INTESTINAL ADENOCARCINOMA AS UNEXPECTED CONTENT OF SPIGELIAN HERNIA: WHEN CT-SCAN IT’S NOT ENOUGH.

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Abstract
Spigelian hernias are only 1-2% of all types and are considered a very uncommon condition: it develops through a weaker area between the semilunar line and the lateral edge of rectal muscle. There is an association with female sex, obesity, prior surgery, chronic obstructive pulmonary disease, abdominal trauma or coexistence of other wall defects. The content is intraperitoneal fat or omentum, sometimes small bowel or colon. Symptoms, like abdominal pain, lateral lump and altered bowel habits are linked to the most common complication: incarceration. The CT scan can be useful in confirming the abdominal wall defect and in discriminating the content and can help in the choice of the best surgical
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procedure, especially if there is an ischemia. In literature are described some case of atypical content, such as testicle in association with cryptorchidism in children or appendicular abscesses or abdominal wall tumor, especially in adults. We report a very unusual finding in a 81-year-old patient: a well-differentiated intestinal adenocarcinoma of left colon as content of a left Spigelian hernia. The patient was admitted to our Department because of abdominal pain, left abdominal lump and altered bowel habits. The preoperative CT-scan showed a large defect in lateral left abdominal wall and a content of sigmoid colon with abscesses and diverticula, but there were not signs of ischemia. It was chosen an anterior direct approach, performed an adhesiolysis, isolated the left colon and identified the abdominal wall defect. Despite the absence of CT or clinical signs of ischemia, the colon was included in necrotic tissue and partially twisted. It was performed a left hemicolectomy through the hernia defect and side-to-side colonic anastomosis reconstruction. After the bowel replacement, the abdomen wall was reconstructed without mesh or other devices in order to reduce the infection risk. The diagnosis of well differentiated (G1) intestinal adenocarcinoma, necrotic, infiltrating the bowel up the subserosa was performed only after histological examination. The CT-scan was not able to differentiate the neoplasm from diverticular abscess, and even during the operation there was no a definite suspect, probably because the loss of normal anatomy due to the chronic inflammatory process. According to some studies the CT-scan has an importance in distinguishing the type of Spigelian hernia, but it is not so clear in discriminating the content, as it happened in our case, when the correct diagnosis of colon cancer concomitant with diverticula and abscess was possible only with the later microscopic examination.

Abstract

L’ernia di Spigelio è una patologia rara, stimata intorno al 1-2% delle ernie della parete addominale: si sviluppa attraverso un’area di minor resistenza tra la linea semilunare e il margine laterale del muscolo retto. Vi è un’associazione con il sesso femminile, l’obesità, la chirurgia pregressa, bronco pneumopatia cronica ostruttiva, traumi addominali o coesistenza di altri difetti della parete. Il contenuto è costituito da adipe intraperitoneale o omento, più raramente da anse intestinali. I sintomi, come dolore addominale, la nausea, il vomito e l’alterazione dell’alvo sono legati alle possibili complicanze derivate dall’incarcerazione.

La TC può essere utile nel confermare il difetto della parete addominale e discriminare la tipologia del contenuto erniario in maniera tale da poter effettuare la scelta della migliore procedura chirurgica, specialmente nel caso in cui si dovessero presentare segni di una incipiente ischemia. In letteratura sono descritti alcuni casi di contenuto atipico, come testicolari in associazione a criptorchidismo nei bambini, o ascesso appendicolare o tumori della parete addominale soprattutto negli adulti. Nella nostra esperienza descriviamo il caso veramente raro di un paziente di 81 anni affetto da un’ernia di Spigelio sinistra con contenuto un adenocarcinoma ben differenziato del colon sinistro. Il paziente era stato ricoverato presso il nostro Dipartimento per dolori addominali, tumefazione addominale laterale sinistra e alterazioni dell’alvo. La TC preoperatoria mostrava un ampio difetto della parete addominale sinistra contenente colone sigma con ascesso diverticolare, ma con assenza di segni di ischemia. È stato scelto un approccio diretto anteriore, eseguita un’adesiolisi, isolato il colon sinistro ed identificato il difetto della parete addominale. Nonostante la negatività alla TC e l’assenza di segni clinici di ischemia, il colon si presentava necrotico e parzialmente ruotato. È stata eseguita un’emicolectomia sinistra attraverso la porta erniaria e ristabilita la continuità intestinale con anastomosi colo-colla.

