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CASE REPORT

Double Torsion of a Continuous Type of a Splenogonadal Fusion Presenting as an Inguinoscrotal Emergency: A Rare Case Report

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Background: Splenogonadal fusion (SGF) is a rare congenital anomaly that presents with an abnormal connection between the gonad and the spleen. Misdiagnosis can lead to unnecessary surgical interventions, underscoring the need for thorough pre-operative assessments and differential diagnoses. Histopathological and imaging evaluations are essential for accurate diagnosis and appropriate management.

Case Presentation: A 35-year-old male presented with an acute swelling and pain in the scrotum and the inguinal region. The patient's medical and surgical history was unremarkable, with no known congenital anomalies. Emergency surgery revealed a fibrous cord extending between the orthotopic spleen and the left testis with multiple tissue formations along it in a rosary-like shape. The reason for the acute presentation was double torsion of the abnormal cord at the internal inguinal ring and near the junction between the fibrous cord and the testis. The abnormal tissue was resected and histopathological examination confirmed splenic tissue. confirming the final diagnosis of splenogonadal fusion (continuous form).

Conclusion: SGF is a rare congenital anomaly which is difficult to diagnose that is difficult to diagnose due to its nonspecific clinical presentation. Most SGF cases are diagnosed during the exploratory surgery, no single definitive diagnostic modality exists; however, imaging and splenic scintigraphy may aid in selected cases. Surgeons should consider this benign condition in the differential diagnoses of left inguinal or scrotal masses to avoid unnecessary orchiectomy.

Keywords: splenogonadal fusion, testicular torsion, inguinal mass, congenital anomaly

Background

Splenogonadal fusion (SGF) is an uncommon congenital anomaly that results from an aberrant connection between the gonad and the spleen.[1] It is believed to result from abnormal adhesion between splenic primordium and the gonadal ridge during early embryogenesis, particularly between the 5th and 8th weeks of gestation.[2,3] This anomaly is classified into two types: continuous and discontinuous. In the continuous type, a cord-like structure connects the orthotopic spleen to the gonad, which may contain splenic or fibrous tissue. In contrast, the discontinuous type lacks any direct connection with the spleen and presents as ectopic splenic tissue attached to the gonad, often resembling an accessory spleen.[2–5] SGF shows a marked male predominance and most commonly affects children and adolescents, with most cases identified before 20 years of age; thus, occurrence in adulthood is unusual and may pose additional diagnostic challenges.[2,3,6] This malformation can be misdiagnosed as inguinal hernia, cryptorchidism, epididymo-orchitis, and testicular malignancy. Notably, cryptorchidism and inguinal hernia represent the most commonly reported associated conditions.[2,3,6] SGF, particularly the continuous type, may be associated with other congenital anomalies.[2–4] Even though SGF is a benign condition, many patients undergo orchiectomy and subsequent histopathological examination to establish an accurate diagnosis. Thus, failing to conduct adequate preoperative evaluation and disregarding such differential diagnosis may result in an unnecessary surgical intervention.[6] Imaging modalities, including ultrasound, CT, MRI, and scintigraphy, assist in the preoperative evaluation of suspected SGF; however, findings are often nonspecific, particularly in acute settings, and definitive diagnosis may still require histopathological confirmation.[2,3,7,8]

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This case reports a continuous type of splenogonadal fusion that was complicated with double torsions in two different locations leading to acute presentation.

Case Presentation

A 35-year-old male presented to the emergency complaining of swelling and pain in the scrotum and the left inguinal region. The onset of symptoms occurred 24 hours prior and has gradually worsened. The patient has no relevant medical or surgical history. On clinical examination, the left testis was palpable and severely tender. The abdomen was soft with mild tenderness; there were no clinical signs of guarding or rigidity. Ultrasonography imaging showed normal testicular and epididymal size and perfusion. However, a septate tubular structure with turbid fluid inside was seen extending superiorly from the testis towards the groin. Based on these findings, a strangulated inguinal hernia, specifically a necrotic omentocele, was the primary preoperative diagnosis, and surgeons decided to perform an emergency exploration. A left inguinal incision was made to access the inguinal canal, subsequently isolating and opening the hernia sac. They discovered an elongated burgundy tissue that extends to the scrotum. The tissue was connected to a fibrous cord that reached the upper pole of the testis without any connection to the epididymis or the spermatic cord. Another fibrous cord extended from the tissue up to the level of the internal inguinal ring Palpation along the fibrous cords revealed multiple similar burgundy formations arranged in a rosary-like shape (Figure 1). The acute clinical presentation was due to double torsion in the abnormal tissue at distinct sites: the first one at the entrance of the internal inguinal ring, and the second near the junction between the fibrous cord and the testis (Figure 2). The abnormal tissue was

