

Acute appendicitis inside a Spigelian hernia. A rare case of a rare disease

Apendicitis aguda dentro de una hernia de Spiegel. Un caso infrecuente de una patología infrecuente

Guadalupe K. Peña-Portillo*, Aarón Zaraín-Rodríguez, Victoria Rendón-Camarillo, Gabriela Ayala-Hernández, Iván Marín-Pardo, and Elías Ballesteros-Suárez

Servicio de Cirugía General, Hospital Universitario de Puebla, Puebla, México

Abstract

Introduction: Spigelian hernia is a rare entity, with higher improbability of acute appendicitis within it. **Case report:** A 75-year-old female with a 30-year evolution hernia, abdominal pain, and fever of 1 week of onset, in whom was found an acute appendicitis within a Spigelian hernia. **Discussion:** Spigelian hernia comprises 0.12-2% of all abdominal hernias. Presurgical diagnosis is established only in 50% of cases, with an hernial ring less than 2 cm and hidden localization. There isn't statistics of this complication because of the lack of case reports.

Keywords: Spigelian hernia. Acute appendicitis. Herniorrhaphy. Complicated hernia.

Resumen

Introducción: La hernia de Spiegel es una afección infrecuente, con una improbabilidad aún mayor de apendicitis aguda en su interior. **Caso clínico:** Mujer de 75 años que acude con una hernia de 30 años, dolor abdominal y fiebre de 1 semana de inicio, en quien se encontró apendicitis aguda dentro de una hernia de Spiegel. **Discusión:** La hernia de Spiegel comprende el 0.12-2% de las hernias abdominales. El diagnóstico prequirúrgico se ha logra en el 50% de los casos, con defecto menor de 2 cm y de localización oculta. No existe estadística de esta complicación debido a la escasez de casos reportados.

Palabras clave: Hernia de Spiegel. Apendicitis aguda. Herniorrafia. Hernia complicada.

Introduction

Spigelian hernia is a rare condition that occurs more frequently in individuals aged 50 to 60, with a predominance in women¹. It is mostly described as isolated cases, with the largest reported series including a total of 162 cases². Acute appendicitis as the content of a Spigelian hernia is one of the less common

complications reported in the medical literature. Pre-operative diagnosis of Spigelian hernia has been reported in only 50% of the cases described to this date³, with computed tomography being the most useful diagnostic imaging modality¹.

This is the case of a 75-year-old woman who presented to the ER with a 1-week history of abdominal pain. She had had a protrusion for 30 years with

*Correspondence:

Guadalupe K. Peña-Portillo

E-mail: gkpp16@gmail.com

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Figure 1. *Spigelian hernia.*

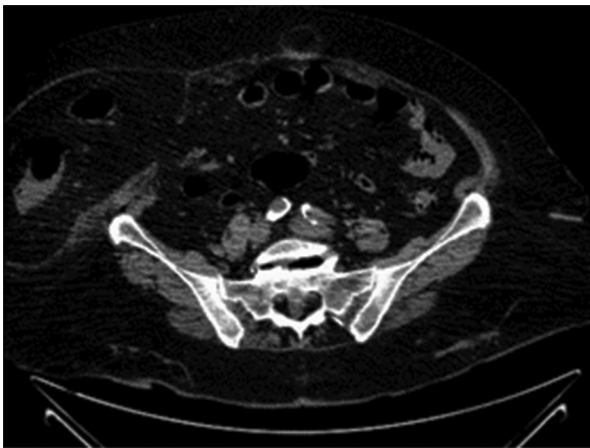


Figure 2. *Plain abdominal computed tomography, transverse view.*

progressive growth, previously asymptomatic. Intraoperatively, acute appendicitis was discovered as a complication. The aim is to contribute information about this condition to facilitate timely diagnosis and proper treatment.

Case report

A 75-year-old woman with a history of hypertension and 15 pregnancies resolved by delivery presented to the ER with a 30-year history of an abdominal hernia. She had experienced abdominal pain 1 week prior to consultation and fever for 48 hours. On physical examination, she was hemodynamically stable without any signs of systemic inflammatory response. She showed a 30 cm × 30 cm protrusion on the right flank, which was tender to palpation plus another 10 cm protrusion in the umbilical region, both non-reducible

