50 Case Report

# **Splenogonadal Fusion Discovered by Testicular Torsion**

Nahla Hmidi<sup>1\*</sup>, Dorsaf Makhlouf<sup>1</sup>, Rachida Laamiri<sup>1</sup>, Nahla Kechiche<sup>1</sup>, Badii Hmida<sup>2</sup>, Amine Ksia<sup>1</sup>, Laassad Sahnoun<sup>1</sup>, Mongi Mekki<sup>1</sup>, Mohsen Belghith<sup>1</sup>, Adellatif Nouri<sup>1</sup>

<sup>1</sup>Department of pediatric surgery Fattouma Bourguiba University Hospital, Monastir, Tunisia

\*Address for Corresponder: Dr. Nahla Hmidi, Department of pediatric surgery Fattouma Bourguiba University Hospital, Monastir, Tunisia (email: nh.nahlahamdi@gmail.com)

#### How to cite this article:

Hmidi N, Makhlouf D, Laamiri R, Kechiche N, Hmida B, Ksia A, Sahnoun L, Mekki M, Belghith M, Nouri, A. Splenogonadal fusion discovered by testicular torsion. Iranian Journal of Pediatric Surgery 2020; 6(1): 50-53.

DOI: https://doi.org/10.22037/irjps.v6i1. 31705

## **Abstract**

## **Keywords**

- Splenogonadal Fusion
- Testis
- Torsion

Splenogonadal fusion, which is adherence of splenic tissue to gonads, is an uncommon congenital anomaly which mainly affects males. Herein, we report a case of splenogonadal fusion in a 20-month-old boy presenting with acute scrotal pain and inflammation. With the suspicion of left testicular torsion, an emergent left scrotal exploration was carried out. It revealed a necrotic left testicle along with a 360° rotation of the spermatic cord and three accessory structures in the lower pole of the testicle. Histology showed the presence of a splenic tissue. Splenogonadal fusion can present as an acute condition mimicking a testicular torsion. But, one should always bear in mind the possibility of this association. Splenogonadal fusion should be included in differential diagnosis of testicular mass to avoid unnecessary orchidectomy.

#### Introduction

Splenogonadal fusion, which is adherence of splenic tissue to gonads, is an uncommon congenital anomaly. <sup>1, 2</sup> This anomaly is further categorized based on splenic attachments in to continuous

or discontinuous; and can interfere with normal gonadal descent and closure of the processus vaginalis.<sup>3</sup> As a result, it is often associated with undescended testis and inguinal hernia.<sup>3</sup> Yet in literature there are very few cases reported of this condition presenting as testicular torsion.

received: 12 February 2019 accepted: 11 August 2019 Published online: August 2020 This open-access article is distributed under the terms of the Creative Commons Attribution Non Commercial 3.0 License (CC BY-NC 3.0). Downloaded from: http://journals.sbmu.ac.ir/irjps

<sup>&</sup>lt;sup>2</sup>Department of Radiology, Fattouma Bourguiba University Hospital, Monastir, Tunisia

Splenogonadal Fusion Hmidi et al 51

## Case report

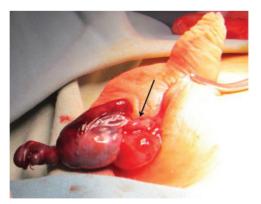
A 20-month-old boy was referred to us with a diagnosis of acute scrotum. There was no history of trauma. Physical examination revealed a sudden and severe pain in the left scrotum. However, the right testicle was palpable high in the inguinal region (undescended testis). Ultrasound of the left

testis demonstrated a loss of blood flow consistent with testicular torsion. It also showed 3 hypoechoic, well-circumscribed lesions. The child was prepared for emergency surgery. It was performed via a scrotal incision that showed findings of a necrotic left testicle with a 360° rotation of the spermatic cord **Figure 1**.



Figure 1: a necrotic left testicle with rotation of spermatic cord

Surgical Exploration showed three accessory structures in the lower pole of the testicle Figure 2.



**Figure 2**: a necrotic left testicle with spleen tissue in the lower pole

Detorsion of testis was performed from the medial to the lateral side and a left orchidopexy was also performed. The accessory structures were removed and were sent for histological examination which revealed the presence of accessory mature splenic tissue. The whole procedure was then completed by right orchidopexy. The patient was discharged on the second postoperative day.

