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Case Report

Splenogonadal fusion presenting as a testicular mass, a case report and literature review☆☆☆

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ABSTRACT

Splenogonadal fusion is a rare, frequently misdiagnosed, congenital anomaly in which the splenic tissue is abnormally attached to the gonadal or mesonephric remnants. It is commonly found as an incidental finding at autopsy, during orchiopexy or hernia repair. However, it can present as a testicular mass or as an acute scrotal pathology such as testicular torsion or epididymo-orchitis. It poses as a diagnostic challenge preoperatively and often leads to unnecessary orchiectomy.

We present a case of a 15-year-old male who presented with a long-standing left testicular mass thought to be a testicular tumor. Resection of the lesion along with partial left orchiectomy was done and histopathologic evaluation revealed splenogonadal fusion.

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Introduction

Splenogonadal fusion is a rare and benign congenital anomaly characterized by the presence of ectopic splenic tissue in the scrotum [1,2]. It is most commonly found incidentally upon surgical exploration for an undescended testicle or hernia. Symptomatic patients present with a testicular mass or a picture of acute scrotum similar to testicular torsion or epididymo-orchitis.

Case report

We present a case of a 15-year-old male who presented to the urology service after a testicular mass was found on routine physical exam by his primary care provider. The patient reported the mass to be painless without significant change in size over a year. The patient had a history of bilateral hydrocelectomy at the age of 1-year-old and was otherwise healthy without any family history of testicular disease. Clinical examination demonstrated bilateral descended testicles with Tanner V stage of development. There was an approximately 2 cm

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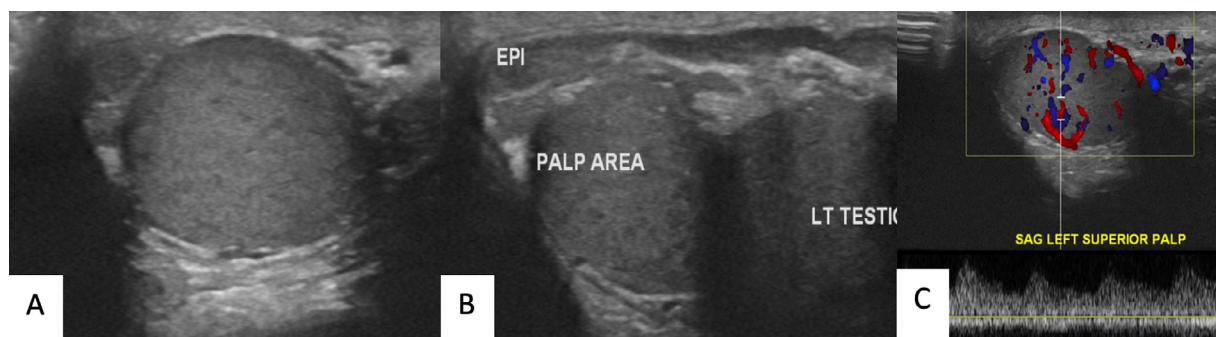


Fig. 1 – (A) Sonographic evaluation of the left hemiscrotum demonstrates a hypoechoic well circumscribed lesion. (B) Sonographic evaluation demonstrating lesion between left epididymis and left testis abutting the testicle. (C) Color doppler evaluation demonstrates venous and arterial blood flow with the lesion.



Fig. 2 – (A) T2 hypointense well circumscribed lesion superior to the left testicle (white arrow). (B) Pre-contrast T1 fat-sat coronal image demonstrating a lesion isointense to the left testicle (red arrow). (C) Post contrast images show enhancement of the lesion (blue arrow).

oval shaped, firm, nontender mass within the left hemiscrotum, superior to and indistinguishable from the left testicle. No inguinal lymphadenopathy was palpated on the exam.

Tumor markers including AFP and B-HCG were within the normal limits.

Sonographic evaluation of the scrotum demonstrated a well-defined 2.1 cm oval-shaped lesion with similar echogenicity and appearance to the left testicle, abutting the upper pole of the left testicle without definite evidence of testicular invasion (Figs. 1A and B). On color doppler, vascular flow was demonstrated within the lesion (Fig. 1C). The remainder of the scrotal sonogram was within normal limits. These findings were reported as a supernumerary testicle with the possibility of a testicular neoplasm.

Follow-up magnetic resonance image (MRI) of the pelvis was obtained with and without intravenous contrast demonstrated a well-circumscribed, 2.6 × 2.3 × 2.9 cm, lesion immediately superior to the left testis. It was hypointense to the adjacent testis on the T2 sequence (Fig. 2A), isointense on T1 fat sat sequences (Fig. 2B) with mild enhancement post contrast administration (Fig. 2C). The remainder of the pelvic MRI was

within normal limits. These findings were reported to likely represent an adenomatoid tumor and less likely supernumerary testicle in the left scrotum.

Surgical exploration of the left hemiscrotum which revealed a large mass arising from the upper pole of the left testicle which was excised along with partial left orchiectomy.

Histopathological evaluation demonstrated circumscribed splenic tissue with lymphoid aggregates in a background of highly vascular parenchyma and fibrous trabeculae, consistent with splenogonadal fusion (Fig. 3).

