Spigelian hernia
A series of cases and literature review

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AIM: *The aim of this case series is to describe the pathophysiological, clinical, and radiological features of Spigelian hernias, and to describe their modern surgical management.*
MATERIAL OF STUDY: *We describe the clinical management of four cases of Spigelian hernias, and we discuss the main issues and controversies in the current scientific literature.*
RESULTS: *In all the cases the clinical suspect arouses during the clinical examination of the patients, and it was confirmed through imaging techniques. All cases were treated surgically by an open approach; no intraoperative or postoperative complications occurred, and no recurrences were registered.*
DISCUSSION: *Our experience and the literature review that we carried out confirm that Spigelian hernia is a rare condition, and its diagnosis should always be based on a thorough clinical examination and possibly a ultrasound or computed tomography, which are important for the study of the exact anatomical features of the defect, and for the planning of the adequate operative approach. The laparoscopic technique is effective but used in few reference centers, both for the rarity of the disease and the long learning curve, whereas the open surgical correction is the most common approach, with excellent results, especially in cases where urgent repair is needed.*
CONCLUSION: *The surgical treatment of Spigelian hernias has excellent results when a thorough clinical – radiological evaluation is carried out, and the appropriate surgical approach is chosen.*

KEY WORDS: Abdominal wall, Spigelian hernia, Surgery

Introduction

Spigelian hernia is a rare surgical condition representing 0.1-2% of all the abdominal wall defects. Its incidence peak occurs between 40 and 70 years of age, with a 1:1.18 male to female ratio [1,2]. Diagnosis is mainly clinical and can be challenging, especially in early stages, due to both the non-specific signs and symptoms and the poor objective findings, given the presence of the prefascial tissue which makes the clinical evaluation difficult. Only when the herniation increases in volume it becomes palpable, and pain is the most frequent symptom, especially when the abdominal wall is contracted. Surgery is the only therapy, as it is unanimously described in the scientific literature, especially in the light of the high risk of incarceration which amounts to 35% of the cases [3]. Surgery avails itself of the common techniques for repairing the abdominal wall, both through the use of autologous tissue or by interposing prostheses; open surgery is the most common operative

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TABLE I - *Demographic and clinical data of the patients involved in the study.*

Patient	Sex	Age	Side	Previous/ other hernias	Prosthesis	Complications	Recurrence
1	F	75	L	Yes	No	No	No
2	F	68	L	Yes	No	No	No
3	M	73	L	Yes	Yes	No	No
4	F	58	L	No	Yes	No	No

approach, even if minimally invasive approaches have been largely adopted in the last decade.
In this article we describe the clinical management of four cases of Spigelian hernias, and we discuss the current issues and trends through a comprehensive review of the scientific literature.

Materials and Methods

Demographic, clinical and surgical data of four consecutive patients who underwent surgical correction of a Spigelian hernia in our institution in the last 2 years were reviewed. All clinical information were obtained from clinical records and referrals; the main data are summarized in Table I.
All patients underwent open elective surgery, with or without the use of a mesh. No laparoscopic approaches were used in this series. All the procedures were performed by experienced general surgeons. A surgical incision was performed in correspondence of the abdominal wall defect; the length of the incision was adjusted on the basis of the diameter of the hernia. The surgical access allowed the isolation of the hernia sac by blunt dissection. After isolating the sac, it was reduced within the abdomen or opened and the adhesions between the sac itself and its content were sectioned (Fig. 1); the intestines involved appeared vital in all cases, and they were easily reduced in the abdomen. The correction of the defect was performed by direct suture or by the use of a preperitoneal prolene mesh fixed with interrupted stitches, and subsequently covered by the aponeuroses of the internal and external oblique muscles. Follow-up was performed through clinical examination at one, six and twelve months.
In order to review the current literature a methodological research was performed for English language articles on Pub Med. The key words used were: "Spigelian Hernia" "Spigelian Hernia repair" "Spigelian Hernia treatment" "Spigelian Hernia management". Only articles in English were selected. Titles and abstracts were evaluated in order to include the most relevant studies. References of the selected articles were cross-checked in order to detect papers missed by the search engine.