#### Discussion

Splenogonadal fusion is an uncommon congenital anomaly with less than 200 cases reported in the literature. 1,2 Its first description was by the German pathologist Bostroem in 1883. 1, 2, 3 It is defined as an anomalous attachment between splenic tissue and the gonads or mesonephric derivatives. Around 82% of patients are diagnosed when under 30 years of age, but 50% of splenogonadal fusion are reported in children (less than 10 years). It is predominant in males with a ratio of 16:1. 1, 2 In female patients, splenic tissue is attached to the ovary or mesovarium. 4

The pathogenesis for this anomaly is not clear <sup>1,2</sup> but several hypotheses have been proposed. Around the 5th week of embryological development, the dorsal mesogastrium shifts to the left where the splenic anlage is situated beside the left urogenital fold. <sup>1,2,3</sup> Any inflammation of the peritoneal surface of the spleen can result in the attachment of these organs and ultimately the caudal migration. <sup>5</sup> This abnormal attachment seems to be the most accepted theory of splenogonadal fusion and teratogens are attributed. <sup>3,4</sup> So, as in our case it is the left testis which is always affected, (98% of the cases). <sup>1,4</sup> <sup>3,5,6</sup> The right sided cases are very rare and may

exist because of situs inversus.<sup>4</sup> Other explanation could be that an abnormal suspensory ligament of testis is colonized by splenic cells.<sup>4</sup> Other authors suggested a possible familial predisposition of this disorder.<sup>4</sup>

This anomaly usually first apears as an inguinal hernia (probably because the aberrant splenic tissue interferes with the normal gonadal decent and/or the closure of processus vaginalis) or a left scrotal mass (represents 10% of scrotal mass) and can be misdiagnosed as testicular cancer.<sup>1, 2, 4</sup>

The attachment between the gonads and splenic tissue can be continuous or discontinuous.<sup>2</sup> The continuous form occurs in 55% of cases, and there usually is a connection of splenic or fibrous tissue, between the spleen and the gonads. In the discontinuous form (which adds up to 45% of cases), ectopic splenic tissue is attached to the gonad yet no connections exist with the spleen.<sup>4</sup> The spleen tissue has a capsule like the native spleen and gonadal tissue are distinct and spate. <sup>7</sup>

Splenogonadal fusion is mostly an incidental discovery while exploring the groin for cryptorchidism, hernia or rarely torsion testis.<sup>3, 7</sup> Mimicking torsion of testis and presenting as an acute condition, such as our case is rare.<sup>7</sup>

There is considerable controversy regarding the role of imaging in diagnosis of splenogonadal fusion.5 In some cases, Doppler ultrasound of the scrotum has been used to diagnose preoperatively.<sup>3</sup>, Computed tomography, magnetic resonance imaging and technetium-99m spleen scintigraphy can also be used for diagnosis.<sup>5</sup> Perioperative frozen section examination is recommended to

Splenogonadal Fusion Hmidi et al 53

avoid misdiagnosing splenogonadal fusion as a testicular tumor and performing unnecessary orchidectomies.<sup>1</sup>

Treatment of splenogonadal fusion involves complete excision of ectopic spleen with preservation of testis and orchidopexy if testis is undescended.<sup>7</sup> Cases were reported of simultaneous presence of splenogonadal fusion and testicular cancer for which orchidectomy was needed.<sup>2</sup>, In literature, only four such cases have been reported.<sup>2</sup> Splenogonadal fusion does not increase the risk of testicular cancer but since it is usually accompanied by cryptorchidism it can explain the risk of associated testicular malignancy.<sup>3,7</sup>

#### **Conclusion**

Splenogonadal fusion is an uncommon and

benign congenital developmental anomaly that is frequently associated with cryptorchidism and hernia. Pediatric surgeons should be aware of this anomaly as a rare differential diagnosis of testicular mass so they can perform excision of the scrotal spleen and spare the testicular tissue.

### Acknowledgment

Not Applicable

## **Funding/Support**

There are no sources of support.

#### **Conflict of interests**

There are no conflicts of interest.

#### References

- 1. Shakeri A, Shakeri A, Rasolmali R, et al: A case of splenogonadal fusion accompanied by accessory spleen in a 4-year-old boy. Urol Ann 2018;10: 406-8.
- 2. Abokrecha A, Almatrfi A: Discontinued splenogonadal fusion and bilateral empty scrotum in an 18-month-old boy. European J Pediatr Surg Resp 2017;5(1): e1-e3.
- 3. Lopes RI, De Medeiros MT, Arap MA, et al: Splenogonadal fusion and testicular cancer: case report and review of the literature. Einstein (Sao Paulo) 2012;10(1): 92-5.
- 4. Chiaramonte C, Siracusa F, Li Voti G: Splenogonadal fusion: a genetic disorder? Report of a case and review of the literature. Urol Case Rep 2014; 2(2): 67-9.
- 5. Sountoulides P, Neri F, Bellocci R, et al: Splenogonadal fusion mimicking a testis tumor. J Postgrad Med 2014; 60(2): 202-4.
- 6. Kocher NJ, Tomaszewski JJ, Parsons RB, et al: splenogonadal fusion: a rare etiology of solid testicular mass. Urology 2014; 83(1): e1-2.
- 7. Sam Aruputha John, Ahamed Faiz Ali, Sunil Kumar, et al: Splenogonadal fusion presenting as acute torsion of testis. American Journal of Pediatrics 2017; 3(5): 28-31.