jejunal perforated gastrointestinal tumor, GIST, Acute abdomen, Stromal tumor, Jejunal tumor

INTRODUCTION

Gastrointestinal stromal tumors (GIST) are the most common mesenchymal tumor of the GI tract. Most cases are asymptomatic, or present as an abdominal mass. Less frequently, they can present as upper GI bleed or acute abdomen. Tumor perforation leading to acute abdomen is rare, and increases the risk of disease progression.[1] An adequate resection and proper adjuvant therapy can improve overall survival and recurrence free survival in this high-risk patient.

We hereby presented the case of a 47-year-old male who visits the ER department complaining of abdominal pain with physical findings of acute abdomen who underwent laparotomy and a perforated jejunal GIST was discovered.

CASE REPORT

This is case report of a 47-year-old male patient with no past history of chronic diseases. He affirms to consume an approximate of 7 cigarettes per day, occasional alcohol consumption. Family history of gastric cancer by fathers' side.

He presented to our emergency unit referring a 1-month history with bloody stools and abdominal pain, previously checked by the gastroenterology department where he was scheduled an upper endoscopy. He refers that 48 hours prior to his admittance in the emergency department, he had presented epigastrium pain, nausea, vomits and oral intolerance., His vital signs upon arrival were as follows: heart rate 90 beats per minute, respiratory rate 20 per minute, blood pressure 120/60, temperature 36°C.

Upon examination, the patient revealed responsiveness, alertness, painful expression, agitation, rhythmic precordium, normal lung ventilation, abdomen with absent peristalsis, voluntary rigidity, tenderness in epigastrium, positive Blumberg sign. Laboratory analysis showed white blood cells 15,000 ul, hemoglobin 10.1, platelets 200 k/ul, glucose 141 mg/dl, creatine 1.1 mg/dl, TP 12 sec, INR: 1.2, CK: 145 U/l, LDH: 250 U/l, sodium 130 mmol/l, potassium 4 mmol/l, chloride 110 mmol/l, blood gas: pH 7.45, and lactate 3.6 mmol/l.

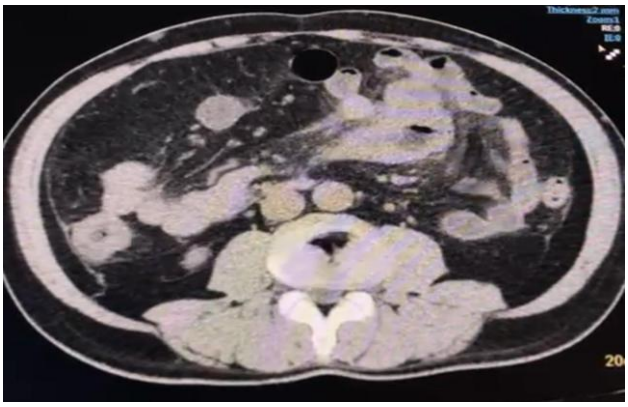

Figure 1: Axial abdominal CT-scan, inflammation of the mesentery is present in the left lower quadrant, an image of a tumor like mass is present with hypodense areas in the left quadrant.

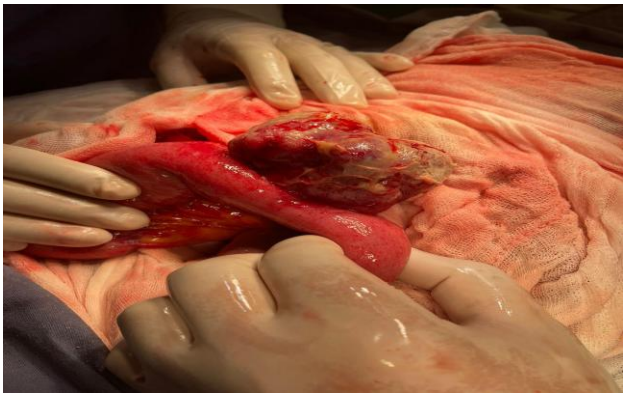

Figure 2: During exploratory laparotomy, we observe in jejunum changes of coloration and tumoral structure in antimesenteric edge representing submucosal hemorrhage and perforation.

An abdominal computed tomography (CT) scan was ordered to complete the formal examination which revealed presence of abdominal free air and the presence of a small fluid collection in the mesentery next to the small bowel in the left upper quadrant. Due to the topographical findings and the presence of acute abdomen, it was decided to perform a laparotomy.