Results

Three of the four patients included in this study were females, and only one was male. The mean age was 68 years (range 58-75). The Spigelian hernia was sited at the left side in all the cases (Fig. 2); 75% of the case had a history of an inguinal hernia, in all the cases sit-

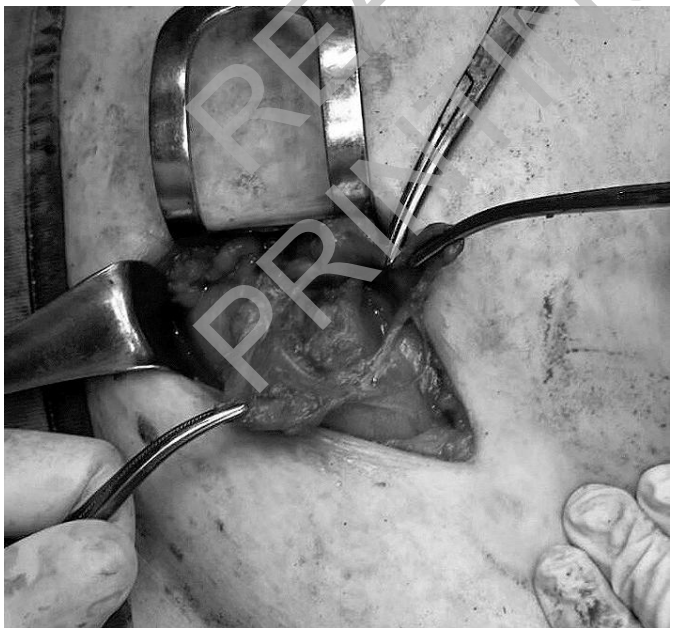

Fig. 1: Open approach for the correction of Spigelian hernia.

Fig. 2: Spigelian hernia of the left side of the abdominal wall.

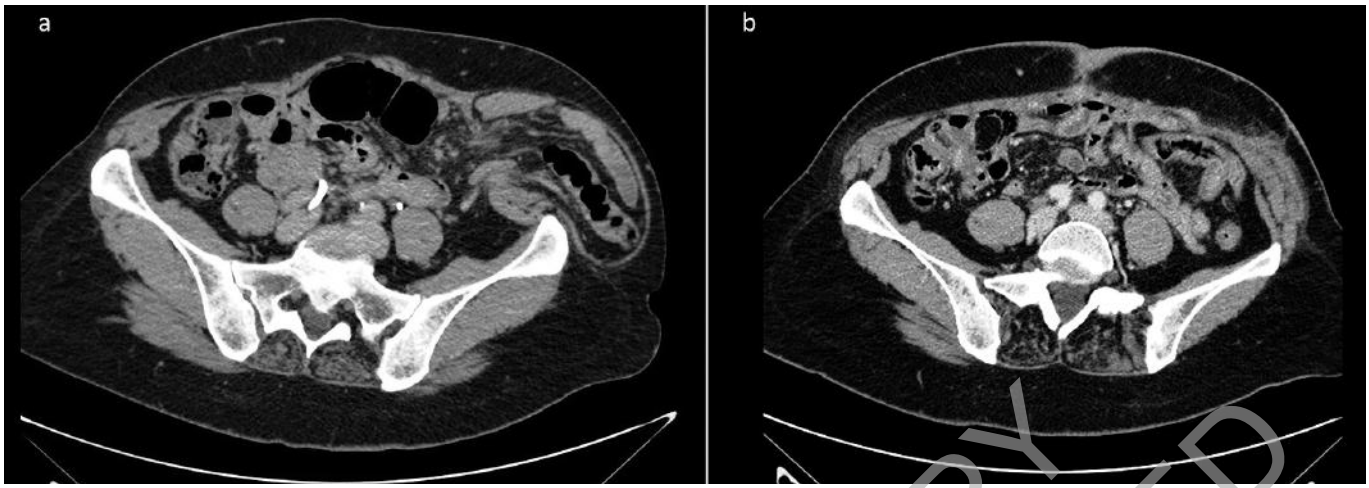

Fig. 3: Preoperative (a) and postoperative (b) CT scan of a left-sited Spigelian hernia.

ed at the right side. Two clinical manifestations were present in all cases: abdominal wall swelling and local pain. Ultrasonography (US) was employed in all the cases, while computed tomography (CT) scan was used for the preoperative evaluation in only one case (Fig. 3). All patients were treated successfully with the surgical technique described above. A prolene prosthesis was used in two cases, while in the remaining cases a direct suture was employed. The mean hospital stay was 3 days, and no local or systemic complications were observed during and after surgery (Table I). Up to date no recurrences occurred (mean follow-up time 9 months).