Discussion

Splenogonadal fusion is a rare congenital anomaly characterized by the fusion of splenic tissue and a gonad or mesonephric remnants [3], which was first described in 1833 by Bistrom and about 200 cases have since been reported in the literature [4,5]. About 72% of the cases have been described in individuals younger than 20 years old and 98% of the cases

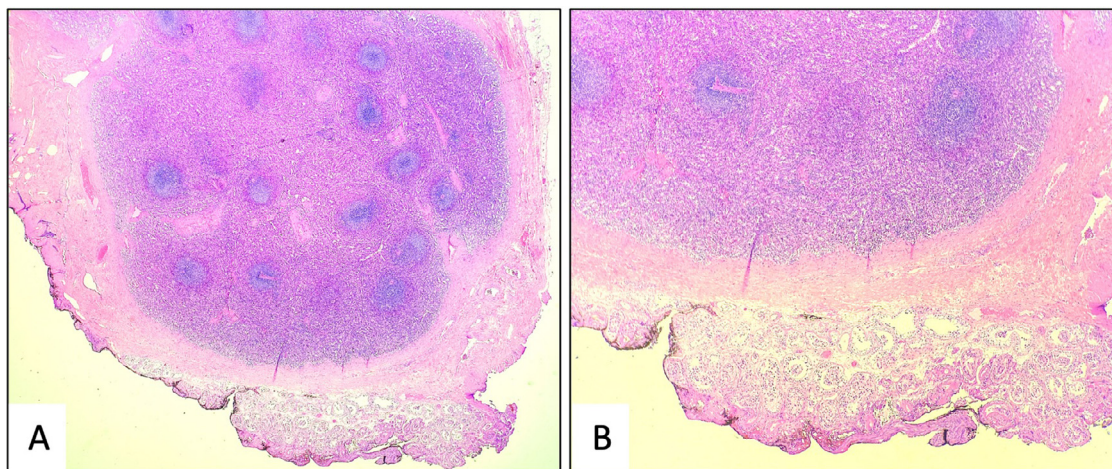


Fig. 3 – (A, B) Hematoxylin and eosin stain (A: 10x, B: 20x) – Low and high-power views of the left scrotal lesion: the circumscribed splenic tissue (with lymphoid aggregates in a background of highly vascular parenchyma and fibrous trabeculae) is shown at the top of the images. Seminiferous tubules are seen at the bottom.

occurring on the left side [5]. Splenogonadal fusion is approximately 16 times more common in males than in females with only 8 cases reported in females in the literature. The discrepancy is thought to possibly be due to the internal nature of the female gonads limiting its accessibility rather than an actual difference in incidence [6].

The exact etiology remains unknown. Theories suggest that fusion occurs between 5 and 8 weeks gestation when the splenic anlage and the gonadal ridge are in close proximity of each other prior to the gonadal descent [7].

Two types of splenogonadal fusion have been described: continuous and discontinuous types. The continuous type has a direct continuous cord-like connection between the “main” spleen and the splenogonadal structure. The discontinuous type is when there is no direct connection between the “main” spleen and the splenogonadal structure [8].

It is most commonly found as an incidental finding during groin exploration for orchidopexy or hernia repair [9,10]. Other presentations include testicular mass and acute testicular pain, mimicking testicular torsion and epididymitis. About 20% of cases have been found on postmortem autopsy [5]. Associated congenital anomalies have been described in 27% of the cases with cryptorchidism being the most common anomaly, often seen with the continuous type [11].

Preprocedural diagnosis of splenogonadal fusion is challenging as it mimics the presentation of any testicular mass. Investigations usually include sonography, computed tomography (CT), MRI and when suspected, 99m-Tc sulfur colloid scintigraphy. Sonography is often the initial imaging modality, most commonly demonstrating hypoechoic extra testicular mass without any specific features. Doppler sonography often demonstrates patterns of central vascularity. CT would demonstrate a homogenous enhancing soft tissue mass without any calcifications, with density values similar to that of the spleen. 99m-Tc sulfur colloid scintigraphy would demonstrate activity in the scrotum consistent with ectopic splenic tissue [11].

Given the rarity of this condition, many patients undergo unnecessary orchiectomy to evaluate for a testicular tumor. Even when confirmed with scintigraphy, some authors still recommend surgical excision of the splenic tissue for confirmation of the diagnosis [10].

Conclusion

Splenogonadal fusion is a rare, often misdiagnosed, benign congenital condition frequently leading to orchiectomy of the affected testicle. Adding this condition to the differential diagnosis list of extra testicular tumors, and when clinical suspicion is high, utilization of 99m-Tc sulfur colloid scintigraphy may salvage the testicle and prevent unnecessary orchiectomy.

Ethics approval

This is a retrospective case report not requiring ethics approval.

Patient consent

The author was unable to obtain written consent from the patient or from the patient's relatives, despite attempts to do so. Because of the public interest in publication, the anonymization of the patient, and that attempts had been made to contact the patient and their relatives, exceptional agreement for publication of the case report was given by the Editor-in-Chief of the journal *Radiology Case Reports*.

Availability of data and materials

Not applicable.

Code availability

Not applicable.

Authors' contributions

All authors contributed to writing the manuscript. All authors read and approved the final manuscript.

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