

# Laparoscopic Diagnosis and Management of Splenogonadal Fusion: Case Report and Review of Literature

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## ABSTRACT

Splenogonadal fusion is a rare congenital anomaly in which there is fusion of the spleen and the gonad or mesonephric derivatives. Approximately, 150 cases have been reported since the condition was first described by Bostroem in 1883.

The diagnosis of this uncommon anomaly is rare even to be suspected preoperatively; I describe a case in which laparoscopic diagnosis and management has been done and review of the literature.

**Keywords:** Splenogonadal fusion, Laparoscopic management of splenogonadal fusion.

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## INTRODUCTION

Splenogonadal fusion is a rare entity with approximately 150 cases reported since the first description of this entity in 1883 by Bostroem. Close proximity of the spleen and gonad during early embryological development allows fusion, whether continuous or discontinuous, of these seemingly unrelated organs. The continuous type of splenogonadal fusion describes the gonad attached to the anatomic spleen. The discontinuous type consists of gonadal fusion with an accessory spleen or ectopic splenic tissue. The diagnosis of this uncommon anomaly is rare even to be suspected preoperatively. Laparoscope is more diagnostic than the ultrasound, computed tomographic (CT) scan, magnetic resonance imaging MRI and helpful in the management. I present a case of continuous splenogonadal fusion presenting as an impalpable left testicle. This case is unique in that the laparoscopic management in such condition after negative open groin exploration.

## CASE PRESENTATION

An 11-year-old boy had impalpable left testis since birth. He had ultrasound, CT scan examinations and open groin exploration which revealed no left testis. Physical examination, apart from the left groin scar and the impalpable left testis, was unremarkable. Routine preoperative laboratory investigations were within normal range.

On laparoscopic exploration, a reddish brown, smooth cord of tissue measuring about 20 mm in diameter was observed to be coming from above to down in a peritoneal fold ending by fusion to the superior pole of the testis intra-abdominal higher to the internal ring of the inguinal canal (Fig. 1).

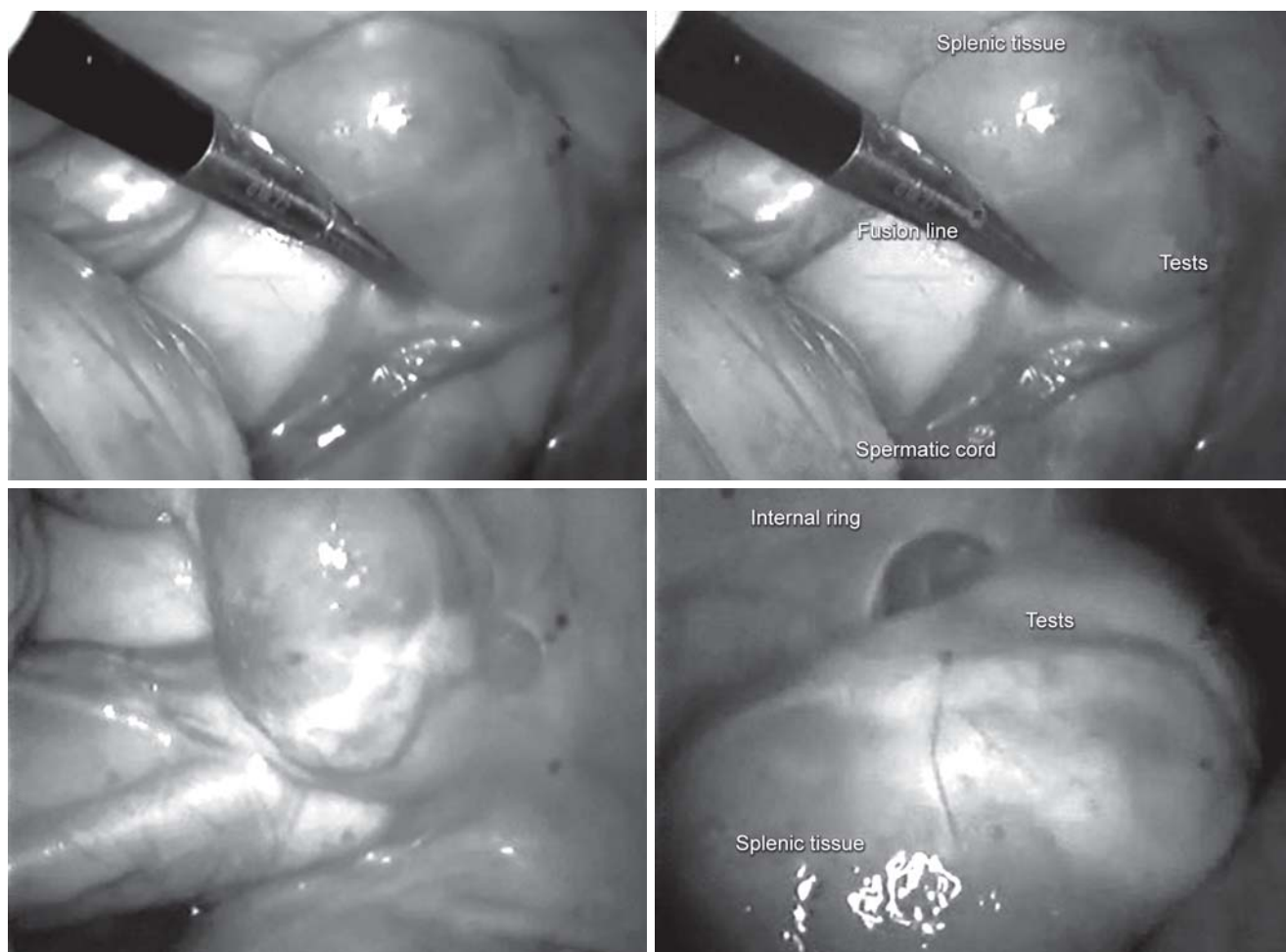
Grossly, the tubular cord had the appearance of splenic tissue having a serosal capsule and fibrous trabeculae and a vascular pedicle running on its medial aspect (Fig. 2). The splenic cord-like tissue fused with upper pole of the testis, there was a line of demarcation between the different tissues. Laparoscopic-assisted left orchidopexy with preservation of the spleen was then performed. He has uneventful postoperative follow-up for 1 year.

