ORIGINAL RESEARCH PAPER
Volume - 14 | Issue - 06 | June - 2025 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

"A RARE ENCOUNTER: SPIGELIAN HERNIA AND CRYPTORCHIDISM SYNDROME EXPLORED"

General Surgery

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ABSTRACT

We have reported a case of spigelian hernia with cryptorchidism in 39 years old male patient. Patient was examined clinically and evaluated radiologically. Radiologically he was diagnosed with the spigelian hernia with cryptorchidism due to absence of right testis in right side of scrotal sac. Patient then planned for onlay mesh repair, during which testis was found in hernial sac without gubernaculum, which suggests a part of new syndrome [2][4]. This case underscores the importance of through clinical and imaging evaluations in case of spigelian hernia to detect the syndromic evidence and to support the unreported syndrome with very limited literature available regarding it.

KEYWORDS

BACKGROUND:

Spigelian hernia is named after Adriaan van der Spieghel, in 1645 [2] but Josef Klinkosch in 1764 first defined the spigelian hernia as a defect in the semilunar line [1]. Spigelian hernia is a rare constituting 0.12% of all ventral hernias [2] defined as herniation of abdominal contents or peritoneum through defect in spigelian fascia.

Spigelian fascia is found between the lateral border of rectus abdominis muscle and the semilunar line, It is formed by the aponeurosis of the internal oblique at its line of division to enclose the rectus. This is reinforced anteriorly by the external oblique, and posteriorly by the transversus abdominis above the arcuate line. it extends from the costal cartilage to the pubic tubercle. Hernia sac usually contains peritoneal fat, in most of the cases testis, followed by small intestine, sigmoid colon, omentum, gallbladder, stomach, Meckel's diverticulum, ovary, while herniation of bladder has been rarely reported. Raveenthiran et al suggested that a defect in the spigelian fascia, combined with the ipsilateral cryptorchidism, may be part of a new syndrome [3,4]. The additional lack of a gubernaculum observed in this patient, as well as the lack of an inguinal canal observed by others in similar cases, may be considered as two additional and closely connected elements of this new syndrome [4]. Traditionally, repair options include open anterior herniorrhaphy, mesh repair via laparoscopy, and orchidopexy or orchidectomy based on risk factors

INTRODUCTION:

Spigelian hernia is a rare condition. Compared to other types of hernias, spigelian hernia accounts 1-2% of all hernias, while it constitutes about 0.1-0.2% of all abdominal wall hernias [2].

It has been reported that 75 % of male infants with Spigelian hernia present an ipsilateral cryptorchidism and commonly present with swelling of the abdominal wall or intestinal obstruction [18,19]. Spigelian hernia with associated cryptorchidism was first reported in 1895 in a child by Schoofs and was implicated to be a syndrome, now termed as Spigelian-Cryptorchidism Syndrome, by Raveenthiran et al in 2004[3]. Since then, more and more cases have been reported, mostly in the paediatric age group [18]. Most spigelian hernias in adults are secondary to trauma or an increase in intra-abdominal pressure, whereas in paediatric age group, these are mostly congenital. [9,19]. The testis is found within the hernia sac in 87 % of those patients [4].

CASE:

A 39 years old male patient clerk by occupation came to our hospital with a complaint of a swelling in the right side of abdomen for 5 months. The patient was hypertensive with no other co morbidities. On clinical evaluation, a 4*3cm, ovoid, soft, non-tender swelling was palpated in right iliac region of abdomen. Patient underwent ultrasound imaging, which was suggestive of defect in the anterior abdominal wall in right iliac region with herniation of omentum and bowel.

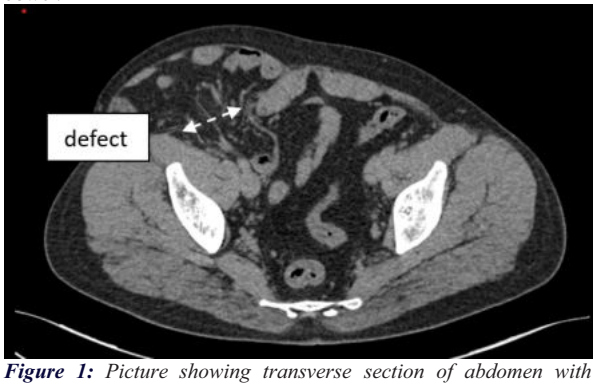


Figure 1: *Picture showing transverse section of abdomen with anterolateral abdominal wall defect.*

