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2173-5077/

Abdominal Cocoon: A Rare Cause of Intestinal Obstruction☆

Síndrome de Cocoon: una rara causa de oclusión intestinal

Sclerosing encapsulating peritonitis (SEP) is a cause of intestinal obstruction of unknown etiology. Also known as abdominal cocoon syndrome, it is characterized by a fibrocollagenous membrane that either completely or partially encompasses the small bowel.[1] This causes patients to repeatedly seek medical care for symptoms of intestinal obstruction. Both symptoms as well as radiological images are non-specific, and diagnosis therefore requires elevated suspicion.[2,3]

We present the case report of a 54-year-old male patient with no prior history of interest, except for bilateral endoscopic hernioplasty that was completely extraperitoneal and recent elective surgery for cholelithiasis. It was during this latter surgery that sclerosing peritonitis was incidentally diagnosed when a whitish membrane that affected the small bowel and descending colon was found intraoperatively, blocking the supramesocolic compartment as well.

Afterwards, the patient came to the Emergency Department on repeated occasions due to colicky abdominal pain and subacute intestinal obstruction, so he was hospitalized once again. Physical examination detected abdominal distension associated with a palpable mass in the mesogastrium. A CT scan demonstrated medialized small intestinal loops, some with wall thickening, that were adhered amongst themselves and to the anterior abdominal wall; there was also free interloop fluid, which was probably related to adhesion-related syndrome. We decided to schedule surgery, and found a membrane covering the jejunum and a large part of the ileum (Fig. 1). We performed almost complete exeresis of the membrane, that could be separated from the serosa of the intestine. The integrity of the bowel loops was confirmed up to the ileocecal valve; no perforations were observed, and resection was not required. The patient's condition progressed favorably, and he was discharged 6 days after surgery with complete resolution of the symptoms.

Abdominal cocoon syndrome is a very uncommon disease of unknown etiology that is classified as either idiopathic or secondary. This latter form is more common, and there have been descriptions of cases of secondary SEP associated with peritoneal dialysis, tuberculosis, treatment with beta-blockers, familial Mediterranean fever, etc. The idiopathic form of SEP is relatively more frequent in tropical countries,

☆ Please cite this article as: Illán Riquelme A, Camacho Lozano J, Abdalahi H, Calado Leal C, Huertas Riquelme J. Síndrome de Cocoon: una rara causa de oclusión intestinal. Cir Esp. 2016;94:417–419.

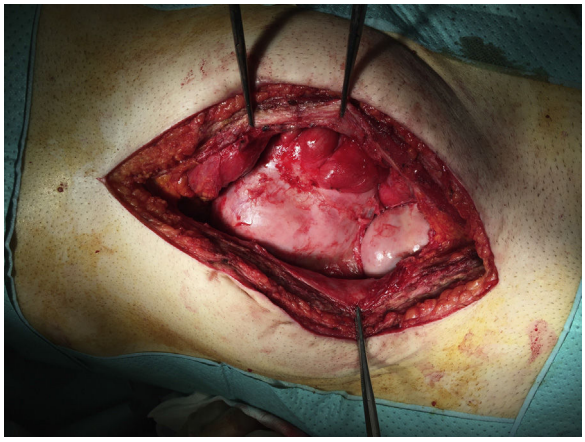

Fig. 1 – Image from the surgery showing a pearly membrane encompassing the small intestine.

and in a greater proportion in young women. Some hypotheses have been proposed as causes of this idiopathic form, such as retrograde menstruation associated with a viral infection or through the Fallopian tubes, although neither has been demonstrated.[3] In some patients, a relationship has been described between SEP and other embryologic alterations like omental hypoplasia and sclerotic retracted intestinal mesentery. This syndrome is characterized by a fibrotic membrane that completely or partially encapsulates the small bowel and may envelop other organs, such as the large bowel, liver and stomach.[4] These patients present recurring episodes of abdominal pain, nausea, vomiting, anorexia, weight loss, malnutrition, recurring crises of total or partial intestinal obstruction, while in some cases an intraabdominal mass is frequently palpated during physical examination. Diagnosis requires a high index of suspicion, and most cases described in the literature are diagnosed during surgery.[5]

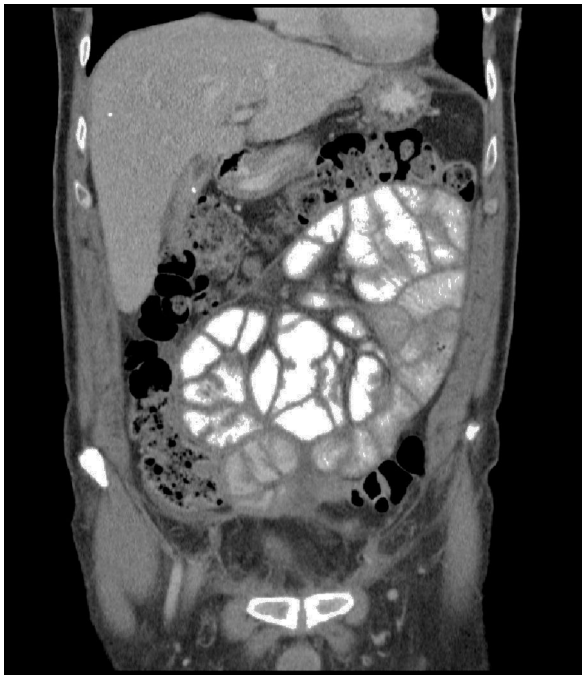

Fig. 2 – CT image: encapsulated conglomeration of small intestinal loops.