The research on Pub Med produced 30 significant articles, published in the period 1984/2014; only seven of these were complete revisions, whereas the remaining articles were sporadic clinical cases. Table II summarizes the articles with the greater number of cases included in this review.

TABLE II - *The main articles pubblished in the last 30 years and the number of Spigelian hernias reported per single article.*

Author	Period	N° of cases
Larson et al, 2002	(1976-1997)	76
Vos et al, 2004	(1980-2002)	25
Ruiz de la Hermosa et al, 2010	(2001-2008)	39
Moreno-Egea et al, 2002	(1994-2001)	28
Moles Morenilla et al, 2005	(1986-2005)	21
Leon et al, 2011	(1998-2008)	23
Perrakis et al, 2012	(2000-2010)	16
Cinar et al, 2013	(2005-2011)	9
Mittal et al, 2008	(1997-2007)	10

Discussion

Spigelian hernia is a rare and particular defect of the abdominal wall.

It represents approximately 0.1-2% of all the abdominal wall hernias [3]. In 1742 Henry-Francis Le Dran reported a case of spontaneous rupture of the band along the linea semilunaris [4], in 1764 Klinkosh published the case of lateral abdominal hernia which name, [5] however, was assigned after Adrian van der Spiegel, Belgian anatomist who dedicated his studies to the description of the linea semilunaris of abdominal wall (lunate) [6]. This line, characterized by a lateral convexity, marks the transition from the muscular abdomen to the aponeurosis of the transversus abdominis; the area located between the semilunar line and the lateral margin of the rectus abdominis muscle is called "Spigelian fascia" [7,8].

Spigelian hernias are more often located below the umbilical line, as occurred in all the cases included in our series. In their early stages of development they are often difficult to diagnose because they origin below the intact fascia of the external oblique muscle, and the hernia sac passes through the aponeuroses of the transversus and internal oblique muscles [9]. The defect is generally located in the confluence between the lunate fascia and the semicircular line of Douglas, in accordance to the most accredited pathophysiological theory of Zimmerman et al. [10] The sac only in sporadic cases reaches the subcutaneous plane, becoming clinically evident and presenting the classic shape of a fungus, classically protruding in the lateral wall [11].

Spigelian hernias have a major impact around the fifth and sixth decade of life; also in our series the ages of the patients were within such range [12-14]. Apparently there is no substantial difference in incidence between the two sexes; Moreno-Egea signals a greater involvement of women and a more frequent left localization compared

