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Gastrointestinal stromal tumor (GIST) presenting as a groin mass mimicking and incarcerated hernia

José Tinoco-González *

N. S. Soledad, Avenue 35, Cantillana, Seville 41320, Spain

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ABSTRACT

INTRODUCTION: The contents of a hernial sac can harbor preperitoneal fat, epiploon or some hollow viscera, with a malignant lesion in its interior being an unusual finding.
PRESENTATION OF CASE: We present the case of a 59 year-old male who arrived in the Emergency Department with an irreducible inguinal hernia, with no local signs of vascular compromise. The diagnosis was made after examining the hernia sac, using an inguinal approach, showing evidence of a largely necrosed gelatinous mass, which was resected by mid-line infraumbilical laparotomy. The mass was diagnosed as a high risk gastrointestinal stromal tumor.
DISCUSSION: GIST are characterized by the absence of symptoms in most cases. The unusual presentation, as incarcerated hernia, allowed the diagnosis of this disease. The tumor measured 6 cm with a mitotic index greater than 5/50 CGA tumor is classified as high risk. The finding of a thickened peritoneum proceeded to opening. The exploration of the sac is critical.
CONCLUSIONS: Exploring the hernia sac in any complicated hernia, especially when the sac has macroscopic alterations and presence of areas of bleeding inside is essential.

1. Introduction

Gastrointestinal stromal tumor (GIST) is the name given to mesenchymal tumors originating in the Cajal cells (mixed neural and myogenic), with the location in the small intestine being second in frequency after the stomach.[1] The immunohistochemistry typically shows the expression of the tyrosine kinase receptor gene (c-Kit or CD117, present in 95% of the cases)[2] with diffuse staining in the cytoplasm, and with membrane strengthening or peri-Golgi accumulates. The prognostic factors of survival in GIST are, tumor size, its location (stomach vs. small intestine), and the mitotic index expressed as mitoses per high-magnification field.[1,3]

The inguinal hernia can collect several viscera in its interior, the most usual contents being epiploic or small intestine loops; if blind, there are usually, appendix, sigma or bladder, due to sliding, where the viscera itself forms part of the hernial sac. There are few references to a malignant disease forming part of the contents of a hernial sac. In the case of complicated hernias (strangulated or incarcerated) the examination of the hernial sac must be assessed.[4,5]

We present the case of a middle aged man who was intervened urgently with a diagnosis of incarcerated hernia, describing the presence of a complicated GIST in the hernial sac.

2. Clinical case

A 59 year-old male, with no medical history of interest, who arrived in the Emergency Department, due to having a mass in the left inguinal region for 10 days, with no fever or gastrointestinal symptoms. The physical examination revealed the presence of an irreducible mass, non-pulsatile, and with no flogistic signs in the left groin. The abdomen was soft, with no pain, and normal bowel sounds. A simple X-ray of the abdomen was performed, which showed a normal intestinal luminogram with no dilation of the small intestinal loops.

With the diagnosis of an incarcerated hernia, urgent surgery was indicated. The exploration of the inguinal canal revealed nodular peritoneal thickening that protruded through the deep inguinal orifice and the entire posterior floor. At the opening of the hernial sac, we found a jejunal loop that contained a cerebroid-like fragmented tumor mass with implants in the left pelvic, prevesical and parietocolic peritoneum. With these findings, we closed the peritoneum and performed a prosthetic repair of the inguinal canal using an anterior preperitoneal technique (Rives technique). A limited infra-umbilical laparotomy was then performed, with the finding of an exophytic and pedunculated tumor mass adhered to the vesical peritoneal reflection. A segmental bowel resection was

* Tel.: +34 625 962 094; fax: +34 955731065.
E-mail address: tinoko243@hotmail.com

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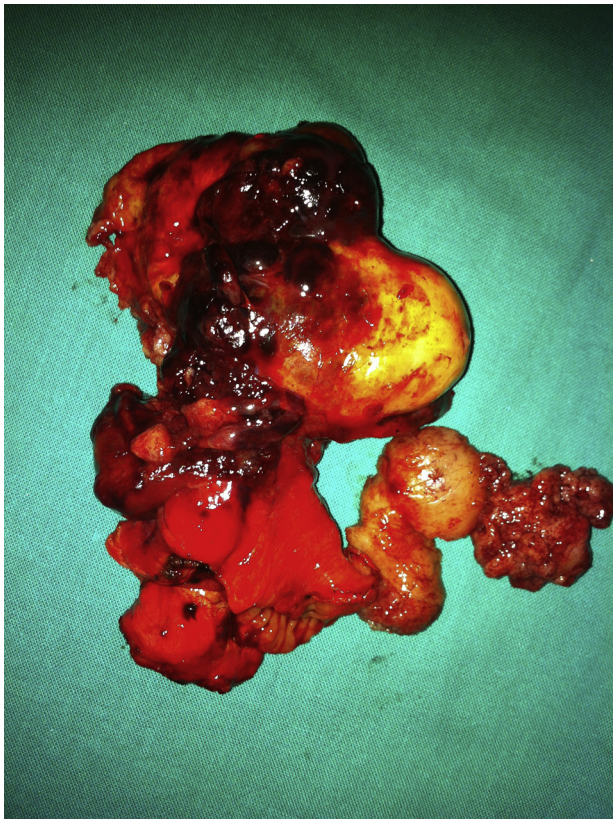

Fig. 1. Surgical specimen. Non obstructive exophytic lesion.

performed with end to end anastomosis of about 10 cm of jejunum, leaving macroscopic disease in the pelvis and bladder (R2).

