

CASE REPORT

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Continuous splenogonadal fusion in a pediatric patient with bilateral cryptorchidism: A case report

Anjali Borsum, Emmett Kennady, Nora Kern

ABSTRACT

Introduction: Splenogonadal fusion (SGF) is a rare condition characterized by abnormal fusion of splenic and gonadal tissue with fewer than 200 cases reported in the literature. While ipsilateral cryptorchidism is an established association in males, bilateral cryptorchidism is exceptionally rare with only a handful of documented cases.

Case Report: A 5-week-old male infant was evaluated and followed in clinic for bilateral undescended testes with the right testis initially being palpable and the left testis being nonpalpable. Later both testes were found to be nonpalpable. Diagnostic laparoscopy at ten months of age revealed a healthy intra-abdominal right testis and an abnormal left intra-abdominal testis attached to an elongated tail of the spleen extending into the pelvis, consistent with continuous SGF. Given the abnormal appearance of the left gonad and the importance of preserving at least one functional testis, a first-stage laparoscopic Fowler–Stephens orchidopexy was performed on the right and definitive management of the left testis was deferred. At a second operation five months later, the right testis remained viable and was successfully brought into the scrotum. After freeing the left testis from the spleen, the testis lacked spermatic vessels and vas and was found to have limited mobility despite extensive dissection. Because the testis could not be positioned within the scrotum, a left orchiectomy was performed. The pathology demonstrated an immature testis with a predominantly Sertoli cell pattern.

Conclusion: Splenogonadal fusion is a rare condition associated with cryptorchidism. Bilateral cryptorchidism associated with SGF is exceedingly rare and may reflect a broader developmental anomaly beyond simple mechanical tethering.

Keywords: Cryptorchidism, Orchidopexy, Splenogonadal fusion, Undescended testis

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INTRODUCTION

Splenogonadal fusion (SGF) is a rare condition where there is a connection between the spleen and the gonad with fewer than 200 cases reported in the literature [1–3]. There are two types of SGF including continuous, where there is tissue linking the spleen and gonad, and discontinuous where there is splenic tissue fused to the gonad without a structural connection. Continuous SGF is more associated with congenital anomalies [1, 2]. About one-third of SGF cases have additional defects including limb defects, micrognathia and microstomia, anal atresia, diaphragmatic hernia, and/or ventricular septal defect [3, 4]. Literature shows that SGF occurs more commonly in males compared to females, but this may be underdiagnosed due to inaccessibility of female gonads [3, 5]. There are no known diagnostic tests for SGF, and SGF is based on clinical, imaging, intraoperative, and histologic findings.

The most common presentations of SGF in males are a scrotal mass, inguinal hernia, scrotal swelling, and/or

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cryptorchidism [4, 6]. Ipsilateral cryptorchidism is common because the testis is fused to the spleen, most often intra-abdominally. Bilateral cryptorchidism, however, is a particularly rare association with SGF with only a handful of documented cases reported [2]. This rare association raises the possibility of a broader developmental anomaly beyond local mechanical effects alone.

In this report, we describe a case of continuous splenogonadal fusion discovered during evaluation of bilateral nonpalpable testes in a 15-month-old boy. To our knowledge, this represents one of a few reported cases of SGF associated with bilateral cryptorchidism.

CASE REPORT

A 15-month-old male initially presented to pediatric urology at five weeks of age for evaluation of penile adhesions following office circumcision. During this visit, examination revealed a nonpalpable left testis and a right testis located within the superficial inguinal pouch. The patient returned for follow-up at five months of age, at which time both testes were nonpalpable. Given this finding, surgical intervention was recommended.

At ten months of age, the patient underwent diagnostic laparoscopy with possible orchidopexy. Intraoperatively, the right testis was identified in an intra-abdominal position and appeared healthy enough for preservation; however, it could not be advanced into the inguinal canal because of tethering by the spermatic vessels. The left testis was located intra-abdominally posterior to the internal inguinal ring and appeared elongated and smaller than the contralateral gonad. At that time, a diminutive vas deferens was thought to be identified. Notably, the left testis was attached to an elongated tail of the spleen that extended into the pelvis, consistent with splenogonadal fusion. Given the abnormal anatomy of the left gonad and the importance of preserving at least one functional testis, the decision was made to proceed with a first stage Fowler–Stephens orchidopexy on the right side and defer definitive management of the left testis until viability of the right gonad could be established (Figure 1).

The patient was seen one month postoperatively and recovered without complication. Plans were made for completion of the second-stage right orchidopexy and definitive management of the left testis.

