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Case Report

Surgery Section

Splenogonadal Fusion Presenting as Acute Scrotum: A Case Report

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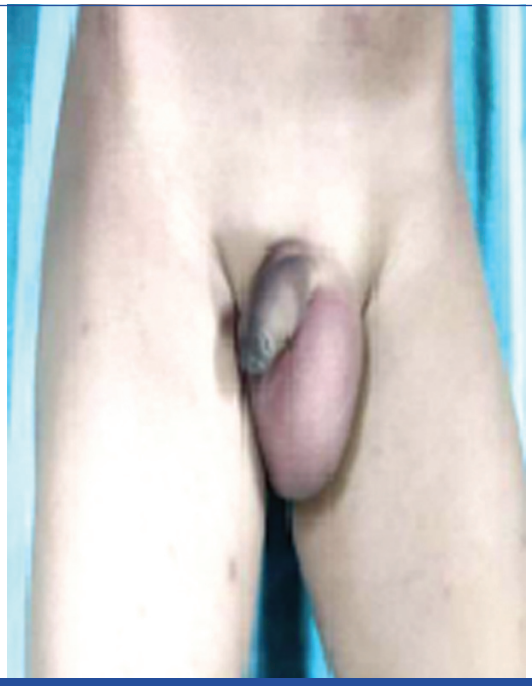
ABSTRACT

Splenogonadal Fusion (SGF) is a rare congenital anomaly characterised by abnormal attachment of splenic tissue to the gonads. We report the case of a five-year-old male child presenting with an acute scrotum, initially suspected to be testicular torsion. During emergency surgical exploration of the scrotum, a gangrenous structure was discovered. This structure was connected to the left testis by a fibrous band that had undergone torsion. It also had another fibrous band at its opposite pole, with the fibrous band extending into the peritoneal cavity. Later, ultrasound confirmed its connection to the spleen. Histopathological examination confirmed the diagnosis of SGF. The patient had an uneventful postoperative course, and follow-up imaging showed normal testicular vascularity. SGF is often misdiagnosed due to its rarity, and early diagnosis through imaging and histopathology is critical in avoiding unnecessary orchiectomies and preserving gonadal function. This case underscores the need to consider SGF as a possibility whenever unusual findings are encountered during scrotal exploration.

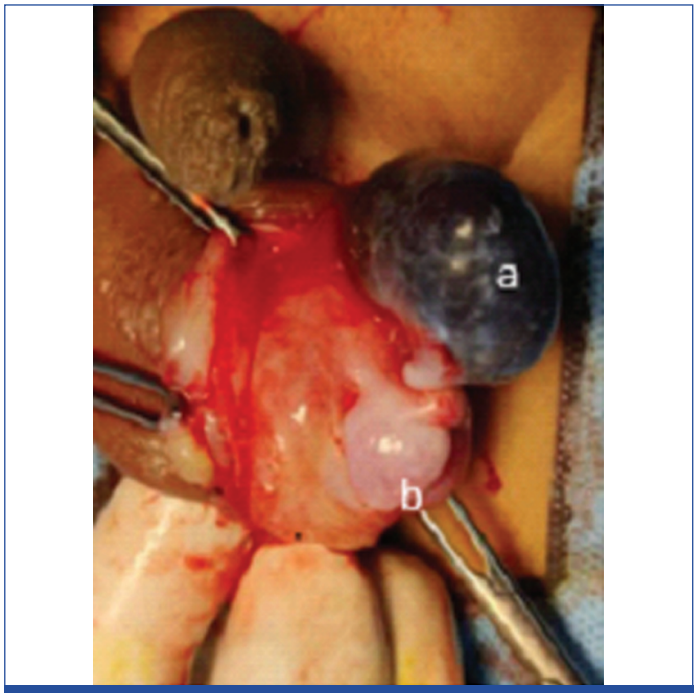
Keywords: Ectopic splenic tissue, Embryological anomaly, Paediatric scrotal emergency, Rare tissue in scrotum, Torsion testis