The preoperative diagnosis is made in patients with repeated symptoms of intestinal obstruction and suspicious CT scans[6] that show images of a small bowel loop conglomeration in the center of the abdomen that is encapsulated in a membrane (Fig. 2). Interloop ascites is also frequently observed.

Treatment of idiopathic SEP in symptomatic patients involves surgery.[7] The most widely used surgical technique is total or partial excision of the membrane and adhesiolysis of the affected small bowel loops. This reduces the need for intestinal resection, which should only be used in cases where it is considered unavoidable.[1,2] Adequate preoperative nutritional support in these patients has been observed to reduce postoperative complications and increase postoperative satisfaction.[8]

Conflict of Interests

The authors declare having received no funding to complete this article, nor were there any conflicts of interests.

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2173-5077/

Oesophageal Leiomyoma With Intense Uptake in Positron Tomography With ^{18}Fluorodeoxyglucose and Computed Tomography☆

Leiomioma de esófago con intensa captación de ^{18}fluorodeoxiglucosa en la tomografía por emisión de positrones-tomografía computarizada

Gastrointestinal stromal tumours (GIST) are the most common mesenchymal neoplasms of the digestive tract (3%). They are most frequently located in the stomach and small bowel, which are involved in 75% and 25% of cases, respectively.[1] Oesophageal locations are rare (5%); the most common tumours in this region are leiomyomas, which represent 70% of benign oesophageal tumours. Given the clinical, endoscopic and radiological similarities between GIST and leiomyomas, they may not be properly differentiated until after resection. We present the case of an oesophageal leiomyoma that was treated as a GIST due to metabolic hyperactivity observed on PET/CT.

The patient is a 59-year-old woman with no medical history of interest, who reported epigastric discomfort that had progressed over the previous 5 years in association with a sensation of premature postprandial fullness in recent months. Upper gastrointestinal (UGI) endoscopy revealed, at the entrance to the gastric cavity, a large impression on the anterior lesser gastric curvature involving a lesion with bilobulated morphology that was submucosal in appearance. Endoscopic ultrasound (EUS) identified a mediastinal mass measuring 4×7 cm, which was well-outlined and suggestive of benign disease, in contact with the left liver lobe, inferior vena cava and left atrium, with no signs of infiltration; the mass was slightly hypoechogenic compared to the liver parenchyma and well vascularised. The result of the EUS-guided fine-needle aspiration (FNA) was compatible with low-grade mesenchymal neoplasm (c-kit [−], actin ML [±], CD31 [−]). Computed tomography (CT) scan demonstrated a hypodense mass in the oesophagogastric junction (OGJ) measuring 9 cm in length that caused circumferential wall thickening in its upper portion of up to 3.5 cm, and somewhat more eccentric at its lower gastric portion, where it showed a clear submucosal component. With homogenous density and absence or very slight uptake of intravenous contrast, it was in contact with the posterior wall of both atriums, but with a defined interface. These findings were compatible with a submucosal tumour of the oesophagogastric junction, whose differential diagnosis included GIST, other mesenchymal tumours or adenocarcinoma. The study was completed with PET/CT, which demonstrated a hypermetabolic mass in the posterior mediastinum over the distal third of the oesophagus and extended towards the cardia, with a maximum SUV of 8.21 (Figs. 1 and 2).

Given the high diagnostic probability of c-kit (−) oesophageal GIST (some 5% do not express c-kit), due to the large tumour size, positive uptake in PET/CT and inconclusive FNA, we decided to perform oesophagectomy.

Midline laparotomy and a transhiatal approach revealed a large mass (10×8 cm) in the distal oesophagus and OGJ that completely surrounded the oesophagus. The tumour was encapsulated, did not affect the serosa, and had several lobulations but no apparent lymph node involvement. Subtotal oesophagectomy was conducted with substitution gastroplasty and cervical oesophagogastric anastomosis.

The postoperative period ran its course without complications. Oral intake was properly re-established, and a follow-up transit study showed no pathological findings. The patient was discharged from hospital on the 7th day after surgery.

☆ Please cite this article as: Álvarez Martín MJ, Garcia Navarro AM, Rodriguez Fernandez A, Jimenez Rios JA. Leiomioma de esófago con intensa captación de ^{18}fluorodeoxiglucosa en la tomografía por emisión de positrones-tomografía computarizada. Cir Esp. 2016;94:419–422.