The patient was discharged on day 5 after the surgery with no complications. The histology confirmed a GIST tumor with affected surgical margins (ruptured anti-mesenteric margin), classifying it as a pT3Nx, with a mitosis index greater than 5/50 HMF (high-magnification fields), a *Ki* 67 of 15%, which could be established as a risk of progression according to the criteria of Miettinem–Lasota[6] of 85% (high risk). The immunohistochemistry showed an intense positive for CD117 (c-kit), with positive foci for CD34, a positive smooth muscle actin, and negative for S100, desmin, chromogranin, synaptophysin and CD56 (Fig. 1).

One month after admission, a CT was performed to stage the residual disease, which was present in the sub-hepatic region, the right flank and in the pelvis. After being started on adjuvant therapy with imatinib (a tyrosine kinase inhibitor), the follow up CT at six months showed evidence of a partial response of 50%, with the patient being asymptomatic.[7]

3. Discussion

Small bowel GISTs are characterized by the absence of a clinical picture in the majority of cases, being large tumors with specific locations that show symptoms such as, gastrointestinal bleeding, ulceration of the mucosa due to growing, or an obstruction syndrome, usually by invagination. In our case, the mainly extrinsic growth fixed the lesion to the peritoneum of the left groin region, leading to this unusual presentation as an incarcerated hernia. In the wide search performed by us, ours is the second case described in the medical literature of a GIST that first presented as a suspicion of a complicated hernial disease. In the case published by Yeung and colleagues a diagnosis of bilateral groin hernia (both of them easy to reduce) existed, and while being in the awaiting list a dominant

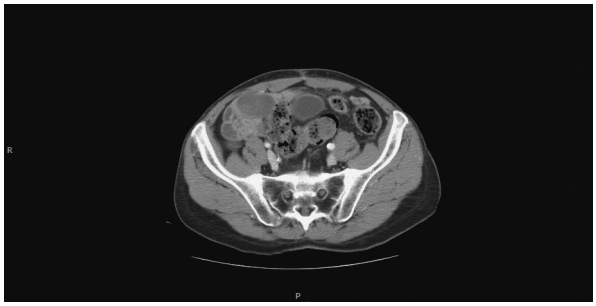

Fig. 2. Image abdominal scan. Residual mass in the left iliac fossa.

mass in the right side appeared but the patient was operated on an elective basis and with swelling in both of the groins[13] (Fig. 2).

Emergency surgery was done because of a diagnosis of a mass in the location of the inguinal external ring, and in our hospital and our digestive surgery the surgical exploration of the groin is the next step in theses cases. When a mass appears suddenly at this level (upper the ilio-inguinal ligament) it's not cost-effective to perform a CT scan because it's well known that in 100% of the cases the diagnosis will be an hernia related disease and the CT scan will show if there is obstruction or not but will not be specific to determine the etiology. In our patient, a theorically performed CT would have shown a mass in the groin area related to bowel.

In our case, the mesentery of the involved loop was not easily exposed through the groin incision and we were not able to perform a safe suture after the resection. In addition, we think that in the urgent setting and once that you have found a non-typical finding as this was, an small laparotomy to properly assess the problem you are dealing with is an attitude that cannot be criticized. We would not expected so much morbidity of performing a small incision and we thought that we were in the need of having as much information as possible to be explained to the patient and to the familiy.

From the histological point of view, it had three typical architectural patterns[6,8,9]: fusiform (70%), epithelioid (20%), and mixed (10%). The CD117 usually expresses other markers, often positive, such as CD34 (60–70%), smooth muscle actin (15–60%), S100 (10%), and desmin (rarely).[10,11] Expression of NSE (neuron specific enolase) has sporadically been observed as well as neurofilaments, chromogranin, and keratins 8 and 18.[12]

If we consider the prognostic classification most used, that of Miettinem–Lasota, the tumor that we present is classified as "high risk", on measuring more than 5 cm (6 cm), having a location other than gastric and, finally, a mitosis index greater than 5/50 HMF, all of these implying a risk of disease progression in the follow-up of 85%. Kinblom completed the Miettinem–Lasota classification by adding the Ki 67 cell proliferation index, which when it is higher than 10% (15% in our case), it indicates an even worse prognosis, with a mean survival of 28 months.[7]

The presence of a painful mass of sudden onset in the groin region leads, in the first place, to the diagnosis of a complicated inguinal hernia, which is one of the few classic emergency surgery indications, with little room for discussion. The absence of a history of hernia disease, associated gastrointestinal symptoms, and flogistic signs, may lead to a debate on whether performing an imaging study would have permitted a diagnosis oriented toward the tumor process with a more aggressive surgical therapeutic approach. An ultrasound or CT would not have changed the initial staging of the process or oriented toward the diagnosis of a jejunal GIST, due to the rarity of the clinical picture presented. In the GISTs with positive c-kit, an R0 surgery is not a determining prognostic factor of survival, given the favorable response to the treatment with imatinib.

4. Conclusion

Finally, generically, with the presence, or a clinical suspicion, of an incarcerated inguinal hernia it is clear that exploration of the hernial sac is obligatory, since it enables the assessment of the presence of vascular involvement, the visceral contents, and the diagnosis, despite the exceptionality of cases like the one we present.

Conflict of interest

None declared.

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Author contributions

Tinoco-Gonzalez wrote the manuscript. Tinoco-Gonzalez also in association with Ramallo-Solis, Ramirez-Plaza, Reguera-Rosal involved in reviewing the bibliography. Pareja-Ciuro and Padillo-Ruiz are the investigation co-directors of the manuscript.

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