CASE REPORT

A five-year-old male presented to the institution with severe scrotal pain for one day following trauma to the scrotum while skating. He also had swelling that gradually increased. He had no similar episodes in the past. On examination, swelling was noted over the left hemiscrotum. The scrotum was oedematous and erythematous, and the left testis was enlarged, measuring 3×3 cm, firm and tender. The left testis was found to be slightly elevated [Table/Fig-1]. Ultrasound and color Doppler were suggestive of torsion of the testis. Emergency scrotal exploration was performed. Intraoperatively, torsion of an unusual structure was observed at its pedicle connecting to the testis. This structure appeared gangrenous, with three clockwise twists [Table/Fig-2]. The abnormal structure was connected to the testis via a fibrous band at its lower end, while its upper end had a fibrous connection that disappeared into the peritoneal cavity, where ultrasound later confirmed its connection to the spleen. The entire



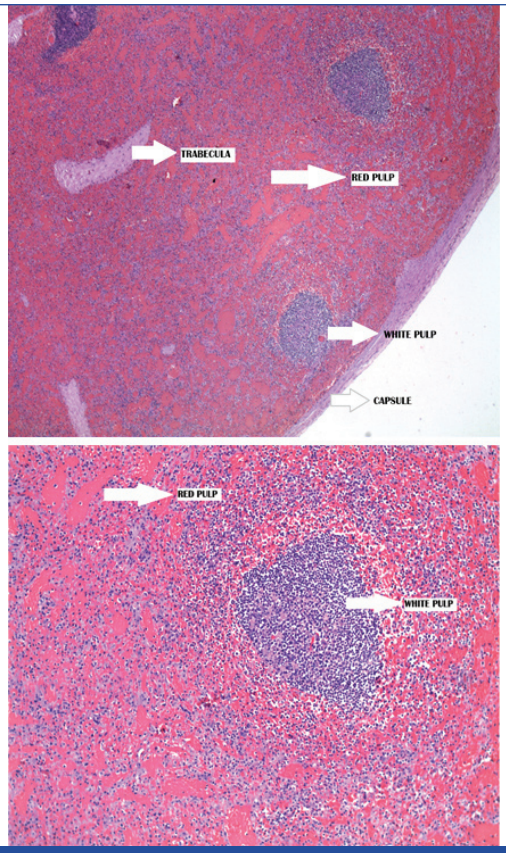
[Table/Fig-1]: Acute scrotum on clinical evaluation.



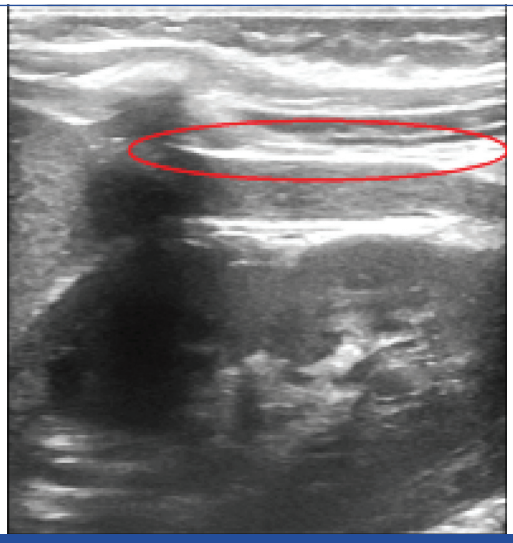
[Table/Fig-2]: a) Splenogonadal fusion with torsion at pedicle and gangrenous splenic tissue b) left testis.

gangrenous tissue was excised, leaving the left testis intact, and the specimen was sent for histopathological evaluation. Grossly, the excised tissue resembled kidney, liver, or spleen. Histopathology revealed splenic tissue with congested sinusoids [Table/Fig-3]. No gonadal tissue was identified. Prophylactic orchiopexy was performed on the contralateral side. On the affected side, the testis was found to be fixed to the surrounding soft tissues, with no risk of further torsion, and thus no additional procedure was performed.

The child had a smooth and speedy postoperative recovery. He was discharged on postoperative day 2 in an alert, active, and stable condition. The surgical wound was healthy and healing well. At a review visit a week later, a follow-up scan showed normal vascularity of both testes and an elongated soft-tissue band extending from the lower pole of the spleen to the left iliac fossa, likely representing a continuous form of SGF [Table/Fig-4].



[Table/Fig-3]: Histopathology specimens tissue resembling spleen showing white pulp and red pulp (H&E, 4x and 40x)



[Table/Fig-4]: Postoperative Ultrasonography (USG) abdomen: Soft-tissue band extending from lower pole of spleen.