Histopathological examination confirmed that specimen was splenogonadal fusion. There was no evidence of malignance.

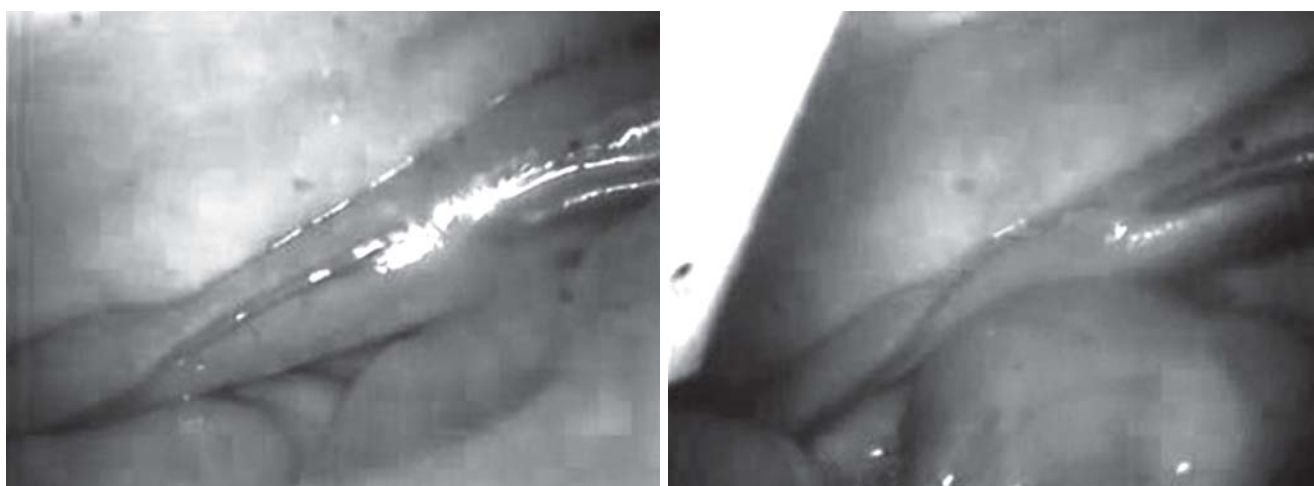
## DISCUSSION

This case presents an unusual presentation of splenogonadal fusion. The case is unique in that it was diagnosed and managed with laparoscope. The meta-analysis of published reports of 111 boys with splenogonadal fusion found that 31% had cryptorchidism. Of these, 59% were bilateral, 26% had right intra-abdominal testes and 65% had left intra-abdominal testes. Of those with continuous splenogonadal fusion, 44% had cryptorchidism. Solely cryptorchid cases with splenogonadal fusion reportedly had bilateral absence of legs, imperforate anus, spina bifida, diaphragmatic hernia and hypospadias.<sup>1</sup>