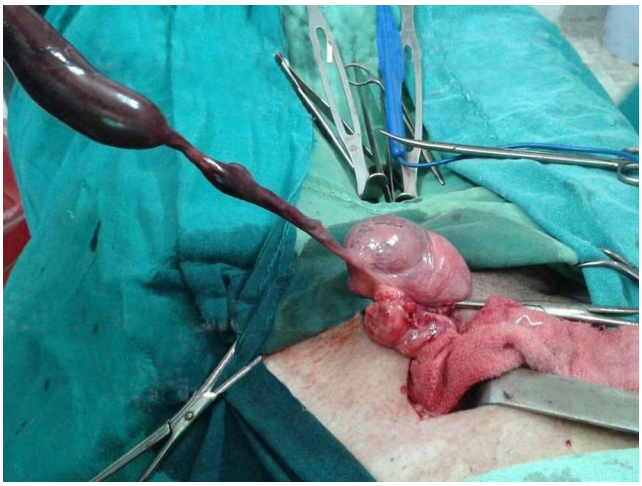

Figure 1 Detorsed fibrous cord with islets of splenic tissue connecting to the left testis.

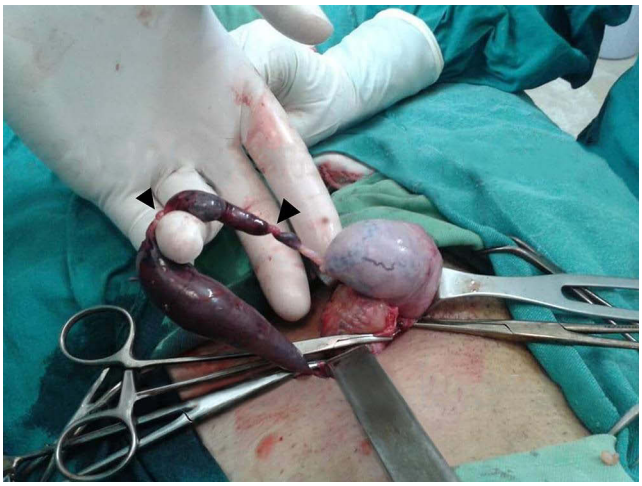

Figure 2 The torsion sites of the splenofibrous cord (arrowheads).

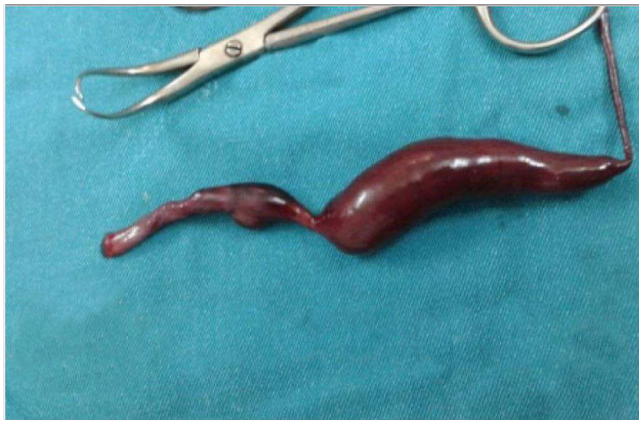

Figure 3 The resected fibrous cord with splenic tissue.

excised from the testis to the internal inguinal ring. Given the absence of intra-abdominal findings and the clear localization of torsions within the inguinoscrotal segment, laparotomy was not indicated. The proximal fibrous–splenic cord toward the orthotopic spleen was therefore preserved to minimize unnecessary morbidity. The resected tissue was submitted for histopathological analysis, which confirmed ectopic splenic tissue (Figure 3). This confirmed the final diagnosis of splenogonadal fusion (continuous form).

Discussion and Conclusions

Splenogonadal fusion (SGF) is a rare congenital malformation defined by an abnormal connection of ectopic spleen tissue between the orthotopic spleen and the gonad during gestation.[2,4,5] It was described for the first time by Bostroem in 1883,[2,7,9] and there have been only about 200 cases reported globally since this malformation was first described.[9]

SGF is classified into continuous and discontinuous types. The continuous type is characterized by the presence of a cord-like structure consisting of fibrous or ectopic splenic tissue connecting between orthotopic spleen and a gonad.[3–6] The connecting cord could be entirely fibrous, entirely consisted of splenic tissue, or fibrous with islets of ectopic spleens,[4] the present case is an example of the continuous type with the last-mentioned type of connecting cords. In most cases this cord extends from the upper pole of spleen to the upper pole of the gonad.[4] On the other hand, in the discontinuous type, the ectopic splenic tissue is directly fused to the gonad but there is no connection to the native spleen.[2,4,5] Previous studies indicate that the frequency of discontinuous type is lower than that of continuous one, but other studies suggest that the frequency of the two types is equal.[4,9]

