



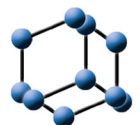
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CASE REPORT

Spigelian Hernia and Appendicitis: A Rare Case Report

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Abstract:

Introduction:

Hernias containing the appendix are well-known entities that preferentially affect the groin. The presence of an appendix within a Spigelian hernia, a rare ventral hernia that presents with nonspecific symptoms and location, adds significantly to the complexity and rarity of its diagnosis. Further, only 18 such cases were reported and published in the literature. We report a case of Spigelian hernia and appendicitis within the hernial sac to highlight the diagnostic challenges associated with this type of hernia.

Case Presentation:

In her late 30s, a female who underwent laparoscopic surgery for a uterine fibroid two and a half years back presented with right lower quadrant abdominal pain for two days. This followed abdominal pain on exertion for the previous nine months. On examination, there was tenderness, guarding, and a positive cough impulse in the right lower quadrant of the abdomen. Her body mass index was 37.53 kg/m². A contrast-enhanced computed tomography imaging of the abdomen reported a right iliac fossa incisional hernia with omental congestion necessitating an emergency laparoscopic surgery. At surgery, a Spigelian hernia was found at least 5 cm away from the port site scar of the previous surgery. The hernia contained an inflamed appendix. Thus, a diagnosis of Spigelian hernia and appendicitis within the hernial sac was made. A laparoscopic reduction and repair of the Spigelian hernia and a laparoscopic appendectomy were performed.

Conclusion:

Whereas Spigelian hernias are considerably rare, the presence of an appendix within a Spigelian hernia sac is a more infrequent occurrence. Although the imaging modalities are often inconclusive, careful interpretation of the computed tomography imaging findings may reveal the pathology preoperatively.

Keywords: Spigelian hernia, Appendiceal hernia, Appendix, Appendicitis, Case report, Surgery.

Article History

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1. INTRODUCTION

Hernias containing the vermiform appendix, otherwise termed appendiceal hernias, are well-known entities described in the literature. These hernias preferentially affect the groin, in particular the right inguinal region [1]. For instance, the eponymously reported Amyand and De Garengeot hernias, which are inguinal and femoral hernias, respectively, containing the appendix, were described nearly three centuries ago [2].

Constituting only 0.1 to 2 percent of all ventral hernias [3], Spigelian hernia was named after Adriaan van den Spieghel, who first described the semilunar line (linea semilunaris) in 1645, almost a century before the hernia was first discovered

by Josef Klingosh in 1764 [4]. It constitutes a diagnostic challenge because of its nonspecific symptoms and location [5]. Further, the presence of an appendix within a Spigelian hernia sac is yet a more infrequent occurrence; and the diagnosis of this entity poses a major problem. We reported a case of Spigelian hernia and appendicitis within the hernial sac to highlight the diagnostic challenges associated with this type of hernia.

2. CASE PRESENTATION

2.1. Patient Information

A female in her late 30s presented with right lower quadrant abdominal pain for two days. There was no associated nausea or vomiting, change in bowel habits, or urinary complaints. The patient had abdominal pain on squatting in the

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preceding nine months. She also had a history of laparoscopic surgery two and a half years back for a big fundal, subserosal fibroid, which measured around 16–18 cm and extended to the left costal margin. The surgery involved laparoscopic myomectomy, endo suturing, and morcellation under general anesthesia. The past medical history and family history were insignificant.

2.2. Clinical Examination

On examination, there was tenderness with guarding and a positive cough impulse at the right lower quadrant. Moreover, no obvious mass could be felt, and there was no abdominal distension. Her body mass index was 37.53 kg/m².

2.3. Diagnostic Assessment

The laboratory investigations showed a normal white blood cell count of 10.81×10^9 cells/L. The absolute neutrophil count was slightly elevated to 7.16×10^9 cells/L, and the C-reactive protein was increased to 12.2 mg/dL. The remaining investigations were within normal limits.

An abdominal ultrasound (US) was done as the initial radiological investigation, and it showed a right iliac fossa incisional hernia but was inconclusive and required further confirmation by an abdominal computed tomography (CT) scan with contrast. The CT scan reported an anterior abdominal

wall muscular defect with herniated bowel loops, which showed focal wall thickening and enhanced vascularity. These findings were thought to suggest a right iliac fossa incisional hernia with omental congestion of the herniated small bowel loop (Figs. 1 and 2).