About 120 to 150 cases of splenogonadal have been reported in the literature. Splenogonadal fusion is most commonly an incidental discover during a routine groin exploration for an undescended testis or hernia. While in our case, the surgeon how had done the open groin exploration could not find the testes because the testes and the cord were totally high intra-abdominal. Nearly, 17% of the splenogonadal fusions were diagnosed at autopsy.<sup>2</sup> Testicular or inguinoscrotal swelling was the most common presentation.<sup>3</sup> In this case, no scrotal or inguinal swelling farther more; there was a scare of negative open groin exploration. Karaman and Gonzales, 37% of 137 cases underwent an unnecessary orchiectomy because of suspicion



**Fig. 1:** These are multiple pictures of splenogonadal fusion



**Fig. 2:** This is splenic tissue with vascular pedicle on its surface

of a primary testicular neoplasm.<sup>2</sup> Only four were reported with a malignant testicular neoplasm and a coexistent splenogonadal fusion. Other presentations include that of an acute painful scrotal swelling secondary to affection of the ectopic splenic tissue by various processes. Talmann<sup>4</sup> and Settle<sup>5</sup> reported cases presented with acute scrotal pain and swelling secondary to malaria involvement of the

ectopic splenic tissue. These patients' symptoms subsided as the malaria resolved. Acute torsion of the splenic tissue,<sup>2</sup> mumps, leukemia and mononucleosis<sup>6</sup> and traumatic rupture of the ectopic spleen<sup>7</sup> also presented as painful scrotal swellings. Mechanical bowel obstruction by the intraperitoneal cord of the continuous splenogonadal fusion was described by Hines and Eggum.<sup>8</sup> Sripathi<sup>9</sup> one case of

macroorchidism was reported. Few cases were diagnosed preoperatively. One of those was reported by Kadlic<sup>10</sup> in 1943. Three cases were diagnosed by 99mTc-sulfur colloid liver-spleen scan, one of them during workup of a patient with an undescended left testicle and associated limb malformations,<sup>11</sup> and two cases during evaluation of intra-abdominal mass.<sup>12,13</sup> Patel<sup>14</sup> diagnosed one case by ultrasonography when he followed a tubular process arising from the upper pole of the spleen down to the upper pole of a left undescended testis. Our case has the same anomalies of Patel case but it cannot be suspected or diagnosed by ultrasound or CT scan prior to the previous surgery had done. He also noted movement of the upper splenic pole when applying traction to the testis. The left side is far more commonly involved than the right side. Only three cases (2%) had a discontinuous right-sided splenogonadal fusion and were all male.<sup>15,16</sup> Half of the cases presented below 10 years<sup>3</sup> and 82% below 30 years.<sup>2</sup> It is predominant in male. Male-to-female ratio is about 1:16.<sup>3</sup>

Two forms of splenogonadal fusion have been described, continuous and discontinuous. The continuous form occurs when the anatomic spleen is connected by a discrete cord to the gonad. The discontinuous form consists of a fused splenogonadal structure that has lost continuity with the main spleen. This is a variant of an accessory spleen. The continuous type seems to be predominant.<sup>17</sup> Our case is continuous type of splenogonadal fusion. A column of splenic tissue comes out from the upper pole of the spleen and passing downward anterior to the anterior splenic border, swing to the left over the splenic flexure of the colon, then passes through left paracolic gutter to fuse with the left testis in the abdominal cavity.

Two theories have been proposed to describe splenogonadal fusion. Von Hochstetter attempted to explain this entity by a retroperitoneal pathway for the splenic angle to come into contact with the developing gonad. In this theory, the splenic cells could potentially be found along the pathway of gonadal descent.<sup>18,19</sup> Sneath<sup>20</sup> proposed that inflammation over two opposing peritoneal surfaces, namely, the gonadal ridge and spleen, could cause fusion. During gonadal migration, the peritonealized adhesion would lengthen and develop as a cord continuous with the spleen or rupture during development, making it discontinuous with the spleen.<sup>18</sup> Because of the rarity of this condition it is infrequent to be diagnosed preoperatively.<sup>21</sup> Techniques of diagnostic imaging is available if there is a clinical suspicion of splenogonadal fusion. The most reliable preoperative imaging, according to published results, is technetium isotope scanning, which detects accessory splenic tissue.<sup>21</sup> Laparoscopic diagnosis of impalpable testes is superior to all investigation including ultrasound, CT scan,

or even MRI.<sup>22</sup> Laparoscope was valuable and highly effective not only in the diagnosis but also in the management of this case. It should be pointed out that orchiectomy has been performed needlessly,<sup>21</sup> the unique in this case is the use of laparoscope in the diagnosis and management. The search of the database shows there is laparoscopic use in splenogonadal fusion.

