Congenital Spigelian hernia associated with undescended testis

Mustafa Inan, Umit Nusret Basaran, Burhan Aksu, Zafer Dortdogan, Murat Dereli
Edirne, Turkey

Background: **A Spigelian hernia (SH) is a ventral interstitial hernia through a defect in the Spigelian fascia; an undescended testis is sometimes associated with this clinical entity in male newborns. The etiopathogenesis, surgical anatomy, diagnostic methods, and treatment for this rare condition are discussed with a review of the literature.**

Methods: **A 20-day-old newborn was admitted to our hospital for a swelling in the right lower abdomen and undescended testis. Physical examination of the abdomen and scrotum revealed a congenital SH associated with cryptorchidism.**

Results: **Herniotomy, herniorrhaphy, and orchidopexy were performed. In the post-operative period, scrotal abscess occurred and was drained. After drainage, the fixed testis was found to be atrophic.**

Conclusions: **This association may be a distinct clinical syndrome. The operation time in cases of neonatal SH with undescended testis should be well planned because of probable surgical complications such as vascular damage, tension, or compression.**

World J Pediatr 2012;8(2):185-187

Key words: **congenital Spigelian hernia; testicular atrophy; undescended testis**

Author Affiliations: Department of Pediatric Surgery, Trakya University Faculty of Medicine, Edirne, Turkey (Inan M, Basaran UN, Aksu B, Dortdogan Z, Dereli M)

Corresponding Author: Mustafa Inan, MD, Department of Pediatric Surgery, Trakya University Faculty of Medicine, 22030, Edirne, Turkey (Tel: +90 284 2357641; Fax: +90 284 2357652; Email: mustafainan@trakya.edu.tr)

doi: 10.1007/s12519-011-0313-5

Introduction

Spigelian hernia (SH) is a rare condition firstly described in 1764, whose typical patient profile is of an overweight 50-year-old with associated respiratory disease.[1] By contrast, its occurrence as a congenital hernia was firstly described in 1935.[2] Since then, 54 cases of SH have been reported in infants or children.[1,3-6] However, SH associated with undescended testis cases are extremely rare in the literature. We present a case with a review of the literature concerning the pathogenesis, diagnostic methods, and treatment of SH.

Case report

A boy born with a non-palpable right testicle was observed by his parents to have an intermittent swelling in the right lower abdomen. An abdominal wall swelling was especially noticeable when crying. The boy was born at full-term after an uneventful pregnancy, weighing 2900 g with a length of 48 cm and a head circumference of 36 cm. The boy was aged 26 days at diagnosis. Upon examination, the left testis was found to be normal in size with hydrocele and situated within the scrotum. The right testis was not palpable. When straining, a reducible hernia containing a 3 cm mass was found at the lateral border of the right rectus abdominis muscle about 3 cm above the inguinal canal (Fig. A and B). Ultrasonography of the swelling revealed a fascial plane defect through the linea semilunaris with herniation of bowel loops between the internal and external oblique muscles, consistent with a SH. Moreover, ultrasonography of the inguinoscrotal region revealed that there was no testis in the right scrotum, but a normal inguinal canal and spermatic cord images were not observed. A presumptive diagnosis of SH was made. During the operation, an incision was made over the swelling and the external oblique fascia was found intact. The internal oblique fascia also appeared to be superficial to the defect, although thinning of the layers made identification difficult. Below these, a 2-cm defect in the transversalis fascia was found 2 cm superior to the inferior epigastric

Case report

artery and vein. The right testis was found within the hernial sac (Fig. C and D). The spermatic cord and vasa were found adherent to the posterior wall of the sac. Once dissected, the hernia was ligated. Thereafter, an inguinal skin crease incision was made and exploration of the inguinal region yielded no evidence of the inguinal canal and gubernaculum. A new internal ring was made through the abdominal wall medial to the inferior epigastric vessels and just lateral to the pubic tubercle. The abdominal wall was reconstructed using absorbable sutures. The testis was then passed down into the scrotum, where it was anchored in a subdartos pouch. The boy's progress was complicated by a scrotal abscess eight days postoperatively. After drainage, the testis was found to be atrophic.

