



Splenogonadal Fusion: Accurate Radiological Diagnosis Avoids Maltreatment

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ABSTRACT

Splenogonadal fusion (SGF) is a rare congenital anomaly in which there is abnormal fusion of spleen and the gonad or the remnants of the mesonephros. Diagnosis of this uncommon anomaly is rare and difficult preoperatively. Radiological expertise is necessary for successful surgical exploratory procedure of SGF. In this article, we report an unusual case of splenogonadal fusion that was initially diagnosed as a solid testicular mass by clinical and inguinoscrotal ultrasound examinations. After which patient was undertaken for orchiectomy, however no mass was found, so a biopsy sample taken. Then patient was sent for complete radiological re-evaluation. Now computerized tomography (CT) scan and complete abdominal ultrasound were performed. In ultrasound examination splenogonadal fusion was suspected which was confirmed on CT examination. So before laparotomy is planned patient should undergo complete radiological evaluation so as to avoid unnecessary failed attempts.

KEYWORDS

Cryptorchidism; mesonephros; orchiectomy; splenogonadal fusion.

Introduction

Splenogonadal fusion (SGF) is a rare congenital anomaly of abnormal fusion between the spleen and the gonad or the remnants of the mesonephros. In 'continuous SGF,' there is a cord-like connection between the two organs, whereas in 'discontinuous SGF,' there is fusion of accessory splenic tissue and the gonad without a distinct structural connection to the spleen itself.^[1] The continuous SGF is more common, compared to discontinuous SGF. Here we described a case of the continuous type of splenogonadal fusion.

Case Report

Ten years old boy came to surgical OPD with complain of hard swelling in left scrotal sac since birth. It was clinically diagnosed as left congenital inguinal hernia. On inguinoscrotal ultrasound examination, it was misdiagnosed as left testicular mass. Patient was taken for laparotomy for suspected congenital hernia/ left testicular mass; but no hernia/mass was identified and a biopsy sample was taken for histopathological examination (HPE). In HPE splenic tissue was found. After one month, the patient was re-admitted with complain of increase in the size of swelling in left scrotal sac and mild pain while touching the swelling since fifteen days. This time repeat ultrasound of inguinoscrotal region along with whole abdomen was done in our department. We found a hypoechoic, and homogenous mass was located within testicular parenchyma in the left scrotal sac just under the epididymis. [Figure 1c] Marked hypervascularity, and increased blood flow were noted within the mass. Also a thin and continuous, splenic parenchymal tissue tract extending was seen extending from spleen to left testicle. [Figure 1a and 1b]. No sign of inflammation was reported. No fluid was seen in either scrotal sac. After that CT scan was performed and the similar findings were found. [Figure 2a and 2b] Once again patient was undertaken for exploratory laparotomy and this time surgeons could identify a splenic parenchymal band continuous with left testicular mass was, thus confirming the radiological findings. [Figure 3a and 3b] So, the accurate diagnosis is necessary to avoid the maltreatment and failed surgeries of splenogonadal fusion.

Discussion

The splenogonadal fusion (SGF) was first described by Boström in 1883, and a detailed study by Pommer followed in 1889.^[2] SGF is a rare congenital anomaly that results from an abnormal connection between the spleen and gonad during the embryonic life, when the organs are in close proximity to each other. When gonadal descent begins, the attached splenic

tissue follows the gonadal path.^[3] In a review of 30 cases, Putschar and Manion categorized splenogonadal fusion into continuous and discontinuous types depending on the anatomical continuity between the principal spleen and the gonad. SGF is two type; either continuous, in which the direct anatomical connection between the spleen and the gonad persists, or discontinuous, in which the ectopic splenic tissue is attached to the gonad. The presented case was of continuous type.^[4]

In the literature, there are approximately 175 cases have been reported and most of them are associated with cryptorchidism.^[5] The half of cases reported under the age 10 years and 82% of cases in younger than 30 years.^[2] The our case age was ten years and was not associated with cryptorchidism.