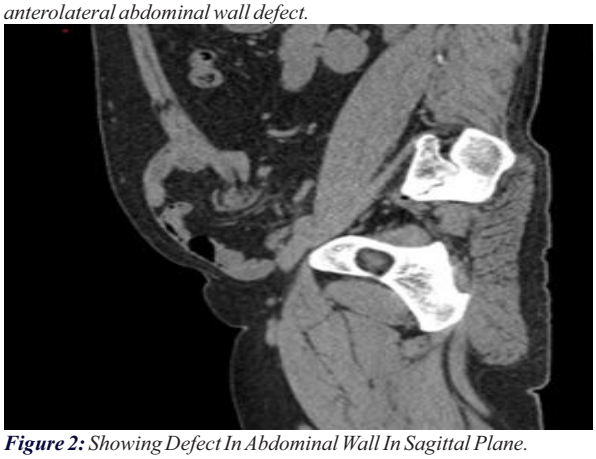


Figure 2: *Showing Defect In Abdominal Wall In Sagittal Plane.*

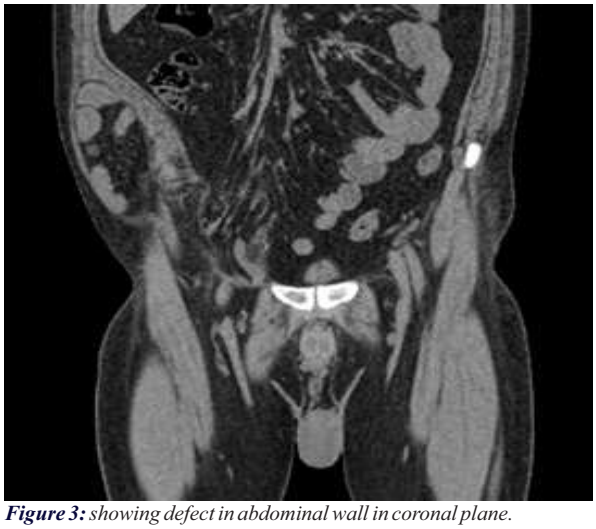


Figure 3: *showing defect in abdominal wall in coronal plane.*

For the precise determination of the defect and subsequent decision on the appropriate surgical intervention, a computed tomography was done which is suggestive of defect of size 4.9*4.7cm in anterolateral abdominal wall in right iliac fossa with herniation of ilieal loops and mesentery with absence of right testis in right scrotal sac or inguinal region evocative of right spigelian hernia with cryptorchidism.

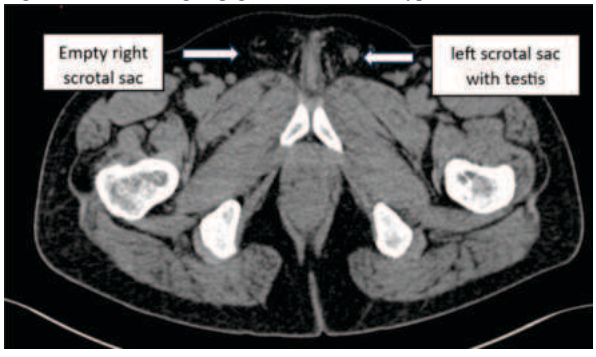


Figure 4: *picture showing empty right scrotal sac and testis in left scrotal sac.*

considering the above clinico-pathological and radiological findings of the patient it was decided to go ahead with onlay mesh repair with right orchidectomy. Intra operatively right testis was found in hernial sac without gubernaculum and ilieal loop with its mesentery as content of hernia which was reduced first followed by repair of defect with onlay mesh hernioplasty and right sided orchidectomy. Specimen was sent for further histopathological evaluation which was suggestive of non- malignant state of testis with atrophic changes.

Patient was discharged after 2 days of procedure with no complication.