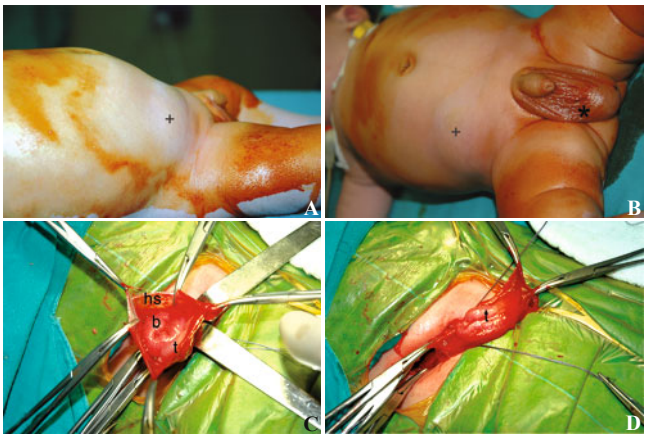

Fig. A, B: a Spigelian hernia in the right lower abdomen and undescended testis in the right side (+: Spigelian hernia; *: undescended testis); **C, D:** During the operation, the testis was found in the hernial sac (hs: hernial sac; b: bowel; t: testis).

Discussion

SH is a ventral interstitial hernia occurring through a defect in the transversus abdominis aponeurosis (Spigelian fascia) between the lateral border of the rectus abdominis muscle and the semilunar line, which extends from the ninth costal cartilage to the pubic tubercle. Clinically, SH occurs in the fascia below the level of the umbilicus, lateral to the junction of the semilunar and arcuate lines.[7] It lies mostly in the "SH belt" and is characterized by the retroperitoneal fat, the peritoneal sac, and organs through a congenital or acquired defect in the Spigelian aponeurosis.[8] In other words, Spigelian line is where the transversus abdominis muscle ends in an aponeurosis. SH does not arise on this line, but the Spigelian fascia is also involved.

Our case resembles all those reported previously about the location of the cryptorchid testes,[1-3,5,7,9-14] which are often found within the defect or situated within a hernial sac just beneath the defect itself (Table). We support the idea that SH occurs secondary to an ectopically descended testis, as reported by Raveenthiran.[6] In the case described here, the testis was placed with difficulty into the subdartos pouch because of relatively short testicular vessels, which resulted in moderate tension at the end of

Table. Reported cases of Spigelian hernia associated with undescended testis

Authors	Year	Age	Side	Comments and associated pathologies
Gravier et al[9]	1978	6 mon	Right	-
		9 mon	Left	Right inguinal hernia
Pul and Pul[10]	1994	18 mon	Right	-
Silberstein et al[11]	1996	10 wk	Left	-
		4.5 mon	Right	-
Ostile and Zerella[2]	1998	5 mon	Right	Identified in newborn period
Al-Salem[7]	2000	3 mon	Left	-
		1 wk	Left	Micrognathia, cleft palate, malformed ears, right clubfoot, deformed left lower limb, exitus before the surgery
Levy et al[12]	2003	1 mon	Right	Bilateral undescended testis, bilateral Spigelian hernia and right strangulated Spigeian hernia
		3 mon	Left	Identified at 5 weeks of age
Raveenthiran[13]	2005	13 mon	Right	Imperforate anus, left inguinal hernia, umbilical hernia
Torres de Aguirre et al[5]	2005	26 d	Right	Strangulated
		40 d	Bilateral	-
O'Sullivan et al[1]	2006	4 mon	Left	Hypospadias
Durham and Ricketts[3]	2006	8 mon	Left	Scrotal abscess after orchidopexy
		13 mon	Bilateral	A mesh used to reconstruct the right sided defect
		14 mon	Right	Sibling of the next patient
		2 mon	Right	Bilateral undescended testis
Fascetti-Leon et al[14]	2009	Newborn	Bilateral	Scalp aplasia cutis
Present case	2009	20 d	Right	Scrotal abscess and testicular atrophy after orchidopexy

the orchidopexy procedure. Moreover, no inguinal canals could be identified in those cases of intra-abdominal testes. The testes could be brought down transabdominally and a new ring, where an external ring would normally be located, was created medial to the inferior epigastric vessels and just lateral to the pubic tubercle. This technique was suggested by Durham and Ricketts.[3] In our case, the progress was complicated by serious scrotal infection and the testis was atrophic. We think that the testicular atrophy may be due to the following four leading points: vascular damage during surgical dissection, vascular compression in the new ring, vascular tension, or scrotal infection.

