



An Unique Case of Spigelian Hernia with the Complicated Appendicitis and Ipsilateral Undescended Testis in an Infant

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Introduction

Lateral ventral hernia, which is also called spigelian hernia, is a rare abdominal wall defect seen in children. This hernia is caused by the herniation of preperitoneal fatty tissue and internal organs through the weakness of the aponeurosis which is between semilunar line and lateral edge of rectus muscle [1,2].

The stomach, urinary bladder, colon, appendix and other intrabdominal organs can be seen in hernia sac. In the literature, a small number of case reports has been described for associa-

tion of spigelian hernia and ipsilateral undescended testis; however, its etiology has not been fully elucidated [3]. The incidence of appendix in the spigelian hernia sac is very rare. Appendix in spigelian sac has been shown in total of 20 adult cases in the literature to date [4-21].

We aimed to present spigelian hernia of a 38-day-old male who had ipsilateral undescended testis and incarcerated gangrenous appendicitis in the sac as the first case in the literature in this study.



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Case presentation

A 38-day-old boy is being evaluated with the complaint of swelling in the right lower lateral region of the abdomen and vomiting. Findings of the physical examination of patient were right spigelian hernia, an empty right hemiscrotum due to an undescended right testis and tenderness in the localization of the spigelian hernia. In addition, atypical gas distribution and partial obstruction appearance were detected in the abdominal X-ray of the patient. (Figure1).

Abdominal ultrasound revealed ‘herniation of an intestinal loop with a wall thickness of 3.5 mm and testicular tissue measuring 11x9 mm from an approximately 8 mm defect in the right lower quadrant’. The patient was operated urgently (Figure 2). A right paramedian incision was made and it was seen that the right testis and associated structures, as well as the cecum and appendix were located in the hernia sac. Testicular appearance was normal, but spermatic cord and epididymis were edematous. The cecum appeared normal, but the appendix was gangrenous. There was no inguinal canal formation on the right. Appendectomy was performed. Orchiopexy was performed by releasing the right testis and associated structures in the retroperitoneum and descending them from the right inguinal region into the dartos pouch formed in the right hemiscrotum. Lateral ventral hernia repair was performed (Figure 3-7). No complications were observed. Patient was discharged on the post-operative third day.

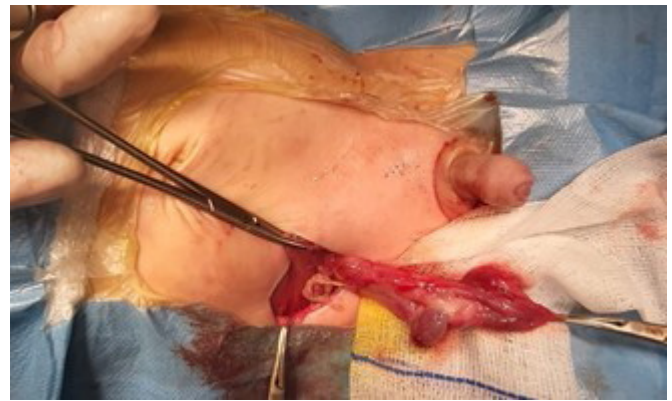


Figure 3: Right testis, spermatic cord and its components are shown in lateral abdominal defect.

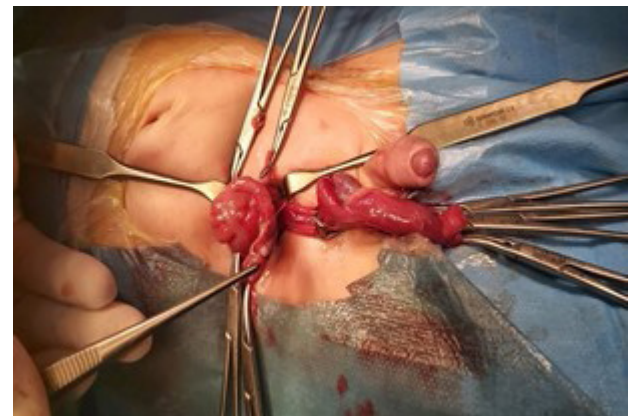


Figure 4: Undescended testis and gangrenous appendicitis are in spigelian hernia sac.



Figure 1: Standing abdominal X-ray of patient, no gas in pelvis and intestinal obstruction.



Figure 5: Performing of appendectomy.



Figure 2: Exploration of defect of spigelian hernia by right paramedian incision.