Li Wan-Fuet al.^[6] reported four cases of children with SGF, three of which were treated laparoscopically. Combined with the relevant literature, they discussed the diagnosis and treatment of SGF and the value of exploration by laparoscopy. The diagnosis of SGF prior to surgery is challenging. Their four cases were inaccurately diagnosed prior to surgery. The lack of awareness of SGF is a major factor in its misdiagnosis. Ultrasonography, computed tomography (CT), magnetic resonance imaging (MRI) and ⁹⁹Tc^m spleen scanning are major imaging methods the diagnosis of SGF. However, laparoscopy can achieve improved diagnosis and management of SGF in one goes. In our case, initially, it was misdiagnosed as congenital hernia or left testicular mass clinically as well as on ultrasound examination leading to a failed surgery.

Santosh Kumar et al.^[7] reported a case of continuous type splenogonadal fusion in a young male with primary infertility for 3 years. On evaluation abdomino pelvic ultrasound and MRI evaluation, his right scrotal testis was atrophied and left intra-abdominal undescended testis. On laparoscopic assessment, a mass was seen on the left side due to continuous type of splenogonadal fusion for which excision and left orchidectomy were done. They found SGF is a rare entity and it is commonly mistaken for testicular tumour. It should be considered in the differential diagnosis of testicular masses especially when there are associated congenital anomalies and preoperative laparoscopic assessment, should be done to avoid unnecessary radical surgery.

R. Fernandez Atuan et al.^[8] reported a case of a two year old child with a continuous type splenogonadal fusion presenting

as an incarcerated symptomatic inguinal hernia. According to them the radiologic identification of SGF is important to prevent unnecessary orchiectomy due to its macroscopic resemblance to cancer. Splenogonadal fusion is a rare malformation that is infrequently diagnosed preoperatively. So the possibility of its sudden apparition must be kept in mind in any surgery of the inguinal area.

Conclusion

The diagnosis of splenogonadal fusion is challenging, because this is not a common congenital anomaly. It is misdiagnosed as congenital hernia or left testicular mass, especially in discontinuous type splenogonadal fusion. However, the condition can be easily diagnosed with ultrasonography, CT scan, magnetic resonance imaging (MRI) and ⁹⁹Tc^m spleen scanning when we keep in mind about this rare differential as this may avoid misdiagnosis and maltreatment.

In our case, initially, the case was misdiagnosed as congenital hernia or left testicular mass clinically as well as on ultrasound examination. Due to which patient had to undergo laparotomy, however no mass found and so, just a biopsy sample was taken for histopathological examination. Patient then underwent a radiological re-evaluation which revealed a thin and continuous splenic parenchymal band, continuous with and connecting spleen and left testicle.

Images with legends:



Figure 1a: Ultrasound axial images showing splenic parenchymal beak downward and medially (white open arrows).



Figure 1b: Ultrasound longitudinal images showing continuous splenic parenchymal tissue tract (white open arrows) from spleen to left testicle.



Figure 1c: Ultrasound sagittal image showing splenic parenchymal tissue (white open arrow) and testicular parenchymal tissue (black open arrow) in left scrotal sac.



Figure 2a: CT coronal image showing splenic parenchymal beak (white open arrow) downward and medially.



Figure 2b: CT sagittal image showing continuous splenic parenchymal tissue tract (white open arrows) from spleen to left testicle.



Figure 2c: CT axial image at the level of scrotum showing splenic parenchymal tissues (white open arrow) and testicular parenchymal tissue (black open arrow) in left scrotal sac.



Figure 3a During laparotomy gross photograph showing continuous splenic parenchymal tissue tract (black open arrow) from spleen to left testicle.



Figure 3b: During laparotomy gross photograph showing continuous splenic parenchymal tissue tract (black open arrow) with left testicle (black solid arrow).

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Figure 2c: CT axial image at the level of scrotum showing splenic parenchymal tissues (white open arrow) and testicular parenchymal tissue (black open arrow) in left scrotal sac.

Figure 3a: During laparotomy gross photograph showing continuous splenic parenchymal tissue tract (black open arrow) from spleen to left testicle.

Figure 3b: During laparotomy gross photograph showing continuous splenic parenchymal tissue tract (black open arrow) with left testicle (black solid arrow).