Considering probable surgical complications such as vascular damage, tension, compression, and infection, we suggest the timing of operation in cases of congenital SH with undescended testis be well planned. Further anatomic development of the infant may prevent surgical complications. For instance, Ostlie and Zerella[2] reported that surgery for a newborn who had SH associated with undescended testis was only undertaken when the infant was 5 months old, similar to the case described by Levy et al.[12]

To our knowledge, this is the first case report discussing the management and timing of surgical treatment for undescended testis associated with congenital SH in light of experience in dealing with testicular atrophy. The following alternatives for treating this entity may be speculated to avoid testicular damage: surgical repair of congenital SH may be performed in the early period, and orchidopexy could be postponed to 1 year of age. In the meantime, the risk of incarceration is decreased to a limited degree. The testis and spermatic cord are enlarged, and the infant's immune response is getting stronger. After a year, the abdominal testis may be anchored by the Fowler-Stephens procedure or another surgical technique. The other speculated therapy is to wait until the end of the first year of life before performing both orchidopexy and surgical repair of congenital SH and conducting a strict follow-up because of the extremely rare risk of incarceration.

We believe infantile SH is indeed congenital in nature and that the presence of congenital SH predisposes the patient to the development of ipsilateral undescended testis. This association may suggest a new syndrome. A higher index of suspicion and preoperative ultrasonographic evaluation may improve the diagnostic accuracy. Once diagnosed, SH should be repaired expeditiously or followed up carefully because of possible incarceration. Finally, it is important to remember that the surgical treatment of SH associated with undescended testis in a newborn carries serious risks.

Funding: None.
Ethical approval: Not needed.
Competing interest: None declared.
Contributors: Inan M proposed the study and wrote the first draft. All authors contributed to the design and interpretation of the study.

References

1 O'Sullivan O, Bannon C, Clyne O, Flood H. Hypospadias associated undescended testis in a Spigelian hernia. Ir J Med Sci 2006;175:77-78.
2 Ostlie DJ, Zerella JT. Undescended testicle associated with spigelian hernia. J Pediatr Surg 1998;33:1426-1428.
3 Durham MM, Ricketts RR. Congenital spigelian hernias and cryptorchidism. J Pediatr Surg 2006;41:1814-1817.
4 Vaos G, Gardikis S, Zavras N. Strangulated low Spigelian hernia in children: report of two cases. Pediatr Surg Int 2005;21:736-738.
5 Torres de Aguirre A, Cabello Laureano R, García Valles C, Garrido Morales M, García Merino F, Martínez Caro A. Spigelian hernia: two cases associated to cryptorchidism. Cir Pediatr 2005;18:99-100.
6 Raveenthiran V. Congenital spigelian hernia and cryptorchidism: a distinct clinical syndrome. J Pediatr Surg 2007;42:1794-1795.
7 Al-Salem AH. Congenital spigelian hernia and cryptorchidism: cause or coincidence? Pediatr Surg Int 2000;16:433-436.
8 Aksu B, Temizöz O, Inan M, Gençhellaç H, Başaran UN. Bilateral spigelian hernia concomitant with multiple skeletal anomalies and fibular aplasia in a child. Eur J Pediatr Surg 2008;18:205-208.
9 Graivier L, Bernstein D, RuBane CF. Lateral ventral (spigelian) hernias in infants and children. Surgery 1978;83:288-290.
10 Pul N, Pul M. Spigelian hernia in children—report of two cases and review of the literature. Yonsei Med J 1994;35:101-104.
11 Silberstein PA, Kern IB, Shi EC. Congenital spigelian hernia with cryptorchidism. J Pediatr Surg 1996;31:1208-1210.
12 Levy G, Nagar H, Blachar A, Ben-Sira L, Kessler A. Pre-operative sonographic diagnosis of incarcerated neonatal Spigelian hernia containing the testis. Pediatr Radiol 2003;33:407-409.
13 Raveenthiran V. Congenital Spigelian hernia with cryptorchidism: probably a new syndrome. Hernia 2005;9:378-380.
14 Fascetti-Leon F, Gobbi D, Gamba P, Cecchetto G. Neonatal bilateral spigelian hernia associated with undescended testes and scalp aplasia cutis. Eur J Pediatr Surg 2010;20:123-125.

Received December 29, 2009
Accepted after revision February 10, 2010