2.4. Therapeutic Intervention

The patient was taken to the operation theater for emergency laparoscopic surgery. Intraoperatively, the hernia was found at the Spigelian aponeurosis at least 5 cm away from the incision of the port site of the previous surgery. It was also seen to contain an inflamed appendix (Fig. 3). Thus, the likely diagnosis was made intraoperatively as follows: Spigelian hernia and appendicitis within the hernial sac. Further supporting our argument that this is a Spigelian, rather than an incisional, hernia is the fact that the 5 mm port site scar was too narrow to allow a hernia to pass through. Additionally, on the reassessment of the abdominal CT scan, the hernia was noted to protrude from the typical Spigelian hernia site. It is, however, important not to negate the unlikely possibility that this hernia was an incisional hernia that herniated through the 5 mm port site. This potential diagnosis should be kept in the differential diagnosis. A laparoscopic reduction and repair of the Spigelian hernia and a laparoscopic appendectomy were done. Ultimately, the hernial defect was closed using prolene 3/0 interrupted sutures.

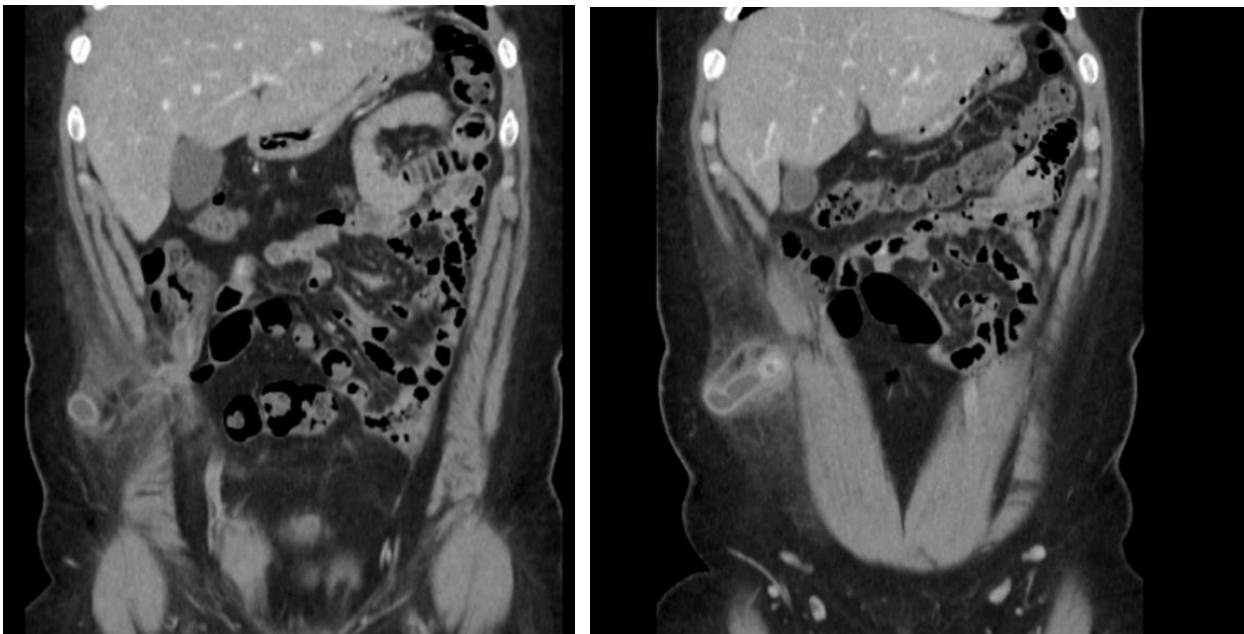


Fig. (1). Sections of the abdominal CT scan in the coronal plane are depicted. It was initially thought to show an incisional hernia containing a loop of the bowel. Intraoperatively, however, an inflamed appendix was seen to herniate through the Spigelian hernia defect on the right side. Thus, the diagnosis was made intraoperatively at the time of laparoscopy.

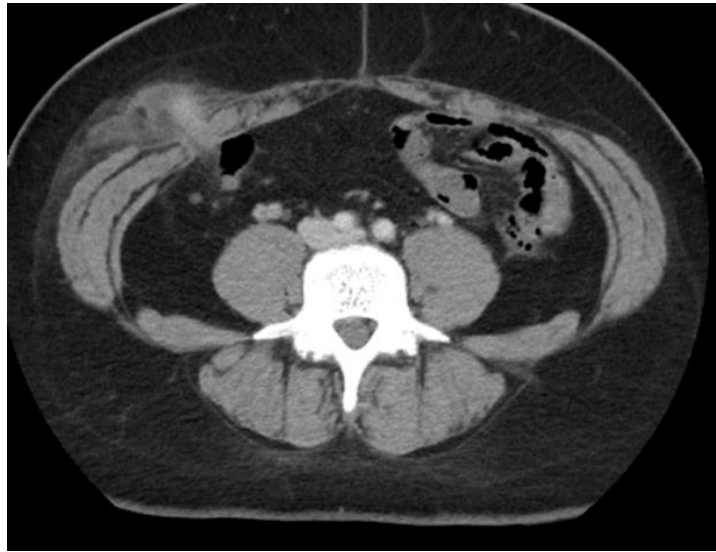


Fig. (2). A section of the abdominal CT scan in the axial plane is depicted. It shows the appendix herniating through the Spigelian hernia defect on the right side.