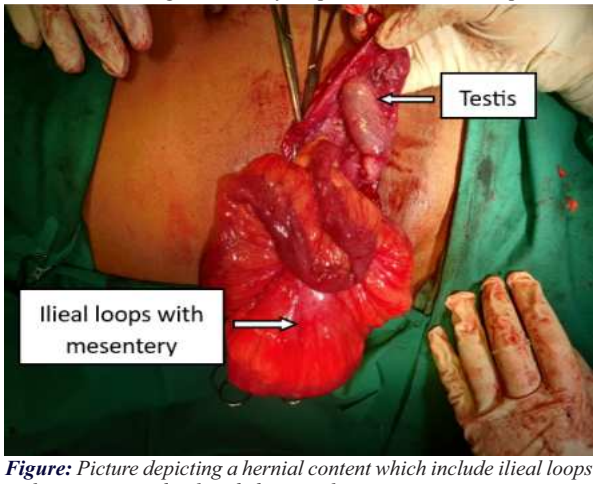


Figure: *Picture depicting a hernial content which include ilieal loops with mesentery and right sided testis in hernia sac.*

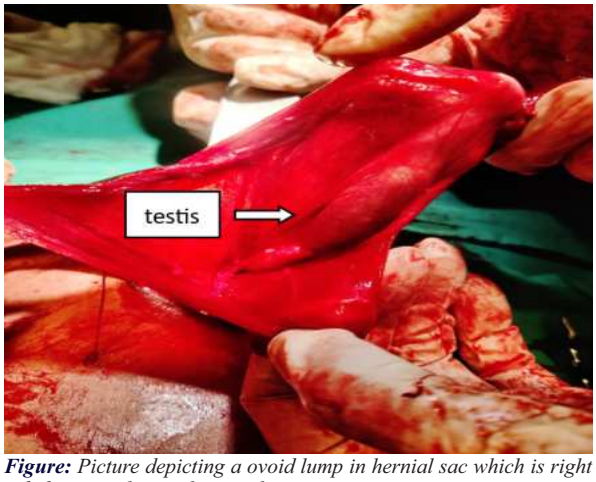


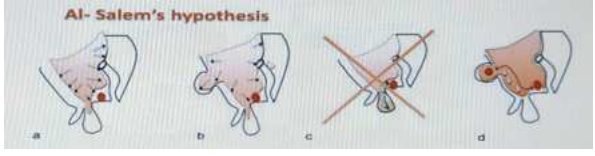
Figure: *Picture depicting a ovoid lump in hernial sac which is right sided testis without gubernaculum.*

DISCUSSIONS:

After thorough investigation of the relevant literature, it has been suggested that spigelian hernia is associated with ipsilateral cryptorchidism at about 28%-75% of male paediatric patients [4,6,7]. the testis is found within the hernia sac in 87% of those patients [4].

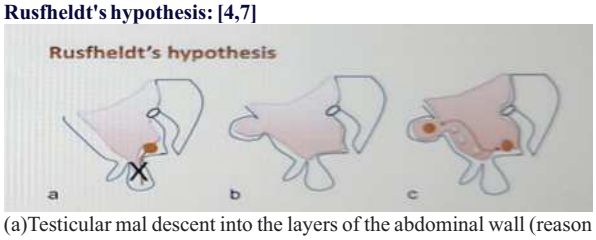
Spigelian-cryptorchidism syndrome remains a relatively uncommon finding in adult males with a few cases being reported past puberty. About 35% cases of spigelian hernia are reported to be associated with other congenital anomalies [5]. There are three theories which supports the cryptorchidism in spigelian hernia[7].

Al-Salem hypothesis: [7,9]



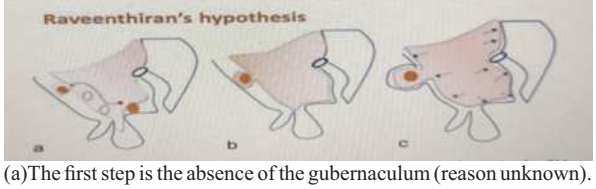
(a)Appearance of the normal intra-abdominal pressure. (b) Spigelian hernia leads to a decrease in intra-abdominal pressure. (c) Because of intra-abdominal pressure insufficiency, the testis could not descend into the scrotum. (d) As a result, ectopic testis occurs in the Spigelian hernia, which is the weak point.

Rusfheldt's hypothesis: [4,7]



(a)Testicular mal descent into the layers of the abdominal wall (reason unknown). (b)Ectopic testis drags down its processus vaginalis during descending, and this peritoneal processus forms a potential hernial sac. (c) Increased intra-abdominal pressure opens up the potential sac. As a result, a Spigelian hernia with an ectopic testis occurs.

