Case of Spigelian hernia with incarcerated appendix

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ABSTRACT

Spigelian hernias are uncommon lateral ventral wall hernias with a significant rate of incarceration; these hernias often produce nonspecific clinical signs and symptoms as well as elusive imaging findings. Although there are reported cases of incarcerated appendices within Spigelian hernias, this case specifically illustrates the diagnostic difficulty these hernias present to both surgeons and radiologists. Additionally, we discuss important anatomy, demographics and risk factors, clinical symptoms, imaging pitfalls and recommendations for repair based on a review of literature.

CASE REPORT

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A 74 year-old Caucasian female initially presented to the emergency room with months of intermittent, cramp-like right lower quadrant and flank pain, which was sometimes worse when standing. Her past medical history included hyperlipidemia, hypertension, scoliosis, as well as prior transvaginal hysterectomy and spinal surgery. Physical examination at the time was unremarkable without point tenderness or palpable hernia sac. A computed tomography (CT) scan was performed, and there were no findings identified to explain the reported pain.

Two weeks later, the patient returned with new onset severe, sharp right lower quadrant pain. On physical examination, there was a subtle irreducible right lower quadrant mass with abdominal guarding and rebound tenderness. Pertinent laboratory values included a WBC count of $10.7 \times 109 / L$ (normal value range: $4.5 - 10.0 \times 109 / L$). Repeat CT scan showed a thickened, inflamed loop of bowel within a ventral hernia sac; retrospective evaluation of the prior CT scan with additional attention to the coronal images

demonstrated a non-dilated appendix extending through a narrow-lipped hernia orifice (Fig. 1a - 1d).

Emergent laparoscopic repair was employed, and the appendix was partially visualized at the base of the cecum herniating through the Spigelian fascia. As the appendix was not reducible with gentle traction, a counter incision was placed over the right lower quadrant mass. The appendix was circumferentially dissected out and amputated with a stapling device (Fig. 2a and 2b). Pathologic tissue results confirmed the appendix to be inflamed with attached fibromembranous connective tissue and a focal abscess. On follow-up evaluation in clinic one month later, the patient was doing well without surgical complication, and her abdominal pain had resolved.

DISCUSSION

Spigelian hernias were first defined by Flemish anatomist Joesph Klinkosch in 1764 as protrusions of preperitoneal fat, peritoneal sac or organ through the aponeurosis between the semilunar line and the lateral edge of the rectus abdominis muscle where the abdominal fascial layers fuse. Klinkosch named this aponeurosis after Belgian anatomist Adriaan van den Spieghel [1]. Collectively, the Spigelian aponeurosis is composed of the transversus abdominis aponeurosis, the transversalis fascia and the aponeuroses of the internal and external oblique muscles.

Etiology & Demographics:

Spigelian hernias occur most frequently in the 4-7th decades with a slight female predominance and account for 1-2% of ventral hernias [1-3]. Although these hernias are uncommon, they have a significant rate of bowel or organ incarceration, reported to be approximately 17-21% [3,4]. Most Spigelian hernias contain fat/omentum, small intestine or colon; however, Spigelian hernia sacs have been reported to contain stomach, gallbladder, a hepatic lobe post cholecystectomy, Meckel's diverticula, and ovaries [1,3-8]. There have been less than ten reported instances of Spigelian hernia with incarcerated appendix (as in our case), some cases of which were associated Crohn's disease [9-14]. These cases were all diagnosed at the point of appendiceal incarceration by examination with palpable mass, by CT scan with obvious inflamed appendix, or during exploratory laparotomy.

Risk factors for Spigelian hernia are related to increased intra-abdominal pressure: obesity, COPD/chronic cough, ascites with multiple paracenteses, weight-lifting, pregnancy. Prior abdominal surgery, placement of drains, or laparoscopic ports are also thought to be strong risk factors [4,15].

Clinical & Imaging Findings:

90% of Spigelian hernias occur near the arcuate line in the area of the umbilicus (below which the transversus abdominis and internal oblique aponeuroses combine); this is the widest and weakest part of the Spigelian fascia and is also known as the "Spigelian hernia belt" [15]. It is important to note that Spigelian hernias are commonly interstitial hernias because they rarely traverse the thick external oblique aponeurosis. Because these hernias often extend between the muscular or fascial layers, the hernia orifice is effectively concealed, and the hernia can be mistaken for normal intermuscular or subcutaneous fat; in fact, Martin et al. report 15 of 35 cases as 'interstitial' rather than subcutaneous in nature. Hernias with an interstitial location can result in a narrow hernia neck and absence of a palpable mass [2,3,7,16].

