Syndrome of Spigelian Hernia and Cryptorchidism: New Evidence Pertinent to Pathogenic Hypothesis

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INTRODUCTION

Lateral ventral hernia (Spigelian hernia) is a rare surgical condition in children. The cases are commonly seen in adult population. It arises because of weakness in fascia. Preperitoneal fatty tissue, intraabdominal organs may herniate through hernial sac. Strangulation or incarceration may rarely occur. Frequency of undescended testis was found much higher in cases with Spigelian hernia than normal population.[1-3] A one-month old boy who had testis in Spigelian hernial sac was found to have gubernaculum and inguinal canal opposing previously described "Spigelian-cryptorchidism syndrome". The aim of this study is to describe details of this case.

CASE REPORT

One month-old boy had left inguinal-lower quadrant swelling and left nonpalpable testis. Physical examination revealed Spigelian hernia and left nonpalpable testis (**Figure 1**). Diagnostic laparoscopy which was performed through umbilicus revealed Spigelian hernia with left testis inside hernial sac and open bilateral internal rings (**Figure 2**). After termination of laparoscopy, left oblique incision was used and hernial sac was found (**Figure 3**). Testis was liberalized after gubernaculum was cut, upon protecting spermatic cord and vessels. Spermatic cord had sufficient length to bring the testis into the scrotum through the inguinal canal. Hernial sac was excised and defect was repaired. Left testis was descended through inguinal canal and positioned into supra dartos pouch. Contralateral inguinal hernia was repaired. There were no complications.

DISCUSSION

Spigelian hernia is a ventral interstitial hernia which occurs as a result of weakness of aponeurosis between semilunar line and lateral edge of rectus abdominis muscle.[1-6] External oblique abdominis muscle and its aponeurosis are usually intact whereas transversalis fascia and internal oblique abdominis muscle are weak.
Spigelian hernia is rare in children and 40 cases of childhood Spigelian hernia have been reported in the literature.[1,3-5] Co-occurrence of Spigelian hernia and cryptorchidism has been reported only in thirty cases in the literature. There are some theories on coexistence of Spigelian hernia and cryptorchidism. Discussions are on whether undescended testes contribute to the development of congenital Spigelian hernia or weakness in abdominal wall somehow

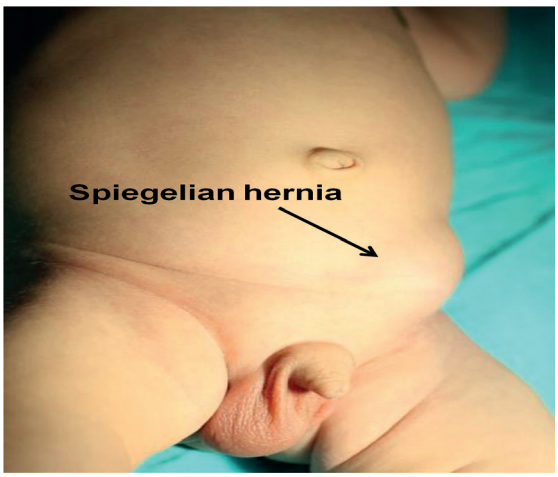

Figure 1. Physical examination revealed Spigelian hernia and left nonpalpable testis.

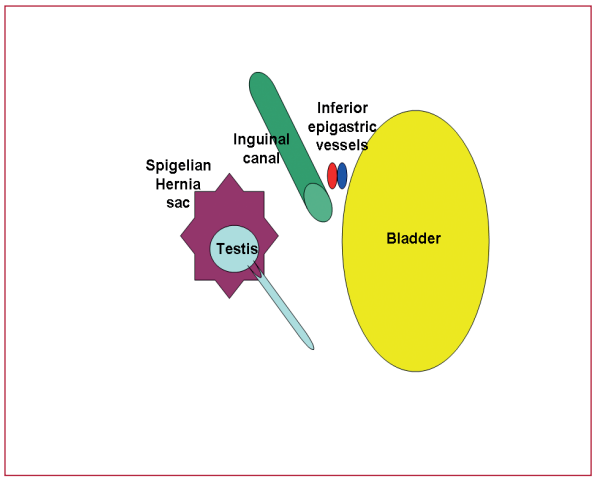

Figure 2. A schema of the laparoscopic view.

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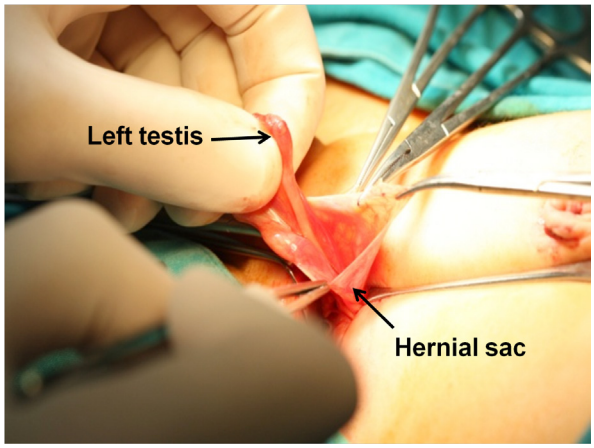

Figure 3. Left testis inside the Spigelian hernial sac (informed consent was taken from the parent for the photographs).

interrupts normal testicular descent and causes testis to settle down in hernial sac.[2,3] According to generally held view, Spigelian aponeurosis is weak point and undescended testis takes path of least resistance, coming to rest in hernial sac and accounting for coexistence.[1,3,7] Another view describes ectopic descent of testis as main problem. While descending to ectopic site, analogous to its descent into scrotum, testis may stimulate formation of processus vaginalis and this peritoneal processus accompanying ectopic testis forms potential sac.[5] According to Rushfeldt and colleagues, Spigelian hernia-cryptorchidism syndrome arises from inability of testis to descend into scrotum because of failure in development of gubernaculum and inguinal canal. Spigelian hernia-cryptorchidism consists of 4 congenital ipsilateral elements: defect in the Spigelian fascia, hernial sac containing testis, absence of gubernaculum and absence of inguinal canal.[3] Existence of gubernaculum and inguinal canal in this presented case has failed to support Rushfeldt and colleagues hypothesis.

Although infantile Spigelian hernia are very rare, they are indeed congenital in nature and there is cause and effect relationship with undescended testis, although this relationship is not yet elucidated.[2]

Physical examination is main diagnostic factor in Spigelian hernia however the intact overlying muscle and aponeurosis can lead to diagnostic challenges. In these cases where hernia is suspected but cannot be demonstrated, ultrasonography can be used to show hernial sac. Once diagnosed, Spigelian hernia should be repaired because of high rate of incarceration.[2,7,8] These defects are repaired primarily, however in unusually large defects, mesh may be required for the repair. Spigelian hernia is rare condition in childhood. The incidence of cryptorchidism is much higher in cases with Spigelian hernia with respect to normal population and the testis is frequently found in the hernial sac. Repair of the hernia and orchiopexy can be easily performed due to the sufficient length of testicular vessels.

CONFLICT OF INTEREST

None declared.

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