At 15 months of age, the patient returned to the operating room for second-stage right Fowler–Stephens orchidopexy and plans for first-stage orchidopexy of the left intra-abdominal testis. The right testis remained small but viable and was successfully mobilized and secured within the scrotum. Evaluation of the left side again demonstrated an abnormal intra-abdominal testis adherent to the spleen. After freeing the testis from the spleen, there was no meaningful vas deferens identified. A pink fleshy fibrous attachment was seen to attach to the testis and proceeded behind the bladder in place of the vas. Assessment of potential mobility in the scrotum

demonstrated that the gonad could reach just beyond the contralateral midline bladder. Due to concerns that the testis would not survive or reach the scrotum, the parents were informed of the findings via telephone call, and a joint decision was made to perform a left orchiectomy. Hemostasis was confirmed following separation of the gonad from the splenic tissue. The patient tolerated the procedure well and recovered without complication. Histopathologic evaluation of the specimen demonstrated an immature testis with a predominantly Sertoli cell pattern (Figure 2).

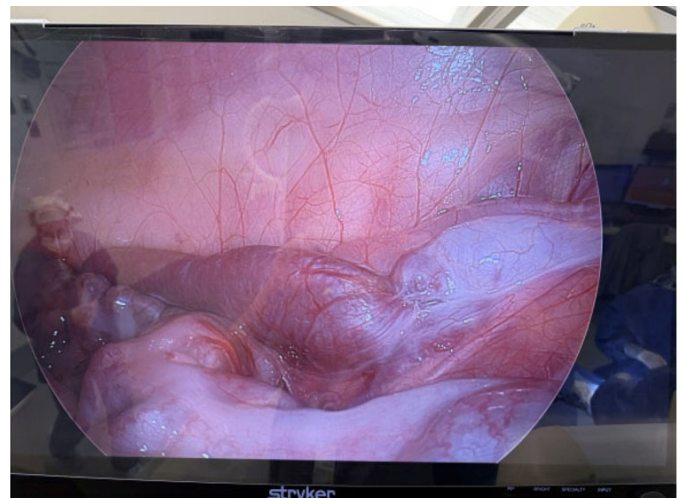


Figure 1: Elongated splenic tissue stretched into the pelvis attached to the left gonad.

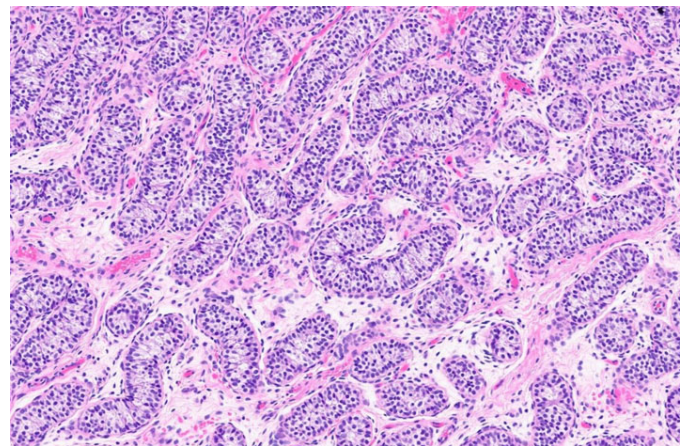


Figure 2: Histology of seminiferous tubules from specimen demonstrating predominant Sertoli pattern.

DISCUSSION

While ipsilateral cryptorchidism is reported in up to one-third of patients with SGF, bilateral cryptorchidism remains exceptionally rare. The rarity of bilateral involvement suggests that factors beyond mechanical tethering may contribute to testicular maldescent in

these patients. It has been proposed that bilateral cryptorchidism associated with SGF represents a broader developmental field defect occurring during early embryogenesis [1].

While exact etiology of SGF is unknown, when associated with ipsilateral cryptorchidism, mechanical tethering of the gonad to the spleen is the most likely hypothesis [2]. For bilateral cryptorchidism, mechanical tethering cannot fully explain this and is more likely associated with a developmental field defect affecting multiple systems [3]. It is hypothesized to be due to a malformation occurring during the fifth and tenth week of gestation when the developing gonads are migrating caudally [3, 4]. The gonads come near the fetal spleen, which is formed at the end of the fourth gestational week. Mechanisms may include vascular disruption, genetic mutations, or chromosomal abnormalities that could be due to a teratogenic disruption during this critical window [3, 4].

The clinical presentation of SGF is often seen as cryptorchidism, and preoperative diagnosis of SGF is often challenging because the condition is often diagnosed visually during surgery [5]. Ultrasound can be helpful in the diagnosis with visualization of the testis adjacent to or continuous with a solid mass with echogenicity similar to splenic tissue [6]. Some reviews recommend Technetium-99m sulfur colloid scintigraphy as a more reliable diagnostic tool demonstrating uptake in the orthotopic spleen and ectopic splenic tissue in the scrotum or inguinal region [7], but SGF may be unidentifiable when the testis is intra-abdominal.