Raveenthiran's Hypothesis: [3,7]



(a)The first step is the absence of the gubernaculum (reason unknown). (b) Concurrent Spigelian hernia independently from absence of the gubernaculum (reason unknown). (c) The testis that does not descend to the inguinal canal uses the Spigelian defect as an escape point. As a result, an ectopic testis occurs in the Spigelian Hernia.

In the hypothesis presented here, the absence of the gubernaculum and the inguinal canal and the observation of the testis in the Spigelian hernia

sac together with the fact that this coexistence cannot be explained support the hypothesis that these findings are components of a syndrome.

A syndrome is a complex of complaints and findings that seem to be independent from each other but appear as a single disease when they come coexist. The development of a finding, disease or disorder is dependent on the development of another disease or disorder in the sequence. Therefore, how one disease or pathology causes another disease or disorder in sequence is obvious. Hence, if the causal relationship between Spigelian hernia and Undescended Testis could be elucidated, the Spigelian Hernia and the Undescended Testis could be evaluated as components of a sequence. In conclusion, neither the theories suggesting that a Spigelian Hernia leads to an Undescended Testis nor those suggesting that a Undescended Testis leads to an Spigelian Hernia are satisfactory. Therefore, we accept the view that this coexistence may be the congenital Spigelian-cryptorchidism syndrome seen in boys [7,8,10].

Diagnosing Spigelian hernia remains a challenge for clinicians due to the inter-parietal location of the hernial sac, especially in adults. Spigelian hernias are differentiated from other ventral wall hernias due to the location of the hernial sac. Direct inguinal hernia protrudes through a weakness in posterior wall of inguinal canal, medial to the epigastric vessels, through the Hasselbach's triangle whereas indirect inguinal arises through the deep ring and enters the inguinal canal[5][11]. In contrast to this, Spigelian hernia is located in the Spigelian fascia, between the lateral border of rectus abdominis muscle medially and semilunar line laterally. The spigelian hernia belt is a transverse band lying 0 to 6 cm cranial to the line joining both anterior superior iliac spines [11]. This is where majority of spigelian hernia are located. However, low spigelian hernia may mimic inguinal hernias. The main point that differentiates spigelian hernia from inguinal hernia is the direction of herniation. Spigelian hernia is laterally and upwards while inguinal hernia is directed downwards, towards the scrotum. [11]

As adults contain an abundance of abdominal fat, the diagnostic challenges increase. In comparison, the scarcity of fat in children enables a quicker diagnosis [12]. Only about 50 % of Spigelian hernias are diagnosed preoperatively. The recommended first line of investigation for spigelian hernia is Ultrasound followed by CT scan [13]. Ultrasound is a highly operator-dependent investigation, but is still considered the first method of imaging due to its accessibility and repeatability. In case of impalpable testes, a definitive diagnosis is obtained via surgical exploration. The sensitivity of ultrasound in localizing the location of testes in case of non-palpable testes is reported to be 45 % and specificity as 78 % [14].

Management of this condition involves repair of the hernia along with either orchiopexy or orchiectomy, depending upon the risk factors involved. Contrary to cryptorchidism in children, orchiectomy has been mainly recommended for cryptorchidism in adults, because of the increased probability of neoplasms [15]. Orchiopexy after puberty does not protect against subsequent malignancy and the risk of death owing to malignancy remains greater than the surgical risk of orchiectomy for abdominal cryptorchids at all ages [18]. In our particular case, the surgical team went ahead with the decision of orchiectomy based on our clinical judgment. Given the history of absence of descent and risk for malignancy after due consent was taken.

CONCLUSION:

In conclusion, the pathogenesis of Spigelian-cryptorchidism syndrome remains elusive. The accurate diagnosis of this syndrome depends both on clinical examination as well as radiological investigation. Sometimes radiological investigations are not enough for diagnosis, though we can estimate the size and location of defect accurately but cryptorchidism can be missed in radiological investigation (Computed tomography). In such cases, confirmation can be done on Magnetic resonance imaging or surgical intervention. The treatment is surgical and varies according to the age of the patient, clinical setup and the expertise of the surgeon. The decision of orchidectomy is depend on surgeon, and the age of patient. If this condition occurs in early age up to 18 months, then orchidopexy can be done as fertility is preserved [17] otherwise orchidectomy is done as a precautionary measure in light of potential malignancy.

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