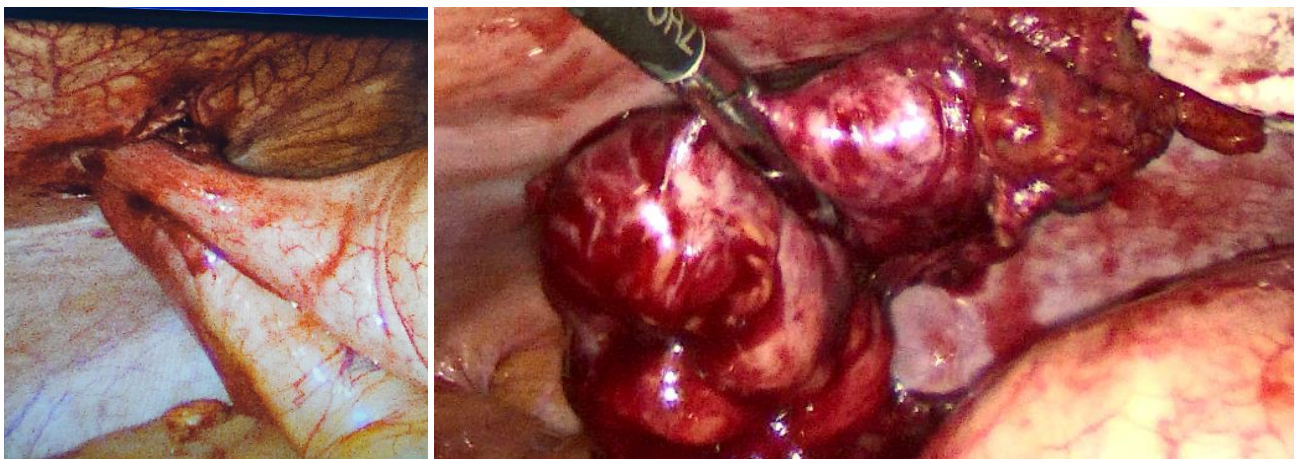


Fig. (3). Laparoscopic views of the inflamed appendix are shown. This appendix was found to herniate through the Spigelian hernia defect at the time of surgery.

2.5. Follow-up and Outcomes

The patient developed pulmonary edema postoperatively and was treated with oxygen and diuretics. Afterwards, she improved, was discharged in two days, and had an uneventful recovery. Gross evaluation of the resected specimen showed fragmentation of the appendix with a blocked lumen. Microscopic evaluation revealed moderate acute inflammation and increased eosinophils in the mucosa, mixed inflammation in the serosa, focal ulcerations, wall hypertrophy, and peri-appendiceal fibrosis. The final pathological diagnosis was acute-on-chronic appendicitis with increased eosinophils.

3. DISCUSSION

Spigelian hernia is a rare type of hernia that occurs when preperitoneal fat, abdominal viscera, or peritoneal sac protrudes through a defect in the Spigelian aponeurosis [4].

The latter is merely the aponeurosis of transversus abdominis muscle environed laterally by the linea semilunaris, which is the point where the transversus abdominis muscle transforms into its aponeurotic tendon, and medially by the lateral edge of rectus abdominis muscle [4, 6]. The so-called Spigelian belt describes a 6 cm zone located transversely in the lower abdominal wall between the interspinous plane and a plane that is 6 cm cranial to it but caudal to the umbilicus [7]. Notably, the Spigelian aponeurosis is widest in this area, and up to 90 percent of Spigelian hernias occur in this region, particularly at the intersection between the semilunar line and the arcuate line of Douglas [7, 8]. The hernial ring is a well-defined defect; the sac is often interparietal, where the hernia passes through defects in the transversus and the internal oblique aponeuroses and appears beneath the intact aponeurosis of the external oblique. Alternatively, the sac can lie in the rectus sheath beside the rectus muscle [7].

Most cases of Spigelian hernias present in the fourth to seventh decades of life with a slight female predominance, and, contrary to our case, they happen in the left side of the abdomen. The risk factors for Spigelian hernias do not differ much from other ventral hernias [9]. It is important, however, to note that one of the risk factors for ventral hernias is prior injury or port site incision and that such cases should be clinically classified as incisional hernias [10]. Despite a prior history of laparoscopic surgery for fibroids on the same side of the hernia, our case was diagnosed with Spigelian hernia as it was found to be unrelated to the incision during laparoscopic surgery. Spigelian hernias commonly present with pain and an abdominal mass. On physical examination, tenderness and a mass may be appreciated in the location of the Spigelian aponeurosis [9]. Physical examination alone is often indecisive due to abdominal fat and the external oblique fascia (the hernia is usually located beneath the intact fascia) [7].