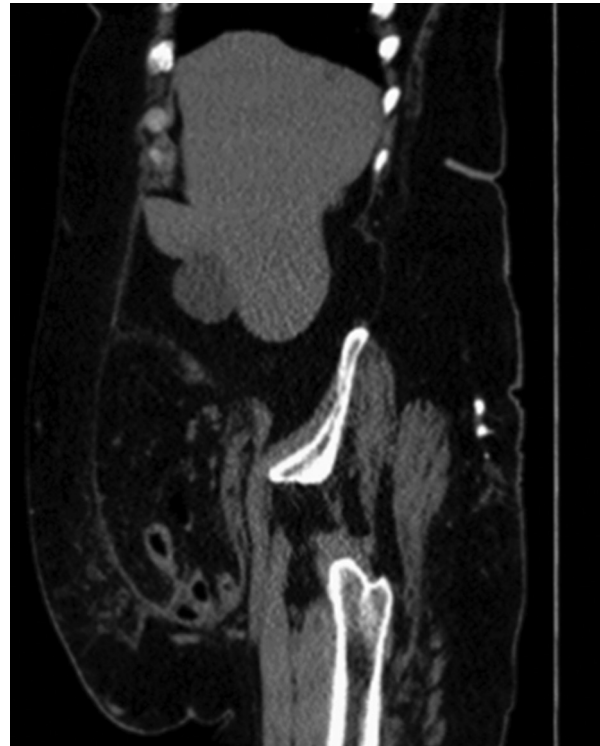


Figure 3. *Plain abdominal computed tomography, sagittal view.*

(Fig. 1). Lab tests showed leukocytosis ($12.6 \times 10^3/\mu\text{L}$) and neutrophilia (85.1%), without other abnormalities. The abdominopelvic computed tomography performed revealed the presence of a defect in the abdominal wall lateral to the right rectus abdominis muscle, extending from the mesogastric region toward the umbilical region, with a ring measuring up to 7.2 cm. A 16.7 cm × 17.2 cm × 15.3 cm sac was observed in its visualized portions. This sac contained peritoneal fat, the ascending colon, a portion of the transverse colon, and the cecal appendix. Striations in the mesenteric fat and minimal free fluid were also noted. Additionally, a defect in the umbilical region wall was observed, with a 10 mm × 8 mm ring and a 62.3 mm × 44.5 mm sac, through which only fatty tissue protruded (Figs. 2 and 3).

The diagnosis of Spiegel hernia with a concomitant abdominal wall abscess was established. An open approach was decided, involving a transverse incision on the right flank. During the surgery, a 30 cm × 30 cm hernia sac was found (Fig. 4). The sac contained loosely adherent intestinal loops, including the ascending colon, transverse colon, cecum, omentum, a dependent omental mass, distal ileum, and cecum. Additionally, there were signs of mesenteric thickening



Figure 4. *Hernia sac.*



Figure 5. *Remnant of cecal appendix flattened at the appendicular base.*

and a circular depression without communication with the cecal lumen (Fig. 5). Perforation of the hernia sac was identified, along with approximately 300 mL of foul-smelling purulent fluid located supraaponeurotically (Fig. 6). Abundant fibrinopurulent plaques were present as well. Dissection of the muscle layers revealed weakness and bulging in that region, with



Figure 6. *Supraaponeurotic purulent fluid, outside the hernia sac.*

thinned and devitalized edges. The surgical procedure included abscess drainage, surgical cleaning, adhesiolysis, partial omentectomy, and debridement of devitalized muscle edges. Herniorrhaphy was performed using X-shaped sutures of 1 polypropylene, preceded by the placement of an intra-abdominal Blake drain directed toward the pelvic cavity and a supraaponeurotic Penrose drain.

In the first 24 postoperative hours, the patient developed compartment syndrome, leading to a reoperation for abdominal decompression through the previous incision. Intestinal loops with color changes were found but recovered upon decompression. The patient was managed as a controlled hernia and admitted to the intensive care unit (ICU) due to septic shock, with a 3-day stay. She received critical care management and antibiotic therapy with meropenem (1 g/8 h). Subsequently, she showed improvement in her condition, with stable hemodynamics, serosanguinous drainage, without any signs of systemic inflammatory response, or abdominal hypertension. She tolerated oral intake well and had overall improvement, which led to hospital discharge 5 days after the ICU stay.