with the contralateral [15]. Also in our series most of the cases occurred in women, and in all cases they were localized in the left side of the abdomen.
As for the etiology, the majority of cases are acquired hernias, because the Spigelian fascia itself is among the less resistant areas of the abdominal wall. Predisposing factors are obesity, COPD, multiple pregnancies, constipation, ascites, previous abdominal surgery (mainly laparoscopic), a rapid weight loss, abdominal trauma and peritoneal dialysis [16]. Differential diagnosis should be taken into account more frequently with conditions such as haematoma, lipoma, abscess, neoplasia, muscle-tendon inflammations and diverticulitis [17-19]. Since in one of our cases hernia was associated with a concomitant uterine malignancy, we assessed if an increased incidence of the disease exists in patients with concomitant gynecological pathology. In this regard there are no specific data, but only a few case reports that do not confirm such incidence increasing [20,21].Instead, the only correlation found was between Spigelian hernia in childhood and cryptorchidism: in this case a consistent evidence exists, and some authors describe it as a specific syndrome [22,23]. Furthermore, in 75% of our patients at least one other defect of the abdominal wall was observed (right inguinal hernia in two cases, a right femoral and inguinal hernia in one other case); this suggests that in individuals with Spigelian hernia a predisposition to abdominal wall defects may exist, and it may act along or in combination with the risk factors mentioned above.
The most common symptoms of Spigelian hernias are: pain located at the level of the palpable swelling, diffuse abdominal pain,[14] occlusive symptoms with closure of the alvus to faeces and gas, nausea and vomiting in cases of strangulation that requires urgent surgical intervention [1,13,24]. Only abdominal wall swelling and local abdominal pain were present in the patients of our series. As already mentioned, the clinical diagnosis of the Spigelian hernia is not always feasibile, especially in small hernias, for this reason instrumental examinations are essential in most cases [25-27]. US of the abdominal wall is the examination most commonly employed (2% false negatives), and it generally allows a prompt detection of the wall defect and evaluation of its size [11,28]; moreover, cases of reduction of incarcerated Spigelian hernias under ultrasound guidance have been reported [29,30]. US was used in all cases in our experience. When the local conditions are difficult to interpret, especially when the defect occurs in conjunction with other abdominal diseases, a CT scan is recommended. CT scan is crucial in identifying the defect of the wall, and evidencing any organs contained in the sac. [3,31]
The exact definition of the location and size of the defect and its content are decisive for the proper treatment of the disease, especially in the case of a possible laparoscopic approach [32].
CT scan was used only in one case in our experience, in a women affected by a synchronous gyneacological tumor. In this case, indeed, CT scan had a double utility: evaluate the anatomical features of the Spigelian hernia, and staging the uterine cancer.
The treatment of Spigelian hernias is essentially surgical, and due to its high incidence of complications (35%, strangulation, incarceration) , surgery is recommended in almost all cases [3]. The optimal surgical approach for repair is not yet well defined, as surgeons can use a traditional open approach or a laparoscopic technique. The former is based on the same general principles used in repairing any type of abdominal wall defect: a pararectal incision is commonly preferred, while a median laparotomy is chosen in cases of complicated Spigelian hernias, as reported by Moles-Morenilla. After freeing the sac and reducing its content in the abdomen, the restoration of the muscle fascia is carried out with or without reinforcement by the use a prosthesis; the latter is generally used in cases of large defects or when the aponeurotic fascia is very loose or atrophic (9 33 bibl kumar hiseh) [9,33]. This is the technique we used in our series, and in two cases we used a prolene prosthesis to reinforce the abdominal wall.
Carter and Mizes treated Spigelian hernias with a laparoscopic approach for the first time in 1992 [34]. The laparoscopic repair can be performed through two different accesses: a transabdominal access and an extraperitoneal access. The latter was published for the first time by Moreno-Egea et al in 1999 [35], and appeared preferable at first; recently the same Authors reconsidered the intraperitoneal technique, which better allows to avoid contact with the intra-abdominal organs, describing it as the best current technique for the laparoscopic treatment of Spigelian hernias [36]. The laparoscopic technique is constantly evolving and became today the gold standard approach for not complicated Spigelian hernias in reference centers; it still requires a good learning curve which may require several years of practice, given the rarity of the disease. For this reason, especially in cases of complicated Spigelian hernias, the open technique with or without implants remains the recommended approach [32,37,38].

Conclusions

Spigelian hernia is a rare condition, with only a limited number of cases pubblished in the scientific literature. Data from our experience, and from the review of the literature suggest that the diagnosis of Spigelian hernias requires a thorough clinical examination, and often imaging evaluation with US or CT scan; such approach is essential for the study of the exact location of the defect and its anatomical characteristics, as well as for the planning of the correct therapeutic strategy. The surgical correction with a traditional open technique is the approach most commonly used, particularly in those cases where urgent repair is needed. The laparoscopic technique is valid and effective, especially when used in specialized centers.

Riassunto

Lo scopo del presente articolo, che include una serie di casi di ernia di Spigelio, è quello di illustrare le caratteristiche fisiopatologiche, cliniche e radiologiche di questo difetto della parete addominale e di discutere il suo management chirurgico, sulla base delle attuali evidenze scientifiche riscontrate attraverso una revisione della letteratura moderna. A questo scopo quattro casi di ernia di Spigelio vengono descritti: in tutti i casi il sospetto è stato posto attraverso l'esame clinico del paziente e confermato con delle metodiche di imaging. Tutti i pazienti sono stati trattati chirurgicamente, con un approccio a cielo aperto; non ci sono state ne complicanze intra- o post-chirurgiche, ne recidive. La nostra esperienza e la revisione della letteratura che abbiamo effettuato ci confermano che la diagnosi di questo raro difetto della parete addominale si deve basare su un attento esame clinico – radiologico, cosi da poter individuare nel dettaglio le sue caratteristiche anatomiche e scegliere l'adeguato approccio chirurgico. L'approccio laparoscopico può essere eseguito in centri con esperienza in queste tecniche, mentre l'approccio tradizionale a cielo aperto è quello più diffusamente eseguito e garantisce ottimi risultati, specialmente in regime di urgenza.

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