Dopo la ricostruzione dell’intestino, la parete addominale è stata ricostruita senza utilizzare la rete tipo “mesh” o altri dispositivi al fine di ridurre i rischi di infezione. La diagnosi di adenocarcinoma intestinale ben differenziato (G1), necrotico, infiltrante la parete intestinale fino alla sottosierosa è stata resa possibile solo con l’esame istologico. La TC non è stata in grado di differenziare la neoplasia dall’ascesso diverticolare, ed anche durante l’intervento non c’è stato un sospetto definito, probabilmente per la perdita della normale anatomia a causa del processo infiammatorio cronico. Secondo alcuni studi la TC ha importanza nel distinguere il tipo di ernia di Spigelio, ma non è in grado di discriminare il contenuto, come è successo nel nostro caso, quando la diagnosi corretta del colon concomitante con diverticoli e ascessi è stata possibile solamente al momento della valutazione istologica.
Background
Among all abdominal wall defects, Spigelian hernia may be considered a very uncommon kind. According to literature, only 1-2% of all hernias appears as antero-lateral mass due to a weaker area between the semilunar line and the lateral edge of rectal muscle, as described for the first time by Adrian van der Spieghel (1). Even if the Spigelian hernia content is frequently represented by intraperitoneal fat or omentum, in several cases, the small bowel or colon are unusual findings. Most of Spigelian hernia are acquired and they are classically associated with female sex, obesity, prior surgery, chronic obstructive pulmonary disease, abdominal trauma or coexistence of other wall defects (2). Usually, the lateral abdominal lump, pain (described as sharp, constant or intermittent) and sometimes nausea, vomiting or altered bowel habits are symptoms that only appear after the most common complication: the incarceration. The clinical diagnosis is quite difficult (3) and the use of CT-scan can be useful to confirm the abdominal wall defect and to analyze the hernia content (4). We report a case with a very unusual finding as content of Spigelian hernia: an adenocarcinoma, correctly diagnosed only after histological examination, despite the use of CT-scan in the preoperative time.

Case Report
An 81-year-old Caucasian patient was admitted to our surgical department because of abdominal pain, left abdominal lump and altered bowel habits. His clinical history was characterized by chronic ischemic heart disease, hypertension, hyperlipidemia, chronic obstructive pulmonary disease, obesity and diverticular disease. In the past he was submitted to coronary stenting for heart stroke, right hip replacement and bilateral inguinal hernia repair. He took a therapy with thiazide diuretics, platelet antiaggregants, beta blockers, calcium antagonists, gastric protectors and alfuzosin for prostatic hyperplasia. At the admission the patient had remittent mild fever and was dyspnoetic, he complained about nausea and inappetence. Blood exams showed neutrophilic leukocytosis, with 18,000 white cells per milliliter and 80% of neutrophils, an high PCR level, with a 17,83 mg/dL value and a decrease in albumin, with 2,6 g/dL. The abdominal mass well defined, with a 30 cm maximum diameter and a hard parenchymatous consistency, slightly painful, covered by normal skin, that could not be reduced in abdomen. There were no clinical signs of obstruction and perforation, only feces were diarrheal: abdomen could be palpable without pain except the lump and digital rectal examination has not revealed blood or other abnormal conditions. He underwent an abdominal CT-scan (Fig. 1-2) who revealed a large defect (94 mm) in the left lateral abdominal wall, in correspondence of Spigelian line, with a content characterized by sigmoid-colon. There were no signs of ischemia, but the incarcerated bowel sections were studded with abscesses and diverticula (Fig. 2). After the CT diagnosis of diverticular abscess herniated as content of a Spigelian hernia, systemic antibiotic therapy with metronidazole was performed in order to reduce fever and create the ideal conditions for the surgical treatment. After three days of hospitalization he had a pulmonary edema basically due to urinary retention, treated with diuretic therapy and placement of a foley catheter. After two weeks the patient underwent surgical treatment through a direct anterior approach under general anesthesia. It was performed a transverse incision over the abdominal mass; after a accurate adhesiolysis the left colon was isolated and the abdominal wall defect was identified: the maximum length of the abdominal gap was about 10 centimeters (Fig. 3). Despite the absence of CT or clinical signs of ischemia, the colon was included in necrotic tissue and partially twisted. A left hemicolectomy through the hernia defect and side-to-side colonic anastomosis reconstruction were performed. After the bowel replacement, the abdomen wall was reconstructed without mesh or other devices in order to reduce the infection risk. Histological examination revealed a well differentiated (G1) intestinal adenocarcinoma, necrotic, infiltrating the bowel up the subserosa, with no peritoneal or lymph node invasion. Resections margins were tumor-free. The patient needed a clinical stabilization in the Intensive Care Unit especially because of the heart and respiratory preoperative conditions. He had a slow recovery of the bowel function, with episodes of vomiting: in the fifth postoperative day the execution of abdomen radiography showed a distension of all colon tract and gastrectasia, but no signs of perforation or obstruction. He was treated with prokinetic agents until the complete recovery of bowel function in the tenth postoperative day. There were no complications linked to the surgical site and the sutures were removed in the tenth postoperative day. The patient was dismissed on twentieth post-operative day in order to achieve a good respiratory and cardiac performance upon returning home.
**Fig. 1** - The axial contrast media CT (MDCT) image well shows the Spigelian hernia with the left abdominal wall defect (blue arrows) localized in the lateral part of rectal muscle. It is also possible to observe herniation of abdominal fat and of part of the sigmoid-colon. CT signs of diverticulosis are present at this level (pink arrow).