Discussion

Spigelian hernia is the spontaneous protrusion of preperitoneal fat, or a peritoneal sac containing visceral content through a defect in the Spigelian aponeurosis or fascia. It is named after the Belgian anatomist and surgeon Adriaan Van der Spiegel, who first described it as the semilunar line marking the transition from muscle to aponeurosis in the transverse abdominal muscle. However, it was Klinkosch who, in 1764, described a lateral ventral hernia specifically located on the Spigelian line^{1,2,4}. These hernias typically occur in the Spangen belt (an anatomical strip 6 cm above the intercrystal line) in 90% of the cases^{1,5}. They can be categorized into superior or inferior, being the former more common². Inferior Spigelian hernias are often confused with direct inguinal hernias in 50% of the cases⁴. They can also be categorized based on the location of the sac: interstitial, intraparietal, or hidden, if they are found between the muscle layers; and superficial, when they are subcutaneously located (0.8%)^{2,4}.

The incidence rate of Spigelian hernia is estimated to be between 0.12% to 2% of all abdominal wall hernias, with a woman-to-man ratio of 1.4:1^{1,4,6}. Predisposing factors have been described in 100% in some studies and involve increased abdominal pressure, including factors such as obesity, chronic obstructive pulmonary disease, multiple pregnancies, constipation, ascites, previous surgeries, trauma, collagen diseases, and weight loss^{1,4,6,7}.

The preoperative diagnosis ranges from 47% to 92%, with only 50% of patients typically being diagnosed before surgery³. Imaging modalities, particularly computed tomography, are recommended for diagnosis¹. Although 90% of the cases are asymptomatic⁶, when symptoms are present, they are often non-specific, including pain, bulging, nausea, and changes to bowel movements if intestinal loops are involved⁴. Acute presentation is uncommon (20% to 30% of symptomatic patients)⁸. Complications occur in 21.1% to 35.5% of the cases, with 10.4% to 20% being due to incarceration, 6.7% to 24% to strangulation, 12.9% to intestinal obstruction, and 5.5% to subocclusions^{3,6,9}. The hernia defect is typically < 2 cm with rigid borders in 75% of the case, which explains the propensity for these complications⁵.

The hernia sac often contains peritoneal fat, but cases have been reported with the presence of the ileum, cecum, appendix, colon, stomach, ovary, uterine leiomyosarcoma, diverticula, and bladder^{3,6,10,11}.

The occurrence of acute appendicitis within a Spigelian hernia is exceptionally rare, with very few cases described. Two possible etiopathogenic mechanisms have been proposed. The more widely accepted theory suggests that the entry of the appendix into the sac exposes it to trauma, eventually leading to erosion and fibrin formation. This, added to extrinsic muscle contraction reduces, or eliminates blood supply, causing inflammation and bacterial overgrowth, ultimately resulting in acute appendicitis. Another less accepted theory suggests the possibility of intraluminal obstruction by an appendicolith in an appendix previously located within a hernia sac, leading to appendicular inflammation⁸.

The management of Spigelian hernia is surgical, either through open or laparoscopic approaches, with or without a mesh. Laparoscopic surgery is the preferred method in most series due to the well-known benefits of this approach, being extraperitoneal the preferred approach⁴. However, it requires experience for successful execution.

In uncomplicated emergency surgery, repair with a mesh via an anterior approach is advised for defects > 2 cm^{6,11}. This procedure involves opening the aponeurosis of the external oblique muscle, resecting the hernia sac, and reducing its content⁵. Herniorrhaphy should be spared for cases with a significant local infectious process to avoid placing prosthetic material in contaminated tissues⁸. Recurrence is typically rare, occurring in < 1% of the cases⁵.

Spigelian hernia is a condition known for its rarity, and the presence of an inflamed appendix within it further adds to its improbability. In this patient, several risk factors can be identified, such as age, gender, obesity, and multiple pregnancies. The significance of this case lies in various factors, including the size of the defect, the duration of evolution, the contents (notably describing the presence of the transverse colon for the first time), and surgical management. In this case, herniorrhaphy was decided due to contamination of the surgical site, the potential for wound dehiscence due to weakness in the abdominal wall at that level, and the patient's comorbidity, which increased the risk of morbidity and mortality in the event of a second intervention. Additionally, the patient experienced appendiceal disintegration due to the prolonged inflammatory process, which is a rare occurrence. This is only the second reported case of such an event¹². Ultimately, after a challenging course, the patient had a favorable outcome.

The preferred surgical treatment for this type of hernia is not yet well-established due to the limited number of cases available for statistical analysis⁴. Therefore, it is essential to continue reporting cases like this one. We should also mention that this type of hernia does not yet have an eponym for quick identification, as is the case with Amyand's hernia or De Garengeot's hernia.

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Conflicts of interest

None declared.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained approval from the Ethics Committee for analysis and publication of routinely acquired clinical data and informed consent was not required for this retrospective observational study.

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