## CONCLUSION

Splenogonadal fusion is a rare condition, seldom to be malignant. Diagnostic imaging has a limited role in the evaluation of boys with undescended testes and it is related condition. I recommend that efforts be developed to increase routine use of laparoscope in the evaluation of a boy with cryptorchidism. Laparoscope is essential for diagnosis and management of simple, complex and rare anomalies associated with undescended testes such as splenogonadal fusion.

## REFERENCES

1. Cartes D, Thorup JM, Visveld J. The pathogenesis of cryptorchidism and splenogonadal fusion: A new hypothesis. *Br J Urol* 1995;77:285-90.
2. Karaman MI, Gonzales ET Jr. Splenogonadal fusion: Report of 2 cases and review of the literature. *J Urol* 1996;155:309-11.
3. Carragher AM. One hundred years of splenogonadal fusion. *Urology* 1990;35:471-75.
4. Talmann IM. Nebenmilzen in nebenhoden und somenstrang. *Virchows Arch Pathol Anat* 1926;259:37.
5. Settle ER. The surgical importance of accessory spleens with a report of 2 cases. *Amer J Surg* 1940;50:22.
6. Andrews RW, Copeland DD, Fried FA. Splenogonadal fusion. *J Urol* 1985;133:1052-53.
7. Halvorsen JF, Stray O. Splenogonadal fusion. *Acta Paed Scand* 1978;67:379.
8. Hines JR, Eggum PR. Splenic-gonadal fusion causing bowel obstruction. *Arch Surg* 1961;83:887-89.
9. Sripathi V. Macro-orchidism may be an indicator of continuous splenogonadal fusion. *BJU Int (England)* 1999 Oct;84(6):733-34.
10. Kadlic T. Nebenmilz in einer angeborenen Skrotalhernie. *Zentralbl All Pathol* 1943;81:49.
11. Guarin U, Dumitrieva Z, Ashley SJ. Splenogonadal fusion—rare congenital anomaly demonstrated by 99mTc-sulphur colloid imaging: Case report. *J Nucl Med* 1975;16:922.
12. Falkowski WS, Carter MF. Splenogonadal fusion associated with an anaplastic seminoma. *J Urol* 1980;124:562-64.
13. Steinmetz AP, Rappaport A, Nikolov G, et al. Splenogonadal fusion diagnosed by spleen scintigraphy. *J Nucl Med (United States)* 1997;38(7):1153-55.
14. Patel RV. Splenogonadal fusion. *J Pediatr Surg* 1995;30:873-74.
15. Gordeef M, Cuenant J. Rate surnumeraire a localisation scrotale. *Maroc Med* 1951;30:744-45.
16. Tiberio G, Colombo F, Panizzari GP. A Proposito di un caso di ectopia splenica nell'emiscroto destro (fusione gonadosplenica). *Chirurgia* 1965;20:193.

17. Putschar WGJ, Manion WC. Splenic-gonadal fusion. *Am J Pathol* 1956;32:15-28.
18. May JE, Bourne CW. Ectopic spleen in the scrotum: Report of two cases. *J Urol* 1974;111:120-23.
19. Von Hochstetter A. Milzgewebe im linken ovarium des linken individualteiles eines menschlichen thoracopagus. *Virchows Arch* 1953;324:36.
20. Sneath WA. An apparent third testicle consisting of a scrotal spleen. *J Anat Physiol* 1912-13;47:340.
21. William L, Duncan Jra, Mark A. Barraza. Splenogonadal fusion: A case report and review of literature. *J Pedia Surg* 2005;40:E5-7.
22. Gregory E Tasiana, Hillary L Coppb, Laurence S Baskin. Diagnostic imaging in cryptorchidism: Utility, indications and effectiveness. *J Pedia Surg* 2011;46:2406-13.

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