Interestingly, more than 70% of the patients are younger than 20 years old,[5,9] and half of the cases are less than 10 years old.[4,9] In our case, the age of the patient with this anomaly was 35 years old, whereas 82% of the cases are reported in men under the age of 30.[4] SGF occurs more frequently in males than females with a reported male-to-female ratio of 16.6:1;[7,9] with only 8 cases described in females.[4] Nevertheless, these rates may be undervalued as a result of underdiagnosis in females due to the inaccessibility of the ovaries to clinical examination.[1,4] SGF occurred on the left side in almost all reported cases (98%).[7,9] G. Chen et al reported that the close proximity between the left gonad and the spleen during embryogenesis interprets this fact.[2]

The clinical presentation of SGF is not defined.[4] In females, it is discovered incidentally on laparotomy or at autopsy.[10] While in males, it usually presents as inguinal herniation or cryptorchidism, which are the two most frequently related anomalies, or may present as scrotal mass or swelling especially in the discontinuous form. Subsequently, it could be misdiagnosed as testicular tumor leading to unnecessary, life-altering orchiectomy.[2,4] The other uncommon presentations include inguinal swelling that gets bigger during viral infections and malaria, torsion testis, or bowel obstruction.[10] In this case, the acute presentation—characterized by abrupt swelling and severe pain—mimicked a strangulated inguinal hernia. Importantly, while the inguinal and scrotal regions were exquisitely tender, the abdomen remained soft and non-distended

without signs of guarding or rigidity. To the best of our knowledge, this may represent the first reported presentation of SGF with these acute symptoms due to double torsion of the fibrous cord that connects the orthotopic spleen with the left testis.

SGF is associated with congenital anomalies, the most common of which are limb defects such as ectromelia, peromelia, amelia, phocomelia, and craniofacial abnormalities such as micrognathia.[2,4] The reason behind the commonness of these anomalies is that the occurrence of SGF coincides with the development of the mandible and limb buds.[10] Less common associated anomalies include cleft palate, Moebius syndrome, hypospadias, osteogenesis imperfecta, persistent mullerian duct syndrome, Potter syndrome, gastrointestinal malrotation, transverse testicular ectopia,[2] cardiac anomalies, sexual ambiguities, varicocele, and spina bifida,[4] imperforate anus.[1] There were no congenital abnormalities associated with this reported case.

Preoperative diagnosis remains a significant challenge. As Oshiba et al recently emphasized, splenogonadal fusion remains a diagnostic challenge preoperatively, as imaging findings are often inconclusive and may mimic testicular or inguinoscrotal pathology, with definitive diagnosis frequently established only after surgical exploration and histopathological examination.[8]

In most reported cases, as in our case, SGF was diagnosed during exploration surgery of the scrotum or inguinal area,[2,7] and 17% of the cases were diagnosed at autopsy.[6] Y. Akama et al[5] found no cases of SGF were diagnosed preoperatively, which is challenging because of the rarity of this anomaly. On the other hand, G. Huang et al[9] reported that only a few cases have been diagnosed preoperatively using ultrasound, computed tomography or magnetic resonance imaging. Ultrasound may depict an extratesticular mass with hypoechoic or isoechoic to the neighboring testis.[10] In our case, ultrasound showed a reduced-vascularity tubular formation above the left testis extending toward the left inguinal region, with preserved testicular perfusion; it was thought to be necrotic omentocele. When SGF is suspected, especially in the case of presence of associated congenital malformations, splenic scintigraphy with technetium-99 m (99mTc) is a useable option due to the similarity of radioactive tracer binding in the spleen and testicular mass and this confirms the ectopic splenic origin of mass.[4]

The treatment of choice of SGF is complete excision of splenic tissue from gonadal structures with preservation of testis, orchiectomy is generally not indicated.[3,4,8] Unfortunately, unnecessary orchiectomy was performed in 37% of reported SGF cases because many surgeons did not know about SGF.[10]

SGF is a very rare congenital anomaly which is difficult to diagnose because of its undefined clinical symptoms. There are no specific definitive preoperative methods to detect this disease, but many investigations may be useful such as ultrasound, computed tomography, magnetic resonance and splenic scintigraphy which is advised in suspected SGF patients who present with limb defects or micrognathia. Educating surgeons about this condition and considering it as a potential cause for solid left inguinal or scrotal masses can help avoid misdiagnosing it as testicular cancer, and prevent unnecessary surgical removal of the testis in cases where it is a benign condition.

Abbreviation

SGF, Splenogonadal fusion.

Data Sharing Statement

The imaging results are available from the corresponding author on reasonable request.

Ethics Approval and Consent to Participate

Ethical Committee of the Faculty of Medicine at Damascus University do not require ethics approval for case reports.

Consent for Publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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Disclosure

The authors declare that they have no competing interests in this work.

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