As might be expected, the clinical presentation of Spigelian hernias is quite variable; the largest case series of Spigelian hernias reported by Larson et al. found that 36% had no physical exam findings and a smaller but significant proportion (6%) had no symptoms whatsoever. Larson also reports that 29 of 81 had a palpable mass (which was easily diagnosed) but that indistinct abdominal pain was the presenting complaint in 20 of 81 [3]. Alternatively, our case (and other reported cases of herniated appendix) involved intermittent, focal flank/abdominal pain [9,13]. Additionally, pain elicited by contraction of the abdominal musculature as well as postural pain, worse when standing and relieved by laying down, appear to be common [12,17,18].

CT scan is by far the imaging modality of choice for evaluation of possible Spigelian hernia; however, due to their unique anatomy, Spigelian hernias are regularly missed. Larson reports as many as 6 of 19 CT scans in his series were false negatives. These are correctable mistakes and presumably remedied by specific clinical information and careful evaluation of multiplanar reformatted images; notice the herniated appendix in Fig. 1a is not easily visible on axial images. If carefully reviewed, CT scan can identify Spigelian hernias and differentiate both subcutaneous and interstitial types. Involved bowel loops are typically seen as tubular structures with a dilated lumen, thickened enhancing wall, and periappendiceal fat stranding. Ideally, oral contrast would be used to optimize sensitivity for bowel involvement.

Ultrasound (US) has also been described for use in diagnosis of Spigelian hernia, and in certain patients can be an effective and low-radiation alternative to CT. Specific ultrasound technique involves scanning in the transverse plane at the lateral margin of the rectus abdominis. Identification of the rectus abdominis muscle and lateral abdominal wall muscle interface is key to identifying a defect within the underlying peritoneal fat stripe [17,19]. Ultrasound can also be used for verification of hernia sac contents or in cases of equivocal cross sectional findings; although in our case, cross sectional imaging clearly demonstrated the underlying pathology and relevant anatomy. Of additional note, as MRI becomes increasingly available, the superior soft tissue contrast it provides should be of particular use for diagnosis and planning of particularly complex cases.

Differential Diagnoses:

As discussed above, mistakes in imaging evaluation are most commonly perceptual errors with missing the finding altogether. A non-dilated appendix within a Spigelian hernia or fat-containing hernia could be easily missed. Other differential considerations for Spigelian hernia include rectus sheath hematoma, abdominal wall abscess, and abdominal wall tumor [17]. In the case of rectus sheath hematoma, CT will typically demonstrate a heterogeneous fluid collection; US would show a low echogenicity collection in acute hematomas or possible calcification / septae formation in chronic hematomas. Abdominal wall abscess will present as a wellcircumscribed fluid collection with ring enhancement on CT; may demonstrate internal echoes and adjacent US hypervascularity. Abdominal wall tumors can be differentiated as the CT will typically show a solid/partially-solid mass extending from the abdominal musculature or fascial layers and internal vascularity on doppler US evaluation.

Treatment & Prognosis:

Spigelian hernias are preferably repaired due to high rates of bowel incarceration and subsequent strangulation. A total extraperitoneal procedure is preferred in cases without large hernia sac or incarcerated contents as it can avoid violation of the abdominal cavity. In urgent cases, laparoscopic approach is preferred over open repair because of its significantly lower morbidity and mortality and shorter hospital stay [18,20]. Mesh may be used in cases involving multiple wall defects or thin muscle aponeuroses [15]. **Journal of Radiology Case Reports**

The general unfamiliarity of many physicians with Spigelian hernias results in missed diagnoses. Our case was one of these misses. Increased physician awareness will likely decrease the time to diagnosis and the need for emergent repair. In the absence of palpable hernia orifice or sac, point tenderness in the area of the Spigelian aponeurosis with a tense abdominal wall should raise the question of an incarcerated Spigelian hernia.

TEACHING POINT

Spigelian hernias are uncommon ventral abdominal wall hernias with significant rates of complication in part due to delay in clinical and imaging diagnoses. Increased awareness of Spigelian hernia presentation, anatomy, and recognizable imaging features, particularly its common interstitial location, will likely decrease the time to diagnosis and the need for emergent repair.