Figure 6: Repair of defect.

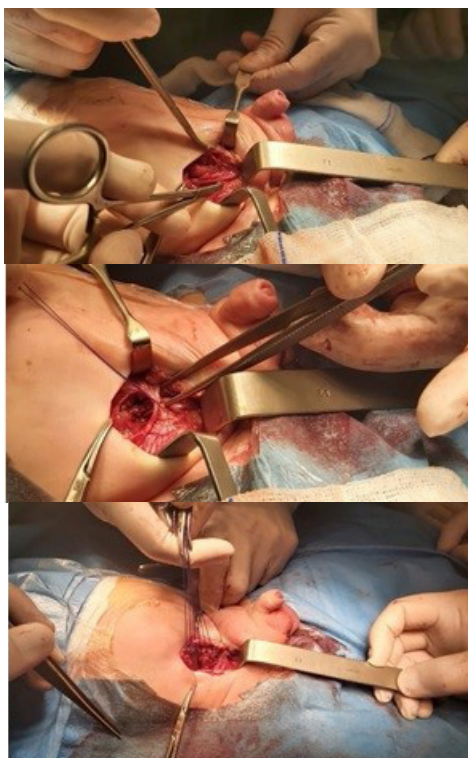


Figure 7: Neoformation of right inguinal canal and orchiopexy.

Discussion

The Flemish anatomist Adriaan van der Spiegel (1578-1625) was the first to describe the semilunar line in the anatomy of the abdomen, and in 1764 Klinkosch described a hernia in the semilunar line and he defined it as a spigelian hernia [22]. In 1895, Schoofs reported the first case of pediatric spigelian hernia by presenting a case of spigelian hernia and ipsilateral undescended testis [22]. In addition, Scopinaro reported a six-month-old pediatric patient who died due to an incarcerated spigelian hernia in 1935 [3].

Spigelian hernia (lateral ventral hernia) is defined as a hernia that develops from a defect resulting from weak aponeurosis of the transverse abdominus which is between the semilunar line and the lateral edge of the rectus muscle [23]. The semilunar line starts from the ninth costal cartilage and extends to the pubic tubercle. The defect is usually not very large, most of them are less than two cm in diameter, therefore, the risk of strangulation is considered to be high. Approximately 8-25% of Spigelian hernias require immediate surgical intervention [23-25].

Spigelian hernia is more common in adults rather than children, accounting for 1-2% of all abdominal hernias. It has been shown to be associated with trauma, previous surgeries, and obesity. Spigelian hernia, which is very rare in children, is mostly congenital. It has been defined in 78 pediatric patients in the English literature search, between 1900 and 2014. 71 of these patients were reported as non-traumatic, 53% of patients had ectopic testis in the hernia sac, and 69% of cases with ectopic testis were found to have no guberneculum and/or inguinal canal [26]. In our case, there was an ectopic testis located in the spigelian sac and there was no inguinal canal on the ipsilateral side. The high prevalence of Spigelian hernia with coexisting undescended testis makes this entity a matter of debate. According to embryology of the testis descending, the fact that abdominal stage occurs due to intraabdominal pressure, spigelian hernia causes undescended testis. It is claimed that the testis may have remained in this region due to low resistance in the hernia localization during the abdominal descent phase

[27]. Another view is that the spigelian hernia defect occurs due to undescended testis. It is argued that the testis remaining in the abdomen and absence of inguinal canal and gubernaculum structures may cause a defect in the abdominal wall, and that the coexistence of spigelian hernia and undescended testis occurs for this reason [28].

In pediatric patients testis, preperitoneal adipose tissue, small intestine, colon, omentum are frequently found in spigelian hernia sac; less frequently, Meckel's diverticulum, gallbladder, stomach, bladder, and ovaries have been reported. All of the cases in which appendix was found in the spigelian hernia sac in the literature were adult cases. In these patients, laparotomy or laparoscopic exploration, hernia repair mostly by primary or rarely patching, and appendectomy if inflamed - strangulated are recommended [4-16,29].

Pediatric spigelian hernia, ectopic testis, absence of guberneculum and absence of inguinal canal is a defined entity even though the developmental theories are still controversial. There is no reported case of incarcerated appendicitis in spigelian hernia in children. An infant patient with ipsilateral undescended testis and gangrenous appendicitis in a spigelian hernia is presented to contribute to the literature, which has no analogues in literature review

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