DISCUSSION

The SGF is an uncommon congenital anomaly in which splenic tissue is abnormally connected to the gonads or remnants of the mesonephros. It is often detected incidentally during procedures such as orchiopexy or hernia repair, or occasionally during autopsy. This condition is frequently misdiagnosed due to its rarity [1]. SGF is classified into two types: continuous, where a direct anatomical connection exists between the spleen and gonad, and discontinuous, where no direct connection is present. In the continuous type, a cord connects the ectopic and orthotopic spleen, which may consist of splenic tissue, beaded splenic nodules, or fibrous tissue. This cord can follow a retroperitoneal or transperitoneal route and may cause bowel obstruction. In the discontinuous type, ectopic splenic tissue fuses with the gonad [2]. The exact cause of SGF remains unclear, although it is believed to arise from an abnormal connection between the gonads and the spleen during embryonic development [3].

It is believed to occur during the fifth to sixth weeks of gestation, prior to gonadal descent. Around the fifth week, the stomach rotates to the left and moves away from the median plane, bringing the spleen and gonadal tissues into close proximity. This process may result in the fusion of the developing genital ridge with the splenic outline, explaining the frequent left-sided occurrence of SGF [4]. Some researchers suggest that the migration of splenic cells via the retroperitoneal route, possibly influenced by an unknown teratogenic agent, could contribute to this anomaly [4]. To date, no environmental factors have been directly associated with this condition. Additionally, a hereditary autosomal recessive pattern has been proposed as a potential cause [4].

In the present case, the fusion anomaly was observed on the left side, consistent with the higher prevalence of left-sided SGF [5]. Histopathological evaluation played a pivotal role in the definitive diagnosis of the condition. While imaging and intraoperative findings may raise suspicion of SGF or other anomalies, only histopathology can confirm the nature of the tissue. This is especially crucial for distinguishing splenic tissue from other possible ectopic tissues, such as renal tissue, and for ruling out neoplastic processes, highlighting the importance of postoperative tissue examination in identifying rare anomalies. This not only rules out other differential diagnoses, such as ectopic renal tissue or malignancy, but also provides valuable insight into the embryological basis of the anomaly [6].

The SGF is often discovered incidentally during procedures for inguinal hernia, undescended testis, or testicular masses [7]. In this case, the anomaly was identified during emergency scrotal exploration for torsion of the left testis. The most frequent clinical presentations in males include scrotal swelling, inguinal hernias, and cryptorchidism [8]. What makes our case notable is that it is among the rare instances presenting as an acute scrotum, initially suspected to be testicular torsion.

Preoperative or intraoperative identification of this condition, supported by imaging modalities such as ultrasound and Doppler studies, can guide surgical decision-making. A postoperative ultrasound on the review visit is recommended to confirm normal testicular vascularity and evaluate the anatomy and morphology of the spleen. Additionally, the presence of any residual connecting band or ectopic tissue should be documented. The continuous type of SGF, as seen in this case, often involves a fibrous or vascular connection between the ectopic and orthotopic spleen. This condition can be associated with other congenital abnormalities in about 30–33% of cases, including limb defects, micrognathia, heart defects, and cleft palate [9]. However, no such associated anomalies were noted in the current patient. Although bowel obstruction is reported in some cases of continuous-type SGF [10], no such complication was observed here.

Due to the rarity of SGF and the lack of specific clinical or imaging characteristics, the condition is frequently missed. As a result, male patients are sometimes subjected to unnecessary orchiectomies. A review of 123 reported cases of SGF indicated that 37% of these patients had undergone orchiectomy [11]. Awareness of this rare congenital anomaly is crucial, as misdiagnosis can lead to unnecessary orchiectomy, resulting in avoidable loss of testicular function.

CONCLUSION(S)

The SGF is a rare congenital anomaly that can present with acute scrotum, as demonstrated in this case. Early recognition and appropriate management, including careful intraoperative evaluation and histopathological confirmation, are essential in preventing misdiagnosis and preserving gonadal function. This case highlights the importance of considering SGF in the differential diagnosis of acute scrotum to avoid unnecessary orchiectomies and ensure optimal outcomes for the patient.

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