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Figure 1: 74 year old female with Spigelian hernia; herniated appendix (a and b) and incarcerated appendix (c and d). FINDINGS:

(a and b) Axial and coronal non-contrast enhanced CT images of the abdomen at initial patient presentation demonstrate a very thin tubular structure with base at the cecum and tail extending into the subcutaneous fat.

(c and d) Axial and coronal non-contrast enhanced CT images of the abdomen two weeks after initial presentation demonstrate a dilated, blind-ending tubular structure with a thickened wall and adjacent fat stranding.

TECHNIQUE:

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Axial and coronal computed tomography images (CT; GE LightSpeed VCT) with the following parameters. Axial: 546 mA, 120 kV, 1.25 mm slice thickness. Coronal: 10 mA, 100 kV, 3 mm slice thickness.

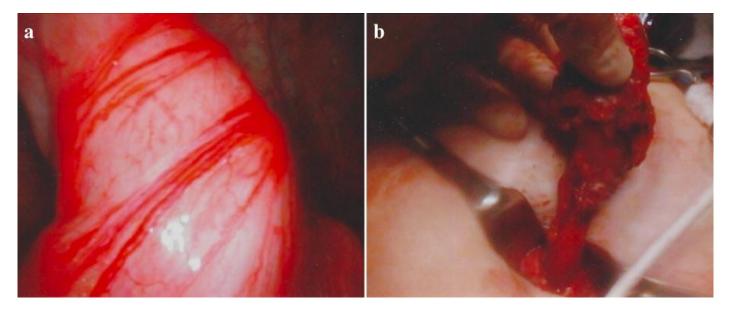


Figure 2: 74 year old female with Spigelian hernia and incarcerated appendix. Intraoperative images during emergent repair (a and b).

FINDINGS:

(a) Intra-abdominal view of the dilated, hyperemic appendix.

(b) Extraperitoneal view of the dissected appendix immediately prior to amputation.

Etiology	Protrusion of preperitoneal fat, peritoneal sac or organ through the aponeurosis between the semilunar line and the lateral edge of the rectus abdominis muscle. 90% near the arcuate line.	
Incidence	Uncommon. 1-2% of ventral abdominal hernias. Most commonly involving preperitoneal fat/omentum, small intestine, colon.	
Gender ratio	Slight female predominance.	
Age predilection	4-7th decades.	
Risk factors	Increased intra-abdominal pressure: obesity, COPD/chronic cough, ascites with multiple paracenteses, weight-lifting, pregnancy; also, prior abdominal surgery, placement of drains, or laparoscopic ports.	
Treatment	Repair required due to high rates of bowel incarceration. Total extraperitoneal repair in cases without large hernia sac or incarcerated contents. Laparoscopic over open repair preferred for urgent cases. Mesh may be used for multiple wall defects or thin muscle aponeuroses.	
Prognosis	Surgical repair is curative.	
Findings on imaging	CT: protrusion of pre peritoneal fat, peritoneal sac or organ through aponeurosis between semilunar line and lateral edge of rectus abdominis. US: best for verification of hernia sac contents.	

 Table 1: Summary table of Spigelian hernia.

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Entity	СТ	US
Spigelian hernia	Protrusion of preperitoneal fat, peritoneal sac or organ through the aponeurosis between the semilunar line and the lateral edge of the rectus abdominis. Subcutaneous and interstitial types. Often small orifice.	Best for use to for verification of hernia sac contents or in cases of equivocal cross sectional findings.
Rectus sheath hematoma	Heterogenous collection with/without adjacent inflammation depending upon age.	Acute: low echogenicity. Chronic: heterogeneous with possible calcification/septae formation.
Abdominal wall abscess	Well-circumscribed, low-intermediate density fluid collection with ring enhancement.	Fluid collection with mobile internal echoes and local hypervascularity.
Abdominal wall tumor	Solid/partially-solid mass extending from musculature or fascial layers.	Internal vascularity on Doppler evaluation.

Table 2: Differential diagnosis for Spigelian hernia.

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KEYWORDS

ABBREVIATIONS

CT = Computed Tomography

US = Ultrasound

Spigelian hernia; appendix; appendicitis; incarcerated; abdominal; computed tomography

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