Management of SGF typically includes surgery, and a common clinical consequence of failure to recognize SGF preoperatively is unnecessary orchiectomy [1, 8]. Surgical management of SGF should prioritize preservation of viable gonadal tissue whenever possible. Historically, many patients underwent orchiectomy because SGF was mistaken for a testicular neoplasm [8]. Increased awareness of the condition has reduced unnecessary gonadectomy, particularly when SGF is recognized intraoperatively. In the present case, preservation of the left gonad was planned; however, the testis was severely abnormal, lacked identifiable spermatic vessels and vas, and demonstrated minimal mobility despite extensive dissection, leading to the unlikely ability to position the testis within the scrotum. Given these findings and the successful preservation of the contralateral testis through staged Fowler–Stephens orchidopexy, orchiectomy was determined to be the most appropriate management strategy. Furthermore, the pathology of the abnormal testis demonstrated an immature testis with a predominantly Sertoli cell pattern, likely reflecting the effects of longstanding cryptorchidism and abnormal gonadal development. The absence of normal germ cell maturation indicates that the testis would not support spermatogenesis [9]. These findings support the decision to perform orchiectomy because the testis would not have meaningful fertility potential.

The case presented represents continuous SGF, characterized by a direct connection between splenic and gonadal tissue [1]. Continuous SGF accounts for approximately half of reported cases and is more frequently associated with congenital anomalies than the discontinuous form [1, 8]. Interestingly, our patient did not demonstrate other major congenital abnormalities besides the presence of bilateral cryptorchidism.

As mentioned, there are only a handful of cases of SGF with bilateral cryptorchidism reported. One case described bilateral cryptorchidism with splenic tissue attached to the left gonad and adrenal tissue attached to the right gonad [10]. Another report describes SGF diagnosed in an adult with bilateral cryptorchidism [11]. More recently, there is a case of discontinuous SGF described in a pediatric patient presenting with bilateral empty scrotal sac [12]. Our case is unique because it demonstrates continuous SGF with elongated splenic tissue extending into the pelvis and required staged management to preserve the contralateral testis, which is a presentation not described in the literature thus far.

CONCLUSION

Splenogonadal fusion is a rare anatomical variation in children with cryptorchidism. Preoperative awareness and appropriate imaging can prevent unnecessary surgery and unnecessary gonad loss, especially in the bilateral setting where both testes are at risk. However, given the rarity, most cases will likely be diagnosed incidentally, and the implications on testis health and fertility should be considered.

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Author Contributions

Anjali Borsum – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Emmett Kennady – Conception of the work, Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content,

Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Nora Kern – Conception of the work, Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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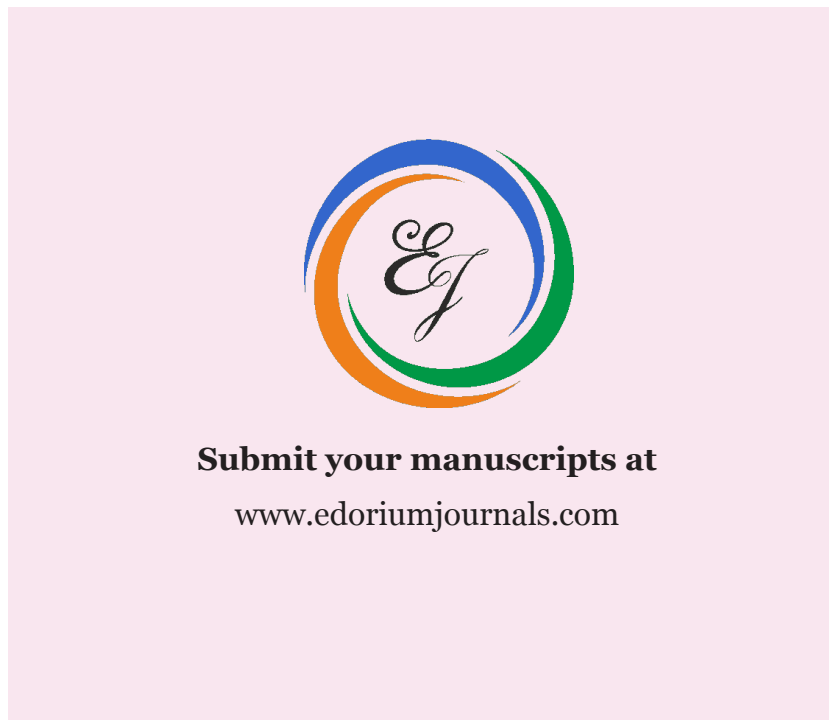
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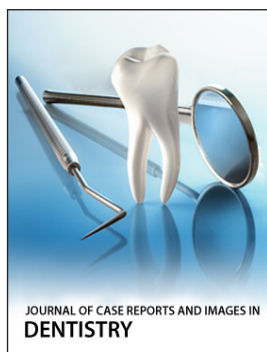
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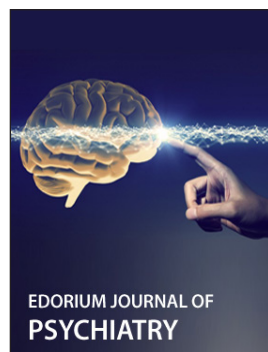
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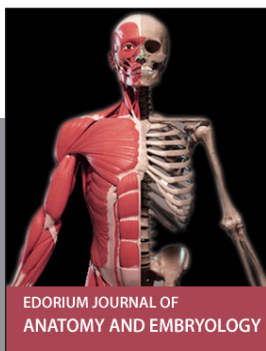
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