**Fig. 2** - The sigmoid-colon with the involvement of the left colic artery branches (Fig A - yellow arrows). The image shows widespread increase in wall thickness of the colon (Fig. B - green arrows).
Fig. 3 - At the beginning of surgical procedure, after the incision and adhesiolysis, it is put in evidence the wall defect.

Discussion and Conclusions
Despite the suggestive clinical presentation and the typical CT scan images, the definition of Spigelian hernia can be difficult, and in some cases the content can be only investigated with a postoperative histological examination. As described, Spigelian hernia can be congenital with a mean age of presentation is 2.7 years. In their review Moles Morenilla et al (5) report the close association with cryptorchidism, in which the usual content is testicle, and with inguinal and umbilical hernia, in which omentum and bowel are often found. In acquired types the most common content is bowel and abscesses, but there are some cases in literature of atypical content. There are some reports of appendicular abscesses: Demetriou et al (6) described an appendicular abscess with appendicolitis which appeared as a caecal volvulus in CT images. As in our case the real diagnosis was clear only after the surgical treatment. Allewart et al (7) reported a case of primary serous papillary carcinoma of the peritoneum. Before the surgical procedure the images showed only an heterogeneous that extended from umbilicus to the right groin, even if associated with cystic pelvic masses and peritoneal masses. Only the histological examination of the herniated omental cake proved its nature of tumor, as well as the peritoneal masses were proven to be tumoral spread. In fact, the atypical content was not the diverticular colon, but the intestinal adenocarcinoma. Neither the clinical examination nor the CT scan were able to differentiate the abscess from a neoplasm. Even during the operation there was no a definite suspect, probably due to the loss of the usual anatomy of the left colon, disrupted by the chronic inflammatory process. There are some studies being published that enrolls CT scan as the instrument to distinguish between the subtypes of Spigelian hernia (8), but it is controversial the real role in discriminate content of this hernia, as shown in literature and in our case (9). Considering that, especially in case of diverticular disease with abscesses that could be considered a risk factor for concomitant colon cancer (10), even if was difficult to identify a cancer (11), a colon cancer as content of a hernia
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should never be excluded. This difficulty increases in a bowel squeezed between abdominal wall and subcutaneous tissue, as in our case, and greatly limits the power of definition of CT scan. Thus, our case supports the choice of the anterior open approach as standard surgical treatment when the viability of the bowel is uncertain or bowel incarceration was suspected (12, 13).

We report the case of an intestinal adenocarcinoma, presented as atypical content of a Spigelian hernia. The correct diagnosis was possible only after surgical treatment and subsequent histological examination. This condition represents a CT scan limitation before surgery, probably provoked by bowel and abdominal wall inflammation that, together with abscess, obscures the tumor identification. In this case in which neither the clinical examination nor the CT scan were able to differentiate the abscess from a neoplasm, the anterior open approach should be adopted as standard surgical treatment.
References


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