It was decided to perform an open exploratory laparotomy, the intraoperative findings were as follows: hemoperitoneum in left upper quadrant, intestinal leakage next to a tumor like lesion found measuring 6 cm by 6 cm in the antimesenteric border of the small bowel, approximately 25 cm from the Treitz angle with signs of exophytic ulcer and perforation.

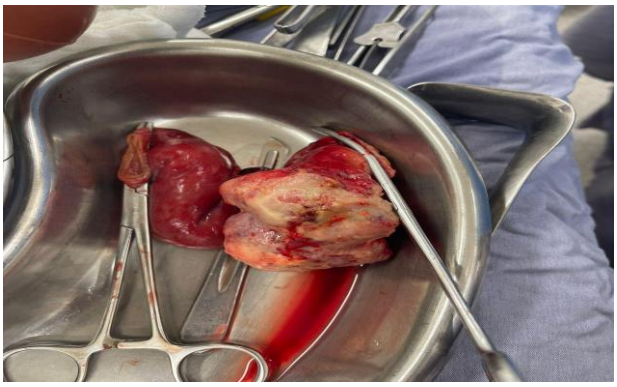

Figure 3: Macroscopic picture of portion 30 cm of small bowel resected where we can appreciate a tumor like lesion with GIST characteristics. We also observe changes in small bowel coloration, indicating ischemia of this portion.

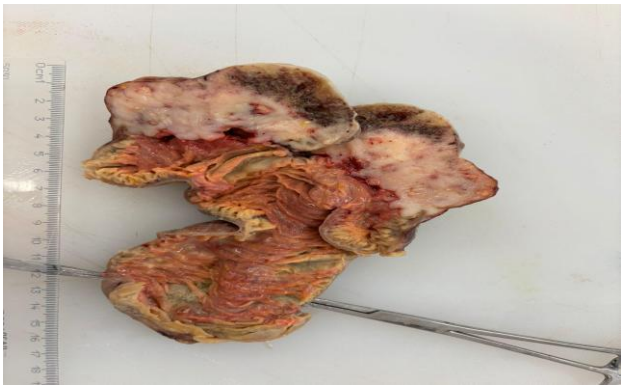

Figure 4: Macroscopic picture in the pathology department showing the gastrointestinal stromal tumor in jejunum.

A jejunum resection 10 cm distal and proximal to de tumor was performed was performed. After the resection, it was decided to perform and end to end jejunum anastomosis. Surgical cleansing of abdomen was performed and a nasojejunal feeding tube was introduced distal to the anastomosis zone.

Post-surgery follow-up was continued with broad spectrum antibiotic treatment régime, oral intake was indicated 24 hours after surgery with the nasojejunal oral feeding tube. A chest X-ray was indicated 24 hours after surgery, demonstrating adequate introduction of feeding tube. Oral intake was indicated 48 hours after surgery without any complications, he was later discharged on the 5th day of surgery postop.

Surgical piece was sent to the pathology department where they confirmed the presence of GIST measuring 6.8 cm by

4.5 cm. Inmunohystoquimic factors DOG-1 positive and CD117 positive.

DISCUSSION

GISTs are the most common mesenquimal tumors of the GI tract. Due to multiple similarities found between GIST tumoral cells and interstitial cells of Cajal (natural pacemakers for bowel peristaltic movements), they are believed to originate them. Historically, they were considered resistant to systemic therapy, with poor response rates, however, identification of key mutations on c-KIT and PDGRF genes, allowed to understand this tumors biology and develop targeted therapies which improved significantly patient's prognosis.

Incidence is calculated 4.3-22/1,000,000, with wide variations around the world. It is located more commonly in the stomach (50-60%), followed by the small bowel (30-35%), the rectum (5%), esophagus (<1%). Malignant potential is variable, clinical presentation goes from small, slow growing lesions to big, aggressive, hastily growing tumors.[1] 20% of cases present metastatic disease at diagnosis, with predilection for the liver, peritoneum and omentum. Metastasis outside the abdomen are extremely rare[1]. It is well established that 70-80% of GISTs harbor a mutation in the KIT gene, which produces a constitutive activation of the receptor. This mutation is an important therapeutic target for this kind of tumors. KIT receptor is part of a family of type III tyrosine kinase receptors; PDGFR receptor also belongs to this family. Oncogenic mutations of KIT produce a ligand independent activation of the receptor.