US, CT, and magnetic resonance imaging (MRI) are the imaging modalities that can be employed in diagnosing Spigelian hernias. Out of these, US is the first-line imaging modality. Accentuation of the hernia by maneuvers that increase intra-abdominal pressure is possible during ultrasonography. If the US proved indecisive, a CT scan with contrast is the investigation of choice. When the diagnosis remains unclear, a diagnostic laparoscopy is indicated; this offers the added benefit of immediate repair in the case of positive findings [11, 12]. Due to the lack of evidence, there are no specific recommendations on whether an open or laparoscopic approach should be employed, and the decision on which method to perform is in the surgeon's hands [13]. There is no difference in recurrence rates between the two approaches. The open approach, however, is associated with more morbidity and a longer hospital stay [14].

The presence of an appendix in the hernial sac of a Spigelian hernia is very rare. On review of the literature, we only found 18 published similar cases [2, 8, 11, 12, 15 - 28]. Similar to our case, most cases were not explained by imaging modalities and exploratory laparoscopy was employed. In cases of appendicitis or gangrene, besides the repair of the hernia defect, an appendectomy was also performed [2, 8, 11, 12, 15, 17 - 22, 24, 28]. It is important to note that in one of the cases where CT was inconclusive, a retrospective study of the same CT scan with attention to coronal scans detected an appendix through the hernial orifice [15]; this indicates that more attention to CT can sometimes reveal the pathology preoperatively. Nevertheless, whether or not an appendix is detected on CT does not change further management, as exploration and repair would still be the line of management.

CONCLUSION

In conclusion, diagnosing a Spigelian hernia is challenging because of its nonspecific symptoms and location; and while it is remarkably rare, the presence of an appendix within its hernial sac merely adds to the complexity and rarity of its preoperative diagnosis. Although the imaging modalities can often be inconclusive, careful interpretation of the CT findings may reveal the pathology preoperatively.

PATIENT PERSPECTIVE

"I started to have severe lower abdominal pain one and a half years back, which I self-treated by resting. About a few weeks before I got admitted, I started weightlifting exercises for my lower body, this was when the pain slowly grew and, at one point, was not relieved. My husband took me to our doctor, who immediately recommended that we visit the hospital. I had a "keyhole" surgery two and a half years back to remove a fibroid in the upper part of my womb. Thus, I felt a bit more prepared for a similar surgery for my hernia. Yet, this surgery was a bit different; the anesthetic worked on me late, and my eyes were still open when they got me inside the operating room. The last thing I remembered was the feeling of my eyes being glued and pipes going in through my throat.

Similar to my post-surgical care in the previous surgery, I had to use breathing practice equipment right after surgery, with the need to use an oxygen mask for a few days to maintain the oxygen rate. I am not worried about my appendicitis, as I felt this problem was waiting to 'burst' anytime. I am more concerned about the hernia. As I am approaching 40, I fear a future hernia may come again. Along with my husband, we are changing our lifestyle to increase the chance of a healthier outcome."

AUTHORS' CONTRIBUTIONS

All the authors met the four criteria of the International Committee of Medical Journal Editors (ICMJE) for authorship. ZE designed and contributed to the conception, drafting, and revising of the article; HWI contributed to the conception and drafting of the article; TK contributed to the acquisition of data and conception and drafting of the article; BTG contributed to the conception, drafting, and revising of the article; and AM contributed to the acquisition of data and revising of the article. All authors read and approved the final version of the manuscript and agreed to the accountability of their contributions.

ABBREVIATION

CT = Computed Tomography

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The study was approved by the RAK Medical and Health Sciences University Research Ethics Committee (RAKMHSU-REC) and the Ministry of Health and Prevention (MOHAP) Research Ethics Committee.

HUMAN AND ANIMAL RIGHTS

No animals were used for studies that are the basis of this research. All the humans were used following the ethical standards of the committee responsible for human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2013 (<http://ethics.iit.edu/ecodes/node/3931>).

CONSENT FOR PUBLICATION

Verbal and written consents were obtained from the patient to publish the text and figures of this article, with anonymity maintained.

STANDARDS OF REPORTING

CARE guidelines were followed.

AVAILABILITY OF DATA AND MATERIALS

The data supporting this study's findings are available within the article.

CONFLICT OF INTERESTS

The authors declare no conflict of interest, financial or otherwise.

FUNDING

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