Type and location of the mutation are prognostic factors for disease progression. The most common mutation occurs in the exon 11 (66%); deletions are associated with a shorter disease free survival and overall survival compared with other kind of mutations of the same exon. Mutation in other exons besides from being a disease progression prognostic factor, also predict tumor sensitivity to TKIs. PDGFR and other receptor gene mutations could also be responsible for tumor formation, although these represent a minor contribution compared to KIT mutations.[2]

Although most patients present with symptoms or an abdominal palpable mass, up to 25% of GISTs are discovered incidentally when performing imaging studies or abdominal surgery for other reason. Most common presentation is GI bleeding with anemia, however, patients can present with vague abdominal symptoms such as dyspepsia, nausea, vomit, constipation, fatigue or acute abdominal pain which may or may not require emergency surgery.[3] Diagnosis is ideally made through an endoscopic US guided biopsy, to avoid the risk of tumor seeding that exists when performing percutaneous biopsies. When it is not possible to obtain an adequate biopsy safely and imaging is highly suggestive of GIST, surgical resection is indicated when feasible. Treatment in tumors >2 cm or in emergency context consists of complete surgical resection (R0) avoiding rupture of the tumor pseudocapsule.[3,4] Due to the subepithelial location of these tumors, endoscopic resection is not advisable because a great risk of incomplete resection exists. Since effective, targeted therapies are available, it is not clear whether microscopic residual disease (R1) after resection affects the prognosis of patients or not, and so the decision to take a patient to re-resection should be taken cautiously. Multiple factor can increase the risk of disease progression. In the past, the tumor size was the only factor taken in to account to predict disease progression, however, with a better understanding of tumor biology and effective, targeted therapies, more precise risk stratification scores where needed to identify those patients who would benefit from adjuvant systemic therapy. Multiple risk stratification scores have been used (National Institute of Health, Armed forces Insistute of Pathology) considering a variety of risk factors. The principal risk factors identified where: tumor size (<2 cm or >5 cm), mitotic count (<5 or >10/HPF), location (gastric GIST has a lower risk of progression than other locations), symptoms at presentation, histology (epitelioid and mixed tumors have a worse prognosis), ulceration, tumor rupture (iatrogenic or spontaneous) and some specific mutations.[8]

In other gastrointestinal tumors, infiltration of the tumor to the peritoneal surface reflects an advanced stage (T4), which has implications in both prognosis and treatment, however, this item is not included in GIST´s risk scales, nor in the TNM classification. It has been identified that "tumor rupture" either spontaneous or iatrogenic predicts a worse oncologic prognosis, however, the term "tumor rupture" is ambiguous, and used to describe different situations causing tumor rupture rates to vary widely across the literature (2-22%).[10] To answer this problematic situation a GIST tumor defect classification was developed. Defects where classified as minor and major defects according to the risk of recurrence they provided. Major defects include: tumoral spillage, tumor fracture, piecemeal resection, intestinal perforation at the tumor site, hematic ascites, tumor infiltration to neighboring organs or prior incisional biopsy. Minor defects include: Peritoneal infiltration, iatrogenic laceration or intestinal microscopic positive margins. Major defects increase importantly the risk of peritoneal recurrence or recurrence at any site (HR: 8.57 and 3.85) while minor defects did not increase the risk of recurrence.[9]

In patients with high risk of recurrence and a R=resection, adjuvant therapy is suggested with oral imatinib for 3 years. 3-year adjuvant therapy has proved to improve overall survival and disease free survival compared to 1-year therapy. In patients with intermediate risk of recurrence decision to start adjuvant therapy should be individualized. Decision to give adjuvant therapy might be supported by genetic risk stratification studies. In patients not deemed resectable, neoadjuvant imatinib might be given for 6-12 months with further evaluation for resectability. TKI´s are also standard treatment in

metastatic or locally advanced tumors, not candidates to surgical resection.

CONCLUSION

GIST´s are common tumors that rarely can present to the emergency room as an acute abdomen. It is important to the surgeon to bear this in mind and have knowledge of the specifics of management of this patients to provide an adequate surgical resection and postoperative risk specific follow up.

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